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An exploratory randomised trial of a nurse-led telephone support intervention for people who support people with multiple sclerosis (The MNTISS trial).

A thesis presented to the University of Dublin, Trinity College Dublin, for the Degree of Doctor of Philosophy.

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Acknowledgement

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Thesis summary

Background

In Ireland, the provision of care by family caregivers contributes substantially to the reduction in annual national expenditure from healthcare budgets. However, the negative impact of providing informal care has been well-documented, with personal and financial cost implications for caregivers. This study focused on the condition of multiple sclerosis and the provision of support to those who provide care to people with multiple sclerosis (PwMS). Although the variety of caregiver support needs require interdisciplinary support, a number of the unmet needs of caregivers of PwMS, such as the need for accurate information and advice, and dealing with the emotional impact of caregiving, can be addressed within the scope of the role of the nurse specialist in multiple sclerosis (NSMS). These nurses have expert knowledge about the PwMS's condition and circumstances, which means they can tailor their support to the individual and the people who support them. Despite the known unmet needs of caregivers of PwMS, there is a dearth of interventions developed and tested to bridge this gap. Therefore, the purpose of this study was to develop and test an intervention to support people who support PwMS (SPs).

Research aims

The aims of this PhD research study were to: 1. Develop an intervention for use by nurse specialists in Multiple Sclerosis (NSsMS) to help support people who support PwMS (SPs), 2. Test the feasibility and acceptability of the newly developed intervention to NSsMS, SPs and PwMS within a randomised trial, 3. Determine SPs' satisfaction with the intervention,4. Evaluate the potential impact of the intervention in terms of measurable SP self-efficacy in problem management and obtaining respite, preparedness to care, quality of life and burden.

The null hypothesis was: There is no difference in self-reported measurable self-efficacy, preparedness for supporting/caring, burden and QoL for SPs who participate in a nurse-led manual based telephone support intervention service when compared to SPs who receive usual care.

Methods

A model that was influenced by the Medical Research Council Framework documents (Medical Research Council 2000, 2008) was developed to guide the development of the tested intervention. Egan's model of the skilled helper, the core components of the

COPE model (creativity, optimism, planning and availing of expert information) and an experiential approach to learning (Quinn 2000) were amalgamated to provide the overarching framework for the intervention. The intervention was tested using an exploratory randomised trial, which comprised a pilot feasibility study carried out in 3 hospital sites. Seventy-one SPs from three research sites were randomised using block randomisation. Thirty-three SPs were assigned to the intervention group and 38 to the control group. Data were collected using a number of questionnaires at baseline, time one (week 6) and time two (week 13). Interviews were carried out with NSsMS, SPs and PwMS to inform the feasibility component of the study.

Results

The acceptability and feasibility of delivering the intervention and carrying out the study were supported. Egan's model of the skilled helper was found to be a suitable model to guide the delivery of the intervention because, while providing a framework for the NSMS to help SPs manage problems, it allowed for flexibility and tailoring of the telephone interaction between the SPs and NSsMS. A number of challenges were identified such as the availability of resources, both human and environmental. While all SPs may benefit from the intervention, in practice offering it to all SPs is unlikely to be feasible. Those most likely to be in greatest need of the intervention were SPs who had an immediate issue or concern to be addressed. According to the SPs, the greatest benefits included increased support, confidence and knowledge. A significant between groups difference (p=0.025) with moderate effect (0.35) was found in support of the intervention group for improved preparedness to care following final data collection. No significant between groups differences were reported for self-efficacy in obtaining respite and problem management, SP burden or quality of life.

Conclusion

The intervention developed and tested in this study was feasible to deliver and test using a randomised trial design. The findings suggest that, with additional tailoring in specific areas, the intervention has the potential to help ameliorate some of the unmet needs of SPs. The findings highlight the potential and unique contribution that NSsMS can make to people who support PwMS. This is the first proactive nurse-led support intervention for SPs of PwMS internationally.

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Glossary of Abbreviations

ANCOVA: Analysis of co-variance

CD: Compact disc

CIS: Clinically isolated syndrome

CRA: Caregiver reaction assessment

EDSS: Expanded disability status scale

GP: General practitioner

MNTISS: Manualised Nurse-Led Telephone Support Intervention

MRI: Magnetic resonance imaging

MS: Multiple sclerosis

MSIS: Multiple sclerosis impact scale

NSsMS: Nurse specialist in multiple sclerosis

NSMS: Nurse specialist in multiple sclerosis

MSQoL: Multiple sclerosis quality of life

OPD: Outpatient department

PPMS: Primary progressive Multiple sclerosis

PwMS: People with Multiple sclerosis

QoL: Quality of Life

RRMS: Relapsing remitting Multiple sclerosis

SP: Support person

SPMS: Secondary progressive Multiple sclerosis

SWL: Satisfaction with life

WHOQoL: World health organisation quality of life

Glossary of Terms

Clinical nurse specialist: A registered nurse who has been approved in a specialist nursing role.

Dyad: A group of two people, which in this study refers to the person with multiple sclerosis and their support person.

Exploratory trial: A trial that enables the researcher to answer specific questions with limited exposure to the intervention and the number of participants.

Interventionists: The nurse specialists in multiple sclerosis who delivered the intervention.

Intervention: A process to support and help people who support people with multiple sclerosis to learn problem management skills.

Intervention manual: The written document containing guidance for the interventionists, the guidebook for the support persons and the training booklet that provided guidance to the nurse specialists in multiple sclerosis on the delivery of the intervention.

Multiple sclerosis: A chronic progressive neurological condition that affects the nerves in the brain and spinal cord leading to loss of vision, balance, sensation and muscle control. Its course is unpredictable.

Nurse-led telephone support: A telephone support system that is initiated and led by a nurse specialist in multiple sclerosis.

Nurse specialist in multiple sclerosis: Nurses who have specialised in the area of multiple sclerosis.

People with multiple sclerosis: People who have been diagnosed with multiple sclerosis.

Pre-exploratory phase: A trial run of the exploratory design to model the delivery of the intervention in the practice setting, evaluate recruitment methods and identify other factors that may impact on the exploratory phase of the study.

Process analysis: a nested study exploring the way in which the intervention was delivered and tested.

Randomised trial: The highest form of an experimental design to elicit cause and effect outcomes.

Support people: Individuals nominated by PwMS whom they identify as providing the most emotional or physical support for them and can be a partner or spouse, other family member or carer.

Chapter 1: Introduction

This thesis presents the process and findings of a study designed to develop and test an intervention to support family members and caregivers of people with multiple sclerosis (PwMS). Multiple Sclerosis (MS) is a chronic neurodegenerative disorder of the central nervous system resulting in physical, mental and psychological symptoms. In many cases the impact of the condition results in significant life changes for both the person with the diagnosis and their family members/caregivers. Some PwMS require the assistance of informal caregivers. Literature reviews that were published prior to the commencement of this study (McKeown *et al.* 2004, Corry & While 2009) highlighted that family members and informal caregivers of PwMS have many unmet needs, a number of which can be addressed within the scope of the role of the nurse specialist in multiple sclerosis (NSMS). Nurse specialists in MS are qualified first level nurses who have specialised in the area of MS nursing. Their role, like that of all nurse specialists, encompasses a holistic approach to care that takes into consideration the needs of patients, family members, caregivers and the community. This study focuses on the aspect of their role that involves supporting family members and caregivers. In order to contextualise the study, a brief overview of the role that caregivers play in supporting people with chronic illness is initially presented in this chapter. This is followed by an overview of MS and how it impacts on people with the diagnosis and those who support them. The reasons for pursuing this topic for doctoral research, along with the approach adopted to develop and test the intervention is then presented. Finally, the outline and structure of the thesis and a brief overview of the content of each chapter is provided.

1.1 Caregivers

Many people with chronic conditions are dependent upon family members and caregivers to provide support and care. The care provided varies according to the nature of the needs of the individual with the illness and the relationship with the care recipient. In a number of instances, people unexpectedly find themselves in a caring role for which they are ill prepared. Providing care may negatively impact on the

caregiver from an emotional, physical, social and financial perspective (Care Alliance Ireland 2010). For some, the adjustment to the role is difficult and requires significant emotional and life changes. Having access to support people with the required knowledge and skill may ease the transition and potential feelings of isolation. The programme for Government 'Towards 2016 – 10 year framework' (Government of Ireland 2006) recognise the importance of caregivers and the need to acknowledge and support them in their role. In 2006, it was estimated that there was a total of 161,000 family caregivers in Ireland (Care Alliance Ireland 2010). This figure increased to nearly 183,000 in 2011, representing a 13.7% increase over a five year period (Care Alliance Ireland 2013). In the United Kingdom (UK) and Republic of Ireland (ROI), family caregivers provide millions of hours of care each week contributing an annual estimated national reduction in health care expenditure of £119bn in the UK (Brindle 2011) and €5bn in the ROI of Ireland (Care Alliance Ireland 2012).

A carer is an individual (family member, friend or significant other) who provides "... unpaid personal help..." (Care Alliance Ireland 2010, p.2). The care provided by family members has been categorised into four different levels according to the intensity of the care provided (Care Alliance Ireland 2010). Care that includes companionship with some caring assistance is categorised as low level caring, care that involves instrumental care (such as cooking and shopping) is categorised as medium level caring with chores. If the care involves providing some personal assistance, such as dressing and washing, it is categorised as medium level caring with some personal assistance. In situations where the person requiring the care cannot provide much assistance to the caregiver in meeting their own needs, the care is categorised as high level caring (Care Alliance Ireland 2010).

Over 7,000 people are living with a diagnosis of MS in Ireland, a condition with a disease progression that is unpredictable in presentation and course. It is estimated that up to 40% of the 7,000 sufferers are in receipt of unpaid care and assistance with 67% of carers of people with MS providing care on their own, and 51% spending more than 45 hours per week providing care (Multiple Sclerosis Society of Ireland 2006). State/agency care was found to provide a very small proportion of care, with many carers reporting that they had difficulty accessing services; 44% having some difficulty and 28% finding it quite difficult (Multiple Sclerosis Society of Ireland 2006).

1.2 Multiple Sclerosis: the condition

Multiple Sclerosis is a chronic progressive neurological condition that impacts on people throughout their lifespan. It is the most common cause of non-traumatic neurological disability in young adults (Edmonds *et al.* 2007, Compston & Coles 2008, Mahad *et al.* 2009). Although Giovannoni *et al.* (2006) purports that many theories exist as to its cause, the exact cause of MS remains unclear. Epidemiologists continue to propose an infectious cause, but there is currently no scientific evidence to support this position (Subramanima & Steiner in Giovannoni *et al.* 2006). Consequently, MS is generally accepted to be an inflammatory autoimmune disease of the central nervous system (Subramaniam & Steiner in Giovannoni *et al.* 2006, Cuddy 2007, Compston & Coles 2008, Ross *et al.* 2008), which results in patches of inflammatory demyelination throughout the central nervous system (Miller & Leary 2007). Progression of the disease has been correlated with degeneration of the demyelinated axons (Mahad *et al.* 2009). Recent research by Mahad *et al.* (2009) suggests that the axonal degeneration and resultant dysfunction that occurs during the progressive stages of MS is due to oxidative changes to mitochondrial deoxyribonucleic acid.

It is argued that MS is a complex disease brought about by an interaction between genetic and environmental factors (Giovannoni *et al.* 2006, Compston & Coles 2008), which triggers a series of events within the immune system, generating an acute inflammatory response leading to post inflammatory gliosis and neurodegeneration (Compston & Coles 2008). The precise environmental factor that triggers onset and progression of MS is unknown (Coyle 2008) but recent research suggests that environmental factors due to urbanisation may be implicated (Kotzamani *et al.* 2012). It is generally accepted that MS affects caucasians more than any other race (Irish Network of Multiple Sclerosis Nurses 2012). Migration studies support the environmental genetic interplay, in that people born in high risk areas have reduced risk if they migrate to a low risk area (Ascherio & Munger 2007, Irish Network of Multiple Sclerosis Nurses 2012). Exposure to environmental risk is believed to occur in early adolescence (Giovannoni *et al.* 2006) and before the age of 15 years (Irish Network of Multiple Sclerosis Nurses 2012), which suggests that the risk of getting MS occurs in early life. This early childhood risk has been linked to low levels of vitamin D in early life, in particular before the age of 20 (Munger *et al.* 2006). Recent writings, however, argue that the latitude gradient feature (higher latitude greater prevalence) of

MS is disappearing (Ascherio & Munger 2007, Alonso & Hernan 2008) and a greater appreciation of the genetic susceptibility theory is emerging (Rosati 2001). Despite this, environmental factors are thought to work at the population level and not the family level (Giovannoni *et al.* 2006) and whether they act as a trigger or are involved in ongoing disease pathogenesis has yet to be established (Giovannoni *et al.* 2006).

MS affects approximately 2.5 million people worldwide (Compston & Coles 2008, Ross *et al.* 2008, Finlayson 2009) and has an incidence rate of approximately 7 per 100,000 per annum and prevalence of 120/100,000 (Compston & Coles 2008). The UK prevalence is 0.1-0.2% of population (Edmonds *et al.* 2007). Lifetime risk is one in 400 (Compston & Coles 2008) or one in 1500 of population in Europe, North America and Australasia (Miller & Leary 2007). In high risk areas, Ascherio & Munger (2007) argue that lifetime risk is about 1 in 200 for women and less for men. Women are roughly twice as likely to have a diagnosis of MS as men (Orton *et al.* 2006, Ascherio & Munger 2007, Compston & Coles 2008), with a trend for even greater incidence in women in recent years (Ascherio & Munger 2007).

Researchers vary in their estimation of the typical age of onset but in general agree, that it is a disease of middle-age, with diagnosis usually between 20 and 35 (Ascherio & Munger 2007, Finlayson 2009) to 45 years of age (Cuddy 2007, Compston & Coles 2008). The average age of onset is 30 years (Sadovnick *et al.* 2009)). People can however be diagnosed from childhood (Ascherio & Munger 2007, Banwell *et al.* 2007), with 2% presenting before the age of 10 and 5% before the age of 16 (Compston & Coles 2008). MS also presents over the age of 55 years, but this is rare (Ascherio & Munger 2007), and comprises fewer than 10% of cases (Finlayson 2009, Sadovnick *et al.* 2009).

The way in which MS presents has significance for the course and prognosis of the disease (Miller & Leary 2007). The disease itself presents as either a relapsing remitting MS type (RRMS) which accounts for 40% of cases, secondary progressive MS (SSMS) accounts for another 40%, primary progressive MS (PPMS) making up 10-15% of cases, and benign MS making up 10% of cases (Cuddy 2007). According to Compston & Coles (2008), 80% of patients present with a relapsing remitting form of the illness and pass through various stages from relapses with full recovery to persistent deficits and finally the secondary progressive type of MS. Relapsing remitting MS generally presents as a clinically isolated syndrome (CIS) (Coyle 2008) which, in 85% of young adults, is usually of the optic nerve, brainstem or spinal cord

(Miller & Leary 2007). Despite this, only 30-70% patients who present with a CIS develop MS, with the risk of this occurring being high for those presenting with clinically silent brain lesions (Miller & Leary 2007). Rovaris *et al.* (2006) report that up to 40% of Canadian patients with RRMS develop secondary progressive MS within 10 years of disease onset, with chronic progression for up to 80% of people with RRMS within two decades (Kremenchutzky *et al.* 2006, Rovaris *et al.* 2006).

In RRMS type MS, individuals experience random episodes of discrete clinical attacks (Rosati 2001), usually at least one year apart at the start. The intervals steadily decrease, and deterioration is slowly progressive (Compston & Coles 2008), leading to a steady build up of fixed disability and culminating in a secondary progressive type of MS (Kremenchutzky *et al.* 2006, Rovaris *et al.* 2006). A relapse is defined as a new or worsening of existing symptoms due to MS which lasts for greater than 24 hours and is separated from previous events due to MS by at least 30 days (McDonald *et al.* 2001). In SPMS, progression becomes relentless and relapses less prominent (Tremlett *et al.* 2010). Although there is no consensus on the mechanism for the transition from RRMS to SPMS, it is hypothesised that it occurs due to the increase in neurodegenerative changes compared to the inflammatory pathological changes. Support for this is found in the limited efficacy of immunomodulating and immunosuppressive treatments as the disease progress (Rovaris *et al.* 2006).

Individuals with PPMS experience severe disability within a short time period (Compston & Coles 2008). This type of MS is usually seen with older onset of illness and in a higher proportion of men (Miller & Leary 2007). Pathologically, the inflammation of white matter which is seen in RRMS is less evident and axonal loss and microglial activation in healthy looking white matter together with cortical demyelination and atrophy on magnetic resonance imaging (MRI) is more likely to present (Miller & Leary 2007).

The existence of a benign form of MS is controversial (Irish Network of Multiple Sclerosis Nurses 2012). In this type of MS, there is complete recovery following attacks which are 10 years or more apart and there is little or no accumulation of disability (Irish Network of Multiple Sclerosis Nurses 2012), with the patient remaining neurologically functional 15 years after disease onset (Lublin & Reingold 1996). This type of MS may go undiagnosed for many years or it may never be diagnosed during life and might only be detected in a post-mortem examination (Irish Network of Multiple Sclerosis Nurses 2012). In general, most MS sufferers develop neurodisability due to a

progressive deterioration in neurological functioning (Miller & Leary 2007, Mahad *et al.* 2009). The increasing disability as the disease progresses is thought to be due to axonal dysfunction resulting from changes within the axonal membrane (Mahad et al. 2009). The presence of an abnormal brain scan and the number of lesions present on the scan effects the course of the condition, with the risk of having a second episode higher in those with abnormal scans (Miller & Leary 2007). Disability is less likely in those with few or no MRI lesions, a long period to the first relapse and no disability after the first five years (Miller & Leary 2007). Irrespective of disease duration or the frequency of early relapse, Scalfari *et al.* (2011) purport that long-term prognosis is primarily determined by the development of SPMS. Progression rates are similar regardless of type of MS at onset, once a patient reaches a disability score of 4 on the expanded disability status scale (EDSS) (Hurwitz 2010). Although age itself does not affect the progression of the course, it independently affects disability by increasing the probability of SPMS onset (Scalfari *et al.* 2011).

1.3 Impact of Multiple Sclerosis on PwMS and carers

Multiple Sclerosis affects people in different ways and this is largely dependent on the functional areas of the brain affected, which can include the cerebrum, brain stem and cerebellar pathway, and the spinal cord (Compston & Coles 2008). Involvement of the brain stem and cerebellar pathways present clinically as problems with coordinated eye movement and limbs, along with bulbar and axial muscular problems. Motor, sensory and autonomic problems present with spinal cord involvement (Compston & Coles 2008). The patchy arrangement of the lesions results in a presentation and course of illness that is unique to the individual and difficult for health care professionals to predict. As the disease progresses and disability increases, symptoms worsen (Higginson *et al.* 2006). More than 50% of patients with increasing disability experience problems using legs/arms, fatigue/lack of energy, spasms, pain and feeling sleepy (Higginson *et al.* 2006). As the disease continues to advance, PwMS encounter what Edmonds *et al.* (2007) describe as "new changes and losses" (p. 106) and commonly experience increasing levels of pain, spasticity, fatigue, depression, bedsores and incontinence; with cognitive impairment reported in up to 50% of PwMS (Edmonds *et al.* 2007) Cognitive impairment can occur at any stage of the disease trajectory and in the absence of physical symptoms (Johnson 2007). It can be mild or severe with

severity increasing as the condition deteriorates and in those with progressive types of MS (Johnson 2007). Memory problems are the most frequently encountered type of cognitive impairment in PwMS, occurring in 40-60% of PwMS (Rogers & Panegyres 2007).

It is important to note that there is no cure for MS but treatment is available to slow progression and onset of disability by slowing the development of new lesions and reducing the number of flare ups (Cuddy 2007, Compston & Coles 2008). As yet, there is no treatment to stop the disease progression (Kleinschnitz *et al.* 2007). The approach to treatment is complex and unique to the individual type and course of the illness, thereby requiring significant patient and family education which usually falls within the scope and role of the nurse specialist in MS. Patients sometimes revert to complementary therapies and alternative medicine for treatment (Cuddy 2007). In some instances, these therapies are uncontrolled and can be offered by practitioners with limited knowledge about the treatments and their side-effects, particularly in the treatment of PwMS. It is important that family members and caregivers have access to professionals who can provide accurate advice and guidance on the use of such therapies. For example, it is important that they are made aware that the use of herbs may have side-effects and that some herbs may interact with other drugs (Cuddy 2007). Likewise, there may be safety concerns for PwMS on some medications who take vitamins (Cuddy 2007). However, some complementary and alternative therapies have been reported to provide benefit, including cooling suits, yoga, meditation, hippotherapy (horseback riding) and massage (Cuddy 2007).

Patients, family members and carers require considerable MS nurse support to manage and cope with the initial diagnosis and subsequent progression and treatment of MS. International research confirms that caregivers, many of whom are family members, have many needs which are not being met by the currently available support systems (Corry & While 2009). The importance of supporting family members/caregivers of PwMS in Ireland, has been highlighted by high profile MS cases such as the Marie Fleming right to die case (Irish Independent 2013) and the Evelyn Joel neglect case. Evelyn was a Wexford woman who died of malnourishment. Her daughter and her daughter's partner were found guilty of manslaughter by neglect (Irish Examiner 2013). Reports from experts during the trial highlighted the deficiencies in the health system, including the multi-disciplinary team's failure to put a community action plan in place (Irish Times [Dublin] 2011). In the Republic of Ireland, NSsMS are

a point of contact for family members and caregivers. They offer a telephone support service to PwMS and their families/caregivers but this service is only provided to those who make contact with the nurse specialist. Family members or carers may not be aware that this service is available to them or they may be reluctant to make initial contact. Issues such as fear of being judged, not wanting to waste the nurse's time or simply fear of making the first contact may prohibit them from making contact. Therefore, the research conducted for this doctoral study set out to develop and test a proactive nurse-led telephone support system that falls within the scope and role of the nurse specialist.

1.4 Context of the study

I am a registered general nurse and I specialised in neuromedical/neurosurgical nursing in the late 1980s at the National Hospital for Nervous Diseases in Queen's Square, London. On completion of the course, I remained in clinical neurology posts before progressing on an educational route in the mid 1990s. Despite adopting an educational career pathway, I maintained a keen interest in neurological nursing and continued to teach the topic while retaining clinical links through my contacts in practice. I was intrigued by the impact of neurological illness on individuals, in particular PwMS and the unpredictable course of the condition. As most of my clinical experience was in the UK and USA, my Irish clinical networks were limited to one main neurological centre where I worked as a registered nurse for a number of years on my return from abroad. These networks were, however, broadened during my time as a lecturer in Trinity College Dublin (TCD), when I co-ordinated a postgraduate nursing course. At that time, a certificate in MS nursing was offered alongside the postgraduate course. Nurses who completed the certificate in MS had the option to complete additional modules to obtain a postgraduate qualification. During this time, I developed close links with the MS nurses in Ireland and a keen interest in the condition itself. This experience, together with the fact that many nurses that I had taught at undergraduate nursing level were now specialising in neurological nursing broadened my clinical contacts in Ireland.

My keen interest in the brain was coupled with my interest in psychology and in particular human motivation and behaviour. This interest led me to study psychology at

BSc. Level with the Open University. Much of this course was research based and the material was primarily presented through the examination of research studies, in particular experimental research studies; which increased my interest in experimental research design. Soon after completing this psychology course, I met with Professor Alison While who had taken up a visiting professor's post at the School of Nursing & Midwifery in TCD. We both had a background and interest in MS, my work was primarily clinical and Alison had published on aspects of the role of the MS nurse (Forbes *et al.* 2003, While *et al.* 2009). We discussed possible approaches to working together and we decided to carry out and publish a literature review on the needs and experiences of caregivers of PwMS. Following this publication, I developed a proposal for a research study to develop and test an intervention that could address the needs of this group of caregivers. As the work progressed, I began to explore potential funding opportunities and in September 2009 decided to register for a research degree at doctoral level while in pursuit of research funding. In March 2011, I was awarded a 'Research Training Fellowship for Healthcare Professionals award' from the Health Research Board in Ireland. This provided the salary and running costs to support the study, course fees and training. It also afforded me the opportunity to focus primarily on refinement and preliminary testing of the intervention that I had developed.

In this thesis, I provide an overview of the development and preliminary testing of this intervention. The primary goal of the intervention was to enable NSsMS help family members and carers of PwMS learn problem management skills in order to be better prepared for their role in supporting a person with MS. The development of the intervention was guided by Egan's Model of the Skilled Helper, elements of the COPE model and the experiential model of learning. These models were used to structure the delivery of the intervention. This doctoral research study presents the development of the intervention and examined the feasibility of delivering it. This included determining the support persons most likely to benefit from receiving it, the outcome measures that are most sensitive to the intervention and the potential benefits to participants.

The objectives of the study were to:

1) Develop an intervention for use by nurse specialists in Multiple Sclerosis (NSsMS) to help people who support PwMS (SPs).
2) Test the feasibility and acceptability of the newly developed intervention to NSsMS, SPs and PwMS within a randomised trial.
3) Determine SPs' satisfaction with the intervention.
4) Evaluate the potential impact of the intervention in terms of measurable SP self-efficacy in problem management and obtaining respite, preparedness to care, quality of life and burden.

1.5 Structure of the thesis

The thesis is divided into eight chapters. In chapter two, the literature on the needs and experiences of caregivers of PwMS is examined. This literature review extends two previously published reviews (McKeown *et al.* 2003, Corry & While 2009) and also includes a re-examination of papers from these reviews in order to provide a comprehensive profile of family members and caregivers of PwMS. Chapters three and four detail the process adopted to develop the intervention, which included a series of literature reviews. To manage word count restrictions, where my research has been published, summaries of the papers are provided with reference to the published papers. Chapters five and six provide an account of the methodology and research methods used to test the intervention. The results are presented in chapter seven. Finally, in chapter 8, the findings are discussed with reference to the international literature, where it exists. In addition, the implications of the study for practice are discussed and recommendations are made for further testing of the intervention in a full-scale randomised trial.

Chapter 2: Literature review

2.1 Introduction

The purpose of this study was to develop and test an intervention for delivery by NSsMS to meet the needs of people who support PwMS. It was, therefore, important that the intervention could be incorporated into the scope and role of the nurse specialists in Multiple Sclerosis (MSsNS). As the most recently published review on the needs and experiences of this group of caregivers included research up to November 2007 (Corry & While 2009), an up-dated literature review of their needs and experiences, and the support services available to them, was required to inform this study. Two previously published reviews have consistently highlighted that caregivers of PwMS have many unmet needs and that caring for a PwMS impacts on the physical, psychological, social and financial wellbeing of caregivers, which results in increased caregiver burden and reduced quality of life (QoL) (McKeown *et al.* 2003, Corry & While 2009). The review published by Corry & While (2009) extended the work of McKeown *et al.* (2003) which had spanned literature from 1990 to April 2002, and provided a review of the literature on the needs and experiences of carers of PwMS from May 2002 to November 2007. These two original pieces of work provided the building block for this current literature review. This chapter, therefore, provides a detailed overview of the needs of caregivers of people with multiple sclerosis (PwMS) and the support services they use.

2.2 Literature search

In order to update the review for this doctoral research, a comprehensive search of the literature on the needs and experiences of carers of PwMS published between November 2007 and March 2010 was carried out in March 2010 using the following search strings: 'multiple sclerosis and carer(s)/caregiver(s)', 'multiple sclerosis and carer(s)/caregiver(s) and experience', 'multiple sclerosis and support systems', 'multiple sclerosis and carer(s)/caregiver(s) and needs', 'multiple sclerosis and burden', 'multiple sclerosis and carer(s)/caregiver(s) and support systems', 'multiple

sclerosis and carer(s)/caregiver(s) and needs assessment' and 'multiple sclerosis and caring'. A broader and supplementary review with no time limits was applied to the search for 'carer burden' and 'QoI' in multiple sclerosis to determine the scope of the literature on how MS impacted on caregivers in terms of these concepts. The following databases were used: Pubmed, Psychlit, British Nursing Index, CINAHL, AMED, Nursing and Allied Health Care, Academic Science Premiere, Cochrane Library, Web of Knowledge, Ovid Nursing Database, Social Science Index and Joanna Briggs Evidence Based Source. A flow diagram illustrates the search strategy and its outcomes (Figure 1).

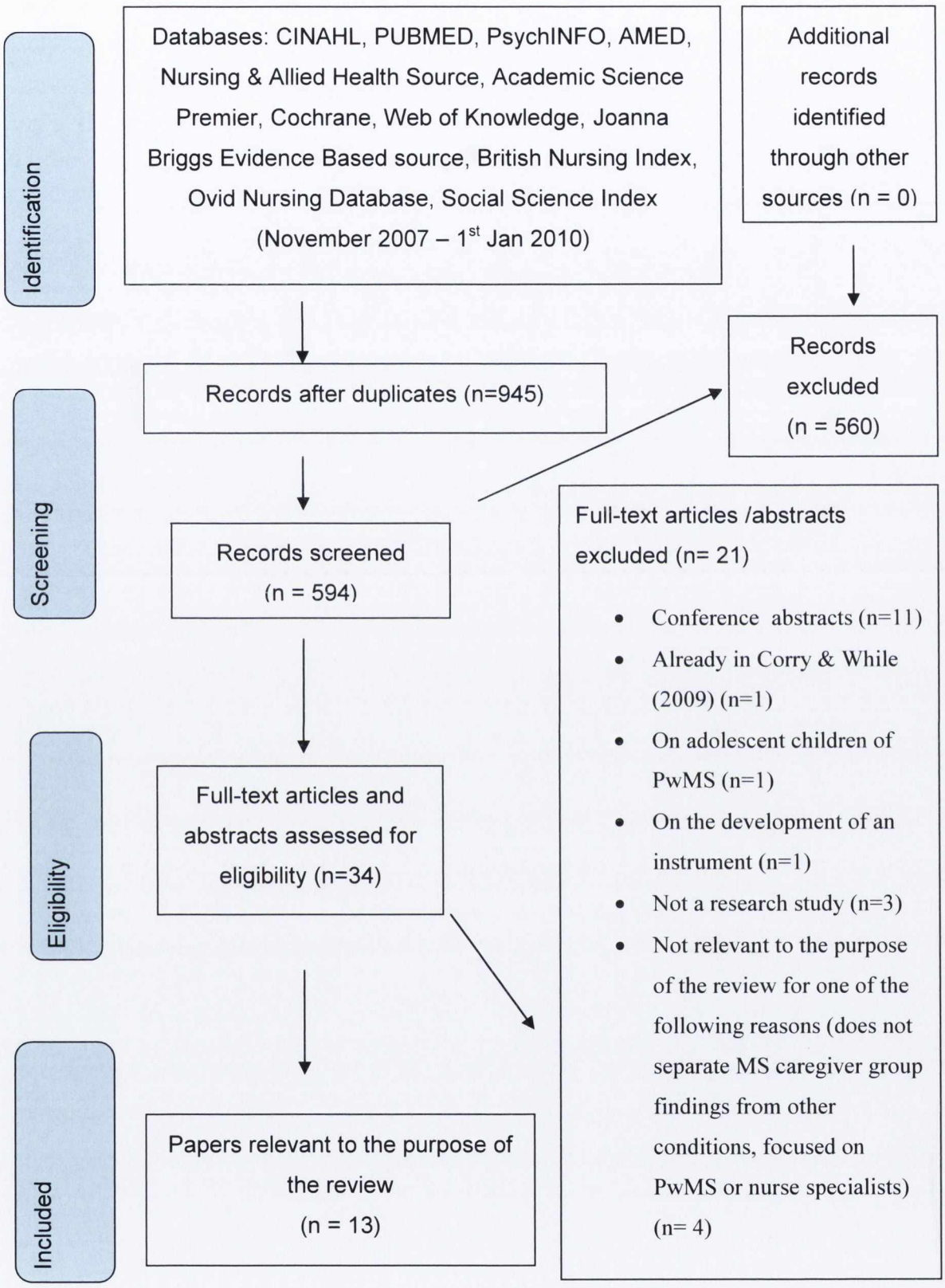


Figure 1: Flow diagram illustrating search strategy for needs and experiences of caregivers of PwMS

A total of 945 papers were found, of which 355 were duplicates. All titles and abstracts were checked for relevance to the purpose of this review. Papers were included if they reported findings on the needs and experiences of caregivers of PwMS. Thirteen original research papers were included. The studies were based on quantitative (n=11) and qualitative (n=2) research approaches (Appendix 1). Quantitative studies were primarily surveys, with one pilot intervention study. The two qualitative studies were descriptive in approach. The methodologies employed included surveys using questionnaires administered via post (n=2), telephone (n= 7), self-completion on-site methods (n=1) or some other self-completion approach that was not specified (n=1), telephone interviews with no identified structured questionnaire (n=1), face-to-face interviews (n=2) and face–to-face interviews using structured questionnaires (n=1).

The studies were carried out in different countries, with original research studies from the USA (n=8), Australia (n=3), Kuwait (n=1) and Spain (n=1). The needs that emerged from this review were identified from the way in which caring for a PwMS impacted upon the support person. These impacts were categorised in terms of the emotional impact (caregiver feelings, burden and QoL and satisfaction with life), social areas of need (effects on work life and recreational activities) and the physical impact (in terms of the need for information and use of support services). The strategies used and found helpful in improving caregiver outcomes were also reported. Furthermore, there was evidence from the literature that the variables of age, gender, marital status, educational level, employment status, length of time caregiving and hours caring/24hour period were similar across studies. These variables have been summarised in Appendix 2.

The impact of caregiving and the strategies used to improve caregiver outcomes are discussed in the next section. The supports available for caregivers of PwMS in the Republic of Ireland are then outlined and the literature to determine the extent to which such support services have been evaluated from the perspective of caregivers of PwMS is examined.

2.3 The effects of caring for PwMS on caregivers

2.3.1 Emotional impact

From an emotional perspective, caregivers reported that caring impacted on the extent of burden they experienced, their quality of life and their level of satisfaction with life.

2.3.1.1: Caregiver feelings and expressed needs

Four researchers reported that caregivers experience negative feelings (Alshubaili *et al.* 2008, McCabe *et al.* 2008a, Buchanan *et al.* 2009, Waldron-Perrine *et al.* 2009). The studies differed with respect to the feelings reported. For example, Alshubaili *et al.* (2008) reported that high numbers of caregivers experienced sadness, disgust, exhaustion and fear of having MS. Buchanan *et al.* (2009) reported that caregiving was demanding, physically and emotionally draining or in some way strained or threatened the relationship. Likewise, McCabe *et al.* (2008a) reported negative feelings about changes to working lives, such as concern about the financial impact of not having paid work, social isolation and insufficient activity to occupy them. These findings from McCabe *et al.* (2008a) must be interpreted with caution, as findings from caregivers of PwMS are reported along with those of other neurological illnesses. Other feelings expressed by caregivers included uncertainty regarding patient illness. Possible ranges on the Mishel Uncertainty in Illness Scale-Community Form (MUIS-C) (a 23 item questionnaire with a 5-point response option ranging from strongly agree to strongly disagree on items pertaining to patient symptomatology, diagnosis and treatment) were 23-115, with greater uncertainty equating with the higher score. Moderate to high scores were reported by Waldron-Perrine *et al.* (2009) (mean 61.9, SD 11.1) and as caregiver income, level of education or social support increased, less uncertainty in illness was found (Waldron-Perrine *et al.* 2009).

Despite the fact that, in general, caregivers experience the role of caregiving in MS as negative, some positive feelings were reported. Feelings such as pride, happiness, a sense of accomplishment and reward from the role were reported (Buchanan *et al.* 2009). Some caregivers felt that the role strengthened their relationship with the PwMS but happiness was linked with the extent to which they could support the PwMS (Buchanan *et al.* 2009).

Expressed needs varied from the need for assistance and information, to access to mental health services (Buchanan *et al.* 2009). The information needs ranged from the caregiver's personal need for information on how to manage their own time and maintain social life and friendships (Buchanan *et al.* 2009) to information on how to care for the PwMS (Finlayson & Dahl Garcia 2008, Buchanan *et al.* 2009). Three of ten respondents who felt that they would benefit from psychological support, reported that they did not wish to engage with a mental health professional (Buchanan *et al.* 2009).

2.3.1.2 Burden and quality of life

One study reported on caregiver burden (Rivera-Navarro *et al.* 2009). Although the majority of caregiver respondents in this Spanish study did not report burden as measured using the Zarit Caregiver Burden interview, for those that did, role emotional function and vitality on the SF-36 as well as the PwMS's level of disability were independent significant predictors of increased caregiver burden (Rivera-Navarro *et al.* 2009).

Three papers reported on caregiver quality of life but all used different instruments to measure the concept, making comparison across studies difficult (Alshubaili *et al.* 2008, Bogosian *et al.* 2009, Rivera-Navarro *et al.* 2009). Alshubaili *et al.* (2008) used the WHOQoL Brief questionnaire, Bogosian *et al.* (2009) reported findings from themes that emerged from a qualitative study, while Rivera-Navarro *et al.* (2009) used the Spanish version of the Medical Outcomes Study 36-item Short Form Health Survey (SF-36). Across all three studies, the severity of the care recipient's needs was a key factor influencing caregiver QoL. Those caring for people with RRMS reported higher QoI scores, thereby indicating greater QoL when compared with those caring for people with PPMS (Alshubaili *et al.* 2008). Similarly, Rivera-Navarro *et al.* (2009) found that the disability score of the PwMS was a statistically significant factor that impacted on caregiver QoL. Caregiver age, co-morbidity and female gender were also important factors that significantly impacted on caregiver QoL (Rivera-Navarro *et al.* 2009).

Another important finding was that caregivers of PwMS reported significantly lower scores to that reported in the general population on all domains of quality of life, except for environment (Alshubaili *et al.* 2008). The MS carer group did however report similar scores for the environment and general facet on health to carers of people with

diabetes and psychiatric patients. Although Bogosian *et al.* (2009) were reporting on the experiences of partners of PwMS in the early stages of MS, the findings reveal experiences that impact on QoL (Appendix 1).

2.3.1.3 Satisfaction with life and quality of care for the PwMS

Caregiver satisfaction was reported in relation to quality of care delivered to the PwMS (Buchanan *et al.* 2010) and caregivers' own satisfaction with life (Waldron-Perrine *et al.* 2009). The majority (60.9%) of respondents were satisfied with their lives (Waldron-Perrine *et al.* 2009). Factors contributing to caregivers' levels of satisfaction with life included their income, the severity of patient illness, social support and uncertainty in illness. Social support related to the extent to which provisions pertaining to attachment, social integration, reassurance of worth, reliable alliance, guidance and opportunity for nurturance was available to the caregivers (Waldron-Perrine *et al.* 2009). In addition, patient characteristics such as the PwMS's awareness of the deficit and their neuropsychological functioning were important contributing variables. These factors contributed 43.6% of the variance in caregiver satisfaction with life (Waldron-Perrine *et al.* 2009). Income, unawareness of deficit, caregiver uncertainty in illness followed by caregiver social support, illness severity and neuropsychological functioning made the most unique contributions to the prediction of caregiver satisfaction with life. A stronger adverse relation was found between PwMS' level of impairment and responses on satisfaction with life scales (SWLS) for caregivers with low scores on measurement of perceived social support, compared to caregivers with high scores on measurement of perceived social support (Waldron-Perrine *et al.* 2009). Caregivers were generally satisfied with the quality of routine care and MS-focused care offered to PwMS but were less satisfied with the access that PwMS had to home care and the quality of mental health care provided (Buchanan *et al.* 2010).

Caregiver satisfaction with life differed by gender. Waldron-Perrine *et al.* (2009) reported on factors that influenced satisfaction with life for female and male caregivers. These included illness severity, duration of caregiving and their experience of social support. Women caregivers experienced less satisfaction with life than men, as illness severity and duration of caregiving increased (Waldron-Perrine *et al.* 2009).

2.3.2 The social impact of caregiving

Two researchers reported on how the illness impacted on the caregiver's work or recreational activities (McCabe *et al.* 2008b, Buchanan *et al.* 2009). Both highlighted the negative impact of caregiving on work commitments. For many, caregiving duties posed difficulties with getting to work, maintaining employment or adhering to previously agreed working hours. Although some positive reactions to work changes were reported, most were negative and focused on the financial implications of the change, fear of social isolation or not having enough to keep them occupied at home (McCabe *et al.* 2008b). Caregivers also reported that illness had a negative impact on their social life and recreational activities (McCabe *et al.* 2008b).

2.3.3 The physical impact of caregiving

Three researchers reported on the time spent by caregivers providing care. The times ranged from an average of 2.5 hours per day (Finlayson & Cho 2008) to a minimum of 20 hours per week for more than 50% of caregivers (Finlayson & Dahl Garcia 2008, Buchanan *et al.* 2009). Instrumental care was the most common type of care/support provided for PwMS by their caregivers. The provision of this type of support ranged from 42% (Finlayson & Dahl Garcia 2008) to 90% (Buchanan *et al.* 2009) of caregivers. A smaller proportion of caregiving time was spent assisting with personal care, with study reports ranging from 16% (Buchanan *et al.* 2010) to 41% (Finlayson & Dahl Garcia 2008). In their study, Buchanan *et al.* (2010) reported that 11% of caregivers spent the largest amount of their caregiving time providing psychological or emotional care and that more than 40% of carers reported that assistance was also provided by other carers, either paid or unpaid. Personal assistance was more likely to be provided by a non-spousal carer (Finlayson & Cho 2008). Along with transport provision, this was one of the most challenging issues.

2.3.3.1 Use of support services

Three researchers (McCabe *et al.* 2008a, Buchanan *et al.* 2009, Buchanan *et al.* 2010) reported on the use of support services by caregivers of PwMS. There was greater use of support services to help carers provide care for the PwMS than use of support services by carers for their own personal needs. This highlights the requirement for the

development of supports for those caring for PwMS. For example, Buchanan *et al.* (2009) reported that only one caregiver in eight participated in support groups for informal/unpaid caregivers, yet Buchanan *et al.* (2010) reported that up to 91% used aids to help them care for the PwMS. The internet was a major source of support and information (Buchanan *et al.* 2009). In most studies there was no evidence of use of professional support services, with McCabe *et al.* (2008b) reporting very little use of respite care or other professional services for advice. Among carers who did use the services, those that provided home help, cleaning and assistance with meals and funding for transport or recreational activities were identified as the most helpful (McCabe *et al.* 2008b). Professional societies and support groups were also reported as helpful. Respite assistance was reported to be less helpful. Ideally, carers would like more help in the home and more respite care (McCabe *et al.* 2008b).

Although no Irish study was identified for this up-dated review, Corry & While (2009) included a paper by McKeown *et al.* (2004) that reported findings specifically related to 'support' from an Irish perspective. This paper reflected services from northern Ireland and the Republic of Ireland, which differ in terms of political governance and health care funding, structure and provision (Table 1). Other services used in southern Ireland were identified through internet searchers and personal communications (see Table 1).

Table 1: Services used by caregivers of PwMS in Ireland

Services used by caregivers of PwMS (McKeown *et al.* 2004)	Other services identified though internet searches and personal communications with NSsMS
• Respite • Occupational Therapy • MS organisations • Carer organisations • Churches	• Professional supports (neurologists, MS nurse, public health nurses) • Services (respite care, social welfare support)

2.3.4 Strategies to improve caregiver outcomes

Few significant findings were reported from the only published study found on the outcomes of an education programme to meet the challenges of MS (Finlayson *et al.* 2009). The programme was found to improve carers' preparedness to care as identified from the Caregiver Preparedness Scale (t=-2.51, df=18, p=0.02). Additional evidence

for the potential benefit of the programme was found in the qualitative findings, where respondents reported increased confidence in their role as carers, feelings of less anger, feeling happier and an awareness of the need to plan and ask for help. The findings from this study are limited by the small sample size (n=19) and the use of a pre-post test design. The provision of social support was reported to positively impact on caregivers' satisfaction with life for women, but not for men (Waldron-Perrine *et al.* 2009).

2.4 The needs of caregivers of PwMS

This up-dated review provides additional supporting evidence for the findings previously reported with respect to the many unmet needs of caregivers (McKeown *et al.* 2003, Corry & While 2009). Providing care to a PwMS was found to negatively impact on caregivers physical, psychological, social and financial wellbeing, which result in increased carer burden and reduced QoL (McKeown *et al.* 2004 , Corry & While 2009). In addition to highlighting the ongoing and recurring impact of caring on caregivers of PwMS, this review identifies new areas of need, such as dealing with uncertainty around the illness and the factors that influence this uncertainty.

Caring experiences ranged from being physically and emotionally draining, to a sense of accomplishment and reward from the role. This supports the theory of 'losses and gains' in MS caregiving. There was no evidence to substantiate the findings that caregiving was a "... state of chronic sorrow" (McKeown *et al.* 2003, p. 243), but the extent of negative experiences reported in the literature give weight to the heavy emotional burden of caregiving. It is important to note that despite the overwhelming evidence supporting the negative psychological impact of caregiving in MS, a minority of caregivers report positive experiences, such as pride and a sense of accomplishment in the caregiving role.

A new finding from this current review relates to caregivers' feelings of uncertainty regarding the PwMS' illness. This feeling is likely to reflect the individual and unpredictable course of the MS condition. Of note, the factors that significantly impact on this feeling of worry included caregiver income, level of education and social support. Although these factors are amenable to intervention, and any attempt to address them has the potential to reduce caregiver uncertainty, it is difficult to surmise

how these factors actually impact on uncertainty. For example, level of education could relate to the caregiver's ability to interpret information on MS or could relate to their own sense of competence to cope into the future. Irrespective of the reasons, it is important to offer support to those engaged in the role of caregiver for PwMS.

2.4.1 The need for measures to address the impact of caregiving on QoL

The impact of caring on caregivers' QoL is a recurring theme in research studies (McKeown *et al.* 2003, Alshubaili *et al.* 2008, Bogosian *et al.* 2009, Corry & While 2009, Rivera-Navarro *et al.* 2009). According to Corry & While (2009), carers who experienced 'strain' also experienced greater reductions in QoL. Factors associated with reduced QoL for carers of PwMS included being a spouse, the duration and frequency of caring, increased severity of recipient symptoms and an unstable disease course (McKeown *et al.* 2003). These findings are reflected in the studies included in this up-dated review and other papers reporting on caregiver burden and QoL for caregivers of PwMS. The factors reported to impact on burden for this group of caregivers include a primary progressive type of MS (Alshubaili *et al.* 2008, Rivera-Navarro *et al.* 2009) female gender, number of carer co-morbidities, increased age of carer and recipient disability (Patti *et al.* 2007, Rivera-Navarro *et al.* 2009), carer burden (Knight *et al.* 1997), coping satisfaction and coping support (Knight *et al.* 1997) and spousal relationship (Figved *et al.* 2007).

The findings from this review suggest that caregivers of PwMS have a reduced QoL relative to the general population. Many of the influencing factors are not easily changed such as age, comorbidity and gender. Other factors, such as the severity of the care recipient needs may be amenable to some intervention but these are likely to be limited in many instances. Despite the use of recognised measures of QoL, only Forbes *et al.* (2007) report on the psychometric properties of their questionnaire. This may be because the instruments are already validated and internationally accepted QoL measurement questionnaires. None of the instruments used to measure QoL were caregiver specific tools, which may reflect the view that carers are not patients. The lack of theoretical exploration of the concept of QoL or clear operational definition for research purposes provides challenges when drawing conclusions from various studies. The limitations of the studies in the caregiver MS literature is reflective of the general literature on QoL, which confirms that the concept is multidimensional, a factor

that has challenged researchers in their attempts to define and contain the concept (Kleinpell 1991, Meeberg 1993, Haas 1999, Mandzuk & McMillan 2005).

2.4.2 The need for measures to address the impact of caregiving burden

Further evidence to support the previously identified link between burden and QoL reported by Corry & While (2009) is provided by Rivera-Navarro *et al.* (2009). Rivera-Navarro *et al.* (2009) reported that along the disability level of the PwMS, the two QoL domains as measured using the SF-36 instrument were the only remaining items found to significantly influence burden scores during the final modelling phase of data analysis. Previous studies demonstrated the impact of burden on QoL, but Rivera-Navarro *et al.* (2009) demonstrated that QoL impacts on burden, which is a new perspective that was not previously considered. It is likely that the relationship between the two concepts is reciprocal and improvements in one would help improve the other. It can be argued that any experience that puts a strain on caregivers could be conceptualised as burden, given that the concept is broad with blurred boundaries. This finding suggests that, in addition to interventions to alleviate caregiver burden and improve their QoL, there is a need for large surveys to further explore and confirm the relationship between the two variables. It is important that caution is used when interpreting the findings from studies pertaining to caregiver burden, as most studies on caregiver burden used convenience sampling methods, which threatens the external validity of the findings. In addition, the concept of burden from the perspective of caregivers of PwMS has not been explored.

Buhse (2008) explored the concept of burden and its assessment in relation to families of PwMS. From the definitions provided, it can be concluded that burden, like QoL, is a multidimensional concept that can be viewed in terms of the experience of strain, stress or load, as a result of the caring role. Burden can also be viewed objectively, in terms of externally observable phenomena, such as the financial impact of caring, or subjectively, in terms of how it is perceived by the individual (Buhse 2008). Despite the focus on assessing burden in families of PwMS, no MS caregiver specific definition of burden is provided. This suggests that either the broader and general concept of burden is immediately transferable to caregivers of PwMS or it has not been conceptualised from the unique perspective of this group of caregivers. The findings from a secondary and comprehensive search and analysis of the literature examining

the range of studies exploring the concept of burden and QoL in MS for the purpose of this doctoral research, failed to provide a working definition of the concept of burden from the unique perspective of carers of PwMS. Of the 41 papers that explored issues that could be categorised as burden-related, only nine focused on the concept of burden encompassing the associated terms such as strain and stress (O'Brien 1993, Knight *et al.* 1997, Chipchase & Lincoln 2001, Ybema *et al.* 2002, Rivera-Navarro *et al.* 2003, Pakenham 2005b, Forbes *et al.* 2007, Khan *et al.* 2007b, Rivera-Navarro *et al.* 2009). Some researchers referred to previous research findings on burden as part of their background and discussion, but none provided a working definition of the concept. This suggests that the concept is poorly understood particularly in relation to caregivers of PwMS. An exploration of the general literature on the concept confirmed a lack of clarity on the concept (Loukissa 1994, Hoffmann & Mitchell 1998).

2.4.3 The need for measures to address the impact of time spent caregiving

As a disease progresses, caregivers are likely to spend more time providing care. The considerable amount of time that caregivers spend caring (McKeown *et al.* 2003) is supported in this updated review by the two studies (Finlayson & Dahl Garcia 2008, Buchanan *et al.* 2009) exploring time spent on caregiving activities. The finding that more than half of caregivers reported spending a minimum of 20 hours/week caregiving is noteworthy, given that both studies included reasonably large sample sizes (302 and 530). Of similar importance is the finding that personal assistance, such as helping PwMS with elimination was more likely to be provided by non-spousal carers (Finlayson & Cho 2008). Assistance with elimination and providing transportation were identified as the two most challenging areas for the provision of assistance (Finlayson & Cho 2008). Overall, spousal carers were found to provide assistance with more activities than non-spousal carers (p=0.01) (Finlayson & Cho 2008). The most common type of care is instrumental in nature, accounting for between 42% and 90% of the care provided. Instrumental care refers to activities such as general housework, cooking and transportation. The variety of tasks caregivers are required to perform are likely to increase as the disease progresses (Corry & While 2009). Likewise caregivers are more likely to experience loss of earnings due to reducing hours of paid employment, while others may find that becoming a carer has a negative impact on their career and employment prospects (Corry & While 2009). It is important that NSsMS have an

understanding of the type of care that is provided by caregivers of PwMS and how this impacts on them, so that they can put strategies in place to minimise caregiver burden.

2.4.4 The need for interventions to improve caregiver satisfaction

Although caregiver satisfaction was not a major point of discussion in the two previously published reviews, researchers identified that social support was elementary to levels of satisfaction (McKeown *et al.* 2003, Corry & While 2009). In fact, social support was identified as the strongest individual predictor of carer satisfaction with life (SWL) (Corry & While 2009). Other satisfaction related factors included the income of the significant other, disease characteristics experienced by the PwMS, the care requirements, caregiver depression and anxiety (Corry & While 2009). These findings are reflected elsewhere with respect to income, severity of patient illness and social support (Waldron-Perrine *et al.* 2009); and are highlighted as key influencing variables for caregiver SWL. In addition, illness uncertainly is of importance, as it was identified as one of the key factors that contributed to 43.6% of the variance in caregiver satisfaction with life (Waldron-Perrine *et al.* 2009). In terms of satisfaction with care offered to PwMS, access to home care and the quality of mental health care provided were identified as areas of concern (Buchanan *et al.* 2010). It is important, however, to balance the reported negative experiences of caregivers of PwMS with the fact that the majority of caregivers were satisfied with their own lives (Waldron-Perrine *et al.* 2009) and were in the main, satisfied with the quality of routine care delivered to the PwMS (Buchanan *et al.* 2010).

The finding of inadequacy and dissatisfaction with available services among caregivers of PwMS has been widely identified (Buchanan *et al.* 2009, Corry & While 2009, Buchanan *et al.* 2010). The finding that 3 in 10 respondents who felt that they would benefit from psychological support but did not wish to engage with a mental health professional (Buchanan *et al.* 2009), suggests a need exists to educate and support carers to come to terms with accepting help.

2.4.5 Supports used and strategies to improve outcomes for carers of PwMS

No published study specifically evaluated services from an MS carer perspective. Service impact on the lives of carers of PwMS is therefore, unknown. It is also unknown if the range of services, their extent of availability and ease of access to them, meets the needs of caregivers of PwMS. From the literature reviewed, ease of access to services was identified as a source of burden for carers (Buchanan *et al.* 2010). Although only 13% of carers spent a lot of time negotiating the health care system, it was a source of considerable burden. In fact 18% of caregivers indicated that large amounts of their burden and frustrations were caused by negotiating the health care system (Buchanan *et al.* 2010). Nurse Specialists in MS are the co-ordinators of care and are therefore in a unique position to assist caregivers and PwMS with negotiating the health system (Corry *et al.* 2011).

The literature suggests that the use of an educational programme improves caregiver outcomes (Finlayson & Dahl Garcia 2008, Finlayson *et al.* 2009a). This educational programme was offered by occupational therapists in the United States of America (USA) in a face-to-face setting. Although findings were promising, they are inconclusive, given the pilot nature of the study and the use of a pre-post-test design. Other strategies to improve outcomes include caregiver coping strategies (O' Brien 1993) feeling appreciated (Wollin & Sato 2001) and getting more support from others, especially friends and relatives (Ybema *et al.* 2002). Carer needs included the need for regular breaks from the caring role (Wollin & Sato 2001). Coping strategies used by caregivers included generating a better understanding of their problems through problem analysis, seeking solutions to problems, following through on planned actions, having respect for others' views, increasing efforts to make things work and seeking information and advice from others (O' Brien 1993). As caregiver stress increased, so too did the use of coping strategies, in particular the use of detachment ($r=0.76$, $p<0.002$), problem focused coping ($r=0.44$, $p<0.02$), wishful thinking ($r=0.45$, $p<0.01$), tension reduction ($r=0.36$, $p<0.05$), keeping to self ($r=0.41$, $p<0.04$), and positive thinking ($r=0.37$, $p<0.04$) (O' Brien 1993). Other variables found to be helpful included interest, support from health professionals and from family and friends, access to information, feeling that their own lives were productive and being encouraged to keep control over their own lives, as well as financial security. These were among the top 10 needs of significant others of PwMS (Koopman *et al.* 2006).

These findings suggest that from the perspective of caregivers, strategies that may be helpful include preparation for the role, mechanisms to improve coping abilities, appreciation and support from family, friends and health professionals, access to information, scheduled breaks from caregiving and the opportunity to develop their own lives. Many of these strategies fall within the scope of the role of the nurse (Sato *et al.* 1996). Indeed nurses can address the needs of this group of caregivers by teaching them about the disease process and how to do technical things (Sato *et al.* 1996). Additional contributions by nurses include listening and demonstrating understanding and sending out newsletters and other information on new developments (Sato *et al.* 1996).

2.5 Chapter summary

The literature presented in this chapter comprised a literature review that was conducted during the development of this study, together with an up-dated review that was conducted in March 2010. This review summarised the effect of caring on caregivers from a physical, social and emotional perspective. Issues surrounding burden and QoL were addressed, together with levels of satisfaction among caregivers of PwMS. The importance of social support to caregiver satisfaction and the notion that MS caregivers spend a considerable amount of time caring was highlighted. The majority of time is spent on instrumental care tasks. Although caregivers of PwMS were generally satisfied with their lives and the quality of routine care offered to PwMS, they were less satisfied with home care provision and the quality of mental health services for PwMS.

From this review, it can be concluded that caregivers of PwMS need professional support, information, help with managing the burden of caregiving, negotiating and gaining access to the health care system, managing their feelings and the uncertainty surrounding the condition. Caregiver reluctance to engage with mental health professionals for counselling and support is of importance and warrants further investigation. There is a need to prepare and support caregivers for their caregiving role in an effort to reduce the burden imposed upon them and improve the quality of their lives. The development of an intervention for use by NSsMS in their supporting role is one means by which caregivers could be supported. Consequently, the

intervention reported in this thesis was developed and tested to enable NSsMS to support caregivers of PwMS.

Chapter 3: The theoretical development of the intervention

3.1 Introduction

Chapter one provided an overview of the literature which highlighted that caregivers have unmet needs. Many of the identified unmet needs fall within the scope of the role of the nurse, in particular that of the NSMS. These needs have also been shown in smaller scale unpublished national surveys. For example, a small scale study (Phelan 2010) using a sample size of 50, found that carers of PwMS in the west of Ireland were not always aware of the support services available to them in the community, such as respite services and education/training on how to provide care for a PwMS. This low use of community services is similar to that reported by carers of PwMS internationally. The findings echo those in an unpublished report from MSI (Multiple Sclerosis Society of Ireland 2006), where it was reported that carers (N=83) identified the need for training, emotional support/counselling and information in providing or developing services.

Despite the large volume of literature identifying needs, there is a lack of intervention studies to address them. With the exception of the work of Finlayson et al. (2009), an occupational therapist who pilot tested an intervention for caregivers of PwMS, no support intervention has been found in the literature for this group of caregivers. It was therefore important to consider how their unmet needs could be addressed, from a nursing perspective. This required the development of an intervention congruent with the role of the nurse specialist in the Irish health care context. This chapter provides an overview of the factors that required consideration when developing and testing such an intervention. This included an analysis of the professional environment within which NSsMS practiced and the scope of the role of the NSMS. It also necessitated the identification of an overall framework to guide the development of the intervention. The literature reviewed to inform the method of delivering the intervention is also presented here, including a review of reviews of caregiver interventions for people with chronic illnesses and a review of telephone support interventions for caregivers.

