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Supporting Families and Carers of people living with Multiple Sclerosis: A Rapid Realist Review and Realist Evaluation

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Title: Supporting Families and Carers of people living with Multiple Sclerosis: A Rapid Realist Review and Realist Evaluation

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Abstract

Supportive interventions are needed for family and carers of people with MS. A Rapid Realist Review and Realist Evaluation explored what helps, who it helps, when and how. Literature analysis was synthesised with thematic analysis of qualitative interviews and focus groups with 49 family and carers of people with MS. The resulting model summarised a family of interventions that could help people develop their capabilities and expand their resources, for more positive outcomes. This may prevent or delay a 'tipping point' where capacity to care is overwhelmed by caring roles.

Key Words

Multiple Sclerosis, Family; Carers; Rapid Realist Review, Realist Evaluation

Word count

7541 With references, reference list, tables and figures.

Background

People living with Multiple Sclerosis (pwMS) are often supported by family and friends who engage in varied caring roles. This can have negative physical, emotional and financial impacts, sometimes limiting the ability of family and carers to continue such roles. Supportive strategies and interventions may improve experiences of caring, but there is a lack of evidence into what these might be. The UK MS Society commissioned a study to generate recommendations for service provision, research, and influencing activities and a Rapid Realist Review and subsequent Realist Evaluation were conducted to explore what helps, who it helps, when and how.

The MS Society defines a carer as “someone who, without payment, provides help and support to a partner, child, relative, friend or neighbour, who could not manage without their help” (MS Society, 2015). Global estimates of people requiring daily assistance from another person are increasing and UK carers save approximately £132 billion annually, close to annual health expenditure (Harwood et al, 2004; Buckner and Yeandle, 2015). A survey of over 5,000 carers highlighted substantial impacts of caring on physical and mental health and well-being, employment opportunities and financial security (Carers UK, 2014). Access to supportive services is limited, however, contributing to over half of carers stopping work (Carers UK, 2014; EUROFAMCARE Research Consortium, 2005).

Carers’ needs can be multifaceted and context-specific, with specific challenges associated with MS. This progressive autoimmune disease of the central nervous system generates varying symptoms, including fatigue, weakness, and posture and mobility difficulties, psychological and cognitive impacts (Corry and While, 2009). UK prevalence of MS is comparatively high at 1:600, approximately 107,000 people (Corry and While, 2009; Rosati, 2001). Just over 40% of pwMS are diagnosed between the ages of 30 and 49 with about a ten-year reduction in life expectancy, therefore living for many years with MS (Mackenzie et al, 2014; Runia, 2012). Approximately 15% of people are initially diagnosed with progressive MS, and for 35% of people initially diagnosed with

relapsing-remitting MS, this becomes progressive (Kremenutzky, 2006). Complications and co-morbidities can include results of falls, infections, and pressure ulcers (Higginson, 2006), with substantial impacts on those living with MS, family and carers.

The substantial evidence base relating to needs of families and carers of pwMS includes a systematic review of 24 qualitative or descriptive survey-based studies published between 1990-2002 (McKeown et al, 2003). In all, participants were the primary carers, spending substantial time on caring responsibilities, including personal care and household tasks. Quality of life reduced over time, particularly where family income was lower and the person with MS was a spouse with more severe or unstable symptoms. All studies noted deteriorating health in carers, with substantial social and psychological impacts. Financial stresses were common due to costly equipment and assistance, alongside reducing personal income. Coping strategies of men and women differed, as did those of people in rural or urban locations. Studies had small participant numbers and lacked geographical diversity. A progression of this systematic review from 2002-2007 found 33 new studies from 11 countries (Corry and While, 2009). All were descriptive, finding similar impacts of caring, and additional insights into some positive experiences of caring and distinct needs as well as greater distress among spousal carers. People needed information, education, professional counselling, support groups and advocacy. More recently, studies have focused on specific transition stages with two focusing on people with substantial caring roles (Borreani et al, 2014; Bowen, 2011). Three focus groups of carers in Italy highlighted increased need for respite and time for personal needs such as health appointments, finding worse experiences among people with lower social support and less affluence (Borreani et al, 2014). In a Grounded Theory study of 25 people, barriers to accessing support included difficulty recognising personal needs, and lack of identification with the word 'carer,' with priority placed on their relationship rather than role (Bowen, 2011).

Fewer studies focus on services and support provided to families and carers of pwMS and no synthesis study was found. In-depth exploration was required, with recognition of complex and

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diverse characteristics, contexts and needs, and an underpinning ethos that family and carers are
persons with multiple perspectives and priorities. The research team prioritised a person-centred
approach and a focus on implementation. Person-centred practice focuses on the perspectives and
individual priorities of people, prioritising sharing of power between providers and those accessing
services (McCormack et al, 2017). This study aimed to provide person-centred recommendations
for practice, with potential for early implementation to enhance service provision. The great diversity
of context was inconsistent with a traditional systematic review approach to evaluating linear
relationships between intervention and outcome which could lead to rejection of potentially useful
interventions when implemented in sub-optimal contexts. The minimal literature available also
prompted a more evidence-generating approach. These considerations led to selection of Rapid
Realist Review (Wong et al, 2013) and Realist Evaluation (Wong et al, 2016) methodologies.

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Rapid Realist Review is an expedited form of realist synthesis which uses systematic search
strategies to identify published and grey literature, emphasising speed over comprehensiveness to
enable early implementation of results, appropriate in a nine-month commissioned study. In-depth
analysis of selected literature develops theoretical understanding of how different interventions or
strategies interact with diverse contexts to generate different outcomes (Saul et al, 2013; Rycroft-
Malone et al, 2012). For example, a confident information-user in a rural setting (context a) may
develop greater confidence and capability in caring roles (outcome) through using a mobile phone
app (mechanism), while a non-computer literate person in a rural setting (context b) may not. New
thinking is generated by enabling people with lived experience and professional expertise to
contribute to validity of the review through participation in an Expert Panel. The remit of this Expert
Panel includes identification of key literature and discussion of emerging findings, contributing to
generation of deeper understanding. Utility of the final product is enhanced through a Reference
Panel, including funder representatives (Saul et al, 2013). Theories developed through the iterative
review process are then evaluated from a realist perspective by seeking evidence that confirms,
clarifies or refutes them, using methods such as qualitative interviews to explore experiences and
views of family and carers.

Rapid Realist Review, followed by Realist Evaluation addressed the question: What kinds of support or intervention help families or carers of pwMS, who do they help, in what circumstances, and why? An overview of each stage is presented, followed by synthesis of the findings and discussion of related recommendations. In order to do explain the full research journey while demonstrating rigour, references to a repository of additional files are included to demonstrate the audit trail of extensive literature and data analysis that support theory development and evaluation [N.B. included in submission for peer review].

Methods

Rapid Realist Review

A three-phase, iterative search of published and grey literature focused on interventions to support family and carers of pwMS, integrated with Expert Panel discussion (see Table 1 and Figure 1 for summary and illustration of phases; Additional File (AF) 1 for key words and AF2 for panel membership). Paper selection prioritised articles relating directly to MS, and if few emerged, included analysis of results that related to other conditions. Data were extracted independently by three reviewers (CHB, DM, GB) to identify: mechanisms and sub-mechanisms which generated a response (M), contexts within which the mechanism took place or which may have influenced the mechanisms (C) and outcomes or impacts (O).

➔ Insert Table 1 here

➔ Insert Figure 1 here

The second phase literature search focused on published and grey literature with search terms based on the early draft working theories and chains of inference (search strategy in Table 1; search terms in AF1). Relevant articles were analysed for information supporting, refuting, or clarifying the draft working theories. These were then discussed by the Expert Panel, clarifying

wording and justification. Areas felt to require further exploration formed the basis for the third phase search (see Table 1 with search terms in AF1).

Realist Evaluation

Working theories were evaluated through analysis of lived experiences of being a family member or carer of someone living with MS using qualitative methods from a realist perspective. This aimed to confirm, refute, and/or clarify the working theories. Ethical approval was granted by the National Health Service London-Chelsea Research Ethics Committee (Reference: 16/LO/1194) and Higher Education Ethics Committee (REP 0130). Strategies summarised in Table 2 led to recruitment of 49 participants to 13 face-to-face interviews, two focus groups of 6 people per group, and 24 telephone interviews. Family members or carers of someone living with MS, or people whose caring roles ended in the past year were included if 18 years or older, and able to communicate sufficiently to be understood in English due to a lack of budget for interpretation.

➔ Insert Table 2 here

Topic guide questions aimed to elicit insights and experiences relating to the working theories and chains of inference (outline in AF3). Face-to-face interviews were conducted in participants' homes and focus groups in a community hall. Where permission was granted (for all but one person) interviews were digitally recorded for transcription. Three telephone recordings failed and detailed notes were used.

Thematic analysis focused on each theory in turn. Transcripts were read and re-read to identify experiences and views that confirmed, clarified or refuted a theory as a whole and chains of inference. Unexpected findings were discussed in relation to whether they were relevant to an existing theory, or might require further theory development. Themes were categorised as mechanisms, contexts or outcomes. Interactions between mechanisms and contexts were explored

in the text and linked outcomes then explored, enabling diagramming of MCO interactions. This enabled conclusions about whether or not evidence supported the strategy or intervention described in the theory as being helpful, and when and who it helps most. Where the intervention or strategy had not been experienced, evidence for chains of inference was used to reach conclusions about whether the intervention might help different people in different contexts.

Results

Rapid Realist Review of the literature

The first search phase resulted in seven publications (details in AF 4). One US-based intervention study and two from Australia included: a six-day intervention for 20 children of pwMS (Coles et al, 2007), a five-week education programme for 19 people (Finlayson et al, 2009) and a psycho-educational programme for 15 people (Hudson et al, 2012). The latter two studies were for carers of people receiving palliative care for cancer. All three studies used pre and post-intervention self-report questionnaires, one with a six-month follow-up (Coles et al, 2007) and one with focus groups (Finlayson et al, 2009). Three cross-sectional studies focused on relationships between different variables such as carer distress, health behaviours and communications with medical professionals about health-promoting behaviours. Outcomes were explored using telephone interviews with 530 informal carers (Buchanan et al, 2012) and questionnaires with 29 couples (Pakenham and Samios, 2013) and 67 carers (Penwell-Waines et al, 2016) of pwMS. Finally, a qualitative study used face-to-face interviews with people with progressive neurological conditions (including MS) and their carers (McCabe et al, 2008).

Debate regarding MCO interactions generated key theoretical principles, synthesised further to generate seven draft working theories that described theoretical principles thought to underpin interventions with potential to support family and carers of pwMS. Draft Working Theories and chains of inference are listed in AF5.

The second and third search phases resulted in 21 and five further articles respectively which supported the theories, summarised in AF6 and AF7. This in-depth, iterative, and discursive process generated seven working theories, presented in Table 3 alongside chains of inference and a summary of key sources, which were taken forward to the Realist Evaluation.

→ Insert Table 3 here

Realist Evaluation

Table 4 summarises participant characteristics. For auditability, detailed notes of the evaluation of each theory are included in AF8, with MCO Diagrams in AF 9a-g.

