

Perceptions of fatigue and fatigue management interventions among people with multiple sclerosis: A systematic literature review and narrative synthesis

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Abstract

Background/Aims: Fatigue is the most common symptom of multiple sclerosis.

Evidence supports the effectiveness of fatigue management interventions for people with multiple sclerosis. This review aimed to identify how people with multiple sclerosis experience fatigue and to examine their perceptions of fatigue management interventions (FMIs).

Methods: The review was conducted systematically, searching electronic databases AMED, CINAHL Complete, eBook Nursing Collection, ERIC, Health Source: Nursing/Academic Edition, MEDLINE, APA PsycArticles, and APA PsycInfo for original peer-reviewed empirical research published in English between January 2000 and June 2020. Qualitative and mixed-methods studies that captured the perspectives of people with multiple sclerosis on their fatigue and and/or their perceptions of FMIs were included. A narrative synthesis was used to synthesise the findings.

Findings: Twenty-three qualitative and eight mixed-methods studies were extracted comprising a total of 662 people with multiple sclerosis (relapse remitting = 293; secondary progressive = 129; primary progressive = 73; relapsing progressive = 3; benign = 2; fulminant = 1; MS type not reported = 161). Fatigue was perceived by people with multiple sclerosis as a debilitating symptom of the condition and which they felt impacted adversely on their lives. A lack of understanding from others about multiple sclerosis fatigue was challenging for people with multiple sclerosis. People with multiple sclerosis valued the physical and psychosocial-based content of FMIs and felt FMIs enabled them to legitimise their fatigue and feel more in control of their fatigue.

Conclusions: From the perspective of people with multiple sclerosis, fatigue is a central and debilitating feature of the everyday experience of living with multiple sclerosis. Research on the potential of FMIs to foster control for people with multiple sclerosis to help them manage their fatigue is warranted. Healthcare professionals should consider how they can empower people with multiple sclerosis to educate others about their fatigue. Some evidence being generated for practice might not be sufficiently contextualised to different forms of multiple sclerosis.

Authors' contribution

All authors conceived the study and designed the research strategy. M. P., N. A., and A.W. conducted the search, screened all records and studies, and extracted the data. All authors contributed to the appraisal and synthesis of the studies. All authors contributed to the writing of the manuscript. Drafts were reviewed by all authors for intellectual content. All authors read and approved the final manuscript.

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Conflict of Interest

The authors report no conflicts of interest.

Key words: Multiple Sclerosis, Fatigue, Perceptions, Experiences, Fatigue Management, Rehabilitation

Introduction

Multiple sclerosis is a neurodegenerative autoimmune condition, whereby the immune system attacks myelin in the central nervous system (Bishop & Rumrill, 2015). Over 2.3 million people worldwide have a diagnosis of multiple sclerosis (Multiple Sclerosis International Federation, 2013). The condition can cause a wide range of symptoms including fatigue, cognitive and emotional disturbances, pain, muscle weakness, spasticity, and incontinence (Bishop & Rumrill, 2015; Hyarat et al., 2018). Fatigue is among the most common and disabling symptoms of multiple sclerosis, affecting approximately 90% of those who have the condition (Arewasikporn et al., 2018). Fatigue in multiple sclerosis can be broadly divided into primary and secondary fatigue. Primary multiple sclerosis fatigue originates from a dysfunction of neuronal circuits in the central nervous system secondary to increased inflammation, diffuse demyelination and axonal lesions, and brain atrophy. Secondary multiple sclerosis fatigue is associated with depression, cognitive impairments, deconditioning, and sleep disturbance in multiple sclerosis (Langeskov-Christensen et al., 2017). Fatigue in multiple sclerosis is exacerbated by conditions such as infection, progression of the disease, sustained or excessive activity, warm temperatures, or sleep disturbance (Bakshi, 2003; National Institute for Health and Care Excellence, 2014). People with multiple sclerosis can experience fatigue at all stages of the disease (Isuru et al., 2012; Renoux, 2011).

Broadly speaking, fatigue management interventions (FMIs) in multiple sclerosis refers to both pharmacological and non-pharmacological treatments aimed at the management of fatigue in multiple sclerosis (National Institute for Health and Care Excellence (NICE), 2014). Different pharmacological agents are used for treatment of multiple sclerosis fatigue (e.g., Amantadine, Modafinil) although there has been insufficient evidence that demonstrates their effectiveness (Braley & Chervin, 2010; Khan et al., 2014). Non-pharmacological interventions are broadly

classified into physical-based interventions (e.g., exercise), behavioural and education-based interventions (e.g., lifestyle training, energy conservation, and work simplification) and cognitive and psychological-based interventions (e.g., cognitive training and psychological support). A systematic review of fatigue management in multiple sclerosis (Khan et al., 2014) identified multiple types of non-pharmacological interventions (including exercise, aquatic therapy, cooling therapy, tai chi, energy conservation interventions, mindfulness and cognitive behavioural therapy) that have proven effective in both reducing fatigue and/or for overall management of fatigue. The above interventions can be delivered as specific standalone rehabilitation interventions, but energy conservation, lifestyle education, psychological support and cognitive training are typically combined and delivered in the form of fatigue management programmes. A randomised-controlled trial of a fatigue management programme in multiple sclerosis has been effective in reducing fatigue severity and increasing fatigue self-efficacy in people with multiple sclerosis (Thomas et al., 2013). Non-pharmacological interventions such as energy conservations programmes have lessened the impact of fatigue and increased self-efficacy and quality of life for people with multiple sclerosis (Mathiowetz et al., 2001; 2007).

Healthcare professionals are tasked with understanding the effects of fatigue on people with multiple sclerosis in order to provide interventions aimed at helping them manage their fatigue. However, poor understanding of how multiple sclerosis-related fatigue impacts on the everyday lives of people with multiple sclerosis from their perspective, and how people with multiple sclerosis perceive the effects of FMIs on their fatigue can make the overall clinical management of multiple sclerosis-related fatigue challenging. Despite the evidence which reports on the effectiveness of FMIs, less is known about the lived experience of fatigue among people with multiple sclerosis or about their perceptions of FMIs (Cowan et al., 2020; Mathiowetz & Busch, 2006; Twomey & Robinson, 2010). Hence, we conducted a review to

identify how people with multiple sclerosis experience fatigue and to examine their perceptions of FMIs.

Methods

Search strategy

We conducted a systematic search of literature published on this topic between January 2000 and June 2020. Databases searched included: AMED; CINAHL Complete; eBook Nursing Collection; ERIC; Health Source: Nursing/Academic Edition; MEDLINE; APA PsycArticles; and APA PsycInfo. Search terms were used which related to the condition in question (MS and Multiple Sclerosis), to perspectives (attitude*, perception*, view*, thought*, belief*, opinion*, perspective*, experience*, satisfaction, and personal satisfaction), to participants (service user*, patient* and client*), and to the substantive area of the review (fatigue, fatigue management, energy conservation and intervention). The asterisk in this case was used in order to capture the plural case of these terms. We used these terms in multiple ‘and/or’ combinations. The search terms were agreed between all authors after generating through mutual discussion a preliminary list of search terms focused on capturing experiences and perceptions of fatigue and FMIs in multiple sclerosis. Our rationale for looking at 20 years of research rather than from inception was to search for evidence situated in current contexts of the experience of fatigue and among people with multiple sclerosis. The search was conducted during the period of March to June 2020.

Inclusion/exclusion criteria

Qualitative studies that captured the perspectives of people with multiple sclerosis on their fatigue and their perceptions of FMIs were included. Mixed-methods studies were also included where the qualitative component of these studies reported on the subjective

experience of fatigue and/or of FMIs for people with multiple sclerosis. We did not limit our inclusion to specific types of FMIs or on the basis of duration or content of a FMI. Nor did we limit our inclusion based on participant age, specific type of multiple sclerosis or disease stage, or fatigue assessed. Studies were included regardless of specific aims once they reported on participants' own experiences and perceptions of fatigue and/or FMIs. Only peer-reviewed original studies published in full and in English were included. Studies which focused solely on the views of healthcare professionals, family or caregivers were excluded because the review aimed to capture the perspective of people with multiple sclerosis only. Details of the systematic search are presented in figure 1.

