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Understanding Poorly Understood Chronic Illness:

Lived Experiences, Healthcare Journeys, and Recommendations for Change

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Declaration

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I, Natalie Woodville (formerly Duda), declare that this thesis has not been submitted as an exercise for a degree at this or any other University and it is entirely my own work.

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Extended Abstract

Chronic health conditions present a substantial and growing challenge for healthcare systems worldwide. While much work has focused on improving care for common chronic illnesses, a significant gap remains in addressing the unique needs of patients with poorly understood or "unexplained" conditions. Employing a multi-method, multi-phase design with Patient and Public Involvement (PPI) throughout, this thesis examines the experiences of individuals living with such conditions in Ireland, with a particular focus on their experiences of healthcare. The research programme embraced the collaborative ethos of PPI, capturing the voices of patients both as participants and co-creators, with a view of developing actionable recommendations for change. Four studies were conducted over four phases of research.

Phase one involved a systematic literature of qualitative evidence concerning the experiences of individuals with Medically Unexplained Symptoms (MUS), Fibromyalgia Syndrome (FM) and Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS), and those of healthcare practitioners working with this group of patients. Meta-aggregation of findings from 143 studies resulted in thirteen synthesised findings, with patient and clinician experiences reflecting a common theme of sensemaking at the limits of biomedical knowledge. Highlighting the challenge of medical uncertainty, the review revealed a lack of person-centeredness in healthcare and pointed to the patient-clinician dynamic as a viable target for change.

Research phase two employed a cross-sectional, mixed-method online survey to explore the knowledge, understanding and perspectives on ME/CFS and FM of a sample adults in Ireland, specifically those with prior recognition of either condition, along with their appraisal of public and clinician knowledge and awareness. A total of 319 individuals completed the survey, and results revealed a good level of understanding among individuals familiar with ME/CFS (*n*=238) and FM (*n*=280). Participants typically highlighted core issues of stigma and invalidation and recognised a myriad of healthcare-related challenges faced by individuals living with these conditions. Notably, the results revealed a largely pessimistic outlook on the diagnosis, treatment and management along with overwhelmingly negative views of clinicians' knowledge and awareness of both conditions. Together, findings highlighted a troubling lack of confidence in healthcare among adults in Ireland, underscoring the need to further explore patients' lived experiences in this context.

The third phase of research comprised of an in-depth exploration of the lived experiences and healthcare journeys of individuals (*N*=12) with various poorly understood conditions in Ireland, including FM, Ehlers-Danlos Syndrome, Postural Tachycardia Syndrome, Functional Neurological Disorder, ME/CFS, and Long-COVID among others. Utilising the Life Grid method and a phenomenological focus, the results provided a rich, multidimensional account of the transformative impact of chronic illness on individuals' lifeworlds. Combined with exploratory patient journey maps, accounts illustrated that healthcare experiences are largely disappointing, as individuals struggle to navigate a poorly coordinated, "broken" system that fails to provide multi-disciplinary care. Participants' accounts of being disbelieved and silenced highlighted complex power dynamics that exist between service users and healthcare practitioners, with overall results suggesting that integrated and patient-centered care is lacking.

The fourth phase of research employed focus groups to explore the healthcare-related needs of individuals with poorly understood conditions (*N*=24), with the specific aim of developing recommendations for change. PPI contributors were involved in the collaborative analysis of data collected, further ensuring that research outcomes were aligned with the needs and priorities of patients. Findings revealed that the healthcare experiences of patients with chronic and poorly understood conditions are unsatisfactory, with recommendations most often targeting consultation dynamics and service structure. Results highlighted a fragmented healthcare system, characterised by poor coordination and communication breakdowns, and a lack of comprehensive multidisciplinary care for individuals with complex needs. Generally, a need for holistic and personalized approaches to care and the development of multidisciplinary clinics, networks and specialised diagnostic facilities were emphasised, as was a need for patient partnership in research.

Collectively, this research reveals major gaps in care for this patient population. The research programme has two overarching implications. First, it provides compelling evidence for a need to adopt an integrated, patient-centered model of healthcare delivery; and second, it highlights the need for meaningful, sustained patient and public engagement in research and healthcare design.

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Dedication

To all those finding their voice and speaking their truth

Abbreviations

CCM	Chronic Care model
CRPS	Chronic Regional Pain Syndrome
EDS	Ehlers Danlos Syndrome(s)
EHR	Electronic Health Records
FM	Fibromyalgia syndrome
FND	Functional Neurological Disorder
GET	Graded Exercise Therapy
HCP	Healthcare Professional
hEDS	Hypermobile Subtype of Ehlers-Danlos Syndrome
HRB	Health Research Board
HSD	Hypermobility Spectrum Disorder
HSE	Health Service Executive
IBS/IBD	Irritable Bowel Syndrome/Disease
ICP	Integrated Care Programme
IRC	Irish Research Council
JBI	Joanna Briggs Institute
MCAS	Mast Cell Activation Syndrome
ME/CFS	Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome
MUPS	Medically Unexplained Physical Symptom
MUS	Medically Unexplained Symptom
NES	Non-Epileptic Seizure
NIHR	National Institute for Health Research
OECD	Organisation for Economic Co-operation and Development
PCOS	Polycystic Ovary Syndrome
PCRI	Patient Centered Research Institute
PEM	Post-Exertional Malaise
POTS	Postural Orthostatic Tachycardia Syndrome
PPI	Patient and Public Involvement
PwC	Person with Condition
PwFM	Person with Fibromyalgia Syndrome
PwME	Person with ME/CFS
PwO	Person without Condition
QoL	Quality of Life
RHA	Regional Health Area
SDM	Shared Decision-Making
WHO	World Health Organisation

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Chapter 1. Introduction

1.1 Background

Chronic health conditions, characterized by their persistent and often progressive nature, have become increasingly prevalent, and their management has emerged as one of the biggest challenges facing healthcare systems today (Holman, 2020; McGrail et al., 2016). Projections indicate that the burden of chronic illness world-wide will increase such that by 2048, chronic noncommunicable diseases will account for 86% of the 90 million deaths each year (World Health Organization, 2023). Individuals with chronic conditions frequently face a range of intricate, fluctuating, and worsening symptoms that substantially affect numerous aspects of their daily lives. They might require interventions and follow-up management from multiple specialists and health professionals, community services and social care providers, which necessitates a high level of integrated care (Borgermans et al., 2017; Gavaldà-Espelta et al., 2023; Lennox-Chhugani, 2021; Martínez-González et al., 2014). Thus, it has long been recognised that, as the number of people living with chronic conditions continues to increase, the healthcare system must adapt to provide comprehensive, coordinated, and patient-centered care to address the complex and multifaceted needs of this population (e.g., Epping-Jordan et al., 2004; Wagner et al., 2005; Zulman et al., 2014).

While most research in Ireland has focused on integrated care for patients with acute or common chronic illnesses, such as cardiovascular disease, cancer, and diabetes (e.g., Darker, 2014; Darker et al., 2015), less is known about the care coordination for patients with poorly understood, under-recognised or rare conditions. For these patients, the medical journey is additionally complicated by diagnostic uncertainty and a lack of rudimentary care pathways for care, as well as a disbelief, invalidation, and a lack of awareness by healthcare providers. For example, patients with Ehlers-Danlos Syndromes, a group of poorly recognised connective tissue disorders impacting multiple systems, often face disbelief, a diagnostic odyssey and a fragmented healthcare system that fails to effectively provide the multidisciplinary care their condition requires (Dijk et al., 2024; Halverson et al., 2021; Julier, 2021; Mittal et al., 2021; Ward et al., 2022). This pattern of disbelief and dismissal is well-documented across a number of poorly understood or "contested" chronic conditions, including Fibromyalgia Syndrome (FM), Myalgic

Encephalomyelitis/ Chronic Fatigue Syndrome (ME/CFS) , Irritable Bowel Syndrome (IBS), Postural Orthostatic Tachycardia syndrome (POTS), chronic Lyme Disease, and more recently, Long-COVID (e.g., Björkman et al., 2016; Clutterbuck et al., 2024; Geraghty & Blease, 2019; Kavi, 2021; Kool et al., 2009; Luty, 2018; Morrison et al., 2021). While distinct, these conditions are often subsumed under the umbrella of "medically unexplained" syndromes; or those with ambiguous physical symptoms that cannot be readily attributed to an underlying disease, referred to as Medically Unexplained Symptoms (MUS) (Baloh, 2020).

Generally, the literature suggests that patients with MUS tend to be high utilizers of healthcare and incur substantially higher costs than other patients (Barsky et al., 2005; Bermingham et al., 2010; Burton et al., 2012). As a group, they are also considered unpopular in healthcare settings: negative attitudes are expressed by practicing physicians and medical students, who frequently refer to this group of patients as "frequent flyers", "doctor shoppers", "attention seekers", "heartsink" or "thick folder patients" (e.g., Aiarzaguena et al., 2013; Czachowski et al., 2012; Stone, 2014). Uncertainty regarding aetiology and detection often leads clinicians to conclude that MUS and contested syndromes are psychosomatic, psychogenic, manifestations of hysteria, or are conversion or functional disorders (Bransfield & Friedman, 2019; Ding & Kanaan, 2017). This practice of attributing symptoms to ill-mental health is increasingly recognised as traumatic to patients who feel psychological explanations are stigmatising, offensive, and undermine the physical reality of their illness (Cheston, 2022). Unsurprisingly, this feeds into a conflictual and mutually disappointing patient-clinician dynamic, with communication practices identified as potential intervention targets (Epstein et al., 2006; Houwen et al., 2017; Stortenbeker et al., 2022).

It is noteworthy that a number of other ethical, conceptual and practical issues with MUS have been raised (e.g., Eriksen et al., 2013; O'Leary, 2018), including the clinical utility of the label, which Rasmussen (2020) describes as a "junk drawer category". Tack (2019) also points out that MUS are often reappraised as medically explained at long-term follow up, which raises concerns regarding missed treatment opportunities for patients living with them. The expanding evidence base for pathophysiological mechanisms underlying many so-called unexplained conditions (e.g., Annesley et al., 2024; Clauw et

al., 2024) serves as a reminder that “unexplained” symptoms likely reflect gaps in medical knowledge rather than an absence of organic cause per se.

With this overview in mind, this thesis takes the position that current scientific understanding is incomplete; thus, redressing medically unexplained symptoms and syndromes as “poorly understood” conditions throughout. The remainder of this chapter provides further context to the research, including an overview of the healthcare system in Ireland, integrated care for chronic illness, the central role of partnership with patients, and finally, a rationale for the research and outline of the structure of the thesis.

1.2 Healthcare in Ireland

The Irish healthcare system operates as a mixed, two-tier public-private model, providing services to a population of approximately 5.1 million people (Central Statistics Office, 2023). Primarily funded through general taxation, the system is under the stewardship of the Department of Health, which oversees policy direction and performance. The Health Service Executive (HSE) acts as the statutory body responsible for managing healthcare delivery and implementing health and social care policies.

The Organisation for Economic Co-operation and Development (OECD) reports that recent trends in health among the Irish population are optimistic. In 2019, 84% of the population reported being in good health, though one in three (28%) adults reported having at least one chronic condition (OECD, 2021). In 2020, mortality rates from preventable causes in Ireland were approximately 20% lower than the EU average, and in 2022, 80% of Irish adults reported being in at least good health, the highest proportion in the EU (OECD, 2023, 2024). However, despite this and significant investment in healthcare in Ireland, the organization also reports that Ireland has one of the lowest numbers of hospital beds per capita in Europe, and that lengthy waiting lists and unequal access to care remain critical issues. In 2022, 2.6% of the population reported having unmet needs, a percentage higher than the EU average, attributable to factors such as excessive cost, travel distance and wait times (OECD, 2023). These capacity issues are exacerbated by fragmentation of care and poor coordination between services, an issue that particularly impacts patients with complex multi-disciplinary care needs.

The need for an integrated, single-tier system has been long recognised in Ireland and is identified as a priority target in Irish policy documents for healthcare reform. *Sláintecare* was a landmark initiative in Irish healthcare reform introduced in 2017 by the Houses of the Oireachtas Committee on the Future of Healthcare to address long-standing inequities and inefficiencies in healthcare. At time of writing this thesis, Ireland's national healthcare system began to implement *Sláintecare's* 10-year plan for reform, articulated in the *Sláintecare Implementation Strategy and Action Plan 2021–2023* (Department of Health, 2021). This government policy document sets out an agenda and roadmap for change, outlining two key targets: system fragmentation and the two-tiered structure of healthcare, discussed below.

1.2.1 System Structure

The two-tiered healthcare system in Ireland has been problematic and has been a point of ongoing criticism. Achieving a health service with a single tier, where patients are treated on the basis of need rather than ability to pay, has been a goal for many years and remains a priority of *Sláintecare*, articulated under Programme 2 in the Implementation Strategy (Department of Health, 2021). While most European countries have achieved universal or near-universal coverage of the population of a core set of health services, such as consultations with doctors and hospital care (OECD, 2024), Ireland has been an anomaly in that it has historically failed to move towards Universal Healthcare despite repeated implementation promises (Connolly, 2024; Wren & Connolly, 2019).

The public tier of the system, funded through general taxation and managed by the HSE, provides a comprehensive range of services to the population. All residents of Ireland are eligible to access this tier, with holders of means-tested Medical Cards or GP Visit Cards entitled to either free or subsidized care. The private healthcare tier exists alongside this and is funded through private health insurance and out-of-pocket payments, serving those at the upper end of the socioeconomic distribution (Goldrick-Kelly & Healy, 2018). This translates to stark inequalities in access to healthcare, as patients relying on the public system face long waiting times for diagnostic services, access to specialists or consultants and procedures, whiles those with private insurance can bypass queues in the public sector (Connolly, 2024). In addition to this, users of the private tier benefit

from access to private accommodations in public facilities, leading to concerns over "crowding out" of public patients' access to care in public hospitals (see Keegan et al., 2018, 2022).

The implication of the dual model through which the system operates at present is that healthcare in Ireland is heavily determined by socioeconomic status. For context, in 2019, the OECD reported a gap in the prevalence of chronic conditions by income groups, such that 42% of adults in the lowest income group reported having at least one such condition, as compared to 17% of those in the highest economic group (OECD, 2021). Highlighting the disproportionate impact of the structure on socioeconomically disadvantage groups, the OECD (2023) also reported that while 3.2% of individuals in the lowest income quintile reported wait-time related unmet medical needs, this figure was only 1.1% for those in the highest quintile. Put more directly by Heavey (2019), this model has resulted in patients dying while waiting for public treatment, when they could have been treated and possibly saved if they had private health insurance. However, others have highlighted that inequalities in the system exist for both public and private patients, for example, with access to allied health professionals such as psychologists, physiotherapists and occupational therapists being problematic for both groups (e.g., Darker et al., 2015).

1.3 Integrated Care

Patients in Ireland have long complained of challenges arising from a lack of integration of services, noting poor coordination between and within services in both acute and chronic care in the Irish healthcare system (Darker, 2014; HSE, 2021). Nearly a decade ago, Darker et al. (2015) highlighted a need to adopt a chronic care model, incorporating patient, provider and system-level interventions to improve care for this population nationally.

The Chronic Care Model (CCM), which emphasises the importance of coordinated, patient-centered care, has been recognised as a promising framework for addressing the complex needs of individuals with chronic illness (Wagner et al., 2005). First published in 1996 and revised in 1998 (Wagner, 1998; Wagner et al., 1996), the CCM is an evidence-based framework that aims to transform healthcare from a reactive to a proactive approach, and encompasses a multifaceted strategy involving patient, provider, and

systemic interventions. This model suggests that optimal care is realized when a well-prepared and proactive healthcare team engages with a knowledgeable and empowered patient, who is seen as a partner in their care (Grudniewicz et al., 2023; Holman & Lorig, 2000; Oprea et al., 2010). Shared decision-making is at the core of patient empowerment (R. Anderson & Funnell, 2010) and is particularly important in chronic care due to the significant levels of self-management required (Lenzen et al., 2015; Montori et al., 2006; Wagner et al., 2001).

Studies have linked the successful implementation of this model to enhancements in healthcare processes, improved patient care and health outcomes, enhanced quality of life, as well as reductions in healthcare costs and service utilization (e.g., Coleman et al., 2009; Davy et al., 2015). However, as emphasised by Darker et al. (2015), the implementation of this model has been fraught with difficulties, particularly in healthcare systems that have been historically focused on acute, provider-centric care, including in Ireland. Some progress has been made in recent years, as the HSE has published the *Integrated Care Programme* (ICP) *for the Prevention and Management of Chronic Disease* (HSE, 2017), along with models of integrated care for four prevalent chronic diseases affecting over one million people in Ireland: cardiovascular disease, type two diabetes, chronic obstructive pulmonary disease and asthma. In alignment with *Sláintecare*, the focus of the ICP is to deliver patient-centered, coordinated services across healthcare settings for this population, whilst ensuring care that is safe, efficient, timely and local.

More recently, *A Model of Care for Rare Disease* (HSE, 2021) was published outlining a vision for integrated care, and a pilot study (Ward et al., 2022) has explored the development of a national rare disease care pathway in the context of twenty-nine more prevalent multi-systemic rare conditions, including EDS and Juvenile Idiopathic Arthritis. This report involved patient and clinical specialist stakeholders who collaboratively identified Nurse Specialists, Medical Social Workers, Psychologists and Registry Manager roles as essential in the care pathway and emphasised a need for holistic approaches to care. Beyond this, however, there appear to be a lack of guidelines and defined pathways for care for patients with other poorly understood chronic conditions in Ireland, complemented by a notable lack of epidemiological data.

For instance, aside from an estimate of 6,000 rare diseases in Ireland reported in the Irish National Plan for Rare Diseases 2014-2018 (Department of Health, 2014), true prevalence of these diseases in the country is unknown (Aymé et al., 2015). One estimate claims that over 2,000 people in Ireland have been diagnosed with EDS (Ehlers Danlos Society, 2023), which is likely an under-estimate given that this term is an umbrella term for at least 14 recognised types, some more rare than others (Dijk et al., 2024). There are no available statistics regarding the number of people in Ireland diagnosed with FM and ME/CFS, though one estimate suggests that one in three people live with non-cancer chronic pain (Raftery et al., 2012) and the Irish ME/CFS Association (2024) estimates that before the COVID-19 Pandemic, between 10,000 and 20,000 individuals were living with ME/CFS in Ireland.

1.3.1 Organisational Reform in the Irish Healthcare System

As previously mentioned, an essential component of the *Sláintecare* reform agenda is the system integration strategy. This is articulated under Programme 1, *Improving Safe*, *Timely Access to Care, and Promoting Health & Wellbeing* (Department of Health, 2021). At time of writing this thesis, the HSE has initiated *The Health Regions Organisational Reform* (Government of Ireland, 2023) as part of this strategy, a transformative move with full implementation envisioned by March 2025. Patient and Service User Partnership is central to the structural reform, reflecting broader international trends towards patient-centered care and Patient and Public Involvement (PPI). This reform focuses on improving the coordination and continuity of care through the establishment of six Regional Health Areas (RHAs) to facilitate the delivery of integrated health and social care designed around people and planned around communities. According to this programme,

> ***"Integration will be achieved through the alignment of hospital-and community-based services and through a holistic partnership with GPs, Pharmacists, and the fuller range of health service providers at regional and at national level."*** (p. 10)

1.4 Patient and Public Involvement (PPI)

Patient and Public Involvement (PPI) has increasingly been recognised as a critical in the pursuit of more patient-centric and impactful research. This initiative recognizes that patients and the public possess invaluable insights and perspectives that can enhance the quality and relevance of research and, in the healthcare context, ensure that services meet patient needs (Brett et al., 2014a; Dudley et al., 2015; Edgman-Levitan & Schoenbaum, 2021; Sharma et al., 2017; Shippee et al., 2015). The involvement of patients in research and healthcare design is not only a way of improving quality, however, but a step towards democratizing the process and empowering communities (Frith, 2023). This partnership reflects a marked departure from the paternalistic model dominant in medicine and is at the heart of patient-centered care (Edgman-Levitan & Schoenbaum, 2021; Taylor, 2009).

A number of initiatives have been established internationally to facilitate this engagement. For example, in the United States, the Patient-Centered Research Institute (PCRI) has placed significant emphasis on the active engagement of patients in healthcare research (Frank et al., 2014). In the UK, the National Institute for Health Research (NIHR) INVOLVE advocates for research to be created “with” or “by” users of services, rather than “for”, “about”, or “to” them (INVOLVE, 2012). The Health Research Board (HRB) and Irish Research Council (IRC) have played a pivotal role in advancing PPI in Ireland by co-funding and supporting the nation’s PPI Ignite Network, which was launched in 2021. The network aims to build capacity and promote meaningful and sustained PPI across the Irish research landscape, and provides leadership, resources and training to researchers (Health Research Charities Ireland, 2023). For example, it has co-developed a *Values and Principles Framework* (2022) to guide effective and inclusive PPI, emphasising core values like respect, trust, transparency and empowerment.

1.4.1 Patient Partnership in the HSE

The HSE recently published its *Patient and Service User Partnership Plan* (HSE, 2024), a document representing the outcomes of a co-designed approach to partnership embedded in the national approach to the aforementioned health regions. The plan outlines a structured multi-tiered model of embedding partnership within the national and regional frameworks of the HSE, establishing a Patient and Service User Partnership

Council to contribute through designated representatives and working groups at various levels, including Regional Executive Officers, Integrated Health Areas, Community Healthcare, and Service Levels (Bonar & Johnston, 2024).

According to this plan,

> *"The continued focus on building a better health system must put in place the building blocks for diverse, meaningful, and sustained patient and service user partnerships recognising them as true partners in decisions about care, service design, research, and governance. The agreed patient and service user partnership structures should deliver on patient and service user's preferences, needs, expectations and values, while also cultivating an appropriate culture."* (HSE, 2024, p. 8)

Thus, in line with patient-centred approaches and PPI, this HSE initiative seeks to ensure that meaningful stakeholder input is integrated across all aspects of health service planning and delivery.

1.5 Rationale and Thesis Structure

In the context of a transforming healthcare system and a growing emphasis on patient-centred care, the overarching aim of this thesis is to provide a rich, multi-layered account of the lived experiences of individuals with conditions that are chronic and poorly understood in Ireland. The research programme embraces the collaborative ethos of PPI, capturing the voices of patients both as participants and co-creators, with a view to identify targets for change in healthcare.

The structure of the remainder of this thesis is as follows:

Chapter 2 provides an overview of the foundational assumptions, methodological approaches, and specific methods employed in the thesis as a whole and for individual studies. The chapter introduces the multi-phase research design providing a detailed account of each of the four interconnected phases along with PPI activities embedded throughout.

Chapter 3 presents the results of a Systematic Literature Review (SLR) of qualitative research conducted in Research Phase 1. It examines the experiences and perspectives of individuals living with MUS while focusing on two exemplar conditions, ME/CFS and

FM, as well as that of healthcare practitioners. Together, the literature review serves to provide a justification for subsequent research in the Irish context.

Attending to the knowledge-based stigma, *Chapter 4* presents the results of a cross-sectional survey conducted in Research Phase 2, exploring the perspectives, attitudes and beliefs about ME/CFS and FM and their clinical management among adults familiar with these conditions in Ireland. It also elicits appraisals of public and healthcare professional knowledge and awareness of these conditions, and explores the perceived challenges faced by individuals living with them.

Extending the research to poorly understood conditions more broadly, *Chapter 5* focuses on patient voices, providing an in-depth phenomenological account of their lived experiences and healthcare journeys as part of Research Phase 3. The qualitative research also explores the use of a specific method of life story interviewing, eliciting participant reflections on this approach to research. The discussion focuses on the transformative impact of chronic illness on individuals' lifeworlds, with an emphasis on the inter-relationship between personal and healthcare contexts.

Chapter 6 extends the research focusing on healthcare system improvement, with an emphasis on collaborative problem solving. It presents the results of a focus group study conducted as part of Research Phase 4, in which individuals with poorly understood conditions were invited to propose improvements and recommendations for change. The results outline a set of targets which could be addressed to improve patient satisfaction with healthcare services in Ireland and improve the experiences of individuals with poorly understood and multi-systemic conditions. These findings are discussed in relation to integrated patient-centered care.

Further demonstrating commitment to PPI, *Chapter 7* presents a worked example of engaging patient contributors in collaborative data analysis, thereby highlighting the potential for and value of PPI at this stage of the research cycle.

Chapter 8 triangulates the main empirical findings across each of the phases of research, providing a multi-layered account of the lived experiences and needs of individuals with poorly understood conditions in Ireland, with implications of research, limitations and future directions for research discussed.

Chapter 2. Methodology

This chapter provides an overview of the underlying philosophical assumptions, approaches and methods used throughout the project, both as a whole and for individual studies. The studies conducted were part of a multi-phase design, with Patient and Public Involvement (PPI) embedded throughout, thereby putting the patient at the centre of the research. Adopting a pragmatist approach with mixed methods, this research illustrates how no single theoretical tradition or methodology can provide the perfect understanding of complex phenomena.

2.1 Philosophical Underpinnings

This thesis adopts a pragmatist paradigm, employing both qualitative and quantitative approaches to meet the research aims and objectives. It aligns with the pluralist worldview, which assumes that there cannot be one, final, "true" account of a phenomenon or experience, but rather, that different layers of meaning can be uncovered by diversity of methods. By focusing on lived experiences and engaging patients as partners, this work draws heavily on phenomenological philosophy and aligns closely with the principles of action research.

2.1.1 Pragmatist Paradigm

Research paradigms are the foundational frameworks that inform and guide formal scholarly inquiries, thereby influencing the direction and methodology of research, as well as the interpretation of findings. Each research paradigm is characterised by its own set of beliefs about the nature of phenomena and what it means to attain knowledge about the phenomenon, in other words, the ontological and epistemological assumptions (Crotty, 1998; Guba & Lincoln, 1994). These varied assumptions are often reduced to contrasts between quantitative and qualitative methods of inquiry, associated with the positivist and constructivist paradigms respectively (Jurich et al., 2014; Poole et al., 2002)

The longstanding divide in academic discourse between positivism and constructivism and/or interpretivism reflects two fundamentally different worldviews: one that relies on objective observation and measurable facts, and another that embraces the subjective, context-dependent and interpretative nature of human experience. Positivism, the

dominant paradigm in the scientific community, is founded on the belief in an objective, singular reality or "truth" which exists independently of the observer (English & Nielsen, 2023; Guba & Lincoln, 1994). Accordingly, assuming that reality is observable, researchers within this paradigm employ quantitative methods which enable them to measure, explain and predict this truth, thereby producing generalizable findings through controlled and systematic investigation (Park et al., 2020; Teo, 2018). In contrast, the constructivist and interpretivist paradigms posit that knowledge is subjective and context-dependent, shaped by individual experiences and social interactions (Crotty, 1998; English & Nielsen, 2023; Shannon-Baker, 2016). The latter paradigms reject the notion of a single, objective truth and prioritise the exploration of local truths and multiple perspectives (Labonte & Robertson, 1996). Thus, research grounded in these paradigms typically relies on qualitative approaches, such as case studies, phenomenological or hermeneutic methods, and ethnographies, which facilitate in-depth exploration of subjective meanings and experiences (Scotland, 2012). Along with recognition of their merits, the assumptions and methods of each paradigm have been subject to criticism, highlighting that while valuable, neither approach on its own can fully capture complex phenomena. With its focus on objectivity and measurement, positivism has largely been criticised for failing to account for the subjective experience of people (English & Nielsen, 2023; Neuman, 2014; Park et al., 2020). Conversely, while rich in depth and contextual sensitivity, the constructivist and interpretivist paradigms are often critiqued for promoting relativism and lacking generalizability (Parker, 1999). A pragmatist approach, employing mixed-methods, offers a middle ground in this paradigmatic debate.

Pragmatism primarily considers beliefs and theory as being linked to one's practical engagement in the world rather than concentrating on the nature of reality. In other words, instead of strict adherence to a specific paradigm of methodology, this perspective focuses on practical outcomes; in other words, what "works" best in a given context, or for answering specific research questions (Allen & Clough, 2015; Feilzer, 2010; Yardley & Bishop, 2017). Acknowledging the importance of both objective and subjective understanding, this paradigm supports the development and use of mixed-methods designs to leverage the strengths of both qualitative and quantitative methods of inquiry (Alam, 2019; Frost & Bailey-Rodriguez, 2020; Tashakkori & Creswell, 2007).

While some concerns have been raised about methodological pluralism, particularly around the compatibility of differing ontological and epistemological orientations, Willig (2022) contends that complementarity between approaches is important in studies aiming for integration, rather than those demonstrating the various ways a phenomenon can be understood.

2.1.2 Phenomenological Tradition

As mentioned, with its emphasis on lived experience, pragmatically oriented research draws significantly on the phenomenological tradition. Phenomenology is a philosophy and a research approach within the constructivist paradigm, and at most basic can be described as an approach which focuses on understanding human experience, rather than of the "reality" of things (Carel, 2011; Finlay, 2009; Guba & Lincoln, 1994). Although views and emphases within phenomenology vary, it can be divided into descriptive phenomenology articulated by Edmund Husserl (1900), and interpretative phenomenology developed by Martin Heidegger (1927), the core distinction being their respective emphasis on the "lifeworld" and "being-in-the-world".

Husserl's lifeworld refers to the taken-for-granted realm of everyday lived experience, while Heidegger's being-in-the-world highlights the inherent interconnectedness between individuals and their lived experiences; that is, their existence as intertwined with the world and not as alongside it (Davidsen, 2013; Finlay, 2009; Tuffour, 2017; Willig, 2017, 2022). In practice, Husserl was concerned with identifying the "essence" of phenomena through phenomenological reduction, which entails bracketing or suspending one's natural attitudes and preconceptions during inquiry. Heidegger instead posited that it is not possible to produce a pure description of experience, and that description and understanding always involves a certain amount of interpretation. This emphasis on hermeneutics, or the interpretation of meaning, is at the core of Interpretative Phenomenological Analysis employed as part of this research, detailed in *Section 2.5.*

The research also draws on Merleau-Ponty's (1968, 1962) concept of embodied interpretation. Merleau-Ponty's interpretative position built upon Heidegger's being-in-the-world, emphasising that our lived experiences are inherently embodied. According to this perspective, the body is not merely an objective physical body, but a "lived body"

that is inextricably linked to our perceptions of the world. Pertinent to this research, Merleau-Ponty emphasised that typically, the body functions as a transparent medium, allowing us to engage with the world and its objects. With illness, this transparency is disrupted such that the previously unobtrusive body demands attention. Carel (2011, 2021) writes extensively on how the interpretative phenomenological method, and Merleau-Ponty's embodied phenomenology offers a fruitful way of exploring how illness changes the relationship between the body and the self, shifting focus from the body as an object of biomedical inquiry. This position is adopted in the studies comprising this thesis, which explore what it is like to live with poorly understood chronic illnesses, with consideration for the different contexts lived and their temporal dimension.

2.1.3 Participatory Approach

As mentioned, the research comprising this thesis places the person at the centre of the work. By engaging patients as partners in developing meaningful recommendations for change in healthcare, it aligns with the values of action research and particularly the participatory approach.

2.1.3.1 Action Research

Action Research is a family of approaches which share the core aim of producing practical knowledge that is useful to people in everyday life (Reason & Bradbury, 2008). Unlike traditional research models which involve individuals as participants or consultees, such approaches involve stakeholders as collaborators, seeking to empower communities and individuals to actively engage in problem identification and work towards positive change (Bradbury, 2015; Macaulay et al., 1999). Though variations in specific methodologies exist, such methods are characterised by a cyclical process of planning, action, observation and reflection, which ensure that the research remains adaptive and responsive to participants' feedback. Participatory Action Research is a specific approach within the action research frame that emphasises and enables the active involvement of community members and stakeholders throughout the entire research process (Kemmis et al., 2014; Kidd & Kral, 2005; Kindon et al., 2007). It acknowledges that expertise comes in many forms and ensures that stakeholders have decision-making power not only in problem and solution identification, but also in research co-design, execution, and change implementation (MacDonald, 2012; Vescey et al., 2023).

2.1.3.2 Patient and Public Involvement (PPI)

Given their focus on stakeholder engagement, collaborative learning and change, action research approaches have become a leading method for research in healthcare (Cordeiro & Soares, 2018; Kjellström & Mitchell, 2019). As discussed in Chapter 1, this is reflected in the growing emphasis on Patient and Public Involvement (PPI) and the establishment of initiatives like INVOLVE (2012) in the UK and PPI Ignite in Ireland (2022), which advocate for research to be conducted "with" or "by" the people who use health services. Generally, although levels of contributor involvement vary, the ambition is that PPI partners contribute to all stages of research, including planning, design, execution, and dissemination, thereby ensuring that the research addresses the needs and priorities of those it aims to serve (Hovén et al., 2020; Ocloo et al., 2021). Although rapidly expanding, however, PPI is still in its formative days as a normalised way of conducting research in Ireland (Gilfoyle et al., 2022). The present research involves key stakeholders, namely individuals with lived experience of poorly understood chronic illnesses, in various capacities across the studies. This involvement encompasses problem identification, study co-design and execution, collaborative data analysis, and review of outcomes.

Several rationales for incorporating PPI in research broadly, and healthcare research specifically, have been outlined throughout the literature (e.g., Boote et al., 2015). From an emancipatory or moral standpoint, it has been argued that those most affected by research outcomes should shape the research agenda, rather than leaving such decisions to elite groups like researchers, funders and policy makers. PPI is therefore seen as a way of democratising research, enabling more equitable power distribution and decision making (e.g., Frith, 2023; Jackson et al., 2020; Pearce, 2021; Wynne, 2006). From an epistemological stance, relatedly, it has been argued that PPI can help bridge the gap between expertise in the form of lived experience, and academic expertise. Incorporating diverse perspectives, which might challenge researchers' assumptions, is seen as critical for enhancing research relevance and quality, ultimately leading to more robust and impactful outcomes (e.g., Brett et al., 2014b; Hovén et al., 2020; Staley & Barron, 2019). Further, by promoting accountability, transparency and autonomy, PPI can also be seen as a means of building trust and enhancing the ethical dimension of research.

2.1.4 Reflexivity

The research was conducted with a reflexive approach, acknowledging positionality and the potential influence of personal and professional experiences on the research process and outcomes. The main researcher and thesis author has lived experience of poorly understood syndromes and symptoms, which informed the research focus and provided insights into the subject matter. Yet proximity to the subject matter necessitated careful reflection to ensure academic rigor and minimise the influence of personal biases on the interpretation of findings (Olmos-Vega et al., 2023; Palaganas et al., 2017). To this end, reflective journals were maintained and revisited by the author to document thoughts and decision-making processes, fostering ongoing critical self-awareness and recognition of personal biases and assumptions, and their role in interpretation. Rigor was maintained through the use of structured frameworks for data collection and analysis and ensured through regular supervisory oversight. Certain analyses were conducted with the help of research assistants who brought diverse perspectives to different phases of the research. Acknowledged in each study, the research assistants were psychology students without personal lived experience of poorly understood chronic illness, but had an academic interest in health psychology, chronic illness and healthcare more broadly. As mentioned above, the involvement of PPI contributors in the research brought unique but complementary perspectives to the research, shaped by their differing demographic and health characteristics, life worlds and varied experiences with healthcare.

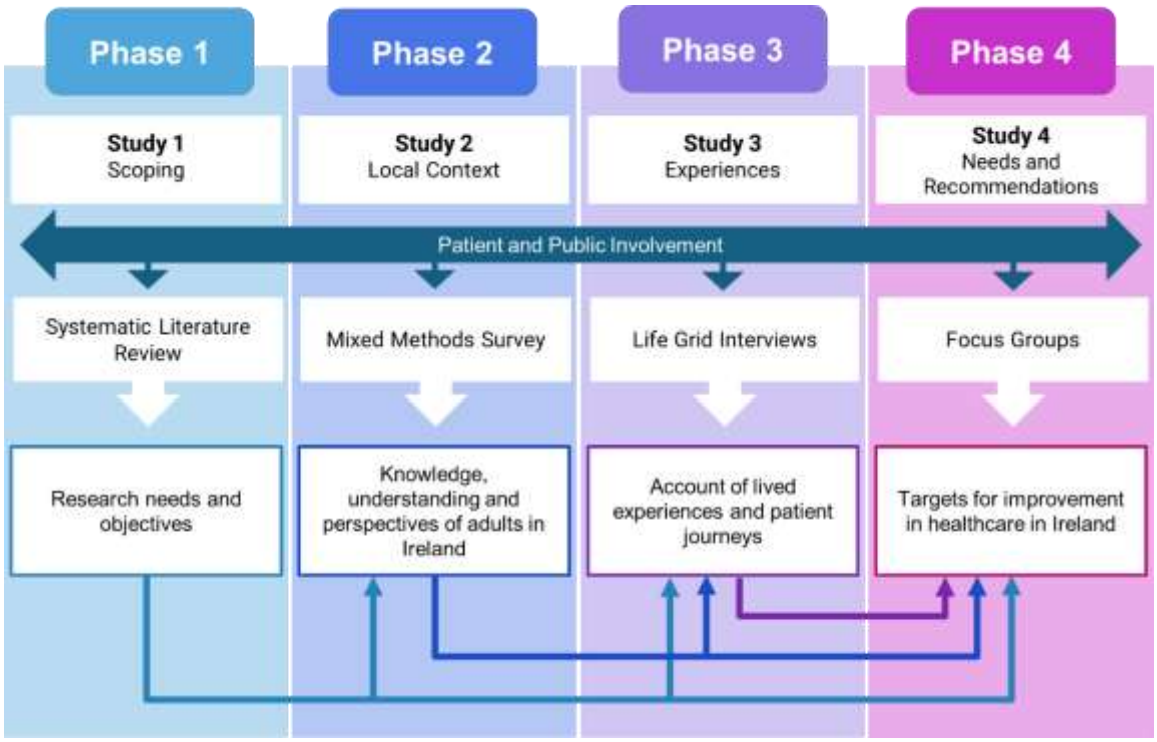
2.2 Muti-Phase Design

The research conducted in this thesis followed a multi-phase design with four inter-related phases of research, whereby each phase was used to inform the design of the following study and further contextualise that which occurred prior. An overview of the phases of research and studies within can be seen in Figure 2.1, with methods, underpinnings and analytic approaches detailed below. As illustrated, the findings of each phase fed into the design and objectives of subsequent studies.

Detailed in *Sections 2.3* to *2.6,* the involvement of stakeholders, specifically patients with poorly understood chronic conditions in Ireland, was sustained through all four phases of research. Their contribution shaped the research programme overall, and input at each phase fed into the design and objectives of the studies at other phases. With regards to study design, both methodological and analytic pluralism were employed, with quantitative and qualitative methods to address research questions, thereby aligning with the pragmatist philosophy. Rather than seeking to integrate findings across all studies, the research within this dissertation sought to present layers of meaning produced by plurality of methods.

Figure 2.1

Multi-Phase Design of Research



Note. PPI Contributors were involved in the conceptualisation, design and execution of studies, providing ongoing feedback and input on the process throughout.

2.3 Research Phase One

The first phase of research comprised of a systematic literature review and stakeholder engagement, with the objective of identifying research needs and involving patients as partners in the research.

2.3.1 Study One

The first study, reported in Chapter 3, sought to aggregate knowledge regarding the experiences of patients and healthcare providers in the context of medically unexplained symptoms and two poorly understood conditions, Fibromyalgia and ME/CFS. A systematic review of qualitative research was conducted between August 2021 and June 2022, and completed in December 2022, to meet this aim. The Joanna Briggs Institute (JBI) approach to meta-aggregation (Lockwood et al., 2020) was used to synthesise the qualitative results of 143 qualitative and mixed methods studies, published between 2001 and 2021. Aligning with pragmatism of the thesis, this approach does not seek to interpret findings nor generate theory, but rather, seeks to synthesise qualitative findings in a manner which maintains the original author's meaning (Hannes & Lockwood, 2011). The JBI Manual for Evidence Synthesis (Aromataris & Munn, 2020) and Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement guided the methodology and reporting of the systematic review.

2.3.2 PPI

Aligning with participatory values, Research Phase One involved extensive consultations with individuals with lived experience of poorly understood conditions in Ireland. Initial stages of PPI commenced in June 2021 and involved informal discussions with founders and members of various patient support and advocacy groups, including Chronic Pain Ireland, Rare Disease Ireland, the Irish ME Association, Fibromyalgia Ireland. This step involved discussions around key challenges faced by patient groups and the possible avenues of research, and recruitment of patient volunteers for a research panel. Across various networks, and via referral by group members, ten patients expressed interest in joining the panel and contributing to the design of research.

The first panel meeting was conducted online in September 2021. Eight patients living with various conditions, including ME/CFS, Ehlers-Danlos Syndrome, Fibromyalgia, Long-

COVID and Lyme Disease attended the two-hour online meeting, during which they were asked about the key issues faced by patients in Ireland, as well as their perspectives on possibilities for change. Group members were asked about research needs in the area, including the core topics, and discussed possible ways of investigation. A mind map of concerns raised during the discussion was created following the meeting and shared with the panel members for feedback. Topics discussed included issues related to the patient journey, the personal impact of illness, terminology used, research-related challenges, and gender-based stereotypes. Four patients attended a second one-hour online meeting in January 2022, which was arranged to discuss progress on and preliminary trends identified in the systematic literature, and to discuss targets for the next phases of research. The remainder of panel members found online meetings too taxing and expressed a desire for further communication via email.

Across these activities, PPI contributors identified stigma, disbelief and invalidation, in both social and healthcare settings, as an initial target for research, highlighting the lack of recognition for the struggles of this patient group in Ireland. Recounting their personal experiences, they suggested that such issues likely arise from inadequate biomedical knowledge and poor understanding of the challenges associated with conditions that have been deemed “psychosomatic” or “medically unexplained”. Given the lack of research and basic data on these conditions in Ireland, it was agreed that the initial step should explore public awareness and knowledge, and that this could be achieved through an online survey within the constraints of the COVID-19 Pandemic. Contributors also expressed dissatisfaction with healthcare services and their journeys through the system, raising concerns regarding diagnosis, treatment and care coordination, as well as scepticism regarding the medical education and training of healthcare professionals regarding these conditions in Ireland. Other significant issues concerned the lack of electronic patient health records, lack of services and supports for patients with rare or undiagnosed conditions, as well as the financial implications of having to seek care abroad. These concerns aligned with results of the systematic literature review, and provided a foundation for Study 2, 3, and 4.

2.4 Research Phase Two

The second phase of research, informed by outcomes of Phase 1, comprised of a mixed-method survey and ongoing collaboration with PPI contributors, with an overall objective of grounding the research in the Irish context.

2.4.1 Study Two

The second study sought to explore the knowledge, understanding and perspectives of adults in Ireland on two poorly understood conditions, ME/CFS and FM, as well as their appraisal of public and clinician knowledge. A cross-sectional, anonymous, mixed-method online survey was conducted between May 2022 and January 2023 to meet this aim. While the survey was intended to reach a broad population sample, knowledge and understanding were of primary interest. Thus, data collection procedures were designed to collect the responses of individuals who were aware of ME/CFS and FM, thereby ensuring that responses were directly relevant to these conditions. In alignment with pluralist ontology, the survey included categorical and open-ended questions, and utilised quantitative and qualitative methods of analysis, outlined below. Chapter 4 (*Section 4.2*) provides further detail of the design, procedure and specific methodology of Study 2.

Briefly, survey participants completed a series of questions concerning their demographic characteristics, self-rated health literacy and questions pertaining to their beliefs, knowledge and understanding of ME/CFS and/or FM. As the study sought the perspectives of individuals on these two conditions, only participants who indicated knowing of the condition could proceed to the relevant questions. These participants were asked a series of multiple-choice and Likert-scale type questions concerning their perspectives on each condition, randomised automatically to reduce order effects and potential response bias. Categorical questions concerned their attitudes and beliefs, ratings of own knowledge and understanding, ratings of public and healthcare professionals' knowledge and awareness. An open-ended question asked participants what they perceived to be the challenges faced by individuals living with the relevant condition.

2.4.1.1 Exploratory Analysis

Participants' responses to categorical questions were analysed descriptively with non-parametric statistics. Chi-squared tests of independence conducted to explore associations between participants' beliefs, ratings of knowledge and understanding, and personal factors. Bonferroni adjustments were applied to account for multiple comparisons and effect sizes calculated.

2.4.1.2 Inductive Thematic Analysis

Participant responses to the open-ended question were analysed thematically by the author and research assistant, using NVivo 12 Plus Software. Thematic analysis, as described by Braun and Clarke (2006, 2021a) is a versatile approach to qualitative data analysis, which seeks to identify, describe and interpret themes within a dataset without being tied to a specific epistemological or ontological framework. As emphasised by the authors, this does not mean that Thematic Analysis is atheoretical; rather, it can be applied across a range of paradigms and frameworks (Clarke & Braun, 2017). Within thematic analysis, themes can be generated either inductively or deductively, with the former being primarily data-driven and latter drawing on pre-existing theoretical frameworks or pre-conceptions (Braun & Clarke, 2006; Nowell et al., 2017). An inductive approach was selected in line with the exploratory stance, the six steps of which are detailed in *Section 4.2.5.2* of Chapter 4.

In parallel with the quantitative constructs of reliability and validity, credibility and trustworthiness in thematic analysis were sustained by drawing on the work of Lincoln and Guba (1985) and pragmatic guidelines provided by Nowell et al. (2017). The following steps were implemented to sustain methodological rigor: *(i)* NVivo software was used to maintain a decision trail, documenting the evolution of themes; *(ii)* a codebook was devised, ensuring coding consistency; *(iii)* inter-rater reliability was assessed using Cohen's kappa; *(iv)* triangulation methods were employed to mitigate potential bias in data extraction and analysis, whereby researchers performing the analysis had different levels of experience with chronic illness and themes were reviewed by the patient panel.

2.4.2 PPI

Based on the needs identified in Phase One, and initial suggestions regarding survey content, the author devised a mixed-method survey concerned with public perceptions of two poorly understood conditions, ME/CFS and FM. PPI contributors were consulted with regards to the design and content of the mixed-method survey. They made suggestions for improvement with regards to phrasing and appropriate terminology, and their feedback was discussed and integrated into the study. For example, contributors highlighted the issue of a distinction between chronic symptoms and syndromes, emphasising that people incorrectly assume "chronic fatigue" is synonymous with "ME/CFS". This distinction was maintained in the survey by the inclusion of questions regarding familiarity with both terms. PPI contributors were also involved in dissemination of the survey among their personal networks, thereby assisting with snowball sampling.

While ongoing panel meetings were unfeasible for those with energy-limiting conditions, informal virtual meetings were arranged with patient advocates from various groups and charity organisations, including Rare Ireland, the Irish ME Association, Chronic Pain Ireland, and the Irish EDS and HSD Group. Preliminary results of Study 2 were shared, and discussions culminated in the parallel design of Study 3 and 4.

2.5 Research Phase Three

Drawing on outcomes of Phase 1 and 2, the third phase of research sought to provide a rich account of the lived experiences of individuals with poorly understood conditions, including an exploration of their journeys with the healthcare system.

2.5.1 Study Three

Study three was a qualitative study conducted in July and August of 2023, involving in-depth interviews using the Life Grid approach. The study is rooted in the interpretative phenomenological tradition which placed emphasis on the situated and embodied nature of experience. Aligning with the pragmatic stance, analytic pluralism was employed, whereby data generated through the interviews and the content of Life Grids were analysed in three ways: phenomenologically, to explore lived experiences; descriptively, to examine patterns in patient journeys; and thematically, to assess the application of the Life Grid tool. Study three, including a detailed account of the interview structure and procedure, is presented in Chapter 5, with the Life Grid approach and three methods of analysis outlined below.

2.5.1.1 The Life Grid Interview

The Life Grid is an interview tool which captures information about participants' life experiences in a chronological, visual format. The tool was originally devised for quantitative health research with older adults, with the aim of addressing issues of recall validity (Berney & Blane, 1997). Later, it was adapted for qualitative research on smoking behaviour by Parry et al. (1999) who employed it not only as a tool for data collection but as a means of facilitating the interview itself. The procedure includes the collaborative construction of a grid which comprises of time intervals on one axis and domains of interest on the other. This is completed and discussed throughout the interview.

Although studies employing the Life Grid are scarce, the limited literature demonstrates its application in exploring individuals' experiences across diverse contexts. For example, the method has been used to explore young people's experiences of parental substance abuse (Wilson et al., 2007) and psychotic illness (Ballal et al., 2021), to explore the experiences of individuals with chronic pain (Richardson et al., 2009), to investigate family meals over a life-time (Harrison et al., 2011), how nursing graduates promote their

psychological well-being (Zhou et al., 2023), and to explore the benefits of recording biographical stories on life grids for comparative European educational research (Abbas et al., 2013).

This method was considered particularly suitable for this research due to its ability to capture both the situated and temporal dimensions of experience, thus providing a holistic account of the experience of living with chronic illness. The handful of studies employing life grids have praised the flexibility and participatory nature of the tool, noting that it fosters relationality between interviewer and participant, promotes engagement, empowers participants to pace and structure the interview, and can therefore aid in the discussion of sensitive topics (Parry et al., 1999; Richardson et al., 2009; Wilson et al., 2007). Ballal et al. (2021) also reported participants' positive feedback on the method, which was regarded as capturing a holistic account of their life. Where participants are distracted or breaks are needed, the visual anchoring of events enables the participant to easily return to the story, and the researcher to redirect conversation back to the interview (Harrison et al., 2011; Richardson et al., 2009). As suggested by Richardson et al. (2009), the interview approach might therefore be particularly suitable for interviewing participants with chronic conditions, where symptom fluctuations over time are a key feature.

Despite its merits, several challenges related to the Life Grid interview have been noted, though these mirror the caveats of narrative approaches more generally. For example, narrative interviews can be hard to predict with regards to their length and the potential emergence of sensitive or unexpected material, thus requiring good rapport and interviewer skill (Anderson & Kirkpatrick, 2015). Richardson et al. (2009) note that the length of the Life Grid interview can be a challenge in the context of prolonged illness. Harrison et al. (2011) and Nico (2016) also point out that the method cannot ameliorate all the limitations inherent in collecting life histories; the authors note that retrospective data and biographical memories are often faulty, and it is not possible to evaluate to what extent the memories are a "true" rendering of experience. Finally, while the method facilitates the collection of both qualitative and quantitative data, available studies provide little guidance on how the spoken narrative and visual content can be integrated beyond description.

2.5.1.2 Interpretative Phenomenological Analysis

Interpretative Phenomenological Analysis (IPA), the first analytic method, was used to explore the life stories and experiences of participants as captured through in-depth interviews. IPA is an established qualitative method of inquiry, concerned with the detailed exploration of personal lived experience, examined on its own terms with a focus on meaning making (Smith, 2019; Smith et al., 1995, 2009). Detailed guidelines on conducting IPA exist (e.g., Eatough & Smith, 2017; Smith et al., 2009; Willig, 2017) and the steps of the approach are detailed in Chapter 5, *Section 5.2.4.3*.

The two commitments of IPA are the phenomenological requirement to "give voice" to participants, and the interpretative requirement to "make sense" of this in relation to the wider social, cultural, psychological and theoretical context (Larkin et al., 2006; Tuffour, 2017). The method was chosen as a suitable analytic approach in the current study, which seeks to explore interviewee's life stories, with a focus on their lived experiences of health and illness in personal, social and healthcare contexts.

A core assumption of IPA is the impossibility of gaining direct access to the research participants' life world. As a result, the analysis produced by the researcher is always the interpretation of the participants' experience, that is, it involves a double hermeneutic (Smith et al., 2009). In recognizing the interpretative role of the research, IPA does not advocate the use of "bracketing" but encourages the researcher to reflect on their presumptions and to arrive at an insightful interpretation through an iterative process of analysis (Biggerstaff & Thompson, 2008; Finlay, 2009).

Smith et al. (2009) discuss several methodological considerations which guided the design of Study 3. The authors emphasise that "good" IPA concerns matters of existential importance to participants, including situations in which they are prompted to reflect on such concerns in an attempt to make sense of their meaning, as in the case of illness. Further, IPA studies typically involve semi-structured or in-depth interviews and small sample sizes, ranging from single case designs to up to thirty participants, with the norm being towards the lower end of approximately six participants (Reid et al., 2005; Smith et al., 2009; Willig, 2022). Large samples are considered inappropriate in light of the idiographic commitment of IPA to deeply exploring phenomena through the lens of

particular individuals, which stands in contrast to the nomothetic focus of psychology (Pietkiewicz & Smith, 2014).

Furthermore, although the hermeneutic focus is the key feature of IPA, the subjectivity inherent in interpretation has raised some concerns regarding evaluating the quality of research (Willig, 2017). According to Smith's (2011) ranking system, "Good" studies should exhibit sensitivity to context, commitment to and rigor in data engagement, transparency in documenting research stages, coherence in narrative development, and a double hermeneutic focus that aims for meaningful interpretation. Nizza et al. (2021) more recently highlighted four markers of high-quality IPA: constructing a compelling narrative, developing an experiential or existential account, close analytic reading of participants' words, and attending to convergence and divergence. These guidelines informed the conduct and write up of Study 3.

2.5.1.3 Descriptive Life Grid Analysis

Data concerning the participants' healthcare experiences over time were extracted from the Life Grid tool. Aligning with the phenomenological focus, the content of the Life Grid was accepted as the participant's truth regarding their healthcare journey. This content was visualised to map out the individual patient journey. Extraction of data drew on the scoping review by Davies et al. (2023) which highlights that visual journey mapping can provide insights into care touchpoints, continuity and transitions, which can highlight barriers and facilitators to care in the health system. The exploratory visual mapping applied is detailed in *Section 5.2.4.2*.

2.5.1.4 Reflexive Thematic Analysis

Data concerning participants' experiences of the Life Grid interview were analysed thematically using NVivo 12 Plus software, specifically employing the reflexive approach (Braun & Clarke, 2021b). Unlike the inductive approach employed in Study 2 and described in *Section 2.4.1.2* above, Reflexive Thematic Analysis encourages the researcher to reflect on how their positionality and worldviews may be shaping the interpretation of data (Braun & Clarke, 2019). Here, the analysis is assumed to reflect the researcher's interpretation, which is shaped by the dataset, theoretical assumptions of the analyst, as well as their analytical skill. This aligns with the phenomenological position of Study 3, as well as the pragmatist philosophy underlying the research as a whole.

As noted previously, analysis followed the six-step process and is detailed in Chapter 5, *Section 5.2.4.4*. Data were coded by two analysts, but unlike in the inductive approach, no codebook was devised and emphasis was placed on sense-checking, rather than inter-rater reliability as per Cohen's kappa.

2.5.2 PPI

As mentioned, consultations with patients in Phase One and Two of research underscored a need to examine the healthcare experiences, and patient journeys, of individual with poorly understood conditions in Ireland. Following discussions regarding design, ethical approval for Study 3 and 4 was sought by the author. One patient contributor expressed interest in piloting the method of Study 3, engaging in a pilot interview. Their feedback regarding the interview process, including question phrasing and presentation, was incorporated into the design. In this process, several possible challenges were identified, including issues related to font size, potential memory difficulties, and participant fatigue during the interview. Thus, the interview procedure was modified to enable participants to prepare in advance of the meeting. Following this, patient contributors from the original panel and those engaged in leading online support groups assisted in the recruitment of participants for Study 3, sharing information about the study among their personal networks.

2.6 Research Phase Four

The aim of the fourth phase of research was to identify targets for improvement with regards to the healthcare system, and to involve patients in developing recommendations for change.

2.6.1 Study Four

Study 4 was a qualitative study centered around collaborative problem solving, thus aligning with both pragmatism and action research. Online focus groups were conducted between September and December of 2023, with individuals living with chronic poorly understood conditions being invited to discuss their healthcare experiences, needs and recommendations for change. Two patient partners who did not participate in the groups were involved in collaborative data analysis. Qualitative data were analysed using the Framework Approach, and analysis supplemented by exploring group dynamics through sociograms. Study 4 is reported in Chapter 6, while Chapter 7 reports on patient involvement in the co-analysis process. The focus group method, including considerations for design, is outlined below along with the analytic methods applied in the study.

2.6.1.1 Focus Group Design

Focus groups are a research method chosen for their well-established ability to facilitate in-depth discussion and provide a comprehensive understanding of diverse viewpoints. The approach involves engaging a small number of people from a target population, typically between four and 12 individuals, in a group discussion 'focused' around a particular topic or issue (Krueger & Casey, 2014; Wilkinson, 2004). A popular tool in healthcare research, focus groups have been used, among others, to glean patients' experiences of health and illness (Kitzinger, 2013; Wilkinson, 1998), to develop recommendations for patient-centered care (e.g., Nyhof et al., 2020), to develop feasible interventions (e.g., Versele et al., 2022) and in health promotion messaging (e.g., Thai et al., 2019). The approach was therefore chosen to capture the diversity of perspectives among individuals with poorly understood condition, with the view of developing meaningful recommendations for change in healthcare.

An advantage of this approach over individual interviews is the ability to capture the dynamics between participants, which can reveal how opinions are formed, challenged, and reinforced. Within this social setting, individuals can explore and challenge their "held truths", co-construct meanings and narratives about topics, or develop solutions for problems (Belzile & Öberg, 2012; Duggleby, 2005; Lehoux et al., 2006) making the method particularly suitable for action research. However, this also raises challenges with regards to choosing the unit of data analysis (Fern, 2001). While some researchers prioritise the analysis of interview content, others point to a need to account for social aspects, exploring issues such as social influence, power, group cohesion, peer validation, and conflict within the focus group dynamics.

Several other methodological considerations informed the design and analysis of Study 3. Available literature provides extensive guidance on focus group design, highlighting the impact of factors like the research setting, group task, group cohesion and moderator characteristics on outcomes (e.g., Fern, 2001; Stewart et al., 2007). For example, with regards to group moderation, it is recommended that moderators possess neutrality, strong facilitation skills, as well as sufficient knowledge about the topic to guide the conversation in meaningful ways (Krueger & Casey, 2014). With regards to group features, homogeneity is recommended, and participants are recruited based on shared characteristics or experiences in relation to the research question. This can help foster a sense of belonging, thus encouraging participants to engage in more open and cohesive discussions (Plummer-D'Amato, 2008). Concerns regarding the quality of interactions in groups conducted virtually, as opposed to in-person, have also been raised (e.g., Bolin et al., 2023) though studies typically suggest that online groups provide a valuable opportunity to reach populations whose participation is limited by social barriers, time or travel distance (Stewart & Shamdasani, 2017; Tran et al., 2021).

2.6.1.2 Framework Analysis

Focusing on discussion content, qualitative data were analysed using the Framework Approach (Ritchie & Spencer, 1994). This approach, which was initially developed to produce actionable insights in applied policy work, has become a well-established method in multidisciplinary healthcare research (Furber, 2010; Parkinson et al., 2016; Ward et al., 2013). Recognised for its methodological rigor and flexibility, it facilitates the

systematic organization and comparison of different data types, such as observational data, policy documents, case studies, as well as data from interviews and focus groups (Goldsmith, 2021). As discussed by Gale et al. (2013), the method holds advantages of both deductive and inductive approaches, making it suitable for answering specific questions as well as allowing for the development of new ideas. The steps of Framework Analysis are discussed in Chapter 6, *Section 6.2.3.1.*

2.6.1.3. Interaction Analysis

In addition to analysis of spoken content, sociograms were constructed to explore the interactions between participants in the focus groups. The Sociogram is a quantitative and visual tool which can be used to examine relationships between different group members, as well as group structure and dynamics (Huang et al., 2006). When used alongside transcribed data, sociograms enable researchers to 'see' group processes and who is shaping and dominating group discourse (Baiardi et al., 2015). The analytic procedure adapted the methods devised by Drahota and Dewey (2008), focusing on the flow of conversation between group members. The visualisations were used to reflect on issues of dominance, thereby providing insights about the recommendations generated by the groups.

2.6.2 PPI

The research conducted in Research Phase Four reflected a culmination of insights and ongoing inputs from phases one to three of research, which together pointed to a need for improvement in the healthcare system. While previous stages involved consultations, co-design and feedback on outcomes, the most significant PPI activity in Study 4 was the involvement of two PPI contributors in collaborative data analysis and interpretation. These contributors, with different types of lived experience, were previously uninvolved in the research programme. Given the lack of clear guidance regarding PPI in the analysis stage, a worked example along with key learnings and reflections is detailed in Chapter 7.

Chapter 3. Study One

Systematic Literature Review

3.1 Introduction

Fibromyalgia (FM) and Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) are complex, debilitating syndromes with a poorly understood pathophysiology and lack of curative treatment. Both are considered syndromes with medically unexplained symptoms (MUS; Baloh, 2020; Luty, 2018), and include symptoms of chronic pain, fatigue and cognitive dysfunction (Carruthers et al., 2011; Rahman et al., 2014; Wolfe et al., 2016). While these are distinct syndromes (Abbi & Natelson, 2013), they are often considered in tandem due to high rates of co-occurrence and similar symptom profiles (Mckay, Martin, et al., 2021; Mckay, Walker, et al., 2021; Ramírez-Morales et al., 2022). Suboptimal management is well-documented, with ME/CFS (Geraghty et al., 2019; Geraghty & Blease, 2019) and FM (Wilson et al., 2022) patients reporting dissatisfaction with the quality of medical care received.

Statistics on the prevalence and incidence of ME/CFS in Europe are scarce (Estévez-López et al., 2020) though an estimated 1.7 to 3.38 million patients are diagnosed in the United States (Valdez et al., 2019). FM occurs in 2-4% in the general population (Häuser et al., 2015a; Sarzi-Puttini et al., 2020) and up to 6.8% in women (Marques et al., 2017). Like other chronic illnesses, FM and ME/CFS are associated with significant individual and familial distress, often leading to socio-occupational disability and poor health-related quality of life (Ghavidel-Parsa et al., 2015; Pendergrast et al., 2016; Valdez et al., 2019; Van Houdenhove et al., 2002, 2010). Profound effects on mental health are well-documented, including increased suicidal ideation and suicide attempts (Pederson, 2020; Pederson et al., 2017), with some literature suggesting that relative to the general population, individuals with FM and ME/CFS are at an elevated risk of suicide mortality (e.g., Gill et al., 2021; Roberts et al. 2016).

Debates about classification, terminology and diagnosis characterise the MUS, FM and ME/CFS literature. Against the backdrop of evidence-based medicine, the phenomenon of uncertainty at the core of “unexplained” illness poses significant conceptual and

practical challenges (Eriksen et al., 2013; Evans & Trotter, 2009; Han et al., 2019; Kim & Lee, 2018). For example, uncertainty regarding aetiology and difficulty detecting pathology with routine clinical testing has led many clinicians to contest these illnesses or conclude that they are psychosomatic (Baloh, 2020; Bransfield & Friedman, 2019; Swoboda, 2008). Questions have been raised about the nosology and clinical utility of the term MUS (Greco, 2012; O'Leary, 2018; Scott et al., 2022; Tack, 2019), with patients considering such labels dismissive (Creed et al., 2010). Similar issues have been raised in the context of FM and ME/CFS (Lim & Son, 2020; Natelson, 2019). However, while FM and ME/CFS technically remain medically unexplained, extensive progress has been made in the search for biomarkers of these conditions (Hackshaw & Yen, 2021; Maksoud et al., 2020; Martínez-Lavín, 2021; Missailidis et al., 2019; Ryabkova et al., 2019; Sarzi-Puttini et al., 2020; Sotzny et al., 2018). At time of writing this review, renewed interest has emerged in the wake of the SARS-CoV-2 pandemic: not only are there remarkable similarities between Long-Covid and ME (Davis, 2021; Komaroff & Bateman, 2021; Mackay, 2021; Wong & Weitzer, 2021), some evidence suggests that FM may emerge as part of the post-Covid syndrome or Long-Covid (Gavrilova et al., 2022; Mathkhor et al., 2022; Ursini et al., 2021).

As discussed in Chapter 1, as medical science continues to make headway, there is a growing impetus to draw on the experiential knowledge of individuals with lived experience of illness, involving patients and the public in healthcare-related research and service co-design (Caron-Flinterman et al., 2005; Sacristán et al., 2016; Sharma et al., 2017). This partnership reflects a marked departure from the paternalistic model dominant in medicine, and is at the heart of integrated, person-centered care (Edgman-Levitan & Schoenbaum, 2021; Taylor, 2009; World Health Assembly, 2016). Shared decision-making is the pinnacle of patient-centered care (Bauman et al., 2003; Institute of Medicine, 2001; Thomas et al., 2020; Tonelli & Sullivan, 2019) as it exemplifies the collaborative nature of healthcare decision-making and ensures that patients have an active role in shaping their treatment plans based on their unique values and preferences.

Given that the needs and priorities of patients and clinicians often differ (Crowe et al., 2015; Stewart et al., 2011), it is imperative to understand the experiences and

perspectives of both stakeholders. Previous qualitative reviews have typically adopted a single syndrome approach or explored patient *or* clinician perspectives (e.g., Crowe et al., 2017; Fennell et al., 2021; Johansen & Risor, 2017; Mengshoel et al., 2018; Pheby et al., 2021; Polakovská & Řiháček, 2022). It appears that only Anderson et al. (2012) explored patient-specific, physician-specific and intersecting themes, but the scope of their review was limited to ME/CFS research. More recently, Polakovská and Řiháček (2022) reviewed qualitative studies concerned with medically unexplained physical symptoms (MUPS), an umbrella term which may technically encompass FM and CFS. However, FM and ME/CFS were not included as search terms due to the volume of literature generated.

3.1.1 Objectives

The objective of this review is to aggregate available evidence regarding the experiences of *(i)* individuals with MUS, FM and ME/CFS, hereinafter collectively referred to as Persons with Conditions (PwC), and *(ii)* healthcare practitioners (HCPs) working with PwC. In line with systems perspectives on health and healthcare (Bergeson & Dean, 2006; Kriz, 2013; Lipsitz, 2012; Sturmberg et al., 2010, 2014) the review is underpinned by the position that both patients and clinicians are integral pieces of the dynamic healthcare landscape. By aggregating findings from both perspectives, unique and shared experiences which arise in the context of medical uncertainty can be identified, which can in turn inform recommendations for practice. As discussed in Chapter 2 (*Section 2.2*), the review was conducted as part of Research Phase One, occurring in parallel with stakeholder engagement, with an overall aim of identifying trends, research needs and objectives for further studies.

3.2 Methodology

A systematic review of qualitative research was conducted using the Joanna Briggs Institute (JBI) approach to meta-aggregation (Lockwood et al., 2020). Underpinned by the philosophy of pragmatism (Hannes & Lockwood, 2011), this approach seeks to generate a descriptive mapping and categorization of findings across a comprehensive range of studies, highlighting commonalities rather than discrepancies (Lockwood et al., 2015; Lockwood & Pearson, 2013; Munn et al., 2021; Sim & Mengshoel, 2023). Notably, the approach seeks to synthesise qualitative findings while maintaining the original authors' intended meaning: that is, it does not seek to interpret nor generate theory.

The JBI Manual for Evidence Synthesis (Aromataris & Munn, 2020) and Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement were employed to guide the methodology and reporting of this systematic review. Confidence in synthesised findings was established using the ConQual approach (Munn et al., 2014) for meta-aggregative qualitative systematic reviews. The review protocol was registered in PROSPERO (Registration Number: CRD42021248028).

3.2.1 Search strategy

Searches were conducted in May 2021 in the following electronic databases: PubMed, PsycInfo, ScienceDirect, CINAHL, Scopus, ProQuest Dissertations & Theses. An initial search was conducted across all selected databases to consult key literature and identify search terms. A comprehensive search was conducted using free-text and MeSH terms combined with Boolean operators. The following terms and their variations were used: *(i)* Fibromyalgia, *(ii)* Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis *(iii)* MUS *(iv)* Experience and *(v)* Qualitative research. A sample of the search strategy used is available in Appendix A1.

Definitions from the International Classification of Diseases [ICD-11] (World Health Organization, 2018) were adopted, whereby FM is a Primary Chronic Pain Disorder and ME/CFS is recognised as neurological condition (Post-Viral Fatigue Syndrome). However, variations in terminology for MUS, FM, and ME/CFS were included to account for evolving diagnostic criteria and conceptual differences between the included studies. It was assumed that medically unexplained symptoms and syndromes are those which

have not yet been adequately explained by medical science and are therefore characterized by a high degree of uncertainty. The choice to include MUS was driven by the use of this label as an umbrella term in the literature, rather than reflecting the researchers' assumptions about the aetiology of included conditions.

3.2.2 Eligibility criteria

Inclusion and exclusion criteria were based on a PICo (population, phenomena of interest and context) framework for qualitative research synthesis (Lockwood et al., 2015). Published, peer-reviewed qualitative and mixed-methods studies exploring the perspectives of PwC and/or HCPs involved in their management were considered, as were grey literature in the form of dissertations and theses. The phenomena of interest were: *(i)* the lived experiences, attitudes, beliefs, and challenges faced by PwC, including their needs and experiences related to healthcare; and/or *(ii)* the experiences, beliefs, and attitudes of healthcare professionals, including any practical challenges and direct experiences of consulting, diagnosing, and managing PwC. Chronic pain and fatigue were the primary focus, both of which present in FM and ME/CFS. No limitations on context with regards to cultural background, country, setting were applied.

Only full-text, English-language studies published between March 2001 and March 2021 were eligible for inclusion. Studies were excluded if they did not report the views of PwC or HCP, or involved older adults (aged 85+), patients in palliative care, or medical students. Single case studies, systematic reviews and quantitative-only mixed methods studies were excluded.

3.2.3 Study Screening and Selection

All records retrieved were imported into Zotero, with duplicates electronically removed. Two reviewers independently reviewed titles and abstracts to identify studies meeting inclusion criteria. Both reviewers were doctoral researchers in Psychology, the author having lived experience of chronic, poorly understood illness, the other (IG) a trained nurse with clinical experience. Full-text articles were retrieved for the chosen studies and where abstracts were insufficient to determine inclusion. These were reassessed independently by reviewers and included or excluded as appropriate. Any disagreements were resolved by discussion or a third reviewer from the supervisory team.

3.2.4 Quality Appraisal

The JBI Qualitative Assessment and Review Instrument (QARI) Critical Appraisal Checklist (Lockwood et al., 2020) was used to assess the methodological quality of all included studies by two independent reviewers, with any disagreements resolved through discussion. In accordance with the JBI Protocol (Lockwood et al., 2020), all qualitative studies were initially ranked as High on a scale of high, moderate, or low on methodological quality (see Lockwood et al., 2015). Studies were not excluded based on the initial ranking as doing so could have led to omissions of potentially valuable content in grey literature.

3.2.5 Data extraction and synthesis

The methods of data extraction and synthesis in JBI Meta-Aggregation have been described extensively elsewhere (Hannes & Lockwood, 2011; Lockwood et al., 2015; Lockwood & Pearson, 2013; Munn et al., 2014) and are presented in Figure 3.1. Briefly, data extraction includes *(i)* study characteristics and *(ii)* study findings, defined as a verbatim extracts of the primary study authors' analytic interpretation. Findings are accompanied by illustrations in the form of participant voice, field-work observations or other supporting data (Lockwood et al., 2015). A level of Credibility (*Unequivocal, Credible, Not Supported*) is allocated to each finding based on the perceived degree of support offered by the associated illustration (Lockwood et al., 2020). Synthesis involves a three-step process in which extracted findings are grouped into categories, and categories aggregated to produce a set of synthesised findings. Unsupported findings are not included in synthesis. Grouping of findings into categories, and categories into synthesised findings, is based on similarity of meaning (Lockwood et al., 2015)

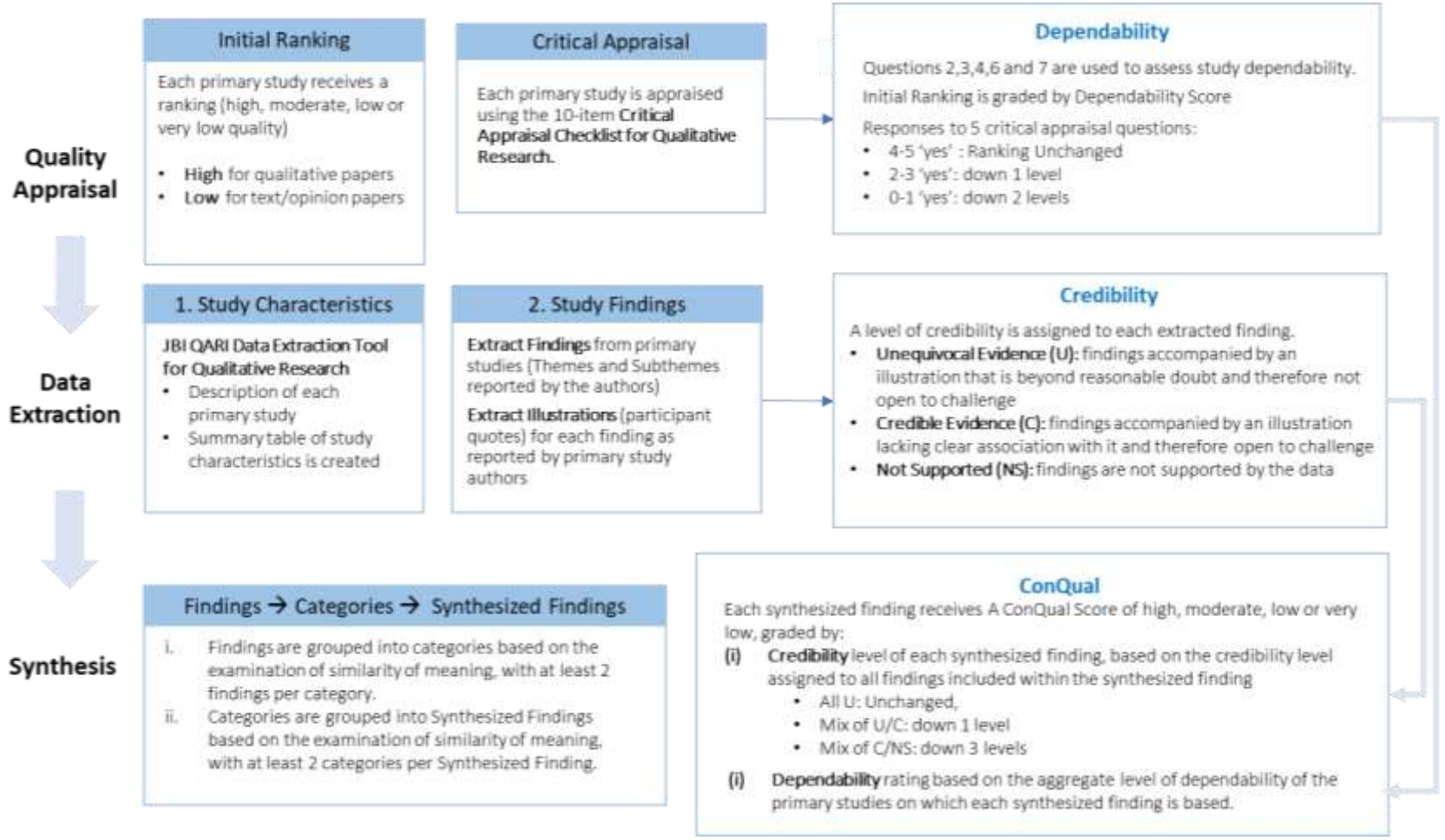
Descriptive data from included studies were assembled using the JBI QARI Data Extraction for Qualitative Research Form (Lockwood et al., 2020). This included publication details, population characteristics, phenomena of interest, study design, methodology and qualitative research methods used, context and geographical setting, and the authors' conclusions. The primary reviewer identified and extracted relevant findings from each study and assessed finding Credibility.

The units of extraction were the themes and subthemes generated by the author of each primary study, accompanied by participant quotes. Where unavailable, themes were generated by repeated reading of results and data supporting those findings. The first reviewer grouped findings into categories based on meaning, which were then aggregated to produce an overarching synthesised finding. Findings were expressed as indicatory statements and were finalized by consensus between the reviewers.

The level of confidence in each synthesised finding was established using the ConQual approach (Munn et al., 2014), whereby each paper is initially ranked on a scale of High to Very Low, with qualitative papers initially ranked High, and then graded for Dependability and Credibility (See Figure 3.1). Dependability is assessed by evaluating each study with regards to five questions on the JBI QARI Critical Appraisal Checklist (Lockwood et al., 2020). The rating is downgraded when included studies do not meet at least 4 of the 5 appraisal criteria. Credibility level is determined by the credibility ratings of the individual findings within the synthesised finding. For mixed Unequivocal/Credible ratings, credibility is downgraded by 1 level. Otherwise, the credibility remains unchanged.

Figure 3.1

The meta-aggregative approach to qualitative evidence synthesis.



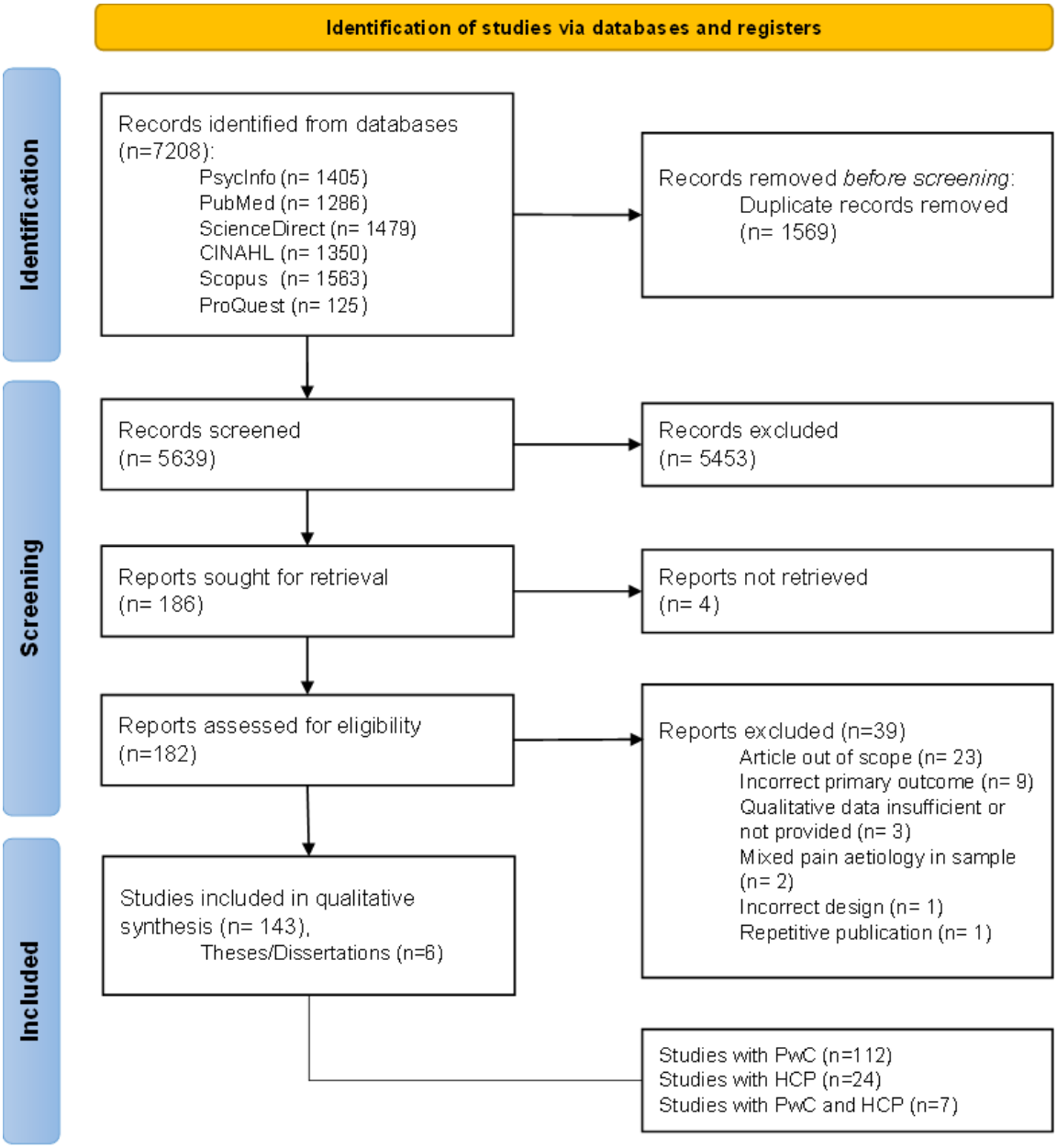
3.3 Results

3.3.1 Study selection

The database search identified 7208 records (Figure 3.2). One hundred and eighty-six studies remained after adjusting for duplicates and screening titles and abstracts. Four full-text records could not be retrieved. One hundred and eighty-two full-text articles were assessed for eligibility. Thirty-nine full-text articles were excluded, 23 of which were deemed out of scope for this review. Other reasons for exclusion were wrong primary outcome(s); qualitative data insufficient or not provided; mixed pain aetiology, including medically explained pain; incorrect design; and repetitive publication. In total 143 papers were included in the meta-synthesis. For ease of reading, the included papers included in the study are labelled numerically (1-143), with the corresponding references presented in Appendix A2.

Figure 3.2

PRISMA flow diagram



3.3.2 Study characteristics

The characteristics of included studies are presented in Appendix A3. Most concerned FM (*n*=81), followed by ME/CFS (*n*=32) and MUS (*n*=29), while four concerned patients with FM *and* ME/CFS (93–96). Studies exploring the PwC perspective were most frequent (*n*=112), 49 of which involved female-only participant samples (Table 3.1). Only six studies explored the illness experience of males, though three of these included the same participants (74,102–104,119,120). Twenty-four studies explored the perspective of HCPs. Of the studies which provided HCP gender (*n*=19), a higher number of male practitioners were involved in the majority (*n*=12). Seven studies included both PwC and HCP perspectives (20,21,29,51,58,61,77). Reporting standards for participant characteristic varied, often lacking clarity on PwC diagnoses (e.g., regarding formal criteria used) or illness duration, HCP demographics (age, gender identity) and professional roles (e.g., qualifications, specialty, years of experience).

Table 3.1

Summary of population characteristics.

			Sample Size			Participant Gender		
Participant Group	*N Studies*	*% of Total*	Total no.	Min	Max	No. Male	No. Female	N/A (N Studies)
PwC	112	78.32	4051	4	1163	1166	2694	5
HCP	24	16.78	492	6	30	194	203	5
PwC & HCP	7	4.89	270	21	101	94	153	1
Total	143		4813			1454	3050	11

All included studies used qualitative design, with 4 employing mixed methods approaches (36,61,65,74). Studies were conducted in 19 different countries, primarily in the UK (*n*=45), Sweden (*n*=18) and the United States (*n*=18). Seven studies involved multi-national samples (17,66,74,81,90,119,140). Phenomenology and grounded theory were the most common theoretical approaches, and most employed semi-structured or unstructured interviews and focus groups or group interviews (Table 3.2). Four studies used both approaches (32,77,78,92). Interviews were also conducted online (40,140) and with video-stimulated recall (64). Eleven studies employed a combination of qualitative methods, including interviews, diaries, questionnaires, and focus groups.

Table 3.2

Summary of study characteristics.

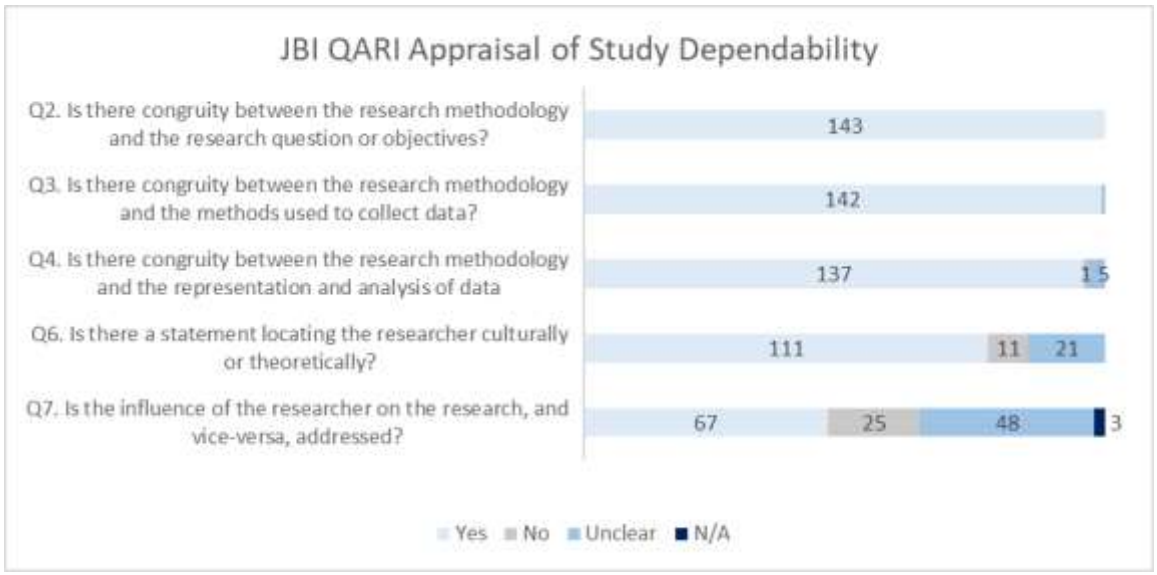
			PwC		HCP		PwC & HCP	
	N Studies Reviewed	*% of total*	*n*	*%*	*n*	*%*	*n*	*%*
Setting								
Community	71	49.65	71	63.39	0	0	0	0
Healthcare	63	44.05	32	28.57	24	100	7	100
Support Group	9	6.29	9	8.04	0	0	0	0
Theoretical approach								
Phenomenology	32	22.37	29	25.89	3	12.5	0	0
Grounded Theory	22	15.38	16	14.2	3	12.5	3	42.86
Framework Approach	7	4.89	4	3.57	3	12.5	0	0
Discourse Analysis	4	2.79	3	2.68	0	0	1	14.29
Ethnography	1	0.69	1	0.89	0	0	0	0
Symbolic interactionism	1	0.69	0	0	1	4.17	0	0
Undefined	76	53.14	59	52.68	14	58.33	3	42.86
Methodology								
Individual Interview	107	74.82	88	78.57	16	66.67	3	42.86
Focus Group	15	10.48	8	7.14	6	25	1	14.29
Survey	4	2.79	4	3.57	0	0	0	0
Other	2	1.39	2	1.79	0	0	0	0
Multi-method	15	10.48	10	8.93	2	8.33	3	42.86
Country								
UK	45	31.46	34	30.36	8	33.33	3	42.86
Sweden	18	12.58	14	12.5	3	12.50	1	14.29
USA	18	12.58	14	12.5	2	8.33	0	0
Norway	13	9.09	11	9.82	2	8.33	0	0
Canada	12	8.39	10	8.93	1	4.17	1	14.29
Australia	7	4.89	4	3.57	3	12.50	0	0
Multi-national	7	4.89	7	6.25	0	0	0	0
Other	9	16.08	7	6.25	3	12.50	2	28.57

3.3.3 Methodological quality

The quality appraisal of all included studies is detailed in Appendix A4. Congruity between the philosophical perspective and research methodology, research objectives, methods of data collection, representation and analysis of data and interpretation of results were generally high across studies (Q1-Q5). Many studies lacked recognition of the position and influence of the researcher on the research, reflecting a common Dependability issue. Figure 3.3 presents the rankings of study dependability as assessed by five items on the JBI QARI Critical Appraisal Checklist. The relationship between the researcher and study participants (Q7) was rated *Unclear* or *Not Addressed* in 73 of the reviewed studies. The researchers' cultural and theoretical orientation and its potential influence on research outcomes (Q6) was *Unclear* in 21 studies and *Not Addressed* in 11 studies. The congruity between participant data and the findings and interpretations reported by the author (Q10) was *Unclear* in 6 of the studies. Results of the critical appraisal were used in the interpretation of the synthesised findings.

Figure 3.3

Dependability of included studies (N=143)



3.4 Main Findings

Table 3.3 provides an overview of the synthesised findings. A total of 708 findings were extracted and aggregated into 82 categories and 13 synthesised findings, 8 concerning PwC perspectives and 5 concerning HCP perspectives. Overall, the credibility rating was high, with Unequivocal evidence identified for 685 of the findings. All 13 synthesised findings received a ConQual Score of Medium or Low. The ranking of 12 synthesised findings was downgraded 1 level based on study dependability.

Table 3.3 JBI Table of Synthesised Finding

Review Title	Patient and clinician experiences of fibromyalgia, ME/CFS and medically unexplained symptoms: A meta-aggregative systematic review
Population of Interest	Adults with MUS, FM, ME/CFS (PwC) and healthcare professionals (HCP)
Phenomena of Interest	The scope and complexity of experience among PwC and HCP
Context	Any country, any gender, natural community settings and healthcare environment

	Synthesised Findings	N Studies*	ConQual Summary of Findings: Type of Research**	Dependability (QARI)	Credibility Ranking: No. of Findings (Rating)	Credibility Ranking: Credibility Level	ConQual Score Synthesised Findings
1	Symptom experience and impact	52	Qualitative [1,2,4]	Downgrade 1 level	121 (U)	Unchanged	Medium
2	The patient journey	64	Qualitative [4]	Downgrade 1 level	120 (U)/4(C)	Downgrade 1 level	Low
3	Identity loss and change	23	Qualitative [1]	Unchanged	43 (U)/ 3 (C)	Downgrade 1 level	Medium
4	Managing chronic illness	26	Qualitative	Downgrade 1 level	41 (U)	Unchanged	Medium
5	Understanding and legitimacy	26	Qualitative	Downgrade 1 level	35 (U)	Unchanged	Medium
6	Support needs and experiences	22	Qualitative	Downgrade 1 level	30 (U)/ 1(C)	Downgrade 1 level	Low
7	Healthcare needs and experiences	51	Qualitative [2]	Downgrade 1 level	94 (U)/ 4(C)	Downgrade 1 level	Low
8	Managing healthcare encounters	6	Qualitative	Downgrade 1 level	15(U)	Unchanged	Medium
9	HCP attitudes towards patients	11	Qualitative [2,3]	Downgrade 1 level	18(U)	Unchanged	Medium
10	Making sense of patients' symptoms	22	Qualitative [2,3]	Downgrade 1 level	57 (U)	Unchanged	Medium
11	Consultation and clinical management	19	Qualitative [2,3]	Downgrade 1 level	46 (U)/ 6(C)	Downgrade 1 level	Low
12	Managing the relationship	16	Qualitative [2,3]	Downgrade 1 level	29 (U)/ 5(C)	Downgrade 1 level	Low
13	Barriers and facilitators to care	13	Qualitative [2,3]	Downgrade 1 level	36 (U)	Unchanged	Medium

Note: Total number of studies included in review *N*=143. Synthesised findings in blue (1-8) concern PwC; those in green (9-13) concern HCP.

* Number of studies contributing to each synthesised finding

**Includes qualitative component of mixed method studies

[1] Crooks (2006) ; [2] Hayes et al. (2010) ; [3] Howman et al. (2016); [4] Kueny et al. (2021)

Key: MUS= Medically Unexplained Symptoms; FM= Fibromyalgia Syndrome; ME/CFS= Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome; PwC= Person with condition; HCP= Healthcare Professional. *Credibility Ranking*: (U)= Unequivocal evidence; (C)= Credible evidence

3.4.1 Experiences of PwCs

A total of 511 findings reflecting the lived experiences and perspectives of PwC were extracted from 116 studies. These were aggregated into 57 categories and 8 synthesised findings encompassing *(i)* The experience of symptoms and their widespread impact; *(ii)* The patient journey; *(iii)* Identity loss and change *(iv)* Managing chronic illness; *(v)* Understanding and legitimacy; *(vi)* Support needs and experiences; *(vii)* Healthcare needs and experiences; and *(viii)* Strategies for managing the healthcare encounter. Findings were supported by direct quotes from PwC, and most frequently encompassed themes related to the patient journey (124 findings, 24%) and lived experiences of illness (121 findings, 23%). Only 3% of all findings (15/511), derived from 6 studies, concerned patients' strategies for managing healthcare encounters (See Table 3.3). The frequencies of extracted findings are presented in Appendix A5. Categories with the largest proportion of findings included the experience of physical symptoms and difficulties (4.31%), the psychological impact of symptoms (3.52%), making sense of complexity in the illness experience (3.52%), and healthcare needs and expectations (3.52%). Illustrative patient quotes for each synthesised finding are presented in Appendix A6. Where reported by study authors, PwC gender, age and diagnosis accompany the quote.

3.4.1.1 The symptom experience

Studies show how individuals with FM and ME/CFS experience a wide range of interrelated physical and cognitive symptoms which are chronic, fluctuating, and unpredictable. Widespread pain and debilitating fatigue are common physical symptoms, typically accompanied by disturbed sleep, paraesthesia, and headaches, among others (7,9,38,54,66,71,74,76,79,86,94,101,115,117,122,128,129,136). Numerous cognitive symptoms are also reported (7,36,40,66,101,128,136), collectively described as 'brain fog' and include disturbances to attention and memory. Symptoms often exacerbate one another, and PwC describe experiencing a cycle of pain, fatigue, poor sleep, depression, and stress (71,115,122,129). Their chronic, unrelenting, and unpredictable character is a major aspect of the illness experience. Widespread pain is a constant presence for many (40,71,74,94,124,128) while fatigue and exhaustion persist despite rest and adequate sleep (66,122,123,136). Many describe struggling to cope with the inconsistent, variable nature of their illness (3,16,40,83,102,136). The symptomatic impact of FM and ME/CFS

is further exacerbated by the fact that the symptoms are vague and invisible (2,47,74,94,107,114). Language used to describe symptoms in patient narratives suggests that the lived experience can be overwhelming (117,119). Pain has been described as unbearable (74), an adversary (94), sinister, violent, punitive (72), fatigue as paralyzing (53) a killer (40) and an invisible foe (128).

Many studies document the impact of FM, ME/CFS and MUS across all domains of life. PwC report their lives being fundamentally altered (21,76,79,95,115,123,136) and describe difficulties in completing everyday activities such as shopping, housekeeping, and participating in social events or leisure activities (19,39,48,53,54,66,73–75,120,122,142). Symptoms also interfere with occupational roles and career progression: many are left unable to work, despite desperately wanting to do so (40,95,142), or are faced with the necessity to reduce their working hours, responsibilities or change career trajectory completely (5,7,95,123). As individuals part with their work identity, their sense of self is threatened (14,36). Family dynamics are affected, with profound changes to roles, responsibilities and relationships among household members (14,19,123,142). Individuals with FM report feeling like a burden to their partners and children (19). Outside of their family, PwC report their social life being significantly restricted (7,123,36,79). They describe losing friends, feeling lonely and becoming isolated from others (15,94,101,142). For many, keeping up with the demands of a social life is taxing, resulting in a withdrawal from social activities (95,117).

The experience of multiple chronic symptoms can take a toll on the mental health of PwC (3,7,95). Sadness, frustration and anger can arise from one's inability to perform daily activities (61,83) and many experience affective changes (101) and psychological distress (47,79) reduced motivation (66,117), low mood, depression and anxiety (33,38,124,136) as a consequence of their physical symptoms. PwC feel overwhelmed by the prospect of symptom progression (16) and experience a sense of loss, sorrow and powerlessness with regards to their condition (15,56,94). Shame, guilt and fear of being a fraud are common themes in the accounts of individuals with medically unexplained symptoms (44,97), likely reflecting internalised stigma.

3.4.1.2 The patient journey

Four broad experiential epochs were identified in the illness narratives of PwC: *(i)* symptom onset and uncertainty; *(ii)* help seeking and diagnosis; *(iii)* acceptance and management; and *(iv)* perspectives on life and the future.

The illness journeys of PwC are often characterised by an initial period of denial, resistance, and uncertainty. Many ignore their tiredness (53) and pain (56,57), and report fighting against their increasingly intrusive symptoms (47). This resistance, combined with an insidious or gradual onset of symptoms (31,40) can lead to delays in diagnosis or hinder help-seeking efforts (26). Increasing disruption in everyday life leads individuals to seek explanations to give meaning to their experience (5,9,89,138). Many actively seek out and evaluate information about their symptoms, leading them to seek a specific diagnosis, Initial attempts to make sense of symptoms are characterised by disappointment, as individuals struggle to come to terms with the possibility that debilitating physical symptoms are not readily explained by an organic pathology (85). The experience of uncertainty is well documented: PwC struggle with not knowing what is wrong (136), what the causes of their symptoms are (9,85) and what to do next (31). Doubts about the possibility of having a stigmatised, controversial condition such as FM are expressed (120,136) and exacerbated by experiences of diagnostic uncertainty and reluctance from HCPs to provide diagnoses (132).