→ Insert Table 4 here

Minimal evidence existed for Theory 1 as a whole, with no reports of experiencing tailored information or advice. Analysis suggested that needs vary with diverse contextual factors, including coping style and unpredictable challenges over time. Analysis indicated that rather than tailored information, people need both proactive information and advice that is sensitive to their needs and responsive to changing needs over time, prompting amendment of the theory. There was clear evidence of the benefits of information and advice, for example, to increase feelings of preparedness for caring roles. As Charlotte (pseudonym) explained, *“The MS Nurse was the most helpful. She was instrumental in helping us understand... when he’s ill, he can’t think straight, so then I had the knowledge to say, ‘now, we know what’s going on here’.”* Some people had fewer capabilities in sourcing and using information effectively and many felt they had not received this during healthcare interactions, for example, Fred indicated *“I’ve gone to (health) appointments before... but they’ve never talked to me.”*

1 Information and advice were frequently used in problem solving and coping – Theory 2. Ralf
2 described his thinking: *“how can we get around this problem/ what’s the way of solving it?”* No
3 participant described receiving training in problem solving or coping, but where people described
4 existing capabilities, there were clear benefits to wellbeing and ability to continue caring. Simon
5 explained *“it’s just listening to how other people cope with that, and building up a toolkit....”* This
6 linked with Theory 6, as people used problem solving skills to make time for their own needs,
7 important for resilience.
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17 There were few experiences of coordinated help and support (Theory 3), as explained by Anne *“He*
18 *[primary carer] spends an inordinate amount of time which he doesn’t have... chasing down,*
19 *rushing around, dealing with professionals who don’t seem to speak to each other.”* The need for
20 practical help and support was clearly demonstrated and necessary for coping over time. Andrew
21 stated *“it is physically and mentally draining. For example, I have to put on slings, take off slings,*
22 *several times a day... that takes quite a bit of physical strength. I’m [over 70] years of age, and*
23 *obviously that leads to a few worries about, well, how long will I be able to keep going?”* Family and
24 social networks helped; where lacking, financial foundations were important. This linked Theory 3
25 with Theory 1, as people often needed information and advice about availability of financial support,
26 followed by practical support to complete relevant documentation. There was substantial evidence
27 that coordination of help and support would be beneficial, particularly for people who are
28 overwhelmed by their caring and other roles, as described by Paula: *“I am totally flummoxed by it... knowing what your rights and responsibilities are... it’s very difficult to get hold of that information,*
29 *so you get emotionally tired, so you just let it slide...”* However, help provided must be trustworthy
30 and flexible to people’s needs.
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53 People rarely experienced strength-based training (Theory 4) but there was evidence that people
54 with strengths such as a positive attitude achieved greater acceptance of their situations and more
55 positive relationships with the person with MS. Ted stated *“MS is not going to beat us, we’re going*
56 *to live a life as normal as MS allows to be possible.”* There was minimal evidence of support to
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engage in health-promoting activities (Theory 6), but where people protected time for themselves, there were clear benefits to wellbeing. Bill explained *"It's a question of looking after yourself... when the time comes to start moving people, you're strong... you'll not break down."* This linked with Theory 2 in relation to problem-solving and coping, and to Theory 3, as making time for self frequently required practical help and support. In particular, people needed time off through different forms of respite, as Mike asserted, *"my most important thing is to get a break. Because without that, I would not be able to cope continuously."* He explained that with respite, *"you can feel the weight coming off you."*

There was more evidence for Theory 5, which addressed emotional support through counselling - most often accessed by participants for reasons unrelated to the caring role. Analysis suggested that while some people found what they needed through social networks, most people could benefit from counselling during challenging periods. Debbie stated *"I find it really helpful to go and just let it all go."* Being supported to cope with negative emotions from any source could impact positively on caring experiences.

Theory 7 was most clearly confirmed, with more people having experienced carer support groups. For some people group interactions were highly beneficial and reduced feelings of isolation, helping people to source information and advice (Theory 1) and problem solve (Theory 2). Bill remembered *"we joined this group and I think that was the best thing we could have done... because you talk to people who have been going through the experience."* The theory was revised slightly to address evidence that some people preferred support from family and social networks.

A clear range of contextual factors played important roles across multiple theories in affecting whether or not a mechanism would be appropriate, or accessible, for the family member or carer, and therefore whether or not outcomes would be achieved. This is synthesised below.

Synthesis of Rapid Realist Review and Realist Evaluation Results

1 The final synthesis brought together all MCO interactions into one model (Figure 2). Common
2 contextual issues were treated as foundations in the model, influencing the potential for benefit from
3 any mechanism, or intervention.
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13 The model suggests that there can be a predominantly negative or positive picture of contexts and
14 outcomes. As these interact, people are more likely to identify with one side of the model. For
15 example, someone who self-identifies as a carer is more likely to seek information, advice and
16 support; if confident in this and tenacious in seeking support, they may implement more supportive
17 strategies that keep their workload manageable. Where someone uses problem-solving skills to
18 justify and identify time for self-care, their social relationships and wellbeing may be enhanced.
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28 While supportive positive contexts were frequently described as existing characteristics rather than
29 intervention outcomes, they provided evidence of positive mechanisms triggering more positive
30 outcomes summarised in the arrow on the right. If people who identify more with unsupportive
31 contexts are facilitated to develop more supportive contexts, their quality of life could be improved.
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40 Some contexts are less amenable to modification and should be respected, for example, stage of
41 disease progression and differences in personality and preference. Coping style affects what people
42 want at each stage; too much information, either at the start of their journey, or when overwhelmed
43 may exacerbate negative emotions. This may undermine a coping strategy of focusing on the
44 positive and not looking too far ahead. For others, mentally preparing themselves and planning for
45 the future has a stabilising effect. Options for intervention delivery should take account of these
46 differences. Other contexts represented in Figure 2 may be modifiable through a cluster of carefully
47 designed mechanisms as described in the centre of the model. These were translated into
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recommendations listed in AF10, cross-referenced according to relevant theories, which are now discussed.

Discussion

The recommendations of this realist evaluation are discussed in turn below.

Person-centred health-care delivery

Healthcare interactions should pay attention to the needs of the family member or carer, supporting them in acknowledging and prioritising their own needs, and identifying when they are at risk. This requires person-centred policy and its implementation. Consideration of the carer should be promoted by health and social care professionals, managers, and educators, advocated by the World Health Organization when stating that carers should be “able to attain maximal function within a supportive working environment” (World Health Organization, 2016).

Prompts to action may enable more proactive provision of information, advice and support to people who may be less confident in researching options, and who may be at risk of deteriorating personal outcomes. A screening tool such as the ‘CSNAT’ – Carer Support Needs Assessment Tool, developed for end of life care at home (Ewing and Grande, 2012), could identify carers’ needs.

Accessible and stable point of contact using different media

People should be directed early to a stable and accessible point of contact which provides signposting to many areas of information and advice. National and local information would be valued, including financial support, and holiday and respite options. Different media should be used, including telephone, website and paper based literature. The potential of social media should be explored as there is evidence of increasing use for seeking health information, with potential to increase engagement, empowerment and community building (Househ et al, 2014).

Increasing access and opportunities for training to support and enhance caring

Training in different capabilities to enhance caring would be valuable, including emotional resilience, information use, assertiveness in seeking support, time management, and moving and handling. It is very important that access and preference are considered, for example, by providing training for family or carers while the person with MS is engaged in a parallel activity at the same location, as advocated in a review of models of care for socially isolated older carers (Winterton and Warburton, 2011).

Personalised coaching through different media

Individual coaching may help, particularly for people who are less proactive in seeking help due to, for example, caring responsibilities, health concerns, and lack of time and energy. This could facilitate access to practical help by identifying possible strategies, access routes, and supporting applications for funding. This could be offered through mobile phone applications and social media. A telephone coaching intervention for carers of people with dementia was compared with respite day care, and a combination of both, in 54 informal carers (Van Mierlo et al, 2012). The group with both interventions reported a significantly lower burden and fewer mental health problems than the other two groups. However, a review of behavioural coaching using telehealth for carers of people with neurocognitive disorders found that self-efficacy scores for obtaining respite were greater than for those who received only basic education and telephone support (Steffen and Grant, 2016). This suggests an interplay between different interventions will provide best results, using varied media.

Well-trained, accessible and flexible care support and respite

The availability of well-trained and flexible care support, and respite facilities that can cater to more substantial caring needs are very important. Currently this can be very challenging for people with substantial caring workloads due to lack of access to facilities with good quality nursing care. Time may not be freed up by care support in the home, as carers stay present to monitor hygiene and

positioning due to lack of continuity and training of paid carers. Influencing activities are needed to increase both the availability of appropriate reserved short-term residential care and increase the level of training of all care staff in the needs of pwMS. A recent realist review focused on improving capabilities and standards in the support workforce for older people, with recognition of personal and organisational complexities and suggestions of practical ways forward (Rycroft-Malone et al, 2016; Williams et al, 2016).

Social and emotional support through peer support groups, volunteering and counselling

Social and emotional support are crucial to wellbeing and could help reduce the impacts of fewer financial resources. Social support was often described by participants as reducing over time, thereby decreasing emotional and practical help as needs increase. For those preferring social interaction, peer support groups can be invaluable, showing most evidence of benefit in this analysis - both emotional and practical. Online interactions were rarely mentioned, but comments indicated that whether face-to-face or online, good facilitation is important. Volunteer support for practical tasks and companionship for pwMS could relieve workload and enable the family member or carer to have time for their own needs. A systematic review of volunteer peer-support schemes for carers of people with dementia showed some benefit from four studies and advocated further research (Smith and Greenwood, 2014). Support for access to counselling - face-to-face or at a distance - would also be valuable for people lacking social support, or needing more skilled support.

The potential for a multicomponent internet based support intervention

The complex interacting unsupportive contexts, and family of interventions recommended from the evidence synthesis in Figure 2, lend themselves to a multicomponent intervention delivered through different media to optimise access. There has been some development and evaluation of such strategies for carers of people living with dementia using online and/or telephone support. A systematic review included 12 internet intervention studies and found increased confidence and self-efficacy, and decreased depression where interventions were individualised and multi-

1 component, such as coaching and peer support (Boots et al, 2014). Another systematic review of
2 telehealth interventions found greater impacts on self-efficacy and mood from multifaceted
3 interventions (Jackson et al, 2016). A more recent study evaluated a 14-week behavioural
4 intervention in 28 people who received educational materials and telephone coaching compared
5 with basic education and telephone support in 38 people (Steffen and Grant, 2016). The
6 intervention group's improvements in depression, mood and self-efficacy for obtaining respite were
7 not sustained beyond six months, suggesting that long-term conditions require longer-term
8 interventions. Within these papers, the contextual challenges and desired outcomes for carers of
9 people with dementia have many commonalities with carers of people with MS.
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22 This supports our conclusions that multicomponent interventions with multifaceted delivery have
23 potential to support carers of people with MS, if accessible as new needs arise. Ideally, components
24 would include many of the mechanisms in Figure 2, including a strategy to prompt action for those
25 at risk, a point of contact for information and advice, personalised coaching, access to online
26 training and counselling, support in accessing technology and equipment, information on respite,
27 links to local volunteer support, and well-facilitated peer support. This is an area with great promise
28 for development and research.
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40 Study considerations

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42 The emphasis on early implementation necessitated a rapid, non-comprehensive but systematic
43 literature review that generated theory which could be tested. Participant diversity was limited and
44 studies focused on engaging specific groups may need to address this (Bonevski et al, 2014).
45 Further studies with people who have left the caring relationship by choice, and young carers are
46 warranted,.
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56 To facilitate implementation, a stakeholder workshop was held at the UK MS Society Headquarters,
57 including people with varied expertise, such as service provision, research, influencing, innovation,
58 policy and IT. The researchers facilitated engagement with results and recommendations, and
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workshop participants then planned forward and organised work-streams and related working groups to include family and carers.