The first three authors ran the search and independently screened each title and abstract based on the inclusion/exclusion criteria. They then systematically cross-checked all results at this point to limit the possibility of omitting records that met criteria for inclusion. Any disagreements regarding inclusion or exclusion based on review of a full-text article were resolved through mutual discussion and multiple readings. A standard template was used to report extraction under the following headings: Study citation; sample (n=) including MS type, study location; methods; focus of study or study aims; and key findings of each study. The template was then refined into a table that tabulated the data concisely under each heading as depicted in Table 1.

Insert Figure 1 here - Figure 1: Systematic search

Insert Table 1 here – Table 1. Summary of studies

Appraisal and synthesis

Although our focus was not to exclude studies based on their quality, all included studies were critically appraised for quality and rigour. We used the Critical Appraisal Programme (CASP)

qualitative checklist (CASP, 2018) to appraise the qualitative studies and the Mixed Methods Appraisal Tool (MMAT) (Hong et al., 2018) to appraise the mixed-methods studies. The CASP is a 10-item checklist for assessing the quality of a qualitative study. The MMAT comprises a five-item checklist for overall appraisal of a mixed-methods study - in addition to a five-item checklist that appraises the qualitative component of a mixed-methods study and a five-item checklist as appropriate to appraise the quantitative component of a mixed-methods study. Neither the CASP nor MMAT are intended for scoring. Rather each criterion is reported as either 'yes', 'no' or 'can't tell'. Using these tools, we found studies to be of medium to high quality in that each study scored 'yes' for majority of criteria assessed. To ensure consistency in our appraisal, each study was appraised by at least two of the four authors.

We conducted a narrative synthesis of the selected studies (Popay et al., 2006). A narrative synthesis adopts a textual approach to the process of synthesis to tell the story of findings from the included studies. First, each of the first three authors looked exhaustively at the evidence in each study that reported on experiences and perceptions of fatigue and/or of fatigue management interventions. We then collated and summarised our evidence from each study. Next, we (all authors) collectively explored relationships in the data (Popay et al., 2006) by comparing the data both within and across the studies. Looking iteratively for similarities and differences in the data resulted in the grouping of findings into categories (Popay et al., 2006) which more broadly encapsulated participants' perceptions of fatigue and fatigue management interventions, how they managed their fatigue, and the factors (including those related to FMIs) which mediated both their experience of fatigue and their management of their fatigue - from their perspective. The grouping of findings into categories or themes and the labelling of them were agreed through multiple rounds of discussion between all authors.

Results

Thirty-one studies (23 qualitative and eight-mixed methods studies) were extracted comprising a total of 662 people with multiple sclerosis (relapse remitting = 293; secondary progressive = 129; primary progressive = 73; relapsing progressive = 3; benign = 2; fulminant = 1; MS type not reported = 161). The characteristics of the studies included in the review and their reported findings in relation to how participants perceived fatigue and FMIs, are presented in table 1. Thirteen studies (Al-Sharman et al., 2018; Aminian et al., 2019; Blaney & Lowe-Strong, 2009; Cowan et al., 2020; Ghahari et al., 2019; Lohne et al., 2010; Kayes et al., 2010; Koffman et al., 2013; Newland et al., 2013; Ploughman et al., 2012; Silverman et al., 2017; Turpin et al., 2018; Yorkston et al., 2003) reported on participants' experiences of fatigue (and/or how they felt they managed their fatigue) but without a focus on FMIs. The remainder of the studies reported on both the experience of fatigue and perceptions of FMIs. Of the studies that reported on MS type, more than half of them focused predominantly on people with relapse-remitting multiple sclerosis. Some qualitative studies did not report on multiple sclerosis type in their sample. Our synthesis resulted in the categorisation of the findings as follows: Feelings of loss; control over fatigue; other's lack of understanding of fatigue; and perceptions of fatigue management interventions.

Feelings of loss

Loss of roles and routine

Fatigue resulted in loss of independence for people with multiple sclerosis, adversely impacting their roles and routines (Newland et al., 2012; Ploughman et al., 2012; Smith et al., 2009). Work performance was affected by also fatigue which frequently led to early retirement and resignation from work (Jellie et al., 2014; Ploughman et al., 2012a; Turpin et al., 2018; Yorkston et al., 2003). In addition, people with multiple sclerosis reported that fatigue impaired their capacity to fulfil responsibilities as a parent or spouse (Cowan et al., 2020; Ploughman et

al., 2012b). Fatigue was identified as a barrier to engaging in many activities, such as social events and physical activities, and was perceived by participants to limit their opportunities and choices in life (Cowan et al., 2020; Ploughman et al., 2012b; Silverman et al. 2017; Turpin et al., 2018).

Loss of control

In many studies, people with multiple sclerosis described fatigue as one of the most distressing and disabling symptoms of multiple sclerosis, depicting fatigue as overwhelming and controlling (Cowan et al., 2020; Koffman et al., 2013; Lohne et al., 2010; Pétrin et al., 2018; Smith et al., 2009, 2011; Thomas et al., 2010). People with multiple sclerosis felt they were losing control of many aspects of their lives, such as spontaneity and ability to plan because of the variable nature, unpredictability, and sudden onset of fatigue (Smith et al., 2009, 2011; Thomas et al., 2010; Turpin et al., 2018; Wendebourg et al., 2016). As a result, fatigue was seen by people with multiple sclerosis to have an adverse effect on their wellbeing (Kayes et al., 2010; Wendebourg et al., 2016). However, perceived control over fatigue was deemed important by people and had the potential to improve their wellbeing (Mulligan et al., 2016; Smith et al., 2009).

Loss of ability

During periods of fatigue, people with multiple sclerosis experienced a decline in their physical abilities which manifested through muscle weakness, imbalance, immobility, and exhaustion (Al-Sharman et al., 2018; Aminian et al., 2019; Cowan et al., 2020; Kayes et al., 2010; Koffman et al., 2013; Lohne et al., 2010; Newland et al., 2012; Ploughman et al., 2012a; Smith et al., 2011). Fatigue also affected cognition, which negatively impacted their ability to think clearly and implement fatigue management strategies (Newland et al., 2012; Yorkston et al., 2003).

Furthermore, people with multiple sclerosis reported that fatigue affected their ability to communicate because it caused dysarthria and impacted their language processing and word finding ability (Blaney & Lowe-Strong, 2009; Lohne et al., 2010; Newland et al., 2012; Yorkston et al., 2003). Exacerbations of fatigue were perceived by people with multiple sclerosis as a deterioration of their health, made it difficult for them to make plans, and as a symptom which resulted in feelings of depression (Smith et al., 2009, 2011).

Control over fatigue

Legitimisation of fatigue and fatigue management strategies

People with multiple sclerosis reported that FMIs played an important role in validating and legitimising their experiences of fatigue and current fatigue management strategies (Holberg & Finlayson, 2007; Mulligan et al., 2016; Pétrin et al., 2018; Twomey & Robinson, 2010). Education about fatigue in multiple sclerosis increased feelings of control over symptoms among people with multiple sclerosis (Holberg & Finlayson, 2007; Jellie et al., 2014; Twomey & Robinson, 2010). Prior to engaging in FMIs, many people with multiple sclerosis had found it difficult to determine whether fatigue was related to their condition or simply a manifestation of the inclination not to engage in activity. However, learning about fatigue enabled people with multiple sclerosis to shift the blame from themselves to their condition, thereby reducing feelings of guilt (Akbar et al., 2018; Holberg & Finlayson, 2007).