Faced with uncertainty, PwC attempt to make sense of their symptoms by developing various biological, psychological and cultural explanations (31,53,59,76,99,124). Multiple, complex causes and triggers are explored in their illness narratives, reflecting varying beliefs about the contribution of internal factors and external influences, such as stress or genetics (40,81). With regards to aetiology, the explanations of individuals with ME are primarily biological and, in many cases, patients can identify a viral infection as the trigger for their illness (5,138). A history of trauma and hardship are speculated to play a causal role in FM (5,46,105,118) though the accounts of PwC also reflect lives with normal day-to-day hassles (118). Symptom flares are attributed to excessive physical and mental effort, diet, chemicals, psychological stressors, climate, and changes in barometric pressure and temperature, among others (74,81,86).

The help-seeking and diagnostic journey is often a long and taxing process (16,32,33). Receiving a diagnostic label is important for several reasons. It can legitimize the symptom experience (32,34), provide a starting point for illness management (88), and commence the process of acceptance (75). However, achieving a diagnosis is a significant challenge. The process can take many years, numerous specialists and investigations, and involve considerable distress (3,9,78,137,138). Diagnoses are made on the basis of exclusion, but diagnostic criteria are overly inclusive, as in the case of FM (17). Patients attribute diagnostic delays to clinician knowledge, poor diagnostic criteria, lack of confirmatory tests, and the assumption that in the absence of physical evidence, symptoms are psychiatric or psychological in origin (34,73,85,87).

Patient narratives indicate that the diagnostic experience is an ambivalent one. On one hand, diagnostic labels provide relief from the turmoil of uncertainty and confirm that something is not right in the body (23,56,73,128,132,140). On the other hand, a diagnosis of contested illness can be burdensome in that it offers little hope for a cure (40,85,91,132). Their quest for an effective treatment is typically unsuccessful (138,9,45).

Coming to terms with a chronic illness is an ongoing daily process, described by some as akin to grieving (23,75). Realising there is no cure, patients must learn to live with their symptoms and the limitations imposed by them (33,44,47,54,70,73,75,89,140,142). This requires ongoing work and involves a transition to symptom monitoring and management (5,26,39,52,104,123,142). Different approaches to management primarily include symptom prevention (57,83) and alleviation (5,57,71,83,129,142). However, many also struggle with acceptance: they long for normality and refuse to give into the 'sick role' (23,75,89,91,104,128,130,136,140). Further, chronic illness can transform individuals' perspectives on life. Many describe redefining life goals, values, and obligations (8,10,56,91,138), shifting goals of recovery from finding a cure to an ability to partake in ordinary living (42,24,68). Uncertainty is prominent in patients' accounts (42,54,88,103,111,112,142) with outlooks on the future varying greatly. While some are hopeful for improvement, others fear their symptoms will only get worse with time

3.4.1.3 Identity loss and change

The limitations imposed by chronic illness can pose a threat to an individual's sense of self and change their relationship with their body. Several studies point to the biographical disruption experienced by individuals with FM and ME/CFS: Patient narratives reflect experiences of identity loss and change, both in relation to the past self and the future they had once anticipated (5,8,10,44,56,59,79,102,114,115,131). Stories individuals tell about their past selves highlight their positive qualities, and by extension preserve their true identities. In their narratives, they present themselves as 'good' persons and patients (46,55,135), having been strong, active and independent individuals before their illness (110,136,89). Individuals' accounts also describe changes in relationships with their body, which is experienced as changed, unreliable and unpredictable (70,83). The language used by PwC indicates that the unwell body is objectified and often perceived as separate from the true self: "it" is an obstruction (104), a burden (122) and stands between an individual and their ability to participate in life (70,83,89,108). Several studies describe a process of identity reconstruction in which individuals begin to let go of their past self and gradually rebuild their identity (8,10,37,93,131,137). Progress in grief work, cognitive realisation, diminished defensiveness, and space for self-reflection can facilitate the restructuring response (131).

3.4.1.4 Managing chronic illness

Coping with chronic illness means making changes and developing acceptable levels of self-management. Patients recognise that coming to grips with illness means taking charge over their own health (26,47,54,124,142), problem-solving (26) and developing skills to manage symptom flares and complications as they arise (128). Lifestyle changes are necessary, and patients describe adjusting their life activities to match limitations imposed by their condition (53,119). Among others, these include developing and keeping strict daily routines, making dietary changes, and engaging in relaxation practices (33,40,52,56,57,129).

The narratives of individuals with FM and ME/CFS illustrate the importance of listening to the body and responding accordingly (83). PwC learn to recognise the warning signs of relapse, highlighting the need to avoid stress and other triggers (52,57,71). Energy and

activity management are crucial and individuals with FM and ME/CFS describe pacing in order to prevent symptom flares (5,47,71,91,129,140). However, achieving an appropriate balance between rest and activity can be a challenge, particularly during low-symptom days. A positive mental attitude (47,68,94,104) and distraction (16,57,70,71). Access to relevant information and knowledge is a recognised enabler of change, empowering patients to take control over their healthcare (12,47,75,142).

3.4.1.5 Understanding and legitimacy

The feeling of not being understood or believed by others is a common experience among PwC (6,15,31,40,52,69,80,115,136). As the symptoms of FM and ME/CFS are not always overtly apparent to others, individuals can become the targets of scepticism and social insensitivity. Stigma and delegitimation occur across all contexts, including family, friends, workplace, welfare, and healthcare settings (43,93,107,110). Dismissive attitudes and growing disinterest are also described (8,15,22). Individuals report being accused of malingering, laziness, attention seeking, exaggerating their symptoms, and exploiting the social welfare system (11,15,35,69,75,107,142). Experienced as an assault on one's moral character (11), accusations of feigning illness can be an additional burden for PwC. Patients respond to delegitimation of their conditions by avoiding social interactions (6,14), limiting what information they share with others (11,76,128,136,142), attempting to conceal illness (71,73). While many actively seek to educate others about their condition (6,15) this can be a draining experience, which prompts PwC to maintain a certain amount of secrecy (121).

3.4.1.6 Support needs and experiences

Across the literature, individuals with FM and ME/CFS express a need for practical, emotional, and social support for themselves (78,130,112,58) and their families (35,78). Patient narratives, however, suggest that these needs are inconsistently met. Multiple studies report that individuals experience a lack of recognition and support from friends, family, workplace settings, physicians, and social welfare services (5,15,78,108,136). Others highlight positive experiences of support: feeling understood, cared for, and appreciated by family (6,33,35,69,75,78), supported in the workplace (2,69) and in healthcare settings (2,23). Some find support in solitude, turning to prayer and spirituality (121). Peer support networks in the form of patient groups and online

communities are a valuable resource for individuals with FM and ME/CFS (23,35,71,88). Such communities foster a sense of belonging, enabling individuals to express their concerns, exchange experiential knowledge, and make better sense of their illness (62,116). However, while the importance of mutual understanding is highlighted in patient narratives (31,116), the threat of delegitimation within support groups lingers (40).

3.4.1.7 Healthcare needs and experiences

Different expectations about healthcare services are expressed by PwC across the literature, but the fundamental need to be believed, validated, and supported by HCPs dominates their accounts. In addition to seeking practical guidance on coping and symptom management, PwC describe a need to be involved in decisions about their care (4,27,45,47,48,88) and expect clinicians to 'do no harm'(49). In contrast, their experiences of healthcare are often an uphill battle (80). Many PwC express dissatisfaction with various aspects of health services healthcare (20,46,48) including a long process of consultations and referrals (20,23) and limited access to specialist care (3). As a group, patients with contested illnesses experience a lack of support from the medical community (136,138): they feel lost or neglected by the system (103), like "medical orphans" (96) or test subjects (103,130). They describe having to repeatedly defend the legitimacy of their illness (61,80) and struggle against reluctant HCPs (14) to secure basic tests, referrals or treatments.

Patient-provider relationships are often suboptimal and a source of discontent. Individuals with FM and ME/CFS struggle to identify suitable HCPs to work with and express dissatisfaction with interactions related to specific aspects of their treatment (4,27,31,34,55,136,142). Doctors are perceived as unable or unwilling to help, and as failing to listen to patients' needs (33,76,90,136). Explanations and treatments offered are often inflammatory, particularly when psychological or psychiatric origins of illness are implied (11,80,90,96). The consequences of inappropriate explanatory frameworks can be tragic, as seen in the case of graded exercise for ME (e.g., (80)).

Much of the dissatisfaction with regards to healthcare can be attributed to lack of knowledge and understanding among HCPs (50,59,90,136). Patients perceive HCP to have major information gaps, lacking basic knowledge about FM and ME/CFS (27,80,136)

and their treatment (20,40,61). Consequently, patients report that their concerns are ignored and dismissed (5,43,50,69,77,80,90,130) and that they are seen as lazy, lacking motivation, lying, or faking their illness (6,15,78,80,86,87,90,114). The impact of gendered stigma on quality of care is described by both men and women with FM. Gendered and racialized stigma was explored by one study (107) which examined the unique experiences of Black women with chronic pain. Women presenting with unexplained pain report their symptoms are minimised or attributed to mental health, particularly when their physician is an older male (4). Men with FM describe issues related to toxic masculinity (103,120) and perceive women to be offered better quality of care (95).

While challenging healthcare experiences dominate patient narratives, positive experiences with specialist services and family physicians are reported (23,79,136). They describe therapeutic patient-provider relationships (73,4,27) though they recognise that they are fortunate to have this experience (23,55).

It is also noteworthy that alongside frustration and dissatisfaction, patients are cognisant of the effect their illness may have on the healthcare encounter. They describe how presenting with complex needs (106), multiple, fluctuating symptoms (46,98) and emotion (4) can overwhelm a consulting physician. They reflect on their own difficulties in accurately conveying their symptom experience, and how these complicate the diagnostic process (98). Despite criticisms of healthcare, patients appear empathic to clinicians' lack of understanding.

3.4.1.8 Managing healthcare encounters

Communication between physician and patient can be strenuous within the context of medical uncertainty. Patients, knowing that the alleviation of their suffering may rely upon productive HCP consultations, develop strategies for managing these encounters. They may attempt to gain control by directly confronting their clinician, managing their appearance and disclosures, or distancing and disengagement. Some describe efforts at educating HCPs on their condition, and demand specific tests, referrals, or treatments (12). Others attempt to "get through" to their HCPs by keeping their interactions direct and succinct (32).

Patients also use more subtle tactics to influence their healthcare process. They can control the consultation direction by selecting what to say, and if necessary, avoiding psychological disclosures (106). Having experienced stigma and delegitimation, they recognise that to be taken seriously it is important to fit the norm of a 'proper patient' (77). They do this by maintaining an appropriate level of assertiveness and the correct appearance. To be a credible patient, they must not look too healthy nor too sick and appear neither too strong nor too weak (134). Patients also describe working to maintain good relationships with their clinicians, and do so by avoiding disagreement, for fear of antagonizing HCPs (4,134). However, when healthcare encounters fail to bring comfort or resolution, patients can assert control, non-compliance, demonstrative distancing, and the ultimate strategy of "exiting" or disengagement (12).

3.4.2 Experiences of HCPs

A total of 197 findings reflecting the experiences and perspectives of HCPs were extracted from 31 studies. These were aggregated into 25 categories and 5 synthesised findings concerning *(i)* Beliefs and attitudes towards patients, *(ii)* Sensemaking at the limits of medical knowledge *(iii)* Consultation and management *(iv)*The patient-clinician relationship; and *(v)* Barriers and facilitators to care. The majority of reviewed studies featured findings linked to biomedical uncertainty, such as clinician attempts at making sense of their patients' symptoms (22 studies, 70%) or themes related to consultation and case management (19 studies, 61%). Findings concerned with clinician beliefs and attitudes towards patients, including positive and negative perspectives, were least frequent, though reported in 11 studies (See Table 3.3). Categories with most findings concerned diagnostic challenges (6.6%), formal education and training (6.6%) and the choosing the course of action (6.09%). The frequencies of extracted findings are presented in Appendix A7. Illustrative quotes for each synthesised finding are presented in Appendix A8 and are accompanied by HCP role, age and gender were reported by study authors.

3.4.2.1 Beliefs and attitudes towards patients

Clinicians' views of PwC are primarily negative. Across the literature, HCPs describe these groups as demanding, frustrating, time-consuming, and difficult to manage (13,61,65,67,84) and suggest that these patients pretend to be ill (18) or deliberately exaggerate the severity of their symptoms (13). Gender stereotypes and prejudice are expressed in the context of FM, with typical patients perceived to be problematic, "complaining women" (18,21,77). However, some HCPs are sympathetic to patients' suffering (18) and recognise the personal, familial and financial burden of illness (63). They acknowledge the strain of a lengthy and inconclusive healthcare journey (51) and recognise that patients come with a "perhaps with an appropriate chip on their shoulder toward the health care system that has failed them so far" (60).

3.4.2.2 Sensemaking at the limits of medical knowledge

The challenge of sensemaking at the limits of medical knowledge featured as a dominant thread throughout the literature. Clinicians lack explanatory frameworks needed to understand MUS, FM and ME/CFS and the link between emotional and physical

symptoms experienced by PwC (28,61,84,126). Inadequate case definitions for ME (30) and FM (61), as well as the complex, non-specific nature of symptoms (63) contribute to sensemaking difficulties, and are further compounded by the lack of clinical indicators of disease. These difficulties can translate into negative attitudes and scepticism towards FM and ME/CFS as legitimate medical conditions (28,58,63).

Experienced practitioners (25,127,141) and medical residents (60) recognise that the biomedical approach provides little guidance on prioritising symptoms and working with PwC. Gaps in knowledge concerning pathophysiological mechanisms of FM and ME/CFS limit options (20,29,58), and while pharmacological treatments for pain exist, clinicians express concern over the potential for misuse, abuse and dependence (61).

Establishing a diagnosis is a further challenge, particularly against a backdrop of inadequate diagnostic procedures and lack of objective clinical tests (21,25,61,63,109). Assessment often relies on patients' testimony, which is perceived as problematic in the absence of objective evidence (61,109). Clinicians describe arriving at diagnoses of FM (20), ME/CFS (30), and MUS (65) by a process of elimination, or by excluding other causes of symptoms. While this often involves extensive testing, HCPs note that patients struggle to accept their diagnosis (63,65).

Challenges experienced in relation to sensemaking, diagnosing and managing patients give rise to personal and professional frustration. An inability to help patients directly challenges clinicians' professional role and can give rise to feelings of unease, incompetence and helplessness (13,61,65,84,113). These frustrations can also accumulate in negative attitudes towards patients and cast doubts about the legitimacy of their symptoms (109). Insufficient medical knowledge leads clinicians to use alternate explanations for patients' symptoms. They describe using biopsychosocial explanations (60,109) and attribute symptoms to psychiatric illness, personality factors and a myriad of stressors, such as social disadvantage, trauma, and childhood adversity (13,60,61,67,84,84,92,139,143). They arrive at explanations by drawing on various sources of knowledge, including personal experience, socio-cultural knowledge, formal education, and clinical practice (28,29,60,63,67). The impact of personal experience on sensemaking is particularly noteworthy, as some HCPs describe truly understanding a condition once someone in their personal life had become ill.

3.4.2.3 Consultation and management

Consulting and managing PwC is a complex and often stressful task. A key challenge described by HCPs is finding a consultation structure when there is no obvious diagnosis or treatment (126), particularly when coupled with patient demands for referrals and investigations. Stressors identified include feeling unprepared for the consultation, making sense of vague or non-specific symptoms, and the potential of missing a serious disease (61,65,113,127). Working with this group of patients essentially requires clinicians to relinquish their quest for medical certainty. Many describe having to alter their process and outcome expectations (127,13,41,63), shifting focus from curing the patient to providing support and facilitating change (126,127). Several useful strategies for consulting patients with MUS are discussed, and include taking a comprehensive medical history, transparency regarding diagnostic interpretations and investigation outcomes, as well as clearly defining the consultation rules and structure (1,139). Clinical tests and examinations can be used to build trust and create structure to consultations by satisfying patients' needs for concrete action (1,13).

Consultations involve a nuanced assessment of risk and HCPs express concerns about potential harms of diagnostic labels and investigations. The clinical utility of diagnosis in the absence of clear treatment pathways is put into question, and similar difficulties are described in the context of investigations and referral practices. While receiving a diagnosis can empower, validate, and provide relief (20,28,58,125,126), clinicians recognise that a label of contested illness can misrepresent or stigmatise patients, negatively affecting the care they receive (30,125). Similarly, while clinical tests can provide reassurance, facilitate diagnosis, and enable access to specialist services, each procedure carries a risk of iatrogenic harm (30,125,133,63). Thus, despite pressure from patients, clinicians often refuse investigations perceived as dangerous and unnecessary. However, HCPs also describe motivations which may not necessarily be to the benefit of the patient. Referrals can serve as a 'way out' of difficult cases (77), while clinical tests can be used to 'speed up' the consultation and appease demanding patients (143). Testing also serves to relieve clinicians' anxiety over missing an organic pathology and the related threat of litigation (126,133). However, HCPs recognise that their referral practices can have a cascade effect and express immense frustration at referrals and admissions that serve no purpose other than the medicalisation of distress (127).

Approaches to symptom management vary depending on clinicians' beliefs and include the use of psychological and complementary therapies as well as education and psychosocial support within consultations (63,141). Many HCPs emphasise the need to establish shared responsibility for management, seeking to facilitate self-management and encouraging patients to take control over their journey (61,63,13). Simultaneously, they recognise their role in supporting patients and their ethical duty to help alleviate suffering. Clinicians provide practical supports by directing patients to appropriate resources and information, while also supporting them in the workplace and providing guidance on symptom management (28,63).

3.4.2.4 The patient-clinician relationship

While experiences vary, many clinicians describe building rewarding relationships with patients, characterized by partnership and founded on mutual trust, respect and unconditional positive regard (60,65,67,84,100,133). They express an awareness of the healing power of validation and make efforts to build a safe environment for patients, even in the absence of medical explanations (25,67,125,127,139). However, conflictual relationships also arise, particularly in cases of poor shared understanding (20,100). A range of strategies for managing patient relationships are discussed, depending on HCPs desire to engage with PwC. Some describe making a conscious effort to interact positively and build a helpful alliance (127), while others manage relationships by asserting a position of power and clearly defining the rules of a patient-doctor dynamic (61). Some distance themselves from patients completely, refusing to consult patients with FM or ME/CFS (13). Generally, HCPs desire to provide good explanations about the causes of patients' symptoms, though this is challenging in the context of MUS. They acknowledge uncertainty and describe attempts at normalising patients' experiences, providing reassurance, and emphasising the benign nature of their symptoms (100,133). HCPs also describe appealing to patients' worldview in the explanations provided, as this is more likely to foster trust and a mutual alliance (1,143).

3.4.2.5 Barriers and facilitators to care

Personal biases, organisational constraints, and consultation dynamics are identified by clinicians as barriers to effective management and care. HCPs describe how stereotyping thoughts (77), personal biases (51) and negative attitudes (139) towards patients can

lead to reduced clinical efforts and potential patient harm. Beliefs about their scope of responsibility (28,143) also affect care, particularly if clinicians are reluctant to support patients with concurrent physical and psychological complaints. At consultation level, appointment structure and the quality of contact with patients were identified as the key factors affecting care. Issues discussed by HCPs include suboptimal communication, characterized by an absence of dialogue and lack of psychosocial exploration (64), and concerns over patients' ability and readiness to self-manage (51).

Organisational constraints are a well-recognised challenge, and clinicians describe several interconnected barriers at this level. These include time pressures (51,133,143), difficult service structures (84) and poor continuity of care (51,133), as well as barriers with social welfare services (61). General practitioners do not feel sufficiently supported to work with patients in the community (84) and voice the need for third-sector support services and specialised treatment units (20,51). Time pressures and limited resources affect how clinicians prioritise patients and tasks, which can lead to the use of clinical shortcuts.

Many clinicians feel insufficiently prepared to work with patients with MUS and attribute this to inadequate formal education and training (60,65). They describe a discrepancy between their expectations of practice and clinical reality, particularly with respect to managing uncertainty. Much of their knowledge is acquired informally, by learning from colleagues and through practice (133,143). While role modelling can contribute to the transmission of negative beliefs and attitudes, senior clinicians (126) also emphasise teaching registrars to value patients and accept their ethical responsibility to care for them. With regards to specific training needs, clinicians describe a need for practical skills training in communicating with patients (143), and in managing difficult consultations (51). However, despite the recognised gaps in knowledge and understanding, many HCPs are confident in their clinical abilities (60,109) and express scepticism with regards to the value and feasibility of additional training (51).

3.5 Discussion

This review aimed to identify and aggregate qualitative evidence regarding the experiences of individuals with MUS, FM and ME/CFS and those of their healthcare professionals. Aligning with the core tenets of systems theory (Cross, 2018; Gomersall, 2018; Lipsitz, 2012; Sturmberg et al., 2014), a range of multifaceted, interconnected themes were identified with respect to both groups. The key findings are summarised below, with consideration of the issue of medical uncertainty in the context of person-centered care. Taken together, results point to patient-clinician dynamics as a viable target for change.

Studies with PwC provided rich accounts of the lived experience of illness, particularly with regards to the burden of symptoms and their widespread impact, transitions in the patient journey, experiences with social others and in the context of healthcare. Studies with HCPs primarily reflected concerns around the clinical management of PwC and the strategies used to manage challenging consultations. Sensemaking at the limits of biomedical knowledge was a common thread across the literature, underpinning patients' experiences of stigma, disbelief, and a general lack of understanding from others. Likewise, sensemaking difficulties coloured HCP's attitudes towards their patients, affecting their clinical efforts and quality of care. Lengthy and inconclusive healthcare journeys, characterised by diagnostic and prognostic uncertainty, and strained patient-clinician dynamics were described by both groups. Furthermore, PwC and HCPs identified gaps in knowledge and understanding as a major impediment to care and emphasised a need for advances in biomedical research.

The findings are largely in line with others: for example, Polakovská and Řiháček (2022) highlighted patients' fundamental need to be understood and believed by others, their experiences of isolation, uncertainty and a general disappointment in healthcare in the context of MUPS. Doebl et al. (2020) similarly illustrated FM patients' difficult interactions with the healthcare system, raising issues of inconsistent and poorly coordinated care. Johansen and Risor (2017) synthesised qualitative studies on family physicians' perception and management of MUS, pointing to an incongruence between patients' symptom presentations and the explanations offered by the biomedical model, while a recent literature review (Pheby et al., 2021) showed that disbelief, lack of

knowledge and understanding of ME/CFS among family physicians is widespread. In the context of ME/CFS and comparative research, Anderson et al. (2012) identified diagnostic issues and consultation dynamics, namely the struggle for power, as shared themes among patients and healthcare providers.

This synthesis highlights that medical uncertainty is a common, if not dominant, experience for both patients and clinicians and poses significant challenges to care. The problematic nature of uncertainty in healthcare has been widely acknowledged, and efforts have been made to elucidate the nature of the concept (Han et al., 2011, 2019; Kim & Lee, 2018). Uncertainty in medical-decision making can arise at various levels, including in the patient, clinician and their interaction (Dhawale et al., 2017). Diagnostic uncertainty, when mismanaged, can result in decreased patient satisfaction, confidence and trust in healthcare; contributes to stress and burnout among clinicians, and increases costs for the healthcare system (Meyer et al., 2021). While the concept of uncertainty *tolerance* is well-researched (Han et al., 2019, 2021), the literature indicates that clinicians are often hesitant to acknowledge or express uncertainty for the fear that this will undermine their professional authority (Etkind et al., 2017; Sallay et al., 2022; Simpkin & Schwartzstein, 2016). The literature also suggests that the manner in which clinicians communicate uncertainty can shape patients' illness experience and satisfaction with care (Etkind et al., 2017; Medendorp et al., 2021). In light of this, an evaluation of communication practices in the context of FM, ME/CFS and MUS might be a productive step for healthcare research.

Many individual-level barriers to shared decision-making have been identified to date, though patient health literacy appears to be the key issue (Brabers et al., 2017; Joseph-Williams et al., 2014; Scholl et al., 2018). For example, a Dutch cross-sectional study (Driever et al., 2020) found that despite preferences for shared decision-making, physicians often perceived patients as incapable of participating in decision-making, and thus practiced paternalistic approaches to care. Yet- as demonstrated in this review- there are circumstances where patients are more knowledgeable, both biomedically and practically, than clinicians. An exploration of consultation dynamics, communication and decision-making practices in this context could prove valuable, particularly as some evidence suggests knowledgeable patients are met with clinician resistance (Snow et al.,

2013). Interestingly, despite support for SDM in chronic illness management (Joosten et al., 2008; Montori et al., 2006) there is an apparent lack of research into SDM practices in the context of FM, ME/CFS and MUS. Bieber and colleagues (Bieber et al., 2008) report outcomes of a randomised controlled trial of SDM training for physicians, pointing to improved patient-clinician dynamics in FM care. Clearly, more work in this area is needed.

Taken together, findings related to PwC experiences of delegitimation and stigma indicate a fundamental need to improve societal understanding and awareness of the burden of FM, ME/CFS and other chronic, invisible, and contested illnesses. This need is supported by a considerable body of literature which points to the psychological and functional consequences of stigma in ME/CFS and chronic pain (Baken et al., 2018; Bean et al., 2022; Ko et al., 2022; Zhang et al., 2021). However, two decades of reviewed research has shown these issues to be persistent, and that deeply ingrained negative beliefs are both enduring and resistant to change. Similar themes have emerged from the narratives of patients with Long-Covid (Kingstone et al., 2020) and recent evidence points to an association between social stigma and poor mental health-related quality of life (Scholz et al., 2023). Thus, it may be more fruitful to identify and support the skills that enable PwC to live well despite uncertainty, invalidation, and disbelief. For example, Goldberg et al. (Goldberg et al., 2022) suggest that a key role for mental health professionals in supporting Long-Covid patients is essentially through *validating* their illness experience, facilitating coping and adaptation, while also managing any pre-existing or secondary mental health issues. Based on the findings of this review, arguably such suggestions can be extended to any illness characterised by disbelief, delegitimation and uncertainty.

3.5.1 Strengths and Limitations

The primary strength of this review lies in its broad scope and the volume of aggregated findings. The heterogeneity of primary studies with respect to qualitative and analytic procedure was expected and meta-aggregation was selected in anticipation of such differences. With its deliberate focus on commonalities (Lockwood et al., 2015), meta-aggregation was suitable for identifying experiences in the context of distinct conditions underpinned by the core issue of medical uncertainty. However, as elaborated

elsewhere (Bergdahl, 2019), the pragmatism of meta-aggregation comes at a cost. The key issues relevant to this review are considered below.

First, meta-aggregation seeks to generate a descriptive categorization and mapping of extracted qualitative findings (Lockwood et al., 2015; Lockwood & Pearson, 2013; Munn et al., 2021; Sim & Mengshoel, 2023). Frequency counts can be used to comment on dominant phenomena, but little more can be said about their meaningfulness or representativeness. Further, owing to the meta-aggregative focus on commonalities, this review falls short in discussion of rare or discrepant findings. Perhaps the most significant criticism of the approach is that it does not seek to generate theory, making it incompatible with the broad aims of meta-synthesis (Bergdahl, 2019). Although the findings are positioned broadly within systems thinking, they should not be assumed to- nor claim to- advance systems theory. This review of patient and clinician perspectives provides insights into the individual and wider social context of chronic illness, but more focused work is needed to move beyond description.

Additionally, the strength of the conclusions is ultimately limited by the quality of included studies. Few studies clearly referred to the researchers' personal or theoretical orientation and their impact on the research conducted. Consequently, the strength of evidence for all but one synthesised finding was downgraded, resulting in Medium or Low ratings of Confidence. While a critique of quality appraisal tools is beyond scope, some concerns have raised with regards to JBI Critical Appraisal tool (Majid & Vanstone, 2018). Among others, the lack of explicit reference to philosophical orientation may simply reflect reporting choice, rather than poor methodological quality. Thus, the ConQual rating for synthesised findings should be regarded with caution.

While that the inclusion of "unexplained" symptoms, or MUS, alongside FM and ME/CFS is controversial, this decision is anchored by the fact that research has, historically, employed MUS as an umbrella term encompassing a large variety of poorly understood syndromes. An attempt to review two decades of research should not shy away from an issue that exemplifies the challenge of medical uncertainty. While efforts were made to include variations of terms relevant to MUS, FM and ME/CFS, many of the reviewed studies used different case definitions, or made vague reference to the duration or origin of symptoms experienced by participants. Thus, it was not always possible to determine

whether participants' symptoms were primary ("unexplained") or secondary fatigue and pain. Overall, it is clear that reporting practices in qualitative research need substantial improvement.

Ultimately, this review highlights the value in conducting multi-perspective research to better capture the complexity of the illness experience. While patients, clinicians and academics are increasingly advocating for the involvement of multiple stakeholders in healthcare-related research (Fitzcharles et al., 2017; Price et al., 2018), very few qualitative studies appear to do this in the context of MUS, FM and ME/CFS. Partnership in research- and in clinical practice- is much needed. If consultation dynamics are to be fully understood and improved, both parties must be represented in research. However, this research should occur in a context of epistemic justice, whereby patients' experiential knowledge is valued and treated as equal to clinicians' scientific expertise.

3.5.2 Conclusion

This meta-aggregative review of qualitative research provides a comprehensive description of the experiences of individuals with MUS, FM and ME/CFS as well as those of healthcare professionals. A range of multifaceted themes with respect to both groups were identified, many of which appear underpinned by sensemaking difficulties. While gaps in biomedical knowledge are clear, this review highlights the need to address the patient-clinician dynamic in the context of uncertainty and calls for the involvement of multiple stakeholders in health-related research. Policies and practices should prioritise a holistic, patient-centered approach, supporting both patients and clinicians to engage in shared decision-making despite medical uncertainty.

Building on the findings of Phase One, particularly with regards to the identified stigma and gaps in knowledge and understanding, Chapter 4 reports a cross-sectional study designed to explore the beliefs and perspectives of a sample of adults with knowledge of FM and/or ME/CFS in Ireland, as well as their perspectives on public and clinician awareness and knowledge of these.

Chapter 4. Study Two

Cross-Sectional Survey

4.1 Introduction

Individuals living with chronic, invisible, and poorly understood illnesses encounter a wide array of personal, social, and systemic challenges. These challenges often stem from the "contested" nature of their conditions, where legitimacy and recognition are frequently questioned. As reviewed in *Chapter 3*, disbelief, invalidation, and stigma emerge as central aspects of the illness experience, particularly for conditions characterized by disparate, and historically "medically unexplained" symptoms, including Fibromyalgia (FM) and Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS). This body of work illustrates how the invisibility of symptoms, compounded by incomplete or inaccurate biomedical understanding, often manifests in stigma, resulting in a lack of empathy and rejection from others, including family members and healthcare professionals.

Stigma associated with concealable illnesses has been long recognised and theorised, with foundational work by Goffman (1963) and more recent contributions by Vickers (2000) and Quinn and Earnshaw (2011, 2013) providing insights on the phenomenon. Goffman's (1963) conceptualisation of stigma as "blemishes of character" is particularly relevant, whereby individuals with invisible conditions frequently face attacks on their moral character, presumably because their presentation deviates from normative expectations about what illness "should" look like. When symptoms are not outwardly apparent, societal biases and misconceptions may lead judgments about character that undermine the legitimacy of the individual's suffering. Vickers (2000) expands on this by introducing the concept of knowledge-based stigma, highlighting how incorrect, absent or incomplete knowledge among the public or professionals allow myths and misconceptions to perpetuate stigmatizing attitudes. Specifically, when faced with the ambiguity of these invisible illnesses, people may rely on myths and beliefs to fill knowledge gaps, potentially leading to discriminatory behaviours.

The profound impact of stigma on the wellbeing of individuals with concealable illnesses is also well-established, with evidence for considerable distress arising as a result of negative societal perceptions, as well as the internalisation of stigmatizing beliefs (Earnshaw & Quinn, 2012; Quinn & Earnshaw, 2013). Enacted, anticipated and internalised stigma have shown, for instance, to negatively impact the quality of life, help-seeking behaviour, treatment adherence and self-esteem of individuals with invisible illnesses (Earnshaw et al., 2012; Engebretson, 2013; Kao et al., 2016; O'Donnell & Habenicht, 2022). Given the role of knowledge and normative beliefs about illness in stigma, targeting public misconceptions and informational gaps appears a viable route to alleviating some of the psychosocial burden experienced by individuals with invisible conditions, including those falling under the umbrella term of "medically unexplained" illness. Patient-led advocacy groups, unsurprisingly, dedicate their efforts to this, seeking to educate the public and raise awareness about specific conditions (Bloom et al., 2021; Hartley & Penlington, 2023; Keller & Packel, 2014). As discussed in *Chapter 2*, a panel of patients involved in this research also highlighted their concerns around stigma, and emphasised the importance of exploring public knowledge about, understanding of and attitudes towards a range of poorly understood conditions.

4.1.2 Objectives

Considering the priorities identified by the patient panel, the deficit in knowledge and understanding identified in the literature reviewed in *Chapter* 3, as well as the relationship between knowledge and stigma, the aims of the present study were *(i)* to explore the perspectives of a sample of adults familiar with FM and ME/CFS in Ireland, with a focus on understanding their personal beliefs and perceptions of these conditions and their clinical management; *(ii)* to explore their appraisals of public and healthcare professional knowledge and awareness of these, and *(iii)* to explore their perceptions of the challenges faced by individuals living with FM (PwFM) and ME/CFS (PwME). These two conditions were chosen as exemplars due to their long history of being dismissed, and in light of a remarkable absence of information on attitudes towards them in Ireland.

4.2 Methodology

4.2.1 Design

The study was designed as a cross-sectional study, employing an anonymous, mixed-methods survey administered online using the survey platform Qualtrics (Qualtrics, Provo, UT). This study adhered to the STROBE guidelines for reporting cross-sectional studies (von Elm et al., 2008). The study design was approved by the Ethics Committee at the School of Psychology, Trinity College Dublin (Appendix B1).

4.2.1.1 PPI

The survey was designed in collaboration with individuals with lived experience of poorly understood chronic illness, who were part of a patient panel involved in the research, described previously in Chapter 2 (*Section 2.4.2*). PPI contributors identified a need to explore the perspectives of the Irish public on conditions with unexplained symptoms, including FM and ME/CFS as exemplars. Their input on survey content was sought, and suggestions regarding appropriate terminology and phrasing were discussed and incorporated into the design. PPI contributors also assisted with survey distribution among their personal networks.

4.2.2 Recruitment

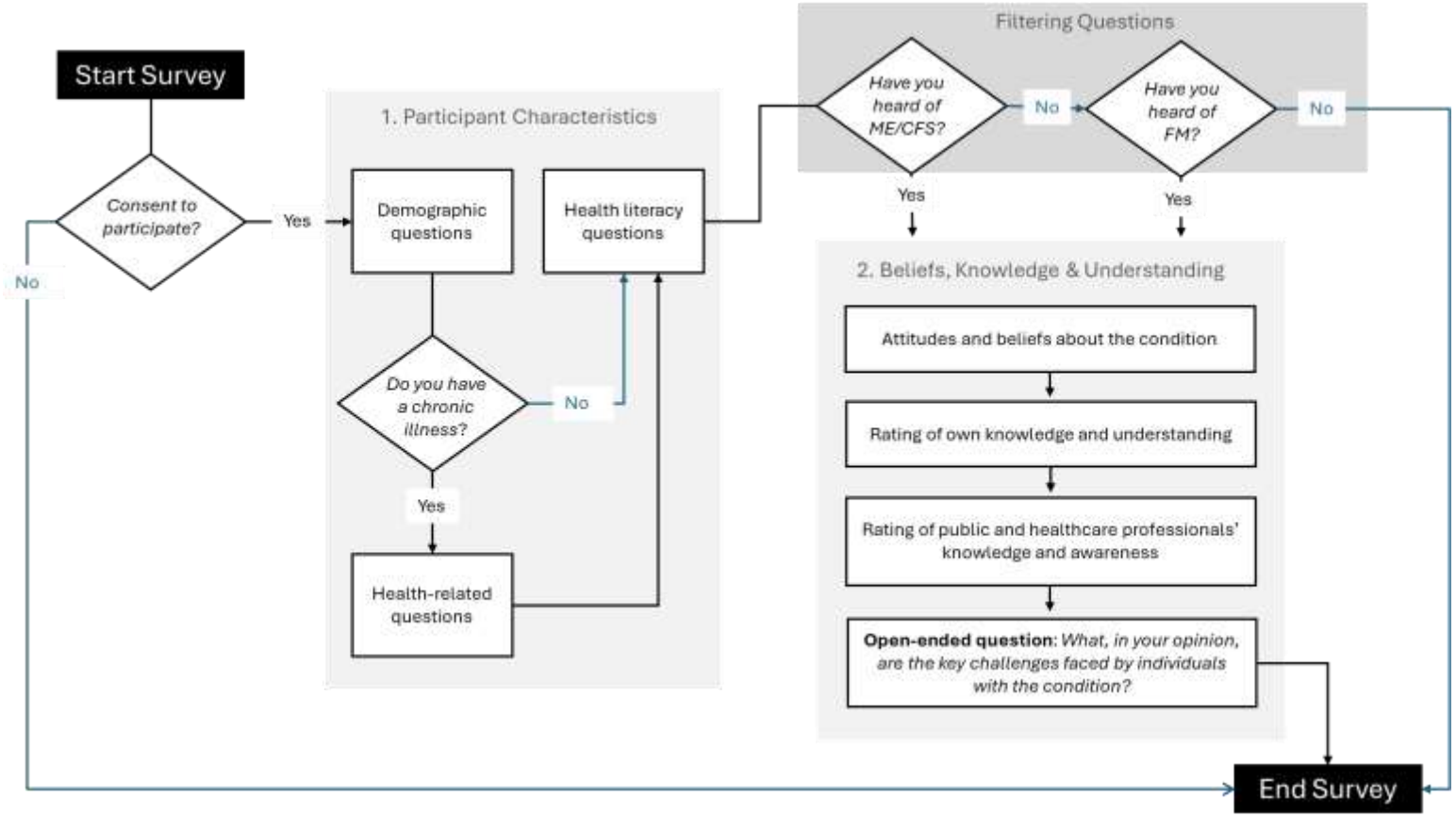
A snowball sampling strategy was used within a purposive framework to recruit participants who were familiar with FM and/or ME/CFS. This non-representative sampling approach facilitated the identification of individuals with a presumed baseline level of awareness, thus ensuring the inclusion of suitable respondents to meet study aims. Recruitment commenced in May 2022 and the survey remained open till January 2023. Participants were recruited via social media, drawing on the researcher's personal networks and the assistance of PPI contributors and patient advocacy groups. All consenting individuals who were aged 18 and over, and residing in the Republic of Ireland, were eligible to take part. Screening questions were included in the survey to identify the sample of interest.

4.2.3 Procedure

Consenting participants were asked to complete questions related to their demographic characteristics, self-rated health literacy, and perspectives on ME/CFS and/or FM. All participants responded to the demographic and health literacy questions. The option to skip questions was available by selecting "*Prefer not to say"*, and while participants were able to return to skipped questions, they were not able to re-access the questionnaire or modify their responses once the survey was submitted. As the study specifically sought to elucidate the perspectives of individuals on these two conditions, mandatory filtering questions were added *("Have you ever heard of Fibromyalgia before?" and "Have you ever heard of ME/CFS before?"*) following the demographic and health-literacy questions, such that only those who indicated knowing of the condition could proceed to the relevant survey sections. Participants who were familiar with both conditions were asked to complete questions on each, with relevant survey sections becoming available in succession. Figure 4.1 presents the survey flow and contents of each section.

Figure 4.1

Flowchart of survey procedure.



4.2.4 Measures

The survey questions were specifically designed and developed for the purpose of this study to address the research objectives and gather relevant data. The measures with sample questions are outlined below, with the full questionnaire included in Appendix B2.

4.2.4.1 Demographic Characteristics and Health

The demographic questionnaire included questions about age, gender identity (Male, Female, Non-Binary, Prefer not to say) and highest level of education achieved (No level of education, Primary level, Secondary level, Third level, Post-secondary training, Prefer not to say). The health questionnaire included binary-response (Yes/No) questions about personal experience of chronic illness ("*I am a person with a chronic, poorly understood condition")* and experience of chronic illness in others *("I know someone with a chronic, poorly understood condition"*, "*I have a relative or significant other with a chronic, poorly understood condition*").

Participants who selected having personal experience with chronic illness (PwC) were asked to disclose the condition(s) they live with and to indicate whether they have been formally diagnosed (Yes/No). Participants were also asked to select the type of symptoms they primarily experience (Physical, Psychological, Cognitive, Other) and whether they experience symptoms that are "Medically Unexplained" (Yes/No/Unsure). They were also asked to rate the severity of their symptoms most of the time (Mild, Moderate, Severe), and to rate the overall impact of chronic illness on their Quality of Life using a 5-point Likert scale (No Impact-Severe Impact).

4.2.4.2 Self-rated Health Literacy

Participants were asked to respond to five health literacy statements which were designed for the purposes of the study. For each statement, participants indicated their level of agreement on a 5-point Likert scale (Strongly disagree- Strongly agree). These statements concerned their self-perceived knowledge about health, wellbeing and healthcare (e.g., "*I consider myself to be knowledgeable about topics related to health and wellbeing*", "*I consider myself health literate- I can obtain, read, understand, and use healthcare information to make appropriate health decisions and follow instructions for*

treatment", *"I have a good level of knowledge and understanding of common chronic illnesses (e.g., diabetes, heart disease)"*)

4.2.4.3 Beliefs, Knowledge and Understanding

Participants who indicated knowing about ME/CFS and/or FM were asked twenty questions about their perspectives on each condition. The order of survey questions in this section was randomized automatically using the Qualtrics survey platform to minimise order effects and reduce potential response bias (see Figure 4.1). Participants were asked multiple-choice questions regarding how common the condition is, what they perceive the primary cause to be, and were asked to indicate their agreement (Strongly disagree- Strongly agree) with six statements gaging their beliefs about ME/CFS and FM and people living with it (e.g., *"Fibromyalgia is a real condition", "People with fibromyalgia exaggerate their symptoms."*).

Participants were also asked to rate their understanding of various aspects of the condition and its clinical management (e.g., "*How would you rate your understanding of the treatment of fibromyalgia?*") on a five-point Likert scale ranging from Very Poor to Very Good, and to estimate average time to diagnosis of the condition. Similarly, participants were asked to rate public and health professionals' knowledge and awareness of the condition. Three comparative questions were included in which participants were asked whether the diagnosis, management and experience of the condition is easier, similar to or harder than of other chronic illnesses. Finally, participants answered an open-ended question about the perceived challenges faced by individuals affected by the condition (e.g., *"What, in your opinion, are the key challenges faced by individuals with ME/CFS?"*)

4.2.5 Analysis

4.2.5.1 Exploratory Analyses

Descriptive statistics (means, frequencies) were calculated for all measures. Only data from participants who completed the survey were included in analyses. Data were then split into participant subgroups, i.e. participants familiar with ME/CFS and participants familiar with FM, and exploratory chi-square tests of independence were conducted to examine associations between participants' beliefs, ratings of knowledge and

understanding, and personal factors such as experience of chronic illness. Bonferroni adjustments for multiple comparisons were applied.

4.2.5.2 Thematic Analysis

Participant responses to the open-ended question were analysed using an inductive thematic approach (Braun & Clarke, 2006, 2021a) with NVivo 12 Plus Software, the assumptions of which are discussed in Chapter 2 (*Section 2.4.1.2*). The credibility and trustworthiness of the analysis was ensured by drawing on the seminal work of Lincoln and Guba (1985) and the pragmatic guidelines outlined by Nowell et al. (2017). Separate analyses were conducted for the answers of the two participant subgroups, with an identical process applied for each dataset. Each qualitative analysis was performed by the author and a research assistant (AH and JH), ensuring the research team comprised of people with various levels of expertise on topics of chronic illness. Both research assistants were final year undergraduate Psychology students with an interest in health psychology and invisible illness, though no previous experience with research on the topic. Research assistants were trained in inductive thematic analysis and NVivo by the author.

Thematic analysis involved six steps: *(i)* First, researchers familiarised themselves with the data and met to discuss initial impressions; *(ii)* Systematic data coding followed, with researchers independently performing initial coding and categorisation according to similarity. Researchers met to compare codes and condensed these into more focused codes and categories; *(iii)* Categories were organised into initial themes and sub-themes and reviewed by researchers using Patton's (1990) dual criteria of internal homogeneity and external heterogeneity; *(iv)* A codebook was devised and preliminary themes were discussed with the patient expert panel. Feedback was incorporated into the thematic structure and codebook, with the labels and descriptions revised to ensure shared understanding among researchers. Free-text responses were coded once again as per codebook and a coding comparison query was carried out in NVivo to ensure reliability of the coding process. A Cohen's cappa coefficient was calculated as a measure of inter-rater agreement between the author and research assistant for each thematic analysis. The coding comparison of qualitative data on ME (ND & JH) achieved a Cohen's kappa of 0.62, corresponding to a good level of agreement. A Cohen's kappa coefficient of 0.75

was achieved for the analysis of FM data (ND & AH), indicating a very good level of inter-rater agreement. *(v)* Researchers refined the codebook to include more precise definitions of themes. *(vi)* Quotations corresponding to each theme were identified and the report produced. Descriptive results were accompanied by illustrative quotes. As member checking was not possible given the anonymous nature of the survey, results of the analysis were presented to the patient expert panel for review.

4.3 Results

4.3.1 Respondent characteristics

The survey was initiated by 349 respondents with a completion rate of 91% (*N*=319). Respondents who completed the survey were aged 19 -74, primarily middle aged (*M*= 46.8, *SD*=12.3), female (80%) and highly educated (87%). In general, participants rated themselves as health literate (90%) and reported having a good level of knowledge about common chronic conditions, though fewer (36%) reported the same for acute illness. Three in four survey respondents reported knowing someone living with a chronic illness, and 66% disclosed having a chronic illness themselves. While the majority were familiar with the terms "*chronic fatigue*" and "*chronic pain*" fewer recognised the clinical entities ME/CFS (94% vs. 75%) and FM (97% vs. 88%). Table 4.1 provides an overview of demographic characteristics of all respondents, including participant subgroups. Of participants living with chronic illness (PwC, *n*=211), similar proportions reported one (49%) or multiple (51%) conditions, and most (90%) indicated having a formal diagnosis for their chronic illness. Symptoms experienced were primarily of a physical nature (69%) and moderate severity (65%), with a moderate or high impact on Quality of Life (35% and 44% respectively). Over half of PwC indicated having symptoms that could be considered "medically unexplained" (54%), and one in five (22%) reported having comorbid physical and mental health conditions (see Table 4.2).

Table 4.1

Demographic characteristics and self-reported health literacy of survey respondents (N=319) and participant subgroups.

	Survey respondents (*N*=319)		Familiar with ME/CFS (*n*=238)		Familiar with FM (*n*=280)	
	n	%	*n*	%	*n*	%
Gender						
Female	256	80.3	201	84.5	234	83.6
Male	57	17.9	33	13.9	40	14.3
Non-Binary	4	1.3	3	1.3	4	1.4
Highest education level achieved						
Primary Level	2	.6	1	.4	2	0.7
Secondary Level	38	11.9	29	12.2	34	12.1
Professional Training or Third Level	276	86.5	208	87.4	242	86.4
Chronic illness						
Personal experience [a]	211	66.1	176	73.9	195	69.6
Know others with [a]	240	75.2	185	77.7	214	76.4
Have relative with [a]	138	43.3	105	44.1	124	44.3
Self-Reported Health Literacy						
Knowledgeable about health [b]	255	79.9	198	83.2	231	82.5
Health Literate [b]	286	89.7	218	91.6	253	90.4
Aware of available health services [b]	221	69.3	170	71.4	198	70.7
Good knowledge of acute illness [b]	114	35.7	84	35.3	98	35.0
Good knowledge of chronic illness [b]	252	79	200	84	230	82.1
Awareness of ME/CFS						
Familiar with term “chronic fatigue” [a]	300	94	238	100	272	97.1
Familiar with ME/CFS [a]	238	74.6	238	100	223	79.6
Awareness of FM						
Familiar with term “chronic pain” [a]	310	97.2	237	99.6	280	100
Familiar with term FM [a]	280	87.8	223	93.6	280	100

Note. Survey respondents: Age 19-74 (*M*= 46.8, *SD*=12.35). Respondents familiar with ME/CFS: Age 20-74 (*M*=48.4; *SD*=12.02). Respondents familiar with FM Age: 19-74 (*M*=47.6; *SD*=12.13)

[a] Reflects the number and percentage of participants answering “Yes” to this question.

[b] Reflects the number and percentage of participants who answered “Agree” and “Strongly Agree” to the statement

Table 4.2

Illness burden among participants with a self-disclosed chronic illness (PwC)

	PwC (*n*=211)		PwC Familiar with ME/CFS (*n*=176)		PwC Familiar with FM (*n*=195)	
	n	*%*	*n*	*%*	*n*	*%*
Participant health						
One condition reported	103	48.8	83	47.2	91	46.7
Multiple conditions reported	108	51.2	93	52.8	104	53.3
Physical and Psychological Comorbidities	46	21.8	39	22.2	44	22.6
Medically unexplained symptoms reported	114	54	98	55.7	105	31.8
Formally diagnosed	189	89.6	159	90.3	174	89.2
Primary Symptoms Experienced						
Physical	145	68.7	120	68.2	138	70.8
Affective	30	14.2	22	12.5	27	13.8
Cognitive	11	5.2	10	5.7	7	3.6
"All"	10	4.7	8	4.5	8	4.1
Other	15	7.1	15	8.5	15	7.7
Severity of Symptoms						
Mild	11	5.2	11	6.3	10	5.1
Moderate	137	64.9	111	63.1	128	65.6
Severe	63	29.9	54	30.7	57	29.2
Impact of Symptoms on Quality of Life						
Low	12	5.7	12	6.8	11	5.6
Moderate	73	34.6	54	30.7	67	34.4
High	93	44	80	45.5	86	44.1
Severe	32	15.2	30	17.0	31	15.9

4.3.2 Knowledge and understanding of ME/CFS and FM

A subsample of 238 participants familiar with ME/CFS responded to questions regarding their beliefs, knowledge and understanding of ME/CFS and their perception of public and clinician knowledge and awareness. Of these participants, 74% (*n*=176) were PwC, of whom 31.2% (*n*=55) reported living with ME/CFS (PwME) and 68.8% (*n*=121) reported other chronic conditions. A subsample of 280 participants familiar with FM responded to questions regarding their beliefs, knowledge and understanding of FM and that of others. Of these participants, 70% (*n*=195) were PwC, of whom 77% (*n*=150) reported living with FM or Chronic Pain (PwFM) and 23% (*n*=45) reported other conditions. Further demographic characteristics are detailed in Appendix B3. Descriptive statistics for key measures for each subgroup are included in Appendix B4. and presented in Figures 4.2 and 4.3.

4.3.2.1 Beliefs about ME/CFS and FM

Most of the participants familiar with "ME/CFS" (*N*=238) agreed that ME/CFS is a legitimate condition (95%, *n*=226), and two in three (65%, *n*=156) considered it to be common or very common. As shown in Figure 4.2 (Panel A and B), there was a strong recognition of the burden of illness experienced by PwME, with 97% (*n*=231) acknowledging the significant suffering and 72% (*n*= 171) agreeing that it is more difficult to live with compared to other chronic conditions.

However, beliefs about the cause of ME/CFS were more varied. Less than half of the participants (48%, *n*=113) believed that the primary cause of ME/CFS is biological, while 42% (*n*=101) were unsure about the aetiology. A minority (10%, *n*=23) believed the cause to be primarily psychological. Only a small proportion (14 %, *n*=34) believed that it is possible to cure ME/CFS, while 32% (*n*=76) felt that it is possible to effectively manage the symptoms. Most participants considered ME/CFS to be more challenging to manage (78%, *n*=186) and more difficult to diagnose (85%, *n*=203) compared to other chronic conditions. When asked to estimate time to diagnosis, only 11% (*n*=27) of participants assumed diagnosis would occur within a 1-year timeframe. Further, a small proportion of participants held negative beliefs about PwME, with 4% (*n*=10) believing that individuals with ME/CFS exaggerate their symptoms and 1% (*n*=3) suggesting that they may be pretending to be ill.

Similar trends were observed in the responses of participants familiar with “Fibromyalgia” (*N*=280). Overall, participants recognised that FM was real, common, and more difficult to diagnose, live with and manage than other chronic conditions. Nearly 50% considered the primary cause to be biological (*n*=133) and almost all (98%, *n*=274) agreed that PwFM are suffering. As shown in Figure 4.2, Panel C, a relatively small proportion believed that the primary cause of FM is psychological (15%, *n*=43), that PwFM exaggerate the severity of their symptoms (3%, *n*=7) or are pretending to be ill (1%, *n*=3). Most respondents estimated that that diagnosis of FM takes up to 3 years (26%, *n*=73). Perspectives on outcomes were unfavourable: Few believed that it is possible to cure FM (12%, *n*=33) or effectively manage symptoms (38%, *n*=106).

Figure 4.2.

Participant beliefs about and attitudes towards ME/CFS and FM

Figure 4.2. Comparison of key measures across participant subgroups. Panels A and B show responses of participants familiar with ME/CFS to questions concerning their beliefs about ME/CFS and attitudes towards PwME, as well as their perspectives on living with, diagnosing, managing and treating the condition. Panels C and D show responses of participants familiar with FM to questions concerning their beliefs about FM and attitudes towards PwFM, and their perspectives on living with, diagnosing, managing and treating the condition.

4.3.2.2 Perceived knowledge, understanding and awareness of ME/CFS and FM

Figure 4.3 shows participants' appraisals of their own knowledge and understanding, and their ratings of public and healthcare professionals' knowledge and awareness of ME/CFS and FM. Overall, participants generally reported a low or average level of self-rated knowledge and understanding of both ME/CFS and FM, encompassing its causes, diagnosis, treatment, and management, though ratings are slightly more favourable in the case of FM (see Figure 4.3, Panels A and B). Only 38% (*n*=107) of participants felt that they had a good level of knowledge about FM, with fewest participants endorsing a good level of knowledge about causes (20%, *n*=55). By comparison, 29% (*n*=68) of participants indicated that they had a good or very good overall understanding of ME/CFS, while fewest (16%, *n*=38) rated themselves as having a good level of knowledge about treatment.

Participant ratings of public and clinician knowledge and awareness of both conditions are predominantly poor. In the case of ME/CFS (Figure 4.3, Panel C) a small minority of participants assessed the knowledge and awareness of healthcare professionals (6%, *n*=15) and the general public (<1%, *n*=2) as good or very good. Similarly, only 3% (*n*=8) endorsed good knowledge and awareness about FM among the public, and 6% (*n*=16) among healthcare professionals (Figure 4.3, Panel D). Approximately one in four, and one in three participants considered healthcare professionals to have an average level of knowledge and awareness of ME/CFS and FM, respectively.

Figure 4.3.

Participant ratings of knowledge and understanding of ME/CFS and FM

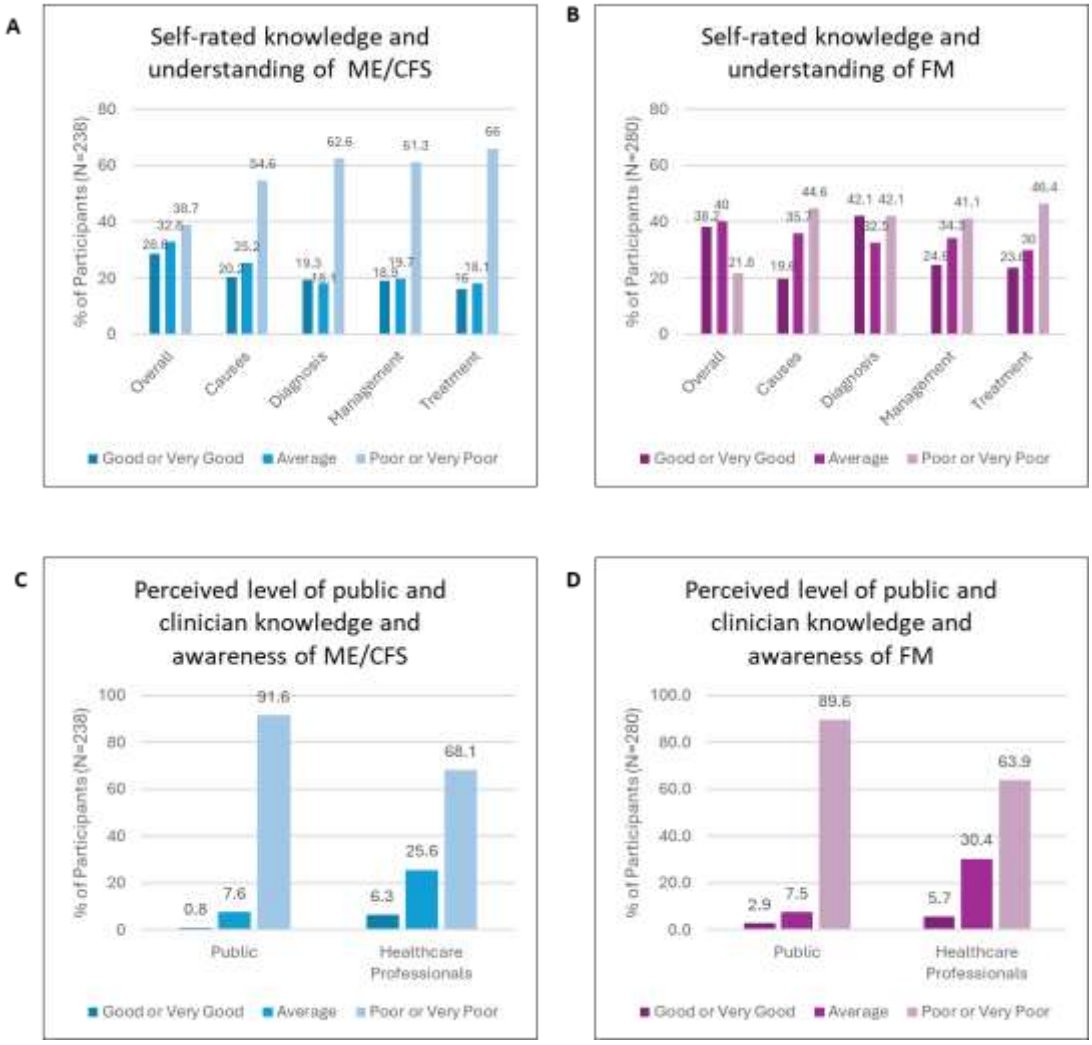


Figure 4.3. Comparison of knowledge and understanding ratings across participant subgroups. Panels A and C show responses of participants familiar with ME/CFS to questions concerning their own levels of knowledge and understanding of ME/CFS (A) and their perception of knowledge and awareness among healthcare practitioners and the general public (C). Panels B and D show the parallel responses of participants familiar with FM.

4.3.3 Exploratory Analyses

4.3.3.1 Knowledge and Understanding of ME/CFS

Chi square tests of independence were conducted to compare the beliefs about aetiology, clinical management and ratings of knowledge and understanding of ME/CFS among participants with a chronic illness (PwC, *n*=176) and those without chronic illness (PwO, *n*=62), detailed in Appendix B5. While the largest proportion of PwC considered the primary cause of ME/CFS to be biological, and the largest proportion of PwO were unsure of cause, no significant association was found between having a chronic illness and beliefs about aetiology.

Although participants with chronic illness were more likely to believe that the effective management of ME/CFS is not possible (28%) than PwO (16%), both groups indicated similar levels of uncertainty (43% and 44%, respectively), and no association between health and belief was found. A small, significant association was found between health status and beliefs about curing ME/CFS, $\chi^2(2) = 7.79$, $p = .02$, Cramer's $V = .18$, with PwO generally being more optimistic, and PwC more pessimistic, about the possibility of curing the condition (Figure 4.4, Panel A). There were no significant differences between the groups in terms of self-rated overall understanding of ME/CFS, nor were there differences in ratings of public knowledge and awareness, with both PwC and PwO rating it as poor to very poor (93% and 87%, respectively).

A significant association was identified between illness status and ratings of healthcare practitioner knowledge and awareness of ME/CFS, $\chi^2(2) = 11.01$, $p = .004$, Cramer's $V = .21$. A higher proportion of PwC rated healthcare practitioner knowledge as poor to very poor (72%), and lower proportion as Good or Very Good (3%) compared to PwO (Figure 4.4, Panel B). These results remained significant after applying a Bonferroni adjustment for multiple comparisons ($\alpha = .008$).

Given the observed differences between participants with and without chronic illness, further exploratory analyses were conducted to compare the perspectives of respondents with ME/CFS (PwME, *n=55*) to those with other chronic illnesses but not ME/CFS (*n*=121) as detailed in Appendix B6. Significant associations were observed in participants' beliefs regarding the aetiology and potential for curing ME/CFS (Figure 4.4, Panel C and D). PwME were more likely to endorse a biological cause for the condition compared to those without ME/CFS, $\chi^2(2) = 17.1$, $p < .001$, Cramer's $V = .31$, and they were also more likely to disagree with the notion that the condition can be cured, $\chi^2(2) = 14.19$, $p < .001$, Cramer's V = .28. A significant association was found between illness status and self-rated knowledge and understanding of ME/CFS, $\chi^2(2) = 41.11$, $p < .001$, Cramer's V = .48, as well as between illness status and ratings of healthcare practitioners' knowledge and awareness of ME/CFS, $\chi^2(2) = 12.8$, $p = .002$, Cramer's $V = .27$. Compared to participants without ME/CFS, PwME were more likely to rate their own knowledge and understanding as good or very good (64%) and were more likely to rate healthcare practitioners' knowledge as poor or very poor (89%), as shown in Figure 4.4 Panels E and F.

Figure 4.4

The clinical management and perceived knowledge and awareness of ME/CFS among participants with and without chronic illness.

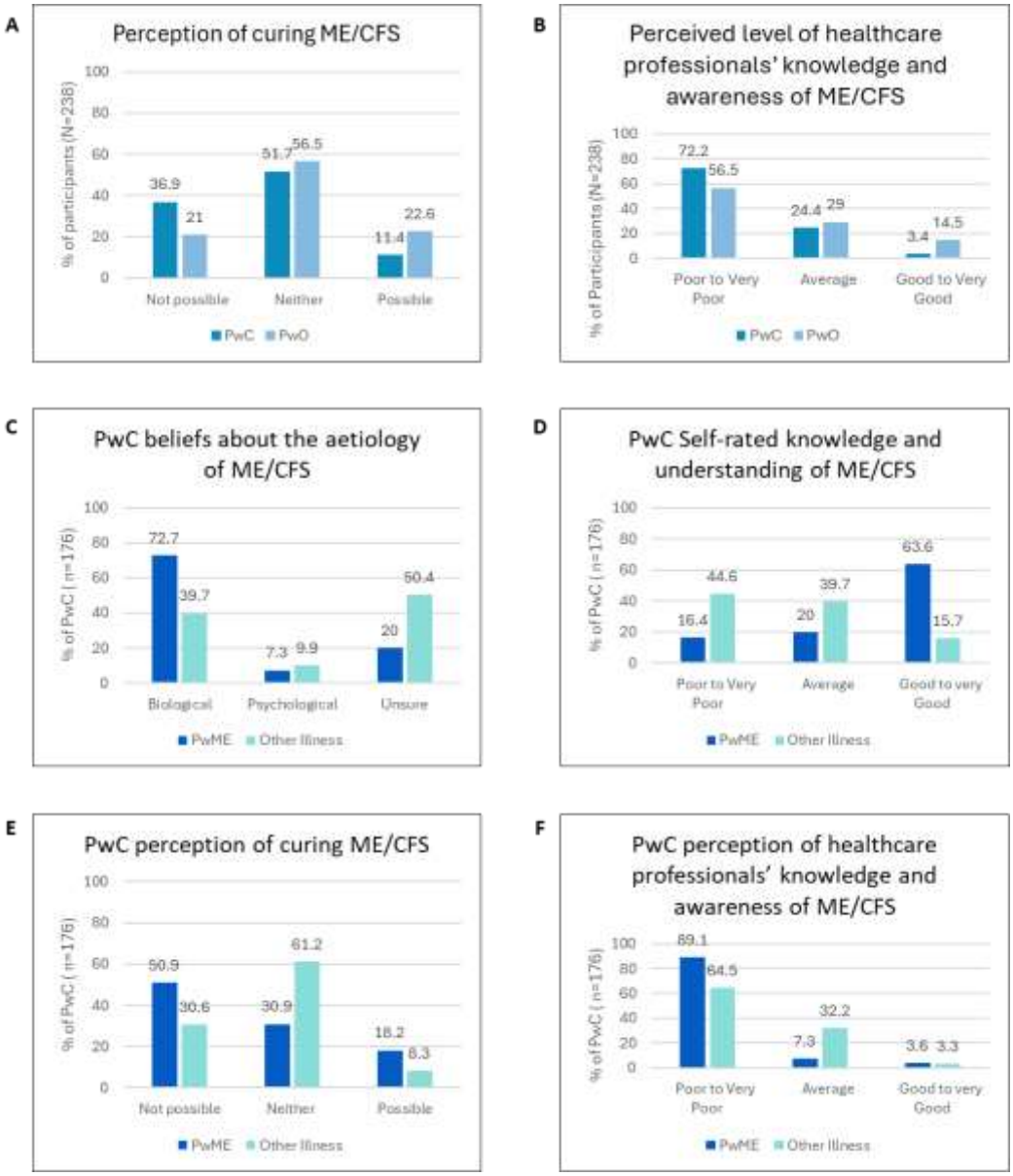


Figure 4.4. Exploratory comparison of responses among participants with chronic illness (PwC) and those without chronic illness (PwO), and between participants with ME (PwME) and with other chronic conditions (Other Illness) across chosen variables. Panel A show participants' beliefs about the possibility of curing ME/CFS, with a significant association between health and belief. Panel B shows ratings of clinician knowledge and awareness, with a significant association between rating and health status. Panels C to F show significant between chronic illness type (ME or Other) and beliefs about the aetiology of ME/CFS, aetiology, and ratings of own and healthcare professionals' knowledge, understanding and awareness of ME/CFS.

4.3.3.2 Knowledge and Understanding of FM

Next, exploratory analyses were conducted to compare the beliefs and ratings of knowledge and understanding of FM between participants with chronic illness (PwC, *n*=195) and without chronic illness (PwO, *n*=85), detailed in Appendix B7. A Bonferroni Adjustment (α=.01) was applied for multiple comparisons.

Generally, PwC were more negative with regards to the possibility of effectively managing and curing FM than PwO. As shown in Figure 4.5 (Panel A), most PwC disagreed that it was possible to cure FM (49%), while the majority of PwO expressed uncertainty (61%). A significant association between health status and beliefs about curing FM was found: $\chi^2(2)=22.18$, $p < .001$, Cramer's V=.281. Similarly, participants without chronic illness were more likely to express optimism or uncertainty about managing the condition, with only 15% believing effective management is not a possibility (Figure 4.5, Panel B). By contrast, 38% of those with chronic illness disagreed. The association between health status and beliefs about management was significant: $\chi^2(2)=14.82$, $p< .001$, Cramer's V=.230.

Compared to PwC, fewer PwO rated their overall knowledge and understanding as Good or Very Good or Average, and more rated it as Poor or Very Poor (Figure 4.5, Panel C). The association between health status and understanding was significant, with a large effect size was found: $\chi^2(2)=67.25$, $p<.001$, Cramer's $V= .490$. Ratings of public knowledge and awareness were not affected by health status, and both groups of participants rated public knowledge and awareness as poor or very Poor (90% PwC, 89% PwO). A significant association was found between health status and rating of healthcare practitioner knowledge and awareness: $\chi^2(2)=26.39$, $p < .001$, Cramer's $V= .307$. As shown in Figure 4.5 (Panel D) compared to PwO, PwC appraised this more negatively and only 3% considered it Good or Very Good.

Given the negative perceptions of healthcare professional knowledge and awareness of FM, further exploratory analyses were conducted on a selection of demographic variables, detailed in Appendix B8. Compared to participants without a diagnosis (*n*=21), a larger proportion of participants with a diagnosis (*n*=174) rated HCP knowledge and awareness as Poor or Very poor (66%) than those without (7.2%). Chi squared tests of independence revealed a significant association between ratings of clinician knowledge and diagnosis, with a small effect size: $\chi^2(2)= 9.95^b$, p=.007*, Cramer's $V= . 226$, though

the analysis is limited by difference in sample size. No significant association was found for having experience with chronic illness other than one's own (i.e. having a relative with chronic illness, or knowing someone with chronic illness), or having experience of Fibromyalgia or Chronic Pain as opposed to other chronic illnesses, and presence of multimorbidity as opposed to its absence.

Figure 4.5

The clinical management and perceived knowledge and awareness of FM among participants with and without chronic illness

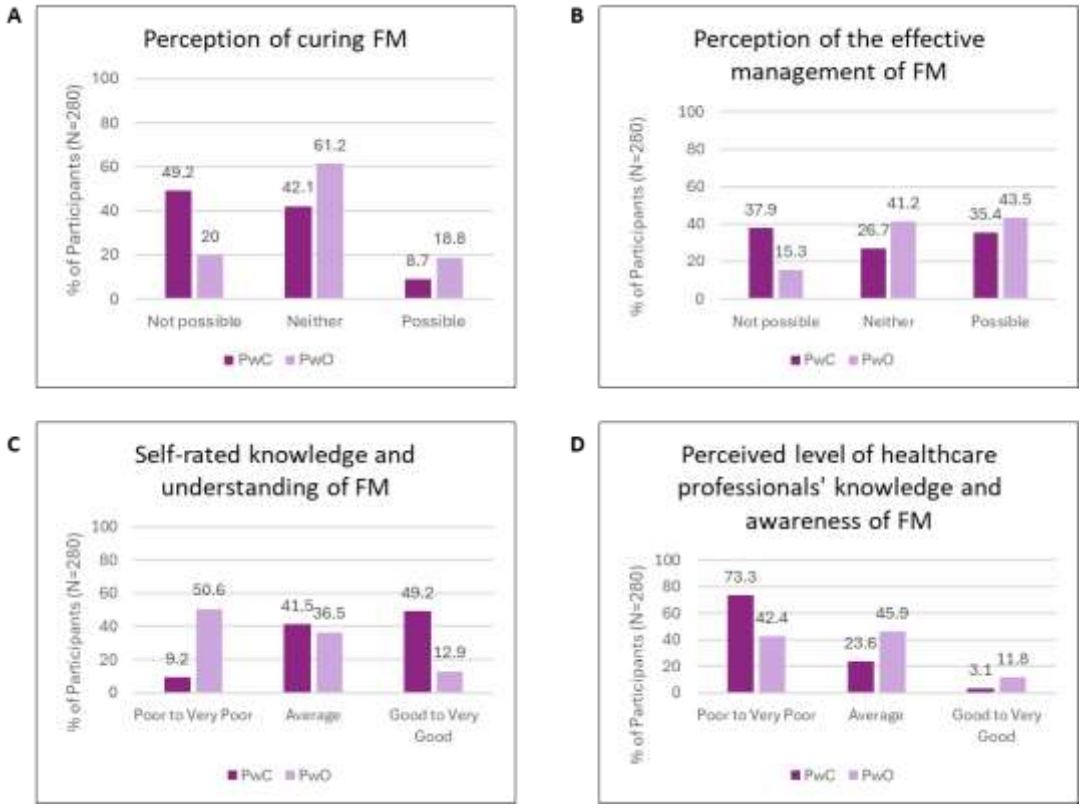


Figure 4.5. Exploratory comparison of responses among participants with chronic illness (PwC) and those without chronic illness (PwO) across chosen variables. Panels A and B show participants' beliefs about curing and managing FM, with a significant association between health and belief in possibility of a cure (A) and effective management (B). Panel C shows self-rated level of knowledge and understanding of FM among participants with and without chronic illness, with a significant association between health status and rating. Panel D shows participant health status and perception of healthcare professionals' knowledge and awareness of FM, with a significant association between the variables.

4.4 Thematic Analysis

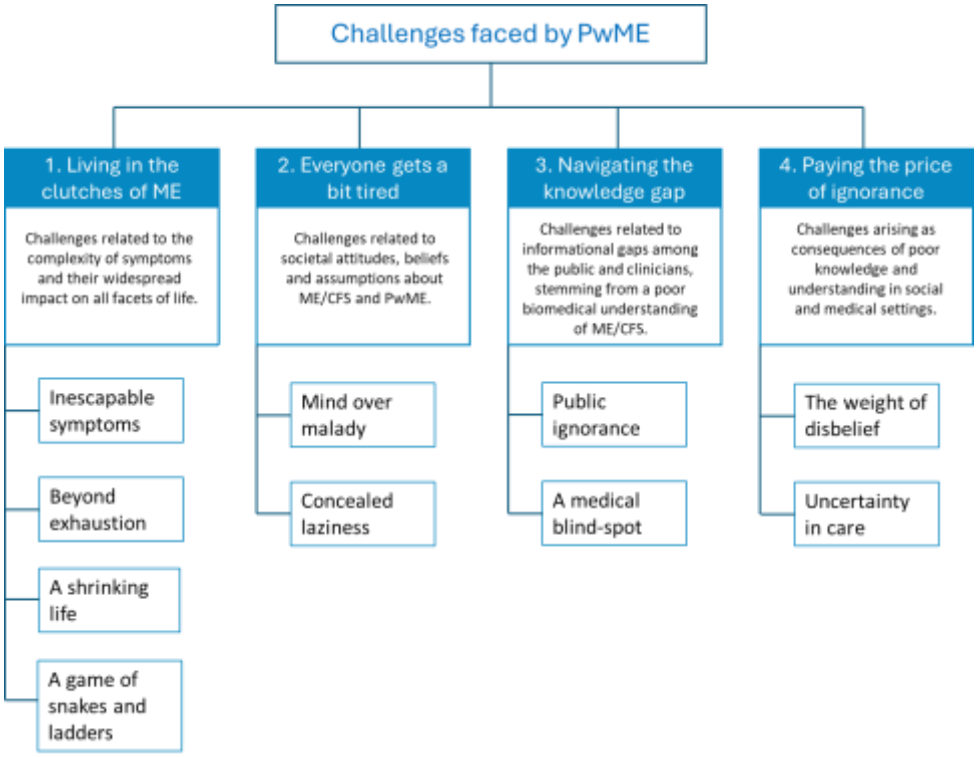
Each participant subgroup was asked to answer a question regarding the challenges faced by individuals living with either ME/CFS or FM. Inductive thematic analysis was conducted on the responses of each participant group, and findings are presented in two separate sections below.

4.4.1 Challenges faced by People with ME/CFS

Of the 238 participants familiar with the condition ME/CFS, 219 provided examples of the challenges they perceived are faced by PwME. These responses were categorised into ten themes and grouped under four overarching types of challenges, presented in Figure 4.6 and discussed below. The four challenges identified were (1) *Living in the clutches of ME*, which captures the profound impact of ME/CFS on PwME, (2) *Everyone gets a bit tired,* which captures societal attitudes, assumptions and beliefs about ME/CFS and PwME, (3) *Navigating the knowledge gap*, which reflects a lack of knowledge about ME/CFS, and (4) *Paying the price of ignorance*, which reflects consequences of poor knowledge and understanding on PwME, both clinically and in social contexts. A selection of illustrative responses is provided in Table 4.3.

Figure 4.6

Perceived challenges faced by PwME and associated themes.



4.4.1.1 Challenge 1. Living in the clutches of ME/CFS

The first set of challenges faced by PwME related to the direct impact of ME/CFS on their wellbeing and daily lives. These challenges include living with inescapable symptoms, primarily persistent exhaustion and post-exertional malaise, and the impacts of ME/CFS beyond exhaustion, such as the effects on activities of daily living, family life and socio-occupational functioning as well as secondary effects on mental health and wellbeing.

Theme 1.1 Inescapable symptoms

Many participants, both those with and without chronic illness, identified severe, debilitating and inescapable symptoms as the primary challenge faced by PwME. They identified a myriad of complex symptoms, including persistent fatigue, post-exertional malaise, pain, physical discomfort, and cognitive difficulties often described as "brain fog". Some emphasised that fatigue in ME/CFS is not akin to tiredness, and is better characterised as an all-encompassing and life-ruining exhaustion (e.g., "*The feeling of being tired all the time, and I don't mean normal tired, I mean so tired that it makes you*

cry but even then you don't have the energy to cry because you literally exhausted to the point where you just collapse")

Theme 1.2 Beyond exhaustion

Many participants also identified challenges that extend beyond the immediate symptoms of ME/CFS, noting the widespread effect it has on individual's lives, including daily activities, family lives and socio-occupational functioning. They noted the chronic and unpredictable nature of the condition as a significant burden, suggesting that these features pose a barrier to understanding, diagnosing and managing ME/CFS. Some discussed the inter-related nature of symptoms, particularly exhaustion, pain and low mood. They also acknowledged the difficulty posed by the invisible nature of the ME/CFS, recognizing how this impairs appreciation of the impact of the condition (e.g., "*Also that it is an invisible disability in many cases - the whole "but you don't look sick!" spiel").*

Theme 1.3 A shrinking life

Participants often recognised that PwME struggle to "live a normal life" and sustain social connections due to the nature of symptoms, highlighting isolation as a major challenge experienced by PwME. Some participants, particularly those with chronic illness, described the psychological toll of living a shrinking life and likened this experience of being in a state of "*living death*".

Theme 1.4 A game of "Snakes and Ladders"

Some participants recognised that managing and coping with the condition is a significant hurdle faced by PwME, noting that learning to manage symptoms requires substantial trial and error. Several participants identified pacing as the key strategy for energy conservation and prevention of post-exertional malaise, but noted that this is an unpredictable science. As described by one participant, "*ME is a game of Snakes and Ladders, and there's always a crash awaiting when you least expect it*". Several participants also referred to loss associated with the condition, and how learning to manage ME/CFS also necessitated learning to cope with a new life. This sentiment was often shared by participants with chronic illness, who discussed feeling that ME/CFS rewrites life narratives (e.g., *"it robs you of things some people can take for granted*").

4.4.1.2 Challenge 2. Everyone gets a bit tired

The second group of challenges identified pertains to societal attitudes, beliefs and assumptions about ME/CFS and PwME, reflecting a general lack of understanding of the severity and impact of the condition. This includes the perception that ME/CFS is psychogenic in nature, and that PwME are feigning their illness.

Theme 2.1 Mind over malady

Participants frequently identified poor understanding and faulty perceptions about ME/CFS as a major challenge faced by PwME. Most commonly, they highlighted that PwME may appear healthy, leading others to conclude that they are exaggerating or malingering, and that PwME should "get over" a condition that is "all in the mind". As put by one participant, *"They are not believed. If recognised at all then their illness is often categorised as being "in their heads".*

Theme 2.2 Concealed Laziness

Much like the assumption that ME is 'in the head', many participants noted that PwME are often branded as lazy or perceived to be lacking willpower. Many participants noted that having to fight such stereotypes in both clinical and personal encounters is a significant challenge faced by PwME (e.g., "*Getting doctors to accept that your symptoms are real and that you aren't "lazy" or "not trying hard enough"").* Such perceptions can lead PwME to doubt the validity of their illness, whereby derogatory attitudes are internalised (e.g., "*Sometimes you will think this yourself which is worse")*. Some highlighted others' naïve understanding of ME/CFS as a condition where one is "a bit tired" and can therefore be resolved with sleep and determination. One participant attributed this to the deceptive terminology of Chronic Fatigue Syndrome: "*The name CFS, just concentrates on fatigue which people universally think is cured by a rest."*

4.4.1.3 Challenge 3. Navigating the knowledge gap

The third category of challenges identified by participants relates to the informational gaps about ME/CFS among the public and clinicians, including a lack of knowledge about the aetiology and suitable treatments for the condition, stemming from a lack of biomedical research.

Theme 3.1 Public Ignorance

In addition to poor understanding, many participants identified a lack awareness of ME/CFS among the public as a challenge faced by PwME (e.g., "*Lack of real understanding of CFS/ME among the general population, understandable perhaps*"). Some participants noted that the knowledge held by the public is inaccurate, stemming from the widespread belief that ME/CFS is a psychogenic condition. As noted by one participant, "*[The] Public are ignorant about the condition. Lack of respect for people with ME*".

Theme 3.2 A medical blind-spot

Many participants also emphasised the challenges faced by PwME in the context of healthcare. They identified ME/CFS as a medical blind-spot, noting a lack of specialist clinics and ME consultants as a challenge faced by PwME. Further, a lack of accurate medical knowledge about ME/CFS was identified as a key issue by participants, who generally perceived clinicians to be inexperienced, disinterested and lacking awareness of the complexity and interconnectedness of symptoms. The failure of medical schools to educate clinicians on ME/CFS was often acknowledged (e.g., "*Medical Professionals total ignorance of the condition. Their laziness at willing to study and learn about this illness.*").

Several participants also described a cycle of misinformation which has resulted in inadequate research into the pathophysiological mechanisms and treatments of ME, contributing to a gap in knowledge among health professionals and the public. As explained by one participant, "*Following years of (sometimes aggressive) misinformation, for example the claim that ME is a psychiatric disorder, there hasn't been enough research into causes and treatments and funding for research hasn't been adequate*".

A number of participants attributed this lack of research to gendered assumptions, for example "*My sense is women predominantly suffer from these conditions and as a result there is little effort put into research on or learning of the conditions"*. Several participants also critiqued the involvement of mental health professionals in the space of ME/CFS (e.g., *"lack of research money for proper biomedical research of ME/CFS and allowing Psychologists to be involved with a biomedical illness.")*

4.4.1.4 Challenge 4. Paying the price of ignorance

The final category of challenges faced by PwME relates to the consequences of poor knowledge and understanding in both social and medical settings. Such consequences include a lack of suitable healthcare services, medical mismanagement, as well as the personal impact of being disbelieved.

Theme 4.1 The weight of disbelief

Participants most often stated that the major challenge faced by PwME is "not being believed" as well as a general lack of empathy from both clinicians and the public. Some participants noted that PwME are often alienated and discriminated against by others, and left to "fend for themselves", contributing to loneliness, isolation and depressive states. This was attributed largely to a poor awareness and understanding of ME/CFS (e.g., *"because it can't be seen by the eye, it is ignored by doctors and society").* Participants also recognised PwME are "fobbed off" in healthcare settings, and face "*Medical scepticism and gaslighting which in turn negatively impacts societal empathy and support."*

Theme 4.2 Uncertainty in care

Many participants highlighted finding the "right" doctor and receiving support is a significant challenge faced by PwME (e.g., "*finding a doctor who doesn't believe that ME is psychological, finding a doctor that understands that exercise is not a treatment and can be dangerous*"). Overall, the process of seeking and receiving care was perceived to be filled with uncertainty, particularly with regards to diagnosis, treatment and management. Participants frequently identified "getting a diagnosis" as a key challenge and recognised difficulties like navigating the welfare system and accessing adaptive technologies (e.g., "*Lack of access to adaptive care such as wheelchair or scooter*"). Several participants recognised that achieving a diagnosis is complicated by assumptions

about the aetiology of ME (e.g., *There is a widespread belief among medical professionals, that ME isn't a physical condition which prevents patients from receiving a diagnosis.")* and commented on a lack of specialist clinics and consultants for ME in Ireland (e.g., *"Dr's unaware and unable to understand diagnosis, with nowhere to refer us to get good sensible management advice and follow-up treatment").*

Some participants also raised concerns about iatrogenic harm emerging from clinician's limited knowledge of post-exertional malaise. As highlighted by one participant, a challenge for PwME is the "*Lack of understanding that patients can get worse potentially permanently if they are pushed too hard (or push themselves too hard)".* The use of outdated clinical guidelines, based on faulty research, was discussed by several participants. For example, referring to the PACE trials, one participant commented that "*the study that recommended this approach was severely compromised and this approach is actively disliked by patients with CFS/ME as it has caused more harm than good*".

Table 4.3

Perceived challenges faced by PwME

Theme	Subtheme	Example Quotes
Challenge 1. Living in the Clutches of ME/CFS		
1.1 Inescapable symptoms	Life ruining exhaustion	"Tiredness, fatigue, emotional stress and anxiety, pain, uncertainty, struggling to experience life"
		"Life-ruining severity of symptoms, inescapable exhaustion and brain fog causing extremely low capability of almost any exertion (including conversation and completing basic low-effort tasks)."
1.2 Beyond exhaustion	An intractable illness	"I sincerely hope one day people will recognise the word chronic isn't used lightly in illnesses."
	It ebbs and flows	"One day headache, next minute pain and brain fog. Inconsistent."
	You don't look sick	"It's also invisible and people don't understand it"
	A cycle of symptoms	"Cyclical nature of challenge- exhaustion creates mental health problems creates exhaustion.. And so on"
1.3 A shrinking life	Trying to live a normal life	"Keeping up a daily regiment, performing general tasks, maintaining social circles"
		"Disability- unable to work or socialise, difficulty daily basics of living. Living death."
	Sustaining connections	"Missing out on social events and time with family. Keeping on top of housework etc"
	Psychological toll	"You become depressed by circumstances, by the manner in which this is shrinking your life. You may no longer be able to work but you have a mortgage so you push through the working day and crash every evening. Your social life & therefore, your support network, shrinks. Your GP will tell you that you are ticking every box for depression and would you

Theme	Subtheme	Example Quotes
		consider antidepressants. You do not need anti-depressants, you are not depressed. You are sick."
1.4 A game of snakes and ladders	Finding the right pace	"Managing the condition- the only treatment I have heard for CFS/ME is the individual pacing themselves, listening to their body and resting as required"
	Losing control	"There will be periods of time, sometimes weeks and even months when you are fine and you begin to believe you've turned a corner but ME is a game of Snakes and Ladders and there's always a crash awaiting when you least expect it. It's hard not to be angry but you do learn to live with it."
		"The fact that it's often triggered by many different conditions, or happenings, and there's a lot of waiting to see if it clears up or changes over time."
		"Do something that seems innocuous and end up floored with PEM. There's an invisible tolerance line that your body can handle for cognitive, physical, emotional or social activity. That line changes and you've absolutely no idea where it is until you fail.."
	Rewritten narratives	"The biggest challenge is accepting that your old life is over. You have to grieve it, accept the new reality and move forward. This takes a long time and a professional counsellor is a great advantage."
Challenge 2. Everyone gets a bit tired		
2.1 Mind over malady	All in the head	"Socially people assume they are exaggerating or it's all in their heads."
		"GPs not clued in and taking symptoms to be 'in the mind', mistaking fatigue for depression ..."
2.2 Concealed laziness	A lack of will	"There will always be a sense that some people think you are just being lazy. Sometimes you will think this yourself which is worse."
		"Perception it is easily treatable with enough willpower"

Theme	Subtheme	Example Quotes
Challenge 3. Navigating the knowledge gap		
3.1 Public Ignorance	Public knowledge and Understanding	“People don’t understand that this kind of fatigue is debilitating. People don’t understand that it ebbs and flows so you can be ok one day or for one part of a day, but that the energy you expend to attend an event/activity can mean you crash for days/weeks afterwards”
3.2 A medical blind spot	Doctors lack knowledge and experience	“Lack of understanding that patients can get worse potentially permanently if they are pushed too hard (or push themselves too hard)/can’t rest enough and similar”
	Failed by medical school	“Medical professionals lack knowledge and training, time and empathy”
	Misinformation	“No medical research into causes and treatments, doctors not being taught about it, doctors still labelling symptoms as psychological”
		“Poor knowledge of ME/CFS amongst Medical practitioners”
Challenge 4. Paying the price of ignorance		
4.1 The weight of disbelief	Fobbed off by others	“Patients presenting with symptoms aren't believed or taken seriously, especially if they're female.”
		“Disbelieved, dismissed and medical professionals and members of the public lacking empathy “
		“They get fobbed off by the "experts" who give up easily on their struggles because these experts don't understand it either so it's easier to just forget about them.”
	Alienation	“rejection and gaslighting both socially and from medical workers not informed about the realities of CFS/ME”.
		“Lack of understanding & support throughout the medical community. Therefore lack of understanding & empathy among lay people. Leading to isolation & loneliness & depressive states.”

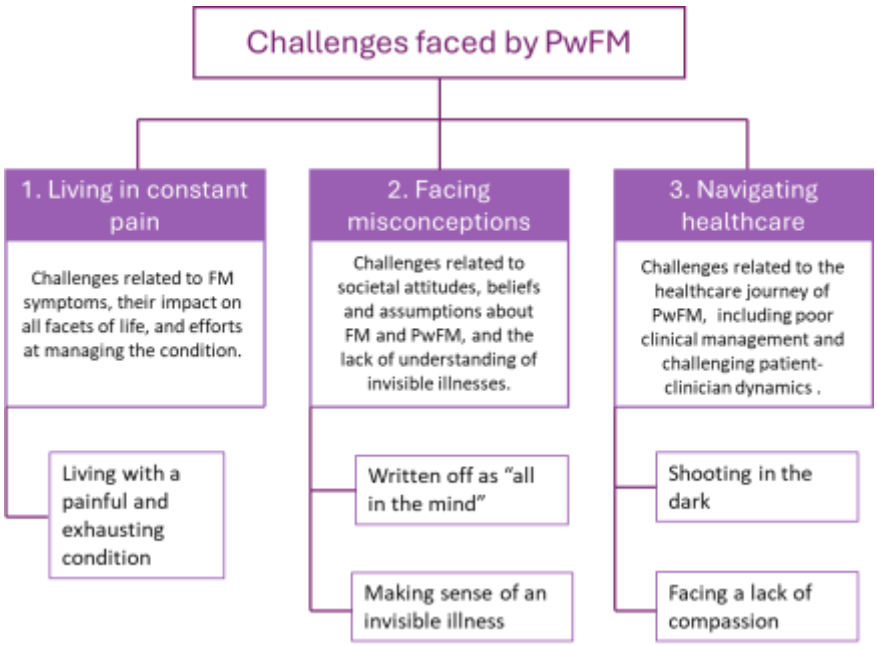
Theme	Subtheme	Example Quotes
4.2 Uncertainty in care	Getting the right doctor	"Finding a doctor who doesn't believe that ME is psychological, finding a doctor that understands that exercise is not a treatment and can be dangerous"
		"It takes time to get the right medial person to continue to work through the diagnosis."
	Medical mismanagement	"Medical mismanagement (forced to exercise, limited access to therapy, limited access to a few knowledgeable staff)"
		"It is too easy to assume they are depressed or malingering. There are no tests that can confirm the diagnosis, so doctors follow a checklist, once convinced the person is really suffering.")
		"One visit and you are sent back to your GP with guidelines to follow the treatment that doesn't really exist"
	Faulty guidelines	"The UK published (now discredited, cognitive behavioural therapy and graded exercise therapy) guidelines on how to treat ME which were totally detrimental to the health of many ME sufferers who attempted to follow them. Although discredited this study still seems to form the basis of treatment for ME."
	Understanding own symptoms	"Struggling to understand why they feel the way they do because of a lack of knowledge from others including health professionals"

4.4.2 Challenges faced by People with FM

All 280 participants familiar with FM provided responses to the open-ended question concerning challenges faced by PwFM, but thematic analysis was conducted on the responses of 270 participants. While all participants responded to the open-ended question, responses of 10 were excluded for the following reasons: 1 participant did not directly identify perceived challenges faced by PwFM ("Yes, definitely") and 9 expressed a lack of knowledge about the topic (e.g. "no idea"). As shown in Figure 4.7, five themes and three broad categories of challenges faced by PwFM were identified from participants' responses. The three overarching challenges were: (1) *Living in Constant Pain*, which captures challenges directly related to Fibromyalgia Syndrome and the theme *Living with a painful and exhausting condition* which reflects the characteristics of FM, its impact, and efforts at coping with and managing the condition; (2) *Facing misconceptions*, which captures challenges related to other's perspectives on FM and includes two themes: *Written off as "all in the mind"*, referring to the underlying beliefs, preconceptions and attitudes held by individuals regarding FM, and *Making sense of an invisible illness*, which reflects suboptimal knowledge and understanding of FM and its consequences; and (3) *Navigating healthcare*, which captures the challenges arising in the context of healthcare and includes two themes: *Shooting in the dark*; a theme concerning practical aspects such as service availability, quality of care, and the clinical management of FM and *Facing a lack of compassion*, a theme concerning interpersonal relationships in the context of healthcare, including consultation dynamics and the patient-clinician interaction. A selection of illustrative quotes are presented in Table 4.4

Figure 4.7

Perceived challenges faced by PwFM and associated themes.



4.4.2.1 Challenge 1. Living in constant pain

The first set of challenges faced by PwFM related to their experience of living with chronic and fluctuating widespread pain, and the impact of the condition across all domains of living.

Theme 1.1 Living with a painful and exhausting condition

Difficulties arising from the nature of FM were perceived as a key challenge faced by PwFM. Most participants recognised the key symptoms and qualities of the condition, the burden of illness and individuals' ongoing efforts to cope with and manage symptoms. Chronic widespread pain, fatigue, cognitive difficulties and other non-specific symptoms were frequently identified, and participants often referred to FM as a disabling and invisible illness (e.g., "*You look okay, but can be suffering. It's an invisible illness*"). Participants also noted that the persistent, unpredictable and subjective nature of symptoms was a challenge for PwFM. As noted by one participant, PwFM can

experience "*Difficulties in explaining the symptoms that can be mostly referred to as subjective experiences and feelings, rather than those physical identifiable.".*

Several participants also emphasised the challenge of living with dynamic disability, for example, *"The varying pain levels and thresholds (i.e., how it was comfortable to sleep yesterday, may not be comfortable today), mobility and access to disability services (because sometimes on a good day you can walk to the store, that must mean you don't NEED a wheelchair)"*

Many respondents also noted that the impact of FM extends beyond the immediate experience of symptoms. The transformative impact of FM was widely recognised by participants, who described the condition as life-altering and profoundly disruptive across all domains of functioning and individuals' sense of self. (e.g., "*The loss of their " normal " life. Having to give up work, hobbies, socializing, traveling, activities with family and friends. Feeling of letting people down when you have to cancel plans last minute.").* The inconsistency arising from fluctuating symptoms was associated with difficulties in holding down a job and maintaining social connections, and many participants acknowledged the psychological toll of living with a painful and exhausting condition. PwFM were perceived by some participants as being of the mercy of their illness (e.g., *"fibromyalgia dictates to the patient how their day will be*").

Accepting, coping with and managing the symptoms of FM were perceived as a significant challenge by many, who highlighted that *"learning to live with the condition is an ongoing battle*". Several participants noted that learning to manage FM is a substantial burden for PwFM, and highlighted that the variability and complexity of symptoms and triggers make the condition particularly challenging to manage. As put by one participant "*It is a painful and exhausting condition, and people with the condition just don't have the energy to pursue diagnosis, treatments, management strategies, or advocate for themselves".* A few participants also noted the challenge of maintaining positive outlooks while living under the pressure of relentless symptoms (e.g., " [The challenge of] *managing it in your mind more than the physical symptoms, always trying to be positive in order to outweigh the negatives*")

4.4.2.2 Challenge 2. Facing misconceptions

Participants often identified public and healthcare practitioners' beliefs and misconceptions about FM as a challenge to individuals living with the condition. They highlighted that debates about the legitimacy and validity of FM result in PwFM experiencing dismissal and disbelief in personal, occupational and healthcare settings.

Theme 2.1 Written off as "all in the mind"

The most frequent challenge discussed by participants is the disbelief experienced by PwFM. Many participants discussed issues of credibility, and noted that PwFM are not taken seriously, particularly in the absence of overt signs of suffering. As put by one participant, *"no one believes you are in so much pain all the time".* Several participants also commented on broader gender-related issues (e.g., "*GPs don't believe women as much as men*"), highlighting the complexity of the challenge faced by PwFM.

Many participants perceived public and clinician assumptions about the aetiology of FM as an obstacle for PwFM. They frequently stated that FM is assumed to be psychological, and that others therefore question its legitimacy as a "real" clinical entity. References to FM as "in the mind" were common, in tandem with gendered assumptions about the illness. As stated by one participant *"the myth that it is 'in the mind' is still around - also among doctors in Ireland".* Some participants also suggested that such attitudes arise due to the invisible nature of symptoms (e.g., "*It is easy for medical professionals and the public to assume it is a psychological issue, since they can't actually see anything (i.e. the sufferer looks well.)"*).