Conclusion

This Rapid Realist Review and Realist Evaluation addressed: what kinds of support or intervention help families or carers of people living with MS, who can they help, when and why. Detailed analysis of limited literature resulted in seven working theories addressing interventions relating to information and advice, problem-solving and coping, coordinated practical support, strengths-based training, emotional support, health-promoting activities and peer interactions. Qualitative evaluation with 49 people confirmed and clarified these theories. Analytic synthesis generated a model summarising a cluster of interventions that could help people with less supportive contexts develop their capabilities and expand their resources, with more positive outcomes. Resulting recommendations were discussed in a stakeholder workshop and aim to improve quality of life for family and carers of people with MS, delaying a 'tipping point' where capacity to care is overwhelmed by caring roles. A key message is: 'one size does not fit all,' supporting a person-centred approach.

Reference List

Bonevski, B., Randell, M., Paul, C., Chapman, K., Twyman, L., Bryant, J., Brozek, I. and Hughes, C. (2014) Reaching the hard-to-reach: a systematic review of strategies for improving health and medical research with socially disadvantaged groups. *BMC Medical Research Methodology*, 14: 1-29.

Boots, L.M.M., de Vugt, M.E., van Knippenberg, R.J.M., Kempen, G.I.J.M. and Verhey, F.R.J. (2014) A systematic review of Internet-based supportive interventions for caregivers of patients with dementia. *International Journal of Geriatric Psychiatry*, 29: 331-344.

Borreani, C., Bianchi, E., Pietrolongo, E., Rossi, M., Cilia, S., Giuntoli, M., Giordano, A., Confalonieri, P., Lugaresi, A., Pattie, F., Grasso, M.G., De Carvalho, L.L., Palmisano, L., Zaratin, P., Battaglia, M.A. and Solari, A. (2014) Unmet needs of people with severe multiple sclerosis and their carers: Qualitative findings for a home-based intervention. *PLoS ONE* 9(10): e109679. doi:10.1371/journal.pone.0109679

Bowen, C. (2011). Advanced multiple sclerosis and the psychosocial impact on families. *Psychology and Health*, 26(1):113-27.

Buchanan, R.J., Huang, C. and Crudden, A. (2012) Use of the Internet by Informal Caregivers Assisting People with Multiple Sclerosis *Journal of Technology in Human Services*, 30(2): 72-93.

Buckner, L. and Yeandle, S. (2015) Valuing Carers 2015: the rising value of carers' support. Carers UK. <http://www.carersuk.org/for-professionals/policy/policy-library/valuing-carers-2015>

Carers UK, 2014. Carers at Breaking Point. <https://www.carersuk.org/for-professionals/policy/policy-library/carers-at-breaking-point-report>

Coles, A.R., Pakenham, K.I. and Leech, C. (2007) Evaluation of an intensive psychosocial intervention for children of parents with multiple sclerosis. *Rehabilitation Psychology*, 52(2): 133-142.

Corry, M. and While, A. (2009) The needs of carers of people with multiple sclerosis: a literature review. *Scandinavian Journal of Caring Sciences*, 23(3): 569-88.

EUROFAMCARE Research Consortium (2005) Overview summary of the EUROFAMCARE Trans-European-Survey Report. The EUROFAMCARE consortium. https://www.uk-e.de/extern/eurofamcare/documents/overview_teusure.pdf

Ewing, G. and Grande, G. (2012) Development of a Carer Support Needs Assessment Tool (CSNAT) for end-of-life care practice at home: A qualitative study. *Palliative Medicine*, 27(3):244-56.

1 Finlayson, M., Preissner, K. and Garcia, J. (2009) Pilot Study of an Educational Programme for
2 Caregivers of People Ageing with Multiple Sclerosis. *The British Journal of Occupational Therapy*,
3
4 72(1): 11-19.
5
6
7 Harwood, R.H., Sayer, A.A. and Hirschfeld, M. (2004) Current and future worldwide prevalence of
8
9 dependency, its relationship to total population, and dependency ratios. *Bulletins of the World*
10
11 *Health Organization*. 82(4):251-8.
12
13
14 Higginson, I.J. (2006) Symptom prevalence and severity in people severely affected by multiple
15
16 sclerosis. *Journal of Palliative Care*. 22(3):158-16.
17
18
19 Hudson, P.L., Lobb, E.A., Thomas, K., Zordan, R.D., Trauer, T., Quinn, K., Williams, A. and
20
21 Summers, M. (2012) Psycho-educational group intervention for family caregivers of hospitalized
22
23 palliative care patients: Pilot study. *Journal of Palliative Medicine*, 15(3): 277-281.
24
25
26 Househ, M., Borycki, E. and Kushniruk, A. (2014) Empowering patients through social media: The
27
28 benefits and challenges. *Health Informatics Journal* 20(1): 50-8.
29
30
31 Jackson, D., Roberts, G., Wu, M.L., Ford, R. and Doyle, C. (2016) A systematic review of the effect
32
33 of telephone, internet or combined support for carers of people living with Alzheimer's, vascular or
34
35 mixed dementia in the community. *Archives of Gerontology and Geriatrics*. 66: 218-36.
36
37
38 Kremenchutzky, M. (2006) The natural history of multiple sclerosis: a geographically based study 9:
39
40 observations on the progressive phase of the disease. *Brain*. 129: 584-94.
41
42
43 Mackenzie, I.S., Morant, S.V., Bloomfield, G.A., MacDonald, T.M. and O'Riordan, J. (2014)
44
45 Incidence and prevalence of multiple sclerosis in the UK 1990–2010: a descriptive study in the
46
47 General Practice Research Database. *Journal of Neurology, Neurosurgery and Psychiatry*, 85(1):
48
49 76–84.
50
51
52 McCabe, M.P., Roberts, C. and Firth, L. (2008) Satisfaction with services among people with
53
54 progressive neurological illnesses and their carers in Australia. *Nursing & Health Sciences*, 10(3):
55
56 209-215.
57
58
59
60
61
62
63
64
65

McCormack, B. and McCance, T. (2017) *Person-centred Practice in Nursing and Health Care-Theory and Practice*. 2nd ed. Chichester: Wiley Blackwell.

McKeown, L.P., Porter-Armstrong, A.P. and Baxter. G.D. (2003) The needs and experiences of caregivers of individuals with multiple sclerosis: a systematic review. *Clinical Rehabilitation*, 17(3): 234-248.

MS Society (2015) Commissioning brief: Families and Carers of people with MS: Evidence Review & Scoping Study. MS Society.

<https://www.mssociety.org.uk/sites/default/files/Family%20and%20Carers%20Commissioning%20Brief%202015-09-24.pdf>

Pakenham, K.I. and Samios, C. (2013) Couples coping with multiple sclerosis: a dyadic perspective on the roles of mindfulness and acceptance. *Journal of Behavioral Medicine*, 36(4): 389-400.

Penwell-Waines, L., Goodworth, M.R., Casillas, R.S., Rahn, R. and Stepleman, L. (2016) Perceptions of caregiver distress, health behaviors, and provider health-promoting communication and their relationship to stress management in MS caregivers. *Health Communication*, 31(4): 478-484.

Rosati, G. (2001) The prevalence of multiple sclerosis in the world: an update. *Neurological Sciences*, 22(2):117-39.

Runia, T.F. (2012) Recent gains in clinical multiple sclerosis research. *CNS and Neurological Disorders: Drug Targets*, 11(5):497-505.

Rycroft-Malone, J., Burton, C.R., Williams, L., Edwards, S., Fisher, D., Hall, B., McCormack, B., Nutley, S., Seddon, D. and Williams, R. (2016) Improving skills and care standards in the support workforce for older people: a realist synthesis of workforce development interventions, HS&DR. <http://dx.doi.org/10.3310/hsdr04120>. <http://www.journalslibrary.nihr.ac.uk/hsdr/volume-4/issue-12#abstract>

Rycroft-Malone, J., McCormack, B., Hutchinson, A.M., DeCorby, K., Bucknall, T.K., Kent, B., Schultz, A., Snelgrove-Clarke, E., Stetler, C.B., Titler, M., Wallin, L. and Wilson, V. (2012) Realist

synthesis: illustrating the method for implementation research. *Implementation Science*, 12(7): 33-33.

Saul, J.E., Willis, C.D., Bitz, J. and Best, A. (2013) A time-responsive tool for informing policy making: rapid realist review. *Implementation Science*, 8(1): 103-103.

Smith, R. and Greenwood, N. (2014) The Impact of Volunteer Mentoring Schemes on Carers of People with Dementia and Volunteer Mentors: A Systematic Review. *American Journal of Alzheimers Disease & Other Dementias*, 29(1): 8-17.

Steffen, A.M. and Grant, J.R. (2016) A telehealth behavioural coaching intervention for neurocognitive disorder family carers. *International Journal of Geriatric Psychiatry*. 31(2): 195-203.

Van Mierlo, L.D., Meiland, F.J.M. and Droes, R.M. (2012) Dementelcoach: effect of telephone coaching on carers of community-dwelling people with dementia. *International Psychogeriatrics* 24(2): 212-22.

Williams, L., Rycroft-Malone, J., Burton, C.R., Edwards, S., Fisher, D., Hall, B., McCormack, B., Nutley, S.M., Seddon, D. and Williams, R. (2016) Improving skills and care standards in the support workforce for older people: a realist synthesis of workforce development interventions *BMJ Open*. 6(8): e011964.

<http://bmjopen.bmj.com/content/6/8/e011964.full?keytype=ref&ijkey=M3UfSPkB6laYvgH>

Winterton, R. and Warburton, J. (2011) Models of care for socially isolated older rural carers: barriers and implications. *Rural and Remote Health*, 11:1678.

Wong, G., Greenhalgh, T., Westhorp, G., Buckingham, J. and Pawson, R. (2013) RAMESES publication standards: realist syntheses. *BMC Medicine*, 11: 21.

Wong, G., Westhorp, G., Manzano, A., Greenhalgh, J., Jagosh, J. and Greenhalgh, T. (2016) RAMESES II reporting standards for realist evaluations. *BMC Medicine*, 14: 96.

World Health Organization. (2016) *Framework on integrated, people-centred health services. Report by the Secretariat*. Sixty-Ninth World Health Assembly A69/39. World Health Organization.