Acceptance

People with multiple sclerosis considered acceptance to be a key element of coping with fatigue, which involved acknowledging their changing ability and adjusting expectations of themselves (Pétrin et al., 2018; Ploughman et al., 2012b; Smith et al., 2009). Acceptance was described as a gradual process rather than a conscious effort, mediated by the individualised

experience of fatigue (Ploughman et al., 2012b; Smith et al., 2009). Due to the progressive nature of multiple sclerosis, acceptance could be challenging, as people were required to repeatedly adapt to decreasing levels of ability (Kayes et al., 2010; Silverman et al., 2017). People with multiple sclerosis found that upholding a positive attitude, such as challenging negative thoughts and maintaining a sense of humour, was essential to feeling accepting of multiple sclerosis and multiple sclerosis-related fatigue (Blaney & Lowe-Strong, 2009; Ghahari et al., 2019; Ploughman et al., 2012a; Silverman et al., 2017; Smith et al., 2011; Thomas et al., 2010).

Readiness for change

Acceptance of fatigue was described by people with multiple sclerosis as important in facilitating a readiness to implement fatigue management strategies (Holberg & Finlayson, 2007; Yorkston et al., 2003). A variety of factors influenced individuals' preparedness for change, such as social support, self-reflection, education and enhanced understanding of fatigue, and increased self-confidence to manage fatigue (Clarke & Coote, 2015; García-Jalón et al., 2013; Mulligan et al., 2016; Ploughman et al., 2012a). Being ready for change was subjective to the individual and although people with multiple sclerosis continued to engage in FMIs, they did so at their own pace until they felt ready to adapt their habits and take control of their fatigue (Moss-Morris et al., 2012). Following completion of FMIs, perceptions of enhanced control over fatigue, and increased frustration tolerance aided participants in their readiness for change (Akbar et al., 2018; Moss-Morris et al., 2012; Pétrin et al., 2018).

Feeling empowered

Feeling empowered was key for people with multiple sclerosis to gain control over fatigue. Through participation in FMIs, people with multiple sclerosis engaged in active problem-

solving to adapt fatigue management strategies to their own lives (Akbar et al., 2018; Barlow et al., 2009; Hersche et al., 2019; Holberg & Finlayson, 2007). Furthermore, many individuals reported positive impacts of FMIs on addressing loss of ability associated with various symptoms of multiple sclerosis (Akbar et al., 2018; Clarke & Coote, 2015; Flensner & Lindencrona, 2002). Participants felt FMIs facilitated coping and planning because of the peer support they received and goal-setting skills they acquired in FMIs to participate in everyday life (Barlow et al., 2009; Clarke & Coote, 2015). Being able to manage day-to-day challenges was reported to be of significant benefit among people with multiple sclerosis, and this newfound empowerment made participants feel an increased sense of control (Mulligan et al., 2016; Smith et al., 2009). As a result, people with multiple sclerosis re-engaged in activities in which they had previously felt no longer capable and felt more competent to re-engage in meaningful activities (Barlow et al., 2009; Clarke & Coote, 2015; Flensner & Lindencrona, 2002).

Other's lack of understanding of multiple sclerosis fatigue

Society's lack of understanding of multiple sclerosis fatigue

People with multiple sclerosis felt society was unaccepting and unaccommodating of people with disabilities, and that society valued independence over dependence (Blaney & Lowe-Strong, 2009). Consequently, this made receiving assistance, living openly with a disability, and using fatigue management strategies difficult for some (Silverman et al., 2017). People with multiple sclerosis suggested that society's lack of understanding of fatigue (and its impact) was perpetuated by the invisible nature of the symptom (Lohne et al., 2010). Due to the challenge of explaining fatigue to others, some people with multiple sclerosis concealed it to avoid portraying themselves as having a disability (Blaney & Lowe-Strong, 2009; García-Jalón

et al., 2013; Turpin et al., 2018). Conversely, others felt that it was not their responsibility if people did not know about fatigue (Turpin et al., 2018).

Friends' and relatives' lack of understanding of multiple sclerosis fatigue

Some people with multiple sclerosis felt they experienced limited support from their friends and family due to their lack of understanding or awareness of fatigue (Blaney & Lowe-Strong, 2009; Cowan et al., 2020; Smith et al., 2011; Thomas et al., 2010; Wendebourg et al., 2016). Participants were fearful of being viewed as a nuisance for cancelling or changing plans and some hesitated to tell family and friends about their fatigue (Turpin et al., 2018). Indeed, some people described their social circles narrowing due to fatigue and others' perceptions of their condition (Ploughman et al., 2012a). As a result, limited opportunities to socialise often led to isolation. Lack of social support had from the perspective of people with multiple sclerosis, an adverse impact on their quality of life and wellbeing (Ploughman et al., 2012b).

People with multiple sclerosis found it difficult to explain to others the impact of fatigue on their lives and its difference from regular tiredness (Pétrin et al., 2018; Turpin et al., 2018) and some participants expressed frustration when others pushed them to do more (Smith et al., 2011). Communication was deemed by participants as necessary to enable relations to understand their experience of fatigue, which reduced the perceived need to meet their expectations (Ghahari et al., 2019; Holberg & Finlayson, 2007; Thomas et al., 2010). Additionally, delegation of tasks was preferred over asking for help, as people with multiple sclerosis felt in control. After participating in FMIs, people with multiple sclerosis reported improved family dynamics and enhanced communication with family (Mulligan et al., 2016).

Perceptions of FMIs

Perceptions of group interventions

People with multiple sclerosis reported that peer support and meeting others with similar experiences were essential for coping with fatigue and adjusting to life with multiple sclerosis (Clarke & Coote, 2015; García-Jalón et al., 2013). Most people expressed a preference for group interventions over individualised training (García-Jalón et al., 2013; Twomey & Robinson, 2010; Wendebourg et al., 2016). The group setting brought about friendships, support and feedback, reduced isolation among members and boosted morale among people with multiple sclerosis (Hersche et al., 2019; Mathiowetz & Busch, 2006; Mulligan et al., 2016; Thomas et al., 2010). Peer support also motivated people with multiple sclerosis to continue with the intervention and keep up with assigned work (Barlow et al., 2009). This resulted in people joining community groups to continue socialising with others (Twomey & Robinson et al., 2010). However, some participants found it taxing to meet others who were at different stages of disease progression, as this highlighted the challenges that they may face in the future (Mulligan et al., 2016). In some circumstances, people with multiple sclerosis felt that an overly diverse group led to unmet expectations and goals for some members of the group (Finlayson & Holberg, 2007; Mulligan et al., 2016).

Perceptions of fatigue management strategies

Primary reasons which people with multiple sclerosis identified the need to engage in FMIs were to acquire strategies for self-management and to take personal responsibility for managing multiple sclerosis symptoms (Akbar et al., 2018). Following completion of FMIs, people with multiple sclerosis described a wide range of strategies they had learned which enhanced their control over fatigue, such as managing energy, balancing a schedule, planning and pacing activities, breathing exercises, regular exercise and realistic goal-setting (Barlow et al., 2009; Mathiowetz & Busch, 2006; Thomas et al., 2010; Twomey & Robinson, 2010). Participating in FMIs was described by people with multiple sclerosis as beneficial in enabling them to

reflect on their own routines and develop an individualised approach to implementing fatigue management strategies and lifestyle changes (Hersche et al., 2019; Moss-Morris et al., 2012). They also found rest to be a beneficial fatigue management strategy when incorporated into their daily routine (Cowan et al., 2020; Ghahari et al., 2019; Thomas et al. 2010). Various relaxation techniques, including sleep, meditation and muscle relaxation were reported by participants to facilitate rest while also enabling a positive outlook and improving their clarity of thought (Barlow et al., 2009).

Some FMIs involved engagement in exercise which provided participants with a sense of increased strength, stamina, and fitness. Participation in physical activities was reported by people with multiple sclerosis to improve health and wellbeing and increase their perceived control over fatigue (Aminian et al., 2019; Smith et al., 2009, 2011). Participants indicated that participation in exercise groups such as aqua jogging and yoga enabled them to regain a sense of normalcy. Engaging in regular exercise was perceived by participants as useful to maintain employment (Smith et al., 2011). Indeed, some participants felt that medical professionals should place greater emphasis on the benefits of exercise in multiple sclerosis (Kayes et al., 2010).