A related challenge raised by participants was the stigmatisation and discrimination faced by PwFM, who were seen as being judged unfairly by others. Participants often felt that PwFM are often judged unfairly by others (e.g. *"Lack of understanding, discrimination, bullying, accusations of being lazy or looking for an excuse to justify perceived laziness, labelled as hypochondriacs who are lazy and wound up").* A few participants linked societal attitudes and perspectives on FM to the dismissive attitudes of clinicians. As stated by one participant, a challenge is *"Medical scepticism & gaslighting which in turn negatively impacts societal empathy & support.".*

Theme 2.2 Making sense of an invisible illness

An added challenge highlighted by participants concerned barriers to sense-making, particularly the lack of knowledge, biomedical research and consequently understanding of FM. Many participants expressed the view that people do not understand FM and highlighted a lack of general awareness about FM among clinicians and the public (e.g., *"Medical professionals don't have an appropriate level of understanding of fibromyalgia. The general public has hardly any understanding of it.")*. This was often attributed to the fact that FM is an invisible illness.

Participants often perceived significant informational gaps about FM among healthcare providers, highlighting this as a significant challenge for PwFM in healthcare settings. Several critiqued the quality of medical education and training and referred to limitations of the biomedical model. For example, one participant identified a challenge as *"GPs not being taught about it in school, GPs taught to not believe their patients and that unexplained symptoms must be psychological"*. Given the biomedical focus on evidence, a lack of objective diagnostic markers was seen as reflective of poor knowledge and understanding of the condition more generally, and as stated by one participant, the condition "*Requires more research*".

Several participants highlighted the repercussions of ignorance on the wellbeing of PwFM. Informational deficits were seen as resulting in a neglect of patient needs and underappreciation of the widespread impact of FM on patients. Many participants noted that a lack of knowledge and awareness directly contributes to a lack of appropriate and timely supports for PwFM, as well as missed opportunities to intervene. For example, one participant highlighted that *"The lack of effective treatments means that individuals with fibromyalgia have to 'wait out' the particularly painful periods and just wait for their symptoms to subside.*". Several other participants noted that PwFM often need to seek alternative supports and treatments, resulting in financial strain (e.g., "*My mother has had to spend thousands trying to get answers here and abroad with little support from Drs, consultants and "experts" in the field.*")

4.4.2.3 Challenge 3. Navigating Healthcare

The third broad type of challenges identified by participants concerns the healthcare journeys of PwFM, particularly the issues faced when navigating healthcare.

Theme 3.1 Shooting in the dark

Many participants identified the practicalities of navigating healthcare as a challenge faced by PwFM, including issues with quality of care, the diagnostic journey and long-term management of FM (e.g., *"Proper health care, prescription medications for other conditions prescribed incorrectly, no medical follow up, not enough real information on how nutrition and exercise can help"*). Many commented on a "trial and error" approach to diagnosis and treatment, recognizing that a diagnosis of FM is often made when other explanations for symptoms fail. They often noted that FM is a "catch all" diagnosis, and there was a sense that clinicians are shooting in the dark. Most participants perceived the diagnostic journey to be lengthy and involving a process of elimination. For example, one participant stated that a challenge for PwFM is *"Diagnosis by exclusion (i.e., having to do every, absolutely exhaustive test before fibromyalgia even becomes an option)."* Only one participant suggested that the diagnostic process is curtailed, contributing to the over-diagnosis of FM: *"Personally I feel fibromyalgia is often over-diagnosed as its supposed to be an elimination process to discount many other potential diagnosis first, but often very few are ruled out before fibromyalgia is thrown at a person."*

Similarly, many participants described the treatment of FM as a process of trial and error. They often commented on a lack of curative or standardized treatment, noting that little is done beyond symptom management (e.g., *"No set treatment so it's hard to find something that works").* Several participants perceived this to be a significant added burden for PwFM. As stated by one participant, PwFM face *"A lack of clarity in their experience with healthcare and constant trial and error in their treatment on top of their pain and tiredness"*). Many participants noted that the healthcare journeys of PwFM are filled with uncertainty, and unmet needs for support in healthcare and beyond were widely identified (e.g., *"Hard because there are not many supports and resources as far as I know."*).

Theme 3.2 Facing a lack of compassion

In addition to the practical aspects of healthcare, participants identified inter-personal relationships as a challenge in faced by PwFM in their journey. Participants often alluded for a need for person-centered care and made statements about PwFM struggling to be seen, heard and treated with respect and empathy (e.g., "*the lack of empathy from the very people who should be able to help you.").* They echoed the sentiment that "Doctors don't listen" and noted that clinicians appear disinterested in FM more generally (e.g., *"When other conditions are ruled out people and medical professionals seem to care less."*). Referring to the patient-clinician alliance, one participant stated that PwFM are faced with "*professional hostility*."

Table 4.4

Illustrative quotes for challenges faced by PwFM.

Theme	Subtheme	Example Quote
Challenge 1. Living in constant pain		
1.1 Living with a painful and exhausting condition	Constantly changing disparate symptoms	"pain every day; fatigue and brain fog; judged unfairly by others; sleep disturbance; unable to control body temperature ie either too cold or too hot; feel very left to manage it myself with no supports"
	Fibromyalgia dictates the day	"Every day life can be hard because you can not plan anything in advance as you never know how you will feel on that day."
		"life slowing down due to pain"
	A constant battle	"Stress management. Pain management, Sleep management. Coming to terms with a limited life. Pacing oneself, Pacing oneself, busy day/slow day. No motivation as one realises when one gets busy so does the fibromyalgia kicks in. It is a constant fight to fight off debilitation and pain, exhaustion."
		"It is difficult to manage because many things can trigger or affect it, for example stress and stress is not easy to avoid. It takes a long time to learn how to cope and manage it effectively."

Theme	Subtheme	Example Quote
Challenge 2. Facing misconceptions		
2.1 Written off as “all in the mind”	Nobody believes you	“Being believed, because a smile can seem like we have no pain whatsoever and that because we look okay then we must be fine”
		“Medics don’t believe it exists so it’s luck with finding someone who does”
	The myth of mental illness	“Doctors assume people (especially women) presenting with symptoms are exaggerating or are experiencing mental illness.”
	Judged unfairly	“Also not easy to explain to friends and family and some people dismiss it, you can face the same stigma and discrimination as people with mental health challenges face”
2.2 Making sense of an invisible illness	People don’t understand it	“overall lack of empathy or understanding of the condition within society, many people don't know what fibromyalgia even is or haven't even heard of the term”
	Doctors don’t know about it	“Medics knowledge/Understanding. Patient understanding pain and how it affects the body and mind. Knowledge of both doctors and also patient is key”
	The repercussions of ignorance	“the poor understanding of the public, including family, who cannot mobilize appropriate support in their lack of understanding”

Theme	Subtheme	Example Quote
Challenge 3. Navigating Healthcare		
3.1 Shooting in the dark	When all else fails	“I know someone who has this. It was very difficult to get a diagnosis in the first place even though they were experiencing significant pain. Doctors often misdiagnose these symptoms in women attributing it to things like period pain. It is not well understood and even if you get a diagnosis you probably won't ever know the cause.”
	Treatment by trial and error	“Inconclusive tests, frustration at lack of understanding or help, trial and error treatment”
	Left In the shadows	“Insufficient research and treatments, insufficient concern among medical profession, insufficient supports (medical, workplace, etc.), inability to access/difficulty accessing social welfare supports, mental health impacts of the symptoms (…)”
		“Uncertainty that appropriate care will be provided”
3.2 Facing a lack of compassion	To be seen and heard	“Getting a health professional to listen and not tell you it's all in your head or that you need to lose weight.”
	A call for empathy	“lack of care and concern among medical community and lack of supports for people suffering this horrid, life-altering pain and fatigue.”
		“Medical professionals lack of knowledge and lack of empathy”

4.5 Discussion

The present mixed-method cross-sectional survey sought explore the perspectives of a sample adults familiar with ME/CFS and FM in Ireland, with a focus on their knowledge and understanding of the conditions, as well as the challenges faced by persons living with them. While the conditions are distinct, similar trends in participants' responses were observed for both. Together, the results suggest cognisance that the conditions ME/CFS and FM are distinct from the symptoms of chronic fatigue and chronic pain, a good awareness of the symptom profile of ME/CFS and FM, and a rich understanding of the challenges faced by PwME and PwFM in both social and healthcare settings. The findings also indicate that respondents aware of these conditions were attuned to the misconceptions surrounding both conditions, particularly regarding assumptions about their psychogenic nature, and the consequent discrediting of patients as suffering from a "real" condition. In contrast to ample literature reporting negative attitudes and beliefs towards ME/CFS, FM and PwME and PwFM, reviewed in Chapter 3 (e.g., *Section 3.4.1.5*), both conditions were recognised as legitimate, and individuals living with them were not assumed to be exaggerating or feigning illness.

Furthermore, participants also highlighted challenges related to diagnosis, treatment and management of ME/CFS and FM, generally demonstrating pessimistic outlooks on the clinical management of both conditions. Relatedly, and most strikingly, results indicated a deep distrust of healthcare practitioners among the public, with only a small fraction perceiving their knowledge about and awareness of either condition as good or very good. While participants were aware of the shortcomings of medicine and the healthcare system, particularly with regards to diagnosis and service availability or accessibility, they were often highly critical of medical professionals' attitudes towards patients with PwFM and PwME, highlighting a need for more humility, compassion and empathy in healthcare as well as more emphasis on biomedical research.

4.5.1 Understanding of ME/CFS and FM

As mentioned, the results of this survey suggest that adults in Ireland who are familiar with the clinical entities ME/CFS and FM possess a good level of understanding of the complex symptom profile and the personal, social and fiscal impacts of each condition. This mirrors the accounts of PwME and PwFM which extensively document highly

heterogenous and fluctuating symptoms, their impact on health-related quality of life (Galvez-Sánchez et al., 2019; Vyas et al., 2022) as well as the experience of biographical disruption and loss (e.g., Åsbring, 2001; Brown, 2018, 2021; Eaton-Fitch et al., 2020; Fennell et al., 2021; Larun & Malterud, 2007; Warner, 2017). The good recognition of these issues by respondents is encouraging, but also somewhat surprising given that patients often highlight others' ignorance, as discussed in Chapter 3. Given the sampling strategy employed in this study, the favourable results most likely reflect the views of individuals with a higher level of knowledge or more personal investment in these conditions, rather than those of the general population.

The results also suggest a good awareness of the challenges emanating from the "contested" nature of both conditions, namely the misconception that they are psychosomatic or psychogenic in nature, and the negative imprint it leaves on patients' social and healthcare experiences. Not only was public knowledge and awareness appraised to be very poor, the qualitative findings emphasised how negative societal attitudes and rejection a major challenge faced by individuals with both conditions. Namely, results highlighted the issues of stigma, invalidation and disbelief, and the historically problematic narrative that that ME/CFS and FM are "all in the mind", or that patients are lazy or lacking willpower. This is aligned with existing literature which highlights debates about the aetiology of "medically unexplained" illnesses (e.g., O'Leary, 2018) and the consequent stigmatisation, dismissal and delegitimation of PwME (e.g., Anderson et al., 2012; Åsbring & Närvänen, 2003; Hunt et al., 2024; Larun & Malterud, 2007; Tschopp et al., 2023) and PwFM (Briones-Vozmediano et al., 2018; Juuso et al., 2011; Ko et al., 2022; Oldfield et al., 2018; Sallinen et al., 2019). Additionally, participants noted that the experience of stigma in social and healthcare settings can lead to the internalisation of negative attitudes by PwFM and PwME, resulting in self-doubt and additional psychological distress. Such impacts of repeated stigmatisation on self-concept are well-described, particularly in the context of concealable or invisible illness (Earnshaw et al., 2012; Earnshaw & Quinn, 2012; Quinn & Chaudoir, 2015; Quinn & Earnshaw, 2013).

Interestingly, while the aforementioned negative views and assumptions about ME/CFS and FM were not shared by the respondent sample, and many considered both to be

primarily organic, there was also a large degree of uncertainty about the aetiology of each condition. In the case of FM, just over one in seven participants considered the primary cause to be psychological, which could suggest a broad shift away from psychosocial explanations of pain, or a growing appreciation of the complex physiological changes associated with the condition (e.g., Clauw et al., 2024; Ovrom et al., 2023). By comparison, a smaller proportion of participants indicated that the primary cause of ME/CFS is psychological, and participants with lived experience of chronic illness including ME/CFS were more likely to endorse the biological explanation for the illness. Similar trends have been noted in the literature whereby patients identify an infection-related, typically post-viral, origin of their condition (e.g., Angelsen & Schei, 2024) despite clinicians' assertions that is it psychosomatic in nature (e.g., Hng et al., 2021; Tschopp et al., 2023). Given the growing body of evidence which links a history of infection with increased risk for developing ME/CFS (Lacerda et al., 2019) and demonstrates that ME/CFS is a complex, multisystemic illness (e.g., Schreiner et al., 2020; Sotzny et al., 2018), it appears that patients in this study also have a more accurate understanding of their illness than the general public and clinicians. Generally, the recognition of a biological aetiology among respondents signifies a positive step forward, perhaps as a reflection of a growing understanding of post-viral syndromes in light of the COVID-19 pandemic. With the growing recognition of Long-COVID in both medical and public discourse, it will be interesting to observe whether patient, public and clinician beliefs about the aetiology of ME/CFS become more aligned with time.

4.5.2 Diagnosis, treatment and management

Despite their familiarity with these conditions, respondents indicated having a comparatively low self-rated understanding of the diagnosis, treatment and management of FM and particularly ME/CFS. However, the findings suggested that they had a largely accurate understanding of the challenges associated with the clinical management of both. The diagnostic odyssey was seen as a significant additional burden for PwME and PwFM, and both conditions were considered as more difficult to diagnose than other chronic illness. Findings highlighted that the lack of objective tests and biomedical understanding of both conditions were a barrier to obtaining a diagnosis, and consequently appropriate care. This interpretation is closely aligned with the reality

reflected in patient accounts throughout the literature, as well as the concerns expressed by healthcare providers (see Chapter 3, *Section 3.4.1.2* and *Section 3.4.2.2*).

Briefly, many studies highlight that misdiagnosis and delayed diagnosis of FM are common, with patients reporting delegitimising experiences when standard medical tests do not identify an organic cause for their symptoms (Baron et al., 2014; Boulton, 2019; Goldenberg, 2009). Similarly, in the context of ME/CFS, diagnosis is often a protracted process that involves the exclusion of other illnesses (Conroy et al., 2023) as well as the involvement of numerous clinicians (e.g., Tidmore et al., 2015). The recognition of diagnostic delays was evident in participants' estimates regarding time to diagnosis, which was most often estimated to take up to three or five years for both FM and ME/CFS. While broadly accurate, this is slightly below estimates recently published Irish data from European ME Alliance Survey (Angelsen & Schei, 2024) which indicate that the average time to diagnosis as 5.5 years for women, and 3.9 years for men with ME/CFS. At present, there are no reported data regarding the prevalence of FM or the diagnostic trajectory of PwFM in Ireland, but some international data suggests that it can take between 2 and 6 years (Choy et al., 2010; Gendelman et al., 2018).

Interestingly, while participants felt that both conditions were more difficult to diagnose than other chronic illness, a larger portion of participants felt it was more difficult to diagnose FM than ME. While the survey did not seek explanations for such ratings, it is possible that the high incidence of pain across various chronic conditions complicates the identification of its specific aetiology. As suggested by others, the heterogenous symptom profile of FM, the frequent presentation of chronic pain alongside other conditions, and lack of defined clinical pathways often result in a burdensome diagnostic experience (e.g., Galvez-Sánchez et al., 2019; Häuser et al., 2017; Mengshoel et al., 2018; Wilson et al., 2022).

Additionally, the findings of this study paint a largely pessimistic view of the treatment and management of ME/CFS and FM among the respondent sample, including both individuals with and without personal experience of chronic illness. Many participants expressed uncertainty with regards to the possibility of curing each condition and views were more often negative than positive. Multiple challenges with regards to finding suitable treatments were identified and were perceived to be a major frustration and

added burden for patients. Such views of poor prognosis reflect the current state of clinical management, given the lack of curative treatment for ME/CFS (Deumer et al., 2021; Toogood et al., 2021) and mixed evidence for the effectiveness of pharmacological and non-pharmacological interventions in the case of FM (e.g., Adams et al., 2023; Bidonde et al., 2023; Climent-Sanz et al., 2023; Doebl et al., 2020; Rico-Villademoros et al., 2020). As PwME often report being told that "nothing" can be done to treat their condition (McManimen et al., 2019), it is unsurprising that the most negative views regarding curing ME/CFS were expressed by respondents living with the condition.

There was a perception that treatment often proceeds in a "trial and error" manner, which can be harmful to both groups of patients, though such accounts were most elaborate in the context of ME/CFS. Specifically, respondents highlighted that the treatments suggested by healthcare professionals, namely Cognitive Behavioural Therapy (CBT) and Graded Exercise Therapy (GET), are ineffective or detrimental to PwME. Indeed, as discussed elsewhere (e.g., Geraghty & Blease, 2019; White et al., 2023; Wilshire et al., 2017) these approaches have been widely criticised and demonstrated to cause harm to PwME but continue to be recommended to patients by healthcare providers (e.g., Angelsen & Schei, 2024; McManimen et al., 2019).

Relatedly, the management of both conditions was perceived to be more difficult than of other chronic illnesses, with qualitative findings again highlighting a lengthy process of trial and error. In the context of FM and ME/CFS respectively, one in three and one in four participants felt that the effective management of symptoms was not a possibility, though perception was associated with personal experience of chronic illness. Challenges identified in relation to self-management mirrored those which are discussed in the literature, including those related to lifestyle adjustment, listening to one's body, and learning to pace to prevent symptom exacerbation (e.g., Angelsen & Schei, 2024; Ashe et al., 2017; Jason et al., 2013; Kengen Traska et al., 2012).

4.5.3 Clinician knowledge and awareness

To reiterate, the most striking results of this study were the vastly negative views regarding clinicians' knowledge and awareness of FM and ME/CFS. Fewer than one in ten participants felt that healthcare professionals had a good or very good level of knowledge and awareness of either condition; interestingly, those with lived experience

of chronic illness were more negative. Although the extent of negative appraisals is surprising, the trend is broadly in line with that reported in other literature. For instance, 89% of PwME in the current survey rated healthcare professionals' knowledge and awareness as poor or very poor; by comparison, a large Australian study found that just under 50% of PwME considered their GPs to be poorly or very poorly informed of the condition (Emerge Australia, 2020).The results of a UK survey with 549 PwFM also highlight similar issues, with healthcare professional knowledge and skills identified as an unmet need for 40% of patients (Wilson et al., 2022).

Literature demonstrates that the self-assessed knowledge and awareness of FM among clinicians is poor (Emorinken et al., 2022), further supporting patients' accounts in the literature, as well as the critiques captured in this research. For instance, participants emphasised a lack of basic knowledge among clinicians about the symptom profile, pathophysiology and impact and of FM and ME/CFS, suggesting that healthcare professionals lack adequate medical training about these conditions. Several studies support this, demonstrating that little or no space is afforded to ME/CFS on medical school curricula (Muirhead et al., 2021; Peterson et al., 2013). Similarly, gaps in training have been identified in the context of FM (Arshad & Ooi, 2007; Emorinken et al., 2022). Such omissions are not only problematic in terms of skills, but as reported by Silverwood et al. (2017) can lead medical students to believe that FM is less important than other conditions.

The overwhelmingly negative perceptions of healthcare practitioners' knowledge and awareness were mirrored in participants' open-ended responses, which indicated an in-depth understanding of the barriers faced by PwFM and PwME in healthcare. Studies often report a large degree of dissatisfaction with healthcare among PwME and PwFM, suggesting that these patient groups are medically underserved (e.g., Byrne et al., 2023; Doebl et al., 2020; Froehlich et al., 2022; Sunnquist et al., 2017; Thanawala & Taylor, 2007; Wilson et al., 2022). Many respondents also noted that PwFM and PwME in Ireland lack access social support or specialist care and are faced with a high level of uncertainty in their healthcare journey, which involves substantial trial and error rather than clearly defined pathways. The critiques of limited support for PwME in the Irish context are in line with the results reported by the European ME Alliance Survey (Angelsen & Schei,

2024) whereby over 80% respondents from Ireland reported receiving little or no support from healthcare services for their condition.

Interestingly, participants with a formally diagnosed chronic illness appraised clinician knowledge and awareness of FM less positively than those without; yet neither the presence of multimorbidity nor experience of chronic illness in friends or relative were associated with their perception. Such results may suggest that patients with chronic illness lose trust or confidence in healthcare professionals' competence as a result of their diagnostic experience, though this warrants further investigation. As a large portion of participants reported living with FM or chronic pain, it is noteworthy that achieving a diagnosis of FM does not always result in relief or satisfaction. Some studies illustrate that a diagnosis of FM might be disappointing or undesirable, being essentially useless in the face of limited options for treatment (e.g., Boulton, 2019; Mengshoel et al., 2018; Undeland & Malterud, 2007), thus possibly explaining the negative appraisals.

Finally, participants often discussed challenging patient-clinician dynamics experienced by PwFM and PwME, with clinicians failing to recognise or invalidating the impact of FM and ME/CFS on their patients' lives, and suggested a need for empathy and compassionate approaches to care. Similar themes are largely reflected throughout the literature examining perspectives of both patients and healthcare practitioners. As discussed previously and in Chapter 3, issues of stigma, scepticism and dismissal were widely acknowledged in the context of healthcare, and these are well-documented in the literature exploring both clinician perspectives and patient experiences (e.g., Åsbring & Närvänen, 2002, 2004; Briones-Vozmediano et al., 2013; Gilje et al., 2008; Hunt et al., 2024; McManimen et al., 2019; Nishikawara et al., 2023; Pheby et al., 2020). Like previous studies, participants also recognised that adverse experiences of healthcare can be a source of significant emotional distress for patients and contribute to negative health outcomes.

4.5.4 Strengths and Limitations

While the results demonstrating good awareness and understanding of ME/CFS and FM among a sample of adults in Ireland are encouraging, several limitations warrant that results are interpreted with caution. Most notably, due to the purposive sampling strategy and data collection procedures employed, the survey is subject to substantial selection bias. Given that the survey specifically targeted individuals with some level of familiarity with the conditions, the results exclude perspectives of those without such knowledge thereby limiting generalisably to the wider adult population in Ireland. Further, recruitment through advocacy groups and patient networks likely resulted in the participation of individuals with greater experience, knowledge, or interest in the topic. This is evidenced in the demographic profile, whereby respondents generally rated themselves as health literate and more knowledgeable about chronic health conditions than acute illness. By comparison, data from the European health literacy survey (Sørensen et al., 2015) suggest that approximately 40% of the Irish public have problematic or inadequate general health literacy. However, as standardized assessments of health literacy were not employed in the present research, instead relying on participants' self-ratings, direct comparisons of this domain are not supported.

It is also noteworthy that while the survey sought to recruit adults from the public, a large portion of respondents disclosed living with chronic conditions, including ME/CFS and FM or Chronic Pain. While the lack of prevalence data from Ireland makes it difficult to comment on sample characteristics, it is likely that results are not representative of the population at large. In a similar vein, those with more negative perceptions of others' understanding, or personal experiences of stigma and negative attitudes, may have been compelled to highlight these issues in both healthcare and social settings. This is exemplified by the discrepancy between participants' self-rated knowledge and understanding, and their perception of the poor awareness and knowledge of both ME/CFS and FM among the general public. As emphasised, the survey was designed to specifically explore the perspectives of individuals who had heard of the clinical entities ME/CFS and FM, excluding those who were unaware of these conditions. It is possible that excluded participants possess some familiarity with the clinical profiles of these conditions without appropriate terminology, thus, an exploration of their lay beliefs and attitudes could be worthwhile in further research on stigma.

Despite the aforementioned limitations, this study is the preliminary attempt at understanding beliefs and perceptions about ME/CFS and FM among adults in Ireland and is driven by patients' perspectives on the need to improve awareness about these and other poorly understood conditions. Like previous research exploring patient experiences, results identify a range of challenges faced by patients in both social and healthcare contexts and reiterate medicine's long-standing problem with conditions that lack a well-defined pathophysiology, and consequently, diagnostic tools and treatments. Collectively, the evidence highlights a troubling lack of confidence in the healthcare system among adults regardless of experience of chronic illness, pointing to a need to explore clinical practices in the local context.

4.5.5 Conclusion

This mixed-methods cross-sectional study investigated perceptions of ME/CFS and Fibromyalgia among a sample of adults in Ireland, revealing a good awareness and understanding of both conditions and the substantial challenges faced individuals living with them. Contrary to documented negative societal biases, empathy for patients was expressed and the legitimacy and burden of these illnesses were widely acknowledged. However, stigma and disbelief were also perceived as a persistent problem in social and healthcare settings, underscoring the need for continued education and advocacy. Healthcare services were frequently criticised for underserving these patient groups, while professionals were perceived as lacking the knowledge, empathy, and skills to adequately support individuals with ME/CFS and FM. Further investigation of clinical practices, medical training and the experiences of healthcare professionals in Ireland is needed, as is research examining the lay beliefs and attitudes of the wider population.

Given the lack of research on patients' experiences and healthcare journeys in the local context, further work is also needed to inform specialised support strategies and to identify targets for change. Finally, while this study focused on ME/CFS and FM, the findings likely resonate with other poorly understood or "invisible" chronic illnesses, highlighting a broader need for research and improved healthcare provision for these often-marginalized patient populations. Building on the findings of Phase Two, Chapter 5 describes a study exploring the experiences of individuals living with a range of poorly understood conditions, with a particular focus on their personal and healthcare journeys.

Chapter 5. Study Three

Lived Experiences of Poorly Understood Chronic Illness

5.1 Introduction

Chronic illness profoundly shapes the lives of those who experience it, influencing not only their physical health but also their emotional, social and existential realities. Studies One and Two illustrate that for individuals with conditions that are poorly understood or unexplained, these impacts are compounded by arduous healthcare journeys marked by uncertainty, diagnostic delays, inadequate clinical management, and strained patient-clinician dynamics. Given that patient experience is recognised as a fundamental pillar of healthcare quality and a critical measure of success, with evidence linking it to improved clinical outcomes and patient safety (Doyle et al., 2013; Wolf et al., 2014), these results are concerning and suggest a need to explore patient perspectives in depth.

The patient stakeholders engaged throughout this doctoral research have also emphasised this, pointing to a need to explore lived experiences of healthcare journeys with a view to elucidating gaps in care, bringing patient voices to the fore of research and initiatives for change. Yet as pointed out by Wilson and Hutchison (2024), while the person-centered philosophy may underpin healthcare service models, this does not necessarily translate to the actual delivery of care; thus, a greater recognition and integration of patient perspectives and values by service providers is still needed.

Several approaches can be used to provide a multi-dimensional, in-depth account of the experience of living with and navigating healthcare with poorly understood chronic illness. As explained in Chapter 2 (*Section 2.2.1*), phenomenology provides an individualistic lens through which to understand the subjective, contextualised and embodied nature of experience; such an approach transcends biomedical or objective accounts of disease to reveal the ways in which individuals' lifeworld's are transformed by their experience of illness (Carel, 2011, 2021). Narrative methods complement the phenomenological position in that they capture temporal dynamics, focusing on the

"stories" people tell about their chronic illness (Frank, 2013; Hurwitz et al., 2004; Jowsey, 2016), the importance of which has been emphasised in healthcare research and the practice of narrative medicine. Attending to both phenomenology and story-telling, Charon (2006, 2001, 2007, 2017) writes extensively on the central role of patient narratives in medicine, arguing that to provide effective and truly patient-centered care, clinicians should engage with the stories and plights of their patients and allow themselves to be deeply moved by them.

In healthcare research, patients' first-hand accounts of their healthcare journeys have been pivotal in driving efforts to redesign and improve services (e.g., Bolz-Johnson et al., 2020; Galvin et al., 2015; Gualandi et al., 2019; Maas et al., 2023; Varley et al., 2011). This drive towards person-centered care has been reflected in the growing field of patient journey mapping. As reviewed by Davies et al. (2023), patient journey mapping research employs a variety of visual, narrative, and descriptive methods to document the full range of patient experiences within healthcare systems, including critical touchpoints, bottlenecks, and unmet needs. Bulto et al. (2024) argue that patient journey mapping has significant potential for transforming healthcare delivery by informing the development of personalized, patient-centered care and addressing communication and collaboration gaps within and across services. Mapping the patient journey, in addition to an in-depth consideration of patient narratives, can therefore form a foundation step towards exploring and developing care pathways for individuals with multi-systemic and poorly understood conditions in Ireland.

5.1.1 Objectives

The present study seeks to provide an in-depth, multi-dimensional account of the lived experiences of individuals with poorly understood conditions in Ireland, including their experiences of healthcare. In light of the needs identified by patient stakeholders in this research, the study also seeks to explore a selection of patient journeys by visually mapping the key touchpoints gleaned from their accounts of healthcare. As discussed in Chapter 2 (*Section 2.5.1.1*) and detailed below, the study employs the Life Grid, a distinctive and infrequently used method of interviewing and data collection. Thus, a secondary aim of the study was to explore participants' experiences of the Life Grid approach.

5.2 Methodology

5.2.1 Design

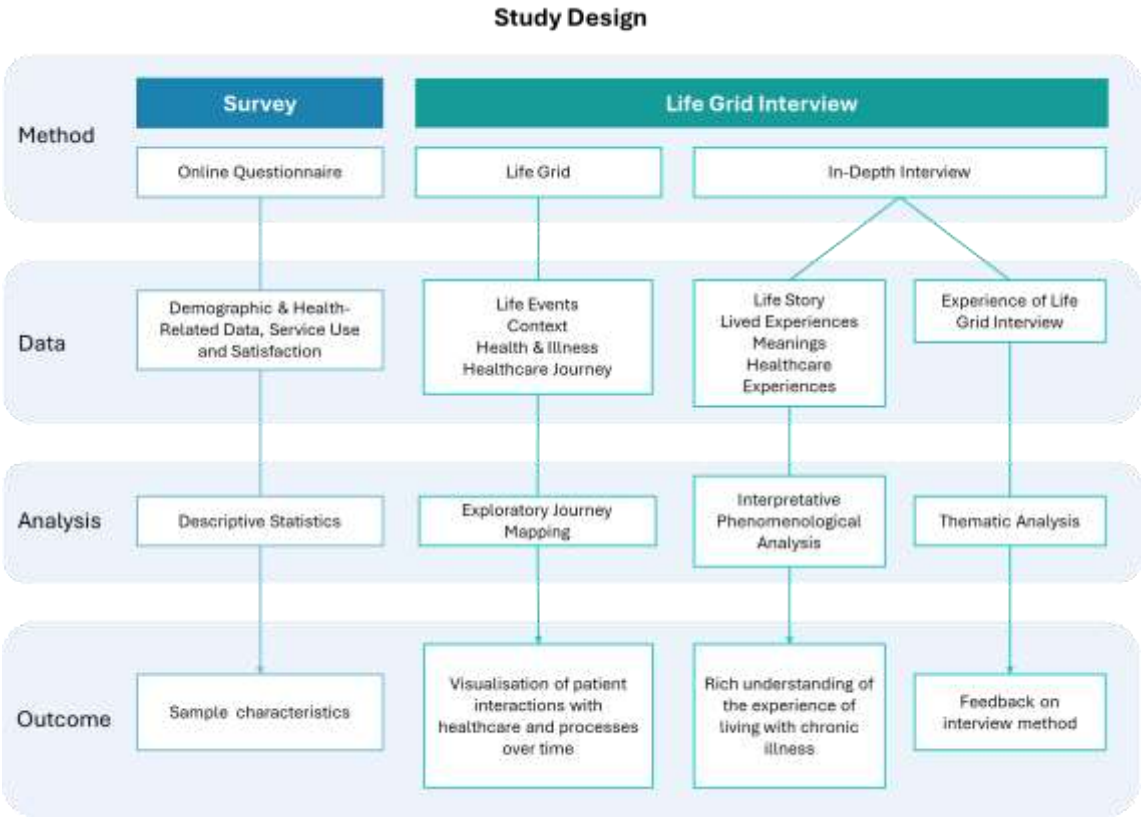
The primary paradigm of this qualitative study was interpretative phenomenological, with an emphasis on the lived experiences of individuals with poorly understood chronic illnesses. The study methods involved a survey of demographic and health-related characteristics and biographical mapping using the Life Grid Interview. Aligning with the pragmatist position, analytic pluralism was employed. Figure 5.1 illustrates the design of the study, with three components (Survey, Life Grid and In-Depth Interview) and the associated analytic approaches, detailed further in the sections below. Ethical approval for this study was granted by the School of Psychology Ethics Committee at Trinity College Dublin, Approval ID: SPREC032022-11, and is included in Appendix C1.

5.2.1.1 Patient and Public Involvement

As discussed in Chapter 2 (*Section 2.5.2*), consultations with patients in Research Phase One and Two underscored the need to examine healthcare experiences and patient journeys, thereby setting the priorities for the present study. PPI contributors emphasised a need to explore and understand the lived experiences of patients in Ireland, with a focus on capturing and illustrating the complexity of their healthcare journeys. In addition to conceptualisation and design, one member of the patient panel (SOC), a person with ME/CFS and Long-COVID, assisted in pilot testing of study methodology. Specifically, a pilot Life Grid Interview was performed to ensure suitability of the approach for in-depth interviewing in the context of fatigue and consequent cognitive symptoms, such as difficulties with memory and articulation. Following feedback, two changes were made to the procedure to make it less taxing on participants. The visual tool was revised to include core interview questions for ease of reference, and the interview procedure was modified to enable participants to complete a preliminary timeline prior to the full in-depth interview.

Figure 5.1

Study design, methods and analytic approach.



5.2.2 Participants

A purposive sampling strategy was used to recruit participants in July and August of 2023. Participants were eligible if they were 18 years or older, living with a self-reported or formally diagnosed condition falling under the broad category of "medically unexplained" or "poorly understood" illness, and had received healthcare for their condition in Ireland. To reach the targeted population, the researcher utilised personal networks and shared the study online with patient communities and advocacy groups. Suggestions on specific communities were sought from the patient panel and PPI contributors involved throughout the research. A total of 21 individuals initially expressed interest in participating in the interview. Of these, three did not consent due to current severity of symptoms, two preferred to share written accounts and declined to be interviewed, and four did not respond to the initial follow-up invitation. A total of twelve participants consented to take part and completed all components of the study.

5.2.3 Materials and Procedure

5.2.3.1 Demographic Survey

Basic demographic and health-related and service use data were collected from each participant prior to taking part in the Life Grid interview. Once participants provided informed consent, they received a link to an online questionnaire hosted on the survey platform Qualtrics (Qualtrics, Provo, UT).

Consenting participants were asked to indicate their age, gender identity (*Male, Female, Non-Binary, Prefer Not to Say),* highest level of education completed *(No education, Primary Level, Secondary Level, Third Level, Post-Secondary Training, Prefer Not to Say)* and their current employment status (*Unemployed, Employed Full Time, Employed Part-time, Student, Other: provide detail, Prefer Not to Say)*. Participants were then asked to list the chronic health condition(s) they live with and to indicate the main symptoms they experience (*Physical, Cognitive, Affective, Other: provide detail*). They were asked to rate the severity of their symptoms on a 5-point Likert Scale (*No Symptoms- Very Severe Symptoms*) as well as the overall impact of the condition on their quality of life (*No Impact- Very Significant Impact*). With respect to the condition(s) disclosed, participants were asked to estimate, in months or years, the age at which their symptoms first started and the age at which they first sought medical advice for the said symptoms.

Participants then answered seven questions relating to their healthcare journey. They were asked to indicate whether they have received a formal diagnosis of their condition(s) (*Yes/No*) and, if yes, to estimate the approximate time it took to receive this diagnosis in months and/or years. Participants were asked to estimate the number of services or different specialists they have seen in relation to their condition(s), to list what services they had used the most, and which services they found to be most useful. Finally, participants were asked to indicate on a 5-point Likert Scale (*Extremely Dissatisfied- Extremely Satisfied*) their satisfaction with the quality of healthcare services currently available to them, and their overall satisfaction with their healthcare journey to date.

5.2.3.2 The Life Grid

Semi-structured in-depth interviews were conducted using the Life Grid approach to interviewing (Parry et al., 1999), described in detail in Chapter 2 (*Section 2.5.1.1*). Briefly, the method was initially developed for the collection of quantitative data while addressing issues of recall validity in health research (Berney & Blane, 1997), and later applied to facilitate biographical interviews in qualitative research on smoking behaviour. More recently, Richardson et al. (2009) emphasised that the Life Grid can be a fruitful tool for exploring the meanings and experiences of health and illness over time, particularly where temporal fluctuations in symptoms are a feature. As previously discussed, the merits of the approach suggest that it is particularly suitable for life story interviews with individuals living with chronic conditions. In the present study, the primary purpose of the Life Grid was to facilitate the participants' sharing of life stories and their lived experiences of chronic illness in a holistic and contextual manner. The specific design and interview procedure employed are described below.

Drawing on the limited methodological literature available, and primarily the work of Parry et al. (1999) and Richardson et al. (2009), the Life Grid was structured as a blank table created in Microsoft Word, with interview topics across the horizontal axis, and a blank time span (year and age) with associated meaningful life event on the vertical axis. Interview topics pertained to *(i)* the participant's life at the time of the event, including the domains of family, friends, work and leisure, and *(ii)* the participant's health at the time of the event, including any symptoms or illnesses they were experiencing, their overall physical or mental health, as well as their healthcare journey, including any key touchpoints with the health system and experiences of these. Table 5.1 provides a template of the grid used, while Appendix C2 provides an example of a completed Life Grid, created from a combination of interviews for illustrative purposes.

Table 5.1

The Life Grid

Life Events			*What was your life like at the time?*		*What was your health like at the time?*	
Year	Age	Significant Life Events	Family & Friends	Work & Leisure	Health & Illness	Healthcare Journey

5.2.3.3 The Life Grid Interview

The Life-Grid interview involved two steps. First, pre-interview, participants completed a partial Life Grid, building a timeline of any significant or meaningful events over their lifespan. Second, participants took part in an in-depth interview, during which they completed the full Life Grid in collaboration with the researcher.

Life Grid Preparation

Participants were emailed with a partial template of the Life Grid to complete and return to the researcher prior to the interview, along with a set of instructions on completing this template and the structure of the interview. The partial Life Grid consisted of the Year, Age and Significant Life Events columns only (see Table 5.1), and participants were asked to reflect on any meaningful events across their lifetime and to note these on the grid, to the best of their ability, in chronological order. Participants were informed that during the interview, they would be asked to elaborate on these events during the interview and would be able to modify the content of the Life Grid should new insights arise. Once returned, the researcher extended the Life Grid by adding the remaining

columns (i.e. "*What was your life like at the time?*" and "*What was your health like at the time?*"), resulting in additional blank cells to be completed with the participant during the interview.

Life Grid Interview

Participants were given a choice of taking part in an in-person interview at the School of Psychology at Trinity College Dublin, or online, using the teleconferencing software Zoom. A total of twelve interviews were conducted, ten of which were online, between July and August 2023. Participants were informed that the interview could last approximately 1 hour 30 minutes but could be longer or shorter depending on how much they wished to share. Interviews lasted an average of 1 hour 40 minutes, ranging from 48 minutes to 2 hours 45 in duration, with breaks offered to participants throughout. During each interview, the participants' personal Life Grid was presented on-screen, either through the Zoom screen-share function where online, or on a computer monitor in-person. In the interest of transparency, each interview commenced with an introduction to the study aims and outline of the interviewing procedure, including the co-construction of the Life Grid during the interview.

Briefly, interviewees were instructed that they would be invited to introduce themselves and tell their story in any order they would like, and that the researcher would follow with probing questions about other aspects of their life and health at the time. The flexibility of the approach was emphasised, with the Life Grid was treated as a conversation guide. Participants were asked whether they preferred to make their own notes on the Life Grid during the interview, but all opted for the researcher to enter information into the grid cells as they spoke. The Life Grid remained on-screen for the duration of the interview, with participants instructing the researcher where new information should be added.

All interviews were audio-recorded, transcribed verbatim to Microsoft Word, and anonymised by the researcher to ensure all identifying data (e.g., names) were removed, after which recordings were discarded. Interview transcripts and complete Life Grids were emailed to participants for review, with opportunity for content to be redacted or corrected in the final transcript.

Interview Structure

The interview approach was semi-structured and largely directed by each participant and the events they had disclosed on the Life Grid. The grid was ultimately used as a visual aid to stimulate the interviewees' reflexivity with regards to their life trajectories rather than to formally structure the interview. A broad questioning strategy was used, with a focus on building rapport and maintaining flow of the narrative. Participants were informed that they could tell their story in any order, return to or move past life events throughout the interview, and add events or modify the chronological order of events on the grid during the interview.

Each interview commenced with an introductory question (e.g., "*Can you tell me a little bit about yourself and what brings you here?"),* followed by an invitation for the participant to begin telling their story with reference to any event on the Life Grid ("*Starting anywhere on the Life Grid, could you tell me more about your life?").* As participants discussed their life events, they were asked to elaborate on additional facets of their life (e.g., "*What was your family life like?*", "*What was your social life like*?") and health (e.g., "*What was your health like*?", *"Did you experience any symptoms?"*) at the time period, with further probing questions in relation to the healthcare journey (e.g., "*Did you see any medical professionals?", "What was your experience like?").*

To maintain flow of narrative, once the participant discussed any event to their satisfaction or concluded the discussion of an event, the researcher proceeded with the interview by asking "*And what happened then*?". As participants' narratives often referred to events not included on the Life Grid, clarification on structure was sought by the researcher at various points of the interview (e.g., "*Do you remember around what age you were at the time?*", "*Do you remember if this happened before or after [another event]?*"), and these were noted. Furthermore, in the case that participants disclosed adverse life events (e.g., history of abuse, sexual assault) on the grid, no request from the researcher was placed on participants to discuss or elaborate on these. Where naturally acknowledged by the participant, participants were assured that they could discuss the event in as little or much detail as they would like, take a break, or discontinue the interview. All participants proceeded to discuss such events with varying degrees of detail.

Once the final event on the Life Grid was discussed, participants were given the opportunity to modify or share additional information about their life or health ("*Is there anything else you would like to add*?"). Finally, participants were asked to share their perspective on and experience of the Life Grid interview, including any personal reflections on the process (e.g., "*What was it like for you complete the Life Grid?", "What was it like for you to discuss your life in this way?")*. A debrief followed the formal interview, with both participant and researcher sharing their experiences, and with the researcher signposting psychological supports should they be needed in light of the content discussed. Immediately following each debrief, the author took reflexive notes of their impressions about how the interview progressed, including the overall atmosphere and takeaway messages, as well as personal reflections on interview content.

5.2.4 Analysis

As previously mentioned, and illustrated in Figure 5.1, analytic pluralism was employed, with four types of analysis conducted: *(i)* descriptive analyses of demographic and health related data, *(ii)* exploratory analyses of the Life Grid contents with regards to the patient journey, *(iii)* Interpretative Phenomenological Analysis of the in-depth interviews, and *(iv)* Thematic Analysis of Participants' reflections on the interview procedure.

5.2.4.1 Descriptive Statistics

Descriptive statistics (means, frequencies) were calculated for all measures of the demographic survey. No formal content analysis was performed on responses to open-ended questions. This data was extracted and manually cleaned (e.g., for spelling errors), then treated as nominal data for descriptive analysis.

5.2.4.2 Patient Journey Analysis

Patient journey data was extracted from a selection of Life Grids purposefully chosen to illustrate journeys of varying lengths and complexities among individuals with similar diagnoses. A small sample of four cases was selected for exploratory analyses based on convenience, owing to their availability within the sample, as well as their potential to illustrate participant trajectories, owing to the detail recounted by participants. These cases concerned the pathways of pairs of individuals with an established diagnosis of

Functional Neurological Disorder (FND) and Fibromyalgia, with one protracted and one comparatively shorter journey in each pair, thus allowing comparison.

Exploratory visual maps were constructed drawing on the scoping review conducted by Davies et al. (2023), who identified six representative types of journey maps across the literature, also highlighting a lack of standardised approaches to mapping. The approach taken in the present study was mapping by events over time, with the aim of illustrating key service touchpoints, continuity or discontinuity between services and any key interventions or delays (e.g., diagnosis and referral).

5.2.4.3 Interpretative Phenomenological Analysis

A selection of transcripts was analysed using Interpretative Phenomenological Analysis (IPA), the premise and assumptions of which are detailed in Chapter 2 (*Section 2.5.1.*2). Twelve in-depth interviews were conducted and transcribed by the thesis author and a sub-sample of five was chosen for analysis in line with methodological principles that favour small sample sizes to enable in-depth idiographic analysis (Smith et al., 2009). These accounts were chosen reflexively by the author, informed by several key considerations, particularly concerning the narrative richness of the data in relation of the study aims. Specifically, cases were purposefully chosen to *(i)* capture a broad spectrum of conditions, recognising that individuals with poorly understood chronic illness often experience multi-morbidity and overlapping symptoms; *(ii)* reflect a wide range of and depth of experiences across the life course, as articulated by participants, and *(iii)* encompass nuanced accounts of the healthcare journey and navigating healthcare in Ireland. A limit of five cases was set to enable deep analytic engagement with narratives, while also ensuring the analysis remained feasible within the scope of the study. Accounts not included in IPA were included in other analyses of this study.

Analysis proceeded via the broad methodological steps outlined by Smith et al. (2009), outlined below. The approach is typically described as an iterative and inductive cycle, which moves from understanding of the particular (i.e. individual) to the shared (i.e. group) experience. Quality of the analysis was ensured by drawing on guidelines (Nizza et al., 2021; Smith, 2011), which emphasise that high quality IPA studies exhibit sensitivity to context, maintain rigor and transparency, construct coherent and compelling narratives with an experiential or existential account, involve close analytic reading of

participants' words, and attend to convergence and divergence. Each interview was analysed in full prior to conducting the cross-case analysis to generate key themes. Throughout the analysis, efforts were made to ensure reflexivity, with the researcher reflecting on their own influence on the interpretative process.

Single Case Analysis

Step 1. Reading and re-reading

First, the interview was read and re-read in full to immerse and engage the researcher with the participants' experience. In this process, initial interpretations and observations were noted in a reflective journal.

Step 2. Line-by-line Analysis

A line-by-line analysis was conducted to identify emergent patterns, or themes, within the experiential material, with exploratory comments handwritten on the left-hand margin of the transcript. As discussed by Smith et al. (2009), this step involves a close reading of the transcript and engagement in analytic dialogue, reflecting on what words, phrases and sentences mean to the researcher, and what they can mean to the participant. The exploratory comments were *(i) descriptive notes,* which summarise the participants' explicit experience (i.e., what is said); *(ii) linguistic notes*, which include observations about how language is used by the participant, including specific words in the narrative (i.e., how it is said); *(iii) interpretative notes*, which offer interpretations of the meaning behind what is said, moving away from the explicit claims of the participant (i.e., what it means); and *(iv) conceptual notes*, which reflect more abstract theoretical concepts of themes that can help explain the participants' experience. Once annotated in full, comments from line-by-line analysis were reviewed and interpreted to develop experiential statements associated with extracts of text, reflecting interpretations of experience whilst remaining grounded in the narrative. These statements were initially hand-written in the left-hand margin, then transferred into Microsoft Excel along with data excerpts (i.e. quotes) and locations (i.e. line numbers) for further stages of analysis.

Step 3. Developing emergent themes

Third, participant emergent themes were developed by clustering the experiential statements on the basis of the researchers' understanding of the core experiences, their patterns and deeper meanings, thus moving towards a higher level of abstraction. As

discussed by Smith et al. (2009), this step is a manifestation of the hermeneutic cycle, whereby the themes reflect not only the original words and experiences of the participant, but the analyst's interpretation regarding their meaning. During this process, emergent themes and associated line numbers from experiential statements within them were tabulated in Microsoft Excel, thus simultaneously reducing the volume of data while maintaining complexity within it.

Step 4. Searching for connections

Fourth, emergent themes were clustered to illustrate patterns and relationships between within each transcript, which was achieved by colour-coding in Microsoft Excel. The emergent themes were organised in a hierarchy with sub-themes and super-ordinate themes, along with data extracts and locations, based on conceptual connections and patterns observed in the data. A table listing the superordinate and subordinate themes was produced for the analysed interview. Descriptive summaries for each subordinate theme were written to assist cross-case comparisons. Reflexive notes were revisited when devising case descriptions and used to supplement case descriptions.

The above steps were carried out to completion on each of the five interviews individually, with the researcher engaging in bracketing of analytical insights between cases. Separate tables of superordinate and subordinate themes were produced for each interview before proceeding to cross-case analysis.

Cross Case Analysis

Step 5. Looking for patterns across cases

Once all five transcripts were analysed, comparisons between cases were conducted to identify shared themes across the interviews. As discussed by Smith et al. (2009), this step seeks to account for convergences and divergences within cases, allowing for an in-depth understanding of idiosyncrasies but also the shared qualities in the data. The author reviewed the descriptive case summaries and thematic structure of each interview with the aim of identifying broad, higher-order themes across the narratives (i.e., Group Experiential Themes). Extracts of data associated with the interpretations were revisited to support cross-case comparisons. A group-level thematic table was constructed by comparing the accounts of participants.

Step 6. Interpretation and synthesis

The process of synthesis and interpretation was iterative and involved moving between individual case findings and wider group-level data, relating this to the research question and literature. Findings were structured into a thematic account with a focus on the meanings and experiences of health and illness within the participants' life narratives. Themes were evidenced by data extracts and accompanied by a detailed interpretative commentary. In line with the double hermeneutic process involved in IPA, the author reflected on their positionality and its influence in interpreting participants' interpretations of their own experiences.

5.2.4.4 Thematic Analysis

Reflexive thematic analysis (Braun & Clarke, 2006, 2021b) was conducted on participants' responses to the question regarding their experience of taking part in the Life Grid Interview. As discussed in Chapter 2 (*Section 2.5.1.4)*, this approach is assumed to reflect the researcher's worldview and interpretation, aligning with the overall phenomenological position of the study. The accounts of all participants were included in the analysis to capture a breadth of perspectives on methodology.

The analysis was performed by the researcher and research assistant (AA), an undergraduate Psychology student with an interest in qualitative work in health psychology. Thematic analysis followed six steps in the manner previously detailed in Chapter 4 and was conducted using NVivo 12 Plus Software. Briefly, the steps of analysis involved *(i)* Familiarisation with data, involving immersion in content and an initial discussion of impressions among analysts; *(ii)* Systematic data coding and categorisation, performed independently and discussed to ensure consistency; *(iii)* Creation of initial themes and subthemes, achieved by grouping of related codes based on observed patterns of meaning; *(iv)* Reviewing and refining themes, ensuring agreement among analysts that themes accurately reflect data; *(v)* Defining and naming themes; and *(vi)* Writing up the thematic findings, accompanied by relevant quotes. Given the reflexive nature, a codebook was not devised and inter-rater agreement was not analysed using the coding comparison query. Instead, all stages of analysis were performed by both analysts in a collaborative manner, whereby coding decisions and thematic findings were discussed to reach agreement on final outcome.

5.3 Results

5.3.1 Participant characteristics

5.3.1.1 Demographics and Health

Sample demographic and health characteristics are summarised in Table 5.2 and detailed in Table 5.3. Twelve participants (4 male, 8 female), aged between 21 to 62 years (*M*=40.1, *SD*=14.8) took part in Life Grid interviews. Most participants were educated to a post-secondary or higher level (*n*=8) and were currently unemployed or not working due to health-related reasons (*n*=7). As a group, participants reported living with a range of 26 conditions, and multimorbidity was common, with 10 participants reporting between 2 and 8 conditions (*M*=3.5) each, detailed in Appendix C3**.** The conditions most frequently reported by participants were Fibromyalgia (*n*=5), Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (*n*=4), Hypermobile Ehlers-Danlos Syndrome/ Hypermobility Spectrum Disorder (*n*=3), and Functional Neurological Disorder (*n*=3).

Participants most frequently reported living with Moderate (*n*=5) or Severe (*n*=6) symptoms and most rated the impact of the condition on their overall quality of life as Significant (*n*=4) or Very Significant (*n*=5). All participants indicated experiencing physical symptoms such as pain and fatigue, though only one participant indicated that their symptoms were solely physical in nature. Others more frequently reported a combination of Physical and Cognitive symptoms (*n*=6), or Physical, Cognitive and Affective Symptoms (*n*=5). Three participants indicated having additional symptoms, one reporting “weakness” and two reporting “non-epileptic seizures”. Most participants reported seeking medical advice for their symptoms within months to one year of onset of symptoms (*n*=10), with one participant seeking help 2.5 years later, and one 12 years later. All participants in the study reported having received a formal diagnosis for their conditions, with time to diagnosis ranging between “a few months” to 11 years, and an approximate average of 5.5 years.

Table 5.2

Participant Demographic and Health Characteristics (N=12)

	n	*%*
Demographic Characteristics		
Age	*M*=40.1	*SD*=14.8
Gender		
Male	4	33
Female	8	66
Non-Binary	0	0
Educational Attainment		
Primary Level	0	0
Secondary Level	4	33
Training (Post-Secondary)	1	8.3
Third Level	7	58.3
Prefer not to say	0	0
Employment Status		
Currently Employed	3	25
Student	2	16.6
Currently Unemployed	3	25
Other: On sick Leave	2	16.6
Other: Retired early due to health	1	8.3
Other: Unable to work due to health	1	8.1
Prefer not to say	0	0
Participant Health		
Symptoms Experienced		
Physical	12	100
Cognitive	11	91.6
Affective	5	41.6
Other	3	25
Symptom Severity		
Mild	0	0
Moderate	5	41.6
Severe	6	50
Very Severe	1	8.3
Impact on Quality of Life		
No Impact	0	0
Mild Impact	0	0
Moderate Impact	3	25
Significant Impact	4	33.3
Very Significant Impact	5	41.6

Table 5.3

Participant Health, Service Use and Satisfaction

ID	*Gender (Age Range)*	*Condition(s)* [a]	*Impact: QoL*	*Symptoms* [b]	*Symptom Severity*	*Symptom onset (Years)*	*Help Seeking (Years)*	*Time to Diagnosis*	*Services Accessed (N)*	*Satisfaction: Services Available*	*Satisfaction: Healthcare Journey*
1	Male (18-24)	POTS, hEDS, ME/CFS, Chronic Migraine	Very significant	Physical, Cognitive	Severe	15	17.5	8 years	10	Somewhat dissatisfied	Somewhat dissatisfied
2	Male (25-34)	Fibromyalgia	Moderate	Physical, Cognitive	Moderate	20	20	Few months	4	Somewhat satisfied	Neither
3	Female (55-64)	Long COVID	Significant	Physical, Cognitive	Severe	59	59	1 year	8	Neither	Somewhat dissatisfied
4	Female (18-24)	Fibromyalgia, CRPS	Significant	Physical, Cognitive, Affective, Other: weakness	Severe	15	16	1.5 years	12	Extremely dissatisfied	Extremely dissatisfied
5	Female (45-54)	Fibromyalgia	Moderate	Physical, Cognitive, Affective	Moderate	38	38	10 years	5	Somewhat dissatisfied	Neither
6	Female (35-44)	ME/CFS; Long COVID (Post-Vaccine)	Very significant	Physical, Cognitive	Moderate	30	30	4 months	6	Somewhat satisfied	Somewhat dissatisfied
7	Male (45-54)	FND	Very significant	Physical	Severe	25	25	11 years	12	Somewhat dissatisfied	Somewhat dissatisfied
8	Female (18-24)	POTS, ME/CFS, hEDS/HSD, Fibromyalgia	Very significant	Physical, Cognitive	Very severe	18	18	2 Years	20	Somewhat dissatisfied	Somewhat dissatisfied

ID	*Gender (Age Range)*	*Condition(s)* [a]	*Impact: QoL*	*Symptoms* [b]	*Symptom Severity*	*Symptom onset (Years)*	*Help Seeking (Years)*	*Time to Diagnosis*	*Services Accessed (N)*	*Satisfaction: Services Available*	*Satisfaction: Healthcare Journey*
9	Female (25-34)	FND	Moderate	Physical, Cognitive, Affective, Other: NES	Moderate	16	16	7 years	Many	Extremely dissatisfied	Extremely dissatisfied
10	Male (45-54)	FND	Significant	Physical, Cognitive, Affective, Other: NES	Severe	42	42	6 months	3	Extremely dissatisfied	Somewhat dissatisfied
11	Female (45-54)	Fibromyalgia	Significant	Physical, Cognitive, Affective	Moderate	53	53	5 months	8	Extremely dissatisfied	Extremely dissatisfied
12	Female (55-64)	Lyme Disease, hEDS, Chronic Pain Syndrome, Chronic Migraine, ME/CFS	Very significant	Physical, Cognitive	Severe	25	37	10 years	10	Somewhat satisfied	Somewhat dissatisfied

Note. [a] Conditions meeting participant inclusion criteria (i.e., poorly understood chronic illness). [b] Symptom Type: Physical (e.g., pain, fatigue), Cognitive (e.g., memory problems), Affective (e.g., Low mood, anxiety), O=Other. Abbreviations: ID= Participant Number, QoL= Quality of life. Conditions: Postural Orthostatic Tachycardia Syndrome (POTS); Hypermobile subtype Ehlers-Danlos Syndrome (hEDS); Hypermobile Spectrum Disorder (HSD); Chronic Regional Pain Syndrome (CRPS), Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS), Functional Neurological Disorder (FND), Non-epileptic Seizures (NES)

5.3.1.2 Service Use and Satisfaction

As detailed in Table 5.3, most participants (*n*=8) were Somewhat or Extremely dissatisfied with the quality of healthcare services available to them, and ten out of twelve were Somewhat or Extremely dissatisfied with their healthcare journey to date. Participants reported accessing between 3 to 20 specialists or health services in relation to their condition or symptoms, with one reporting "too many to count". As a group, participants reported that the most frequently used services were their GP (*n*=11), Physiotherapy (*n*=5) and Psychology or Counselling (*n*=5), while Physiotherapy (*n*=5) and GP services (*n*=4) were the most useful to them. When cross-tabulated, there was partial overlap among individual participants with regards to the services they accessed most and those perceived as being most useful, with several instances of useful services being accessed infrequently (see Table 5.4).

Table 5.4

Mapping of participants' reported service use vs. perceived usefulness

	Participant											
Service or Specialist	1	2	3	4	5	6	7	8	9	10	11	12
General Practitioner (GP)	✓	✓●	✓	✓	✓	✓●		✓	✓	✓●	✓	✓●
Physiotherapy	✓	●		✓			✓●		✓●	✓●	●	
Psychology/Counselling	✓			✓●					✓	✓●	✓	
Occupational Therapy			✓									
Hydrotherapy			✓									
Neurology	✓						✓●			✓●		
Rheumatology	✓●											
Cardiology	✓●											
Genetic Counselling	✓											
Nutritionist	✓											
Pain Consultant					✓●							
Consultant (Unspecified)						●		✓				✓●
Alternative Medicine										✓●		

Note: ✓ denotes most frequently used service(s) ● denotes most useful service(s)

5.3.2 Patient Journeys

Four patient journey maps were constructed and are presented in Appendix C4. The illustrative cases concerned two participants with a diagnosis of FM, and two with FND, with each pair varying in terms of journey length (see Table 5.4 for details). The patient journeys, focusing on service touchpoints, continuity in care, diagnoses and referrals, are outlined below.

Participant 2

Participant 2 is a male with FM who reported a relatively short journey from symptom onset to diagnosis, approximately 4 months, and 4 key services used: GP, Rheumatology, Neurology and Physiotherapy. While the GP was a key touchpoint in relation to earlier mental health and pain concerns, the FM-related healthcare journey commenced with A&E following sudden onset pain and inability to walk. Discharge with no abnormalities detected through imaging resulted in a sequence of referrals initiated by the GP. Assessment by Rheumatology confirmed FM diagnosis, which was previously speculated by the GP, resulting in a referral for Physiotherapy and discharge back to the GP for pain management. Intensifying and new symptoms in the subsequent year, specifically numbness, resulted in the participant seeking further care from their GP, which resulted a referral to Neurology. Assessments by Neurology found no abnormalities, resulting in discharge back to GP care, with Physiotherapy ongoing. The participant has been experiencing ongoing pain in the four years following, with management involving the GP and Physiotherapy at present (See Figure A, Appendix C4).

Participant 5

Participant 5, a female with FM, detailed a 10-year journey to diagnosis which was complicated by long-standing back pain and widespread pain consistent with FM emerging in the aftermath of an accident. Five key healthcare services were identified: the GP, Rheumatology, Pain Clinic, Physiotherapy and Counselling. The GP's critical role in initiating referrals was highlighted, though poor continuity of care was seen throughout the journey. The participant sought assistance from her GP regarding widespread pain following the accident, receiving pain medication and referral to Physiotherapy. A subsequent visit to the GP involved a failed referral for MRI, though this was corrected by a third GP. Within two years, the participant returned to A&E due to

acute pain but was discharged once again as no abnormalities were detected. This led to subsequent GP consultations and a referral to Rheumatology, where she was diagnosed with Failed Back Syndrome, and referred for surgery and to the Pain Clinic within the same year. The participant continued to experience ongoing pain and engaged in self-management for 4 years. She then sought help from GP due to progressing walking difficulties and fatigue, which resulted in a referral to a hip specialists and pain clinics for further management. Discharge from the Pain Clinic occurred within one year despite ongoing pain, with the participant returning to the GP for further guidance and referral. At this stage, she was given diagnosis of FM by the GP. She disengaged from healthcare despite ongoing problems, reflected by a 9-year gap in her journey map. The GP remains the primary touchpoint with regards to progressing symptoms and disability, and at present, no further referrals have been made (See Figure B, Appendix C4).

Participant 10

Participant 10 is a male with FND, who described a short 6-month journey to diagnosis and three key services used: the GP, Neurology and Physiotherapy. Attending the GP at the onset of symptoms, he was referred for and accesses an MRI within several months. As lesions were identified by MRI, the GP subsequently referred him to a private Neurologist whom he attended within the year. He was discharged back to the GP with a speculative FND diagnosis and self-management guidelines. Several months later, loss of consciousness lead to re-engagement with services through A&E and referral for a Neurology in-patient hospital stay, where FND was diagnosed. There was no follow up nor subsequent imaging and the participant was discharged back to the GP. The GP continued symptomatic treatment with a referral to public Neurology services, for which the participant had been waiting 3 years at time of interview (See Figure C, Appendix C4).

Participant 7

Participant 7, a male with FND, described an extensive 11-year journey to diagnosis, during which he engaged with a wide range of healthcare services. These included the GP, Physiotherapy, Rheumatology, Neurology and Cardiology among others. His patient map underscores the pivotal role of the GP in facilitating referrals and coordinating access to multiple specialties, however, extended delays between referrals and first contact with services were a significant issue. For example, there was a 3-year delay

between referral and a Rheumatology appointment, and a 4-year delay to a Neurologist specialised in movement disorders, who eventually diagnosed FND. Continuity of care was illustrated for Physiotherapy over a number of years as well as ongoing Botox injections for dystonia at a hospital-based Neurology service. The GP also played a crucial role in referring to mental health services, both privately and through public primary care, for the management of ongoing anxiety and depression. Referrals for imaging were made by specialists (e.g., Urologists, Neurologists) and due to private health insurance occurred without delay (See Figure D, Appendix C4).

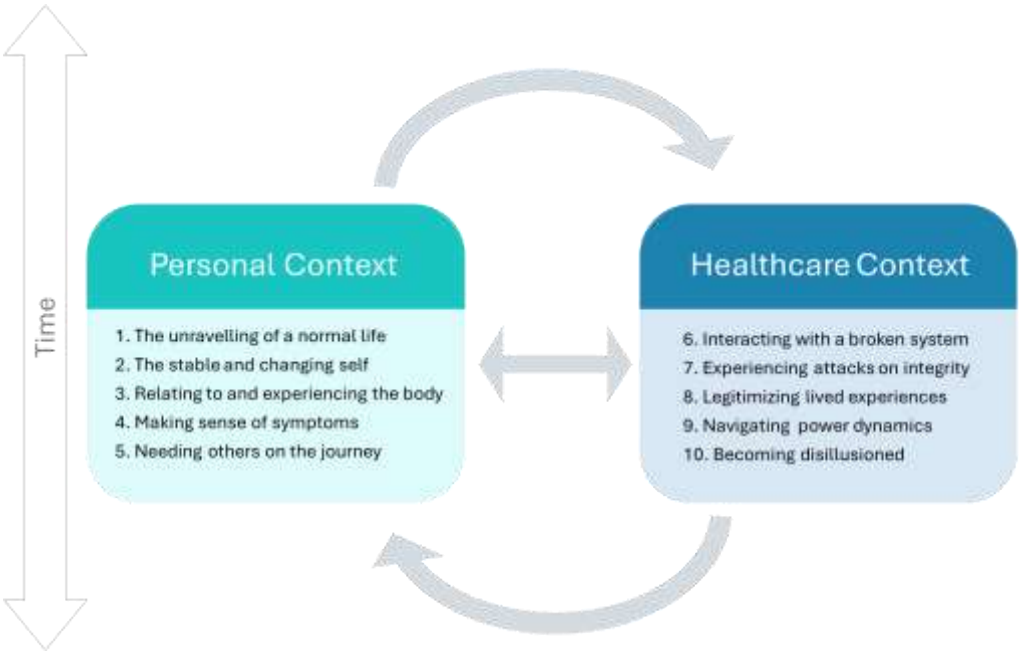
5.4 Findings I. Interpretative Phenomenological Analysis

The interpretative phenomenological analysis (IPA) of participants' life stories revealed 10 interrelated superordinate or group experiential themes (GET), which capture shared patterns of meaning across all participants. As illustrated in Figure 5.2, superordinate themes concern participants' lived experiences of *(i)* their personal contexts, encompassing subjective experiences of their identity, embodiment, and the evolving process of adapting to chronic illness, and *(ii)* the healthcare context, reflecting their lived experiences of navigating the patient journey and engaging with the healthcare system.

While these themes offer a structured lens for exploration, each was situated within participants' broader life course, shaped by the passage of time and shifting lifeworlds. Thus, the dynamic, relational and temporal nature of participants' subjective experiences is emphasised throughout.

Figure 5.2

Superordinate themes (GET) representing the lived experience of poorly understood chronic illness over time, across personal and healthcare contexts



In the following section, results are presented thematically, with superordinate themes and subthemes, and illustrative quotes throughout. In line with quality guidelines (Smith, 2011; Nizza et al., 2023), the results are presented in the participants' context, focusing on experiential and existential narratives and with close analytic reading of their words. Convergent and divergent accounts are emphasised and linguistic particularities retained. Table 5.5 provides brief contextual information about each participant.

Table 5.5

Participant Case Summary

Case	Background
1	Dan is a male in his early 20s, navigating life with a range of poorly understood conditions, including EDS, POTS, ME/CFS and chronic migraine. He experienced generally good health in childhood, marked by minor injuries, surgeries and hypermobility. His symptoms began to emerge and intensify during adolescence with a significant impact on his schooling, leisure activities, social interactions and school life. As he transitioned to young adulthood, the symptoms grew more severe and at times life-threatening, leading to multiple hospitalisations and a complex diagnostic journey which continues.
2	Lisa is a female in her early 40s who experienced remarkable good health during her childhood, adolescence and young adulthood. Her health took a drastic turn following a viral infection at age 30, after which she developed ME. Following the misinformed guidance of clinicians, she initially practiced Graded Exercise Therapy, leading to significant deterioration and disability. Becoming unable to work and bedbound, she began to engage in pacing and entered a 10-year period of remission. Her symptoms were recently reactivated following vaccine with significant impact on all aspects of her life at present.
3	Sue is a female in her mid-20s, who has experienced a lifelong journey with chronic pain. She recalls early onset pain around the age of 5 due to a rare spinal condition. During adolescence, her pain gradually became more widespread, significantly affecting all aspects of life. Preliminary diagnoses included Juvenile Arthritis or Fibromyalgia, though these were not definitive, and symptoms progressed to severe nerve pain despite efforts at management. She is awaiting further evaluation for a possible diagnosis of Chronic Regional Pain Syndrome (CRPS).
4	Isabel is a female aged in her early 20s, living with ME/CFS, EDS, POTS and chronic pain, among other symptoms. She recalled good health throughout childhood and adolescence with the exception of tonsilitis and glandular fever from age 15 onwards. Her health began to decline significantly during young adulthood in the aftermath of a viral infection, affecting her college, social and work life, and further declined following vaccine, resulting in hospitalisation. Her symptoms continue to grow more severe and complex, and her diagnostic and treatment journey is ongoing.
5	Sharon is a female in her mid-50s who experienced minor health issues during her childhood and adolescence, including anaemia and dizziness consistent with POTS. Her symptoms began to accumulate from young adulthood onwards, as she developed hypothyroidism, underwent preventative surgery for cancer, and developed widespread chronic pain following a medical procedure. Diagnosed with Fibromyalgia, her symptoms have continued to grow more severe, impacting her personal, social and work life. She is awaiting further investigation to explain her progressing symptoms and verify her diagnosis.

Note. Potentially identifiable characteristics, such as rare diagnoses and exact age have been removed to protect participant confidentiality.

5.4.1 GET 1. The unravelling of a normal life

When sharing their life stories, participants often reflected on their childhoods and spoke of the many ups, downs and unexpected turns throughout their lives. Despite facing setbacks and minor illnesses in their youth, most painted pictures of living "normal" lives and being largely active and healthy. However, as their narratives progressed, there was a noticeable shift, with symptoms and illnesses emerging as increasingly dominant aspects of their experiences, suggesting a disruption to the sense of normalcy they once knew. All participants reported initially attempting to downplay or deny their symptoms, both to themselves and others. When the symptoms became unavoidable, they expressed a profound sense of injustice in becoming chronically ill, vividly describing the losses and far-reaching impacts of their conditions across all areas of their lives.

5.4.1.1 Living a normal life with twists and turns

Four participants' accounts portrayed a deep sense of gratitude for their pasts, as they warmly counted their blessings and shared memories of "*great*" and "*lucky*" childhoods and reflected on their experiences of supportive families and communities. For instance, Dan described an almost idyllic childhood expressing gratitude for his adoptive family, who offered love, nurturance, opportunities for exploration and personal growth: "*it was like a great childhood, like just all fond memories of a lot of love and acceptance and maybe being a bit spoiled by our parents"*. He recalled being active, athletic and driven; his family's strong academic focus bolstered high expectations of himself regarding resilience and achievement. Like him, Sue spoke warmly of her upbringing, "*Like, like, I do think I had a great childhood, you know*", and recounted her love for Irish dancing. Isabel spoke of "*always being like, full of energy",* describing herself as a "typical" Irish teenager with a vibrant social life. Lisa reflected on her rural upbringing, painting a picture of being physically strong, healthful and active from the outset. She vividly recounted her social life, sharing her experience of being "present" throughout college.

> *[…] Yeah, very good time in college, and I never drank I never drank, never smoked, never did drugs.* ***I was a good girl****. But I had a very good social life. […] So I went out every night. I had my group of classmates. I had my group of housemates. And then I had my housemate's mates. So I'd go with any group that was going out that night.* ***I was there****. So yeah, yeah.*

Together, all participants emphasised the normality of their life, regardless of circumstances and backgrounds. Yet "*normal*" life was not devoid of challenges. Lisa shared stories of a life filled with many unexpected turns, "*chapters*" opening and closing. Sharon also recounted the ups and downs of her life, though her experiences were generally not as positive as those of others. For her, "*normal"* meant coming from what she described as a *"weird"* family or collection of *"weird"* people, something she became aware of as a child. Unlike that of others, her story conveyed a profound sense of being abandoned and betrayed by others during her youth; she shared painful stories of being let down by family and other adults when she needed them most.

> *It was, it was kind of rough. Because like I didn't, you know, you. If you have a mother like mine, you feel like burden anyway. So to have no place to go and have to ask people can you stay there? Or, you know, it was, it was kind of awful. Like,* ***I didn't feel like belonged anywhere****, you know.*

Feeling rejected and unwanted, she spoke of how this led her to distance herself from home and *"get in trouble*" or act out. She explained the legacy of being labelled a "*bad kid"*: "*So that kind of has stuck with me. They still think I'm 14/15 and I'm still bad. Like, no matter what I do".* Though like others, she spoke of blessing that came albeit at a later point in life, such as becoming a mother, finding the right partner, and graduating college despite the odds: *"I was so proud of myself. I was I was doing everything myself".*

> *He's [son] still, he's still my best thing. I still think he's the best. You know? It's like you, you see what's really important kind of thing […] My son was great. Like, I was so worried, because I know what it was like to have a broken home and everything, you know, but he was he, it didn't seem to faze him.*

Throughout their normality, all participants discussed having experiences of injury or illness in their early life, though accounts suggested their experience was being "*fine*", or otherwise healthy enough. For example, Lisa spoke of childhood digestive issues, which she managed successfully throughout college. Dan spoke of a minor surgery and sprain, though noting an unusually slow healing process. Sue spoke of having grown up in pain due to her rare spinal condition ("*I remember being like five or six years like quite young complaining of pains in me back")*, while Sharon shared growing up with anaemia, at the time referred to as "*tired blood*". However, these ailments seemed to stay in the background of participants' experiential fields; they seemed to simply "*get on*" with things before becoming chronically ill.

[…] but even when I had the frequent bouts of tonsillitis, I was on quite a lot of antibiotics. But it didn't interfere. Like I was still able to go to school, I was still able to go to work. I was still able to go out and do things and I didn't have any like, side effects really […] (Isabel)

5.4.1.2 Life and health coming undone

Participants' narratives conveyed a sense of unravelling as their life stories progressed, with symptoms and illnesses coming to the foreground of their experience. There was a sense that their ordinary way of living started to change, where for some, symptoms accumulated over time, while for others, sudden illness introduced a sharp cut to normality. Dan spoke of how his condition gradually unfolded in school, a particularly painful experience given drive for academic achievement. He shared how initially he became exhausted from the *"littlest of challenges"*, such as walking or cooking, and how this quickly "*took off",* progressing to an inability to participate in any activity.

*And then just every day that went by, **it was just gradually** I'd think twice a day faint three times a day. I'd faint in school. The school would have to send me home and I just yeah. 2017 2018 was when everything just took off. So it was a very, very difficult time […]*

[…] I was fainting like 6 to 8 times a day sleeping, not like, I was either I was in bed or I was unconscious. It was, there was no like getting out of bed, going for walks. I was completely bed bound.

Now in her 50s, Sharon similarly noted that her POTS symptoms likely began to surface during her teenage years, accumulating in the later stages of life. While Sue had experienced pain from an early age, she spoke of how this began to transform into something more overwhelming. She talked of experiencing a rapid change in her symptom profile, unlike the progressive accumulation described by Sharon. Despite being somewhat used to a life in pain, ("*And I can usually deal with muscle pain, or like, achy joints or whatever*"), her new nerve pain was overwhelming to manage ("*that's the worst type of pain*").

***I don't know when**, but I started getting, I started getting like POTS kind of syndrome symptoms, but I don't know when that actually started. But like, I would get really dizzy in places that I have to sit down or whatever. And then I, I know like when I was 18 or 19, it was happening a lot. (Sharon)*

*Yeah. Because it was still quite in just my hands at this point. But then, like, **quick enough kind of spread** to other hips and ankles and most of the joints really, I think I remember at one point, it was just me elbows that weren't sore, everything else was... (Sue)*

Isabel and Lisa, who experienced an acute infection before developing ME, spoke of a sudden onset of symptoms and gradual realisation that they were ill. Their experience was that of an initial interruption to life, followed by an ongoing decline with cycles of remissions and crashes. Isabel explained:

> ***I was fine.*** *Now, I was on the go like 24/7, I was in college every day, I was out most nights or I was up to like 4 am every night. So I had come you know, I thought at the time that my body... it was just really rundown. But I got this, like, awful bout of flu. I remember, it was like, it lasted for about a week. I was so dizzy. I couldn't stand, my vision and my cognitive symptoms were just like, awful at the time. And I didn't know what it was […]*
>
> *[…] And I remember* ***day by day****, I felt like I was getting* ***worse and worse****. And I remember my breaking point was probably the January of 2019.*

Over time, for all participants, life became completely dominated by their illness. They often spoke of how they could no longer ignore the cascade of their symptoms. Sue and Sharon both remarked that everything was "*happening at once*" with their condition. The overwhelming nature was evident in participants' struggle to articulate their symptoms, or to identify the main issue. Sharon, for example, apologised for "*talking too fast*" when trying to recount her symptoms, while Dan struggled to remember all the relevant diagnoses, evident in his hesitation and pauses.

> ***Um. Oh lord.*** *Gastro… slow gastric emptying. So not necessarily gastroparesis, I was told, but it's in that direction […], um, MCAS activation****. So, um, I'm trying to think there's so many****. Um, migraine. Chronic fatigue syndrome. And yeah, that's, I think the main ones.*

5.4.1.3 Denying and minimising the problem

Interestingly, participants often attempted to minimise or deny the weight of their initial and progressing symptoms, before their accumulation became impossible to ignore. Dan's minimisation often came across in his regular use of the word "*just*" to describe his acute symptoms, perhaps in an attempt to derive the simplest explanations. Explaining his tachycardia and frequent fainting, he speculated *"maybe I was just dehydrated from the heat"* and explained his overwhelming fatigue as being *"just excessively tired".* Explaining his MCAS, he corrected, "*Oh, maybe it's just allergies*". At the same time, his account reflected experiencing seizures and fainting as traumatic- something he needed to block out: "*I think it was like so traumatic and it was like go, go, go that like, I think my brain is like, blocked out like every single moment."* His tendency to minimise symptoms

transpired in the interview. Experiencing an increased heart rate from speaking, he remarked "*Yeah. No, I'm used to all the tachycardia and breathlessness, so I'm good to go"*. He later explained,

> *Like I tend to still to this day kind of not dismiss my symptoms, but like **downplay how I feel** because I'm so used to having to downplay how I feel, because I believe that I was over-exaggerating, thinking like if I dial it down and ignore the symptoms, what he (doctor) was saying to do, I'm still kind of getting out of it now […]*

Perhaps seeking to comfort himself, he compared his current state of health to the worst-case scenario, again denying the seriousness of his condition:

> *[…] I'm like healthy. Like I don't have autonomic failure. I don't have like severe epilepsy that I thought like, I'm not going to die. I don't have like anything wrong with the structure of my heart. It's just the rhythm. So they're like, I think there's relief in that regard.*

Sharon also tended to downplay some of her symptoms, for example, negating the impact of her cysts by saying its "*not cancer or anything",* hence signalling that her concerns were not that important. While acknowledging the severity and impact of her current symptoms, she simultaneously appeared to deny her experience of *"just kind of"* suffering. Similarly, Lisa discussed clinging to the belief that her ME was *"no big deal",* being initially convinced that *"this isn't anything, it's a blip",* only later coming to terms with her normal life coming apart: "*and it's only when I say them all out loud that I'm sort of realizing just how bad I am"*

> *I'm just kind of suffering with a lot of muscle pain. And I have been kind of going, it was kind of a weird beginning of it, like it just, it's like muscle and joints and everything. But I didn't have that before I had no pain. And all this started happening at the same time, like at the same time […] (Sharon)*

> *[…] It's like I get maybe three hours in a day where I can actually do anything and then I'm done. You know? So it's, I've now, I feel overwhelmed because everything's kind of adding up and adding up and I'm not, I can't handle it all. (Sharon)*

> *[…] I'm struggling to walk upstairs. I'm no longer running. I'm no longer walking. I'm turning up at work but I'm falling asleep at work. I'm having to pull off on my way home and my 14 minute drive to sleep on the way home before I complete my drive. I'm getting confused at traffic lights, I can no longer read emails that are coming at me at work. […] And suddenly I'm just zombified. (Lisa)*

In addition to minimising their symptoms to themselves, participants also gave accounts of others minimising them for them. Dan, for example, spoke of how his family reacted to his acute seizures by instinctually minimising their severity ("*you're okay", "you're fine*"), perhaps attempting to comfort him. Yet this appeared to invalidate his real experience, in other words, of not being "*fine*". Isabel recalled the weight of symptoms being minimised in an exchange with a nurse, who suggested that "*everyone*" else was dealing with similar issues, stating *"sometimes you need to* ***get up and get on*** *with your life. Everyone has to do it""*

Participants seemed to want to protect others from the reality of their condition. For example, attempting to protect her husband from worry, Lisa spoke of using humour to offset tension following her taking of the contraindicated COVID-19 vaccine, referring to it as *"giving blood"*, and avoiding the topic in the aftermath: "*It's all finished. We don't talk about it. There's nothing to talk about."* Dan spoke of immediately comforting his family following any noise that could imply a seizure: "*[…] like instinctively like I'm fine, I'm awake. I didn't faint",* while Sue spoke of *"staying quiet"* regarding her pain, presumably as an attempt to spare her mother some suffering.

> *[…]* ***it was rough for my mom*** *to deal with as well. So, you know, just like I was young, but it's like, kind of aware that it's not necessarily me, like she felt guilty for whatever reason, some, like her own guilt, and then just me trying to be constantly* ***quiet over it****. It's just, I tried not to... […]*

5.4.1.4 The injustice of becoming chronically ill

Once participants began to acknowledge the weight of their condition, their accounts painted a picture of a cosmic injustice imposed on their lives by chronic illness; they often grappled with the question of why they had been singled out for hardship, having tried to do everything "right". This was perhaps most clearly illustrated by their comparisons of life before and after becoming unwell, seen in their statements about normal life *then*, and life as it is *now.* Taken together, their accounts reflected the unfairness of not having a choice but to live their new reality.

> *I had a lovely, happy, normal life.* ***And then at the age of 30, I, got sick*** *and ended up with a diagnosis of ME. (Lisa)*

> *[…] overall, there's nothing that sticks out that would have been significant in terms of and then traumatic or triggering* ***up until obviously I got sick****. (Isabel)*

Participants often implied the unfairness of having to accept and adapt to the unremitting, fluctuating and often progressive symptoms that characterised their current life. While emotionally exhausting, however, all spoke of learning to tolerate a new baseline of symptoms. For example, Sharon spoke of living with a baseline level of pain, speaking of *"really bad headaches"* as *"nothing unusual",* while Lisa spoke of having to come to terms with a new baseline level of energy: "*I'm still not able to function at the level I was. I'm about less than 50% of where I was […]"*

Reflecting on how her life has changed over time, Isabel spoke of how the meaning of *"feeling good"* and "*normal functioning"* changed drastically between then and now. She explained that with time, she learned to tolerate symptoms that others might experience as unbearable or urgent, stating that these are now "normal" to her. Speaking of her dangerously high heart rate, she commented:

> *Like, I didn't think it was that high compared to what I'd seen before. It was gone to like 180s, but like I had seen it higher. So I was like "**that that's fine**" like.*

A similar desensitisation to severe symptoms was evident in Dan's account, who spoke of his morning ritual in a manner which implied he had resigned to his hypermobility.

> *[…] my morning ritual is like I wake up, I don't move and it's like, what's dislocated and what do I need to put back into place before I get out of bed? And it's always my hips, It's always my shoulders, always my elbows, always my two thumbs […]*

Further highlighting the injustice of becoming ill, participants provided nuanced accounts of their losses, capturing what their condition has taken away. They described loss in terms of practical difficulties and spoke of their experience of diminishing independence, as well as losing connection with themselves and their social worlds. The loss of physical strength and ability was often most perceptible, uncovering other losses such as those of autonomy and identity. For example, Sue spoke of losing grip in her hand, literally, her account implying a loss of control over life due to the uncertainty of her condition.

> *Yeah, my mock exams, I missed the rest of my, missed, like two or three of them. Because **I literally couldn't hold…I had no grip** whatsoever. And my hands were in a lot of pain. So like, even if I could, I couldn't for long. Anyway, it's quite scary. And **especially like, not having any answers** is why we even like straight away or, yeah, yeah, it was, it was weird. Yeah.*

Her story conveyed a sense of unfairness when she spoke of missing out on normal life due to her stays in hospice, "*I felt like it took me a whole summer from me* ", describing it as "*dehumanising in a way".* She also spoke of how she had to become more emotionally mature as a result of her ongoing pain and treatments, conveying an unfair loss of youth when compared to her peers. This was magnified by the fact that clinicians often expected her to understand and cope with her condition like an adult would, adding to her frustration: "*I wasn't an adult like, I didn't understand that in a way*". She reflected:

> *I have like a very small tolerance for people's bullshit. Sorry, I don't know which other way to put that. But I think it's because* ***I used to get so frustrated that I felt like I had to mature quicker*** *cause of my pain as well. So I had a very short... I wouldn't snap at anyone, it was just like, I can't be around them.*

Lisa, who prided herself for once being her doctor's "*healthiest patient"*, spoke of her loss of athletic abilities, which was particularly painful as being strong and active was a major component of her identity. Her account conveyed a sense of injustice and disbelief when she realised that chronic illnesses can and do happen to previously energetic and healthy people. She spoke of her doctor treating her as a "*walking wreck*".

> *I'm saying "But I ran the marathon. I ran the marathon in October" and she's looking at me going "you can't have, based on what I'm seeing […] And I'm looking at it going "what? I did, I was healthy then I was not the person I am now"*

Sharon also spoke of loss in terms of activity, explaining how her pain impacted her ability to express herself artistically. Yet her account also reflected concern of *becoming* a loss due to her condition, particularly when she grappled with the possibility of having developed a terminal illness. The injustice of becoming unwell was further magnified by the fact that she made tremendous efforts over her life to escape from and break familial patters, re-building her life to provide a different upbringing to her son.

> *You know, it didn't... I was worried about how the whole thing, its, I was worried about having cancer, because if I died, my son is going to be an orphan. And he's not even 30 like, that was something that was my like,* ***I can't orphan my son****.*

Emphasising her extroverted and social nature, Isabel spoke at length of her loss of connection, experiencing a feeling of disconnection *"from everything from my body like from absolutely everything".* Her account highlighted that for this reason, some symptoms are easier to tolerate than others, regardless of their severity.

> ***The exhaustion- whatever**, but the feeling of like, being disconnected, the feeling of like, vision being all like spaced out just like it's probably the worst symptoms.*

The loss of a social life was similarly discussed by Dan, whose account illustrated the injustice of having to miss out on key transitions in life. He expressed his frustration that Ehlers-Danlos had "*kind of taken everything away*" and talked of *"feeling very left out of like the social aspect of like becoming a young adult",* constantly being *"just in bed"* due to his symptoms. He provided a painful account of how his healthy brother served as a sort of reminder for his own lost ability. Growing increasingly reliant on his brother's support, Dan' narrative also conveyed that chronic illness had taken away his personal dignity, and how he felt he had negatively impacted his family.

> *But it's always like a bit difficult to like, you know, like seeing like my brother cooking dinner for the family. But my version of cooking dinner is like, I'll have to sit down in my stroller or like, sit in like a stool to like, make something really quick because, you know, if I make dinner for everyone, I'm not going to be able to eat dinner because I'm going to be so exhausted.*
>
> *So my twin brother, if I had to go to the bathroom, he'd come to the bathroom with me and stand outside the stall because, you know, I was fainting so much.*
>
> *[...]* ***and I always feel so bad*** *that like, I almost feel* ***like my brother has like this PTSD*** *aspect of like, while I might not even be home and he'll hear a noise and he'll think I'm after fainting even though I'm not home. So like, everyone is so like ingrained in like being hyper aware of how I feel, what my symptoms are that like, it just upsets me sometimes that like, sometimes my brother doesn't want to leave the house because I'm going to be home alone and he feels like it'll affect me.*

All participants indicated feeling like a burden to their families, implying the loss of another "normality" due to becoming chronically ill. For some, there was a strong sense that their close ones were heavily invested – both practically and emotionally- in their health and healthcare journeys. For example, Sharon spoke of how her current symptoms affected her "poor" family, while Lisa recalled the emotional impact of her severe condition on her husband, who was *"absolutely petrified"*.

> *Yeah, so, and like if I do anything, I'm out. Like, I'm laying on the couch and I'm miserable. I have hot water bottles all over my body. My, my poor boys, my husband and my son are like," what can we do? Like you don't even know?" And I'm like, "Oh, no." So I just feel like* ***I feel like a burden*** *on people, you know? (Sharon)*

5.4.2 GET 2. The stable and changing self

The interplay of stability and change in their sense of self emerged as a central theme in participants' narratives. Many reflected deeply on their identity, often contrasting who they were before the illness and the aspects of themselves that remained intact despite their health challenges. Participants spoke of their determination to maintain a sense of authenticity and individuality, highlighting the tension between feeling different because of their illness and striving to stay true to their core selves.

5.4.2.1 Being, feeling and becoming different

All participants described being and feeling in some way different from others, whether due to their personality, interests or their chronic illness. Dan and Sue spoke of being and feeling different from early on due to the inherited nature of their condition, which manifested in various ways throughout their childhoods. Isabel, on the other hand, emphasised that she was like other teenagers, feeling that her condition forced her to become different. Lisa emphasised being different due to her personality, while Sharon spoke of her long-standing feeling of being "other", which was magnified in her healthcare journey when clinicians referred to her results as *"unusual".*

Experiences of being different varied in emotional tone, for some participants being a source of pride while for others, embarrassment and shame. Dan spoke of feeling different in a positive way due to his hypermobility, having always been told he was "*super double jointed"* and doing all the *"party tricks".* He later explained that this difference later impacted on his recovery from a surgery or physical activity; he was different from a "*normal person*".

By contrast, Sue also described a feeling of not "fitting in" but expressed shame in being different, or "just being" the way she is. She highlighted how her physicality differed from others, including the researcher, but at the same time gave painful accounts of the impact of being "seen" as different. Her discomfort and shame of being unusual was clear in her visible upset when recounting having to use a medical brace, a visible signal of her issues.

> *So like, if me and you stand up and compare, I'm more, a lot more hunched, or more curved, than natural, right. So I guess that was caused me pain the whole time.*
>
> *[…] I was, like, really embarrassed to wear it, but I always found the horrible because the moulded one, it was like it was rock hard […] And because it was against something hard I was just felt like I was hurting even more. So it was looked really ugly as well. So I was really embarrassed. It was like, a fleshy kind of colour. So it was just like, it's just disgusting.*

Her sense of feeling different was amplified through her experiences with healthcare, particularly when she recalled being a child utilising adult rheumatology services.

> *[…] oh, you know, the guys in the waiting room, I remember it was men and women sitting around me in their six, seventies or older. And like, I was walking into like a transition year at the time. I was like, I should be making stuff with clay right now. And these people are like, like, really old. So that kind of messed me up. Like mentally a bit.*

Unsurprisingly, having emphasised being a "typical" teenager, Isabel strongly rejected the possibility of becoming or being seen as different due to her chronic illness. She recalled her frustration at having to use a wheelchair, and later walker, emphasising that there was "nothing wrong" with her legs. Like Sue, she felt embarrassed at the prospect of being signalled out as different from her school peers.

> *I came home using a walker. So not great. And for me that was most embarrassing thing because I was like, I'm 20 Like, why do I have this geriatric walker? So yeah, I transferred over to crutches then because I thought crutches looked a little bit better than the walker. But either way, I was still embarrassed to use them.*

The stories shared by Lisa often captured a sense of pride in being different; she spoke of her independence, resourcefulness and strong sense of justice; called herself "unconventional", always marching to the beat of her own drum. These attributes permeated her life and set her apart from others in personal and work contexts, though a sense of being unique was most evident in her interactions with healthcare. She spoke of how her clinicians remarked on her unusual emotional resilience:

> *On an emotional level…yeah, she is fascinated by me as an individual. Because she keeps saying to me, "you have to have a breaking point" and I'm saying "I don't think I do". Yeah, she is absolutely astounded that mentally I'm not floored […]*

Sharon also spoke of feeling like she did not belong or fit in with others. While being different was at times highlighted as her strength, she also shared painful accounts of being treated as "other" by her family. She highlighted this in her comparisons of herself to her chaotic siblings and mother, who acted "out of control". Not fitting her family mould, however, often translated to being unwanted or feeling like she was "bad": "*I'm kind of like my mother's scapegoat. Like everything's my fault.* ***I'm terrible.*** *I don't do anything right".*

5.4.2.2 Staying authentic, resilient and headstrong

When telling their stories, participants often painted a picture of their "real" or authentic self and illustrated how their personalities impacted on and manifested themselves in their healthcare and illness experience. Participants often referred to themselves as "*stubborn*" or "*defiant*" and emphasised their determination to overcome life's hardships; their accounts reflecting that they did not want to be seen as vulnerable or weak. For instance, Lisa often spoke of her defiant nature, giving examples of determination and self-assurance in work, sport and her relationships with others. She illustrated her stubbornness: *"And I'm a fighter by nature. So I'm just fighting, making all the wrong decisions in my fight".* Her rejection of the sick identity is evident in her accounts of refusing to be seen or to be treated as "*broken*", and in her refusal to allow her condition to determine her identity, evident in her consideration of "*ME as separate from me*".

> *[…] and I am stubborn, I am planting these (flowers) today, I don't care how sick I am, they are going in the ground, I'm sick of being sick. It's like repeat all over again 10 years ago. So I go out to plant them. And I know I have this massive chest pain, and I can catch my breath and I nearly collapse. So I come back inside.*

Similarly, Dan spoke of pushing through symptoms until he was no longer able to do this. He attributed this attitude to his upbringing, noting that he had to be very ill or "dying" to take time off school.

> *I think it was always from growing up, you know, I was I'm still to this day very stubborn with my health. I find that if I get it's like unless I'm on death's door, I won't do anything about it because I think growing up, having health complications being dismissed so much, I'm kind of just like it'll pass*

Isabel reflected on her defiance with regards to dismissing her clinicians' advice to rest following an infection. Demonstrating personal insight and perhaps a feeling of regret, she commented on how this tendency to push through was not the best decision; likely contributing to her poor recovery, and later development of ME/CFS. Like Lisa, there was a sense that she wanted to be seen as strong and resilient by others. Speaking of a difficult clinician encounter, she recalled hiding her vulnerability , *"obviously I went to the bathroom and cried afterwards".*

> *And then as she did the blood, she said, you're best to go home and I said, "Oh, I'm going into town", like to meet my boyfriend, like I was living my best life. And she was like, "trust me, you're gonna want to go home".* ***I didn't go home, obviously****.*

For Sue, her determination was evident in her accounts of reaching out for help, and fighting for herself, utilising the supports that were available: *"anytime I've done it, it's been of my own accord".* For Sharon, her sense of resilience and determination to fight through was evident in her accounts of breaking away from a difficult family past, looking to rebuild her life and start fresh in different contexts. Her tendency to push through was also evident in her accounts of being unable to rest and of overextending herself despite her injuries or symptoms, illustrated by her account of being post-surgery. When asked if she took time to recover, she said:

> *No, I, Okay. I can't like sit still. So I was sitting there trying to recuperate and I was looking at my wall that had fake stones on it and thinking how much I really hate this fake stone. So ended up (laughing) a little chisel, you know, so I was I ended up taking off all the stones on the wall like*

5.4.3 GET 3. Relating to and experiencing the body

When speaking of their life and experience of becoming chronically ill, all participants made references to their body, giving the impression that it was separate from the self. Their narratives captured different ways in which they perceived and related to the physical body, sometimes as passive, an object that "gets in the way", and at others as an agent, or an entity that "acts out" and makes demands. Speaking of their experiences, participants also referred to control over the body, namely feeling powerlessness, and described how their relationship to their body has changed, moving from fighting *against* it to working *with* it.

5.4.3.1 The body as an object or agent

Unsurprisingly, participants' accounts often painted a picture of their symptomatic or malfunctioning body as an obstacle; a broken object to be fixed. Their narratives often conveyed an experience of the body being in the way, preventing them from performing normal day-to-day activities. For example, Sharon, referring to her chronic pain, spoke of how her body prevented her creating art; something that was both her livelihood and defining aspect of her "*whole*" identity. Her accounts often implied a sense of being stuck and fragmented – both in terms of her career progression in Ireland, and in terms of living a body that doesn't fully "*work*". Similarly, Lisa described how her body was broken and refused to "work" as it should, leading to failures at work and tensions with colleagues.

> *And I, I was kind of just feeling lost and stuck. And then then this happened, like, my, **my hands don't work**. They're not doing what I want them to do. So I'm not doing anything. (Sharon)*

> *So he's standing in front of me absolutely raging, because there's managers on his back, because this project has been stalled.[…] And so he's at my desk. He's got steam coming out of his ears. He is screaming at me. I'm laughing, I'm just sitting there laughing. It's so, you know, it's like this **cogs in your brain just don't work** anymore. You're taking in the information and you're hearing it, which you just don't understand how serious it is. It's really hard to explain what it's like. (Lisa)*

Interestingly, the language used by participants sometimes suggested a sort of separation between self and body, indicating that it was an object. For example, Dan spoke at length of how "*it*", the body destabilised by EDS and POTS, prevented him from

fulfilling his passion for gymnastics; while Isabel spoke of how "*it*" was making it impossible for her to work.