<http://www.who.int/servicedeliverysafety/areas/people-centred-care/framework/en/>

List of Tables, Figures, Additional Files

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Additional Files (N.B. these have been included in this manuscript but would be moved to an online repository if the article is accepted):

Additional File 1. Literature Search Terms: Phases 1, 2 and 3

File name: Additional File 1 RRR Search Terms [Microsoft Word doc; contains lists of typed key words for literature searches]

Additional File 2: Membership of the Research Team, Expert Panel and Reference Panel
[anonymised for initial submission]

File name: Additional File 2 Panel membership [Microsoft Word doc; contains a list of members of the research team, expert panel and reference panel]

Additional file 3. Interview questions in relation to theories

File name: Additional File 3 Realist Evaluation Topic Guide [Microsoft Word doc; contains lists of questions / topics used for qualitative interviews]

Additional File 4. Results of Phase 1 Literature Search and Theoretical principles identified

File name: Additional File 4 Results Phase 1 Literature Search [Microsoft Word doc; contains results of the Rapid Realist Review phase 1 literature search with the theoretical principles derived from each article]

Additional File 5. Draft Working Theories and Chains of Inference

File name: Additional File 5 Draft working theories and chains of inference [Microsoft Word doc; contains tabulated working theories and chains of inference]

Additional File 6. Results of Phase 2 Literature Search

File name: Additional File 6 Results of phase 2 literature search [Microsoft Word doc; contains results of the Rapid Realist Review phase 2 literature search]

Additional File 7. Results of Phase 3 Literature Search

File name: Additional File 7 Results of phase 3 literature search [Microsoft Word doc; contains results of the Rapid Realist Review phase 3 literature search]

Table 8. Summary of evaluation of each theory through qualitative analysis

File name: Additional File 8 Summary of evaluation of each theory [Microsoft Word doc; contains results of the qualitative analysis in relation to each theory]

Additional File 9. MCO Interaction Diagrams

9a. Theory 1 – Information and Advice: MCO Interactions

9b. Theory 2 – Coping and Problem-Solving: MCO Interactions

9c. Theory 3 – Practical Help and Support: MCO Interactions

9d. Theory 4 – Acceptance and Attitude: MCO Interactions

9e. Theory 5 – Emotional Support: MCO Interactions

9f. Theory 6 – Looking After Self: MCO Interactions

9g. Theory 7 – Social Contact: MCO Interactions

File name: Additional File 9a-g Realist Evaluation MCO mind-maps [Microsoft Word doc; contains diagrams of MCO interactions for Theories 1 - 7]

Additional File 10 Recommendations for Policy, Practice, Influencing Activities and Research

File name: Additional File 10 Recommendations for Policy, Practice, Influencing Activities and Research [Microsoft Word doc; contains tabulated recommendations]

Figure 1

Figure 2

Table 1. Summary of Rapid Realist Review Phases and Outcomes, cross-referenced to tables and figures

Stage	Process (took place in 2016)	Outcomes
Preparation phase	Use of the research question identified by the funders to develop a 'purpose statement'; exploratory search to confirm initial search strategy; identification of key literature known to Expert Panel	Initial search strategy for the Phase 1 literature search
Phase 1 literature search:	<p><u>Databases:</u> Full-text, English language articles published 2000-2016 using databases: ASSIA (Applied Social Sciences Index and Abstracts); CINAHL Plus (Cumulative Index of Nursing and Allied Health Literature); NHS e-library; Web of Science; Cochrane Library and Embase;</p> <p><u>Search terms</u> in Additional File 1.</p> <p><u>Inclusion criteria:</u> articles focusing on the carer; initial focus on the condition of interest only, opened to further conditions later if there are gaps; ability to identify context, mechanism and outcome</p> <p><u>Exclusion criteria:</u> clinical setting; medical intervention / medication-related; articles focusing on the person with the condition</p>	Figure 1 summarises the process and the result of 2037 hits, narrowed down to seven articles for detailed analysis. Results and theoretical principles identified (Table 3); Theoretical principles extracted from each paper synthesised into draft working theories with clearly identified MCO and chains of inference (Table 4);
Phase 2 literature search	<p><u>Databases:</u> as above; additional grey literature sources: Scottish Government; Health Scotland; GreyLit.org; Kings Fund; Joseph Rowntree Foundation; Carers UK; Carers of East Lothian; OAlder; MS Society Australia; MS Research Australia; Multiple Sclerosis International Federation; MS Society UK/Scotland; VOCAL; Department of Health Social Care blog; OpenGrey.eu</p> <p><u>Search terms:</u> in Additional File 1.</p> <p><u>Inclusion criteria:</u> as above</p> <p><u>Exclusion criteria:</u> as above</p>	Figure 1 summarises the process and further 3203 hits identified, with 21 relevant articles and documents identified to further inform the draft working theories (Table 5).
Expert Panel meeting	Discussion of draft working theories and contributory analysis as well as areas that may not have been sufficiently addressed.	Identification of specific changes to the phrasing of draft working theories and of areas for further searching.
Phase 3 literature search	<p><u>Databases:</u> as for phase 1;</p> <p><u>Search terms:</u> in Additional File 1, designed to address issues raised in the Expert Panel Meeting.</p> <p><u>Inclusion criteria:</u> as above</p> <p><u>Exclusion criteria:</u> as above</p>	Identification of five additional papers that supported the draft working theories (Table 6). Articulation and agreement of Working Theories to take forward to the Realist Evaluation (Table 7).

Table 2. Summary of Realist Evaluation Data Collection

Data collected	Recruitment strategies	Quantity of data
13 face-to-face individual interviews: Lothian	Recruitment optimised through Expert Panel members who identified possible participants and provided information packs; this represented: Astley Ainslie Hospital and Lanfine Unit, Western General Hospital, Voices of Carers Across Lothian (VOCAL) and Carers of East Lothian (CoEL).	688 minutes of interviews; mean of 53 minutes per interview
24 telephone interviews: UK-wide	Recruitment optimised through MS Society (website and discussion forum; emails to local branches, attendance at the 'Carers' Area' at MS Life event; advert in 'Team Spirit' bi-monthly newsletter to branch volunteers); social media tweets and blogs; CoEL magazine, mailshot to VOCAL members; poster in Edinburgh MS Centre; press releases	estimated 795* minutes of data; mean of 33 minutes per interview
12 focus group participants: Macclesfield	Recruitment facilitated by Expert Panel member through a carers' group at a specific branch of the MS Society	173 minutes of focus group data; 2 focus groups; mean of 87 minutes per group

* one telephone interview was not recorded at the request of the participant and the mean of all other telephone interview durations was used in calculations

Table 3. Working Theories and Chains of Inference

Working theories	Chains of inference	Key sources <i>(note: references lists are in the appropriate Additional File)</i>
Theory 1: Information and advice to family or carers of people with MS (m) that is individually tailored (c) may mitigate the negative impacts of caring (o)	M: Information and advice, knowledge and understanding, caring role C: Carer characteristics, disease progression, location, mode, intensity, educator skills O: Confidence, knowledge, preparedness, well-being (physical and emotional)	Phase 1 search (see AF3): Buchanan et al. (2012); Coles et al. (2007); Finlayson et al. (2009); Hudson et al. (2012); Phase 2 search (see AF6): Banbury et al. (2014); Francobandera (1992); Lancer (2007); Ray and Street (2006); Takamoto and Hori (2016)
Theory 2: Training in problem solving and coping strategies (m) taking into account characteristics and emotional state of family or carers of people with MS (c) may result in greater confidence and resilience as a carer (o).	M: Training in coping strategies and problem solving skills C: Carer characteristics, existing emotional state, face to face interventions O: Creativity, resourcefulness, capacity, resilience and reduce stress	Phase 1 search (see AF3): Buchanan et al. (2012); Coles et al. (2007); Finlayson et al. (2009); Hudson et al. (2012); Phase 2 search (see AF6): Banbury et al. (2014); Eisler et al. (2016); Masoodi et al. (2013); McConigley et al. (2014); Tams et al. (2016); Phase 3 search (see AF7): Horner (2013);
Theory 3: Practical help and support for family or carers of people with MS that is coordinated (m) and tailored to the carers situation (c) may result in an improved experience of caring (o).	M: Access, coordination, availability, comprehensiveness, technology, communication skills C: Coordination, location, availability, carer communication skills, technology O: Access to tailored services, tailored action plan, capacity to care, experience of caring	Phase 1 search (see AF3): McCabe et al. (2008); Phase 2 search (see AF6): Boland et al. (2012); Kristjanson et al. (2006); Rollero (2016);
Theory 4: Strengths based training for family or carers of people with MS (m) particularly where there is a spousal/partner relationship (c) may result in improved family relationships and acceptance of their situation (o).	M: Relationship management, acceptance, reframing, family relationships, family communication, mindfulness C: Person centred, relationship to people with MS, emotional/cognitive status of people with MS and carer, credibility and training O: Mental attitude/family relationships, acceptance, family communication, emotional state	Phase 1 search (see AF4): Pakenham and Samios (2013); Phase 2 search (see AF6): Boland et al. (2012); Carletto et al. (2016); Liedstrom et al. (2010); Nordin and Rorsman (2012); Weisser et al. (2015);

<p>Theory 5: Individual sharing of experiences and emotions by family or carers of people with MS with a third party (m) in a safe environment (c) may result in increased acceptance of their situation (o).</p>	<p>M: Discussion with third party, sharing, emotional expression, validation, communication C: Interactive, tailored mode, person centred, safe environment, third party O: Acceptance, validation, understanding, capacity, reduced distress</p>	<p>Phase 1 search (see AF4): Buchanan et al. (2012); Coles et al. (2007); Finlayson et al. (2009); Phase 2 search (see AF6): Banbury et al. (2014); Busch et al. (2014); McConigley et al. (2014); Rollero (2016); Phase 3 search (see AF7): Horner (2013); Nagl-Cupal et al. (2015);</p>
<p>Theory 6: Opportunities to engage in health promoting activities for family or carers of people with MS (m) facilitated by an appropriate range of support (c) may result in improved physical and mental health and well-being (o).</p>	<p>M: Respite care, step out of caring role, social/recreational activities, self-preservation, communication with HCPs C: Carer characteristics, disease progression, location, timing, existing stress levels, expertise of HCPs O: Health and well-being (physical and emotional), mental attitude, resilience</p>	<p>Phase 1 search (see AF4): Coles et al. (2007); Penwell-Waines et al. (2016);</p>
<p>Theory 7: Opportunities for interaction between family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o).</p>	<p>M: Social contact, physical/virtual interaction, discussion groups, self-help groups C: Carer characteristics, technology, degree of interaction, synchronous/asynchronous, location O: Social contact, emotional state, recognition 'not alone', capacity to care</p>	<p>Phase 1 search (see AF4): Buchanan et al. (2012); Coles et al. (2007); Finlayson et al. (2009); Phase 3 search (see AF7): Bjorgvinsdottir and Halldorsdottir (2014);</p>

Table 4. Participant Characteristics

Characteristic	Summary
Sex of the family member or carer	Female:23 Male: 26
Age bracket of the family member or carer	31-49: 6 50-65: 25 66+: 18
Relationship of the family member or carer to person with MS	Spouse: 40 Partner: 3 Sibling: 2 Child: 4
Living situation of the family member or carer	In the same home as the person living with MS: 39 In a different home to the person living with MS: 10
Occupation of the family member or carer	Employed full-time: 7 Employed part-time: 7 Engages in voluntary employment: 2 Not employed: 30 Not revealed: 3
Occupation of the person living with MS	Employed: 6 Not employed: 42
Mobility and independence of the person living with MS	Able to get around independently outside the home: 10 Able to get around with help outside the home: 19 Able to get around independently inside the home: 14 Able to get around with help inside the home: 23 Reliant on a wheelchair most / all of the time: 26 Unable to change position without help: 24
The family member or carer has other carer roles	Yes: 7 No: 39 Not revealed: 3
Ethnicity of the family member or carer	White: 43 Other ethnic group:2 Declined to answer: 4
Sexual orientation of the family member or carer	Heterosexual: 44 Bisexual:1 Declined to answer:4

Additional File 1. Literature Search Terms: Phases 1, 2 and 3 – implemented in 2016