Perceptions of method of delivery

Healthcare professionals were valued by people with multiple sclerosis as facilitators of FMIs because they trusted their advice and received accurate information about multiple sclerosis (Finlayson & Holberg, 2007). However, some people wanted more freedom to listen to their own bodies in implementing fatigue management strategies (Smith et al., 2009). Lay facilitators who had multiple sclerosis themselves were perceived as understanding and sympathetic, but some people with multiple sclerosis suggested it would also be helpful to have a healthcare professional who could provide more in-depth information (Barlow et al., 2009).

Some participants felt it was important to take an active role within group interventions, for example, having an input into the intervention and homework content and actively participating during each session (Twomey & Robinson, 2010; Wendebourg, 2016). However, some people with multiple sclerosis also raised concerns about intervention facilitation. For instance, some participants who participated in FMIs found it stressful to complete homework following sessions and to practically implement fatigue management strategies into daily life (García-Jalón et al., 2013, Mathiowetz & Busch, 2006).

Discussion

Participants' descriptions of fatigue in this review were similar across studies in that fatigue was described as all-consuming and overwhelming, and interfered in many aspects of their lives. Participants across studies described the experience of a decline in their physical and cognitive ability in the context of fatigue and they experienced anxiety and fear because of fatigue. Their experiences are consistent with descriptions of multiple sclerosis fatigue in the literature, as a symptom which can manifest both mentally and physically and can impact an individual's social, emotional, and cognitive functioning (Krupp, 2003; Morrison & Stuifbergen, 2016; Powell et al., 2017). Multiple sclerosis fatigue is understood to impact people's ability to engage in everyday life (Krupp, 2003; Morrison & Stuifbergen, 2016; Van Kessel & Moss-Morris, 2006), and which is confirmed from the perspective of people with multiple sclerosis in this review. Indeed, one of the key findings of this review is the extent to which people's (with multiple sclerosis) perceptions of fatigue are embedded in their overall experience of multiple sclerosis because fatigue can from their perspective, exacerbate the physical and cognitive symptoms of their condition.

Participants across studies in this review perceived fatigue as controlling and unpredictable, and which they felt had an adverse effect on their psychological health. It is important to note

that fatigue can be significantly influenced by people's subjective thoughts and beliefs (Krupp, 2003). Perceptions of being controlled by fatigue also have a strong association with multiple sclerosis fatigue (Van Kessel & Moss-Morris, 2006). For example, people with multiple sclerosis who worry that their symptoms signify a deterioration of health are more likely to experience fatigue (Morrison & Stuifbergen, 2016). In contrast, having a sense of control over fatigue can help people with multiple manage the symptom (Van Kessel & Moss-Morris, 2006). In this review, participants described a sense of empowerment and enhanced self-efficacy upon gaining some control over their fatigue. The experiences of fatigue among people with multiple sclerosis in this review are comparable to those of people with cancer, rheumatoid arthritis, and stroke (Mock, 2011; Repping-Wuts et al., 2008; Whitehead et al., 2016). A lack of understanding in society about multiple sclerosis fatigue and participants' perceived stigma in relation to their fatigue were seen by participants as significant barriers to implementing fatigue management strategies, and in some cases caused them to try and conceal their fatigue. The findings resonate with those of Valeras (2010) who also found that people with various 'hidden' symptoms can conceal their condition because of the stigma they perceive in relation to their disability.

The review captured perspectives among people with multiple sclerosis on a wide variety of FMIs, including for example energy conservation and self-management courses, exercise programs, vocational rehabilitation, and the effects of a cooling suit on managing fatigue. Of note, group-based interventions primarily involving self-management and education were highly regarded by participants. However, perceptions of FMIs were captured primarily in studies involving self-management and education, and only a few interventions that participants reported on comprised other contents (e.g., exercise). Patient perceptions of FMIs are perhaps contextualised primarily within a self-management approach to fatigue. The findings of this review suggest overall, positive perceptions of FMIs among participants. From

the perspective of people with multiple sclerosis, FMIs helped them to manage their fatigue and feel in control of their fatigue. These findings are consistent with views on fatigue management interventions among other diagnostic groups (e.g. acquired brain injury, rheumatoid arthritis) (Cooper et al., 2009; Dures et al., 2012).

Across some studies, participant heterogeneity for multiple sclerosis type was limited. Although 10 of the 31 studies did not report on multiple sclerosis type for their sample, of the remainder that did, 12 studies sampled predominantly people with relapse-remitting multiple sclerosis (Akbar et al., 2018; Aminian et al., 2019; Clarke & Coote, 2015; Finlayson & Holberg, 2007; Ghahari et al., 2019; Jellie et al., 2014; Mathiowetz & Busch, 2006; Mulligan et al., 2016; Newland et al., 2012; Petrin et al., 2018; Smith et al., 2009; Thomas et al., 2010). Only seven of the extracted studies included a substantial mix of people with different types of multiple sclerosis (Cowan et al., 2020; Flensner & Lindencrona, 2002; Kayes et al., 2010; Koffman et al., 2013; Moss-Morris et al., 2012; Smith et al., 2011; Twomey & Robinson, 2010). Two studies (García-Jalón et al., 2013; Ploughman et al., 2012a, b) focused predominantly on people with progressive multiple sclerosis. The findings of this review suggest that some research on the lived experience of fatigue among people with multiple sclerosis and/or on their perceptions of FMIs, may focus somewhat less on people with progressive forms of multiple sclerosis.

Strengths and limitations

Only peer-reviewed original research published in full and in English was included in the review and we limited the review to a 20-year period. A wider ‘scoping’ review from inception, incorporating in addition the grey literature, would have reported more evidence on this topic. Nonetheless, limiting our review to original research allowed us to appraise the methodological quality of each item of evidence included in the review. Limiting our review

to the last 20 years allowed us to synthesise studies contextualised to contemporary experiences of fatigue and FMIs among people with multiple sclerosis. As stated, we generated through mutual discussion a list of search terms focused on capturing experiences and perceptions of fatigue and FMIs in multiple sclerosis and so there is a possibility that some studies that met the inclusion criteria were not found. Neither did we undertake a hand search for articles based on reference lists of articles included in the review. That said, we were highly systematic in our extraction of the studies. Three of the four authors independently screened/assessed each title and abstract and then systematically cross-checked all results at this point to limit the possibility of omitting records that met criteria for inclusion. Moreover, full-text reviews for a high number of results were completed by the first three authors and any disagreements regarding inclusion or exclusion following a full-text review were resolved through multiple readings and discussion between the first three authors. All authors contributed to the synthesis of the studies included in the review.

Conclusions

From the perspective of people with multiple sclerosis, fatigue can have a significant impact on their everyday life and wellbeing. However, having a perceived sense of control over fatigue can be valued highly by people with multiple sclerosis and can from their perspective, impact positively on their health and wellbeing. FMIs can be viewed by people with multiple sclerosis as a valuable tool to help them adapt to their fatigue because they can empower them to manage their fatigue and help them feel in more control of their fatigue. Further research on how people with multiple sclerosis can best adapt to their fatigue, in particular on the role of FMIs in legitimising fatigue and fostering control for people with multiple sclerosis, would be beneficial to expand our understanding of how people with multiple sclerosis can best manage their fatigue in their everyday life. Particular attention to the medium of delivery could be

useful to understand how FMIs can help people with multiple sclerosis gain a sense of control over their fatigue.

Across studies, people with multiple sclerosis highlighted the negative impact of fatigue on their roles and daily routine and the importance of addressing the physical and psychological impact of fatigue. Of the studies that reported on MS type, more than half of them focused predominantly on people with relapse-remitting multiple sclerosis. Progressive multiple sclerosis often results in increasing levels of physical and cognitive disability which can limit ability to engage with physical and cognitive-behavioural components of FMIs. Some more focus on the experience of fatigue among people with progressive multiple sclerosis and their needs in relation to FMIs, could be beneficial. Some of the evidence that is currently being generated for practice might not be sufficiently sensitive to different forms of multiple sclerosis.