> *My body physically couldn't keep working even from home.* ***It was,*** *by the end of every shift****. I was*** *literally like keeled over like ready to puke. (Isabel)*

An objectification of the body was also illustrated in dialogues with healthcare practitioners, which often focused on removing or fixing a "bad" or malfunctioning part. Sharon recalled,

> *He was like, "Don't worry about it." He was like," I think we're gonna need* ***to take the ovaries out****". And he's like, "That way. You can't have ovarian cancer. Like, if you don't have ovaries, you're not gonna have cancer."*

There was also a sense that the body became an obstacle through its loss of transparency and demand to be seen. Sue explained how she initially attempted to ignore her symptoms and body (*"always been really ignorant towards it"),* in this case hand pain, but could no longer do this ("*they look like I broke both my hands. Like there were huge, like swollen")* . Sharon also alluded to the loss of transparency, discussing becoming more aware of her physicality through illness.

> *[…] and now I'm like,* ***I didn't even know your bones can hurt****, but like your bones can hurt. Like, I didn't know that. I thought that was muscle, you know? So he did like, he did the pressure point thing on me and stuff.*

Participants' accounts also conveyed their experience of the body as an animate entity, at times troublesome or demanding. For example, they spoke of how their body essentially said "no", forcing them to listen and accept its limits. They described how their body responded by shutting down, demanding that a "price" be paid for overactivity. For instance, Isabel and Lisa described themselves being *"floored"* and unable to keep their eyes open having pushed their body to the limit. There was also a sense that participants were at the mercy of their body, implying its punitive rather than passive role. A sense of betrayal was evident in the account of Dan, who spoke of pleading with his body to stop acting out.

> *I think it was like a month after that I noticed that like when I'd walk up the stairs I was having tachycardia and I was like, "****Please, no, please don't be doing this again****." And then I noticed that like everything that used to be easier was gradually getting harder again, like before. And I was like, Here we go again.*

5.4.3.2 Being powerless over the body

All participants' narratives reflected the experience of losing control over their body, which in turn resulted in a feeling of being dominated by their illness. The powerlessness accompanying this was particularly evident in Dan's stories, which conveyed loss of control through loss of consciousness due to seizures. Although Dan could predict pre-syncope, speaking of a precedent "*feeling of unsafety and dread*", the outcome was in essence a complete loss of control, resulting in a disconnect between the world and himself.

> *[…] So I remember waking up and the ambulance were there and I could like hear I like remember being able to hear things, but like I was so weak and like still so out of it. Like I wasn't able to like, respond and I could hear them like shaking me like, "no, you can wake up, wake up for me." And I was just like in my brain, I was like," I'm here, I'm awake, I'm awake". But like **I couldn't** communicate.*

Participants also provided examples of losing control over *themselves*; Lisa, for example, recalled her behaviour in a state of fever-induced delirium.

> *[…] I basically just made an absolute **fool of myself.** I always remember lying across his (doctor) examination bed telling him I was just going to go and sleep like as if it was my bedroom and "Go away. Leave me alone". And the poor man was just standing there at his doorway, he didn't come near me. He's just looking at this weirdo going "When is she going to leave my room?" (laughing)*

> *[…] On this particular day, I remember waiting for two handbags to come from America. And the courier arrived with them. And in front of the courier, and I always remember it. It's one of those experiences where you know you're doing it **but you can't stop yourself,** but you need to stop yourself. In front of him. I proceeded to open up the bags, pull them out, put them around my neck and start swinging them, showing him these wonderful bags" (laughing)*

All participants spoke of attempting to re-gain control of their bodies and struggling or failing to do so. Isabel, for example, experienced her body as resisting change, while she made attempts to exert influence over it, unsuccessfully. This was exemplified by her relationship with eating and weight, ranging from an inability to eat and maintain weight, to managing her body's increased demands for food while trying to lose weight, a struggle attributable to insulin resistance. At different points in her narrative, she recalled:

*And I remember that whole day, I literally felt like I was going to collapse. Like **I couldn't** take in any food.*

*So I was rapidly gaining weight in that timeframe, as it says there (life grid). Which I found, my symptoms at that time, which were the worst, where **I had to** eat every hour, **or** I was literally shaking.*

While participants often engaged in lengthy periods of pushing back against their symptoms, they spoke of eventually coming to a realisation that they could re-gain control by working *with* rather than *against* their bodies. This was illustrated by their narratives of having reached a limit and then trying to "*do everything right*". Sharon spoke of reaching her limit of coping and then seeking help, while Dan and Lisa often referred to following guidance, despite at times being disappointed by unpredictable results of this attempt at staying in control.

And she (doctor) said, like, "anybody with oestrogen that high would have high anxiety." She said, "I don't know how you're coping". And I said," I'm not". (Sharon)

But I kept like implementing my like strategies again of like nutrition, making sure I was sleeping every single day, didn't drink a single ounce of caffeine, did everything to like help myself. It just got worse and worse. (Dan)

Over time, participants appeared to develop a different way of relating to their bodies, becoming more protective of them, particularly in the context of the COVID-19 pandemic. For example, Lisa described becoming "*militant*", having to draw the line and discipline *herself* in relation to pacing, thereby protecting her body from herself, while Sue spoke of how she made a conscious effort not to aggravate her symptoms through infection.

Yeah, I was like, I wasn't one of these likes, germ freaks or anything. I was just afraid to catch and COVID in case it made me worse while I was already in a flare or something. (Sue)

5.4.4 GET 4. Making sense of symptoms

A significant aspect of participants' narratives was their effort to make sense of their symptoms, a process often described as lengthy, isolating, and exhausting. By piecing together personal timelines, they recounted how they gradually began to connect their accumulating symptoms, seeking to identify potential triggers or factors that could explain progression of their condition. Narratives revealed how diagnosis, even if tentative, played a key role in their personal journey to understanding and enabled them to legitimise their experiences.

5.4.4.1 Looking for pieces, patterns and triggers

All participants expressed frustration related to "not knowing" what was wrong, or what was causing their symptoms, and described their efforts at piecing together the puzzle that was their illness. Living in uncertainty was experienced as distressing, and when combined with the lack of explanations from clinicians, motivated participants to explore various avenues, searching for clues that would enable them to develop "a theory". At times, an inability to make sense of their experience led participants to question their own judgments, and ultimately, the validity of their illness; "*maybe like there's nothing wrong with me and I'm just crazy" (Dan).*

> ***As I say, you're dealing with all of these new symptoms you've never had to deal with before; you're getting questions off colleagues asking you what is ME, and you can't even give a proper answer. You're getting questions from family, especially my parents, "when will you be better" you know, you're then wondering, "will I get better?" (Lisa)***

A key aspect of the path to understanding was personal research. For instance, Sharon, who had always been independent, spoke of her intense attempts at "figuring it out" alone by reading and learning as much as she could, trying to find connections between her symptoms: " *I'm just like, "No, I'm just hyper focusing, and I want to understand it". Like, I have to understand that, everything, you know […]".*

In addition to looking for clues independently, some participants attempted to make sense of their conditions with others, for example by speaking to family members who had witnessed the progression of their disparate symptoms over time.

Dan spoke of how he learned about Ehlers-Danlos Syndrome through online communities, and later made sense of his symptoms by speaking with his mother, like Sue in the context of her ADHD.

> *The slow wound healing was like the red light bulb. And I was like, "Mom, remember appendicitis? You always told me that my wound wouldn't heal." And she was like, "Oh, yeah". So I was kind of like, I think it's Ehlers-Danlos syndrome (Dan)*

Participants' accounts also suggested a need for a point of comparison against which they could validate or make sense of their symptoms. All participants attempted to understand their conditions by reflecting on the experiences of others. Such comparisons were at times confusing, particularly when symptoms did not align; or were helpful, enabling participants to grasp the implications of their illness. Isabel discussed how, when trying to understand her experiences, she struggled to explain her exhaustion and feelings of being disconnected from reality. By comparing her symptoms to others, she started to recognise that her condition might be more complex than she thought initially, leading her to seek further answers. For Lisa, recalling another person's experience of ME and relapse led to a realisation that she too might be living with a chronic and possibly disabling condition.

> *Because when I was comparing my symptoms to other patients, I was like, "they don't have these cognitive symptoms". Like they don't like they, they don't have this absolute, like, sheer utter exhaustion, like I was comparing the symptoms to people I have met, who just, their, that was their primary diagnosis. (Isabel)*

> *And then what happened was, I remembered a girl that my sister went to school with, she got ME, and she still lives in our hometown. And so my mom knows her parents. So there's conversations there.* ***And then the picture becomes clearer.*** *This girl is never, she only ever got to like 80% of where she was, she has what's called relapses.* ***I'm learning that****. (Lisa)*

At other times, knowledge derived from such comparisons was inaccurate or misleading, adding confusion to participants' sensemaking journeys. Attempting to understand his tachycardia and syncope, Dan found comfort in knowing of others with similar experiences, which initially gave him false hope that he would outgrow what was then assumed to be a *"teenage"* condition.

> *So he (friend) was like, "Oh, you'll grow out of it" because he grew out of it. That's all he knew. So I kind of was in this mindset of, "Oh, I'm just where he was and then I'm going to get to where he is now".*

Having attempted to compare her experiences of pain to those of others, Sue commented on the impossibility of understanding chronic illness in this manner. Her narrative highlighted the subjectivity of experience:

> *Because like, I understood very quickly that everyone experiences it differently. Could be 10 people and all experiencing differently. So you can't really take other people's stories for it. So. Yeah, it was just getting to know me, body, really?*

During their sensemaking journey, participants often sought to identify specific triggers for their condition, reflecting on patterns throughout their life course. For example, Isabel and Lisa discussed how over time, they were able to trace the origin of their ME to an early infection, also explaining the reactivation of their viral symptoms post-vaccine.

> *I actually figured out how I got it. And it was actually from a, I was in business class with a friend. And we always used to share water bottles. And I only found out once afterwards, that he actually was positive […] (Isabel)*

> *Yeah. And then what transpired, now this is that what I've been told in hindsight, is that the swine flu damaged my system. And that when I got sick in 2011, that my body couldn't handle that virus. And that's what led to my ME. That's, that's the thought process now as to as to what happened. (Lisa)*

Participants often struggled to make sense of the fluctuations of their symptoms, which varied in impact from minimal to devastating. Thus, sensemaking required them to become attuned to patterns in their condition, observing and making connections between events surrounding flares and remissions. Sue, for example, spoke of learning about the relationship between her lifestyle, stress and her pain. Isabel and Lisa spoke of how over time, they became better at monitoring and predicting their symptoms, demonstrating a better "*knowing*" of their own body.

Often referring to a "*boom and bust*" cycle, Lisa explained how she developed an understanding of the *"crashes"* she experienced over time. Giving examples from multiple phases of her life, she spoke of slowly coming to recognise patterns of symptoms, learning that *"exercise is definitely making it worse".*

> ***And I can tell, obviously**, I don't feel well for the month, but I can tell the week before I'm getting this, because the symptoms are, its literally my brain just goes completely, like mush. (Isabel)*

> *At this stage, I obviously know more about my body now than I did 10 years ago. And I randomly track my heart rate myself […] if my heart rate is in the high 90s, that's kind of* ***the sign that I've been pushing things*** *and I need to be really careful. I'm on the verge of a crash. (Lisa)*

Participants also spoke of developing a deeper understanding of the relationship between the body and mind as part of their sensemaking journey. Ensuring that this was well-understood, all strongly emphasised that their conditions were *not* psychogenic in nature, but instead, had impacts on their mental health.

> *Ah, I think the mental aspect of it, it's having a physical I know it's an invisible illness, but having a physical thing. I think people don't realise how much it affects your mental health as well (Sue)*

> *No one with these kind of issues and these kind of stories would be perfectly healthy mentally. There's always there's always going to be an aspect of like anxiety, depression, feeling down like stress. It's, it's going to be there. (Dan)*

Again demonstrating an acute awareness of the patterns of her symptoms, as well as her needs, Lisa commented on her affective and cognitive abilities during a crash.

> *Now, when I would you know, when I'm at my worst in the crash, I mean, I'm mentally gone as well. I am like, don't throw any sort of emotional problem at me because I can't handle that mental problem at me. I can't handle that, I am just, leave me alone in my bedroom for 10 days.*

5.4.4.2 Connecting the dots through diagnosis

A crucial stage in participants' sense-making journeys was that of receiving a diagnosis, even if tentative, which could help them make sense of their symptoms. For example, Isabel spoke of how she came to understand the relationship between her hormones and intensifying symptoms, speaking of the interaction with hypermobility syndrome, following a diagnosis of PCOS. She spoke of the immense validation and relief provided by a diagnosis of a physical, organic illness, and the impact this had on her relationship with her family, explaining with disappointment that until diagnosis *"Nobody really believed me, including my mom."*

> *[…] He validated this, he said, "Oh, yeah, definitely you have this". And because of that my whole family kind of had this outlook of "oh my god, like she's actually not well", though, everyone believed me from that point onwards.*

Receiving a diagnosis of dyslexia and ADHD was a source of relief for Sue in that it helped explain some of her previous and current symptoms. There was a sense that such validation gave her relief from self-blame, enabling her to become more compassionate towards herself, "*Like, it's not my fault that I spell things wrong."*

> *[…] because I like growing up, put everything down to brain fog. I can't remember that. That's brain fog. My spelling was bad. Oh, that's brain fog ...which was actually dyslexia the whole time.*

Similarly, Dan, spoke of the relief when his symptoms were explained by something organic, and how a diagnosis of EDS helped him understand the fluctuations in his symptoms ("*it can like get worse and then get better and then get worse again")*. He spoke of becoming more compassionate towards himself after learning that the origin of his condition was outside of his control. He also spoke of becoming stronger through the journey of sense-making, learning to advocate and stand up for himself in other aspects of life.

> *[…] knowing that like this isn't something that like, I brought on myself, this isn't my fault. Like these are just the cards I've been dealt and like, especially like the, like knowing that it, like it is a genetic underlying factor, like being able to be told like you can't give yourself a genetic mutation […]*

5.4.5 GET 5. Needing others on the journey

The role of the social environment in lived experiences of chronic illness was another key theme. Participants' stories generally conveyed a deep sense of loneliness as they tried to make sense of their changing lives and symptoms. Despite their tendency to "*push through"* everything alone, narratives reflected a longing for understanding, recognition and support from others.

5.4.5.1 Loneliness and longing for support

Participants spoke of needing support from family, friends, healthcare professionals and the community, and there was a sense that in the absence of diagnosis or cure, they simply wished for others to validate their experience and accompany them on their journey. For example, Sharon discussed how she got through problems in life on her own, yet her account conveyed a sense of helplessness: *"I can't do it all. And I don't like asking people for help. So I'm kind of just don't know, it sucks".*

A longing for the understanding and care of others was reflected in participants' gratitude for having supportive friends and family members, as well as accommodating work or school environments. Dan described being lucky to have a supportive friend group, who validated his experiences. Sue spoke of how assistance and emotional support was crucial for navigating healthcare challenges, particularly when underage. She expressed gratitude for accommodations and supports received in school, following an absence due to her hospice stay.

> *And then I was only missing one week of school, but like the school would understand that? You know, the teachers were just like, "yeah, do this while you're there", you know, they were pretty nice. I didn't have to do too much schoolwork, although thankfully. (Sue)*

> *[…] my friends would sit with me at lunch if I fainted. They'd stay with me. They'd go get help. So they were all so supportive, which was so great, you know, They were like, "You're not crazy". (Dan)*

In contrast, Isabel discussed receiving competing messages in school. An initial experience of support and empathy from her teachers when she was unwell, paired with a lack of understanding and pressure to perform.

> *They were kinda, now they did say, "oh, gosh, that's a awful one". But they were also kinda like, "you're gonna fail the leaving cert because you've been out for so long" […] so they were quite good when I was in the hospital. But when I came out, it was like, "Okay, it's leaving cert now"*

Similarly, Lisa spoke warmly of the support received from her employer, giving the impression that this was an unusual or unexpected kindness. She described how with the progression of her ME she became an inconvenience at work, making serious financial mistakes and causing delays in her company. She described her colleagues and manager as "*very understanding*", though her account portrayed that this was largely a matter of politeness.

> *And my colleagues are still **so lovely** to me, even though they know I'm sick. And they know I have a diagnosis at this point. They know I've been to hospital. They're incredibly supportive, you know, on that side of things, to be fair to them**. But, they're glad to see the back of me**.*

Participants also spoke of their desire for support from healthcare professionals, particularly in the absence of a diagnosis or cure. Generally, their fantasies involved simply having someone who would listen or accompany them on their journey; "*say, "sit down, tell your story"" (Isabel), or "help you navigate like being chronically ill" (Dan).* This need was implied in Sue's account, as she spoke of being let down by counsellors, leaving her feeling like she was beyond help due to her pain.

> *And then I went down and they told me they couldn't help me. They told me I needed pain management, which I do agree with. But at the time I was like, **"I just need to talk to someone",** just constantly getting frustrated about the pain, everything frustrated me. So then it was like, I'm going to actual counsellors and **they're telling me that they can't help me** […]*

Other participants spoke of the support which was, or could be, provided by mental health professionals, though their accounts conveyed hesitation and worry that this could be misinterpreted as implying they had a primary mental health condition.

Isabel shared her ambivalence, emphasising a need for specialist mental health supports, but also noting her reluctance to seek counselling due to previous experiences with healthcare.

> *Yeah, mental health professions are important, but I do think there needs to be designated specific ones for chronic illnesses, because the general run of the mill counsellors I've heard don't really have an open mindset when it comes to chronic illness.*
>
> *[…] And so I know, for me, I think that's probably one of the reasons why I haven't gone to the mental health professional yet is because of the kinds of trauma that the doctor has left me with in regards to mental health.*

5.4.5.2 Relating to others who understand

Participants often spoke of the importance of connecting with others who have a shared understanding of what it is like to live with a chronic illness. Their stories reflected a desire for belonging; they spoke of the emotional comfort derived from connecting with patient communities. They also shared examples of practical benefits gleaned from communities, such as learning about specific symptoms and their connections, treatments, and gaining information regarding services and healthcare providers. Dan highlighted how online communities were instrumental to him achieving a diagnosis, and his account conveyed a being truly understood. Similarly, Lisa emphasised the importance of online communities in her journey, highlighting that others' lived experiences, including her own, can be a valuable source of information. She described being comforted by the ME community, particularly given the lack of understanding and support she had experienced from friends and family. Again, there was a sense of being truly understood by others due to their shared experiences.

> *And then also having people who like understood how I felt because as much as I could tell my parents tell my brother, tell my boyfriend, my friends how I felt, they'd never really experienced it […] (Dan)*
>
> *So, because nobody in my family fully understands it, I'm also keeping a blog alongside of all of this. So I'm trying to educate people from my own learnings about my own body, what my symptoms are, what makes me worse. And it's kind of a learning curve tutorial for myself also. So it's kind of like, you know, catapulting myself into understanding what's going on[…] (Lisa)*

5.4.6 GET 6. Interacting with a broken system

A key theme in participants' experience of becoming and being chronically ill was their interaction with the healthcare system over time. The system was perceived as both stable and dynamic, and participants' accounts of their healthcare experiences painted a picture of an ongoing struggle; a "blur" or referrals and delays, going in circles and getting nowhere. Their accounts revealed an experience of being abandoned to navigate the uncertainty of their conditions alone, highlighting a healthcare system fundamentally unfit for purpose.

5.4.6.1 Fighting against an army

All accounts vividly conveyed the patient journey as a lengthy, frustrating, and exhausting battle for care. Participants regularly described having to *"push"* and *"fight"* against healthcare, feeling that they were going up against an *"army"* in a bid to be taken seriously. Together, their narratives captured a clear sense of 'us' against 'them', and a sense of unfairness in having to fight the system whilst being chronically ill.

> *[…] I felt like **everyone turned on me**, then that's how I felt at the time. Because. They were all defending her (physiotherapist). So that was pretty frustrating. (Sue)*

> *So I was like, "Well, you didn't test my iron or anything?" […] And he's like, "this isn't (home country), so **you don't get to just get tested** for anything you want". (Sharon)*

Each step of the journey appeared to be going uphill in the sense that participants described having to *"beg"* for appointments, referrals and explanations. For instance, recalling an exchange with her consultant, Lisa described having to *"fight"* from her hospital bed to see a specialist in ME.

> *[…]And I'm looking at him going "no, but I want to go to number two, because you've already told me you don't know enough about this. If anything, you've told me you're not the expert. I want to see the expert." **I have to fight** with him on my hospital bed **to get a referral** to a doctor, he originally told me he was going to refer me to! So at the end, or at the end, yeah, he wasn't happy.*

Over time, most began to anticipate and prepare themselves for some kind of conflict on their journey. Some discussed their efforts at keeping meticulous records of their symptoms and medical histories, implying this was a defence against attacks of invalidation and dismissal. The emotional toll of this battle was palpable in participants'

accounts. Many described exhaustion of constantly having to fight for care while simultaneously dealing with their illness. Isabel spoke of her journey, conveying exhaustion, disappointment and giving an impression of her needing to protect herself from the system. As illustrated in the excerpts below, she experienced her healthcare journey as traumatic, more so than becoming chronically ill or disabled, and later expressed that it was unsurprising that many patients with poorly understood conditions die by suicide.

> *Because before that, it was just me saying, "I don't feel well", like, whereas since gone to him, I was able to properly like, articulate my feelings, my symptoms, I was able to* ***prepare in advance****. […] I, every appointment I go to now I have a list of my symptoms in categorical order.*
>
> *[…] Because when you go through a journey, and when you look back at your journey, and everything that you've been through, and* ***it's not just simple****, you don't go to doctors, and they say, "Oh, this is what's wrong with you here, take this".* ***You're fighting.*** *And when* ***you're ill and fighting****, you physically can't do it. So you're fighting the healthcare system, when you're not feeling well. So it's basically you up against this massive army.*
>
> *[…] I just think that the whole process as well in general, it's just painful and it's quite traumatic. And I feel like a lot of people that have chronic illnesses need like extensive counselling, I probably need to go (laughs) but, you do need a lot of counselling after just the journey process, never mind actually having to live with the condition.*

An unfairness was also implied by Sharon, who spoke of having to push herself to attend appointments, an unrecognised struggle of patients with invisible illnesses.

> *Like, I'm lucky if I can get up, but that's the other thing. To like keep going into the GP, like, I'm like dragging myself like to get in the car and go to you. It takes everything the rest of my day like, I'm done.*

5.4.6.2 Going in circles, towards nothing

Participants' language strongly captured the percept of a system which fails to achieve outcomes. Participants frequently described their healthcare journeys as cyclical and stagnant, leaving them feeling like objects, both pushed around by and trapped in a broken system. Their accounts captured a sense of them of being "*bounced*" around between specialists and their GPs, giving the impression that they were constantly going in circles between doctors yet getting nowhere. Participants spoke of being "*pawned off on people*", giving the impression that clinicians were simply trying to get rid of them by referring them back into the same system, with little communication or follow-up. Isabel,

for instance, spoke of feeling "*like a Yo-Yo*", capturing the poor coordination of her care, along with a sense of defeat about the possibility of change. It appeared that the endless cycles of referrals created an illusion of movement; perceived as an incredibly wasteful and draining process which requires patients to undergo the same tests, repeatedly explain their symptoms, with little progress beyond this.

> *I think at the moment, and, I just wish all the doctors were in one place, because it's frustrating trying to like when you're under five or six different consultants,* ***and it's hard to look at one and be like, "Okay, this doctor said this" and going back and forth****. You're like a Yo Yo, you know, […] I haven't seen reports and I just wish they could all sit down and have a chat together. Obviously they can't, I get that.* ***It would just make life easier.*** *(Sharon)*

> *[…] I was just kind of, sent on the way really, okay, there was no like referral back to [hospital] or anything to see how I got on there or anything. I was just,* ***if I had a problem, back to my GP,*** *it was just kind of felt like I was kind of* ***going around in the circle****. (Sue)*

In addition to their experience of going nowhere, participants expressed frustration and disappointment, concluding that "nothing" happens or can be done, particularly for patients with chronic, multi-systemic and poorly understood illnesses. As expressed by Sharon, *"But there's not anything here when you have a thing."* This nothingness referred to a lack of options regarding medication, referrals and treatment.

Participants' narratives also placed the GP in the centre of this system, sometimes creating a sense that they were in an omnipotent position, deciding on all the steps of the journey. Yet, highlighting systemic barriers, participants' accounts generally acknowledged that "*the GP can only do so much*", and how in certain cases, the doctor's hands are tied. This is frustrating for both clinicians and healthcare providers, though as expressed by Lisa, can lead the patient to feel like they "*just have to suck everything up".*

> *[…] There was not really much more she could do because I wasn't like a simple like low blood pressure, high heart rate, take some sodium, take some beta blockers. You know, when I go into her with like these complex, like severe chest pain and like migraine, she was like,* ***"I can't, I can't do anything. Like I don't know what to do****" (Dan)*

> *But there's no overall treatment that she can offer me and she's honest about that. And she's empathetic, you know, she's agreeing that she doesn't have all the answers. She's agreeing.* ***It's incredibly frustrating. But there's nothing she can do for me****. (Lisa)*

Participants' accounts also illustrated how navigating the healthcare system is "*impossible".* They commented on how they spend most of their time being on waiting lists, with "*not a whole lot happening*", highlighting that delays can pose major risks to patients, aptly captured by Lisa's statement, "*well in three weeks, you could be bloody dead".* Sharon spoke of the endless cycle of waiting lists, expressing frustration at having wasted time and money during her journey. Like the others, her account conveyed exhaustion with navigating the broken system, having eventually given up on jumping through hoops to receive care.

> *[…] it's impossible here to get help as an adult.* ***It's impossible****. And it's very expensive. If you even try to go for that. It's like 1000 euros to get properly assessed, the way they, the way they have the assessment you have. […] But you have to go to psychiatrists get assessed again, get you know, like, it's and then they have to go to like group therapy like,* ***you have to jump through all these hoops****. And I'm just like, I've lived this long with it.* ***I'm just gonna live more like that****.*

Participants' experiences of a broken system were also evident in their comparisons of public and private healthcare, which revealed their expectations about quality of care. Generally, they perceived the public system as chaotic and unreliable, and their narratives implied that by paying for private services, they were more likely to receive appropriate care. Sharon spoke of how she attempted to navigate her clinician's refusal to run basic blood tests by offering to pay, giving the impression that she had to "buy" their care and concern: "*And I was like, "Okay, I'll pay for it". Like, I've never had to ask a doctor to take my iron.".* Contrasting her experiences of private and public care, Isabel highlighted the thoroughness of a clinician accessed privately, versus the "usual" delays.

> *He was very, very thorough. I was in there for about two hours with him. So yeah, so because when you hear that, "Oh, you got to diagnosed in-the first time you met them?" Like, it wasn't just I walked in and said, Hey, this is how I feel like he literally felt every bone in my body. Every bone. […]*
>
> *Whereas with the public one I was waiting for three years now […] But the only problem with public is they've cancelled my appointment that I was supposed to get the bloods back first. So I'm waiting for it to be rescheduled, you know,* ***the usual bullshit.***

5.4.7 GET 7. Experiencing attacks on integrity

Another key theme in all participants' accounts related to their interactions and dialogues with specific clinicians. Narratives illustrated how experiences of being disbelieved and dismissed in healthcare settings profoundly undermined their sense of personal integrity. The act of having their symptoms minimised or disregarded by medical professionals was described as an assault on their identity and a denial of their lived experience.

5.4.7.1 Being unheard and invalidated

A core component of all narratives was the experience of being unheard throughout their healthcare journeys, repeatedly captured by the statement that clinicians "didn't listen". This was impactful and distressing; for instance, Isabel spoke of how it contributed to her sense of alienation, also leaving an imprint on her relationships with others.

> *So in that year is out no one was really listening. You know what, that was probably one of my .... that one sticks out immediately because* ***it was probably one of like, the loneliest and worst periods of my life****.*

Participants often shared their experiences of invalidation, recounting dismissive comments from clinicians, implicit in which were their assumptions about what qualifies as a *"real"* condition. They gave distressing examples of being accused of "*making it up",* their symptoms being dismissed as ones "*everyone*" has or "*in their heads*", illustrated in the excerpts below. Discussing her interactions with clinicians while underage, Sue spoke of what it was like to be a young person in pain. Speaking of how she needed her mother's support in appointments, her account illustrated that young people's voices aren't heard: *"He didn't listen to me. My mom would say it and he would listen more. That was frustrating".*

> *So I just felt very invalidated during that time period, because of my symptoms as well. […] she said, "Oh, it's because you're a woman. This is the second puberty. You, It's your* ***just your hormones****" like […] (Isabel)*

> *She was, it was just the way she spoke to me. It felt like she was trying to tell me the pain was in my head,* ***I'm sorry I'm starting to get upset again****. (pause). It just frustrates me. Because I felt like this was because* ***it was me age****. (Sue)*

> *So they kind of blamed it on like they said, it was like a psychological like you faked the seizure. But they said it was like a non-epileptic kind of like they were like you did have a seizure,* ***but it wasn't a real seizure*** *(Dan)*

Isabel gave a particularly vivid example of how a consultant implied she was simply convincing herself that she was ill.

> *And I had mentioned, I mentioned some other condition I had mentioned like ME or CFS, which are now diagnosed. But whatever I mentioned at the time, he turns and goes, "Can I tell you a story?" And I said, "Yeah" […] He said, "I had a dog before" and I was just went, I literally closed my eyes and I just went, I had a feeling I know where this is going. And he goes, "I had a dog before" and he said "my dog had fleas". And I said, "okay", and he goes, "I started scratching myself because my dog had fleas". And he said****, "I was convinced myself that I had fleas because my dog was scratching."*** *And I said "okay", I said "what's your point?" And he goes," I think you just need to take the diagnosis that you have now and go back to college, and go back to work and get back out in society […]*

As she continued, her account conveyed a sense of not being valued as a person:

> *[…] His biggest concern wasn't my health, his biggest concern was that I would be withdrawn from society and basically become like, anxious.*

Dan spoke of misleading explanations for his condition, and how his symptoms were initially and regularly assumed to be related to age, stress, anxiety or a desire for attention. He was often told he would simply "grow out of it", though his symptoms progressed. He described being outrightly accused of choosing to be unwell, or "faking it", his narrative also capturing gendered assumptions about hypermobility spectrum disorders, the clinician asserting that EDS was a "*teenage condition*" that *"only women"* get. Illustrating further, he spoke of how his pain and daily experiences of joint dislocations were undermined.

> *[…] he said back in 2019 he said that, um, it was an attention seeking disorder that he'd seen a lot of teenage girls have and he thought that I was using it as a way to get out of the stress of the leaving cert.*
>
> *And he (consultant) was like, "You'd know if you were dislocating every day you'd be in pain." I was like, "I am in pain." And he was like, "No, if you can put the joint back in yourself, it's not a dislocation*

Repeated negative experiences also impacted engagement with care, as participants became increasingly distrustful and anxious about experiencing invalidation again. Sharon shared her feelings of anxiety over a "normal" blood test result, fearing this would cast doubt on the validity of her symptoms as a whole: *"I actually went home and*

cried for like, the whole day because I was like, it was like, they don't believe you.". Her doctor's invalidation of fibromyalgia contributed to disengagement from care.

> *And then my GPs reaction to the diagnosis was kind of shocking,* ***and I don't think I could go back*** *there with knowing that she thinks it's* ***...nonsense****.*

5.4.7.2 Being dismissed as "crazy"

Participants' accounts of their healthcare experiences captured their struggle and fight against clinicians who assumed that they were manifesting mental health issues through their symptoms. All expressed frustration and anger when speaking of this issue, experiencing psychological explanations as an insult, an attack on their integrity, or the clinician's way of minimising the impact of their condition. For instance, Sue recounted feeling a complete lack of understanding from her physiotherapist, who offered simplistic advice for managing pain, while Isabel recalled the dismissive tone of a clinician who suggested she had a psychiatric condition.

> *But like, every time she talked to me, it was "Go out with your friends because then you'd be happier and then you won't be in pain" To me... So you're telling me it's it me head like.* ***But if I'm happier, the pain doesn't go away****? (Sue)*

> *The next day, then a doctor came in and he goes, "aw, my wife too, she suffers from mental illness". And I said, and he goes, "she, she suffers. And I suffer,* ***we all suffer****". (Isabel)*

Participants expressed hurt and anger at such assumptions, frustrated at their clinician's lack of knowledge and understanding. Isabel spoke of the tendency to apply a somewhat trendy label of anxiety, prematurely dismissing patients' symptoms: *"I think to the doctors are at the moment, everything is anxiety, everything, you know, the same problem, you think anxiety"*. Her account suggested that labels of anxiety and depression were seen as an attack rather than genuine diagnosis, often delivered to dismiss rather than explain symptoms. She emphasised the long-term impact of such assumptions, possibly preventing her from seeking mental health supports if ever needed.

> *[…] when I've heard the words anxiety and depression so many times throughout my journey. If I even hear a doctor even say them words, I'm like, "Get away from me". Because it's just almost a trigger, because of how many times it's been said when it wasn't.*

Some participants felt that having a previous mental health history or diagnosis contributed to their current being disbelieved. Sharon spoke of her previous mental

health history having an impact on her current treatment; being seen as "crazy" or just anxious by her doctors.

> *Like, I would be like hyper vigilant just like, like freaking out. And I'm like, "this, this is actually ridiculous". I'm afraid of my own shadow practically, you know. So it's a different, it's…**now I don't feel that**. And this new GP, he's like, you know, "I'll give you an anti-anxiety medicine. You were on them before". It made me feel like I'm just being treated like this anxious, crazy person and like that. **Now I'm labelled this**, and that's what it's gonna always be, you know. (Sharon)*

5.4.7.3 Being looked at but not seen

While participants typically experienced being unseen, their accounts also implied that they were looked *at,* in the sense that their outwardly appearance was used as measure of their suffering. Such comments were not only confusing, but unwelcome and experienced as crossing boundaries, illustrated in Sharon and Isabel's accounts. Sharon recounted,

> *[…] And I was sitting there talking to her and she's like, "you know, you can get that turkey neck fixed. We could do it in the office or you could go to a Skin Clinic and they can do it for you. No problem". And I was just like, "what? Oh, sorry, why are we talking about my turkey neck? Like, it's like, you're just saying it or like what's the story?". It was just so odd, you know?*

Isabel extensively discussed how her appearance, rather than expression of suffering, was used to judge the validity of her condition. She further explained that with chronic illness, appointments became her "*outings*", highlighting how misleading such assumptions can be. At attempt to retain her sense of self despite illness was palpable.

> *So like, yeah, I wore makeup. And I think to doctors that came across as "Well she looks good. She's fine". And that is such a shitty misconception. Because you could be so far from fine. […] **when the one thing you hear all the time is "Oh but you look great".** Its like what like what you want me to do, like? when I don't wear any makeup everyone goes, "Oh, you don't look great". **You can't win. Like***

> *[…] one thing I always had throughout my whole journey is.... I love my eyelashes. I love my makeup. I love my tan. I'd said over the years you'd have to pry it out in my dead hands. Like that's how much I love it. So you bet, You could could bet everything that when I walk into an appointment, I'm gonna have like a full face of makeup on. […] because since ...I quit, my, my thing was like, I couldn't go anywhere. They were like, the appointments were my outings.*

5.4.8 GET 8. Legitimizing lived experiences

A key component of participants' healthcare journeys was their experience of searching for diagnostic validation. Narratives revealed how subjective experiences of chronic illness clashed with the evidence-based focus of medicine, leaving participants in a liminal space where their symptoms were assumed "*real*" if objectively measurable. The need to "prove" their suffering through diagnosis was a recurring theme, with many describing their healthcare journey as a relentless battle for credibility.

5.4.8.1 Proving the truth

All participants described an arduous journey towards proving that something was indeed wrong with their bodies, therefore deeming them worthy of care. Their narratives revealed a profound frustration with the medical system's reliance on objective evidence and the emphasis on diagnosis as the arbiter of legitimacy. For instance, Isabel conveyed that having a diagnosis "on paper" was the only way to get clinicians to listen and take their patients' subjective experience seriously.

> *But you shouldn't have to have a **name of something** on a piece of paper for somebody to listen to you. They should look at you and say "I believe you" and not just say "oh, she's a teenager, she's just putting it on"*

Dan described how clinicians were focused on finding real "proof", and how this was used to invalidate his experience. He spoke of how his symptoms were dismissed because his bloods were "perfect", and of having to put in "work" to demonstrate that his condition was real, both to others and himself: *"and now at least I know that I'm not crazy".* A diagnosis was validating and legitimising, later reflected in his wish to differentiate himself from patients who feign illness to abuse accommodations.

> *They were more than happy to accommodate and they were like, "Of course. Do you have any like proof of diagnosis?" Because there was a lot of people pretending they were sick to try and get through.*

Given the poorly understood nature of their conditions, a reliance on measurable data often left participants experiencing a state of diagnostic limbo. All expressed emotional turmoil of *not* being able to find definitive evidence for their condition, despite numerous diagnostic tests and procedures. Sue spoke of how following repeated blood tests, clinicians were unable to specify her condition. She spoke of struggling to make

sense of a new diagnosis: "*Like every time I went, I heard something different. So I had like my mom worried...[pause] Sorry I'm remembering all these things.".* Lisa spoke of how a test was used to prove something was wrong, without necessarily providing answers: "(it) *proves apparently that there's like a viral infection in you, but it can't prove, it just proves that there's something going on*" Like others, she found some solace in "bad" test results, as this meant she could prove she was unwell.

> *He's telling me, "there is anomalies". He's telling me that straightaway, he's telling me "this is not in your head". And I'm going,* ***"Okay, this is good". I'm saying "this is great.*** *This guy knows something.". So I was immediately reassured,* ***and I liked him****.*

As previously mentioned, achieving a diagnosis was a key step in participants' personal sense-making journeys, in that it enabled them to draw connections between their complex symptoms. Yet, while participants typically desired a diagnostic label, their accounts gave the impression that a diagnostic label later became a sort of a curse, in that it impacted their subsequent care. In all cases, a diagnosis was regarded as an "end point" of their journey, which left them feeling abandoned to figure out the next steps alone. Sue spoke of how at a later point all of her ailments were dismissed as fibromyalgia: "*But like, I need to, like I could have a limb hanging off into like, "that's your fibromyalgia". Do you know what I mean? Like*". Similarly, Sharon spoke of how she struggled to sway her doctors to consider anything else once she had fit the diagnostic criteria; that is, fibromyalgia was used as an explanation for "everything". She also expressed uncertainty regarding the validity of her diagnosis, stating multiple times that she was unsure if her clinicians "*ruled everything out*".

> *Like I was saying, I said, like, "Look, I know, I tick all the boxes for fibromyalgia, I get it. But I am having like, so like, this same arm is causing, like, it's intense pain, like, it's, this all hurts, too. But like, this has never stopped. It's, and no one's looked at my arm", do you know? And I was, like I had, I said, "I have this theory about the B12. Because all this stuff is the exact symptoms of this B12 deficiency. And now I have an abnormal pap smear". And I was like, "Can you please test my B12?" […] And he was just shaking his head****. And he was like," No."***

5.4.8.2 A hierarchy of suffering

Participants' accounts of their diagnostic journeys implied there was a hierarchy of diagnoses, leaving them feeling like their condition was not interesting enough. There was a sense that some conditions will always receive more attention and care than others, also suggesting that there is a ranking of levels of suffering. Dan recalled his conversations with clinicians, who explicitly acknowledged this, stating *"I'm going to give you like a rheumatological diagnosis of EDS rather than a neurological one because she's like, it'll be taken a little bit more seriously".* Sharon spoke of how she experienced different approaches from clinicians depending on diagnosis, contrasting her experience of being a "*VIP*" with possible cancer, or being dismissed with fibromyalgia. This hierarchy was mirrored in her clinician's attitude towards referrals.

> *It was like when I they thought I had cancer, I was like VIP treatment. You go in there like, "oh, let's give you this test and this and that and that" and I was like, " eh okay, like what's going on?" And now it's like, "here, let me give you some antidepressants, you'll be fine".*
>
> *When she was trying to say it could be cancer, that's another thing, she was like," Oh, I would have sent you to her - I mean, the first gynaecologist that she sent me to- for those type of things. But I'll send you to him for this kind of thing". Like she kept like, really stressing this. Like "**this is a different story**" kind of thing, you know?*

Alluding to a similar hierarchy of conditions, Lisa shared her experiences with both acute and chronic illness. There was a sense that ailments are only taken seriously if they are likely to kill a patient. She recalled the urgency she was treated with when experiencing possible cardiac issues ("*So it's all panic stations*"), a stark contrast to ME ("*They think I'm just a little bit tired*.")

> *[…] I told him all of that. So just in case any of it could have fed into where I was right now. And again, he was saying "if your stomach issues were really bad, **you'd be dead with cancer"**. Yeah. So yeah.*
>
> *So lo and behold, **they actually take everything really seriously**. I'm not expecting this. But of course, it's just, Why wouldn't you know? See in my head, I'm not taking it seriously. Because I'm so tired. It still doesn't register with me that this could be serious.[…] They **immediately** send me for an echo. They send me for bloods, for a chest X ray, they do a blood test to see if I've actually had a heart attack.*

5.4.9 GET 9. Navigating power dynamics

Captured in their interactions with clinicians, participants' healthcare stories illuminated the pervasive power imbalance between doctor and patient, a dynamic which left them feeling like they were navigating unsaid rules of healthcare. Narratives highlighted participants' efforts to be "good" patients, following guidance and authority, but also instances of challenging their clinicians by speaking up. Experiencing being silenced by healthcare providers, they spoke of adjusting their strategies for managing healthcare encounters.

5.4.9.1 Obeying the unwritten rules

All participants acknowledged an inherent asymmetry of power in healthcare, recognizing their reliance on clinicians' medical expertise, interventions, and decisions, particularly in moments of acute vulnerability. In light of this assumed expertise, they made efforts to adhere to their clinicians' guidance and judgment, following instructions and accepting decisions without question, essentially obeying the unsaid rules of healthcare. Dan gave examples of making efforts to improve and recover, following his doctor's guidance: "*I was able to just completely put every bit of energy into like recovering to the best of my ability*". Sharon spoke of being given medication to take, and "blindly" doing so, without undermining clinicians' instructions: *"So and then they told me, "You got to take them again". So that's just what I do."* Her account generally portrayed that she played by the "*rules*", perceiving the clinician as an authority.

> *Like, okay, like, **I like I was literally clueless** if, if the doctor would tell me something, I would just blindly be like, "okay, they're the doctor". But like, I didn't ask questions. I didn't think to ask questions.*

Lisa's account conveyed a desire to be a successful and unproblematic patient; one who follows guidance, not one who runs for or abuses medication at every opportunity, nor one of the "*random lunatics*" who never recover from ME. She spoke of seeing herself as stronger and more resilient than them, initially as a special case that defied the rules of ME. Her account conveyed respect for clinician authority and expertise. She spoke of how she was essentially given new "rules" for life by her clinician, and how she adopted her life around her illness ("*So I do as I'm told*").

Being a good patient, her account captured a sense of obeying medical authority despite receiving advice against graded exercise from other patients and her husband.

> *[…] I'm going "they're nut cases. Who are these people?" Like, why wouldn't you? So I'm steadfast, these people, my consultants, and the doctors know what they're doing. They're doctors, why wouldn't they?*

Despite coming across as headstrong and defiant throughout her life, Isabel also presented herself as someone who typically follows rules and behaves as expected, describing instances where she didn't argue back. This desire to be seen as respectful of clinicians was reflected in her interview; she was apologetic for harsh criticisms:

> *And I know that sound sounds like I'm like this, you know, I'm shitting on the healthcare system, but obviously**, like I have had good experiences** […]*

5.4.9.2 Speaking up and being silenced

Having grown increasingly frustrated and disappointed with care, participants spoke of how they attempted to share personal insights with clinicians, inquired about alternative tests and treatments, or suggested potential diagnostic avenues. They spoke of how with time and experience, they grew in confidence and began to assert their needs. For instance, Sue gave an example of confronting a clinician who assumed her symptoms were due to fibromyalgia *("I was like, "that's not what I'm here for")* having received an incorrect referral, and similarly, despite the power imbalances, Sharon spoke of confronting her clinician when she failed to disclose possible side effects of medication. Dan also discussed finding his voice and learning to stand his ground in healthcare, something he wished he had done earlier.

> *I wish I could have like if I could go back now with the knowledge I know now, like I would **have been a lot more like vocal** and saying like, this is what you like, you're missing something instead of kind of like accepting it […] (Dan)*

> *And then after, I was like, you know, **"so I guess you weren't going to tell me** about the side effects of this statin, with my muscles thing happening" Like that didn't occur, you know?[…] She was gonna ignore that one. (Sharon)*

However, these attempts at challenging the clinician were usually met with significant pushback. Participants shared instances of when they experienced being silenced, highlighting their lived experience of being looked down upon in healthcare.

There was a general sense that their voice was unwelcome, with their suggestions perceived as undermining clinician authority or expertise. Accounts illustrated that not only were clinicians unresponsive, but their reactions were both defensive and aversive, leading participants to feel that they were not allowed to speak up, even when dissatisfied with their care.

> *And I remember a doctor came in one day and **roared at me**. And he goes, "Stop saying you have this, you do not have this!" (Isabel)*

> *So I'm telling him this, **I'm saying very politely.** "I have got a treadmill in my home. I don't have an exercise bike, can we amend it (exercise plan) because I cannot get in the car to drive to the gym, to then go on an exercise bike to drive home again, I am not well enough to do that." **He throws a hissy fit.** (Lisa)*

Participants spoke of how through their reactions, clinicians highlighted their status as that of above the patient. Their rejection of patients' voice was embodied, their gestures revealing a complete denial of alternative explanations or possibilities. Together, such responses conveyed a strong message that the clinician is always right and is entitled to the "final" say.

> *[…] So I was saying this to my GP and he would not listen to me. He was like, "That's ridiculous. **Who you think you are**?" (Sharon)*

> *[…] Yeah, because he wasn't, he wasn't listening, like, and he was literally like **closing his eyes and doing this** (puts hands up), like, he wasn't gonna listen. (Sharon)*

Such dismissive responses were experienced as hurtful and at times enraging, triggering unanticipated responses. Lisa spoke of how she "lost control" over her temper, enacting what the clinician assumed of her, namely that she was out of control or had a mental health condition. She recalled trying to regain composure, when her consultant suggested she see a psychotherapist; an insult added to injury, given that she expressed frustration at being disbelieved.

And every time I started asking a question, he not only doesn't answer, but ***he puts his hand up****. And he says "I'll do the talking. I'm the consultant. I'll do the talking." So I'm sitting there going "Oh, okay, that's fine. But I still have loads of questions". And every time I go to ask he puts his hands up […]* ***So then finally, I just I lose it. Pounding his desk, and just screaming, I just completely lose it.*** *Of course, he's looking at me going, "this woman is insane."*

[…] I'm regaining composure, because ***I don't want to make whole scene*** *of myself. And so I glance at the card at that stage. And I discover it's a psychotherapist he wants me to see. And I'm saying, "you want me to see a psychotherapist, but why?". And he's saying, "oh, to discuss your anger issues". And at that stage,* ***I'm just biting my tongue****, because I'm thinking he wants me to pay someone to discuss my anger towards him, because that was my anger was completely directed towards him.*

5.4.9.3 Adjusting to the rules

Reflecting the imbalance of power, setting boundaries within the context of healthcare was a challenge for all participants, as they struggled between the wish to be regarded as a good patient and assert their needs. In light of their clinician's negative reactions, participants spoke of how they broke the rules of healthcare by quietly going against their guidance. For example, Lisa spoke of realising that exercise was harmful to her health ("*the penny's dropped*"), and how she decided to rest, going against the prescribed Graded Exercise Therapy. She also spoke of later exchanges with consultants, where she adapted her strategy having learned that confrontation is non-productive. In one example, she spoke of her strategy or mirroring her doctors' attitude, becoming "equally flippant", and hiding her disappointment and true feelings as a way of avoiding conflict. Learning to play by the rules, she withheld her frustration with the outcome in anticipation of being dismissed.

*She sticks a stethoscope on me, she straightaway says my pains are nothing. And I'm going "okay, that's fine". Nothing, great****. I'm not going to press the issue****. She's saying that she's not going to send me for any further tests. And she is really flippant, really flippant.*

[…] And I'm really cross with her but I don't want to show her on cross because she, I'm convinced, already thinks I'm psychosomatic. ***So if I get cross, that's not gonna help me*** *[…]*

Participants acknowledged that they sometimes had to appear to play by unsaid rules and tolerate disappointments, primarily due to their need to have access to any type of

service. Sue spoke of how despite her disappointments she continued to attend her appointments as she was "*desperate*", essentially safeguarding her future in the system.

> […] *Like, I think I was just under the impression that if I didn't go to all these appointments that they set out to try to help me, and then I went back in a few years or whatever, to try to help and they'd be like, "Well, yeah, we gave it to you before and you didn't take it" so I think it was just I was doing...**I guess just worrying about yourself in the future.***

There was also an impression that certain rules were simply intolerable, leading participants to completely disengage from services. Recalling a particularly aversive encounter with a clinician, Dan spoke of being "done" listening and subsequently refusing to follow through on a referral.

> *He said something to me and I was really like the moment where I was like, I'm done with this cardiologist. I'm trying to think what he said. It was like it was something obscene. Like, um. Like you need to move on with your life. You've had your time in the spotlight or something […] He referred me to a psychologist and I literally turned around to my dad and I was like, **I'm not going. It's not happening.** There's something wrong. **I'm not listening to this**.*

5.4.10 GET 10. Becoming disillusioned with healthcare

Over the course of their narratives, there was a sense that participants' relationships with healthcare, namely their expectations and attitudes towards it, shifted significantly over time. Participants seemed to move from a state of initial hopefulness, with expectations that they could and would be helped, becoming disillusioned in the face of repeated disappointments and failures. They came to realise that most clinicians were not experts, but "just people" with limited knowledge about their condition.

5.4.10.1 Being left in the dark

Taken together, all participants' accounts repeatedly conveyed that they felt let down by the system and healthcare professionals, resulting in the subjective experience of being unwanted and alienated as a patient. For example, Isabel gave a vivid account of being pre-maturely discharged from hospital, suggesting that the system is focused more on closure than patients' wellbeing. She implied that this likely compromised her recovery, contributing to her later outcomes. Her distrust and frustration with the system was evident as she spoke of how information about her health status was withheld from her until recently, referring to the healthcare system as the "*puppet master*".

> *They sent me home **they should have kept me in for longer**. But looking back now, knowing what I know now. And kind of the conditions that I have now. They really should have kept me, and I couldn't even walk out to the care when I was coming out […] they had me on back-to-back 72 hour drips from the minute that I walked in. And then one day they just walked in and they said "your liver count is down a bit now so we're gonna let you home". And like it was just awful. Like I was, one of my friends came over to visit the day I came home and she said I was literally grey. So the fact that they thought that it was okay to let me out. Obviously looking back. I don't think it was right […]*
>
> *[…] But anyways, then I was in there for over a week, I want to say because of the... I only found that recently-- So another thing about the healthcare system, **they don't tell you things when you're in there**, which, if they tell you these things, it might help prevent things in the future. I only found out recently that I had viral hepatitis. And at the time, they just said, "Oh, your liver enzymes are four times what they should be", like I was yellow. **But I only found out recently**.*

Sue similarly expressed her frustration with the system, commenting that she was left to deal with a "*mess*", without adequate information or support on her journey.

> *Ah I think a common theme is frustration. Like I don't really have too much work going on now to be frustrated up, or like I said, they're frustrated, when they think back on it or like even talking about today, it was getting upset over it […] So I think, I don't think that dealing with the healthcare system helped with understanding my condition […]*

She explained that while not explicitly discharged, her spinal condition went unmonitored for over a decade, despite being possibly related to her current pain. Her account implied that the manner in which the system operates is inexcusable: *"I was never told "You're being discharged today" or anything. It was just kind of that was that. And you can't blame any of that on COVID, because it was so long Before COVID as well."*

She also recalled being let down at the time of her fibromyalgia diagnosis, feeling abandoned without information or support in the face of uncertainty. This sentiment was highlighted by her clinician's insensitivity and attempt to escape the room following the consultation.

> *And she (mother) was like, "can you tell us what's wrong her?" And she's like, "Oh, well, it's not arthritis" and then like, "okay, then what is it?" And she's Like, I'm not 100% Sure. Like, I think that's fibromyalgia and then like that was, that was my diagnosis, […] but like, she was trying, the door was behind us. And so she would have been sitting in front of us or like me**, my mom kind of had to stand up to keep her from leaving**, but it was just like, **"you're telling us we can leave and you're not telling us anything?"** You know, so and it's hard, Like to say on the way out, with other strangers in the room that "oh, yeah, we think it's Fibromyalgia" like that was my diagnosis. (pause).*

A similar experience of feeling let down, without explanations or support, was illustrated by Lisa.

> *I'm diagnosed with ME but nobody has yet to definitively sit me down and explain what ME is. Nobody has told me what my prognosis is. I still don't understand what I have. I can't explain it to friends, I can't explain it to family. I can't explain it to work. I'm standing in limbo land. And I'm trying to deal with the symptoms that I'm dealing with on top of all of that, figuring everything out**. So I'm desperate for someone to just tell me, what do I have? And where am I headed**?*

Feeling that they have been left in the dark, all participants spoke of how they had to take matters into their hands to "get somewhere", essentially doing the "work" that should have been done by clinicians. Their accounts conveyed a sense of role reversal, as

expressed by Isabel, *"sometimes it feels like you need to hold their [doctor's] hands".* Personal research was a key aspect of the work done by participants, who felt they were given little information, guidance and support on their healthcare journey. Dan commented how he had been let down by his doctors, and eventually had "*done so much research from being so left in the blue"* that the eventual diagnosis did not surprise him.

5.4.10.2 Unfulfilled promises

Disillusionment was evident as participants often compared their attitudes "then" and "now", explaining how healthcare experiences changed their perspectives. For example, speaking of her relationship and expectations of healthcare then and now, Isabel implied she progressed from initially viewing her clinicians as figures of authority ("*And at this time, I still thought that the doctors were Gods* ") to normal people who might not have their patients' best interests at heart. She spoke of learning to expect challenging dynamics, becoming less fearful when navigating the process. Generally, her account portrayed an exhausting journey with many disappointments, giving an impression of broken promises.

> *[…] I feel like I was, because of all the healthcare professionals, I was mentally at my weakest. Like, I had absolutely no hope, I, whereas now.... I have come so far, I'm like, I know exactly what needs to happen if I go to a doctor, I have a plan out,* ***I'm not afraid of them anymore.*** *Whereas back then, I was putting on my trust and almost worshipping them as gods.*

> *[…] So I just It goes to show that every healthcare professional aren't these like amazing, amazing, amazing people. They're just regular people like us. The only difference is, is they have piece of paper say they went to college. They're still,* ***some of them are still cruel****.* ***And some of them are lovely****. Like I've met lovely ones, but some are quite horrible and they don't really care.*

Summarising her change in outlook, she spoke of how negative experiences led her to limit her trust in healthcare providers. Capturing the sense of being abandoned and without care, Isabel reflected on the lessons she learned over time, deducing that the only way to get through the patient journey is to fight for yourself, without expecting help from others.

> *[…] Because, over time, health care professionals, they really made **me lose my faith** in the system I'm ever getting anywhere. It's still a damaged relationship at the minute, but I do have a tiny, tiny bit more trust in them, but only when I'm telling them what tests I want done. **I know it sounds crazy, but I don't really trust them otherwise**.*
>
> *[…] You need to **paddle your own canoe** when you have chronic, any type of chronic illness, because you just left on the shelf.*

A similar initial idealisation of clinicians was evident in the narratives of other participants. For example, Lisa referred to a specific consultant as the "*ME God*", explaining how she anticipated that an appointment with him would be "*the best day of my (her) life*". An added component of this hopefulness related to her beliefs about the private healthcare system, presumably providing a higher level of care, though her experience was ultimately disappointing. She spoke of how this "God" immediately dismissed her, a particularly frustrating incident given she had anticipated answers and life-changing care.

> *[…] What I'm saying to myself, "don't worry, you're seeing consultant number two in another week or two? **All will be fine. He's going to answer your questions**.". We (laughs) used to call him in this house, "The ME God", you're seeing "ME God", **he's going to be great. It's fine.***
>
> *[…] Now, this is a private consultant. And this is no longer public. This is a private consultant. **So I am paying them** 180 Euro, **and I'm expecting to get answers**. So I walk in his door. I immediately allude to the email that I've sent prior to my arrival, he immediately dismisses it and says, " **Sorry, I didn't read it, couldn't be bothered**. By the way, it's not the longest email I've ever read."*

She spoke of her further disappointment, stemming from a realisation that there were no other or better options for care. There was a sense that her illusion of a fair healthcare system had dissipated.

> *[…] So I go back to my GP, I tell her about the horrendous meeting I've had with consultant number two, she apologises and says, "unfortunately, he has record for such a complaint." But unfortunately**, she doesn't have anyone else that she could send me to.** He was the only one that is classified, as an ME Expert, you know, the only one that would have the knowledge to take me forward.*

Experiences of being let down repeatedly by consultants shaped her attitudes towards and expectations of healthcare when seeking help a decade later.

> *So I go into it on the Wednesday. It's the first time now that I've engaged with consultants since all those years 10 years ago. **So I'm not really expecting much** say by way of treatment or by way of anything.*

Taken together, participants' accounts of their healthcare experience gave the impression of being one of broken promises, namely, the implied promise of health. For Dan and Lisa, the promise of becoming well again was explicitly made by clinicians who suggested that they could, and would, get better. While this likely stemmed from poor recognition of the nature of their patients' illness, participants expressed disappointment and frustration at the false hopes they held about their trajectory. For instance, Dan expressed his bewilderment following subsequent seizures, which he assumed would improve over time: *"Um, and then I said, "**Why** am I having another seizure like you told me? I'd never have another one again.".* His later reflection on the POTS diagnosis conveyed a certain scepticism regarding his clinicians' position, conveying that trust has been eroded.

> *[…] so I think it was a year later that my cardiologist diagnosed like teenage POTS. So even then I don't even know if that was like an official. Like I don't even know if he even believed I had POTS.* ***Maybe he just saying it to say it.***

All participants spoke of becoming increasingly aware of their clinician's limited understanding of their condition, leading them to lose trust in medicine and healthcare. Speaking of the lack of understanding of ME, Lisa emphasised that a little bit of knowledge could go a long way. Her comment suggested that acknowledgement of suffering, and a basic understanding of the condition were needed. Even in the absence of a cure, some direction from her clinicians would have positively impacted her healthcare journey.

> *You know, they're seeing... there's such a lack of understanding as to how badly impacted your life is over it. That I'd love to know, just that a doctor would have* ***just that basic grasp*** *as to how it impacts on life.*

Reflecting on her journey, Isabel spoke of how things could have been easier or different and spoke of a greater need for supports for patients with chronic and poorly understood conditions, suggesting this was dependent on improvements in education.

> ***I think it could have been a lot different****. Things could have went a lot different. It shouldn't have taken that long to get where I am now. And I know I'm probably one of the lucky ones.*

[…] And I do think the healthcare system needs to change. They've to change their views. They need to get more educated, the amount of doctors I've walked into, that I've had to tell them what POTS is, or that you've had to explain your conditions to them, to healthcare professionals. There needs to be more education, like barely anyone in Ireland knew what POTS or ME is […]

5.4.10.3 Encountering saviours along the journey

Participants' accounts of their healthcare journeys seemed to follow a pattern, where against a backdrop of primarily negative experiences, they met the "*right*" person who created a turning point in their journey. There was a sense that they met the right clinician by accident, whether because they were a "*filler*" or appeared on their path as a saviour. Corrective experiences with these clinicians were meaningful to participants, who then emphasised the qualities of good doctors, even in the absence of a cure. Following a lengthy journey of being dismissed, and accused of faking, Dan spoke of a turning point in his journey where he, whilst in hospital, met his *"godsend",* implying a need to be rescued from the system.

And my friend's dad, who's a neurologist who was ***the Godsend saviour to everything*** *passed by […] And he was like," I'm paging him. I'm going to take care of you". I, like, because he knew me through like his son. I was friends with his son, so he was like,* ***"I'm going to take care of you."***

Highlighting this landmark event, Dan extensively praised this clinician, highlighting what seemed to be core qualities of a good doctor. Recalling his experience, he recalled how this clinician was shocked to hear about his history of being dismissed, perhaps in this way validating his experience. In addition to being believed, he highlighted how this doctor engaged him in shared decision making and set transparent goals, or a "*game plan".* Taken together, his account suggested that a good doctor is one who takes control and leads, conducts investigations, and treats the patient as a partner. Critically, Dan explained how, after a long journey, this clinician finally connected the dots and made sense of his complex symptoms, bringing some initial closure to his illness experience.

[…] So he got a few more neurologists and rheumatologists to come in and he was like, We're doing an EEG. We're doing, I think some scan with like contrast of like dye to like go around my heart. He did another tilt table, he did all the tests and he was the one that sat down with like all this team of like cardiologists, neurologists, rheumatologists, gastroenterologists […] He, he said, "You're not leaving until we have it sorted because he was looking through my charts and he was like, Why did they not take you seriously?" […] he was like, "We're not, you're

not leaving this hospital until you're sorted, until we have a game plan. You can, you know, work with this."

[…] And it was because of like my resting tachycardia, my severe high heart rates that he was like, "Oh, you have it. SVT and vasovagal syncope was what- they, they were like "It's not the part that's making you faint. It's like the vasovagal syncope." And then he also diagnosed me with, um, convulsive syncope and then like, seizures. So he said that because my oxygen levels are going so low, like into like the 70s or 60s, that they've been able to see from the when the ambulance arrived, and my heart rate was so high he said "the only way your body is like able to like cope or like get oxygen where it needs to go is like your brain is so starved of oxygen that it's triggering a seizure"

Like Dan, Sue also identified meeting the right clinicians as a landmark event in her healthcare journey. She praised healthcare providers who truly listened to her, involved her in decision making, made referrals and used age-appropriate language. Again, there was a sense that such doctors were a godsend, appearing on the journey out of nowhere. She also made certain observations about "good" and "bad" doctors, emphasising her feeling of not being taken seriously as a patient.

[…] But like I had two GPs, so I was always trying to see the nicer one. Yeah, so that GP is a bit more understanding. Yeah, the other one, not so much. […] He's just, he's just a ***horrible, rude person****, you know, he'd kind of like, diminish what you're saying or play down on what you're saying, you know? Nothing's ever like serious or anything to him.*

And then ***when someone filled in for him, he was amazing****. I went to him. I was like, I've like "I'm having really bad headaches". "I'll send you for an MRI "and I was like, yeah, like I was.* ***He was a lifesaver.*** *The few weeks or he was there a few months. He was great.*

Similarly having access to a multidisciplinary team was a major turning point for Sue. She spoke of having a whole team of professionals who appeared to listen and have her best interests at heart, a stark contrast to her previous experiences in healthcare. She spoke of how meeting a clinician who engaged in a real conversation and considered her perspective made her feel valued and was deeply meaningful. She described becoming emotional after meeting this team, for the first time feeling listened to and safe to speak.

I kind of went in and thinking like, "they're not gonna listen to me. And they're gonna just talk about the fibromyalgia even though that's not what I'm here for.[…] And they were so understanding, which was a first for me. I was 21 at that point. And that's the first time I'd ever been like, "Oh, I've been listened to". […] It was weird. I think ***I actually cried because I was so happy.*** *Yeah. Which wasn't something I did often. Yeah, I was just worried because he listened to me like, I'm sorry. It's even. Getting emotional. Sorry. (pause).*

Like the others, Isabel's account illustrated that meeting the "right" doctor is a key turning point in the healthcare journey. She described certain doctors as "saviours", suggesting that they could save her from a bad or broken healthcare system. Speaking of her surprise regarding blood test results, she implied that "good" doctors are ones who pick up on symptoms that their patients dismiss. Again, meeting a helpful doctor appeared to be accidental:

> *And I ended up going to a GP but it wasn't my normal GP, it was a **filler** […] And this GP kind of had a look at everything and looked at my symptoms. And she kind of went, "I'm gonna do a couple of blood tests". And I was like, "I thought it was my normal tonsilitis".[…] A week later, within that week, they got the tests back. So it was the glandular fever. But my situation was just getting progressively worse, like my throat was closing up, couldn't take in, any fluids, any food. And then it got to a point where my GP gave a letter and said, "Bring her straight in" and we went into, obviously the A&E and sat there. But the doctors basically said that we should have went straight in because they had like an isolation room setup.*

In a powerful account of her experience of medication-induced psychosis, Isabel also implied that a good clinician defends the patient from other doctors. Having been accused of suffering from a mental health condition, she recounted how she agreed to a mental health consultation to prove the point that she was not mentally ill. Her account seemed to convey that there are "good" an "bad" specialists, differentiated by whether or not they listen to their patients' experiences.

> *And I actually said, "Can you please get me a psych consult?" And he goes, "You", they were so happy. "You want to psych consult?" And I was like, "yeah, so they can, So they can actually like write on a piece of paper and say that I don't have anything", which is one thing they gave me. […] They gave it to me. I think it's because **they thought they were going to get somewhere.** And they were going to get like, and fucking anxiety, sorry for cursing. Or like a depression diagnosis. And that will be it.*
>
> *[…] But no, psych came down, And I remember I was with her for five minutes, and chatted to her about my family life, about like, my journey and everything. And she was like, "okay, okay". And then she was like, "and your parents haven't been together, has that affected you?" And I was like, "Well- I was like-I have four sisters. I love them". So I was like, "I don't really care". And after the five minutes, she looked down at her page, and she goes, "I have to go get somebody", called in the nurse. And I was like," oh," and she, she looked me up and down and goes, "**they need to take you off their medication** and put you on something that's not a beta blocker".[…] **And she looked at the doctor, she walked out the door and she goes**, "if you even looked at the side effects, you'd see the beta blockers can cause psychological symptoms".*

Later, meeting a doctor who helped explain what was happening, her account illustrated some qualities of a "good" clinician. Again, her account conveyed that being seen and heard is emotionally transformative. She described an initial apprehension, and then the relief of a positive experience.

> *And I'm not joking. I went in and I sat down, and I make a point. I was sitting there and I was going**, "this man's going to look at me and think, "Oh, well, how could she possibly not feel great?** Like how could she possibly have all these things?" And within the first five minutes of talking to him, he literally said**, "I see you. I hear you. And I'm here."** And it was... So I came out of that appointment, nearly crying like, I was so happy with the level of care from him. And that was a first.*

Sharon also discussed her few positive experiences with healthcare practitioners, highlighting how being given some thought was unusual against her typical experience of being disbelieved and dismissed.

> *And she sent me to a different gynaecologist, who was very calm, and was very good. Like, he's, **he's the best**. He's very good. So **luckily** have him. […]*
>
> *I didn't know what it was and I was happy that someone was actually giving it some thought, like, "what's going on with you? Let's get this guy to check it out. Because this is what he does". Do you know what I mean? So I was like**, happy that he considered it? You know, because my GP wasn't,** and I was giving her like, I was like seeing her twice a month and coming out with nothing new, you know?*

Her narrative also captured the qualities of what she felt was a "good" doctor and expressed feeling "lucky" to meet someone curious, attentive and calm. She emphasised the impact of feeling listened to and cared for even in the absence of a solution, showing appreciation for her clinician's transparency regarding this.

She praised her clinician for putting effort into making sense of her disparate symptoms and test results, and also, for taking her side against a disbelieving other.

> *So he was lovely and he listened. And he looked at my X rays, MRIs and all that and you know, the symptoms, but the thing was to at the time, my symptoms kept changing. Like it would get, I would get like really tight muscles and then really, like, bad nerve pain and then really...*
>
> *[…] Yeah. And I told him about what my GP said, because I said she didn't think, like I said," She sounded really annoyed to be getting referred to you." And he was like, "**Oh, she's one of those**." And then he wrote, like this lovely letter to her to explain why he diagnosed me with that. So there's that.*

Despite having largely negative experiences, Lisa also spoke of experiencing turning points where she felt lucky with healthcare, for example, being seen to quickly, and getting an ME diagnosis on a "lucky" day. Recalling her positive experiences, her account conveyed that a "good" clinician believes their patient, validates them and are genuinely interested in their wellbeing. She noted that a good clinician does not necessarily have to have a solution, but rather, is willing to provide ongoing support to the best of their ability. In another instance, she highlighted the importance of transparency and willingness to change.

> *She's a really, really good GP in the sense that she wants to find something. You know, she really is wanting to find something, she's wanting to treat me if she can treat me, she's almost in tears sometimes listening to me listing out my varying symptoms, like she's nearly more upset about things than I am. […] She feels her hands are tied. She's tried to send me where she sent me. There's nowhere else in Ireland she has an offer to send me and she's got a blank wall. She's, **she's, she's doing what she can and I'm...her door is open**. She's telling me if I ever have, you know, low mood or any sort of issues that are door's open, she's telling me that you know, as I say, get my bloods done every year. If I worsen any further to let her know, […]*
>
> *[…] She says that from all of my notes, from my symptoms, exercise has made me worse. And we need to pull the plug. And this ME thing is more serious than she thought […] She's, again, patting me on the back saying that, you know, I've been really good, you know, handling it, blah, blah, blah, how I've been coping. She again empathizes with how bad my symptoms are. She, you know, says all the right things, essentially. And then she agrees that **if an ME patient ever comes to her again, she will not take them on.***

Like the others, Lisa spoke of the importance of meeting a clinician who understands and takes time to explain their patient's condition. Her account conveyed that it is so rare, and her expectations are so low, that she was almost shocked at empathy and being understood.

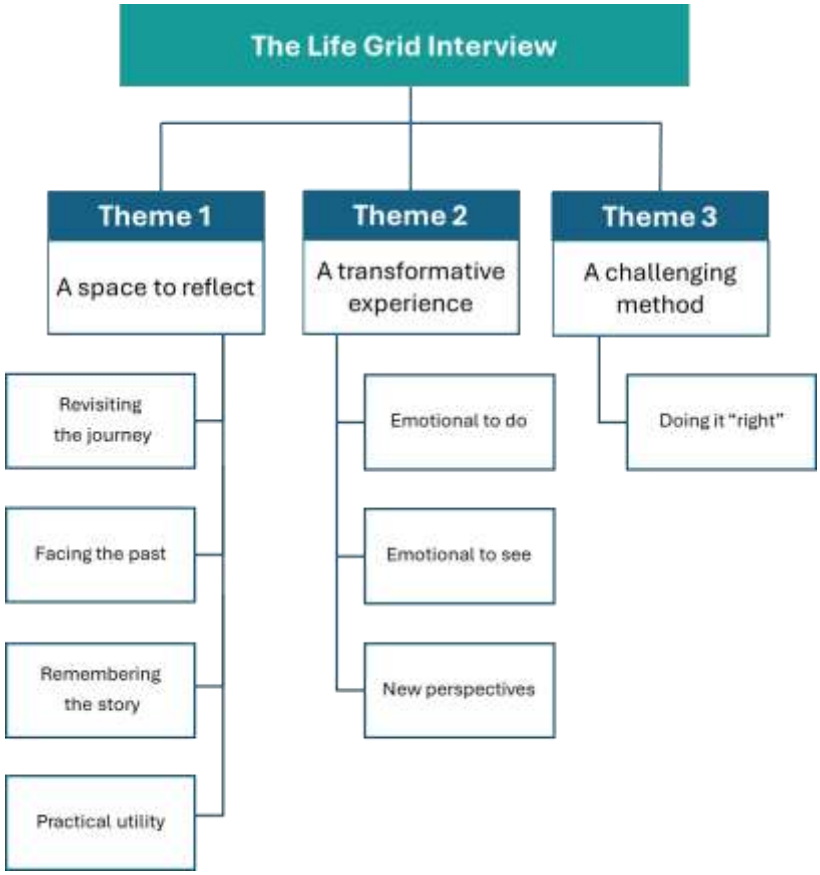
> *So they're really empathetic, which **I'm shocked at, they actually do have a clue** about ME, the doctor does actually understand it. And for the first time, in my life, a doctor explains ME to me. […] So yeah, the consultant, a lovely one, I forgot his name, he was lovely. He is sitting or standing at my hospital bed, and he's telling me you "You've got ME, which means that you have an overactive immune system to begin with. It's post viral" you know, he goes through my case history, he again says "you have this, this is what happened. […] So yeah, so he's going through all of that. And, as I say, I'm sitting there going, "Whoa, that's interesting.". That's the first time the doctor is actually linked things put things in front of me for the first time in ever.*

5.5 Findings II. Thematic Analysis

As the concluding step of the interview protocol, participants (*N*=12) were asked about their experiences of the Life Grid Interview. Reflexive Thematic Analysis was conducted on their responses, which were grouped into 3 themes and 8 subthemes presented in Figure 5.3 and discussed below. Results are presented with illustrative quotes from participants in response to the question and are supplemented by the authors' observations on the overall interview process.

Figure 5.3

Participants' experiences of the Life Grid Interview



5.5.1 Theme 1. A space to reflect

The first theme relates to participants' experiences of the Life Grid interview as dedicated time and space for reflection with regards to their life course. In discussing their experiences, many spoke of the Life Grid interview as one which gave them a chance to revisit specific periods in life and reflect on their biography. While this process was insightful for many, it also required confrontation with unpleasant realities, both those which were forgotten and actively avoided within personal narratives. Taking part in the interview, which required participants to elaborate on specific periods of their life, also triggered the recollection of other events which may have not been included on the timeline. By stimulating recall, the Life Grid tool was seen as a suitable qualitative research method in the context of chronic illness, and possible applications of the constructed timeline were identified.

Sub-theme 1.1 Revisiting your journey

A key experience among participants was that the Life Grid interview enabled them to retrace, and engage with, their journeys in a holistic manner. This biographical mapping encouraged a sense of continuity, whereby participants were able to locate and narrate their experiences within the broader contexts of their life trajectory. For some participants, this resulted in a recognition of the journey they have been on:

> *"I think I think when you go through like so much like with chronic illness, when you're like have these rare illnesses, you kind of forget like where you've come from" (Participant 1)*

> *"You sometimes think "is, this is really all happening?". So actually, all that really happened in my life. I mean, with all my work abroad, with all the troubles I had, with all the things I've been through privately, in my family all chaotic times, and stressful and traumatic times, privately. And to see where I am now. And sometimes it looks like I lived the life of what, 10 people". (Participant 12)*

While some participants were comforted by such realisations, others expressed dissatisfaction with the manner their life, and health, had progressed.

> *"Like, it just doesn't seem fair. You know?" (Participant 11)*

Completing the Life Grid and in-depth interview sometimes highlighted the biographical disruption by illness in narratives, resulting in disappointment or frustration. A few

participants expressed this in relation to their healthcare journey, noting the impact it had on their overall life course and identity.

> *"And I think it's in a great way, but all that shouldn't have to happen for me to be the way that I am now. And, I feel like everybody who has a chronic illness goes through it, and you don't get it until you get it, really." (Participant 6)*

Sub-theme 1.2 Remembering the story

In addition to providing a means to revisiting the life story, the Life Grid served as a memory aide, enabling participants to better recall and articulate their life events and experiences. Throughout the interviews, participants often remembered life events and entered these into the life grid as the narrative progressed.

> *"It's authentic, you know, there's just like, "oh, like, I remember that" (Participant 5)*

They were often surprised at having forgotten milestones and critical health events, as expressed by one participant.

> *"Oh my God, I actually remember I had a seizure at that time and like I forgot that I even had a seizure in school." (Participant 1)*

Several participants found the challenge of reconstructing a chronological timeline enjoyable and fruitful, in that it enabled them to better tell their story during the interview.

> *"It was good to kind of sit down and then kind of go, okay, so would have been like, this year and this thing. So it was nice to have a picture of that and the timeline." (Participant 2)*

Sub-theme 1.3 Facing your story

The Life Grid also challenged some participants to face aspects of their life stories which they would not have willingly or typically engaged with otherwise. Several participants noted that the Life Grid pushed them to confront and process aspects of their lives that previously evaded deeper consideration, or which they had actively avoided.

> *"I never looked back. I had it, done it, got the T-shirt and I moved on, I put it the back of my head, put it on the shelf. And never talked about it, never analysed it, because there's no point, well, and you have to get up and get on with it." (Participant 9)*

> *"[you] are springing up like old memories, I guess which like I don't like to deal with. I do think that was great, kind of been able to see it because like, I like I said, like, if I, if I don't think about it then it's not there, kind of thing. So I was just kind of having to deal with it again" (Participant 5)*

Interestingly, the process of recognizing and recounting significant events did not necessarily result in experiences being fully integrated into personal narratives. As noted by one participant, even though they revisited parts of their life during the interview, there remained a disconnect in fully internalizing these as part of their identity:

> *"Yeah, I haven't thought about it really, I think as a coping mechanism. It's like, it's all there and I can see it all there, but it doesn't fully register that that's me. Do you know what I mean?" (Participant 7)*

Sub-theme 1.4 Practical utility

Some participants commented on the practical utility of the Life Grid tool in the context of qualitative research and beyond. Interestingly, several participants who attended the interview disclosed having kept chronological lists of their health changes and appointments in preparation for engaging with the healthcare system, which assisted them in filling out the Life Grid. When asked about this list, one participant explained:

> *"I keep it going. Because I know it's easier for a hospital procedure. And you just, they ask you what you had and the surgeries and I just handed over the list because it was getting too complicated." (Participant 12)*

Another participant commented on how the tool can be particularly useful for research with people who may have memory problems or experience fatigue, enabling a more complete in-depth interview.

> *"[…] you can have like poor memory and that kind of thing... I find in a lot of other interviews and things. I'll go," Oh, I forgot to mention this." […] So it was good to kind of fill this out in advance, go through it, and like talk a bit more about it and be able to add things in. But I kind of know then that I haven't left anything out. And I haven't missed anything." (Participant 2)*

They further suggested that a completed Life Grid can be a tool for engaging with healthcare professionals:

> *"It was it was almost useful for just me as like a reference […] And I'm like, "Okay, I'm gonna write that down somewhere that when these things happen, because if I go into a GP or something, and they asked my medical history, I'm like, I blank." (Participant 2)*

5.5.2 Theme 2. A transformative experience

Taking part in the Life Grid interview was an emotional and transformative experience for many participants. The task of recounting complex life events and re-living memories during the interview, as well as witnessing a timeline of these events in a physical form, often brought about deeper reflections on one's life course and a recognition of personal strength and resilience.

Sub-theme 2.1 Emotional to do

As mentioned, the task of narrating one's life story through the Life Grid interview proved to be deeply evocative for many participants. While the structure allowed for control over disclosure, many chose to share both positive and painful life events, revisiting personal triumphs and unexpected turns alongside challenging experiences. Participants often found themselves re-experiencing emotions associated with these events, highlighting the powerful nature of the interview process.

> *"[…] so yeah, it was…it was it was different, to feel it, to go through it." (Participant 3).*

Re-telling stories from childhood often prompted participants to access and re-examine formative experiences within the broader context of their lives. The Life Grid provided a framework for participants to explore their evolving relationships with family members, revisiting key events, conflicts, and reconciliations that shaped their family dynamics. This process of revisiting early memories evoked a range of emotions, as participants grappled with the lasting impact of these experiences on their present selves. One participant, reflecting on their family dynamics through the Life Grid, expressed surprise at being able to contain their feelings during the interview.

> *"[…] it's very hard to like, you know, I mean, let's say even to speak about my mother, and I'm surprised now I didn't get as much, you know, upset, but like it does, it brings back, it brings back the emotions, you know, even going through that when I was doing it now, you know, and the things that she did. You know what I mean?" (Participant 4)*

For some, the act of revisiting past wounds, whether from family life or the burden of chronic illness, re-ignited a grief over a life they had lost:

> *"And I, I thought it was weird because I felt really almost ...sad for old me because as I was going through it, I kept getting this like, feeling like, as I was filling it out because I was remembering like, how I felt at that time. And how like that teenage girl like didn't know what was ahead of her like, I just want to go back and like be able to hug her, so it was it was nostalgic to fill it out. But it was, it was kind of sad at the same time because I was also thinking, even though I am further than I was by a massive pile, but, I just didn't think that I would still be unwell, but yeah looking back It was kind of sad.." (Participant 6)*

For others, the opportunity to voice their pain and the experience of being listened to were a source of relief, highlighting the transformative effect of the interview.

> *"I think it's really good. It's very cathartic. I think it's really, really good. And, you know, I know you're doing this for your study and everything, but I really appreciate you listening. […] whatever I'm doing for you, you're doing 100 times more for me by listening. So I really appreciate it" (Participant 8)*

Sub-theme 2.2 Emotional to see

In addition to the emotions evoked during the interview, some participants commented on witnessing their life story condensed in a visual format. For some, reviewing the journey as a chronology of primarily negative life events was a jarring experience.

> *"Oh, God, when I did it, when I was filling it out yesterday, I was like, "oh my god, it's just miserable". It's not, it's kind of, it's kind of depressing to look at it all together. Do you know?" (Participant 11)*

For others, seeing the Life Grid served as a reminder of how life could have been different:

> *"But um it's not the nicest thing to look at. I mean, it's like, well, yeah, this is the reality. This is how it is (pause). But it could have been better." (Participant 12)*

Sub-theme 2.3 New Perspectives

Some participants remarked on how completing the Life Grid and interview was a transformative learning experience. Several made reference to having "realisations" during the process, which led them see themselves in a new light. Some participants discussed this in the form of a change in mindset:

> *"I'm starting to see things because like, I don't really think about these times too often, but like, having a different mindset looking back, and I'm like, oh, like, you know, I'm kind of starting to see it differently now. So I guess it was nice. Just for me, personally, kind of looking back on it [the life story]." (Participant 5)*

Often, these realisations involved recognition of one's own strength and resilience.

> *"That I'm probably resilient that like, looking back, like even just as I had, thinking about the mindset that I had at that time, it made me realise like, how mentally strong I am now." (Participant 6)*

Some reflected on their agency, expressing a sense of amazement at what they had overcome:

> *"Like, wow, like, I've been through, I went through this, I went through this, and I'm still here to this day. […] But I'm pretty amazed and I don't want to be like, "Oh, look at me, me, me, me", but I'm proud of myself. I'm really proud of myself of how I've handled it, accomplished […]." (Participant 9)*

Such realisations gave participants comfort, or confidence, in their own abilities to face future challenges:

> *"And like thinking back, I was like, "Oh my God, that happened". And like remembering like the journey you've gone through and kind of like. Helping you feel as strong as you are today, knowing that like if you can get through all of that, you're going to be able to get through so much more like you've done it already." (Participant 1)*

5.5.3 Theme 3. A challenging method

The final theme concerns the challenges faced by participants when taking part in the Life Grid interview, primarily in relation to their concern over completing it correctly. Throughout the interviews, participants often sought validation about whether or not they were proceeding in the "right" way and were apologetic over perceived mistakes or amendments to the timeline they had constructed. Only two participants made explicit reference to these concerns when asked about their experience of the interview.

Sub-theme 3.1 Doing it "right"

Participants' concern over whether or not they were completing the Life Grid in a correct manner were typically expressed at the start of the interview, before participants became familiar with the flexibility of the tool and non-directive nature of the interview.

Despite being given full control of developing an initial blueprint of their life, participants were often unsure of which significant or meaningful events to speak of, and whether or not these were relevant to the research question.

> *"I probably have too much in this I think, you know, but I was trying to pick." (Participant 4)*

> *"Yeah, so it was actually kind of… it's not so much that it was difficult. But it took me longer than I expected, because when I did open it, and I was like, this is really a really good, like, I was really interested in the method. I was like, "Oh, this is such a cool way to kind of look at the overview on how to go back and forth". And what then I looked at it now, I actually don't know what's been going on for my entire life. Like, I was like, "Okay, I'll start at the start. I was like, I was born. And then also, then what happened?" Like I had to actually rethink, and like, it's probably not what you're supposed to do. But I literally Googled like, what do you put in a life grid, and I couldn't find any results anyway, so I didn't, but I was like, "what happened in my life?""(Participant 2)*

5.6 Discussion

The present study provided an in-depth account of the lived experiences and healthcare journeys of individuals with poorly understood conditions in Ireland, and further, explored participant experiences of the Life Grid approach to interviewing. Findings illustrated the deeply transformative impact chronic illness has on individuals' lifeworlds, providing a holistic view of personal, social and healthcare contexts. The results also provided a nuanced experiential account of healthcare journeys, highlighting themes of disbelief and invalidation, power imbalance, and disillusionment and distrust among others. Together with quantitative data and exploratory patient journey maps, results suggest a lack of patient-centered, coordinated and multi-disciplinary care for this patient group in Ireland. The Life Grid approach was positively regarded by participants, suggesting it can be a fruitful tool for research exploring illness experiences over time. Several findings in relation to lived experiences and healthcare journeys are revisited below, along with considerations regarding Life Grid methodology.

5.6.1 Lived experiences and Healthcare Journeys

A core component of the phenomenological experience of poorly understood chronic illness is the disruptive impact on an individual's "normality". Participants' accounts conveyed how developing symptoms reshaped their trajectories, progressively or acutely impacting their life, often forcing them to re-evaluate their expectations about the present and the future. Their narratives illustrated a profound sense of loss and injustice as they grappled to retain their sense of self while navigating their new reality as a person with chronic illness. Their personal journey was characterised by initial attempts at minimising the symptoms, both to themselves and others, and then confrontation with and attempts at making sense of their condition. Such experiences are well recognised throughout the literature (e.g., Åsbring, 2001; Frane, 2023; Saetre & Eik, 2019; Wilde et al., 2020), exemplifying what Bury (1982) described as biographical disruption, or the re-shaping of how individuals view their identity, roles, relationships and aspirations when they experience a destabilising event such as chronic illness. Further, in line with Merleau-Ponty's (1968) embodied phenomenology, participant narratives also repeatedly illustrated how chronic illness leads to the disruption of embodiment, or the loss of transparency of the body. Their accounts conveyed how their

symptoms forced a shift in how they related to their bodies, which was at times experienced as obstructive, punitive or out of control. With greater insight into the patterns and triggers of their symptoms, participants spoke of how they had to succumb to their body's demands for rest or otherwise pay the "price" of subsequent symptom flares.

In terms of healthcare experiences, the findings present a multi-layered account of disappointment with the patient journey and quality of care. Most participants (83%) were dissatisfied with their healthcare journey to date, while over two in three were dissatisfied with the quality of healthcare services available to them. Despite seeking help within a year of symptom onset, most reported delays in diagnosis and noted lengthy wait lists for specialist services. In terms of service use, patients generally reported seeing a GP most frequently despite the fact that this was not always the most useful service for their condition. The central role of the GP in coordinating care was further reflected in a selection of patient journey maps. Phenomenological accounts mirrored these findings, further illustrating patients' experiences of "fighting" their way through a broken system, with a sense of going in circles between the GP and other services or being left without guidance, stuck in limbo on waiting lists.

At an interpersonal level, the findings highlighted that patients' encounters with healthcare providers were marked with disbelief, invalidation and stigma, and were underpinned by complex power dynamics whereby patients were silenced when expressing their concerns or needs. The rich accounts demonstrated an ongoing battle for care and legitimation through diagnosis. As illustrated, a focus on objective evidence was particularly dehumanising due to the "unexplained" nature of symptoms, with participants being dismissed, told "nothing" is wrong, or accused of faking their illness due to a lack of "proof" despite disclosures of ongoing suffering. At the core of this was the experience of not being seen as a person, which resounds strongly with Foucault's (1973) concept of the medical gaze, a phenomenon where professionals view patients and their bodies as objects of knowledge while distancing themselves from lived experiences of individuals. It is also noteworthy that while participants' narratives typically illustrated a progression from hopefulness to distrust in healthcare, they also identified turning points or landmark events, conveying a sense of being saved from a

broken healthcare system. Participants discussed the transformative experiences of meeting the "right" doctor and differentiated good and bad healthcare providers by whether or not they listened to them and engaged in shared decision-making. Together, participants' accounts underscored a need for compassion and empathy in addition to coordinated multi-disciplinary care.

5.6.2 Life Grid experiences

Participant reflections on the Life Grid interview revealed that it is a powerful and versatile tool with potential applications extending beyond research. The approach allowed participants to recount their life stories, while its flexibility enabled the revisiting and remembering of important life events, and consequently, the restructuring of personal narratives. This aspect was particularly valued by participants due to their convoluted medical histories, many of whom noted that constructing an initial timeline prior to the interview encouraged deeper reflection. Furthermore, the method facilitated a holistic exploration of life histories, illustrated by the depth of phenomenological accounts, which while occasionally challenging, was overwhelmingly regarded as a positive experience.

Beyond this, the Life Grid interview was a deeply emotional experience for participants, bringing to the fore a range of complicated feelings. Participants reported feelings of sadness, grief, and frustration but also expressed pride and recognition of their personal strength and resilience. For some, the process of telling of their stories was deeply transformative; it enabled them to speak of events they had never shared before. For others, seeing the mapping of their life course was unsettling in that it necessitated facing unpleasant truths and disappointments. Interestingly, underscoring its utility for patient journey research, some participants commented on the value of a tangible output of the interview, i.e. a mapping of their journey on a grid, and how this could be informative for their future medical appointments. Overall, participants' experiences aligned with the merits of the Life Grid in the literature, which has underscored its empowering and flexible nature as well as suitability for in-depth explorations of the chronic illness experience (Ballal et al., 2021; Parry et al., 1999; Richardson et al., 2009; Wilson et al., 2007). Methodological considerations are discussed below.

5.6.3 Methodological considerations

While the Life Grid was an effective tool for the collection of in-depth life course data, as perceived by the researcher and experienced by participants, several methodological considerations related to this approach and narrative methods warrant further reflection. These include the practical aspects of applying the Life Grid, the type of data generated, as well as the emotional labour and impact on both participants and researchers. From a practical standpoint, the participatory and visual nature of the Life Grid was particularly useful in that it helped "anchor" the interview, whilst prompting a multi-dimensional exploration of experience over a lifetime. Such reflections have been reported by others (e.g., Harrison et al., 2011; Richardson et al., 2009) who note that the Life Grid enabled participants return to their story if distracted, also making it possible for the researcher to guide the conversation back to the interview where needed. Interestingly, despite the freedom to start anywhere on the timeline, all participants decided to tell their stories chronologically and began their accounts with childhood experiences; this tendency to start "at the beginning" was also observed by Richardson et al. (2009) in the context of chronic illness research. Thus, this approach might productively address some limitations of narrative interviews which tend to result in unstructured accounts, making it difficult to establish timelines and sequences of life events (see Anderson & Kirkpatrick, 2015). The tool might therefore be particularly useful for qualitative healthcare journey research.

With regards to the data generated, one important critique of life story research concerns the inaccuracies inherent in retrospective and autobiographical accounts. In other words, it is important to remember that the past is reinterpreted and constructed through the lens of the present. Several authors have noted that it is not possible to evaluate to what extent the stories captured by the life grid are a "true" rendering of experience (e.g., Bell, 2005; Nico, 2016). In the present study, the Life Grid enabled a reconstruction of life histories and participants' healthcare journeys, with a focus on lived experiences rather than accuracy. It would be interesting to align participant timelines of their healthcare journeys against those recorded in their medical records. However, the underdevelopment of electronic patient records in Ireland (Walsh et al., 2021) combined with system fragmentation significantly limits the tracking patient

journeys through the system, particularly where prolonged and requiring multidisciplinary input.

It has also been suggested that the task-based nature of the Life Grid interview can make it particularly suitable for research concerning sensitive topics, as it reduces the pressure for eye contact (e.g., Harrison et al., 2011; Parry et al., 1999). Although most interviews in the present study were conducted online, the researcher observed that the task of completing a virtual grid while speaking improved conversation flow, requiring less interruption or prompting during the interview, with emotional content coming to the fore more organically than in other interview formats. The utility of task-based approaches to interviewing, particularly regarding sensitive topics, has been illustrated by others (e.g., Figoureux & Gorp, 2021; Marshall, 2019).

A noteworthy consideration specific to this study design was the benefit of preparing an initial timeline prior to the interview. While this was helpful for participants in that it reduced the cognitive load of the interview, the opportunity for the researcher to review the content prior to the interview afforded the possibility to reflect on and prepare for the discussion. This was felt to be particularly important given the researchers' positionality, which as discussed was one of close proximity to the research topic. The emotional impact of qualitative work on the "insider" researcher has been widely acknowledged (e.g., Hoffmann, 2007; Ross, 2017) with calls for a focus on training in reflexive practices which stretch beyond ensuring academic rigor (Kinitz, 2022). The transformative impact of methodology was captured by participants' reflections and mirrored by the researcher's experience underscores that choice of method is not only a question of pragmatism, but one with ethical implications.

5.6.4 Strengths and Limitations

A particular strength of the present study was its analytic pluralism and methodological rigor. The design of the study allowed for a multi-faceted exploration of participant experiences of chronic illness, of healthcare and of the participatory approach, as well as providing preliminary insights into patient journeys and the potential for patient journey mapping in this domain. These approaches complemented one another, providing unique insights that could not be captured by qualitative of quantitative data alone. As previously discussed, the IPA adhered to quality guidelines in that it provided a rich

experiential and existential account, focusing both on similarities and contrasts between participants, whilst remaining contextually sensitive. Putting patient voices at the centre of the research, the study was co-developed and piloted with individuals with PPI Contributors.

However, several limitations of this study should also be acknowledged. While accuracy of accounts was not the premise, it is important to note that the retrospective data collected regarding healthcare journeys limited the elaboration of patient journey maps. Given the complexity of accounts, and reliance on memory, it was not always possible to establish where referrals to services materialised, where discontinuity occurred, and how information regarding clinical processes was communicated or fed back to providers. An added critique is that the study involved individuals with a variety of poorly understood conditions, thus limiting the generalisability of findings. However, generalisability is not a key aim of the interpretative phenomenological approach, which as discussed by Smith et al. (2009) seeks to identify shared experiential themes among individuals with a shared experience. In the context of this study, the shared reality among participants was living with a "contested" or "medically unexplained" condition. An additional consideration relates to participant literacy and digital literacy. Some participants expressed anxiety about writing content into the Life Grid due to fears around spelling, highlighting the need for flexible data collection approaches. While most participants preferred online interviews due to convenience, some preferred to take part in-person, which raises broader questions of sampling and accessibility.

5.6.5 Future research

Future research should detail the patient journeys of patients with specific poorly understood conditions to identify gaps in care, assessing continuity, coordination and breakdowns of communication. By examining and understanding what the patient journey is like in practice, the research can identify targets for change and assist in developing refined care pathways for individuals who fall through the cracks of existing models of care. Further, as the central role of the GP in care coordination is well recognised (Vassbotn et al., 2018), including in the context of rare disease in Ireland (Byrne et al., 2020; Department of Health, 2014), established pathways for individuals with complex or unexplained symptoms could better support GPs in decisions regarding

referrals and investigations. As an initial step, studies could gather retrospective data on GP referral practices based on primary care charts in addition to patient accounts of their journeys, capturing both the practical and experiential aspects of the patient journey. Prospective studies could systematically track patient groups as they move through the healthcare services from when they first seek help for their symptoms, through to diagnosis and long-term management. This would provide data on key transitions and longitudinal trends in service use, also identifying resource allocation needs. A key focus of future work should be amplifying the patient voice to inform system improvements and change, thereby ensuring care models are responsive to patients' priorities. This is crucial given the high dissatisfaction with healthcare expressed by patients in the present study, and the association between patient experiences of care and their outcomes.

5.6.6 Conclusion

The findings of this study illustrate the complexity and situated nature of the lived experience of chronic illness and highlight that healthcare journeys of patients with poorly understood conditions in Ireland are fraught with difficulties, with barriers to care at inter-personal and systemic levels. This research underscores the transformative impact of patient narratives on both research and medical practice, paving the way for a more holistic, person-centered approach to care and challenging outdated, reductive perspectives.

Expanding on the findings from Phase Three, which reveal a range of barriers to care for patients with poorly understood conditions, Chapter 6 presents a focus group study that explores service users' perspectives on healthcare improvement targets and proposes actionable recommendations for change.

Chapter 6. Study Four

Healthcare Needs and Recommendations for Change

6.1 Introduction

As discussed in Chapter 1, the prevalence of chronic noncommunicable diseases is rising, posing a significant challenge for healthcare systems worldwide (McGrail et al., 2016; World Health Organization, 2023). A key factor contributing to this challenge is the oftentimes progressive multi-systemic nature of these conditions, which often necessitates the coordinated involvement of different medical professionals and specialties, thereby requiring a high level of integrated care. Various models for integrated care have been developed, including the Chronic Care Model (CCM) (Wagner et al., 1996, 2005) and *the Integrated Care Programme* (ICP) *for the Prevention and Management of Chronic Disease* (HSE, 2017) in Ireland, both of which emphasise the provision of patient-centered and coordinated care for this population in particular.

