

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

Authors: Cathy Bulley^{1*}, Gillian Baer¹, Dawn Mahal¹, Christina H Buckton¹, Stewart Donald², Keith Lugton²; David Gillespie³, and Brendan McCormack¹

***Correspondence**

Addresses:

1 School of Health Sciences, Queen Margaret University, Musselburgh, EH21 6UU

cbulley@qmu.ac.uk

gbaer@qmu.ac.uk

bmccormack@qmu.ac.uk

dmahal@qmu.ac.uk

cbuckton@qmu.ac.uk

2 Astley Ainslie Hospital, 133 Grange Loan, Edinburgh, EH9 2HL

Stewart.Donald@nhslothian.scot.nhs.uk

Keith.Lugton@nhslothian.scot.nhs.uk

3 Department of Clinical Neurosciences (DCN), Western General Hospital, Crewe Road South, Edinburgh, EH4 2XU

David.Gillespie@nhslothian.scot.nhs.uk

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, 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. Caring can have negative physical, emotional and financial impacts, which sometimes limits 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 on 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 the economy 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 some challenges particularly associated with MS. This progressive autoimmune disease of the central nervous system generates varying symptoms that include fatigue, weakness, posture and mobility difficulties and 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). Over 40% of pwMS are diagnosed between ages 30 and 49 with about a ten-year reduction in life expectancy, therefore living for many years with MS is not uncommon (Mackenzie et al, 2014; Runia, 2012).

Approximately 15% of people are initially diagnosed with progressive MS; of the 85% diagnosed with relapsing-remitting MS, 35% later develop progressive MS (Kremenutzky, 2006).

Complications and co-morbidities can include results of falls, infections, and pressure ulcers (Higginson, 2006), with substantial impacts on people with MS and carers.

The substantial evidence base relating to needs of the families and carers of pwMS includes a systematic review of 24 qualitative or descriptive survey-based studies published between 1990 and 2002 (McKeown et al, 2003). In all studies participants were the primary carers. Their caring responsibilities were time consuming and included 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 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 reduced personal income. 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). New insights emerged, including greater distress when someone was caring for their spouse than a person who was not their

spouse. 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. They found worse experiences among people with lower social support and less affluence (Borreani et al, 2014). In a Grounded Theory study of 25 people, carers found it hard to recognise their own needs and did not all see themselves as carers, which formed barriers to accessing support (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 diverse characteristics, contexts and needs. Family and carers have multiple perspectives and priorities and therefore the research team prioritised a person-centred approach, which focuses on the perspectives and individual priorities of people, prioritising sharing of power between providers and people accessing services (McCormack et al, 2017). The current 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. Evaluating linear relationships between intervention and outcome can lead to rejection of potentially useful interventions which have been evaluated in unsuitable 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.

Rapid Realist Review (RRR) is an expedited form of Realist Synthesis; both aim to explore complexity. Realist Synthesis focuses on identifying the causal mechanisms within an intervention, which are the underlying actions that lead to change. It then explores the differences in context that affect how mechanisms work, for example, a context could relate to differences in participant characteristics, geographical location, or service model. Finally, the interaction between

mechanisms and contexts in producing different outcomes is explored. This enables the reviewer(s) to propose an explanation - or theory - rather than a judgement of efficacy. In-depth analysis of selected literature enables identification of 'mechanism, context, outcome (MCO) chains' which are written in the form of hypotheses (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. RRR uses systematic search strategies to identify published and grey literature, emphasising speed over comprehensiveness. The research team can then continue into a second stage of RE, where theories developed through RRR are then evaluated from a realist perspective. This involves seeking evidence that confirms, clarifies or refutes them, using methods such as qualitative interviews to explore experiences and views of family and carers. This journey enables early implementation of results, appropriate in a nine-month commissioned study. The process also generates new thinking by enabling people with lived experience and professional expertise to contribute to validity of the review and evaluation through participation in an Expert Panel. Their remit includes identification of key literature and discussion of emerging findings. Utility of the final product is enhanced through a Reference Panel, including funder representatives (Saul et al, 2013).

In our study, RRR, followed by RE 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 explain the full research journey while demonstrating rigour, we make reference to additional files which are all located in a data repository ([10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)). These files demonstrate the audit trail of extensive literature and data analysis that support theory development and evaluation.

Methods

Rapid Realist Review

Expert Panel discussion supported a three-phase, iterative search of published and grey literature focused on interventions to support family and carers of pwMS (see Table 1 and Figure 1 for summary and illustration of phases; Additional File (AF) 1 for key words and AF2 for panel membership: [10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)). 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: [10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)). 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: [10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)).