Engaging with healthcare professionals who were knowledgeable about multiple sclerosis and fatigue in multiple sclerosis was important for participants. However, often of more significance for people with multiple sclerosis was the lack of awareness of non-healthcare professionals about multiple sclerosis fatigue. It is beneficial to consider how FMIs can enable people with multiple sclerosis not only to manage their fatigue but also how they can educate others about their fatigue. Empowering people with multiple sclerosis to educate others about their fatigue could address their difficulty of coping with the stigma they may perceive in relation to their fatigue.

References

Akbar N, Turpin K, Petrin J, et al. A pilot mixed-methods evaluation of MS INFoRm: A self-directed fatigue management resource for individuals with multiple sclerosis. *Int J Rehabil Res.* 2018;41(2):114-121.

Al-Sharman A, Khalil H, Nazzal M, et al. Living with multiple sclerosis: A Jordanian perspective. *Physiother Res Int.* 2018;23(2):1-9.

Aminian S, Ezeugwu VE, Motl RW, et al. Sit less and move more: Perspectives of adults with multiple sclerosis. *Disabil Rehabil.* 2019;41(8):904-911.

Arewasikporn A, Turner A, Alschuler K, et al. Cognitive and affective mechanisms of pain and fatigue in multiple sclerosis. *Health Psychol.* 2018;37(6):544-552.

Bakshi R. Fatigue associated with multiple sclerosis: Diagnosis, impact and management. *Mult Scler.* 2003;9(3):219-227.

Barlow J, Edwards R, Turner A. The experience of attending a lay-led, chronic disease self-management programme from the perspective of participants with multiple sclerosis. *Psychol Health.* 2009;24(10):1167-1180.

Bishop M, Rumrill PD. Multiple sclerosis: Etiology, symptoms, incidence and prevalence, and implications for community living and employment. *Work.* 2015;52(4):725-734.

Blaney BE, Lowe-Strong A. The impact of fatigue on communication in multiple sclerosis: The insider's perspective. *Disabil Rehabil.* 2009;31(3):170-180.

Braley TJ, Chervin RD. Fatigue in multiple sclerosis: Mechanisms, evaluations and treatment. *Sleep.* 2010;33(8):1061-1067.

Clarke R, Coote S. Perceptions of participants in a group, community, exercise programme for people with multiple sclerosis. *Rehabil Res Pract*. 2015;2015:123494.

Cooper J, Reynolds F, Bateman A. An evaluation of a fatigue management intervention for people with an acquired brain injury: An exploratory study *Br J Occup Ther*. 2009;72(4):174-179.

Cowan CK, Pierson JM, Leggat SG. Psychosocial aspects of the lived experience of multiple sclerosis: Personal perspectives. *Disabil Rehabil*. 2020;42(3):349-359.

Critical Appraisal Skills Programme. CASP Qualitative Checklist. [internet]; 2018. Available from <https://casp-uk.net/casp-tools-checklists/> (accessed 12 February 2020)

Dures E, Kitchen K, Almeida A, et al. “They didn’t tell us, they made us work it out for ourselves”: Patient perspectives of a cognitive-behavioural program for rheumatoid arthritis fatigue. *Arthritis Care Res*. 2012;64(4):494-501.

Finlayson M, Holberg C. Evaluation of a teleconference-delivered energy conservation education program for people with multiple sclerosis. *Can J Occup Ther*. 2007;74(4):337–347.

Flensner G, Lindencrona C. The cooling-suit: Case studies of its influence on fatigue among eight individuals with multiple sclerosis. *J Adv Nurs*. 2002;37(6):541-550.

García-Jalón E, Lennon S, Peoples L, et al. Energy conservation for fatigue management in multiple sclerosis: A pilot randomized controlled trial. *Clin Rehabil*. 2013;27(1):63-74.

Ghahari S, Forwell SJ, Suto MJ, et al. Multiple sclerosis self-management model: Personal and contextual requirements for successful self-management. *Patient Educ Couns*. 2019;102(5):1013-1020.

Hersche R, Weise A, Michel G, et al. Development and preliminary evaluation of a 3-week inpatient energy management education program for people with multiple sclerosis-related fatigue. *Int J MS Care*. 2019;21(6):265-274.

Holberg C, Finlayson M. Factors influencing the use of energy conservation strategies by persons with multiple sclerosis. *Am J Occup Ther*. 2007;61(1):96-107.

Hong QN, Fàbregues S, Bartlett G, et al. The Mixed Methods Appraisal Tool (MMAT) version 2018 for information professionals and researchers. *Educ Inf*. 2018;34(4):285-291.

Hyarat SY, Subih M, Rayan A, et al. Health related quality of life among patients with multiple sclerosis: The role of psychosocial adjustment to illness. *Arch Psychiatr Nurs*. 2018;33(1):11-16.

Isuru I, Constantinescu CS, Gran B. Fatigue in multiple sclerosis: A brief review. *J Neurol Sci*. 2012;323(1-2):9-15.

Jellie B, Sweetland J, Riazi A, et al. Staying at work and living with MS: A qualitative study of the impact of a vocational rehabilitation intervention. *Disabil Rehabil*. 2014;36(19):1594-1599.

Kayes N, McPherson K, Taylor D, et al. Facilitators and barriers to engagement in physical activity for people with multiple sclerosis: A qualitative investigation. *Disabil Rehabil*. 2010;33(8):625-642.

Khan F, Amatya B, Galea M. Management of fatigue in persons with multiple sclerosis. *Front Neurol*. 2014;5:177.

Koffman J, Gao W, Goddard C, et al. Progression, symptoms and psychosocial concerns among those severely affected by multiple sclerosis: A mixed-methods cross-sectional study of Black Caribbean and White British people. *PloS one*. 2013;8(10):e75431.

Krupp LB. Fatigue in multiple sclerosis. *CNS Drugs*. 2003;17(4):225-234

Langeskov-Christensen M, Bisson EJ, Finlayson ML, et al. Potential pathophysiological pathways that can explain the positive effects of exercise on fatigue in multiple sclerosis. A scoping review. *J Neurol Sci*. 2017;373:307-320.

Lohne V, Aasgaard T, Caspari S, et al. The lonely battle for dignity: Individuals struggling with multiple sclerosis. *Nurs Ethics*. 2010;17(3):301-311.

Mathiowetz V, Busch M. Participant evaluation of energy conservation course for people with multiple sclerosis. *Int J Mult Scler Care*. 2006;8(3):98-106.

Mathiowetz VG, Matuska KM, Finlayson ML, et al. One-year follow up to a randomised controlled trial of an energy conservation course for persons with multiple sclerosis. *Int J Rehabil Res*. 2007;30(4):305-13.

Mathiowetz V, Matuska KM, Murphy ME. Efficacy of an energy conservation course for persons with multiple sclerosis. *Arch Phys Med Rehabil*. 2001;82(4):449-456.

Mock V. Fatigue management. *Cancer*. 2011;92(S6):1699-1707.

Morrison JD, Stuijbergen AK. Predictors of fatigue impact in persons with long-standing multiple sclerosis. *J Neurosci Nurs*. 2016;48(3):143-150.

Moss-Morris R, McCrone P, Yardley L, et al. A pilot randomised controlled trial of an internet-based cognitive behavioural therapy self-management programme (MS Invigor8) for multiple sclerosis fatigue. *Behav Res Ther.* 2012;50(6):415-421.

Mulligan H, Wilkinson A, Snowdon J. Perceived impact of a self-management program for fatigue in multiple sclerosis. *Int J MS Care.* 2016;18(1):27-32.

Multiple Sclerosis International Federation. Atlas of MS 2013: Mapping Multiple Sclerosis Around the World. [internet]. London: UK; 2013. Available from <https://www.msif.org/wp-content/uploads/2014/09/Atlas-of-MS.pdf> (accessed 14 April 2020).