PHASE 1 search - developed following a scoping search					
Condition	AND	Caring Person	And	Type of intervention	
“multiple sclerosis” OR		“carer” OR		“support” OR	
		“caregiver” OR		“burden” OR	
		“famil*” OR		“unmet need*”	
		“partner”			
PHASE 2 search – developed from Phase 1 daft theories and chains of inference					
Condition	AND	Mechanism	AND	Context	AND Outcome
“multiple sclerosis” OR “Parkinson*” OR “Huntington*” OR “motor neurone” OR “neurodegenerative”		“Education” OR “knowledge” OR “understanding” OR “caregiving role”		“Carer age” OR “years of caregiving” OR “relationship to” OR “disease progression” OR “location” OR “mode” OR “intensity” OR “educator skills”	“Confidence” OR “knowledge” OR “preparedness” OR “emotional state”
As above		“Coping strateg*” OR “problem solving”		“Carer age” OR “years of caregiving” OR “relationship to” OR “disease progression” OR “existing emotional state”	“Creativity” OR “resourcefulness” OR “capacity”
As above		“Access” OR “coordination” OR “availability” OR “comprehensiveness” OR “technology” OR “communication skills”		“Coordination” OR “location” OR “availability” OR “carer communication skills” OR “technology”	“Tailored services” OR “management strategies” OR “capacity”
As above		“Relationship management” OR “acceptance” OR “reframing” OR “family relationships” OR “family communication” OR “mindfulness”		“Person centred” OR “relationship to” OR “emotional status” OR “cognitive status” OR “credibility” OR “training”	“Mental attitude” OR “family relationships” OR “acceptance” OR “family communication” OR “emotional state”
As above		“Discussion” OR “sharing” OR “emotional expression” OR “validation” OR “communication”		“Interactive” OR “tailored” OR “mode” OR “person centred” OR “safe environment”	“Acceptance” OR “validation” OR “understanding” OR “capacity” OR “not alone”
As above		“Respite care” OR “step out of caring role” OR “social” OR “recreational” OR “self-preservation” OR “communication with HCPs”		“Carer age” OR “years of caregiving” OR “relationship to” OR “disease progression” OR “location” OR “timing” OR “stress levels”	“Health promoting behaviour” OR “mental attitude” OR “resilience”
As above		“Social contact” OR “physical interaction” OR “virtual interaction” OR “discussion groups” OR “self-help groups”		“Carer age” OR “years of caregiving” OR “relationship to” OR “technology” OR “interaction” OR “synchronous” OR “location”	“Social contact” OR “emotional state” OR “not alone” OR “capacity”

PHASE 3 search – to explore ideas emerging from Expert Panel Discussion of the draft working theories					
Condition	AND	Mechanism	AND	Context	AND Outcome
“multiple sclerosis” OR		“family therapy” OR		“transition” OR	“resilience”
“Parkinson*” OR		“letting go” OR		“marital breakdown” OR	
“Huntington*” OR		“coping” OR		“divorce” OR	
“motor neurone” OR		“education” OR		“young carers” OR	
“neurodegenerative”					
OR					

Additional File 2: Membership of the Research Team, Expert Panel and Reference Panel

Team / Panel	Name	Representing
Research Team: conducting the project and contributing to Expert and Reference Panel discussions	ANONYMISED	FOR ADDITION IF ACCEPTED
Expert Panel: contribution to validity of the product		
Reference Panel: contribution to utililty of the product		

Additional file 3. Interview questions in relation to theories

Primary question and example prompt questions (used as appropriate)	Key draft working theory being explored (n.b. there is overlap)
What is your relationship with the person in your life who has MS? How long have they had MS for? <i>Example prompts:</i> Do you consider yourself to be this person's carer? How much care do you give just now?	<i>Awareness and acceptance in family or carers of people with MS (m) through strength based approaches (c) may result in improved family relationships (o)</i>
Firstly we would like to explore how people deal with the diagnosis of MS, both as a carer of that person and the person receiving the diagnosis. How did you feel when your (partner / relative / spouse) received the diagnosis of MS? <i>Example prompts:</i> What sort of impact did that have on you? Do you think it has changed your relationship? What strategies / approaches do you think were useful in helping you deal with the diagnosis?	<i>Developing resilience in family or carers of people with MS (m) through face-to-face interaction and opportunities for discussion (c) may result in a greater capacity to care (o)</i>
Can you talk about how you have found being a family member or care of a person with MS? How does being a carer / family member affect your day to day life? <i>Example prompts:</i> Can you tell me about some of the challenges you have faced? When you encounter a new problem or difficulty or come across a situation you haven't faced before, how do you try and solve it? What about difficulties which you haven't been able to resolve yourself - at what point would you look for outside help? Who would you ask for help from?	<i>Education of family or carers of people with MS (m) delivered through individually tailored modes (c) may result in improved ability to care and reduced distress (o)</i>
We are interested in exploring the educational support carers and family member have had receive. Can you tell me about any educational support your or your family have had? <i>Example prompts:</i> Was it about the disease or being a carer or how to cope as a family? Was it aimed at giving you practical support, like how to physically help some with movement or at giving you information about available services or the practical, emotional or economic support available to you? Were you offered this education support or did you have to seek it out? Thinking about that support – how was it delivered - Individual or group, location, via the internet / technology? What elements were helpful, why. If it wasn't helpful, why not? Was there anything that could have been done differently or added? Was it offered at the right time? What sort of impact did it have on you? How would you have liked to receive information or education? How would that have helped you with your caring role?	<i>Access to practical help and support for family or carers of people with MS (m) through coordinated services (c) may result in improved management strategies and overall experience of caring (o)</i>

We are interested in exploring further your experience of services which support you and your person with MS. Do you currently get any practical help or support? *Example prompts:* By practical help I mean someone to come and help you at home with the shopping or gardening for instance, or does your person with MS attend somewhere such as a day center or respite care; How does it impact you – physically and emotionally? Does anyone co-ordinate the available services? In an ideal world what practical help and support would you like? If no do you have any idea what sort of practical help and support would help you? How much would you like and when? Format – face to face, online? Someone to co-ordinate all the available services and to help you access them?

Holistic self-management by family or carers of people with MS (m) with an appropriate range of support (c) may result in health promoting behaviours (o)

We are also interested in looking in more depth about the impact of your caring role on you. Does the support you get allow you to look after yourself, to take care of your own physical or mental health? *Example prompts:* – what kind of support do you get? How does it impact on you? If no, is that because of limited support? how does that affect you? What difference do you think that ability to take care of yourself would make you feel? Do you receive respite care for the person with MS – can you tell me about that? How much / how long/ availability/ difficulties accessing – what would be your ideal? Do you find taking care of yourself easier or harder than looking after other people? Why is that? What do you do to help improve your physical or mental health? Have any HCP discussed these kind of behaviours with you or advised you?

Sharing experiences and emotions of family or carers of people with MS (m) in safe environments (c) may result in increased acceptance (o)

We would like to know more about people feel about support groups. Are you a member of any support groups? Either MS or a general carers support group? *Example prompts:* if yes, can you tell me about the group and how you have found being a member? How has that helped you? Have your requirements from such groups changed over time? Do you have experience of online social media? Have you used online support groups or forums? Can you tell me about your experience of them?

Opportunities for interaction for family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o)

Thinking more about the ideas of support from others in a similar situation - do you have opportunities to talk to other carers / family members of people living with MS? *Example prompts:* If yes, could you tell me more about this – what sort of interaction is it, who arranges it, is it face-to-face or over social media? How does this impact on you? If no – is this by choice or because of your circumstances, or because you are unaware of how to access others? Do you think it could be helpful? In what circumstances. Is there anything help or support which we haven't touched on today that you feel could have helped you in your journey as a carer?

Opportunities for interaction for family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o)

Finally, thinking about what we have talked about today, have you had any experiences which we haven't had a chance to talk about which you would like to discuss?

Additional File 4. Results of Phase 1 Literature Search and Theoretical principles identified

Draft theory	1	2	3	4	5	6	7
PAPER	Education of family or carers of people with MS (m) delivered through individually tailored modes (c) may result in improved ability to care and reduced distress (o)	Developing resilience in family or carers of people with MS (m) through face-to-face interaction and opportunities for discussion (c) may result in a greater capacity to care (o)	Access to practical help and support for family or carers of people with MS (m) through coordinated services (c) may result in improved management strategies and overall experience of caring (o)	Awareness and acceptance in family or carers of people with MS (m) through strength based approaches (c) may result in improved family relationships (o)	Sharing experiences and emotions of family or carers of people with MS (m) in safe environments (c) may result in increased acceptance (o)	Holistic self-management by family or carers of people with MS (m) with an appropriate range of support (c) may result in health promoting behaviours (o)	Opportunities for interaction for family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o)
Buchanan et al. (2012)	✓	✓			✓		✓

Identification of factors associated with internet use by informal carers assisting people with MS (US);

Theoretical principles identified:

- Younger carers of people with MS(c) are more likely to find the internet (m) a useful source of information (o).
- More educated carers of (c) are more likely to find the internet (m) a useful source of information (o).
- Carers who provide a high level of care (c) are more likely to use the internet (m) as a source of information (o)
- Number of years of caring (c) is likely to decrease the internet (m) as a useful source of information (o).
- Carers of people with MS (c) may find use of the internet (m) reduces their sense of social isolation (o).

Coles, et al. (2007)	✓	✓			✓	✓	✓
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Evaluation of an intensive psychosocial intervention for children of parents with MS with aims of increasing knowledge, reducing negative impact of caring and producing better adjustment (Australia);

Theoretical principles identified:

- Young carers of people with MS (c) may benefit emotionally (o) from the opportunity to share their experiences with people in the same situation (m).
- Young carers of people with MS (c) may benefit from meeting people in the same situation (m) resulting in a reduced sense of social isolation (o).
- Young carers of people with MS (c) may be more able to develop coping strategies (o) from an increased understanding of the condition (m).
- Young carers of people with MS (c) may benefit emotionally (o) from opportunities to step out of the caring role (m).
- An intensive face to face group based (c) educational programme (m) may result in an increased knowledge and understanding of MS in young carers and thereby increase their supportiveness to a parent with MS (o).

Draft theory	1	2	3	4	5	6	7
PAPER	Education of family or carers of people with MS (m) delivered through individually tailored modes (c) may result in improved ability to care and reduced distress (o)	Developing resilience in family or carers of people with MS (m) through face-to-face interaction and opportunities for discussion (c) may result in a greater capacity to care (o)	Access to practical help and support for family or carers of people with MS (m) through coordinated services (c) may result in improved management strategies and overall experience of caring (o)	Awareness and acceptance in family or carers of people with MS (m) through strength based approaches (c) may result in improved family relationships (o)	Sharing experiences and emotions of family or carers of people with MS (m) in safe environments (c) may result in increased acceptance (o)	Holistic self-management by family or carers of people with MS (m) with an appropriate range of support (c) may result in health promoting behaviours (o)	Opportunities for interaction for family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o)
Finlayson, et al. (2009)	✓	✓			✓		✓

Pilot educational programme to enable carers of people ageing with MS to feel more prepared, reframe their experience and seek more practical assistance (US);

Theoretical principles identified:

- Mode of delivery (m) may be important when seeking to develop coping strategies and problem solving skills (o) for older carers of people with MS (c).
- Group interaction (m) may be important for older carers of people with MS (c) when seeking to reduce the negative emotions associated with caring (o).
- The opportunity to meet others in a similar situation (m) may help to alleviate social isolation and validate feelings (o) for older carers of people with MS (c).
- Professional background and expertise of the educator (m) may be important in order to successfully engage (o) older carers of people with MS (c) in psychoeducational programmes.
- A psychoeducational programme (m) may help to increase an older carer of a person with MS (c) preparedness (managing change, identifying services, problem solving) (o).

Hudson, et al. (2012)	✓	✓					
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Pilot of a psycho-educational group intervention delivered to carers supporting a relative receiving hospital based palliative care (Australia);

Theoretical principles identified:

- Stage and manifestation of condition (c) may be important in determining type of information and emotional support required (m) that is seen as useful by carers of people with MS (o).