3.2 Context influencing the development and delivery of the intervention

The process for formal recognition of the existence of nurse specialists in Ireland began in 2000 (Health Service Executive and National Council for the Professional Development of Nursing & Midwifery 2008). The first nurse specialist role was ratified by the National Council for Nursing & Midwifery in 2000 (National Council for the Professional Development of Nursing and Midwifery 2010) and the first NSMS was appointed soon after 2002 (personal communications via email with Orla Coady, An Bord Altranais, Ireland). Since then, nine nurses have specialised in MS (Duggan & Mckenna 2011). NSsMS are nurses who have specialised in caring for PwMS and their families/carers [hereafter referred to as nominated support persons (SPs)]. Their role includes the provision of support and education to PwMS and SPs (Duggan & Mckenna 2011). All newly diagnosed patients in the public health care system are referred to the neurology nurse specialist (personal communications, NSMS). In addition to face-to-face contact with PwMS and their SPs, as part of usual care, NSsMS provide a telephone support service on demand. In general, SPs use the service for issues pertaining to the care of the PwMS. Issues pertaining to their own needs are usually only raised when a situation has reached or is near crisis point. To date, there is no evidence in the literature that the service has been evaluated in terms of caregiver outcomes. More importantly, however, is the need to address the unique needs of SPs so that they make contact with NSsMS in a timely manner and address issues/concerns at an early stage.

NSsMS are in a unique position to provide individualised information and advice to PwMS and their SPs. They have knowledge about the individual patient's medical diagnosis and are professionally trained to provide information, advice and support on MS and its treatment. They have ready access to the patient's neurologist and other members of the multi-disciplinary team. Therefore, unlike other voluntary organisation or health care workers, NSsMS can provide individualised and specialised support to SPs. To identify what role NSsMS play in providing support to caregivers of PwMS and to identify what contribution they can make to reduce caregiver burden and increase caregiver QoL, as part of this doctoral study, a literature review was carried out on the role of the clinical nurse specialist in multiple sclerosis (Corry *et al.* 2011). This review (Corry *et al.* 2011) highlighted that although there is a dearth of quality research

evaluating the impact of the NSMS, there is increasing evidence on the potential of the role in terms of quality patient care. The review also highlighted the lack of formal acknowledgement of the contribution that NSsMS make to the wider family and caregivers who provide on-going care for PwMS at home.

3.2.1 Scope of nursing practice in Ireland

In order to enable the NSsMS in this study to work within the boundaries of safe practice and be in a position to impact on the study outcomes, it was essential that the goal of the intervention was within the scope of the role of the NSMS. Likewise, if the impact of the intervention on outcomes was to be shown, the outcomes tested had to be amenable to nursing intervention (Corry *et al.* 2013). This meant that the goal and theoretical model guiding the intervention had to reflect the beliefs and values underpinning the practice of nursing and be within the scope of nursing practice (An Bord Altranais 2000). The beliefs underpinning nursing practice in Ireland can be found in the definition of nursing as agreed by An Bord Altranais (2000), the professional body governing nursing practice in this country:

> "Nursing helps individuals, families and groups to determine and achieve their physical, mental and social potential, and to do so within the challenging context of the environment in which they live and work..... Nursing promotes the active involvement of the individual and his or her family, friends, social group and community, as appropriate, in all aspects of health care, thus encouraging self-reliance and self-determination while promoting a healthy environment".
>
> (An Bord Altranais 2000, p. 3)

The core values underpinning the practice of nursing, in particular the first three values, were crucial in guiding the design and content of the intervention for this study.

> "1. In making decisions about an individual nurse's scope of practice, the best interests of the patient/client and the importance of promoting and maintaining the highest standards of quality in the health services, should be foremost.

2. Nursing care should be delivered in a way that respects the uniqueness and dignity of each patient/client regardless of culture or religion.

3. Fundamental to nursing practice is the therapeutic relationship between the nurse and the patient/client that is based on trust, understanding, compassion, support and serves to empower the patient/client to make life choice"

(An Bord Altranais, 2000, p.3)

It was essential that the model selected to guide the intervention enabled the NSsMS to individualise their help and promote involvement of the SPs within the scope of the role of the nurse. Other key factors that guided the selection of the model for this intervention included the reported potential benefits of theories and models guiding published studies on caregiver interventions, and the methods used to deliver them. The successful implementation of the intervention required a method of delivery that was feasible to implement in the environment within which NSsMS practiced. To identify these key factors, a series of literature reviews were carried out, to inform the selection of the intervention model used.

3.3 Approach to reviewing the literature for the development of the intervention

A systematic and incremental approach was used to review the literature (Figure 2). The focus of each successive review was informed by the outcomes of the preceding one and concurrent field work, which involved interviews with NSsMS. The developmental process commenced with a review of frameworks and guidelines for developing interventions. This was followed by a review of literature reviews on interventions for caregivers of people with chronic health conditions. This, along with an analysis of the context of care within which the intervention was to be delivered led to the identification of the telephone as a suitable medium through which the intervention could be delivered. Models of communication were considered to guide the delivery of the intervention, however, no suitable model was identified from the literature reviewed. The findings from the literature review on telephone interventions revealed that models

of problem-solving had the potential to impact change for caregivers. The review of models of problem-solving is presented in chapter 4. Through my on-going reflection on the findings of the reviews, personal educational knowledge, clinical experience of caring for PwMS and their families, and the scope of the role of the NSMS, Egan's Model of the Skilled Helper emerged as a suitable model to guide the development and delivery of the intervention.

Due to the number of literature reviews carried out to inform the development of this intervention (Figure 2) and the resultant volume of literature reviewed, the search strategies are presented using flowcharts (Figures 5-7). The presentation of the findings of these literature reviews are presented as summaries of the included studies as provided in Tables 2-4 and Appendix 3, and Tables 6-7 in chapter 4. The discussion of the findings for each review is confined to those aspects of the reviews that guide the identification of the method of delivering the intervention for this study.

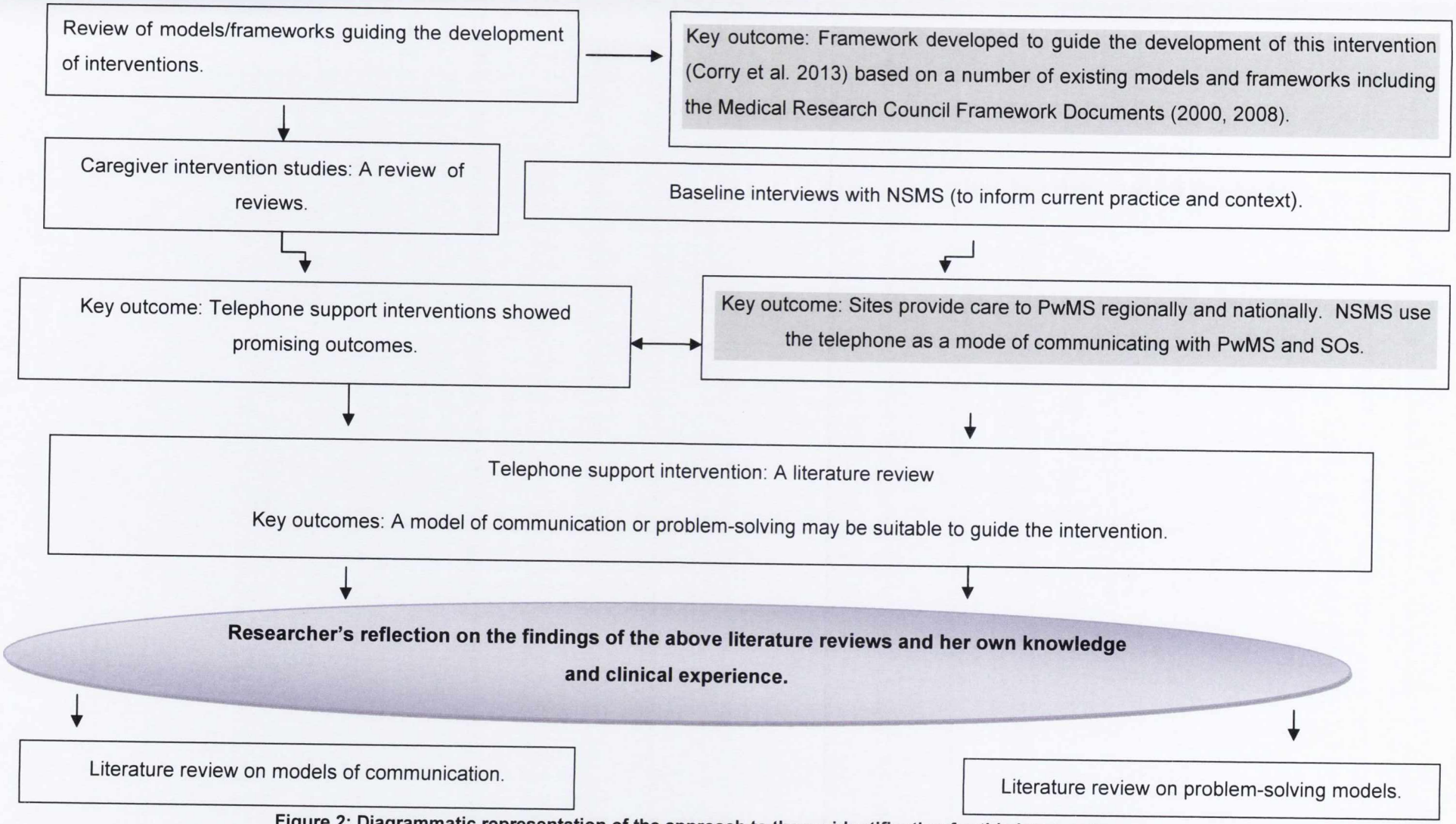


Figure 2: Diagrammatic representation of the approach to theory identification for this intervention

3.4 Quality appraisal criteria for literature reviews and original studies

The quality assessment criteria used to evaluate the included papers differed according to the nature of each review. For example, the quality of the systematic reviews were evaluated using R-AMSTAR, an 11-item assessment tool with the inclusion of a 4 point quality rating against unique criterion for each item (Kung *et al.* 2010). The quality ratings on this scale range from 11 [satisfies no criterion] to 44 [satisfies all criteria] (Appendix 4).

Systematic reviews, in particular those of randomised trials, are reviews of strong quality literature and are considered to provide the strongest evidence on the effects of interventions (Akonbeng 2005). The inclusion of only those reviews with strong quality systematic review ratings significantly limits researchers synthesising the available evidence in areas where a number of reviews exist but the research available is limited by research design and sample size. As the number of strong quality systematic reviews on the selected topic for this review was limited and the purpose of this review was to inform the development of an intervention, all systematic reviews published in peer reviewed journals were included and were rated using R-AMSTAR.

The R-AMSTAR does not differentiate between reviews using strong research designs only, such as the randomised trial and those including designs without concurrent control groups, for example, pre-post-test or post-test designs. In addition, the R-AMSTAR gives equal weighting to each of the 11 items and therefore does not differentiate between their importance [eg. comprehensiveness of the search and rating of the quality of the included papers versus reporting conflict of interest]. To address this shortcoming, additional criteria were devised for the purpose of this study. Five essential R-AMSTAR items were identified [Items 2,3,6,7 and 8] (Appendix 5) and categorised alongside research study designs, which were ordered in accordance with the hierarchy of evidence (Akonbeng 2005). Reviews receiving a rating of B+ or above were categorised as strong quality [at a minimum research designs included a control group with positive rating for all key items on R-AMSTAR], those that received a rating of between C+ and ≤ B were considered to be of moderate quality [at a minimum reviews included research designs with a control group and those rated below C+ were

considered as weak quality [reviews included before and after studies with no control groups]. (Appendix 5). Table 2 presents a summary of the included reviews.

The CONSORT 2010 checklist of information to include when reporting a randomised trial (www.consort-statement.org) and a modified version of the Critical Appraisal Skills Programme (CASP) (Appendix 6) were used to inform the appraisal of the quality of the studies included in the telephone intervention literature review (see 3.6.2.1 for further details)

The process for the development of the intervention for this study commenced with a review of models, frameworks and guidelines for developing interventions.

3.5 Frameworks and guidelines for developing interventions

In order to identify a framework to guide the development of the intervention for this study, a literature review was carried out on the area of complex interventions (Corry et al. 2013). The MRC (2000) framework guidelines are used widely for developing complex interventions and were developed to help researchers provide transparency in the development of interventions for health care by enabling them to provide a comprehensive account of their intervention development. Adhering to the guidelines facilitates the development of interventions that are grounded in best practice. Although the MRC (2000) framework and more recently updated MRC (2008) guidelines provide good guidance for intervention development, other guidelines also provide important guidance for the development of interventions for nursing practice (Corry et al. 2013). These published guidelines were reviewed and synthesised to create a model to guide the development of the intervention for this study (Figure 3) (Corry et al. 2013). Underpinning this model were several core principles underpinning intervention development, which include identifying the theory and research, building and representing the intervention, determining acceptability and planning, and intervention delivery (Corry *et al.* 2013). This model (Corry et al. 2013) differed from other published models in that it made explicit the need to base all stages of intervention development and testing on available evidence and to take cognisance of the scope of the role of the nurse. The model also makes explicit the influence of the researcher's experience and expertise in the development of the intervention. In order to increase the likelihood of the successful implementation of the intervention, it was essential that it was within the scope of the role of the NSMS. Consequently, the

model presented in Figure 3 was used in conjunction with the MRC (2008) guidelines (Figure 4) to frame the development of the intervention in this study.

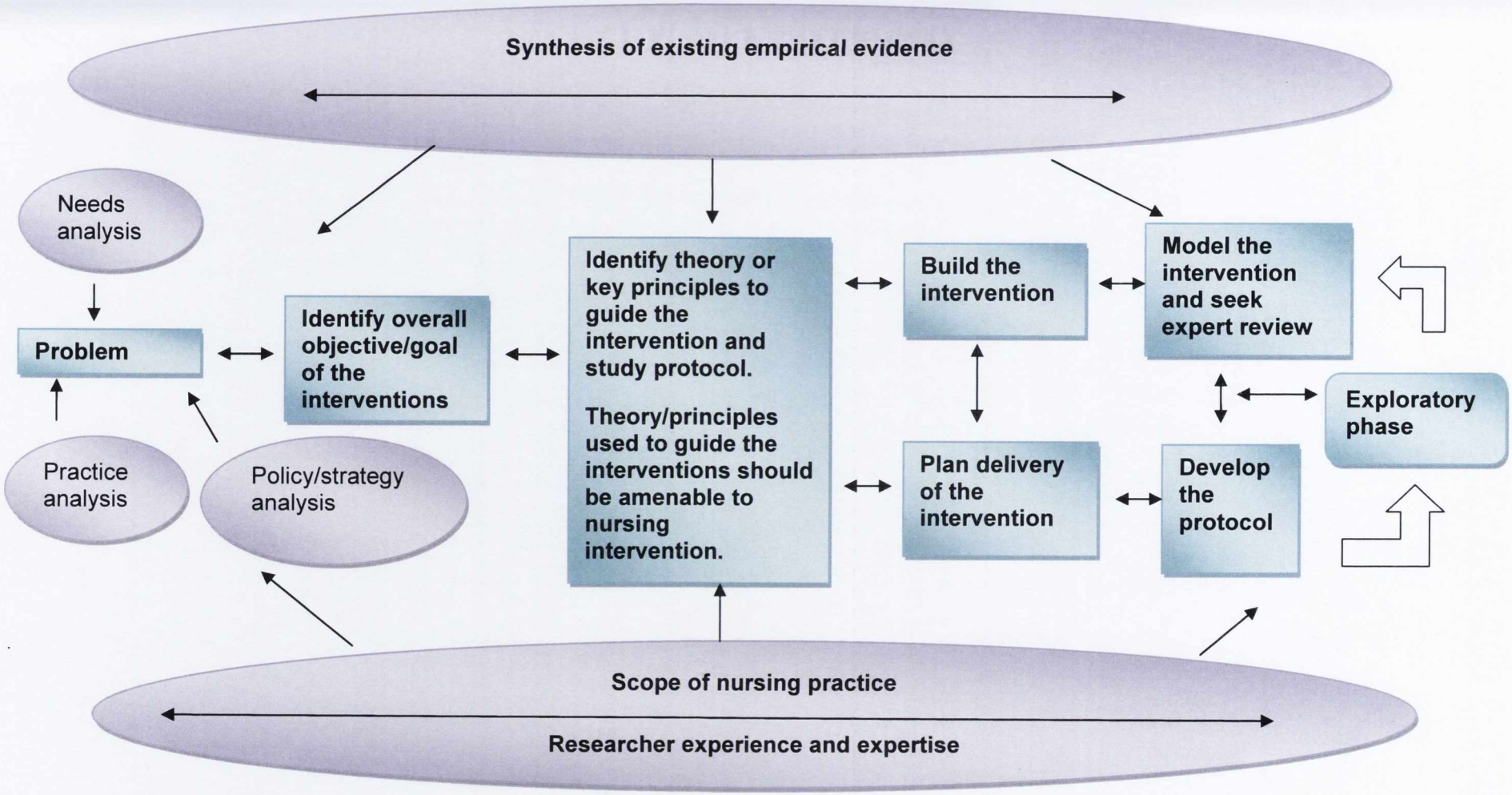


Figure 3: Model for developing complex interventions for nursing (Corry *et al.* 2013, p.2381)

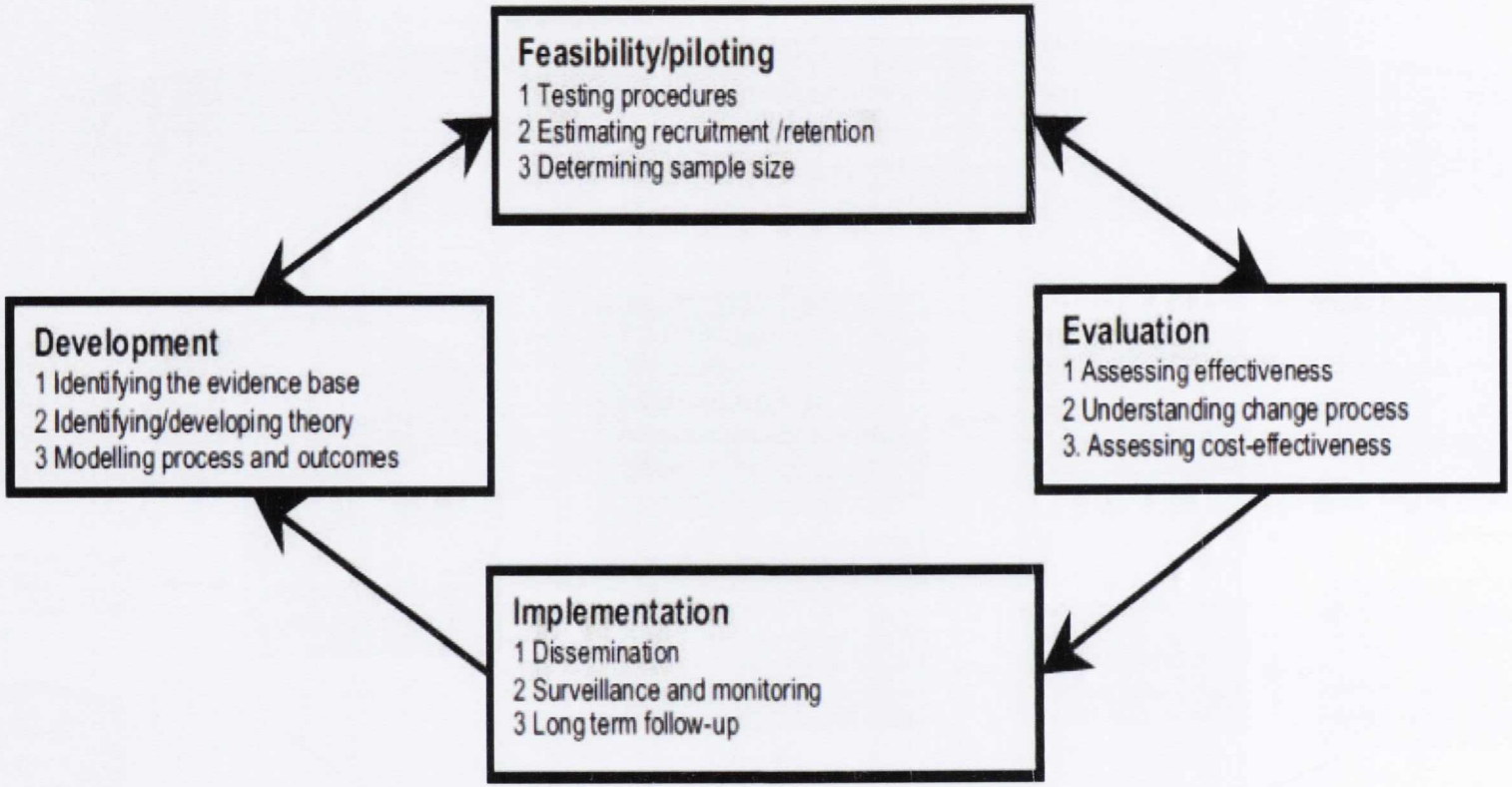


Figure 4: Key elements of the development and evaluation process (MRC 2008, p.8)

In this study, the problem was identified from the literature (presented in chapter 2), which confirmed that internationally caregivers have unmet needs, some of which fall within the scope of the role of the NSMS, such as providing information and advice that empowers them to make decisions (An Bord Altranais 2000). The overall purpose of this study was, therefore, to develop and test a nursing intervention to meet these caregiver needs. Development of the goal and objectives of the intervention was an ongoing process informed by the overall purpose of the study, the approach for delivering the intervention and the scope of the role of the NSMS.

3.6 Identifying/developing theory for the development of the intervention

In order to identify and develop a theory for this study, it was necessary to determine what type of intervention was likely to produce positive outcomes for caregivers. The process, therefore, commenced with a review of systematic reviews of caregiver interventions.

3.6.1 Caregiver interventions in chronic conditions: A review of reviews

Review papers were included if they reported on the outcomes of interventions for unpaid caregivers of people with chronic conditions who reside in the community. To be included, the primary focus of the interventions had to be to assist the caregivers in their role in supporting people with chronic conditions, with a view to improving the physical and psychological well-being of the caregiver. Chronic conditions were defined as long term conditions which are incurable, progressive, constant or relapsing/episodic (Rolland 1987) and involved some functional impairment or disability which lasted for 6 months or longer (Department of Health and Children (Ireland) 2008). As the purpose of the review of reviews of interventions for caregivers was to inform the development of this intervention for NSsMS to support caregivers of PwMS, papers that focused solely on the outcomes of respite care, the introduction of a health care service or peer support, were excluded. Papers were included if they referred to issues that NSsMS were likely to encounter and influence in their nursing roles. These included quality of life, burden and supporting caregiver abilities (e.g. knowledge, preparedness and self-concept). Likewise, as the purpose of this review of reviews was also to provide an overview from a range of interventions and their methods of delivery,

along with providing guidance on the topic for more focused literature reviews, no restrictions were placed on the research designs of reviews included. The search was carried out in September 2011. A flow chart outlining the literature search and selection of systematic reviews is shown in Figure 5.

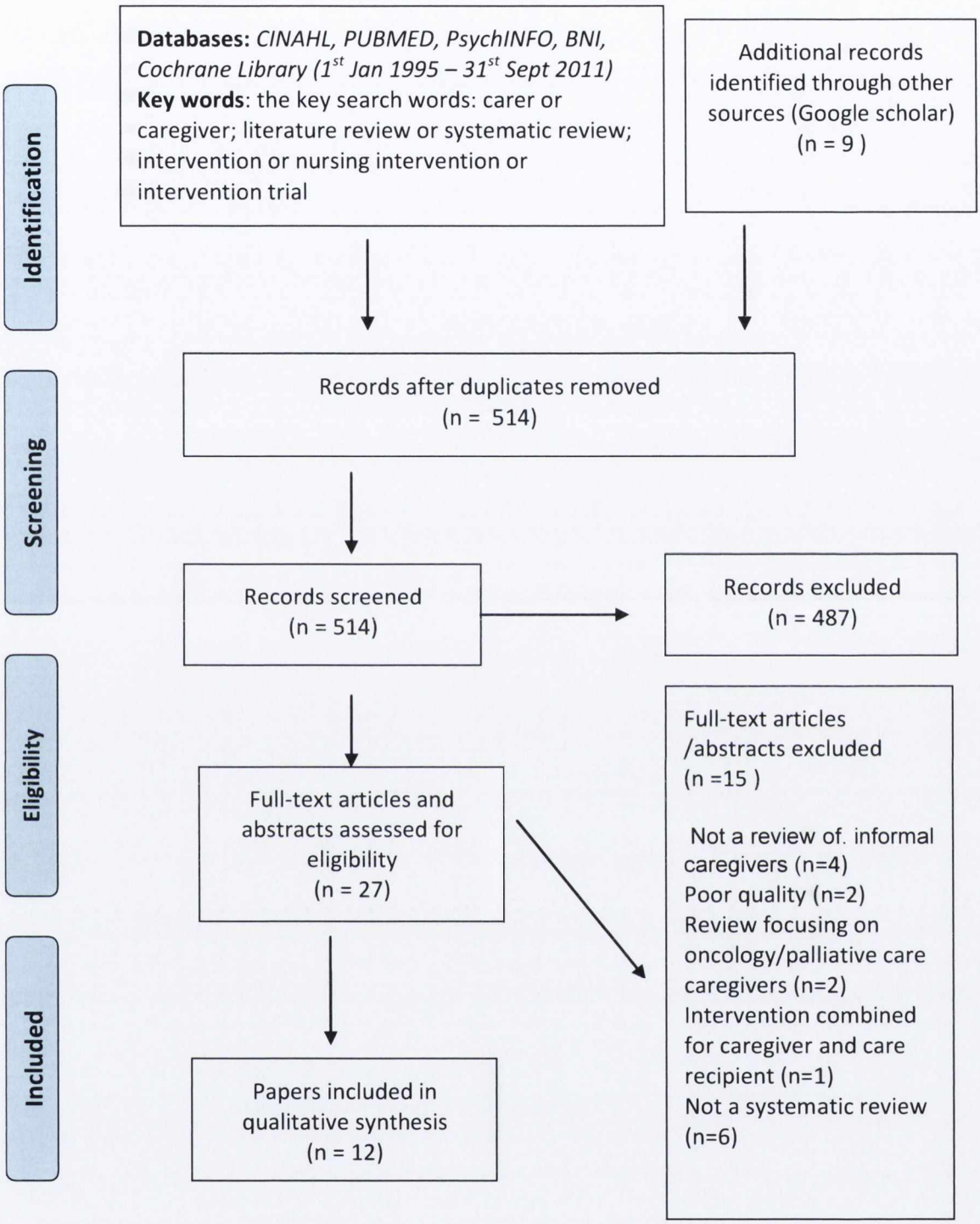


Figure 5: Flow chart for the literature search and selection of systematic reviews for the review of reviews of interventions for caregivers

Twelve systematic reviews fulfilled the criteria for inclusion; five were reviews of interventions for a range of caregivers of people with dementia including Alzheimer's disease, one for older people, one for mixed chronic illnesses and five related to stroke survivors. Most reviews employed descriptive and critical analytical qualitative methods. There was one quantitative meta-analysis (Sörensen *et al.* 2002) and one Cochrane Review (Smith *et al.* 2009). Five reviews referred to quality appraisal of the included papers (Pusey & Richards 2001, Peacock & Forbes 2003, Brereton *et al.* 2007, Schoenmakers *et al.* 2010, Cooper *et al.* 2007) and all reviews used different approaches for quality assessment and differed in the amount of detail given (see Table 2).

Table 2: Summary of systematic reviews of caregiver intervention studies

Author, (Year)	Review details	Key findings	R-Amstar rating (additional criteria) & comment
Cooke *et al.* (2001)	**Caregivers:** of people with dementia. **Aim:** To identify the type of components utilised in psychosocial/psycho-educational. Interventions for dementia caregivers and to evaluate the success of the different components or combinations of components in producing positive outcomes for dementia caregivers. **Type of review:** systematic review. **Years:** Varied according to database (up to 2009/10). **Number of studies included:** 40 **Type of studies:** Quantitative evaluation of the effectiveness of a psychosocial intervention with a focus on improving caregiver psychological well-being or social well-being directly. **Method of study appraisal:** Not stated. **Method/type of analysis:** Qualitative, critical appraisal. **Interventions:** Intervention components varied widely. Individual components were rarely used in isolation; combinations of components tended to be employed.	***Caregivers of people with Dementia*** **Education interventions** - Provision of information at one time-point resulted in significant improvement in general well-being. - ↑ Coping ability. - ↑ knowledge of illness and quality of life. ***Information technology*** ↑ Knowledge of illness [telephone lectures > peer telephone networking]. - ↑Assertiveness [cognitive behavioural + phone contact intervention]. - ↑ General well-being [telephone support intervention]. - ↑ Decision confidence [computer link intervention]. ***Training Interventions*** ***Caregivers of people with dementia*** - ↓ Depression, burden and ↑ knowledge of illness. - Improved relationship issues for problem-solving intervention. **Support interventions** - Burden, general well-being, social support (findings not significant). - Carer-relative relations. - Improved anxiety state and knowledge of illness.	23(E^+) Weak No pooled results

		Improved depression. **Combined Education and Support Interventions** - Knowledge of illness. **Psychological intervention** - Significant improvement in depression for group receiving emotional support when compared to group receiving information and controls. - Improved perceived stress, preparedness for caregiving, caregiver competence, coping and general well-being.	
Sörensen *et al.* (2002)	**Caregivers:** Family caregivers of older adults. To determine the effectiveness of interventions for family caregivers of older adults. **Years:** Used ancestry method. **Type of review:** Systematic meta-analysis. **Number of studies included:** 78 **Type of studies:** All tested interventions with an untreated control condition. **Interventions:** Psycho-educational, supportive interventions, psychotherapy, respite/day-care, training of care recipient, multi-component, miscellaneous.	**Multi-component interventions** Effective in improving care-giver burden, well-being, ability/knowledge but not for caregiver depression or care receiver symptoms. **Psychological interventions** - Psychotherapy based interventions were effective for all outcome measures [burden, depression, well-being, ability/knowledge and care recipient symptoms] [n= 10 studies in the review]. - Effective for all outcome measures in comparing different types of interventions using only RCTs except well-being and care receiver symptoms. **Training interventions** - Improved subjective well-being and care receiver symptoms, not significant for carer burden, depression, ability/knowledge.	26(C^+) Moderate

Peacock & Forbes (2003)	**Caregivers:** Of elderly persons with dementia living in the community. **Aim:** To determine the effectiveness of a range of interventions to enhance the well-being of caregivers of elderly persons with dementia living in the community. **Type of review:** Systematic [1992- April 2002]. **Number of studies included**: 11 (36 but only 11 strong ones discussed). **Type of studies:** RCTs **Method of study appraisal:** Used a relevance and validity tool developed for this review based on that of previous researchers. **Method/type of analysis:** Qualitative, critical analysis. **Interventions:** Education (either in group setting or individually) (n=4); case management (n=4); psychotherapy (n=2); computer networking (n=1).	***Caregivers of people with dementia*** **<u>Education Interventions</u>** - Educational interventions were insufficient to improve overall caregiver psychological well-being, albeit the reviewers do highlight a few exceptions [(n=4 of the studies in the review]. **<u>Psychological Interventions</u>** - Psychotherapy based interventions resulted in a delay in institutionalisation of the care recipient although the findings differ in terms of age [older care recipient in one of the studies and younger carer greater likelihood; age was not a significant factor in the other study] and gender [one of the studies reported that female carers were more likely to institutionalise spouses]. Illness severity was also reported as a predictor of institutionalisation. **<u>Information technology</u>** - Increased confidence in decision making skills for caregivers [n=1 study] in a review reporting on computer networking, but no significant outcomes were reported in terms of decision-making skills, social isolation and use of health services.	28 (A^-) Strong No pooled results
Pusey & Richards (2001)	**Caregivers:** Of older people with dementia **Aim:** To assess the evidence of effectiveness of psychosocial interventions with caregivers of people with dementia. **Number of studies included:** 30 **Type of review:** Systematic[varied according to database-1998/99]. **Type of studies:** RCTS OR controlled trials without randomisation.	***Caregivers of people with dementia*** ***<u>Information Technology Interventions</u>*** - Significant improvement in outcomes [areas of confidence and decision making n=1 study] and subjective measures of social support and knowledge (n=1 study). No evidence was provided by either study on specific mental health of carers.	26(C^+) Moderate No pooled results

	Method of study appraisal: Ranked according to methodological hierarchy (no details of the criteria used given). **Method/type of analysis**: Qualitative narrative critical synthesis. **Interventions:** Technology based (n=2) (computer networking providing carers with education, decision support and communication and a network of telephone support); group based interventions (n=14); individually based interventions (n=9); service configuration (n=5).		
Visser-Meily *et al.* (2005)	**Caregivers:** Of stroke survivors. **Aim:** To evaluate the effectiveness of different types of intervention programs for caregivers of stroke patients. **Years:** Varied with database – 2003 **Type of review:** Systematic **Number of studies included:** 22 **Type of studies:** Intervention studies (RCTs (n=18), uncontrolled trials (n=2), non-randomised comparative studies (n=2). **Method of study appraisal:** Not stated **Method/type of analysis:** Qualitative critical appraisal. **Interventions:** Provision of specialist services (n=12), psycho-educational (n=6), counselling (n=4) and social support by peers (n=1).	***Caregivers of people with stroke*** **<u>Psychological Interventions</u>** - 4 studies [psycho-educational interventions] reported a positive effect (3 reported increased knowledge, one increased knowledge but decreased social functioning, one improved mental health. For counselling interventions: 3 reported a positive effect [in one of the studies a positive effect was reported during but not post the intervention]. - Increased knowledge (3/4 studies), increased knowledge but decreased social functioning (1/4 studies) and improved mental health (1/4 studies).	32 (D^+) Weak No pooled results

Brereton *et al.* (2007)	**Caregivers:** Adult family caregivers of people with stroke. **Aim:** To determine which interventions specifically designed for carers of people with stroke are effective in improving carer outcomes. **Type of review:** Systematic **Years:** Varied according to database (up to 2005/06). **Number of studies included:** 8 **Type of studies:** RCTs **Method of study appraisal:** Critical Appraisal Skills Programme (CAPS) tool for RCTs. **Method/type of analysis:** Qualitative, critical appraisal. **Interventions:** Caregiver training (n=1), home and telephone social problem-solving partnership (SPSP/SPTP) (n=2, same intervention), a psycho-educational telephone support group (n=1), a nurse – led support and education programme (n=1), a support programme (delivered by nurses to a groups at hospital or individuals at home) (n=2 same intervention), a combination of education and counselling (n=1).	***Caregivers of people with stroke*** **Psychological Interventions** - Combined education and counselling interventions [one of eight studies included in the review] were found to improve knowledge and result in significantly less deterioration in family functioning than either education alone or routine care. **Training Interventions** - Improved caregiver well-being and quality of life (QoL) but the improvement followed reduction in caregiver depression, anxiety and burden n=1 study. **Combined Education and Support Interventions** - Confidence in knowledge of patient care and seeking social support. - An intervention comprising support groups and home visits led by health education nurses to improve well-being and coping skills in terms of self-efficacy were not found to be effective for stroke caregivers. ***Information Technology Interventions*** - Combined home and telephone social problems solving partnership was found to significantly improve caregivers' health, problem-solving and preparedness to care (n= 1 study in the review). -No significant effects were found for caregiver well-being and competence following a psycho-educational telephone support group intervention (n= 1 study in the review).	30 (A⁻) Strong No pooled results

Cooper *et al.* (2007)	**Caregivers:** Of people with dementia. **Aim:** To synthesise evidence regarding interventions that reduce anxiety in caregivers. **Type of review:** Systematic **Years:** Search done in June 2005 (between dates not stated) **Number of studies:** 24 **Type of studies:** Primary research reporting effects of an intervention on anxiety using validated outcome measures. **Method of study appraisal:** Studies were assigned levels of evidence according to the Centre for evidence based medicine guidelines (CEBM). **Method/type of analysis:** Critical appraisal **Interventions:** Level 2 rated intervention studies: Cognitive Behavioural Therapy (CBT) (n=3); Behaviour Management Therapy (BMT) (n=3); Provision of IT support for caregivers (n=1); exercise therapy (n=2); providing additional professional support for CGs (n=1); Respite care (n=1); relaxation/yoga (n=2). Level 4 rated intervention studies (n=10): psycho-education and CBT and resource information (n=1); psychotherapy groups (n=10); 5 x 90 minute weekly groups including psycho-education, relaxation training and discussion sessions (n=1); weekly for 6 weeks session manualised yoga meditation including discussion about application (n=1); home based intervention for 6-12 hours over 4-14 weeks (n=1); treatment in memory clinic, CMHT or day hospital (n=1); volunteer befriending & in-home respite (n=1); care home (n=1); care home admission within the	***Caregivers of people with dementia*** **<u>Psychological Interventions</u>** - One study reported that combined cognitive behavioural therapy and relaxation-based intervention specifically devised to treat anxiety provided significant effects. **Relaxation Techniques** - Combined cognitive behavioural therapy and relaxation-based intervention specifically devised to treat anxiety, significantly reduced anxiety. - One study [testing relaxation techniques – no control group] and another one [yoga – no control group] provides preliminary evidence to suggest that these two techniques are effective without cognitive behavioural therapy. - Review authors caution that there are no RCT studies to confirm this.	23 (D^-) Weak No pooled results

	past year (n=1); CR placed in fulltime care during 18 months follow-up period for an RCT (n=1). Level 5: coping skills, meditation, assertiveness and cognitive appraisal (n=1).		
Lee *et al.* (2007)	**Caregivers:** Of elderly stroke patients **Aim:** To examine the effectiveness of the interventions for improving mental health of caregivers of people with stroke by synthesising individual studies. **Type of review:** Systematic **Years:** Database inception -2005 **Number of studies:** 4 trials **Type of studies:** RCTs **Method of study appraisal:** Used items selected from a published quality measure with a total score range from 0-16. **Method/type of analysis:** Meta-analysis **Interventions:** Support and education interventions which varied as follows: one education program, two support programs, one in-home meeting with follow up telephone sessions.	**QoL** Significant difference in favour of interventions across all 4 trials (p<0.01) 95% CI 0.118 TO 0.435). Education programs: Significant positive effect t of interventions (p<0.01) 95% CI 0.087, 0.621, 2 trials. Support programs: Significant positive effect t of interventions (p =0.02) 95% CI 'k 0.037, 0.432, 2 trials. Atheroetical interventions were found to be more effective than theory based intervention studies: (p<0.01) 95% CI - 0.098, 0.477, 2 trials.	30 (A) Strong

Eldred & Sykes (2008)	**Caregivers:** Informal adult carers (age 16 years and above) of a survivor of stroke living in the community where the interventions provided to the carer or to the patient and their carer. **Aim:** To analyse interventions that target psychosocial functioning in carers of stroke survivors to understand how such interventions can reduce the burden of caring. **Type of review:** Systematic **Years:** Not provided **Number of studies:** 7 **Type of studies:** RCTs **Method of study appraisal:** Original quality assessment tool (not validated). **Method/type of analysis:** Narrative synthesis of results. **Interventions:** Psychosocial interventions that provide counselling, support or training including education packages along with counselling based on different models, telephone based interventions, group and individual face-to-face/in-home (the interventions were based on different models such as family systems theory, cognitive behavioural therapy, social problem-solving theory and stress and coping model).	**QoL** SF 36 Mental: SMD: 0.307, 95% CI: -0.194 to 0.808, p=0.230) (62 participants, 1 trial). SF 36 Physical: SMD: 0.093, 95% CI: -0.405 to 0.591, p=0.714) (62 participants, 1 trial). **Caregiver competence** SMD: 0.326, 95% CI: -0.095 to 0.747, p=0.129) (86 participants, 1 trial).	26 (A^-) No pooled results Strong
Glasdam *et al.* (2010)	**Caregivers:** Of chronically ill patients (stroke, diabetes, cardiovascular disease and cancer). **Aim:** To describe and critically evaluate clinical interventions targeting families of chronically ill patients. **Type of review:** Systematic	**Findings are not Intervention specific** - 10 studies showed no effect of intervention (9 were for stroke caregivers).	24 (C^+) Moderate No pooled results

	Years: 1997-2007 **Number of studies included**: 32 (stroke n=19; CVD n=3; cancer n=10 and diabetes n=0) **Type of studies:** All controlled studies, mostly RCTs but 3 non-randomised studies included. **Method of study appraisal**: Not stated **Method/type of analysis:** Qualitative/critical appraisal. **Sample sizes:** ranged from n=30- n=520 caregivers. **Interventions:** Home care, education counselling (individual, couples and group), some compare different forms of interventions.		
Schoenmakers *et al.* (2010)	**Caregivers:** Family caregivers of people with dementia **Aim:** To provide a quantitative analysis of the effect of the different types of professional dementia home care interventions. **Number of studies:** 28 **Type of review:** Meta-analytic review-1980 -2007 **Type of studies:** Quasi-experimental or RCTS in dementia home care with intervention and control groups. **Method of study appraisal:** Delphi Criteria **Method/type of analysis:** Quantitative, using meta-analysis. **Interventions:** Cognitive behavioural family therapy; group training (psychosocial interventions); respite care; telephone or internet based support; case management; physical exercise; communication	***Caregivers of people with dementia*** **Psychological Interventions** - In a meta-analysis psychosocial interventions resulted in ↓ burden [6 studies included in the review] and depression [15 studies included in the review] but the findings were not statistically significant (effect sizes 2.94, 95% CI -6.28-0.40 and 0.03, 95% CI -0.2-0.35 respectively). ***Information Technology Interventions*** - Telephone support produced a slight but non-significant increase in depression (effect size 0.07, 95%CI – 2.62-2.75).	31(B) Moderate

	skills.		
Smith *et al.* (2008)	**Caregivers:** Of patients with a clinical diagnosis of stroke or TIA. **Aim:** Assess the effectiveness of information provision strategies in improving the outcome for stroke patients and/or their identified caregivers **Type of review:** Systematic **Years:** varies according to database **Number of studies:** 17 **Type of studies:** RCTs **Method of study appraisal:** Unnamed rating scale ranging from 0-16 **Method/type of analysis:** Meta-analysis **Interventions:** Interventions that provided information with the intention of improving the outcome of patients or their caregivers or both. Education and counselling (1 trial), Information provision plus counselling (n=1).	***QoL*** One trial reported significant outcome in favour of the intervention group (IQR at 1 year: intervention 80 (70-90), control 70 (60-80), $p<0.0001$, 268 participants, one trial). Two trials did not find any difference between the intervention and control for active information interventions and one for passive information interventions. **Depression** Passive information giving (Not significant for one trial) Active information giving significant result reported [(IQR): intervention 2 (1 to 3), control 3 (2 to 5); $p<0.0001$. (300 participants, one trial)]. **Burden** Active information intervention: Median score at 12 months (IQR): intervention 32 (27-41), control 41 (36 to 50); $p=0.0001$. (268 participants, one trial). ***Caregiver competence*** Information giving (active and passive interventions pooled) SMD: 0.74, 95% CI 0.06 to 1.43, $p=0.03$ in favour of the intervention (336 participants, 4 trials). **Stress** OR 1.13, 95% CI 0.65 to 1.97, $p=0.65$ (498 participants, 4 trials). **Social activities/functioning** Active information studies: No significant differences reported (339 participants, 2 trials).	41 (A) Strong

Mean Difference (MD); SMD (Standardised mean difference); RCT (Randomised Controlled Trial); RR (Risk ratio); OR (Odds Ratio)

A wide range of study designs were included in the reviews, with three limited to randomised trials. Most included a combination of study designs including those without control groups (Table 2). R-AMSTAR ratings ranged from 23-41 and the additional criteria developed for this review enabled the categorisation of the reviews according to the included designs as strong (n=5), moderate (n=4) and weak (n=3) (see Table 2).

Due to the quality of the studies included in the reviews and the range of study designs, most of the reviews used qualitative analyses rather than meta-syntheses. Even in reviews that employ a meta-analysis, the findings should be interpreted with caution when designs other than the randomised trials are employed. The randomised trial is the design which produces the strongest form of evidence on the effects of interventions (Akonbeng 2005) and failure to retain positive findings when designs of weaker quality are removed from the analysis is an important factor when interpreting a review's findings. Additionally, many of the reported positive findings from the quantitative analyses across the reviews need to be considered with caution due to the use of study designs without control groups. The reported changes from such studies may have been due to temporal or testing effects, or regression to the mean (Gerrish & Lacey 2010), rather than being due to the intervention itself. Consequently, in the absence of high quality systematic reviews with meta-analysis and synthesis, the findings from all of the sourced systematic reviews were included, as they provided information to help focus the approach to developing the intervention for this study.

3.6.1.1 Findings from the review of reviews of interventions for caregivers and their contribution to the development of the intervention for this study

A wide range of interventions were included within and across the reviews (Table 2). These were categorised in terms of education, training, combined education and support, psychological [included psycho-educational/psycho-social and combined education and counselling interventions], relaxation techniques, information technology [Information technology interventions included telephone, computer based interventions] and multi-component interventions. These categories were not always mutually exclusive and a number of interventions had components from more than one category. In general, the different categories of interventions were linked to specific outcomes, although Cooke *et al.* (2001) did not link the outcomes to specific interventions.

This review of reviews of interventions for caregivers highlighted that studies have tested a range of interventions across an array of outcomes to identify helpful ways of supporting family members/caregivers of people with chronic conditions. Although positive outcomes were reported for all intervention categories, in some instances the findings did not reach statistical significance [see for example, information technology interventions (Brereton *et al.* 2007) and psychological interventions (Schoenmakers *et al.* 2010)].

Interventions using the telephone as a mode of delivery were of particular relevance because this is the mode of communication used in practice by NSsMS in Ireland. In addition, potential recipients of the intervention for this current study were regional and nationally based, which meant that a face-to-face intervention was unlikely to be feasible. It was therefore encouraging to note the consistent positive outcomes for interventions based on information technology across the reviews, which suggested that this mode of delivering interventions to support caregivers had potential (Cooke *et al.* 2001, Pusey & Richards 2001, Peacock & Forbes 2003, Brereton *et al.* 2007).

Although the nature of the telephone support interventions differed across the reviews, the consistent findings from two reviews [strong (Brereton *et al.* 2007) and weak (Cooke *et al.* 2001) suggested that using the telephone to deliver an intervention had potential for improving caregiver outcomes. In addition, the reported improvement in outcomes of caregiver knowledge of illness, assertiveness, general well-being (Cooke

et al. 2001), and health, problem-solving and preparedness to care (Brereton *et al.* 2007), were of interest because these outcomes are within the scope of the role of the NSMS.

3.6.1.2 Individual and group based interventions for caregivers of people with chronic illness

Positive findings were reportedly associated with individual (Sörensen *et al.* 2002) and group-based interventions (Brereton *et al.* 2007). The strength of the evidence varied across reviews. Sörensen *et al.* (2002) reported that individual or combined individual and group interventions were more effective than group interventions only. Conversely, Pusey & Richards (2001) did not find strong evidence in their review to support the use of individual-based interventions and argued that many of the included studies had methodological weaknesses. However, they did assert that despite methodological weaknesses, there was weak evidence to support the value of group-based interventions. Further support for group-based interventions is provided by Brereton *et al.* (2007) who suggested that group-based interventions are effective in improving coping skills for caregivers of people with stroke. Likewise, Cooper *et al.* (2007) asserted that there is preliminary evidence that group-based interventions involving yoga and relaxation techniques can be potentially beneficial.

Despite the positive findings for group-based interventions, it is not possible to determine if they are more effective than those offered to people on an individual basis. This may, as suggested by Pusey & Richards (2001), be due to methodological weaknesses in the included studies. It may also be explained by the ad hoc approach to testing the impact of individual or group-based interventions within studies. None of the reviews reported that the included studies set out to test its effect.

3.6.1.3 Application of the findings of this review of systematic reviews of interventions for caregivers to the development of the intervention for this study

This review of systematic reviews of interventions for caregivers of people with chronic illness highligted a number of considerations for the content when developing the

current intervention. These included the use of information technology, multi-component interventions, psychological interventions, support and education or training. However, some of these (such as psychological interventions) were outside the scope of the role of the NSMS and it was important to be cognisant of what was feasible and likely to be acceptable for implementation in practice by NSsMS. As funding for the study was not available at the time of intervention development, the cost of set-up and delivery of the intervention was another important consideration. PwMS were regionally and nationally based and it was not feasible to have nominated support people travel long distances for a face-to-face intervention. Furthermore, the extent to which they had access to or familiarity with computers was unknown. It was clear that the telephone was the mode of communication used by the NSsMS to make contact with PwMS and their family members or caregivers. Therefore, the telephone was identified as the most likely mode of delivery for the intervention.

Two key documents were found which provide an overview of the contribution of telephone care in the area of chronic illness, one American based (Piette 2005) and one from the United Kingdom (Royal College of Nursing (RCN) (RCN 2006). Piette (2005) purport that telephone care can help both patients and carers by addressing some of the challenges to effective care management, as it allows healthcare workers to communicate with patients in a timely manner without the need for face-to-face contact. However, the author (Piette 2005) argues that such services must be carefully organised in order to avoid them becoming costly add-ons that deliver no true benefits. In their report, Piette (2005) provides an overview of the benefits of telephone care services primarily from patients' perspectives for a number of chronic conditions such diabetes, heart failure and chronic pain. They argue that carefully structured telephone support that is linked to usual outpatient care can significantly contribute to better outcomes (Piette 2005). However, they also report that findings based on a Cochrane review (Currell *et al.* 2000) provides mixed evidence to support the use of telephone support, due to the "...low quality of most telephone care..." (Piette 2005, p. 11). The RCN (2006) primarily provides guidance on setting up telephone support services. Important advice on programme evaluation (Piette 2005) and guidance on setting up a service (RCN 2006) was integrated into the manual for this intervention.

The importance of telephone support, which is linked to clinically based care and offered by clinically based nurses or other health staff, is highlighted by Piette (2005). Although Piette (2005) was referring to telephone support for patients, the principle has

application for family members and carers of patients. Clinically based NSsMS are in a unique position to provide an individualised professional telephone support service for family members/carers of PwMS. They have legitimate access to case notes, are familiar with the care recipient's unique circumstances and stage of illness, may have prior contact with the caller in outpatient's department and are professionally educated and registered health care providers, with specialised expertise in providing care for PwMS and supporting their family members and carers. The provision of telephone support was therefore clearly within the scope of the role of the nurse and in particular the NSMS. In order to further inform the development of the intervention for this study, a comprehensive search of the literature was undertaken on caregiver telephone support interventions.

3.6.2 Caregiver telephone support interventions: a review

This literature review sought to: a) identify how caregiver telephone support services are delivered and b) determine the overall outcomes from telephone support interventions. A flow chart outlining the literature search and selection of telephone intervention studies is presented in Figure 6.

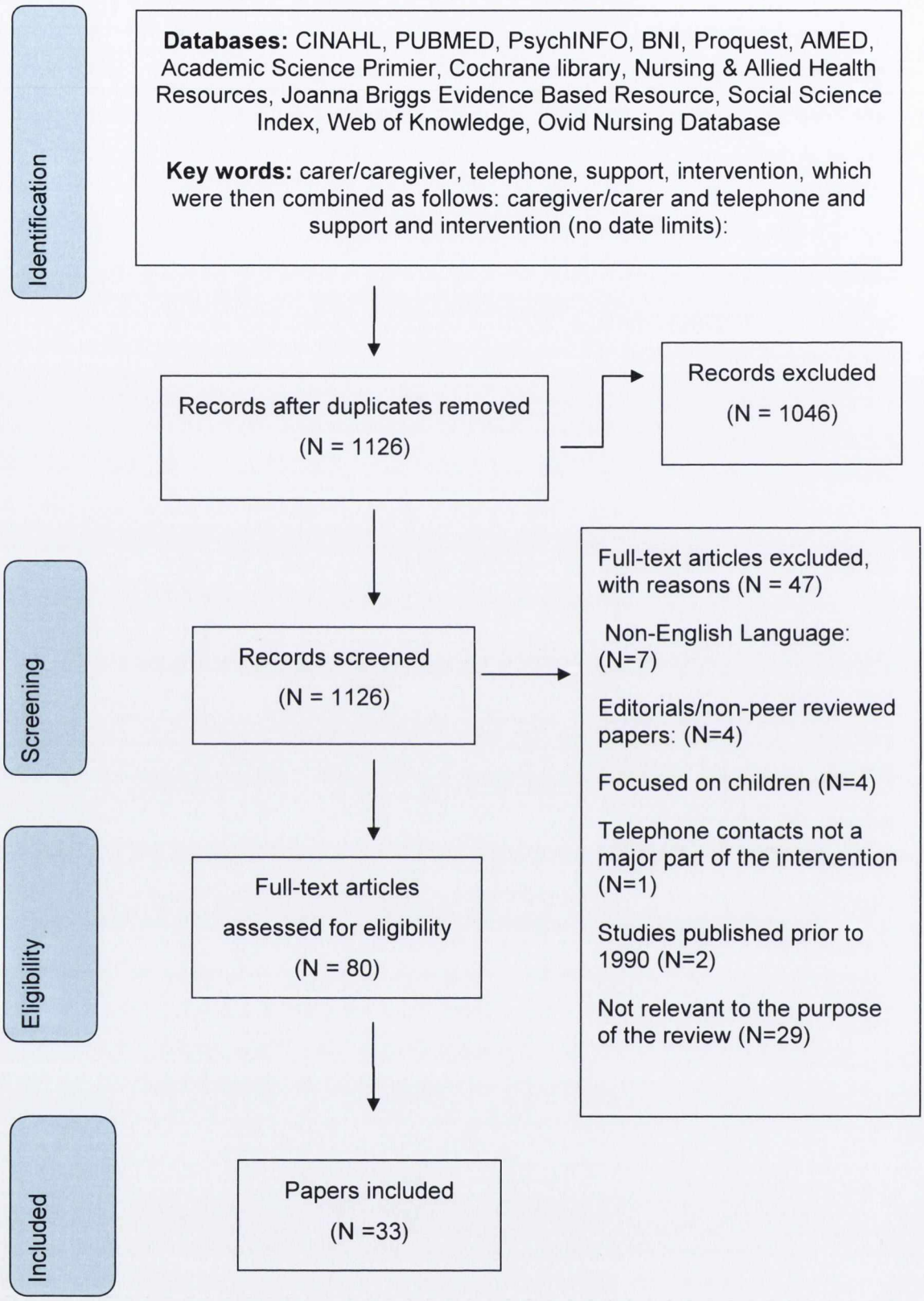


Figure 6: Flow chart for literature search and study selection for the literature on caregiver telephone interventions

The papers were categorised into those using a randomised trial (n=8) (Table 3), those reporting on non-randomised interventions studies (n=7) (Table 4) and those reporting on studies in progress/ feasibility aspects of an intervention/ qualitative arm of an intervention study (Appendix 3).

3.6.2.1 Methods used for quality appraisal and synthesis

The CONSORT 2010 checklist of information to include when reporting a randomised trial (www.consort-statement.org) was used to inform my appraisal of the reports of the randomised trials. Other intervention studies (Table 4) were appraised using a modified version of the Critical Appraisal Skills Programme (CASP) (Appendix 6). Scores ≥17 were categorised as high, ≥14 medium, ≥12 low, and ≤11 very low quality.

Intervention intensity is conceptualised in terms of length of the intervention, method of delivery, variety of practitioners and/or number of people receiving the intervention at one time [this was in reference to cognitive behavioural therapy] (Bennett-Levy *et al.* 2010). In the absence of a more general explanation of the concept, this understanding was used to inform the categorisation of the interventions in this review with the intervention intensity categorised as high, medium and low according to the frequency and duration of the intervention. Weekly interventions lasting 12 weeks or more or bi-weekly interventions lasting 24 weeks or more were classified as high intensity interventions, those delivered weekly for 8 -12 weeks or bi-weekly for 12-24 weeks were categorised as medium intensity. Weekly interventions of less than 8 weeks duration or bi-weekly of less than 12 weeks duration, were categorised as low intensity.

3.6.2.2 Findings of the telephone review and their contribution to the development of the intervention for this study

The papers reported on studies which included caregivers of patients with specific illnesses such as dementia, older people or mixed groups of caregivers (Table 3, 4 and Appendix 3). A number of aspects of telephone support were reported upon, which included demonstrating the usefulness of technology (Bank *et al.* 2006), implementing telephone support (Shanley *et al.* 2004), evaluating the impact of telephone support

service (Goodman & Pynoos 1990, Brown *et al.* 1999), determining helpfulness (Chang *et al.* 2004), feasibility of an intervention (Smith *et al.* 2004, Herman *et al.* 2006), exploring or describing telephone support (Smith *et al.* 2004), examining the pros and cons of using telephone support (Goelitz 2003), investigating the use of tele-rehabilitation (Hauber & Jones 2002) and describing and exploring caregiver concerns (Walsh *et al.* 2004). Others reported on experiences of implementing (Shanley 2008) or skills needed (Toseland *et al.* 2007, Shanley 2008) for implementing the services. Despite the descriptive nature of some of these studies, they provided valuable insight into the introduction and benefits of such services to carers. Several studies introduced an intervention with the intent of enabling the caregiver through education, empowerment or behaviour change (Strawn & Hester 1998, Mahoney *et al.* 2003, Smith & *Toseland* 2006, Glueckauf *et al.* 2007, Rivera *et al.* 2008, Tremont *et al.* 2008, Bormann *et al.* 2009, Connell & Janevic 2009).