National Institute for Health and Care Excellence (NICE). Multiple sclerosis in adults: Management. [internet]. London: UK; 2014. Available from: <https://www.nice.org.uk/guidance/CG186/chapter/1-Recommendations#ms-symptom-management-and-rehabilitation-2> (accessed 27 April 2020).

Newland PK, Thomas FP, Riley M, et al. The use of focus groups to characterize symptoms in persons with multiple sclerosis. *J Neurosci Nurs.* 2012;44(6):351-357.

Pétrin J, Akbar N, Turpin K, et al. The experience of persons with multiple sclerosis using MS INFoRm: An interactive fatigue management resource. *Qual Health Res.* 2018;28(5):778-788.

Ploughman M, Austin M, Murdoch M, et al. Factors influencing healthy aging with multiple sclerosis: A qualitative study. *Disabil Rehabil.* 2012a;34(1):26-33.

Ploughman M, Austin MW, Murdoch M, et al. The path to self-management: A qualitative study involving older people with multiple sclerosis. *Physiother Can.* 2012b;64(1):6-17.

Popay J, Roberts H, Sowden A, et al. Guidance on the conduct of narrative synthesis in systematic reviews. *A Product from the ESRC Methods Programme, Version 1*, b92; 2006.

Powell DJ, Lioffi C, Schlotz W, et al. Tracking daily fatigue fluctuations in multiple sclerosis: Ecological momentary assessment provides unique insights. *J Behav Med.* 2017;40(5):772-83.

Renoux C. Natural history of multiple sclerosis: Long-term prognostic factors. *Neurol Clin.* 2011;29(4):293-308.

Repping-Wuts H, Uitterhoeve R, van Riel P, et al. Fatigue as experienced by patients with rheumatoid arthritis (RA): A qualitative study. *Int J Nurs Stud.* 2008;45(7):995-1002.

Silverman AM, Verrall AM, Alschuler KN, et al. Bouncing back again, and again: A qualitative study of resilience in people with multiple sclerosis. *Disabil Rehabil.* 2017;39(1):14-22.

Smith C, Hale L, Olson K, et al. How does exercise influence fatigue in people with multiple sclerosis? *Disabil Rehabil.* 2009;31(9):685-692.

Smith C, Olson K, Hale LA, et al. How does fatigue influence community-based exercise participation in people with multiple sclerosis? *Disabil Rehabil.* 2011;33(23-24):2362-2371.

Thomas S, Thomas PW, Kersten P, et al. A pragmatic parallel arm multi-centre randomised controlled trial to assess the effectiveness and cost-effectiveness of a group-based fatigue management programme (FACETS) for people with multiple sclerosis. *J Neurol Neurosurg Psychiatry.* 2013;84(10):1092-1099.

Thomas S, Thomas PW, Nock A, et al. Development and preliminary evaluation of a cognitive behavioural approach to fatigue management in people with multiple sclerosis. *Patient Educ Couns.* 2010;78(2):240-249.

Turpin M, Kerr G, Gullo H, et al. Understanding and living with multiple sclerosis fatigue. *Br J Occup Ther.* 2018;81(2);82-89.

Twomey F, Robinson K. Pilot study of participating in a fatigue management programme for clients with multiple sclerosis. *Disabil Rehabil.* 2010;32(10):791-800.

Valeras A. “We don’t have a box”: Understanding hidden disability identity utilizing narrative research methodology. *Disability Studies Quart.* 2010; 30(3/4).

Van Kessel K, Moss-Morris R. Understanding multiple sclerosis fatigue: A synthesis of biological and psychological factors. *J Psychosom Res.* 2006;61(5):583-585.

Wendebourg MJ, Feddersen LK, Lau S, et al. Development and feasibility of an evidence-based patient education program for managing fatigue in multiple sclerosis: The “Fatigue Management in MS” program (FatiMa). *Int J MS Care.* 2016;18(3):129-137.

Whitehead L, Unahi K, Burrell B, et al. The experience of fatigue across long-term conditions: A qualitative meta-synthesis. *J Pain Symptom Manage.* 2016;52(1):131-143.

Yorkston K, Johnson K, Klasner E, et al. Getting the work done: A qualitative study of individuals with multiple sclerosis. *Disabil Rehabil.* 2003;25(8):369-379.

Table 1. Summary of studies

Study	n=	Location	Methods	Primary Focus of Study	Findings
Akbar et al., 2018	n=23 (RRMS=16; PPMS=3; SPMS=4) Mean age: 48.8 years	Canada (specific location not reported)	Mixed methods Qualitative component: semi-structured interviews and content analysis Pilot Study	To explore the need for further research into the intervention MS INFoRm; to determine if PwMS experienced improvements in fatigue management, self-efficacy, and quality of life after participation in this FMI.	Participants found the fatigue management strategies useful. Participants reported self-perceived improvements in communication, quality of life, and knowledge about fatigue management after participation in the FMI.
Al-Sharman et al., 2018	n=16 (MS type not reported) Mean age: 36.6 years, age range: 22-57 years	Irbid & Amman, Jordan	Qualitative; focus groups and thematic analysis	To gain insight into the everyday experiences of PwMS and the challenges they face which can impact on their lives.	Participants identified three main challenges associated with the everyday experience of MS including physical decline, psycho-social withdrawal, and fear of the future. Fatigue underpinned physical decline and psycho-social withdrawal.
Aminian et al., 2019	n=15 (RRMS=10; PPMS=2; SPMS=3) Mean age: 43.1 years, age range: 23-61 years	Edmonton, Canada	Qualitative; semi-structured interviews and thematic analysis	To explore the perspectives of adults with MS about sedentary behaviour, physical activity, and ways to change behaviour.	Participants were aware of the benefits of physical activity for their overall health and in the management of fatigue and muscle stiffness. Due to fatigue, they often chose sitting to regain their energy. Barriers to mobility included perceived fear of losing balance and embarrassment while walking. Activity monitoring, educational and individualised programs were motivational strategies for participants.
Barlow et al., 2009	n=10 (MS type not reported) Age range: 35-60 years	Coventry, UK	Qualitative; semi-structured interviews and framework analysis	To explore the perceived value and experience of attending a chronic disease self-management course among people living with MS.	The FMI provided participants with new fatigue management strategies, but some participants also felt the course focused on strategies they already used. Peer support was valued by participants. The lay facilitator was perceived as beneficial by participants, but they also identified the need for professional input. Participants requested more culturally sensitive content for the course.

Blaney & Lowe-Strong, 2009	n=10 (MS type not reported) Mean age: 54.6 years, age range: 45-64 years	Ulster, UK	Qualitative; phenomenological approach; in-depth interviews and inductive analysis	To explore and understand the insider's perspective of the impact of fatigue on communication in MS.	Fatigue impacted communication impairment, participation, and activity for participants. Barriers to participation arose from the complex interplay between the range of impairments experienced by an individual, the coping strategies they employed and by other people's attitudes towards fatigue.
Clarke & Coote, 2015	n=14 (RRMS=11; SPMS=3) Mean age: 53.9 years	Limerick, Ireland	Qualitative; focus groups and thematic analysis	To explore perceptions among PwMS of a 10-week community-based group exercise programme including factors affecting participation in the programme.	Participation in the exercise programme provided physical and psychological improvements with functional carry-over reported. The group format provided motivation and peer support to participants and participants found it difficult to sustain motivation post-intervention without group support. Participants valued having a professional facilitator.
Cowan et al., 2020	n=15 (RRMS=4; PPMS=6; SPMS=4; Fulminant MS=1) Mean age: 45.6 years, age range: 25-64 years	Melbourne, Australia	Qualitative; semi-structured interviews and thematic analysis	To explore the perspectives of PwMS on the psychosocial aspects of living with MS following inpatient rehabilitation; to examine the experiences of rehabilitation hospital stay and discharge home among PwMS.	Participants considered fatigue to be a major issue for them and they felt it limited their ability to maintain employment and to engage in activities of daily living. Participants experienced both physical and mental fatigue, which resulted in loss of independence and feeling fearful about the future. Participants perceived inpatient rehabilitation as beneficial in educating them about fatigue management strategies as well as providing guidance on exercise and healthy eating.
Finlayson & Holberg, 2007	n=31 (HCPs n=3; PwMS n=28) [RRMS=17; PPMS=3; SPMS=3; Not known=5] MS participant mean age: 47 years, age	Chicago, USA	Qualitative; semi-structured interviews and pragmatic evaluation approach Pilot Study	To explore the strengths and limitations of delivering FMI through the medium of teleconference.	Teleconference was a favourable method of FMI delivery for participants. Social support and normalisation of fatigue were valued by participants. Participants found the information provided was useful for their everyday lives. Technical issues, lack of sharing and homework proved a challenge for some participants. The diverse range of ability levels and stages of MS disease progression among group members impacted negatively on intervention experiences for some participants. A longer intervention duration was desired by some participants.