Draft theory	1	2	3	4	5	6	7
PAPER	Education of family or carers of people with MS (m) delivered through individually tailored modes (c) may result in improved ability to care and reduced distress (o)	Developing resilience in family or carers of people with MS (m) through face-to-face interaction and opportunities for discussion (c) may result in a greater capacity to care (o)	Access to practical help and support for family or carers of people with MS (m) through coordinated services (c) may result in improved management strategies and overall experience of caring (o)	Awareness and acceptance in family or carers of people with MS (m) through strength based approaches (c) may result in improved family relationships (o)	Sharing experiences and emotions of family or carers of people with MS (m) in safe environments (c) may result in increased acceptance (o)	Holistic self-management by family or carers of people with MS (m) with an appropriate range of support (c) may result in health promoting behaviours (o)	Opportunities for interaction for family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o)
McCabe et al. (2008)			✓				

Survey of satisfaction with different types of services and support networks accessed by people with progressive neurological illnesses and their carers (Australia)

Theoretical principles identified:

- There may be a range of basic services (m) that are required by carers of people with MS that need to be individually tailored at different stages of MS progression (c) in order to effectively provide support (o).
- Respite care (m) may be essential for carers of people with MS (c) to promote resilience and allow people with MS to be cared for at home for as long as possible (o).
- Integration of services (m) provided for people with MS (c) may help to reduce the burden and/or provide a more efficient service for their carers (o).
- There may be more specialised services (m) required for carers of people with MS (c) (eg: advocacy, employment assistance, financial services, peer support) that will allow them to cope better with their caring role (o)

Pakenham & Samios (2013)				✓			
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Examination of the effects of mindfulness and acceptance on adjustment (depression, anxiety, life satisfaction, and relationship satisfaction) in couples coping with MS (Australia);

Theoretical principles identified:

- In "spousal" carer relationships (c) mindfulness and acceptance (m) may be important in maintaining a positive mental state (o)

Penwell-Waines et al. (2016)						✓	
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To examine perceptions of distress, health behaviours, provider health promoting communication, and impacts on stress management in carers of people with MS (US);

Theoretical principles identified:

- Carers of people with MS who are suffering high levels of stress (c) may be less likely to engage in health promoting behaviours (m) and therefore more likely to be at risk of ill health (o)

(m= mechanism, c = context; o = outcome)

Reference list:

- Buchanan, R.J., Huang, C. and Crudden, A. (2012) Use of the Internet by Informal Caregivers Assisting People with Multiple Sclerosis *Journal of Technology in Human Services*, 30(2): 72-93.
- Buckner, L. and Yeandle, S. (2015) Valuing Carers 2015: the rising value of carers' support. Carers UK. <http://www.carersuk.org/for-professionals/policy/policy-library/valuing-carers-2015>
- Coles, A.R., Pakenham, K.I. and Leech, C. (2007) Evaluation of an intensive psychosocial intervention for children of parents with multiple sclerosis. *Rehabilitation Psychology*, 52(2): 133-142.
- Finlayson, M., Preissner, K. and Garcia, J. (2009) Pilot Study of an Educational Programme for Caregivers of People Ageing with Multiple Sclerosis. *The British Journal of Occupational Therapy*, 72(1): 11-19.
- Hudson, P.L., Lobb, E.A., Thomas, K., Zordan, R.D., Trauer, T., Quinn, K., Williams, A. and Summers, M. (2012) Psycho-educational group intervention for family caregivers of hospitalized palliative care patients: Pilot study. *Journal of Palliative Medicine*, 15(3): 277-281.
- McCabe, M.P. and O'Connor, E.J. (2012) Why are some people with neurological illness more resilient than others. *Psychology, Health & Medicine*. 17(1): 17-34.
44(1): 87-90.
- Pakenham, K.I. and Samios, C. (2013) Couples coping with multiple sclerosis: a dyadic perspective on the roles of mindfulness and acceptance. *Journal of Behavioral Medicine*, 36(4): 389-400.
- Penwell-Waines, L., Goodworth, M.R., Casillas, R.S., Rahn, R. and Stepleman, L. (2016) Perceptions of caregiver distress, health behaviors, and provider health-promoting communication and their relationship to stress management in MS caregivers. *Health Communication*, 31(4): 478-484.

Additional File 4. Draft Working Theories and Chains of Inference

Draft theories	Chains of inference
Education of family or carers of people with MS (m) delivered through individually tailored modes (c) may result in improved ability to care and reduced distress (o)	M: Education, knowledge and understanding, caring role C: Carer characteristics, disease progression, location, mode, intensity, educator skills O: Confidence, knowledge, preparedness, emotional state
Developing resilience in family or carers of people with MS (m) through face-to-face interaction and opportunities for discussion (c) may result in a greater capacity to care (o)	M: Coping strategies, problem solving skills C: Carer characteristics, existing emotional state O: Creativity, resourcefulness, capacity
Access to practical help and support for family or carers of people with MS (m) through coordinated services (c) may result in improved management strategies and overall experience of caring (o)	M: Access, coordination, availability, comprehensiveness, technology, communication skills C: Coordination, location, availability, carer communication skills, technology O: Access to tailored services and development management strategies, capacity
Awareness and acceptance in family or carers of people with MS (m) through strength based approaches (c) may result in improved family relationships (o)	M: Relationship management, acceptance, reframing, family relationships, family communication, mindfulness C: Person centred, relationship to people with MS, emotional/cognitive status of people with MS and carer, credibility and training O: Mental attitude/family relationships, acceptance, communication, emotional state
Sharing experiences and emotions of family or carers of people with MS (m) in safe environments (c) may result in increased acceptance (o)	M: Discussion, sharing, emotional expression, validation, communication C: Interactive, tailored mode, person centred, safe environment O: Acceptance, validation, understanding, capacity, 'not alone'
Holistic self-management by family or carers of people with MS (m) with an appropriate range of support (c) may result in health promoting behaviours (o)	M: Respite care, step out of caring role, social/recreational activities, self-preservation, communication with HCPs C: Carer characteristics, disease progression, location, timing, stress levels O: Health promoting behavior, mental attitude, resilience
Opportunities for interaction for family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o)	M: Social contact, physical/virtual interaction, discussion groups, self-help groups C: Carer characteristics, technology, degree of interaction, synchronous/asynchronous, location O: Social contact, emotional state, recognition 'not alone', capacity

Draft theory	1	2	3	4	5	6	7
PAPER	Education of family or carers of people with MS (m) delivered through individually tailored modes (c) may result in improved ability to care and reduced distress (o)	Developing resilience in family or carers of people with MS (m) through face-to-face interaction and opportunities for discussion (c) may result in a greater capacity to care (o)	Access to practical help and support for family or carers of people with MS (m) through coordinated services (c) may result in improved management strategies and overall experience of caring (o)	Awareness and acceptance in family or carers of people with MS (m) through strength based approaches (c) may result in improved family relationships (o)	Sharing experiences and emotions of family or carers of people with MS (m) in safe environments (c) may result in increased acceptance (o)	Holistic self-management by family or carers of people with MS (m) with an appropriate range of support (c) may result in health promoting behaviours (o)	Opportunities for interaction for family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o)
Masoodi et al 2012	✓	✓					
Effects of family centered empowerment model on perceived satisfaction and self-concept of multiple sclerosis patients care givers (Masoodi R. et al. 2012)							
McConigley et al 2014					✓		
Staying just one step ahead: providing care for patients with motor neurone disease (McConigley et al. 2014)							
Nordin & Rorsman 2012				✓			
Cognitive Behavioural Therapy in Multiple Sclerosis: A Randomized Controlled Pilot Study of Acceptance and Commitment Therapy (Nordin and Rorsman 2012)							
Perry 1995				✓			✓
A study of women caregiving to husbands who have Alzheimer's disease: family know-how as a process of interpretive caring (Doctoral dissertation) (Perry 1995)							
Ray & Street 2007	✓						
Non-finite loss and emotional labour: family caregivers' experiences of living with motor neurone disease (Ray R.A. and Street 2007)							
Rollero 2016			✓		✓		
The experience of men caring for a partner with multiple sclerosis (Rollero 2016)							
Takamoto & Hori 2016	✓						
Relationship between stay-at-home patients understanding of information regarding their Parkinson's disease and the burden of spouse caregivers (conference abstract) (Takamoto H. and Hori 2016)							
Tams et al 2016		✓					
Helping families thrive in the face of uncertainty: Strengths based approaches to working with families affected by progressive neurological illness (Tams R. et al. 2016)							
Tams et al 2015		✓					
A resilience-building day for children who have a parent with MS (poster)							
Weisser et al 2015	✓			✓		✓	
Experiences of burden, needs, rewards and resilience in family caregivers of people living with Motor Neurone Disease / Amyotrophic Lateral Sclerosis: A secondary thematic analysis of qualitative interviews (Weisser F.B. et al. 2015)							

Key: "Author" = published academic literature; "**Author**" = conferences /dissertations / grey

Reference list:

- Banbury, A., Parkinson, L., Nancarrow, S., Dart, J., Gray, L. and Buckley, J. (2014) Multi-site videoconferencing for home-based education of older people with chronic conditions: the Telehealth Literacy Project. *Journal of Telemedicine and Telecare*, 20(7): 353-359.
- Boland, P., Levack, W.M., Hudson, S. and Bell, E.M. (2012) Coping with multiple sclerosis as a couple: 'peaks and troughs' - an interpretative phenomenological exploration. *Disability and Rehabilitation*, 34(16): 1367-1375.
- Busch, A.K., Spirig, R. and Schnepf, W. (2014) Coping with multiple sclerosis in partnerships. A systematic review of the literature. *Nervenarzt*, 85(6): 727-37.
- Carletto, S., Borghi, M., Francone, D., Scavelli, F., Bertino, G., Cavallo, M., Malucchi, S., Bertolotto, A., Oliva, F. and Ostacoli, L. (2016) The efficacy of a Mindfulness Based Intervention for depressive symptoms in patients with Multiple Sclerosis and their caregivers: study protocol for a randomized controlled clinical trial. *BMC Neurology*. 01/13(16): 1-8.
- Eisler, I., Ellisoin, M., Flinter, F., Grey, J., Hutchison, S., Jackson, C., Longworth, L., MacLeod, R., McAllister, M., Metcalfe, A., Murrells, T., Patch, C., Pritchard, S., Robert, G., Rowland, E and Ulph. F. (2016) Developing an intervention to facilitate family communication about inherited genetic conditions, and training genetic counsellors in its delivery. *European Journal of Human Genetics*, 24(6): 794-802.
- Feicke, J., Sporhase, U., Kohler, J., Busch, C. and Wirtz, M. (2014) A multicentre, prospective, quasi-experimental evaluation study of a patient education program to foster multiple sclerosis self-management competencies. *Patient Education & Counselling*, 97(3): 361-369.
- Francabandera, F.L. (1992) *Self-directed learning by family members of individuals with multiple sclerosis*. Phd thesis. Columbia University Teachers College.
- Kristjanson, L.J., Aoun, S.M. and Yates, P. (2006) Are Supportive Services Meeting the Needs of Australians With Neurodegenerative Conditions and Their Families? *Journal of Palliative Care*, 22(3): 151-157.
- Lancer K.M. (2007) Immune function and psychological distress in familial dementia caregivers: A controlled trial of a cognitive-behavioral intervention. *Dissertation Abstracts International: Science and Engineering*, 68: 1931.
- Liedstrom, E., Isaksson, A. and Ahlstrom, G. (2010) Quality of life in spite of an unpredictable future: The next of kin of patients with multiple sclerosis. *Journal of Neuroscience Nursing*, 42(6): 331-341.
- Masoodi, R., Soleimani, A.M, Alhani, F., Rabiei, L., Bahrami, N. and Esmaeili, A.S. (2012) Effects of family-centered empowerment model on perceived satisfaction and self-concept of multiple sclerosis patients care givers. *Koomesh*, 14(2): 240-8.
- McConigley, R., Kristjanson, L.J., Aoun, S.M., Oldham, L., Curow, D.C., O'Connor, M. and Holloway. K. (2014) Staying just one step ahead: providing care for patients with motor neurone disease. *BMJ Supportive & Palliative Care*, 4(1): 38-42.
- Nordin, L. and Rorsman, I. (2012) Cognitive behavioural therapy in multiple sclerosis: a randomized controlled pilot study of acceptance and commitment therapy. *Journal of Rehabilitation Medicine*, 44(1): 87-90.