Table 3: Papers using RCT designs to test a telephone support intervention

Author, date, country & study design	Population Intervention (I) and control (c) group	Type of intervention (single/combined; proactive/reactive; tailored/fixed; interventionist/caller initiated)	Duration & frequency (Intensity & group/individual)	Model used & Interventionist(s)	Outcomes	Comment
Bakas *et al.* (2009a) Indianapolis Pilot RCT	Caregivers of people with stroke I: N (26) TASK C:N (24) attention control group (brochure on family caregiving and 8 weekly calls from a nurse)	Single intervention: Task is completely delivered by telephone and is initiated within one month after the stroke survivor is discharged home. TASK intervention (to develop skills based on own assessment of their own needs in 4 main areas. In addition it was designed to increase caregiver optimism and reduce threat of appraisal by helping cares deal with own personal responses to providing care	Medium : eight sessions weekly Length of calls: Not stated; gives an overall mean contact time in minutes with the nurse for the TASK group Number in group: N/A (one-to-one intervention).	Model derived from Lazarus's transactional theory of stress Nurse (no further details given) Training: Using standardised protocols and treatment manuals for both intervention and control groups. Nurses had to 'pass' role-playing sessions using structured check lists. Individualised. Calls were recorded and individualised re-training was provided based on evaluations	Greater satisfaction with TASK group compared to the attention control group	Attrition rate not reported Intervention content validity reported separately in (Bakas *et al.* 2009b)

Elliott & Berry (2009) US RCT	Caregivers of people with spinal cord injuries I: N (30) C:N (30)	Brief Problem-Solving Training for Family Caregivers of Persons with Recent Onset of Spinal Cord Injuries	Low: 12 months (three times at 6 monthly intervals first session 2-3 hours in a face-to-face session, then at 6 and 12 month sessions).	Not explicitly stated. Appears to be based on problem-solving as in project FOCUS(Facts, Optimism, Creativity, understanding and the steps involved in solving a problem) Not clear who the interventionists were. It is stated that two project co-coordinators had doctorates in clinical psychology and received training in PST.	A statistical significant decrease in dysfunctional problem-solving over time for carers who received PST and an increase among carers in the control condition. SF-36 health-related subscales (physical and social functioning):interaction effects that approached statistical significance Both groups had a decline in physical functioning over time but the rate of decline was much steeper for carers assigned to the control group. Social functioning increased for carers who received PST and declined among carers assigned to the control group.	8/30 discontinued the intervention 10/30 discontinued from the control group

Glueckauf *et al.* (2007) US 2-group RCT	Rural caregivers of people with dementia I: N (12) C:N (8)	Telephone Based Cognitive Behavioural Intervention for distressed Rural Dementia Components of the intervention: overview of 6 basic characteristics of dementia; relaxation training; effective thinking about caregiving challenges; building in pleasant daily activities as a guard against emotional distress; assertiveness in caregiving situations and with family members; developing problem-solving skills through personal goal setting; Caregivers met online for the group sessions and received calls for the individual sessions.	High: weekly for 12 weeks (group sessions week 1-6 and week 12; week 7-11, 5 individual telephone sessions). Length of sessions: group sessions (45 minutes); individual (1 hour). Number in group: 3	Not stated but is most likely to be based on cognitive behavioural therapy. Psychologist or counsellors Trained doctoral or master's level counsellor	Partial support was obtained for the differential benefits of cognitive-behavioural (CB) intervention over routine education (ES) interventions for this population	Findings presented were on a very small sample from the halfway point of an ongoing 3-year RCT. Reported high level of attrition (65%) in the control group. Does not report attrition for intervention group.

Grant *et al.* (2002) US RCT	Family caregivers of stroke survivors N =74 participants in total (not clear how many assigned to each group	Social Problem-solving Intervention Family caregivers were trained to use: social problem-solving skills to manage caregiving problems and cope with the stresses of caregiving. 3-hour face-to-face session with a trained nurse in the home (taught to use a positive problem orientation and 4 systematic steps when solving caregiving problems) This session was followed by weekly (occurring during weeks 2, 3, and 4 after discharge) and biweekly (occurring during weeks 6, 8, 10, and 12 after discharge) telephone contacts to develop and maintain these skills over a 12-week period.	Medium-high: 3-hour face-to-face session followed by weekly (occurring during Weeks 2, 3, and 4 after discharge) and biweekly (occurring during Weeks 6, 8, 10, and 12 after discharge) telephone contacts to develop and maintain these skills over a 12-week period.	D'Zurilla's problem-solving model Trained research nurses (for intervention) Graduate research assistant (sham group)	Intervention group: statistically significant improvements in vitality, mental health, and role limitations related to emotional problems. Control group had ↓ levels of mental health over time. Intervention group reported significantly better problem-solving skills than other 2 groups; also reported significantly less negative orientation and impulsivity/carelessness but more rational problem-solving skills. Satisfaction with healthcare services decreased over time in the control group but remained comparable in the intervention and sham intervention groups.	Data were collected by academic health sciences research fellow who was blinded to the purposes of the study, intervention and group assignments

Hartke & King (2003) US RCT	Older stroke caregivers I: N (43) C:N (45)	Telephone Group Intervention Telephone support group followed a structured psycho-educational format. A group manual that outlined the 8 topics of discussion was mailed to each participant (this included an audiotape of a relaxation procedure and a publication on stress management). Conference call initiated by group facilitator	Low: 8 weeks in duration (Frequency: not stated (possible weekly) Length of sessions: 1 hour Number in group: 3-6 (total of 16 groups)	Stress and coping model Most groups had 2 group facilitators who were clinicians in areas of psychology, social work or nursing.	Intervention group: more distressed at T1 with significant higher levels of depression, burden, loneliness, and stress and lower sense of competence Significant reduction in stress as measured by the PPI over time (change significant between T1 and T2 And T1 and T3). Significant gain in their sense of competence from T1 to T3. Control group: significant greater change in burden between T1 and T3. Significant overall effect on stress over time for the 2 groups but not between group and time. High levels of satisfaction with aspects of the group	Attrition: 29% drop out rate. Significantly more treatment than controls dropped

Mahoney *et al.* (2003) US RCT	Caregivers of people with Alzheimer's disease I: N (49) C:N (51)	Automated Telephone Support System consisting of an integrated telephone network system with an interactive voice response (IVR) computer network system. The technology application used a multisystem telecommunications-based approach to help carers manage AD related to disturbing behaviours that are commonly associated with carer stress. The IVR-mediated system provided carer stress monitoring and counselling information, personal voice-mail linkage to AD experts, a voice mail telephone support group and a distraction call for care recipients.	Duration: one year. Frequency: use of the system was on demand; also received weekly caregiver conversation Length of calls: N/A could use the system on demand Group size: N/A appears to be a one-to-one system	Pearlin's model of AD caregiver's stress (Pearlin *et al.* 1990) Research assistant was a certified REACH project interviewer otherwise not addressed.	Significant intervention effect for those with low mastery at baseline on all 3 outcomes: bother (p=0.04), anxiety (p=0.01) and depression (p=0.007). Wives with low mastery and high anxiety benefited most. Wives exhibited a significant intervention effect in the reduction of the bothersome nature of caregiving (p=0.02).	Did not report numbers lost to follow up but appears to have lost at least 10 from the intervention group and 8 from the control group by the 19 month follow up time point.

Rivera *et al.* (2008) US RCT	Caregivers of persons with traumatic brain injuries (TBIs) I: N (33) C:N (34)	Problem-solving training program or telephone follow-up programme. All received brief telephone calls on a monthly basis and project staff was available to them on a toll-free telephone number. Problem-solving training group received a combined in-home and telephone sessions	Low: Telephone sessions for 8 months of the study at monthly intervals) Length of calls: Intervention group: not stated; control group :10-15 minutes Number in groups: N/A one-to one	no specific theory identified Doctoral degree in administration with experience in volunteer work. Training: was provided by first author in individual sessions that included instruction in problem-solving model and PST group protocol. Also supervised 2 home visits. Total training 30 hours	Significant decreases in depression, health complaints and in dysfunctional problem-solving styles over time. No effects were observed on caregiver well-being, burden or constructive problem-solving styles	No loss to follow up. Intervention: One did not receive the intervention as he/she withdrew Control group: 6 withdrew before receiving allocated sequence

Tremont *et al.* (2008) US RCT	Caregivers of people with Dementia I: N (16) C:N (17)	The Family Intervention: Telephone Tracking-Dementia (FITT-D) Model The three underlying theories were geared towards enhancing coping with the caregiver through active problem-solving and facilitating positive changes within the family system (Original intervention modified by increasing the duration from 6 to 12 months along with changes to the key areas to focus on). Control group: standard care (no detail provided)	High: 1 year (weekly for 6 weeks; every 2 weeks for the next 12 calls; monthly for the next 4 termination calls. Length of calls: initial contact approx 60 minutes; follow up 15-30 minutes. Number in groups: N/A one-to one.	Theories based on psychosocial transition, transactional stress and coping and a systems view of family functioning Non-specified master's level prepared therapist	Lower burden scores for those receiving the FITT-D intervention	FITT model was originally tested and demonstrated efficacy for stroke caregivers. Attrition: 27/60 (45%)

RCT:Randomised controlled trial; I:Intervention group, C: Control group

Table 4: Papers reporting on telephone intervention studies (not using RCT design)

Author, date, country & study design	Population Intervention (I) and control (c) group	Type of intervention (single/combined; proactive/reactive; tailored/fixed; interventionist/caller initiated)	Duration & frequency (Intensity & group/individual)	Model used & Interventionist	Outcomes	Quality rating (CASP score /30) & Comment
Bank *et al.* (2006) US Intervention study using post-test design	Caregiver s of people with dementia I: (N=41) SET + CTIS C: N/A	Combined Intervention (SET + CTIS) SET: family based psychotherapy intervention CTIS system an information network that relies on computer-telephone technology Who initiated contact: Carer logged into the service	Low-high: Bi-monthly at beginning and then monthly for 18 months (number of contacts: ranged from 1-23 (depended on the carer) Length of calls: approx. One hour Carers took part in 1 of 6 concurrently run groups (group size: maximum 6 in addition to the group leader	No model identified for the telephone support groups. In-home sessions appear to be based on a model of family therapy (none specified) Sessions facilitated by study certified therapists (who were not providing the family therapy intervention who held at least a Masters degree in psychology or related discipline	91% of caregivers benefited because of the greater social and emotional support they received as well as the information they obtained from other group members Majority reported that their participation had increased their knowledge and skills as caregivers.	CASP =15 (medium) Evaluates the usefulness of the technology for conducting telephone-based support groups in an ethnically diverse sample of dementia caregivers. 41/51 caregivers completed the study

Bormann *et al.* (2009). US Exploratory using a prospective, mixed-methods, 3-time repeated measures design.	Caregivers of people with Dementia I: N (16) C:N/A	Teleconference-delivered with face to face meetings in week 1 and 8, teleconference calls in weeks 2 through 7. Individual calls week 10, 12 and 14 to assess adherence and encourage practice. Face-to-face meeting: Participants given course materials, "the mantra handbook", reading assignments and phone instructions for weekly conference calls were provided. 8-week program of mantra repetition with 3 additional cognitive behavioural skills	Low to medium: 14 weeks (calls weekly for 6 weeks and further individual calls week 10,12 and 14). Length of calls: Not stated Number in group: tele-conference calls in groups of 3-5	Based on concept of spirituality Program was co-facilitated by a geriatric nurse practitioner and licensed clinical social worker.	Carer burden scores high at baseline (m: 23.2, SD=9.85) ↓ significantly showing linear trend at weeks 8 (m=17.94, SD=7.74) and 16 (m=16.75, SD=7.66) Significant effect for time and linear terms for perceived stress, depression and rumination (not in trait-anger) Significant effect for time and linear terms for quality of life but not for mindfulness attention awareness Satisfaction: majority satisfied.	CASP =15 (medium) Original intervention delivered as an 8 week (60 minute/week) group meeting. 16/21 (76%) completed the study.

Brown *et al.* (1999) Canada Quasi-experiment	Family caregivers of adult persons with caregivers I: N (52) C:N (39)	Distance education and caregiver support groups: comparison of Traditional and Telephone Groups. Group counselling and education was supplemented by telephone and newly produced videos and discussion guides Group members selected topics for discussion Groups were conducted via conference call technology	Medium: weekly for 9 or 10 weeks. Length of calls: between 1.5 and 2 hours. Number in group: 4-11.	None stated Groups were led by a psychologist, a neuro-psychologist, or social worker from within the brain injury program.	Similar amounts of improvements for outcomes for onsite group (OG) and telephone support group (TSG). Rural carers had fewer difficulties on all measures at all measurement intervals Both groups: a statistically significant improvement in POMS scores and a trend toward improvement in FAD and CBI results). Both groups: rated experiences highly (rural carers were generally more satisfied)	CASP =16 (medium) Held in the evening, once per week 18 from onsite group 22 from TSG

Connell & Janevic (2009) US Intervention study (group randomisation	Caregivers of people with Dementia I: N (86) C: N (71)	Problem-solving During calls participants set various types of goals. A problem-solving process was used to address any barriers to goal attainment	Medium: 6 months (14 calls in total, weekly for 2 months; biweekly for 2 months; monthly for 2 months Length of contact: Not stated) Number in group: N/A (one-to-one intervention)	Social cognitive theory (SCT; (Bandura 1986) and motivational interviewing (Miller 1996). Trained behaviour change counsellors Training: Trained to help participants problem solve in event that participant had difficulty finding the time to exercise. Day-long training session to address program fidelity that included opportunities for role-playing and performance feedback to promote appropriate an accurate delivery of program.	Increases in physical activity	CASP =18 (High) Exercise intervention Study attrition: 15.9% (n=26) was lost to follow up.

Davis *et al.* (2004) US 3 group repeated measures design (groups were randomised)	Caregivers of Persons With Dementia Group1 (in-home training: n (24) Group 2 (Telephone: N (23) Group 3 (Friendly calls: N (24)	Socially supportive (friendly) phone call contacts with 3 group (comparison group: in-home contact, telephone contact and friendly calls) Carers in both telephone and in-home training groups participated training sessions that included general problem-solving skills, carer appraisal of behaviour problems). Carers received a standardised loose leaf notebook of suggestions for managing frequently encountered AD situations and were asked to fill out weekly log on problems they had managed over the past week.	High: weekly for 12 weeks Length of contact: averaged 37 minutes for telephone contacts) (initial contact home visit of approx 45 minutes to introduce themselves and familiarise the carer with the use of the notebook and log sheets). Number in group: N/A (one-to-one intervention)	Intervention was grounded in the stress appraisal and coping theory (Lazarus, 1999) 3 staff members (no further details given) Training: Using real and simulated contacts with AD carers and persons with AD, three staff members were trained to 85% agreement in identifying caregiver problems and recommending appropriate home care management strategies. They conducted both in-home training and telephone training sessions.	All 3 groups experienced initial reduction in carer distress, only in-home training group demonstrated a significant reduction (F (2, 41) 3.01, *p*=0.023) Carers with higher burden scores were more likely to remain in the study. More time was spent with the in-home training group, who averaged 65 min per contact (SD = 29 minutes). All groups reported similar levels of satisfaction with the project.	CASP =19 (High) Only 44 (64%) of the original 71 dyads were retained to T3 follow up. Drop outs were spread across the three groups.

Goodman (1990) US 2-group comparison study (both groups received both interventions in sequence)	Caregivers of people with Alzheimer's patients Group I: N (22) Group 2:N (18)	Care-Line: A Model Telephone Information and Support Program for Caregivers of Alzheimer's Patients, a caregivers Phone Network A hybrid program (professional intervention played an important role for information and referral crisis counselling, and coordination as well as program initiation). Two components: 1. Peer networks over the phone 2. An informational lecture series, twelve in total, accessed over the phone that covered AD and care management topics.	High: weekly for 12 weeks Length of sessions: not stated and it's not clear if the peer support call is the same as the weekly call for a brief (15 minute) supportive conversation. Number in group: 4 or 5 carers.	No model identified Carers rotated making the calls No evidence of the presence of a facilitator	Psychological effect showed significant time effect between time 1 and 2 (benefits not continued for second period). Mental Health index tended towards significance. Burden increased from time 2 & 3. Both components of the program increased the social support. Post hoc tests showed gains during first period but for perceived social support this declined during second period Social conflict ↑ between time 2 and 3 Informal supports grew in second period	CASP =18 (High) Of an original 81 subjects in the study 32% (n=13) did not complete round 3 data collection as the study ended, 32% (n=13) dropped out between time 2 and 3.

Goodman & Pynoos (1990) US 2-group comparison study (both groups received both interventions in sequence and using randomisation .	Caregivers of Alzheimer's patients Group I: N (31) Group 2:N (35)	Care-Line: a model telephone program: Two components: 1. Peer telephone networks 2. Telephone accessed informational lectures on Alzheimer's disease.	High: weekly for 12 weeks Length of sessions: not stated and it's not clear if the peer support call is the same as the weekly call for a brief (15 minute) supportive conversation. Number in group: 4 or 5 carers.	No model identified Carers rotated making the calls No evidence of the presence of a facilitator	Significant group effect for perceived social supports for caregiving and satisfaction with support for caregiving (participants on the network component scored higher on this variable regardless of time). For network participants (trend towards greater impairment among relatives of network participants (p=0.076) and greater distress with relatives' problems (p=0.087).	CASP =18 (High) Attrition: 7/38 (18%) dropped out of the original network group. 8/43 (19%) dropped out of the original lecture group. Overall about 18.5% dropped out between first and second interview.

POMS: Profile of Mood States, FAD Family Assessment Devise (FAD), CBI: Caregiver Burden Instrument

3.6.2.3 Method of delivering the telephone interventions

Few (n=5) studies used the telephone as the sole means of support for the caregiver. Several telephone support interventions were hybrid interventions that incorporated other types of education or training, in addition to the telephone contact (Brown *et al.* 1999, Chang *et al.* 2004, Bank *et al.* 2006, Smith & Toseland 2006, Glueckauf *et al.* 2007, Rivera *et al.* 2008, Sepulveda *et al.* 2008, Tremont *et al.* 2008, Bormann *et al.* 2009). Table 5 illustrates the components of hybrid interventions which were additional to the telephone.

Table 5: Components of hybrid interventions additional to the telephone

- Group counseling and videos (Brown *et al.* 1999).
- In-home family therapy sessions (Bank *et al.* 2006).
- Four in-home sessions (Smith & Toseland 2006).
- Twenty minute video tape providing information at start of programme) (Chang *et al.* 2004, Glueckauf *et al.* 2007).
- Five telephone-based individual caregiver goal setting and implementation sessions delivered in the home setting (Rivera *et al.* 2008).
- Workbook to each member in the telephone support group arm of the study (Tremont *et al.* 2008).
- Face to face meetings (Bormann *et al.* 2009).
- Supplemented with newly produced videos and discussion guides (Sepulveda *et al.* 2008).

Overall, there were roughly equal numbers of one-to-one (n=18) and group facilitated (n=14) telephone support services. Both approaches were evaluated using a variety of

designs and there was no conclusive evidence to suggest either approach was superior to the other.

3.6.2.4 Characteristics of the interventionists and fidelity to the intervention

Although most studies identified the interventionist, their training and ongoing performance monitoring was often not reported. Likewise, no rationale was provided for the selection of interventionists. Although several telephone interventions were delivered by nurses (Hauber & Jones 2002, Walsh & Schmidt 2003, Chang *et al.* 2004, Walsh *et al.* 2004, Bakas *et al.* 2009b, Radziewicz *et al.* 2009), many were offered by disciplines other than nursing, particularly disciplines such as psychology and counselling (Strawn & Hester 1998, Glueckauf *et al.* 2007, Connell & Janevic 2009) and social work (Smith *et al.* 2004). Some studies used a mixture of interventionists/facilitators such as nurse and social worker (Bormann *et al.* 2009), social workers, clinical psychologists and nurses (Herman *et al.* 2006), psychologists, neuropsychologist and social worker (Brown *et al.* 1999) or health and community workers (Shanley *et al.* 2004). Others were offered by non-health care professionals (Rivera *et al.* 2008, Tremont *et al.* 2008).

No evidence was found in the studies reviewed to suggest that having interventionists from any particular discipline produce better outcomes. None of the studies isolated the effects of the interventionists. This may be due to the complex nature of the interventions, which makes it difficult to test components in isolation.

A variety of methods were used to monitor the delivery of the intervention to ensure that it was delivered consistently and correctly. These included audio-taping, direct monitoring by project manager, analysis of notes and the use of treatment fidelity checklists.

3.6.2.5 Models/theories used to guide the telephone interventions

Eleven papers included a named theory and provided some detail of the underpinning theory (Grant *et al.* 2002, Walsh & Schmidt 2003, Walsh *et al.* 2004, Herman *et al.* 2006, Tremont *et al.* 2008, Bakas *et al.* 2009a, Radziewicz *et al.* 2009). In general, theoretical models were used when the intervention was designed to modify caregiver behaviour or skill. Such studies focused on areas such as problem-solving (Grant *et al.* 2002, Chang *et al.* 2004, Rivera *et al.* 2008, Connell & Janevic 2009), family therapy (Bank *et al.* 2006), spirituality (Bormann *et al.* 2009) stress, coping and family functioning (Tremont *et al.* 2008), stress [Lazarus's transactional theory of stress (Bakas *et al.* 2009a), cognitive behavioural changes (Glueckauf *et al.* 2007), models of self-help (Hauber & Jones 2002), and bereavement or grief (Walsh & Schmidt 2003, Walsh *et al.* 2004). In some studies there was an indication that the intervention was based on a theory but no specific theory was identified (Brown *et al.* 1999, Grant *et al.* 2002, Chang *et al.* 2004, Bank *et al.* 2006, Dollinger *et al.* 2006, Smith & Toseland 2006, Rivera *et al.* 2008, Bogosian *et al.* 2009, Connell & Janevic 2009). Details of the theory were either not included, [for example, Bank *et al.* (2006) who identified family therapy sessions], or scant details were provided [for example, Brown *et al.* (1999) who mentions that the intervention was to provide education, skill development and counselling]. Others state that the training was based on problem-solving and although they provided some detail on the steps taken, they fail to identify a specific theory on which the intervention was designed and delivered (Chang *et al.* 2004, Rivera *et al.* 2008, Connell & Janevic 2009). Grant *et al.* (2002) identified a problem-solving model, but gave very little detail on the model specifics. Similarly, Smith & Toseland (2006) stated that a multi-component telephone support group intervention was provided for caregivers based on an adapted face-to-face support intervention that included education on illness, emotion coping strategies, problem-solving and support, while the intervention tested by Dollinger *et al.* (2006) was a multi-component intervention based on the Perceived Stress Model of Caregiver Burden.

No differences were noted in terms of outcomes between those studies that used a specific model (n=19) to guide the intervention and those that did not (n=16), with both approaches producing positive outcomes. In this review, none of the interventions

delivered by nurses were framed using theories from a nursing perspective. It is considered good practice to use pre-existing and, where possible, tested models to guide intervention development and delivery (Medical Research Council 2008). This poses a challenge for nurses providing support to family members and caregivers as the majority of nursing models that may be of relevance to guide such interventions are the level of grand theories. In addition, there are few off the shelf models to guide telephone interventions for use within the scope of nursing practice. Mason & Harrison (2008), in a review of telephone interventions for family caregivers of patients with dementia, frame the family telephone support for people with dementia in Watson's theory of human caring. They argue that Watson's theory focuses on the caring processes and provides an overview of the key processes within the model. Although Mason & Harrison (2008) provide a reasonable argument for the use of the model, for example that using this model for telephone support interventions enables such interventions to be theory-based and relevant to caregiver burden, the link created by the authors between Watson's theoretical framework and the concept of burden is weak. Both the framework and concept of burden are presented separately and other than a few examples of how the model applies to telephone support interventions for caregivers of people with dementia, the application of the model to telephone support interventions is limited. In order for potential users to comprehend how the model can be effectively applied, a more detailed explanation of how the model can be used in the design, delivery and evaluation of a service is required. This may have been achieved through the use of a model case or by providing suggestions for application of the model in the design of interventions in order to impact on caregiver burden.

3.6.2.6 Methodological strengths and weaknesses of the telephone studies

All the studies in this review used convenience sampling methods, which is not unusual given the contact details for caregivers are not easily obtained. Caregivers are not required to register their existence and registers are not readily available. Despite this limitation, several studies used a randomised trial design (Grant *et al.* 2002, Hartke & King 2003, Mahoney *et al.* 2003, Glueckauf *et al.* 2007, Rivera *et al.* 2008, Tremont *et al.* 2008, Bakas *et al.* 2009b, Elliott & Berry 2009). The use of this design adds to the

confidence researchers and practitioners can have in the estimates for the potential benefits of telephone support interventions for client groups. The 8 randomised trials in this review were of weak methodological quality when evaluated using the CONSORT 2010 checklist of information to include when reporting a randomised trial (see appendix 7 for the findings of the evaluation based on 7 of the key items on the CONSORT 2010 checklist)

Sequence generation was unclear for all the trials and there was little information on allocation concealment. There was no information on sample size calculations and, in general, sample sizes were small. Although small sample sizes may be acceptable for the pilot randomised trial (Bakas *et al.* 2009a), full scale trials require adequately powered samples to determine efficacy (Torgerson & Torgerson 2008). It is important to consider the potential for bias in the included randomised trials when interpreting the findings and their implications for practice.

Although not presented as randomised trials, other studies also carried out feasibility and pilot work using small sample sizes (Bormann *et al.* 2009, Stewart *et al.* 2001, Walsh & Schmidt 2003). In these studies generalisability of the findings are also limited in terms of sampling methodologies employed. Failure to report evidence of intervention fidelity in a number of studies limit researchers' and practitioners' abilities to interpret the validity of the findings, as outcomes may be due to the intervention or some other extraneous variable.

In addition to considering the study design used, other areas of interest include data collection points and instruments used to collect data. None of the studies provided rationale for data collection points. In general, measurement instruments used were recognised and validated instruments. Along with other outcome measures, a number of studies used recognised measures of burden. For example, the Zarit Caregiver Burden Scale (Smith & Toseland 2006, Tremont *et al.* 2008, Bormann *et al.* 2009), the subjective caregiver burden using a modified version of the Revised Memory and Behaviour Problem Checklist (RMBPC) (Connell & Janevic 2009), the 19-item caregiver appraisal inventory (CAI) (Glueckauf *et al.* 2007), the Caregiver Burden Inventory (CBI) (Brown *et al.* 1999) and the burden Interview (Hauber & Jones 2002). However, other than previously published psychometric properties, little additional

information is given on the validity and reliability of these tools in the context of the studies presented.

3.6.2.7 Overall effectiveness of the telephone interventions

All studies reported at least some positive outcomes, in particular for perceived support, burden, stress, knowledge, satisfaction and depression (see Table 3,4, Appendix 3), but some findings were not statistically significant. For example, although positive changes were reported for a number of variables, Bormann *et al.* (2009) did not report significant changes in caregiver trait-anger and mindfulness attention awareness. Similarly, Glueckauf *et al.* (2007) reported partial support for the outcomes of a telephone-based cognitive behavioural intervention, with significant improvement in caregiving problems from initial goal setting to 1-week post-treatment across all three measures of problem change. Likewise, Rivera *et al.* (2008) found a significant decrease in depression, health complaints and dysfunctional problem-solving styles but no changes in caregiver well-being, burden or problem-solving abilities over time. Some negative outcomes from telephone interventions were also reported (Hartke & King 2003, Chang *et al.* 2004). For example, Chang *et al.* (2004) found that several participants perceived the telephone calls to be lacking in helpfulness, while Hartke & King (2003) reported that the treatment group was more distressed overall at their first assessment, with significantly higher levels of depression, burden, loneliness, and stress and lower sense of competence.

Despite the possible negative effects, these findings provide general support for the potential contribution of telephone interventions in the provision of caregiver support. They are consistent with a review showing the benefits of telephone support for caregivers of people with dementia (Mason & Harrison 2008), which concluded that frequent telephone calls should be made to caregivers who require additional support. The authors of that review concluded that evidence exists to support the theory that telephone support can reduce caregiver burden and other symptoms of stress for family carers, especially older women.

3.6.2.8 Impact of intervention intensity on study outcomes

In the studies reviewed here, the original researchers did not provide a rationale for the duration of the telephone interventions, length of time allocated/given for each call or the frequency of the calls, nor did a clear pattern emerge from the studies reviewed. The length and frequency of calls varied from an average of 4 minutes (with frequency of calls determined by the caregiver caller) to 2 hours (weekly for 12 weeks) (Brown *et al.* 1999). Most intervention studies designed to change or modify caregiver behavior delivered calls over an 8-10 week timeframe or longer and this pattern was similar for both one-to-one and group sessions

Intervention intensity was not consistently related to reported outcomes, with both low and high intensity interventions reporting positive and significant outcomes. However, there was a tendency for the interventions designed to promote caregiver change, such as coping (Walsh & Schmidt 2003, Dollinger *et al.* 2006) and problem-solving (Grant *et al.* 2002, Rivera *et al.* 2008, Connell & Janevic 2009, Elliott & Berry 2009) to be of low to medium intensity, with the exception of Tremont *et al.* (2008), which was of high intensity. Other studies focused on skill development, education, coaching, providing support and counselling. The aim of most of the interventions (n=12) was to provide support which varied in intensity: high (n=5) (Goodman & Pynoos 1990, Strawn & Hester 1998, Davis *et al.* 2004, Smith *et al.* 2004, Smith & Toseland 2006), medium (n=3) (Meier *et al.* 1995, Brown *et al.* 1999, Goelitz 2003), low (n=4) (Stewart *et al.* 2001, Moore 2002, Walsh & Schmidt 2003, Shanley 2008) and low-medium (n=1) (Hauber & Jones 2002). Similar variability in the impact of intervention intensity was reported for interventions designed to help caregivers cope (Dollinger *et al.* 2006, Tremont *et al.* 2008) and those of psychological therapeutic interventions such as counselling and cognitive behavioural therapy (Mahoney *et al.* 2003, Bank *et al.* 2006, Glueckauf *et al.* 2007) and interventions with an educational focus (Davis 1998, Brown *et al.* 1999, Mahoney *et al.* 2003, Herman *et al.* 2006).

Likewise, there was no clear pattern in terms of the type of intervention, length or duration of interventionist training, or training in relation to the professional expertise of the interventionists. Due to the variety of interventionists used and outcomes measured across the studies in this review, it is not possible to link positive outcomes to any one

discipline or group. One study (Hartke & King 2003), which used a mixture of interventionists [psychology, social work or nursing], reported that the treatment group was more distressed overall at the first time point with significantly higher levels of depression, burden, loneliness, and stress and lower sense of competence. However, positive findings were reported in terms of stress, sense of competence and satisfaction.

Some studies did not identify who the interventionists were (Moore 2002, Goelitz 2003, Davis *et al.* 2004, Dollinger *et al.* 2006, Elliott & Berry 2009). Of those that did, positive findings were reported for those facilitated by professionals and calling carers. Studies facilitated by non-professional callers were, however, primarily to provide support with little therapeutic intervention.

3.6.2.9 Summary and implications for intervention development

This review demonstrated the potential positive contribution of telephone support interventions for caregivers. Technology based interventions and those based on models of problem-solving have demonstrated potential for positive outcomes in the caregiver population. However, developing and testing an intervention for nominated support persons of PwMS is a complex activity and is limited by context and resources. This review on telephone interventions revealed the piecemeal approach to testing telephone support interventions for caregivers and the challenges encountered by researchers when doing so. Additionally, the sketchy approach to reporting the processes within publications that primarily focus on findings was highlighted. For the purpose of the intervention tested in this study, the importance of testing outcomes in relation to intervention intensity, the need to consider the professional discipline of the interventionist, the intensity and duration of interventionist training, the requirement to monitor fidelity to the intervention and potential negative effects of the intervention were identified.

3.7 Chapter summary

Nurse Specialists in Multiple Sclerosis are nurses with specialist knowledge and expertise in the area of MS. They practice within the scope of nursing practice (An Bord Altranais 2000) within which the scope of their role in the provision of support to PwMS and their SPs, is determined. As PwMS and their SPs are nationally based, outside of scheduled healthcare contacts, support is provided via telephone and on demand. There is a dearth of quality research evaluating the impact of the role of the NSMS and there is a lack of formal recognition of their contribution to supporting family members and caregivers.

The wide geographical location of PwMS and their caregivers was a major factor influencing the method adopted for the delivery of this intervention. The review of reviews of interventions for caregivers of people with chronic illnesses revealed that interventions based on information technology had potential. A subsequent review of telephone support interventions for caregivers highlighted that telephone interventions based on models of problem solving had potential. The review highlighted the challenge of developing and testing an intervention for caregivers and key issues to consider when developing and testing the intervention for this study emerged. These issues are combined in the next chapter, which presents the process of selecting a specific theory to guide the development and delivery of the intervention for this study, and the development of the intervention itself.

Chapter 4: Selecting the theory and developing the intervention for this study

4.1 Introduction

The first stage in the theoretical development of the intervention was presented in Chapter 3. The review of systematic reviews of interventions for caregivers of people with chronic illness highlighted that interventions delivered by telephone had potential to improve caregiver outcomes. Likewise, the literature review of telephone interventions for caregivers provided further support for the use of telephone as a medium for the delivery of the intervention and highlighted that problem-solving models had potential to positively impact on caregiver outcomes. This chapter will focus on the identification of a specific model to guide the development and delivery of the intervention. The resources [human and financial] available at the time of intervention development and the geographical location of patients were key issues that needed to be considered when designing the intervention. The selection of the model was therefore influenced by the feasibility of its delivery and consequent likelihood of successful implementation. This required fieldwork to determine usual practice, the characteristics of the NSsMS and the availability of resources. Simultaneously, a literature review was carried out to determine the potential of communication models to guide the delivery of the intervention, but no suitable model was identified. Therefore, the review on communication models has not been included in this thesis. However, a literature review on problem-solving models was informative and is included in this chapter.

In this chapter, the findings from the fieldwork carried out to inform the development of the intervention, such as the availability of resources and usual care practices, is presented. The goal of the intervention, the model selected to guide the delivery of the intervention and how, based on that model, the intervention was developed, are presented. The intervention and the training provided to the interventionists are also described.

4.2 Fieldwork to develop the intervention

In order to guide the selection of the model most likely to be feasible for this intervention it was important that the resources available for its delivery and usual care were identified. To achieve this, interviews were carried out with the NSsMS, using semi-structured interviews (Appendix 8). Following receipt of ethical approval and access to the sites, these interviews were carried out at the hospital site, at the request of the NSsMS. Details of the methods used for this qualitative component of the study are presented in the methods chapter (chapter 6).

All three sites were large teaching hospitals providing services to people on a regional basis. Although the sites had different numbers of MS patients registered with them (site 1 N=280, site 2 N=2000, site 3 N=360), there was a single NSMS at each site. Community support services were similar across the sites and included public health nurses, MS carer support groups, MS care workers, community physiotherapy and occupational therapy. Access to these services varied and depended on demand at any given time. Analysis of the interviews with the NSsMS indicated that although access to services varied for PwMS and SPs, the professional characteristics of the NSsMS and the type of care they provided was similar. Their roles, the systems of referral and composition of the multi-disciplinary team were similar across sites. Most decisions regarding the care of PwMS were made in consultation with the medical team.

The nurses were certified MS nurse specialists who had been certified as competent to care for PwMS and their family members/carers (SPs). Two NSsMS had 12 years and the third had 15 years of nursing experience prior to taking on the NSMS role. They were similar in terms of the length of time they were doing the role (9, 11 and 12 years of experience). They had all completed the postgraduate diploma in nursing (which incorporated the certificate in MS nursing course) and this was the highest level of education they had achieved. They differed in the type of exposure to post-qualifying training on providing support to PwMS and their families. One of the NSsMS had completed a certificate in counselling skills since qualifying as a nurse, one had

attended in-house workshops run by the psychologists, and the other NSMS did not identify any further training in communication skills since registering as a NSMS.

Each hospital had only one NSMS in post, which limited the time that she/he could commit to delivering the intervention. These NSsMS provided care to PwMS within a wide geographical area, which created a barrier to face-to-face contact. Outside of scheduled hospital appointments, the NSMS provided care to PwMS and SPs via telephone. Across sites, the number of calls the NSsMS received from family members or caregivers were similar relative to the number of attending PwMS. Approximately 2 in 10 calls received from PwMS or support persons were from SPs. SPs usually left a voice message and in two of the sites they got a call back within 24 hours or the next working day, if the message was left on a Friday. At busy times, the NSsMS prioritised the call backs and responded to the most pressing ones, leaving the others to a later time. In the third site, the NSMS initially provided a call back service within 24 hours, but this was no longer always possible. All calls were responded to within the week or the following week if the call was left on a Thursday evening or Friday.

The NSsMS indicated that SPs raised a variety of issues pertaining to the PwMS and the SPs themselves, when they made contact. These included the physical and psychological care needs of PwMS and how they impacted on the SP in terms of burnout and the need to talk to someone. Due to the population affected by MS, it was not unusual for parents of young people diagnosed with MS to make contact. Issues pertaining to PwMS included concern regarding change in symptoms in relation to elimination and cognitive impairment, about their reaction to diagnosis and other psychological concerns, community supports and the need for extra help. SPs themselves raised issues regarding their own ability to cope, their emotional concerns, and how the PwMS's ability to cope was impacting upon them. The type of support they provided is captured in the following extracts from NSsMS interviews:

> "...there's a whole myriad of issues that people would ring up about. I suppose the most common would be usually people are in trouble ..." (NSMS site 1).

"...very often ...family members who are primary carers of patients would ring and it could be just a conversation that's more problem-solving, often it can be solved on the phone" (NSMS site 2).

"But it's there for family members when they call, it's usually more to do with personal, and how they are coping with their emotional issues...and the impact it has on the carers themselves" (NSMS site 3).

NSsMS provided a range of services to SPs, including listening to their concerns, pointing them to appropriate resources, imparting knowledge and providing advice, questioning, intervening and arranging for services. All three NSsMS stated that a large part of the support provided to SPs was reassurance and they were all cognisant of the PwMS's right to confidentiality. One NSMS raised a concern in relation to the potential for conflict of interest and had concerns regarding the motives of some individuals who may call them on behalf of PwMS. Examples provided in the interviews include:

"You have to ask the carer family member, does the person know that they're calling on their behalf... (NSMS site 2)

"...And it's usually working through issues with them, you know... Some of it can be reassurance. You could say to the support person or carer, no she's not having a relapse... (NSMS site 2)

"... sometimes I'd be 20 minutes to half an hour on the phone just having a listen" (NSMS site 1).

"And I was trying to reassure him that I think, well at least I hope what I did was reassure him that anything he was talking

about we can probably do something about. We can certainly improve it, I don't know if we are going to be able to resolve it completely but we can certainly improve the situation at home" (NSMS site 3).

"And sometimes she would just ring because she wants somebody to talk to..." (NSMS site 3).

All NSsMS felt they were well positioned to provide support to SPs. They felt that their experience and knowledge of the PwMS enabled them to provide this support.

"...I've twelve years doing the job ... I hopefully provide a service that people are happy with and that I can actually make a difference to whatever situation they present me with ...one of the biggest things I've learned over the years is just ...listening ...over the years I've learned the right approach..." (NSMS site 1).

"... I suppose with experience ... I think I'm about nine years now in the post ... So over the years I've learned how to better manage and prioritise, and your problem-solving skills get better .....Because you always have to think laterally in the context of individual cases because everyone's scenario [is different], I've probably come across every single scenario you could think of now at this stage..." (NSMS site 2).

"...I think because the years I've been doing this now, ... puts me in a good position to do this" (NSMS site 3).

These NSsMS did however identify barriers to providing such support, including getting the consent of the PwMS or SP not wanting support, availability of resources (including time), community resources, lack of administrative support (which meant they had to do non-nursing administration duties) and internal supports such as neurologists and psychologists.

These nurses also had a number of years clinical experience in the specialty and were experienced at managing telephone enquiries from PwMS and SPs. My challenge was to develop a structure within which that guidance could be delivered and tested. The range of issues that NSsMS deal with meant that it was essential that the model selected to guide the delivery of the intervention allowed for flexibility and tailoring in the implementation phase. The exploratory nature of this study provided an opportunity for ongoing evaluation, development and refinement of the intervention over time. All these factors, along with previously reported positive benefits from telephone interventions, influenced the decision to use the telephone as the mode of intervention delivery. They also guided me in determining the goal of the intervention and identifying a theory to guide the delivery of the intervention.

4.3 Goals of the intervention

The primary goal of the intervention was to enable NSsMS to help family members and carers of PwMS by enabling them to learn problem management skills and helping them feel prepared for their role in supporting a person with MS. A secondary goal was to determine the potential impact of the intervention on reducing burden and increasing the QoL of family members/caregivers of PwMS.

4.4 Selecting a model to guide the Intervention

The goal of the intervention called for a structured but flexible approach to enable NSsMS to prepare family members and caregivers for supporting PwMS. It was important that the model selected to guide the intervention enabled NSsMS to explore issues/concerns that family members/caregivers were encountering, in addition to empowering them to manage issues that may arise into the future. Although communication is at the core of telephone contact, no suitable model was identified from a comprehensive search of the literature on communication models. As shown in

chapter 3, interventions based on problem-solving models demonstrated potential benefits. In a further attempt to identify a suitable model, I carried out a focused literature review on problem-solving models used in caregiver research.

4.4.1 Review of problem-solving models used in caregiver research

The purpose of this review was to examine the potential benefits of caregiver interventions based on problem-solving models and identify a potentially suitable model of problem-solving for this study. I undertook a detailed search of the literature on the use of problem-solving models in caregiver research using the following key words 'carer' 'caregiver' 'problem-solving' (see figure 7 for databases searched and search details).

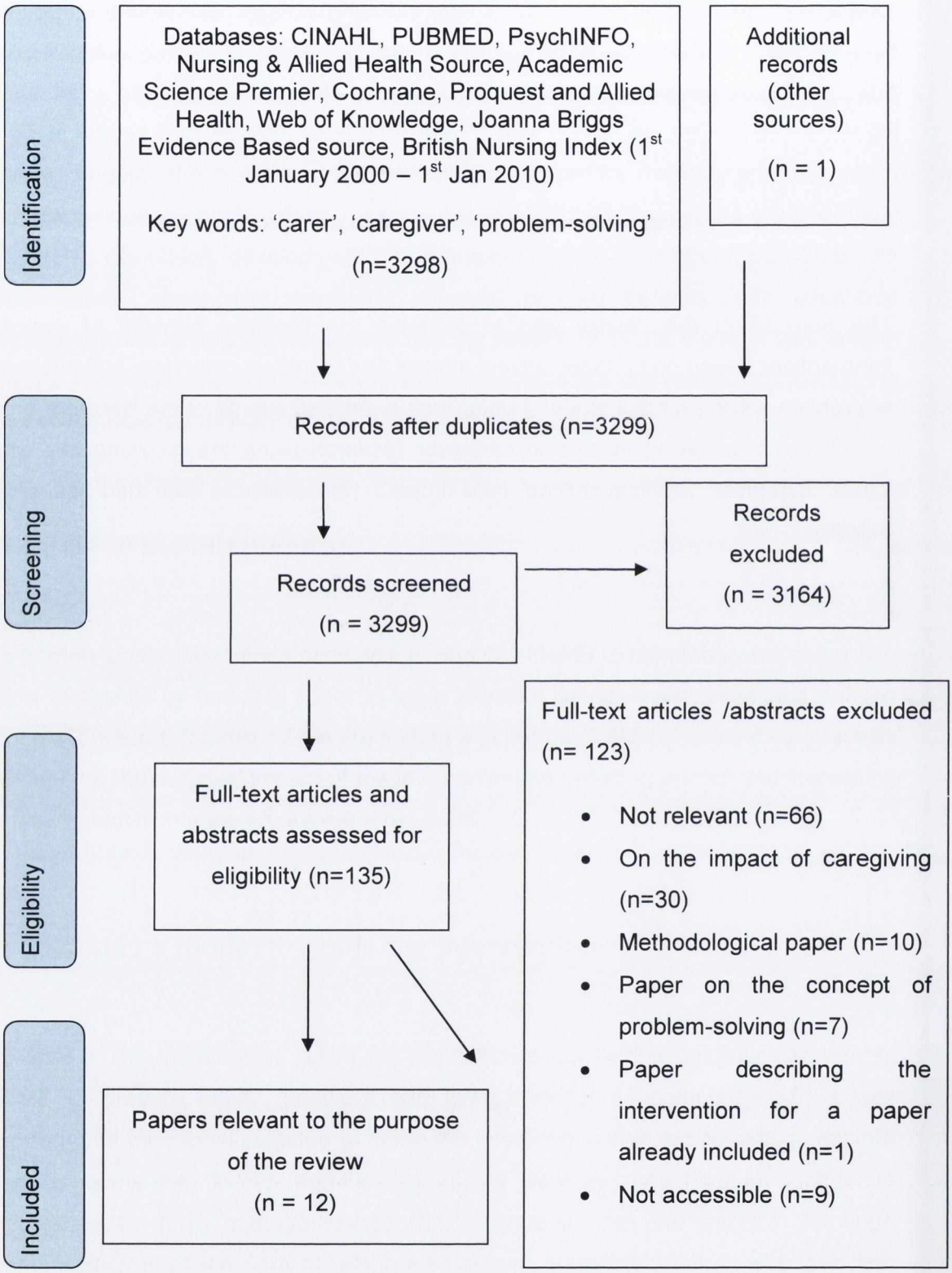


Figure 7: Flow diagram for literature search and selection for problem-solving interventions

4.4.2 Outcomes of problem-solving interventions

Twelve studies evaluated the outcomes of problem-solving interventions and one paper (Kurylo *et al.* 2001) provided an overview of a problem-solving model (Table 6). All the studies were carried out in the USA and all demonstrated some positive outcomes, although these outcomes varied according to the overall purposes of the studies and outcome measures used. In general, they support the potential value of using problem-solving as a means of supporting caregivers across a number of illnesses [cancer, spinal cord injury, women with disabilities, multiple sclerosis, stroke survivors, and traumatic brain injuries]. Some problem-solving interventions were part of multi-component interventions that included, for example, an education (Finlayson *et al.* 2009a) or cognitive behavioural therapy (King *et al.* 2007) component.

It is difficult to extract which outcomes resulted from the problem-solving component of such interventions, especially if the components are integrated into one larger intervention and researchers do not explore which elements of the interventions are active during intervention delivery. Despite this, the positive outcomes produced from single component problem-solving interventions (Grant *et al.* 2002, Cameron *et al.* 2004, Elliott & Berry 2009, Elliott *et al.* 2009, Demiris *et al.* 2010) support the potential value of problem-solving interventions for caregivers. Outcomes for which a positive effect was found include physical health (Demiris *et al.* 2010), burden (McMillan *et al.* 2006, King *et al.* 2007, McMillan & Small 2007, Demiris *et al.* 2010), improvements in emotional tensions (Cameron *et al.* 2004), stress and rumination (Dollinger *et al.* 2006), depression (Grant *et al.* 2002, Dollinger *et al.* 2006, King *et al.* 2007, Elliott *et al.* 2009), increased social functioning (Grant *et al.* 2002, Elliott & Berry 2009), improvements in vitality, role limitations related to emotional problems (Grant *et al.* 2002) and improved preparedness to care (Grant *et al.* 2002, King *et al.* 2007, Finlayson *et al.* 2009a). These positive findings support findings for caregivers as reported by D'Zurilla & Nezu (2007), where caregivers of people with physical and cognitive impaired older adults reported improvements in assertiveness, burden, depression and distress, and those of people with dementia experienced less psychological distress and greater psychosocial adjustment. Similarly, families of children with severe traumatic brain injury were found

to benefit in terms of reduced burden, parental psychiatric symptoms, depression and parenting stress (D'Zurilla & Nezu 2007).

Table 6: Papers evaluating outcomes for problem-solving interventions

Author, Year, Country	***Participants & study design***	***Intervention/comparison***	***Findings***
Cameron *et al.* (2004) Canada	Family caregivers of people with advanced cancer. N=58 (55.2%) of the available population (34 (32.4%) analysed). **Design:** One-sample pre-test, post-test design.	**Intervention:** COPE (Address problems using a five step approach. Introduced to the problem-solving techniques using example from the Home Care guide for Advanced Cancer). **Mode of delivery:** face-to-face (one-to-one). **Frequency:** once **Duration**: 1 hour session **Control:** N/A **Outcomes measured:** Problem-solving, emotional well-being, caregiver assistance, caregiver self-efficacy	Improvements in emotional tension (p =0.024). Caregivers reporting higher levels of fatigue at baseline were more likely to report an increase in impulsiveness-carelessness problem-solving between baseline and follow-up assessment (r=0.40, p<0.02).

Demiris *et al.* (2010) USA	Hospice caregivers. N=29 **Design:** Pilot study: pre-test, post-test design.	**Intervention:** Problem-solving training adapted from previous intervention studies (e.g. Grant *et al.* 2002, Nezu *et al.* 2003). **Mode of delivery:** Three in-home intervention visits between day 5-16 of hospice admission (as close to days 5, 11 and 16 as possible). **Frequency:** Every 5-6 days. **Duration:** 16 days. **Outcomes measured:** Quality of life, anxiety, problem-solving abilities, burden.	Quality of life: Only subscale that decreased was the physical dimension. CRA: Increase of positive esteem average and decrease of the average value of lack of family support, impact on finances, impact on schedules and on health. Qualitative outcomes: All caregivers: Intervention was useful. Advantage: It had structure and provided specific tools. Convenience of delivery in own home, opportunity to take time and reflect on one's options and strategise/prioritise to prepare for challenging time. Disadvantages: (n=2 respondents). • Discussing patient problems in a residential setting that did not provide privacy for these conversations (n=1). • All items on the CRA did not apply in the person's case (n-1).

Dollinger *et al.* (2006) USA	Rural Family Caregivers (no specific illness). N=36 carers in the analysis. **Design:** Intervention trial with 2-group assignment (ongoing study). Paper focuses on the needs and challenges for informal caregivers in rural settings.	**Intervention:** Tele-Help Line for Caregivers (TLC): A Comprehensive Tele-health which included a social problem-solving component incorporating the problem-skills training program. **Mode of delivery:** Via a helpline (one-to-one). Intervention group received a fully manualised and structured programme with regular appointments. **Frequency:** Set regular appointments or call interventionist at a convenient time to complete the different components. **Duration:** 8-week (but can take longer due to caregivers' schedules or time constraints). **Length of contact:** Not stated. **Group assignment:** Random. **Control:** Helpline condition. **Outcomes measured:** Quality of life and problem-solving.	Caregivers in the treatment group reported less psychological distress upon completion of the intervention, and this improvement was maintained at the six-month follow-up ($p <$ .05). Symptom distress was less after eight weeks and maintained at six months. Improved social role functioning was evident in the eight-week treatment group. At the 8-week time-point caregivers in the treatment group indicated greater perception of attachment support and social integration on the 'social provision scales' (all $p <.05$).

Elliott *et al.* (2008) US	Caregivers and persons with spinal cord injury (SCI). N=61 (m=7, f=54). **Design:** RCT.	**Intervention:** Problem-solving training program based on techniques demonstrated by Nezu et al. (2003). **Mode of delivery**: One 2-3 hour face to face in-home session. Follow up sessions via videoconferencing. **Frequency:** Monthly (additional contacts as requested by participants) (one-to-one). **Duration:** 1 year. **Control:** Education only (received telephone contact from research staff member and discussed educational concerns, educational materials provided based on need and monthly face-to –face videoconferencing contact provided. **Group assignment:** Random assignment (29 Control, 32 intervention). **Outcomes measured:** Social problem-solving abilities, depression, life satisfaction, quality of life.	Only outcome with significant interaction over time was depression (p=0.044). Pre-treatment depression was a significant covariate (p<0.01). Positive effect on care recipient social functioning over time (p<0.05).

Elliott *et al.* (2009) USA	Family caregivers of women with disabilities. N=91 **Design:** Intervention study- 2 group design.	**Intervention:** Problem-solving training (PST) adapted from previous intervention studies. (e.g. Grant et al. 2002; Nezu et al. 2003). **Mode of delivery:** In-home PST sessions at months 1, 4, 8, 12. Telephone sessions once a month on the alternate 8 months (one-to-one). **Frequency:** Monthly. **Duration:** 1 year. **Control:** Education only (received telephone contact from a control group specialist. Structured telephone conversation 10-15 minutes in length where previously mailed health education materials were reviewed. **Group assignment:** Simple random assignment (43 Control, 38 intervention). **Outcomes measured:** Depression, life satisfaction, caregiver health complaints, social problem-solving abilities.	Baseline higher dysfunctional problem-solving among caregivers in the intervention group which approached significance (p=0.09). Baseline constructive problem-solving was not significantly associated with depression across time. Baseline dysfunctional problem-solving was significantly associated with depression across time (p<0.01). Findings suggest that the impact of PST on depression over time was due, in part to the effects of PST on constructive problem-solving. Effects of PST on caregiver depression were unrelated to caregiver age or functional deficits of the care recipient.

Elliott & Berry (2009) USA	Caregivers and persons with spinal cord injury (SCI). N=60 (m=11, f=49) **Design:** RCT	**Intervention:** Problem-solving training program based on techniques demonstrated by Nezu et al. (2003). **Mode of delivery:** One 2-3 hour face to face in-home session/clinic office session. Follow up sessions, telephone contacts and booster sessions and mailings of educational materials as needed. **Frequency:** Not stated (one-to-one). Duration: 1 year. **Control:** Usual access to OPD clinic staff and information provided by rehabilitation staff concerning SCI management. **Group assignment:** Random assignment (30 Control, 30 intervention). **Outcomes measured:** Social problem-solving abilities, depression, quality of life.	Two SF-36 health-related subscales had interaction effects approaching statistical significance: physical functioning (p=0.07, effect size 0.73) and social functioning (p=0.09, effect size 0.44). Caregivers who received the PST intervention reported an increase in social functioning. Control group reported a decline in social functioning.

Finlayson *et al.* (2009a) USA	Caregivers of people ageing with multiple sclerosis. N=19 (12 completed all sessions). **Design:** Pilot using pre-post design.	**Intervention:** Meeting the Challenges of Multiple Sclerosis: Providing Support through Problem-solving (PS) (group based educational and PS programme). **Mode of delivery:** 2 hour face to face group sessions. **Frequency:** weekly. **Duration:** 5 weeks. **Group assignment**: N/A. **Outcomes measured:** Problem-solving, coping, preparedness to care, self-efficacy.	Caregiver problem-solving self-efficacy: no changes. Coping with Multiple Sclerosis caregiving Inventory: practical assistance ($p<0.02$) and positive re-framing ($p<0.01$) showed significant improvement. Preparedness to care: significant findings – participants reported they felt more prepared to care ($p<0.02$). No significant improvement in caregiver self-care self-efficacy. Qualitative focus group findings: Positive feedback: Participants reported that the content was relevant, length of the programme was appropriate, they liked the methods of delivery. Some did not like the homework assigned but others found it helpful. Provided an opportunity to learn the problem-solving process and its associated techniques. Group interaction (felt not alone). The programme helped them feel more confident in their caregiving role, less angry and happier and more aware of need to ask others for help and plan for the future. Others found that it helped to increase their knowledge of MS and resources. It helped them to see value of delegating, asking for help and thinking about the present and future needs of the person with MS and themselves. The course also validated some strategies that they were already using.

Grant *et al.* (2002) US	Primary family caregivers of stroke survivors. N=74 (11 lost (15% attrition and mortality, does not clearly state how many participants in each group). **Design:** Randomised, 3-group, repeated-measures experimental design.	**Intervention:** SPTP model . **Mode of delivery:** 3 hour face to face in-home session followed by weekly (weeks 2, 3 and 4 post-discharge) and biweekly (weeks 6,8,10 and 12 post-discharge telephone contacts). **Interventionists:** Trained research nurse (one-to-one). **Frequency:** Weekly and bi-weekly. **Duration:** 12 weeks. **Sham intervention** same number of contacts as the intervention group and identified services the stroke survivor received since last contact. **Control:** No details provided. **Group assignment:** Random. **Outcomes measured:** General health, social problem-solving abilities, satisfaction with health care, depression, caregiver preparedness, caregiving burden.	Significant improvements in vitality (p=0.13), mental health (p<0.001), and role limitations related to emotional problems (p=0.007), for the intervention group. Statistically significant group by time effect for problem-solving (p=0.0059). Intervention group had significantly better social problem-solving skills than either the sham intervention or the control group. They had statistically significant less negative orientation in impulsivity/carelessness (p=0.007), but more rational problem-solving skills (p=0.025). Problem orientation: no change over time for the intervention or sham group. Control group deteriorated. Intervention group showed statistically significant greater caregiver preparedness (p<0.001), and less depression (p<0.001) than either the sham intervention or the control group. No statistical significant difference amongst the groups for caregiver burden. Satisfaction with health care services decreased over time in control group while remaining comparable in the intervention and sham groups. Sham group – no significant changes over time on any of the dependent variables.

King *et al.* (2007) US	Caregivers of stroke survivors. N=25 (14 completed the final assessment). **Design:** A single-group repeated measures design.	**Intervention:** CPSI (10-session intervention combined PS (problem orientation and problem-solving skills) and CBT (relaxation training, reframing negative thoughts). **Mode of delivery:** Initial 2- 3 in-person sessions during survivor's hospitalisation. 7 or 8 post-discharge sessions via telephone (one-to-one). **Frequency:** Weekly. **Duration:** Lasted up to 7-8 weeks post discharge. **Comparison group:** 15/136 matched caregivers who participated in a descriptive, longitudinal study of stroke caregivers and their stroke survivors. **Group assignment**: N/A (Single group using comparison group from matched longitudinal study). **Outcomes measured:** Helpfulness, burden, coping, depression, anxiety, perceptions of life changes, healthy caregiving.	CPSI – significant improvement over three times in the following: Depression: F (2,26)=13.74, $p<0.001$; Preparedness: F (2,26)= 20.20, $p<0.001$; Anxiety: F (2,26)=3.67, $p<0.001$; Cognitive function: F (2,26)=13.40, $p<0.001$; The life change scale: F (2,26)=1.45, $p<0.001$; Burden: F (2,26)= 0.83, $p<0.001$; Taking care of own needs (not significant): F (2,26)=1.13, $p>0.001$; Hypothesis that severity and lower rate of depression in the CPSI group post-intervention was supported. Burden and taking care of own needs worsened over time.

McMillan *et al.* (2006) US	Primary family caregivers of hospice home care patients N=329 **Design:** Randomised, 3-group, repeated-measures experimental design.	**Intervention:** COPE intervention. **Mode of delivery:** In-home session (one-to-one) (McMillan & Small 2007). **Interventionists:** Nurses hired from the hospice. **Frequency:** 3 visits (visit 1 45 minutes, visit 2 & 3, 30 minutes (McMillan & Small 2007). **Duration:** not stated (overall duration of the study 30 days). **Treatment group 1:** (experimental group) (n=111) standard care plus teaching on the COPE intervention. **Treatment group 2:** (n=109) Standard care plus three friendly visits of same schedule and length as COPE intervention. **Control:** (n=109): standard care from hospice staff. **Outcomes measured:** Caregiver quality of life, burden, coping, mental impairment.	Significant findings/trends in that direction for time and intervention group for QoL (p=0.054), burden related to patient symptoms (p=0.001) and caregiving task burden (p=0.021). Caregiver task burden: main effect of time (p=0.014) with increases in burden scores being seen with time. Treatment group by time interactions for QOL, symptom burden, caregiver task burden was not significant for the comparison between usual care and support condition but for the comparison between usual care group and the COPE intervention group, the group by time interactions for caregiver was all significant. Only the COPE intervention group showed statistically significant improvements in QOL ratings over time, usual care group experiences ns over time.