	range: 22-64 years				
Flensner & Lindencrona, 2002	n=8 (RRMS=1; RPMS=2; SPMS=5) Mean age: 50 years	Western Sweden	Mixed methods Qualitative component: open-ended/semi-structured interviews and thematic analysis Pilot Study	To explore if the use of a cooling suit by individuals with MS influenced their experience of fatigue and consequent restrictions in daily life.	Participants reported a reduction in fatigue during the test period and experienced fatigue less often and for shorter periods. The cooling suit was perceived to promote balance and mobility, and improve cognitive fatigue as well as physical fatigue. Participants reported that the cooling suit improved their mental health and well-being and enabled them to engage in meaningful everyday activities.
García-Jalón et al., 2013	n=23 (RRMS=5; PPMS=3; SPMS=13; Not known=2) Mean age: 48.9 years	Northern Ireland, UK	Mixed methods Qualitative component: structured interviews and qualitative analysis Pilot Study	To develop and test the feasibility and effectiveness of an energy conservation programme to manage fatigue in MS.	Peer support was valued by all participants and education enhanced the long-term effects of the FMI. Energy conservation was perceived as helpful and relevant to daily living. Fatigue was reported as a barrier to a balanced lifestyle and impacted quality of life for participants. Some practical-based activities of the course were difficult for participants to implement into their daily routine.
Ghahari et al., 2019	n=18 (RRMS=9; PPMS=3; SPMS=3; Not known=3) Mean age: 49.8 years; age range: 24-70 years	Canada (specific location not reported)	Qualitative; grounded theory approach; semi-structured interviews and grounded theory analysis	To explore self-management strategies used by PwMS to inform the development of a MS self-management model.	The resulting model included self-management strategies classified under six person-related categories and three context-related categories. Fatigue was one of the most common symptoms highlighted by PwMS. Among the person-related self-management strategies, planning and prioritising were perceived to be important in managing fatigue. In context-related categories, participants expressed a need for healthcare professionals to provide greater support with day-to-day fatigue management.
Hersche et al., 2019	n=12	Valens, Switzerland	Qualitative; focus groups and content analysis	To develop a group-based Inpatient Energy Management Education (IEME) program for PwMS; to complete an IEME	The IEME programme was deemed feasible and effective in an inpatient setting. Participants perceived the format and content of the intervention appropriate to learn about MS symptoms and behavioural strategies. Participants also felt the programme increased their perceived sense of control. However, participants revealed their confidence was

	(PwMS=9; OTs=3) (MS type not reported) MS participant mean age: 48 years, age range: 32-56 years		Pilot Study	pilot program to evaluate the experience of participants and of occupational therapists (OTs) facilitating the programme.	unstable in relation to implementing energy conservation strategies in their daily routine outside the inpatient setting.
Holberg & Finlayson, 2007	n=8 (MS type not reported) Mean age: 46.2 years, age range: 40-54 years	Illinois, USA	Qualitative; semi structured interviews and thematic analysis Pilot Study	To examine factors influencing the implementation and continued use of fatigue management strategies among persons with MS after a FMI.	Participants identified that fatigue impacted adversely their valued activities. The physical, cognitive, and emotional impact of fatigue was a significant barrier in everyday life. Participants perceived environmental factors and the value of education as relevant in management of their fatigue. Participants reported the use of at least seven fatigue management strategies five months after the FMI had ended.
Jellie et al., 2014	n=19 (RRMS=14; PPMS=3; SPMS=2) Mean age: 40 years; age range: 24-63 years	London, UK	Qualitative; semi-structured interviews and comparative analysis	To examine the impact of a patient-designed vocational rehabilitation intervention on the experiences of people with MS who were experiencing work instability; to identify aspects of the intervention that were most helpful from the perspective of PwMS.	Participants expressed that fatigue was particularly problematic in relation to work and it severely impacted their memory. Education about fatigue led to adapting tasks within the work and home environment. Emotional support from employers and co-workers regarding fatigue management for demanding tasks was important for participants.
Kayes et al., 2011	n=10 (RRMS=4; SPMS=3; PPMS=3) Mean age: 44.3 years, age range: 34-53 years	New Zealand (specific location not reported)	Qualitative; grounded theory approach; semi-structured interviews and constant comparative analysis	To explore the barriers and facilitators to engagement in physical activity from the perspective of PwMS.	Participants identified fatigue as a barrier to physical activity. Fatigue resulted in a need to balance exercise with other activities of daily living and to preserve energy. A significant barrier to engaging in physical activity for participants was their conflicting beliefs about exercise and fatigue.

<p>Koffman et al., 2013</p>	<p>n=87 (RRMS=24; PPMS=30; SPMS=33) (Qualitative component n=30; MS type not reported for participants interviewed in qualitative component) Mean age: 52.6 years</p>	<p>South London, UK</p>	<p>Mixed methods Qualitative component: structured interviews and framework analysis</p>	<p>To examine the experience of symptoms, disease progression and psychosocial challenges among Black Caribbean and White British PwMS who are severely affected by MS.</p>	<p>Many participants described rapid deconditioning as the aspect of MS which troubled them the most. This perspective was more common among Black Caribbean participants compared to White British PwMS. Fatigue was identified as one of the main symptoms related to severe MS and included both physical and mental fatigue.</p>
<p>Lohne et al., 2010</p>	<p>n=14 (MS type not reported) Age range: 39-66 years</p>	<p>Norway (specific location not reported)</p>	<p>Qualitative; phenomenological-hermeneutic approach; semi-structured interviews and hermeneutic analysis</p>	<p>To identify how PwMS experience and understand dignity and violation in the context of a rehabilitation ward.</p>	<p>Participants described fatigue as sudden, unpredictable, and draining which challenged their ability to maintain employment and made them feel fearful about the future. Participants suggested that the invisible nature of fatigue made it difficult for others (including healthcare professionals) to understand the disabling nature of MS fatigue. Participants expressed a desire to be treated with respect and dignity.</p>
<p>Mathiowetz & Busch, 2006</p>	<p>n=134 (RRMS=90; PPMS=7; SPMS=21; RPMS=1; Not known=15) Mean age: 49.1 years, age range: 29-70 years</p>	<p>Minneapolis & Chicago, USA</p>	<p>Mixed methods Qualitative component: open-ended interview questions and descriptive analysis</p>	<p>To examine how individuals who had participated in the FMI evaluated aspects of it in terms of content, format, and instruction.</p>	<p>Resting, balancing a schedule, and strategies to communicate fatigue including group discussion were identified as useful strategies for fatigue management. The least helpful aspect of the course was body mechanics. Participants felt the quality of homework assignments and the presentation of material could have accommodated their needs better.</p>