Realist Evaluation

Working theories were evaluated through analysis of lived experiences of being a family member or carer of someone living with MS. Qualitative data were analysed from a realist perspective 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: [10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)). Face-to-face interviews were conducted in participants' homes and focus groups in a community hall (CHB, DM). Where permission was granted (for all but one person) interviews were digitally recorded for transcription. Three telephone recordings failed and detailed notes were used. The researchers were able to provide contact details for someone who the participants could talk to if the interactions raised any concerns or distress.

Thematic analysis by three researchers (CHB, DM, CB) 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. Conversations between CHB, DM and CB took place throughout data collection and analysis to ensure reflexivity.

Results

Rapid Realist Review of the literature

The first search phase resulted in seven publications (details in AF 4:

[10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)). 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 included pwMS. Telephone interviews with 530 informal carers (Buchanan et al, 2012) explored influences on carer use of internet-based information, while Samios (2013) used questionnaires with 29 couples to explore the impacts of mindfulness and acceptance on adjustment to MS. Penwell-Waines et al (2016) surveyed 67 carers about their stress and engagement in health-promoting activities. 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 ([10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)).

The second and third search phases resulted in 21 and five further articles respectively which supported the theories, summarised in AF6 and AF7 ([10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)). 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

The working theories in Table 3 were evaluated; for auditability, details are included in AF8, with MCO Diagrams in AF 9a-g ([10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)). Table 4 summarises participant characteristics.

→ 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. 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.”*

Information and advice were frequently used in problem solving and coping (Theory 2). Ralf described his thinking: *“how can we get around this problem/ what’s the way of solving it?”* No participant described receiving training in problem solving or coping, but where people described existing capabilities, there were clear benefits to wellbeing and ability to continue caring. Simon explained *“it’s just listening to how other people cope with that, and building up a toolkit....”* This

linked with Theory 6, as people used problem solving skills to make time for their own needs, important for resilience.

There were few experiences of coordinated help and support (Theory 3), as explained by Anne: *“He [primary carer] spends an inordinate amount of time which he doesn’t have... chasing down, rushing around, dealing with professionals who don’t seem to speak to each other.”* The need for practical help and support was clearly demonstrated and necessary for coping over time. Andrew stated *“it is physically and mentally draining. For example, I have to put on slings, take off slings, several times a day... that takes quite a bit of physical strength. I’m [over 70] years of age, and obviously that leads to a few worries about, well, how long will I be able to keep going?”* Family and social networks helped; where lacking, financial foundations were important. This linked Theory 3 with Theory 1, as people often needed information and advice about availability of financial support, followed by practical support to complete relevant documentation. There was substantial evidence that coordination of help and support would be beneficial, particularly for people who are 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, so you get emotionally tired, so you just let it slide...”* However, help provided must be trustworthy and flexible to people’s needs.

People rarely experienced strength-based training (Theory 4) but there was evidence that people with strengths such as a positive attitude achieved greater acceptance of their situations and more positive relationships with the person with MS. Ted stated *“MS is not going to beat us, we’re going to live a life as normal as MS allows.”* There was minimal evidence of support to 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 them 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 in the discussion.

Discussion

In synthesising the results of the RRR and RE, all MCO interactions were brought together into one model (Figure 2). Common contextual issues were treated as foundations in the model, influencing the potential for benefit from any mechanism, or intervention.

→ Insert Figure 2 here

The model suggests that there can be a predominantly negative or positive picture of contexts and outcomes. As these interact, people are more likely to identify with one side of the model. For example, someone who self-identifies as a carer is more likely to seek information, advice and support; if confident in this and tenacious in seeking support, they may implement more supportive strategies that keep their workload manageable. Where someone uses problem-solving skills to justify and identify time for self-care, their social relationships and wellbeing may be enhanced.

While supportive positive contexts were frequently described as existing characteristics rather than intervention outcomes, they provided evidence of positive mechanisms triggering more positive outcomes summarised in the arrow on the right. If people who identify more with unsupportive contexts are facilitated to develop more supportive contexts, their quality of life could be improved.

Some contexts are less amenable to modification (see the middle-top lines of Figure 2) and should be respected, for example, stage of disease progression and differences in personality and preference. Coping style affects what people want at each stage; too much information, either at the start of their journey, or when overwhelmed, may exacerbate negative emotions. This may undermine a coping style of focusing on the positive and not looking too far ahead. For others, mentally preparing themselves and planning for the future has a stabilising effect. Options for intervention delivery should take account of these differences. Other contexts represented in Figure 2 may be modifiable through a cluster of carefully designed mechanisms as described in the centre of the model. These were translated into recommendations listed in AF10, cross-referenced according to relevant theories, which are now discussed ([10.6084/m9.figshare.12996683](https://doi.org/10.6084/m9.figshare.12996683)).

Person-centred service 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 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

These 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.

Point of contact for information or advice

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).