Rivera *et al.* (2003) US	Caregivers and persons with spinal cord injury (SCI). **Design:** Intervention study-2 groups.	**Intervention:** Problem-solving intervention (FOCUS) using a tailored approach. **Mode of delivery:** Via TV Videophone (one-to-one). **Interventionists:** Master's level psychologist, counselor, or nurse. **Frequency**: Monthly. **Duration:** 1 year. **Control:** No-problem-solving treatment. **Group assignment**: Randomised. **Outcomes measured:** Social problem-solving, depression, health.	Interest and satisfaction remained high for participants. Some technological connection problems. Case study supports the hypothesis that caregivers of people with chronic illness need skills to negotiate a wide array of problems.

Rivera *et al.* (2008) US	Family caregivers of persons with traumatic brain injuries (TBIs). N=67 **Design:** RCT.	**Intervention:** PST based on D'Zurilla and Nezu social problem-solving model. **Mode of delivery:** 4 in-home sessions (month 1, 4, 8, 12) and 8 follow-up telephone calls (for the remaining 8 months) (one-to-one) (one-to-one). **Frequency:** Monthly. **Duration:** One year. **Treatment group**: (experimental group) PST (n=33). **Control:** Telephone based education group (n= 34). **Group assignment:** Simple randomisation. **Outcomes measured:** Depression, well-being, health complaints, caregiver burden, problem-solving abilities, coping.	Caregivers in the PST group reported a significant decrease in depression scores over the 12-month period ($p<0.005$) effect size Cohen's d =0.- 062 (moderate effect). Well-being: no statistically significant interaction between treatment and time on caregiver scores on the SWLS. Health complaints: Significant treatment by time interaction effect for the caregiver scores on the health complaints ($p<0.005$). Caregiver burden: NS.

Caregiver problem solving interventions (CPSI), Social Problem-solving telephone partnership (SPTP), Outpatient department (OPD), Problem solving (PS), Problem solving training (PST).

4.4.3 Problem-solving models

Three problem-solving models were identified from the studies reviewed: the social problem-solving model, Project FOCUS and COPE (see table 7). All three models are based on the work of D'Zurilla and Nezu, which originates from psychology in the 1960s and 1970s (D'Zurilla & Nezu 2007). The concept of social problem-solving relates to problems "... that occur in the natural social environment" (D'Zurilla & Nezu 2007, p.12). The authors point out that the use of the term social does not limit or restrict the use of the model but refers to "...problems that occur in the natural social environment" (D'Zurilla & Nezu 2007, p.12).

The social problem-solving theoretical model is based on two main components: 'problem orientation' and 'problem-solving skills'. Problem orientation refers to one's own awareness and views on problems of living and ability to solve them. Within the model, there are five aspects to problem orientation: Problem recognition (identification of the problem), problem attribution (causal beliefs), problem appraisal (person's evaluation of the significance of the problem for well-being which can be negative or positive), perceived control (belief in own ability to solve the problem and belief that problems are solvable) and time/effort commitment (realistic estimate of time required to solve the problem and willingness to give the time and effort required to solve the problem) (D'Zurilla & Nezu 2007). The skills required for problem-solving include problem definition and formulation, generation of alternative solutions, decision making, solution implementation and verification (D'Zurilla & Nezu 2007). In the studies summarised in Table 7 the interventions based on this model were primarily delivered via in-home face-to-face sessions for a number of sessions followed by videoconferencing or telephone follow-up.

In the 1990s, the Social Problem-Solving Inventory (SPSI) was developed based on 2 factors, problem orientation and problem-solving skills. This was further developed in the mid 1990s, when the two factor model was found lacking and was replaced by a 5-factor model. The 5-factor model consisted of two dimensions for problem orientation (positive and negative) and three dimensions from problem-solving styles (rational (effective) problem-solving; impulsivity/carelessness style, and avoidance style).

Individuals who score high on positive orientation and rational problem-solving are likely to report positive problem-solving outcomes while those that score high on negative problem orientation, impulsivity or carelessness style and avoidance style demonstrate negative/maladaptive outcomes (D'Zurilla & Nezu 2007).

Although the social problem-solving model was the model most extensively used in research studies (Grant *et al.* 2002, King *et al.* 2007, Rivera *et al.* 2008, Elliott & Berry 2009, Finlayson *et al.* 2009a, Demiris *et al.* 2010), other models such as Focus (Dollinger *et al.* 2006, Rivera *et al.* 2008) and the COPE model (Cameron *et al.* 2004, McMillan *et al.* 2006, McMillan & Small 2007) were also used, albeit to a lesser extent. All three models include problem identification, goal setting, identification of possible solutions and their application. They all rely on participants remaining positive and actively engaging in the process (Table 7). The COPE model was delivered in face-to-face sessions and the FOCUS via teleconferencing. In order to determine the suitability of these models to guide the intervention in this study, the models are examined in more detail in the next section.

Table 7: Problem-solving models used in caregiver research studies

Title and original authors of the Model	Definition o f Problem-solving	Model overview	Structure/design of interventions based on the model
D'Zurilla and Nezu social problem-solving model.	"Social problem-solving refers to the process of problem-solving as it occurs in the natural environment or "real world" (D'Zurilla & Nezu 2007, p.11). Problem-solving is conceived as a conscious, rational, effortful and purposeful activity.	3 major concepts a. Problem-solving b. Problem c. Solution Model identified 4 major skills: a. Problem definition and formulation b. Generation of alternative solutions c. Decision making d. Solution implementation and verification.	Demiris *et al.* (2010) Addresses four skills: (1) Problem definition and formulation (data and information gathering; articulating the issues in clear terms; identifying the challenge and setting realistic goals (2) generation of alternative strategies (3) decision making (4) solution implementation. The intervention was designed using the acronym ADAPT. A= Attitude (positive, optimistic attitude). D= Define (the problem by obtaining facts, identify obstacles and specify realistic goals). A = Alternatives (to overcome obstacles and achieve goals). P= predict +/- consequences of each alternative and choose best one to achieve goal. T= Try Out (implement the solution in real life and monitor its effects) (Demiris *et al.* 2010, p.1006).

			Elliott *et al.* (2008) Aim: developed a problem-solving training program (PST) for family caregivers based on techniques demonstrated by Nezu *et al.* (2003). Adapted key elements and techniques of PST to ensure that the major theoretical components of PST were used to address the specific and immediate problems reported by each caregiver over time. 1st session: manuals containing information about problem-solving principles were provided. Participant getting the training were firstly orientated to the basic steps of the problem-solving process: problem definition, optimism and orientation towards problem-solving, creativity and generating alternatives, understanding and decision making, solving the problem with implementation and evaluation of a solution. During videophone sessions, carers were asked about progress made in the resolution of problems in previous session and given the opportunity to ask questions or discuss any new concerns. The 5 basic steps of PS were then discussed as they pertained to the immediate problem. Carers were presented with different problem situations for which they were asked to provide potential solutions. Twelve different scenarios were used, one for each monthly session. Elliott *et al.* (2009) Similar to Elliott *et al.* (2008). Telephone sessions were based on scripts used in the protocol by Grant *et al.* (2002).

			Elliott & Berry (2009) - similar to Elliott *et al.* (2008). Finlayson *et al.* (2009a) The program comprised written, verbal, visual and experiential activities. This provided the opportunity to discuss problems participants' experienced with caregiving, to develop new skills and identify resources. The course content was applied via planned homework with opportunity to discuss it in subsequent sessions. The researchers also developed resources such as videos and checklist for participants to refer to as required. Grant *et al.* (2002) Family caregivers were taught to use a positive problem orientation (viewing problems as a normal part of life, that problem-solving is a viable way to cope, and that problem-solving efforts will be successful). They were taught to use 4 steps when solving problems: (1) identify and define the problem, (2) decide what needs to be accomplished and list possible solutions to the problem, (3) Choose and test the best solution(s), (4) evaluate outcomes of problem-solving. King *et al.* (2007) Participants identified their most difficult problems with the counsellor and developed plans to solve them using the problem-solving steps (steps not

			outlined in the paper). They were asked to complete home assignments, e.g. applying problem-solving steps to problems, rating their mood and practising relaxation. The interventionists were flexible and rescheduled meetings as required (often held them in the evening or at weekends). Rivera *et al.* (2008) The intervention was tailored to deal with problems of immediate concern to the caregiver. Each participant received 4 home visits with a staff member who taught them problem-solving skills. Previous protocols were adapted and used in the protocol. Initial visit: interventionists discussed the 5 basic principles of the social problem-solving model (identify the problem, brainstorm solutions, critique the solutions, choose and implement a solution and evaluate the outcome. A list of problems obtained from a focus group with caregivers of traumatic brain injury was used to help guide the delivery of the intervention. Using a 'card-sort task' caregivers identified their most pressing problems. This was used to guide the discussion of feelings the carer experienced with problems and generated a list of possible solutions and goals to address the problems. Interventionist followed a similar format to structure interaction during the follow-up sessions. Telephone session – based on similar format to Grant *et al.* (2002). Guidelines and prompts were provided on a worksheet.
FOCUS		A five-step model of problem solving consisting of 1.defining the problem, 2. achieving a positive	Rivera *et al.* (2003) – Used the traditional five steps to problem-solving. Active participation in problem-solving is encouraged. Visual materials and worksheets are used. All aspects of the caregiver's behaviour are used to maximum therapeutic effect, including resistance and denial.

		frame of mind, 3. generate possible solutions, 4. implement a solution with the greatest potential for efficacy and 5. evaluate the results. These steps can be summarised under the principles of Facts, being Optimistic, the benefits of Creativity, the importance of Understanding all aspects of a potential solution and the steps associated with solving the problem (FOCUS).	Kurylo *et al.* (2001) Used a card-sorting (or Q-sort) task based on 24 problems identified by a focus group of 7 caregivers. Problem-solving training consists of 5 major components FOCUS: facts/problem definition, optimism/orientation; creativity/generation of alternatives/ understanding/decision making; solve/implementation and verification. These five components are viewed as a continuous and interlinking process rather than five stages to be followed in serial order. Dollinger *et al.* (2006) The social problem-solving component incorporates the FOCUS social problem-solving skills training program (Kurylo et al. 2001). In the full, 8-week manualised intervention, the interventionists work with the caregiver to determine the preferred order in which to complete the four treatment components (i.e., knowledge, social problem-solving, social support, and affect).
COPE Derived from problem-solving therapy.		COPE • Creativity • Optimism • Planning • Expert Information	Cameron *et al.* (2004) The intervention was based on COPE (Creative, Optimistic, Plan, and obtain Expert information). Problems were addressed as follows: (1) Define the problem (e.g., When does the problem occur? What are the consequences of the problem?), (2) Identify when to get professional help with the problem (e.g., Which symptoms need emergency attention and which do not?),

			(3) Identify what they could do to help (e.g., prevention or amelioration of symptoms), (4) Consider possible obstacles to their caregiving plan (e.g., What could prevent the success of this solution?), (5) Carry out and adjust the plan. On a one to one basis and using a slide presentation the interventionist described the steps and applied them to the general problem of "caregiving." Participants were encouraged to apply their own experiences to the process and given the opportunity to ask questions. The intervention lasted approximately 1 hour. McMillan & Small (2007) Caregivers were given training on assessment techniques and given three symptom assessment tools (one for constipation, pain and dyspnoea). Although 23 patient problems are in the Home Care Guide for Advanced Cancer this intervention emphasised three and use of these three served as a model for plans for other problems. Caregivers were educated on the need for careful assessment of symptoms and how to apply the tools. They were encouraged to assess for the three symptoms targeted. They received 3 visits during the 9 day intervention. They also received copies of the 'Home care guide for Advanced cancer' for personal use. The nurse demonstrated the use of COPE intervention problem-solving principles in caring for a patient with cancer. McMillan *et al.* (2006) – as above.

4.4.4 Reflection on literature reviewed and selection of the model to guide this intervention

Despite the extensive literature reviews conducted, the theory identified to guide the development of the intervention for this study, was incomplete. I felt that it was important to have a theory or model to guide the interventionists in the delivery of the intervention. Although problem-solving emerged as a core component of the intervention, a mechanism for its delivery using the telephone as a mode of communication remained elusive. While D'Zurilla & Nezu's model of problem-solving demonstrated potential, in all studies that used the model, both in-home, face-to-face sessions and follow-up telephone calls were used. I therefore reverted to my educational knowledge and experience of practice.

It was important that the model selected could guide the development of an intervention that could be delivered using the telephone, taking cognisance of the role of the NSMS, the feasibility of delivering the intervention in practice and the resources available. The theories reviewed did not make provision for these essential factors. In this study, SPs of PwMS were nationally based and therefore face-to-face methods were unlikely to be feasible. I reflected on these matters, and keeping the mode of delivery (the telephone) and the scope of the role of the NSMS at the fore of my thoughts considered that the ideal would be a communication model that facilitated use of a problem-solving approach to provide support. As ideas formed I asked myself 'How will that work?' 'What else is required?' Egan's model of the skilled helper (Egan 2010) emerged as a possible model as I had encountered this model as part of my ongoing educational and professional development. The model had the advantage of having the two key components that were required for this intervention: a problem management and a communication component.

4.4.5 Egan's model of the skilled helper

Egan's Model of the skilled helper was originally developed and first introduced in counseling by Gerard Egan in the 1970s (Wosket 2006). Although the model has

traditionally been applied in professional counseling, it can be used by individuals providing help in a number of settings, including nursing (Egan 2010). The generic nature of the model meant that it could be easily adapted and applied for the purpose of this intervention. The concept of helping, which underpinned the model, was congruent with the definition of nursing (see section 3.2.1). Egan (2010, p.8) defines helping as "...constructive change that makes a substantive difference in the life of the client". A competent helper is described as one who uses the required knowledge and skill to help others manage problems and create opportunity (Egan 2010). This model was particularly suitable to guide the delivery of this intervention because of its congruence with the role and definition of nursing. It is unique in that it combines the principles of communication and problem-management within the one model. Although not based on any specific theoretical model, Egan's model draws on a number of theoretical perspectives including Bandura's theory of self-efficacy, quoting Bandura's definition of self-efficacy, "...Perceived self-efficacy refers to beliefs in one's capabilities to organize and execute the courses of action required to manage prospective situations" (Egan 2010, p.346). This model was congruent with the requirements of this intervention and provided the overarching framework for development of the manual.

Although Egan's model was appropriate to guide the delivery of the intervention, it was also important that the intervention was delivered in a manner that respected the participants as adults. It is widely recognised and accepted in educational settings that adults prefer to learn through problem-solving and experience, rather than learning through specific content (see for example Barker & Doran 2006). Although I found no research evidence on the learning preferences of support persons of PwMS, similar learning preferences have been reported in research studies exploring the learning preferences of other support persons, for example, carers of asthmatic children (Dinakar *et al.* 2005) and carers of older persons (Zarit & Femia 2008). Consequently, a model of experiential learning, a well recognised approach to learning, which is congruent with adult learning theory, was integrated into the intervention.

4.4.6 Theory of learning: Androgogy

Experiential learning differs from traditional methods of learning (such as listening or reading) in that learning occurs through a process of active involvement (Quinn 2000) and working with real life problems and experiences. The best known experiential learning model is "Kolb's cycle" (Quinn 2000, p. 62) which is termed the Lewinian Experiential Learning Model (Figure 8) (Quinn 2000).

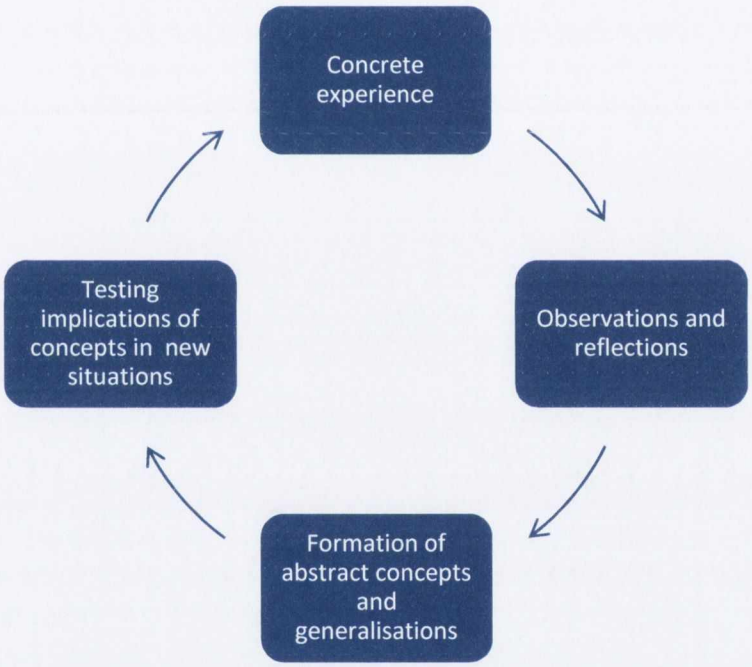


Figure 8: Stages of Lewinian Experiential Learning Model (Quinn 2000)

Although the model is traditionally applied to adult classroom learning, the stages of the model are readily applicable to learning for adult support persons. Using this model, the NSMS and support person could explore a real life problem that the support person is experiencing. Using the framework outlined in Egan's model of the skilled helper, the NSMS helps the support person to reflect on the problem by looking at it from different perspectives. The NSMS then helps the support person to develop new

perspectives and understanding of the problem and to test them out by applying them to the management of the problem.

4.4.7 Use of COPE model of problem-solving

To assist in achieving the objective of the intervention, it was also important to provide a structure to assist the nominated SP to engage in the process. Egan's model alone was insufficient to achieve this, as it was a guide for the helper and was not composed of tangible guides or worksheets for participants. The COPE model is represented diagrammatically (see support person guidebook in the accompanying CD rom) and provides an overview and structure to thinking about problem-solving. It was possible to incorporate the diagram representing the COPE model, which lets participants know that, problem-solving requires some level of creativity, optimism, planning and availing of expert information.

4.5 The intervention: building and modelling (representing) the intervention

The intervention that was developed and tested in this study is presented in compact disk format (CD) and is part of this thesis. The intervention comprises an intervention guide for the NSsMS, which includes the interview guide, the SP's guidebook and the intervention training manual. Prior to modelling (representing) the intervention, I reflected on the theories identified and asked myself 'How can I enact these models so that they can provide guidance for the NSsMS in helping SPs and teaching them problem management skills?' Thinking about this question, I recalled a telephone support intervention study that tested an intervention for people with the human immuniodeficiency virus (HIV) and their caregivers (Herman *et al.* 2006). The outline provided on the telephone component of the intervention as described by the study author (Herman *et al.* 2006) suggested to me that this intervention was worthy of

further examination. I emailed the main author (Debra Herman), who sent me a copy of the telephone component of the intervention. The process for delivering the content during the telephone contacts was scripted and it became clear to me that a script guided by Egan's model of the skilled helper may be suitable for the intervention for this study. Inspired by Herman's intervention, I mentally created a scripted image of the intervention. Due to the unique and varying needs of SPs in this study, the intervention script was not content specific. The NSsMS already had the subject specialist knowledge and expertise which underpinned the intervention. All of the NSsMS were involved in writing and had access to the Guide to Best Practice Multiple Sclerosis Specialist Nursing in Ireland, which was updated in 2012 (Irish Network of Multiple Sclerosis Nurses 2012). The Guide to Best Practice Multiple Sclerosis Specialist Nursing in Ireland provides information and guidance on all aspects of MS and its treatment including the management of psychosocial issues and patient and caregiver general wellness. They had also completed a postgraduate diploma in clinical practice, which included a certificate in MS nursing. A resource pack was included in the intervention manual to enable the NSMS to source any additional content specific material they may require during the delivery of the intervention or refer the SPs as required. For example, how to contact the community social worker, addiction and mental health websites such as sites for depression, suicide, bereavement and eating disorders (see CD rom, intervention manual).

The intervention was then modelled in a written manual format (see CD rom). The guide for the NSMS provided guidance on best practice in providing telephone support, an overview of the models guiding the intervention, information on the knowledge and skill required to deliver the intervention, a scripted interview guide for the four calls and documentation to be completed at the end of the calls. The scripted interviews were designed to provide a focus for the telephone contacts and help the NSMS and support persons structure their interaction using a problem management approach. During call one, the script guided the NSMS to firstly listen to the SP and hear their story. The SP was then facilitated to identify any problems or issues that he/she would like help to address. If there was more than one problem, the NSMS asked the SP to identify the most pressing ones and focused on one or two of the priority problems. Firstly, realistic goals were identified and agreed between the SP and the NSMS. The NSMS then,

using skills such as active listening, probing and summarizing, assisted the SP to identify ways to manage the problem, using the key questions to consider for problem management (CD rom, SP guidebook, Figure 1, p.14) and the steps for problem-solving (CD rom, SP guidebook, Figure 2, p.16). If the SP did not have any problems at the time of delivery of the intervention, the NSMS discussed how the SP was managing and provided education and advice on how to go about managing a problem, if one arose. They drew on potential problems such as the need to find information or manage care needs of the PwMS. Each of the successive calls (call 2, 3 and 4) referred to issues discussed in the preceding call, progress with problem management was discussed and new problems identified and explored as appropriate. In advance of receiving the intervention calls, the SPs received the SPs guidebook and during the calls the NSMS referred to the guidebook. The support person guidebook was designed to facilitate the process and enable nominated support persons prepare for the calls from the nurse specialists. It consists of a self-evaluation problem checklist, worksheets to explore the problem identified, brief guidance on how to solve a problem, key questions to consider when problem managing and the steps to managing problems as in the COPE model. During call four, the NSMS explained that the fourth call was the last call as part of the intervention and advised all SPs that they could make contact with him/her in the usual way. Persons with ongoing problems were advised to keep in contact with the NSMS, as required. The manual including the support person guide book was subjected to content validation (see section 4.5.2 for details).

The intervention manual was then simulated by me and a colleague to determine how it could function in practice. Further simulation of the intervention was observed prior to the exploratory phase by recording a NSMS deliver the intervention on at least three occasions. This confirmed that it was possible to deliver the intervention, that all of the theories were helpful in its delivery and use by SPs, and that the intervention had the potential for positive outcomes for both PwMS and nominated support persons. During this phase, positive outcomes were reported by the PwMS and the SPs. These included the SP having someone to talk to and getting information that they required. The PwMS who participated in the post-intervention interview was pleased that the SP had somebody to talk to.

4.5.2: Content validity of the intervention

Content validity is the extent to which an instrument covers what it is supposed to measure (Yaghmaie 2003). The concept of content validity as applied to interventions is similar to content validation of measurement instruments (Bakas *et al.* 2009b) which suggests that it is the evaluation of the extent to which the content of the intervention is relevant to meeting the aims and objectives of the intervention. Content validity of the manual was determined by presenting the manual to a panel with four areas of expertise. They comprised a nurse educator, three NSsMS, a nurse with counselling expertise and three family members of PwMS. Family members were recruited via the MS Society and were asked to provide a detailed evaluation of the SP guidebook. All other expert members were asked to evaluate the entire manual. The experts were independently asked to rate the relevance of each item on the manual to the aims and objectives of the intervention, using a four point scale form, based on the recommendations of Waltz & Bausell (1981). The index of content validity (CVI), the proportion of items given a rating of 3 or 4 by raters, was then calculated. The CVI for this intervention manual ranged from 0.66-1, with the majority of items scoring a 1(see Appendix 9 and 10). This score was well above the unacceptable CVI level of 0.50 (Waltz & Bausell 1981), indicating a high level of content validity. Minor word changes were made to the manual using the comments on the CVI forms and feedback following the pre-exploratory phase (Appendix 10).

4.6 Intervention delivery

This section provides an overview of the process that I followed to provide training for delivery of the intervention, in accordance with the intervention manual. NSsMS were expected to comply with the best practice guidance in delivering telephone support (see CD rom, intervention manual, section 2, page 6). I provided training on how to deliver the intervention (full details provided in the intervention manual) and use the equipment. An on-going support system was set-up to ensure that the NSsMS were supported throughout the implementation phase (section 4.6.4).

4.6.1 Training of the interventionists

Each NSMS received a minimum of two hours one-to-one training. This was offered at a time and location of convenience to the NSsMS and close to the start of recruitment. All of the NSsMS requested on-site training. This enabled the training to be delivered at a time of convenience to the NSMS and for the equipment to be set up and tested. It also allowed for site specific issues and questions to be addressed. At least two weeks before the training, the NSMS received a copy of the intervention manual, support person guidebook and the accompanying training manual, and was asked to read the manuals prior to the training. During the training, I went through the key issues within the manual, in particular the models underpinning the intervention, the purpose of the intervention, the process of delivery, for example, how to use the equipment and the need to document the calls using the associated documentation. Each NSMS was offered the opportunity to role play the delivery of the intervention. Two of the NSsMS availed of this opportunity. The other NSMS felt that she was 'Ok' and could deliver it without the role play. As NSMS participation was voluntary, I was not in a position to insist that the NSsMS participated in role play. The NSMS in question, did role play the use of the equipment. During the training, I helped the NSMS with the installation and use of the equipment, such as the head set and call recording equipment. Advice was provided on how to set up for the calls and laminated prompt sheets were provided to assist with the application of the principles underpinning the intervention during the calls.

4.6.2 Sourcing call equipment

It was essential that the equipment selected was easy to set-up and resulted in minimal disruption to the NSMS current systems. The equipment also needed to be compatible with the telephone systems at the clinical sites and the recording equipment needed sufficient memory to avoid cut-offs or power outage during calls. Sourcing this equipment was a time consuming activity as it was difficult for me to source headsets with voice tubes for landlines. A relatively inexpensive headset and call recording equipment was eventually sourced.

4.6.3 Call organisation and set up

This necessitated communicating with the NSsMS and nominated support persons to agree a mutual time for call delivery/receipt. As the NSsMS had busy clinical schedules, their availability to deliver the calls was usually limited to one or two evenings a month. When the calls were due, I contacted the NSMS a week or two in advance to agree her availability. The NSMS was asked to offer at least two days that she could make herself available for the call. I made sure that I was available on those days to provide support to the NSMS, if required. Therefore, for efficient use of time for both the NSsMS and myself, I attempted to set all the calls for the same evening. Contact was then made with each participant to agree the day and time of the call. The agreed dates, times and call schedule were communicated to the NSMS by email using site specific code numbers. On the day the calls were due to be delivered, as a reminder of the time of the call, I sent an email to the NSMS and a text to the family member/caregiver.

4.6.4 Support systems

I was available for the NSMS to contact immediately before, during and after the calls should she require assistance or have difficulty making contact with participants. A flow chart was provided on how to manage challenges in making contact with SPs. Following the call, I confirmed with the NSMS that the call was delivered and logged

the call. The due date for the next call was then calculated. The NSsMS were colleagues working in different hospitals and communicated with each other on a regular basis. The NSMS from site one was the most experienced NSMS and the first to complete delivery of the intervention. She agreed to provide support to the other interventionists, if they wished to contact her.

4.7 Chapter summary

In this chapter, a focused literature review on the outcomes of problem-solving models and interventions used to guide the development of problem solving interventions was provided. The findings suggest that problem solving interventions have potential to improve outcomes for caregivers across a range of outcomes including physical health, burden, stress, depression, emotional tensions, social functioning and preparedness to care. The approach to identifying the most suitable model to guide the development and delivery of this intervention was described. In addition to the information obtained from the series of literature reviews in this and previous chapters, my clinical and educational expertise was instrumental in informing the development of the intervention. The intervention provides a framework that guides NSsMS in helping nominated support persons to manage problems and learn problem management skills. The theoretical foundations of the intervention are eclectic in that it incorporates a number of pre-developed models and frameworks including Egan's model of the skilled helper, the COPE model of problem-solving and Lewinian Experiential Learning Model. The intervention differs from current approaches to telephone support offered by NSsMS in that it provides a structured and pro-active approach to delivering their services. This intervention was tested to determine its potential to improve outcomes for nominated support persons of PwMS. Chapters 5 and 6 provide an overview of the methodological approach and methods adopted to test the intervention

Chapter 5: Methodology and research design

5.1 Introduction

The previous chapter provided detail on how the intervention for this study was developed. In this chapter, the research design, the methodology underpinning this design and the methods used for the preliminary test of the intervention are discussed. As the study tested a new intervention, the frameworks guiding the development and testing of complex interventions (Medical Research Council 2000, 2008) were instrumental in guiding the overall objectives of the study and influenced the methodology and research design selected to test the intervention (Corry *et al.* 2013). Early phase testing of a new intervention necessitates evaluation of its acceptability to clients and practitioners, along with determining the feasibility of outcome testing in the real world of practice (Medical Research Council 2008). The stage of intervention development and testing of this complex intervention, (i.e. a new intervention with no prior testing) limited this study to exploratory research, which consisted of pilot feasibility testing.

In this chapter the theoretical issues relating to the evaluation of complex interventions are explored, together with the philosophical and methodological considerations given to choosing the approach adopted in this study. Recommended approaches to evaluating exploratory trials are examined and those selected in this study are presented. The concept of pilot feasibility testing is explored along with the experimental research design, which forms the foundation for the randomised trial design. Variations in the randomised trial design, which is the design adopted to test the intervention developed, are examined together with justifications for decisions taken in this research study. Where appropriate, application within this study is explicated. The key principles underpinning the randomised trial are discussed prior to explicating their application within this study, in chapter 6.

5.2 Aims and hypotheses

The aims of this PhD research study were to:

1. Develop an intervention for use by nurse specialists in Multiple Sclerosis (NSsMS) to help people who support PwMS (SPs).
2. Test the feasibility and acceptability of the newly developed intervention to NSsMS, SPs and PwMS within a randomised trial.
3. Determine SPs' satisfaction with the intervention.
4. Evaluate the potential impact of the intervention in terms of measurable SP self-efficacy in problem management and obtaining respite, preparedness to care, quality of life and burden.

The following null hypothesis was tested in the evaluation of the intervention:

There is no difference in self-reported measurable self-efficacy in problem solving and obtaining respite, preparedness for supporting/caring, burden and QoL for support persons who participate in a nurse-led manual based telephone support intervention service when compared to support persons who do not participate in the intervention.

5.3 Theoretical and operational definitions

Self-efficacy: "...is concerned with people's beliefs that they can organise and execute courses of action to manage situations" (Zeiss *et al.* 1999, p. 222). For the purpose of this study, managing situations refers to managing problems encountered in caregiving and seeking respite as measured using the 4-item Self-Efficacy for Problem-solving scale (as used by Zeiss *et al.* (1999)) and the Self-Efficacy for Obtaining Respite subscale of the revised scale for Caregiving Self-Efficacy scale (Steffen *et al.* 2002).

Preparedness: "... [is] a caregiver's assessment of his or her readiness to provide care rather than to the adequacy with which he or she is providing care" (Schumacher *et al.* 1998, p. 66). This was measured using the 'Caregiver preparedness scale', an 8-item

subscale with a four point rating scale that was developed as part of The Family Care Inventory in the early 1980's (Archbold *et al.* 1990).

Burden: A support person's subjective perception of overload in one or all of the following domains: physical, psychological, social or financial, as measured using the caregiver reaction assessment (CRA) questionnaire (Forbes *et al.* 2007).

Quality of Life: "Individuals perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (World Health Organisation 1996, p. 5) as measured using the WHOQOL BREF instrument (WHOQOL BREF).

Satisfaction: is a "positive attitude" (Pascoe 1983, p. 185), where attitude is defined as a "...feeling of favourableness" (Fishbein & Ajzen 1975) towards the intervention, as measured using the Client Satisfaction Questionnaire-8 (Attkisson & Greenfield 1994)

5.4 Evaluating complex interventions

Complex interventions are "...multifaceted, organisationally elaborate, and socially mediated" (Munro & Bloor 2010, p.700). The outcomes are influenced by many factors including context (Wells *et al.* 2012). Complex interventions should be evaluated using the best available and most appropriate methods in order to determine how the intervention works and how effective it is in practice (Medical Research Council 2008). The approach adopted is influenced by the degree of control the researcher has over the intervention and its delivery and the ethical implications of randomisation or withholding of an intervention. When assessing an intervention for effectiveness, a design that includes randomisation should be considered to prevent selection bias, which negatively impacts on the validity of outcomes (Medical Research Council 2008). In studies where randomisation is not possible, observational studies may be employed, although the reliability of the findings of such studies might be more limited (Medical Research Council 2008). The choice of design, therefore, depends on the

research question, the purpose of the evaluation and the practicalities of carrying out the most robust approach.

As the aim of this study was to explore and test the feasibility of a new intervention in order to generate evidence to support its use or otherwise, current evidence based implementation methods such as action research (Bellman 2003), were unsuitable. The randomised trial design was selected to test the intervention in this study, as it is the gold standard method for testing the effects of different interventions (Sackett *et al.* 1997, Bellman 2003, Hodgson *et al.* 2007). It is the only design that allows for the balancing of unknown prognostic factors at baseline (Stolberg *et al.* 2004), thus minimising the influence of these factors on the outcomes (Stolberg *et al.* 2004). Like other designs, randomised trials are limited in terms of their ability to identify safety in that they are not always of sufficient size or duration to detect adverse events (Yazici 2008). Despite this, early phase testing gives some indicator of safety and feasibility, prior to progressing to full scale intervention testing.

5.5 Philosophical assumptions underpinning this study

Positivism is based on the scientific approach where the researcher is required to set aside their pre-conceived notions and collect data in an objective and replicable manner that is free from researcher bias. Researchers operating from this paradigm aim to develop general laws (McEvoy & Richards 2006, Parahoo 2006, Weaver & Olson 2006) that predict what is happening in the world (Weaver & Olson 2006). As a result of philosophical arguments that reality can never be fully known (Weaver & Olson 2006) and an extensive critique of positivism, post-positivism emerged in the 1980s (Giddings 2006). Although the ontological and epistemological underpinnings of post-positivism are broadly similar to that of positivism, they differ in that post-positivism sets about to falsify hypotheses as opposed to testing hypothesis and replicating findings (Weaver & Olson 2006). Like positivism, post-positivism aims to generate findings that can be generalised (Parahoo 2006) and reduces humans to component parts that can be measured using numerical scores (Weaver & Olson 2006).

Intervention testing within an exploratory trial is entrenched in the post-positivist paradigm because it requires the adoption of approaches and methods that fit both the randomised trial and the concurrent process evaluation. This requires the use of more than one method of data collection. This trial component which is based on the experimental research design, sets out to falsify the hypothesis which is formulated in advance (Parahoo 2006). In this study, I set out to evaluate the potential value of a research design that aims to falsify the research hypothesis using objective and numerical outcome measures. Having identified that the support service offered to family members/carers of people with MS was unstructured and lacked evaluation, an attempt was made in this study to address this gap by developing an intervention to provide a structured pro-active problem management support intervention. The intervention was tested using an experimental research design, which is a rigid and objective methodological approach. The outcomes of the support service were primarily evaluated using structured self-reporting questionnaires. Using outcomes that cannot be observed and are evaluated using self-reporting, is congruent with the underpinnings of post-positivism (Parahoo 2006). Although I collected the views and opinions of stakeholders and participants using semi-structured interviews, a method usually attributed to the interpretative paradigm, this was done within the overall framework of the post-positivist paradigm. Thus, a multi-method approach was adopted within the overall framework of the post-positivist paradigm.

5.6 Early phase intervention testing

According to Munro & Bloor (2010), the traditional method of testing using the randomised trial is limited in terms of explaining why an intervention, did or did not produce the desired effects. They argue that this lack of explanation, especially when the intervention is found to be ineffective, is unfortunate as the researcher will not know why it was ineffective. It is therefore, important to do preliminary testing to determine how the intervention will unfold in the practice setting and make the necessary refinements to the intervention and the study protocol before full scale testing (Medical Research Council 2000, 2008). Early phase testing allows the researcher to test the

intervention to determine its potential to bring about the desired change, how best to deliver it during full phase testing and provides an opportunity to experiment with different approaches. During this phase, modifications can be made to the study protocol that might not be acceptable at later stages. For example, the researcher can alter the components of the intervention, alter the mode of delivery, the intensity or frequency of delivery, the interventionists, the outcome measures and the context for delivery.

Early phase exploratory testing may be limited to acceptability and feasibility testing or can extend to include pilot testing. Testing following feasibility or pilot exploratory work is usually a much larger and more costly exercise (Whittemore & Grey 2002). The exploratory phase precedes the definitive trial (Medical Research Council 2000) and is important because the findings have implications for the success or failure of a full scale trial. The exploratory phase findings may deem larger scale testing to be impractical because the cost might outweigh the potential benefits or likelihood of success. It is common during this phase to test for feasibility by incorporating a process evaluation (Medical Research Council 2008) alongside the design chosen to test the intervention, as it provides "...insight into why an intervention fails unexpectedly or has unanticipated consequences" (Medical Research Council 2008, p.12). A process evaluation helps to inform researchers why an intervention works or not and whether to proceed to a full scale trial. It might also suggest ways to modify the intervention or the evaluation process (Munro & Bloor 2010). The intervention developed for this study was tested using a pilot study and a concurrent process analysis was carried out to determine acceptability and feasibility.

5.6.1 Acceptability

Acceptability refers to the extent to which those who deliver and receive the intervention perceive it as appropriate. It is determined by the extent to which it is viewed as agreeable or favourable (Sidani & Braden 2011). It can be assessed before, during and after intervention delivery using both quantitative and qualitative methods (Ascherio & Munger 2007). Approaches to determining intervention acceptability vary,

with published papers generally failing to provide an operational definition of the concept. Satisfaction with the intervention is a frequently cited indicator of acceptability (Hosek *et al.* 2011, Dombrowski *et al.* 2012, Juraskova *et al.* 2013). Acceptability and satisfaction are however distinct concepts, although they are related in terms of social desirability, that is, the extent to which an intervention is desired, valued, considered useful and important (Sidani & Braden 2011). Satisfaction relates to the extent to which participants believe that the intervention is effective in addressing issues of concern and is likely to influence adherence and continued use of the intervention (Sidani & Braden 2011). In addition to satisfaction, researchers also use indicators such as participation rate (Zeltzer *et al.* 2002, Pincus *et al.* 2013), intervention tolerance (Zeltzer *et al.* 2002) or use (Habib *et al.* 2013), comments on the materials (Dombrowski *et al.* 2012) and relevance of content and activities (Hosek *et al.* 2011), logs recorded by interventionists and focus group (Hosek *et al.* 2011), ongoing and post-intervention interviews with stakeholders (Hosek *et al.* 2011, Iribarren *et al.* 2013, Pincus *et al.* 2013) and researcher designed rating scales to rate satisfaction and acceptability (Juraskova *et al.* 2013, Zarit *et al.* 2013).

Reports vary in the detail provided in particular regarding researcher designed scales, with Juraskova *et al.* (2013) providing scant detail while Zarit *et al.* (2013) was more specific by indicating the focus of the scale 'perceived helpfulness' and the scoring range of 0 'not very helpful' to 3 'very helpful'. Sidani & Braden (2011) express caution about the use of indicators such as enrolment, retention/attrition rates and adherence or continued use of an intervention for evaluating acceptability. Such indicators can only be used if designed and tailored to assess acceptability as part of the study design. According to Sidani & Braden (2011), enrolment is only a good indicator when evaluated relative to the potential participants' overall understanding of the intervention. They highlight that retention/attrition is influenced by many factors other than the acceptability of the intervention and should be used only if explicitly linked to the reason for remaining in a study or not. Evaluations of acceptability are made on the basis of beliefs about health and issues of importance to clients, perceived understanding of the intervention and awareness of treatment alternatives (Sidani & Braden 2011). Participants and interventionists therefore need education on such

influencing factors in order to make judgements on acceptability before experiencing the intervention.

5.7 Feasibility and pilot

5.7.1 Feasibility studies

Feasibility studies focus on the practical aspects of implementing the intervention (Arain *et al.* 2010, Sidani & Braden 2011) and in some cases, the study design. They are important for determining whether the intervention can be delivered as planned. Failure to deliver the intervention as planned can lead to a conclusion that an intervention is ineffective when the lack of effect is actually because the intervention was not implemented correctly (Sidani & Braden 2011). Feasibility work relates to the technical and practical aspects of the study and is achieved by establishing acceptability of the intervention and the likelihood of being able to test the intervention fully. The extent of feasibility work varies from testing how realistic aspects of the process are, to the inclusion of specific elements, such as a cost analysis (McConnell 1998). Feasibility studies do not have to resemble the main study, do not need a primary outcome, power calculation or control group (Arain *et al.* 2010). Their main function is to inform the practical aspects of implementation of interventions in the full scale study, such as the availability of resources, the identification of factors that interfere with implementation, such as client and interventionist reaction to the intervention, fidelity to the intervention and, where appropriate, the comparator, context and ability to reach the target population and clarity of the protocol (Sidani & Braden 2011). They can also be used to estimate important parameters for the design of the main study (Arain *et al.* 2010), including degree of co-operation from stakeholders. This might include willingness of participants to be randomised and to adhere to the study requirements, clinicians to recruit, follow-up rates, outcome measures including their standard deviation rates, the number of eligible participants, response and follow-up rates (Arain *et al.* 2010). The feasibility stage may include a concurrent pilot study to establish if the full-scale trial is workable (McConnell 1998).

5.7.2 Pilot studies

Writers differ in their conceptualisation of a pilot study. Arain *et al.* (2010) argue that a pilot is a small version of the main study to determine if the methods of recruitment, randomisation, follow-up assessment and other protocol processes run efficiently. The Medical Research Council (2008) guidelines discuss these two aspects of intervention testing together and argue that a "A pilot study need not be a 'scale model' of the planned mainstage evaluation, but should address the main uncertainties that have been identified in the development work" (p.10). Pilot studies can therefore incorporate feasibility work or be stand alone following feasibility testing. In this study the pilot study was a small version of the main study which included a concurrent process analysis to determine feasibility.

As the objectives of my study included testing the feasibility of both the intervention and study design, a Phase 2 exploratory study was done. This included both pilot and feasibility testing using an overall process evaluation based on the work of Linnan & Steckler (2002) (Section 6.2.17 provides an overview of the process evaluation). A randomised trial was chosen because it is based on the experimental research design, the design most suited to testing the effects of an intervention.

5.8 The experimental research design

The uniqueness of the experiment lies in its ability to provide explanation (Davis & Rose 2000) and it is the best method for the identification of causal relationships (Moses & Knutsen 2012). Experiments are used to determine the outcomes of interventions by testing hypotheses that predict cause and effect relationships between variables (Parahoo 2006).

An intervention is an essential ingredient for experimental research designs (Parahoo 2006) and, together with a control for comparison and random assignment, allows the researcher to generate strong evidence about the relationship between the intervention (the independent variable) and the outcomes (dependent variables) (Moses & Knutsen

2012). Control, random assignment and manipulation therefore underpin the experimental design and, when these studies are carried out in the healthcare environment they are referred to as clinical trials (Parahoo 2006). Clinical trials are defined as "Any research project that prospectively assigns human subjects to intervention and comparison groups to study the cause and effect relationship between a medicinal intervention and a health outcome" (DeAngelis *et al.* 2005, p. 2927). Medicinal interventions include treatments or interventions designed to bring about changes in care or behaviour (DeAngelis *et al.* 2005).

Clinical trials can be categorised as explanatory and pragmatic (Torgerson & Torgerson 2008). Pragmatic trials are designed to measure the benefit of the intervention in routine practice (Roland & Torgerson 1998). Explanatory trials are not designed to be similar to routine care (Torgerson & Torgerson 2008) or administered under normal practice conditions (Schwartz & Lellough 1967). In this study, a pragmatic design was chosen because context is a key factor influencing outcomes in trials of complex interventions (Medical Research Council 2008, Wells *et al.* 2012). The design of a clinical trial is determined by factors such as the variables to be tested, the availability of participants and the need to minimise bias. Bias refers to the potential for factors other than the intervention to bring about the desired changes. Designs available include the post-test only, the pre-test post-test, single subject/single case design, quasi experimental, the true experiment (the randomised trial) and variations on this (Polit & Beck 2012). A number of these designs were considered for the testing of this intervention, but the randomised trial was selected because of its advantage over the others in minimising the risk of bias.

5.8.1 The randomised trial

In clinical research, the experiment is termed a randomised trial, which is a clinical trial that tests at least two alternative approaches to treatment. The randomised trial is based on the experimental design and enables researchers to isolate an intervention, test it against a comparator by random assignment of participants to one or other of the groups and measure the outcomes using pre-determined and bias free outcome

measures (Meinert 2012). Participants are allocated to one of the forms of care using methods that are free from bias (i.e. randomisation) and the intervention is evaluated using pre-determined outcome measures (Meinert 2012). The randomised trial is recognised as the gold standard for comparative research (Sackett *et al.* 1997, Lindsay 2004, Akonbeng 2005, Torgerson & Torgerson 2008). On the hierarchy of evidence for the effects of interventions, it is second only to systematic reviews of randomised trials (Akonbeng 2005). It is the only research design that allows researchers to "balance known and possibly unknown factors" that may affect research outcomes (Watson *et al.* 2002, p.132). Findings from designs other than the randomised trial can generate 'misleading results' (p.7) which have led to the implementation of harmful practices (Torgerson & Torgerson 2008, p.7).

In order to have a high level of confidence that the findings are due to the intervention and not some other factor, it is essential that mechanisms such as the use of a control group, manipulation of the intervention and random assignment of participants are applied (Schneider *et al.* 2003). Manipulation, control and randomisation are the hallmarks of the true experiment (Schneider *et al.* 2003).

Manipulation is a strategy used in randomised trials to expose the treatment group to the intervention while the other group, the control group, does not receive it (Schneider *et al.* 2003). By ensuring that all variables that could impact on the outcome, other than the intervention, are balanced across the groups, the researcher can attribute the findings to the intervention. This can be achieved using randomisation to determine which participants are allocated to the intervention or the control group. The purpose of a control group is to neutralise the effects of factors other than the intervention when the effects of the intervention are being estimated. The type used is determined by factors including practical and ethical concerns (Polit & Beck 2012).

Random allocation of participants to the intervention or control group is the cornerstone of the randomised trial (Kendall 2003), because it rules out the effects of systematic bias (Schneider *et al.* 2003) and is essential for internal validity (Torgerson & Torgerson 2008). It involves the allocation of participants to the groups on a random or chance basis (Schneider *et al.* 2003), thereby ensuring that each person has an equal chance of being assigned to the intervention or control group (Davis & Rose 2000,

Akonbeng 2005). This balances known and unknown variables (other than the intervention), which may have an effect on the outcome between the control and the intervention groups (Torgerson & Torgerson 2008).

The application of manipulation, control group and randomisation means that researchers can have confidence in attributing cause and effect to the intervention as other possible explanations (Topping 2007) such as selection bias, regression to the mean and temporal effects (Torgerson & Torgerson 2008) are outruled. However, it is still possible that the detected effect is due to chance. This potential can be minimised through the inclusion of a sufficiently large number of participants.

5.8.1.1 Sample size estimation

One purpose of the estimation of the sample size for a Phase 2 (exploratory) trial is to obtain a sufficient number of participants to inform the recruitment rate and protocol refinement for a full-scale, Phase 3 trial, where adequate power is essential for determining the effects of the intervention. Determining these effects is not the primary objective of an exploratory study, but is the primary objective of a Phase 3 trial (Stallard 1998). The objectives of the study for this doctoral research are in line with the overall aims and objectives of Phase 2 trials generally, and can therefore be achieved with or without a sample that is sufficiently powered to detect the true effects of the intervention (Stallard 1998). When determining a sample size, an estimate of the expected difference between the two groups must be made and a statistical significance level decided upon. The expected difference between the two groups can be informed from previous research about the target population (Proctor & Allan 2007). The sample size for a feasibility study should be large enough to allow for estimates of important parameters in future studies (Arain *et al.* 2010, Pincus *et al.* 2013) and to achieve this a minimum of 30 participants in each arm has been recommended (Pincus *et al.* 2013). Estimating important parameters in full scale intervention studies is important for ensuring that the sample size is large enough to detect a difference between the intervention and control group, if such a difference exists (Breakwell *et al.* 2000). It is unlikely that significant differences will be detected with small sample sizes

(Breakwell *et al.* 2000, Parahoo 2006), unless the effect is very large (Parahoo 2006). This can lead to failure to detect a real difference where one exists: a Type 2 error (Jones *et al.* 2003, Proctor & Allan 2007, Banerjee *et al.* 2009). The probability of finding a significant difference due to chance, when there is truly no difference between the interventions is known as a type 1 error (Proctor & Allan 2007, Banerjee *et al.* 2009). The likelihood of these errors occurring can be reduced by increasing the sample size (Banerjee *et al.* 2009). It is however important to avoid recruiting more participants than required to detect a difference as to do so is unethical (Parahoo 2006) and costly (Proctor & Allan 2007).

Using critical parameters such as anticipated recruitment rate (Arain *et al.* 2010), and measures of central tendency and variability (Lancaster *et al.* 2004) obtained from prior research or feasibility work, the required sample size can be calculated. To avoid a Type 1 error, a cut-off point (α) at which the null hypotheses is rejected is set in advance (Banerjee *et al.* 2009). The cut-off point depends on the seriousness of the effect of the treatment. For example stringent cut-off points are used for drug testing, whereas in studies where the treatment is less likely to be detrimental to the recipient less stringent ones can be used (Polit & Beck 2012, LoBiondo-Wood & Haber 2014). A significance level set at 5% means that an effect of the size detected or greater will occur by chance one time in twenty when there is truly no difference between the interventions. To avoid a type 2 error, a decision is made regarding willingness to accept missing an effect when one exists. This risk, β, is usually set at 0.1 (10%) or 0.2 (20%) and this is used to calculate power (1- β) (Banerjee *et al.* 2009). Most studies use either 90% or 80% power (Proctor & Allan 2007).

5.8.1.2 Internal validity

Internal validity is the extent to which the differences between the intervention and control group is due to the intervention and not some other confounding factor (Sedgwick 2010). Although the application of stringent measures of control, manipulation and randomisation minimises the risk of bias, a number of threats to internal validity remain. These include testing effects (Schneider et al. 2003), resentful

demoralisation, the hawthorne effect (Torgerson & Torgerson 2008) and attrition bias (Watson *et al.* 2002, Torgerson & Torgerson 2008). The impact of these threats to a study can be lessened through careful design of the study (Torgerson and Torgerson, 2008).

Testing effects can be minimised using, where possible, different forms of the same test, as it reduces the effect of prior exposure to the same instrument (Schneider et al. 2003). A variation of the true experiment such as the Solomon 4 design or the after only design also helps reduce testing effects (Schneider *et al.* 2003). The Solomon 4 design involves randomly assigning participants to 4 groups, with 2 groups given a pre- and post- test and the other two groups given a post-test only. When pre-testing is unacceptable or there are insufficient participants to allow the use of the Solomon 4 design, the post-test design is the only option available to the researcher (Schneider *et al.* 2003). As this was an exploratory trial and recruitment rates were unknown the effects of pre-testing were not considered.

Resentful demoralisation occurs when participants have a preference for one treatment over the other and are not randomly assigned to their preferred treatment (Torgerson & Torgerson 2008). This may impact on outcomes because treatment effects can be exaggerated or diluted (Torgerson & Torgerson 2008). Treatment contamination can also occur if participants who are not assigned to their preferred group seek alternative treatments or fail to comply with the study protocol (Torgerson & Torgerson 2008). In this study, SPs preferences were determined at consent so that they could be considered during the interpretation of the findings. In addition, during final data collection SPs were asked to identify any other supports received or training attended since the start of the study.

Attrition bias can also impact on the internal validity of a trial by changing the direction of the effects, even when the rate of attrition is equal across study groups (Torgerson & Torgerson 2008). It may lead to incorrect conclusions regarding the effectiveness of a treatment (Torgerson & Torgerson 2008). It occurs when there is "... inappropriate handling of dropouts, withdrawals and protocol violations" (Watson et al. 2002, p.133). The impact might be reduced by making adjustments for known covariates such as age and gender at analysis, however, in situations where covariates are unknown such

adjustments are not possible (Torgerson & Torgerson 2008). Maximising participant retention (see section 6.2.9) is the best method of managing attrition bias (i.e. by preventing it). Furthermore, an intention to treat analysis (ITT) can be used (Watson *et al.* 2002, Torgerson & Torgerson 2008). This requires the retention of participants for outcome measurement even if they have not received the full treatment and analysing their outcomes in the group to which they were originally assigned. While the use of ITT may lead to dilution of treatment effects, it avoids an outcome that is due to attrition bias rather than the effects of the intervention (Torgerson & Torgerson 2008). In this study participant retention was maximised by the use of a research incentive and an ITT approach to data analysis was used.

The research setting has a great influence on the risk to the internal validity of the study. Field researchers have less control over the environment and study conditions than laboratory researchers, with a higher probability of study contamination due to factors beyond the control of the researcher (Schneider *et al.* 2003). This is further complicated by the fact that many interventions in the practice setting are complex, in that they comprise a number of different components all having a potential impact on the outcomes. Examination of the effect of different components may require the use of multiple intervention designs in the same study (Polit & Beck 2012). In order to determine the impact of the environment in this study, a process analysis that examined the influence of context was carried out.

5.9 Intervention fidelity

Intervention fidelity is the extent to which the intervention is delivered in a consistent manner (Bellg *et al.* 2004, Mars *et al.* 2013) and in accordance with the intervention trial protocol (Gearing *et al.* 2011, Mars *et al.* 2013). Monitoring adherence is important as it determines the extent to which the intervention was delivered in accordance with the intervention protocol and can provide explanations for the outcomes, such as intervention effectiveness or ineffectiveness or implementation failure (Mars *et al.* 2013). The extent to which the intervention protocol was adhered to, therefore, determines the extent to which the outcomes can be attributed to the intervention,

rather than some other variable (Bellg *et al.* 2004, Borrelli 2011). Failure to implement the intervention as intended leads to variations that weaken the effects of the intervention (Waller 2009) and this decreases the likelihood of detecting outcomes that are of statistical significance (Bellg *et al.* 2004, Resnick *et al.* 2005). It is therefore important that strategies are in place to optimise intervention fidelity. These include minimising and monitoring contamination, selection and standardisation of training to the interventionists, standardisation and monitoring the delivery of the intervention, monitoring receipt of the intervention and the ability of participants to use the skills (Bellg *et al.* 2004, Resnick *et al.* 2005, Mars *et al.* 2013). Interventionist training is key to optimising intervention fidelity. This means providing similar training to all interventionists and evaluating their knowledge and skills against pre-defined performance criteria (Bellg *et al.* 2004). This will facilitate the standardisation of the delivery of the intervention through adherence to the intervention protocol, while allowing for interventionist differences. It is however, important to minimise what Bellg *et al.* (2004) refer to as drift in the interventionists' skills. This can be avoided by ongoing monitoring of the delivery of the intervention and ongoing training. The ability of participants to use the skills can be optimised by ensuring that they understand the content of the intervention and that they are able to use the information or skill. The strategies used to optimise intervention fidelity in this study are discussed in section 6. 2.15.

5.10 Conclusion

Early phase intervention testing allows for understanding of how the intervention works in practice and for adjustments to be made to the intervention and study protocol prior to full-scale testing. The approach used to evaluate a complex intervention depends on the degree of control the researcher has over the intervention and its delivery. The experimental design is at the heart of the randomised trial and was selected for the testing of the intervention for this study because of its superiority over other designs in ruling out the effects of possible confounders. The experimental design is the only design that allows for the testing of hypotheses that predict cause and effect

relationships. It is, however, dependent on the researcher's ability to control for threats to internal validity including resentful demoralisation, attrition bias and maintaining intervention fidelity. The key components of a randomised trial which includes the population, sampling, sample size, eligibility criteria and randomisation are explored in chapter 6. The application of these components to this study along with the methods used to examine the feasibility of the intervention and study are presented.

Chapter 6: Methods

6.1 Introduction

In this chapter, the research methods used, their application to this exploratory trial and the concurrent process analysis are presented. Firstly, a summary of challenges encountered during the pre-exploratory stage of this trial and how they informed the protocol for the study, is presented. The recommended approaches to the selection of the population, sampling, recruitment and retention, randomisation, data collection and analysis within an exploratory randomised trial are examined along with their application to this study. This is followed by an overview of the approach adopted in this study to the monitoring of intervention fidelity and determining overall acceptability and feasibility of the intervention and study design. This chapter will conclude with an overview of trial registration, quality control, gaining ethical approval and trial management.

6.2 Methods

The philosophical underpinnings of the randomised trial permeate all aspects of the study and inform the approach to population and sample selection, subject group allocation, data collection, analysis and interpretation of the findings. The methods adopted during the exploratory phase were modified in accordance with the lessons learned during the pre-exploratory phase. The pre-exploratory phase was a trial run of the exploratory design to model the delivery of the intervention in the practice setting, evaluate recruitment methods and identify other factors that may impact on the exploratory phase of the study.

6.2.1 The pre-exploratory phase

Prior to embarking on this exploratory study a pre-exploratory pilot was carried out using the same methods as outlined for the randomised trial for this exploratory study. Recruitment commenced on the 20th June 2011 and ceased in December 2011. This phase of the study highlighted that the main challenge was related to the method used for recruitment and participant response rate.

Study invites containing information packs for the PwMS and their nominated support persons were handed out on attendance at the OPD. There were challenges with the distribution of the packs as the NSMS was not always available to distribute them. Using the OPD as the method of recruitment was dependent on the NSMS handing out the information packs to potential participants. When the NSMS was absent for any reason, packs were not distributed. This meant that some potential participants did not have the opportunity to consider joining the trial. In addition, the NSMS felt that the full information pack may be overwhelming for the PwMS especially at an outpatient department (OPD) visit when they were most likely to be stressed and had limited time to discuss the study with the NSMS due to her work commitments and the PwMS's travel times (as many travelled long distances). The PwMS information pack also contained a full information pack for the nominated support person. As there were a number of aspects to the study, the information sheet contained a large amount of detail. On reading the information pack, some participants may have felt that it was very time consuming and therefore decided not to participate.

In addition to the challenge with the distribution of the study invites and information packs, the response rate was very low (by 3rd August 10 packs were distributed with no response and only 2 responses received by September 2011). One of the respondents did not fulfil the inclusion criteria (6.2.5.1). The SP of the second respondent was willing to take part in the study but stated that the PwMS had difficulty with the personal nature of the study. This may have been related to the nature of the PwMS's relationship to the SP as they were in a relatively new personal relationship. By early November 2011 34 full information packs were distributed with 2 further responses. In consultation with the primary academic supervisor for this study, a decision was made

to trial the use of a letter of invite at the OPD and seek authorisation to use database recruitment. Three further statement of interest forms were returned in late November 2011 and by December 2011, three participants were recruited. This phase of the study highlighted that database recruitment using a letter of invite and statement of interest form was more acceptable to PwMS at that site. It also demonstrated that participants were willing to be randomised and provided an opportunity to model the intervention in the real world of practice. Recruitment for this phase of the study closed in December 2011. Intervention delivery and data collection continued until April 2012 when all 4 intervention calls and outcome data were collected from the 3 participants recruited. Analysis of the findings demonstrated that it was possible to deliver the intervention in practice and that there were benefits for both the SP and PwMS. The PwMS was happy that the SP had an independent person to discuss areas of concern with. The SPs reported that it was helpful to have the opportunity to discuss areas of concern with the NSMS and found the intervention calls helpful. The changes made as a result of the lessons learned from this phase of the study are presented in section 6.2.6.