To date, most research on integrated care in Ireland has focused on acute or commonly occurring chronic conditions, such as cardiovascular disease, cancer, and type 2 diabetes (e.g., Darker, 2014; Darker et al., 2015). Relatedly, models of integrated care have primarily addressed these chronic conditions with well-established diagnostic markers and treatment protocols (HSE, 2017, 2021). While the HSE has published a vision for integrated care for rare disease (HSE, 2019), and work on care pathways is beginning to emerge (e.g., Ward et al., 2022), comparatively little is known about the provision of care for patients with under-recognised conditions or syndromes, including those that have historically been deemed “medically unexplained”, such as those described in previous chapters.

The studies conducted in phases one, two and three of this research indicate that patients with poorly understood syndromes, including FM, ME/CFS, and EDS among others, are routinely let down in their care. For example, as illustrated in Chapter 5, the phenomenological experience of healthcare among patients is one of disillusionment, frustration and deep distrust.

Highlighting the challenges of a fragmented system, patients describe “fighting” for care, “going in circles” yet “getting nowhere”, which leaves them feeling abandoned, undervalued, and ultimately betrayed by a service that was meant to help them. Furthermore, patient accounts reveal nuanced issues of power and voice, underscoring the imbalance in shared decision-making and ultimately, the lack of a truly patient-centered approach to care. Together with the issues highlighted by PPI Contributors involved in the previous phases of research, evidence suggests that improvements in care for this patient group are desperately needed. Further, involving patients in the design of care improvements is essential for creating systems that are both effective and responsive to real-world needs. Patient and consumer involvement are now widely recognised as a key aspect of developing clinical practice guidelines, driving quality improvements, and reforming healthcare systems (e.g., Armstrong et al., 2017; Fønhus et al., 2018; Sharma et al., 2017).

6.1.1 Objectives

Considering this, along with limited research in the Irish context, the present study seeks to *(i)* explore the experiences of patients living with diverse and poorly understood chronic conditions, specifically those related to healthcare; and *(ii)* to explore their needs and recommendations for improving the healthcare system in Ireland. Aligning with the values of participatory research and espousing patient partnership, the present study takes an initial step toward creating more patient-centered care by eliciting the views of service users, and additionally, involving patients in the co-analysis of collected data.

6.2 Methodology

6.2.1 Design

An exploratory, qualitative design with online focus groups (Krueger & Casey, 2014) was used, with data analysed using the Framework Approach (Ritchie & Spencer, 1994). The rationale and suitability of these methods is detailed in Chapter 2 (*Section 2.6.1*). Reporting follows the Consolidated Criteria for Reporting Qualitative Research (COREQ) guidance (Tong et al., 2007). Ethical approval for this study was granted by the School of Psychology Ethics Committee at Trinity College Dublin, Approval ID: SPREC032022-11 (see Appendix D1).

6.2.1.1 Patient and Public Involvement

Patient contributors played a crucial role in identifying and formulating potential research questions and objectives throughout the thesis, including this study, thereby ensuring the research addressed relevant issues from the patient perspective. As discussed in Chapter 2 (*Section 2.6.2*), this study was patient-led and co-designed by PPI contributors with poorly understood chronic illnesses. Two PPI contributors were involved in the process of collaborative data analysis as well as the interpretation of results, the selection of illustrative quotes and in manuscript write-up.

6.2.1.2 Sampling and Recruitment

Purposive and snowball sampling strategies were used to recruit focus group participants between September and December 2023. Individuals were eligible to participate if they were aged 18 years or older, living with a self-reported or formally diagnosed condition within the broad category of “medically unexplained” or “poorly understood” illness, and had received healthcare for their condition in Ireland. To reach the target population, the researcher utilized personal networks and shared the study online with patient communities and advocacy groups. A total of thirty-five individuals expressed interest in participating in the focus groups. Of these, one was not eligible to participate due to being a carer without lived experience of the illness, one did not consent due to technological difficulties, and eight did not respond to initial follow-up invitation. Twenty-five individuals consented to take part, though one withdrew due to decline in

health. A total of twenty-four participants were included in the study and focus groups were arranged in sequence, according to participant availability.

6.2.2 Procedure

6.2.2.1 Focus Groups

Participants were given a choice of taking part in-person or online focus groups, but all preferred online meetings due to travel distance, risk of COVID-19 infection and the energy expenditure required to attend in-person meetings. The focus groups were therefore conducted online, using the teleconferencing software Zoom. Five focus groups were conducted online in September, October and December of 2023, each lasting approximately 90 minutes. Groups consisted of between 3 to 8 participants and were not segregated by any characteristic but were homogenous for the experience of poorly understood illness. To maintain group dynamics, participants were encouraged to keep their video on and to speak spontaneously rather than sequentially, as they would during in-person gatherings. To maintain anonymity, each participant was afforded the opportunity to change their Zoom display name to a pseudonym before joining the group, though none chose to do so.

Basic demographic and health-related data were collected from each participant prior to participating in a focus group. Once participants provided informed consent, they received a link to an online questionnaire, hosted on the survey platform Qualtrics (Qualtrics, Provo, UT) and were asked to provide detail regarding their chronic illness and the primary symptoms experienced. Symptom severity, impact on quality of life and overall level of satisfaction with healthcare were rated using 5-item Likert Scales. The questionnaire is included in Appendix D2.

6.2.2.2 Moderation

Focus groups were led by the thesis author, an experienced moderator with lived experience of chronic illness, and supported by a co-facilitator with an interest in healthcare improvement (JM). It has been suggested that choosing moderators who share similar characteristics to group participants will promote rapport and a sense of safety, particularly when discussing sensitive topics (Morgan, 1993, 1995). While moderator's personal experience was disclosed to participants prior to the group in

order to maintain transparency and build trust, no further discussion or reference to this was made during the interview. The co-facilitator was involved in the first three focus groups; their role was to observe and take notes of group processes, and to debrief with the moderator following the interviews.

6.2.2.3 Interview Structure

Following the triangular structure for focus group questioning (Hurworth, 1996) each focus group began with the moderator reviewing the study purpose and discussing confidentiality, followed by a round of personal introductions. The moderator then initiated the focus group with an open-ended question, followed by a series of transition questions, leading to the key questions outlined in Table 6.1.

Within the triangular approach, different types of questions are strategically employed to steer participants from a broad discussion to key research questions: Opening questions are used to initiate discussion and gauge initial thoughts or experiences related to the topic, key questions explore central issues, while transition questions are used to bridge the different phases of discussion. Primary topics of discussion included participants' experiences of the patient journey, the ingredients of a positive patient journey, as well as specific recommendations for change in which the patient journey could be better supported.

This topic guide was developed based on previous literature and consultations with the patient panel and individual PPI contributors in earlier phases of the research. Throughout the focus group sessions, participants were encouraged to speak spontaneously, share their ideas without pressure to reach consensus, and freely express agreement or disagreement with the issues discussed. The moderator adopted a facilitation strategy based on principles of active listening (Rogers & Farson, 1957), which include techniques like reflecting, paraphrasing, and summarizing the speakers' thoughts. The closing question encouraged any participant in the group to share their reflections and insights on the discussion, or to provide additional information they wished to add.

Table 6.1

Focus group interview guide

Question Type	Question
Opening	As a person with a poorly understood condition, what has your healthcare journey been like?
Transition	Were there any positive experiences, and what made them so? Were there any negative experiences, and what made them so? What has helped you the most in your journey? What was the biggest barrier or challenge in your journey?
Key	What do you think are the key features of a good patient journey or good healthcare system? What would be the perfect healthcare experience? Based on your personal experiences, can you suggest one major improvement? How might it be achieved?

6.2.2.4 Data Extraction

All focus groups were recorded via Zoom and only the audio files were downloaded and retained. The audio content was transcribed verbatim by the moderator, with all potentially identifying information (e.g., clinician names) removed. Each audio recording was erased following transcription, and an anonymized transcript was sent to participants for member-checking. Participants were invited to choose a pseudonym to be included in the manuscript, or alternatively, one would be assigned to them. Participants were asked to review the transcript and invited to redact content that they felt did not reflect their experience or opinion. Only one such redaction was requested, with the participant making a correction regarding potentially identifying expressions.

6.2.2.5 Data Saturation

A reflective journal was maintained by the moderator following each focus group. While formal analysis was not yet completed at this stage, key issues arising from the discussion were noted and patterns in content observed, allowing the identification of novel material. After the fourth focus group, the researcher experienced that no new information emerged. In order to confirm that data saturation had occurred, a fifth focus group was conducted, verifying saturation and thus concluding data collection.

6.2.3 Data Analysis

Two types of data analysis were conducted, based on the methodological concerns regarding unit of analysis, as discussed in Chapter 2 (*Section 2.6.1.1*). According to Morgan (1996), focus group data can be analysed by concentrating either on the content of what participants say or on the process of interaction among participants, i.e. the "what" and the "how". Focusing on discussion content, qualitative data were analysed using the Framework Approach (Ritchie & Spencer, 1994), while group dynamics were explored visually by constructing Sociograms (Drahota & Dewey, 2008). An analysis of dynamics was conducted to ensure that interpretations and recommendations were representative of multiple viewpoints, rather than being skewed by any dominant voices within the groups.

6.2.3.1 Group Dynamics

An analysis of participant interactions was conducted through the construction of sociograms. The Sociogram is a quantitative and visual tool that can be used to examine relationships between group members, as well as overall group structure or dynamics (Huang et al., 2006). When used alongside transcribed data, sociograms enable researchers to 'see' group processes, including who is shaping and dominating group discourse (Baiardi et al., 2015). We followed the methodology described by Drahota and Dewey (2008) who provide an example of using sociograms to examine the flow of conversation between focus group members. Sociograms for each focus group were constructed sequentially by ND and JM during data collection to explore interactions between participants. These were constructed as data collection progressed, allowing for ongoing reflection on moderation strategy. Drawing on the work of Baiardi et al. (2015), we also used sociograms to explore and reflect on issues of power and the role of the

focus group moderator. The PPI contributors involved in co-analysis of interview transcripts, discussed below, were not present during data collection and were therefore not involved in this process.

Sociogram Construction and Analysis Procedure

To construct sociograms, we calculated and represented the contribution (words spoken) and directionality of interactions between all participants, including the moderator, throughout the entire interview. Weighted arrows between participants were used to illustrate flow of conversation, with weightings related to the number of interactions between people. We also weighted the participants' overall contribution (% of total words spoken) to the focus group discussion, indicating their dominance on the sociogram. Once interactions were visualised, we characterized the groups by the response patterns described by Drahota and Dewey (2008): *(i)* a *"perfect"* group with evenly weighted and distributed arrows; *(ii)* a *"normal"* group with non-symmetrical, irregular arrows; *(iii)* a serial *"serial turn-taking"* group, with flow regularly passing from one person to another, or *(iv)* a *"serial interviewing"* group, with heavily weighted moderator to participant arrows, and less exchange between other members. As groups were conducted online, we excluded the *"proximal turn-taking"* pattern proposed by the authors. Interpretation of sociograms was supplemented by reviewing reflective notes from each interview.

6.2.3.2 Framework Analysis

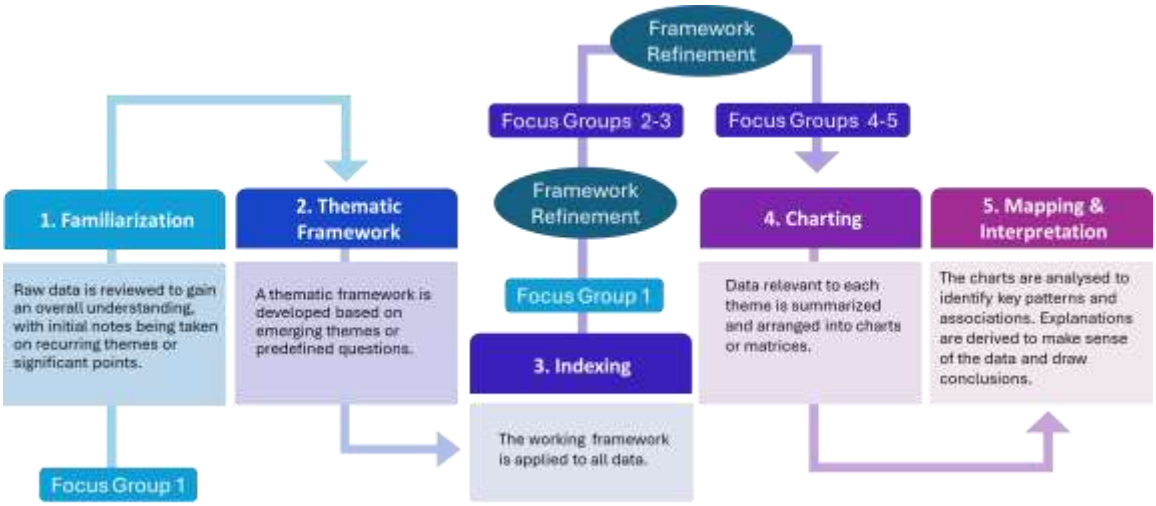
Focus group discussion content was analysed using Framework Analysis. Initially developed for use in applied policy research, Framework Analysis has become a well-established method in multi-disciplinary healthcare research (Furber, 2010; Gale et al., 2013; Parkinson et al., 2016; Ward et al., 2013). The approach is highly systematic and valued for its rigor and adaptability; it also facilitates the organisation and comparison of various types of data, including observations, policy documents, case studies, individual interviews and focus groups (Goldsmith, 2021). Framework Analysis holds advantages of both deductive and inductive approaches, making it suitable for answering specific questions while allowing the development of new ideas (Gale et al., 2013). The method involves five key steps that allow researchers to uncover and interpret complex patterns and themes in a systematic manner: *(i)* familiarization, *(ii)* identifying a thematic framework, *(iii)* indexing, *(iv)* charting, and finally, *(v)* mapping and interpretation (Ritchie & Spencer, 1994).

Framework Analysis Procedure

Anonymized focus group transcripts were collaboratively analysed by the moderator and author (ND), research assistant (JM) and two patient contributors (LM, EF), both of whom were not previously involved in the research. Audio recordings were not shared with contributors to maintain the confidentiality and anonymity of focus group participants. Each team member analysed a combination of three of five focus group during this process. Steps of the approach are outlined in Figure 6.1, which shows the sequence of analysis steps as applied to the five group transcripts.

Figure 6.1

Steps of the Framework Approach applied to focus group interviews (N=5)



During the familiarization phase, all members of the team (ND, JM, EF, LM) reviewed and coded the first interview (Focus Group 1) noting key ideas and possible themes in the transcript. A thematic framework was developed collaboratively by all to capture these insights. We agreed to organise the thematic framework and subsequent revisions according to three broad categories: *(i)* Participants' experiences of living with chronic illness, *(ii)* Participants' experiences of healthcare, and *(iii)* Participants' needs and recommendations for improving healthcare. Framework development was an iterative process, with three revisions made before a final structure was reached. The initial thematic framework was applied back to the first interview by JM, LM and EF through indexing. Revisions were agreed upon by the team and the refined framework was applied to the other interviews by pairs of team members, with weekly analytic meetings to guide the process. Each member of the team completed charting summaries which were then discussed and finalized in meetings. Charting summaries were mapped by ND and JM onto a single matrix representing all five focus groups. A detailed worked example of the co-analysis process is provided in Chapter 7.

Drawing on Goldsmith's (2021) worked example, a "strength" rating was assigned to each theme, reflecting the strength of signals from the data within groups. To achieve this, ND and JM reviewed the mapping matrix and charting summaries for each focus group, focusing on the relative dominance of each topic in the discussion. Themes were rated as *high*, *medium* or *low* intensity, depending on whether they were perceived as a strong component of the focus group, an occurring component but not a dominant thread, or a weak component of the discussion. Importantly, this rating was not derived quantitatively through an analysis of frequency, but rather the impression of supporting evidence charted by the co-analysts. The mapping matrix and strength ratings were initially reviewed independently by all team members, followed by collaborative interpretation and agreement on results. Word templates for all steps of the process were developed by ND to ensure consistency of indexing and charting among team members. Charting summaries were collated in an Excel spreadsheet for mapping and interpretation.

6.3 Results

The results section is structured as follows: *(i)* description of participant demographics, *(ii)* an analysis of focus group dynamics, and *(iii)*, results of the framework analysis and thematic discussion.

6.3.1 Participant Demographics

A total of twenty-four participants (4 male, 20 female), aged between 21 to 63 years (*M*=47.9, *SD*=11.3) took part in the five online focus groups. The composition of four groups was mixed-gender while one group, Focus Group 1, consisted of all females. The demographic composition of groups is summarised in Table 6.2 and further detailed in Table 6.3. Most participants across the groups were educated to a higher level (79%, *n*=19). Just 25% (*n*=6) were currently employed, with a further 29% (*n*=7) employed and on sick leave. Collectively, participants reported living with over 65 different conditions and were homogenous in terms of living with a poorly understood illness, detailed in Appendix D3. Most participants reported living with 2 to 4 conditions (*M*= 4.25, *SD*= 3.34), though six participants (25%) reported five or more diagnoses, thereby illustrating that multimorbidity was common in the sample. Fibromyalgia Syndrome or Chronic Pain (38%, *n*=9), ME/CFS (29%, *n*=7) and POTS (20%, *n*=5) were the most frequently reported conditions across the groups. Overall, participants most frequently indicated living with symptoms of moderate severity or higher and rated the impact of these symptoms on quality of life as significant to very significant (83%, *n*=20). Only four participants (17%) were somewhat or extremely satisfied with the overall quality of their healthcare journey.

Table 6.2

Focus group composition and demographic characteristics

	Focus Group					
	1	2	3	4	5	*Total*
	(*n*=5)	(*n*=4)	(*n*=4)	(*n*=8)	(*n*=3)	*N*=24
Demographic Characteristics						
Age						
M (*SD*)	44.6 (*11.7*)	50.5 (*5.7*)	48.7 (*15.7*)	46.25 (*12.6*)	53 (*11.8*)	47.9 *(11.3)*
Gender						
Male	0	1	1	1	1	4
Female	5	3	3	7	2	20
Educational Attainment						
Primary Level	0	0	0	1	0	1
Secondary Level	0	0	0	0	0	0
Training (Post-Secondary)	0	0	1	2	0	3
Third Level	5	4	3	4	3	19
Prefer not to say	0	0	0	1	0	1
Employment Status						
Employed	2	2	2	0	0	6
Student	1	0	1	1	0	3
Unemployed	0	1	1	0	0	2
Retired	1	1	0	2	0	4
Sick Leave/Disability	1	0	0	4	2	7
Other: Carer	0	0	0	1	0	1
Prefer not to say	0	0	0	0	1	1
Health and Healthcare						
Conditions reported						
Number Reported	15	8	8	42	13	66
Fibromyalgia/Chronic Pain Syndrome	2	1	3	3	0	9
ME/CFS	2	2	1	1	1	7
POTS	1	2	0	2	0	5
Symptom Severity						
Mild to Moderate	5	2	4	3	2	16
Severe to Very Severe	0	2	0	5	1	8
Impact on Quality of Life						
Mild or Moderate	2	0	1	1	0	4
Significant or Very Significant	3	4	3	7	3	20
Satisfaction with Healthcare Journey						
Satisfied	0	1	1	1	1	4
Neutral	2	0	0	2	0	4
Dissatisfied	3	3	3	5	2	16

Table 6.3

Focus group participant demographics (N=24).

FG	Participant (Pseudonym)	Gender	Age Range	Conditions Reported [a]	Symptoms	Symptom Severity (self-rated)	Impact on QoL (self-rated)	Satisfaction with Healthcare
1	Millie	Female	35-44	ME/CFS, Long Covid	Physical, Cognitive	Moderate	Significant	Neutral
	Amy	Female	55-64	Chronic Lyme Disease, FM, Sjögren's Syndrome, POTS.	Physical, Cognitive	Moderate	Significant	Extremely dissatisfied
	Sarah	Female	25-34	Bladder Pain Syndrome	Physical	Moderate	Moderate	Extremely dissatisfied
	Julia	Female	45-54	ME/CFS, FM, EDS, Lyme Disease	Physical, Cognitive, Affective	Moderate	Moderate	Extremely dissatisfied
	Chloe	Female	45-54	Long Covid	Physical, Cognitive, Affective	Moderate	Significant	Neutral
2	Anna	Female	45-54	ME/ CFS, POTS, MCAS	Physical, Cognitive	Mild	Very significant	Extremely satisfied
	Ellen	Female	35-44	Lyme Disease, FM	Physical, Cognitive, Affective	Severe	Significant	Extremely dissatisfied
	Alex	Male	45-54	Chronic Lyme Disease	Physical, Cognitive, Affective	Severe	Very significant	Somewhat dissatisfied
	Emily	Female	45-54	ME/CFS, Long Covid, POTS	Physical, Cognitive	Mild	Significant	Extremely dissatisfied
3	Sophie	Female	55-64	ME/CFS, MCAS	Physical, Cognitive, Affective	Moderate	Significant	Extremely dissatisfied
	Maureen	Female	45-54	FM	Physical, Cognitive, Affective	Moderate	Significant	Extremely dissatisfied
	Liam	Male	25-34	FM	Physical, Cognitive	Moderate	Moderate	Somewhat satisfied
	Ashley	Female	45-54	FM	Physical, Cognitive, Affective	Moderate	Significant	Somewhat dissatisfied
4	Niamh	Female	18-24	EDS, POTS, MCAS, Migraine	Physical, Cognitive, Affective	Very severe	Very significant	Extremely dissatisfied
	Kelsey	Female	45-54	EDS, POTS	Physical, Cognitive, Affective	Severe	Significant	Somewhat dissatisfied

FG	Participant (Pseudonym)	Gender	Age Range	Conditions Reported [a]	Symptoms	Symptom Severity (self-rated)	Impact on QoL (self-rated)	Satisfaction with Healthcare
	Robert	Male	35-44	FM	Physical, Cognitive, Affective	Mild	Moderate	Neutral
	Laura	Female	55-64	Chronic Pelvic Pain, Lipoedema	Physical, Cognitive, Affective	Moderate	Very significant	Extremely dissatisfied
	Caitlyn	Female	45-54	Centralised Pain Processing Disorder, IBS/IBD	Physical, Cognitive, Affective	Severe	Very significant	Neutral
	Amber	Female	45-54	Chronic Pain Syndrome	Physical, Cognitive, Affective	Severe	Very significant	Somewhat dissatisfied
	Mia	Female	45-54	FM, Long Covid, Lupus, Chronic Migraine	Physical, Cognitive, Affective	Severe	Significant	Somewhat satisfied
	Lucy	Female	45-54	ME/CFS	Physical, Cognitive	Moderate	Significant	Extremely dissatisfied
5	Shane	Male	55-64	Hereditary Neurodegenerative Disorder [b]	Physical, Cognitive, Affective	Mild	Significant	Somewhat dissatisfied
	Caroline	Female	35-44	Sjögren's Syndrome, CFS, Chronic Pain Syndrome	Physical, Cognitive	Severe	Very significant	Somewhat dissatisfied
	Dawn	Female	55-64	Hereditary Neuromuscular Disorder [b]	Physical, Cognitive, Affective	Moderate	Very significant	Extremely satisfied

Note. [a] Conditions meeting inclusion criteria (i.e., poorly understood chronic illness); [b] Rare disease, details redacted for confidentiality. Abbreviations: FG (Focus Group), FM (Fibromyalgia Syndrome), ME/CFS (Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome), POTS (Postural Orthostatic Tachycardia Syndrome), MCAS (Mast Cell Activation Syndrome), EDS (Ehlers-Danlos Syndrome(s)), IBS/IBD (Irritable Bowel Syndrome/Disease)

6.3.2 Group Dynamics

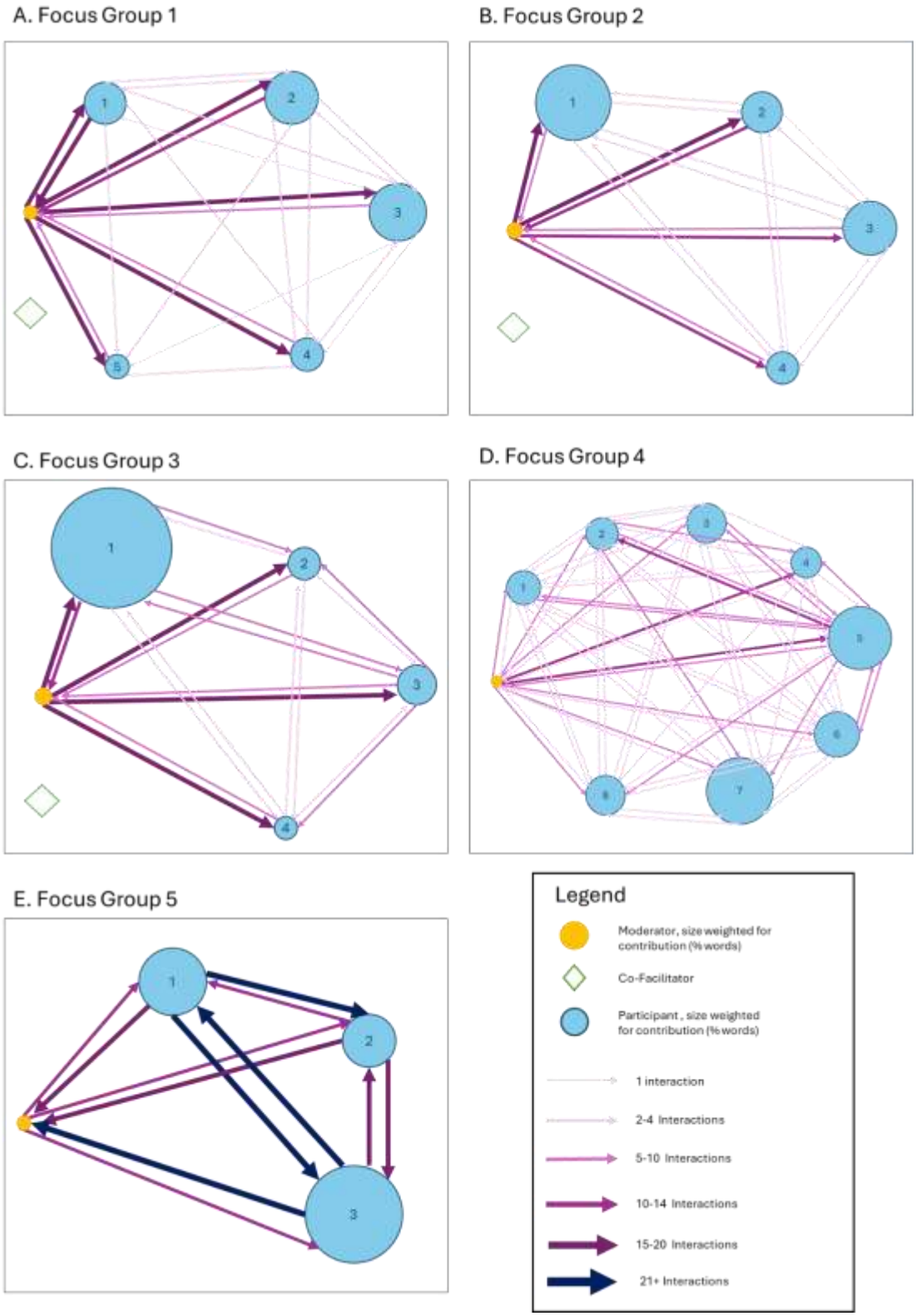
Sociograms were constructed to explore participant interactions in each focus group and are presented in Figure 6.3. Overall, interactions between most participants were bi-directional, indicating good group dynamics and cohesion of the groups. All five focus groups exhibited a "*normal group*" structure characterized by non-symmetrical and irregular interactions among all participants, including the moderator. Moderator input aside, Focus Group 2 (Fig. 6.3, Panel B) was most akin to the "*perfect group*" structure, with regular and symmetrical interactions between group participants. While the majority of participants actively engaged with one another during the interview, the sociograms revealed that certain participants were less embedded in the discussion. For instance, one-directional interactions were observed with Participant 5 in Focus Group 1 (Panel A) and Participant 4 in Focus Group 4 (Panel D).

Focus Group 5 (Panel E), the smallest group (*n*=3), displayed the strongest interactions between participants and less interaction with the moderator, suggesting a well-flowing discussion with ample elaboration among participants. The heavily weighted feedback from Participant 3 to the moderator in this group reflected Participant 3's role in summarizing the discussion, reducing the need for clarification from the moderator. In all focus groups except Focus Group 4, the moderator's interactions were evenly dispersed among participants, reflecting a tendency for broad, group-wide questions rather than serial questioning of individuals. In contrast, interactions in Focus Group 4 were less balanced. Upon reflection, the larger group size (*n*=8) seemed to encourage participants to engage in discussions unrelated to the interview questions, necessitating more frequent re-direction from the moderator.

In addition to mapping interactions, we also visualised participants' relative contributions to the focus group discussion by analysing word count (Appendix D4). As expected, certain participants led the discussion, particularly in Focus Group 3 (Panel C), where Participant 1 contributed over 50% of the conversation. The moderator's input ranged between 3% and 7% of the total word count, reflecting a balanced approach that allowed participants to steer the discussion while still providing structure.

Figure 6.2

Focus Group Sociograms



Note. Participant and moderator size is weighted for number of words contributed to the discussion, suggesting dominant voice in the group. Arrows are weighted for number of interactions between participants, reflecting development of ideas during the discussion. The co-facilitator in Focus Groups 1-3 had no verbal input into the group and functioned as an observer, debriefing with the moderator following the interview.

6.3.3 Framework Matrix

Framework analysis resulted in a final matrix of 11 themes organised into three broad categories. Results are described below, grouped according to the two research questions. The category *Experience of Chronic Illness* addresses the participants' lived experiences of their condition and encapsulates symptom-related challenges, coping strategies, and the social implications of poorly understood illness. *Health experiences* reflects participants' accounts of their interactions with the healthcare system, their overall experiences of the journey, as well as barriers to effective care. The category *Needs and Recommendations* addresses participants' aspirations and solutions for improvements in their care. The framework mapping and relative thematic strength across all five groups is presented in Figure 6.3. The charting matrix, summarising thematic findings across all groups, is included in Appendix D5. Themes are described below with illustrative excerpts extracted from the interview transcripts. Additional quotes supporting each theme are included in Appendix D6.

Figure 6.3

Framework Analysis Mapping

Category	Theme	Subtheme	Focus Group 1	2	3	4	5
Experience of Living with a Chronic Illness	Living with chronic illness	Burden of illness/disease	***	***	***	***	***
		Coping with chronic illness	***	***	***	**	***
		Positive outlook	0	*	0	*	*
	Community and Social Support	Value of community	***	**	0	***	**
		Positive interpersonal experiences	**	*	*	*	*
		Negative interpersonal experiences	*	***	**	*	**
Healthcare experiences	Experiences with healthcare	Positive experiences (clinicians)	***	***	***	**	**
		Positive experiences (other professionals)	*	*	***	**	*
		Negative experiences (clinicians)	***	***	***	***	***
		Negative experiences (other)	0	*	0	0	0
		Negative (treatments/therapy)	**	**	**	**	**
		Power dynamics	*	0	***	***	***
	The patient journey	Negative experiences of the journey	***	***	***	***	***
		Positive experiences of the journey	*	*	*	*	*
		Systemic issues	*	**	*	***	***
		Burdens associated with healthcare	**	***	***	***	***
	Barriers to effective care	Lack of continuity & poor coordination	***	***	**	***	***
		Inconsistency of care	**	**	**	**	*
		Resources	*	**	*	**	**
		Stereotypes/ Assumptions	***	***	***	***	***
		Financial constraints	*	*	0	***	**
		Social welfare	*	*	0	*	*
Needs and Recommendations for Change	The consultation	Patient-clinician relationship	***	**	***	**	***
		Guidance / Signposting	**	*	*	**	***
		Supports at different stages	**	*	**	**	**
		Improved knowledge	**	**	*	**	**
		Improved understanding	*	*	**	**	**
		Improved efforts	0	*	*	*	0
	The service	Comprehensive care	**	**	*	***	**
		Personalised care	*	*	*	**	*
		Improved coordination	***	**	*	***	**
		Linking beyond healthcare	*	*	0	**	*
		Increase resources & incentives	0	*	0	0	**
		Diagnostic services	0	**	0	*	**
	Training and development	Knowledge/Understanding	**	*	*	*	*
		Continuous professional development	*	*	*	*	*
		Medical training	*	0	*	**	**
	Clinician wellbeing	Targeting burnout	*	0	*	**	**
		System/Culture change	*	0	*	*	**
	Knowledge generation	Need for research	*	*	*	*	**
		Lived experience/patient voice	*	*	**	*	**
		Public awareness	0	*	0	0	*
	Implementation	Coordinated efforts	0	**	0	*	0
		Application of research/knowledge	*	*	*	0	*

Note. Colours depict strength signal per theme in each focus group.

*** *Indicates high intensity theme/subtheme (strong component of interview)*
** *Indicates medium intensity; occurs but not a dominant thread*
* *Indicates low intensity (component present but not often or is implied)*
0 *Indicates no data supporting this component is present*

6.4 Findings

6.4.1 Category I. Experiences of Living with Chronic Illness

Across all five groups, participants often discussed the *Burden of Illness or Disease*, referring to the widespread impact of chronic illness on their lives, as well as strategies for *Coping With Chronic Illness* and managing their symptoms, both of which were the highest intensity themes. While some participants discussed developing *Positive Outlooks* and adjusting to their condition, this topic was mentioned infrequently, and not at all in two of the groups (See Figure 6.3). Participants in four of five groups discussed the *Value of Community,* emphasising the role of advocacy groups and patient communities in their journey. Several participants across the groups gave examples of *Positive Interpersonal Experiences,* noting instances of support from family, friends and employers, though such accounts were generally weak and infrequent. *Negative Interpersonal Experiences* were more common and were an important theme in three groups, and a weaker component of two. Thematic findings are discussed below.

6.4.1.1 Theme 1: Living with a chronic illness

The experience of life with a poorly understood chronic illness was an important theme shared by participants across all groups. All described the multifaceted impacts of their conditions on their lives, often referring to physical symptoms such as persistent pain, fatigue and cognitive symptoms collectively referred to as “brain fog”. In all groups, participants also expressed sadness and frustration over the loss of their health, passions, relationships, ability to work and the future they had once anticipated. They highlighted the invisible and dynamic nature of their conditions, with periods of remission, symptom flares and debilitating crashes, and often bonded with other participants over their lived experiences.

> *Emily was talking about the crashing. And I was in the crash for five months. And if I did something that, if I had a conversation with somebody for 10 minutes, I couldn't function the next day, at all. Like that's how wiped I was. There was no healthcare, that was all finding stuff to help myself, pretty much alone. (Ellen, Group 2)*

Despite diversity in their conditions and symptoms, several participants were taken aback by similarities in their overall experience.

> *What struck me is how similar we all are. Even though our conditions are quite different. (Amy, Group 1)*

Many described having to shape their lives around their illness, which not only affected daily activities but also long-term decisions around childbearing, career choice and marriage. Participants' narratives revealed a poignant picture of life restricted by illness, accompanied by a sense of wasted potential and personal failure.

> *And we're all banjaxed. Not banjaxed enough to be in a hospital, not banjaxed enough to be fully on disability, but banjaxed enough to- as Amy said- you know, you have to pare back your life, live defensively, and kind of go "right, what's my energy envelope for this" (Julia, Group 1)*

> *But it's kind of like, I can't talk to anybody. I didn't have a social life. There's no, I don't do anything. I go to work (…) But I don't do anything because I don't have any energy to do anything else. I don't have a life. D'you know what I mean? "How are you doing?", "Umm, just don't ask me. Because I can't answer you. Because it's crap. (…) I don't have any news about achievement. Because I haven't achieved anything other than surviving." (Sophie, Group 3)*

While cognitive and physical symptoms were most frequently discussed, participants in the groups also referred to mental health impacts of chronic illness. However, they carefully emphasised the secondary nature of psychological symptoms, stressing that poorly understood conditions are not psychogenic illnesses. Anxiety and depression were an outcome of loss, uncertainty and systemic ableism:

> *I'm 20 years old, I've just found out I have a chronic illness for the rest of my life that I can't do anything about, how am I... How am I supposed to live my life? Because it's very much like there's a, there's an effect on your mental health as a result, and I think that gets forgotten about. And I know, for me, I'd be almost slightly reluctant to bring it up in those situations, because of the kind of stigma or idea that it's all in your head, because it's not (Liam, Group 3)*

Others, particularly those with ME/CFS, Long Covid or Lyme Disease, explained their symptoms were a direct outcome of neuroinflammatory sequelae, post infection.

> *Yeah, there is a problem. But the problem has been caused by the disease. (…) I'm always very careful to say, "look, yeah, I'm not, I'm not that happy person I used to be, but I'm coping fine, generally. But my brain is not operating normally, because it got infected. There is an infection". (Alex, Group 2)*

Participants often described developing unique coping mechanisms and "home-made solutions" to manage their symptoms, largely due to a lack of support and guidance from medical professionals. They discussed facing a steep learning curve, initially hiding their symptoms or pushing through pain and fatigue to compensate for the limitations of their illness. Such resistance often led to the exacerbation of symptoms. Learning to pace themselves and constantly monitor energy levels was a significant burden for many participants.

> *Because even though, to be honest, it's after taking me three years to get that into my head, to not batter my body, because my head thinks that I can still do all the things they always did, I can think the way I did, I thought I could go hiking or whatever. But the fact of the matter is, I couldn't. (Anna, Group 2)*

For most, the process of adjustment was complicated by the prognostic uncertainty of their illness:

> *I would actually prefer to have a terminal condition because at least you know, it's over. But this business of you're just going to be in chronic pain, trying to come up with weird ways to kind of get through the week, I think that's far worse (Julia, Group 1)*

Despite significant hurdles, a small number of participants from three focus groups also reflected on their personal growth and resilience, expressing hope and optimism for recovery or advances in medicine. However, such accounts were infrequent and usually provided by those who had succeeded in managing their symptoms.

> *It's... your life's not over basically, there are things you can do. Plenty of people live with this condition and manage it (Liam, Group 3)*

6.4.1.2 Theme 2: Community and Social Support

An important though less consistent category of findings related to experiences of the social context, whereby the daily challenges faced by participants were either compounded or alleviated by their social experiences. Unfortunately, negative or adverse interpersonal experiences were common, and many participants discussed a lack of social support as key components of their illness experience. Generally, participants felt misunderstood by others, and discussed how they often faced questions about the legitimacy of their illness. They spoke of being accused of laziness and malingering, and discussed how family and friends often misattributed their symptoms to mental illness:

> *Yeah, that's, that's…the biggest challenge is the perspective from people that don't know about these unusual kinds of illnesses. I'm definitely in the "Mad" group. Particularly with my family, because they phoned my GP -two members, an aunt and a sister- and asked...would he kind of send me away to one of those, you know...psychiatric facilities. (Ellen, Group 2)*

A lack of compassion among friends and relatives was especially difficult to deal with, and many discussed others' apparent indifference, at the same time highlighting the burden of being ill.

> *Yeah, and it's really hard to, it makes me feel like bawling, crying. Basically, we are the living dead, like we just fall out of life and nobody notices. And it's not their fault. I don't expect them to understand because I know if I was in their position, I wouldn't understand and I might doubt as well. (Anna, Group 2)*

> *And you see, because we're not terminal, no one cares. But if you kind of middle aged women who are just a bit tired, and that's what they see that it's seen as a psych condition, because it's not terminal (Julia, Group 1)*

Though less common, some participants also shared stories of positive experiences with others. They spoke of practical and emotional support from friends and family, and some were offered accommodations from their employers. Participants commented on an increasing level of empathy and understanding which emerged as a consequence of the COVID-19 pandemic:

> *And then when COVID came along, people started to really kind of say, "Oh, so this is real", (…) People were looking at me and saying, "Yeah, we understand now what you were going through". Now, it doesn't make it any easier. (Alex, Group 2)*

In four of five groups, participants reported seeking support from patient communities and advocacy groups and highlighted the positive effects such networks had on their wellbeing. For many, virtual communities were the sole avenue for reducing isolation. They emphasised how these groups fostered a deep sense of belonging, creating a supportive space where they felt both validated and understood. Connecting with peers offered participants an opportunity to share personal learnings and insights, and many spoke of exchanging “tips and tricks” for managing their conditions as well as navigating the healthcare journey. The educational materials and self-management courses offered by registered charities were also recognised as a valuable resource.

> *And without the groups on Facebook, you're really lost at sea on your own. And you'll wonder "Will I ever get out of this?" (Ellen, Group 2)*

> *And then, the HSE has, actually, developed a lot of courses, and I've done some of their courses with Chronic Pain Ireland. And they've been really, really supportive. And I've actually made friendships from them that I've carried on seeing, a support outside of that (Robert, Group 4)*

6.4.2 Category II. Healthcare Experiences

When asked about their experiences of healthcare, *Negative Experiences with Clinicians* emerged as the predominant theme across all focus groups. All participants in each group recounted a range of issues they had faced, such as feeling dismissed, not being listened to or heard by GPs and consultants. Participants in all groups also discussed *Positive Experiences with Clinicians,* though this theme was less consistent and highlighted primarily by Groups 1, 2 and 3. Participants also shared their experiences with allied health professionals, such as psychologists, occupational therapists, and physiotherapists. *Negative Experiences* with these professionals were mentioned briefly by participants in only one group, while *Positive Experiences* were more commonly reported across the groups. A number of participants across all five groups discussed their *Negative Experiences with Treatments or Therapy*, whereby they felt interventions were ineffective, harmful or not tailored to their needs. Multiple participants emphasised issues related to *Power Dynamics* in healthcare settings, though this theme was weaker in Group 1 and not mentioned at all by participants in Group 2.

Further, all participants discussed their *Negative Experiences of the Journey,* a theme reflecting issues of cost, journey length, and a general sense of lack of support. Similarly, most participants discussed *Burdens Associated with Healthcare,* or the negative impacts of the healthcare journey on their lives and wellbeing. While *Positive Experiences of the Journey* were less frequent, a small number of participants across all groups mentioned positive aspects of their care, such as meeting helpful individuals along the way. *Systemic Issues*, or challenges related to the organisation and responsiveness of the healthcare system, were discussed by the majority of participants but mostly by those in Groups 4 and 5.

With regards to barriers to effective care, *Stereotypes and Assumptions* were a key challenge highlighted by most participants across all groups, reflecting a high intensity theme overall. The majority of participants also emphasised the challenges arising from *Lack of Continuity of Care and Poor Coordination* between and within services, as well as inconsistent testing, diagnostic and referral practices within services, captured by the theme *Inconsistency of Care.* Other barriers to care, such as limited *Resources,* personal

Financial Constraints and challenges of navigating and accessing *Social Welfare* were discussed, though with less consistent emphasis across the groups.

6.4.2.1 Theme 1: Experiences with healthcare

Speaking of their healthcare experiences, as mentioned, participants often reflected on their interactions with general practitioners (GPs), specialists and other healthcare professionals, as well as their overall experiences of the patient journey. Despite mixed accounts, the overall picture was negative with supportive and understanding clinicians being an exception. Participants often felt their plight was unappreciated by healthcare practitioners, who appeared to lack compassion and investment in their care:

> *I think doctors and other health professionals forget that we're the ones who have to go home at the end of the day and live with this. They don't, they're seeing people in and out of their office, they don't have to go home and deal with it at the end of the day (Niamh, Group 4)*

Generally, negative interactions with GPs and Consultants were discussed far more extensively than interactions with other professionals, like nurse practitioners, though some participants also noted suboptimal encounters with allied health professionals (e.g., Occupational Therapists and Physiotherapists). A notable issue was healthcare professionals' unwillingness to engage in dialogue, listen to patient needs and involve them in decision-making. Participants in all groups described being dismissed, disbelieved and invalidated.

> *Now, I don't want to doctor bash, but you know, it's been, like Millie said, I've had mixed experiences. I've met some lovely people along the way. But like I had one consultant in particular who I was paying privately and paying a lot of money. And it was a year and a half in and it wasn't working. And I said, "Look, I don't think this is working. I think there's an issue with my stomach". And, and he went "who's in charge here?" (Julia, Group 1)*

Clinicians appeared reluctant to consider their patients' perspectives and knowledge, presumably due to a difference in status:

> *We're very good at figuring stuff out, but nobody's listening to the stuff that we're figuring out. (Julia, Group 1)*

> *So if you're, if you're, unless you're medically trained, if you go in and say "I'm a doctor, I'm a nurse", that's fair enough, (…) But when you're not medically trained. And you use the lingo because you've self-educated yourself. And that's such a red herring for some of them (Mia, Group 4)*

Appointments were often a source of anxiety, contributing to participants' reluctance to engage with services:

> *So I'm literally trying to build up the courage to go back and see him. So that's that would explain high anxiety, for a very good reason (…). (Sophie, Group 3)*

Participants also described how repeated invalidation and disbelief affected their self-confidence, leading them to doubt their own judgment and experiences. Over time, an accumulation of negative experiences eroded their trust in the Irish healthcare system:

> *I think that was a really important point there about like the not being believed. I think that is it's actually almost like a form of torture like that, You feel like you're going mad, that like it, and I did begin to question like, is this in my head? (…). Because I, because I doubted myself and trusted her medical expertise, I did what I knew was wrong, like, I could feel that this was the wrong thing to do, you know. And I think that's what not being believed does, that you actually don't even trust yourself anymore. (Caroline, Group 5)*

> *So these people are portrayed to us as the people we're supposed to trust and go to when we are in the worst of our worst and in dire needs. (…) And then when you go into that environment, and you're being accused yourself or not being believed, and that you're making it up, how are you supposed to want to go back there to get help? (Niamh, Group 4)*

Participants often attempted to explain clinicians' dismissive attitudes and behaviours as defensive, suggesting that poorly understood conditions pose a threat to their expert identity.

> *So rather than them saying, "I actually don't know with, with the limited knowledge with the knowledge that I have, I do not know what's wrong with you." They seem to go down the route of actually invalidating and dismissing the patient. (Lucy, Group 4)*

Negative interpersonal experiences with healthcare professionals were further compounded by adverse experiences with various treatments, therapies and diagnostic procedures. Participants recounted numerous medical misadventures, giving examples of rushed solutions offered without adequate investigation. Participants felt that clinicians followed a "one size fits all" approach, with little consideration for individual needs or circumstances. Fragmented, symptom-based treatments often resulted in secondary complications.

> *(…) I was put on morphine, and I was put on fentanyl, I was pushed, and I was put on morphine for six years. And all these things that actually led to more syndromes (Chloe, Group 1)*

When declining particular routes of treatment, some participants were accused of non-compliance, labelled as problematic, or threatened with discharge from services.

> *And again, it was put down to me being a difficult patient. Not that these drugs weren't actually suitable for me to manage my health. (Julia, Group 1)*

Sweeping recommendations for increased physical activity were particularly concerning for patients with post-exertional malaise:

> *And they said they reckon I was fit for a 45-minute exercise class. I said, "Are you sure about this? Because I have extreme post-exertional malaise". I said, "and I have", you know, "I'm very debilitated afterwards. And in pain from the neck down". They assured me I was. This was after emails and phone calls to confirm. I thought, "well, this is the Long COVID clinic in a very prominent institution, they must what they're talking about". And I did the 45-minute exercise class. I did, I couldn't get out of bed the next day, surprise, surprise (…) And when I subsequently said it back to the Senior Consultant when I got back into Long COVID clinic, he said "Yeah, it's not for everybody." Now, exercise, it's for nobody with ME. (Lucy, Group 4)*

A core theme of power dynamics came to the fore in participants' accounts of their experiences with healthcare. Reflecting on their encounters, many felt that clinicians interacted with them from a position of superiority and acted as gatekeepers of information.

> *I think some of them just think that their patients are stupid, and that they are just smarter. Some of them, I think some of them are very nice. They are genuinely very nice. But I do think some of them just have a very overly, you know, they just think that they're very superior (…) You know and there's a, there's a power differential (Sophie, Group 3)*

Such an attitude was evident particularly when participants made suggestions about diagnostics and treatments based on personal research. In a number of groups, participants discussed being ridiculed for using *"Doctor Google"* to make sense of their symptoms, yet noted that *"Often they're on Google themselves"* (Liam, Group 3)

> *They don't like that, they don't like when you, it's like…. If they're not helping you. Say you, like, look things up yourself. And then like, you'll be like, "Oh, I read this medical study" where you're Googling, like every word that you don't understand. And, you know, like, "this is what I think is happening to me." They're like, like, "don't use Google". It's like, they don't want to help you. But then they don't want you to look. (Maureen, Group 3)*

Many participants also spoke of having to manage the power dynamics in order to receive adequate care. Some described trying to stay on their doctors' "good side", refraining from disagreement or confrontation for fear of retaliation.

> *We give a lot of power to doctors […] I mean, I do all, all the time. Why? I know why. Because I suppose we think they're… I feel personally that we're so dependent on staying on their good side to get the help that we need. If you find, you're vulnerable, who else are we supposed to trust? (Caitlyn, Group 4)*

Others spoke of emphasising their status as professionals during consultations, hoping this would equalize power imbalances, and alter clinicians' presumptions about them as patients.

> *I remember, like going to consultants, and my wife will be like, "Oh, you're very dressed up Caroline". And I was like, "Yeah, I have to dress up, I have to show them that I'm a professional person who, em, isn't mad, and is like taking control" (Caroline, Group 5)*

While negative experiences were more common, some participants also described positive relationships with clinicians who were empathic, attentive and proactive. They highlighted instances of clinicians taking time to provide guidance, to explain diagnoses and outline possible treatment paths. Interestingly, positive relationships were discussed in the absence of curative treatment, whereby certain professionals were seen as making a difference despite this. For instance, participants' frustrations with the limits of medicine were offset by clinician warmth, accountability and transparency about their insufficient knowledge. They appreciated practitioners who treated them as partners in their care.

> *Like, even though the system would let you down, there are people that I've kind of met on, on my healthcare journey that have kind of made a difference. And it's, even if they're not in a position to help you or cure you, it's the fact that you're listened to, is what makes a difference and there's that empathy. (Ashley, Group 3)*

> *I mean, nowadays, I have a wonderful GP who says, "Look, I'm sorry, I'm, I'm learning from you", which is just so refreshing for a medical person to admit they don't know it all. It's just wonderful. (Amy, Group 1)*

Though not solely the case, positive experiences were often associated with nurses rather than other healthcare professionals:

> *Yeah, I find often that nurses for me anyway tend to be a lot more open to listening to things like that. Whereas doctors are very quick to dismiss. But I've, I've had so many good experiences with nurses, I find it very different from doctors, not necessarily any specific experience, but I just find stuff they're like, their manner that they interact with patients is just even much more like empathetic. Whereas doctors, it's almost like, "you have your 10 minute slot, hurry up and tell me your problems. I'll prescribe something and then leave", whereas the nurses even if they are, you know, pressed for time, they're just, it's just kind of different? (Liam, Group 3)*

Some were hopeful for changes in clinician attitude as a consequence of the pandemic, highlighting the transformative power of lived experience on their professional practices.

> *But I think that what's been quite illuminating, I think for, for quite a lot of doctors is that some of them have got Long COVID. And then they know what it's like. Directly. Themselves. And they've completely changed their view. (Sophie, Group 3)*

6.4.2.2 Theme 2: The patient journey

In each group, participants painted a disheartening picture of their patient journey, with negative experiences being the highest intensity theme in this category of findings. The predominant sentiment was one of systemic failure: despite living with different chronic conditions, participants gave similar accounts of their healthcare experiences. For example, they often described a lengthy, costly and complicated journey that left them feeling hopeless and alone, and collectively their accounts conveyed of endless uphill battle for basic care.

> *So I think we're very much out on our own. On the scrap heap, as some people call it, to fight and look for yourself, and like (…) it was just very daunting and scary and lack of support. (Ellen, Group 2)*

> *We're all fighting this journey, and there is no support to help you get out of this abyss of pain and depression. It does get you down. (Dawn, Group 5)*

> *(…) you're battling for everything, you know, every referral, every test every, emm, trying a new treatment, you know, that it's always a fight. (Caroline, Group 5)*

Overall, participants across all five focus groups emphasised that navigating the system was an added burden to their already burdensome condition. Many described the weight of having to be their own advocates, having to push for tests, seek out treatments and coordinate their own care. Costs of private care or care abroad, the practicalities of travel and unnecessary paperwork were discussed at length:

> *You know, we all work so hard. And we, even with our impairments, to educate ourselves and to research. And there's so little work and the professional end, it would save so much time and energy, you know, I'm particularly bitter (…) It just takes a phenomenal amount of effort. (Emily, Group 2)*

> *But yeah, that also, I think adds on to the burden, the burden of providing your own care, of finding the money, the time to travel, see this one specific person. Like, there's, there's a lot of burden on the patient side to make the system work for us. (Robert, Group 4)*

Very few participants spoke of positive experiences of patient journeys and when they did, examples related to others' experiences of acute services. Further illustrating their disappointment, many compared their journeys to that of patients with other chronic or terminal conditions, expressing frustration at the lack of similar supports in their own care.

> *When she [friend] came home, she got sepsis. They looked after her amazingly, cuz she was ready to die. (Caitlyn, Group 4)*

> *And it's a terrible thing to say, but I'd often have people, you know, friends or whatever, that have cancer. And I just kind of feel a bit envious of their, the thoroughness of their, their treatments, and the support that they have. (Ellen, Group 2)*

6.4.2.3 Theme 3: Barriers to effective care

Participants' discussions of their healthcare experiences revealed a number of interconnected barriers contributing to suboptimal care in the context of poorly understood chronic illness. Along with identifying issues related to stereotypes and assumptions, participants highlighted the issue of fragmented services within the healthcare system, noting poor coordination between services, lack of follow-up, and breakdowns of communication. Both of these themes were frequent topics of discussion, and taken together, participants often suggested that the Irish healthcare service was simply not cut out for their care and remains resistant to change.

> *Yeah, I mean, nothing has changed. I think that's one thing that's really depressing. (…) The system is not good at dealing with chronic or unknown conditions. (Robert, Group 5)*

Participants often spoke of being "*bounced around*" between services, "*fobbed off*" by clinicians, and eventually "*falling through the cracks*" of the system due to the "nonspecific" nature of their symptoms.

> *Once there is a system, they follow the system, and it's excellent if there is. But if you don't fall into a category or a system, there isn't one, you're just left hanging. (Amy, Group 1)*

They also experienced a lack of continuity in their care, and described seeing different GPs or consultants at each appointment, which lead to what they described as a "*pot*

luck" of services. This discontinuity disrupted the development of a consistent and comprehensive medical history, leading to fragmented care and repetition of tests, procedures and referrals. Consequently, it hindered the establishment of a trusting patient-doctor relationship, often resulting in delays and suboptimal treatment plans.

> *And then when you go back to the GP, it's like, it's I've never spoken to him before, he doesn't know who I am. And you're starting again. And it's just, it gets so frustrating. And, you know, you kind of begin to think "what is the point of going back again, and again, I'm just, you know, getting absolutely nowhere, nowhere with it". (Ashley, Group 3)*

Despite being sent down a trial-and-error path of referrals, participants described being stuck in limbo, waiting for consultations that never came. They experienced situations where important medical information was not effectively shared with them, and among their healthcare providers. The lack of electronic patient records in Ireland was seen as a significant source of these problems, essentially placing the burden of coordination on the shoulders of patients. For instance, participants spoke of having to collate their own medical histories to ensure clinicians had an up-to-date records.

> *I waited three years for this appointment on public system. And there was no report. There was no image sent back to the GP. I was just dismissed back to my GP, and it said normal (…) And I waited three and a half years. And it said "normal". But no image. No results. Nothing. My GP said it never even said what area of your body they did the MRI. Just said "MRI. Normal". And I tried to follow it up for months to get anything off this consultant. (Ellen, Group 2)*

> *(…) [I] had to email them all my symptoms because I knew it was a case of if it's not documented, there's no paper trail, and it's not said, I basically- before every apply appointment. I literally emailed him all my symptoms. And I, I also brought a hard copy with me, because more often than not, they said" No, actually I didn't get them" or "I didn't receive them". (Lucy, Group 4)*

Poor coordination of care were even more frustrating in light of participants' explicit need for multidisciplinary care, thus marking it as a significant obstruction. They often felt there was a lack of "joined up thinking" among clinicians, and discussed how different aspects of their health were treated separately despite being interconnected. They also spoke of challenges related to hyper-specialisation among consultants, noting silos and a lack of collaboration.

> *I think, for, for me, because everything has to be multidisciplinary approach. I think enough of it is down to luck. And it's the luck of which doctor or specialist you get and how interested they are, and whether they'll make the referrals and whether they'll try to join the dots together. (Mia, Group 4)*

> *They've all gone into these like silos, and they don't talk to each other. So then when you have a condition like mine, that is rheumatology, neurology, you know, autoimmune, you know, it's like a cross between like maybe five different specialties. Nobody knows anything about it, you know, what I mean? (Caroline, Group 5)*

The compartmentalisation of services and disconnection between them often resulted in further delays, despite the potential to advance care. As expressed by Robert in Group 4, *"Even though their advocacy or investigations could impact further tests. They just drop you."*

Sharing their frustrations, participants also referred to barriers in the form of limited national resources, mainly staffing and medical facilities, and described the resultant necessity for them to go abroad to seek help. Some participants discussed demand-supply issues, providing examples of being unable to register with general practitioners within their catchment areas due to these centres not accepting new patients.

In relation to consultations, many felt short appointments were insufficient for patients with complex and chronic health problems and lengthy medical histories. While such arrangements were seen as suitable or necessary for basic tests and brief follow ups, participants felt that short consultation times limited their opportunity to fully explain and make sense of their health issues. This was an added barrier to achieving holistic care in an already fragmented healthcare service, further compounding the uncertainty felt by participants.

> *If we go in to see a GP, they have to see a person in approximately nine minutes. So I was about two minutes to get into the chair, two minutes to get out of the chair, most people present with around 2.5 to three issues. So the, the physician is supposed to diagnose, and come up with the prognosis for two and a half condition in less than five minutes, that's impossible. That cannot be done. And there is no bureaucrat that can make that happen. It just cannot happen. […] my whole life has gone to hell. There's no way I'm going to be able to summarize in five minutes. It just can't be done. (Robert, Group 5)*

A further concern raised by participants related to the inconsistency of care, most notably, the varying standards and practices among clinicians in public and private services, but also among expert consultants. Again, there was a sense that care quality

was a case of luck, and in light of this, many participants spoke of turning to the private tier of healthcare or travelling abroad in the hope of finding answers or help. Cost was a significant concern, with participants indicating that financial constraints often prevented them from accessing comprehensive care, including specialist consultations and holistic therapies.

Some described having no choice but to sacrifice their livelihoods to secure diagnostic tests and treatments, highlighting the injustice of the two-tiered system.

> *(…) it's that lottery, that there should be some level of consistency. Now we know, consultants will have different personalities, backgrounds and training, but there should be some kind of standards, I basically paid 250 euros to get gaslit (Julia, Group 1)*

> *Um, Yeah, like, like the first girl said, it was unnecessarily expensive, I spend 70,000 in three years. I had to do GoFundMe, I had, I spent my own life savings, inheritance. And I would, you know, because I've had to go abroad a few times, four times, I've been abroad for various types of treatment to boost it. It was very frustrating. (Ellen, Group 2)*

Those who sought care abroad often compared their experiences to that in Ireland, highlighting significant gaps in knowledge among other barriers. Their experiences in other countries further magnified the failures of the healthcare system, at the same time illustrating that care for their conditions *is* a possibility. For instance, many participants referred to inadequate diagnostic services in Ireland, as compared to other European countries, noting this as a significant hurdle to the provision of care.

> *So I think the experience in different countries, the knowledge level and the understanding is vastly different. Its... incomparable in a lot of aspects. Whereas in Barcelona, the hospital is treated like... it's treated like it's your home. And they're, they're there to look after you for every need you may have. And there was never a doubt put towards you- they would always listen to your concerns or whatever, same in the UK, very understanding consultants. And they knew a lot more about what was going on. (Niamh, Group 4)*

> *I had to go to Cyprus, and their healthcare is second to none. "You've got a pain down there? We'll get you in for an ultrasound tomorrow". Everything's tomorrow. Everything's cheap. (Ellen, Group 2)*

> *And I think I've a brilliant GP, you know, he's, he did everything he could and he never stopped looking (…) And he's, he tried his best to find answers. But he just didn't have diagnostics. And when I sent the blood away to Germany, and I got the results, he was, "Oh, my God, I'm so sorry. I wish I had access to these tests". (Ellen, Group 2)*

As mentioned, clinicians' assumptions and biases concerning poorly understood conditions were a strong component of discussions, and significant barrier to care identified by participants in all groups. Participants often described how stereotypes and assumptions based on age, gender and mental health significantly impacted their patient journeys. Many felt that these biases influenced the attitudes and practices of healthcare providers, leading to suboptimal diagnosis and management.

> *And I hate saying it, would, if these conditions were more male focused, I think there would... we'd be doing a lot better from a medical perspective. (Kelsey, Group 4)*

They often described how clinicians applied stigmatizing labels or "waste basket" diagnoses to conditions they could not understand, which negatively impacted on their subsequent care.

> *And then similar then with like, when they see something on your file, like a medication, or even like fibromyalgia, I find that they tend to get very dismissive of anything you come in with. So if I go in with like, a new symptom for anything else, everything, and I know Fibromyalgia has so many symptoms anyway, but it's always put to, "oh, that's just part of the Fibro" and then that then leads to, "there's no treatment for Fibro", like, "can't do anything there". (Liam, Group 3)*

Age and gender-related assumptions were seen as particularly detrimental, and participants recounted instances where their health concerns were minimised or attributed due to the fact that they were "*too young*" and "*too female*". Concerningly, one participant gave examples of implied promiscuity when seeking help for pelvic pain, presumably due to their gender and age. Others reported similar comments of a gendered nature.

> *I, just like, oh my gosh, I'm so tired of being told that I'm "too young" or "too female" to have things wrong with me. That's kind of though, like part of the other thing, my age actually is a huge thing. A lot of the time like, this like, not to be TMI in or anything but like, I got to the point where like if I went in, obviously with the urinary stuff, and I actually had to be like, "if the words chlamydia or gonorrhoea come out of your mouth, I am leaving your office. I do not have an STI. I've been with the same partner for nearly eight years now. So unless he has something earth shattering to tell me, I guarantee you, I do not have an STI, please stop assuming that's what's wrong with me just because I happen to be in my mid 20s". (Sarah, Group 1)*

> *I was told, you know, I call it 'lovely girl syndrome', you're lovely, you're just a wee bit anxious and just stressed out too much, you just need to be a little bit less stressed and less anxious, and it'll be grand. And so I went to my GP, and he just kind of gave a little laugh and just said, "have a baby". (Julia, Group 1)*

Some spoke of being diagnosed with low mood, prescribed antidepressants and referred to mental health services, again presumably as a function of their gender:

> *I don't have low mood. But they basically were saying, like, "this happens to women, you know, they just go mad." (laughs) "They imagine that there's something wrong with their body that's not actually there." (Caroline, Group 5)*

6.4.3 Category III. Improving Healthcare

As a core component of focus group discussions, participants were invited to reflect on their healthcare-related needs and challenges, and to make suggestions for changes that would improve the healthcare journeys of patients with poorly understood chronic conditions. A visual mapping of recommendations made participants by is presented in Figure 6.4, which illustrates the interconnections between targets for change as identified by participants in the focus groups. The categories of recommendations and their subcomponents are outlined below, with thematic findings discussed in subsequent sections.

Though there was less consistency across groups with regards to specific recommendations, participants most often emphasised a need for improvement at the level of The Consultation, particularly the *Patient-Clinician Relationship*, highlighting a need for *Guidance and Signposting* and *Support at Different Stages of the Journey*. At this level, participants in all five groups discussed a need for *Improved Knowledge* and *Improved Understanding* of their conditions, noting that this would improve their clinical encounters. While several participants recommended that clinicians should *Improve Efforts*, particularly with regards to diagnosis or investigation, such suggestions were infrequent, and not proposed at all by participants in Groups 1 and 5. Overall, participants often focused on improving the patient-provider relationship by targeting *system-level* factors that culminate in a largely negative experience, as illustrated by Figure 6.4.

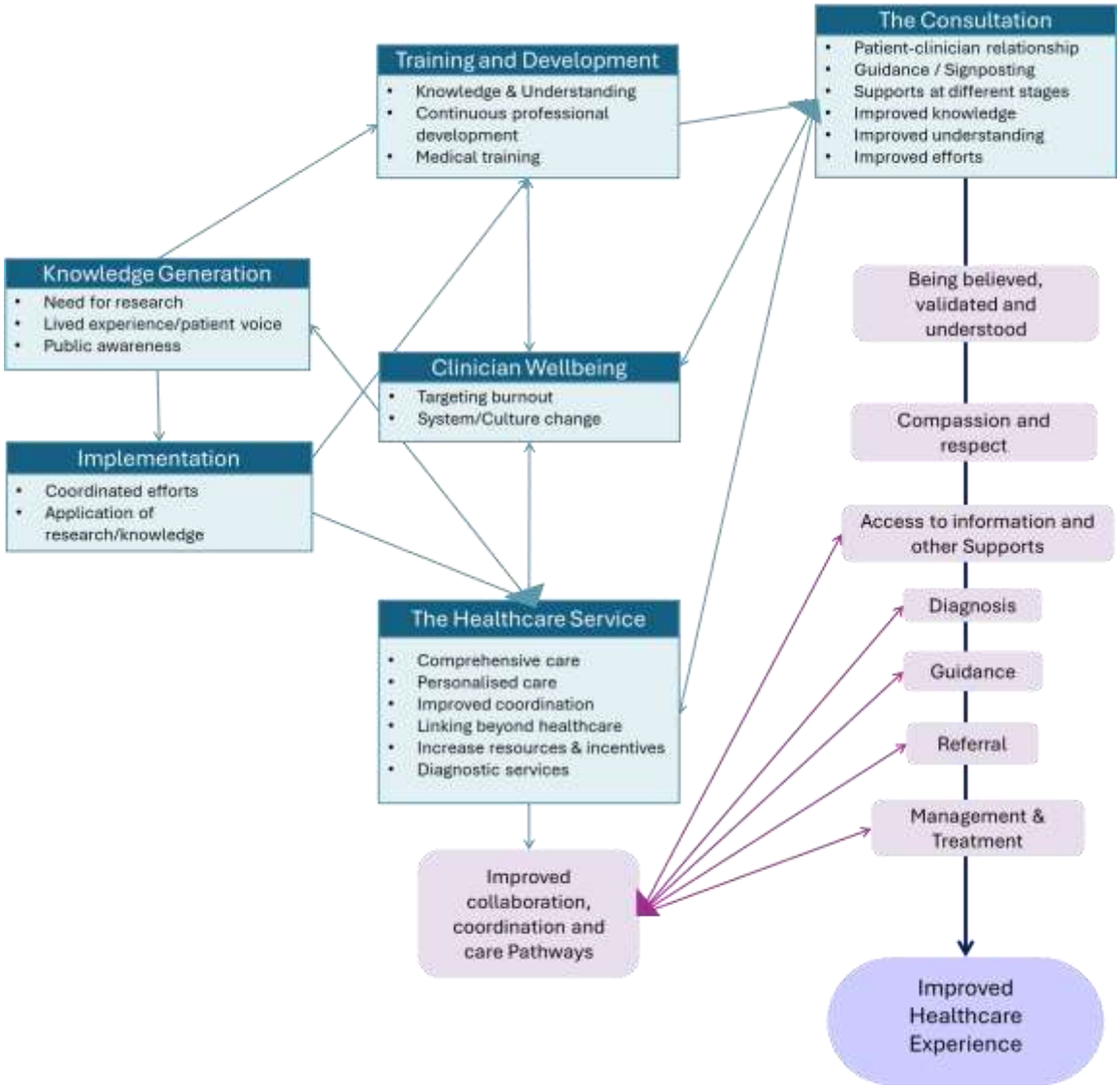
At the level of The Service, most participants emphasised a need for *Improved Coordination* and *Comprehensive Care* to improve patient outcomes, and less frequently

made recommendations around *Personalised Care.* Participants in four groups recommended *Linking Beyond healthcare*, emphasising that patient communities and advocacy groups, as well as alternative therapists, can play an important supportive role in healthcare. A few participants from Group 2 and 4 suggested a need to *Increase Resources and Create Incentives* for improving services for patients with poorly understood chronic conditions. Given their poor experiences of diagnosis, several participants in three groups emphasised a need to improve *Diagnostic services* and facilities in Ireland.

Recommendations related to Training and Development, Clinician Wellbeing and Knowledge Generation varied between the groups, with less agreement among participants on how to achieve change in these domains. A small number of participants in each group discussed a need to address clinicians' *Knowledge and Understanding* of their conditions, with *Continuous Professional Development* targeting soft skills like bedside manner. Participants in four groups suggested that changes should be made to *Medical Training*, with greater emphasis on kindness and compassion. Participants in all but Group 2 also recommended *Targeting Burnout* to improve the manner in which clinicians deliver care, along with *System and Culture change* to reduce the pressures placed on medical students and practitioners. With regards to knowledge generation, a few participants in all groups discussed a *Need for research*, preferably biomedical, as well as an emphasis on *Lived Experience and the Patient Voice.* A few participants also suggested that research is needed to improve *Public Awareness*, though emphasis on this was weaker. Finally, a number of participants discussed the challenges of and possible solutions for Implementing Change, highlighting the need to *Apply research and knowledge* that is already available. *Coordinated Efforts* involving key stakeholders, both nationally and internationally, were discussed by a small number of participants in Groups 2 and 4.

Figure 6.4

Patient recommendations for an improved healthcare experience



6.4.3.1 Theme 1: The consultation

As highlighted previously, participants often discussed negative experiences with healthcare providers, with many recommending change at the level of the consultation. Participants in all groups emphasised the importance of fostering a strong, positive relationship with clinicians. Generally, they stressed a need for compassionate care where they are listened to, believed and treated with respect. Participants emphasised a need for mutual trust between patients and healthcare providers, and explicitly recommended that clinicians demonstrate more empathy.

> *I suppose the fundamental thing is to be treated as a person and not a number. You know, and like the others were saying, is that they actually listen to the patient and respect the patient. You know, and to be believed, because no one is there for attention, no one is there for the, you know, there's always a root cause to whatever is going on. And it's not that, it's not that we expect answers straight away. But we do deserve to be treated with respect, kindness and empathy (Chloe, Group 1)*

Participants felt that being believed and treated as partners in care would significantly improve the consultation experience. Reflecting on their experiences of a power imbalance, they often spoke of a desire for clinicians to demonstrate humility, recognizing that *"the patients know their body better than anybody else"* (Niamh, Group 4).

> *What would make a huge difference is just not having the eye rolling and huffing and puffing and being laughed at. That'd be a start. That'd be a big start. I mean, that'd be a big help. (Sophie, Group 3)*

The most basic recommendation was for practitioners to engage with patients in a professional manner, regardless of their personal beliefs and biases. As expressed by Amy in Group 1, "*I think we're all saying "believe us (…), and "try and put your personal biases aside"*. Many wished for clinicians to acknowledge their suffering and emphasised a need for effective communication during the consultation. They acknowledged that time constraints and other limited resources may affect the nature of the clinical encounter, but suggested that improvement *within* these limits is still possible:

> *And there isn't time. But when there is time, they can do so much better, but they waste so much time having no time. So time, time is a big thing, because they... it, they just wasted it because of the wrong professional, or the wrong communication. You know, even when they're using the time, they're not using the time they have well, through poor communication. (Emily, Group 2)*

Participants recommended that clinicians provide them with clear guidance during consultations and stressed the importance of being directed to relevant information and resources, particularly those provided by charity or advocacy groups. Related to this, many emphasised a need for *concrete* guidance rather than broad recommendations about lifestyle modification.

> *(…) they don't have to print up a manual, they don't have to do anything. Even just a website of "these are the groups associated with you", "these are triggers", like just a list something because I had absolutely nothing like. (Robert, Group 4)*

> *It's not enough just to diagnose the person, you have to tell them where to go to next and be specific. (Julia, Group 1)*

As previously highlighted, many spoke of being left in limbo while waiting for diagnosis or referrals. Referring to this, participants discussed how receiving transparent communication about the next steps in their journey would improve the overall consultation experience while waiting.

> *You know, sort of, say, "right, we know we can't cure it. But we have a pathway. There's a few drugs we can try". You know, "there's actually something we can do", as opposed to just saying "it's a blip, suck it up" (laughs). (Millie, Group 1)*

> *And maybe even, I don't know, offer suggestions. Or maybe "it could be this, or maybe it could be that" (Maureen, Group 3)*

Patients also expressed a need for tailored support throughout various stages of their journey. They suggested that appropriate assistance should be available from initial diagnosis through to follow-up care, catering to their evolving needs. However, the initial stages of the healthcare journey were identified as particularly critical, requiring a high level of support from clinicians and allied health services.

> *You get diagnosed, you're in shock, you don't know what it means you don't know what it means for your life, depending on how it's impacting you. You need psychological support, or some sort of information support, support of some description (Caitlyn, Group 4)*

Support at early stages was also seen as crucial for effective symptom management and harm reduction, even in the absence of a formal diagnosis.

> *(…) a lot of us have done the wrong thing and our beginnings that have perhaps hampered us now. So it really is the most important part of the journey, is to get that beginning right (Millie, Group 1)*

Recommendations related to clinicians' understanding of poorly understood chronic illness and multi-systemic conditions were frequently raised by participants. There was a consensus that clinicians should possess more comprehensive knowledge in order to provide nuanced and effective care. Participants stressed the importance of clinicians being attentive, responsive and open to learning from the patient:

> *And... working on the basis of... the person who's sitting there in front of you, the* patient *knows more- and will know more- than you ever will. (Robert, Group 5)*

At the very least, participants recommended that clinicians should have enough awareness to identify the conditions and to make appropriate referrals.

> *But I think that like a major stumbling block for a lot of these conditions is that the GP isn't actually aware of it, or what the symptoms are, or what the tests are or that kind of thing. (Caroline, Group 5)*

> *And it would just be so refreshing if they turn around to you and say, "I don't know what's wrong with you, but I'm going to look it up". Or "I'm going to send you to somebody who I think might know what's wrong with you" or, or anything?. (Caroline, Group 5)*

Participants often urged clinicians to demonstrate greater diligence and commitment to their care and recommended that clinicians should make efforts in the way of actively educating themselves and exploring all possible avenues to address their patients' health concerns. Most participants felt that clinicians should be more willing to fully review their medical histories and investigate complex symptoms, rather than apply stigmatizing labels and issue a premature discharge. They emphasised a need for a patient-centered approach to management, rather than the consumerist "conveyor belt" system:

> *Whereas if someone had spent quality 40 minutes, and they're going through my case history, they could have joined the dots, But we have a conveyor belts thing, get them in and get them out. (Julia, Group 1)*

6.4.3.2 Theme 2: The healthcare service

Participants made a number of suggestions for how to improve barriers to care at a system level. While identifying the best solution was a challenge, participants most often suggested that enhancing coordination of care, fostering multidisciplinary collaboration, and improving communication were essential steps towards improving healthcare. Given the complexity of their conditions, a need for comprehensive, multidisciplinary care was repeatedly emphasised. Many discussed a need for improved access to specialist care as well as enhanced collaboration and knowledge-sharing among those specialists.

> *And so I don't know whether that means we need a multi-disciplinary clinic so that people have knowledge about loads of different stuff. And then you can get loads of people and together to help loads people at the same time and then maybe send to another, or is it that we need ME specialists? I mean, for CFS, POTS Lyme, whatever, I don't know. (Anna, Group 2)*

> *I think more likely, they just need to start talking to each other and actually use teamwork to help patients. And that doesn't sound like such a big deal. But it really is like, it really seems like the hardest thing (laughs) is for doctors to talk to each other. (Caroline, Group 5)*

As previously illustrated, participants recognised a general lack of specialists in their conditions nationally, and consequently suggested that creating incentives could persuade practitioners to develop interest in conditions that are dismissed and stigmatized. They felt that such incentives would lead to improved services in the future.

> *And every one of those people told me it's career suicide, that no one will want to touch it [ME/CFS] with a bargepole. So we're not just jammed between lack of medical knowledge, we're also jammed with there's hierarchies within the medical profession. (Julia, Group 1)*

Others highlighted a need to reduce hyper-specialisation, as this practice leads to patients being “bounced around” the healthcare system. Instead, participants suggested a need for clinicians with a broad set of skills and knowledge who would “join up the dots” between seemingly unrelated symptoms.

> *I suppose I went to, I don't know how many consultants over the years, and what the different specialities were, but there, they were just really going, "not my area, not my issue, buh-bye" (Amy, Group 1)*

And basically, the situation with the doctors and consultants here is they don't understand that Ehlers Danlos, or hypermobile spectrum disorder are multisystemic condition. They don't get that. They treat it that a rheumatologist treats X, cardiologist treats this. What they don't realise that everything is connected. And that sometimes when you're treating one, you also need to be treating the other. (Kelsey, Group 4)

Participants made specific recommendations for designing specific care pathways for patients with undiagnosed poorly understood conditions. They emphasised a need for personalised care, moving away from a "one size fits all" approach to management. In the interim, a need for quality care was voiced:

Why not have a specialist category for them? You know, we have the "we don't know yet". Why not say "we don't know what's wrong with you? So we'll put you in this holding category where we're still doing an investigation, still doing whatever you, But we can give you the excellent care that you would get if you got a heart attack or you know, a broken leg or something" (Amy, Group 1)

Discussing the burden of having to coordinate their own care, participants made recommendations around information flow through the system. They suggested that addressing breakdowns of communication between departments is necessary to support timely, multidisciplinary care.

Well, the first thing I think, is to fix the breakdown of communication between departments. Because there is no communication between different departments in, in the system, and also that, like the system, when you're sent to a consultant, and if they can't find anything wrong with you, you're sent back to your GP, and then the waiting list starts over again. And if you could take it takes years to get actually proper diagnosis. (Chloe, Group 1)

Most participants also spoke of a need for continuity of care and emphasised the importance of having a single and consistent point of contact, specifically their assigned GP, in their journeys. Stressing this, several participants explained that seeing a new GP or specialist at each appointment impacts on quality of care, emphasising that consultation time is wasted on the re-telling and synopsizing of medical histories.

Your general practitioner should be the central person and with everything, everything should come back to the GP, to have a proper overview of you and the health of your whole body. But that just doesn't seem to be happening. It just seems to me that like there's a lack of communication, and you're just kind of ended up kind of falling through the cracks a little bit (Ashley, Group 3)

I don't have any, any one doctor (…) Whereas if I had the one GP It would make life so much easier both for them and for me, because they're going to know me,

you know, to begin with, and I'm not wasting half of their time either. It's a win for both sides. (Millie, Group 1)

Noting significant delays in healthcare, participants also suggested general changes to referral practices. Improved flow through the system could be achieved if specialists and allied health professionals could refer to one another, essentially bypassing the GP. They also discussed a need for improved follow-up, and suggested the creation of new roles to help manage this.

And actually, I think the way we can do it here is you have your GP, but then you can find your specialist say in your area as well, like your physio, or if it's women's issues, and they can also refer-- so there is a way of working the system where you can find different people that will refer you back into the system. (Amber, Group 4)

And I just thought maybe something like having a… a patient rep. That has you know, works for the GP, they can kind of…. the GP doesn't have time, but it's literally somebody that can kind of follow up with the patients just to see how they're doing. It's obviously like it'll be a huge task and a huge job. But like, I suppose it creates like a separate role, like a patient rep in healthcare centre, that their job is to follow up with patients and how they're doing so. (Ashley, Group 3)

Importantly, when discussing coordination of multi-disciplinary care, participants often recommended a need to introduce electronic solutions, namely electronic patient records, into the Irish healthcare system.

And so I think, joined up thinking is a definite one, sharing, like if this if our information if every test that we have, and every consultation if those medics just entered into a database, what they did, it, what the results are, what the tests are, and that we have access to it, obviously. And then it can be shared out. There'll be a lot less duplication and a lot less second guessing. And I think it might just be a tool towards more, as I call it, "the joined-up approach" or the multidisciplinary approach, because right now, they're hiding behind that and saying, "oh, sorry, you've that done in such and such a hospital. I have no access to that. I haven't got those results". And then the GP six weeks later has gotten nothing. So anytime that I've been referred into A&E, my GP is unaware of it unless he referred himself. He doesn't get anything. And so he's relying on me to tell them or if I'm admitted from A&E into the hospital, I have to ring him and tell him I'm actually in hell. (Mia, Group 4)

Digital solutions, particularly telemedicine, could also reduce barriers to accessibility. Many participants suggested building on the services that were implemented throughout the pandemic, emphasising that this was a realistic possibility.

I found COVID...It was great. When they did online calls it was a big help (Caitlyn, Group 4)

> *Being told for years that this, this kind of accessibility was impossible, it would be too much of a burden, it's you can't be done. And the second it's necessary we have it in two months. (Robert, Group 4)*

When discussing their experiences of diagnosis, participants often made broad recommendations about the need to improve the scope of diagnostic services in Ireland. They noted that delays in diagnosis are partially due to their clinician's lack of knowledge and understanding, and partially due to the need to outsource tests to facilities abroad. Reflecting on the general state of facilities, participants suggested developing national centres of excellence as a way of improving access to specialist and quality of care.

> *For the country, that's supposed to be, as you said, Shane, "forward thinking", we don't have access to facilities to make things easier for people to diagnose things as well, the doctors don't have it either. (Dawn, Group 5)*

Many participants suggested there should be stronger links between the healthcare system, patient communities and complementary services. Some criticised that useful services are being underused, largely due to clinicians' reluctance to refer to them:

> *The GPs are, and consultants are fast, in some ways, to refer you to other doctors, but very slow to refer you to things that might actually help you like physio, OT, psychology, social work, all these things that, like, that's what you actually need is somebody to help you figure out how social welfare systems works or you know, how. (Caroline, Group 5)*

Reflecting on the value of peer support networks and advocacy groups, participants discussed the possibility of integrating such communities into standard healthcare services:

> *Sorry, just thinking about peer support groups. (…) But I'm thinking that that's a model that could be used, you know, for the various different particularly also have these, as I call them the "weird and wacky" stuff that don't fit neatly into a little parcel. Just like even this today, just chatting and we can see immediately the connections that we have and the similarities of our experiences and just to be able to have a voice to see what's happening, that if some of the consultants had that and brought people together(…) , how much more productive it will be if we were sitting at home, online, with people with similar conditions to ourselves, that we can support each other, we can learn from each other and what's more, better again, we could bring that back to these specialists as the expert in our fields, because we've the lived experience (…) (Mia, Group 4)*

6.4.3.3 Theme 3: Training and development

While participants often emphasised a need to improve clinician knowledge and understanding around their specific conditions, their recommendations for training focused more strongly on values and person-centered care. For instance, participants suggested that medical education should teach the clinician to learn from the patient, listen, and take their experience at face value: as expressed by Laura in Group 4, *"pain is what the patient says it is.".*

Participants often felt that aptitude or intelligence were not sufficient qualities for a healthcare professional. Many of the suggested changes related to humanizing the patient, and focusing on clinician's bedside manner, empathy and compassion.

> *And I think the one thing that we could do that would have everybody is teach them to listen, and their patients are patient experts in their pain, because we'll never be able to teach everybody everything about every disease. (Caitlyn, Group 4)*

> *I know that kind of sounds very simplistic, but I think kindness goes a long way. And it's brilliant that you're in medical school, and you're learning how to name all of these body parts. And you know how to do this condition or how to perform this procedure. But I honestly think there needs to be more emphasis in medical training on how to just be kind to people. (Sarah, Group 1)*

Again reflecting on inter-personal factors, participants recognised that while certain clinicians are "excellent diagnosticians" and researchers, they lack the qualities that would make them a compassionate doctor. Some suggested a need to re-evaluate medical school intake practices, with more emphasis on interpersonal characteristics.

> *The problem is, who are we shunting in to do medicine? Yeah, this kinda system. Now I have a niece who is doing her consultant training. And I love her. I love her. However, she does not have a bedside manner, nor will she ever. Yeah, she's the coldest creature (…) And she's really intelligent, this girl, and has written some fabulous research papers already. And she's got to be a great, you know, she, she's great. That's what she should be at. She shouldn't be near people at all, really. But unfortunately, she will be near people. (Laura, Group 4)*

Some participants also suggested a need to re-evaluate the content and delivery of medical school curricula, ensuring that misinformation about stigmatized conditions is not being perpetuated through training.

I really do think they have to start back at base back when they're in when they're doing their studying, they need they need these lectures coming in from all over the world that are world experts on these and educating the new up and coming doctors or student doctors. So they will then come in and teach some of the old dogs new tricks, you know, because unfortunately, it's the old dogs that are teaching. The old ones, the old tricks, and that's no good. You know, teaching them that, giving them bad habits. (Kelsey, Group 4)

Reflecting on limited resources, some also suggested a need to incentivize specialist training in chronic conditions, with added efforts to retain medical graduates in Ireland.

And the problem there is that we're training, we're training GPs, we're training doctors here and we're paying for their education, and we're letting them off the minute they qualify. They should be, they should have to give back to the state for a couple of years and take away some of the stress that the medics, the medics are under at the moment (…) They should be- okay if we're looking to fix something that's broken, the first place is to start with supporting the medical staff and giving them, giving them extra staff. (Dawn, Group 5)

6.4.3.4 Theme 4: Clinician wellbeing

Participants often suggested that their negative experiences with clinicians and healthcare could reflect deeper issues related to culture and the nature of the clinical training environment, or more broadly, the institution. They suggested that a focus on clinician self-care could lead to improved relationships between patients and providers:

So maybe their own self-care. If they, if they really are not capable of being caring and compassionate and believing us, maybe they need to look more at their own self-care. (Amy, Group 1)

They suggested a need to address clinician burnout, emphasising the consequences of the tremendous pressure many healthcare professionals are under:

I really feel that part of the…part of the problem is like, how much…emm… how much pressure doctors are under themselves, you know, and how high the rate of burnout is. And, emm… like that they're really badly, badly treated in their training. You know, it's almost abusive of some of the things they have to do in terms of the hours they work with no sleep and making these life and death decisions. Like it's, it's cruel. And I think that what, where that ends up is them having a very, a great amount of difficulty to empathize with patients, you know, and I have often wondered that if we were kinder to doctors, could the doctors be kinder to us? (Caroline, Group 5)

Some participants who were also practitioners echoed this sentiment, providing a glimpse into the pressure faced by practitioners:

> *I remember one situation where I was working in a spinal cord injury treatment unit. And it's the nurse's station. And one of the docs on call came to the nurse's station and the doc had been working for 72 hours non-stop sat down in the nurses station and tried to figure out medication, and what dose is this person, and I saw him putting his head in his hands, just crying, saying "I'm going to kill somebody". Somehow, we think that that level of education is going to help. We can take a human being and do all that kind of crap to them, and say, "this, this is a good thing, and this is forging them". You can't do that to a human being. You just can't. (Robert, Group 5)*

6.4.3.5 Theme 5: Knowledge generation

As illustrated, focus group participants often referred to poor levels of awareness, knowledge and understanding of their conditions, and relatedly, spoke of a need to improve the quality of research concerning them. They agreed that conditions falling under the umbrella term of "medically unexplained" have historically been neglected by research, and that the research conducted has failed to address underlying pathophysiology but instead focused on lifestyle modification. Reflecting a need for improved diagnostics and curative treatments, participants suggested a need for funded research focusing primarily on biomedical advances.

> *I think too much in the past, it's been kind of these things have been kind of... there's the research has gone into the psychological side of them, which I mean, there is some benefit in and it is good to think about, but like, that's, we can all agree that's never going to fix anyone, you know, that's never going to solve the problem. The only thing that is hopeful, and actually, like, being a cure would be biomedical research. And that hasn't been happening enough I think. (Caroline, Group 5)*

They also emphasised the importance of patient voice in knowledge generation, highlighting the practice of patient and public involvement in research:

> *It's great when you see that it's also led by people lived experience of these conditions, or, or even if it is healthcare professionals who have these conditions, or maybe they know someone or they do know it. So I think I am seeing that a bit more often, and I think that's a really positive change, and to see where patients have more of a voice and are working alongside healthcare professionals rather than just you know, they're going to talk about us to each other. (Liam, Group 3)*

6.4.3.6 Theme 6: Implementing change

Critical of failures to implement strategies for improvement, several participants spoke of the challenge of implementing change in the Irish healthcare system and made suggestions for a multi-level approach to improving healthcare. For instance, they highlighted a necessity for coordinated efforts involving stakeholders like physicians, policy makers and government representatives. They spoke of a need for rigorous epidemiological research to quantify the scale of the problem and suggested initially identifying the cost of chronic illness to the system.

> *It isn't until there's a coordinated group of people with bulletproof information, and the threat of financial implications for people in power, that something gets done. It's just the way my experience of how things work in this country. And it's probably in most countries, to be honest. I think by the very nature of the illness, we're all really tired and whacked. We have enough on our plate. But until somebody is able to pull everybody together, or a large percentage of the people together, a representative, and try to get some consultants on board, try to centralize the treatment plans and see over a period of time, can we adopt treatments and processes that help, that actually help us in the long term to get better. (Alex, Group 2)*

Some were hopeful that the impact of the pandemic on the male workforce would motivate change, primarily by highlighting the cost of the problem. Others were less optimistic about improvements in healthcare, and commented on how change is particularly slow in Ireland. While they voiced a desperate need for change, their accounts conveyed a sense of defeat regarding progress, despite advances in science.

> *But like, when I'm watching the numbers of Long COVID, I'm there thinking, "that is actually going to help us" because it's not only women, but it's men that have fallen out of their jobs as well. And its costing companies an absolute fortune. (Anna, Group 2)*

> *When I got hit with COVID, and Long COVID, I was asking about Epstein Barr Virus, and has it been reactivated? (…) And now the science has caught up and saying, yes, it can be reactivated and you should be tested for it. But we're still not doing it in this country. (Mia, Group 4)*

Others emphasised the need to implement changes emerging from patient involvement in research. Many felt that advocacy, whether it be related to research or policy, should not be the sole responsibility of the patient, though it often is.

> *(…) if they would have actually implemented what the ME and fibromyalgia advocates asked, we would have actually been in a better position to deal with COVID. (Julia, Group 1)*

> *Yeah, I think like, a part of the... part of the reason that these things have gone on for so long as they have is that the people who have these conditions are too ill to advocate for themselves, you know. (…) there needs to be research, there needs to be awareness campaigns, there needs to be blah blah blah because I think the people who have it themselves are too sick, you know? (Caroline, Group 5)*

6.5 Discussion

6.5.1 Key findings

The overarching aim of this study was to elicit the views of patients with diverse poorly understood chronic conditions regarding their healthcare needs and experiences, and further, to involve them in identifying targets and making recommendations for change. Like the previous studies comprising this thesis, results magnified the multifaceted impacts of chronic illness on patients' lives and highlighted the issues of delegitimation and disbelief as central to the patient experience. Results further illustrated patients' dissatisfaction with healthcare services in Ireland, with the majority (83%) of participants expressing dissatisfaction with the *overall* quality of care received. This is a stark contrast to the over 70% of patients that were satisfied with their care in the six preceding months reported by Darker et al. (2015), though their results represented a larger sample of participants with common chronic conditions. Regrettably, like in study three, participants in the present study reported how negative experiences with health services providers led them to lose trust in the Irish healthcare system, leading some to disengage from care altogether. Participants' accounts highlighted a need for changes in healthcare broadly, and particularly for patients with multi-systemic, rare, or "unexplained" conditions.

Overall, the findings underscore significant obstacles to care at both the level of individual consultations and the broader design of healthcare services, with particular emphasis on the challenges stemming from system fragmentation. Taken together, results suggest that the Irish healthcare system fails to follow chronic care models when providing care for this group of patients. The study's implications are discussed below, centered around the two broad targets for change identified by participants: the consultation and care coordination.

6.5.2 Implications

6.5.2.1 Targeting the consultation

Previous literature has highlighted that the patient-physician relationship is the cornerstone of effective healthcare (e.g., Kelley et al., 2014). The findings of this study demonstrate that when illness is fraught with uncertainty, the patient-clinician relationship becomes vital. Regrettably, as evidenced by the Study Three and shown in the present study, experiences of patient-clinician relationships are typically negative and marked by invalidation, poor communication and an imbalance of power. Unsurprisingly, then, patients' recommendations primarily focus on improving the consultation dynamics.

Interestingly, while patients emphasised the importance of expertise among healthcare practitioners, the findings also show that clinicians' disclosure of and transparency about the limits of their knowledge can be positive in the context of compassionate care. Participants stressed that being validated and truly listened to by healthcare providers was crucial, and some felt that their doctors' ability to make them feel heard mattered more than their technical expertise. This aligns with previous studies, which highlight that clinician listening and communication skills are often the subject of patients' formal complaints (O'Dowd et al., 2020, 2022), and illustrate the importance of kindness, deep listening (Bradshaw et al., 2022) and transparent communication (Bontempo, 2023) in healthcare. Thus, while biomedicine makes headway in the realm of rare or "unexplained" conditions, clinician humility, compassion and communication skills appear to be a key target for improvement.

Relatedly, participants suggested reviewing medical school admission criteria to incorporate an assessment of soft skills and personality, in addition to academic proficiency. Many highlighted contrasts in their experiences with nurses and other healthcare professionals, suggesting that training for GPs and consultants could incorporate elements from nursing curricula, particularly surrounding respect for the patient and bedside manner. This suggestion aligns with previous work showing that training on rapport building and patient-centered care (Kee et al., 2018), or more broadly empathy (Paulus & Meinken, 2022), could be effective.

The results of this study further demonstrate a need to address healthcare professional's preconceived notions about individuals with poorly understood or controversial health issues. Patients stressed a need for targeted training in this domain: they often reported facing significant challenges stemming from age and gender-based stereotypes, as their conditions were frequently misinterpreted or unjustly characterized as functional or psychological in origin. Similar findings are widely discussed in the literature, particularly in the context of chronic pain (Peate, 2021; Windrim et al., 2024) and ME/CFS (Geraghty et al., 2019), as illustrated in Study 1. Given the adverse impacts on patient outcomes, a need to target bias and discrimination in healthcare has been underlined (Mateo & Williams, 2020).

Though their experiences with the healthcare system and individual healthcare practitioners were predominantly negative, participants also recognised the systemic nature of these challenges and expressed empathy for those working within an imperfect system. They acknowledged that the challenges healthcare providers face, such as during training and in practice, can shape the clinical consultation in ways that limit their capacity for compassion and kindness. Indeed, declines in healthcare professionals' empathy have been attributed to increased workloads, burnout and empathic distress fatigue (Razi et al., 2023). Studies in the Irish context have found that doctors and trainees express high levels of dissatisfaction with their working conditions and training programs, and often report experiencing burnout, psychological distress, and bullying within the medical field (e.g., Clarke et al., 2017; Crowe et al., 2017; Hayes et al., 2019). Consequently, a high proportion of junior doctors intend to leave Ireland (Brugha et al., 2020), adding further strain to the limited resources within the healthcare system. While improving clinician wellbeing through targeted interventions could promote greater empathy and a more patient-centered approach to healthcare delivery, it is evident that more wide-reaching changes, particularly to medical culture, are needed.

6.5.2.2 Targeting care coordination

As discussed, the findings of this study illustrated the barriers posted by a fragmented healthcare system, highlighting patients' painstaking efforts to navigate disjoined care. At a system level, the key recommendations for change centered around achieving integrated multidisciplinary care and improving patient and information flow within and between services, as well as enhancing links between primary care services, allied health and patient advocacy groups. As stressed by participants, by providing clear signposting to resources, as well as guidance on navigating the next steps in their healthcare journey, the burden on patients to independently seek out information and "figure things out" alone would be alleviated. The available literature similarly indicates that that support from multiple sources is essential to effective self-management among patients with chronic illness (Vassilev et al., 2013). Such interconnections are recommended by chronic care models, the ICP (HSE, 2017) and emphasised in emerging care pathways for rare disease (Walton et al., 2022; Ward et al., 2022).

Previous studies have shown that inadequate care coordination can negatively impact patients with rare conditions, affecting physical health, psychological wellbeing, social functioning and finances (Simpson et al., 2021). Likewise, the present study revealed that a fragmented health system imposes substantial burdens on individuals already grappling with debilitating symptoms and diminished quality of life. For instance, many patients reported feeling overwhelmed by the demands of self-advocacy and spoke of the added burden of having to manage their own care coordination and follow-ups, at the same time maintaining their own paper-based medical histories to inform clinicians on their care.