Perry, J. (1995) *A study of women caregiving to husbands who have Alzheimer's disease: family know-how as a process of interpretive caring*. Phd thesis, University of Washington.

Ray, R.A. and Street, A.F. (2007) Non-finite loss and emotional labour: Family caregivers' experiences of living with motor neurone disease. *Journal of Clinical Nursing*, 16(3): 35-43.

Rollero, C. (2016) The Experience of Men Caring for a Partner With Multiple Sclerosis. *Journal of Nursing Scholarship*, 48(5): 482-489.

Takamoto, H. and Hori M. (2016) Relationship between stay-at-home patients' understanding of information regarding their Parkinson's disease and the burden of spouse caregivers. *Parkinsonism and Related Disorders*, 22:E65.

Tams, R., Prangnell, S.J. and Daisley, A. (2016) Helping families thrive in the face of uncertainty: Strengths based approaches to working with families affected by progressive neurological illness. *Neurorehabilitation*, 38(3): 257-70.

Tams, R., Daisley, A., Prangnell, S.J., Wilson, H., Russell, H., Hewitt, P. and Reeves, N. (2015) A resilience-building day for children who have a parent with MS. MS Trust 2015 Conference, UK.

<https://www.mstrust.org.uk/health-professionals/learning-and-development/highlights-ms-trust-2015-annual-conference>

Weisser, F.B., Bristowe, K. and Jackson, D. (2015) Experiences of burden, needs, rewards and resilience in family caregivers of people living with Motor Neurone Disease/Amyotrophic Lateral Sclerosis: A secondary thematic analysis of qualitative interviews. *Palliative Medicine*. 29(8): 737-45.

Additional File 7. Results of Phase 3 Literature Search

Draft theory	1	2	3	4	5	6	7
PAPER	Education of family or carers of people with MS (m) delivered through individually tailored modes (c) may result in improved ability to care and reduced distress (o)	Developing resilience in family or carers of people with MS (m) through face-to-face interaction and opportunities for discussion (c) may result in a greater capacity to care (o)	Access to practical help and support for family or carers of people with MS (m) through coordinated services (c) may result in improved management strategies and overall experience of caring (o)	Awareness and acceptance in family or carers of people with MS (m) through strength based approaches (c) may result in improved family relationships (o)	Sharing experiences and emotions of family or carers of people with MS (m) in safe environments (c) may result in increased acceptance (o)	Holistic self-management by family or carers of people with MS (m) with an appropriate range of support (c) may result in health promoting behaviours (o)	Opportunities for interaction for family or carers of people with MS (m) through a variety of modes (c) may reduce the sense of social isolation (o)
Bjorgvinsdottir & Halldorsdottir 2014			✓			✓	✓
Silent, invisible and unacknowledged: experiences of young caregivers of single parents diagnosed with multiple sclerosis (Bjorgvinsdottir and Halldorsdottir 2014). Aim: To study the personal experience of being a young caregiver of a chronically ill parent diagnosed with MS. Conducted in Iceland.							
Horner, R.		✓			✓		
Interventions for children coping with parental multiple sclerosis: A systematic review (Horner 2013). Aim: Systematic Review exploring the effects of caring for a parent with MS on children and to identify positive coping strategies.							
Lackey and Gates 2001			✓				
Adults' recollections of their experiences as young caregivers of family members with chronic physical illnesses (Lackey and Gates 2001). Aim: To examine positive and negative experiences of caregiving as a child, from the perspective of the adult. Conducted in the US.							
McCabe & O'Connor 2012		✓		✓			
Why are some people more resilient than others? (McCabe and O'Connor 2012). Aim: To evaluate coping strategies of people living with a chronic progressive neurological illness and their carers. Conducted in Australia.							
Nagl-Cupal et al. 2015			✓		✓		
Experiences of being a former young carer: effects in the transition into adulthood and in the present life situation (Nagl-Cupal et al. 2015). Aim: To describe the experiences of adults who had undertaken caring responsibilities during childhood. Conducted in Austria							

Key: "Author" = published academic literature; "Author" = conferences /dissertations / grey

References

- Bjorgvinsdottir, K. and Halldorsdottir, S. (2014) Silent, invisible and unacknowledged: experiences of young caregivers of single parents diagnosed with multiple sclerosis. *Scandinavian Journal of Caring Sciences*, 28(1): 38-48.
- Horner, R. (2013) Interventions for children coping with parental multiple sclerosis: A systematic review. *Journal of the American Association of Nurse Practitioners*, 25(6): 309-313.

Lackey, N.R. and Gates, M.F. (2001) Adults' recollections of their experiences as young caregivers of family members with chronic physical illnesses. *Journal of Advanced Nursing*, 34(3): 320-328.

McCabe, M.P. and O'Connor, E.J. (2012) Why are some people with neurological illness more resilient than others. *Psychology, Health & Medicine*. 17(1): 17-34.

Nagl-Cupal, M., Metzing, S. and Mayer, H. (2015) Experiences of being a former young carer: effects in the transition into adulthood and in the present life situation. *Revista Eletronica De Enfermagem*. vol. 17, no. 4.

Additional File 8 Summary of evaluation of each theory through qualitative analysis		
Theories	Supporting evidence	Clarifying CMO Interactions
<u>Amended</u> Theory 1: Information and advice to family or carers of people with MS (m) that is <u>individually tailored both proactive and responsive</u> (C) may mitigate the negative impacts of caring (O)	Full theory: unsupported due to lack of experience in participants Chains of inference: highly supported, with some clarification leading to amendment of the theory	<p>Being acknowledged and included in healthcare interactions and receiving clear explanations from people with expertise (M), for people who like to understand and prepare, at the point of diagnosis and later (C), may reduce anxiety and increase feelings of coming to terms with the diagnosis and increase feelings of preparedness (O).</p> <p>Being acknowledged and included in healthcare interactions and receiving carefully selected information from other people with expertise or experience (M), for people who like to focus on the present and on positives, at the point of diagnosis and where caring roles are minimal (C), may help to minimise feelings of anxiety and depression (O).</p> <p>Being able to access information and advice on demand from an accessible point of contact (M), responsively to challenges that emerge as the condition of the person living with MS progresses, for people who like to focus on the present and on positives (C), may reduce anxiety and increases feelings of confidence and preparedness when needed (O).</p> <p>Being able to access information and advice on demand from an accessible point of contact (M), as the health and wellbeing needs of the family member or carer and the person living with MS change (C), may enable changes of scenery and respite and can increase ability to access financial and practical help and support (O).</p> <p>Being able to access information and advice on demand from varied modes of information and advice such as paper-based, online, events, and face-to-face (M), at all points in the progression from minimal to substantial caring roles, for people who have more confidence or capability in seeking and evaluating information proactively (C), may reduce anxiety and increases feelings of confidence and preparedness when needed (O).</p> <p>Proactive provision of information or advice by people with expertise (M), at all points in the progression from minimal to substantial caring roles, for people who have less confidence or capability in seeking and evaluating information proactively (C), may help to reduce feelings of anxiety, stress, and desperation (O).</p> <p>Proactive provision of information or advice by people with expertise (M), for people who are too tired or busy with substantial caring roles to be proactive in seeking information (including people who have caring roles for others in their lives, e.g. children) (C), may contribute to the development or exacerbation of health conditions for the family member or carer, including physical and psychological (O).</p> <p>Training in skills and capabilities such as moving and handling, strategies to make caring easier, and time management (M), for people who perceive themselves to be carers, who are able to leave the person living with MS for a period of time, and who can be in the right place at the right time despite their carer roles (C), may increase feelings of being prepared for changing caring roles (O).</p> <p>Learning tips and strategies that relate to caring roles from other people with expertise or experience in support group contexts (M), for people who perceive themselves to be carers, wish to be with other people, are able to leave the person living with MS for a period of time, and who can be in the right place at the right time (C), may increase feelings of being prepared for changing caring roles (O).</p>

<p>Theory 2: Training in problem solving and coping strategies (m) taking into account characteristics and emotional state of family or carers of people with MS (c) may result in greater confidence and resilience as a carer (o).</p>	<p>Full theory: unsupported due to minimal experience in participants Chains of inference: highly supported, with clarification of CMO interactions</p>	<p>Training to enable an approach to challenges that involves planning, strategy, and creative problem-solving (m) for people with fewer capabilities in problem-solving and planning, with different financial foundations, who need to respond to challenges emerging as the condition of the person living with MS progresses and their own health needs change, at all points from minimal to substantial caring roles (c) may be more resilient and prepared for changing aspects of their caring role; may have greater capacity, partially due to spending more time on their own needs; may have greater physical and emotional wellbeing, and may have greater variety of experiences (o).</p> <p>Assertiveness training to enable development of tenacity, confidence in seeking information, advice, practical and financial help and support (m) for people with fewer capabilities in clearly and confidently expressing their needs and tenaciously pursuing desired outcomes, who need to respond to challenges emerging as the condition of the person living with MS progresses and their own health needs change, at all points from minimal to substantial caring roles (c) may be more resilient and prepared for changing caring roles; may have greater capacity, partially from spending more time on their own needs; may have greater physical and emotional wellbeing, and greater variety of experiences (o).</p> <p>Thinking through challenges and solutions in discussion with others (e.g. person with MS, friends, peer support) to generate and test ideas (m) for people who need to respond to emerging challenges as the condition of the person living with MS progresses and personal health needs change, at all points from minimal to substantial caring roles, who wish to be with other people, can leave the person living with MS for a time, and who can be in the right place at the right time (c) may be more resilient and prepared for changing caring roles; have greater capacity, partially due to spending more time on personal needs; have greater physical and emotional wellbeing, and more experiences (o).</p> <p>Cross-reference to all MCO interactions in Theory 1.</p>
<p>Theory 3: Practical help and support for family or carers of people with MS that is coordinated (m) and tailored to the carer's situation (c) may result in an improved experience of caring (o).</p>	<p>Full theory: unsupported due to minimal experience of coordinated or tailored practical help or support in participants Chains of inference: highly supported, with clarification of CMO inter-actions</p>	<p>Technological support such as alarm buttons/phone support (m) for people who can leave the person with MS as long as emergency support is accessible, who may be in employment or have other caring responsibilities (c) may provide the family member or carer with confidence to leave the person with MS knowing help can be accessed (o).</p> <p>Supportive equipment and home conversions which are appropriate to the home and to caring needs (m) at all points from minimal to substantial caring roles, but particularly where physical and emotional demands are increasing, and health and wellbeing needs changing (c) may enable the family member or carer to continue caring for the person with MS within a home environment, if they choose to while supporting their emotional and physical wellbeing (o).</p> <p>Access to appropriate practical help and support which adjusts flexibly to the needs of the person with MS and their carer (m) at all points from minimal to substantial caring roles, particularly for people experiencing increasing physical and emotional demands of caring and changing health and wellbeing needs (c) may enable continued care for the person with MS within a home environment, if preferred, while supporting their emotional and physical wellbeing (o).</p> <p>The availability of practical help to aid the carer to run and maintain a household (m) for people who may have substantial caring roles, and as their health and wellbeing needs change (c) may enable the carer to continue to support and care for the person with MS within a home environment, if they choose to by supporting their emotional and physical wellbeing (o).</p> <p>The availability of practical help to facilitate seeking financial help or advocacy (m) for people who may feel overwhelmed and fatigued due to their caring roles and who do not have the financial foundations to support the practical help they require (c) may enable the carer to continue to support and care for the person with MS within a home environment if they choose to while supporting personal emotional and physical wellbeing (o).</p>