6.2.2 Population

The population is the group of people that the researcher is interested in studying (Breakwell *et al.* 2000, Polit & Beck 2012). The population under study is known as the theoretical population (Parahoo 2006) and includes those who are and those who are not easily accessible to the researcher. In determining the population, the eligibility (inclusion) and exclusion criteria (Polit & Beck 2012) need to be defined. These criteria are usually based on consideration of how much the resulting sample will represent the population, but criteria such as costs, practical constraints, people's ability to participate in a study and design considerations such as the need for a homogenous population to control confounders also need to be considered (Polit & Beck 2012).

The target population is the population to which the researcher wishes to generalize the findings to (Topping 2007). Access may not be possible to the entire population of interest. The sample may have to be drawn from the accessible population (i.e. those accessible to the researcher for the study who reflect the characteristics of the target

population) (Polit & Beck 2012). This is known as the study population (Topping 2007). The target population for this study is all people who support people with multiple sclerosis, hereafter referred to as 'support persons' (SPs). Support persons are those individuals nominated by PwMS whom they identify as providing the most emotional or physical support for them and can be a partner or spouse, other family member or carer. The study population comprised of the PwMS and SPs at the study sites. The sites selected were sites of convenience where NSsMS volunteered to participate. Five of the nine neurology centers where NSsMS were in post expressed interest in the study. The original intention for this research was that the study population would be all SPs of PwMS attending the outpatient clinic in one large teaching hospital in Ireland. However, due to participant recruitment challenges as outlined in section 6.2.6 during the pre-exploratory phase of this study, the study population was extended to all support persons of PwMS registered in three neurological outpatient clinics in teaching hospitals in Ireland, where neurological nurse specialists were employed. A total of 3070 PwMS were registered in the three clinical sites for this study [(site one (n=390), site 2 (n=2000) and site 3 (n= 680)].

6.2.3 Sampling

A sample is a subset of the population and sampling is a method of selecting elements of the population (Polit & Beck 2012). In a randomised trial, the aim is to select a group of people within which the relative difference in the effects of the intervention versus control is likely to be applicable to the wider population. Representativeness is, therefore, a key consideration but is difficult to achieve in practice, in particular when carrying out randomised trials. True representativeness can only be achieved with random selection of elements based on a pre-specified probability that an element will be included in the study (Polit & Beck 2012), and the use of probability sampling techniques, for example, simple random, stratified random, multi-stage cluster and systematic random sampling (Proctor & Allan 2007), which are designed to eliminate sources of bias (McEvoy & Richards 2006, Polit & Beck 2012). This requires the use of a sample-frame which comprises a list of all the elements of the accessible population (Proctor & Allan 2007). It may, however, be difficult, if not impossible, to construct a

sample frame (Fife-Schaw 2000) and in some instances (as in researching sensitive topics), it may not be desirable (Parahoo 2006). This prohibits the use of random sampling methods and limits generalization of the findings beyond the group studied. Generalisation of findings are further restricted when researchers are limited to the use of convenience sampling methods (Proctor & Allan 2007).

The nurse specialists were recruited using convenience sampling. The chairperson of the Irish Network of MS nurses was initially approached to determine overall interest from the nurse specialists. Nine NSsMS were invited to engage in early phase discussions during the initial development of the study. Of the nine NSsMS, five engaged in early discussions and supported my funding application and the five NSsMS indicated interest in being involved in the testing of the intervention. Ethical approval was obtained for five sites and medical access was secured in four of these sites. Initial access proved challenging because of the novel nature of the research, which required a time commitment from the NSsMS over and above that of usual studies carried out within nursing in the Republic of Ireland (ROI). Four NSsMS consented to participate in the study and deliver the intervention. One medical consultant denied access to PwMS and consequently that site was not used for the testing phase of the intervention. The NSMS at that site did participate in the pre-intervention interview, which informed the development of the intervention. The participants in this study comprised NSsMS, PwMS and SPs. PwMS and SPs were selected from sites of convenience where the NSsMS had indicated interest in partaking in the study. The target population of SPs for this study was all support persons who support PwMS registered at these clinics. In addition to the use of sites of convenience, the nature of the population for this study meant that it was impossible to construct a sample frame and employ a random sampling technique. For ethical reasons, the main study participants (nominated support persons) had to be invited via the PwMS and the number of PwMS who had a SP was unknown. It was also possible that some PwMS may not have given the invitation to an eligible SP. Table 8 depicts the participants and their role in this study.

Table 8: Participants and their role in the study

Participants	Their role
Nurse Specialists in Multiple Sclerosis (NSsMS)	Interventionists Participated in an interview prior to delivery of the intervention to inform the development of the intervention and to identify usual care (n=4) Participated in a post-intervention interview for the evaluation of the intervention (n=3).
People with Multiple Sclerosis (PwMS)	Nominated a support person Provided quantitative data (on the impact of MS and their quality of life). to contextualize the study findings Fifteen PwMS whose SPs were assigned to the intervention participated in a pre-intervention interview to inform usual support from the nominated support person. They also participated in a post-intervention interview to inform the evaluation of the intervention in terms of its potential benefit to them (PwMS) and their SP and to identify risks, if they existed
Support persons (SPs)	Participated in the trial component of the study. Completed outcome quantitative data at three time points. Those assigned to the intervention group received the intervention and participated in a short telephone interview to inform the evaluation of the intervention.

6.2.4 Sample size

Due to the exploratory nature of this study, the sample size was chosen to determine the likelihood of recruiting a powered sample in a phase 3 study. The preliminary power analysis was based on the Caregiver Preparedness Scale, because published estimates of critical parameters were available (Finlayson *et al.* 2009a). Based on an alpha level of 0.05, a power of 80% and a previously reported effect size of 0.55 (Finlayson *et al.* 2009a), the required sample size without loss of any follow up would be in the region of 53 participants per group, for a one-tailed (directional) hypothesis and 64 participants per group, for a two-tailed (non-directional) hypothesis. A minimum sample size of 100 participants in each of the intervention and control group was decided upon to allow for loss to follow up.

As discussed in chapter 5 (5.8.1.1) adequate power is essential to determine the effects of the intervention in a full-scale trial. As this was a new intervention and there were no published trials from which parameters for the primary outcome measures could be obtained, the parameters from this exploratory trial were used to calculate the sample size for a full-scale trial. The statistical power analyser G*Power http://www.gpower.hhu.de/ was used as it computes the statistical power for a number of tests including the ANCOVA F-test (Faul *et al.* 2007). To calculate an ANCOVA F test using G*Power the power (1-beta) and significance level must be set in advance. In addition, the number of groups, number of covariates, effect size and degrees of freedom (df) must be specified. For this power calculation, alpha was set at 0.05 and power 80. The parameters obtained for the preparedness to care questionnaire (effect size (0.35) were used giving a total minimum sample size of 320 participants (160 per group). This population represents 4.5% of the estimated 7,000 known suffers of PwMS in Ireland.

6.2.5 Eligibility criteria

Nominated support persons were the target population for this study. As the number and details of the support persons at all sites were unknown they could not be

approached directly. The only method of identifying participants was via the PwMS. This, along with the fact that it was likely that some discussion would centre on the care of the PwMS, it was essential that PwMS were approached to partake in the study before contacting support persons. As the intervention was delivered by the NSsMS supporting the PwMS, participants had to be recruited via the participating neurology sites. Therefore, PwMS who attended the participating outpatient department (OPD) clinics were approached and invited to take part in the study.

6.2.5.1 Inclusion criteria

Due to the novel nature of this intervention, a lack of prior testing and limited resources, those most vulnerable were excluded, in the interest of participant safety. I did not have the resources or expertise to manage situations that may arise for children who support PwMS or those with mental health issues. At one site, I had to provide and demonstrate resources to fund psychological support for participants, if required. Therefore, PwMS with severe cognitive impairment or those < 18 years were not included. As the intervention targeted SPs, those SPs with severe mental illnesses were also excluded. In order to identify whether the intervention may be most useful to support persons of people at different stages of progression of MS, no limitations were applied in terms of the type of MS diagnosed. The following criteria were applied:

PwMS

- People with a diagnosis of MS and who were at least > 18 years of age.
- PwMS whose diagnosis did not include a severe cognitive impairment.
- PwMS who nominated an adult support person (> 18 years of age).

Support persons

Informal adult (> 18 years of age) support persons of PwMS who did not have a serious mental health problem and could be contacted by telephone.

Nurses

- Nurse Specialists caring for PwMS

6.2.5.2 Exclusion criteria:

PwMS

- PwMS who could not nominate a support person.
- PwMS who were already engaged in similar research.
- PwMS who were diagnosed with cognitive impairment.
- People with a diagnosis of MS who were < 18 years of age.

6.2.6 Recruitment and site specific challenges

Sample recruitment is important for all research but particularly so for randomised trials, as studies that fail to recruit sufficiently large samples are at risk of failing to detect clinically important outcomes and lead to findings that are of limited value to patients or practitioners in clinical decision making (Mapstone *et al.* 2002). The difficulty of recruiting to randomised trials has been highlighted by several researchers (Watson & Torgerson 2006, Rendell *et al.* 2009, Gul & Ali 2010, Treweek *et al.* 2010), with up to 60% of trials running into difficulties and failing to finish on target (Treweek *et al.* 2010).

Strategies to improve recruitment included monetary incentives (Mapstone *et al.* 2002, Caldwell *et al.* 2010), telephone reminders (Watson & Torgerson 2006, Treweek *et al.* 2010), designs in which participants were aware of treatment group (Watson & Torgerson 2006, Tremlett *et al.* 2010), the use of an educational approach (Rendell *et al.* 2009, Caldwell *et al.* 2010) and taking cognisance of cultural issues in the design of study materials (Watson & Torgerson 2006). Where clinicians were involved in

recruitment, the impact on their workload was a significant deterrent (Foy *et al.* 2003). Participant eligibility was also an important factor where the use of too stringent inclusion criteria impacted on recruitment rates (Foy *et al.* 2003).

The challenges encountered with participant recruitment during the pre-exploratory stage meant the revisions and amendments were required. Site one, which was used for the pre-exploratory phase was retained for participation in the exploratory stage of the study. During the pre-exploratory stage PwMS were initially recruited by handing out packs during OPD visits. This proved to be slow, unreliable and labour intensive. Following discussions with the primary academic supervisor, I decided to change the method of recruitment. As the NSMS at the site was handing out the packs and knew who had received one, it was possible to change the approach to sending a postal letter of invitation along with a statement of interest form (SOI), without concern that PwMS would be subjected to repeated invites. As there was no pre-existing database of PwMS at the site, this necessitated the construction of a database of PwMS at the site, by the NSMS. Ethical approval was granted to facilitate this. The NSMS constructed the database and I contacted PwMS who returned the SOI and gave them a brief overview of the study. Following this and the PwMS's approval, a full information pack was sent by post. As ethical approval was obtained for recruitment via the handout of packs in OPD at the second site and appeared to be working reasonably well at the beginning of the trial, I decided to continue with this approach as it allowed further evaluation of that approach in the context of this exploratory trial.

Despite this, challenges to recruitment presented as the trial progressed. Receptionists varied in their diligence in the handing out the packs and during times of leave packs were not always handed out in a systematic way. No details were retained of individuals who had been offered a pack at the OPD site. Following consultation with the steering committee, I decided to continue with this approach to recruitment, because changing to a postal invitation letter might mean that people who had already received a pack during the OPD visit would be approached for a second time. In addition, as this was an exploratory trial, use of OPD at one site provided valuable information regarding the best approach to recruitment in a subsequent phase 3 trial. As access and ethical approval were not yet applied for at site 3, ethical approval for the use of postal letters of invitation was sought and granted for recruitment at that site.

Packs were delivered and replenished as required for OPD recruitment at site 2 and stamped letters of invitation to be addressed and posted by the NSsMS were provided to site 1 and 3. Table 9 depicts the flow of work from project initiation to the end of data collection.

Table 9: Flow of work from project initiation to commencement of the study

Site	Informal discussions with NSsMS	Ethics submission	Ethics Committee Meeting	Ethical amendments submitted	Ethical approval
Site one	Throughout 2009	27th Jan 2010 (School Advisory) Submitted Feb 26th 2010 Response received requesting amendments 12th April 2010	March 2010 .	20th April 2010	17th May 2010
Site two	Throughout 2009	8th February 2011	6th April 2011 (Responded May 5th 2011)	7th December 2011	20th December 2011
Site three	Throughout 2009	31st Jan 2012	1st March 2012 (Insurance amendment required)	29th March 2012	Received 20th March (letter dated 1st March 2012)
Site four	As per site one	As per site one	Site one ethical approval accepted at this site	As per site one	Site one ethical approval accepted at this site
Site five	As per site one	As per site one	Site one ethical approval accepted at this site	As per site one.	Site one ethical approval accepted at this site.

Site	Ethical amendments	Final ethical approval	Access to site	Access from Director of Nursing	NSMS consent
Site one	7th Jan 2011 (following manual development) 3rd May 2011 (additional amendments requested)	Email approved17th Jan 2011. Email approved 6th May 2011. Site ethics approval (Oct 5th 2012)	Access via Director of Nursing (DoN)	8th July 2010	1st March 2011
Site two	Amendments sought in December 2011	12th January 2012	Access via DoN	14th June 2011	25th May 2012
Site three	29th March 2012	10th April 2012 (Valid until 1st March 2014)	Access via DoN	24th November 2011	16thJuly 2012
Site four	Site one ethical approval accepted at this site	Site one ethical approval accepted at this site	Access via DoN	24th may 2011	Consent Not Returned
Site five	Site one ethical approval accepted at this site	Site one ethical approval accepted at this site	Access committee	29th June 2010	20th December 2010

Site	Date of access approval from the medical consultants	NSMS baseline interview	Manual training	Data collection: start date	Data collection: end date
Site one	5th December 2010	20th October 2010 (for pre-exploratory trial) 6th July 2011 (for exploratory trial)	6th July 2011 (for exploratory trial)	9th November 2011	29th April 2013
Site two	April and October 2011	Friday 22nd June 2012	Friday 22nd June (Completed 16th July 2012)	4th September 2012	12th March 2014
Site three	28th June 2012	December 2012	15th November 2012 Finished it in Dec 2012	31st January 2013	28th February 2014
Site four	22nd June 2011	Site one ethical approval accepted at this site	N/A	N/A	N/A
Site five	Consent not given	Site one ethical approval accepted at this site	N/A	N/A	N/A

N/A: Not applicable as the study was not carried out in the site.

6.2.7 Randomisation

Random allocation is the cornerstone of the randomised trial (Kendall 2003), because it rules out the effects of systematic bias (Schneider *et al.* 2003) and is essential for internal validity (Torgerson & Torgerson 2008). It involves the allocation of participants to the groups on a random or chance basis (Schneider *et al.* 2003), thereby ensuring that each of them has an equal chance of being assigned to the intervention or control group (Davis & Rose 2000, Akonbeng 2005). This balances both known and unknown variables (other than the intervention) which may have an effect on the outcome between the control and the intervention group (Torgerson & Torgerson 2008). Random allocation techniques include simple randomisation, block randomisation, matched randomisation and pairwise randomisation (Torgerson & Torgerson 2008). A list of random numbers using simple randomisation techniques such as coin tossing, a random number table or computer package is easy to prepare and the participants can be assigned to the groups in a number of ways (Torgerson & Torgerson 2008, Polit & Beck 2012). When sample sizes are large, computerized random number generation is helpful and methods of delivering the assignment include using opaque sealed envelopes, randomly pulling the number or having the list retained by an independent third party (Polit & Beck 2012). Although generally robust, simple randomisation techniques are particularly vulnerable to chance effects especially for small samples sizes which can lead to group imbalance on key variables (Kendall 2003, Torgerson & Torgerson 2008). Davis & Rose *(2000)* argue that this is less likely to occur with larger samples as potential confounding variables are more likely to be equally distributed between the two groups. Despite this, imbalances on key variables may still occur (Torgerson & Torgerson 2008).

Subversion is likely to occur when the researcher has a vested interest in the outcome and manipulates or interferes with the randomisation process in order to influence which participants go into which group. This may be avoided by using robust methods of allocation concealment, such as employing an independent third party to generate the random numbers and inform the researcher of group allocation following baseline data collection (Torgerson & Torgerson 2008). Using simple randomisation will, by definition, produce statistically significant (at $p<0.05$) imbalances in one or more

important covariates in 5% of trials and imbalances between the groups lead to loss of power (Torgerson & Torgerson 2008). This can be reduced with block randomisation strategies. Allocation concealment and blocking or stratification procedures are recommended when there is concern that unequal distribution of potential confounding variables across the groups could have an important influence on the results of the trial (Torgerson & Torgerson 2008).

6.2.7.1 Block randomisation

The risk of unequal numbers and imbalances of key variables can be reduced by using a block randomisation strategy, which is also needed for stratification purposes (Torgerson & Torgerson 2008), a process known as stratified block sampling (Akonbeng 2005). The researcher needs to have two or more randomisation lists using blocks of random numbers where variables of key importance to the study outcomes such as age, gender and ethnicity are identified and factored into the randomisation process (Torgerson & Torgerson 2008). This ensures approximately equal distribution of participants with different characteristics across the groups (Akonbeng 2005, Torgerson & Torgerson 2008). Caution is recommended when considering stratification on more than two variables as it can lead to large numerical differences between the groups (Torgerson & Torgerson 2008). When faced with such challenges, matched randomisation can be considered.

In this study participants were assigned to the two groups following baseline data collection, by a person independent to the trial. A random list of numbers for the control and intervention groups was generated by a statistician independent to the study, using blocks of 10, so that there would be 5 intervention and 5 control allocations for each 10 participants. This ensured a roughly equal distribution of participants to the control and intervention group across sites. Participants recruited to the trial were assigned a study number, which was used to track them throughout the trial. When baseline data were collected, the participant's study number was sent to the person holding the random allocations. The researcher was then informed of participant's group assignment.

6.2.8 Blinding

Blinding can be used to minimise bias which may occur when knowledge of group allocation influences process (Akonbeng 2005, Torgerson & Torgerson 2008) Researchers, interventionists or participants may alter their behaviours to consciously or unconsciously influence outcomes in either direction if they know the allocated group. This is particularly likely to happen if any of the stakeholders have a vested interest in producing a particular outcome. It is however, not always possible to implement blinding. In this Phase 2 trial, blinding was not possible.

6.2.9 Retention

In this study it was important that all participants (NSsMS, PwMS and SPs) were retained so that the potential impact of the trial could be determined. While monetary incentives may be helpful to trial recruitment, evidence supporting their value for questionnaire response rate is conflicting (Edwards *et al.* 2002, Nakash *et al.* 2007). In a review by Edwards *et al.* (2002), the use of monetary incentives more than doubled the odds of returning postal questionnaires, while a review by Nakash *et al.* (2007) found no evidence to support the use of incentives which included cash, prize draws, lotteries or phone cards. Both of these reviews reported on the importance of questionnaire design for improving response rate, in particular on the benefits of shorter questionnaires. The use of personalised questionnaires and letters, coloured paper, first class post, recorded delivery and stamp addressed reply envelopes were reported as effective methods to improve postal responses (Edwards *et al.* 2002). The ordering of the questions or inclusion of an information brochure with the questionnaire was not found to be beneficial (Nakash *et al.* 2007).

In this study, I personalised the letters for data collection by handwriting the participant's name and putting an original signature on each letter sent for data collection. As the SPs received a number of questionnaires at each data collection time-point, I hand wrote guidance on each questionnaire, as appropriate. For example, I indicated which questionnaire I would be phoning them to complete over the telephone so that they knew they did not have to fill it in by themselves. A small

monetary gift token was also sent to the SPs and PwMS as a token of gratitude for their participation (see section 6.2.23.1 for further discussion on the use of incentives in this study).

6.2.10 Data collection

Data collection methods appropriate to the conduct of an exploratory trial include structured interviews, focus group interviews, questionnaires and observation (McEvoy & Richards 2006, Medical Research Council 2008). Due to the exploratory nature of this study, multi-methods of data collection were used.

6.2.10.1 Quantitative data collection

Quantitative methods of data collection were used to contextualise the findings and evaluate intervention outcomes. PwMS were asked to complete baseline questionnaires to determine how their MS impacted on them and their quality of life. Demographic data on participants' age, gender, diagnosis and disability were obtained by the NSsMS from medical records. To provide an objective measure of the potential effectiveness of the intervention, nominated SPs completed questionnaires evaluating their ability to seek respite and problem solve. Other variable measures included nominated SP's preparedness to care, burden and quality of life. These questionnaires and the rationale for their use are described in the next section.

6.2.10.1.1 PwMS questionnaires

To contextualise the findings, quantitative and qualitative data were collected from PwMS. Two questionnaires were used to collect baseline data from PwMS (table 10).

Table 10: PwMS baseline questionnaires and data collection points

	Baseline	***Time 1(week 6)***	***Time 2 (week 13)***
Multiple Sclerosis Impact Scale (MSIS-29) (Hobart *et al.* 2001).	√*		
Multiple Sclerosis Quality of Life (MSQOL-54) questionnaire (Vickrey *et al.* 1995).	√*		

* Intervention and control

The Multiple Sclerosis Impact Scale (MSIS-29) (Appendix 11) was used to determine the impact of MS on the PwMS. The MSIS-29 is a patient based disease specific psychometrically validated outcome measure of the impact of multiple sclerosis which is suitable for use in clinical trials (Hobart *et al.* 2001). The instrument has been widely used in MS research and is validated internationally.

Data on Quality of Life (QoL) for PwMS were collected using the Multiple Sclerosis Quality of Life (MSQOL-54) questionnaire (Appendix 12). The MSQoL includes 18 MS-specific questions in addition to the 36 item SF-36 questionnaire, and has been widely used in studies evaluating QOL in PwMS (Nortvedt & Riise 2003). It is likely to be acceptable to respondents as it takes around 20 minutes (Fischer *et al.* 1999) to complete (half the time of other instruments) and has acceptable validity and reliability scores which ranged from 0.75-0.96 (Vickrey *et al.* 1995).

6.3.10.1.2 Support persons questionnaires

Six questionnaires were used to collect baseline and outcome data from the SPs (table 11).

Table 11: Support person outcome questionnaires and data collection points

Questionnaires	***Baseline***	***Time 1(week 6)***	***Time 2 (week 13)***
Support person Self-Efficacy: Self-Efficacy for Problem-solving scale (Zeiss *et al.* (1999) Self-Efficacy for Obtaining Respite subscale (Steffen *et al.* 2002).	√*	√*	√*
Support person preparedness scale (Archibold *et al.* 1990).	√*	√*	√*
WHOQOL BRIEF instrument (WHO 1996)	√*	√*	√*
Caregiver Reaction Assessment (CRA) (Given *et al.* 1992)	√*	√*	√*
Client Satisfaction Questionnaire-8 (Attkisson & Greenfield 1994)			√**

* Intervention and control **Intervention only

As the intervention was based on Egan's model of the skilled helper, which provided a framework for helping clients manage problems, I considered it important to test this variable. No validated instrument was found for use with caregivers of PwMS. However, a caregiver self-efficacy instrument comprising a caregiver self-care self-efficacy and caregiver problem-solving self-efficacy sub-scale was developed and tested with caregivers of frail or cognitively impaired elders. The definition of self-efficacy provided by the originator of that instrument "... concerned with people's beliefs that they can organise and execute courses of action to manage situations" (Zeiss et al. 1999, p.222) was congruent with the helping process that underpinned Egan's model of the skilled helper (Egan 2010), which included encouraging client self-efficacy.

Although the scale was revised by Steffen et al. (2002) with caregivers of people with a dementia diagnosis including individuals with Alzheimer's disease, Stroke/Multi-infarct, Parkinson's and other non-specified patients with dementia, it did not include the self-efficacy in problem-solving sub-scale. However, it did contain a sub-scale for self-

efficacy in obtaining respite, an outcome variable which the intervention in this study might impact upon. Therefore, I used these two sub-scales, the 4-item caregiver self-efficacy in problem management, as used by Zeiss et al. (1999) and the 5-item Self-Efficacy for Obtaining Respite subscale of the revised scale for Caregiving Self-Efficacy scale (Steffen et al. 2002) (Appendix 13). Good reliability scores were demonstrated for the 4-item Self-Efficacy for Problem-solving scale, Cronbach's alpha (0.83) test-retest ($r=0.683$, $p<0.001$) (Zeiss et al. 1999). Cronbach's alpha >0.80 were reported for the Caregiving Self-Efficacy in obtaining respite sub-scale and two-week test-retest results ($r_{12}=0.76$) (Steffen et al. 2002).

In this study, the content validity of the self-efficacy in obtaining respite and problem management was determined by asking a panel of experts consisting of one caregiver, one nurse teacher, and one psychiatric nurse with counselling expertise to evaluate the questionnaire in relation to its validity for evaluating the intervention for SPs of PwMS. A modified version of Sacks (no date) survey content validation instrument was used (Appendix 14). The experts were independently asked to rate each item on the questionnaire in terms of its representativeness and clarity on a four point rating scale, and focus (on the item being assessed) and comprehensiveness (whether the item should be deleted or retained) on a two point rating scale. A Content validity index (CVI) was calculated to determine the validity of each item on the questionnaire. A value of 0.80 is expected and if one third of the items require revision, the instrument incorporating modifications should be subjected to another content validation. In this study these two subscale received a CVI rating of 1, which is well above the acceptable cut off of 0.8 (Sacks No Date).

6.2.10.1.3 Instrument used to measure SPs preparedness to care

Caregiver preparedness was evaluated for its potential to detect effects of the intervention using the 'Caregiver preparedness scale' (Appendix 15). Caregiver preparedness is defined as "... a caregiver's assessment of his or her readiness to provide care rather than to the adequacy with which he or she is providing care" (Schumacher *et al.* 1998, p.66). The 'Caregiver preparedness scale' is an 8-item

subscale with a five point rating scale developed as part of The Family Care Inventory in the early 1980s (Archbold *et al.* 1990). The measure focuses on domain-specific preparedness and on role strain (Archbold *et al.* 1990). This instrument has demonstrated construct validity and crohnbach's alpha in three studies ranged from 0.88-0.93. (Schumacher *et al.* 2007), and internal-consistency reliability using crohnbach alpha of 0.71 at 6 weeks and 0.72 at 9 months (Archbold *et al.* 1990). It also demonstrated sensitivity to change in a recent study with carers of PwMS by Finlayson *et al.* (2009a) who collected outcome data at the 7 week time-point. It was also suitable because, the approximately 10 minutes it took to complete it, was unlikely to create a significant burden for participants.

6.2.10.1.4 Instrument used to measure SP burden

Burden is an objective and subjective (Hoffmann & Mitchell 1998) multidimensional construct comprising biological, psychological and social components (Chou 2000). The need to build a greater understanding of the underlying variables and restrict the concept in order to have theoretical relevance for research studies in nursing was purported by Chou (2000). There have been some attempts to define the concept (Loukissa 1994, Chou 2000), but the broad range of tools used to measure caregiver burden [over 40 burden measurement instruments were found to be used to evaluate carer burden both within and across disease specific illnesses] which included concepts of stress, strain, impact of caregiving, sense of competence and distress, indicates that the concept remains inclusive rather than restrictive.

In choosing an instrument to measure SP burden in this study, a number of instruments were considered. The most commonly used instruments to measure caregiver burden in MS included the caregiver Strain Index (CSI) (Chipchase & Lincoln 2001, Khan *et al.* 2007a), caregiver Reaction Assessment (CRA) (Pakenham 2001, Forbes *et al.* 2007), carer perceived burden visual analogue scale (Khan *et al.* 2006), caregiver Burden Scale (Knight *et al.* 1997), caregiver self-reported burden (SRB) (Khan *et al.* 2007a), the Zarit Caregiver Burden interview (Rivera-Navarro *et al.* 2003), the Spanish version

of the Zarit Caregiver Burden Interview (Rivera-Navarro *et al.* 2009) and the Maslach Burnout Inventory (adapted to care for an ill spouse) (Ybema *et al.* 2002).

Although some researchers did not report the psychometric properties of the instruments used (Chipchase & Lincoln 2001, Khan *et al.* 2006), others referred to previously reported psychometrics (Pakenham 2001, Khan *et al.* 2007a, Rivera-Navarro *et al.* 2009) or provided brief reports on psychometric properties for their study (Ybema *et al.* 2002, Rivera-Navarro *et al.* 2003, Forbes *et al.* 2007, Khan *et al.* 2007a). Although several of these instruments were suitable for use in this study, the CRA (Given *et al.* 1992) (Appendix 16) was selected because it measures both negative and positive subjective burden, is widely used in research for measuring carer burden, and has been well developed and tested (Deeken *et al.* 2003). The CRA is a 24-item scale, which assesses how carers react to caring for an ill person in 5 domains: how caring affects caregivers health, daily schedule, finances, their sense of self-worth (self-esteem) and their family (Pakenham 2001, Forbes *et al.* 2007). Forbes *et al.* (2007) provide supporting evidence for the psychometric properties of the CRA by replicating previously reported good internal consistencies for all 5 subscales (alpha >0.07) except for 'lack of family support' (alpha 0.53). Forbes *et al.* (2007) also purport that the CRA was previously used with carers of PwMS and reported overall satisfactory reliability, although no specific scale data were reported. It also had the advantage over other instruments because Forbes *et al.* (2007) demonstrated that the CRA was responsive to change over a 24 month time-frame.

6.2.10.1.5 Instrument used to measure SP quality of life

Quality of Life is defined as "Individuals perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (World Health Organisation 1996, p. 5). A variety of instruments have been used to measure QoL of carers of PwMS including: General Health Questionnaire (GHQ) (Figved *et al.* 2007), WhoQol Brief (Alshubaili *et al.* 2008), Assessment of QoL (Khan *et al.* 2007a), 36-item Short Form Questionnaire (Patti *et al.* 2007, Rivera-Navarro *et al.* 2009), UK version of the Short Form General

Health Survey (SF-36) version 2 (Forbes *et al.* 2007) and an 18-item, 3 point rating scale giving a total QoL score (Knight *et al.* 1997). Only one study, Forbes *et al.* (2007), report on the psychometrics properties of the questionnaires. This may be because most of the tools were already validated and internationally accepted QoL measurement tools. Knight *et al.* (1997) used a less well recognised instrument but did not provide details on item construction, validity or reliability of the instrument. None of the tools are caregiver specific tools, which may reflect the belief that carers are not patients. A further limitation in all studies of instruments on QoL for MS caregivers is a lack of theoretical exploration of the concept or clear operational definition for research purposes.

In this study, I used the WHOQoL Brief instrument to measure carers' QoL (Appendix 17). It comprises 26 questions covering four domains (physical, psychological, social relations and environmental) and takes approximately 20 minutes to complete. The tool is a global measure of QoL (Huang *et al.* 2006) which acknowledges the multidimensional nature of QoL (World Health Organisation 1996) and has good to excellent psychometric properties (Skevington *et al.* 2004). It is the most widely used tool for evaluating QoL in carers and has been used in studies measuring QoL for caregivers of PwMS. It has been proposed for use in intervention studies (World Health Organisation 1996) and demonstrated sensitivity to change in QoL following liver transplantation (O'Carroll *et al.* 2000). In this study, 15 minutes was required to complete the WHOQoL. It was therefore unlikely to be too burdensome for the SPs.

6.2.10.1.6 Instrument used to measure satisfaction with the intervention

Satisfaction with the telephone support intervention was measured using the Client Satisfaction Questionnaire-8 (Attkisson & Greenfield 1994) (Appendix 18). This 8 item questionnaire with a four option response has been widely used in studies measuring satisfaction with services offered and has known validity and reliability scores. It takes 5-10 minutes to complete. Further details are available at

http://web.mac.com/cliffattkisson/TamalpaisMatrix/CSQ_Scales.html.

6.2.11 Validity and reliability of the data collection instruments

6.2.11.1 Validity of the data collection instruments

The Preparedness to care and the CRA instruments had demonstrated validity for use with caregivers of PwMS in other research studies. The WHoQol Brief instrument is an international instrument used to measure QoL in the general population. In this study, the language used in these instruments was reviewed by a lay person and no issues were identified. No issues were identified with their use during the pre-exploratory stage of the study. During the study, however, some challenges were encountered with the CRA and WHoQoL Brief instruments. When responding to the CRA, participants felt forced into making responses within the response range options of strongly agree to strongly disagree for items that they felt were not relevant to them. For example, question 22 on the CRA which relates to help from family (brothers, sisters and children) in supporting/caring for the PwMS posed a difficulty in how to respond for one SP who did not have siblings. Another SP felt that their response to question 4 on the CRA was between the response options rated 3 'neither agree/disagree' and 4 'agree'. Likewise, for question 20, 'supporting/caring for the PwMS is important to me' one participant was forced to score 3 'neither agree/disagree' but clarified the response by stating "I feel I don't have a choice". In the absence of a 'not applicable' option and detailed guidance on dealing with responses outside of the scoring range, I emailed the authors of the tool to determine how to analyse questions that were not relevant to a particular participant. The advice received included entering the lower response score of 3 when the respondent scored an item between scores and use missing values such as mean score for 'non applicable' responses. The authors also cautioned against computing a total score as each subscale measured a different dimension and computation of a total score would make no conceptual sense (Personal communications from Professor Bill Given, via email, October 25, 2013). Despite these issues, the majority of participants did not report problems with the content of this questionnaire.

The WHOQoL Brief is a generic measure of quality of life, therefore, the issues raised by the SPs were surprising. Some felt that some of the items were not relevant to them.

For example question 22 asked about how satisfied the respondent was with support from friends to which one participant responded that he did not have any friends. At final outcome data collection, one participant commented that the questions were "too big" and gave the following question from the questionnaire as an example 'How satisfied are you with yourself?' Another SP had difficulty understanding some questions and put a question mark after them. Example of such questions included 'How safe do you feel in your daily life? and 'Are you able to accept your bodily appearance?' No problems were verbalised or noted with the preparedness to care questionnaire.

The self-efficacy in obtaining respite and problem-solving subscales were administered by telephone. Some participants felt that the questions were broad and that their response would differ depending on the problem. Once it was clarified that it was an overall assessment of their confidence that was required they were able to provide a response. There were no issues raised with the wording of the questionnaires. In relation to the questionnaires for outcome data collection for people with MS, no issues emerged in relation to the MSIS-29. Questions pertaining to sexuality caused difficulty for some PwMS who felt that due to their age or marital/partner status some of the questions were not applicable. Others left the response options blank. Missing data were particularly high for question 46 (lack of sexual interest) which had a combined missing/non applicable non response of 10% (n=7). Seventy-one percent (n=50) PwMS did not respond to question 47 (difficulty getting or keeping an erection). Three participants did not respond to question 23 (Do you feel full of pep?). One participant phoned the researcher stating that she had difficulty understanding the word 'pep'.

6.2.11.2 Reliability of the data collection instruments

Questionnaire reliability was tested for internal reliability using Cronbach's alpha on all baseline questionnaires for the PwMS and SPs. Table 12 presents the reliability results for the questionnaires administered to the PwMS. Cronbach's alpha score for the MSIS was 0.958 and the MsQoL subscales ranged from 0.647 to 0.950. The scores for all subscales, except social functioning are above the recommended 0.7 (Pallant 2013).

Table 12: Reliability test results for PwMS outcome questionnaires

PwMS Instruments	Cronbach's Alpha (n=70)
MSIS	**0.958**
MSQoL	
Physical health	**0.950**
Role limitations due to emotional problems	**0.917**
Role limitations due to physical problems	**0.854**
Pain	**0.916**
Emotional well being	**0.832**
Energy	**0.798**
Health perceptions	**0.773**
Social functioning	**0.647**
Cognitive functioning	**0.888**
Health distress	**0.925**
Satisfaction with sexual functioning	**0.858**
Overall QoL	**0.898**

Table 13 presents the reliability results for the five data collection questionnaires for the SPs. Cronhabh alpha scores for all the questionnaires ranged from 0.802 to 0.943, which are well above the recommended 0.7 (Pallant 2013). Temporal stability was measured for SP instruments administered to the control group at baseline and T1 (week 6). Test re-tests using the spearman rho correlation test was carried out on SP data from the control group. Test re-test scores varied considerably across the questionnaires with most sub-scales or items on the questionnaires demonstrating statistically significant correlations. One item on the self-efficacy in problem-solving sub-scale and two items on the preparedness to care had low test-re-test results. Scores ranging from 0.601 to 0.796 for the CRA and WHOQoL subscales and one item on the self-efficacy in obtaining respite were acceptable. Lower test re-test correlations (i.e. <0.7) between baseline and T1 scores for self-efficacy in problem management (item correlations ranging from 0.278 to 0.397) suggests that these constructs are less stable and more likely to fluctuate over time. Reliability co-efficients range from 0.00-1,

the higher the value the more stable the measurement instrument (Polit & Beck 2012). Shorter time-periods between testing produce higher scores with optimum time between 2-4 weeks (Polit & Beck 2012). The 6-week gap between testing may have negatively impacted on the reliability co-efficients achieved. Temporal stability for the CRA and WHOQoL subscales and one item on the self-efficacy in obtaining respite were acceptable, with correlations ranging from 0.601 to 0.796. The lower test re-test correlations between baseline and T1 scores for self-efficacy in problem management (item correlations ranging from 0.278 to 0.397) suggests that these constructs are less stable and likely to fluctuate over time. Tests of stability for such items may not be the most appropriate measure of reliability (Pallant 2013).

Table 13: Reliability test results for support person outcome questionnaires

Caregiver Instruments	Cronbach's Alpha (n=70)	Test re-test (r) Control group only
Self-efficacy in obtaining respite Self-efficacy 1-item	0.852	 0.744*
Self-efficacy in Problem-solving Clear and specific in looking at problems List possible solutions to a supporting problem Pick the most effective method of solving a problem Carry out a plan	0.908	 0.397* 0.372* 0.278 0.687*
Preparation to Care	0.943	0.659* (T0 and T1 mean sum score)
Item 1	_	0.442*
Item 2	_	0.462*
Item 3	_	0.526*
Item 4	_	0.304
Item 5	_	0.283
Item 6	_	0.436*
Item 7	_	0.385*
Item 8	_	0.516*
CRA		
CRA subscale 1 (interrupted schedule)	0.844	0.692*
CRA subscale 2 (Financial problems)	0.824	0.693*
CRA subscale 3 (Lack of family support)	0.727	0.601*
CRA subscale 4 (Health problems)	0.809	0.632*
CRA subscale 5 (Self-esteem)	0.770	0.693*
WHOQoL Brief		
WhoOoL subscale 1	0.842	0.775*
WhoOoL subscale 2	0.753	0.796*
WhoOoL subscale 3	0.836	0.784*
WhoOoL subscale 4	0.844	0.662*
Overall WhoQol	_	0.665*
Satisfaction (all 8 items)	0.802	_

*This result shows a significant correlation at p≤0.05

6.2.12 The intervention

A full description of the intervention, interventionist training and delivery was provided in chapter 4 (sections 4.5 and 4.6).

6.2.13 Usual care

Usual care was provided to the intervention and control groups. Usual care comprised NSMS responding to SPs calls on demand. The nature of usual care was identified from the pre-intervention interviews with the NSsMS (section 4.2). This consisted of SPs making contact with the NSMS if they required her assistance. Calls were only followed up if a voice message was left and further follow-up occurred only in situations where the NSMS needed to communicate specific information.

6.2.14 Data analysis

6.2.14.1 Quantitative data analysis

Data were analysed using SPSS version 18, using an intention to treat (ITT) approach, to limit bias. It requires the researcher to analyse outcome data from all participants including those who do not receive the intervention, part of the intervention, or those who withdraw from the intervention (Akonbeng 2005, Torgerson & Torgerson 2008). Every effort should be made to collect outcome data where possible and within the ethical principles of good research practice. The process of data analysis included database preparation, data cleaning, missing data imputation, subscale computation, identifying baseline sample characteristic, data exploration and examination to determine effectiveness of the randomisation procedures adopted, and characteristics of the data so that the appropriate inferential statistical test can be carried out.

6.2.14.1.1 Database preparation and data cleaning

The SPSS database was prepared for data entry by coding the questionnaires and questions and assigning variable names. The first 15 anonymous data sets were entered by a transition year student. I entered the remaining data sets. I checked the first 20 data sets and a number of data entry errors were noted. Consequently, I double checked all data for accuracy. Additional cleaning was performed using descriptive and exploratory statistics. Missing data were detected using the SPSS missing data computation function within SPSS. According to Graham & Donaldson (1993), the application of missing data procedures can produce unbiased conclusions even when there is substantial attrition, although this is questioned by some statisticians. In this study, where appropriate, missing data were imputed using published instrument guidance and additional guidance, where required from the authors of the instrument. Missing data were imputed as follows:

Missing data were imputed using the mean score for the self-efficacy in obtaining respite and problem management subscales (Appendix 13).

Self-efficacy in obtaining respite: For this subscale a number of items were not applicable (N/A) due to the nature of the needs of the PwMS. Most of the PwMS in this study were mobile and did not need to have a person with them when the SP was unavailable, especially for one day. At the first data collection point, 35 responses were missing due to 'N/A' for four items and 32 responses missing for one item. At the second data collection point, 27 responses were missing for 4 items and 24 for 1 item. Due to the large number of missing data, only 1 item (question 5) was analysed for this subscale questionnaire, as it had the least amount of missing data.

Self-efficacy in problem management subscale: For this subscale only 1 response was missing for 2 items at the first data collection point and 2 responses were missing for 4 items at the second data collection point.

Preparedness to care: The method adopted by Carter *et al.* (1998) was applied. Responses with < 75% of items responded to were excluded. For responses where a score was given for at least 75% of items, missing responses were estimated based on

the average of responses for the scale. Only one item response was missing for all questions at the first data collection point and 3 item responses were missing for all responses at the second data collection point, due to participant non-response. These questionnaires were excluded from analysis. All other questionnaire item responses were complete.

CRA: The approach adopted by Nijboer *et al.* (1999) was applied. Item scores up to a maximum of three (≤ 12.5% (n=3) of the total number of items) were replaced by the mean item score of that subscale. Participants with over three missing scores were excluded. Three respondents had complete missing data sets and were excluded. In this study only 1 item response was missing for 17 questions, 2 for 5 questions and 3 for 1 question at the first data collection point (T1). At the second data collection point (T2), 3 questionnaires were missing all items for this questionnaire and were excluded. One of the remaining respondents had 3 items missing and was excluded from the data analysis. The remaining respondents had <3 items missing.

WHOQoL: Missing data were imputed in accordance with the WHO syntax guidance for participants with at least 21 questions responded to. Where 1 item was missing, it was subsidised using the mean of the remaining domain items. Where 2 or more items were missing from a domain, the domain score was not calculated except for domain 3 where the domain score was only calculated if < 1 item was missing. Questionnaires with responses for less than 21 questions were excluded from the analysis (World Health Organisation 1996). In this study only one item response was missing for 24 of the questions due to one participant non-response at T1 and 4 responses on one item which pertained to 'satisfaction with sex life' and 2 responses for one other item 'satisfaction with support from friends'. At the second data collection point, three responses were missing for 23 questions, 4 for one question pertaining to 'ability to accept bodily appearance' and 6 responses were missing for the question pertaining to 'satisfaction with sex life'.

6.2.14.1.2 Subscales and other instrument transformations

The data were further prepared for analysis by the creation of syntax for total and subscale computation, as appropriate. Sum scores for the preparedness to care questionnaire and subscales for the self-efficacy in obtaining respite and problem-solving were computed. The WHOQoL subscales were computed using the WHO syntax provided along with the questionnaire. SPSS syntax was generated for the CRA data and this included the recoding of responses to 5 questions (questions 3,7,13, 15 and 19) (Given *et al.* 1992) as required and the computation of subscales according to the subscale guidance (Given *et al.* 1992, Nijboer *et al.* 1999). A higher score on the self-esteem subscale indicated a greater positive effect of caregiving and higher scores on the other 4 domains indicated more negative effects (Malhotra *et al.* 2012).

6.2.14.1.3 Data checking for normality and variability

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The randomisation method was evaluated by checking if the background and essential variables were equally distributed between the two groups. Descriptive statistics including frequency counts, percentages and measures of central tendency, such as the mean, median and mode were computed, as appropriate to the scale data.

The transformed data were analysed to determine whether the data met the criteria for parametric or non-parametric testing. Parametric testing is more powerful than non-parametric as it includes all relevant detail on the sample. In order to use parametric testing, the data from the two groups must be normally distributed and of similar variability. To meet the criteria data can be transformed into logged data, if required. Parametric tests such as the t-test and ANCOVA are robust and can accommodate some violations of these criteria. In this study, I computed descriptive statistics (mean and SD) and normality checks (skewness, kurtosis, histograms and normality plots). Finally, I computed inferential statistics. On visual examination, the data appeared normal and group variability was similar. With cognisance of the computed data and sample size and in consultation with a statistician, parametric tests were used for

interval and scale data. When data measurement did not meet the criteria for parametric testing, I used non-parametric versions of the tests.

6.2.14.1.4 Statistical tests performed

This study comprised two groups with data for each group collected at three time points, baseline (T0), six weeks (T1) [the primary outcome data collection point] and 13 weeks (T2) [the important secondary outcome]. The characteristics of the baseline data for the intervention and control groups were analysed for similarities and differences between the groups using t-test and categorical tests as appropriate for two tailed hypotheses. Categorical data were analysed using chi-squared analysis and Fisher's exact test. In order for the chi-squared test to be valid, 80% of the expected frequencies should exceed 5 and all should exceed 1. When expected values are too small the Fisher's produces the correct *p* value as that produced by the chi-square test is too small (Bland 2005). The paired t-test was used to test within group differences at different time points. The analysis of co-variance (ANCOVA) test was used to test for differences between the two groups because it adjusts for the baseline measure (Darwin 2005).

6.2.15 Intervention fidelity

Fidelity to the intervention was monitored and evaluated using the treatment fidelity guidelines (Bellg *et al.* 2004), as discussed in section 5.9. Standardisation and monitoring of the intervention was obtained by the development of an intervention manual which guided the delivery of the intervention. A call monitoring instrument was devised based on the manual. Ten percent of all intervention calls were recorded and reviewed for adherence to the intervention guide. A random selection of the first 18 recorded calls was independently evaluated. Due to the uniqueness of each intervention call, the flexibility in intervention delivery, the broad scope of the skills underpinning the principles of the intervention and individual NSMS practice style, it was not possible to apply a standard method for fidelity assessment. Overall

evaluation decisions were based on the appropriateness of call content, the appropriateness of the skills applied and adherence to the principles of the intervention. In addition, written records of the calls maintained by the NSsMS were analysed and further detail on fidelity to the intervention was obtained during the semi-structured interviews with the nominated SPs, PwMS and nurse specialists themselves.

Standardisation of interventionist training was maximised through the inclusion of a training section in the intervention manual (see CD rom, training manual). The training section of the manual standardised the detail provided on the models underpinning the intervention and provided standard guidance on their application through the use of scripted scenarios. I was the only person who provided training to all interventionists. Although no two interactions are ever the same, because interventionists' needs and questions differed slightly, the training was relatively similar across all sites. In accordance with the fidelity guidelines, the progress of nominated SPs in acquiring knowledge and skill from the intervention was adhered to by ongoing interventionist coaching. Prior to each set of intervention calls, the interventionists were reminded of the overall objectives of the intervention, provided feedback on the application of the intervention and monitored participant progress throughout the calls. Monitoring improvement in the ability of participants to use the skills was more challenging as outcomes were not evaluated during the intervention. Where possible, I noted this when monitoring the treatment fidelity calls and provided feedback to the interventionist regarding the application of the principles underpinning the intervention.

I monitored contamination through a review of records of all NSMS contact with control group participants. NSsMS were informed of the importance of adhering to usual practice when interacting with these participants and they were advised not to make initial one-to-one contact with nominated SPs in the control group, in keeping with usual practice.

6.2.16 Acceptability and feasibility

As this was a new intervention, delivered in a manner which differed considerably from current practice, it was necessary to determine its acceptability and feasibility. Acceptability referred to the extent to which the intervention was considered

appropriate by the PwMS, nominated SPs and NSsMS. Feasibility referred to whether it was technically possible for the NSsMS to deliver the intervention and how likely it was that a full scale trial would be concluded. Acceptability and feasibility were evaluated as part of the process analysis.

6.2.17 Process evaluation

A process evaluation enables the researcher to monitor the quality of delivery of the intervention and identify what aspects contribute to success or failure (Linnan & Steckler 2002, Medical Research Council 2008). It can help explain the results that are obtained and whether any lack of success is due to implementation issues, rather than actual ineffectiveness (Medical Research Council 2008). This is achieved through an evaluation of the quality of the delivery of and fidelity to the intervention, along with an examination of the contextual factors that influence application and outcomes (Medical Research Council 2008). It also affords the researcher the opportunity to consider the mechanisms that impact on the delivery and outcomes of the intervention (Hulscher *et al.* 2003, Medical Research Council 2008), as it enables identification of which components of an intervention are likely to be most effective, for whom, and under what conditions (Linnan & Steckler 2002).

At the exploratory stage, a process evaluation can inform the feasibility and acceptability of the intervention (Hulscher *et al.* 2003). It can also inform the ongoing development of the intervention and refinement for further exploratory testing or progressing to a full-scale trial. In addition, it enables an estimate of potential change that the intervention can bring about (Hulscher *et al.* 2003), informing the researcher as to how much, if any, adjustment is likely to be required to bring about the desired change. In this study, the process analysis enabled the determination of the feasibility of implementing the MNTISS, the identification of potential barriers and facilitators to testing the intervention using a randomised trial design and the overall acceptability to NSsMS, PwMS and their nominated support persons. Due to a lack of guidance in relation to how to perform a process evaluation (Linnan & Steckler 2002), some researchers simply nest a qualitative evaluation into their trial to answer areas of

interest. This approach has limitations in that key aspects of the process such as influence of context or intervention implementation may be omitted. It is, however, important that the volume of data collected is balanced with the resources available by focusing on collecting data that are needed to inform the outcomes (Linnan & Steckler 2002). The process evaluation in this study was based on the work of Linnan & Steckler (2002) and was influenced by the way in which other authors (Driessen *et al.* 2010, Hasson 2010) applied the framework/approached the process analysis (Haut *et al.* 2009).

Table 14 presents the components of the process evaluation, together with key process questions and data sources. A researcher log of events and qualitative interviews with key stakeholders [NSsMS, PwMS, nominated support persons] were the main sources of data for the process evaluation. Interviews with nominated support persons were conducted throughout the study and with the final trial outcome evaluation. Interviews were carried out with PwMS and NSsMS at the beginning of the study and on completion of the intervention delivery.

Table 14: Process evaluation components, questions and data sources (based on the work of Driessen *et al.* (2010) and Hasson (2010))

	Process questions	Data sources
	Acceptability and feasibility of research methods	
Recruitment	• What number of sites was invited to partake in the study? • What number of sites participated/did not participate in the study? • How many NSsMS consented to take part? • How many NSsMS participated/did not participate? (reasons)	• monitoring packs/letters sent, method and outcomes/response rate/uptake to the study
Acceptability	• What methods were used to recruit? • What were the enablers/barriers to participant retention?	• Method (database/out-patient) • Statement of interest forms, researcher running log of events, nurse specialist (post intervention interviews), communications with receptionist handing out the packs.
Logistics	• What number of participants (PwMS) were invited/responded/recruited/consented? • What number of participants (nominated support persons) was invited/responded/recruited?	
	• What were NSsMS and participants' attitude towards random allocation to the groups? • Was it possible to apply the randomisation procedure as planned?	• Researcher records • Researcher records/post intervention interviews.
Screening	• Are the inclusion/exclusion criteria appropriate? • Are they practical: Ease with which screening is carried out (Staff/participant burden)? Time screening is scheduled (relative to recruitment, consent, baseline data collection)?	• Researcher reflections

	Extent to which tools used for screening are valid and easy to use? • Is the screening done in an ethical manner?	• Procedure followed
Retention	• How many consenting participants completed the study	• Records of retention rates
	• Were the strategies used to aid retention effective?	• Researcher's reflections
Data Collection	• Are the instruments used easy to understand, content relevant and response options appropriate? • Are they relevant to the population under study? • How long do they take to complete? • How easy are they to administer? • Is the method of administration appropriate? • Can the data be collected within the specified time-frame for the study? • What contributes to delays? • Are there other logistical issues with data collection?	• Responses/comments from participants • Researcher observations/notes, delays in sending out questionnaires
	Acceptability and feasibility of delivery of the intervention	
Context	• What organizational factors impacted positively/negatively on program implementation?	• Nurse specialist (post intervention interviews)
	• What were the barriers/enables for the delivery of the intervention?	• Researcher's database record and log of events
Reach	• What proportion/% of the target audience received all/part of the intervention?	• Researcher and nurse specialist records
	• What nominated support persons received the intervention (age, level of disability of PwMS, working/not working/reasons for partaking)?	• Researcher records

Fidelity	• To what extent did the nurse specialists adhere to the principles and process of Egan's model of the skilled Helper? • To what extent were the participants helped to manage problems?	• Call recordings • Nurse specialist records • Qualitative interviews • Self-efficacy in problem management questionnaire
Dose delivered	• Did the nurse specialists deliver four phone calls (two in month one, one in month two and one in month three)? • What was the average length of the call to each participant?	• Researcher records, nurse specialist records, post-intervention telephone interview (fidelity to intervention component) with nominated support person. • Recordings, nurse specialist records
Intervention delivery (tracking)	• Were the calls delivered as scheduled in the protocol? • What were the reasons for delays to delivery on schedule? • What was the average and range of days to delayed delivery?	• Recordings/nurse specialist records/post-intervention fidelity recordings with nominated support persons • Researcher records

Dose received	• How many calls did the nominated support person receive? • On average how long was the call to each participant? • What was the average length of the calls overall?	• Researcher records, nurse specialist records, post-intervention telephone interview (fidelity to intervention component) with nominated support person. • Recordings, nurse specialist records
Acceptability of the intervention	• Was the intervention acceptable to the participants? • Was the method of delivery of the intervention acceptable to the participants? • What changes did participants recommend to the intervention?	• Interviews with SPs and NSsMS
Satisfaction	• Were participants satisfied with the service offered?	• Satisfaction questionnaire

6.2.18 Application of the qualitative approach to feasibility testing

The qualitative component of the study was to inform the process evaluation as described above, which included evaluating the support persons' experiences of receiving the intervention and NSsMS' experiences of delivering the intervention. In addition, qualitative data were collected to identify qualitative improvements, if any, for PwMS. To achieve this, the views, opinions and experiences of the NSsMS, PwMS and their nominated support persons were elicited using the principles of qualitative descriptive research (Sandelowski 2000, 2010). As the purpose of the qualitative component of this study was to describe usual care and determine feasibility, low level interpretation was employed.

6.2.18.1 Qualitative data collection

Qualitative data collection methods include collection of textual or verbal data using methods such as interviews and focus group discussions. Interview guides can be structured, unstructured or semi-structured and are used for interviews and focus groups. The degree of structure depends on the purpose of the study and interview. Questions for structured interviews vary from precisely worded questions that the interviewer is only permitted to repeat, to those which the interviewer is permitted to provide an explanation or re-word in order to aid interviewee understanding. Responses are usually controlled in some way and may be restricted to pre-determined response options (Burns & Grove 2005). Unstructured interviews are those that enable the respondent to determine the content of the response (Burns & Grove 2005). Semi-structured interviews lie somewhere between structured and un-structured in that the questions focus on a specific area that guides the respondent in terms of the content of the response but the content of that response is determined by the subject. In this study, to inform the process analysis, one-to-one post-intervention semi-structured interviews were carried out with NSsMS, PwMS and nominated support persons. Interview guides (see Appendix 8, 19, 20) were designed based on key components of intervention feasibility and acceptability (Appendix 21). The guide was broad so that I could probe and follow lines of discussion, as appropriate to the site.

In this study, a semi-structured interview guide was used to describe current practice (usual care) and the support offered by the NSMS to SPs (Appendix 8). These pre-intervention interviews with the NSsMS were used to inform the ongoing development of the intervention, describe site specific usual care and guide protocol development. NSsMS were interviewed after delivery of the intervention in order to determine their experiences of delivering the intervention. They were invited by a gatekeeper (the site neurology nurse manager) as part of the overall invitation to the study (see Appendix 22) for NSMS letter of invite and information sheets). Interviews with the NSsMS and PwMS were conducted face-to-face in a location of convenience for the participants. All the NSsMS asked to be interviewed at their place of work.

PwMS had been informed of the likelihood of being invited to participate in the qualitative component of the study if their nominated support person was assigned to the intervention group, when they were asked to give their consent to participate in the trial (Appendix 23). As a consequence, I was able to make direct contact with PwMS and invite them for interview (see Appendix 24 for letter of invite and participant information sheet). A letter of invitation and information sheet was sent to each potential interviewee. Fifteen PwMS whose SPs were assigned to the intervention group were invited to partake in an interview prior to the delivery of the intervention to the SP and the same PwMS was invited for interview following the SP's receipt of the intervention. The purpose of the PwMS pre-intervention interview was to determine the type of support they received from their SP. The purpose of the post-intervention interview was to determine if there was any change in their experience of being supported as a result of the nominated support person taking part in the intervention, and to determine any negative effects of the intervention for the PwMS, if they existed.

When interview participants had made initial contact, I contacted them by telephone to make sure that they fully understood the contents of the information sheet and answered any questions about the interview and study. If the person gave their consent, the interview took place in a location of convenience for them. For PwMS, I had to travel to their homes or locations close to their homes. The PwMS were from counties in the catchment areas for the study sites and included Wexford, Tipperary, Drogheda, Carlow, Wicklow and the Dublin region. Before embarking on each journey,

I ensured that all the necessary transport requirements, equipment and information were prepared (see Appendix 25 for checklist prior to interview).

Only two PwMS requested the interview to be done outside their homes and they recommended a local hotel where a quiet room could be sourced. On both occasions, the private rooms for the interviews were provided without charge. Interviews with nominated SPs were conducted by telephone in conjunction with the final outcome data collection call.