Relatedly, as recommended by participants, implementing electronic patient records (EPRs) could greatly improve care coordination for those with chronic, multisystemic conditions and multimorbidity. Well-designed electronic patient record platforms are systems are considered crucial to achieving patient-centered care (Butler et al., 2020) and have been demonstrated to significantly improve chronic disease management by supporting the critical elements of the Chronic Care Model (Drawz et al., 2015). These digital systems facilitate seamless information sharing, collaborative care planning, and continuous treatment monitoring across diverse healthcare settings, and enhance

communication and decision-making among healthcare providers (Adeniyi et al., 2024; Pérez-Stable et al., 2019). They provide an opportunity for integration of patient information, which has the potential to enhance both the efficiency and quality of care across diverse patient populations (Harrison & Palacio, 2006). While some efforts have been made to incorporate single EPRs in Ireland (e.g., Sheehan et al., 2023), substantial gaps have been identified in the country's health information systems, with evidence demonstrating Ireland is significantly behind other nations in this regard (Walsh et al., 2021).

6.5.3 Strengths and Limitations

A key strength of the present study was the involvement of patient stakeholders in identifying barriers and solutions to effective healthcare, thereby bringing the patient voice to the forefront of the research. As previously emphasised, engaging patients in healthcare research is increasingly emphasised in the literature, as it provides unique insights and firsthand experiences that can uncover practical issues and solutions often overlooked by researchers and medical professionals (Bolz-Johnson et al., 2020; Brett et al., 2014b, 2014a; Graffigna et al., 2020; Rolfe et al., 2018; Vat et al., 2020). The focus group design enabled participants to share their individual experiences and perspectives, whilst also collaboratively developing suggestions for improving healthcare for patients with a wide range of chronic conditions. The collaborative approach taken by this study ensures the study's outcomes and recommendations effectively address patients' priorities and needs. Further, patient stakeholders were involved in co-analysis of data ensuring that the interpretation of needs was grounded in firsthand experiences.

The use of the framework approach to data analysis was a strength of this study, as the method provides a flexible yet structured and highly transparent methodology that can be systematically applied by co-analysts regardless of academic or professional background (Ward et al., 2013). Additionally, as previously discussed, focus group research tends to focus on either the content or dynamics of groups. Incorporating both, the construction of sociograms to assist in the interpretation of findings, ensuring that outcomes represented multiple perspectives rather than one dominant view was an asset of the study. This technique also allowed reflection on the moderation strategy,

which was especially important given the proximity of the researcher to the research topic.

Nonetheless, a number of limitations in the present study warrant consideration. The self-selection of study participants may have introduced potential biases in the findings. Individuals with negative healthcare experiences may have been more motivated to take part, therefore painting a largely negative picture of the Irish healthcare service. Participants in this study reported living with over sixty specific conditions, and while they often discussed commonalities in their experiences, the high heterogeneity in participants' health and background may limit the strength of the recommendations made. Thus, while this study can provide a set of higher-level recommendations for improving healthcare in this group of patients, further research with specific groups will be needed to create tailored pathways for care. Further, focus groups consisted of mostly females and while not necessarily a limitation, this may have restricted the range of perspectives captured. This group composition is unsurprising; it aligns with broader epidemiological patterns which suggest that poorly understood conditions like Fibromyalgia (Häuser et al., 2015b) and Irritable Bowel Syndrome (Kim & Kim, 2018) tend to disproportionately affect women compared to men. As gender and age-related assumptions were often discussed, it is important to recognise the possibility of different healthcare experiences depending on gender (e.g., Samulowitz et al., 2018).

With regards to focus group design, a potential limitation is that focus groups could be skewed by a few outspoken participants, potentially overshadowing the perspectives of more reserved members (Goodwin & Happell, 2009). To address potential biases from dominant participants, the moderation strategy involved using probing and direct questions during the focus groups to ensure all participants had an opportunity to contribute. The online format of the focus groups may have introduced additional barriers to participation for some participants, thus restricting our sample to individuals with higher levels of computer literacy. However, participants were given the option to attend groups remotely or in-person, and all preferred the online focus groups. Some raised concerns regarding mobility and travel to in-person locations, and many found the risk of COVID-19 infection as a deterrent from in-person participation.

Previous research comparing virtual and in-person approaches has suggested that online focus groups provide a valuable opportunity to reach populations whose participation is limited by social barriers, cost, time or travel distance (Stewart & Shamdasani, 2017; Tran et al., 2021). Our experience aligns with this, highlighting significant advantages of the online format. However, some researchers raised concerns about the quality of participant interactions such groups and the need for adjusted moderation strategies (Bolin et al., 2023). To encourage active involvement and interaction, we followed suggestions developed by others (e.g., Santhosh et al., 2021) and encouraged participants to keep their cameras on and speak freely, using probing questions as needed. Further, sociograms revealed good group dynamics with bi-directional interactions among most participants, despite the online format. While participants encountered technical difficulties such as microphone issues or connectivity problems, this was infrequent and did not disturb the overall group process.

6.5.4 Future research

While no single best-practice model for integrated healthcare exists (Darker, 2014), the key focus should be on tailoring healthcare more effectively to the specific needs of service users, especially in cases where care is delivered by multiple professionals and organizations. The present study explored the needs and recommendations of patients, but further development of recommendations would benefit from input from a wider range of stakeholders, including clinicians, policymakers, and community organizations, to develop comprehensive, coordinated, and patient-centered models of care. Ideally, such efforts should reduce rather than perpetuate power imbalances between patients, academics and professionals, maintaining a position of epistemic humility. Adopting this collaborative approach would enhance the feasibility and effectiveness of the proposed solutions, ensuring they are both acceptable and impactful at a systemic level.

6.5.5 Conclusion

The study highlights the significant challenges faced by patients with poorly understood chronic conditions in obtaining accurate diagnoses and accessing coordinated care. These difficulties are exacerbated by the fragmented healthcare system, communication breakdowns, and the absence of comprehensive electronic health records. Patients have emphasised the critical need for healthcare providers to improve communication and demonstrate a better-informed understanding of their conditions, as well as a need for links with allied health services and patient support groups. They desire meaningful dialogues with clinicians where they are believed, and their symptoms, treatment options, and long-term prognosis are thoroughly discussed. The findings suggest the need to establish care pathways and implement robust communication mechanisms among providers to deliver integrated care for this patient population. Taken together, the results of this study underscore that integrating the chronic care model into the Irish healthcare system should be a priority, especially for complex and hard-to-diagnose conditions. Finally, incorporating patient input and tailoring healthcare to the specific needs of individual patients should be the key focus of future work, and the meaningful, sustained involvement of patients in healthcare-related research is needed. To this end, Chapter 7 provides a worked example of involving PPI Contributors in analysis phase of the research cycle.

Chapter 7. Collaborative Analysis

PPI in Co-Analysis of Qualitative Data

7.1 Introduction

As discussed in Chapter 2, from a moral standpoint, PPI is grounded in the view that those most affected by research outcomes should have a say in shaping research agendas, rather than leaving these decisions to elite groups such as researchers, funders, and policymakers (Greenhalgh, 2009; Harting et al., 2022); thus, PPI represents a move towards epistemic justice and the democratization of research (e.g., Liabo et al., 2022; Pearce, 2021). Further, as emphasised throughout this thesis, a crucial aspect of PPI is the meaningful involvement of contributors throughout the research cycle. Yet this level of embeddedness appears largely aspirational at present. Sustained involvement is infrequently achieved due to systemic barriers and tokenistic practices on the part of researchers who involve contributors for political correctness or in a consultative role rather than treating them as true collaborators or partners (Brett et al., 2010; Colomer-Lahiguera et al., 2023; Jackson et al., 2020). For instance, in the context of healthcare innovation, contributors are most often involved in the early stages of research such as problem identification (Cluley et al., 2022).

While substantial progress has been made in including PPI in the planning, design, and data collection phases of research, there remains a notable gap in the involvement of contributors in the collaborative analysis of data in particular (Cowley et al., 2019; Gilfoyle et al., 2022; Hannigan, 2018; Jennings et al., 2018; Powell et al., 2021). This gap is intriguing given that several studies involving PPI contributors in data co-analysis have demonstrated benefits in both quantitative and qualitative research contexts. For example, Staley (2009) illustrated that PPI involvement in mixed-methods research not only enhanced data interpretation but also improved the design and implementation phases, making the research more aligned with public needs. Hannigan (2018) noted that although PPI is infrequently utilized in quantitative analysis, contributors can add

significant value by providing contextual knowledge that enhances statistical modelling, and ensures that outcomes and measurements are aligned with patient priorities. Being more prevalent in qualitative research, several authors have highlighted how contributor involvement enriches the research by challenging researcher assumptions, thereby adding depth and nuance to interpretation. For instance, while acknowledging the extra resource commitments required for collaborative analysis, Garfield et al. (2016) highlighted that lay contributors and researchers worked "synergistically" when coding data, which ultimately enhanced the knowledge gained through the study. Similarly, Jennings et al. (2018) reported that in PPI co-researchers offer a unique and overlapping perspective compared to academic researchers, therefore helping to reduce interpretative bias in qualitative research.

Despite the reported benefits of PPI generally and collaborative analysis specifically, there is little practical guidance on how to involve patient contributors in this step, with only a handful of detailed worked examples available (e.g. Cowley et al., 2019; Hemming et al., 2021; Powell et al., 2021). A need for these examples has been highlighted (e.g., Staley & Barron, 2019) as researchers' subjective accounts of learning through involvement can support wider learning about patient and public involvement, providing insight about *when* PPI works, *how* it works and *for whom*.

7.1.1 Objectives

Given the calls for meaningful involvement of PPI contributors in all stages of research, and the lack of detailed guidance with respect to the analysis stage in particular, this chapter seeks to *(i)* to provide a worked example of meaningfully involving people with lived experience in qualitative data analysis, and *(ii)* to reflect on the process and lessons learned, including barriers and facilitators to co-analysis.

7.2 Methodology

7.2.1 Research Context

This chapter reports on the process of collaborative data analysis using the Framework Approach (Ritchie & Spencer, 1994) as part of the previous study exploring the needs and healthcare experiences of individuals with poorly understood chronic illnesses in Ireland (see Chapter 6). The purpose of involving patient and public contributors in the co-analysis process was to incorporate their unique perspectives and experiential knowledge into the researcher's analysis, thereby producing a more grounded and nuanced understanding of the finding

7.2.2 Analysis Team

7.2.2.1 Contributor Recruitment and Selection

In the context of the present research, the researcher decided to involve two contributors in co-analysis as a larger group was considered impractical in terms of limited resources. The choice to involve two contributors was supported by previous literature (e.g., Hemming et al., 2021), which suggests that involving more than one PPI contributor may reduce power imbalances and tokenistic input. Two contributors were sought as it was felt that the constraints on time available to build relationships with each contributor would negatively impact the overall depth of involvement.

PPI Contributors were recruited online via the PPI Ignite Network Opportunities Noticeboard (n.d.), a platform where researchers can share and the public can discover prospects for involvement. PPI Contributors were eligible to participate in co-analysis if they had lived experience of a poorly understood chronic illness. No previous experience with research or PPI was required of the contributors involved in the analysis. Contributor selection was based on their availability and capacity to complete analyses over a five-week period, willingness to use video-conferencing software for online meetings, and self-assessed proficiency with using word processing tools to review and analyse written transcripts. Eight individuals expressed interest in the PPI Opportunity, though four responded to follow-up information about the study and were subsequently interviewed. Of these, two were unavailable to contribute for the full duration of the analysis at the time of the study.

7.2.2.2 The Co-Researchers

In addition to the primary researcher and thesis author (ND), two patient contributors (LM, EM) and one research assistant (JM) were involved in co-analysis. Throughout this chapter, patient contributors are referred to as "*PPI contributors*", while the primary researcher and research assistant are collectively referred to as researchers. This differentiation is made whilst acknowledging the distinct roles these individuals hold beyond the scope of the current work. The term "*co-researcher*" is applied to all individuals involved in co-analysis.

As acknowledged in Chapter 2 (*Section 2.5.1*), ND has lived experience of poorly understood syndromes and symptoms which has driven the present research into the subject matter. JM is a master's student in Psychological Science and participates in projects mainly in public health, focusing on person-centered approached. EM is a PPI contributor and medical student with personal experience of chronic illness. She has previous experience in stakeholder involved research, promoting the involvement of those being researched in methodology. She is passionate about projects that improve long term outcomes for patients with chronic disease, both at a qualitative and quantitative level. LM is the co-founder and co-president of GACI Global, an advocacy organisation dedicated to supporting patients and families worldwide affected by the rare diseases ENPP1 deficiency and ABCC6 deficiency. Before co-founding GACI Global, LM spent over 20 years in the telecommunications sector, where she held various management roles in product sales, development and marketing. Her journey into advocacy was inspired by her family's personal experience with rare disease, igniting her passion for PPI and driving her commitment to making a difference for patients and families affected by rare and chronic conditions.

7.2.2.3 Terms of Reference

Terms of reference were adapted from resources offered by the PPI Ignite Network at University College Dublin (n.d.) and circulated to PPI contributors for review and feedback over a one-week period. This document outlined the purpose of PPI in the project and terms of collaboration, including the required time commitment, tentative schedule, number of meetings, preferred mode of communication, and the conflict resolution processes. Details of compensation for involvement were discussed during

recruitment and detailed in the terms of reference. Funding was secured from Science Foundation Ireland (SFI) to provide an honorarium to each PPI contributor in the form of a 150 EUR Gift Voucher, in line with institutional guidance on compensation (Trinity Centre for Ageing and Intellectual Disability, 2021). Contributors were also given the opportunity to co-author manuscripts emerging from their involvement. The terms were then revised and agreed upon by the team prior to commencement of the analysis period.

7.2.2.4 Induction and Training

A bespoke induction and training session was created and delivered by the author to ensure a shared understanding of the research project and analytic procedure. This two-hour online session was the initial team meeting whereby all members of the co-analysis team were introduced and reacquainted with the scope of the research study. The session was designed to give a brief overview of quantitative and qualitative research methods and to familiarize the team with the principles and steps of the Framework Analysis (Ritchie & Spencer, 1994) in particular.

Training in Framework Analysis included a step-by-step practice example, focusing on steps of familiarization, coding, framework development and indexing. As part of the training, co-researchers were provided with a two-page sample interview and were asked to read and reflect on content. The sample interview was created specifically for this study and included mock questions and answers from a group of participants discussing their experience of healthcare.

While the training focused on methodology, emphasis was placed on the co-researchers' lived experiences and reflexive practice during the co-analysis process. Here, the importance of reflexivity, the process by which researchers critically reflect on and consciously acknowledge their influence on the research process (Braun & Clarke, 2023) was emphasised. Reflexivity requires ongoing self-examination and awareness of how one's experiences, values, assumptions and positionality shape the research process and interpretations. Sample reflexive questions were provided to contributors and initial impressions of the data discussed among the group.

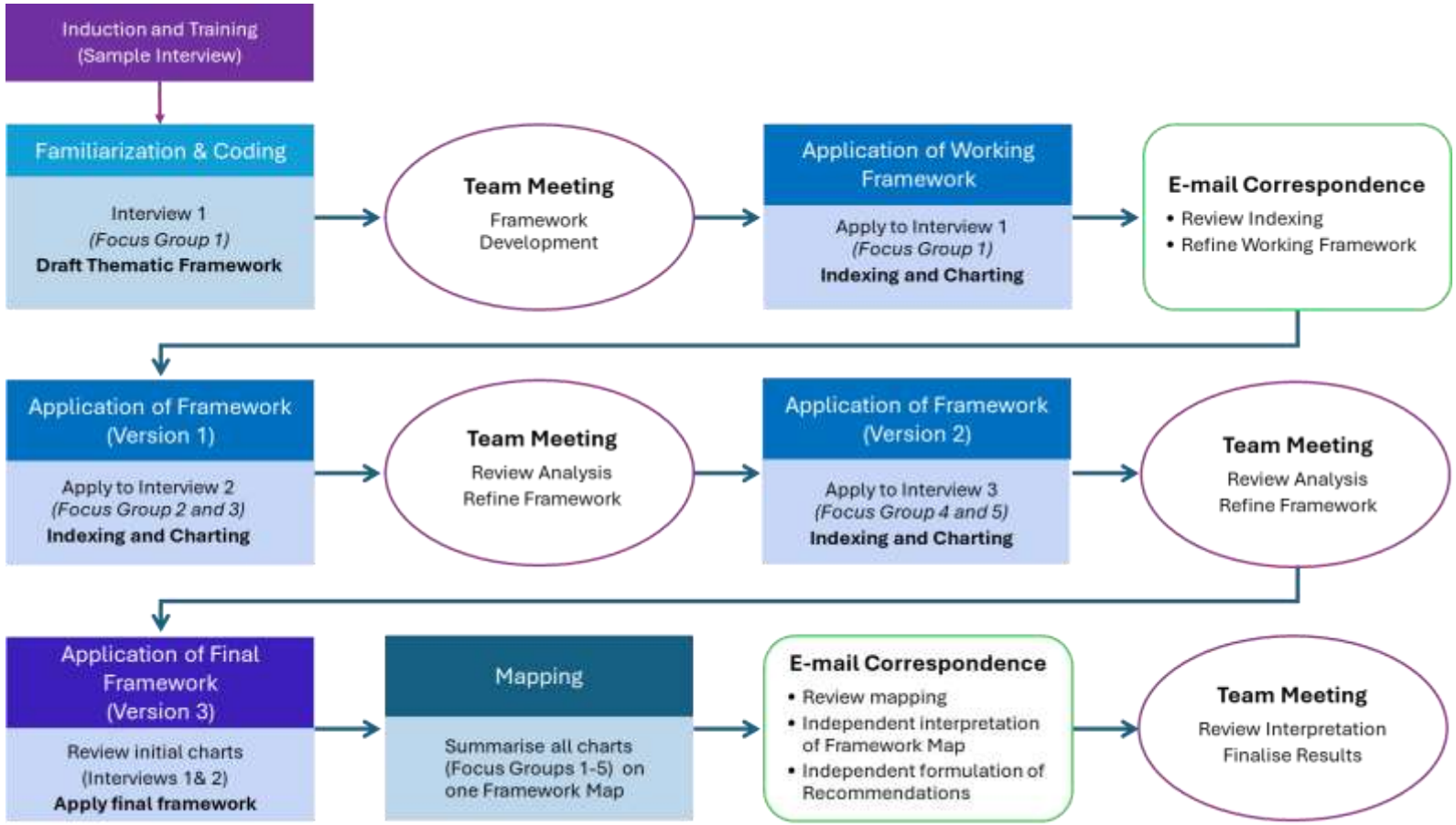
Co-researchers were asked to carry out a brief initial coding on the transcript and to discuss the codes created with the group. Next, they were asked to devise a sample framework, discuss their reasoning and provide feedback to the group. An example of coding and framework generation using the sample transcript was developed by the lead author and presented following the exercise. The charting, mapping and interpretation steps of Framework Analysis were demonstrated and the process of refining the framework discussed. Workshop files, including a step-by-step guide to Framework Analysis and the worked example were shared with the contributors after the training session.

7.2.3 Collaborative Analysis Process

The co-analysis was conducted over a 5-week period with three analytic meetings and one interpretation meeting arranged online. Each team member analysed a combination of three of five focus group transcripts during this time. Transcripts were an average of 17 pages long and were shared with co-researchers for annotation in Word Document form. Details of the procedure are outlined in Figure 7.1. Briefly, Interview 1 was coded by all co-researchers and used to establish an initial working framework for analysis. The framework was subsequently applied back to the transcript by LM, EF and JM and outcomes collated by ND. Revisions were agreed upon by the team and the updated framework was applied by pairs of co-researchers to Interviews 2 and 3, with a weekly group analytic to guide the process. Agendas, coding templates and updated working frameworks were circulated weekly to all to ensure alignment and transparency of the process. A shared OneDrive folder was used to store all interviews, charting summaries and working frameworks.

Figure 7.1

The co-analysis process employed in Study Four



7.3 Worked Example

The steps of Framework Analysis and PPI Contributor involvement are detailed below and summarised in Table 7.1.

Table 7.1

Steps of Framework Analysis and PPI Contributor Involvement

Step	Description	PPI Contributor Involvement
Transcription	Focus group audio recordings converted into written text	None
Familiarization	Analysts immerse themselves in data by repeated reading of transcripts. Notes of initial impressions, ideas and observations about data and reflexive notes are made.	Full
Coding	Coding transcripts used to develop thematic framework	Full
Development of Framework	A working framework aligned with research questions is generated from key issues, concepts and themes identified in the data.	Full
Indexing	Analysts apply the working framework to data. Data is tagged with codes from the thematic framework to organise it systematically.	Full
Charting	Data is summarised into charts which condense and structure information for analysis.	Full
Mapping & Interpretation	All data from charting summaries is collated (mapped) on a chart.	None
	Mapping results are interpreted to identify patterns, connections and key insights about the phenomenon of interest.	Full

7.3.1 Familiarization

During familiarization, researchers ‘immerse' themselves in the data, gaining insights through repeated engagement with content. The purpose of this phase is to thoroughly understand or 'know' the data, achieving a holistic sense of the phenomena (Ritche & Spencer, 1994). In practice, this step involves transcribing interviews, repeated listening to recordings and reading of transcripts and making notes to capture initial thoughts and reflections (Parkinson et al., 2016). Study aims and objectives must be referred to in order to ensure that data relate to these from the outset (Ritchie et al., 2003).

All focus groups were conducted and transcribed by ND and written transcripts were shared with co-researchers on a weekly basis. While transcription and listening are an important facet of familiarisation (Silverman, 2024), interview recordings were not shared with PPI contributors to maintain the confidentiality and anonymity of focus

group participants. Instead, PPI contributors familiarized themselves with data by repeated reading of written transcripts. This decision was made due to the possibility that the PPI contributors, being patients themselves, could identify individuals taking part in the focus groups. JM co-facilitated three of five focus groups during data collection, thus providing an added opportunity for immersion.

When datasets are small, it is possible for the familiarization phase to include all study data. However, this is neither practical nor required in the framework approach, especially when working with voluminous datasets (Ritchie et al., 2003; Srivastava & Thomson, 2008). One focus group transcript (Interview 1) was reviewed by all co-researchers in the familiarization stage of analysis. Co-researchers were asked to read, re-read and reflect on the interview over a period of one week, making notes of any impressions or main ideas that appeared to recur in the data. While not an explicit component of framework analysis, reflexivity was emphasised at all steps of the analytic process. Reflexivity was considered particularly important among co-researchers with both academic and lived experience of the topic. Team members were guided to engage in reflexive practice throughout the analysis of each interview. A document containing sample reflexive questions was provided and co-researchers were encouraged to reflect on these before undertaking analysis, during familiarization and after coding of each interview. Guiding questions were developed by drawing on existing literature (e.g., Olmos-Vega et al., 2023) and are included Appendix E1. It was stressed that sharing of personal reflections with the team was not required, but that a discussion of impressions would be invited at the subsequent meetings. In addition to engaging in reflexivity during data analysis, co-researchers were invited to continuously reflect on their experience of involvement in co-analysis process.

7.3.2 Framework Identification

7.3.2.1 Coding

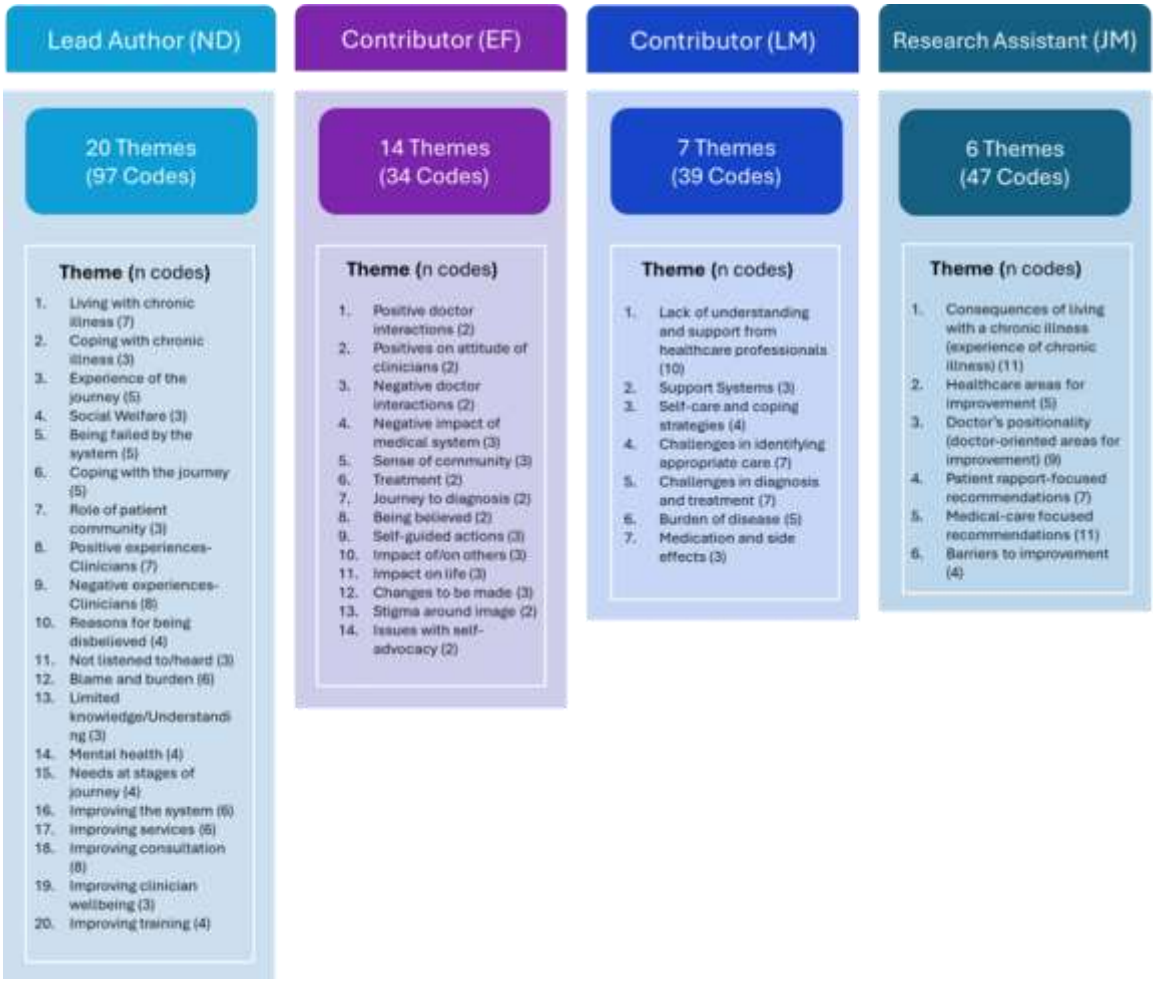
The second step of Framework Analysis is coding of data. Following familiarization, the researcher reads the chosen transcript(s) line-by-line and applies a code or label which describes a meaningful or important component of the passage. The aim of this phase is to classify data so that it can be organised and compared systematically with other parts of the transcript. Key themes are identified to construct a working framework that categorises significant aspects of the data in alignment with the research questions.

All co-researchers independently coded Interview 1 and devised a possible thematic framework for discussion. Co-researchers were given an option of completing coding via pen and paper, but all opted to use the Microsoft Word comment function instead. Codes, descriptions and example quotes were transferred by contributors into a Coding Template provided by ND. Each researchers' codes were compiled side-by-side in a single document which was shared and reviewed with the team at the framework development meeting.

It has been suggested that involvement of various stakeholders can be productive at the initial coding stage, particularly with regards to ensuring one perspective or viewpoint does not dominate the analysis (e.g., Gale et al., 2013). Some differences between PPI contributors and academic researchers with regards to coding were observed and discussed among the team and are outlined in Figure 7.2. The highest number of initial themes (*n*=20) were generated by the lead author, reflecting a granular analysis of content in Interview 1. Fewer themes and codes were generated by PPI contributors and the research assistant, who were more inclined to note broad categories of patient experiences and challenges related to healthcare. We noted that despite coding differences, there was a large degree of overlap in proposed thematic structures and overarching concepts among the team

Figure 7.2

Initial coding and development of a thematic framework



7.3.3 Framework Development

The third step of analysis involves developing a working framework. This is an iterative process, and the term "working" highlights the framework's flexible and evolving nature during the early stages of data analysis. After coding selected transcripts, researchers collaborate to compare and decide on a set of codes and a structure to apply to subsequent transcripts. Themes and concepts can be grouped, ranked or ordered in a way that helps address the focus of the study (Goldsmith, 2021). The goal is to organise the data into meaningful categories, facilitating effective retrieval, exploration, and examination in later stages. Regular team discussions are recommended to ensure mutual understanding of codes and categories (Parkinson et al., 2016).

All co-researchers were actively involved in developing the working framework. Following the initial comparison of coding, co-researchers were asked to reflect on their notes and to identify the most pertinent or meaningful themes in the dataset. According to Ritchie and Spencer (1994), the process of framework development should be informed by both predefined concerns and issues that arise during the familiarization stage of analysis. We therefore decided to base the initial framework structure around the three key areas of the focus group interview schedule, as discussed in detail in Chapter 6 (*Section 6.2.2.3*): participants' experiences of chronic illness, their healthcare journey and needs and recommendations for change.

To facilitate the process of framework development, ND prepared a digital whiteboard with blank sticky notes and the broad topic headings outlined above. Co-researchers were asked to enter key themes on the sticky notes, and to arrange them under the relevant heading to develop a working structure (Figure 7.3). During this exercise, it became apparent that some topics were difficult to organise and spanned broad issues (e.g., stigma) and questions arose with regards to the exclusivity of the broad framework categories. For example, co-researchers were unsure of whether the healthcare journey is part of the general chronic illness experience, or whether themes related to the journey these represent a separate category of data. The arbitrary divide between such categories was discussed, ideas were further fleshed out and agreement about an initial structure reached, resulting in a working framework with ten categories, 27 themes and 81 codes. The working framework is presented in Table 7.2.

Figure 7.3

Collaborative development of an initial working framework

Note. Sticky note colours represent prominent themes within framework categories (Yellow= Experience of chronic illness, Blue= Experiences of healthcare, Pink and Gree*n*= Needs and Recommendations for Change). Orange sticky-notes represent topics which can span multiple categories or themes.

Table 7.2

Working Framework Structure

Category	Theme
1. Experiences of Living with Chronic Illness	Living with chronic illness
	Community and support
2. Healthcare Experiences	Experiences with healthcare
	The patient journey
	Barriers to effective care
3. Needs and Recommendations for Change	The consultation
	Service improvement
	Training and development
	Clinician wellbeing
	Knowledge generation

7.3.4 Indexing and Charting

The next phase of the framework approach is called “indexing”. Once a draft working framework is developed, it is systematically applied back to the transcripts to explore “fit” (Ritchie et al., 2003). In practice, indexing involves reading through the transcript text and assigning specific codes from the working framework to the data. As researchers index the data and gain a deeper understanding, they might also rename, collapse, delete develop new themes or categories and ultimately refine the working framework. Indexed data are summarised and condensed into tables in a process called “charting”. The process has been described as a way of organising data into coherent groupings, or more broadly, as secondary data reduction (Kiernan & Hill, 2018). To chart an interview, the researcher works through each framework category or subcomponent and summarizes the indexed data per unit of analysis (e.g., participant or focus group). The aim is to reduce original data into manageable sections of text that capture the essence without losing context or detail. Brief summaries are entered into charts and example lines, page numbers or quotes from the transcript can be noted. This process facilitates the retrieval of data in later stages of analysis and underscores the systematic nature of the framework approach. Indexing and revisions continue in an iterative process until all data are coded on the final framework (Goldsmith, 2021).

In the current project, the co-analysis process involved three phases of indexing and framework refinement. Three co-researchers (JM, EF, LM) indexed and charted interview

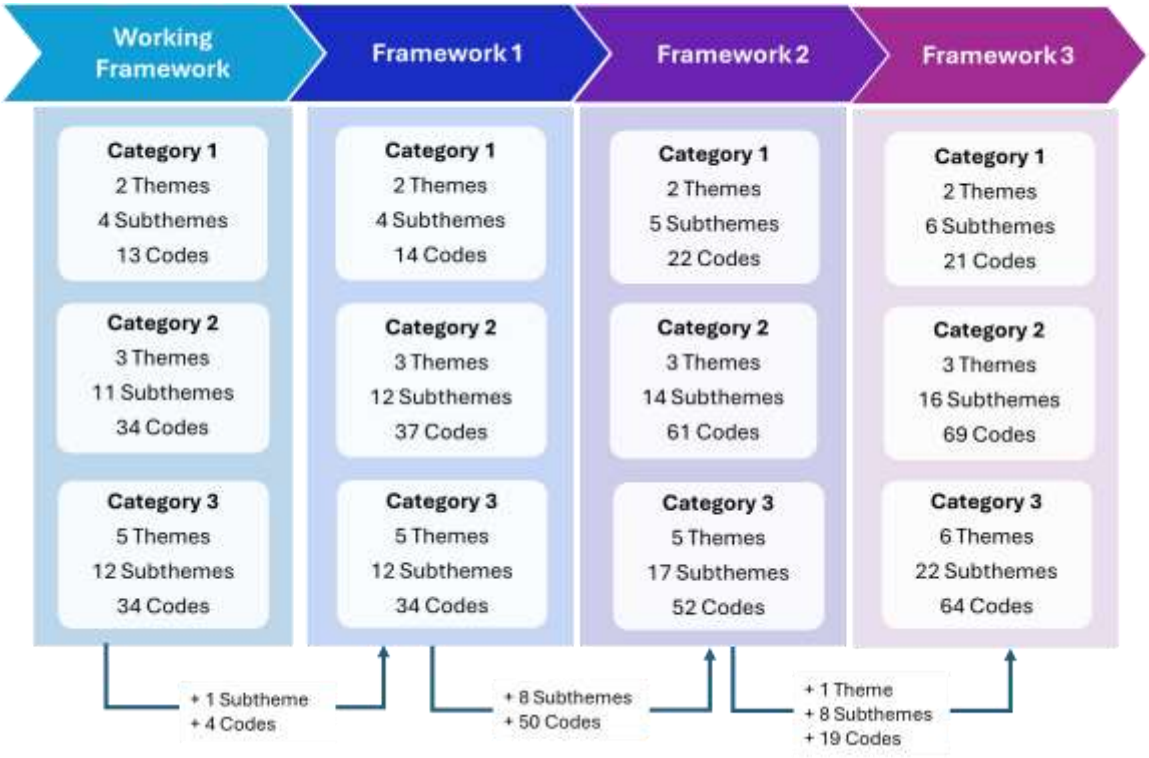
1. Each of the remaining interviews were indexed and charted by two co-researchers, pairs consisting of one PPI contributor (EF, LM) and one researcher (ND, JM). When indexing, co-researchers were encouraged to apply an 'Other' code to data that did not fit the structure of the working framework. They were also encouraged to consider possible codes, themes or categories that apply, and to include these in their charting summaries. Charting summary templates were developed and provided by ND (Appendix E2). In the summary charts, co-researchers entered data relating to the presence or absence of a theme and summarised the data along with example quotes or line numbers from the assigned transcript. Charts for each interview were collated by ND in a single document and discussed at team meetings. Suggested modifications to the framework were agreed and applied to the subsequent transcript, and the cycle repeated with the next set of data.

For the most part, there was good overlap in indexing and charting between co-researchers, indicating similar interpretations of data and approaches of coding of data. Any discrepancies in coding were a key point of reflection and discussion, resulting in modification of theme contents or agreement on coding practices. For example, differences in coding emerged between the PPI contributors and researchers in relation to the subtheme *Additional Burden of Healthcare.* Upon discussion, PPI contributors were initially more likely to code burdens like care coordination and travel to appointments as part of the subtheme *Burden of Illness or Disease.* Following consultation, a distinction was agreed that the latter subtheme related primarily to the direct impacts of chronic illness, or those attributed to symptoms. Healthcare-related burdens were considered those emerging as a result of suboptimal care rather than the symptoms or illness. However, it was recognised that the divide between healthcare-related and symptom-related challenges was somewhat arbitrary, as both elements contributed to the totality of participants' lived experience of chronic illness. Similarly, some questions arose with regards to coding of text into multiple categories. For instance, assumptions and biases were key to the participants' illness experience, primarily lack of support from others, yet these were also a major component of the category *Healthcare Experiences*, especially negative experiences with clinicians. In such cases, we decided to include parallel codes in each subtheme within the framework.

While substantial changes to the thematic structure were not made through the iterations, 17 subthemes and 73 codes were added as the framework evolved. Most changes related to elaboration of subthemes within the existing thematic structure, as well as re-naming of themes and subthemes. For instance, the initial working framework failed to capture 'Negative experiences with treatment' as independent from 'Negative experiences with clinicians', resulting in the addition of the theme to the subsequent iteration. Framework development was an iterative process, with three revisions before a final structure was reached. The evolution of the framework is outlined in Figure 7.4, and the final framework structure presented in Table 7.3.

Figure 7.4

Iterative development of the framework structure



Note. Category 1= Experiences of Living with Chronic Illness; Category 2= Healthcare experiences; Category 3= Needs and Recommendations for Change.

Table 7.3.

Final Framework Structure

Category	Theme
1. Experiences of Living with Chronic Illness	Living with chronic illness
	Community and social support*
2. Healthcare Experiences	Experiences with healthcare
	The patient journey
	Barriers to effective care
3. Needs and Recommendations for Change	The consultation
	The service*
	Training and development
	Clinician wellbeing
	Knowledge generation
	Implementation of change**

Note. * change in theme name **change in thematic structure

7.3.5 Mapping and Interpretation

The final step in framework analysis involves the synthesis of insights from earlier stages of analysis to achieve a deeper understanding of the phenomenon. Here, researchers combine key learnings from earlier steps with comparisons across and within units of analysis and framework components. This step is often considered the most challenging and difficult to articulate, as it requires an intuitive and imaginative approach rather than a purely mechanical one. According to Ritchie and Spencer (1994), researchers must not only manage or examine the data but also engage in a creative and insightful process to produce meaningful knowledge. There is no single outcome of this process, and results can be represented in various forms, including a description or explanation of key concepts or phenomena, or a demonstration of patterns and associations within the dataset (Ritchie & Spencer, 1994). While the importance of documenting the interpretation has been stressed (e.g., Srivastava & Thomson, 2008), little guidance is available with the regards to the practicalities of this phase of analysis. Some researchers do not describe their mapping or interpretive process at all.

To facilitate the process of interpretation by all co-researchers, ND devised a single mapping matrix to collate summaries of key findings across the five focus groups in the dataset. JM and ND transferred data from the focus group charts to this map, revisiting interviews that were indexed using an earlier version of the working analytic framework and applying the final framework. As discussed in Chapter 6 *(Section 6.2.3.1),* drawing on the work of Goldsmith (2021), an indicator of strength of evidence was included to highlight patterns in data both within and across the five focus groups. The complete map of evidence (see Appendix D5) was circulated among all team members for review and feedback. Co-researchers were asked to review the matrix and consider connections within and between focus groups and categories, and consideration of specific recommendations was encouraged. They were also asked to revisit the interviews they had coded and identify any meaningful quotes that would support any of the framework components. A guiding document was provided to assist independent interpretation over a three-week period (see Appendix E3).

Following this, a final meeting was arranged with the team to discuss the mapping framework and study outcomes. During this meeting, interpretative ideas were explored and fleshed out by the team and key results were discussed in preparation for manuscript write-up. While all co-researchers were involved in the interpretative step, the input of PPI contributors was emphasised. PPI Contributors were surprised at the low strength ratings of the theme *Community and Social Support,* which was interpreted as a strong component of the focus groups. This prompted ND to review all charting summaries and the framework matrix, but changes to strength ratings were not justified by the data.

7.3.6 Write up

All members of the analysis team were invited to contribute as manuscript co-authors. An initial complete draft was written by the first author, drawing on feedback and ideas generated during co-analysis and the interpretation phase in particular. The draft was circulated to the team for feedback, and PPI contributors were actively encouraged to suggest changes or make revisions. Final manuscript preparation is planned for post-thesis completion.

7.4 Discussion

The present chapter provides a worked example of involving PPI Contributors in the collaborative analysis of data from study four, reported in Chapter 6, which explored the experiences and healthcare-related needs of individuals with poorly understood chronic illnesses. The overarching aim was to illustrate and reflect on the process od co-analysis. The sections below provide reflections of all co-analysts, lessons learned with regards to challenges and facilitators, as well as recommendations for future projects.

7.4.1 Process Reflections

Each member of the analysis team provided a reflection on their experience of involvement in the project within one month of completion. Co-researcher reflections are included first and presented as quotes to preserve integrity of their experience, followed by the author's reflections on the process.

7.4.1.1 Co-researcher one

> *"The involvement of patients in the analysis was a great opportunity to deepen the richness of information and outputs. It was interesting to see a great overlap of codes and quotes from one person to another despite the diversity of our backgrounds. Our conversations on the data helped me to shift my perspective regarding the strength of many participants' statements, particularly whether the patients were minimising their experiences or not. As a researcher, I was very concerned with the volunteer bias, and I initially started working under the assumption that the patients who volunteered to participate in the study had worse experiences than the average patient with a chronic illness. However, the co-analysts helped me give more credit to the criticisms, as some themes could emerge from them for potential improvements. More precisely, the patient's perspective in the co-analysis helped me understand that the participants could potentially be controlling their choices of words and opinions to not appear too negative despite how negative their experiences were. This position was echoed in the interviews where several patients shared how they were labelled negatively when not behaving or looking a certain way. Their expertise prevented me from overlooking some ideas and invalidating patients' experiences as a researcher.*
>
> *It was also interesting to have a fresh eye on the data from people who were discovering sequentially the interviews, whereas the researchers had more of a vision of the ensemble and had already started to compare the focus groups with one another. The diversity of impressions emerging allowed us to have insightful conversations and inputs on the different elements of the focus groups."*

7.4.1.2 Co-researcher two

"Before working on this project with Natalie, I didn't have any experience with coding interviews. Reading the interviews was very insightful, and it was interesting to see that most patients had similar positive and negative experiences, even though their medical conditions and journeys were quite different. The interviews were longer than I imagined, and the coding also took a lot longer than I anticipated. However, as time went on, I found that I could more easily identify the recurring themes, and this was quite satisfying. Natalie was very helpful and supportive throughout the process. Overall, it was a very positive experience working with the team on this project, where everyone was very engaged and invested in the process."

7.4.1.3 Co-researcher three

"It was fascinating to read others' stories. Some stories I connected with a lot more as a young person and it brought personal comfort to realise I wasn't the only person suffering from some of these issues. It highlighted the importance of studies like this to me to create a collaborative voice and also highlighted the need for support groups and information sources that can get drowned out when only single voices request them. You get a very unique view when you read the scripts from multiple focus groups that have occurred separately. It was interesting to see that bringing together unconnected individuals on different days produced so many of the same themes overall. Coding was time-consuming at times, but the gratification that came out of seeing a final script finished, and hearing within scripts the importance that participants placed on patient-centred projects like these consistently made everything worth it, and I would contribute to a similar project again in a heartbeat. As someone with experience on both the patient and healthcare sides, a lot of the time when trying to formulate recommendations from what we had heard in meetings I had to remind myself to balance what is feasible in our current system and also to allow situations to be put forward that think outside the box, i.e. larger healthcare centres for chronic diseases where all care is integrated. This seemed to be echoed by all participants in coding regardless of their background."

"The collaborative format was useful- particularly after we had coded all of the scripts and came together to discuss the results. I felt having consistent Zoom calls allowed us to play off of each other and our ideas, and hearing other's suggestions brought up thoughts I hadn't had before or things I may have forgotten during interviews. Having these calls also allowed individuals who had reviewed different interviews to share what was similar, and what was different during these, and pick out the overarching themes that encompassed all participants' views well. The PPI aspect of the project was present in all parts, and I feel was very important when coding script and coming up with final recommendations. I understood everything individuals were asking for from a personal level from my own experience, and I think this prevented ideas or suggestions from getting lost in translation as many of the asks of participants are things I wish for myself".

"I feel that this project has made me realise the importance of PPI work, which is something I will take with me in future ideas. I have never been involved in a PPI

project to this extent and have usually been consulted in the beginning and do not get to see the finished outcome. I appreciated our involvement from the beginning, and the genuine consideration of our input throughout the project, and hope this is something that is carried forward. Qualitative work is something I have not participated in, so I was starting from scratch in framework analysis. I feel I probably still have a lot more to learn but this project has given me great insight! I also learned a lot while forming codes and themes around interviews. The suggestion for us to write reflections while coding allowed me to point out any biases I may have and catch them while doing this qualitative work. This was helpful and something I would not have considered doing before and possibly prevented any of my ideas from becoming too far integrated into themes and codes."

7.4.1.4 Doctoral researcher

The opportunity to involve patient contributors in the co-analysis of focus group data was an immensely valuable experience, both personally and academically. While the analysis was grounded in data and followed the rigour of Framework Analysis, ongoing discussions and reflections with PPI contributors provided novel insights and an in-depth appreciation of challenges faced by individuals with poorly understood chronic illness. The PPI contributors generously shared their personal perspectives on the data yet provided insights from very different positions to that of my own: one of being a parent, with experience of navigating and coordinating complex care, and one of being a medical student, with insider experiences of medical culture, training and the challenges faced by healthcare practitioners. The unique lenses of the contributors expanded and complimented my perspectives and led to a greater understanding of issues surrounding training, development, and system integration. Overall, I was often taken aback by the remarkable strength, resilience and drive to advocate of the PPI contributors, not unlike the research participants involved throughout the research. Furthermore, the involvement of a research assistant with an interest in healthcare design expanded the conversations about targets for change, enriching the overall analysis. Discussions of public health issues and sociological perspectives more broadly challenged me to "take a step back" and consider alternative lenses through which system-level issues could be viewed.

With regards to practical aspects of the process, involving PPI co-researchers in analysis was no more challenging than onboarding and involving an academic collaborator in the

project. While several considerations were important, including that of the contributors' availability and understanding of research methods, I did not perceive this as a barrier but rather an expected part of collaboration. What was of most importance was to ensure that PPI contributors felt empowered in the process of co-analysis and did not perceive the researchers as experts, and that involvement in the project did not add considerable strains to their daily schedules. Although a schedule and weekly agenda were set ahead of time, we consistently evaluated progress and revised the schedule as needed. Such practices are in line with PPI guidelines, such as the Practical Guide for Researchers published by the HSE (2021), which emphasize respect, transparency, flexibility and adequate preparation for PPI activities.

Some authors have suggested that it may be easy to lose contributors' voice in the analytic process and that researchers should encourage patient contributors to primarily draw on their lived experiences when analysing and interpreting data and "step back" to highlight their contributions (Goodman & Thompson, 2018; Sweeney et al., 2013). Deliberate efforts were made to ensure that PPI contributor's inputs were the leading voice of the analysis and were supported by the doctoral researcher and research assistant. For example, I did not code all the transcripts, and each interview was analysed in dyads of PPI Contributor and Researcher. My main role was to facilitate data exploration, guide the process, and synthesise the findings and viewpoints of the entire team. Furthermore, the team meetings focused on the process of analysis, co-researchers' reflections on the data and framework, as well as an exploration of personal perspectives on recommendations for improving healthcare. To this end, I explicitly invited the PPI contributors to lead discussions prior to offering my own interpretations of data. Maintaining a sense of epistemic humility was key to ensure all contributions were given equal weight and consideration, and this was the leading position from which I approached the co-analysis, and the research conducted as part of the thesis overall.

7.4.2 Lessons Learned

While the experience of co-analysis was a valuable and positive experience, several important considerations- namely challenges and facilitators- were noted during the process. Previous literature has highlighted a number of barriers to PPI more broadly, including conceptual and practical challenges to sustained contributor involvement.

Among key barriers are those related to recruitment of PPI contributors, funding for PPI activities, time pressures and perspectives on the value and importance of PPI, as well as the possibility of professionalising the contributor (e.g., Blackwell & Rowbottom, 2020; Coupe & Mathieson, 2020; De Simoni et al., 2023; Jackson et al., 2020; Ocloo et al., 2021; Staley et al., 2019). The section below considers several of these in relation to the collaborative analysis conducted, providing an overview of the lessons learned and implications for future research.

7.4.2.1 Recruitment of co-analysts

The recruitment of PPI contributors was not a challenge in the present research and, as discussed earlier, was facilitated by the PPI Opportunities Noticeboard. However, this method of recruitment may contribute to the marginalisation of individuals who are less likely to engage in online forums or are unaware of such participation opportunities (e.g., Blackwell & Rowbottom, 2020). Several authors have highlighted that recruitment practices, contributor diversity and diversity of methods employed are a concern in PPI more broadly, impacting the type of knowledge that is co-produced (Gilfoyle et al., 2023; Hatch et al., 2024; Rose & Beresford, 2024; Tierney et al., 2021). Thus, alternative strategies of recruitment, for example through local community-based services, should be considered specifically to reach patient populations with no prior experience of or awareness of PPI initiatives.

7.4.2.2 Compensation and guidance

While it is recognised that PPI contributors should be compensated for their time, in reality, money to support PPI typically arrives once a project has been funded, therefore affecting how PPI contributors can be involved at early stages of the proposal (Fox et al., 2023; Jackson et al., 2020; Ní Shé et al., 2020). Similar challenges have also been discussed in the context of doctoral research (e.g., Coupe & Mathieson, 2020) whereby funding specifically for PPI activities is limited. In the context of the present study, the doctoral researcher obtained an honorarium from Science Foundation Ireland specifically for co-analysis with patient contributors during the third phase of research, enabling compensation in the form of a gift voucher. Non-financial compensation in the form of co-authorship was also offered to emphasize shared ownership over the research outcome. Although a voluntary patient panel contributed to the conceptualization of the

study, funding at earlier stages could have enabled fuller and sustained involvement in the execution, write-up and dissemination of research, for instance. A key recommendation specifically for early-career or doctoral research is therefore for clarity on funding allocation for PPI activities, as well as transparent and accessible guidance on compensation procedures in the local context, national and institutional, prior to commencement of research activities.

7.4.2.3 Training of co-analysts

The literature on patient and public involvement has highlighted the "professionalisation paradox" (Ives et al., 2013), where public collaborators need training to contribute effectively to research, but this learning process can undermine their 'lay' status. In light of this is finding a balance between enhancing PPI contributors' research knowledge and skills to support their role as partners, while avoiding the risk of professionalising their involvement (Cowley et al., 2019). As discussed previously, PPI contributors in the present study were provided with brief training in methodology and the Framework Analysis in particular, with analytic templates provided during the process to ensure tasks were clear and accessible. This training was necessary to ensure all team members had the technical skills to engage in the full analysis process. However, reflexive practice was heavily emphasised, and personal perspectives were extensively discussed in meetings to capture the contributors' unique expertise, particularly during data interpretation. Future efforts should similarly seek to balance practical skills and lived experience in analysis, though additional considerations and resources might be needed depending on data type and analytic methods (e.g., specialised software), as well as PPI contributor confidence and literacy.

7.4.2.4 Analytic method

The adoption of a structured analytical approach, such as the framework analysis, acted as a key enabling factor by providing a systematic framework to guide the planning and execution of the collaborative data analysis process. As emphasised by literature (e.g., Dixon-Woods, 2011; Gale et al., 2013; Ward et al., 2013) the Framework Approach provides clear steps to follow and produced highly structured, summarized data outputs. This transparent methodology enabled ongoing collaboration between all co-analysts, and did not require PPI contributors to have an academic background or qualitative

research experience to partake in the process. PPI contributors were fully involved in most steps of the analysis, apart from transcribing the interviews and manually transferring all the charting summaries into a single framework matrix (i.e. mapping). Involvement in the iterative process of developing a thematic framework, indexing of data and developing charting summaries allowed PPI contributors to go beyond offering their insights in a consultative manner, but rather engage with the data on a deeper level, facilitating a richer dialogue that ultimately led to more meaningful interpretations of the findings.

However, it is also important to recognise that contributors' familiarisation in this study was limited to anonymised transcripts only. Some qualitative researchers (e.g., Brandenburg & Davidson, 2011; McLellan et al., 2003; Vanover, 2021) have argued that listening to audio recordings and transcribing interviews is in itself a valuable step of analysis, and crucial to becoming more immersed in the data. In the present study, audio recordings were not shared in order to protect participant confidentiality, given the small and specialised nature of the sample population in the Irish context. Provided that such data sharing is transparent and included in ethical applications and consent forms, larger studies could easily involve contributors in all stages.

Several authors have recommended early involvement of PPI contributors to enable a sense of ownership (e.g., Cashman et al., 2008; Pearson et al., 2024; Selman et al., 2021). In the analysis phase, this could include involving PPI contributors in discussions about the choice of analytic method. While the contributors received training on various qualitative analysis approaches, the lead author selected the Framework Approach prior to engagement with PPI contributors. Where possible, future collaborative approaches should encourage PPI contributors to reflect on and assist in selecting the analytic approach to be used.

Finally, some have cautioned that consideration should be given to the matter in which use of technology can exacerbate marginalisation (Adeyemi et al., 2022; Blackwell & Rowbottom, 2020). On agreement with PPI contributors, the co-analysis process in this study was carried out fully online whereby materials were shared digitally, and meetings were held using a video-conferencing platform. This approach was discussed with the PPI contributors, who had the necessary computer skills to participate. However, such an

approach could pose a barrier to contributors with lower levels of computer literacy or access to online resources and thus warrants consideration in future PPI endeavours.

7.4.2.5 Process management

Another often discussed barrier to PPI relates to time constraints, both on the side of the researcher and contributor (e.g., Coupe & Mathieson, 2020; Domecq et al., 2014; Jennings et al., 2018). While many authors have emphasised that engaging stakeholders in the co-analysis of data as part of PPI initiatives requires additional planning and flexibility, this is not necessarily a barrier to success. Building on insights from previous research that effectively incorporated patient and public involvement (e.g., Coupe & Mathieson, 2020; Dawson et al., 2020; Jones & Hunt, 2022), key enabling factors in the present study included intentionally planning PPI activities and objectives from the project's inception, setting all analytic meetings ahead of time with flexibility for personal circumstances, and documenting the progress of activities that were shared among the team. However, the analysis was scheduled over a 5-week period, and while the process was manageable, ideally a less pressured timeline would have been preferred should adequate compensation for sustained involvement be available. The literature has also highlighted challenges related to uncertainty about the goals of involvement, value, and responsibility of contributor (Ocloo et al., 2021). Goals and responsibilities were outlined in a Terms of Reference document, ensuring all contributors understood their roles and responsibilities in the project.

7.4.2.6 Rapport

The importance of building and maintaining trust between PPI contributors and researchers has been emphasised (Dawson et al., 2020; Staley & Barron, 2019) yet building such an alliance may be uniquely challenging as it requires researchers to be open to critique and disagreements and to share power (Goodman & Thompson, 2018; Pearce, 2021). This requires reflexive consideration of, and respect for, the diverse epistemological and ethical stances held by members of the team (Park & Zafran, 2018). As previously discussed, a key enabling factor of collaboration in in the present study was the lead researchers' positionality and personal experience with the subject matter, and an explicitly stated relinquishing of "expertise" in the analytic process. In addition to taking a "step back" and encouraging the co-researchers to share their perspectives first,

PPI contributors were also encouraged to actively challenge the researchers' views and voice any disagreements, which were recognised as essential to enhancing overall understanding. Such discussions were largely constructive, though an unanticipated challenge arose when PPI contributors and researchers had mixed interpretations of the final framework analysis results. To address this, the lead author reviewed all charting summaries to address findings that were unexpected or surprising to the PPI contributors. While this process did not ultimately alter the final results, it is important to consider this possibility in future research and explore ways to resolve such challenges or criticisms should they arise. The goal should be to maintain meaningful contributor input while also preserving academic rigor, thus researchers should consider strategies to maintain this balance.

7.4.3 Conclusion

The overarching aim of this chapter was to provide a worked example of, and reflection on, the involvement of PPI contributors as co-analysts in research. This work contributes to the expanding body of research (e.g. Cowley et al., 2019; Hemming et al., 2021; Powell et al., 2021) by offering a transparent step-by-step example of collaborative analysis using the Framework Approach. It also provides a reflective account on the process, the need for which has been underscored (e.g., Staley & Barron, 2019) to advance PPI practices in research. Study-specific barriers and facilitators are highlighted for consideration in future co-analysis endeavours. As illustrated and captured by the reflections, such engagement is feasible and valuable for and beyond the final outcomes of research, providing a unique learning opportunity for both the contributors and researchers.

Chapter 8. Conclusion

Since each chapter in this thesis included its own specific conclusions, this final chapter serves as a concise synthesis, providing an overarching consolidation and summary of the key findings and implications for future research. The chapter begins by providing a summary of the key findings of each study, then highlights core themes identified across the four studies comprising the thesis. The discussion situates the results within the broader context of integrated and patient-centered care, highlighting core experiences, needs and recommendations articulated from the patient perspective, emphasising their implications and potential impact on service design and delivery. Subsequently, the overarching strengths and limitations of the research programme as a whole are discussed. The chapter concludes by outlining directions for future research and opportunities for advancing work in this domain.

8.1 Research Findings

The research conducted in this thesis offers a nuanced exploration of the experience of living with and navigating healthcare in the context of chronic, poorly understood or "medically unexplained" illness. Collectively, the studies conducted across four phases of research reveal the complexity of patient journeys and provide compelling evidence for the need to address gaps in care delivery and adopt an integrated, patient-centered approach to healthcare. Embedding PPI as a central component throughout, the research programme ensures that the insights and priorities of individuals living with the conditions shaped the research focus and outcomes. The objectives and key findings of each study of the research programme are summarized in Figure 8.1 and discussed below. For detail of PPI at each stage of research, see Figure 2.1 (Chapter 2, *Section 2.2*).

8.1.1 Phase One

Study One, the systematic literature review, aggregated qualitative evidence from a total of 143 studies regarding the experience of individuals with MUS, FM and ME/CFS (PwC) and of healthcare practitioners (HCP) working with this patient group. The study identified a range of multi-faceted and interconnected themes with respect to both groups, unified by the challenge of sensemaking at the limits of biomedical knowledge.

For PwC, the most common aggregated findings related to the patient journey, particularly their healthcare needs and expectations, and experiences of illness, particularly physical symptoms, the psychological impact of chronic illness, and making sense of the complexity of their condition. For HCPs, the most prevalent findings in the literature concerned making sense of patients' symptoms and experiences of and concerns surrounding consultation management, including diagnostic challenges and establishing a course of action. Together, the results magnified challenges related to medical uncertainty and revealed a lack of person-centeredness in healthcare, thereby pointing to the patient-clinician dynamic as a viable target for change.

8.1.2 Phase Two

Study Two was a cross-sectional mixed-method survey conducted to explore the knowledge, understanding and perspectives on ME/CFS and FM among a sample of adults familiar with these conditions in Ireland, as well as their appraisal of public and clinician knowledge and awareness. The results revealed a good level of understanding of these conditions among respondents, who highlighted core issues of stigma and invalidation, the burden of symptoms, and healthcare-related challenges faced by PwFM and PwME. Respondents were attuned to the misconceptions surrounding both conditions and the consequent discrediting of patients as suffering from a "real" condition in both social and healthcare settings. The results also revealed pessimistic outlooks on clinical processes, with challenges identified in relation to diagnosis, treatment and symptom management, which were attributed to inadequate biomedical knowledge and perpetuated by poor quality research. Notably, participants expressed overwhelmingly negative views on clinicians' knowledge and awareness and emphasised deficits in formal education and compassion among healthcare practitioners working with this patient group. Together, these findings revealed a troubling lack of confidence in the healthcare system among the sample of adults, both those with and without personal experience of chronic illness, underscoring the need to further explore patients' lived experiences in this context.

8.1.3 Phase Three

Study Three comprised of an in-depth exploration of the lived experiences and healthcare journeys of individuals with diverse chronic poorly understood conditions in Ireland. The findings provided a rich, multi-dimensional phenomenological account of the transformative effects "unexplained" chronic illness has of individuals' lifeworlds, including their relationships with their identity, their bodies, and others. Narratives revealed a deep disappointment in and frustration with healthcare and the patient journey, highlighting core issues of disbelief, dismissal, and a sense of an ongoing battle for care in a broken system. Combined with patient journey maps, the findings illustrated patients' experiences of navigating a cycle of unsuccessful appointments or of prolonged waits for care. The central role of the GP in this sequence was evident, highlighting a need for better care pathways and clinical decision-making supports for referrals and investigations. Results pointed to complex power dynamics in consultations, highlighting issues of voice and silence, bringing to light a lack of person-centered approaches to care. However, there was also evidence of "landmark" events or turning points in patient journeys, casting light to what a satisfactory system could be like. Narratives revealed qualities of "good" doctors, illustrating the positive impact of being and feeling listened to and believed, as well as the importance of multidisciplinary care even in the absence of established curative treatments. Overall, by focusing on lived experiences and patient narratives, this study met the objectives of putting patient stories and needs at the centre of the research.

8.1.4 Phase Four

The final study of the research programme, Study Four, utilized focus groups to explore the perspectives and healthcare-related needs of patients. Adopting a patient-centered approach, participants identified key areas for improvement and collaboratively proposed recommendations to enhance healthcare experiences and journeys for this patient population. PPI Contributors were involved in collaborative analysis and interpretation of focus group data, ensuring that final outcomes were closely representative of patient experiences and needs.

Findings revealed that the healthcare experiences of patients with chronic and poorly understood conditions are largely unsatisfactory, with recommendations most often

targeting consultation dynamics and service structure. Together, the results highlighted a fragmented healthcare system, characterised by poor coordination and communication breakdowns, which fails to provide comprehensive multidisciplinary care for individuals with complex needs. With regards to consultation dynamics, a need for meaningful dialogues with healthcare practitioners as well as transparency about and involvement in decisions surrounding care were identified. Participants highlighted a need for better guidance regarding their conditions and care from healthcare providers and suggested that, in the absence of specialised clinics in Ireland, formal links between healthcare services and patient advocacy groups should be established. While clinician training regarding specific conditions was important, participants stressed that initiatives should target soft skills of clinicians, such as their empathy, communication, and delivery of compassionate care. Improvements in healthcare culture were seen as important to the patient experience, reflected in suggestions made regarding improving clinician wellbeing and targeting burnout, for example by addressing staff shortages. While systemic issues were emphasised, specific recommendations for improving coordination, communication and care quality varied. Generally, a need for holistic and personalized approaches to care and the development of multidisciplinary clinics, networks and specialised diagnostic facilities were emphasised. Communication breakdowns could be mitigated by the introduction of electronic patient records as opposed to the paper-based system with which the services operate with at present. The need for patient partnership in healthcare-related research was emphasised as a way of tailoring healthcare to the specific needs of patients.

Figure 8.1

Summary of research findings

Research Phase 1: Study One			
Objectives	To aggregate findings of qualitative research regarding (i) The experiences of individuals living with MUS, FM and ME/CFS (PwC) (ii) The experiences of healthcare providers' (HCP) working with patients with MUS, FM and ME/CFS		
Key Findings	*PwC themes*: Experiences of illness and its impact on domains of life; experiences of the patient journey *HCP themes*: Experiences of making sense of symptoms and managing the consultation *Shared theme*: Challenge of uncertainty and sensemaking at the limits of biomedical knowledge		
Implications and Future Research	• The patient-clinician relationship as a target for improvement • Research with multiple stakeholders to identify challenges and co-design solutions • Improved reporting practices in qualitative research		
Strengths	Volume of literature reviewed Perspectives of PwC and HCP	**Limitations**	Limitations of aggregative approach, Inclusion criteria: ascertaining primary vs. secondary pain and fatigue
Research Phase 2: Study Two			
Objectives	(i) Explore perspectives of adults familiar with FM and ME/CFS in Ireland, including their beliefs, attitudes and understanding of the conditions (ii) Explore appraisals of public and HCP knowledge and awareness of FM and ME/CFS; (iii) Explore perceived challenges faced by people living with FM and ME/CFS in Ireland		
Key Findings	Good level of understanding of FM and ME/CFS and the challenges faced by individuals living with these conditions. Core challenges identified include the experience of symptoms and their impact, experiences of stigma, disbelief and invalidation in social and healthcare settings, and challenges related to the diagnosis and management of FM and ME/CFS. Appraisals of public and clinician knowledge and awareness of these conditions are very poor. Respondents are highly critical of healthcare for this group of patients.		
Implications and Future Research	• A lack of confidence in healthcare among adults in Ireland points to a need to explore patients' perspectives and experiences in more depth. • Focused research should investigate clinicians' practices, their attitudes, beliefs and knowledge about the aetiology, diagnosis and management of ME/CFS and FM.		
Strengths	Exploratory survey in Irish context, Co-designed with PPI Contributors	**Limitations**	Selection bias, non-representative sample Lack of standardised measures (e.g., health literacy)
Research Phase 3: Study Three			
Objectives	(i) Provide an in-depth account of the lived experiences of individuals with diverse poorly understood conditions in Ireland, including their experiences of healthcare; (ii) Explore healthcare journeys through journey mapping (iii) Explore participant experiences of Life Grid methodology		
Key Findings	*Phenomenological findings*: Transformative experience of chronic illness: life, sense of self, body, relationships; *Healthcare experiences and Journey maps*: invalidation, stigma, navigating a broken system, power dynamics, disappointment with healthcare *Life Grid experiences:* A memory aide, a transformative experience, a challenging but useful method		
Implications and Future Research	• A need for patient-centered approaches in both research and medical practice • Further journey mapping research to identify gaps in care and to inform care pathways • Integrating Life Grid content and interview data		
Strengths	Involvement of PPI Contributors in design and piloting Methodological rigor and analytic pluralism Narrative approach	**Limitations**	Retrospective journey data and rudimentary mapping Lack of standardised measures (e.g., satisfaction with healthcare, impact of illness)
Research Phase 4: Study Four			
Objectives	(i) Explore the healthcare experiences and needs of patients living with diverse poorly understood chronic conditions in Ireland (ii) Identify targets for change and propose recommendations for improving healthcare (iii) Involve PPI Contributors in data analysis and interpretation		
Key Findings	Patients express a high level of dissatisfaction with healthcare. *Key themes:* Burden of chronic illness, negative experiences with clinicians, negative experiences of the patient journey. *Key barriers to care:* Stereotypes and Assumptions, Lack of continuity of care, Poor coordination of care *Recommendations/Targets:* Consultation dynamics and processes (patient-clinician relationship, decision making) and System Integration (comprehensive and coordinated care)		
Implications and Future Research	Future research should seek input from wider range of stakeholders. Validation workshops to refine findings and actionable recommendations		
Strengths	Involvement of PPI Contributors in analysis Patient-centered approach (research and outcomes) Methodological rigor and analytic pluralism	**Limitations**	Limited generalisability/ specificity of recommendations Stakeholder involvement

8.2 Key Insights

Each study in this research programme offered valuable insights into the experiences and needs of patients, providing multiple layers of evidence highlighting a need to address critical gaps in healthcare. Table 8.1 provides a summary of the key themes identified across the studies, demonstrating that, despite their diverse focus areas, ranging from public perceptions to lived experiences and system improvements, several recurring themes emerged consistently. The profound and multifaceted impact of chronic illness on individuals' lifeworlds was evidenced across all studies, with repeated findings of delegitimation and stigma in social and healthcare contexts. Key insights regarding healthcare experiences as discussed below, with a focus on what patients value in and expect from healthcare, along with implications of the research on service design and delivery.

Table 8.1

Overview of key themes

Finding	Study 1	Study 2	Study 3	Study 4
Experience of symptoms and their impact	XX	XX	XX	XX
Existential themes (Loss, Identity, Embodiment)	X	X	XX	X
Life narrative (Past, Present, Future)	X	X	XX	X
Sensemaking	XX	X	XX	X
Knowledge/Understanding	X	XX	XX	XX
Stigma	XX	XX	XX	XX
Stereotypes (Age, gender, appearance)	XX	XX	XX	XX
Disbelief	XX	XX	XX	XX
Not being listened to/heard	X	X	XX	XX
Evidence, legitimacy, medical uncertainty	XX	X	XX	X
Research needs		X	X	X
Diagnostic journey/ challenges	XX	XX	XX	X
Treatment/management challenges	X	X	X	X
Consultation/ Patient-clinician relationship	XX	X	XX	XX
Negative experiences of healthcare	X	X	XX	XX
Positive experiences of healthcare	X		X	X
Burden/ negative impact of healthcare		X	XX	XX
Delays (diagnosis, referral)	X	X	XX	X
Navigating the system		X	XX	X
Navigating power dynamics	X		XX	X
Distrust of healthcare		X	XX	X
Role of mental health professionals			X	X
Need for guidance and information			XX	XX
Empathy/Compassion		X	XX	XX
Support needs and experiences	X	X	X	X
Patient communities	X	X	X	XX
Continuity of care			XX	XX
Coordinated care			XX	XX
Specialist service needs	X	X	X	X
Comprehensive/Multidisciplinary care needs			XX	XX
System fragmentation			XX	XX
Patient-centered care	X	X	XX	XX

Note. X Denotes theme/finding identified; XX Denotes a strong theme/finding. Theme strength derived reflexively.

8.2.1 Person-Centered Care

Patient-clinician dynamics were highlighted as one of the key challenges and targets for change with regards to improving the healthcare experiences of patients with contested or poorly understood chronic conditions. Expectations about what the dynamic should look like were not unreasonable, however, but mirrored the central tenets of person-centered care (e.g., Grover et al., 2022; Mudiyanse, 2016; Stewart et al., 2024). Specifically, patients across all studies emphasised a need to be listened to and believed; for their concerns to be taken seriously, discussed and appropriately investigated. At the core of this was a desire to be seen and recognised, treated with compassion and respect, and involved in decisions about their care.

Patients also emphasised a desire for transparency, particularly in relation to the clinicians' limits of knowledge and understanding. They discussed positive experiences with certain healthcare providers who listened and demonstrated openness to and a willingness to learn about their conditions. These findings are not surprising, as the broad literature highlights that transparency fosters trust (Montgomery et al., 2020) and supports patient autonomy by ensuring patients feel engaged in their care (Robins et al., 2011).

Essentially, patients expect healthcare professionals to follow the principles of the Hippocratic Oath (Lasagna, 1964), with an emphasis on humanistic approaches to care:

> *"I will remember that there is art to medicine as well as science, and that warmth, sympathy, and understanding may outweigh the surgeon's knife or the chemist's drug.*
>
> *I will not be ashamed to say "I know not," nor will I fail to call in my colleagues when the skills of another are needed for a patient's recovery. […]*
>
> *I will remember that I do not treat a fever chart, a cancerous growth, but a sick human being, whose illness may affect the person's family and economic stability. My responsibility includes these related problems, if I am to care adequately for the sick."*

8.2.2 Guidance and Support

The importance of access to guidance, informational and practical supports throughout the patient journey was highlighted in studies three and four. Patients expressed a need for clear, accessible information about their diagnosis, treatment options and possible avenues for investigation, and noted that such information should be made available by the clinician, thereby reducing the burden on the patient to "figure things out" on their own. Patients also suggested that stronger formal links should be made between primary care practices and established patient charities or advocacy groups, particularly given the lack of specialist services available and range of psychosocial and practical supports offered by patient communities. Such suggestions have been made by patients elsewhere, for example in the context of rare disease pathways in the UK (Walton et al., 2022), while the literature suggests that online patient communities can play an important role in chronic disease management through informational support (Sharma & Khadka, 2019). However, patients' accounts highlighted that communities and advocacy groups should not be seen as a substitute for healthcare services. Participants often spoke of a certain injustice of having to carry the burden of advocacy on top of their chronic illness, highlighting that these efforts were largely a necessity due to systemic failures and inadequate knowledge about their conditions. The research also suggested that mental health professionals can play an important role in the care of individuals with poorly understood conditions, though this should be carefully considered given the harmful narratives surrounding psychogenic aetiology. Patients' accounts indicated that taking a treatment role, counsellors and psychologists can have a positive impact by supporting patients as they navigate the personal and healthcare journeys with chronic illness.

8.2.3 Coordinated Multi-disciplinary Care

Key insights from the research pertained to a critical need for coordinated, multi-disciplinary and comprehensive care for patients with poorly understood chronic illnesses. Such improvements ultimately necessitate structural changes within the healthcare system, including greater investment in resources and infrastructure and the design of clinical pathways. Patients emphasised a need for improved continuity of care, both within primary care setting and specialist services. Related to this, they expressed a

need for consistent follow up, noting that they often feel abandoned after initial consultations with little support or guidance on subsequent steps in their journey. With regards to clinical pathways, for instance, patient contributors recommended the development of guidelines for patients which fall into, as was suggested, the "I don't know" category. The pathway could involve a referral from the GP to a multi-disciplinary team sharing knowledge to establish a diagnosis or pathways of investigation, rather than requiring the patient to progress from specialist to specialist sequentially. The introduction of electronic patient records or other digital solution was seen as crucial to improving coordination and continuity of care, reducing duplication of procedures and information loss, while at the same time reducing the administrative burden placed on patients. Similar findings have been noted previously in the context or rare disease in Ireland (Byrne et al., 2020; Ward et al., 2022), suggesting that larger-scale changes are needed to improve care for those with under-recognised, chronic and multisystemic conditions.

8.3 Implications

Taken together, the findings of this research reflect that at present, the healthcare system is still misaligned with principles of patient-centered care and provide strong support for the implementation of programmes articulated in *Sláintecare* (Department of Health, 2021). The repeated identification of disjoined services as a pervasive barrier to care among individuals with poorly understood chronic conditions underscores the need for system integration. This has been recognised previously (Connolly, 2024; Darker et al., 2015) and is central in *The National Framework for the Prevention and Management of Chronic Disease* (HSE, 2021) which states that *"The full spectrum of care services are required and need to be provided in an integrated way through agreed clinical pathways between community, ambulatory care hubs and hospitals"* (p. 22). The framework also highlights that information must be shared across services in a timely and accurate manner for true integration to occur and includes education and training for staff to support patients in managing their conditions as a component of implementation.

The healthcare reform policies highlight that the design and implementation of well-structured clinical pathways is needed to address issues of care coordination, thereby ensuring that patients receive continuous, personalised care. However, care or clinical pathways are evidence-based, multi-disciplinary care plans that describe, chronologically, the relevant diagnostic and management steps in the care of patients (Lawal et al., 2016; Seckler et al., 2020), making their establishment problematic in the case of poorly understood conditions. As illustrated, these conditions are associated with prognostic and diagnostic uncertainty, making clinical management a challenge. Additionally, Grocott (2019) points out an "uncomfortable truth" that clinical pathway designs are often structured to the convenience of healthcare providers rather than patients. While feedback on current care processes and interventions are a key aspect of pathways design, patients are seldom involved in the process (Cassidy et al., 2024; Wind et al., 2022).

This brings to the fore the second major implication of this thesis, specifically, the emphasis on meaningful and sustained engagement of patient and public contributors in both research and healthcare design. As previously discussed, the practice of PPI is widely recognised as a means to enhance the quality and relevance of research, to democratize the process and improve the quality of healthcare (e.g., Brett et al., 2014b; Edgman-Levitan & Schoenbaum, 2021; Frith, 2023). Several authors have discussed and development of specific PPI Frameworks (Armstrong et al., 2017; Greenhalgh et al., 2019) and patient-centered engagement practices (Sheridan et al., 2017), while guidance provided by national initiatives like PPI Ignite can be helpful in terms of planning the process of PPI, including that of financial compensation for contributors. However, PPI should not be driven by institutional mandates or stakeholder involvement for political correctness, but rather a genuine dedication to engaging patients and the public as equal partners. This requires courage to challenge traditional hierarchies and power dynamics, and a willingness to let go of standard ways of doing research, instead striving for epistemic justice (Liabo et al., 2022; Pearce, 2021).

The present research demonstrates how PPI can, and should, move beyond a consultative role, going beyond asking patients about their experiences and towards involving them in the research process and design of recommendations. This type of

research is particularly timely in the Irish context given the current transformation of healthcare. As discussed in Chapter 1 (*Section 1.4.1*), the HSE has recently made significant strides to embed PPI in their organisational reform processes, publishing the *Patient and Service User Partnership Plan* (HSE, 2024) which outlines how a Patient and Service User Partnership Council will contribute to national and regional frameworks. This is an assuring step forward in line with international trends, aiming to recognise service users as true partners in decisions about care, service design, research, and governance. While this development holds great promise, it also raises questions of who the partners will be and how representative the Council will be of the needs of "lay" patients, or those individuals who might not be aware of PPI or be privileged to take part. It will also be interesting to observe the level of contributor involvement in, for example, the analyses of outcome data. Ultimately, transparency about the engagement processes involved will be crucial and, as demonstrated in Chapter 7, can offer insights about what works in practice, for whom, and how it can be improved to align with patient-centred ethos.

8.4 Strengths and Limitations

The strengths and limitations of each individual study are discussed in the relevant Chapters and are outlined in Figure 8.1 above.

As emphasised, the research programme embraced the collaborative ethos of PPI, capturing the voices of patients both as participants and co-creators, identifying targets for change in healthcare. By engaging patient stakeholders, the research ensured that the perspectives and needs of individuals most affected by its outcomes were central. However, several improvements could be made. For example, involving a more diverse group of stakeholders, including healthcare professionals and policymakers, would have enriched the study's outcomes by providing another layer of nuance, enriching the solutions and providing alternative perspectives. Further, while PPI Contributors were involved in the conceptualisation of research, co-design, analysis and interpretation, engagement at the data collection and finding dissemination stages were limited. The scope of poorly understood conditions was both a strength and limitation in this research. By including a wide range of conditions typically labelled "medically unexplained", the research illustrated common patterns in patients' healthcare

experiences, therefore identifying higher-level targets for change. A focus on the needs of patients with specific conditions, such as EDS, POTS, FM or ME could inform targeted interventions instead. However, the high level of comorbidity and symptom overlap between various unexplained conditions, as well as challenges with regards to ascertaining a clear diagnosis, would likely impact the homogeneity of the sample despite this. This research makes a significant contribution by bridging the nuanced lived experiences of individuals with chronic and poorly understood conditions and transforming these insights into actionable, impactful healthcare recommendations.

The plurality of analytic approaches, methodological rigor and transparency were a key strength of this research programme. As discussed in Chapter 2 (*Section 2.*1), the research was underpinned by phenomenological philosophy and aligned with participatory action research principles, which was its particular strength. Taking this approach, it not only shed light on existential dimensions and lifeworlds of individuals with poorly understood conditions, but also facilitated practical engagement and collaborative problem solving, bringing the work back to the "real world". The researcher' positionality and philosophical orientation were defined, with reflexivity maintained throughout. Further, the use of novel methods to data collection and interrogation, for example in the case of Life Grid methodology, enriched the results and provided an opportunity to explore participant reflections on research methods, thereby informing future designs.

The use of mixed-methods was a key strength in that it allowed for a multifaceted exploration of the research domain. However, the research programmed leaned primarily on qualitative approaches and could be further complemented by quantitative data and standardized measures, for example in relation to quality of life or health literacy. While the qualitative exploration identified numerous challenges and barriers to care, it is difficult to establish which contributes most to patient dissatisfaction with healthcare. Thus, specific and primary targets for improvement are difficult to ascertain beyond broad recommendations.

8.5 Future Research

The findings of this thesis point to several avenues for further investigation. Fundamental research is needed to determine the prevalence rates of various poorly understood conditions in Ireland, offering crucial insights into the scale and scope of the issue. Studies that provide a more detailed mapping of patient journeys, including healthcare pathways and associated costs, would be invaluable and could inform strategies to optimise resource allocation. Such studies should seek to combine data on patient flow along with their experience of the journey. Given that patient-clinician dynamics emerged as a significant challenge throughout the research, future work should examine ways to improve interactions and care quality, particularly in the context of uncertainty and complex needs. As stigma and disbelief were a persistent theme throughout, more work needs to be done to target public and clinician attitudes towards this heterogenous patient group. Research should seek to engage multiple stakeholders, both in a contributor and participant capacity, but will need to address issues of power imbalance. While the present research focused on the experiential and systemic dimensions, biomedical research is needed to progress the diagnosis and treatment of these conditions, so that they are no longer "poorly understood" but widely recognised as legitimate health concerns.

8.6 Concluding Remarks

This thesis employed multi-phase design with Patient and Public Involvement embedded throughout. Through a systematic literature review, a cross-sectional mixed-method survey, in-depth interviews, and focus groups, this research programme provided a holistic exploration of the lived experiences of individuals with chronic and poorly understood conditions in Ireland, attending to personal, social and healthcare contexts. Patients' healthcare-related experiences and needs were elucidated, and insights transformed into impactful recommendations for change. This research is especially timely in the context of evolving PPI practices and system transformation in Ireland. It highlights the importance of stakeholder engagement and collaboration, elevates the voices of individuals frequently overlooked in healthcare, and embodies patient-centered research practices.

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Appendices

Appendix A. Study One

Appendix A1. Sample Search Strategy

Table A1.

Sample Search Strategy (PsycInfo)

	Line	Limiters/Expanders
1	(DE "Fibromyalgia" OR "fibromyalgia syndrome" OR "primary fibromyalgia" OR "primary chronic pain" OR "persistent pain disorder" OR "widespread chronic pain" OR "persistent pain" OR DE "Somatoform Pain Disorder" OR "psychogenic pain" OR "pain disorder")	Limiters - Published Date: 20010301-20210331; Peer Reviewed; Publication Type: Peer Reviewed Journal, Dissertation Abstract; English; Age Groups: Adulthood (18 yrs & older); Document Type: Dissertation, Journal Article
2	(DE "Chronic Fatigue Syndrome" OR "chronic fatigue" OR "idiopathic fatigue" OR "myalgic encephalomyelitis" OR CFS OR ME OR CFS/ME OR ME/CFS OR "chronic fatigue disorder" OR DE "Encephalomyelitis")	Expanders - Apply equivalent subjects Search modes - Boolean/Phrase
3	("medically unexplained symptom*" OR "medically unexplained physical symptom*" OR "medically unexplained syndrome*" OR "medically unexplained illness" OR MUS OR MUPS OR MUI OR DE "Somatization" OR DE "Somatization Disorder" OR DE "Somatoform Disorders" OR "somatic symptom*" OR "psychosomatic" OR "psychosomatic symptom" OR "functional disorder")	
4	(Experience* OR DE "Life Experiences" OR impress* OR thought* OR think* OR subjectiv* OR perspective* OR belief* OR believ* OR attitude* OR DE "Attitudes")	
5	(DE "Qualitative Methods" OR qualitativ* OR DE "Focus Group" OR "DE "Phenomenology" OR DE "Grounded Theory" OR DE "Interpretative Phenomenological Analysis" OR DE "Narrative Analysis" OR narrat* OR "Content analysis" OR "Discourse Analysis" OR DE "Semi-Structured Interview" OR interview* OR ethnograph* OR DE "Action Research" OR DE "Thematic Analysis")	
6	1 OR 2 OR 3	
7	4 AND 5	
8	6 AND 7	

Appendix A2. Reviewed Studies

Table A2

Study number and corresponding reference

	Study Number and Full Reference
1	Aamland, A., Fosse, A., Ree, E., Abildsnes, E., & Malterud, K. (2017). Helpful strategies for GPS seeing patients with medically unexplained physical symptoms: A focus group study. *British Journal of General Practice*, *67*(661), e572–e579.
2	Aamland, A., Werner, E. L., & Malterud, K. (2013). Sickness absence, marginality, and medically unexplained physical symptoms: A focus-group study of patients' experiences. *Scandinavian Journal of Primary Health Care*, *31*(2), 95–100.
3	Abokdeer, S. A. M. (2019). *Patients' and Practitioners' Views and Experiences of Chronic Widespread Pain (Fibromyalgia) and Its Management in the UK and Libya* [University of Northumbria at Newcastle (United Kingdom)]. https://nrl.northumbria.ac.uk/id/eprint/40268/
4	Acker, S. M. (2008). *The doctor -patient relationship in the treatment of fibromyalgia*. Massachusetts School of Professional Psychology.
5	Anderson, V. R., Jason, L. A., & Hlavaty, L. E. (2014). A Qualitative Natural History Study of ME/CFS in the Community. *Health Care for Women International*, *35*(1), 3–26.
6	Armentor, J. L. (2017). Living with a Contested, Stigmatized Illness: Experiences of Managing Relationships among Women with Fibromyalgia. *Qualitative Health Research*, *27*(4), 462–473.
7	Arnold, L. M., Crofford, L. J., Mease, P. J., Burgess, S. M., Palmer, S. C., Abetz, L., & Martin, S. A. (2008). Patient perspectives on the impact of fibromyalgia. *Patient Education and Counseling*, *73*(1), 114–120.
8	Arroll, M. A., & Howard, A. (2013). "The letting go, the building up, [and] the gradual process of rebuilding": Identity change and post-traumatic growth in myalgic encephalomyelitis/chronic fatigue syndrome. *Psychology and Health*, *28*(3), 302–318.
9	Arroll, M. A., & Senior, V. (2008). Individuals' experience of chronic fatigue syndrome/myalgic encephalomyelitis: An interpretative phenomenological analysis. *Psychology and Health*, *23*(4), 443–458.
10	Åsbring, P. (2001). Chronic illness—A disruption in life: Identity-transformation among women with chronic fatigue syndrome and fibromyalgia. *Journal of Advanced Nursing (Wiley-Blackwell)*, *34*(3), 312–319.
11	Åsbring, P., & Närvänen, A. (2002). Women's experiences of stigma in relation to chronic fatigue syndrome and fibromyalgia. *Qualitative Health Research*, *12*(2), 148–160.
12	Åsbring, P., & Närvänen, A. (2003). Ideal versus reality: Physicians perspectives on patients with chronic fatigue syndrome (CFS) and fibromyalgia. *Social Science and Medicine*, *57*(4), 711–720.
13	Åsbring, P., & Närvänen, A. (2004). Patient power and control: A study of women with uncertain illness trajectories. *Qualitative Health Research*, *14*(2), 226–240.
14	Ashe, S. C., Furness, P. J., Taylor, S. J., Haywood-Small, S., & Lawson, K. (2017). A qualitative exploration of the experiences of living with and being treated for fibromyalgia. *Health Psychology Open*, *4*(2).

	Study Number and Full Reference
15	Barcroft, R. (2016). *Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis and Fibromyalgia :A Social Model of Disability Perspective* [Lancaster University (United Kingdom)]. https://doi.org/10.17635/lancaster/thesis/50
16	Bee, P., McBeth, J., MacFarlane, G. J., & Lovell, K. (2016). Managing chronic widespread pain in primary care: A qualitative study of patient perspectives and implications for treatment delivery. *BMC Musculoskeletal Disorders*, *17*, 1–11.
17	Boulton, T. (2019). Nothing and Everything: Fibromyalgia as a Diagnosis of Exclusion and Inclusion. *Qualitative Health Research*, *29*(6), 809–819.
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138	Whitehead, L. C. (2006). Quest, chaos and restitution: Living with chronic fatigue syndrome/myalgic encephalomyelitis. *Social Science and Medicine*, *62*(9), 2236–2245.
139	Wileman, L., May, C., & Chew-Graham, C. A. (2002). Medically unexplained symptoms and the problem of power in the primary care consultation: A qualitative study. *Family Practice*, *19*(2), 178–182. https://doi.org/10.1093/fampra/19.2.178
140	Wilson, L., Whitehead, L., & Burrell, B. (2011). Learning to live well with chronic fatigue: The personal perspective. *Journal of Advanced Nursing*, *67*(10), 2161–2169.
141	Woivalin, T., Krantz, G., Mäntyranta, T., & Ringsberg, K. C. (2004). Medically unexplained symptoms: Perceptions of physicians in primary health care. *Family Practice*, *21*(2), 199–203. https://doi.org/10.1093/fampra/cmh217
142	Wuytack, F., & Miller, P. (2011). The lived experience of fibromyalgia in female patients, a phenomenological study. *Chiropractic and Manual Therapies*, *19*.
143	Yon, K., Nettleton, S., Walters, K., Lamahewa, K., & Buszewicz, M. (2015). Junior doctors' experiences of managing patients with medically unexplained symptoms: A qualitative study. *BMJ Open*, *5*(12).