<p>Theory 4: Strengths based training for family or carers of people with MS (m) particularly where there is a spousal/partner relationship (c) may result in improved family relationships and acceptance of their situation (o).</p>	<p>Full theory: minimal support due to few participants having experienced strengths-based training Chains of inference: highly supported, with clarification of CMO inter-actions</p>	<p>Support to develop and maintain a positive attitude (m) for people experiencing increasing physical and emotional demands of caring and time spent in the caring role (c) may help to strengthen or maintain the quality of relationship with the person living with MS, enabling that relationship to contribute to greater resilience and ability to respond to challenges (o).</p> <p>Support to reframe the situation (m) for people experiencing increasing physical and emotional demands of caring and time spent in the caring role (c) may facilitate acceptance of the situation and reduction of negative emotions (o).</p> <p>Open communication within the relationship (m) at the time of diagnosis and as new challenges emerge (c) may help to strengthen or maintain the quality of relationship with the person living with MS, enabling that relationship to contribute to greater resilience and ability to respond to challenges (o).</p> <p>Prioritising a parental role where possible (m) at all points in the progression from minimal to substantial caring roles, for people who have dependent children (c) may help maintain feelings of normality and positivity within the family (o).</p>
<p>Theory 5: Individual sharing of experiences and emotions by family or carers of people with MS with a third party (m) in a safe environment (c) may result in increased acceptance of their situation (o).</p>	<p>Full theory: some support due to a few participants having experienced counselling for reasons unrelated to caring Chains of inference: highly supported, with clarification of CMO inter-actions</p>	<p>Talking about feelings and experiences with a person who has expertise in counselling within a safe environment (m) for people who are able to leave the person living with MS for a period of time, and who are able to access interactions despite their carer roles, when existing coping strategies and existing support networks are not providing adequate support at any point in time, possibly triggered by changes in circumstances (c) may facilitate externalisation of emotions and subsequent validation of these emotions, leading to increased acceptance.</p> <p>Note: This theory was found to have a lot in common with Theory 7, which focuses on interactions between family and carers of people living with MS. Analysis of Theory 5 prioritised expertise of the third party in the cultivation of a safe environment for families and carers of people living with MS to share their emotions, independently from the person living with MS. This led to a focus on opportunities for counselling in particular.</p>
<p>Theory 6: Opportunities to engage in health promoting activities for family or carers of people with MS (m) facilitated by an appropriate range of support (c) may result in improved physical and mental health and well-being (o).</p>	<p>Full theory: minimal support due to few participants having experienced facilitation to engage in health-related opportunities Chains of inference: highly supported, with clarification of CMO inter-actions</p>	<p>Strategies enabling a person to be away from the person living with MS, such as emergency alarms and appropriate response (m) for people with less substantial caring roles, who are confident in the level of care, and who are able to prioritise their own needs for a period of time (c) may enable the family member or carer to engage in their own chosen health-promoting activities, facilitating preservation of sense of self and ability to care (o).</p> <p>Appropriate levels of support in the home for short periods of time, from a person providing company and/or care (m) for people at all points from minimal to substantial caring roles, who are confident in the level of care, and who are able to prioritise their own needs for a period of time (c) may enable the family member or carer to experience a period of mental and physical respite and increased feelings of wellbeing (o).</p> <p>Access to appropriate levels of support and care in residential respite settings for longer periods of time (m) for people with more substantial caring roles, who are confident in the level of care, and who are able to prioritise their own needs for a period of time (c) may enable the family member or carer to experience a period of mental and physical respite and increased feelings of wellbeing, and to engage in their own chosen health-promoting activities, facilitating preservation of sense of self and ability to care (o).</p> <p>Parallel or joint social and recreational activities with the person living with MS, facilitated by organisations and /or companies with appropriate facilities (m) for people with more substantial caring roles, who are confident in the level of</p>

		<p>care, who have access to this support financially and geographically, and who are able to prioritise their own needs for a period of time (c) may support healthy relationships with the person living with MS and enable the family member or carer to experience a period of mental and physical respite and increased feelings of wellbeing (o).</p> <p>Permission and encouragement to engage in health-promoting activities, during healthcare interactions where there is trust and rapport (m) for people at all points in the progression from minimal to substantial caring roles, who are less aware of their own needs, and who find it difficult to prioritise their own needs for a period of time (c) may develop problem-solving and planning strategies that can be used to find ways of engaging in health-promoting activities, with positive impacts on wellbeing and helping to preserve ability to care for longer (o).</p>
<p>Theory 7: Opportunities for interaction between <u>for</u> family or carers of people with MS (m) through a variety of modes (c) may <u>provide emotional support</u> and reduce the sense of social isolation (o).</p>	<p>Full theory: substantial support, but analysis revealed similar benefits from wider social networks for some, leading to amendment of the theory Chains of inference: highly supported, with clarification of CMO inter-actions</p>	<p>Access to a range of carer group activities, that are effectively facilitated/ organised and well attended by like-minded people (m) for people who do not have an existing supportive social/family network and have a preference for being able to interact with others and share their experiences/ 'offload' (c) may reduce the sense of social isolation and the feeling of being alone (o).</p> <p>Opportunities to engage with people in similar situations and with shared interests (m) for people who are at a similar stage in the MS journey or are able to cope with the uncertainty of the journey (c) may provide emotional support and enable carers to cope better with their situation (o).</p> <p>Reduced barriers to participation in terms of access, location, timing and provision of alternative care arrangements (m) for people who have practical issues with attending carer groups (c) may enable carers to access a source of emotional respite and enhance their ability to cope with their situation (o).</p> <p>Existing access to a supportive social and/or family network (m) for people with a preference for interaction with others (c) may enable carers to retain a sense of normality for periods of time (o).</p> <p>Access to well facilitated and supported virtual interactions (m) for people who prefer their own company and are comfortable using technology (c) may reduce the sense of social isolation and the feeling of being alone (o).</p>

Additional File 9a. Theory 1 – Information and Advice: MCO Interactions

Additional File 9f: Theory 6 – Looking After Self: MCO Interactions

Additional File 10. Recommendations for Policy, Practice, Influencing Activities and Research

Recommendation [addressing emotional (E), information (I) or wider (W) needs]	Theories addressed	Type of activity: Policy / practice / influencing activity / research
Person-centred service delivery: Acknowledgement of family member or carer in healthcare interactions, sensitive explanation and information provision, using face-to-face and through telehealth strategies; potential for home visits where necessary [I, W]	Theory 1: Proactive and responsive information and advice Theory 4: Acceptance and attitude	Influencing activities: to advocate for more person-centred healthcare interactions
Prompts to action: Strategy (e.g. screening tool) for raising awareness of the family member or carer in healthcare interactions and facilitating proactive support; highlighting the need to prioritise personal and health needs [W, I, E]	Theory 1: Proactive and responsive information and advice Theory 4: Acceptance and attitude Theory 5: Looking after self	Research: to develop and evaluate a strategy Influencing activities: to implement a strategy
Point of contact for information or advice: Stable and accessible point of contact for the family member or carer which can signpost to national and local information, using varied media: face-to-face including events, telephone, paper-based and online [I, possibly relating to E and W]	Theory 1: Proactive and responsive information and advice Theory 2: Problem-solving and coping Theory 3: Practical help and support	Policy: to consider the role of local MS Society Branches; to consider developing further avenues for information provision Practice: implementation if appropriate
Personalised coaching: Source of personalised advice and support for the family member or carer in seeking appropriate information, practical, financial and emotional and social support, and in looking after the self, using varied media: face-to-face, telephone, website, app [I, E, W]	Theory 1: Proactive and responsive information and advice Theory 2: Problem-solving and coping Theory 3: Practical help and support Theory 4: Acceptance and attitude Theory 5: Emotional support Theory 6: Looking after self Theory 7: Social contact	Policy: to consider the role of personalised coaching Research: to explore and evaluate use of personalised coaching Practice: as above

<p>Training in capabilities to enhance caring: For the family member or carer in emotional resilience, information seeking and assertiveness, time management, moving and handling, with consideration of strategies to overcome barriers: considering the role of local branches of the MS Society and other voluntary organisations; parallel activities in the same location for the person with MLS; using varied media: face-to-face, paper-based and online [I, E, W]</p>	<p>Theory 1: Proactive and responsive information and advice Theory 2: Problem-solving and coping Theory 3: Practical help and support Theory 4: Acceptance and attitude Theory 5: Emotional support Theory 6: Looking after self Theory 7: Social contact</p>	<p>Policy: considering roles in provision of training and roles of local branches Influencing activities: potential for collaboration with and signposting to other voluntary organisations Research: evaluation of developed / existing training interventions Practice: potential to integrate in practice</p>
<p>Respite options for all: Increased provision of residential care for people who have greater nursing needs with signposting to available facilities and their capabilities for family members or carers and consideration of financial support options; improvement in the capabilities of staff in respite facilities [I, E, W]</p>	<p>Theory 3: Practical help and support Theory 6: Looking after self Theory 7: Social contact</p>	<p>Influencing activities: to increase provision of respite care for all and increase training and capabilities for staff Policy: to evaluate current support for those who require more respite Practice: to develop signposting to capabilities of facilities in different locations Research: to consider development and evaluation of a method of increasing capabilities in residential care staff</p>

Volunteer support: Offering practical help and support to the family member or carer, e.g. with shopping, ironing, cleaning or gardening and by offering companionship to the person with MS [E, W]	Theory 2: Problem-solving and coping Theory 3: Practical help and support Theory 5: Emotional support Theory 6: Looking after self Theory 7: Social contact	Policy: to evaluate current volunteering roles in relation to support for the family member or carer Practice: implementing volunteer support for family members or carers
Well facilitated peer support: Peer support groups for family members or carers, providing opportunities for learning, training and social activities with consideration of barriers to face-to-face interactions and of the role of local branches of the MS Society, with opportunities using different media: face-to-face and online [I, E, W]	Theory 1: Proactive and responsive information and advice Theory 2: Problem-solving and coping Theory 5: Emotional support	Policy: considering supporting the roles of local branches Influencing activities: potential for collaboration with and signposting to other voluntary organisations Practice: reduce barriers to participation; improve facilitation of peer support strategies
Counselling: Financial support for, signposting to, and/or provision of access to counselling with consideration of barriers to face-to-face interactions and using varied media: face-to-face and/or online opportunities [I, E]	Theory 4: Acceptance and attitude Theory 5: Emotional support	Policy: to evaluate current policy and provision relating to counselling Practice: implementation of any decisions Research: exploration of alternative modes of offering counselling e.g. via skype

Multicomponent online support intervention: To prompt action for the family member or carer; provide a point of contact for information and advice, personalised coaching, relevant training, access to volunteer support, facilitated peer support, and access to counselling; this should be based online, but with different 'access routes' than enable introduction and support to engage with the online medium [I, E, W]	Theory 1: Proactive and responsive information and advice Theory 2: Problem-solving and coping Theory 3: Practical help and support Theory 4: Acceptance and attitude Theory 5: Emotional support Theory 6: Looking after self Theory 7: Social contact	Research: development and evaluation of an online resource Practice: implementation if found to be helpful
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