Interviews with PwMS were short and ranged from 6 minutes to 27 minutes. Despite this, I was often with them for at least one hour, as they wished to chat and get to know me. This usually occurred after the interview and some participants invited me to have tea with them and their partner (if present) or they wished to show me parts of their home, especially if they had a hobby or business of interest. This indicated the development of rapport and a level of trust and comfort in my company. The benefit of face-to-face meetings with participants was evident. Despite having consented to take part in the interviews, on initial sighting, I saw that some participants were anxious about meeting me. However, they visibly relaxed quickly. The trust built as a result of having met the PwMS and their family member, if present on the day, may have led to greater dyad commitment to the study.

All SPs were informed that they would be invited to participate in a post-intervention telephone interview, if they were randomised to the intervention group (Appendix 26). This component of the outcome evaluation was to elicit their experiences of receiving the intervention and to determine how it may be modified. It was conducted by telephone during the final post-intervention data collection phase (Appendix 20). All SPs who participated in the intervention agreed to participate in this interview.

The NSMS pre-and post intervention interviews took between 22-57 minutes to complete. The pre-intervention interview at site one took 40 minutes to complete, at site 2 it took 28 minutes and site 3, 22 minutes. The post-intervention interview at site one took 28 minutes to complete, at site 2 it took 57 minutes and site 3, 42 minutes. The NSsMS appeared relaxed in my company and two of them offered to show me the OPD suite and introduced me to some of the staff at the site.

6.2.18.2 Qualitative sampling

Sampling in qualitative research is guided by the purpose of the qualitative study or component of a study and can be convenience, probability or random (Parahoo 2006). In general, qualitative researchers want to interview those who have the knowledge or experience to answer the research question. Sampling is usually purposive in that individuals are selected because of their expertise pertaining to the topic under examination. In this study, all the participants were considered to have the requisite information to inform the aims of the study. All NSsMS in this study had the knowledge and experience required to inform the description of usual care and those who participated in the delivery of the intervention had the experience required to inform the feasibility of the intervention and study protocol. Four NSsMS consented to take part in the study and were interviewed to inform the description of usual care. Three of these were involved in the delivery of the intervention and all three agreed to be interviewed.

As all PwMS whose SP was assigned to the intervention group had the experience required to inform the study outcomes, it was important that all PwMS were given an equal opportunity to participate in this component of the study. All SPs assigned to the intervention group were therefore eligible for inclusion in the pre-post intervention interviews. Simple random sampling was used in the initial stages of the study. However, as the study progressed and fewer participants than anticipated were recruited to the study, it became clear that the number of participants for selection from those assigned to the intervention group, would not be adequate to inform the feasibility component of the study. This issue was raised with the members of the steering committee (which comprised the academic supervisors for the study), and it was agreed that consecutive participants in the study could be invited to participate in the interviews. This was a procedural change and all the ethical principles underpinning the study continued to be upheld, including the right to informed consent, to self-determination and to fair treatment. A sample of 15 PwMS was invited to take part in the interviews. This was 15% of the original estimated sample size of 100 participants in the study. It represents 45% of the 33 participants assigned to the intervention group. All 33 SPs assigned to the intervention group were invited to take part in the post-intervention evaluation interview.

6.2.18.3 Qualitative data analysis

Data analysis involves making sense of the data collected and presenting it in a manner that can be easily assimilated by the readers and users of the research. A process evaluation which forms a component of an exploratory trial informs the researcher as to what works, what might work better and what doesn't work (Medical Research Council 2008). Elaborate interpretation of the views and opinions of stakeholders is not required to achieve this objective; therefore, I did not need to move far beyond the data as presented. However, remaining close to the data as collected does not mean a total absence of interpretation (Sandelowski 2000). There are many ways to analyse qualitative data and many of these are aligned to specific approaches which are categorised within the qualitative paradigm. For studies that are not aligned to a particular approach, content analysis is the usual method adopted. Krippendorff (2004) describes content analysis as a "...technique..." (p.18) and provides a framework for analysing content so that the researcher can make "...replicable and valid inferences from texts (or other meaningful matter) to the contexts of their use" (p.18). Qualitative content analysis is a method of reducing the frequently large volumes of data so that they can be summarised for the reader. It is the main method of analysis for descriptive qualitative research, as it enables the researcher to categorise, summarise and interpret large volumes of written, verbal or visual content (Elo & Kyngäs 2007). Content analysis sits on the border of the quantitative and qualitative approaches and allows for both interpretation and quantification of data (Jackson 2009). The degree of interpretation required depends on the purpose of the qualitative study and content analysis provides flexibility because it helps the researcher to provide analysis of the manifest and latent content as required (Graneheim & Lundman 2004).

6.2.18.4 Framework for analysis

In this study, I had difficulty selecting any one framework to meet the requirements of the process analysis. As the purpose of the process analysis did not require me to

move far beyond the manifest data the process was guided by an adaptation of the work of Krippendorff (2004) and Elo & Kyngäs (2007).

The overarching framework for the analysis was provided by the three phases of content analysis: preparation, organising and reporting (Elo & Kyngäs 2007) with elements of Krippendorff (2004) framework for analysis used to provide more detailed guidance on some aspects of the analysis of the transcribed interviews. While the two frameworks had many similarities (in that they both refer to identifying units of analysis, coding and interpretation of the data), they differed in the degree and depth of explanation provided on how to do this, in particular the method of interpretation. For example, Krippendorff (2004) recommends data reduction as a means of managing large volumes of data and the process of abductive inference when making inferences about the meaning of the data. Abductive inference refers to the process of making inferences which are backed up by evidence through the use of analytical constructs (Krippendorff 2004). Analytical constructs provide a method to enable the researcher to ensure that the analysis does not violate the data provided within the texts. Elo & Kyngäs (2007) provide guidance on deductive and inductive approaches to analysis and the process of category generation. Detail on how to carry out the process, in particular data interpretation is, however, scant. Likewise, the process for data reduction is not explicit in the Elo & Kyngäs (2007) guidelines.

Within this study, these two approaches to content analysis complemented each other. Their application provided a comprehensive framework for the overall analysis of the qualitative data as it, along with the use of the computer aided qualitative data analysis system (CAQDAS), NVivo 9 (QSR International 2012), allowed me to consolidate the results from all three perspectives (NSsMS, support persons and PwMS). In addition, my understanding of the terms and concepts of content analysis was guided by illustrations on the use of content analysis presented by Graneheim & Lundman (2004).

6.2.18.4.1 Application of the framework for analysis of qualitative data

6.2.18.4.1.1 Phase 1: Preparation phase

Each interview comprised the unit of analysis (Graneheim & Lundman 2004) or recording unit (Krippendorff 2004). During this phase, the researcher reads through the material several times to make sense of the data and prepare for data reduction. In this study, this included the transcription of most of the qualitative data, which provided three perspectives (NSsMS, PwMS and nominated support persons), at both pre- and post-intervention interviews and importing audio files or transcripts for direct coding into Nvivo.

As the interviews were done to answer specific questions, a deductive method of analysis was employed. This was carried out at two levels, firstly from each of the three perspectives separately and secondly by a process of consolidation, which allowed for the representation of the findings from all three perspectives.

Each question on the interview schedule comprised the overall category for the analysis. A category produces a descriptive level of content (may include subcategories) and are expressions of manifest content (Graneheim & Lundman 2004). In accordance with the guidance of Graneheim & Lundman (2004) structured categorisation matrixes (Appendix 27) were devised based on the interview schedules. Sentences and paragraphs comprised the context unit, which are *"...units of textual matter that set limits on the information to be considered in the description of recording units"* (Krippendorff 2004, p.101).

The categories were further reduced and consolidated to enable the representation and interpretation of the data from all three perspectives, using the overall framework for the process analysis.

6.2.18.4.1.2 Phase 2: Organising

This step included coding by assigning each segment of the data to each category and subcategory in accordance with the categorisation matrix (Appendix 27). This was done separately for each type of interview (pre- and post-intervention), and from the

three perspectives. The coded content from all interviews were then grouped into the relevant categories and further distilling, re-labelling and merging of the categories ensured that labels accurately reflected the coded content. This was necessary to create the descriptive accounts of participants' experiences pre- and post-intervention. Data reduction involved consolidating and refining interview coding frameworks from all three perspectives into the more condensed and higher level codes of the framework developed for the acceptability and feasibility of the intervention and research design. Where necessary, I developed higher level codes to summarise the content of each category.

6.2.18.4.1.2.1 Rigour

The procedures adopted in a particular study must be made explicit for validation of the study processes and findings, and to give readers confidence in the findings. Confidence in the findings is important for qualitative studies and transparency in procedures is all the more critical with greater degree of interpretation. Just as there is no one right way to do content analysis, there is no single way to determine rigour. The approach adopted must suit the purpose of the study and the extent of data interpretation. Elo & Kyngäs (2007) use the term 'trustworthiness', which means clarity regarding the analysis process and validity of the results. This requires the researcher to provide a detailed description of the context and procedures used to analyse the data together with a trail for how the results emerged from the data.

The strategies used to demonstrate trustworthiness depend on the particular qualitative approach adopted along with the purpose of the study and methods used (Krefting 1991). Irrespective of the approach, the researcher must demonstrate rigour, which requires thoroughness and transparency in the application of the research methods.

Rigour refers to thoroughness and transparency in the application of the research methods. It is used to judge the quality of a study and determines the degree of confidence that can be placed in the findings. In qualitative research, rigour is obtained through the use of strategies that demonstrate trustworthiness in the findings i.e. truthfulness of the findings in relation to the data collected. The greater the degree of data interpretation, the greater the requirement for stringent measures to demonstrate,

that the findings are true to the participants (Williams & Morrow 2009). In this study rigour was established using strategies that demonstrate trustworthiness. These include credibility, transferability, dependability and confirmability (Krefting 1991, Shenton 2004, Koch 2006).

6.2.18.4.1.2.2 Credibility

This refers to confidence that the findings are reflective of the data collected (Krefting 1991, Shenton 2004, Koch 2006). In this study, credibility was demonstrated by the application and written account of a well- established research methods suitable to the purpose of the study, the selection of participants most likely to be reflective of the overall sample, highlighting cases that were at odds with the majority of the findings including negative cases, creating an environment that enabled the respondents to be truthful (e.g. being neutral in my reactions to both positive and less neutral or negative feedback), peer scrutiny of the process and report by engaging my supervisors in the process.

6.2.18.4.1.2.3 Transferability

Transferability of the findings to other sites depends on the degree to which the study site is reflective of other settings. The responsibility for transferability lies with both the researcher and reader. The researcher must provide sufficient information for the reader to allow them to make an inference as to the appropriateness of the findings to another setting. In line with recommended strategies, (Krefting 1991, Shenton 2004, Koch 2006) transferability was addressed in this study by providing detailed information on the context and settings (sections 3.2 and 4.2). In addition, and as recommended by Krefting (1991), demographic data of the participants was compared to that of the overall sample within the study and that more widely available from the literature and voluntary organisations in relation to the study population.

6.2.18.4.1.2.4 Dependability

Dependability enables a researcher to replicate the study and is achieved by the researcher providing detail on the research design, methods of data collection, analysis and interpretation including detail of all decisions made throughout the study. In addition, I used the code-re-code method and peer examination (Krefting 1991) whereby I coded a section of the data and re-coded the same data two weeks later. This allowed me to demonstrate dependability in the analysis process. The research plan and implementation was open to examination by my research supervisors.

6.2.18.4.1.2.5 Confirmability

This is similar to credibility in that it requires the researcher to demonstrate that the findings are those of the participants and their experiences, rather than the preferences of the researcher. In addition to measures noted in the credibility section above, I ensured that the overall findings of the process analysis was reflective of the views of all three groups (NSsMS, PwMS and nominated SP) and made explicit all decisions underpinning the analysis and interpretation of the data. According to Koch (2006), confirmability is established when credibility, transferability and dependability are achieved.

6.2.18.4.1.3 Phase 3: Reporting:

This phase involved the application of the above methods to demonstrate the trustworthiness of the study. It involved a process of examination for bias in the interpretation through a process of self and peer audit. In doing so, I had to validate the analysis by seeking evidence within the data to support the findings. It also required the synthesis of the analysis into coherent and well supported outcomes and the presentation of these outcomes in the findings and discussion section of the thesis.

6.2.19 Study protocol

The protocol was developed following the design of the intervention. Cognisance was taken of the nature of clinical practice and the demands on the nurse specialists' time. The protocol submitted for ethical approval is in Appendix 28.

6.2.20 Clinical trial registration

Under the declaration of Helsinki researchers are required to register all clinical trials in a publicly accessible data base prior to recruiting the first participant to the trial (World Medical Association Declaration Of Helsinki 2008). It is also a requirement by many medical journal editors that trials are prospectively registered before they will be considered for publication (DeAngelis *et al.* 2005). This trial was registered on clinical trials.gov, a service of the US National Institutes of Health. The trial registration number: is NCT01302431.

6.2.21 Quality control

Monitoring for quality was an important part of the study. The process began by carrying out a risk analysis at the start of the study and putting controls in place to avoid, minimise and detect problems, as appropriate. Measures in accordance with recommendations of Kendall (2003) were adopted. These included the use of previously tested and validated data collection instruments, where possible, and evaluation of the quality of data collected by these instruments (user friendly, self-explanatory, clearly formatted and collect only data that is needed) prior to full trial testing. In addition, the database for data entry was designed in a similar format to the forms, to allow ease of entry and to minimise the potential for data entry error. As there was more than one interventionist delivering the intervention, training was standardised using a training manual. Once the trial protocol has been devised, only changes that are necessary should be made (Kendall 2003). Consequently, in this study, only necessary changes were made to the protocol following consultation with the advisory committee.

6.2.22 Ethical approval

Ethical approval was sought and granted by the Faculty of Health Sciences research ethics committee (see Appendix 29 for final letter of approval and amendment approvals) and all relevant hospital research ethics committees (HRECS). I was required to be present to answer any queries regarding the study at two of the HRECS.

6.2.23 Ethical considerations

The ethical principles of respect, justice, beneficence and non-maleficence, and rules based on these principles in accordance with the Nuremberg code, Helsinki Declaration and the Belmont report (Dooley & McCarthy 2012) guided the ethical conduct of this study. These rules include participants' rights to confidentiality and veracity (Newham & Hawley 2007) voluntariness, self-determination, informed consent, anonymity, privacy and fair treatment (Dooley & McCarthy 2012). In Ireland, nurses are professionally accountable to patients, research participants, their employers and the registering body (An Bord Altranais 2000). Furthermore, they are legally accountable to the State (Dooley & McCarthy 2012). This means that when doing research, they must be able to demonstrate how they adhered to the ethical principles and give due consideration to the ethical rules.

In this study, the ethical principles and rules were applied taking cognisance of the dyadic nature of the participants and the design of the study. Due consideration was given to the rights of nurse specialists, PwMS and their nominated support person. The fact that the study was testing a new intervention that was not previously subjected to evaluation was also an important consideration.

6.2.23.1 The principle of respect

The principle of respect relates to participants' right to self-determination and includes autonomy, confidentiality and informed consent (Polit & Beck 2012). Autonomy means that the researcher respects research participants' rights to self govern and is a moral and legal right (Dooley & McCarthy 2012). Autonomy can be upheld by adhering to the rules governing informed consent, self-determination and voluntary participation. Voluntariness, capacity and information are required for a valid informed consent (Dooley & McCarthy 2012). To achieve this, potential participants must have the mental capacity to understand what the study involves, the possible consequences of their decision to partake or not and be able to communicate their decision. To do this they must be given sufficient information to consent or refuse to partake in the study. Information about the purpose of the research, what is involved if they decide to take part, the possible risks and benefits of partaking in the study and of their right to withdraw without penalty are central to giving informed consent.

In this study, the principle of autonomy for PwMS was upheld by making contact with the PwMS in the first instance. As the primary focus of the study was to provide advice, support and teach problem management skills to nominated support persons in order to help them to continue supporting and caring for the PwMS, it was essential that the PwMS was in agreement that the person who supports or cares for them could be contacted by me, as the researcher, and the nurse specialist in the hospital that they attended. This was achieved by giving PwMS an information pack comprising a cover letter and information sheet (Appendix 23) which outlined the nature, purpose and procedures of the study. In the pack, they also received an information pack for their nominated support person, which contained a letter of invite and an information sheet (Appendix 26). The letters of invite (database recruitment) or information packs (OPD recruitment) were distributed by a gatekeeper (the NSMS or receptionists). When statement of interest forms were returned by the potential participants they were contacted by me. The PwMS was contacted in the first instance. When I was satisfied that the PwMS understood the contents of the information sheet and was in agreement to proceed, on receipt of a statement of interest form from the SPs, I contacted them to explain the nature of the study and answer their questions. All participants were

required to provide written consent. They were given a consideration period of no less than 7 days from receipt of the information pack to decide whether or not to participate in the study. During the initial consenting process, I made every effort to explain the consent form and ensure that the potential participant understood what was involved if they agreed to participate. Potential participants were informed of their right to withdraw at anytime without penalty. Refusals were accepted at face value. At each data collection time-point verbal consent to continue was obtained. Process consent was particularly important given the complex and longitudinal nature of the study. I was in contact with all participants on a number of occasions throughout the study and explained the study processes. For example, at the end of the initial discussion I ensured that participants understood the information sheet. I re-iterated what the participant could expect to happen next (i.e. receive the first round of questionnaires). At the end of the baseline (first round) data collection telephone call, I reminded participants that when the baseline postal questionnaires were returned, they would receive a letter informing them about which group they had been assigned to and what to expect in this regard. Verbal consent to proceed was obtained at all points of contact.

Privacy was maintained by minimising intrusion into participants' personal lives (Polit & Beck 2012). Participants determined the location for the interviews. They were always informed that a room in a location of convenience for them could be booked. This gave them the opportunity to keep their home private and limit any unnecessary pressure that could occur by a visit to their home. In addition, I was always cognisant of how I interacted with the participants. At all times, I ensured that I was polite and tactful in my interactions. At the beginning of each interview, participants were informed of their right to participate or withdraw. I reminded them of their rights, for example, to refuse recording or to ask for the recorder to be turned off at any point. Verbal permission to partake in the interview and to record was sought in advance. At the end of each interview, I thanked the participant, advised them that they could contact me if they had any concern or for clarification about the interview or study. If I felt that the participant was feeling pressurised during the interview, I informed them that it was okay if they could not think of anything right now and if they did think of something at a later time

that they could always let me know by telephone or in writing. All participants were advised of their right to review a transcript of their interview.

To uphold the right to autonomy participants must freely volunteer to partake in the study free from "... coercion, manipulation or inducement" (Dooley & McCarthy 2012, p.31). They should therefore not be forced or induced to consent to participate in the study and informed consent should be given free of any threat, harm or inducement (Sears 2001). During the pre-exploratory phase, potential participants were informed that they would receive a 15 Euro gift voucher per dyad at the second point of data collection, as a token of the researcher's gratitude for their commitment to the study. Given the time commitment required to complete the study, this was a small token that did not constitute an excessive or inappropriate reward. Following an independent academic review of the study by the funding body, where the reviewers recommended that the token be increased, the token of gratitude was increased to 30 Euro per study dyad. Each NSMS received a 40 Euro gift voucher for each SP in the intervention group. These small gift tokens were approved by the ethics committees governing two of the study sites. One site refused to grant approval for use of gift tokens. Instead, they required that the NSMS be paid for delivering the intervention outside her core working hours. The nurse specialists were viewed as research participants (in that their decision to get involved in the study was voluntary and I was studying their implementation of the intervention). In the sites where nurse specialists were permitted to integrate the study into their everyday practice, it was considered that the token of gratitude was sufficient reward for supporting the research study, yet not large enough to constitute an undue inducement. In the site where tokens of gratitude were refused, the NSMS was paid to do the study outside of her core working hours.

Confidentiality refers to research participants' right to have personal information kept private. Maintaining confidentiality obliges the researcher to ensure that personal information given to them as part of the research study is disclosed only to those that participants have consented to have access to it. This normally includes those involved with the research study and those who require access to confidential information. The researcher is also obliged to ensure that participants are not identifiable through any of the material that is shared with a third party, such as research reports and publications. Confidentiality can be maintained by anonymising all collected data as part of the

research study, thereby ensuring that individual's names are not linked to the study findings (Polit & Beck 2012). Any findings that could lead to the identification of individuals even after anonymisation of the data should be withheld by the researcher or presented in a manner that ensures the participant cannot be identified. Participants' right to confidentiality is enshrined in law and professional codes of practice and is important to build trust, protect participant privacy and enable the researcher to uphold the principle of autonomy (Dooley & McCarthy 2012).

In this study, participant privacy and confidentiality was maintained by adhering to the Data Protection Act of 1988 and 2003, which legally requires the researcher to ensure the safe storage and proper use of data obtained as well as maintaining confidentiality. The most secure method of maintaining participant confidentiality is by anonymising all data (Polit & Beck 2012). All participants were assigned a code number which was retained by me in a locked filing system. The code number was used for all subsequent stored records. I was the only person with access to all of the study data including the list of assigned code numbers. Nurse specialists were informed of participants who consented to participate in the study and were given their contact details by me in order to identify the correct person and they had access to their data in the usual manner. Research interviews were transcribed and anonymised using the participant code number. As PwMS and SPs know their code number there is a risk that they may recognise the content of the interviews if code numbers were used. Therefore, all code numbers were removed from direct quotes. Each site was assigned a unique study number known only to the researcher.

All hardcopy data were stored and will continue to be stored in a locked cabinet in a secured room until 5 years after study completion. All computerised data has been anonymised and password protected.

6.2.23.2 The principle of justice

Justice refers to participants' right to fair treatment and privacy and requires equal distribution of the benefits and burdens of the research (Polit & Beck 2012). This principle was upheld by recruiting participants based on the requirements of the study and randomly assigning them to the study groups. It was not necessary to include

those most susceptible to distress in order to test the potential benefits of this intervention. Therefore, on this occasion vulnerable groups were not invited to participant in the study. This did not violate what Polit & Beck (2012) term "distributive justice" (p.155), which is the duty to avoid neglecting or discriminating against those who may benefit from the research. Although support persons who have serious mental health issues may well have benefited from participating in this study, their inclusion in the study was not necessary in order to answer the research question. Should this intervention demonstrate positive findings in a healthy population, then it could be tested for use with a more vulnerable population in a follow-up study, involving a larger team of researchers with greater supports and resources.

The principle of justice was also upheld by ensuring that those who declined to participate or who withdrew from the study were not penalised in any way. As all patients were invited to participate and the nurse specialists were informed as to who agreed to participate, there was a risk that the nurse specialist would be aware of who declined to participate. However, this was limited by the fact that a dyad had to agree to participate and therefore the PwMS may be willing to consent but not be in a position to nominate a support person or the support person nominated may not be willing to participate. Nurse specialists were informed of these possible scenarios and this made it more difficult for them to identify who refused to participate in the study. In addition, although the nurse specialists were interventionists, they were also registered nurses and therefore their duty of care to the patient superseded all aspects of this research study. Although the nurses were participants, the research study was overseen by the principal investigator of the study. The study was not the nurses' priority, which meant that although they supported the study, they were unlikely to prioritise it over patient needs. Although I had a vested interest in ensuring the study progressed in terms of my own academic progression, my role as a registered nurse had to supersede the needs of the research study. This dual role raised an ethical issue when participants who initially responded to the study invite declined to take part in the study. This challenged me in terms of my need to recruit and retain participants in order to develop high quality evidence for practice (Polit & Beck 2012) and achieve my research award. This issue was limited by the fact that the study was an exploratory research study and this included exploring all the issues around recruitment and retention. Therefore, even

if participants decided not to participate following contact with me, or if they chose to withdraw during the study, this too contributed to the study findings.

6.2.23.3 Beneficence and non-maleficence

Beneficience refers to the duty to do good and, in research, this means maximising the benefits for participants (Polit & Beck 2012). Non-maleficence is the duty to prevent harm and minimise risks to participants (Polit & Beck 2012). Researchers are obliged to ensure that their actions and behaviours do not bring about harm. The overarching purpose of this study was to do good by testing an intervention that may help nurse specialists improve the support offered to PwMS and the people who support them. As the intervention was not previously subjected to testing and there were few examples of nurses delivering interventions of this nature, the risk to participants was unknown. Nurse specialists in MS offer support and advice via the telephone, as part of routine care when they are contacted by PwMS and their family members. Therefore, the risk to participants from advice and support offered by nurse specialists was not expected to be any greater than that which may occur during usual care. Usual care was not affected and the study provided an opportunity for SPs to have access to professional advice and support.

It was envisaged that the intervention might do good in that it may have positive effects on the lives of PwMS and SPs. The study included a component for teaching problem management skills to the support person at a level beyond routine nurse specialist practice. Previous research studies underpinned by problem management theories demonstrated positive outcomes (section 4.4.2), as did studies of the use of telephone support (section 3.6.2.2). I therefore considered the teaching of problem management skills to be more likely to bring about good than harm and that the risk to participants from the intervention itself was minimal. Despite this, it was essential to assess this intervention because although risk to participants may be minimal, there was uncertainty as to whether integrating pro-active nurse-led problem management telephone support to practice would be any better than usual practice. This uncertainty as to which of the treatments in a trial is more beneficial is known as clinical equipoise

and should exist before recruiting participants to a clinical trial (Djulbegovic 2009). Participants randomised to the intervention group could still access usual care and were therefore not denied access to support outside of the pro-active telephone calls. As this intervention had not previously been subjected to testing or use in any setting, it was unknown whether it was more favourable than usual care, the use of a 1:1 randomisation ratio was appropriate. This meant that all participants had an equal chance of being assigned to the more favourable treatment, if one existed. Given the uncertainty as to the benefit of the pro-active telephone calls alone, continued participant access to usual support ensured that they were not put at risk of injury or harm that may have occurred if they could not access the nurse specialist at times outside the pro-active calls.

People with Multiple Sclerosis who agreed to take part in the pre-and post-intervention interviews were at risk of becoming upset as they talked about their condition and their need for support. Support persons assigned to the intervention group of the study were at risk of becoming upset while thinking about their role as SPs and how this impacted upon their lives. These risks were not considered to be harmful and talking about problems can be beneficial. These anticipated risks and the participant's right to stop interviews or withdraw from the study were included in the information sheets (Appendix 24 and 26).

Polit & Beck (2012) highlight that research should be done by suitably qualified people who should be prepared to stop a study if it results in undue distress or other serious harm to participants. As the study progressed, I was vigilant about reporting adverse events to the steering committee and processes were put in place to ensure participant safety. Due to the novel nature of the intervention in nursing practice and lack of prior testing, the inclusion criteria helped to ensure that those most at risk of psychological distress (such as those with severe mental illness) were excluded from participating. In addition to subjecting a vulnerable group to potential unnecessary distress, the researcher had limited resources to support such individuals in the event of an adverse event resulting from participation in the study. For all other participants, I secured the support of a qualified counsellor. In two of the organisations, the resident psychologists agreed to provide this service, while funding was secured to offer this service in the third site, although it was not availed of by participants.

6.2.24 Trial management

In this study, trial management included the overall management and co-ordination of the study as well as site co-ordination at three sites. I attended work experience at the York Clinical Trials Unit in the UK, where I observed how experienced trial managers set up systems and dealt with challenges encountered in trial management. I also had the opportunity to get expert advice on various aspects of this trial and how best to manage them. These included database set-up in the absence of resources to fund building a study specific database, outcome data input and storage and the importance of not statistically analysing the outcome data during the study unless interim analysis was required. Where appropriate, I also availed of the opportunity to attend research team meetings and observed how principal investigators monitored trial progress and team members contributed and reported on various aspects of trial progress.

Although the nurse specialists provided some assistance with site trial co-ordination, overall responsibility and associated work rested with me. Despite this, maintaining good working relationships with staff in the sites (in particular the nurse specialists who were central to the study) was crucial to site retention and successful delivery of the intervention. I maintained contact with the nurse specialists by telephone and email. I met each nurse specialist at the study site at intervals that suited their work schedules. I also visited each site to deliver recruitment packs or sensitive information to the nurse specialists. I was always contactable on the study specific mobile telephone and was available to provide support and advice to the nurse specialists while the intervention was being delivered and in the immediate period afterwards.

As the trial manager, I sourced and learned how to use the equipment that was necessary for delivery of the intervention. This included head sets and recording devices that were compatible with the telephones at the study sites. I also provided training for the nurse specialists. In the role of trial co-ordinator and in the absence of administration support for the study, I arranged mutually convenient times for the nurse specialists and the SPs for delivery of the intervention. Agreed dates and times were communicated to both parties and email reminders/texts were sent to them on the day the calls were due to be delivered. On the day of the calls, I informed the nurse

specialists which calls were to be recorded and always made contact with the nurse specialist after delivery of the intervention to ensure that all went well. In addition, I posted all questionnaires and collected outcome data by telephone. I also set up dates and times for interviews and travelled to various parts of the country to carry out the interviews. As the study was longitudinal in nature, it was essential that the database was set-up to allow me to identify work that needed to be done on a daily and weekly basis.

6.2.25 Chapter summary

In conclusion, this chapter provided an overview of the application of the research methods to this exploratory trial. The lessons learned from pre-exploratory work informed the design of the exploratory phase, in particular the incorporation of elements in the process analysis before embarking on a full-scale randomised trial. The randomised trial that was used to test the intervention is a pragmatic trial, which enabled the testing of the intervention in the real world of practice. The concurrent process analysis enabled the monitoring of the quality of delivery of the intervention and the identification of contextual factors that influenced its application. It also provided a method of evaluating acceptability of the intervention to key stakeholders. Barriers and facilitators to testing the intervention using a randomised trial design were also identified.

In line with recommended practice, the results of the process analysis were analysed before the analysis of the trial component of the study. The results of this process analysis which include the findings on recruitment and response rate during the exploratory phase are presented in the next chapter.

Chapter 7: Findings and results

7.1 Introduction

The study findings and results are presented in this chapter. It is divided into three sections. In section one, the number of participants invited to take part in the study, their group allocation, the number of participants available to follow up and consequently, whose data were analysed, is presented. The baseline characteristics of the participants are presented and the characteristics of those allocated to the intervention group (IG) are compared to those allocated to the control group (CG). In section two, the findings from the process analysis are presented. The process analysis was carried out alongside the trial in order to determine the feasibility and acceptability of implementing the MNTISS and to identify potential barriers and facilitators to progressing the study to a full-scale trial. This included examination of the acceptability of the intervention to NSsMS, PwMS and their nominated support persons. The qualitative findings are presented under the headings of the key process evaluation components selected for evaluation in this study using the structure presented in chapter 6, table 14. Firstly, issues pertaining to recruitment are examined. This is followed by an overview of the context, reach, intervention fidelity, dose of the intervention delivered and received, acceptability of the intervention to the key stakeholders and overall satisfaction with the intervention. In the third and final section of this chapter, the results from the randomised trial component of the study are presented.

7.2 Section One: Recruitment, response and baseline participant characteristics

Recruitment strategies varied between the three research sites, yet the response rate and numbers who consented to take part in the study were relatively similar across the sites (Table 15). Outpatient Department (OPD) recruitment yielded a lower response

rate (6%), relative to the numbers invited, compared to sites that used a letter of invitation and statement of interest form, where responses were 20% for site 1, and 17% for site 3. The number of responses to invitations at the three study sites respectively was 44, 55 and 56. The numbers that consented to participate in the study across sites 1-3 respectively were 23, 24 and 34. Figure 9 presents the CONSORT flow diagram which provides an overview of the overall number of participants enrolled in the study, their group allocation, follow-up and analysis.

Table 15: Flow of recruitment at each site

Site	*Number invited/Number at site*	*Received letter of invite*	*Responded to invite* *Response proportion*	*Received full information pack*	*Consented to participate*	*Completed baseline data*	*Withdrew prior to randomisation*	*Assigned to Intervention and control groups*
One	280/360	280	55 (0.20)	30	24	24	0	I: 12 C: 12
Two	888/2000	N/A	56 (0.06)	888	34	27	2	I: 11 C: 14
Three	261/600	261	44 (0.17)	44	23	23	1	I: 10 C: 12
Total invited to participate	1429		155	962	81	74	3	I: 33 C:38

I (intervention), C (control)

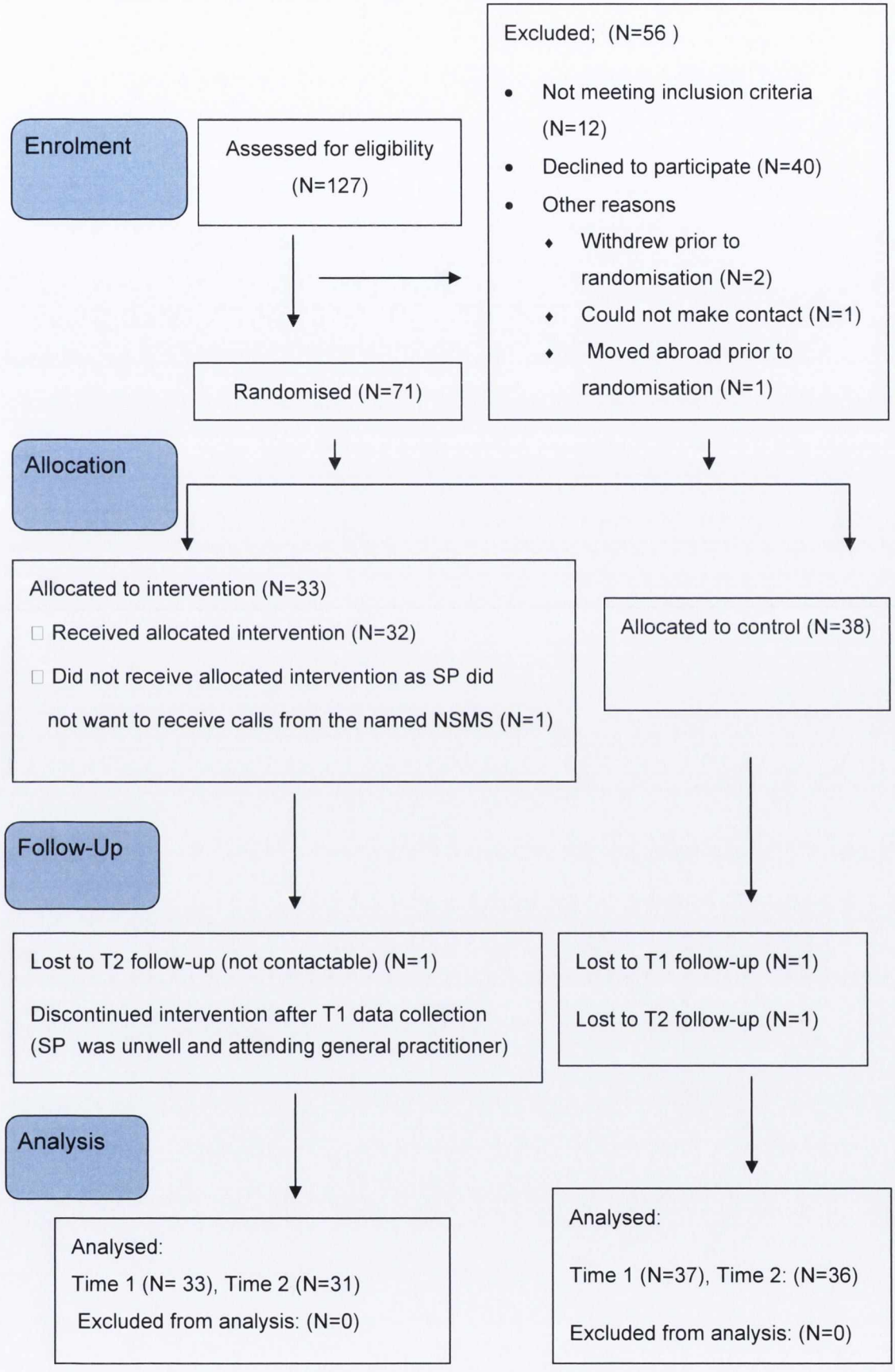


Figure 9: Consort 2010 flow diagram

7.2.1 Reasons for non-participation in the study

Fifty four PwMS and 2 SPs returned the statement of interest form with their contact details stating that they did not wish to participate. Of those who responded 39 provided reasons for not participating in the study, one self-excluded on the basis of the inclusion criteria and no reason was given by 10 respondents. Six PwMS were interested in the study but contact could not be made with their SPs (Table 16).

Table 16: Reasons for non-participation in the study

	Site 1 (N=28)	**Site 2 (N=7)**	**Site 3 (N=21)**
Self-excluded on the basis of inclusion criteria	1	-	-
No time/a lot of other things going on at the time of the study	1**	1**	7**
No support person to nominate	6	1	2
Did not feel the study applies to them	12	-	3
SP was not interested in participating	1	1	3
SP does not like doing research studies	–	1	
No reason given	3 2 **	-	5
Researcher could not make contact with SP	2	3	1

**** Response was from SP**

7.2.2 Baseline biographical data

Tables 17-20 present the baseline characteristics of the intervention and control groups for SP and PwMS. Significant differences were found between the intervention and control group SPs in terms of education [Intervention group > number educated to third level than control group], the time spent caring and the length of time supporting PwMS: the control group had been providing support for significantly greater number of years and spent more time in a 24 hour period supporting the PwMS, than the intervention group. The physical health subscale of the MSQoL was the only subscale that demonstrated a significant difference between the two groups at baseline: the intervention group reported better physical health QoI compared to the control group at baseline.

Table 17: Demographic characteristics of dyads at baseline

Demographic variable	Overall group (N=70)		Intervention (n=33)		Control (n=37)		*p* value	
	SP	PwMS	SP	PwMS	SP	PwMS	SP	PwMS
Age (years+/- SD)	51.34 (13.37) Range 22-84	50.3 (11.37) Range 25-71	48.88 (13.43) Range 25-78	49.61 (12.78) Range 25-70	53.54(13.11) Range 22-84	50.91(10.15) Range 29-71	t-test *p*=0.147	t-test *p*=0.633
Gender Male Female	33 (47.1%) 37 (52.9%)	19 (27.1%) 51 (72.9%)	15 (45.5%) 18 (54.5%)	11 (33.3%) 22(66.7%)	18(48.6%) 19(51.4%)	8 (21.6%) 29(78.4%)	Pearson's Chi-Square *p*=0.789	Pearson's Chi-Square *p*=0.271

SP=support person; PwMS=person with multiple sclerosis ***This result shows a significant correlation at $p \leq 0.05$**

Table 18: Biographical data for support persons

Variable	Overall	Intervention	Control	*p* value
Education				
Primary	5(7.1%)	1(3.0%)	4(10.8%)	Fisher's
Secondary	36(51.5%)	13(39.4%)	23(62.2%)	Exact:
Tertiary	29(41.4%)	19(57.6%)	10 (27.0%)	*p*=0.024*
Marital Status				
Single	12(17.1%)	5(15.2%)	7(18.9%)	
Married	50(71.4%)	24 (72.7%)	26 (70.3%)	Fisher's
Living as married	2(2.9%)	1 (3%)	1 (2.7%)	Exact:
Separated/Divorced/	3(4.3%)	1(3%)	2(5.4%)	*p*=0.756
Widowed	3(4.3%)	2 (6.1%)	1 (2.7%)	
Time/24hour period supporting PwMS (hours+/-SD)	8.814 (9.04)	5.63 (6.68)	11.29 (9.22)	t-test: *p*=0.020*
0-4 hours	22 (33.3%)	13 (41.9%)	9(25%)	
5-10 hours	8 (12.1%)	2 (6.5%)	6 (16.7%)	
> 10 hours	21 (31.8%)	7 (22.6%)	14 (38.9%)	
Number of years supporting PwMS	11 (7.69)	8.35 (5.95)	13.18 (8.36)	t-test: *p*=0.013*
Living arrangements				
Live in	49 (69)	20 (57.1)	29 (80.6)	Fisher's
Live out	13(18.3)	10 (28.6)	3 (8.3)	Exact *p*=0.035*
Employment				Pearson's
Paid employment	36 (51.4)	18 (51.4)	18(50)	Chi Square
Not employed	25 (35.3)	11 (31.5)	14 (38.9)	*p*=0.526
Time spent in paid employment (hours per week)	Range 6-90 ≤ 20 (n=7) > 20- ≤40 (n=21) >40 (n=6)	Range 14-45 ≤ 20 (n=3) > 20- ≤40 (n=13) >40 (n=2)	Range 6-90 ≤ 20 (n=4) > 20- ≤40 (n=8) >40 (n=3)	t-test *p*=0.296

***This result shows a significant correlation at $p \leq 0.05$**

Table 19: Biographical data for PwMS

Biographical variable	Overall Group	Intervention (n=33)	Control (n=37)	*p* value
Type of MS Relapsing/remitting Secondary progressive Progressive relapsing Primary progressive Benign MS	 37 (52.9%) 30 (42.9%) 1 (1.4%) 1 (1.4%) 1 (1.4%)	 18 (54.5%) 14 (42.5%) N/A N/A 1 (3%)	 19 (51.4%) 16 (43.2%) 1 (2.7%) 1 (2.7%)	Fisher's Exact p=0.973
Months diagnosed: mean (SD) (N=69)	162 (112.78) Range 14-540	138.18 (85.07) Range 14-360	178.21 (131.88) Range 24-540	t-test: p=0.134
Disability (Other) Walking Uses stick Wheelchair Missing	 49(70%) 11(15.6%) 9(13%) 1 (1.4%)	 23 (69.7%) 6 (18.2%) 4 (12.1%) N/A	 26(70.3%) 5(13.5%) 5(13.5%) 1 (2.7%)	Fisher's Exact p=0.931

SP=support person; PwMS=person with multiple sclerosis, *This result shows a significant correlation at $p \leq 0.05$

Table 20: Baseline PwMS (Quality of life as measured by MSQoL and impact of MS as measured by MSIS)

PwMS QoL and impact of MS data	Overall Group (N=68 unless otherwise stated)	Intervention (n=33 unless otherwise stated)	Control (n=35 unless otherwise stated)	*p* value for t-test
MSQoL-subscales				
Physical Health	46.52 (33.14)	55.30 (33.11)	38.47 (31.48)	0.034*
Role limitations due to physical problems	34.42 (42.68	38.64 (44.23)	30.55 (41.45)	0.436
Role limitations due to emotional problems	65.7(41.99)	63.64 (42.79)	67.59 (41.77)	0.699
Pain	65.77(24.73)	64.17 (23.09)	67.27 (26.39)	0.606
Emotional well-being	65.78 (64.73)	69.58 (19.13)	72.89 (16.51)	0.433
Energy	71.30 (17.75)	46.42 (19.77)	44.86 (22.08)	0.759
Health perceptions	45.60 (20.87)	49.92 (19.67)	47.07 (23.23)	0.672
Social function	48.77 (21.47)	66.29 (22.54)	65.28 (22.58)	0.853
Cognitive function	65.76 (21.04)	70.15 (20.10)	75.83 (21.79)	0.266
Health distress	66.37(23.82)	68.33 (21.42)	64.58 (26.00)	0.518
Sexual function	63.14(33.49) (n=63)	61.30 (35.93) (n=29)	63.23(31.99) (n=34)	0.822
Change in health	43.47 (18.52)	43.94 (17.71)	43.06 (19.47)	0.845
Satisfaction with sexual function	50.00 (32.95) (n=62)	49.14 (34.38) (n=29)	50.76 (32.16) (n=33)	0.849
Overall quality of life	66.30 (19.27)	67.32 (17.33)	65.37 (21.10)	0.678
Physical Health Composite Score	53.21(20.07) (n=63)	56.54 (18.69) (n=29)	50.26 (21.09) (n=34)	0.219
Mental Health Composite Score	68.64 (18.82)	67.66 (18.73)	69.54 (19.12)	0.681
MSIS	69.1 (23.87)	65.3 (21.7)	73.0 (25.5)	0.178

SP=support person; PwMS=person with multiple sclerosis; MSQoL = multiple sclerosis quality of life scale; MSIS=multiple sclerosis impact scale. *This result shows a significant difference between intervention and control at $p \leq 0.05$

7.3 Section two: Process analysis findings

Data recorded in my log of events and interviews with NSsMS, nominated SPs and PwMS informed the process analysis. The findings from the content analysis of these data sources are presented under the headings of the framework for the process analysis (chapter 5, Table 15): recruitment, context, reach, fidelity, dose delivered, intervention delivery, dose received, acceptability of the intervention and satisfaction with the service.

7.3.1 Trial recruitment

Issues pertaining to site and participant recruitment emerged during the early stages of the study. At the start of this study, it was anticipated that more than one site may be needed because of the time commitment required from each NSMS. However, denial of access to the PwMS at site 5 (the site originally selected for pilot work), meant that the site originally planned for the exploratory trial was used for the pre-exploratory phase. Continued challenges to the recruitment of PwMS and SPs during the early stage of the exploratory phase contributed to the need to seek ethical approval and access in all five sites.

The process of seeking site ethical approval and access was incremental and the final number invited to participate was based on success in obtaining nursing or medical access at the sites and NSMS consent. The challenges encountered included denial of access to PwMS at site 5, the unanticipated early retirement of a NSMS at site 1 due to a Health Service Executive incentive for early retirement to reduce the workforce, and non-receipt of NSMS consent at a site with full ethical approval, nursing access and medical consultant approval to proceed (site 4). Although site 1 was used for the pre-exploratory testing of the intervention, it was agreed with my primary supervisor that given the challenges with gaining access to additional sites, that the exploratory stage could also proceed at that site. As the NSMS at site 1 was not being replaced immediately and her replacement might not have had the skill required to deliver the intervention, a request was made to the NSMS to continue to deliver the intervention

following retirement. She agreed to do this and ethical approval, medical approval and site management approval was obtained to facilitate this.

Obtaining ethical approval and access at site two was a lengthy and challenging process. The novel nature of the intervention and study design within the Irish nursing context raised concerns regarding the potential impact of the study on service delivery. This resulted in one NSMS having to deliver the intervention outside of core working hours, requiring re-imbursement for her time and additional protocol changes including the inclusion of a suspected abuse protocol (Appendix 30) and the need for me to provide funding for psychological support services, if required during the study. I addressed all of the committee concerns and requested amendments to the protocol based on pre-exploratory challenges. Final ethical approval was granted in December 2011 and the amendments were approved on 12th January 2012. Despite this, a further delay with recruitment of the interventionist was encountered due to the method agreed for payment of the NSMS. The agreed method of payment posed problems for the department in terms of staff number returns, which in turn impacted on staffing and funding for the department. Following further discussions and a meeting with the Director of Nursing and others, a solution was agreed and the ethics committee was informed. Despite having obtained approval to access the NSMS from the Director of Nursing in June 2011 and final site ethical approval in January 2012, challenges remained in terms of NSMS receipt of the information pack and NSMS consent was not obtained until May 2012.

7.3.2 Factors that impacted on participant recruitment during the exploratory phase

The acceptability of the OPD and database recruitment methods varied across the sites. The NSsMS at sites 1 and 3 found the database recruitment to be acceptable but the NSMS at site 3 had difficulty getting the time to address the envelopes. When asked about OPD recruitment, the NSMS at site 2 did not report any concerns. This may be because OPD recruitment did not cause additional work for her, because the receptionist at that site was giving PwMS the information packs. The NSsMS differed

in their views of OPD recruitment (see comments page 240). Contextual factors, such as the research culture at the site within which systems were in operation to assist with recruitment, and other aspects of trial work impacted on the acceptability of OPD recruitment. At site two, clinical trials were ongoing and the centre had systems in place that supported research, including recruitment. A member of the administration team distributed the information packs to PwMS on attendance at the site. The NSMS at site 1 found OPD recruitment cumbersome and felt that some PwMS did not receive the study invite because not all of them were reviewed by the NSMS. No administrative support was available for recruitment purposes at this site.

From my perspective, database recruitment was the more efficient method of recruitment. Fewer resources were used because full information packs were sent to those who expressed interest only. In addition, the use of database recruitment using a letter of invitation and SOI form yielded greater expression of interest from the participants. Database recruitment was, however, labour intensive in sites where no database or a clean database did not exist. The database approach required the NSsMS to create clean databases and address the envelopes. Due to lack of administrative support and the requirement to maintain patient confidentiality, this increased the NSsMS' time commitment to the trial, beyond that originally planned. Despite these challenges with recruitment, similar numbers of participants responded to the study invites and were recruited from both methods. Table 21 presents a summary of the overall findings pertaining to recruitment and retention in this phase of the study.

Table 21: Summary of overall findings on issues pertaining to recruitment and retention

Recruitment	Outcome	Explanation
Sites **Participants** **Method** **Enablers/barriers to recruitment** **Enablers/barriers to retention**	• 3 of 5 sites recruited agreed to participate • 4 of 5 NSsMS consented to participate • Sites: convenience sampling • Participants via PwMS: Consecutively based on a database using letters of invite or attendance at OPD clinic using full information packs <u>Enablers:</u> • use of database and letter of invite <u>Barriers:</u> • no pre-developed database at some sites • Staff change or shortage • Participants perception of the study • Study inclusion criteria • <u>Enablers</u> Organisational support from management, having agreed systems in place prior to commencement of the study <u>Barriers</u> NSMS life events, NSMS other work commitments, time commitment required for intervention delivery, number of NSMS at a site, requirement to provide cover for other specialties during staff leave/absences	In one site the Consultant neurologist did not consent to access to PwMS. The second site, the NSMS did not return the consent form. No nominated support person contact details available. Requirement to get PwMS consent prior to inviting family member. Proportion of responses higher for the letters of invite (0.17 and 0.20) compared to proportion for pack (0.06). Overall participant recruitment was low relative to the number of participants invited (see table 15). Overall study retention was high. All of the NSsMS who began delivering the intervention completed the delivery to those recruited at the site. At one site recruitment stopped earlier than intended due to staff shortages and NSMS life events.

The following comments from NSsMS provided insight into factors that were likely to have impacted upon recruitment. The NSMS at site 2 found aspects of OPD recruitment such as having the opportunity to explain the study to the PwMS and answer any questions they may have to be helpful. This opportunity was not always present as not all PwMS met with the NSMS during OPD appointment.

> "...I'd only see a small portion of those patients in the clinic because there's so many of them that would be coming out with their envelopes, or coming in to me with the envelopes...you'd explain the study further where other team members mightn't have been as aware of the study or felt they were able to explain it....you know that way? (NSMS, site 2).

Many of the potential participants that may meet the study inclusion criteria did not routinely attend the OPD.

> "There's a proportion of patients that mightn't come for a couple of years, when they are in trouble again they get in contact". (NSMS, site 2).

The NSMS at site 2 felt that distributing the full information pack at the site did not adversely affected recruitment.

> "...no I don't think it would make a major difference because the fact that they would have taken the information home with them so either way they would have time to reflect on it, you know that way, whether it was sent to their home or they brought it out of the clinic with them..." (NSMS, site 2).

The reasons given by the NSMS for factors which may have negatively impacted on recruitment included the restrictive nature of the inclusion criteria, research fatigue, patient concerns regarding confidentiality and the NSMS work commitments.

The exclusion of PwMS with cognitive impairment was considered a factor by the NSMS at one site:

> "...patients who are cognitively impaired their carers are the ones who ring the most. Because they have to be their memory and their support. ..., I think it would have been beneficial to have people actually with cognitive impairment included in the study. Because their carers require a much larger element of support" (NSMS, site 2).

> "...it's not only memory with cognitive impairment, there also can be behavioural problems, behavioural changes and that's a huge caregiver burden..." (NSMS, site 2).

The NSMS at that site also expressed concern regarding PwMSs' confidentiality

> "... in some context it could have been the patients themselves, ...being afraid of what they might say about them. .....I got a bit of that impression in the clinic as well ..." (NSMS, site 2).

Research fatigue was also a factor which the NSsMS felt may have impacted upon recruitment.

> "...because this is a research centre in MS, ...patients are called in to do various outcome measures ...of various studies over the years ...particularly the patients that are coming more long term. They get asked to do quite a lot, you know" (NSMS, site 2).

> "I'm honestly, not sure,...was it the paper work, we've done, ..satisfaction surveys and that before, and our return hasn't been great. So is it something to do with that, I don't know. ... (NSMS, site 3).

> "...there were some people who said to me in the clinic, oh yeah I received that information from Margarita Corry, I

haven't answered it yet. And I think sometimes it's just, people just don't, I don't think there's anything in particular about the set up with the research study or whatever, I think people just didn't bother" (NSMS, site 3).

At one site, staff shortages (for both administration and NSMS) was a major factor that impacted on sending out the letters of invitation and this eventually led to early cessation of recruitment at that site:

"Was it time or was it patients, it was both of them really, I think initially because, ... a colleague wasn't replaced, it was just timing and it was covering other services.... It could have gone on for longer and we probably could have recruited more but... from my own time perspective it was just impossible" (NSMS, site 3).

"No administrative back up. I mean you did offer that you could give somebody money to do it but ... and I had to get somebody who worked within the neurology field and I had asked 3 separate secretaries but none of them were interested in doing it" (NSMS, site 3).

"It literally was because our service is so busy" (NSMS, site 3).

7.3.3 Reach

The number of people in Ireland and at the individual research sites who provide support to PwMS is unknown. It is therefore not possible to determine the proportion of the target population that received the intervention. Assuming that each PwMS has a support person, the proportion of the population of PwMS registered at the research sites for this study, whose SP received the intervention was 2.3% (n=33). The majority of support persons recruited to the intervention agreed to participate and the overall

attrition rate from the intervention group was low (n=2) out of 33 who were randomised to this group.

The expectation of gaining some benefit from participating in the study and a desire on behalf of nominated SPs to be helpful were the top two reasons that SPs assigned to the intervention group, gave for agreeing to participate (Figure 10). The majority of those assigned to the control group wanted to be helpful (Figure 11).

The acceptability of the OPD and database recruitment methods varied across the sites. The NSsMS at sites one and three found the database recruitment to be acceptable but the NSMS at site 3 had difficulty getting the time to address the envelopes. When asked about OPD recruitment, the NSMS at site two did not report any concerns. This may be because OPD recruitment did not cause additional work for her, because the receptionist at that site was handing out the information packs to PwMS. The NSsMS differed in their views of OPD recruitment (see section 7.3.2 for comments from NSMS). Contextual factors such as a site research culture within which systems were in operation to assist with recruitment and other aspects of trial work impacted on the acceptability of OPD recruitment. At site two, clinical trials were ongoing and the centre had systems in place that supported research, including recruitment. A member of the administration team distributed the information packs to PwMS on attendance at the site. The NSMS at site one found OPD recruitment cumbersome and felt that some participants were missed because not all PwMS were reviewed by the NSMS. No administrative support was available for recruitment purposes at this site.

From my perspective, database recruitment was the more efficient method of recruitment. Fewer resources were used because full information packs were sent to those who expressed an interest only. In addition, the use of database recruitment using a letter of invitation and SOI form yielded greater expression of interest from the participants. Database recruitment was, however, labour-intensive in sites where no database or a clean database did not exist. The database approach required the NSMS to create clean databases and address the envelopes. Due to lack of administrative support and the requirement to maintain patient confidentiality, this increased the NSMS time commitment to the trial, beyond that originally planned.

Despite these challenges with recruitment, similar numbers of participants responded to the study invites and similar numbers of SPs were recruited using both methods.

Recruitment bias is an issue of concern for both methods of recruitment. This was evident when one of the two NSsMS operating database recruitment decided not to give an information pack to one PwMS because her condition had deteriorated so much in the recent past. Likewise, when setting up the database, if the PwMS are known to the person creating the database, a PwMS who may be eligible for inclusion could be excluded. This may occur for a variety of reasons, for example, if the person creating the database has had previous negative encounters with the PwMS or their SP.

Several factors emerged throughout the study that may have impacted upon recruitment, including PwMS and SPs identification with the role of caregiver and perceived personal relevance of the intervention. Following the pre-exploratory phase of this study, the term 'caregiver' was replaced by the term 'nominated support person'. During the early stages of recruitment, a number of participants indicated that they did not have a caregiver and that they were "...able to look after themselves". After explaining the purpose of the study and the meaning of terms such as 'caregiver', the PwMS appeared more receptive of the study and the fact that their partner or spouse or others may benefit from the study. When potential participants were invited to participate in the study, they were informed of a number of unknowns about the intervention, including who was most likely to benefit and how they might benefit from it. Many potential participants (both PwMS and SPs) were unsure of the relevance or potential benefit of the intervention to themselves. When it was explained that this was a new support intervention and part of the purpose of the study was to determine who might be most likely to benefit from receiving it, participants were better able to understand the relevance of the study and some said that they could now understand how they could contribute.

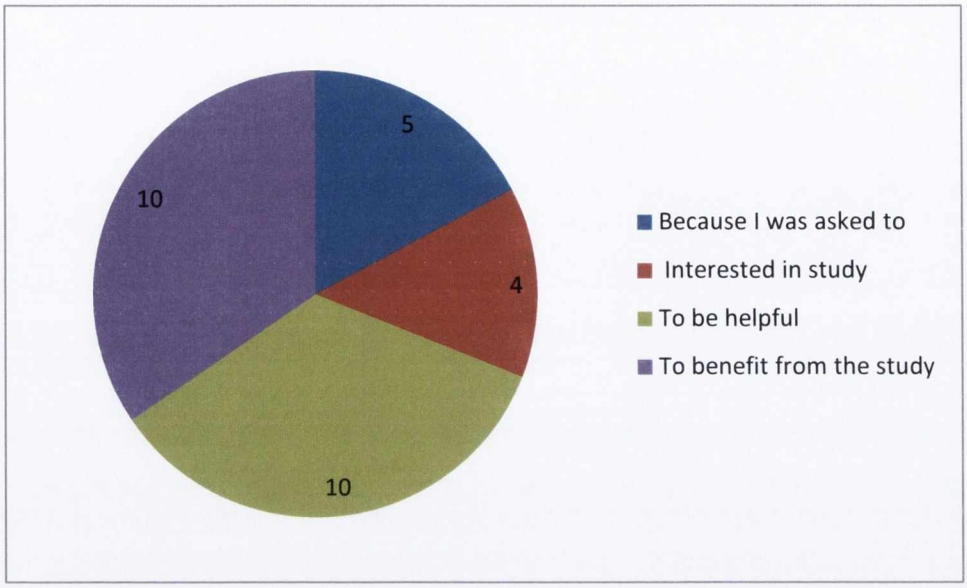


Figure 10: Nominated SPs' reasons for participating in the study (Intervention group)

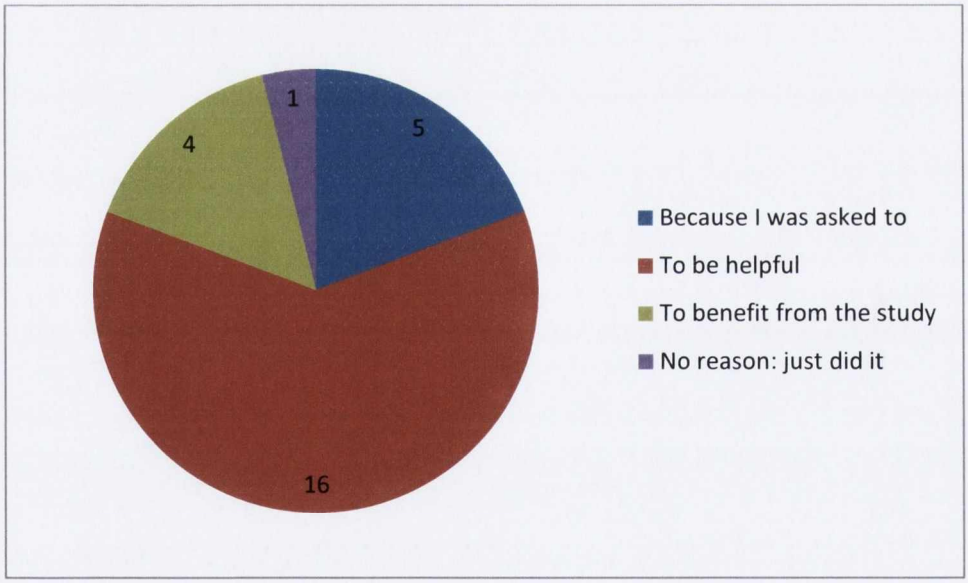


Figure 11: Nominated SPs' reasons for participating in the study (control group)

7.3.4 The impact of context on the delivery of the intervention

Contextual factors that impacted on the delivery of the intervention included the human resource and support available for delivery of the intervention and the study. Delivery of the intervention by the NSMS was restricted to a designated evening once a month in site one and two, and two evenings of a particular week per month in site 3. Scheduling calls for the evening time suited the majority of SPs but there were occasions when this was difficult to arrange due to SPs work schedules. Although the NSsMS were flexible in most instances in terms of arranging the calls outside of the day and time scheduled, this often meant the call was delayed by a week or two because of NSMS work commitments or scheduled leave.