Personalised coaching:

Individual coaching may particularly help 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. Coaching could be offered through different 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 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 and using varied media will provide best results.

Training in capabilities to enhance caring

Training would be valuable in relation to emotional resilience, information use, assertiveness in seeking support, time management and moving and handling. Access and preference must be 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. This was advocated in a review of models of care for socially-isolated older carers (Winterton and Warburton, 2011).

Respite options for all

The availability of respite through well-trained, accessible and flexible care support is crucial, as are respite facilities that can cater to more substantial caring needs. Currently this can be very challenging for people with substantial caring workloads due to lack of facilities with good quality nursing care. Carers frequently stay present during care support in the home, 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 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).

Volunteer support

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).

Well facilitated peer support

Participants often described social support 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.

Counselling

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.

Multicomponent online support intervention

The 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-component, such as coaching and peer support (Boots et al, 2014). Another systematic review of telehealth interventions found greater impacts on self-efficacy and mood from multifaceted interventions (Jackson et al, 2016). A more recent study evaluated a 14-

week behavioural intervention in 28 people who received educational materials and telephone coaching compared with basic education and telephone support in 38 people (Steffen and Grant, 2016). The intervention group's improvements in depression, mood and self-efficacy for obtaining respite were not sustained beyond six months, suggesting that long-term conditions require longer-term interventions. The contextual challenges and desired outcomes for carers of people with dementia have many commonalities with carers of people with MS.

This supports our conclusions that multicomponent interventions with multifaceted delivery have potential to support carers of people with MS, if accessible as new needs arise. Ideally, components would include many of the recommended strategies described above. This is an area with great promise for development and research.

Study limitations

A key limitation in this study was the small number of studies focusing on support for carers of people with MS. We repeated the initial search strategy in July 2020 and found limited new results. Tzitzika et al (2020) conducted a questionnaire study with 909 Greek spouses/partners of pwMS, finding that commitment to their relationship was the strongest predictor of continuing to provide care. Maguire and Maguire (2020) reviewed literature addressing experiences of carers of pwMS, and their key finding reinforced our own identification of varied contextual factors as strongly influencing experiences.

Participant diversity was limited and studies focused on engaging specific groups may need to address this (Bonevski et al, 2014). We noted retrospectively that some questions used in our topic guide tended to be closed in nature. These were followed up by probing and prompting questions, which may be responsible for the in-depth data collected. During discussions with the Funders, a focus on adults was decided. There is literature, however, which notes the impacts of caring on children (e.g. Coles et al, 2007) and further studies focused on young carers are needed. Members

of the Expert Panel also raised the need for further studies with people who have left the caring relationship by choice, which can be fraught emotionally.

To facilitate implementation, a stakeholder workshop was held at the UK MS Society Headquarters, including people with varied expertise, such as service provision, research, influencing, innovation, policy and IT. The researchers facilitated engagement with results and recommendations, and workshop participants then planned forward and organised work-streams and related working groups to include family and carers.

Conclusion

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. This may delay 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., Zaratini, 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): 133-42.

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.uke.de/extern/eurofamcare/documents/overview_teasure.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.

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.

Harwood, R.H., Sayer, A.A. and Hirschfeld, M. (2004) Current and future worldwide prevalence of dependency, its relationship to total population, and dependency ratios, *Bulletins of the World Health Organization*. 82(4):251-8.

Higginson, I.J. (2006) Symptom prevalence and severity in people severely affected by multiple sclerosis, *Journal of Palliative Care*. 22(3):158-16.

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): 277-81.

Househ, M., Borycki, E. and Kushniruk, A. (2014) Empowering patients through social media: The benefits and challenges, *Health Informatics Journal* 20(1): 50-8.

Jackson, D., Roberts, G., Wu, M.L., Ford, R. and Doyle, C. (2016) A systematic review of the effect of telephone, internet or combined support for carers of people living with Alzheimer's, vascular or mixed dementia in the community, *Archives of Gerontology and Geriatrics*, 66: 218-36.

Kremenutzky, M. (2006) The natural history of multiple sclerosis: a geographically based study 9: observations on the progressive phase of the disease, *Brain*, 129: 584-94.

Mackenzie, I.S., Morant, S.V., Bloomfield, G.A., MacDonald, T.M. and O'Riordan, J. (2014) Incidence and prevalence of multiple sclerosis in the UK 1990–2010: a descriptive study in the General Practice Research Database, *Journal of Neurology, Neurosurgery and Psychiatry*, 85(1): 76–84.

Maguire, R., and Maguire, P. (2020) Caregiver burden in multiple sclerosis: Recent trends and future directions, *Current Neurology and Neuroscience Reports*, 20:18.

McCabe, M.P., Roberts, C. and Firth, L. (2008) Satisfaction with services among people with progressive neurological illnesses and their carers in Australia, *Nursing & Health Sciences*, 10(3): 209-15.