Moss-Morris et al., 2012	n=23 (RRMS=10; PPMS=2; SPMS=7; Not known=4) Mean age: 40.1 years	UK (specific location not reported)	Mixed methods Qualitative component: semi-structured interviews and thematic analysis Pilot Study	To assess the feasibility, potential efficacy, and cost-effectiveness of the programme MS Invigor8 in reducing fatigue severity and impact.	Online method of intervention was positively regarded by participants, and telephone communication with the intervention facilitators augmented the sessions. Participants indicated that FMI participation increased their sense of control over fatigue and their understanding of fatigue. Participants also reported significant improvements in anxiety, depression, and quality of life after the FMI.
Mulligan et al., 2016	n=23 (RRMS=14; PPMS=2; SPMS=3; Not known=4) Mean age: 48.9, age range: 37-63 years	Dunedin, New Zealand	Qualitative; semi-structured interviews and thematic analysis	To explore the perceived impact of the 'Minimise Fatigue, Maximise Life: Creating Balance with Multiple Sclerosis' (MFML) fatigue self-management programme from participants' perspective.	Participants reported the intervention enabled them to accept their limits, set boundaries, prioritise, build resilience, and have a positive outlook. Group support was valued by participants and helped them improve their communication about fatigue and adapt to lifestyle change.
Newland et al., 2012	n=16 (RRMS=16) Mean age: 42 years, age range: 25-58 years	Mid-western USA	Qualitative; focus groups and content and comparative analysis	To characterise symptoms experienced by persons with RRMS in patients' own words.	Fatigue was experienced by majority of participants which they felt interfered with many of their activities of daily living and engagement in life roles including parenting. Fatigue and cognitive impairment were described by participants as co-occurring symptoms, and where one impacted the other. Participants also experienced heat intolerance whereby high temperatures exacerbated their fatigue.
Pétrin et al., 2018	n=23 (RRMS=16; PPMS=2; SPMS=4; Not known=1) Mean age: 48.9 years	Ontario, Canada	Qualitative; semi-structured interviews and interpretive description Pilot Study	To explore participant's views on the self-guided fatigue management program (MS INFoRm)	Participants experienced a shift in their knowledge and their everyday behaviour in relation to self-management of fatigue. This led to enhanced quality of life, confidence and empowerment for participants who completed the programme.

Ploughman et al., 2012(a,b)	n=18 (RRMS=3; PPMS=2; SPMS=10; Benign=1; Not known=2) Mean age: 66.5 years, age range: 56-80 years	Glasgow, UK	Qualitative; semi-structured interviews and thematic analysis	To explore older people's views and experiences of ageing with MS; to describe self-management of fatigue from their perspective.	Participants recounted how they learned to accept fatigue through adapting their daily activities in the context of fatigue. Participants reported that fatigue was often a sign of exacerbation of MS. For self-management of fatigue, participants utilised fatigue management strategies such as resting, managing schedules, modifying exercise routine, and using adaptive equipment.
Silverman et al., 2017	n=32 (PwMS=12; care partners=11; community stakeholders= 9) (MS type not reported) MS participant mean age: 52.3 years, age range: 36-62 years	USA (specific location not reported)	Qualitative; phenomenological approach; focus groups and inductive analysis	To understand the meaning of resilience, the factors which facilitate resilience and the barriers inhibiting resilience from the perspective of PwMS, their care partners and community stakeholders.	Participants understood resilience as an ability to focus on the positive and to thrive rather than merely survive. Participants identified a range of factors which facilitated resilience, including psychological adaptation to MS, social connections, finding meaning in life, and ability to plan ahead. Barriers to resilience included fatigue, resource depletion, negative thoughts, social limitations, and social stigma. Participants described fatigue as negatively impacting their participation in meaningful activities.
Smith et al., 2009	n=10 (RRMS=10) Mean age: 46.4 years, age range: 32-61 years	New Zealand (specific location not reported)	Qualitative; semi-structured interviews and interpretative description	To explore in depth the influence of a 8-week exercise programme on perceptions of fatigue amongst PwMS.	Fatigue was described as one of the worst symptoms of MS, impacting participants' ability to perform activities of daily living and to plan for their future. Participation in the exercise programme was perceived by participants to improve their physical and mental energy and enabled them to feel a sense of achievement. However, exercising beyond limits led to perceived deterioration and reduced balance and mobility.
Smith et al., 2011	n=9	New Zealand (specific location not reported)	Qualitative; semi-structured interviews	To describe the experiences of people with MS-related fatigue who engaged in community-based exercise activities;	Perceived control over fatigue influenced exercise choices for participants. The fluctuating and unpredictable nature of fatigue could lead to perceived loss of control for participants. Participants described continual adjustment to MS fatigue through pacing and planning. Different dimensions to fatigue were described including muscle weakness, mental

	(RRMS=5; SPMS=3; Unknown=1) Age range: 28-70 years		and inductive thematic analysis	to discover how fatigue influenced their exercise participation.	tiredness, feelings of depression and an all-consuming fatigue. Many participants felt exercise in a safe and supportive environment improved their well-being and increased their sense of control over their fatigue.
Thomas et al., 2010	Intervention: n=16 (RRMS=11; PPMS=2; SPMS=3) Qualitative component: n=8 (RRMS=5; SPMS=3) Mean age: 45.5 years, age range: 34-59 years	Dorset, UK	Mixed methods Qualitative component: focus groups and thematic analysis Pilot Study	To design a group-based MS fatigue management group programme incorporating cognitive behavioural and energy effectiveness approaches; to evaluate the effectiveness and participants' experience of the programme.	Participants described fatigue as overwhelming, limiting and poorly understood by others. Peer support and learning from others' experiences were perceived as important aspects of the FMI. At follow-up, participants reported the cognitive-behavioural aspects of the program were helpful and that the program facilitated them to make beneficial changes to their lifestyle.
Turpin et al., 2018	n=13 (MS type not reported) Mean age: 46.2 years, age range: 25-67 years	Queensland, Australia	Qualitative; semi- structured interviews and thematic analysis	To explore the experience, understanding and management of fatigue in everyday life for PwMS in order to increase society's understanding of MS-related fatigue.	Participants felt better able to understand their fatigue by gaining information about fatigue, by being more aware of their fatigue, and by seeking healthcare professional support for their fatigue. A better understanding of their fatigue enabled them to understand the impact of fatigue on their lives and make decisions about how to accommodate fatigue in their lives. Participants reported challenges informing others about the impact of MS-related fatigue on their everyday life.
Twomey & Robinson, 2010	n=8 (RRMS=3; SPMS=2; Benign=1; Unknown=2) Mean age: 42.9 years,	Limerick, Ireland	Qualitative; semi- structured interviews and constant comparative analysis Pilot Study	To explore the experience of participating in a community- based fatigue management program for PwMS.	Participation in the program facilitated lifestyle and occupational change for participants. Participants felt that positive outcomes of the programme for them such as empowerment, active participation, legitimisation of fatigue and increase in social support, were due to the group-based format of the programme.

	age range: 29-55 years				
Wendebourg et al., 2016	n=16 (MS type not reported) Mean age: 47.5 years, age range: 24-61 years	Hamburg, Germany	Mixed methods Qualitative component – semi-structured interviews & focus groups and framework analysis Pilot Study	To integrate the concepts of CBT and evidence-based patient information in order to develop a group-based fatigue management intervention for PwMS.	Participants could experience fatigue differently from each other and in some cases, had unique requirements for managing fatigue. Participants reported that other people's lack of understanding about fatigue had a significant and negative impact on them. Participants reported a need for education-based FMIs and preferred sessions to be group-based and in person.
Yorkston et al., 2009	n=14 (MS type not reported) Mean age: 43.9 years, age range: 32-58 years	Seattle, USA	Qualitative; phenomenological approach; semi-structured interviews and interpretative phenomenological analysis	To examine the experiences of participation in everyday work activities, both inside and outside the home, among people with mild to moderate MS.	Participants described how MS changed their perspective of working, causing them to prioritise the work (both at work and at home) which was most important to them. Taking ownership and being in control of work was important to participants. Participants developed a variety of strategies to enable them to manage fatigue and cognitive difficulties in the workplace including obtaining support, prioritising, and planning their activities.

RRMS=relapse-remitting MS; PPMS=primary progressive MS; SPMS=secondary progressive MS; RPMS=relapsing progressive MS