Having one NSMS at each site to deliver the intervention meant that the study calls were at times delayed due to unexpected work commitments or when NSsMS were absent. Life events for either the NSMS or nominated support person impacted on the delivery of the intervention. For example, at one site the NSMS had to take two months leave due to family commitments, which meant that calls could not be scheduled during that time. In another instance, the nominated SP was pregnant and her baby was delivered pre-maturely. This delayed the delivery of the final call by a further 6 weeks, which had already been delayed due to scheduling problems. These contextual factors and life events resulted in protocol violations in terms of call scheduling, and this led to a delay in data collection.

The availability of experienced NSsMS impacted positively on the delivery of the intervention. All NSsMS in the study were qualified nurses for over 20 years, two had over 10 years experience working as a NSMS, and one was in the role for 9 years. They reported that their background and experience enabled them to deal skillfully with issues raised by the SPs.

> "...I feel I had the experience to make the suggestions ... to take things on board. .... I'd say if I had a been twenty [*years old*] doing this intervention, I wouldn't have had a clue" (NSMS site 1, post intervention interview).

"...you are experienced in a job and deal with persons with MS and all sorts of aspects of MS your experience and knowledge obviously, is a help...?" (NSMS site 2, post intervention interview).

7.3.5 SP group assignment preference

The intervention and control group did not differ significantly ($p>0.05$) in terms of their preference for study group, with similar numbers having no preference across the two groups [n=24 Intervention and n=25 control]. Slightly more participants in the control group (n=11) had a preference to receive the intervention compared to the intervention group (n=9).

7.3.6 Intervention fidelity

Fidelity to the intervention was analysed according to the NSsMS adherence to the principles underpinning the intervention, the appropriateness of the content to the purpose of the calls, the training of the interventionists, the intervention dose delivered and received, and call tracking.

7.3.6.1 Fidelity to the intervention

In this study, there was evidence from both the recorded intervention calls and the notes maintained by the interventionists that the NSsMS adhered to the principles of Egan's Model of the skilled helper and, where possible, used an experiential approach to problem management. It was also evident from the recorded content of the intervention calls (section 7.3.6.2) that the conversations were primarily focused on the concerns of the SPs and any additional conversation material, which was minimal, served to strengthen the therapeutic relationship between the NSMS and SPs, and afforded opportunities for the SP to raise their concerns. For example, at the start of a

follow on call the NSMS asked about the trip away, which was discussed in a previous call. This enabled the NSsMS to create continuity between the calls, indicating that content discussed in the previous call was noted and thus afforded the SP the opportunity to raise any issues that emerged during that time. The longitudinal nature of the study and the skills of the NSsMS provided an opportunity for a trusting relationship to be built during the first intervention call and this may have enabled the SP to discuss sensitive issues during the second or third call. The content discussed during the calls was appropriate to the overall goal of the intervention and the issues identified support adherence to the principles underpinning the intervention.

From an analysis of the recorded interviews, there was evidence of application of elements of the skilled helper as in Egan's Model of the Skilled Helper, such as active listening, processing and responding with empathy (Egan 2010). There was also evidence of probing and summarising, assistance with problem management, education and professional advice on how to problem manage/solve. The NSMS referred to the SP's guide book, went through the problem management process and where appropriate offered professional advice and assistance within the scope of the role of the NSMS.

It was also evident from listening to the recorded calls that, although the principles underpinning the intervention were adhered to in general, it was not always done in a consistent manner. There were times during the calls that the application of the problem management process could be strengthened by having greater continuity between calls, less repetition within and across calls, and the taking of opportunities to teach problem management. Analysis of the delivery of the intervention provided evidence of a learning curve, with the NSsMS demonstrating greater familiarity with the process, more confidence, and greater adherence to the principles underpinning the intervention over time. While some repetition was appropriate in that it emphasised important points, unnecessary repetition was less evident when the SP had an important issue to address. Following my feedback to the NSsMS following call monitoring, NSsMS self-evaluation by listening back to the calls recorded and increased experience of delivering the intervention, there was a decrease in unnecessary repetition. At the post-intervention interview, one NSMS said that after the first call with each person it became more comfortable to deliver the intervention

and after the first five calls "things were fine" (NSMS site 3). In general it was evident from the intervention interviews and my experience of running the trial, that once the NSMS became comfortable with delivering the intervention, greater fidelity to the intervention was evident when the SP had a relatively important issue to resolve.

7.3.6.2 Appropriateness of call content

The content of the calls was appropriate in that they focused on issues and problems that the SP voiced (Table 22). Some important SP concerns did not emerge until the second or third call. For example, one SP revealed an alcohol and depression problem in the second call while another SP revealed issues pertaining to fertility in the third call. The NSsMS were not always aware of the existence of these issues before the intervention calls. The issues discussed during the calls were categorised in terms of a) the emotional needs of the SPs which included concerns regarding their relationship with the PwMS/other family members and worries about the future, and b) issues pertaining to PwMS which included concerns about the services on offer, impact of MS on their relationship with the PwMS, the symptoms of MS, treatments for MS and work related issues. The analysis of the intervention call content also revealed that the advice offered by the NSMS was related to the issues raised by the SPs and the purpose of the calls. Some comments from the post-intervention interviews highlighted the learning or action taken by the SPs while in the study.

Table 22: Overview of the call content of intervention calls (from recordings and nursing notes)

Category of call content	Sub-category of call content (number of participants and number of comments made)	Sample content	Exemplar
The emotional needs of the support person	Impact on relationships (4 participants, 5 comments)	With partner/spouse With children With PwMS	Wife of PwMS (SP) feels she is on a different wavelength to PwMS. Providing care for PwMS who was a parent impacted on SPs relationship with partner
	Worries for future (4 participants, 18 comments)	Worried Stressed Bereavement issues	Support person looked it up [referring to medication option] and worried about it in case it makes MS worse. NSMS identified slight anxiety problem for both SP and PwMS.
	Other SP concerns (12 participants, 37 comments)	Varied across participants	Concerns regarding media cutbacks Fear of speaking about future with PwMS

Issues pertaining to PwMS	MS related services (5 participants,10 comments)	Allowance Cut backs Services available Issues raised pertaining to services around current care	Support person was concerned regarding cutbacks as reported in the media- PwMS disability worsening. Concerned that PwMS will not receive adequate care package NSMS offered education regarding supports if required (website, contact numbers etc)
	Relationship issues (2 participants, 4 comments)	No subcategories	Pregnancy issue including in vitro fertilization (IVF) PwMS's relationship with partner Changed relationship with PwMS due to carer role
	Symptoms(11 participants, 41 comments)	Sleep Mood and related issues Fatigue and Mobility Elimination Cognitive issues Others	 PwMS's behavioural issues (other family member confirms deterioration), paranoia Fatigue, restless legs at night PwMS not picking up information about his condition or what to do as part of therapy. A range of issues such as dealing with travel insurance for the PwMS

	Treatments (e.g. medications, other options) (4 participants, 11 comments)	Medications Other issue pertaining to treatment	Spoke about medication change. PwMS had MRI, both worried about side effects of medications. Felt that present treatment was not working. PwMS continues to refuse some treatment e.g. to be seen in neurology clinic.
	Work related issues (4 participants, 5 comments)	Work related worries No longer able to work or unemployed	PwMS stresses over financial issues Full recovery not made, now unable to farm.
Action	NSMS advice (13 participants, 35 comments)	Advice pertaining to services (e.g. on how to or to contact a service) Advice on managing aspects of PwMS care	Advised where to find details for MS care worker Advised SP to send in respite form to respite care centre. Advised regarding travel insurance, thermal socks and gloves Advice given on managing fatigue Advised to read SP guidebook and identify any questions for discussion at next call. Mum reads lots of articles – questionable content e.g. babies in April more likely to get MS

		Advice on preparing for the next call Caution regarding sources of information Encouragement and self-care issues	Advised regarding importance of checking information from internet. Encouraged to think outside box- what to do if help withdrawn. NSMS re-assured SP regarding an issue relating to blood tests.
	Progress gleaned from interviews (3 participants, 13 comments)	Positive steps forward SP did not take action	Feels that being in study helped look at future. PwMS more positive about respite. PwMS made contact with MS Ireland, IWA and voluntary group locally so is looking for help which helps SP. PwMS improved. Is now aware of support services available Did not attend GP.

7.3.6.3 Training of the interventionists

In general, the NSsMS reported that the intervention manual, which included the training manual, was clear and comprehensive. One NSMS felt that it was probably "...a bit too comprehensive" and would have liked a shorter version.

> *"Something more simplistic ... and shorter you know ...but I think that might have been part of the whole ethical protocol for it as well..., it has to be very comprehensive. ...the laminated stuff for the prompts was very good.* (NSMS site 2)

Two NSsMS reported that the length of time given to training on the use of the intervention was adequate, but the other NSMS felt that she would have liked a little more training, or to have it closer to the time of delivery of the intervention. Even though additional training was offered to all NSsMS on a one-to-one basis, they did not avail of this offer. Training was delivered once recruitment commenced at the sites, so that the NSsMS were ready to deliver the intervention when the participants were recruited. The length of time from training to first call varied across sites and was dependent on the time it took to recruit and complete baseline data collection. In site one time from training to 1st intervention call was 6 months, site 2 (3 months) and site 3 (4 months). All NSsMS reviewed the training manual prior to the 1st intervention call. Refresher training was offered but they felt that they were ready to deliver the intervention. I reminded them about the core principles underpinning the intervention and of the importance of using the SP guidebook during the calls.

Although the interventionists felt that they had received adequate training in this study, feedback following call monitoring was required to ensure that the problem management process was being addressed during the intervention calls. The NSsMS demonstrated the ability to listen, give advice and help SPs resolve a problem when one existed. It was, however, only with time that the NSsMS began to embed the teaching of the problem management cycle into the intervention calls. The intervention was new to all involved, including me, and therefore full knowledge of what was required in order for the NSsMS to be competent with delivery of the intervention was unknown before commencement of the study. There was evidence of a learning curve,

with NSsMS becoming more confident and consistent in the application of the intervention over time.

7.3.6.4 Intervention dose delivered by the NSMS and received by SPs

Almost all the SPs (n=31/33, 94%) received the four intervention calls from the NSsMS. One participant withdrew following the first two calls. One SP did not receive their third call, which had been arranged but the SP was not contactable on the relevant evening. Table 23 presents the reported receipt of the calls based on the participant recall. Although one SP could not recall when the calls were delivered, the NSMS and my record of call delivery confirms that four calls were delivered. The average length of the intervention calls at the sites was 17.5, 20 and 23 minutes in the three sites respectively (range across sites 4 to 60 minutes)

Table 23: SPs responses to fidelity questions

	Intervention	Control	Intervention	Control
	Yes	**Yes**	**No**	**No**
Received the 2 calls in first month	31 (91.4%)	N/A	1 (2.9%)	N/A
Get the next two calls	29 (93.5%)	N/A	1 (3.2%)	N/A
Received other training on problem managing during the study	0%	7 (20%)	30 (96.8%)	27 (77%)
Received other help with managing problems in supporting PwMS	0%	1 (2.9%)	30 (96.8%)	34 (97.1)
Did the MSNS make initial contact with you at any time during the study?	N/A	0	N/A	33 (94.3%)

Intervention group: One intention to treat participant not included in analysis

7.3.6.5 Intervention delivery (tracking)

The intervention calls were not always scheduled per protocol. Delay in the delivery of calls varied from 1 week to 6 weeks. Calls were frequently delayed by at least 2 weeks due to problems scheduling a suitable time for the NSMS and nominated SP. Some calls were cancelled and rescheduled at short notice, mainly due to the NSMS availability. The reasons for delay in scheduling the calls were mainly due to NSMS annual leave, sick leave, and workload for the week that the call was scheduled to be delivered. The availability of SPs due to work commitments or holidays was also an influencing factor. Other less frequent reasons included last minute cancellation due to unexpected work commitments for the NSsMS on the day or a few days prior to the scheduled call. Last minute cancellations and arranging the calls for times outside of

the scheduled call time period, created additional administration work for me as the process had to be replicated repeatedly.,

7.3.7 Acceptability of the intervention

In general, the intervention was acceptable to the majority of the participants. This was evident in the analysis of the findings for the intervention group in relation to the study retention rate, perceptions of the helpfulness of the calls, overall satisfaction with the service offered and perceived effectiveness of the intervention.

It was clear that the intervention was acceptable to PwMS who took part in the study as the majority of respondents were positive about the study and consented to take part. However, some PwMS or their nominated SPs did not feel that they were in need of this type of support at the time of invitation and therefore declined to participate. Others stated that they did not have a person to nominate.

7.3.8 Study retention

Study retention was high in both groups. In the intervention group, one person assigned to the intervention group was retained for data collection but did not receive the intervention. Following randomisation to the intervention group she voiced concerns about taking the calls for the NSMS at that site. Another person withdrew from the study after the second call. Other indicators of acceptability were generated from the interviews with the nominated SP, PwMS and nurse specialists. The issues that emerged include the participants' experiences of being in the study including the perceived helpfulness of the intervention, their perceptions of its appropriateness and perceived benefits of being in the intervention group.

7.3.9 Perceived helpfulness of the calls from the NSsMS

Figure 12 presents the number of intervention participants who perceived the intervention calls to be helpful or unhelpful. Most of the nominated SPs (n=24) made over 60 comments that indicated that the calls from the NSsMS were helpful or

extremely helpful. Six SPs made 9 comments that suggested that the calls were unhelpful.

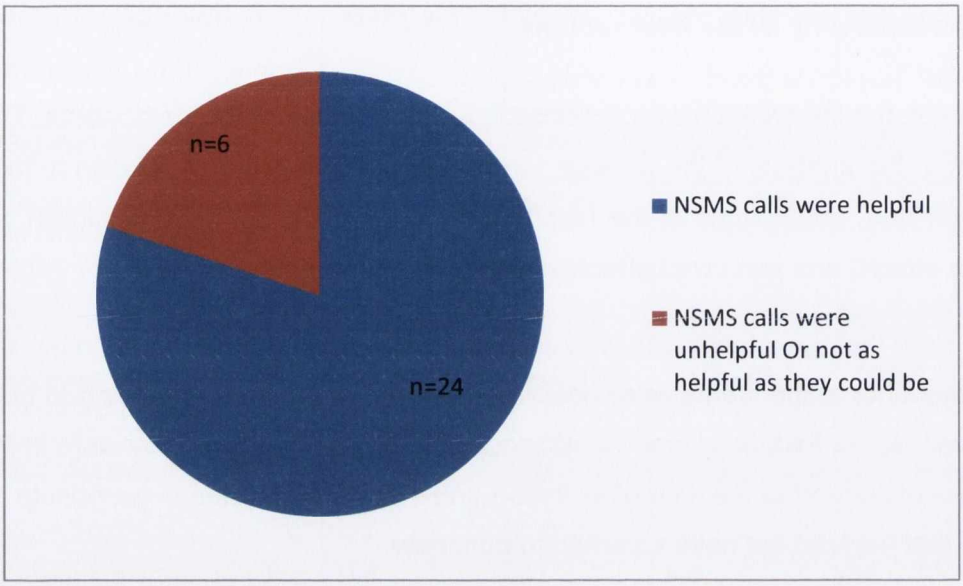


Figure 12: SPs perception of the helpfulness of the calls from the NSsMS

The following comments provide a brief overview of participants' perceptions of helpfulness of the calls.

> "*...it has taken a load off my shoulders and I know that if ever I am stuck ...all I have to do is pick up the phone and I can phone up the NSMS for advice*" (Nominated SP).

> "*...it was great ... talking things over with NSMS, it definitely helped*" (SP).

> "*...but she helped just identify the tools that are out there for us to use and made sure that you know that when I do need to reach out I will be able to find them quickly enough*" (SP).

> "*But that makes him feel better. It reassured him*". (PwMS).

Comments from the support persons who did not find the calls helpful include:

> "*...I didn't need help so therefore NSMS questions weren't of any relevance to me*" (SP)

> "*...at the time I wasn't in great need of support because [Name of PwMS] was in such good health, the NSMS was fantastic though, she was very grounded, lots of common sense...*" (SP)

> "*...depend on what phase you are and [name of PwMS] has been relatively stable during the period of the calls and there's not been anything particular concerns ...I'd say if we were going through some sort of crisis period, they would have been much more valuable.* (SP)

7.3.10 Acceptability of the intervention to the key stakeholders

Findings pertaining to the acceptability of the intervention to key stakeholders (NSsMS, SPs and PwMS) were gleaned from the post-intervention interviews.

7.3.10.1 Acceptability of the intervention to the NSsMS

All three NSsMS felt that the support intervention should continue because they felt that some SPs were in need of their help. They felt that the intervention needed to be streamlined. Their comments included

> "In an ideal world yeah it would be great but its time constraints ...I think it's necessary actually" (NSMS site 2)

> "It's also recognising the benefit of it and the insight it gives me into the person as well. So it would be something certainly worth continuing...There is a huge amount of people out there who I would say are in the same

situation as the ones involved in the research study.... it's certainly needed, there's no doubt about it" (NSMS site 3)

"I think, yeah, probably because there is a lot of people looking after somebody who may not have, who may have issues or problems or things they just need to get off their chest and they don't know where to go with it.... It's very hard for you to approach somebody sometimes...whereas if contact is made from somebody to the person who is caring I think maybe it might allow them to express their own feelings and fears about it" (NSMS site 1).

"I think it still allows them to know that if an issue arises that they have somebody to contact ...or they have a way of managing that problem themselves, if they can't contact somebody they now have a framework to use. But if it doesn't work then they know they can contact us" (NSMS site 3).

For an intervention to be acceptable to practitioners and participants it has to be feasible to deliver and of relevance to the people who receive it. The NSsMS provided insight regarding the changes required and the challenges of implementing the intervention in practice (Figure 13).

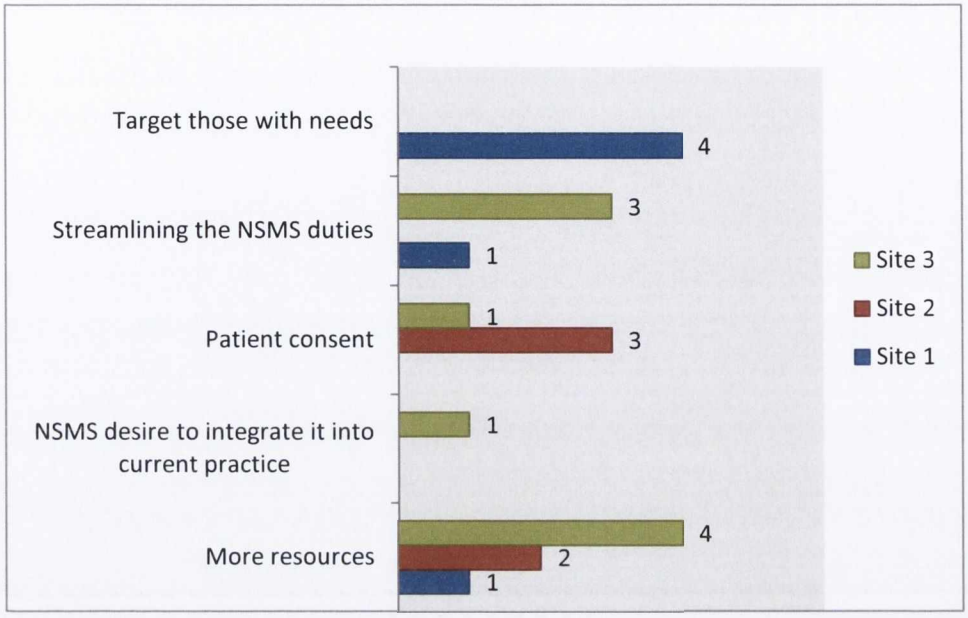


Figure 13: NSMS comments (by site) regarding what is required to integrate the intervention into practice

In order to implement an intervention of this nature in practice, the NSsMS raised issues pertaining to resources, administration support and an environment conducive to maintaining confidentiality. Two NSsMS (site 1 and 2) mentioned the need for more time and more NSsMS. The NSsMS at site 2 and 3 highlighted the restrictions caused by having to share offices and how that impacted on confidentiality and privacy. They said that it was sometimes difficult to get a quiet space to carry out the intervention calls, especially if the call was scheduled during core working hours. For this reason, evening calls were more suitable for the NSsMS because other staff with whom they shared an office were likely to have left. On a few occasions, however, evening calls were cancelled because the NSsMS could not locate a quiet and private room to carry out the study. The need for administration support was highlighted by the NSsMS at site 1 and 3. None of the NSsMS had administrative support for their role and, in order for the intervention to be integrated into practice, one NSMS suggested streamlining of duties by the appropriate delegation of administrative duties and having a structure in place including protected time for the role.

> "If you had, I think if I had a certain evening in the week that I could allow for that" (NSMS site 3).

> "And setting up a phone time and a service for provision of this which I think I need to look at..., you know because when you have a set time ..., you're not ringing somebody because they've left a message and you're rushing through a phone call, ..., it allows you to listen to them. And it allows you to [make] proper suggestions....Whereas if you're rushing to a phone call because you're looking at your diary and it's the end of the day and I need to answer that phone call, it's not a proper service" (NSMS site 3).

This NSMS did feel that the service was needed

> "... So while feasibility, it will be difficult but I do think it's necessary though" (NSMS site 3).

The NSsMS were mixed in their responses as to who to target (Figure 14), with one NSMS feeling that it should be targeted to both PwMS and nominated SPs. This NSMS felt that it was a flexible intervention that could be applied in a number of settings. She commented (off recorder) that no training is given for the provision of telephone support and that this intervention provided structure. All NSsMS said that the intervention was suitable for SPs of people who were newly diagnosed, those who have issues to manage, or at particular times in the stage of MS where the care needs of the PwMS changes, such as when medications have been reviewed and changed.

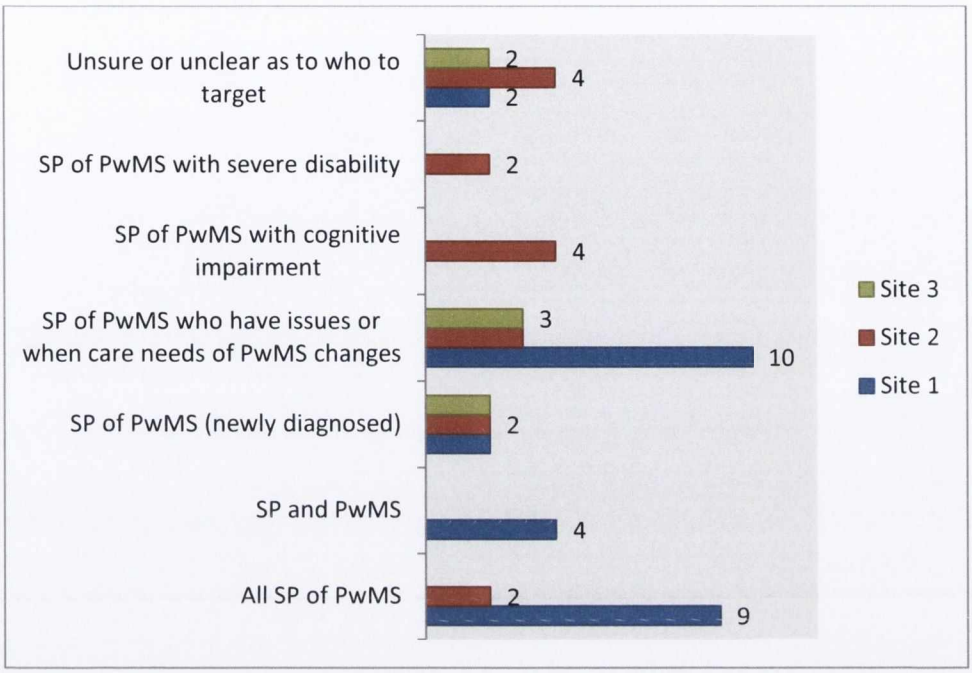


Figure 14: NSMS comments (by site as to who should receive the intervention)

7.3.10.2 Acceptability of the intervention to SPs and PwMS

The acceptability of the intervention to the SPs and PwMS was evident in the comments made in relation to the effectiveness of the intervention and their view on the adequacy of the number of calls. The fact that the majority of participants felt that 4 calls were the right number suggests that they felt the calls were necessary. The following provides a sample of the comments made by SPs regarding the adequacy of the number of calls from the NSMS.

> "For my case anyway it was spot on, enough time in-between to actually, ask kind of reflect, on what you [the nurse specialist] talked about and then maybe ask NSMS or do research and things so that next time you are talking you actually have the time to do something about it and the intervals were short enough that you didn't forget

about things that you did, you had enough time but it was also that you weren't just left alone too long and you'd end up just forgetting about it and the momentum would be lost" (SP)

"I think they were. Yes four is enough. Once you can work through a problem. Like I did have a problem at the start and I was speaking to NSMS about it and at each phone call she just asked how were we getting on and you know, has the problem got any better or has it got worse. She just followed up on it and I suppose I thought that was very helpful. So you know that helps a lot". (SP)

"Yea I think so, anymore seem a bit intrusive or something" (SP).

Others felt that they could have done with more calls (n=4 comments) or that it depended on the circumstances (n=8 comments). Five participants (9 comments) felt fewer calls were required. Two felt that 2-3 calls would have been sufficient, while two felt that they had no issues to discuss and therefore one or two calls would have been sufficient.

The majority of the SPs (n=22) felt that it was important that the nurse delivering the intervention was the one providing care to the PwMS. Although some SPs stated that any nurse or nurse specialist could deliver the intervention (Figure 15) they qualified this by stating that they would need to have the required knowledge.

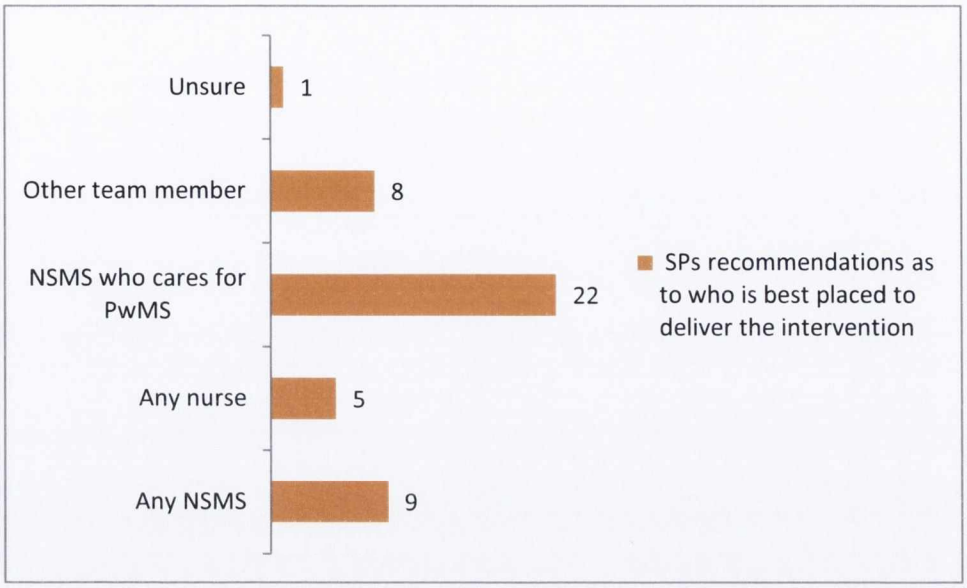


Figure 15: Who is best placed to deliver the intervention?

None of the participants found taking part in the study to be intrusive or time consuming. One person in the control group stated that she would have liked to be in the intervention group because she wanted "to experience getting some support and see what that was like" (SP).

7.3.11 Perceived effects of the intervention

People with MS and SPs indicated that many positive benefits were received from participating in the intervention (Figure 16). The most frequently reported outcomes of receiving the intervention for nominated SPs were feeling more knowledgeable (n=8), having an opportunity to discuss their concerns (n=8) and feeling supported (n=8). Some MS participants said that they initially found it difficult to identify how they or their SPs had benefited from the telephone support intervention. Most of them (n=10) would have initially stated that they didn't notice any change. However, as the interview progressed they mentioned that they had noticed changes such as the SP being more understanding towards them (n=7), being less worried or more relaxed (n=3) and

confident (n=3). Two PwMS commented that because the SP was being supported that was helpful to them.

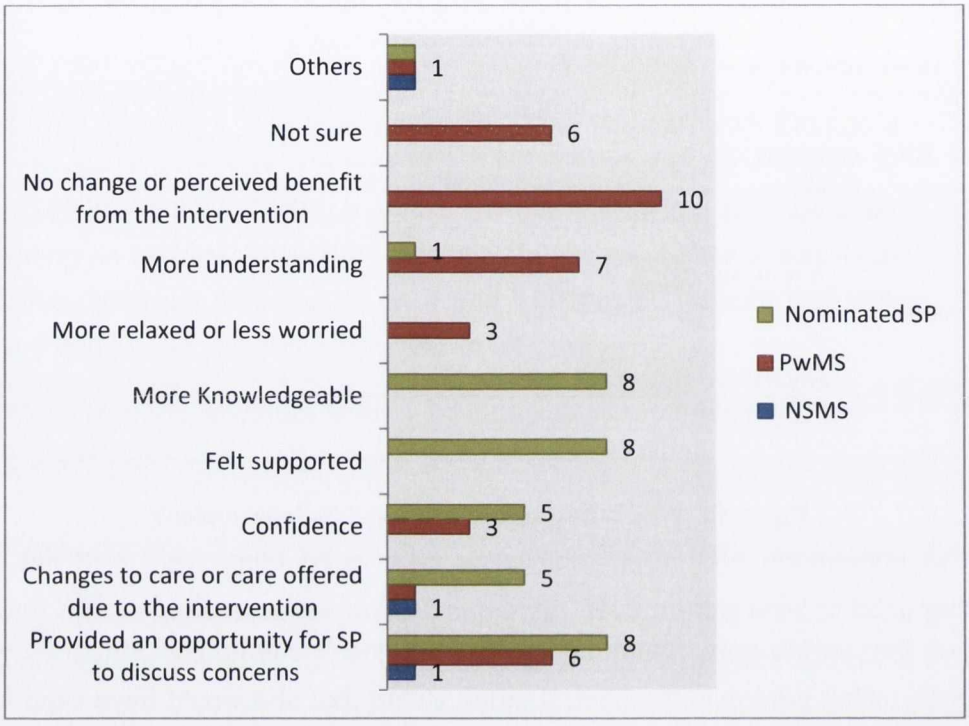


Figure 16: Participant reported outcomes (NSMS (n=3), SPs (n=25), PwMS (n=15)

The SPs views on contacting the NSsMS changed as a result of being in the study. The majority (n=21) felt that they would make contact with the NSsMS sooner as a result of being in the study (Figure 17). A number of respondents were of the view, prior to taking part in this study, that the NSMS was only available to support the PwMS. This differed from the perception of the NSsMS who generally felt that the SPs were aware that they could contact them if needed.

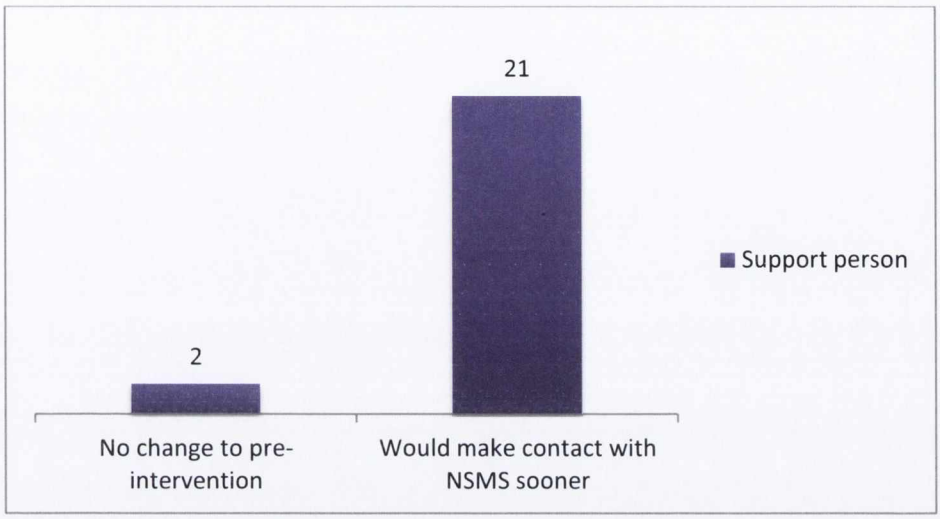


Figure 17: SPs view of contacting the NSMS post-intervention

Evidence of SP benefit from the intervention was volunteered by 7 SPs during the post-intervention interviews. They recounted the changes made to the care offered by the NSsMS, for example referral to services, advice given or information sent by post. Further information regarding the type of support provided by the NSsMS was evident in the analysis of the recordings of the phone calls between the NSsMS and the nominated SPs and the records maintained by the NSsMS. It was evident from the calls that the NSsMS provided general advice to the SPs regarding services available and how to access the services. They also provided advice to the caregiver in relation to their own worries about future care needs of the PwMS and their own self-care needs.

The following post-intervention interview comments from NSsMS and SPs highlight their views on the benefits of participating in the intervention.

Sample comments from NSsMS

> "Since the research, through talking with her sister [the SP] ...and the sister [the SP] herself looking at it as well, she has started a FAS course. ...And is now seeing a counsellor as well. It's made a huge change and it is as a result of the research that this has happened" (NSMS site 3).

"...they're just 2 examples but there are more. ...I'm not saying everyone is coming back and saying oh that we solved problems, but I suppose they were 2 specific ones which I felt were ....considerable and they've made a big, big difference" " (NSMS site 3).

Sample comments from SPs

"You know she [the NSMS] also sent out a book specifically for (name of PwMS)'s young son ..., dealing with... children with parents with MS so... All that was very helpful". (SP)

"Oh, extremely so, in fact we had a physical meeting with [Name of NSMS] on the basis of those conversations and one of the things she did was arrange this session tomorrow in (name of Hospital) to get this pain-killing injection, ...it's been very proactive, ...it's got its desired result" (SP).

"...things were getting quite stressful ...until all this [phone calls with NSMS] started. ...it wasn't in my head when I started this, that we were going to kind of approach that problem but as it turned out it did and (name of NSMS) was very helpful in getting us ...looking at relationship stuff" (SP).

7.3.12 Risks

The NSMS at site 3 raised concern regarding patient confidentiality. One PwMS phoned this NSMS and raised concerns about what may be revealed to the SP during the intervention calls. During the post-intervention interview, the PwMS told me that although she had consented to take part, she was worried that details of every visit, such as being upset during a consultation, would be revealed to the SP during the

intervention call. She told me at the interview that the NSMS had re- assured her that this would not happen, when she contacted her. From the post-intervention interview, it was evident that while the PwMS could see the value of the SP talking to the NSMS to address her particular concerns, that she (the PwMS) wanted a certain amount of privacy and confidentiality maintained regarding her care. Some aspects of care were considered more personal than others. For example, the PwMS stated that there was no problem with the SP knowing what her pain score was but that she would have difficulty with her diet or body weight being divulged to the SP, although she had no issue with the concept of weight and diet being discussed with the SP in a more general way. In relation to confidentiality the SP commented:

> "...I think permission is needed, I do think that. I don't know, it's a hard one, isn't it? I think it depends on the situation, doesn't it, and how bad someone's MS is. I definitely think if things are reasonably okay I think confidentiality should be taken into account whereas if... say if my MS was bad and I wasn't, I was refusing to talk to my SP about it, yeah I think maybe in some cases like that, I don't know, maybe a line needs to be crossed a little bit, does it? (PwMS)

There were two reported incidents that came to light during data collection. A 'SP' contacted me before randomisation to indicate withdrawal from the study because the PwMS became upset when completing the baseline questionnaires.

The second incident related to a SP whom the NSMS could not make contact with for a scheduled intervention call. Following the second call from the NSMS, I contacted the SP in question, had collected outcome data and agreed a date for the third call. There was no evidence of distress or concern and I awaited return of the postal questionnaires but the SP did not respond to the NSMS calls or my follow up calls. Around two weeks later the PwMS phoned the researcher in confidence and said that the SP was unwell and attending the GP. The SPs behaviour had changed and this had been picked up in the work area. I offered to provide psychological support but the PwMS stated that the SP was attending the GP and work place counselling services were available. I thanked the PwMS for calling and as no further contact was to be

made by me, I thanked her and the SPs for participating in the study. The NSMS had identified the SPs as someone who was anxious and excessively worried about what could happen in the future. This is an important finding as it has implications for study inclusion criteria for future testing of this intervention. The incident was reported to the research advisory committee and following analysis of the event, it was not deemed to be a study adverse event. Following this event, a protocol to deal with non-response to intervention calls was devised (Appendix 31).

7.3.13 Satisfaction with the intervention

The response options for the Client Satisfaction Questionnaire (CSQ-8) ranged from 1-4, with higher scores indicating greater satisfaction. Each item had a different response descriptor, and the means and standard deviations are presented in table 24.

Table 24: Outcome of CSQ-8 (satisfaction with the service) questionnaire

Questionnaire items (1-8)	N	Mean (SD)
How would you rate the quality of the service you received?	31	3.61 (0.56)
Did you get the kind of service you wanted?	30	3.63 (0.49)
To what extent has our service met your needs?	30	3.46 (0.50)
If a friend were in need of similar help, would you recommend our service to him or her?	31	3.83 (0.45)
How satisfied are you with the amount of help you received?	31	3.70 (0.59)
Have the services you received helped you deal more effectively with your problems?	31	3.61 (0.56)
In an overall, general sense, how satisfied are you with the service you received?	31	3.64(0.55)
If you were to seek help again, would you come back to our service?	31	3.90(0.40)

Note: number varies from 30-31, depending on missing data.

The results indicate high satisfaction with the intervention. Most participants rated the quality of the service received as good (n=10) and excellent (n=20). One participant rated this item as fair. Respondents reported that in general they received the kind of service they wanted [yes generally (n=11), yes definitely (n=19)]. They reported that either most of their needs had been met (n=16) or that all of their needs were met (n=14). Almost all would recommend the service to others in similar need [Yes, definitely (n=27), Yes, I think so (n=3)]. Only one respondent stated, No, I don't think

so. Most (n=24) were very satisfied with the amount of service they received, five responded that they were mostly satisfied and two were indifferent or mildly dissatisfied. Similarly, participants reported that in general they were very satisfied with the service (n=21), mostly satisfied (n-9) and indifferent or mildly dissatisfied (n=1). If they were to seek help again, the majority would come back to the service [Yes, definitely (n=29), Yes, I think so (n=1) and No, I don't think so (n=1)].

7.4 Section 3: Findings from the trial outcome questionnaires

Tables 25 to 30 present the results from the quantitative data collected. Table 25 gives the mean scores at all three data collection time-points and the t-test result for the difference between the intervention and control at baseline, for the self-efficacy in problem-solving and the preparedness to care questionnaires. No significant differences between the two groups on these two outcome questionnaires were found at baseline.

Table 26 gives the within and between group differences time 1 and time 2 data collection points. Preparedness to care was found to significantly differ both within group and between groups. The intervention group showed statistically significant within group difference between baseline and time 1 (p=0.004), baseline and time 2 (p=<0.001) and time 1 and time 2 (p=0.005). The difference between the intervention when compared to the control group was also statistically significant at time 2 data collection point (p=0.025).

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Table 25: Self-efficacy for problem management, preparedness to care (prepsum) mean score for each time-point and T-scores for baseline between group analyses

MNTISS Self-efficacy Problem Management PREPSUM	Baseline Mean (N)		Time 1 (6weeks) Mean (N)		Time 2 (12 weeks) Mean (N)		Difference between (I) (C) at baseline Independent t-test value p-value
Intervention (I) Comparison (C)	*I*	*C*	*I*	*C*	*I*	*C*	
Self-efficacy Problem Management	73.85 (33)	75.35 (37)	75.55 (32)	73.78 (37)	72.29 (32)	73.40 (36)	0.656, 0.514
PREPSUM Sum of Items mean (SD)	2.04 (33) 16.61 (SD=6.58)	2.32 (37) 18.54 (SD = 6.63)	2.45(33) 19.41 (SD=5.94)	2.39(36) 19.28 (SD=6.07)	2.63 (31) 21.03 (SD=5.51)	2.41 (36) 18.94 (SD= 6.35)	-1.420, 0.160
*This result shows a significant difference between intervention and control at p≤0.05							

Table 26: Self-efficacy problem management and preparedness to care (prepsum) mean scores for each time-point and T-test scores for baseline between group analyses

MNTISS Self-efficacy Problem Management PREPSUM BKOS	Difference between (I) (C) at time 1 ANCOVA F, *p*-value (CI)	Difference between (I) (C) at time 2 ANCOVA F, *p*-value)	Difference between baseline and time 1 Paired t-test value *p*-value		Difference between baseline and time 2 Paired t-test value *p*-value		Difference between time 1 and time 2 Paired t-test value *p*-value	
Intervention (I) Comparison (C)	F,p	F,p	I	C	I	C	I	C
Self-efficacy Problem Management	($F_{(1,66)}$=0.430, 0.514)	-0.168,0.868 ($F_{(1,65)}$=0.028, 0.868)	-0.359, 0.722	0.609, 0.546	0.565, 0.576	0.610, 0.546	0.838, 0.409	0.355, 0.725
PREPSUM	$F_{(1,66)}$=1.271, 0.264	$F_{(1,64)}$=5.282, **0.025*** (0.047-0.666)	-3.071, **0.004***	-0.609, 0.546	-3.915, **<0.001***	-0.642, 0.525	-3.006, **0.005***	0.045, 0.656

***This result shows a significant difference either between intervention and control at p≤0.05 or a significant change within each group over time at p<0.05**

Table 27 and 28 gives the within and between group differences at time 1 and time 2 data collection points for the WHOQoL. Baseline scores on the WHOQoL total score were found to significantly differ between the groups, with the intervention group reporting statistically significant lower initial QoL scores than the control group (p=0.049) (Table 28). This between groups difference was not sustained over time nor was there a significant change between groups over time. Significant within group differences were however noted between baseline and time one scores for domain 4 (environment) of the WHoQoL with the intervention group reporting statistically significantly increased quality of life in this domain (p=0.042). This difference was not sustained at time 2 data collection point despite a continued increase in mean score on this domain (Table 28).

Tables 29 and 30 gives the within and between group differences at time 1 and time 2 data collection points, for the CRA. Significant differences were found within groups for CRA subscales but no significant differences were found between the two groups. Significant within group differences were found with the control group reporting significantly increased burden scores for four of the CRA subscales [Disrupted schedule, Lack of family Support, Health problems, Impact of caregiving on self-esteem] between baseline and time 1 data collection. This increase was also found to exist between baseline and time 2 data collection for one of these subscales [Impact of caregiving on self-esteem] (Table 30)

Table 27: WHOQoL scores for each time-point and t-test scores for baseline differences between groups

MNTISS Four subscales of WHOQol and WHOQoL total.	Baseline Mean N		Time 1 (6weeks) Mean N		Time 2 (12 weeks) Mean N		Difference between (I) (C) at baseline Independent t-test value *p*-value
Intervention(I) Comparison (C)	I	C	I	C	I	C	
WHOQoL domain 1	16.14 29	16.90 35	15.93 31	16.44 35	16.28 31	16.72 35	-01.152, 0.254
WHOQoL domain 2	15.11 30	15.66 35	15.31 31	15.44 35	15.63 31	15.47 35	-0.864, 0.391
WHOQoL domain 3	14.43 31	14.07 35	14.37 31	14.97 35	15.14 31	15.2 35	-0.725, 0.471
WHOQoL domain 4	14.92 31	14.93 35	15.16 31	14.63 35	15.73 31	15.01 35	-0.012, 0.990
WHOQoL Total	25.52 31	25.94 35	25.94 31	25.94 35	25.94 31	25.91 35	-2.006, **0.049***

domain 1: Physical health; domain 2: Psychological health; domain 3: Social relationships, domain 4: Environment

***This result shows a significant difference between intervention and control at p≤0.05.**

Table 28: WHOQoL scores for within and between group analysis

MNTISS	Difference between (I) (C) at time 1 ANCOVA F, *p*-value	Difference between (I) (C) at time 2 (ANCOVA F, *p*-value	Difference between baseline and time 1 Paired t-test value *p-value*		Difference between baseline and time 2 Paired t-test value *p-value*		Difference between time 1 and time 2 Paired t-test value *p*-value	
Intervention (I) Comparison (C)	F,*p*	F,*p*	I	C	I	C	I	C
WHOQoL domain 1	$F_{(1,61)}$=0.009, 0.924	$F_{(1,63)}$=0.371, 0.959	0.766, 0.450	1.733, 0.092	-0.325, 0.747	0.531 0.599	-1.043, 0.305	-1.042, 0.305
WHOQoL domain 2	$F_{(1,62)}$=0.331, 0.567	$F_{(1,62)}$=1.656, 0.203	-0.519, 0.608	0.783, 0.439	-1.275, 0.212	0.802, 0.428	-1.095, 0.282	-0.083, 0.934
WHOQoL domain 3	$F_{(1,63)}$=-0.063, 0.802	$F_{(1,63)}$=0.543, 0.464	0.202, 0.841	0.275, 0.785	-1.716, 0.096	-0.340, 0.736	-1.967, 0.059	-0.613, 0.544
WHOQoL domain 4	$F_{(1,63)}$=2.159, 0.147	$F_{(1,63)}$=3.033, 0.086	-1.048 0.303	0.923, 0.362	-2.123, **0.042***	-0.250 0.804	-1.837, 0.076	-1.493, 0.145
WHOQoL Total	$F_{(1,63)}$<0.001, 0.989	$F_{(1,63)}$=0.123, 0.727	-1.895 0.068	0.000 1.000	-1.895 0.068	0.373 0.711	0.373 0.711	0.373 0.711

domain 1: Physical health; domain 2: Psychological health; domain 3: Social relationships, domain 4: Environment

***This result shows a significant difference either between intervention and control at p≤0.05 or a significant change within each group over time at p<0.05.**

Table 29: CRA mean scores for each time-point and t-test for baseline between group differences

MNTISS Five subscales of CRA	*Baseline* *Mean* N		*Time 1 (6weeks)* *Mean* N		Time 2 (12 weeks) *Mean* N		*Difference between (I) (C) at baseline* *Independent t-test value* *p-value*
Intervention (I) *Comparison (C)*	*I*	*C*	*I*	*C*	*I*	*C*	
CRA subscale 1	2.47 33	2.38 37	2.31 33	2.63 36	2.45 31	2.48 36	-0.424, 0.673
CRA subscale 2	2.97 32	3.07 37	2.71 33	3.03 36	2.77 31	2.87 36	-0.422, 0.675
CRA subscale 3	2.29 33	2.11 37	2.24 32	2.27 36	2.38 31	2.25 36	1.090, 0.280
CRA subscale 4	2.17 33	2.03 37	2.14 33	2.29 36	2.28 31	2.04 36	0.776, 0.440
CRA subscale 5	3.93 33	3.92 35	3.85 32	3.75 36	3.85 31	3.81 35	-0.077, 0.939

subscale 1:Disrupted schedule; subscale 2: Financial problems; subscale 3: Lack of family Support, subscale 4: Health problems, subscale 5: Impact of caregiving on self-esteem, ***This result shows a significant difference between intervention and control at p≤0.05.**

Table 30: CRA (F and T-test scores for between and within group analysis)

MNTISS Five subscales of CRA	*Difference between (I) (C) at time 1* ANCOVA F, *p*-value	*Difference between (I) (C) at time 2* (ANCOVAF, *p*-value)	*Difference between baseline and time 1* Paired t-test value *p*-value		*Difference between baseline and time 2* Paired t-test value *p*-value		*Difference between time 1 and time 2* Paired t-test value *p*-value	
Intervention (I) Comparison (C)	F,p	F,p	*I*	*C*	*I*	*C*	*I*	*C*
CRA subscale 1	$F_{(1,66)}$=6.71, 0.12	$F_{(1,63)}$=0.371, 0.545	1.406, 0.169	-2.090, **0.044***	0.217, 0.830	-0.808 0.424	- 1.019, 0.316	1.152, 0.258
CRA subscale 2	$F_{(1,65)}$=2.08, 0.154	$F_{(1,63)}$=0.069, 0.793	- 3.071, 0.053	0.424, 0.674	1.161, 0.255	1.441, 0.158	- 1.282, 0.210	0.755, 0.455
CRA subscale 3	$F_{(1,64)}$=2.34, 0.131	$F_{(1,64)}$=0.002, 0.964	0.62, 0.541	-2.129, **0.040***	-0.735, 0.468	- 1.394, 0.176	- 1.597, 0.121	0.569, 0.573
CRA subscale 4	$F_{(1,66)}$=4.559, 0.36	$F_{(1,64)}$=1.872, 0.176	0.389 0.700	-2.824, **0.008***	-1.067, 0.294	0.000 1.00	- 1.361, 0.184	2.639, **0.012***
CRA subscale 5	$F_{(1,64)}$=1.59, 0.212	$F_{(1,61)}$=0.681, 0.412	1.22, 0.23	2.562, **0.015***	0.839 0.408	-2.184 **0.036***	0.186, 0.853	- 0.061, 0.952

subscale 1:Disrupted schedule; subscale 2: Financial problems; subscale 3: Lack of family Support, subscale 4: Health problems, subscale 5: Impact of caregiving on self-esteem, ***This result shows a significant difference either between intervention and control at p≤0.05 or a significant change within each group over time at p<0.05**

7.5 Chapter summary

Worry about the future was the main concern raised by SPs in this study. The NSsMS helped SP of PwMS to address a number of issues pertaining to their emotional needs including how the support role impacted upon their relationships and dealing with their worries about the future. Other areas of concern were usually linked to the services available, the emergence or deterioration of the PwMS's symptoms, change to their medication or other treatment changes. Where appropriate the NSsMS provided information, advised on services available or arranged services for the SP or PwMS (7.3.6.2). The NSsMS reported that those most likely to be in greatest need of the intervention were SPs who had an immediate issue or concern to be addressed.

Egan's model of the skilled helper was found to be a suitable model to guide the delivery of the intervention because, while providing a framework for the NSMS to help SPs to problem manage, it allowed for flexibility and tailoring of the telephone interaction between the SPs and NSsMS to meet the unique needs of the SPs. There was clear evidence of adherence to the principles of Egan's model of the skilled helper. Fidelity to the intervention was acceptable given the time spent on training and the novel nature of the intervention, and all three NSsMS stated that the training provided was adequate. Despite this, my analysis of the intervention calls suggests that aspects of adherence to the intervention can be improved upon. More emphasis needs to be placed on the teaching of problem management skills to the SPs.

Preparedness to care was the only outcome measured from which a significant between groups difference with moderate effect was found in support of the intervention group, at the second data collection time point (T2) (p=0.025, partial eta squared = 0.076). This equates to a Cohen d effect size of 0.35 (Cohen 1992). No significant differences were found between the intervention and control groups at time 1 or time 2 data collection for self-efficacy in obtaining respite, SP burden and QoL.

Within group significant differences were found for the intervention group for preparedness and QoL, and for the control group for SP burden. A significant improvement was found in the intervention group between baseline and T1 (p=0.004), baseline and T2 (p=0.001), and T1 and T2 (p=0.005) for preparedness to care. A

significant within group improvement was found for the intervention group at T2 data collection (p=0.042) for domain 4 [Environment] of the WHOQoL. A significant within group increase (negative effect of caregiving) was found for the control group at T1 for CRA subscale 1 (disrupted schedule, p=0.44), CRA subscale 3 [lack of family support], (p= 0.040) and CRA subscale 4 [health problems], (p=0.008). A significant within group decrease (negative effect) was found for the control group for CRA subscale 5 [Impact on self-esteem] at T1 (p=0.015) and T2 (p=0.036).

A discussion of the findings and results presented in this chapter is provided in chapter 8.

Chapter 8: Discussion

8.1 Introduction

The purpose of this study was to develop and test an intervention that nurse specialists in multiple sclerosis (NSsMS) could utilise in practice to meet the unmet needs of people who support (SPs) people with multiple sclerosis (PwMS). The primary goal of the intervention was to enable NSsMS to help SPs of PwMS by enabling them to learn problem management skills and helping them feel prepared for their role in supporting PwMS. A secondary goal was to determine the potential impact of the intervention on reducing burden and improving the QoL of SPs of PwMS. The intervention was guided by Egan's model of the skilled helper (Egan 2010). It was presented in a manualised format and supplemented with a training manual for the NSsMS and a guidebook for the SPs. The SP guidebook provided an overview of what was involved when participating in the intervention arm of the study. Furthermore, the guidebook enabled the SPs to engage in the process of problem management as it included a caregiver problem self-evaluation checklist and worksheet for completion prior to receiving the first intervention call, along with guidance on how to solve a problem. The steps to managing problems as outlined in the COPE model (creativity, optimism, planning and expert information) (Houts *et al.* 1996) were used to facilitate the SP's understanding of the concept of problem-solving in this study.

People who support PwMS have a number of unmet needs, many of which can be addressed within the role of the NSMS. Despite the many unmet needs of caregivers of PwMS, no nurse-led intervention had been developed prior to the development of this intervention. This is the first nurse-led intervention to be developed and tested with a view to supporting this group of SPs. It was designed and developed so that it would be applicable to the everyday role of the NSMS in the clinical setting. It has the potential to be used as a single or multi-component intervention and could be applied across multiple specialties, in any cultural setting. Due to the innovative nature of this intervention, the study was limited to feasibility testing within a pilot trial. A process

analysis was carried out to determine the ease or challenges encountered during the delivery of the intervention, the methodological challenges that need to be addressed and the protocol changes required prior to conducting a full-scale trial.

In this chapter, the findings from the process analysis and trial component of the study are discussed in the context of international literature as is the feasibility of further testing of the intervention in a full-scale trial. As the main objective of this study was to determine the feasibility and acceptability of the intervention and its evaluation within a randomised trial, it was important to establish potential effects of the intervention prior to full-scale trial planning. Firstly, therefore, the perceived effectiveness of the intervention which was uncovered during the process analysis and the trial's quantitative results are discussed. The characteristics of the sample of SPs are then examined to demonstrate the international relevance of the study findings and results. The findings from the process analysis along with the contextual and protocol changes required to optimise success in full-scale testing are also discussed. This chapter concludes with a discussion of the implications, limitations and recommendations of this trial.

8.2 Effects of the intervention

The potential for this intervention to be beneficial is evident from the quantitative and qualitative findings and results. The exploratory nature of the study and the sample size obtained, mean that it was underpowered to detect differences for the outcomes that were investigated. The study was powered on a sizeable benefit on the preparedness to care questionnaire, which would require 106 participants (53 in the control and 53 in the intervention group). Although the full sample size was not obtained, the statistically significant improvement for the intervention group on the preparedness to care questionnaire at the T2 (week 13), time point when compared to the control group, supports the potential effectiveness of the intervention. A moderate effect size, partial eta squared (the proportion of variance in the dependent variable explained by the independent variable) score 0.076 was noted (Pallant 2013). This finding is encouraging as it supports the previously reported finding that the caregivers' of PwMS preparedness to care is amenable to intervention (Finlayson *et al.* 2009a).

The potential impact of this intervention is also encouraging in that the mean sum score change of 4.42 on the preparedness to care questionnaire for this intervention was greater than the 1.63 sum score change reported by Finlayson *et al.* (2009a). An effect size of 0.35 (a medium effect) (Cohen 1992) at the T2 time point is important and has implications for data collection points for a full scale study. The T2 data collection time point in this study is close to the optimum 4-month data collection time point recommended by Schlarmann *et al.* (2011).

Preparedness to care [feeling of readiness to care], is an important concept for caregivers as greater feelings of preparedness have been linked to reduced caregiver burden for caregivers of older persons (Zwicker 2010) and those suffering with cancer (Scherbring 2002, Zwicker 2010). It has also been found to help ameliorate some aspects of role strain (Archbold *et al.* 1990) and was the strongest predictor for lowering caregiver stress in stroke caregivers (Ostwald *et al.* 2009). The change at all time points on the preparedness to care questionnaire for the within group analysis for the intervention group in this study suggest that an early effect of the intervention on this outcome was sustained over time. The increased readiness to support the PwMS is likely to have occurred because of the reported SPs' expressed benefits from participating in the intervention. The response changes from 'somewhat well prepared' or lower, to scores of 'pretty or very well prepared' over the three time points are also reflected in the qualitative comments from the SPs in the intervention group following receipt of the intervention. The feeling of being supported, of having more knowledge, greater confidence, greater understanding and the opportunity to discuss their concerns with the NSsMS may have increased their belief in their readiness to provide care. A feeling of preparedness to care does not mean that the person has the skill or ability to provide the care (Schumacher *et al.* 1998). It does, however, mean greater belief in ability to take on aspects of the caring role, which may explain the improved belief in their readiness to take on aspects of the caring role evaluated in the preparedness to care questionnaire, such as organising services, managing stress, and dealing with physical and emotional needs of the PwMS.