Appendix A3. Characteristics of Included Studies

Table A3

Characteristics of Studies (N=143) included in Systematic Literature Review

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Aamland et al. (2017)(1)	Qualitative, Purposive sample, Focus groups, Thematic cross-case analysis, Systematic text condensation	GPs' reflections on action and the strategies they had experienced as helpful when treating patients with MUPS.	Participants: 24 (M= 17, F= 7) HCP, Age: 33-68, Norway (Healthcare education)	GPs use tangible strategies in consultations with patients with MUPS. Important strategies include a thorough review of the patient's story, along with mutual interpretation and negotiation of symptoms.
Aamland et al. (2013)(2)	Qualitative, Purposive sample, Focus groups, Systematic text condensation	To explore experiences of sickness absence in MUS patients, focusing especially on marginalization.	Participants: 12 (M= 6, F= 6) PWC, Age: 24-59, Norway (Healthcare, rural)	Invisible symptoms and lack of objective findings are perceived as an additional burden to sickness absence.
Abokdeer (2019)[a] (3)	Qualitative (Study phase 1), Purposive sample, Semi-structured interviews, Framework analysis	To explore the experience of service users in the process of obtaining a diagnosis, the emotional experiences of sufferers, experiences of support received, coping and managing daily activities, interventions and experiences of living with pain.	Participants: 12 (F= 12) PWC, Age: 27-60, UK (Healthcare)	Participants faced difficulties regarding a perceived lack of understanding by healthcare professionals and the general public of the management of FMS.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Acker (2008)[a](4)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis; Grounded Theory	To examine the experience that patients diagnosed with fibromyalgia have in relation to their physicians.	Participants: 10 (F= 10) PWC, Age: 33-60 (*M*=44), USA (Healthcare)	The majority of the patient's experiences that were relayed were negative and even traumatizing.
Anderson et al. (2014)(5)	Qualitative, Purposive sample derived from an epidemiological study, In-depth interviews, Constant comparative method; Grounded Theory	To examine the natural history of people with ME/CFS and the events occurring over time that may contribute to the course and progression of the illness	Participants: 19 (M= 3, F= 16) PWC, Age: *M*=51 (*SD*=11), USA (Community)	The illness experience and the physical construction of ME/CFS are key themes in the natural histories of people with ME/CFS. Identifying the community response to ME/CFS is a critical part of understanding the systems involved with the people experiencing the illness as well as how people think about or work with this illness.
Armentor (2017)(6)	Qualitative descriptive, Convenience snowball sample (women), In-depth interviews, symbolic interactionism and ethnomethodology, Modified Grounded Theory approach	To explore how individuals with FM manage their social roles and negotiate their relationships with others including family members, friends, coworkers, and medical professionals.	Participants: 20 (F= 20) PWC, Age: 32-80 (*M*=49), USA (Community)	Participants describe their illness experience through direct and educational approaches. An unspoken awareness of the illness effects was present in relationships with close family and friends, and support was offered. However, scepticism and lack of comprehension often led to social withdrawal among participants.
Arnold et al. (2008)(7)	Qualitative, Purposive sample, Focus groups, Grounded Theory	To identify the key symptom and functional domains associated with fibromyalgia from the patients' view and to elicit their perspectives on the impact of fibromyalgia	Participants: 48 (F= 48) PWC, Age: 31-72 (*M*=51), USA (Community and university-based services)	From the patients' perspective, fibromyalgia is more than a painful condition. It is associated with multiple symptom domains and has a substantially negative impact on function and quality of life. A comprehensive assessment of the multiple symptom domains and the impact of fibromyalgia on multidimensional

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
				aspects of function should be a routine part of the care of fibromyalgia patients and in trials of treatment.
Arroll et al. (2013)(8)	Qualitative phenomenological, Purposive and Snowball sampling, Semi-structured interviews, IPA	The phenomenon of identity change and subsequent post-traumatic growth (PTG) in individuals with myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS).	Participants: 10 (M= 3, F= 7) PWC, Age: *M*=39.6, UK (Community, Support groups)	Participants were required to redefine their sense of self due to the physical limitations ME/CFS placed upon them and their personal and social activities. The interviewees also compared their former and present selves and had to confront the reality of their 'new selves'. Some, but not all, experienced positive and/or transformational growth.
Arroll et al. (2008)(9)	Qualitative phenomenological, Purposive sample, Semi-structured interviews, IPA	The process by which individuals conceptualise their bodily signs and sensations as consistent with the label CFS/ME	Participants: 8 (M= 2, F= 6) PWC, Age: 35-67 (*M*=55.5, *SD*=9.4), UK (Community/Support groups)	Symptomatology and illness course, interference with daily and working life, frequency of symptoms, external information, diagnosis and treatment each played a part in the recognition of individuals' symptoms as CFS/ME.
Åsbring (2001)(10)	Qualitative, Purposive and discriminate sample (women), Semi-structured interviews, Constant comparative method, Grounded Theory	To show that CFS and FM often comprise a biographical disruption in the women's life that has profound implications for their identity in relation to various activities, especially with regard to work and social life. To show that women come to terms with a newly arisen identity to different degrees, and that some	Participants: 25 Female PWC (12 with CFS, 13 with FM) , Age: 32-65 (*M*=46), Sweden (Healthcare)	CFS and Fibromyalgia often seem to entail a radical disruption in women's biography that leads to consequences for identity, in particular in relation to work and social life. However, identity transformations appear to be partial. The biographical disruption and illness experience may be seen as a paradox that consists of two parts: suffering/losses and illness gains.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		also define their illness experience in positive terms.		
Åsbring et al. (2003)(11)	Qualitative, Purposive sample, Semi-structured interviews, Constant comparative method, Grounded Theory	To explore how physicians in a Swedish sample describe and categorise patients with CFS and fibromyalgia; what the character of CFS and fibromyalgia mean to the physicians in encounters with patients; and which strategies physicians describe that they use in the encounter with these patients.	Participants: 26 HCP (M= 14, F= 12), Age: 41-67 (*M*=50), Sweden (Healthcare)	The physicians' professional role encompasses both ideal notions and everyday realities and difficulties can arise in living up to the ideal image in the interaction with patients who have CFS and fibromyalgia. The strategies to manage the situation of patients with CFS and fibromyalgia are partly patient-focused and partly physician-focused.
Åsbring et al. (2004)(12)	Qualitative, Purposive sample (women), Semi-structured interviews, Constant comparative method, Grounded Theory	To describe how women with CFS and FM define their potential for gaining control over their situation during the health care process and of influencing the latter in their encounters with health care providers.	Participants: 25 Female PWC (12 with CFS, 13 with FM), Age: 32-65 (*M*=46), Sweden (Healthcare)	Patients perceive knowledge to be an important way of attaining control, to be able to influence health care providers as well as to increase the possibility of using power in the interaction with the health care provider. Patients believe that they can influence their health care providers by employing various strategies, that is, they perceive themselves to have a certain amount of power and control in relation to the health care providers
Åsbring et al. (2002)(13)	Qualitative, Purposive sample (women), Semi-Structured interviews, Constant comparative method, Grounded Theory	To investigate whether women with CFS and FM experience the illnesses as stigmatizing and to	Participants: 25 Female PWC (12 with CFS, 13 with FM), Age: 32-65 (*M*=46), Sweden (Healthcare)	Stigma arose primarily as a consequence of the uncertainty connected with the illnesses of CFS and fibromyalgia. Stigmatization was greater before the

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		examine the strategies they use to avoid enacted stigma		illnesses were confirmed through diagnosis.
Ashe et al. (2017)(14)	Qualitative, Purposive sample, Semi-structured interviews, IPA	To explore the life and treatment experience of people in the United Kingdom with fibromyalgia	Participants: 14 (M= 2, F= 12) PWC, Age: 29-58, UK (Online community)	FMS is immensely disruptive to individuals' lives. There are challenges in accessing treatments and many treatments are inadequate. Pain and fatigue often dominate individuals' lives. Pain is often misunderstood by others, largely due to its permanence.
Barcroft (2016)[a](15)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis	To explore the experiences of people with CFS/ME from a social model of disability perspective in order to gain a deeper insight into some of the difficulties and discrimination faced	Participants: 12 (M= 4, F= 8) PWC, Age: 23-68, UK (Community)	Participants described the discrimination and stigma that they had encountered from many areas of society as being as disabling as the condition itself. Recommendations for practice and ideas for future research are proposed.
Bee et al. (2016)(16)	Qualitative, Purposive sample nested in RCT, Semi-structured interviews; Constant comparative method, Framework approach	To explore illness and treatment experiences in participants with chronic widespread pain with a view to understanding their potential influences on intervention acceptability	Participants: 44 (F= 75%) PWC, Age: M=58 SD=13, UK (Healthcare)	The acceptability of clinically recommended, evidence-based treatments for chronic widespread pain is influenced by a complex interplay of illness, social and self-identities.
Boulton (2019)(17)	Qualitative, Purposive and snowball sampling, In-depth interviews, Narrative analysis	To examine the experiences of the diagnostic process among individuals with FM and	Participants: 31 (M= 6, F= 25) PWC, Age: 21-69 *(M*=43), Canada and UK (Community)	FM is a diagnostic category based on the absence of disease, but, at the same time, the FM label includes an almost exhaustive list of possible symptoms. FM diagnosis is largely an empty promise because it fails to

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		how they feel about receiving this label		provide definitive answers or confer meaning and legitimacy to their illness experience
Briones-Vozmediano (2017) (18)	Qualitative, Purposive sample, Multimethod (policy review, semi-structured interviews), Qualitative content analysis, situational analysis	To explore the social construction of FM from the perspective of health policies, patients, and health professionals involved in their medical attention	Participants: 28 (16 patients, 12 HCP); Gender: 12 Male (3 patients, 9 HCP), 16 female (13 patients, 3 HCP); Age: Patients 24-61; HCP 35-55, Spain (National health service)	Political, professional and individual spheres have an influence on how FM is constructed on a social level: as one of the "invisible women's diseases". It is recommended to resolve the disease's lack of recognition by i) implementing specific policies for FM and ii) increasing the training and sensitization of health providers about the severity of FM and the existence of gender prejudices biasing the attention.
Briones-Vozmediano (2013)	Qualitative, Convenience sample, Semi-structured interviews, Discourse analysis(19)	To explore three aspects of the clinical management of fibromyalgia (diagnostic approach, therapeutic management and health professional-patient relationship) from two different points of view – that of the professionals and that of the patients – to identify specific areas of the healthcare process that are deficient or unsatisfactory	Participants: 21 (9 HCP, 12 patients); Gender: 9 Male (6 HCP, 3 patients), 12 female (3 HCP, 9 patients) Age: 40-55 HCP, 21-61 patients, Spain (National health service, patient associations)	Health care services should offer greater support for these patients in the form of specific resources such as fibromyalgia clinics and health professionals with increased awareness of the disease.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Briones-Vozmediano et al. (2018) (20)	Qualitative, Purposive and snowball, Semi-structured interviews, Qualitative content analysis	To explore health service providers' perceptions regarding fibromyalgia patients in Spain and to analyse possible consequences of these perceptions in terms of how health service providers construct the disease and treat their patients	Participants: 12 (M= 8, F= 4) HCP, Age: 39-55, Spain (National health service)	The uncertainty surrounding fibromyalgia together with the fact that those affected are primarily women seem to influence professional practice in terms of lack of recognition of fibromyalgia as a severe disease. The definition of fibromyalgia as a women's disorder may exacerbate gender bias at various levels, since diseases suffered primarily by women are considered to be of "low prestige" by health professionals
Briones-Vozmediano et al. (2016) (21)	Qualitative, Convenience Snowball sample (women), Semi-structured interviews, Thematic analysis	How gender shapes women´s experiences of living with FM and how it affects their private lives	Participants: 13 (F= 13) PWC, Age: 24-61, Spain (Community)	How women experience and cope with the consequences of suffering from FM in their private lives is influenced by gendered norms that constrain women to be the carer of the home and the family. Women with FM often respond by renegotiating their identities as women.
Brooks et al (2014)(22)	Qualitative, Purposive (Patient + spouse dyads), Semi-structured interviews, IPA, dyadic analysis	To explore the in-depth the beliefs and experiences of both patients and significant others in relation to chronic fatigue syndrome/myalgic encephalomyelitis	Participants: 4 (M= 2 PWC, F= 2 spouses) , Age: Mid 50s, UK (Healthcare)	Significant others play an important role in the lived experience of CFS/ME. For both patients and significant others, the wider social world and interactions with outside others may be important influences on dyadic coping in CFS/ME.
Broughton et al. (2017)(23)	Qualitative (Participatory paradigm), Purposive sample, Semi-structured interviews, Constant comparative method, Thematic analysis	To explore the experiences of CFS/ME patients who were completing programmes of treatment at three NHS	Participants: 16 (M= 2, F= 14) PWC, Age: 24-62, UK (Healthcare)	Specialist CFS/ME services played a vital role in patients' journeys towards an improved quality of life. This improvement came about through a process which included validation of patients'

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		specialist CFS/ME services in England.		experiences, acceptance of change, practical advice and support, and therapeutic outcomes
Brown et al. (2017)(24)	Qualitative, Purposive sample, Semi-structured interviews, Grounded theory approach	To explores the experiences of 16 people claiming to have recovered from ME or CFS using the concept of liminality	Participants: 16 (M= 5, F= 11) PWC, Age: unavailable, UK (Community)	Recoverees moved from the liminality of illness to a second, and less legible state of sustained liminality in recovery, described as having one foot in the ill world, one foot in the well world
Brownell et al. (2016)(25)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis	To examine and understand the experiences of clinical practitioners dealing with patients with MUPS within their clinical practice and to propose an interim practical management guide that practitioners may find useful to facilitate the clinical care of patients with MUPS	Participants: 30 HCP, Age/gender unavailable, Canada (Healthcare)	The care of patients with MUPS can be characterized in terms of a 4-part framework: (1) the challenge of diagnosis; (2) the challenge of management/treatment; (3) the importance of communication and (4) the importance of the therapeutic relationship.
Chen (2016)(26)	Qualitative, Convenience sample, Semi-structured interviews, IPA, Constructivist Grounded Theory	To examine how health management, information seeking, and information consumption and use processes are related throughout fibromyalgia	Participants: 23 (M= 1, F= 22) PWC, Age: 21-79, US (Community)	Over time, participants became clearer and more accepting of their condition. These developments were accompanied by an increased awareness of and ability to use information sources to improve their health management, as well as improved communication with physicians and other health care providers.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Chen et al. (2020)(27)	Qualitative, Convenience sample, Semi-structured interviews, IPA, Constructivist Grounded theory, constant comparative method	To explore from the patients' perspective on the factors that influence the formation of effective patient–provider relationships.	Participants: 23 (M= 1, F= 22) PWC, Age: 21-79, US (Community)	Bridging information gaps, collaborative information seeking and problem solving, and partnership medicine can be invaluable in forging successful clinical relationships.
Chew-Graham et al. (2008)(28)	Qualitative, Purposive sample drawn from RCT, Semi-structured interviews, Constant comparative method, thematic analysis	To explore how patients with CFS/ME and family physicians understand this condition and how their understanding might affect the primary care consultation.	Participants: 38 (24 patients, 14 HCP) Gender: 18 Male (11 patients, 7HCP), 20 female (13 patients, 7HCP); Patient age: 25-78, HCP age: 26-63; UK (Healthcare)	Family physicians obtain information about CFS/ME from their nonprofessional world, which they incorporate into their professional realm. Patients and physicians describe the use of the discourse of science within consultations about CFS/ME. This form of shared understanding could lead to a positive collaborative interaction
Chew-Graham et al. (2009)(29)	Qualitative, Purposive sample drawn from RCT, Semi-structured interviews, Thematic analysis	To explore Practice Nurses' understanding and beliefs about CFS/ME and its management	Participants: 29 HCP (F= 29) Age: unavailable, UK (Healthcare)	Practice Nurses have little understanding of the evidence-base for treatment of CFS/ME, particularly psychological therapies, describing management options in terms of advice giving, self-help or counselling. Practice Nurses largely welcomed the potential development of their role in this area, but identified barriers and training needs which must be addressed to enable them to feel confident managing of patients with this condition
Chew-Graham et al. (2010)(30)	Qualitative study nested in a multi-center RCT, Purposive sample, Semi-structured interviews, Thematic analysis	To explore GPs' views on their role in making the diagnosis of CFS/ME and subsequent management of patients in primary care.	Participants: 22 HCP, Age and gender unavailable, UK (Healthcare)	Until GPs feel comfortable making the diagnosis of CFS/ME and facilitating initial management, and have appropriate services to refer patients to, there will continue to be delays in confirming the diagnosis and patients presenting in

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
				primary care with fatigue may not receive appropriate care.
Cipolletta et al. (2020)(31)	Qualitative content analysis, Purposive sample, Online conversations and interviews, rounded Theory	To understand the role of online communities relating to fibromyalgia syndrome (FMS) patients' illness experiences and their attitudes towards medication	Participants: 90 PWC (14 interviewed), Gender: 1 male, 13 female, Age: 29-55 (*M*=45.5), Italy (Online community)	Online communities represent a resource that allows users to express and share their needs, especially in terms of legitimacy and recognition.
Clarke et al. (2014)(32)	Qualitative, Purposive sample, Individual and group interviews, Framework Analysis	To explore how older people living with chronic musculoskeletal pain in the community perceive their experiences of interacting with health professionals, seeking factors that might optimise these interactions	Participants: 23 (M= 7, F= 16) PWC, Age: 66-89 (*Mdn*=73), UK (Community)	In common with people of all ages, an effective partnership between an older person in pain and health professionals is essential if pain is to be reported, appropriately assessed and managed, because of the subjective nature of pain and its treatment responses.
Clarke et al. (2007)(33)	Qualitative, Convenience sample, Multimethod (interviews and diaries), Hermeneutic phenomenological approach	To examine the apparent ability/inability of patients to manage their chronic pain through an exploration of their living and lived experiences	Participants: 8 (M= 4, F= 4) PWC, Age: 36-71, UK (Healthcare)	Further research is needed in this field to understand why some patients with chronic pain are able to successfully accept pain management, whereas others relentlessly seek a cure. It is vital that the health professionals as researchers allow the patients' perspectives to emerge authentically and strive not to impose their own expectations and conceptual categories.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Cooper et al. (2017)a(34)	Qualitative, Purposive, snowball sample, Semi-structured interviews, narrative inquiry, Thematic content analysis	To explore the role of social support in addressing the various challenges presented by the complexity of fibromyalgia in this context, through a discussion of the narratives of people living with fibromyalgia	Participants: 15 (F= 15) PWC, Age: 23-59, South Africa (Community)	Sources of social support constitute important mechanisms for coping with the illness experience of fibromyalgia. Support from family, partners, and peers plays an integral role in the process of accepting fibromyalgia diagnosis, adapting to the demands of the condition, and seeking help from healthcare providers
Cooper et al. (2017)b(35)	Qualitative, Purposive, snowball sample, Semi-structured interviews, narrative inquiry, Thematic content analysis	To explore the complexity of attaining a diagnosis of fibromyalgia in SA through the narratives of people living with the condition	Participants: 15 (F= 15) PWC, Age: 23-59, South Africa (Community)	The currency of fibromyalgia as a diagnosis and the inequalities present in the South African health care system characterise the experiences of symptom recognition, diagnosis and treatment. The findings of this study confirm the existing evidence that shows fibromyalgia to be a challenging illness experience, which is attributed to the lack of clarity and legitimacy, and high contestation that surrounds the condition.
Crooks (2007)(36)	Mixed-methods, Purposive, snowball sample (women), In-depth interviews & Sickness Impact Profile, Constant comparative method; case study analysis	How women's lives change after the onset of FMS and how their changing bodies and locations in society and space shape such altered lifeworlds.	Participants: 55 (F= 55) PWC, Age: 30-88 (*M*=57.6), Canada (Community)	Changes in the women's bodies precipitated some of the most significant life changes experienced, including altered identities and diminished incomes, and that altered bodily realities facilitated or denied access to socio-spatial life. At the same time, the women's changing locations in society and space also played a role in bringing about such changes

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Crooks et al. (2008)(37)	Qualitative, Purposive, snowball sample (women), Semi-structured interviews, Constant comparative method	To elucidate the various responses women have to being, or not being, categorized as disabled within specific spheres (e.g., medical, state) or places (e.g., doctor's office, work) after developing a contested chronic illness (fibromyalgia).	Participants: 55 (F= 55) PWC, Age: 35-88 (*M*=58), Canada (Community)	Negotiating disability was, for many, an emotionally charged and complex process, drawing on one or more strategies: reluctantly employing some meanings associated with 'being disabled' to achieve material ends, creating an understanding of disability that is most in keeping with one's sense of self, embracing other meanings to the extent that they offer a legitimate basis for identity, and/or rejecting disability in the interests of sustaining an existing identity
Cudney et al. (2002)(38)	Qualitative, Purposive sample gleaned from an intervention; Thematic content analysis of online communications; constant comparative approach	To explore and gain an understanding of the common components of the personal experiences of 10 rural women as they attempt to deal with FMS.	Participants: 10 (F= 10) PWC, Age: 38-55 (*M*=49), USA (Community)	The women described themes of pain, fatigue, depression, and sleep disturbances; expressed views on the experience of rural isolation; and shared positive philosophies of dealing with this disease.
Cunningham et al. (2006)(39)	Qualitative, Purposive, snowball sample, In-depth interviews, Constant comparative analysis	To explore individuals' descriptions of life with FM, and to gain insight into aspects of the illness and its treatment that would provide some direction for future nursing practice and research.	Participants: 8 (M= 1, F= 7) PWC, Age: 30s-70s, Canada (Community)	Individuals' experiences of living with fibromyalgia are intensely challenging. Living with FM involves experiencing and managing a host of symptoms whose invisibility and lack of predictability contribute to a number of issues affecting access to and experiences of health care. Given the personal, social, and economic toll of this illness, health professionals require greater knowledge of its dynamics and of the issues faced by clients with FM

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Dennis et al. (2013)(40)	Qualitative, Purposive sample, E-mail interviews, IPA	The variation in patients' interpretations of their own symptoms.	Participants: 20 PWC, Age: 18-64, UK (Support groups)	Three key issues were discussed. There was not one overall symptom (e.g., pain) driving the unpleasantness of fibromyalgia; participants spent excessive time and energy trying to manage forces outside their control; because there is no definitive 'fibromyalgia experience', each diagnosis is unique. Issues of stigma and legitimacy need to be considered carefully by health professionals in the context of the complex and uncertain experience of patients
Devendorf et al (2020)(41)	Qualitative, Purposive sample, Semi-structured interviews, Deductive thematic analysis	To understand how patients with CFS/ME conceptualize recovery – regarding the definition and possibility of recovery.	Participants: 10 (F= 10) PWC, Age: 52-75, US (Community)	Recovery is viewed as functioning without fear of relapse, returning to previous roles and identities, and achieving a sustained absence of symptoms. Recovery is more than just symptom reduction. Outcome research should incorporate well-being measures like identity, meaning and quality of life, and personal empowerment to enhance recovery definitions.
Devendorf et al. (2019)(42)	Qualitative, Purposive sample, Semi-structured interviews, Deductive thematic analysis	To explore the views of physicians with expertise in the ME and CFS field. Particularly (1) defining recovery from ME and CFS and (2) measuring recovery from ME and CFS.	Participants: 10 (M= 8, F= 2) HCP, Age: *M*=65 (*SD*=12), USA (Healthcare)	Recovery from myalgic encephalomyelitis and chronic fatigue syndrome should be viewed as multidimensional, considering patients' daily life, psychosocial functioning, and overall physical functioning.
Dickson et al. (2007)(43)	Qualitative, Purposive sample, Semi-structured interviews, IPA	To explore the delegitimising experience of living with CFS,	Participants: 14 (M= 6, F= 8) PWC, Age: 21-68, UK (Community)	Participants perceived their GPs to be sceptical, disrespectful and to be lacking in knowledge and interpersonal skills. They

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		particularly the way in which people with CFS experience stigma and delegitimation through their interactions with a broad range of key individuals, namely their GPs, friends and partners		found delegitimising encounters with their partners more difficult to deal with. Participants viewed such delegitimation as a form of personal rejection; they were hurt by their loved ones' reactions and subsequently pondered the price of love, respect and friendship.
Dickson et al. (2008)(44)	Qualitative, Purposive sample, Semi-structured interviews, IPA	The experience of living with CFS/ME from the participants' own perspectives	Participants: 14 (M= 6, F= 8) PWC, Age: 21-68, UK (Community)	Participants reported an ongoing sense of personal loss characterised by diminishing personal control and agency. An inability to plan for the future and subsequent feelings of failure, worthlessness and insignificance ensued. Scepticism in the wider social environment only heightened the consequential identity crisis. The importance of acceptance for adjusting to a life with CFS was highlighted
Durif-Bruckert et al. (2015)(45)	Qualitative, Purposive sample, Semi-structured interviews, Qualitative content analysis	To understand the medication experience of patients with fibromyalgia and their relationship with the doctors derived from treatment negotiation	Participants: 35 (M= 3, F= 32) PWC, Age: 25-70 (M=49), France (Healthcare)	By mediating their relationship with medication, patients gain access to a state of co-expertise and they put sense into the collaboration they develop with their doctors.
Dwamena et al. (2009)(46)	Qualitative, Purposive sample, Semi-structured interviews, Grounded Theory	To describe and analyze perceptions and lived experiences of high utilising primary care patients with MUS	Participants: 19 (M= 3, F= 16) PWC, Age: 31-65 (*M*=48), USA (Healthcare)	High utilising primary care patients are a heterogeneous group with similar experiences and different perceptions, behaviours and needs. Recognizing these differences may be critical to effective treatment and reduction in utilisation.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Edwards et al. (2007)(47)	Qualitative, Convenience sample, Semi-structured interviews, IPA	To examine the subjective experiences of living with CFS/ME through the use of IPA	Participants: 8 (F= 8) PWC, Age: 37-55, UK (Community, Self-help group)	Living with CFS/ME can be an overwhelming and isolating experience. Participants acquiring social support and greater knowledge were key mediating factors in the emergence of control and acceptance.
Escudero-Carretero et al. (2010)(48)	Qualitative, Purposive sample, Focus groups, Content analysis	To understand the experiences and expectations of persons with fibromyalgia towards the health system and its professionals.	Participants: 21 (M= 1, F= 20) PWC, Age: 33-62, Spain (Healthcare)	Health care for patients with fibromyalgia should be targeted to improving their quality of life, streamlining the diagnosis, reducing anxiety, facilitating the process of care and offering support, caring and understanding from the professionals
France et al. (2008)(49)	Qualitative, Convenience sample, Triangulated phenomenological approach (Interviews, focus groups, observations), Thematic analysis	The woman's perspective on what it was like to live with fibromyalgia (FMS).	Participants: 147 (F= 147) PWC, Age: 25-73 (*M*=50), US (Community)	Individuals with FM revealed the everyday unending struggle of living with FMS with progressive disability further complicated by lack of trust and not feeling safe with the healthcare provider and system. Clinical Nurse Specialists (CNS) are crucial in educating healthcare providers on FMS, individualizing care to slow disability and promote quality of life, and influencing policy-making bodies to improve healthcare services
Gilje et al. (2008)(50)	Qualitative, Purposive sample, Multimethod (Group interview, questionnaire, follow-up); Case Study, Systematic text condensation	To explore obstructions for quality care from experiences by patients suffering from CFS.	Participants: 12 (M= 2, F= 10) PWC, Age: 22-54 (M=41), Norway (Community)	Current medical scepticism and ignorance regarding CFS shapes the context of medical care and the illness experiences of CFS patients, who may feel they neither get a proper assessment nor management
Gordon et al. (2017)(51)	Qualitative, Convenience and Purposive sample, Focus groups, Thematic analysis	To examine the opinions of primary care healthcare professionals	Participants: 101 (63 patients (54+9 carers), 38 HCP) (M= 35 (20 patients, 15HCP), F=	A number of interconnecting barriers were found, presenting difficulties to both HCPs and patients in the facilitation and

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		(HCPs) and people with chronic pain and their carers, in order to identify possible barriers to the facilitation and adoption of self-management.	66 (43 patients, 23HCP)) Both, Age: unavailable, UK (Healthcare)	implementation of self-management. If self-management is to be an important approach to chronic pain, primary care services need to be designed to address the barriers identified.
Grape et al. (2015)(52)	Qualitative, Purposive sample, Narrative interviews, Thematic narrative analysis	What it is like to stay healthy after recovering from fibromyalgia syndrome (FMS).	Participants: 8 (F= 8) PWC, Age: 34-59, Norway (Community)	The findings indicate that although women reported that life was better than before, they also reported investing considerable effort in remaining healthy. Being healthy again, they put great effort into avoiding illness through diet, exercise, and relaxation. In conclusion, remaining healthy requires ongoing hard work to maintain the body, as well as profound changes in everyday life.
Grape et al. (2017)(53)	Qualitative, Purposive sample, Narrative interviews, Thematic narrative analysis	To explore how tiredness and fatigue feature in the narratives of women who recovered from FM: before they fell ill, during their illness, during their recovery process, and after they regained their health	Participants: 8 (F= 8) PWC, Age: 34-59, Norway (Community)	The findings highlight participants' different understandings and meanings of tiredness and fatigue and the ways in which these link the past, present, and future. Significantly, a clear distinction between tiredness and fatigue was not always found. The storyline that emerges from the narratives is about balancing tiredness/fatigue with everyday life.
Gray et al. (2003)(54)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis	To explore stories of the illness experiences, occupational disruption, and adaptation of people with CFS from their own (emic) perspectives.	Participants: 5 PWC, Age: 16-44, Australia (Community)	CFS can significantly alter people's experiences of occupations, necessitating a process of adaptation People living with CFS face issues, for which occupational therapy can be of assistance.

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Guise et al. (2010)(55)	Qualitative, Convenience sample, Discourse analysis,	to explore ME/CFS sufferers' descriptions of interactions with medical professionals taken from an asynchronous, online sufferers' support group.	Participants: 38 PWC, Age: unavailable, UK (Online support group)	Much of the participants' accounts are concerned with attending to issues of illness and identities.
Gullacksen et al. (2004)(56)	Qualitative, Purposive sample, Semi-structured interviews, Inductive phenomenological analysis	To describe and identify significant aspects and themes in the course of life change because of chronic pain; and to propose a clinically useful model for that adjustment.	Participants: 18 (F= 18) PWC, Age: 23-55 (*M*=42), Sweden (Healthcare)	The investigation of adjustment focused on the importance of the individual's biographical wholeness for understanding the adjustment to chronic pain and its consequences. It also demonstrated that the women had power of their own to change and create a new satisfying life role.
Hallberg et al. (2011)(57)	Qualitative, Purposive sampling (2 subsamples), Semi-structured interviews, Constant comparative method; Grounded Theory	To gain a deeper understanding of what is the main concern in daily life for women living with fibromyalgia	Participants: 23 (F= 23) PWC, Age: M= 53.8 Sweden (Community)	The main concern for women with fibromyalgia was to reach a balance in their daily life. This concern was resolved by them using different strategies aimed at minimizing the dysfunctional interplay between activity and recovery.
Hannon et al. (2012)(58)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis, modified grounded theory	To develop an education and training intervention to support practitioners in making an early diagnosis of CFS/ME and supporting patients in the management of their symptoms.	Participants: 44 (18 HCP; 16 patients) Gender: (9 male (4 HCP, 5 patients), 25 female (14 HCP, 11 patients); Age: patients 28-61, HCP unavailable; UK (Healthcare)	Patients and carers stressed the importance of recognising CFS/ME as a legitimate condition, and the need to be believed by health care professionals. GPs and practice nurses do not always have the knowledge or skills to diagnose and manage the condition.
Harding et al. (2005)(59)	Qualitative, Purposive and theoretical sample, In-depth interviews, Framework approach	The experiences of diagnosis and management among	Participants: 15 (M= 3, F= 12) PWC, Age: 21-71 (*M*=57), UK (Healthcare)	Developing, implementing, and evaluating approaches to address patients' spoiled identities might allow us to improve

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		patients who attended a specialist musculoskeletal pain clinic and the factors influencing their interpretation of these experiences		patient-centred outcomes in chronic musculoskeletal pain.
Harsh et al. (2016)(60)	Qualitative, Purposive sample, Semi-structured interviews, Transcendental Phenomenological Method, phenomenological analysis	Medical residents' experiences of caring for patients with MUI/S and the personal and professional factors that contributed to their clinical approaches.	Participants: 10 (M= 8, F= 2) HCP, Age: unavailable, USA (Healthcare)	Residents often experience a lack of confidence in their ability to effectively treat patients with MUI/S, as well as frustration surrounding their encounters with this group of patients.
Hayes et al. (2010)(61)	Mixed-methods, Purposive sample, Multimethod (Discussion groups, semi-structured interviews, online survey), Grounded theory	To assess challenges in providing care to people with fibromyalgia, to examine knowledge and attitudinal challenges affecting optimal care	Participants: 32 PWC and HCP; , Age: Patients 41-60; HCP unavailable; Canada (Healthcare)	Findings revealed the presence of GP attitudinal and confidence challenges in caring for fibromyalgia patients. These fundamental competencies must be addressed to assure that all patients receive the quality of care necessary to manage their disease and to empower physicians to be more professionally effective.
Homma et al. (2016)(62)	Qualitative, Purposive sample, In-depth interviews, hermeneutic-phenomenological analysis	To explore and describe the meaning of attending a self-help group and its contribution to reconstructing the biographies of persons with FM.	Participants: 13 (M= 2, F= 11) PWC, Age: 29-73 (*Mdn*=46), Japan (Self-help groups)	Our findings indicate the importance of hearing the life stories of peers which can repair informants' disrupted biographies through the fu-ni-ochiru experience, a form of comprehension of one's illness by which the informant obtains a clear perspective through physical sensation, or full acceptance rather than logical understanding

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Horton et al. (2010)(63)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis	To explore the nature of professional 'best practice' in working with people with CFS/ME	Participants: 6 HCP, Age: unavailable, UK (Healthcare)	Listening skills, respect and trust are key for establishing a therapeutic relationship which recognises key features of the patient trajectory and promotes effective person-centred management of this CFS/ME. The findings of this study indicate the need to build such skills and knowledge more systematically into professional training informed by the experience of specialist services and those living with the condition
Houwen et al. (2019)(64)	Qualitative, Purposive sample, Video-stimulated recall interviews, constant comparative analysis	To identify GPs' difficulties in communication during MUS consultations.	Participants: 18 HCP, Age: unavailable, Netherlands (Healthcare)	GPs were aware of the importance of good communication. According to them, they could improve their communication further by paying more attention to psychosocial exploration, the structure of the consultation and communicating in a more person-centred way.
Howman et al. (2016)(65)	Mixed-methods, Purposive sample, Multimethod (Questionnaire, semi-structured interviews), thematic analysis, framework analysis	To explore GP trainees' clinical and educational experiences of managing people presenting with MUS.	Participants: 15 (M= 2, F= 13) HCP, Age: unavailable, UK (Healthcare training)	Despite evidence that clinicians struggle with managing patients with MUS, the current biomedical model of medical education continues to produce clinicians who rely on a biomedical model of decision making and in turn role-model this to students through the informal curriculum.
Humphrey et al. (2010)(66)	Qualitative, Purposive sample, Open-ended interviews, Grounded theory approach	to better understand aspects of fatigue that might be unique to FM as well as the impact it has on patients' lives.	Participants: 40 (M= 12, F= 28) PWC, Age: 25-69 (*M*=48.7), US, Germany, France (Community)	In addition to pain, fatigue was an important symptom for individuals with FM.The majority of individuals with FM who participated in this study experience fatigue and describe it as more severe than normal tiredness.

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Ivetić et al. (2013)(67)	Qualitative, Purposive sample, Focus groups, Qualitative content analysis	To explore the views of Slovenian family physicians on MUS and potential forms of treatment for patients with MUS	Participants: 24 (M= 7, F= 17) HCP, Age: *M*=44.9, Slovenia (Healthcare)	The results highlight the importance of good communication and trust between physicians and their patients with MUS, draw attention to the systemic barriers of the Slovenian health system and propose interactive education courses and participation of family physicians within the group for professional decision-making (the so-called 'family counsel').
Juuso et al. (2013)(68)	Qualitative, Purposive sample, Narrative interviews, phenomenological–hermeneutic interpretation	To elucidate meanings of feeling well for women with FM	Participants: 13 (F= 13) PWC, Age: 38-64 (*Mdn*=55), Sweden (Community)	For women with FM meanings of feeling well can be understood as having strength to be involved. The women's experiences of feeling well meant being in control, having power, finding one's own pace, experiencing feelings of belonging.
Juuso et al. (2011)(69)	Qualitative, Purposive sample, Narrative interviews, phenomenological hermeneutic interpretation	To elucidate meanings of pain for women with FM.	Participants: 15 (F= 15) PWC, Age: 38-64 (*Mdn*=54), Sweden (Community)	meanings of pain for women with FM can be understood as living with a double burden; living with an aggressive, unpredictable pain and being doubted by others in relation to the invisible pain. pain. To support the women's needs and help them to feel well despite their pain, it is important that nurses and health care personnel acknowledge and understand women with FM and their pain experiences.
Juuso et al. (2014)(70)	Qualitative, Purposive sample, Narrative interviews, phenomenological hermeneutic interpretation	To elucidate meanings of being received and met by others as experienced by women with FM	Participants: 9 (F= 9) PWC, Age: 40-65 (*Mdn*=59), Sweden (Community)	The meanings of being received and met by others as experienced by women with FM include being seen as a malingerer because of the invisibility of FM. Simultaneously, to

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				be acknowledged by others meant to be confirmed as the person one is.
Kengen Traska et al. (2012)(71)	Qualitative, Purposive sample, Group interview, Descriptive, content analysis	To describe how persons with fibromyalgia manage their lives given the multiple symptoms they experience.	Participants: 8 (F= 8) PWC, Age: 54-81 (*M*=61), USA (Community)	Women with fibromyalgia can develop strategies that enable them to cope with a life encumbered with chronic pain and fatigue.
Kirkham et al (2015)(72)	Qualitative, Purposive sample, Semi-structured interviews, IPA	Understanding individuals' lived experiences of chronic pain through pictorial representations	Participants: 7 (F= 7) PWC, Age: 36-52, UK (Community)	The images and accounts provide a powerful insight into the internal world of the pain sufferer and the subjective experience of chronic pain.
Kornelsen et al. (2016)(73)	Qualitative, Convenience sample, Open-ended interviews, Phenomenological analysis	To explore patients' experiences of prolonged uncertainty in diagnosis.	Participants: 38 PWC, Age and gender unavailable, Canada(Community)	We identified three experiential periods including the active search for a diagnosis, living with MUPS, and, finally, acceptance/resignation of their condition. Findings point to the heightened importance of the therapeutic relationship when dealing with uncertainty.
Kueny et al. (2021)(74)	Cross-cultural mixed methods, Purposive sample (men), Multi-method. Qualitative content analysis with descriptive group comparisons	To describe the pain and fatigue experiences of men with FMS from two Western countries	Participants: 17 (M= 17) PWC, Age: 30-63 (*M*=52), Spain, USA (Healthcare)	Men experience distinct facets of pain and fatigue compared with women, with notable similarities and differences across the Spanish and U.S. samples
LaChapelle et al. (2008)(75)	Qualitative, Convenience sample, Focus group interviews, Thematic analysis	To explore personal definitions of acceptance and the factors that facilitate or hinder acceptance (outside of therapy) in women with fibromyalgia and arthritis.	Participants: 45 Females (20 with FM), Age: 23-75 (*M*= 51.4 *SD*=12.2), Canada (Community)	Acceptance appears to be a vital part of adjusting to chronic pain but, to maximize adjustment, effective self-management is also necessary.

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Lee et al. (2001)(76)	Qualitative, Purposive sample, Semi-structured interviews, content and thematic analysis	To examine patterns of distress (namely, clinical symptoms and their social contexts), self-perceived stigma, perceived causes of illness, and various aspects of prior help-seeking among Chinese immigrants	Participants: 50 (M= 22, F= 28) PWC, Age: 20-64 (*M*=40.4; *SD*=10.7), Canada (Healthcare)	Findings from this clinical cultural epidemiological study highlighted the impact of migration and the stress of adjustment to life in Canada on clinical disorders of chronic fatigue and weakness among Chinese immigrants
Lehti et al. (2017)(77)	Qualitative, Purposive sample, Focus group & individual interviews, Grounded theory	To analyse patient and professional perceptions about (in)equity of care and rehabilitation of chronic pain patients from primary health care to assessment at a specialty rehabilitation clinic.	Participants: 25 (10 patients, 15 HCP), Gender: 11 Males (5 Patients, 6 HCP), 14 females (5 patients, 9HCP), Patient age: 35-65, HCP age: 27-63; Sweden (Healthcare)	From an equity and gender perspective, the study highlights the complexity in rehabilitation of chronic pain patients – both from patient and professional perspectives. Awareness of gendered and the biased preconceptions and norms is crucial when professionals struggle to offer equitable health care and rehabilitation
Leite (2011)(78)	Qualitative, Purposive sample, Focus group and semi-structured interviews, inductive thematic analysis	The needs for equity in health and social care expressed by adults living with CFS/ME.	Participants: 35 (M= 8, F= 27) PWC, Age: 18-56, UK (Community)	Changes in attitudes of health practitioners, policy makers and general public and more flexibly organised health and social care provision are needed to address equity issues in support needs expressed by people with CFS/ME, to be underpinned by research-based knowledge and communication, for public and professional education.
Lempp et al. (2009)(79)	Qualitative, Purposive sample, Semi-structured interviews, Content and discourse analysis	To understand patients' experience of living with Fibromyalgia syndrome	Participants: 12 (M= 1, F= 11) PWC, Age: 20-69 (*M*=49), UK (Community)	FMS is a condition that intrudes upon many aspects of patients' lives and is little understood. Greater attention needs to be paid to the links between the explanatory models of patients and staff, and most

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				important, to the interrelationship between the complex physical, psychological and social needs of patients with FMS.
Lian et al. (2017)(80)	Qualitative, Purposive sample, Qualitative survey (email), inductive qualitative thematic analysis	To explore the interactional dynamics of clinical encounters riddled by medical uncertainty, as experienced by people living with long-term medically unexplained fatigue in Norway	Participants: 256 (M= 26, F= 230) PWC, Age: 16-72 (*Mdn*=47), Norway (Community)	When physical symptoms cannot be detected, explained and managed by biomedical knowledge and technology, good doctor-patient partnerships are crucial.
Liedberg et al. (2006)(81)	Qualitative, Convenience sample, Structured interviews, Content analysis	To describe and compare difficulties young and newly diagnosed women in Sweden and the United States experienced during their first year after diagnosis.	Participants: 94 Female PWC (49 Swedish, 45 US); Age: 18-39; Sweden, USA (Healthcare)	In general, difficulties decreased and coping strategies increased over the 1-year period in both groups of newly diagnosed, young women
Löfgren et al. (2006)(82)	Qualitative, Purposive sample, Multi-method (Diaries, Focus groups, individual interviews), Content analysis & grounded theory	To understand more about how women with FM who manage to keep on working are coping with their symptoms. To explore the strategies they used for control of pain, fatigue and other symptoms.	Participants: 12 (F= 12) PWC, Age: 20-63, Sweden (Community)	The informants fought a constant struggle against the symptoms and the consequences of their fibromyalgia. Their strategies were action-oriented and evinced a positive spirit
Lombaard et al. (2005)(83)	Qualitative, Purposive sample, in-depth interview, autobiographical sketch, thematic analysis	To understand the subjective illness experience of those who	Participants: 4 (F= 4) PWC, Age: 20-50, South Africa (Community)	The profound changes instigated by CFS transform the relationship between the self and the body into a frontline where

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		suffer from CFS with a focus on the complex personhood of the sufferer		conflict prevails, and inner dissension threaten.
Maatz et al. (2016)(84)	Qualitative, Purposive sample, Semi-structured interviews, Content analysis	To explore specialists' experiences of and attitudes towards MUS and the patients who present with them; to examine their use of the term 'difficult'	Participants: 17 (M= 12, F= 5) HCP, Age: unavailable, UK (Healthcare)	This study shows that blanket statements such as 'difficult patients' mask the complexity of doctors' experiences in the context of MUS
Madden et al. (2006)(85)	Qualitative, Purposive sample, Semi-structured interviews, documentary analysis, induction–abduction method	To examine how patients diagnosed with fibromyalgia syndrome (FMS) construct meaning in the diagnosis and interpret this in relation to their illness experience	Participants: 17 (M= 1, F= 16) PWC, Age: 30-55, UK (Healthcare)	Whilst FMS may be an acceptable diagnosis to some, the problems encountered by many with the diagnosis suggest that it should be used sparingly within the medical community until future studies have explored the long-term impact of having a diagnosis, rather than simply the illness, of FMS.
Martinez et al. (2003)(86)	Qualitative, Purposive sample, Semi-structured interviews,	To examine how Brazilian fibromyalgia patients perceive their disease	Participants: 15 (F= 15) PWC, Age: *M*=41.9, Brazil (Community)	The perceptions of Brazilian fibromyalgia patients are similar to those of patients in other countries. One of the principal problems reported was seeking diagnosis and help from health professionals
McCue (2004)(87)	Qualitative, Purposive sample, Semi-structured interviews, Grounded Theory	To assess the illness experiences of women who recovered from CFS/ME in order to examine the extent to which the diagnoses they were given took a mental	Participants: 14 (F= 14) PWC, Age: 21-70 (*M*=42), UK (Community)	Individuals with CFS experience problems with diagnosis in terms of organic versus a mental health approach, and with acceptance and belief that they are actually ill.

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		health perspective, and whether or not it was accepted that they were suffering a genuine illness		
McDermott et al. (2011)(88)	Qualitative, Purposive sample, Semi-structured interviews, Constant comparative method	To explore the hopes and expectations of patients newly referred to a CFS/ME Service in the South of England	Participants: 20 (M= 3, F= 17) PWC, Age: 22-60 (*M*=39), UK (Healthcare)	Patients with symptoms of CFS/ME are looking for diagnosis, guidance, support, and hope from specialist services. They recognize that treatment involves self-help and have often proactively begun to reflect on multifactorial causes for their illness.
McMahon et al. (2012)(89)	Qualitative, Purposive sample, Narrative interviews, Narrative analysis	To explore how individuals with FM make sense of the illness experience and integrate it into their personal biographies	Participants: 10 (F= 10) PWC, Age: 25-70 *(M*=48), UK (Healthcare)	The narrative is characterized by a lack of movement and resolution, with participants engaged in an enduring struggle against the challenges of FM.
McManimen et al. (2019)(90)	Qualitative, Convenience sample, Online questionnaire, Thematic analysis, cross-group comparison	To analyse the patient perspective and to elucidate any issues in the doctor–patient relationship that may be leading patients to perceive HCPs as dismissive	Participants: 464 (M= 56, F= 407) PWC, Age: 18 years +, International (Healthcare)	Results are consistent with previous research. Many participants reported being told their ME or CFS symptoms were the consequences of a psychological issue. Women were more likely to report experiencing disbelief and insensitive treatment from HCPs. There is a need for further training and education for HCPs.
Menghoel et al. (2009)(91)	Qualitative, Purposive sample, Individual interviews, Thematic content analysis	To explore what patients that had completely recovered from FM experienced as being important for their recovery.	Participants: 5 (F= 5) PWC, Age: 37-49, Norway (Community)	Patients can recover from FM. The information from these informants suggests that to struggle against a role of chronic patient and keep up with their social obligations and goals were of great importance

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Mik-Meyer et al. (2012)(92)	Qualitative, Purposive sample, individual and group interviews, Thematic analysis; symbolic interaction approach	To understand how GPs classify and recognise patients with MUS	Participants: 21 HCP, Age and gender unavailable, Denmark (Healthcare)	the GPs' evaluation of the legitimacy of individuals with MUS, who are suffering and therefore unable to work and take on daily duties, relates first and foremost to an assessment of the social background and particular personality type of patients with MUS.
Monsivais (2008)[a](93)	Qualitative, Purposive sample, In-depth interviews, Thematic analysis, Focused ethnography	To describe the cultural constructions of chronic pain held by Mexican-American women.	Participants: 15 (F= 15) PWC, Age: 21-65 (*M*=45.6), US (Community)	Cultural constructions of chronic pain reflect influences of the legitimacy of invisible illnesses, and stigmas related to pain medication. If these ideas are not explored and addressed with patients, they may interfere with treatment.
Munday et al. (2021)(94)	Qualitative, Purposive sample, Focus group interviews, Thematic analysis	How chronic pain sufferers use language to describe their pain experience	Participants: 16 (M= 6, F= 10) PWC, Age: 22-74 (*M*=46.6), Australia (Community)	This study underscores research indicating the complexity of pain experience and hence pain language, and suggests that single word adjectival measures are inadequate to completely capture its complexity
Muraleetharan et al. (2018)(95)	Qualitative, Convenience sample, Survey (online & in-person), Thematic analysis	To understand why less men than women are diagnosed with FM, as well as whether the experiences of men with FM are different than their women counterparts	Participants: 1,163 (M= 805, F= 358) PWC, Age: 45-64, USA (Community)	Men with FM have negative experiences with physical and mental health, quality of life, relationships, and careers as a result of FM. There is a communication gap between men with FM and their HCPs.
Nettleton (2006)(96)	Qualitative, Purposive sample, In-depth interviews, narrative approach, Thematic analysis	To identify and elaborate on key issues experienced by individuals living with medically unexplained illness; to make	Participants: 18 (M= 5, F= 13) PWC, Age: 28-67, UK (Community)	The findings resonate with those of other sociological studies of illness that has no identified organic basis. Three themes were identified: morality, chaos and ambivalence.

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		sociological sense of emergent themes by drawing on sociological and cultural analyses of risk and the body.		
Nettleton et al. (2005)(97)	Qualitative, Purposive sample, in-depth narrative interviews, Narrative analysis	To explore the narratives of patients, who live with medically unexplained symptoms (MUS) and who have not secured a diagnostic label.	Participants: 18 (M= 5, F= 13) PWC, Age: 28-67, UK (Community)	Three features of the patients' narratives identified are: the 'chaotic' structure of their illness narratives; concern that symptoms may be 'all in the mind'; and their status as 'medical orphans'. An understanding of both the structure as well as the content of patients' narratives of undiagnosed illness may contribute to the development of more effective and sensitive patient centred care
Nilsen et al. (2009)(98)	Qualitative, Purposive sample, In-depth interviews, Narrative analysis & thematic analysis; hermeneutic phenomenological approach	To investigate how individuals experience their daily life with persistent pain; to investigate how individuals experience the temporal aspects of persistent pain & how this experience affects their meetings with health care providers	Participants: 20 (M= 10, F= 10) PWC, Age: 26-63 (*M*=42.7), Norway (Healthcare)	The study revealed disjunctions in the participants' lives between the temporal structure of pain experience, and the common, social time. Professionals should give attention to the temporal development of pain, by listening with empathy and imagination to gain an understanding of how their patients' lives have evolved.
Nunes et al. (2013)(99)	Qualitative, Purposive sample, Semi-directive interviews, Thematic content analysis	To explore patients' explanations of the medically unexplained physical symptoms that they were experiencing at	Participants: 15 (M= 4, F= 11) PWC, Age: 26-75, Portugal (Healthcare)	Patients with MUPS have explanations and fears associated with their complaints. The patient comes to the consultation not because of the symptom, but because of what he or she thinks about the symptom.

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		three time points (before, during and after the consultation).		The therapeutic relationship, therapeutic listening, and flexibility should be the basis for approaching patients with MUPS.
olde Hartman et al. (2009)(100)	Qualitative, Purposive sample, Focus group interviews, Constant comparative analysis	To explore GPs' perceptions about explaining MUS to patients and about how relationships with these patients evolve over time in daily practice	Participants: 22 (M= 14, F= 8) HCP, Age: 31-58, Netherlands (Healthcare)	GPs are aware of the importance of explaining MUS adequately to their patients, however they have difficulties in doing so. GPs report that, when patients keep presenting with MUS, they focus on maintaining the doctor-patient relationship by using ritual care.
Olson et al. (2015)(101)	Qualitative, Purposive sample, Unstructured interviews & card sorting; ethnoscience, Componential analysis	To learn how people with chronic fatigue syndrome describe fatigue	Participants: 14 (M= 1, F= 13) PWC, Age: 37-68, Canada (Community)	Fatigue in CFS is characterized by three separate domains—tiredness, fatigue, and exhaustion—with five segregates, including affective, muscular, somatic, cognitive, and social changes. Health care providers could utilize the findings of this study to help their patients understand the nature of CFS.
Paulson et al. (2002)(102)	Qualitative, Purposive sample (men), Narrative interviews, phenomenological hermeneutic interpretation	The meaning of men's lived experiences of living with pain of fibromyalgia type	Participants: 14 Male PWC, Age: 41-56 (*M*=47), Sweden (Community)	Overall, the meaning of men's lived experience of chronic pain was experienced as change in the body, self, and relationships. Striving to live life required achieving balance during both calm and difficult phases of the illness—struggling for a tolerable existence
Paulson et al. (2001)(103)	Qualitative, Purposive sample (men), Narrative interviews, Content analysis	To explore men's descriptions of non-malignant pain of fibromyalgia type	Participants: 14 Male PWC, Age: 41-56 (*Mdn*=48), Sweden (Community)	Men had specific experiences of bodily pain that was difficult to describe precisely. Therefore, they used metaphorical expressions to make their pain visible. Men's pain experience fell in 2 themes:

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
				perceptions of diversified body pain, and perceptions of fluctuating pain.
Paulson et al. (2002)(104)	Qualitative, Purposive sample (men), narrative interviews, Content analysis	To describe how men living with fibromyalgia-type pain experienced being patients in the Swedish health care system	Participants: 14 Male PWC, Age: 41-56 (*Mdn*=48), Sweden (Community)	This study indicates that there should be more attention paid to the care of men living with FM type pain. The waiting time, continuity and follow-ups have been noticed as areas that need to be paid more attention to in the health care system
Peplinskie (2016)[a](105)	Qualitative, Purposive sample, Interviews, narrative inquiry, Thematic analysis	Do patterns exist in chronic pain sufferers' subjective experiences, which may contribute to the onset and/or persistence of their somatic symptoms?	Participants: 12 (M= 4, F= 8) PWC, Age: 27-71, Canada (Community)	When invited to speak about momentous life events, all participants disclosed hardships they had endured. Furthermore, respondents had a strong propensity to downplay, or even deny, the severity of their often-traumatic life events.
Peters et al. (2009)(106)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis	To examine patient experiences of GPs' attempts to reattribute MUS in order to identify potential barriers to primary care management of MUS and improvement in outcomes	Participants: 23 (M= 3, F= 20) PWC, Age: 32-84 (*M*=53), UK (Healthcare)	Patients' decisions over how much, and what, information they present to GPs limits the effectiveness of communication training. Improving GP explanation of unexplained symptoms is insufficient to reduce patients' concerns.
Pryma (2017)(107)	Qualitative, Purposive sample (women), Semi-structured interviews, Inductive thematic analysis	To explore how women with fibromyalgia claim chronic pain by doing moral boundary-work and how race shapes the moral-boundary work performed	Participants: 24 Female PWC, Age: late 20s- mid 60s, USA (Community)	Claiming disability requires chronic pain sufferers to do moral boundary-work. However, the moral boundaries referenced in this process differ based on sufferers' intersectional identities

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Råheim et al. (2006)(108)	Qualitative, Purposive sample, Life-form interviews,, hermeneutic-phenomenological Analysis	To describe women's lived experience of chronic pain and fibromyalgia.	Participants: 12 (F= 12) PWC, Age: 34-51 (M=43.3), Norway (Community)	The women's stories point to a world experienced as fundamentally changed by a body in chronic pain, describing a struggle in which they feel that their existence is at stake.
Rasmussen et al. (2018)(109)	Qualitative, Purposive sample, Focus groups, Thematic analysis	To explore how GPs understand and handle MUS; how medical frames organise GPs' understanding of MUS, and how this enables (or disables) patient management.	Participants: 23 (M= 11, F= 12) HCP, Age: unavailable, Norway (Healthcare)	Biopsychosocial framing, combined with clinical experience, enables GPs to understand and handle MUS better than biomedical framing does. Medical students should spend more time learning biopsychosocial medicine, and to integrate the clinical knowledge of their peers with their own.
Richardson (2005)(110)	Qualitative descriptive, Purposive sample, Life-grid interviews	To explore the use of 'extraordinary' stories by women with chronic widespread pain as one aspect of the process of establishing a positive identity in the face of delegitimation and threats to identity	Participants: 6 (F= 6) PWC, Age: 51-88, Australia (Community)	The accounts of women suffering from chronic widespread pain are constructed to portray a positive identity
Richardson et al. (2006)(111)	Qualitative, Purposive sample, Multi-method (lifegrid interviews, diaries and diary interviews), IPA	To explore how sufferers of chronic widespread pain perceive and plan for their futures, given the underlying and pervasive uncertainty of the condition.	Participants: 8 (M= 4, F= 4) PWC, Age: 40-58, UK (Community)	The pervading uncertainty of chronic widespread pain, in which there is no framework for the trajectory of the condition, affects perceptions of the future and makes planning for the future difficult

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Richardson et al. (2007)(112)	Qualitative, Purposive sample, Multi-method (lifegrid interviews, diaries and diary interviews), Thematic and narrative analysis	To examine the experience and meaning of support for people with widespread chronic pain in the context of their families	Participants: 8 (M= 4, F= 4) PWC, Age: 40-60, UK (Community)	There is a varying, dynamic and reciprocal nature of practical and emotional support in the family. Family members may provide support but are also receivers of support from the person with chronic widespread pain
Ringsberg et al. (2006)(113)	Qualitative, Purposive sample, Focus group interviews, Phenomenography	To elucidate GPs' perceptions of patients with MUS, focusing on stressing situations, emotional reactions and coping strategies	Participants: 27 (M= 11, F= 16) HCP, Age: 33-57, Sweden (Healthcare)	GPs had difficulties in managing their own stress when working with patients with MUS.
Rodham et al. (2010)(114)	Qualitative, Purposive sample, semi-structured interviews, IPA	To explore the lived experiences of both those with FMS and their spousal carers.	Participants: 4 Female PWC (+ spouse), Age: 38-65, UK (Community)	We identified a number of practical and attitudinal barriers that had led to the diminution of social networks for both members of the couple
Russell et al. (2018)(115)	Qualitative, Purposive sample, Focus group interviews; Thematic content analysis; analysis of non-verbal interactions	To explore the perceptions of fatigue, sleep dysfunction, and exercise in people with FMS.	Participants: 14 (M= 2, F= 12) PWC, Age: unavailable, UK (Community)	This study has identified that those with FMS perceive a lack of understanding of their condition and experience a profound sense of loss as a result of their FMS. This sense of loss extends to their inability to engage with physical activity and exercise
Sallinen et al. (2011)(116)	Qualitative, Purposive sample, Narrative life story interview, Narrative analysis	To explore how fatigue was experienced and explained in life stories of women with a long history of fibromyalgia	Participants: 20 Female PWC, Age: 34-65 (*M*=54), Finland (Community)	In fibromyalgia, fatigue is a transient, extreme and intensive experience, which causes major disability and distress and which has consequences on every aspect of life.
Sallinen et al. (2012)(117)	Qualitative, Purposive sample, Individual narrative interviews, Narrative analysis	To explore the structure and content of narrated life stories of women with fibromyalgia to elucidate	Participants: 20 Female PWC, Age: 34-65 (*M*=54), Finland (Community)	The invisible symptoms and unheard experiences describe in this study help us to understand the individual suffering that is associated with fibromyalgia. The model

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		the different life courses patients with fibromyalgia may have		narratives and their counternarrative presented in this study reflect the diversity of meanings ascribed to fibromyalgia
Sallinen et al. (2019)(118)	Qualitative, Purposive sample (men), Narrative life story interviews, Narrative analysis	To elucidate the impacts of fibromyalgia on men's daily life and work ability	Participants: 5 Male PWC, Age: 24-51 (*M*=43), Finland (Community)	Living with fibromyalgia had forced the men to look at their lives, social roles and work from a new perspective. Adjusting ones activities had helped to manage the fluctuating symptoms and to support work ability in many cases.
Sallinen et al. (2011)(119)	Qualitative, Purposive sample, Narrative life story interviews, Narrative analysis	To explore and analyse the experiences related to peer support among women with a long historyof fibromyalgia.	Participants: 20 Female PWC, Age: 34-65 (*M*=54), Finland (Community)	Long-term fibromyalgia patients saw peer support as an impetus to an ongoing process of reconstruction of identity, illness acceptance and coping with fibromyalgia
Sallinen et al. (2019)(120)	Qualitative, Purposive sample, Narrative life-story interviews, Narrative analysis	To elucidate the interplay between illness and gender by exploring life-stories of men who suffer from fibromyalgia	Participants: 8 Male PWC, Age: 24-61 (*M*= 47), Finland/Norway (Community)	Fibromyalgia shapes the way men see themselves as men. The interplay between illness and gender seems to lead to re-constructing their masculinity in terms of what is possible.
Schaefer (2005)(121)	Qualitative, Purposive sample, Interviews, Thematic analysis, Phenomenological approach	To learn what it is like for African American women to live with fibromyalgia	Participants: 10 Female PWC, Age: 37-59 (*M*= 46.5), USA (Community)	Managing an illness involves maintaining the ability to think, coping with physical changes due to treatment and the illness, and acknowledging the need to nurture the spirit. It is important that healthcare providers in all situations care for the whole person—body, mind and spirit—to help achieve the best possible condition to restore their integrity.
Söderberg & Lundman (2001)(122)	Qualitative, Purposive sample, Narrative interviews,, Thematic content analysis	To illuminate the transitions experienced by women with FM.	Participants: 25 Female PWC, Age: 35-60 (*M*= 46.8), Sweden (Community)	This study shows that transitions occur in different areas of the women's life—in daily life pattern, in working life, in social

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
				life, in family life—but the women expressed the view that they were learning to live with the changes. The experience of transitions is apparently something that is invisible to almost everyone except the women themselves.
Söderberg et al. (2002)(123)	Qualitative, Purposive sample, Narrative interviews, Phenomenological-hermeneutic interpretation	To elucidate the meaning of the lived experience of fatigue and tiredness in women with FM and healthy women.	Participants: 25 Female PWC, Age: 35-60 (*M*=46.8), Sweden (Community)	The findings of this study show that fatigue narrated by women with FM is a quite different experience from tiredness narrated by healthy women
Sowińska et al. (2018) (124)	Qualitative, Purposive sample, Semi-structured interviews, Qualitative content analysis	To explore Polish patients' perspectives on living with MUS.	Participants: 20 (M= 8, F= 12) PWC, Age: 18-57 (*M*=37.4), Poland (Healthcare)	This study recognizes the importance of patients' personal accounts of living with MUS and the necessity of appropriate and acceptable explanation of symptoms. Improving the management of patients with MUS in primary care requires the allocation of increased financial resources for GP training and facilitated access to psychologists and psychotherapists
Stone (2013b)(125)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis	To explore the strategies general practitioners use to manage patients with mixed emotional and physical symptoms and no diagnosis.	Participants: 24 (M= 13, F= 11) HCP, Age: 20 to ≥ 60, Australia (Healthcare)	Managing patients with medically unexplained symptoms involves professional and personal challenges. Shifting the emphasis from cure to coping without a disease name is challenging for both the doctor and the patient.
Stone (2014)(126)	Qualitative, Convenience sample, Semi-structured interviews, Constructivist grounded theory; Iterative inductive coding	To explore how novice and experienced GPs manage patients with MUS and how these skills	Participants: 24 (M= 13, F= 11) HCP, Age: 20 to ≥60, Australia (Healthcare)	Negative feelings and a lack of diagnostic language and frameworks may prevent registrars from managing these patients effectively. This study suggests that supervisors have rudimentary frameworks

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		are taught and learned in GP training		which are idiosyncratic. They rely on coping strategies based on personal experience and teach registrars through experiential strategies.
Stone (2013a)(127)	Qualitative, Purposive sample, Semi-structured interviews, constructivist grounded theory, Iterative inductive coding	To explore how GPs make sense of medically unexplained symptoms	Participants: 24 (M= 13, F= 11) HCP, Age: 20 to ≥60, Australia (Healthcare)	GPs were aware of the ethical relevance of psychiatric diagnoses and attempted to protect their patients from stigma. Managing patients with MUS can be uncomfortable and requires examination of core professional and personal values.
Sturge-Jacobs (2002)(128)	Qualitative, Purposive sample, Unstructured interviews, Phenomenological approach, Thematic analysis	To describe and enhance the understanding of the experience of living with FM	Participants: 9 Female PWC, Age: 30-56, Canada (Community)	The essence or "what makes this experience what it is" was captured as "living with or continually confronting an invisible disability.
Theadom et al. (2010)(129)	Qualitative, Purposive sample, Semi-structured interviews, Thematic analysis, interpretative phenomenological approach	To explore perceptions of sleep quality and the influence sleep has on symptoms and daily life in people with Fibromyalgia	Participants: 16 (M= 2, F= 14) PWC, Age: 21-61 (*M*=50.95; *SD*=11.94), UK (Community)	People with FMS find that sleep difficulties are one of the most challenging symptoms to cope with, since exacerbation of pain, fatigue and cognitive difficulties seem to be strongly related to poor sleep.
Thorne et al. (2004)(130)	Qualitative, Purposive sample, In-depth interviews, Constant comparative analysis, Grounded theory	To portray how individuals with fibromyalgia (FM) describe interactions with health care providers and to highlight patterns of communication that were perceived as helpful or unhelpful during these encounters	Participants: 11 (M= 1, F= 10) PWC, Age: 21-76, Canada (Healthcare)	Through their communications with patients, health care professionals, including physiotherapists, are in a position to make an important difference in the lives of persons with FM.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Travers et al. (2008)(131)	Qualitative, Convenience, purposive, discriminate, Semi-structured interviews, Narrative analysis, Grounded Theory	To examine the illness experiences, and specifically the experiences of self, for people affected with CFS living in Australia.	Participants: 19 (M= 9, F= 14) PWC, Age: 20-75 (*M*=49), Australia (Community)	Understanding the intricate nature of the CFS experience provides guidance to people with the condition and their practitioners. Explication of the process of the struggling self seeking renewal brings to the fore the threatening, monotonous, chaotic, protective and transforming aspects of the CFS experience.
Undeland et al. (2007) (132)	Qualitative, Purposive sample, Focus group interviews, Systematic text condensation	To explore experiences and consequences of the process of being diagnosed with fibromyalgia	Participants: 11 (F= 11) PWC, Age: 42-67, Norway (Self-help groups)	A diagnosis may be significant when it provides the road to relief or legitimizes the patient's problems. The initial blessing of the fibromyalgia diagnosis seems to be limited in the long run.
Warner et al. (2017)(133)	Qualitative, Purposive sample, In-depth interviews, Thematic analysis, Framework approach	To explore the ways in which doctors working in secondary care approach and manage patients with medically unexplained symptoms.	Participants: 20 (M= 12, F= 8) HCP, Age: unavailable, UK (Healthcare)	There is a need for serious consideration as to how the management of patients with medically unexplained symptoms is included in medical training and in the planning and delivery of services.
Werner et al. (2004)(134)	Qualitative, Purposive sample, in-depth interviews, Discourse analysis, Feminist interpretation, Narrative theory	To explore issues of self and shame in illness accounts from women with chronic pain.	Participants: 10 (F= 10) PWC, Age: 26-58 (*M*=24.5), Norway (Healthcare)	The stories of women with chronic muscular pain have a moral plot and an argument about biomedical causes and reality of their pain, appealing to both the interviewer, and a medical and general public. Behind the informants' stories about their own strength and the others (negative) talk of illness, we hear whispered accounts relating to the medical narrative about hysteria.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
Werner et al. (2003)(135)	Qualitative, Purposive sample, In-depth interviews, phenomenological analysis	To explore the nature of ''work'' done by the patients in order to be believed, understood, and taken seriously when consulting the doctor.	Participants: 10 (F= 10) PWC, Age: 26-58 (*M*=41.5), Norway (Healthcare)	The women patients' accounts indicated that they engage in hard physical and gendered work in order to appear as ''just right'' under normative, biomedical eyes.
Wheeler (2011)[a](136)	Qualitative, Purposive sample, Semi-structured interviews, phenomenological hermeneutic interpretation	To describe the meaning for several FM sufferers of their own lived experiences with a controversial illness; To explore the concept of unknown aetiology as one factor in illness uncertainty	Participants: 10 (M= 1, F= 9) PWC, Age: 43-86 (*M*=59.1), US (Community)	Only a few participants in this study cited concern over the unknown cause of their illness. The participants in this study want their illness experience to be understood and to be acknowledged. They are focused on increasing coping skills as well as their knowledge regarding their illness. Many described deficits in their support system and numerous negative experiences with medical practitioners
Whitehead (2006)(137)	Qualitative, Purposive sample, In-depth interviews, Interpretive analysis; hermeneutic phenomenological approach	To examine the reconstruction of self-identity for those experiencing CFS/ME.	Participants: 17 (M= 6, F= 11) PWC, Age: 13-63, UK (Community)	Understanding the patients' experience and recognising that different stages may exist is important for health professionals. This awareness can enhance shared understanding and opportunities to work with people in negotiating the impact of illness.
Whitehead (2006)(138)	Qualitative, Purposive sample, In-depth interviews, Interpretive analysis; hermeneutic phenomenological approach	To explores how people with chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ ME) describe and interpret their illness experience by applying	Participants: 17 (M= 6, F= 11) PWC, Age: 13-63, UK (Community)	A trajectory of narrative typologies is experienced, starting with a restitution narrative, moving to a chaos narrative and, for most, back to a restitution narrative and on to a quest narrative.

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		Arthur Frank's narrative typologies		
Wileman et al. (2002)(139)	Qualitative, Purposive sample, Semi-structured interviews, Constant comparative approach	To explore GPs attitudes to the management of patients who present with MUS	Participants: 15 (M= 11, F= 6) HCP, Age: unavailable, UK (Healthcare)	Patients with MUS were seen to be presenting with inappropriate symptoms that were a manifestation of emotional or social distress. GPs felt ill-equipped to deal with the presentations
Wilson et al. (2011)(140)	Qualitative, Convenience sample, Narratives (online), Inductive thematic analysis	To understand how people can live well in spite of the presence of chronic fatigue.	Participants: 43 (M= 3, F= 39, 1 unknown) PWC, Age: 18-65+, International (Community)	People can and do find ways to live well with chronic fatigue and that to do so requires a great deal of effort on their part. Understanding how the person with chronic fatigue has come to conceptualize his/ her experiences will be a more fruitful starting point than providing recipes for successful living.
Woivalin et al. (2004)(141)	Qualitative, Purposive sample, Focus group interviews, Phenomenographic approach	To explore GPs' perceptions and ways of managing patients with medically unexplained symptoms (MUS).	Participants: 27 (M= 11, F= 16) HCP, Age: 33-57, Sweden (Healthcare)	In their work with patients with MUS, GPs need support and further training to improve the way the biomedical frame of reference is integrated with the humanistic perspective.
Wuytack et al. (2011)(142)	Qualitative, Purposive sample, Semi-structured interviews, descriptive phenomenology, Phenomenological analysis	To gain a better understanding of the subjective experience of fibromyalgia, focusing on the personal, occupational and social impact of the condition on patients' lives	Participants: 6 (F= 6) PWC, Age: 36-66 (*M*=51), Belgium (Healthcare)	The study revealed the negative impact of fibromyalgia on patients' lives as comprising of great complexity and individuality. Several implications for health care practitioners can be extrapolated, including the need of a more efficient diagnostic process and increased education about the fibromyalgia experience.
Yon et al. (2015)(143)	Qualitative, Purposive sample, In-depth interviews, Framework approach	To explore junior doctors' views and experiences of managing patients with	Participants: 22 (M= 9, F= 13) HCP, Age: 20-49, UK (Healthcare)	Current training does not equip junior doctors with the necessary knowledge and skills to effectively and confidently manage

Author (year) (Numeric value in Chapter 3)	Study design, sampling, data collection and analysis methods	Phenomena of Interest	Participant number, gender, age, country (setting)	Authors' conclusions
		MUS, and their recommendations for future training		patients with MUS. There is an urgent need to improve postgraduate training about the topics of MUS and avoiding over-investigation.
[a] Thesis/ dissertation	IPA= interpretative phenomenological analysis	MUPS= Medically Unexplained Physical Symptoms; MUS= Medically Unexplained Symptoms; FM= Fibromyalgia Syndrome; CFS/ME= Chronic Fatigue Syndrome/Myalgic Encephalomyelitis	HCP= Healthcare practitioner; PWC= Person With Condition	

Appendix A4. Quality Appraisal of Studies

Table A4

Quality appraisal of included studies (QARI)

		JBI Checklist for Qualitative Research									
Author (year)	Design	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Aamland et al. (2017)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Aamland et al. (2013)	Qualitative	U	Y	Y	Y	Y	Y	N	Y	Y	Y
Abokdeer (2019)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Acker (2008)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Anderson et al. (2014)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Armentor (2017)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Arnold et al. (2008)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Arroll et al. (2013)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Arroll et al. (2008)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Åsbring (2001)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Åsbring et al. (2003)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Åsbring et al. (2004)	Qualitative	Y	Y	Y	Y	Y	U	Y	Y	Y	Y
Åsbring et al. (2002)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Ashe et al. (2017)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Barcroft (2016)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Bee et al. (2016)	Qualitative	Y	Y	Y	Y	Y	U	N/A	Y	Y	Y
Boulton (2019)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Briones-Vozmediano (2017)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Briones-Vozmediano (2013)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Briones-Vozmediano et al. (2018)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

Author (year)	Design	JBI Checklist for Qualitative Research									
		Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Briones-Vozmediano et al. (2016)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Brooks et al (2014)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Broughton et al. (2017)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Brown et al. (2017)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Brownell et al. (2016)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Chen (2016)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	N	U
Chen et al. (2020)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Chew-Graham et al. (2008)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Chew-Graham et al. (2009)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Chew-Graham et al. (2010)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Cipolletta et al. (2020)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Clarke et al. (2014)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Clarke et al. (2007)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Cooper et al. (2017)a	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Cooper et al. (2017)b	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Crooks (2007)	Mixed method	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Crooks et al. (2008)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Cudney et al. (2002)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Cunningham et al. (2006)	Qualitative	Y	Y	Y	Y	Y	U	Y	Y	U	U
Dennis et al. (2013)	Qualitative	Y	Y	Y	Y	Y	U	U	U	Y	Y
Devendorf et al (2020)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Devendorf et al. (2019)	Qualitative	Y	Y	Y	Y	Y	U	Y	Y	Y	Y
Dickson et al. (2007)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

		JBI Checklist for Qualitative Research									
Author (year)	Design	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Dickson et al. (2008)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Durif-Bruckert et al. (2015)	Qualitative	U	Y	Y	Y	Y	Y	Y	Y	Y	Y
Dwamena et al. (2009)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Edwards et al. (2007)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Escudero-Carretero et al. (2010)	Qualitative	U	Y	Y	Y	Y	N	N	Y	Y	Y
France et al. (2008)	Qualitative	Y	Y	Y	U	U	Y	N	N	Y	U
Gilje et al. (2008)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Gordon et al. (2017)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Grape et al. (2015)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Grape et al. (2017)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Gray et al. (2003)	Qualitative	Y	Y	Y	Y	U	Y	Y	Y	Y	Y
Guise et al. (2010)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Gullacksen et al. (2004)	Qualitative	Y	Y	Y	U	U	Y	U	U	Y	Y
Hallberg et al. (2011)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Hannon et al. (2012)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Harding et al. (2005)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Harsh et al. (2016)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Hayes et al. (2010)	Mixed methods	Y	Y	Y	Y	Y	U	U	Y	Y	Y
Homma et al. (2016)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Horton et al. (2010)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Houwen et al. (2019)	Qualitative	Y	Y	Y	Y	Y	U	U	Y	Y	Y
Howman et al. (2016)	Mixed methods	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

Author (year)	Design	JBI Checklist for Qualitative Research									
		Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Humphrey et al. (2010)	Qualitative	Y	Y	Y	U	Y	Y	U	Y	Y	Y
Ivetić et al. (2013)	Qualitative	Y	Y	Y	Y	Y	U	Y	Y	Y	Y
Juuso et al. (2013)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Juuso et al. (2011)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Juuso et al. (2014)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Kengen Traska et al. (2012)	Qualitative	Y	Y	Y	Y	Y	U	U	U	Y	Y
Kirkham et al (2015)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Kornelsen et al. (2016)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Kueny et al. (2021)	Mixed methods	U	Y	Y	Y	Y	U	U	Y	Y	Y
LaChapelle et al. (2008)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Lee et al. (2001)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	U	Y
Lehti et al. (2017)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Leite (2011)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Lempp et al. (2009)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Lian et al. (2017)	Qualitative	Y	Y	Y	Y	Y	Y	N/A	Y	Y	Y
Liedberg et al. (2006)	Qualitative	U	Y	Y	Y	Y	U	U	U	Y	Y
Löfgren et al. (2006)	Qualitative	Y	Y	Y	N	U	Y	Y	N	Y	U
Lombaard et al. (2005)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	U	Y
Maatz et al. (2016)	Qualitative	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Madden et al. (2006)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Martinez et al. (2003)	Qualitative	Y	Y	Y	U	U	N	N	U	Y	U
McCue (2004)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	U	Y
McDermott et al. (2011)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

		JBI Checklist for Qualitative Research									
Author (year)	Design	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
McMahon et al. (2012)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	U	Y
McManimen et al. (2019)	Qualitative	Y	Y	Y	Y	Y	N	U	Y	Y	Y
Menghoel et al. (2009)	Qualitative	Y	Y	Y	Y	Y	U	Y	Y	Y	Y
Mik-Meyer et al. (2012)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Monsivais (2008)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	U
Munday et al. (2021)	Qualitative	Y	Y	Y	Y	Y	U	N	Y	Y	Y
Muraleetharan et al. (2018)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	U	Y
Nettleton (2006)	Qualitative	Y	Y	Y	Y	Y	N	N	Y	Y	Y
Nettleton et al. (2005)	Qualitative	Y	Y	Y	Y	Y	U	N	Y	Y	Y
Nilsen et al. (2009)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Nunes et al. (2013)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
olde Hartman et al. (2009)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	U	Y
Olson et al. (2015)	Qualitative	Y	Y	Y	Y	Y	Y	U	U	Y	Y
Paulson et al. (2002)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Paulson et al. (2001)	Qualitative	Y	Y	Y	Y	Y	N	U	U	Y	Y
Paulson, Norberg et al. (2002)	Qualitative	Y	Y	Y	Y	Y	N	N	Y	Y	Y
Peplinskie (2016)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Peters et al. (2009)	Qualitative	Y	Y	Y	Y	Y	U	U	Y	Y	Y
Pryma (2017)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Råheim et al. (2006)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Rasmussen et al. (2018)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Richardson (2005)	Qualitative	Y	Y	U	U	Y	U	U	Y	Y	Y

Author (year)	Design	JBI Checklist for Qualitative Research									
		Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Richardson et al. (2006)	Qualitative	Y	Y	Y	Y	Y	U	U	Y	Y	Y
Richardson et al. (2007)	Qualitative	Y	Y	Y	Y	Y	U	U	Y	Y	Y
Ringsberg et al. (2006)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Rodham et al. (2010)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Russell et al. (2018)	Qualitative	U	Y	Y	Y	Y	U	Y	Y	Y	Y
Sallinen et al. (2011)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sallinen et al. (2012)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sallinen et al. (2019)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Sallinen et al. (2011)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sallinen et al. (2019)	Qualitative	Y	Y	Y	Y	Y	Y	Y	U	Y	Y
Schaefer (2005)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Söderberg & Lundman (2001)	Qualitative	Y	Y	Y	Y	Y	N	N	Y	Y	Y
Söderberg et al. (2002)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Sowińska et al. (2018)	Qualitative	U	Y	Y	Y	Y	Y	Y	Y	Y	Y
Stone (2014)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Stone (2013a)	Qualitative	Y	Y	Y	Y	Y	U	N	Y	Y	Y
Stone (2013b)	Qualitative	Y	Y	Y	Y	Y	U	N	Y	Y	Y
Sturge-Jacobs (2002)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Theadom et al. (2010)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Thorne et al. (2004)	Qualitative	Y	Y	Y	Y	Y	N	U	Y	Y	Y
Travers et al. (2008)	Qualitative	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Undeland et al. (2007)	Qualitative	U	Y	Y	Y	Y	Y	U	Y	U	Y
Warner et al. (2017)	Qualitative	U	Y	Y	Y	Y	N	U	Y	Y	Y

Author (year)	Design	JBI Checklist for Qualitative Research									
		Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Werner et al. (2004)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	U	Y
Werner et al. (2003)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	U	Y
Wheeler (2011)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Whitehead (2006)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Whitehead (2006)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Wileman et al. (2002)	Qualitative	U	Y	Y	Y	Y	N	N	Y	U	Y
Wilson et al. (2011)	Qualitative	Y	Y	Y	Y	Y	Y	N/A	Y	Y	Y
Woivalin et al. (2004)	Qualitative	Y	Y	Y	Y	Y	Y	U	Y	Y	Y
Wuytack et al. (2011)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Yon et al. (2015)	Qualitative	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

Note. Y = yes, U= Unclear, N= No, N/A= Not Available

JBI Checklist for Qualitative Research Questions

Q1. Is there congruity between the stated philosophical perspective and the research methodology?

Q2. Is there congruity between the research methodology and the research question or objectives?

Q3. Is there congruity between the research methodology and the methods used to collect data?

Q4. Is there congruity between the research methodology and the representation and analysis of data?

Q5. Is there congruity between the research methodology and the interpretation of results?

Q6. Is there a statement locating the researcher culturally or theoretically?

Q7. Is the influence of the researcher on the research, and vice- versa, addressed?

Q8. Are participants, and their voices, adequately represented?

Q9. Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body?

Q10. Do the conclusions drawn in the research report flow from the analysis, or interpretation, of the data?

Appendix A5. Frequencies of Extracted Findings: Patients

Table A5

Frequency Table of Synthesized Findings, Categories and Findings reflecting Patient Perspectives

	Findings (N)	Proportion (%)
Synthesized Finding 1: Symptom experience and impact		
Physical symptoms and difficulties	22	4.31
Cognitive symptoms and difficulties	7	1.37
Symptoms are chronic and unrelenting	9	1.76
Symptoms are variable and unpredictable	8	1.57
Symptoms are overwhelming and threatening	8	1.57
The cyclical nature of symptoms	5	0.98
The burden of invisibility	4	0.78
Impact on everyday life	15	2.94
Impact on family life and relationships	6	1.17
Impact on social life and leisure activities	11	2.15
Impact on work life	8	1.57
Psychological impact of symptoms	18	3.52
Synthesized Finding 2: The patient journey		
Initial resistance and gradual interference	10	1.96
Facing uncertainty	10	1.96
Making sense of complexity in the illness experience	18	3.52
Seeking information and diagnosis	16	3.13
The experience of having a diagnosis	9	1.76
Active help-seeking	6	1.17
Acceptance and realization	14	2.74
Transition to symptom management	9	1.76
Experience of an ongoing struggle	12	2.35
Changing perspectives on life	5	0.98
Perspectives on the future	9	1.76
Recovery	6	1.17
Synthesized Finding 3: Identity loss and change		
Experience of identity loss and change	10	1.96
Narratives about the true self	7	1.37
Experience of the body as changed, obstructive and unreliable	12	2.35
Reconstructing and accepting a new identity	10	1.96
Factors that support identity reconstruction	7	1.37
Synthesized Finding 4: Managing chronic illness		
Taking charge	8	1.57
Gaining control through knowledge	4	0.78
Making lifestyle changes	8	1.57
Energy and activity management	9	1.76
Avoiding triggers	5	0.98
Positive thinking	4	0.78

	Findings (N)	Proportion (%)
Distraction	3	0.59
Synthesized Finding 5: Understanding and legitimacy		
Lack of understanding	5	0.98
Delegitimizing experiences	8	1.57
Social insensitivity and disinterest	10	1.96
Strategies for navigating social insensitivity	12	2.35
Synthesized Finding 6: Support needs and experiences		
Support needs	5	0.98
Unmet support needs	7	1.37
Positive experiences of receiving support	10	1.96
Experience of peer support	9	1.76
Synthesized Finding 7: Healthcare needs and experiences		
The healthcare experience as an uphill battle	9	1.76
Feeling lost and neglected by the system	7	1.37
Challenging interactions with HCP	10	1.96
Inappropriate explanations and treatments	6	1.17
Lack of knowledge and understanding among HCP	10	1.96
Experience of being disbelieved and dismissed	14	2.74
Stigma and prejudice in healthcare	8	1.57
Complexity of symptoms as a challenge	7	1.37
Positive experiences with healthcare	9	1.76
Healthcare needs and expectations	18	3.52
Synthesized Finding 8: Managing healthcare encounters		
Confronting healthcare providers	5	0.98
Appropriate behavior and appearance	7	1.37
Disengagement	3	0.59

Appendix A6. Illustrative Quotes: Patients

Table A6

Experiences of PwC: Illustrative quotes for synthesized findings

1.1 The Symptom Experience
"[Besides the pain and swelling in my feet I also] get migraine headaches, and heart burn, and ulcers. And [we] wonder why we are depressed!" – Female, Fibromyalgia (Cudney et al., 2002, p. 40) *"This illness is so unpredictable, you cannot pin it down, you cannot describe it. Each day is different from the previous one. And because of this I've lost confidence in myself, because I cannot… define myself, I cannot say, this is how I am."* – Female, CFS (Lombaard & Mouton, 2005, p. 298) *"When it comes to the family, it's hard to accept and deal with. I remember the day I got up and found that my legs wouldn't work, and my children put my knickers on. I'll never forget the look of disbelief on their faces as they pulled my knickers up, and the sight of seeing my bum. They said "something really serious is going on here.""* – Female, Fibromyalgia (Briones-Vozmediano et al., 2016, p. 844) *"I started to think to myself ''Am I just making this up? Is it all in my head?'' And I started to worry about it like ''Am I just using this as an excuse because I don't like my life?'' And there's nothing wrong with my life but because you're so low and you're not yourself, these doubts creep in, you know? Who am I and am I turning into a malingerer?"* – Female, CFS (Dickson et al., 2008, p. 466)
1.2 The Patient Journey
"because I didn't understand what was happening to me I'd try to carry on with my normal everyday life, which really was a bad thing to do" – Female, 37, CFS/ME (Edwards et al., 2007, p. 207) *". . . one of the doctors actually turned to me and said, do you realize in the last 6 months you've been in this hospital 26 times?"* – Female, Chronic Pain (K. Clarke & Iphofen, 2007, p. 106) *''My doctor told me I was a bit of a nightmare because he didn't think I was psychologically ill, but didn't quite know what to make of my symptoms.''* – Female, CFS/ME (recovered) (McCue, 2004, p. 197) *Once I read about the condition, I realized that I couldn't really ever be fully better again. Knowing what was wrong made it easier to deal with, since I had a definite answer, however emotionally, it was a lot harder'* – Female, Fibromyalgia (Dennis et al., 2013, p. 772) *I think it's learning about accepting what you can do and trying not to get upset about what you can't do. Trying to accept what you've got and, well you're still alive, you've still got all your body parts and what not. (Sharon)* – Female, 35, Fibromyalgia (McMahon et al., 2012, p. 1363) *"So I work on myself all the time. Because it never ends. Even though I'm well now, it's quite a job to stay that way. You need to work on the physical, the mental. You must be careful about what you eat. What you do. How far you can walk. How little you can walk. The cold, the heat. How much you can endure. How many people are around you. It's a huge job"* – Female, Fibromyalgia (recovered) (Grape et al., 2015, p. 684) *"It frightens me it does. I can have some really good days and I feel fantastic and then, I can have some really, really bad days and think, 'I could be like this for the rest of my life' when I'm older. If it is like this now at 40, what is it going to be like, it does worry me that."* – Female, 40, Fibromyalgia (Richardson et al., 2006, p. 215)
1.3 Identity Loss and Change
"I mean sometimes just a feeling that em… I had no purpose. I was an empty shell. I am a fragment of the man I used to be. I can do nothing now. I'm useless to everyone. I'm useless to myself. My self-esteem is rock bottom" – Male, CFS (Dickson et al., 2008, p. 464) *"At times I see my body . . . often . . . as unreliable because I cannot predict how my state of health will fluctuate between now and an hour from now . . . Although it is irrational, one starts to see oneself as a person who cannot be depended upon. You cannot rely on yourself, others cannot rely on you. You struggle to make appointments and it feels for you as though you have become an unreliable person. It silently breaks down your faith, your self-confidence . . . and you indeed feel inferior to some degree"* – Female, CFS (Lombaard & Mouton, 2005, p. 298)
1.4 Managing Chronic Illness

"To cope with ME physically, I was compelled to listen to my body and to become aware of its warning signs when I was close to a relapse . . . I have made it my responsibility to look after my body." – Female, CFS (Lombaard & Mouton, 2005, p. 301)

"When I have more energy, I do more. The hardest thing in learning to manage the fatigue is to not overdo on my higher energy days. Moderation is highly essential to my well being." (Wilson et al., 2011, p. 2165)

"… just do little things and find all the little positives. That seems to be helping me much better… and I hold on to them and I celebrate them too" – Female, 55, chronic pain (Munday et al., 2021, p. 359)

1.5 Understanding and Legitimacy

" It's a world I never thought I would live in. It's a condition that you don't look sick. It's kinda like convincing the public on the same tone, but it's totally different, that the neighborhood rapist is somebody who looks like a rapist. You don't look sick so there can't be anything wrong with you or you're making it up or you've got psychological problems or you want sympathy or no, I don't want that. I don't feel well. I don't like sleeping when I need to be doing something. I don't think a lot of people even understand it, even my spouse. Because I don't look sick, don't act sick, and you know if you're a cancer patient you got a bandana around your head . . . I don't know anyone who has this condition who looks like that. You just look like the everyday person." – Female, 62, Fibromyalgia (Wheeler, 2011, p. 80)

"You see, I'm a super honest and sincere person and have been strictly brought up not to lie, not to deceive, not to steal, not to do wrong. I have a very moral family on the whole and then one has to listen to that sort of thing, that one is not believed, huh, it is so hard that it is almost the worst thing. It has been worse than the pains, actually." – Female, Fibromyalgia (Åsbring & Närvänen, 2002, p. 152)

"I tend not to discuss my illness with anyone because most people say 'what is that?' They don't believe that something can make you totally exhausted. And trying to explain it takes more energy than just leaving it alone... you start explaining it and they think you are a hypochondriac." – Female, Fibromyalgia (Schaefer, 2005, p. 21)

1.6 Support Needs and Experiences

"I do ask [my mum] for things and she'll come and she'll help me with the children, and she'll either take them out or have them stay at hers or something, so she helps me with the children a lot. I've already had complaints from my dad about my mum's always here and helping and things." – Female, 42, CFS/ME (Hannon et al., 2012, p. 8)

"I think this is a me society. I don't think we live in a culture today that really gives a lot to neighbours and family like we used to. . . My son in law says to me just last night, well you just got to deal with it mom. Thanks!" – Female, 62, Fibromyalgia (Wheeler, 2011, p. 109)

'finding people that understand and have the same experiences that you do are really what help get you through it.' – Female, Fibromyalgia (Kengen Traska et al., 2012, p. 631)

1.7 Healthcare Needs and Experiences

"It's incredible how much I've had to fight to be believed, as well as to get what I´m entitled to [...] It feels as if I'm fighting an uphill battle, being disbelieved about every single thing ." – Participant with ME (Lian & Robson, 2017, p. 7)

"At first when I queried ME, I was told by a GP it was probably depression, and even if it was ME, the cure was the same—graded exercise, CBT and antidepressants. I diligently followed this program and went from being mildly affected to losing my mobility." – Participant with ME (McManimen et al., 2019, p. 248)

"I know when I sit down and I start, the first thing that comes into a man's head is hypochondria. A woman I'd start off by saying I have fibromyalgia….but I won't do it with a man because he already knows better than I do because he's a doctor." – Female, Fibromyalgia (Acker, 2008, p. 47)

"I have been told men don't know what pain is like in women. Or told to suck it up and it is not a real thing just all in my head. Then everywhere in between. I have a friend (female) with [fibromyalgia] and people are so much more supportive including doctors. The only doctors that seem supportive to me is my family doc and specialist. Other doctors (ER especially I get the eye rolls)." – Male, 36, Fibromyalgia (Muraleetharan et al., 2018, p. 956)

"I went back to my GP who I'm fortunate is very supportive indeed…when I heard about some people's experiences with their GP I was so grateful for the fantastic GP that I'm lucky to have…because it makes a tremendous difference" – Participant with CFS/ME (Broughton et al., 2017, p. 3)

"Of course they may often think that it is something you imagine. I went to the doctor, and suddenly the pains were gone. Immediately afterwards they were back again. It is not easy to understand." – Male, Persistent Pain (Nilsen & Elstad, 2009, p. 55)

1.8 Managing Healthcare Encounters
"*You'll have to trust me," I say. "After all, I'm not sitting here paying a lot of money because I think it's fun and to pretend, but because I'm ill. I need help dammit!" Then you have to say, "Don't you get it?" Then they take you more seriously.* – Female, FM (Åsbring & Närvänen, 2004, p. 234) *"Sometimes I feel that I should look groggy, my face should be grey, and I should wear no make-up; that I perhaps appear to be too strong."* – Female, 37, Chronic Unexplained Pain (Werner & Malterud, 2003, p. 1414) *"you have to save some good face...some fragment of a good relationship and some fragment of respect for the time when you're going to have some kind of real problem that you're going to need to come back."* – Female, Fibromyalgia (Acker, 2008, p. 56) *"You have to tread rather softly; because once you antagonise them it's not certain that you are any better off. So there are in fact some comments and events that you just have to accept"* – Female, 31, Chronic Unexplained Pain (Werner & Malterud, 2003, p. 1413)

Appendix A7. Frequencies of Extracted Findings: Clinicians

Table A7

Frequency Table of Synthesized Findings, Categories and Findings reflecting Clinician Perspectives

	Finding (N)	Proportion (%)
Synthesized Finding 9: HCP attitudes towards patients		
A troublesome group of patients	8	4.06
Gender stereotypes and prejudice	4	2.03
Patients are burdened by illness	6	3.05
Synthesized Finding 10: Making sense of patients' symptoms		
The challenge of sense-making within the bio-medical model	9	4.57
Alternate explanations for patients' symptoms	11	5.58
The challenge of managing illness at the limits of medical knowledge	9	4.57
The challenge of diagnosis in the absence of objective evidence	13	6.6
Professional frustrations	7	3.55
Developing and using different sources of knowledge	8	4.06
Synthesized Finding 11: Consultation and clinical management		
Stressors related to the consultation	7	3.55
Managing the consultation structure	6	3.05
Managing process and outcome expectations	7	3.55
Naming the illness vs. iatrogenic harm	7	3.55
Course of action and balancing the costs/benefits	12	6.09
Co-responsibility for management	8	4.06
Approaches to management	5	2.54
Synthesized Finding 12: Managing the relationship		
Rewarding relationships with patients	9	4.57
Conflictual relationships with patients	4	2.03
Strategies for managing the relationship	4	2.03
Building trust and validating the patient	7	3.55
Providing reassurance and negotiating a happy medium	10	5.08
Synthesized Finding 13: Barriers and facilitators to care		
Attitudes and willingness to work with patients	6	3.05
Organizational constraints	9	4.57
Barriers to management within the consultation	8	4.06
Formal education, training, confidence	13	6.6

Appendix A8. Illustrative Quotes: Clinicians

Table A8

Experiences of HCPs: Illustrative quotes for synthesized findings

2.1 Beliefs and attitudes towards patients
"You see them, as a doctor, as very, very troublesome. They... I don't know... You find them to be querulous. They complain in expressive ways for long moments. They are fixed on themselves. It's definitely nothing that you can dismiss with a simple explanation. Everything shall be worked out down to the bottom. Yes, I suppose they have a somewhat special personality." – Female, Physician (Åsbring & Närvänen, 2003, p. 715)
"their way of life is a continual complaint… the typical patient says, 'I ache everywhere, everywhere, and I don't rest, and nothing makes it go away', and they tell you this with a lot of self-pity, I don't know, it's the way they act, it is very similar to when you are with a patient with depression…these people are also very, well very demanding, 'you have to solve this for me because as you can see I can't live this way.'" –Male, Rheumatologist (Briones-Vozmediano et al., 2018, p. 1681)
2.2 Sensemaking at the limits of medical knowledge
"We are used to clinical markers of disease in our work, usually. And here the markers are distress and suffering, and we are not trained to treat distress and suffering." – Physician (Specialist) (Hayes et al., 2010, p. 388)
"How do you teach people, in an environment of evidence-based medicine, to live with the fact that either there is no evidence, or the evidence doesn't apply in your particular patient's case?" – Family doctor (Brownell et al., 2016, p. 4)
"GPs and practice nurses want to do the best for their patients but often they don't know what to do. We bandy around things like (sleep) hygiene and pacing and lifestyle management but do we actually know what we're talking about, exactly what the narrative is in the consultation" – CFS/ME Specialist (Hannon et al., 2012, p. 7)
"I think it is frustrating…because you like to be able to help people. You like to be able to find a diagnosis and say this is what will make it better, or here is your diagnosis we can't do anything, this won't make it better, but then these people we don't know what the diagnosis is we can't do anything and that is frustrating for me as a doctor" – Consultant cardiologist (Maatz et al., 2016, p. 5)
"I have to say, I still did not completely believe in it until I have had a couple of people, one of them, my ex-mother-in law, got chronic fatigue. And I think that made it very real, because she was never someone that I would have ever have dreamt of having something like that in a million years, and you couldn't slow her down, and for her to be, not caught with it, but afflicted with it and to watch the difference in her, it suddenly becomes much more real." – Family Physician (Chew-Graham et al., 2008, p. 345)
2.3 Consultation and Management
"They simply present a flora of symptoms, and they surprise you. You can't see any structure and you think, Oh, God, I have 15 minutes to get out of this'." – Female, General Practitioner (Ringsberg & Krantz, 2006, p. 111)

"What I do for a lot of somatising patients when it's obvious what is happening is, I explain how the practice works, explain how I work as a doctor, how I come to a diagnosis, I explain how much time they have with me, and I ask them what they think it would be reasonable to do in ten minutes. (...)" – General Practitioner (Wileman et al., 2002, p. 180)

"If I actually give a name, I want it to be able to somehow help me ... It's got to help me understand where you're coming from and what your issues are .... It's got to help me with how I manage you. ... There's [also] got to be something that allows you to grow. There are some labels that, whilst it may sound helpful in understanding a process, that may give other doctors a significant misrepresentation of the person." –Supervisor (Stone, 2013a, p. 105)

"It makes you feel you're being used. You swallow and you accept it up to a certain point. You prostitute yourself. Sometimes you have no other choice. But there is a point where the patient's demands are beyond all reason. They are not medically justifiable." – Male, General Practitioner (Ringsberg & Krantz, 2006, p. 112)

"So I said, "Look. Listen to me very, very carefully. This woman somatises like mad and you must not send her to another specialist at all, ever! I am not responsible for these medications. In fact, I just cannot get her off them and she's on them as a result of having seen multiple other people, so please take that on board and do not send her to a specialist." … In hindsight, I hope that I'd handle that a little bit better nowadays." – General Practitioner (Stone, 2013b, p. 4)

2.4 The patient-clinician relationship

"A proportion make me feel quite positive and good because I actually do feel able to explain the situation to them and they are taking it on board and you know, I think that they have actually benefitted from the consultation and will go away with a more sort of, with a greater ability …and understanding to deal with it. And then there's another small minority I said are just angry people and it's pretty unrewarding and I mean I'm sure they find me unrewarding and I find dealing with them unrewarding because we don't really get anywhere." – Consultant gastroenterologist (Maatz et al., 2016, p. 5)

"I know that everyone hates it. It is, you know, the worst thing you can have at your surgery. […]We have doctors who refuse to see such patients. They say merely, ''I cannot help you, go somewhere else"" – Female, Physician (Åsbring & Närvänen, 2003, p. 716)

"They have got to feel that their own anxieties and their symptoms are being taken seriously, otherwise you are not going to get anywhere with them at all. If they are absolutely convinced that they have got something wrong with them, and you dismiss that out of hand, they'll just feel you are not doing your job properly . . . they are never going to believe you, whereas if you have got some trust there they will start to feel that bit safer." – General Practitioner (Wileman et al., 2002, p. 179)

2.5 Barriers and facilitators to care

"You can get yourself into the position where you will never spot an illness in this patient if it was staring you in the face and they were dead on the floor, because you will feel it's just their bloody somatising." – General Practitioner (Wileman et al., 2002, p. 181)

"I don't think we should be taking on psychological problems... I'd be like – I'd be looking at the clock thinking, you know, we've all got problems...get over it. And that sounds harsh." – Female, Practice Nurse (Chew-Graham et al., 2009, p. 7)

"I am very against that it is only about medicine. I am convinced we are not doing the right thing with that but we are in spiral at the minute that that is the only thing we can do in 10 minutes — "Let's give you a prescription."" – General Practitioner (Gordon et al., 2017, p. e214)

"In medical school you're taught that patients have things wrong with them and there's always a medical cause for it and you've got to try and find it." – Trainee General Practitioner (Howman et al., 2016, p. 7)

"This is a person, and even though they're a bit shabby, sweaty, whatever, doesn't matter whether you want them living next door to you or not, they're a person, so you've got to work out their context and how to help them negotiate the system. Otherwise, you're in the wrong job. Get a job as a pathologist." – Supervisor (Stone, 2014, p. 7)

" Having a satisfactory explanation to give a patient, and expressing the uncertainty I've got without necessarily rubbing them up the wrong way would certainly be a good start" – Male, Junior Doctor (Yon et al., 2015, p. 5)

Appendix B. Study Two

Appendix B2. Survey Questions

Section 1: Participant characteristics

1. Age [Free text option]
2. Gender Identity [select one]
   male/ female/ non-binary/ prefer not to say/ [free text option]
3. Highest level of education completed [select one]
   No level of schooling / primary school/ secondary school/ post-secondary school training/ third level education/ other [free text option] / prefer not to say

Please indicate whether the following statements apply to you:

4. I am a person living with a chronic, poorly understood condition
   Yes/No*
5. I know someone with a chronic, poorly understood condition
   Yes/No
6. I have a relative or significant other with a chronic, poorly understood condition [select one]
   Yes/No

**If answer to Question 4 is 'Yes'*

7. What condition do you live with? [select all that apply]
   Fibromyalgia or Chronic Pain
   CFS/ME
   Other- Physical [free text]
   Other- Psychological [free text]
8. What are your main symptoms? [select one]
   Physical Symptoms (e.g. pain, fatigue)
   Cognitive Symptoms (e.g. brain fog)
   Affective Symptoms (e.g. low mood)
   Other [free text]
9. Do you have any medically unexplained symptoms? [select one]
   Yes/ No/ unsure
10. How severe are your symptoms *most* of the time? [Likert scale]
    1 [mild] — [moderate] —[severe] 3
11. Rate the impact of your symptoms on your overall quality of life [Likert scale]
    1 [no impact] – [severe impact] 5
12. Have you received a formal diagnosis?
    Yes/ No

Section 2: Health Literacy

Please indicate how strongly you agree or disagree with the following statements:

Likert Scale

[1] Strongly disagree---------strongly agree [5]

1. I consider myself to be knowledgeable about topics related to health and wellbeing.
2. I consider myself to be health literate- I can obtain, read, understand, and use healthcare information to make appropriate health decisions and follow instructions for treatment.
3. I am aware of the various health services available to me.
4. I have a good level of knowledge and understanding of common illnesses (e.g. cold and flu)

5. I have a good level of knowledge and understanding of common chronic illnesses (e.g. diabetes, heart disease)

Section 3. (a) ME/CFS

1. I have heard about 'Chronic Fatigue' before.
   Yes/No

2. I have heard about Chronic Fatigue Syndrome/Myalgic Encephalomyelitis (ME/CFS) before.
   Yes/No*

   If Q2 is No, end survey or proceed to FM section*

3. How common is ME/CFS?
   1 Very rare-------- Very common 5

4. What, in your opinion, is the primary cause of ME/CFS? [choose one]

   Biological/psychological/social/unsure

Likert Scale

[1] very poor ---------Very good[5]

5. How would you rate your overall understanding of ME/CFS?
6. How would you rate your understanding of the causes of ME/CFS?
7. How would you rate your understanding of how ME/CFS is diagnosed?
8. How would you rate your understanding of the treatment of ME/CFS?
9. How would you rate your understanding of the management of ME/CFS?
10. How would you rate public knowledge and awareness of ME/CFS?
11. How would you rate health professionals' knowledge and awareness of ME/CFS?
12.

Please indicate how strongly you agree or disagree with the following statements:

Likert Scale questions

[1] Strongly disagree---------strongly agree [5]

13. ME/CFS is a real condition
14. People with ME/CFS are suffering
15. People with ME/CFS exaggerate their symptoms
16. People with ME/CFS are pretending
17. It is possible to cure ME/CFS
18. The symptoms of ME/CFS can be effectively managed

Compared to other chronic illnesses (e.g. diabetes, heart disease), ME/CFS is
[select one]

19. Easier to diagnose/ about the same/ harder to diagnose
20. Easier to manage/ about the same/harder to manage
21. Easier to live with/ about the same/ harder to live with

Free-text answers

22. How long, on average, do you think it takes for a patient to receive a diagnosis of ME/CFS? [short answer]
23. What, in your opinion, are the key challenges faced by individuals with ME/CFS? [free text]

Section 3. (b) Fibromyalgia

1. Have you heard about Chronic Pain before?
   Yes/No

2. Have you heard about Fibromyalgia before?
   Yes/No*

*If Q2 is No, end survey

3. How common is fibromyalgia?
   1 Very rare-------- Very common 5

4. What, in your opinion, is the primary cause of fibromyalgia [choose one]
   Biological/ psychological/ social/ unsure

Likert Scale

[1] very poor ---------very good [5]

5. How would you rate your <u>overall</u> understanding of Fibromyalgia?
6. How would you rate your understanding of the <u>causes</u> of fibromyalgia?
7. How would you rate your understanding of how fibromyalgia is <u>diagnosed</u>?
8. How would you rate your understanding of the <u>treatment</u> of fibromyalgia?
9. How would you rate your understanding of the <u>management</u> of fibromyalgia?
10. How would you rate <u>public knowledge</u> and awareness of fibromyalgia?
11. How would you rate <u>health professionals'</u> knowledge and awareness of fibromyalgia?

Please indicate how strongly you agree or disagree with the following statements:

Likert Scale

[1] Strongly disagree---------strongly agree [5]

12. Fibromyalgia is a real condition
13. People with fibromyalgia are suffering
14. People with fibromyalgia exaggerate their symptoms
15. People with fibromyalgia are pretending
16. It is possible to cure fibromyalgia
17. The symptoms of fibromyalgia can be effectively managed

Compared to other chronic illnesses (e.g. diabetes, heart disease), fibromyalgia is…
[choose one]

18. Easier to diagnose/ about the same/ harder to diagnose
19. Easier to manage/ about the same/harder to manage
20. Easier to live with/ about the same/ harder to live with

<u>Free-text answers</u>

21. How long, on average, do you think it takes for a patient to receive a diagnosis of FM? [short answer]
22. What, in your opinion, are the key challenges faced by individuals with fibromyalgia? [free text]

Appendix B3. Demographic Characteristics

Table B3

Conditions and symptoms reported by participants with chronic illness

Conditions reported by PwC [a]	Symptoms reported [b]
Abdominal Adhesions	Allergies (food-related)
Acquired Brain Injury	Anxiety
Anaemia	Brain fog
Ankylosing Spondylitis	Breathing issues
Arthritis	Confusion
Asthma	Constipation
Limp with Left-Side Body Weakness	Digestive symptoms (e.g., GERD, IBS symptoms)
Behçet's Disease	Fatigue
Benign Brenner Tumour (removed)	Inflammation
Bulging Discs (back, nerve damage)	Irritation and soreness
Cardiomyopathy	Low mood
Carpal Tunnel Syndrome	Menopause-related symptoms (e.g., low libido)
Cervical Spine Damage	Migraine
Chronic Back Pain	Muscle spasms
Chronic Lyme Disease	Mysterious viruses
Chronic Lymphocytic Leukemia (CLL)	Negative association with visual appearance to others
Chronic Myofascial Pain Syndrome	No saliva or tears
Chronic Pericarditis	Non-stop brain activity
Complex Regional Pain Syndrome (CRPS)	Overwhelming, inescapable exhaustion (both physical and mental)
Congenital Heart Conditions (hole in heart, valve leak)	Pain
Costochondritis	Panic attacks
Crohn's Disease	Poor coordination
Degenerative Arthritis	Post-exertional malaise
Degenerative Disc Disease (DDD)	Rashes (occasional)
Depression	Raynaud's
Dystonia	Skin blistering
Ehlers-Danlos Syndrome	Skin complaints
Endometrial Hyperplasia	Visual disturbances
Endometriosis	Vomiting
Excessive Thirst	
Failed Spinal Fusion	
Fluctuating High Blood Pressure	
Frozen Shoulder	

Conditions reported by PwC [a]	Symptoms reported [b]
Gastritis	
Gastroesophageal Reflux Disease (GERD)	
Hashimoto's Hypothyroidism	
Heart Failure	
High Cholesterol	
Hypermobile Joint Disorder	
Hypermobile Spectrum Disorder (HSD)	
Hyperthyroidism	
Hysterectomy at 37 Years of Age	
Idiopathic Female Edema	
Insomnia	
Irritable Bowel Syndrome (IBS)	
Lipoedema	
Long COVID	
Long QT Syndrome	
Lupus	
Mast Cell Activation Syndrome (MCAS)	
Metabolic Syndrome	
Migraine	
Migraine with Aura	
Mild Cognitive Impairment	
Mild Dysphasia	
Multiple Sclerosis (MS)	
Myoclonic Jerks	
Neuropathic Pain	
Old Injuries from Road Traffic Accident	
Osteoarthritis (OA)	
Osteopenia	
PCOS	
Peripheral Neuropathy	
Plantar Fasciitis	
Polycystic Ovary Syndrome (PCOS)	
Possible Antiphospholipid Syndrome (APS)	
Possible Multiple Sclerosis (MS)	
Postural Orthostatic Tachycardia Syndrome (POTS)	
Psoriasis	
Psoriatic Arthritis	
Raynaud's Disease	
Reactive Arthritis	
Restless Leg Syndrome	

Conditions reported by PwC [a]	Symptoms reported [b]
Rheumatoid Arthritis (RA)	
Sarcoidosis	
Sciatica	
Shoulder Subluxation	
Sjogren's Syndrome	
Sleep Apnoea	
Small Nerve Fibre Neuropathy	
Supraventricular Tachycardia	
Symphysis Pubis Dysfunction (SPD)	
Traumatic Brain Injury (TBI)	
Type 1 Diabetes	
Type 2 Diabetes	
Underactive Thyroid	
Unspecified Autoimmune Condition (under investigation)	
Urge and Stress Incontinence	
Vitiligo	

a Conditions other than Fibromyalgia or ME/CFS

b Symptoms reported other than choice of "Affective", "Cognitive", "Physical", or "All"

Note. Conditions and symptoms presented are as reported by participants to preserve integrity of responses.

Appendix B4. Descriptive Statistics

Table B4.1

Participant beliefs, knowledge and understanding of ME/CFS

	Total (N=238)		PwC (*n*=176)		PwO (n= 62)	
	n	%	*n*	%	*n*	%
Beliefs about ME/CFS						
ME/CFS is Real [a]	226	95	168	95.5	58	93
Primary cause of ME/CFS is Biological	113	47.5	88	50	25	40.3
Primary cause of ME/CFS is Psychological	23	9.7	16	9.1	7	11.3
ME/CFS is common or very common	157	65.9	127	71.3	30	48.3
Possible to cure [a]	34	14.3	20	11.4	14	22.6
Possible to effectively manage symptoms [a]	76	31.9	51	29	25	40.3
Self-rated knowledge and understanding [b]						
Overall	68	28.6	54	30.7	14	22.6
Causes	48	20.2	38	21.6	10	16.1
Diagnosis	46	19.3	35	19.9	11	17.7
Treatment	38	16	29	16.5	9	14.5
Management	45	18.9	34	19.3	11	17.7
Rating of others' knowledge and awareness [b]						
Public	2	.8	1	.6	1	1.6
Healthcare professionals	15	6.3	6	3.4	9	14.5
Challenges faced by PwME compared to other chronic illness						
More difficult to live with	171	71.8	132	75	39	62.9
More difficult to manage	186	78.2	140	79.5	46	74.2
More difficult to diagnose	203	85.3	146	83	57	91.9
Estimated time to diagnosis						
<1 Year	27	11.4	16	9.1	11	18.0
Up to 3 years	49	20.8	20	32.8	29	16.6
Up to 5 years	47	19.9	35	20.0	12	19.7
More than 5 years	28	11.8	25	14.3	3	4.9
Many	40	16.9	33	18.9	7	11.5
Don't know	45	19.1	37	21.1	8	13.1
Beliefs about PwME						
PwME Suffer [a]	231	97.1	171	97	60	96.8
PwME Exaggerate [a]	10	4.2	4	2.3	6	9.7
PwME Pretend [a]	3	1.3	1	0.6	2	3.2

[a] Reflects the number and percentage of participants who '"Agree" to this question

[b] Reflects the rating "Good or Very good"

Table B4.2

Participant beliefs, knowledge and understanding of FM, comparing participants with a *poorly understood illness (PwC) and those without (PwO)*

	Survey respondents (N=280)		PwC (*n*=195)		PwO (*n*=85)	
	n	%	*n*	%	*n*	%
Beliefs about FM						
FM is Real [a]	268	95.7	189	96.9	79	92.9
Primary cause of FM is Biological	133	47.5	93	47.7	40	47.1
Primary cause of FM is Psychological	43	15.4	27	13.8	16	18.8
FM is common or very common	213	76	169	86.6	44	51.6
Possible to cure [a]	33	11.8	17	8.7	16	18.8
Possible to effectively manage symptoms [a]	106	37.9	69	35.4	37	43.5
Self-rated knowledge and understanding [b]						
Overall	107	38.2	96	49.2	11	12.9
Causes	55	19.6	51	26.2	4	4.7
Diagnosis	71	25.4	66	33.8	5	5.9
Treatment	66	23.6	56	28.7	10	11.8
Management	69	24.6	59	30.3	10	11.8
Rating of others' knowledge and awareness [b]						
Public	8	2.9	8	4.1	0	0
Healthcare professionals	16	5.7	6	3.1	10	11.8
Challenges faced by PwFM compared to other chronic illness						
More difficult to live with	166	59.3	125	64.1	41	48.2
More difficult to manage	226	80.7	163	83.6	63	74.1
More difficult to diagnose	259	92.5	181	92.8	78	91.8
Estimated time to diagnosis						
<1 Year	28	10	12	6.2	16	18.8
Up to 3 years	73	26.1	46	23.6	27	31.8
Up to 5 years	67	23.9	45	23.1	22	25.9
5 to 10 years	34	12.1	31	15.8	3	3.5
More than 10 years	7	2.5	6	3.1	1	1.2
Many	49	17.5	41	21	8	9.4
Don't know	20	7.1	12	6.2	8	9.4
Beliefs about PwFM						
PwFM Suffer [a]	274	97.9	191	97.9	83	97.8
PwFM Exaggerate [a]	7	2.5	3	1.5	4	4.7
PwFM Pretend [a]	3	1.1	0	0	3	3.5

[a] Reflects the number and percentage of participants who "Agree"

[b] Reflects the number and percentage of participants indicating a "good or very good" level of knowledge and understanding.

Appendix B5. Exploratory Analyses ME/CFS (PwC v. PwO)

Table B5

Comparison of beliefs, knowledge and understanding of ME/CFS among PwC and PwO.

Measure	PwC (n=176)		PwO (n=62)		Chi Square Test of Independence		
	n	%	*n*	%	χ^2	*sig*	*Cramer's V*
Beliefs about ME							
Primary cause of ME/CFS					χ^2 (3)=4.34[a]	ns	
Biological	88	50	25	40.3			
Psychological	16	9.1	7	11.3			
Social	-	-	1	1.6			
Unsure	72	40.9	29	46.8			
Possible to cure					χ^2 (2)=7.79	*p*= .02	V=.18
Disagree	65	36.9	13	21			
Neither	91	51.7	35	56.5			
Agree	20	11.4	14	22.6			
Possible to manage					χ^2 (2)=4.6	ns	
Disagree	50	28.4	10	16.1			
Neither	75	42.6	27	43.5			
Agree	51	29	25	40.3			
Rating of knowledge and understanding							
Overall- own					χ^2 (2)=2.59	ns	
Poor to very poor	63	35.8	29	46.8			
Average	59	33.5	19	30.6			
Good to very good	54	30.7	14	22.6			
Rating of others' knowledge and awareness							
Public					χ^2 (2)=2.32[b]	ns	
Poor to very poor	164	93.2	54	87.1			
Average	11	6.3	7	11.3			
Good to very good	1	.6	1	1.6			
Healthcare practitioners					χ^2(2)=11.01[c]	*p*=.004*	V=.215
Poor to very poor	127	72.2	35	56.5			
Average	43	24.4	18	29			
Good to very good	6	3.4	9	14.5			

*Sig after Bonferroni Adjustment (α=.008)

[a] 2 cells (25%) have expected count less than 5.

[b] 3 cells (50%) have expected count less than 5

[c] 1 cells (16.7%) have expected count less than 5

Appendix B6. Exploratory Analyses ME/CFS (PwME v. PwC)

Table B6

Comparison of beliefs, knowledge and understanding of ME/CFS among PwME and other chronic illnesses.

Measure	Other (n=121)		PwME (n=55)		Chi Square Test of Independence		
	n	%	*n*	%	χ^2	*sig*	*Cramer's V*
Beliefs about ME/CFS							
Primary cause of ME/CFS					χ^2 (2)=17.10	*p* <.001*	V=.31
Biological	48	39.7	40	72.7			
Psychological	12	9.9	4	7.3			
Unsure	61	50.4	11	20.0			
Possible to cure					χ^2 (2)=14.19	*p* <.001*	V=.28
Disagree	37	30.6	28	50.9			
Neither	74	61.2	17	30.9			
Agree	10	8.3	10	18.2			
Possible to manage					χ^2 (2)=4.5	ns	
Disagree	29	24.0	21	38.2			
Neither	57	47.1	18	32.7			
Agree	35	28.9	16	29.1			
Self-rated knowledge and understanding							
Overall					χ^2 (2)=41.11	*p* <.001*	V=.48
Poor to very poor	54	44.6	9	16.4			
Average	48	39.7	11	20.0			
Good to very good	19	15.7	35	63.6			
Rating of others' knowledge and awareness							
Public					χ^2 (2)=3.09 [a]	ns	
Poor to very poor	112	92.6	52	94.5			
Average	9	7.4	2	3.6			
Good to very good	0	0	1	1.8			
Healthcare practitioners					χ^2 (2)=12.8[b]	*p* =.002*	V=.27
Poor to very poor	78	64.5	49	89.1			
Average	39	32.2	4	7.3			
Good to very good	4	3.3	2	3.6			

*Sig after Bonferroni Adjustment (α=.008)

[a] 3 cells (50%) have expected count less than 5

[b] 2 cells (33%) have expected count less than 5

Appendix B7. Exploratory Analyses FM (PwC v. PwO)

Table B7

Comparison of beliefs, knowledge and understanding of FM among PwC and PwO.

Measure	PwC (n= 195)		PwO (n=85)		Chi Square Test of Independence		
	n	%	*n*	%	χ^2	*sig*	*Cramer's V*
Beliefs about FM							
Possible to cure					χ^2 (2)=22.18	$p < .001$*	V=.281
Disagree	96	49.2	17	20			
Neither	82	42.1	52	61.2			
Agree	17	8.7	16	18.8			
Possible to manage					χ^2 (2)= 14.82	$p < .001$*	V=.230
Disagree	74	37.9	13	15.3			
Neither	52	26.7	35	41.2			
Agree	69	35.4	37	43.5			
Rating of knowledge and understanding							
Overall- own					χ^2 (2)=67.25	$p < .001$*	V=.490
Poor to very poor	18	9.2	43	50.6			
Average	81	41.5	31	36.5			
Good to very good	96	49.2	11	12.9			
Rating of others' knowledge and awareness							
Public					χ^2 (2)=5.04[a]	ns	
Poor to very poor	175	89.7	76	89.4			
Average	12	6.2	9	10.6			
Good to very good	8	4.1	9	10.6			
Healthcare practitioners					χ^2(2)=26.39[a]	$p < .001$*	V=.307
Poor to very poor	143	73.3	36	42.4			
Average	46	23.6	39	45.9			
Good to very good	6	3.1	10	11.8			

*Sig after Bonferroni Adjustment (α=.01)

[a] 1 cells (16.7%) have expected count less than 5

Appendix B8. Exploratory Analyses (Demographic)

Table B8

Association between rating of HCP knowledge and understanding of FM and demographic variables.

Variable	Chi Square Test of Independence
Know others with chronic illness	$\chi^2(2)= 1.22^a$, $p=.54$
Have relative with chronic illness	$\chi^2(2)= 4.09^b$, $p=.13$
Have a diagnosis	$\chi^2(2)= 9.95^b$, $p=.007$*, Cramer's $V= .226$
Presence of multiple conditions	$\chi^2(2)=.225^b$, $p=.96$

*Sig after Bonferroni Adjustment (α =.01)

[a]1 cells (16.7%) have expected count less than 5

[b] 2 cells (33.3%) have expected count less than 5

Appendix C. Study Three

Appendix C1. Ethical Approval

Coláiste na Tríonóide, Baile Átha Cliath
Trinity College Dublin
Ollscoil Átha Cliath | The University of Dublin

F.A.O. Natalia Duda

Approval ID: SPREC032022-11

School of Psychology Research Ethics Committee

19th May 2022

Dear Natalia,

The School of Psychology Research Ethics Committee has reviewed your application entitled "The healthcare journeys and experiences of individuals with medically unexplained symptoms and poorly understood syndromes", and I am pleased to inform you that it was approved.

Please note that you will be required to submit a completed Project Annual Report Form on each anniversary of this approval, until such time as the research is complete and the thesis is submitted. The form is available for download from the Ethics section of the School website.

Adverse events associated with the conduct of this research must be reported immediately to the Chair of the Ethics Committee.

Yours sincerely,

Richard Carson
Chair,
School of Psychology Research Ethics Committee

Scoil na Síceolaíochta
Dámh na nEolaíochtaí Sóisialta agus Daonna,
Áras an Phiarsaigh,
Coláiste na Tríonóide,
Baile Átha Cliath,
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Appendix C2. The Life Grid

Figure C2 Sample Life Grid constructed during an interview

Participant Code: ___SAMPLE_____

Year	Age	Significant Life Events	*What was your life like at the time?*		*What was your health like at the time?*	
			Family & Friends	Work & Leisure	Health/Illness	Healthcare Journey
1980	10		Mother was grieving Didn't have much support	School was very good- was recommended for gymnasium. Began to struggle socially and academically	Stomach ulcer, bleeding Very stressed Sick all the time- flu like symptoms	Hospital for stomach bleeding- a week Not really figuring out what was happening
		Mother had a partner	Partner passed away 2 years after father- witnessed the heart attack	Struggling in school	Ongoing health issues	GP
1984	14	Mother remarried	Moved to house of partner-village, had 3 sons Difficult situation at home for next years- parents fighting for about 15 years	New social circle	Constant flu-like symptoms- symptoms just changed with time	GP
	15 / 16 years old		School /friends difficulties due to acne	Hated primary school Secondary school- missed majority of 5th/6th Had a supportive teacher in secondary school	2nd year in school Cystic Acne (around 15/16 years old) Not feeling well one day- probably a cold? Around age 15/16 went to A&E Pain started Losing power Tiredness	GP-→dermatologist for acne Months on wait, went privately A&E and admitted to hospital - Meningitis? Glandular fever? - Epileptic unit - Mini-stroke? Told to go to physio and hope for the best,
1990	20 / 21 years old	parents killed by drunk driver became 21 but did not celebrate	Life changed. Aside from brother, siblings supported each other; rest of family didn't really help	Was working when found out about crash	Movement	

Note. Sample is based on a combination of participants' responses.

Appendix C3. Participant Demographics

Table C3

Health conditions among the participant sample (N=12)

Disclosed condition	*n*
Fibromyalgia	5
Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome	4
Functional Neurological Disorder	3
Chronic Back Pain	2
Chronic Migraine	2
Hypermobile Ehlers-Danlos Syndrome/ HSD	3
Long COVID	2
PCOS	2
Postural Orthostatic Tachycardia Syndrome	2
Arthritis	1
Chronic Fatigue	1
Chronic Pain Syndrome	1
Chronic Regional Pain Syndrome	1
Dystonia	1
Gallstones	1
Gastroparesis	1
Gynaecological Issues	1
Inappropriate Sinus Tachycardia	1
Insulin Resistance	1
Lyme Disease	1
Neurocardiogenic Syncope/Vasovagal Syncope)	1
Osteoarthritis	1
Overactive Bladder	1
Sinus Tachycardia	1
Spinal Condition- rare	1
Supraventricular Tachycardia	1

Appendix C4. Patient Journey Maps

Figure A.

Participant 2 Journey

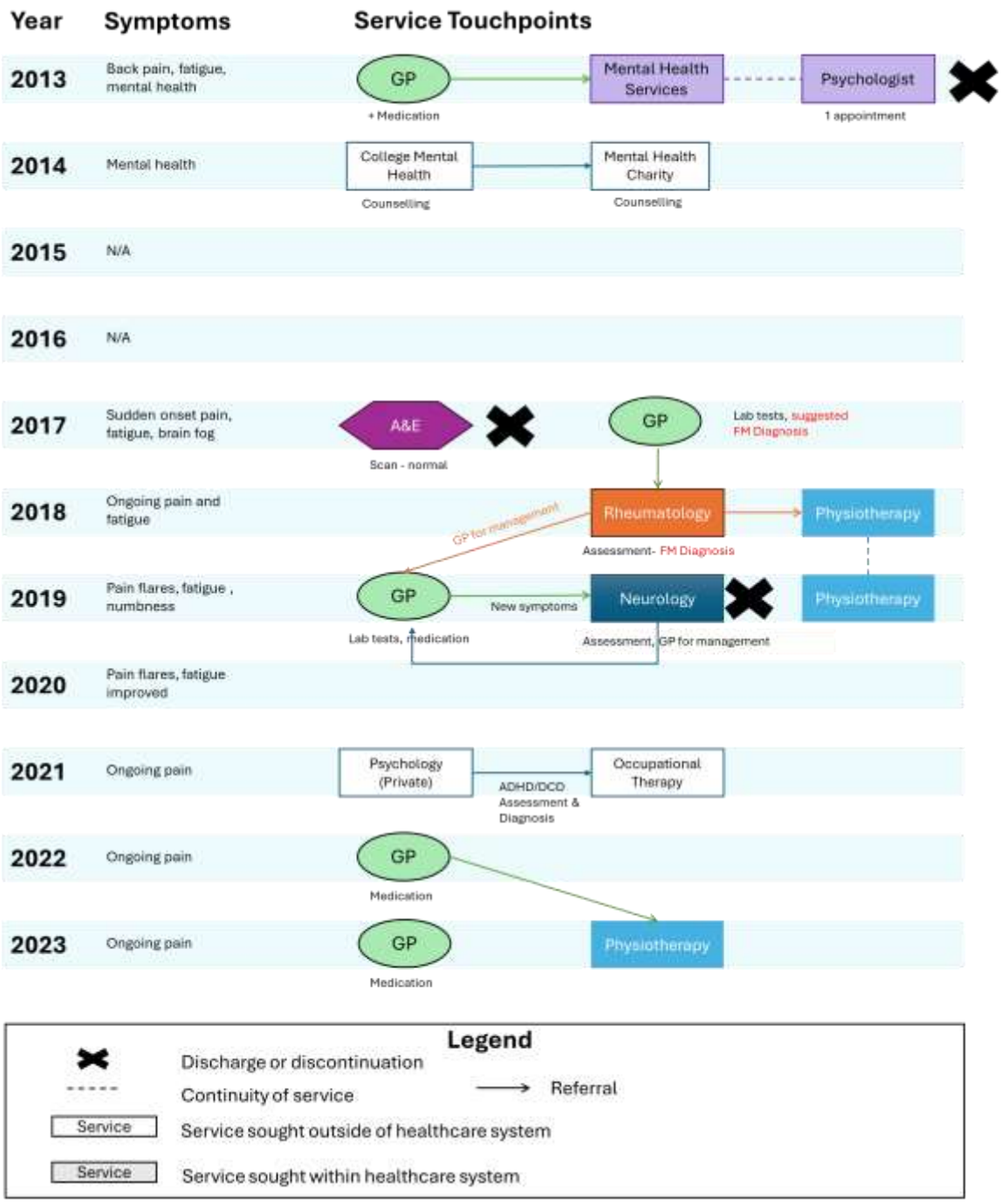


Figure B.

Participant 5 Journey

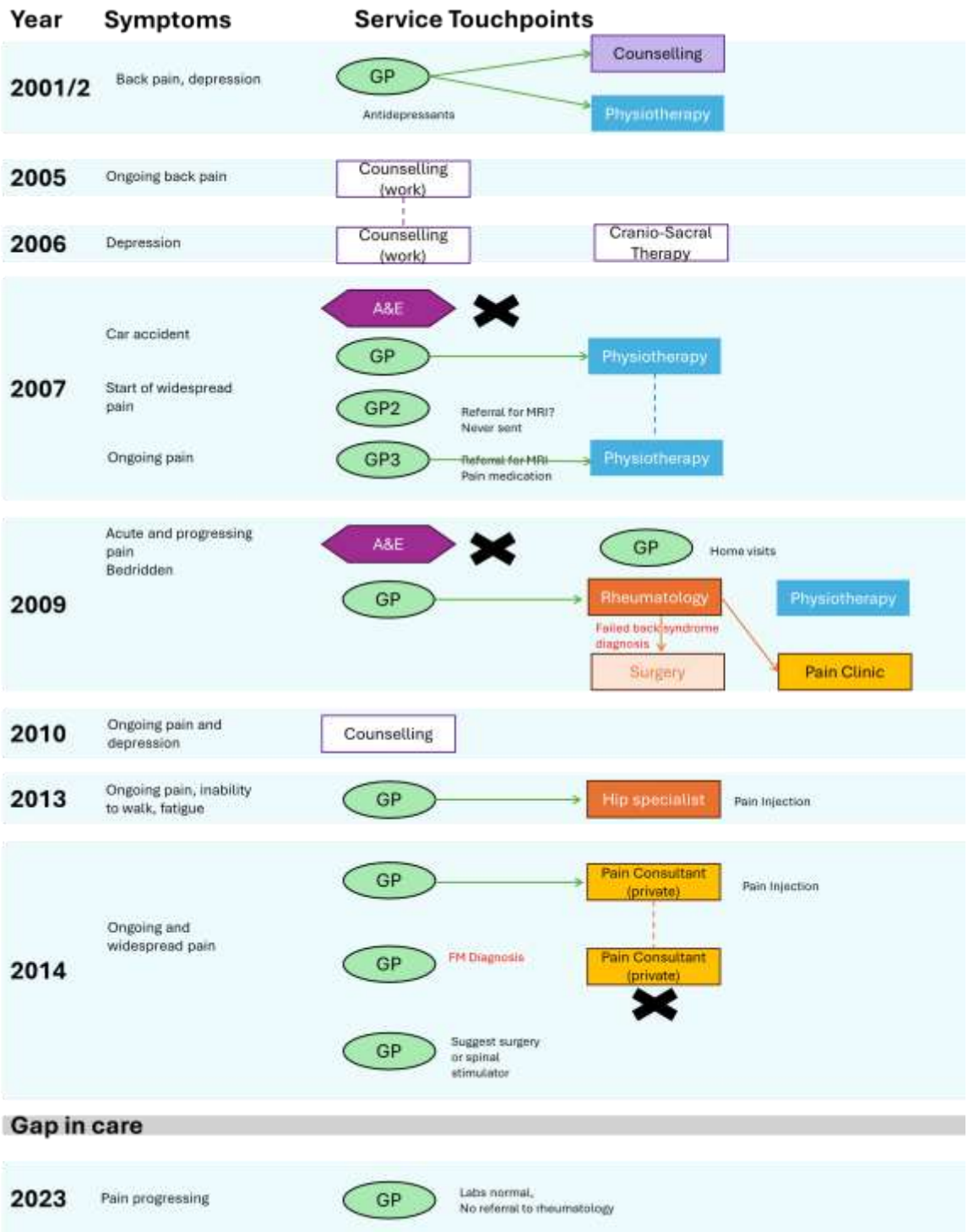


Figure C.

Participant 10 Journey

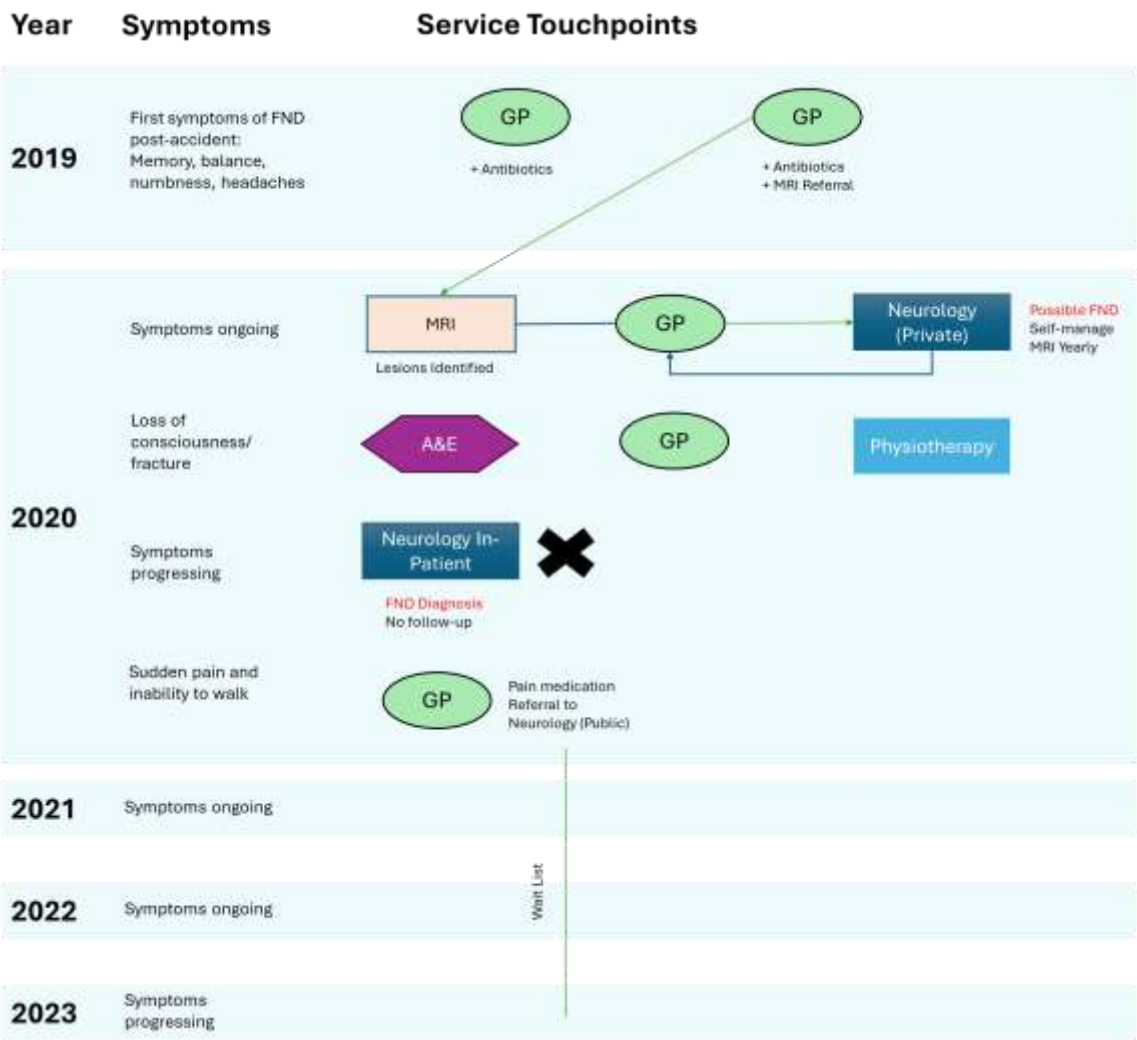
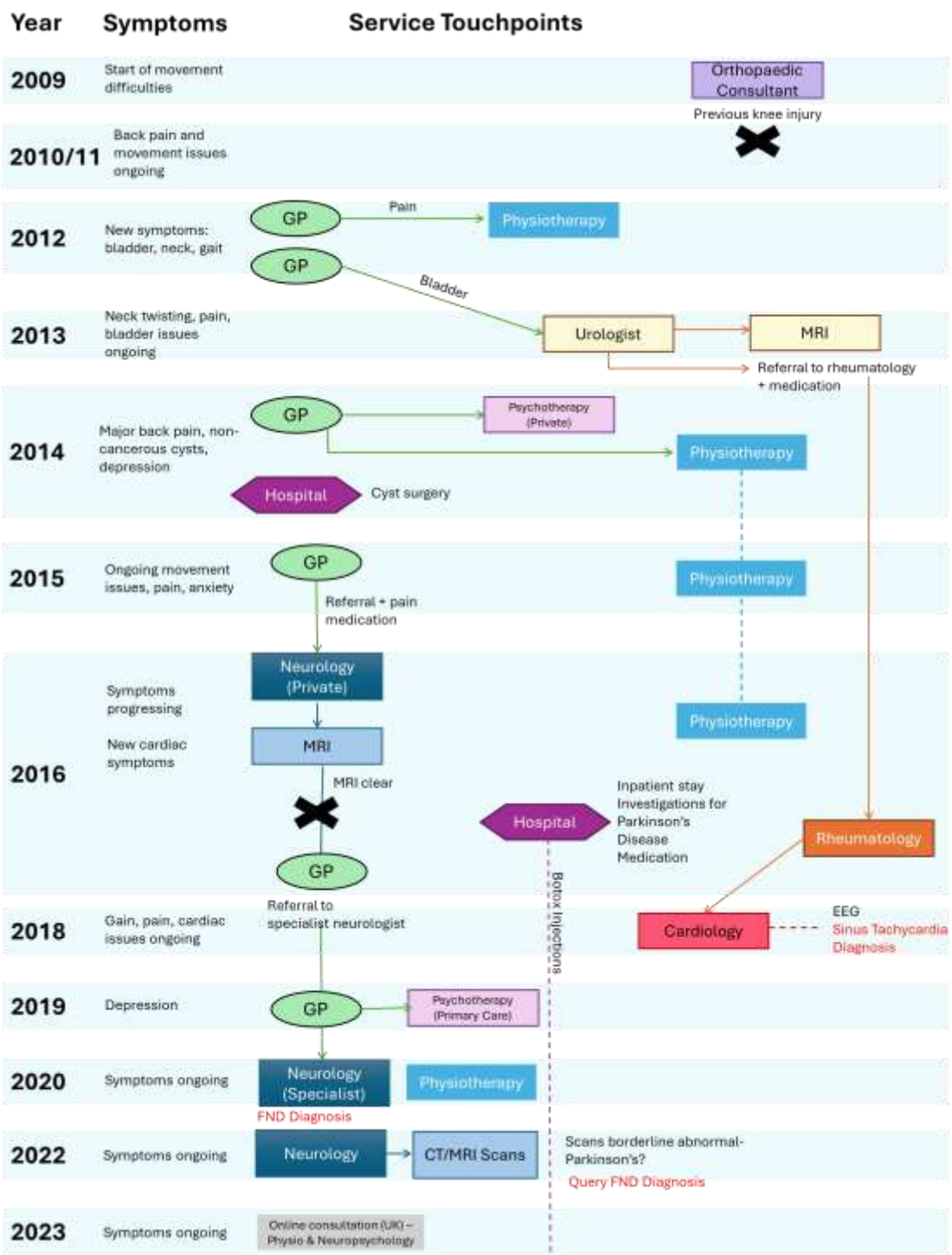


Figure D.

Participant 7 Journey



Appendix D. Study Four

Appendix D1. Study Four Ethical Approval

Coláiste na Tríonóide, Baile Átha Cliath
Trinity College Dublin
Ollscoil Átha Cliath | The University of Dublin

F.A.O. Natalia Duda

Approval ID: SPREC032022-11

School of Psychology Research Ethics Committee

19th May 2022

Dear Natalia,

The School of Psychology Research Ethics Committee has reviewed your application entitled "The healthcare journeys and experiences of individuals with medically unexplained symptoms and poorly understood syndromes", and I am pleased to inform you that it was approved.

Please note that you will be required to submit a completed Project Annual Report Form on each anniversary of this approval, until such time as the research is complete and the thesis is submitted. The form is available for download from the Ethics section of the School website.

Adverse events associated with the conduct of this research must be reported immediately to the Chair of the Ethics Committee.

Yours sincerely,

Richard Carson
Chair,
School of Psychology Research Ethics Committee

Scoil na Síceolaíochta
Dámh na nEolaíochtaí Sóisíalta agus Daonna,
Áras an Phiarsaigh,
Coláiste na Tríonóide,
Baile Átha Cliath,
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Faculty of Arts, Humanities and Social Sciences,
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Appendix D2. Study Four Questionnaire

Section 1. General information

1. Your age [free text]
2. Gender Identity [select one]
   male/ female/ non-binary/ prefer not to say/ Other [free text option]
3. Highest level of education completed [select one]
   No level of schooling / primary school/ secondary school/ post-secondary school training/ third level education/ other [free text option]
4. Employment [select one]
   Currently employed (part-time)/ Currently employed (full-time)/ Currently unemployed/ Never employed/ Other [free text option]

Section 2. Health Journey

5. What condition(s) do you live with? [Free text]
6. What are your main symptoms? [select as many as apply]
   Physical Symptoms (e.g. pain, fatigue)
   Cognitive Symptoms (e.g. brain fog)
   Affective Symptoms (e.g. low mood)
   Other [free text]
7. How severe are your symptoms most of the time? [Likert scale]
   1 [mild] — [moderate] —[severe] 3
8. Rate the impact of your symptoms on your overall quality of life [Likert scale]
   1 [no impact] – [severe impact] 5
9. What age were you when your symptoms first started? [free text]
10. What age were you when you first sought medical advice for your symptoms? [free text]
11. Have you received a formal diagnosis? [choose one] Yes/No
12. If YES, How long (approximately) did it take for you to receive a diagnosis? [free text]
13. Approximately how many services or specialists have you seen in relation to your condition/symptoms? [free text]
14. What service(s) have you used the most? (e.g. GP, physiotherapy) [free text]
15. What service(s) have been the most useful to you? (e.g. GP, physiotherapy) [free text]
16. How satisfied are you with the quality of healthcare services currently available to you? [Likert Scale] 1 [Completely dissatisfied] ---7 {Completely satisfied]
17. Overall, how satisfied are you with your healthcare journey to date? [Likert Scale]
    1 [Completely dissatisfied] ---7 {Completely satisfied]

Appendix D3. Participant Demographics

Table D3

Chronic illnesses and symptoms reported by focus group participants

	Focus Group				
	1	**2**	**3**	**4**	**5**
Conditions Reported					
Number Reported in Group	15	8	8	42	13
Condition (n participants)	Fibromyalgia/ Chronic Pain Syndrome (n=2) ME/CFS(n= 2) Long-Covid (n= 2) Lyme Disease (n= 2) Ehlers-Danlos Syndrome (n= 1) POTS(n= 1) Other Pain: Bladder Pain Syndrome (n= 1) Skin: Eczema (n=1) Allergies (n= 1) Cancer (n= 1)	ME/CFS(n= 2) POTS (n= 2) Lyme Disease (n=2) MCAS (n=1) Fibromyalgia/ Chronic Pain Syndrome (n=1) Long-Covid (n= 1) Coeliac Disease (n=1) Paroxysmal atrial fibrillation (n=1)	Fibromyalgia/ Chronic Pain Syndrome (n=3) ME/CFS (n=1) MCAS (n=1) Asthma (n=1) Diabetes (n=1) Osteoarthritis (n=1) Iron deficiency (n=1) Thyroid: Hyper (n=1)	Fibromyalgia/Chronic Pain Syndrome (n=3) Migraine (n=3) POTS (n=2) EDS (n=2) Bronchiectasis (n=2) ME/CFS (n=1) Long Covid (n=1) Other Pain: Chronic Pelvic Pain (n=1) Other Pain: Centralized Pain Processing Disorder (n=1) MCAS (n=1) GERD (n=1) Coeliac Disease (n=1) IBS/IBD (n=1)	ME/CFS (n=1) Spastic Paraplegia Type 1 (n=1) Osteoporosis (n=1) Sjogren's Syndrome (n=1) Thyroid: Hypo (n=1) Migraine (n=1) Dysmenorrhea (n=1) Pain: Lower back (n=1) Allergies (n=1) Tarlov Cyst (n=1) Ovarian Cyst (n=1) Hereditary neuropathy with liability to pressure palsies (HNPP) (n=1) IBS (n=1)

	Focus Group				
	1	**2**	**3**	**4**	**5**
	Skeletal: Scheuermann's Disease (n= 1) Autism (n=1) ADHD (n=1) Arrhythmia (n=1) Sjogren's (n=1)			Lupus (n=1) Lipoedema (n=1) Rheumatoid Arthritis (n=1) Psoriatic Arthritis (n=1) Osteoarthritis (n=1) Osteopenia (n=1) Osteoporosis (n=1) Psoriasis (n=1) Hypertension (n=1) Reynaud's Syndrome (n=1) Gastroparesis (n=1) Skeletal: Instability (n=1) Tethered cord syndrome (n=1) Autonomic dysfunction (n=1) Von Willebrand Factor 1 (n=1) Skin: DSAP (n=1) MDD (n=1) GAD (n=1) Insomnia (n=1) Allergies (n=1) Asthma (n=1) ILD pneumonitis (n=1) Degenerative disc disease (n=1) Radiculopathy (n=1) Cushing's Syndrome (n=1) Nerve damage (post-injury) (n=1)	

	Focus Group				
	1	**2**	**3**	**4**	**5**
				HPV (n=1) Adrenal insufficiency (n=1) pituitary adenoma (n=1)	
Other Symptoms (n participants)					
	Sensitivity (temperature, light) (n=2) Fatigue (n=1) Swelling (n=1) Post-exertional malaise (n=1)		Gastrointestinal symptoms (n=1) Food intolerances (n=1)	Chronic fatigue (n= 2) Chronic Pain (n=1) Post-herpetic itch/pain (n=1) Gastric (other) (n=1) Neurological (n=1) Post-exertional malaise (n=1) Sensitivity (multiple) (n=1)	Gastric (n=1) Sensitivity (light) (n=1) Post-exertional malaise (n=1)

Appendix D4. Sociogram Word Contribution

Table D4

Word counts of focus group participants

Participant		Group Contribution	
Number	Pseudonym	Total Words	%
Focus Group 1			
1	Millie	2591	19%
2	Amy	3304	24%
3	Sarah	3573	26%
4	Julia	2041	15%
5	Chloe	1453	11%
Moderator		775	6%
Focus Group 2			
1	Anna	3371	35%
2	Ellen	1778	19%
3	Alex	2353	25%
4	Emily	1438	15%
Moderator		651	7%
Focus Group 3			
1	Sophie	5798	52%
2	Maureen	1553	14%
3	Liam	1880	17%
4	Ashley	1147	10%
Moderator		779	7%
Focus Group 4			
1	Niamh	1234	9%
2	Kelsey	1173	9%
3	Robert	1450	11%
4	Laura	1094	8%
5	Caitlyn	2311	18%
6	Amber	1627	12%
7	Mia	2442	19%
8	Lucy	1462	11%
Moderator		393	3%
Focus Group 5			
1	Shane	2976	29%
2	Caroline	2398	23%
3	Dawn	4252	42%
Moderator		583	6%

Note. Total words : Focus Group 1 =13737, Focus Group 2= 9591, Focus Group 3= 11157, Focus Group 4= 13186, Focus Group 5= 10209

Appendix D5. Framework Charting

Table D5.

Framework Charting Matrix

	Focus Group				
	1	2	3	4	5
Category 1. Experience of Living with a Chronic Illness					
Burden of Illness or Disease	Description of cognitive and physical impact (Cognitive issues, fatigue, loss of physical activity, and difficulty managing daily tasks (e.g., hair care); impact on social, personal and work life (e.g. isolation, marriage breakup) and decisions impacted by illness (e.g. not having children). Regular crises with overbearing pain; managing symptoms causing distress. People	All participants describe burden of illness/disease- extensive impact on daily tasks, cognitive abilities/communication, ability to socialize, disabling symptoms, sensory overload. All agree that it is unnecessarily expensive (financial impact).	Strong component. Participants discuss various hardships due to symptoms - examples of how illness affects life e.g., work, struggles of lone parenting, impacts on ADLs. Experience of loss- life/achievements, future, control over the body/weight. Strong theme around mental health consequences of chronic illness- becoming anxious, stressed as a result of	Mention of having to continue to function in life despite symptoms, in particular pain. Loss of mobility and impact on social life. Impact on mental health (e.g., feeling miserable) Illness and comorbid mental health diagnosis can be very difficult.	Description of physical impact (loss of ability to participate in social life, difficulty with daily tasks), social life impact (isolating condition), psychological impact (breakdown, suicide watch, self-doubt especially because of not being believed). Impact on career and financial consequences of working less and having costs due to illness management.

	Focus Group				
	1	2	3	4	5
	with different conditions but similar experiences.		illness/its impacts and interactions with healthcare. Participants discuss avoiding the topic due to mental health stigma.		
Coping with and Managing	Description of lifestyle adjustments and need to pare back on life; efforts to manage and cope with symptoms (e.g., logging symptoms, finding unusual ways to cope) trying different/alternative treatments and therapies or home-made solutions. Adjusting life to illness (e.g. choice of job that accommodates physical limitations). Descriptions of managing or coping with psychological and emotional challenges (e.g. self-blame and self-doubt). Doing own research about condition and management. Patients have to find ways to cope because they are	Participants described efforts at adjusting their lives to make managing illness easier- both in terms of own activities (learning to pace) and adjusting others' expectations (e.g. about not being able to socialize). Learning to cope is a very difficult process- participants describe struggling to adjust/come to terms with limitations and fighting against them. Making efforts to prevent crashes means cutting out activities that are not necessary. Making efforts to learn about condition/ find information (from books, online groups, clinicians) and use of	Strong component. Having to take things into own hands is a major theme. Participants are let down by system/clinicians and have to do own research/ figure out how to cope ('you have to become your own GP'). They develop their own ways of managing (you have to focus on the symptoms) and shape their life around the illness (cutting down on socializing, staying busy to avoid dwelling on it).	Journey from denial to acceptance and finding resources to deal with the condition, mental health impact especially with prior mental health diagnosis. Patients have to manage their energy and limit some activities (e.g., number of calls), their diet (e.g., to prevent flares). Mentions of self-educating a lot.	Description of a psychological journey to learn acceptance, persistence and positivity. Mention of lifestyle changes like buying equipment (e.g., cars, electric bikes, etc.) or hiring help. Description of symptoms management like pain through active techniques (over passive) such as yoga or tai chi.

	Focus Group				
	1	2	3	4	5
	left to figure it out. Also hiding illness from others.	alternative therapies and technology for pacing (e.g., smart watches and monitoring heart rate).			
Positive Outlook					
	Only positive that is implied is developing creative ways of managing symptoms (i.e., becoming resourceful)	Some participants describe a positive outlook- staying hopeful about possibility of recovery and improvements in medical treatment/research in the future, particularly post-pandemic.	Only positive implied is feeling like they can contribute to research/help others.	Mention of feeling of hope when seeing that good experiences exist. Finding quality resources for understanding their condition, life management, and medical literacy.	Some description of feeling stronger and more positive
Community and Social Support					
V*alue of Community*					
	Support networks and advocacy groups provide learning opportunities and support as well as friendship. Gaining knowledge through the experiences of others; learning about treatments and strategies. Other patients are most helpful. Use of	Patients value groups (seeing others improve on those groups gives them hope- would be "lost at sea " without them)- emphasis on meeting people who are similar and supportive. Online webinars are a good resource. By contrast, there is mention of an absence of	No reference	Support groups are very important as patients make friends there, receive information and support (e.g., about their rights). Support groups are also a place for action and advocacy. Learning about having a choice.	Empowerment from sharing and receiving information/knowledge from the support groups. Peer support as a valued support.

	Focus Group				
	1	2	3	4	5
	courses on pain management.	community supports (offline).			
Positive interpersonal Experiences					
	Supportive relationships with friends and family. Encouragement and support (emotional and practical) from partners, family members. Accommodations made in school (e.g. PE exemption)	Weak description of support from friends (e.g., close friend as a confidant) and 'help' from others. Some experience of an understanding employer. There seems to be more understanding post-pandemic.	Minor- one example of speaking to a friend and getting advice to ask about a specific test.	Family member helped push to action (e.g., see a psychologist).	Supportive family member once living a similar experience.
Negative Interpersonal Experiences					
	Suggestions of negative experiences broadly- e.g., isolation; people think it is just a joke (with regards to bathroom trips)	Most participants describe negative experiences with family and social others- lack of support, awareness and knowledge about unexplained illness (because people can't see it, they don't believe you). Description of losing friends and family; family attempting to have participant admitted into a psychiatric facility. Help withdrawn (seen as 'enabling'). Statement that people will always question these	Some discussion of negative experiences with others, particularly feeling let down by friends and family (ignored, disbelieved, no help, not really understood), experiences of gaslighting (about MH issues), feeling judged for lifestyle adjustments (e.g., a strict diet) .Having to hide mental health impacts (e.g. feeling anxious/stressed) because you're not supposed to show struggles/how you really	Brief mentions of the journey being an isolating experience and lacking support from others.	Lack of belief and understanding at work pushing patient to resigning

	Focus Group				
	1	2	3	4	5
		conditions. People won't or can't understand until they are affected themselves.	feel. Clinicians can contribute to family behaviour (turn people against you).		
			Category 2. Healthcare Experiences		
Experiences with healthcare					
Positive experiences (Clinicians)					
	Descriptions of clinicians who understand, believe and are empathic towards patients' conditions, who are proactive, take time to explain diagnoses, help with navigating the healthcare/welfare system. This can occur when a clinician has no answer explanation for the patients' symptoms. Positive experiences are marked by clinicians who listen to patients' insights, admit mistakes and follow up on care. Clinicians who are genuinely interested in a problem outside of their area of expertise.	Strong component. Most describe some positive experiences with clinicians- e.g., good/explicit advice, transparent and apologetic (when not knowing answers), high effort (e.g., being thorough/attentive, a 'brilliant' GP who did everything he could and didn't give up on patient). Some references to "good" consultant who makes sure people have access to testing (for Lyme). Experience of being heard and involved in decision-making. One participant attributes recovery to clinician ('she	Examples of positive experiences are described in the context of a mostly negative journey (i.e., it's been bad, but it's not all bad, some doctors are nice). Description of excellent treatment when there is a readily identifiable issue (on bloodwork) that could be fixed. Positive experiences include attentiveness (feeling that the clinician is thorough, takes time to figure things out) and clinician learning from patients (admitting to and apologizing for mistakes). Emphasis that clinicians are not bad people- some	Description of clinicians who communicate honestly about their lack of knowledge. Positive experiences where patient felt at home (hospital in Spain), believed, listened to, with clinicians presenting good knowledge and listening to advocates' recommendations	Description of clinicians who listen and demonstrate empathy towards patients' conditions, who demonstrate a baseline knowledge of the condition, communicate transparently, and try to come up with solutions.

	Focus Group				
	1	2	3	4	5
		saved me', support stopped her from becoming depressed). Some mention of positive experiences that were ultimately useless (nothing done in the end, no follow through).	are genuinely nice, but lack knowledge, training or skill.		
Positive experiences (Other professionals)					
	Some mention of positive experiences. E.g., physiotherapist being responsive (not continuing due to patients' pain levels)	Weak component. Some mention of good experience with alternative therapies (e.g., ozone).	Participants highlight that positive experiences with healthcare are due to other staff (not clinicians), particularly nurses. Nurses take time to listen and try to help patients despite time pressures; are compassionate and empathetic. Positive interactions are very helpful particularly when there are deficits in an initial consultation. Learning about bedside manner from nurses would be beneficial.	Physiotherapists often described positively: communication equal to equal, transparency, clear pathways, proactive care and communication with other clinicians.	Description of a physio who was transparent about their powerlessness.
Negative experiences (Clinicians)					
	Strong component. Many descriptions of lack of listening or	Strong component. All participants described having and gave examples	Strong component. Main pain point is not being listened to or heard, All	Descriptions of not believing or listening to the patients, therefore	Descriptions of failure around the lack of knowledge and failure to

	Focus Group				
	1	2	3	4	5
	understanding and no want to try and understand further. 'Not being believed' is a major code. dismissal and disregard of patient insights and needs; doing more harm than good; gaslighting behaviour; ill-suited advice (have a baby). Failure to consider patients' medical history. Dismissive attitudes regarding treatment and painful/unsuitable tests. Not 'bothering' to read a medical history before an appointment.	of negative experiences with clinicians, primarily related to assumptions/biases and not being taken seriously. Example of being thrown in the "Mad-Bad-Sad Basket", dismissed as having mental health issues, being told symptoms are anxiety, being told to "find a willie and have sex" (complaint made), no listening skills despite training. Experiences of being questioned, doubted or undermined. Having to fight for basic tests and support (e.g. sick leave). Misattribution of symptoms (to FM, when it was Lyme). Gaslighting of children and teenagers.	participants described having negative experiences with doctors- Describe appointments as traumatic (being laughed at, dismissed); Clinicians are only willing to look at one issue at a time; are in a rush to prescribe and get you out the door. You feel like you are wasting their time. They refuse to do basic tests if a patient asks for them and don't bother to read your medical history. Clinician behaviour is a source of more anxiety on top of symptoms- you have to build up the courage to go back to them.	mislabelling them, giving them wrong referrals, interpreting their symptoms as psychological, giving them meds without checking for potential interactions. Clinicians regularly give unprofessional comments on patient's personal life or give negative and infantilising labels. Clinicians lack knowledge but refuse to admit it, therefore do not follow guidelines.	admit it, notably when designated specialists. Instances of misdiagnosis and subsequent dismissal. Common gas lighting, failure to believe the patients, and misguidance.
Negative experiences (Other professionals)					
	No reference to negative experiences with allied health; only in relation to consultants, specialists/ 'experts in the field' and GPs	Weak component. One description of insufficient help from OT (dismissed symptoms as 'in her head')	No reference	No reference	No reference

	Focus Group				
	1	2	3	4	5
Negative experiences (unsuitable treatments/therapy)	Reports of treatments/therapies with negative consequences (e.g. GET) or those which are not suitable for the patient (unadopted to their abilities, e.g. Stress testing when a patient is saying No). Trial and error approach. Medications/treatments causing side effects (e.g., being on multiple antibiotics) Painful procedures when simpler solutions are available. Being told to come back 'if you're suicidal'.	Negative experiences with tests (expensive), issues around screening (silly questionnaires) and common-sense advice (patronising cartoons about time management). Specialised diets that were difficult to follow or had negative consequences (high cholesterol as a result). Mention of "charlatans"	Example of being recommended exercise or unsuitable treatment as a catch-all (despite having expressed that these will not work). Lack of consideration for patients' ability (told to exercise when she couldn't peel a potato). Talk of a 'trial and error' approach- try a medication and see what happens.	Descriptions of bad experiences with tendency to systematically medicate, exercise therapies, surgery. Patients distrust some treatments and consider them to be prescribed for financial gain.	Description of a tendency to medicate patients ('like smarties') and in particular antidepressants despite no diagnosis of depression
Power dynamics	Issues related to power dynamics are described. E.g. doctor saying "who is in charge here"; being discharged and accused of non-compliance when challenging a clinician or treatment;	No description of power imbalance or dynamics.	Issue of power is dominant; topics of control and disempowerment (of patients). There is a sense that clinicians feel superior/better than other people and look down on their patients (disregard their	Description of 'appalling' and unbalanced dynamics where patients are feeling vulnerable and are not in the loop for referrals and data. Doctors discount self-education (Rd. Google) and patient expertise (feel threatened).	Doctor's ego and a need to "know better" than the patient prevents them from listening to the patient and their expertise. They prefer pretending they know due to their status than acknowledging they don't. Descriptions of

		Focus Group		
1	2	3	4	5
		skills/profession); they act like they are special (e.g., using better PPE/masks than 'normal' people). Doctors treat patients like they're stupid (patients don't or won't understand things they read); intelligence is undermined. They seem to have their own agenda and are focused on following it (e.g., dry would not test for anaemia but seemed obsessed about pre-diabetes). Clinicians don't have the answers- but don't want you to research ('they don't want to help you. But then they don't want you to look'). They disregard or belittle information patients have gathered on their conditions (Rd. Google) but then clinicians end up Googling things themselves in front of you. Status of medical knowledge vs lived		power dynamics in relation to the passive vs. active patient- passive is rewarded and expected.

	Focus Group				
	1	2	3	4	5
			experience (' I went to medical school'). Power imbalance is clear in the expression of anger- doctors are allowed to show anger/frustration but patients are not.		
The patient journey					
Negative experiences of the journey					
	Lengthy and suboptimal journeys (Primarily negative); lack of continuity, being bounced around, limited options for care, long waiting times and lack of resources for management. Symptoms are often dismissed as psychological, especially in women and younger patients (age and gendered assumptions- e.g. being assumed to have an STI, due to being young and female). Being blamed for treatment failures. Lack of options for care or treatment. Experience of having 'nowhere else' to go.	Mostly negative and lengthy journeys with lack of support and experience of uncertainty- described as torture, hell, brutal; daunting and scary. Descriptions of being abandoned to fend for oneself (left on the scrap heap). Many examples of assumptions and biases affecting journey; more broadly, when women are affected- nobody cares what happens. Journey described as 'nonsense'- people have to go through a lot to get basic care for conditions that are not uncommon. Only believed once there	The journey is lengthy and mostly negative, with some positive experiences along the way. Participants describe issues like short appointment times, assumptions and biases (e.g. if a doctor sees fibromyalgia or anxiety, they attribute all your symptoms to it and say they can't help you). Experience of blame and feeling like a burden- when they can't explain it, you are made feel like you are wasting their time. Negative experiences are related to lack of support- feeling abandoned after a	Descriptions of patients having to take care of their own journey and fight every step of the way (e.g., documenting all the symptoms) and long waiting times. Everything is a fight even with medical literacy (even if it helps) like to find knowledge on supports. Patients are blamed for their advocacy (self or others) or when treatments have negative effects. Assumptions (on age, gender, being only child, young mother, weight, psychological history) interfere with their healthcare experience.	Most descriptions of the journey negative about the time spent in healthcare from one service to another. Patients forced to self-educate and advocate for themselves. Symptoms often dismissed when women/younger or patients are blamed for them.

	Focus Group				
	1	2	3	4	5
	Requests/feedback associated with anger or being treated like a 'bad' patient.	is proof (i.e. blood test/ evidence).	diagnosis (diagnosis seen as an end point), with no guidance about management, and nowhere to go. The journey is dominated by a need to 'prove' illness (e.g., in bloodwork)- no abnormality means you are fine and nothing is wrong. The journey often leaves you feeling like there is no hope. All participants described instances of having to take charge in their journey (you have to 'save yourself', become your own GP, educate yourself, find things that help on your own).		
Positive experiences of the journey					
	Descriptions of some positives along the journey (e.g., Generally negative, but meeting some lovely people along the way; making friends with patients). Some examples of being surprised about having a	Positive experiences of the journey abroad. Mention of treatment in other countries as a comparison to the Irish experience. E.g., immediate and Affordable care and	Weak theme. Some mention of positive experience even when there is no cure- meeting good people along the journey, eventually learning how to manage symptoms.	Positive experience in a rheumatology rehab with a multidisciplinary team. Experience of care following patients' recommendations.	Some descriptions of becoming stronger, more positive, and accepting. Development of patient expertise as an empowering experience.

	Focus Group				
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	positive healthcare experience with clinicians once in a while.	testing (‘everything is done tomorrow')			
Systemic issues					
	Systemic issues implied and discussed to some extent- include lack of comprehensive care and holistic treatment approaches (focus on individual symptoms rather than overall condition); patients 'falling through cracks'). System is not set up for chronic illness- clinicians don't specialize in these because it is 'career suicide'.	References for the impact of healthcare on health (mental and physical). The system is not set up to deal with chronic illness or uncertainty (short appointment times, focus on proof/evidence). There is more support for people with known conditions (e.g., cancer). Need to be a good patient to get ahead. Nothing gets done by people in power unless there are financial implications or a crisis.	Systemic issues are implied throughout ('a dysfunctional system'). Reference to 'falling through the cracks' as a patient with these conditions. No system to deal with people who have complex issues; only one issue at a time.	Patients see negative experiences as more systemic issues than personal issues. Failure of the system to learn from patient expertise (disease, helpful treatments,). System is not set for chronic illness and considers it low priority: hyper specialisation, compartmentalisation. Common expectation of very unwell, passive patient fitting into categories. Rigidity of public system for clinicians. Feelings of distrust in the system.	Descriptions of systemic inertia with a focus on acute conditions with known fixes over chronic and unknown illness. Preference for passive patients. Lack of centralisation of patient record and activity (lack of communication between doctors). Lack of linking with other services (other therapies, psychology, etc.). Multiple mentions that the system is cut out for acute, not chronic care.
Barriers associated with healthcare					
	The healthcare system adds to the burden- e.g. stress/ negative emotions, financial constraints, and the need to take care into their	Patients discuss needing backup- they are too sick to have to fight/help themselves all the time, having to fight for basic tests. Lengthy diagnostic	All participants discussed the impact of healthcare on their illness experience- it is hard enough being chronically ill, but you have to	Description of an important burden because of patients needing to force their way back into the system, find solutions of their	The lack of diagnosis and support leaves patients to self-diagnose advocate for themselves. Often too ill to do so. Patients must sometimes manage their

	Focus Group				
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	own hands. Patients must often advocate for themselves, navigate complex systems, and coordinate their care. Linked to burden of illness and coping with chronic illness.	process takes a toll. Having to educate yourself on top of being sick, having to advocate for yourself ("we have enough on our plates"). Implications of this on mental health. Burden of seeing many doctors/consultants (e.g., saw 34 consultants over 16 years). it takes a financial and emotional toll. Added paperwork that is draining and not necessary.	manage the system on top - it is described as a 'war' to get somewhere. Burden of having to chase up results, coordinate care- otherwise you are waiting and nothing happens. It takes a lot of energy/time to get to appointments (particularly when you have an energy limiting condition), so it is disappointing when you don't get anywhere. Managing clinical encounters is another burden- you have to plan how to interact/confront a clinician in a way that will not jeopardize your care; you cannot show anger or they will shut you down; you have to hide your stress/anxiety and worry. This takes a significant toll on the patient emotionally.	own, advocate, manage consultations and communications, travel, etc. Sometimes patients are too ill to do so. Psychological burden (e.g., cry at end of consultations, feelings of self-doubt, desperation) because of gaslighting and lack of support. Detrimental effect on their illness because of medical decisions (e.g., crash, overdose)	appearance to be taken seriously. Their physical health sometimes suffers from bad medical decisions (egg, surgery) and mental health is impacted from being dismissed.
Barriers to effective care *Lack of continuity & poor coordination*					

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	Major pain point. Patients frequently see multiple doctors, experience breakdowns in communication, and lack continuity in care (e.g. always a different GP) or sudden discontinuation (e.g., doctor leaves and replacement never materialize). Care quality is a case of 'potluck'. Systemic disorganization- Issues include wasted time explaining medical history, and poor follow-up on appointments and treatments.	Multiple references to issues around continuity (not able to see your own or one GP); Poor coordination- e.g., No report or image from consultant to GP after 3.5 years of waiting for appointment; couldn't get more information. No follow-up despite promised tests. Communication between doctors is also very poor- they don't listen to and learn from each other.	Participants discuss being 'bounced around' when things don't work out- referrals are used to get rid of the patient. There is no continuity (You see a new doctor every time and they have no idea who you are). You have to coordinate everything yourself.	Compartmentalisation leads to seeing multiple specialists and being dropped by a lot of them, especially when tests return negative. Long waiting times. Clinician potluck. Lack of communication between hospitals and between public and private.	Patients frequently see multiple HCPs and struggle to identify who's who, experience breakdowns in communication, and are often 'abandoned' when hyper specialism does not work. No centralisation. Lack of EHPs mentioned.
Inconsistency of care	You can see a variety of different qualities of doctors ("potluck"), some not as good as others. Diagnosis/solutions offered 'by chance'. Strong links to poor continuity and coordination.	Inconsistent standards of care. Participants described having both positive and negative care experiences. Some conditions (e.g. cancer) get better care than others. 3 patients mentioned travelling abroad for treatment and diagnostics as these are not available in Ireland	Some GPs are better than others- hard to know who you will end up with. Some doctors will do the bare minimum to get by. Some experiences come down to issues of visibility. Participants describe different experiences when they have a visible issue that can be fixed (treated like	Care depends on how interested the doctor is, the "postcode lottery", generation of the doctor. Care mostly if extremely unwell, not before it gets bad.	Mention of care depending on age of the doctor; different levels of care in different countries.

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		(and care is better, faster, cheaper).	a VIP) and completely different experiences when the issue is not visible (e.g., complex pain is ignored). Differences between Ireland and care in another country are described- here, you can't get a test just because you want to.		
Resources					
	There is a high need for more GPs and specialists. Demand is higher than supply; long wait times to be seen are common. Public healthcare systems are often under-resourced, leading to insufficient care availability. Issues around catchment areas and GPs not accepting new patients- therefore having to rely on A&E	Lack of national resources evident particularly with regards to diagnostics. Many examples of waiting for care- e.g., 420 weeks for ENT, but appointment was reduced to 3.5 years by outsourcing. Specialist clinics are being closed down (funding pulled). Even 'brilliant' doctors lack resources (e.g. knowledge and time)	Resource-related issues are implied. Some mention of time constraints as a barrier to care. E.g., a doctor has 10 minutes to figure things out and then you're out the door. You can only discuss one concern in the appointment.	Small island' constraints with lack of specialists (mention of it being better now), appropriate testing, accessibility to medication especially niche drugs. Complain of short appointments.	Small island' constraints with lack of specialists and need to travel. Lack of retention of Irish doctors and international students. Time constraints discussed in detail. Demand-supply issues- lot of patients, not enough clinicians. Lack of IT solutions, despite claims.
Stereotypes/ Assumptions					
	Major stereotypes around mental health; gender and age bias are a common experience and strong theme. Women	Example of 'Mad-Bad-Sad'. Mental health is blamed. Participants agree that stereotypes are the biggest challenge	Stereotypes are an issue- e.g., a doctor will no longer listen to you if you are diagnosed with FM (even if symptoms are	Assumption of health when results return negative or patients look 'too' well. Bias of pain being "in the head" for	Gender and Age Bias: labelled troublemaker especially if not passive patients, pain assumed 'in their head'. Demeaning

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	and younger patients often face stereotypes, being labelled as neurotic or having psychological conditions instead of physical ones. Pain and symptoms in women are often not taken seriously, leading to misdiagnosis and inadequate treatment	in healthcare- people categorize you and won't look any further. Issue of looking normal (you need to be older and sicker to be taken seriously). Problem for females is either too much or too little sex. Hysteria is still being used as an explanation (even in books). Doctors are aware of biases (some acknowledge that other doctors don't believe in Lyme, for e.g.).	not related), they will not engage with alternatives because they "already know the answer". If you have a mental health history/anxiety, doctors will assume everything is related to anxiety- they 'seize onto mental health' as soon as it is mentioned (tunnel vision). Labels applied- e.g., "worried well". Gendered experiences- e.g. "this is due to hormones and nothing can be done. Psychologization of ME/CFS and doctors 'ganging up' against patients- these impacts on medical care offered/provided.	young women or diagnosed FND. Bias on mental health history.	views on chronic illness. Stereotypes lead to not being listened to or believed.
Financial constraints	Patients often face high costs when seeking private care due to the inadequacies of public healthcare. The financial burden is a significant barrier to accessing	Mention that testing and seeing many doctors is expensive. Going abroad is an option but it's extremely expensive and not everyone can do this.	No references	A lot of specialists are or go in the private system and are particularly expensive. Patients frequently the cost barrier that most of the specialists available	Patients face regular costs when seeking multiple opinions or specialists, when testing (in particular genetic testing), or for therapies (e.g., physio), when buying

	Focus Group				
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	necessary/adequate and timely healthcare.			represent on top of travel costs.	equipment/services for help at home. Costs increased by lack of accessibility and quality in public care.
Social welfare					
	Patients face stringent criteria for qualifying for disability or special access to care, making it difficult to receive necessary support. One example.	Weak component. Home help and financial help is needed.	No references	Patients sometimes have to stay with a specific doctor to sign off for income maintenance and disability.	Patients rely on disability payment. Navigating the social welfare system is hard. Patients need guidance.
			Category 3. Needs and recommendations		
The consultation					
Patient-Clinician Relationship					
	Emphasis on need for improved relationships/dynamics. Clinicians should understand and believe patients, treating them as individuals rather than numbers. Clinicians should be honest/transparent (e.g. if they don't know something) take the time to explain and answer	Some examples of issues /need to improve the relationship. E.g., need to be believed and listened to ("really listening"), need to be heard and patient suggestions/knowledge to be considered. Need to improve how time is being used given the limits; better	All participants expressed needs/made suggestions for improvements related to the relationship. Strong need for doctors to listen, be more compassionate and be willing to engage with the patient. Participants discussed how patients are disempowered (clinicians take control and take away patient	Patients mentioned the need to be listened to with empathy, believed, and their experience validated. "Pain is what the patient says it is".	Patients insisted on being believed and their experience validated, with clinicians listening to them and willing to collaborate with their expertise. Emphasis on transparent communication (e.g., knowledge or system failures). Patients need doctors to take time and to be clearly identifiable

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	questions, adapting care to patient needs. Importance of a holistic approach, taking time to listen to patients, explicitly validating their experiences, and showing empathy. Less blame of patients needed. Many examples of positive statements about clinicians, followed by 'but'.	communication during the time available.	voice). Need for empathy and improved bed-side manner is emphasised in the absence of a cure- a good relationship can improve the experience. Aversive body language (e.g., eyerolling, huffing and puffing) needs to be addressed. There is a need for explicit acknowledgement of struggle/validation of experience. Patients should not be made feel like they are wasting a doctors' time. Collaboration- openness and willingness to work with the patient.		(e.g., clearly labelled badges)
Guidance/Signposting	Patients describe need for improved access/signposting to resources (e.g. information/ self-management/ support groups) and concrete guidance at the time of diagnosis and throughout the treatment journey.	Weak component. Some reference to needing a 'guide' or "single source of truth".	Some reference to a need for guidance and concrete solutions; suggestions for next steps (rather than 'shrugging shoulders') and suggestions on how to manage symptoms.	Emphasis on the need to lighten the patient's burden by providing a diagnostic bundle with information and resources about their condition, navigating the system, receiving practical support, and mental health support.	Patients insisted on help to navigate the social welfare system, to access information and resources, notably on concrete and feasible solutions to improve their life quality

	Focus Group				
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	Assistance with applications for disability services and navigating the healthcare system. Emphasis is on not focusing just on a diagnosis.				
Supports at different stages					
	Importance of connecting patients to support groups and advocacy groups, especially at the beginning of the diagnosis journey. Ensuring psychological support is available for patients with chronic illnesses. Support needs during the treatment, after diagnosis and while waiting.	Weak component. Some reference to needing free testing/ improved care during diagnostic phase. Guidance early on is important (e.g., being told not to exercise)- prevents the patient from doing more harm.	Participants discussed a need for continued support (e.g. mental health) post-diagnosis- no supports are in place. It is hard to come to terms with a chronic illness diagnosis, particularly as a young person. Need regular appointments (check-ins) throughout the journey to monitor whether treatment is working. Desire to be given hope -your life is not over, there are ways to manage the condition and live.	Importance of signposting and providing support, in particular post-diagnosis. During the consultation, patients wished for an optimisation of the time to take advantage of every minute (e.g., questionnaires).	Importance of referring patients to therapies/alternative therapies even while waiting for diagnosis. Providing support to avoidance dependence on family. Example of residential therapy for chronic conditions in Poland.
Improved knowledge					
	Clinicians need better knowledge to ensuring	Knowledge among clinicians and more	Not a dominant component. Implied need	Emphasis on increasing the awareness of	Clinicians need better knowledge and new

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	consistent and accurate diagnosis and treatment- there needs to be consistency/standards among specialists/consultants. Need to differentiate between physical and mental illness /symptoms (e.g. anxiety vs. chronic illness) Need to learn from patients and put biases/beliefs aside.	broadly needs to be improved- suggestion to develop centres of excellence, to do longitudinal research. Improved knowledge among clinicians would take the burden off of patients who are doing their own research and educating clinicians.	for more knowledge, particularly in relation to ME/CFS. Emphasis is on the pathophysiology of ME, moving away from psychological explanations.	guidelines to all clinicians and not just specialists to avoid misclassification of illness.	specialties should be created (e.g., neuroimmunology). Doctors need to be willing to learn including from patients, notably about system and testing.
Improved Understanding					
	Implied that clinicians should try to understand the suffering/patient experience from their patients.	Some reference to a need for clinicians to gain a 'real' understanding. To believe the patient/ work *with the patient, believe patients and give them a sense of hope.	Emphasis on 'real understanding'. Having a doctor with lived experience would be ideal but not feasible- instead, doctors need to be open/willing to learn from patients and people with lived experience; they should an effort to truly understand ('shut up and listen'). Need recognition that multiple complex conditions co-exist and are not mutually exclusive- you can be stressed, mentally ill, and	Clinicians need to be less dismissive when they don't know how to treat an illness and be more open-minded about chronic illness. They need to remember that they're not the ones that go home and live with this illness all the time.	Clinicians need to change their outlook on how common chronic illness is compared to psychosomatic issues. Need more willingness to admit to limits and listen/learn from the patient, who lives with condition

	Focus Group				
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			have a real/physical chronic illness.		
Improved efforts	There is a sense that clinicians 'should be doing more' but no specific comments/suggestions.	There is a sense that clinicians should be trying harder/ doing more tests than they are. This should be available without a fight. Improved clinical efforts- e.g., taking a thorough history and really trying to figure things out.	A need for improved efforts is implied throughout but not specifically recommended. Clinicians should be more attentive and thorough (investigate things, read the history) and be willing to run basic tests (e.g., for anaemia). Taking time to listen. Make an effort to call back/follow up.	Clinicians need to be more proactive in looking for the current knowledge and guidelines when they're facing a condition they don't know about.	No reference
The service					
Comprehensive Care	Need for MDTs, including nurse practitioners and specialists for misunderstood conditions. Need access to general specialists for patients who fall through the cracks in the system; clinicians who are interested in the patient as a whole (not just their one specialization).	Need for MDTs and clinics, with specialists who have knowledge of multiple conditions, not just one. There needs to be agreement on how patients with poorly understood conditions (broadly) should be treated.	Weak component. Mention a need for a 'one stop shop' (like a primary health centre) with various specialists in one place.	Need for multidisciplinary and holistic approaches to treat multi-system conditions, notably in pain clinics.	Need for MDTs and rehab centres to cater for the needs of chronic illness patients and compensate for the lack of specialists

	Focus Group				
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Personalised Care	Treatment plans should consider patient-specific factors such and provide personalized care rather than a one-size-fits-all approach. Patient work/life commitments should be considered. Need to adapt faster processes r patients who need multiple referrals.	Care needs to be suited to patients- e.g., longer appointment times for patients with complex issues. Specialist clinics need funding.	Weak component. Some mention of need for specialists-- for example, mental health practitioners that specialize in chronic illness (not general counsellors).	Need for the clinicians to 'meet the patients where they're at'. Need for increasing accessibility to experts and specialised clinics, in particular when unwell (e.g., online calls).	Mention of the need of new specialties in healthcare.
Improved coordination	Need and preference for a single point of contact (e.g. one GP, liaison nurse) to streamline/coordinate care. Suggestion for patient registry- Implementing a single patient information system for full visibility and improved communication between departments. Patient should not have to coordinate everything and chase after documents.	Need to centralise treatment plans and adopt treatments/processes that help. Information should be shared between doctors and with patient. We should be working with other countries and putting data together to help people.	Need for continuity (not a new doctor every time) and regular follow up. Participants discuss a need for a central point of contact (GP or patient rep) who has a proper overview of their needs/ case and is able to coordinate care.	Need for a specialised middleman to help coordinate. Patients insisted on regular follow-up, integrated model of their data with patient ownership. Need for joined-up care with improved teamwork and network. Move to digitalised records.	Mention of the need to improve teamwork, communication, and MDTs, centralisation of a digitalised patient record. Examples of this in other countries.
Linking beyond healthcare					

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	Need healthcare to connect with other services- e.g. link with and guide patient to community supports and patient communities, especially while waiting for diagnosis or treatment. Some links already exist (e.g., meals on wheels)	No explicit reference to a need to link health services with others (e.g. support groups). Example of clinician linking in with Meals on Wheels- this was helpful.	No reference	Connecting patients to support/patient communities and to psychology services. A major suggestion is bringing people together online- to talk to one another.	Connecting patients to therapies/alternative therapies and advocacy groups.
Increase resources and incentives					
	No reference to creating incentives for clinicians to specialize- some reference to how chronic illness/care is unappealing (no one will touch it with a bargepole)	No explicit reference, but it is implied that identifying financial implications of chronic illness will be an incentive to increase funding/work in the area.	No reference	No reference	Facilitating access to the studies. Retaining students by asking for payback once trained. The end-goal is to increase the number of doctors available for shorter waitlists.
Diagnostic Services					
	No reference to specific diagnostic services.	Emphasis on a need to improve diagnostic processes/practices. Need to have a thorough approach ("an NCT"), and resources for specialized tests (instead of going abroad). Diagnostics need a major update- outdated generally (e.g.	No reference	Patients mentioned a need to improve accessibility of diagnosis services.	Increasing the accessibility of the services and forward thinking about the possibilities of testing. Discussion of limitations of genetic testing and specific variants of conditions- needs update.

	Focus Group				
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		diagnosis of ME) and in Ireland (diagnosis of Lyme). Early diagnosis can reduce harm/cost (e.g., a diagnosis of ME may stop patients from pushing self/ decline)			
Training and development					
Knowledge and Understanding					
	Training to combat bias-reduce assumptions (e.g. urinary issue= STI), misinformation, and stigma around chronic illnesses, and improving understanding of different conditions. Clinicians need to educate to prevent harmful stereotypes/eliminate this from patient experience.	There is a broad sense that more knowledge and understanding is needed; clinicians/experts should be listening to each other and learning from each other. There needs to be more knowledge/understanding/recognition of how these conditions affect children.	Weak component. Need more knowledge about specific conditions-doctors get a very small amount of exposure to ME/CFS in their training.	Current guidelines for conditions need to be put forward. Specialised conferences on rare diseases should be offered.	Training to update their knowledge and understand progress in research.
Continuous Professional Development					
	Enhancing Skills: Ongoing professional development to enhance communication skills, empathy, and understanding of chronic	Update competencies-combatting misinformation, learning not to jump to conclusions (e.g. about hysteria/anxiety) if they	Emphasis on exposing clinicians to people with lived experience. More training in empathy/compassionate	Intergenerational exchange of information/expertise between older and younger doctors. Need	Doctors would need increased ongoing professional development to improve testing knowledge and diagnosis.

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	illnesses. Encourage learning from patients.	can't make a diagnosis. Need to develop understanding about links between mental and physical health.	care and bedside manner; learning from nurses.	for international experts training younger doctors.	
Medical Training/Education					
	More emphasis needed in medical training on how to be kind to people and how to speak to people (intelligence is not enough)	Not explicitly addressed. General statements about updated knowledge/understanding.	Current training is brutal and creates doctors who think their patients are stupid. This needs to be addressed. Need for entry criteria that emphasizes empathy and passion more than academics/intelligence	Emphasis in training on non-emergency and/or non-traditional situations. More updates on knowledge. Emphasis on teaching bedside manner (e.g., training for nurses as an inspiration).	Need some changes in medical training for learning empathy and changing the bias around chronic illness. Some mention of a relationship between training and practitioner kindness as an outcome.
Clinician Wellbeing					
Targeting Burnout					
	Increasing support and self-care measures for clinicians to prevent burnout and ensure they can provide the best care. Targeting compassion fatigue.	No reference to clinician wellbeing.	Weak component. Mention that clinicians need to relinquish control- they are burned out because they take on too much responsibility/try to control the patient.	Some discussion about burnout and consultant practices. Mention of burn out since Covid preventing clinicians to look outside the box and provide specific treatment. Compassion fatigue for both patients and doctors provokes adversity.	Need to target the length of work shifts, lack of doctors, pressure that led to overworked doctors who make more mistakes. Need support during and after training
System/Culture Change					

	Focus Group				
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	Supervision needed for clinicians.	No reference	Implied need to address culture in medical school/healthcare. Large amounts of hierarchy, bullying, dysfunctional training which does not see sift skills (people skills) as important. This creates clinicians who look down on patients and do not value patient experiences/expertise.	Need to target the tendency to overwork doctors that were pushed into fields that they don't have experience in.	Need to target the culture of poor trainees' treatment. Brutal training practices are dangerous. Need for more acceptance that not knowing is ok. Doctors expected to pull themselves up by the bootstraps and somehow get by.
Knowledge Generation *Need for Research*					
	More research, especially inclusive trials involving women, to address the scepticism around female subjects and outcomes.	Some reference to a need to conduct research to improve things for people in the future. There is a need to find commonalities among different patients- symptoms and treatments that work- in order to have targets for change. Some research on new medications is promising.	Some references to a need to improve research practices. Participants noted a historical lack of medical research on ME, underfunding, reluctance of pharmaceutical companies to engage with complex and poorly understood conditions (because there are no easy fixes, the area is not seen as lucrative/worth the research).	Implied need for research- discussion of statistics and attempt at quantifying issue (e.g. EDS) in Ireland.	More research and emphasis on specific illness, especially for treatments, managing symptoms, and biomedical solutions. Using patient data should be considered even if need a workaround GPDR.
Lived Experience/Patient Voice					

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	Implied throughout that clinicians need to listen to patients and learn from them.	Not a specific recommendation. Implied throughout that clinicians need to listen to patients and learn from them.	Participants describe a need to capture patient voice/involve patients as collaborators in research (PPI). Clear need to incorporate lived experience/ involve patients in training development. Some mention of taking part in research as a way of contributing.	Encouraging listening and learning from patient experiences.	Knowledge from patient groups should feed the research (e.g. for pain management). Emphasis on listening to the patient who knows their body and condition better than any clinician will.
Public Awareness					
	No reference	No specific recommendation about raising awareness. Things may change now that kids and men are affected and there are millions with ME after the pandemic- you can't ignore millions.	No reference	No reference	Patients expressed how public awareness should be increased. For patients to be believed and supported in terms of advocacy, information about chronic illness should reach the public (e.g., campaigns)
Implementing Change					
Coordinated efforts (National/International)					
	No specific recommendations about implementation/change by involving multiple stakeholders.	Some agreement about a need for coordinated (national and international) efforts that get all stakeholders on board. Information/data (epidemiological, cost),	No specific recommendations about implementation/change by involving multiple stakeholders.	Meetings between coordinators of support groups and different stakeholders who have the power to act on the system.	No reference

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		politicians and clinicians. Need someone to coordinate this and to challenge the norms in order for anything to change.			
Application of research and knowledge					
	Statement that we would be better suited to deal with COVID if patients were listened to and knowledge/outcomes from the PACE trials was applied	Need to use knowledge and data we already have instead of just waiting for treatments to be developed. Need to use data that has already been collected (e.g., from a clinic that is being shut down- rich source of info).	Reference to training of nurses- content is there and can be used with doctors.	No reference	We have knowledge- need to stop re-inventing the circle and use it (i.e. re. alternative therapies).

Appendix D6. Supplementary Quotes

Table D6

Illustrative quotes

Theme & Subtheme	Illustrative Quote(s)
Living with a chronic illness	
Burden of Illness/ Disease	Because it's very, my life is actually quite stressful. But that is the, it's being chronically ill, and chronically poor, and chronically, everything else. And chronically, my life has been banjaxed. And I'm supposed to be like….you know, that it isn't sensible to kind of go, "Okay, your life has been kind of wiped, you've been majorly gaslit by your family and the doctors. And you've had to do your own DIY health care". And there's not supposed to be any kind of, you know, fallout from that? (Sophie, Group 3)
Coping and Managing	I remember, after that, hiding my limp, and it was something- so I was still, you know, teenager living at home, trying to pretend that I wasn't in pain, trying to pace trying to manage myself. So it's a very, very lonely time. very distressing. (Amy, Group 1)
	And I know everybody with chronic illnesses, we go, but I do this. "Should I have done that? Did I do overdo it? What about this? What about that?" because you're haunting ourselves and it's exhausting. On top of being sick. (Caitlyn, Group 4)
Positive Outlook	What I look at is, you see...I've become a stronger person because of this. It has taught me to be persistent. (Dawn, Group 5)
	It's... your life's not over basically, there are things you can do. Plenty of people live with this condition and manage it (Liam, Group 3)

Theme & Subtheme	Illustrative Quote(s)
Community and Social Support	
Positive interpersonal experiences	So no matter how bad my days were, like, I had these people helping me. So all I had to do was do what I have to do, and eventually I'd get better (Anna, Group 2)
Negative interpersonal experiences	So, so that, the biggest challenge was, yeah, what people think of, of this, They don't understand because we look normal. That's the biggest challenge, I think (Ellen, Group 2)
Value of Community	There's all sorts of really useful tips and tricks from the from the groups (…) And I'm still in touch with the patients in Germany, who were mostly American. And I'm still in touch with them all the time, what they're doing, what treatments are using, and and how they're getting along (Amy, Group 1)
Experiences with Healthcare	
Positive experiences- clinicians	And I actually nearly started crying in front of this man because I couldn't believe that he cared enough to do that. And then he's just been really proactive and really good and he follows up immediately (Sarah, Group 1)
	My GP, to be fair, she was good. She didn't have a clue and she didn't pretend… pretend to have a clue. But she at least did listen to you, you felt like she did believe you, she was in your corner. (Millie, Group 1)
Positive experiences- other professionals	And then I ended up going to a different physiotherapist in there because the original one also left. And I took the new one. I just looked at her and she looked at me and was like, "Are you good?" And I'm like, "No, I'm not". And she went, "well, there's no point in me doing anything with you right now because you're in so much pain that I'm not going to be able to loosen anything". So she was very good (Sarah, Group 1)
Negative experiences- clinicians	I would go to doctors and consultants, whether it be in hospital or A&E, it was always a lot of them. Got the "only child" thing a lot, "Princess", "spoiled", that kind of thing. And eventually an intervention, Psych, I got sent to the psychologists a lot, usually being told it was all in my head. (Niamh, Group 4)
	But then when they can't figure out what's wrong with you, there is nothing wrong with you. And then you're left either with a, you know, you have to go along with this, “there's nothing wrong with you”. Or it's made very clear, you're gonna get a psychiatric diagnosis. So its almost like a threat. (Amy, Group 1)

Theme & Subtheme	Illustrative Quote(s)
Negative experiences- other professionals	An occupational therapist came in, I had a shower stool, I had a walking aid, aids around the house, stools in the kitchen and whatever, because I couldn't stand up. Em, like, but that was a five-minute thing. Someone came in, and the occupational therapist basically told me that she thought it was all in my head. (Ellen, Group 2)
Negative experiences- treatment, therapies	I was seeing lots of different people all the time. And I was, I kept trying to tell them "lads, like, I've been on antibiotics about five or six times this year, like there has to be like", and they'd be like, "oh, yeah, just come back. If there's a problem." and I'm sitting, staring up and being like, "Dude", I've been on six antibiotics this year. (Sarah, Group 1)
	But I was on antidepressants and that, and the reason why I didn't want to go on the anti-psychotic was because they tried that on me before. And it had an awful lot of side effects that I couldn't function with. But it was taken as if that I was gonna deny their course of action. (Chloe, Group 1)
Power Dynamics	And I think also, I think it works against them. Because they take on all the responsibility and they think "look, I have to hold on to all, I have to control everything. I can control you, you are my patient". (Sophie, Group 3)
	And maybe we're seen as being threatening when we go into these meetings and consultations then because we know more stuff. And very often I'm finding I know more than the treating doctor on my condition because I literally after studying it so much and trying to keep up all the time. (Mia, Group 4)
The patient journey	
Negative experiences of the journey	The amount of times I came out of consultations and just cried as I drove off thinking "that door just closed", and I'll find another door, and I'll open another door, but it's... it's a, it's a journey. (Amber, Group 4)
	It's a very, very lonely time, because you try, and you don't want to be there. But it's as if you're a burden that you're, you know, that you're, you're coming in, and you're just looking for attention (Chloe, Group 1)
Positive experiences of the journey	Once he got the hospital, he had perfect health care experience, we are all struck by how good it was. And still is, the aftercare he is getting is superb. Just a few weeks ago, he was asked to be a patient representative. And he said," Why do they choose me" and I said, "because you got a good outcome". (Amy, Group 1)
Systemic issues	So (laughs) it's just, yeah. Waiting list, waiting for phone calls, but they don't come. You're young, it's probably fine. (Sarah, Group 1)

Theme & Subtheme	Illustrative Quote(s)
	The system now is set up for an acute type situation, 'Fix me I'm broken'. Everything is about passivity. If the patient just, if the patient does something that they're not supposed to do, they're told they're noncompliant, but they're not believed. (Shane, Group 5)
Burdens Associated with Healthcare	She wanted me to do more paperwork to monitor...and I can't even do my own paperwork, I'm behind with my own, you know, paperwork, to get invoices to get paid for stuff. I'm not going to start filling in silly little childish questionnaires. She has with little cartoons about what I do with my time. It's too patronizing. And it's also more work and more exhausting. One of the most tiring things for me is paperwork. (Emily, Group 2)
	Because we have to go in and advocate, we have to educate ourselves, we have to demand the blood tests demand the scans, even if we don't have the language for like, I was lucky I could draw, I had the idea to draw a pain diagram, I was lucky, I understood physiology and medications. If I didn't push, if I didn't come in proactively with work, I would have taken much longer to get my diagnosis. (Robert, Group 4)
Barriers to effective care	
Lack of continuity and Poor coordination	I was sent into an acute medical assessment unity, and only for the doctor telling me I wouldn't have known, but apparently there was no cross back from there to my GP. So anything that was found out would never be sent back to my GP. I had to physically remember to get on email after the fact and say, "Okay, this is the consultant I saw, this is what was said, and I want it updated on my file." Because obviously, as a patient, I want everything recorded and on the one file so that if someone reads it, they know where I stand, and they know who I've seen. So that's another issue is that continuity. (Millie, Group 1)
	My experience is similar to yours of like being dropped once I got my diagnosis, even though it was very, very quick, considering like, the gauntlet of tests we did and everything. And having a doctor who was very aware- there was no follow up. There wasn't. I didn't know there was a pain clinic in the city, I didn't know there were support groups, I didn't know, there were treatment options, it was just go. I was completely denied any pain management. Because of my age and the long term risks of opioid use. I was, it was just "Sink or Swim". (Robert, Group 4)
Inconsistency of care	You're your own advocate in this, if you're fortunate enough, which I am, but a lot of people aren't, to have a good GP who listens and believes and doesn't, you know, gaslight you? And I think there's, there's hope. But then once you get back into the hospital systems and the consultants it is completely and utterly potluck. (Mia, Group 4)

Theme & Subtheme	Illustrative Quote(s)
Resources	And because they will not accept new patients, like myself and my partner both, neither of us can get into surgery, my parents are actually moving up. And they're, um, a few months ago, they can't get into surgery for another reason, because they're outside of catchment area for literally everybody because they live in the middle of the country. Like, I understand there's like a demand and everything. But like, figure it out. Dyou know, like have a nurse practitioner that people can go to in a walk in service, once a once a week or something (Sarah, Group 1)
Stereotypes and Assumptions	I collapsed in May of 2011. I was brought to the [redacted] in an ambulance. And for a couple of days, one of the doctors said to me, he says "I think you might end up in the MBS Basket". And I was looking at him like, MBS basket? And he smiled, and he said, "You need to be careful." He says, "With doctors," he says "you cannot be put in a mad, bad or sad basket. Look, " he says, "you need to be careful". And that's something that I've seen repeatedly with doctors , and I think Ellen mentioned that where a lot of people will try a lot of consultants and if they can't fix you, then it's not there. It's not their fault. It's in your head. And I would have faced a lot of that up until the blood results. (Alex, Group 2)
	They already know the answer. That's it. You see, they already know the answer, because they already know, as far as they're concerned, that all my issues are...that I'm simply "the worried well", "overly health conscious" and I have no issues. Whereas actually, I'm dealing with unbelievably complicated issues. (Sophie, Group 3)
Financial constraints (for patients)	Yeah, he's private and costs a fortune. Just as a separate piece? 450 a session for the first assessment. And 300? I don't know. 300 to 350 subsequent dates, and supplements. A lot of money isn't it? Yeah. Well I think he does know his stuff. To be fair, I think he does. But I don't know.... that's very exclusive. It's not accessible to everyone. That's just. That's not the kind of money (Mia, Group 4)
Social Welfare	So, the criteria for disability and everything is too stringent, my GP, I suppose, had to lie a little bit, too, to get me a disabled parking permit. (Amy, Group 1)

Appendix E. Co-Analysis

Appendix E1. Guidance for reflexivity and familiarisation

Reflexivity

Reflexivity in the context of qualitative research refers to the practice of researchers examining their own contributions to the research process. This involves recognizing and reflecting upon the researcher's potential biases, beliefs, values, and experiences that could influence various stages of the research—from data collection and interpretation to the final presentation of findings. Reflexivity can help:

1. Enhance credibility and rigor of research. Researchers examine how their own perspectives might shape the analysis and acknowledge and manage their biases.
2. Improve transparency. Documenting reflexive thoughts can help researchers track the path of reasoning behind thematic development and conclusions drawn.
3. Promote ethical research practices: It makes researchers more aware of their responsibilities towards participants, especially in terms of how they represent other people's experiences and the implications of their interpretative acts.

Questions

Before starting the analysis

Before you read the first transcript, consider some of the questions below and make some notes. You do not have to answer them or share your answers with me if you would prefer, so please be as honest as you can be in making notes.

1. How might my personal experiences with the topic influence my interpretation of what interviewees are saying?

2. In what ways might my hopes or concerns about the research outcome affect my analysis of the interviews?

First reading of the transcript

Make some notes of what comes into your mind as you are reading the transcript for the first time. Don't worry about your notes being messy- they do not have to be fully developed ideas, simply thoughts that come up.

Here are some questions to consider as you reflect on the transcript before you begin coding. Again, these serve as a guide and you do not have to share your reflections with anyone if you prefer not to.

1. How did the interview make me feel?

2. Why might I feel this way?

3. How might my emotions influence my analytical decisions?

4. Did anything resonate with my own experiences?

5. Was anything very different from my experiences?

6. Was anything surprising?

7. How did I feel about the interviewees, and why could this be?

8. How might my assumptions about the interviewees affect my understanding of the data?

9. Are there any themes or comments I strongly agree or disagree with?

10. How might my aims for the research influence the way I am reading and interpreting the interview?

11. How might my personal needs (healthcare or otherwise) influence the way I am reading and interpreting the interview?

Coding and generation of themes

Here are some questions to consider once you have coded the transcript:

1. What influenced my coding decisions?

2. How did my experiences or identity influence my interpretation of the data?

3. Did I encounter any information or perspectives that challenged my views? How did I handle this?

4. Do I feel particularly attached to any of the codes/themes? Why might this be?

5. What have I learned from this coding process about the topic, the participants or myself?

6. How might my coding and interpretations affect the outcomes of the research?

Appendix E2. Charting Summary Template

Experience of Living with a chronic illness					
Theme	Subtheme	Occurred?	Key points/Summary of data	Lines for Quote	Notes/Comments
Example	*Delete examples*	*Yes/No*	***Briefly describe/bullet point**- this is a summary of the findings for this interview.* *for e.g., participants in the group discussed experiences of X, P1 suggested Y, others suggested Z..,*	*Relevant quotes used to illustrate the summary* *Any important quotes can be emphasized (e.g., highlighted)*	*Observations- e.g. theme is not sufficient, include codes like X, needs to be expanded…*
Living with chronic illness	Burden (of illness/disease)				
	Coping with chronic illness				
	Add theme/subtheme if needed				
Healthcare experiences					
Needs and recommendations for change					

Appendix E3. Guiding Document for Interpretation

Part 1

Please take some time to reflect on the analysis and your thoughts/feelings about the focus groups. At this point, it might be useful to review the charting summaries and your personal reflexive notes.

Reflection Questions

1. What is the most important takeaway message (or messages) from the focus groups? Consider, for example, the experience of living with a poorly understood chronic illness, experience of healthcare journey).
2. What needs to change/improve?

Mapping Questions

Please review the Framework Map which outlines the findings across all five interviews.

- Are there any interesting or unusual patterns?
- Is anything consistent/inconsistent?
- What does the data tell us/mean?
- Anything else?

You can consider the data across focus groups (horizontal) and within themes or categories (vertical).

Part 2

The aim of this research is to create some recommendations for change. These can be higher level, but ideally, a pathway on how to achieve them can be suggested (i.e. recommendations that are feasible, actionable)

We can base these recommendations on the data and add suggestions from our own knowledge and experience.

Below is a template- but please feel free to expand, add, or change anything in a way that might be more useful to you.

Recommendation	Why is change needed?	What can be done to achieve it?