Feelings of greater ease in seeking the help of the NSMS may also have enhanced this feeling of readiness to care, as the majority of SPs in the intervention group (n=21) stated that they would contact the NSMS sooner than prior to the intervention.

Increased knowledge of the availability of support from a skilled professional who was knowledgeable about their unique circumstances is likely to have been important to them. The finding that the majority of SPs in the intervention group (n=21/33) stated that it was important that it was the NSMS providing care for the PwMS that delivers the intervention implies that the unique expertise of the NSMS was important to them. This suggests that they were either unaware of the availability of this support prior to the study or they were reluctant to use it. This intervention provided an opportunity for the development of a therapeutic relationship between the NSMS and the SPs, which may enable them to more readily seek assistance into the future. It may therefore go some way towards helping ameliorate the previously reported poor up-take of professional support services by caregivers (McCabe *et al.* 2008a, Buchanan *et al.* 2009, Buchanan *et al.* 2010). The need for access to professional support for caregivers has been highlighted in recent studies (Bowen *et al.* 2011, Uccelli 2014) and in particular young carers of PwMS who find themselves abandoned and unacknowledged (Bjorgvinsdottir & Halldorsdottir 2014). This intervention provided an opportunity for the NSsMS to help SPs negotiate access and use health care services when it was required.

Caregiver between group burden scores in this study did not differ significantly. This may be due to the underpowered nature of the study. Although there were no significant within group differences across time for the SPs in the intervention group, significant within group differences were found in the control group between baseline and time 1 data collection time point. The burden scores for the control group increased significantly at time 1 data collection point for subscales 1(disrupted schedule), 3 (lack of family support), 4 (health problems) and decreased for subscale 5 (impact of caregiving on self-esteem). The direction of these significant changes indicated increased burden scores for the control group during the study. A similar trend in relation to burden scores was reported by Hartke and King (2003) who reported on the findings of a randomised trial testing the effects of a telephone support group intervention for older stroke caregivers. The authors proposed the increased control participant burden scores and relatively stable intervention group scores may have been due to the emotional outlet provided by the intervention. Although the intervention tested by Harte and King (2003) was different in that it was a telephone

psycho-educational intervention, the similarity in the findings on burden outcomes to this study support the potential for telephone support to impact on caregiver perceived burden scores.

Like burden, SPs between groups QoL scores in this study did not differ significantly. A significant intervention within group improvement was found at T2, for domain 4 [Environment]. This environment subscale contained items that were likely to have been addressed during the intervention such as health and social care accessibility and opportunities for acquiring new information and skills. A similar statistically significant improvement for within intervention group QoL scores [using the SF36] was reported by Grant *et al.* (2002) who tested a hybrid intervention [face-to-face and telephone] with a total of 73 participants in a randomised trial. Likewise an improvement that approached significance on the social functioning of the SF 36 subscale was reported by Elliott & Berry (2009) who tested a hybrid telephone support intervention with a total of 60 participants in a randomised trial for family caregivers of persons with recent onset of spinal cord injuries in the US. While the within group findings in this study suggest that the intervention may have potential to impact on SPs QoL, it is important to determine such an improvement within a full randomised trial, as comparisons against baseline data are otherwise misleading and invalid (Bland & Altman 2011).

Although comparing findings is difficult and must be done with caution, especially when comparing studies with relatively small sample sizes, the positive finding across studies that used both problem-solving models and telephone intervention components (Grant *et al.* 2002, Elliott & Berry 2009) is encouraging and support the view that this intervention has potential to positively impact on QoL when tested in a larger trial. Unlike the findings of other studies using problem-solving models and the telephone as a medium for delivery (Grant *et al.* 2002, King *et al.* 2007) where participant problem solving abilities were improved, this study found no significant effects on self-efficacy in problem solving. This may have occurred because of the NSMS focus on supporting and helping SPs to resolve problems with less emphasis on the teaching of problem management skills. This limitation can be addressed in a larger study by providing more intensive training and certification of competence for the NSsMS.

8.3 International relevance of the study findings and results

The similarity of the needs of SPs of PwMS, internationally, was highlighted in chapter two. Although many of these needs can be met within the scope of the role of the NSMS, no nurse-led intervention study was found to address them. The findings and results from this study, which tested a nurse-led intervention, have therefore, important implications for supporting caregivers of PwMS, internationally. Caregiver characteristics such as age (Alshubaili *et al.* 2008, Rivera-Navarro *et al.* 2009), female gender (Patti *et al.* 2007, Rivera-Navarro *et al.* 2009) and spousal relationship (Figved *et al.* 2007) have been linked to increased caregiver burden and reduced QoL. Therefore, to facilitate transferability of the findings to SPs of PwMS internationally, the characteristics of the SPs in this study are compared to those in published studies as summarised in Appendix 2.

In this study, SPs age, gender, marital status and education level were similar to those described in the literature. The mean age of 51.3 years (range: 22-84) is similar in variability to the mean ages reported elsewhere, which ranged from 35.7 years to 61 years (Winslow & O' Brien 1992, O'Brien *et al.* 1995, Boeije & van Doorne-Huiskes 2003, Cheung & Hocking 2004, Courts *et al.* 2005, Finlayson *et al.* 2009b) for quantitative studies. Likewise the finding that the majority of SPs (n=52, 74.3%) were married or living as married, has similarity to other studies where the majority of caregivers were spouses (Alshubaili *et al.* 2008, Buchanan *et al.* 2009, Finlayson *et al.* 2009a). The percentage of female 53% (n=37) to male 47% (n=33) SPs in this study differed slightly. Most of the published studies, with the exception of Sato *et al.* (1996) and Rivera *et al.* (2003) (Appendix 2), had higher proportions of men. The lower number of male SPs in this study is surprising given that women are twice as likely to have MS (Orton *et al.* 2006, Ascherio & Munger 2007, Compston & Coles 2008). This may be due to the small sample size or it could be due to reluctance amongst Irish males to seek help. Studies elsewhere highlight men's reluctance to seek health-related help (Galdas *et al.* 2005, O'brien *et al.* 2005). This has implications for recruitment to a full-scale trial and warrants further investigation in advance of full-scale testing.

Caregiver employment can be adversely affected by the PwMS's cognitive abilities and increased caregiving hours (Buchanan *et al.* 2013). The partner nature of the relationship between SPs and PwMS in this study made it difficult to separate support/caregiving from what they would naturally do in a supportive relationship. In this study, 23% of respondents did not provide an estimate of the amount of time spent supporting/caregiving in a 24 hour period. Estimating time spent caring was easier for the SPs when physical care was required but more challenging when emotional support was the primary support function. This difficulty in quantifying caregiving may reflect the view that family members are more likely to view caregiving as an "...extension of the relational role" (O'Connor 2007, p. 157). Family members are unlikely to recognise the work that they do as caregiving, until they position themselves as caregivers, which only happens with time (O'Connor 2007). Despite this, the overall mean reported time of 8 (SD 9.04) hours per 24-hour period spent caregiving in this study was in keeping with that reported in the international literature which varied from 2.5 hours (SD 4.1) (Finlayson & Cho 2008) to >10 hours (Gregory *et al.* 1996).

The length of time SPs spend caregiving has important implications for caregiver employment as, longer hours caregiving negatively impacts on employment (Buchanan *et al.* 2013). Over half (n=36) of the SPs in this study were employed and worked between 20 and 40 hours/week. This figure is in keeping with international figures as in the majority of studies most carers were employed either full or part-time (see Appendix 2 for summary of characteristics of caregivers in published studies). To minimise the impact of caregiving on caregiver employment, Buchanan *et al.* (2013) emphasise the importance of referring PwMS and caregivers to support programs and of encouraging their use to help keep the PwMS living as independently as possible.

In this study, the greater mean number of years providing support in the control group (13.18, SD. 8.36), compared to the intervention group (8.35, SD.5.95) is worthy of note, as the difference was statistically significant (p=0.013). It is not, however, reflective of the PwMS's disability level or length of time since diagnosis. Both groups had similar levels of disability, but the number of years since diagnosis was slightly higher ($\bar{x}$ 15) in the intervention group compared to the control group ($\bar{x}$ 11.5). The length of time since diagnosis or progression of the illness is not necessarily linked to being better prepared to care. Studies have found that SPs of people with advanced MS (i.e. those limited to

a wheelchair or bed) (Bowen *et al.* 2011), families of newly diagnosed PwMS (Wollin *et al.* 2000) and young caregivers (Bjorgvinsdottir & Halldorsdottir 2014) require professional help and personal contact to address their informational needs

As the majority of PwMS in this study had relapsing remitting MS (RRMS) (52.9%, n=37), and secondary progressive MS (SPMS) (42.9%, n=30), the findings are likely to be more reflective and therefore transferable to SPs who care for PwMS with these types of MS. People with primary progressive MS (PPMS) account for 15% of the MS population and therefore have been poorly represented in this study. The effects of the intervention tested in this study for SPs of PwMS with PPMS have therefore, yet to be tested. Despite this, PwMS who have PPMS have greater disability and therefore greater illness severity, a factor associated with increased caregiver burden (Pakenham 2005a, Rivera-Navarro *et al.* 2009, Waldron-Perrine *et al.* 2009) and deterioration in QoL (Acaster *et al.* 2013). It is therefore likely that their SPs would benefit from this intervention. The need for information is a repeated need across studies that include SPs of PwMS across the disease trajectory (Koopman *et al.* 2006, Bowen *et al.* 2011, Lorefice *et al.* 2013). The intervention has, therefore, potential to meet some of the needs of SPs of people with PPMS, given the similarity between the needs reported in this study and those in a study of caregivers of PwMS in the advanced stages of the condition (Bowen *et al.* 2011). The importance of providing information and support to the wellbeing of a family of a PwMS has been emphasised by Uccelli (2014) who urges healthcare professionals to make use of the advances in technology and become "...more savvy and creative in utilising these potentially empowering tools" (Uccelli 2014, p. 184). Nurse specialists in MS are subject specialists who can provide information tailored to the unique circumstances of the SPs. The similarities in SPs personal characteristics across studies, along with the established impact of MS on PwMS and caregivers as identified in chapter 2 (Corry & While 2009), provides support for the transferability of the findings from this exploratory study to SPs of PwMS internationally.

This exploratory study suggests that the intervention has potential benefit for both SPs and NSsMS, but some uncertainties remain. A number of support persons had their concerns addressed during the intervention and were grateful to be given a voice within the health care system. This intervention provided a structure to the NSMS

communications with SPs and therefore has addressed a gap in practice where no formal training on telephone interactions between nurses and patients or family members is provided. The feasibility of testing this intervention using a randomised trial design is discussed in the next section.

8.4 Feasibility of the intervention and study

In this study feasibility was evaluated in terms of the ease of delivering the intervention and testing it within the randomised trial. The intervention was found to provide a formalised structure for NSsMS to provide proactive telephone calls to SPs of PwMS. Both the SPs and NSsMS reported benefits from the use of the intervention (section 7.3.11 and 7.3.13). The findings from this exploratory trial, which was carried out in a clinical practice setting, indicate that with some additional tailoring of the process, it is feasible to implement the intervention in practice. The number of intervention calls could be tailored to the needs of each SP and the intervention could be offered at specific points along the disease trajectory. This would enhance the long-term likelihood of embedding the intervention in practice.

Issues pertaining to recruitment, context and fidelity monitoring posed challenges during the implementation and testing of this intervention. Further tailoring of the intervention itself would create additional challenges for testing in a randomised trial. In order to identify the protocol changes required for full-scale testing, the challenges encountered in this study are examined in the next section.

8.4.1 Study recruitment

In this study, the novel nature of the intervention posed several challenges in relation to recruitment. As the intervention was aimed at SPs of PwMS, it was necessary to recruit both the PwMS and the SP. PwMS had to nominate the SPs and also consent to their inclusion in the study. This, along with the SPs identification with the role of 'caregiver', their perception of the personal relevance of the intervention and PwMS's fears regarding confidentiality, provided challenges to recruitment.

During the early stages of the study, SPs identification with the role of caregiver provided the greatest challenge to recruitment. The problems encountered due to lack of identification with the concept of caregiver among the SPs may have occurred because of their perceptions and beliefs about the caregiver concept, and potential participants' lack of identification with the term 'caregiver'. Other studies involving caregiver recruitment have identified with this challenge (Preissner *et al.* 2012, Hughes *et al.* 2013). For example, in a qualitative study of 40 relatives or friends, Hughes *et al.* (2013) concluded that "...self-identification with the role and label of carer is nuanced, shifting and variable" (p. 78) and refers to two other studies (O'Connor 2007, Bowen *et al.* 2011) that show variation in how relatives identify with the term carer. Hughes *et al.* (2013) found that some participants who were clearly engaged in caring activities preferred to be identified in terms of their relationship with the family member and rejected the term carer. Likewise, in a study exploring aspects of recruitment with caregivers of PwMS, participants suggested using the term 'support' or 'assistance' in place of the term 'caregiver' and highlighted that many people would not consider themselves to be caregivers (Preissner *et al.* 2012).

The importance of participants' perceptions of the personal relevance of the intervention for recruitment to MS caregiver research was highlighted by Preissner *et al.* (2012). In this study, initial difficulty in identifying with the caregiver role may have negatively impacted upon perceived relevance of the intervention. As it was a new intervention, uncertainties about the lack of previous testing made it impossible to provide concrete examples of how other SPs benefited or experienced partaking in the study. When the term 'caregiver' was changed to 'support person', PwMS and SPs responded more positively. They could identify with the fact that family members may need advice and support and had greater understanding as to how they may benefit from participating and contribute to testing the effects of the intervention. The lessons learned from this study should be included in future recruitment campaigns when recruiting caregivers of PwMS. Information about the average time on the intervention call, the positive benefits reported by SPs and some PwMS, and the demands of the intervention would help SPs make an informed decision as to the personal relevance of the intervention to them. The importance of these details to research participants was highlighted by Preissner *et al.* (2012) who reported that having details on the

helpfulness and benefits of the intervention and clarity around the logistical aspects of the study was important when determining personal relevance.

The recruitment challenges experienced within this trial with the use of OPD clinics to recruit reflect those reported by Stuardi *et al.* (2011) who provided evidence to support database recruitment as an efficient method of recruiting large numbers of participants in a timely manner. In this study the use of OPD recruitment was labour-intensive and delayed recruitment. Despite the recruitment of an additional site, the recruited sample of 71 was 35 participants short of the original target sample of 106. This reflects the impact of the early cessation of recruitment at one site due to workload demands on the NSMS and the use of OPD recruitment at another site. OPD recruitment also yielded a lower response rate (6%) when compared to database recruitment (17%). This difference cannot be attributed solely to the use of OPD as a method of recruitment, as the materials used also differed. Initial invite via OPD comprised a full information pack, while a letter of invite with statement of interest form was used for database recruitment.

In this study, both methods of recruitment had the potential to introduce bias as recruitment was not independent of the trial interventionists. Recruitment bias was evident when one NSMS decided that a SP should not receive the study invite as her condition had deteriorated so rapidly. Likewise, not all PwMS who attended OPD received the information pack. To minimise recruitment bias in a full-scale trial, patient databases would need to be updated on a regular basis and information packs and letters of invitation would need to be distributed by an administrator with no vested interest in the outcome. This would help eliminate the subjectivity and consequent risk of bias that can occur where nurses and other health care personnel are involved in the research study (Stuardi *et al.* 2011). It will also reduce the potential for selection bias in terms of who is offered participation in the trial (Altman & Dore 1990).

Both NSsMS' and PwMS' fears regarding confidentiality may pose barriers to recruitment during full-scale testing of the intervention. In this study, the NSsMS felt that some PwMS were reluctant to participate in the study due to concerns about their privacy. While PwMS were consenting to their SP's involvement in the research, the focus of the intervention calls was on SPs' concerns in relation to their role in

supporting or caring for the PwMS. It was not the intention that NSsMS would discuss issues pertaining to the PwMS beyond the boundaries of what would be discussed in usual care practice. The extent to which the personal information about the PwMS can be shared with the SP depends on the status of the consent obtained. Nurses' scope of practice and maintaining the best interest of the patient should always guide clinical judgement. Slade *et al.* (2007) highlight the ethical dilemma between non-maleficence and beneficence when respecting a patient's right to confidentiality. Focusing on the challenges professionals face regarding confidentiality in the mental health setting, Slade *et al.* (2007) distinguish between general information and personal information which is new to the carer and requires consent. In this study, the risk of breach of patient confidentiality was low, as the NSsMS were providing direct care to the PwMS and were familiar with those SPs who attended the clinic with the PwMS. They were also experienced in managing the information needs of family members and carers of PwMS. Their management of issues as they arose demonstrated a high level of skill at managing PwMS's fears regarding their confidentiality and their ability to re-assure participants as required. This was evident from the way in which issues discussed during the calls were managed. As the NSMS delivering the intervention was the person who also provided care to the PwMS, in most instances they had knowledge of what was previously discussed with the SP and to the extent to which the PwMS had consented. When there was doubt as to how much the SP already knew, guidance was provided on issues raised without discussing personal details regarding the PwMS. The level of skill and experience which the NSsMS had was essential for the safe and effective delivery of the intervention, in particular in the early stages of intervention development and testing. The level of skill and experience required has implications for full-scale intervention testing. This is further explored in 8.4.13

8.4.2 Effectiveness of screening

The broad inclusion criteria for this study were appropriate to the purpose of the study, which included identifying potential participants most likely to require and benefit from the intervention. Use of such a broad inclusion criteria reflected the need to be

cautious with SPs who appear over anxious about the PwMS's condition and the impact of excluding PwMS with cognitive impairment on trial recruitment was also established. The findings revealed that although NSsMS felt that all SPs were likely to receive some benefit from the intervention, they argued that in practice human resource challenges would not support its delivery to all SPs. The unpredictable nature of the condition (Barnes 2007) makes it challenging to know when exactly to offer such a service. Targeting participants at the key time-points suggested by the NSsMS and reported by the participants (see section 7.3.10.1 and 7.3.10.2) in full scale testing will ensure less variability within the groups and greater detection of benefit or otherwise from the intervention for specific groups of SPs of PwMS. For example, in a full scale trial SPs of people who are newly diagnosed could be targeted or those of PwMS who are at the advanced stage of the condition. These groups of SPs have been specifically identified in the literature as needing professional support (Wollin & Sato 2001, Bowen *et al.* 2011). As SPs of PwMS who are having a relapse were identified as a group that is likely to benefit from this intervention, they should also be included in a full scale randomised trial.

Another factor worth noting was the exclusion of PwMS with cognitive impairment. The exclusion of the group of SPs limited the testing of the potential benefits of this intervention as up to 50% of PwMS have cognitive impairment (Edmonds *et al.* 2007), which is a predictor of worse caregiver QoL (Labiano-Fontcuberta *et al.* 2014). The NSsMS felt that this exclusion criterion negatively impacted upon recruitment, as a number of SPs may have self-excluded on the basis of the PwMS having been diagnosed with cognitive impairment. They felt that this was one group of SPs that were in need of the intervention. Also, the exclusion of those with severe mental health issues is likely to have an impact on recruitment. The emotional burden of caregiving and perceived threats to the caregiver/care recipient relations increases the likelihood of caregivers requiring mental health treatment (Buchanan & Huang 2013). The inclusion/exclusion of SPs with severe mental health issues from a full-scale trial would create a dilemma in that it would remain unknown whether or not they would benefit. However, the ethical challenges are greater as the risk of adverse events for SPs with severe mental illness is increased and greater demands may be placed on already stretched psychological support services. The unknown risk associated with the

intervention undergoing testing in this exploratory study made securing psychological support for participants, if it was required, challenging.

8.4.3 Method and logistics of randomisation procedures

In this study, the use of concealed block randomisation was effective in that roughly equal numbers of participants across all 3 sites were assigned to the intervention (n=12, n=11 and n=10) and control group (n=12, n=14 and n=12). The process of randomisation ran in a smooth and timely manner. The statistically significant group differences that occurred are likely to be due to chance and the relatively small sample size. The variables that differed are important in that they have been associated with greater caregiver burden and lower QoL. The PwMS in the intervention and control group were statistically similar in all respects other than the physical health subscale of the MSQoL, where PwMS in the intervention group reported statistically significant greater physical health QoL scores than those assigned to the control group.

The SPs in the two groups were similar in many respects. Differences included living arrangements, where significantly more SPs in the intervention group reported 'living out' than 'living in' (p=0.035) with the PwMS. In addition, more of the intervention group attended third level education when compared to the control group (p=0.024). Greater numbers of SPs in the control group provided longer hours of care (p=0.020) and had spent a greater number of years providing support (p=0.013). Higher levels of education have been associated with greater caregiver uncertainty (Waldron-Perrine *et al.* 2009) and have a negative impact on the caregiver and care recipient's relationship (Buchanan & Huang 2011). Likewise, increased length of time caring (Sato *et al.* 1996; Rivera-Navarro *et al.* 2003a) and hours spent caring (Rivera-Navarro *et al.* 2003a) were factors that impacted negatively on caregiver burden. As both groups had variables that could negatively impact on outcome, it is difficult to determine what effect they had on outcomes in this study. Their impact was minimised by the use of the statistical test ANCOVA, which adjusts for the baseline measure (Darwin 2005). Within a larger randomised trial, such differences due to the chance nature of the randomisation process are expected to be smaller (Torgerson & Torgerson 2008).

8.4.4 Retention

Retention following randomisation was high in both the intervention and control group. Study retention was not affected by group assignment, as those who had preference for assignment to the intervention group remained in the study despite their allocation to the control group. Although 3 participants were lost to T2 outcome data collection, this may have been influenced by delays in the delivery of the intervention or factors other than the intervention itself such as personal life events or fatigue with responding to three rounds of data in a period of 13 weeks. Schlarmann *et al.* (2011) argue that the best time to collect outcome data is when the initial enthusiasm of the participants has subsided and the intervention has reached a plateau. Although this varies across interventions, Schlarmann *et al.* (2011) suggest that the 4 month time-point might be a good marker.

8.4.5 Threats to internal validity in a full-scale trial

As discussed above, the use of database recruitment and a larger sample size will help eliminate the risk of bias in a full-scale trial. Of greater concern, however, is the risk of subversion when progressing to full-scale testing. Evidence from the field of criminology demonstrated that larger effect sizes are reported when those who developed the intervention were involved in the testing phase (Petrosino & Soydan 2005). This trial was managed and coordinated by the person (me) who had developed the intervention. Consequently, I had a vested interest in obtaining a positive outcome from the trial. In addition, during the trial, there were times following initial contact with SPs that I had a preference for assigning the person to one or other of the two groups. This occurred as I felt that a SP may benefit more from the intervention or be more co-operative within the intervention group than another SP. However, because I had no involvement in the random allocation and this was done by a person independent of the trial following study number assignment and baseline data collection, I could not influence whether a SP would be in the intervention of the control group. Although the

risk of subversion is less likely to exist within a larger randomised trial, where researchers other than the developer of the intervention might be involved in the practical aspects of recruitment, baseline data collection and study randomisation, it is prudent to ensure that independence is maintained, monitored and reported at all stages of the study.

8.4.6 Risks associated with the study

In general, the intervention was reported to be helpful to the majority of participants and provided a structure within which NSsMS can communicate with and help SPs. The risks associated with this study might be no greater than those that may be encountered during longitudinal survey research, where some questions on the questionnaires may be a source of harm leading to increasing worry and anxiety (Evans *et al.* 2002). The SP who withdrew from this study following T2 data collection was identified by the NSMS as excessively anxious about what could happen to the PwMS into the future, and was advised by the NSMS to attend the GP. Identifying those with mental health issues is challenging, because this requires self-reporting of the existence of a mental health diagnosis by the SP or the possibility that an undiagnosed mental health issue may be present.

The provision of support by the telephone may not be the most appropriate method of supporting SPs who suffer from anxiety, although psychologists and counsellors use the telephone as a method of providing counselling to persons with mental health issues (Masi & Freedman 2001, Campos 2009). According to Campos (2009) issues such as confidentiality, anonymity and severity of the presenting problem are of importance to client use of tele-psychology but these have yet to be examined. Other than the findings of a study by Hartke & King (2003) who reported that the treatment group were more distressed overall at the first assessment time point with significantly higher levels of depression, burden, loneliness, stress and lower sense of competence, no additional negative outcomes from telephone support interventions were found in the literature. No studies or discussion papers were found which examined the risks associated with telephone interventions. Given the lack of insight into those who may

be at risk from telephone interventions and the likelihood of an absence of robust psychological organisational back-up to support the intervention, it would be prudent to screen for anxiety and exclude potential participants demonstrating moderate to high levels of anxiety, from a full scale trial. However, if robust psychological support back-up services were available, this would facilitate the testing of this telephone support as a method of supporting SPs of PwMS with anxiety or those with diagnosed mental health conditions.

8.4.7 Data collection methods

The instruments used in this study, other than the self-efficacy in obtaining respite and problem management subscales have been used internationally within the disease specific group or general population. In general, the questionnaires were acceptable to the participants but minor modifications are recommended to the MSQol, CRA and WHoQoL questionnaires. For the MSQoL questionnaire, question 3, the term 'pep' can be replaced with 'high spirits' or 'energy', which are words more familiar to people in Ireland. The inclusion of a 'non-applicable' option on the questionnaires is not recommended, as to do so would alter the psychometric properties of the instrument and so would require re-validation of the instrument. The high rate of missing data for questions pertaining to sexuality in the MSQoL questionnaire may be explained by the sensitive nature of the questions and the lack of anonymity. It is not unusual for questions of a sensitive nature to pose problems with missing data in survey research (Kays *et al.* 2012) and this is more likely to happen if there is any uncertainty regarding anonymity (Land & Ross 2014). In this study, confidentiality was guaranteed but complete anonymity was not possible and would be unlikely in full-scale testing, due to the longitudinal nature of the study and the need to connect data collected at different time points within the participants and between the people in each dyad.

The WHOQoL instrument is internationally validated and designed for use in the general population. Use of this instrument has the advantage of enabling the comparison of caregiver QoL scores to that of the general population. Since the commencement of this study, an MS carer specific QoL questionnaire has been

developed and tested by Benito-León *et al.* (2011) and a recent validation of the questionnaire confirms its validity in this population (Hoefman *et al.* 2013). These advances in the area of MS caregiver QoL measurements are encouraging and worthy of exploration in future studies where MS caregiver QoL is an outcome of interest.

8.4.8 Context

A number of factors impacted on the timely delivery of the intervention, such as the cancellation of intervention calls and synchronising of the availability of the NSMS and SP at a mutually convenient time for the intervention calls. These challenges are unavoidable and given the pragmatic nature of the trial will remain a challenge during full scale testing. To accommodate these tasks it would be necessary to factor administration time for organising and re-arranging calls in the trial's budget. Likewise, the research team will need to be flexible and this is likely to have cost implications which should be factored into the budget. The resource concerns raised by management in terms of the impact of the study on usual service delivery from both the NSMS perspectives and other supports required reflect the challenges of doing research in practice.

The challenges encountered in this study are similar to those identified elsewhere in a study exploring clinicians' attitudes to research participation (Paget *et al.* 2014). In a survey (n=208) of doctors, nurses and allied health care workers attitudes towards research in a tertiary general hospital in Australia, lack of time, resources, infrastructure and institutional attitudes to research were reported barriers to participating in research (Paget *et al.* 2014). The finding that clinicians experience a lack of support and encouragement in participating in research (Paget *et al.* 2014) reflect those found in an study on nurse practitioners in Ireland (O'Keeffe *et al.* 2013). At practitioner level, NSsMS are required to provide evidence based practice and collaborate and implement research findings (Health Service Executive and National Council for the Professional Development of Nursing & Midwifery 2008). These barriers have implications for future testing or implementation of the current intervention, if services such as additional psychological support is required at clinical sites in order to embed the research in clinical practice

The term collaborate is loose and open to different interpretations and there is little protected time for Irish based nurses to take part in research, even for those who are required to engage in research activities such as nurses at nurse practitioner level (O'Keeffe *et al.* 2013). Although health care managers were supportive of this study, the fact that one of the NSMS had to deliver it outside of her core working hours suggests that taking part in research of this nature was not viewed as a priority of the NSMS role. The study was carried out at the peak of the recent recession in Ireland, a time when there was an embargo on staff replacement and staff were under pressure to deliver the same service with limited resources. This added to the challenges and pressures on the NSsMS who had to provide cover for other NSsMS, in addition to their own clinical duties. Such pressures also impacted on the smooth delivery of the intervention and led to the delay or late cancellation of scheduled calls. Human resource issues are therefore a major barrier to the advancement of development of clinical nursing research and future testing of this intervention.

Layering the clinical workforce with nurses with a research focus is unlikely to be the solution, in particular for the advancement of practice where the practitioner is integral to the research study. Testing the intervention in the 'real world' of practice is important to determine clinical effectiveness and ease of embedding the intervention in practice, which will determine the extent to which it can be implemented in practice and long-term outcomes determined (Murray *et al.* 2010). According to Paget *et al.* (2014), infrastructure and inter-departmental collaboration is required in order to embed research in clinical practice. If interventions of this nature are to be tested for effectiveness, there needs to be a culture shift within the clinical environment from one of research use to one of new knowledge generation. Nursing clinical environments need to be supportive of new initiatives beyond the traditional survey and interview approach to knowledge generation. Like that of Australia (Paget *et al.* 2014), ensuring that protected time is given to clinicians for the collaboration in projects to generate new knowledge and practices, requires changes to working practices and a shift in attitudes of both managers and clinicians towards facilitating and participating in research of a more complex nature.

Further testing within a full scale randomised trial requires both organisational and interventionist commitment, which should be secured at the outset, with a site specific

contract. This could be achieved through shared responsibility for the success of research studies. Commitment to achieving collaboration of this nature for future testing of this intervention presents challenges in the current Irish health care system where the rewards for work completed differ between clinical sites and Universities. University status is judged on the quality of research outputs. Clinical sites are rewarded for patient care outputs. A shared work culture is required so that both organisations value, and are rewarded for, the generation of new knowledge. This calls for protected research time for clinicians, a change that requires major change to their working practices (Paget *et al.* 2014). An effective research and, therefore, work culture can be created for both researchers and clinicians through the sharing of rewards for research outputs (Meek 2014). Such a working culture would optimise the success of a full-scale trial for this intervention. A shared research culture for researchers and clinicians would reduce the human resource cost and increase the likelihood of success of such a trial. It may also enhance site recruitment and long-term implementation of the intervention in practice.

8.4.9 NSMS fidelity to the process

An analysis of the content of the calls and the processes applied by the NSsMS support the view that, in general, the NSsMS adhered to the principles of Egan's model of the skilled helper within the scope and role of the NSMS. Although the core principles were not always applied in a consistent way, there was clear evidence of the provision of support (7.3.6.2). The teaching of the problem management skills was less evident but was integrated to a greater extent over time. Feedback to the NSsMS helped reinforce the application of the problem management component of the intervention. As this research study was conducted in the clinical setting, a high degree of flexibility was required. Adhering to a rigid approach was therefore not a requirement and doing so may be counterproductive in a tailored intervention of this nature. Mars *et al.* (2013) assert that while deviation from the protocol may lower adherence, it may optimise the impact of the intervention. Given the nature of the intervention tested in this study, flexibility is an essential component and standardising

the number of calls may impact negatively upon outcomes. Future testing of the intervention developed for this study requires additional guidance on how to adapt the intervention based on the initial assessment of the SPs needs during the first intervention calls. The intervention scripts need to be adjusted and guidance provided on how to adapt the scripts to the unique needs of the SPs.

8.4.10 Approach to fidelity monitoring

The variability and uniqueness of the issues explored during the calls necessitated a large degree of flexibility in the delivery of the intervention to meet the needs of the SPs. This created challenges for monitoring fidelity to the intervention. Given the absence of a pre-existing monitoring instrument and the exploratory nature of the study, within which the full scope of the intervention and requirements for its application in practice emerged during the delivery phase, application of a rigid structure to fidelity monitoring would have been premature and restrictive. Complete lack of structure would, however, have made the process too wieldy and unfocused. The use of a call monitoring instrument based on the core principles of Egan's model of the skilled helper was appropriate, in that it provided a guide to what was required and a mechanism by which fidelity could be independently monitored by a second person.

Use of the same monitoring instrument provided a focus for the persons monitoring the fidelity to the intervention as to what was considered important at the pre-testing phase, yet allowed for flexibility in its application. This required an advanced level of skill in the application of the model and specialist expertise from the interventionist. The focus of the calls, the range of issues raised, and more importantly the uniqueness of the impact of these issues on the SPs, together with the readiness of the SPs to engage in the process, meant that monitoring for adherence depended heavily on the professional judgement of the person monitoring the call. It required the persons monitoring fidelity to the intervention calls to have a good understanding of the intervention itself, the scope of the role of the NSMS and the application of professional judgement in determining the appropriateness of use or otherwise of skills during each intervention call. It also called for openness to each NSMS's unique approach to

practice delivery and a recognition that interventions will differ in accordance with the interventionist's personality and practice style.

The principle based approach to monitoring fidelity to the intervention in this study was, therefore, appropriate and congruent with the principles of the model guiding the intervention. The call monitoring instrument used served for what is recommended as a best practice template.

Although the lack of quantification of the outcome of the fidelity to the intervention in this study poses a difficulty when describing the degree of fidelity to the intervention, it was appropriate at this stage of intervention development. Further testing of the intervention should include a refinement of the call monitoring instrument to accommodate the additional tailoring of the number of calls for each participant and the application of ratings to the individual items on the call monitoring instrument. To monitor the delivery of the intervention effectively, researchers assigned to monitor its delivery need to have a good understanding of Egan's model of the skilled helper, subject specialist expertise and a working knowledge of the scope of the role of the interventionist. While careful monitoring of the delivery of the intervention and ongoing training and feedback to interventionist will optimise the potential impact of the intervention, the impact of ongoing monitoring on effective implementation needs to be considered for the delivery of the intervention during routine practice. In real-life settings, such interventions may not be effective due to poor implementation in practice (Waller 2009). Future testing of this intervention could include an analysis of the amount of ongoing monitoring and training required for optimum intervention delivery outside of a trial setting.

8.4.11 Dose delivered and received

Adhering to the delivery of four calls over the duration of the study was unnecessary in some instances, and counterproductive in others. When the intervention calls were finished, the SP reverted to usual care. In situations where all needs were met, this was appropriate. However, in situations where issues were on-going or the NSMS and

SP felt that follow-up was warranted, proactive contact may have been terminated prematurely. The process analysis confirmed that for some participants, up to two calls was sufficient, but more than four may be required for others. This suggests that flexibility is also warranted in determining the number of calls required. The findings suggest that a minimum of two calls are required, one to begin the process of developing a therapeutic relationship between the NSMS and SP, and the second to allow for emerging issues to be examined. Following the second intervention call, a decision should be made by the NSMS and SP as to the need for further intervention calls. The calls could be terminated at a mutually agreed time when the SP is in a position to manage issues which are ongoing, or they have grasped the principles of problem management.

The requirement for a greater degree of tailoring for this intervention reflects the findings of a review by Ryan & Lauver (2002) where participants were found to have a preference for tailored over standard informational interventions. There is increasing evidence supporting the effectiveness of tailored interventions (Baker *et al.* 2010, Krebs *et al.* 2010, Gagliardi 2011, Bernhardsson *et al.* 2014). The challenge is how to tailor interventions to enhance effectiveness (Gagliardi 2011). The findings from this study suggest that tailoring the content has the potential to enhance effectiveness. Although not tested in this study, the recommendation from participants to tailor the number of calls suggests that this may also be of value. A Cochrane review to determine what interventions are effective in improving outcomes for adult family caregivers of people with stroke, did not find any study attributes that were significantly associated with effectiveness (Baker *et al.* 2010). The findings from two reviews, suggest that the inclusion of a feedback system may contribute to effectiveness (Ryan & Lauver 2002), but repeated versus single administration were not found to impact on health behaviours (Ryan & Lauver 2002). In this study, the use of more than one call provides an opportunity for feedback to SPs. For example, during follow-up calls the NSsMS discussed problems raised in the previous calls and gave reassurance or reported on progress as appropriate. The importance of providing feedback of this nature as part of the intervention can be emphasised to the NSsMS and specific training can be incorporated as part of the intervention training programme.

8.4.12 Availability and quality of the interventionists

To test this intervention in a full-scale randomised trial, the availability and quality of interventionists would need to be considered. The nurse specialists in MS were among the most experienced and highly skilled NSsMS in Ireland, as they were all in post in 2002 when the specialist nurse role was first introduced in Ireland. By 2003 they had completed the postgraduate diploma which included a specific module on MS nursing. I focused on the intervention process and was confident that the NSsMS had the knowledge and expertise to manage the clinical elements of the intervention. It was evident from the process analysis that experienced NSsMS are required to deliver the intervention. The analysis of the delivery of the intervention demonstrated that the NSsMS in this study managed issues raised by SPs in a skillful and professional manner. In order to deliver a professional service, it was important that the people they telephoned felt confident in their knowledge and abilities. This did not always require an immediate answer but it did require skill in managing the call; a skill that a novice to the role may not have. Given that the intervention is a guide and the specific content of the intervention call is dependent on the NSsMS' knowledge and skill, to ensure a professional service is offered to SPs in full-scale trial, experienced NSsMS will be required to deliver the intervention. Since the 1st September 2010 all nurses appointed to specialist posts require a minimum of 5 years post-registration experience, a post registration diploma related to the specialist area of practice and 2 years practice in the specialist area (Doody & Bailey 2011). All NSsMS should therefore be eligible for the delivery of the intervention in a full-scale trial. The two years experience in managing the information needs and concerns of PwMS and their SPs and the specialist knowledge gained during post-registration study, should be sufficient to enable them to manage issues raised in the delivery of this intervention.

8.4.13 Training of the interventionists

Given the nature of this complex intervention, the extent of training required was difficult to judge in advance. The novel nature of the intervention meant that its full potential and most appropriate approaches to implementation were unknown at the commencement of the study. The degree of flexibility required for implementation became evident as SPs with different needs entered the study. After completing the first few intervention calls, the NSsMS became more confident in the application of the intervention and in the provision of proactive support and advice. In general, the training given was adequate for early phase exploratory testing of a novel intervention. On-going feedback and reminders throughout the study addressed gaps in the delivery of the intervention, as identified from the monitoring of the calls. The teaching of problem management skills remained a challenge throughout the entire study. Prior to each set of intervention calls I reminded the NSsMS to re-iterate the problem management process during the intervention calls. In order to impact on SP problem management, in a full-scale trial interventionists would have to provide the same standard of intervention delivery to all calls, although the content of those calls would differ. To achieve this, a more intensive training programme would be required. Health care organisations would therefore need to facilitate on-site and off-site training. Furthermore, the NSsMS will need to allocate sufficient time to the training sessions and be open to assessment for competence in the delivery of the intervention to SPs with a wide range of needs. In addition to pre-reading the intervention and training manual, the NSsMS should attend a compulsory half day off site training. This could include an overview of the intervention, allow for group discussion and provide the opportunity for the trainer to answer questions. During the training, the NSsMS can take part in simulated role modelling and develop a peer support network. The half day should be followed by on-site training on the use of the equipment installed at the site, and assessment of competence on the delivery of the intervention. It is important to remember that the amount of training required may differ for interventionists but all should be able to deliver the intervention to a specified standard (Miller & Rollnick 2014). This would require a greater commitment from site managers to facilitate the NSMS training and from NSsMS to do the training and be agreeable to additional training until certified as competent by the trainer. This level of commitment creates

challenges in practice. As NSsMS are key players in the delivery of the intervention, they are instrumental to the research study. Full-scale randomised trials can be costly and the voluntary nature of NSMS participation in the delivery of the intervention puts this investment in the study at risk. While this was acceptable during the exploratory phase of the study when there were so many unknowns, including the effects of the intervention, its use within a full-scale trial puts the study at unnecessary risk of collapse.

8.4.14 Intervention acceptability

The findings reveal that although the intervention is acceptable, acceptability can be improved by tailoring the amount of the intervention delivered (i.e. the number of telephone calls) to the recipient needs, targeting PwMS and SPs at specific times throughout the course of the disease trajectory, and addressing NSMS workload, in particular aspects of the role that can be delegated to administration staff. Acceptability is essential for the successful testing and integration of interventions in practice. The findings in this study indicated that the majority of SPs, irrespective of their concerns, experienced some benefit from receiving the intervention itself. Although the NSsMS recommended targeting specific groups they also indicated that in an ideal setting with sufficient human resources that all SPs would potentially benefit. This may reflect the fact that some participants did not require the full dose of the intervention, which led to unnecessary repetition of advice. If the dose of the intervention had been reduced for participants whose concerns were addressed within the first two calls, their overall evaluation of the intervention may be different to that of this study.

In this study, the needs of the PwMS were not factored into the recruitment process. Targeting SPs at times known to be challenging for them, such as when the PwMS is first diagnosed (White 2012) or during the transition of the PwMS to care (Bowen *et al.* 2011) would enable a more focused approach to testing for the acceptability of the intervention. Some participants in this study commented on the lack of support that was available to them at the time of receiving the diagnosis and the NSs

MS found that some participants were confused about the information they received from media sources. This could be ameliorated through the provision of accurate information on diagnosis, using an intervention such as the one described and tested in this study. The key role of the NSMS in supporting patients and families at diagnosis is highlighted by White (2012) who raised a concern regarding the use of websites, particularly those that are unregulated for obtaining information about a condition. Although participants can be targeted during time of relapse or change in the course of the disease, offering this intervention when the PwMS is diagnosed may ameliorate the lack of support experienced by SPs at later stages in the PwMS's illness. In a qualitative research study, exploring the impact of advanced MS on families, Bowen *et al.* (2011) found that relatives were ill informed and were rarely given the name of a key worker. A structured and proactive approach to providing support to SPs from the time of diagnosis would foster the development of a therapeutic relationship from an early stage of the illness. This will ensure that information provided to the SPs is accurate and they may feel more comfortable in contacting the NSMS when problems emerge. High levels of satisfaction with the intervention in this study support the overall acceptability of the intervention. The fact that almost all the participants in the intervention group (31/33) stated that they would recommend the intervention to a friend if they were in need provides further evidence of perceived benefit.

8.5 Conclusion, implications and recommendations

The intervention was tested in an exploratory randomised trial using multi-methods of data collection. This enabled the testing of this new intervention to determine its feasibility, acceptability and potential effectiveness. The findings showed that with additional tailoring, the intervention is feasible to deliver and acceptable to PwMS, their SPs and NSsMS. The findings also highlight the unmet needs of SPs of PwMS in Ireland and that these needs, which are within the scope and role of the NSMS, may be ameliorated through use of this intervention. Although the findings are preliminary, they are encouraging, as potential benefits include increased SP preparedness to care. Feelings of preparedness to care helps SPs believe in their ability to take on aspects of

the caring role (Schumacher *et al.* 1998). While this will not address all of the needs of SPs, preparedness to care has been reported to help ameliorate burden and stress in other groups of caregivers. Caregiver burden and stress have many negative consequences, including the negative impact on caregivers' mental health. With the increasing need for family members and caregivers to assume a caring role, it is incumbent on the health service to provide support. While the full potential benefits of this intervention have yet to be established, they include the potential for a reduction in hospital admissions and repeated hospitalisations. Troubleshooting can often prevent problems arising. In addition, the availability of this intervention may help SPs embrace the caregiving role when they know that back-up and support are available to them.

Although this research has shown the potential impact of this intervention, the limitations need to be acknowledged. These are primarily associated with the exploratory nature of the study, which limits the conclusiveness of the findings. Larger scale testing is required to substantiate the findings from this study and to obtain a reliable estimate of the effects of the intervention.

8.6 Study limitations

The criteria for SPs to be eligible for this study required that they be without a severe mental health condition. This limited the extent to which the potential impact of the intervention could be tested. Likewise, SPs of PwMS with cognitive impairment are likely to have unmet needs and these were identified by the NSsMS as a group that could potentially benefit from their support. These exclusion criteria limited the extent to which the intervention could be tested across the full range of SPs. Although the intervention and control groups had more than 30 participants (which is acceptable for an exploratory trial) (Lancaster *et al.* 2004), a fully powered trial would require many more patients to substantiate the findings. Based on the parameters from this exploratory study a total minimum sample size of 320 participants (160 for each group) would be required.

The novel nature of the intervention for both the researcher and the interventionists meant that as the study progressed, the full extent of the intervention unfolded. Although this did not result in changes to the intervention manual, it did lead to tailoring of the application of the intervention to the unique circumstances of the SPs. This meant that the application of the delivery of the intervention was not standardised to all SPs. Although this might be viewed as a study limitation, it may also be a strength of the intervention, as it has application across a broad range of SPs. Prior to full-scale testing, the intervention manual could be adjusted to allow for further tailoring of the intervention so that the number of intervention calls can be individualised to the needs of the SPs.

The study was at risk of bias from a number of perspectives. The risk of recruitment bias limits the generalisability of the findings to the target population. The fact that the researcher developed the intervention meant that the entire study was at risk of bias. Although the risk existed, it was minimised by the recording of outcome telephone calls and independence in the randomisation process. The potential for bias also existed during the pre-post intervention interviews with PwMS. I met in person with almost half of the PwMS and a number of their support persons when doing the pre-post intervention interviews with the PwMS. This was beneficial in that it helped to develop rapport but may have biased the findings, as on many occasions the researcher met the SP while doing the interview with the PwMS. This interview was, however, essential to ensure that there were no negative effects for PwMS as a result of the SPs engaging in the study. It is unlikely that these meetings had any additional benefit for the SPs in the intervention group as the meeting with them was short and not related to their role in supporting the PwMS. It is important that these types of bias are addressed for a phase 3 study.

Although the instruments used to collect outcome data were validated questionnaires and demonstrated acceptable internal consistency reliability scores in this study, the SPs had difficulty answering some of the questions on the CRA and WHOQoL questionnaires. These difficulties may have adversely impacted on the ability of the study to detect the effects of the intervention.

The NSsMS delivering the intervention were also providing support to the control group and, although there was no evidence of overt contamination in this study, the fact that the NSsMS had learned a structured way to communicate with SPs may have influenced how they communicated with the control group. Given the number of NSsMS at individual sites and the challenge of randomising SPs to particular NSsMS within a site, where more than one exists, makes elimination of this limitation in a full scale trial, difficult. If this risk of contamination were to affect the outcomes then where statistically significant findings are found in favour of the intervention group, its presence would only further confirm the power of the intervention.

I made telephone contact with the control group on three occasions to collect outcome data. As a result of this, I developed a relationship with the SPs in the control group. The SPs were aware that I was a neurology nurse and at times chatted with me. Although, where possible, I tried to limit discussions beyond the completion of the questionnaires, this was unavoidable at times, because cutting off a SP who wished to speak about an issue would have been rude. In addition, it could have adversely impacted on study retention. Although no support or advice was provided, the fact that the SP was listened to could have contaminated the study. The risk of bias from these discussions are limited as outcome data collection was carried out in the same way for both groups and any benefit would have been equalised across the groups. Likewise, some SPs reported benefit from completing the questionnaires but the impact of this on any of the groups was limited, as the effect should have balanced out between the intervention and control groups.

The intervention calls were not always delivered on schedule. Although this was unavoidable, it does impact on the standardisation of the intervention.

8.7 Study strengths

Despite the aforementioned limitations this study has a number of strengths. The model used to guide the intervention is congruent with the definition of nursing and beliefs and values of the discipline. This optimises the likelihood of NSsMS impacting

on outcomes. In addition, the tailored nature of the intervention allows for the unique needs of SPs to be addressed, an approach that values the uniqueness and dignity of the SP which is congruent with the values underpinning nursing. Testing the intervention within an exploratory trial design was appropriate for early phase testing of the intervention as the finding enabled refinement of the intervention and study protocol prior to full-scale testing. The inclusion of SPs of PwMS with varying degrees of disability and impact (other than those with cognitive impairment) meant that the potential of the intervention for a wide range of SPs was revealed. The study has demonstrated that it is possible to deliver and test this intervention using a randomised trial. Use of the randomised trial design is important, as it allow readers of the trial outcomes to be confident that the findings are due to the intervention and not some other variable. It has also highlighted the resources required for full scale testing. The pragmatic nature of the trial enabled the identification of the extent to which the intervention could be embedded in practice.

8.8 Implications of the study

The findings from this study have implications for research, practice and policy. Given the exploratory nature of the study, the implications for practice and policy are tentative. The findings show that SPs of PwMS in the Irish health care system have unmet needs. The literature review on the needs and experiences of caregivers of PwMS (Chapter 3) supports the view that these unmet needs are likely to negatively impact on both the PwMS and the SP. Caregivers provide millions of hours of care each year. This is an international trend, which results in reduced health expenditure for health care budgets (Wilkinson & Lynn 2005, Corry *et al.* 2014). Caregivers' health and well-being should therefore, be a priority for healthcare planners and managers.

In a qualitative study investigating the impact of disease on family members of patients with a wide range of chronic conditions, Golics *et al.* (2013) found that family members across a range of illnesses have similar experiences in the United Kingdom. The emotional impact of worry, frustration, anger and guilt on the family members as a consequence of the patient's illness affected the majority of lives (Golics *et al.* 2013).

As in my study, Golics *et al.* (2013) recognised that not all family members identify with the role of caregiver, and consequently the study focused on family members in general. The study identified the family member "...as a hidden patient group..." and argues that the potential negative impact may be ameliorated through the provision of support, in particular emotional support (Golics *et al.* 2013, p.795). As the purpose of this intervention was to provide support to caregivers, the positive outcomes identified suggest that this intervention may help to support the needs of caregivers in general. Support persons in the intervention group in this study reported feeling supported, more knowledgeable and that the intervention provided them with the opportunity to discuss their concerns. In addition, the intervention was found to positively impact on SPs preparedness to care.

In order to reap maximum return on funded research in areas such as MS, health research funders need to be cognisant of the type of intervention research likely to positively impact on the needs of family members and carers. In a recently published update of the review of reviews of caregiver interventions carried out to inform this research study (Corry *et al.* 2014), little evidence was found on interventions to support caregivers across a wide range of conditions. The review found evidence that education and support programme interventions improved stroke caregiver quality of life, and that information-giving interventions improved their knowledge. There was no evidence of benefit from psychosocial interventions or interventions designed to improve their social functioning. Although referring to stroke caregivers, the findings of the systematic review of reviews (Corry *et al.* 2014) together with the positive outcomes reported in this study, suggest that interventions that provide support for family members and caregivers are most likely to have a positive impact. The similarity of impact of patient illnesses on family members as reported by Golics *et al.* (2013), suggests that the intervention tested in this study has potential for family members and caregivers across a range of chronic conditions. Interventions that provide support and education for caregivers should be a research priority for caregiver strategy and funders of caregiver research.

The findings of this exploratory randomised trial have implications for future testing of this intervention with caregivers. A number of areas have been identified that need to be addressed in order to maximise on resources and the potential impact of the

intervention. The findings, however, have wider implications for intervention research in the practice setting in Ireland, in particular the testing of complex interventions. In order to support intervention testing of this nature, the nursing research culture in the Irish Health care system should be revised. Research infrastructure should be developed to include the availability of psychological support for study participants, and to secure protected research time for staff engaging in research projects. This would be of particular importance where a staff member is an integral component of the intended intervention. Such interventions, while costly to test, might have greater long-term benefits, as the potential to embed the intervention in practice can be determined. In order to achieve this, the benefits and rewards for research outputs must be shared across organisations (Universities and hospitals). This could be achieved through the allocation of additional funding for nursing staff to clinical sites where certain categories of nursing staff are actively engaged in quality research studies and where quality research outputs could be demonstrated.

In terms of practice, this intervention has the potential to improve the quality of care offered to family members and caregivers across a wide range of conditions. Although NSsMS provide telephone support to PwMS and their family members or caregivers as part of current practice, they do not receive any formal telephone communication training. This intervention, which provided a structured approach to communicating with SPs of PwMS, along with guidance on best practice in the provision of telephone communication, might help to fill this gap. In addition, following further testing to determine its effectiveness, the intervention has the potential to improve the quality of support that NSsMS provide. If it cannot be offered to everyone who may benefit, its implementation may be rationalised by offering it to those known to have specific needs, for example, around the time of diagnosis or symptom relapse. Its implementation could also be triggered by those SPs who make contact with the NSMS, should their concerns warrant further follow-up by the NSMS. In addition, interventions that may impact on other outcomes or strengthen the impact of this intervention can be nested within this intervention, and tested in future research studies.

As this was an exploratory study, making recommendations beyond those required for further testing is premature. The recommendations therefore focus on the changes

required to both the intervention and the study design, to optimise the successful implementation of a full-scale randomised trial. The recommendations include those changes required for recruitment, changes to the delivery of the intervention, including interventionist training and fidelity monitoring, and minimising bias.

8.9 Recommendations

8.9.1 Recruitment

- To avoid recruitment bias, it is important that databases of PwMS are used for recruitment and that these are generated independent of the research team and clinical staff involved in the study. Clinicians caring for PwMS (independent of the interventionists) should check the databases for accuracy.
- The findings from this study in terms of the reported benefits for SPs and their experiences of participating in the study provide concrete information that can be used in future recruitment campaigns. These include:
  - Some PwMS were pleased that their SP had someone to talk to. They reported that their SP was more understanding, had more confidence, was more relaxed, less worried and that the study provided an opportunity for them to discuss their concerns.
  - The intervention provided an opportunity for SPs to have access to a specialist nurse who had experience in MS and could tailor the support required. Those SPs who received the intervention reported being more knowledgeable, feeling supported, more confident and felt that the intervention provided an opportunity for them to discuss their concerns.
  - The intervention provided a structure to giving support which can be evaluated and therefore the impact of their role can be measured. On average the intervention calls took 20 minutes but lasted for up to 60 minutes depending on the SPs needs.

- The nurse specialists' interactions are focused on SPs' issues. The interactions are limited to the issues pertaining to the PwMS that have been previously discussed with the SP and to which the PwMS had consented. This information should be clearly stated in the participant information leaflets and consent forms. Guidance can be provided for the NSsMS by the inclusion of best practice guidance on PwMS confidentiality as part of the training manual.
- For future trials, the inclusion criteria could be amended to include SPs of PwMS with cognitive impairment. Support persons of PwMS could be targeted at times that they are most likely to need support from a NSMS (for example, during a relapse or change in treatment plan). This would ensure that those in greatest need of the intervention are targeted and therefore most likely to benefit from this intervention. Future trials could exclude dyads where the PwMS's condition is stable, as this group of people is least likely to need the intervention.
- In the absence of robust psychological back-up services, it would be prudent to screen all participants for anxiety and exclude potential participants demonstrating moderate to high levels of anxiety. However, if the service can offer a robust psychological support back-up service, this would facilitate the testing of this intervention as a method of supporting SPs with anxiety or those with other diagnosed mental health conditions.

8.9.2 The intervention

Intervention dose

- Each participant should receive at least two intervention calls and the number of follow-on calls should be tailored to the needs of each participant. Guidance on how to individualise the number of calls required by each SP could be included in the intervention manual.

Fidelity to the intervention

The following changes should be made to the protocol for monitoring fidelity to the intervention:

- Record all intervention calls and monitor the first five calls from each centre and a randomly selected 10% of all subsequent calls.
- Allow for flexibility in the number of calls delivered to each SP, based on the SP's needs.
- Refine the call monitoring instrument to incorporate flexibility in the monitoring of the number of calls delivered and include a scoring system to facilitate the quantification of the analysis of fidelity. The scoring system could allow for flexibility at the point of intervention delivery. For example, a three point scoring system applied to different components of the intervention may suffice. Such a point scoring system may include the following: Non-adherent (1 point), somewhat adherent (2 points) and fully adherent (3 points). This would enable the calculation of an inter-rater level of agreement.

Interventionist training

- The time available for training of the interventionists should be increased to a half day off-site, and 1-2 hours on-site as required. Health care organisations need to facilitate on- and off-site training and the NSsMS need to allocate more time to the training sessions and be open to assessment for competence in the delivery of the intervention to SPs with a wide range of needs. In addition to pre-reading the intervention and training manual, the NSsMS should be required to attend a compulsory half day off site training. The half day could include an overview of the intervention, allow for group discussion and provide the opportunity for the trainer to encourage peer support. Video demonstrations

of delivery of the intervention could be developed and used for interactive learning.

- To maximise resources, minimise participant research fatigue and avoid the peak of SPs' enthusiasm for this intervention during full scale testing, just two data collection time points could be used: baseline and post-intervention data collection only. In order to enable comparison of outcome data between the intervention and control group this data collection time-point should be fixed at the 4 month data collection time-point.

- The administration support required to facilitate the arranging and re-arranging of the telephone calls needs to be factored into budgets for future testing. The additional administration time required for delivery of the intervention needs to be factored into delivering the intervention outside of a trial.

- The interventionists need to have protected research time during core working hours and be flexible in the administration of that time in order to facilitate SPs availability to take the calls. Likewise, work-time flexibility is required from the research team for data collection.

Intervention delivery monitoring

- To effectively monitor the delivery of the intervention, researchers assigned to monitor its delivery should have a good understanding of Egan's model of the skilled helper, subject specialist expertise and a working knowledge of the role of the interventionist.

8.9.3 Avoiding bias in a full scale, definitive trial

In this study, the researcher who carried out the study was the same individual that developed the intervention. The study was carried out by this researcher, which increases the potential risk of bias. This risk is likely to be lower in a larger study,

where researchers other than the designer of the intervention are likely to be involved in the practical aspects of recruitment, baseline data collection and study randomisation. It is prudent to ensure that independence is maintained, monitored and reported at all stages of the study.

While it would also not be possible to blind the interventionists or participants to their allocation in a Phase 3 trial, knowledge of group assignment could be restricted to those research team members least likely to influence outcomes, while other team members such as data collectors, could be blinded to group allocation. This would minimise the risk of the research team influencing the outcomes.

8.10 Dissemination

To date three of the literature reviews carried out to inform this study have been published in international peer reviewed journals (Corry *et al.* 2011, Corry *et al.* 2013, Corry *et al.* 2014). In addition, a number of conference presentations were delivered at national and international level (see Appendix 32 for list). An educational resource has been developed on the development and testing of complex interventions and has been disseminated in a blog http://complexinterventions.blogspot.ie/, 2014. A summary of this resource has been published in the World of Irish Nursing (Corry 2014).

A report on the findings from this exploratory trial will be presented to the Health Research Board (Ireland) and to the participating organisations. A meeting will be arranged to discuss the findings and their implications for policy, practice and research. It is anticipated that the meeting will provide an opportunity to demonstrate that the development and testing of a complex intervention of this nature is feasible and that it has potential to improve the care offered to SPs of PwMS. It will also provide an opportunity to debate the challenges that researchers and clinicians face in testing new interventions in the real world of clinical practice.

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