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): 234-48.

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): 478-84.

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): article number 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): article number 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.

Tzitzika, M., Lampridis, E., and Kalamaras, D. (2020) Relational satisfaction of spousal/partner informal caregivers of people with multiple sclerosis, *International Journal of MS Care*, 22: 60-6.

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=M3UfSPk6laYvgH>

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/service-delivery-safety/areas/people-centred-care/framework/en/>

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Conflict of Interests

The authors declare that there is no conflict of interest.

Ethics approval and consent to participate

National Health Service ethical approval was received for face-to-face interviews (REC reference: 16/LO/1194) and the Higher Education Institution's Ethics Committee approved all other aspects of data collection (REP 0130). All participants provided informed consent to participate in the study and no identifiable information has been included.

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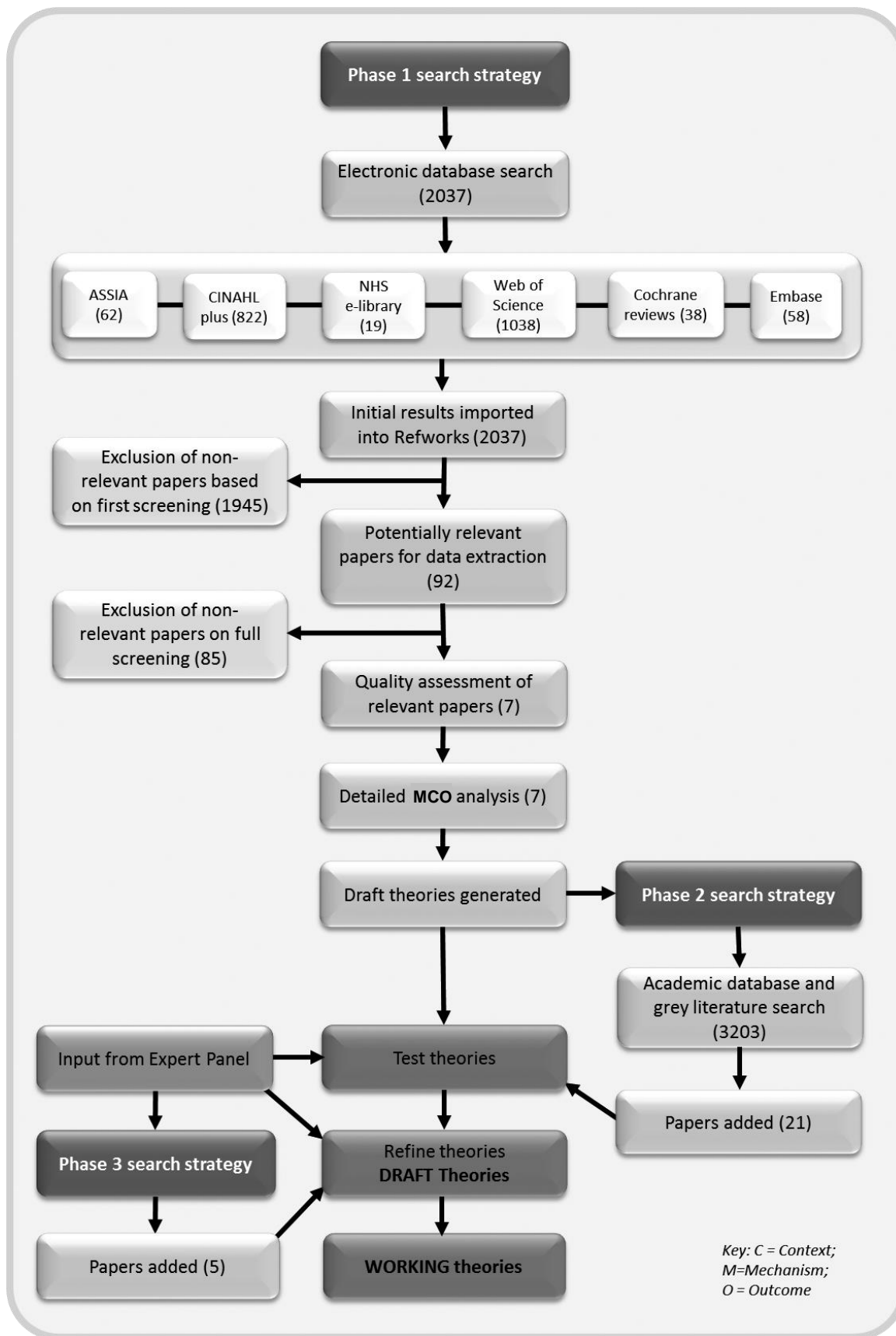


Figure 1: Rapid Realist Review Process

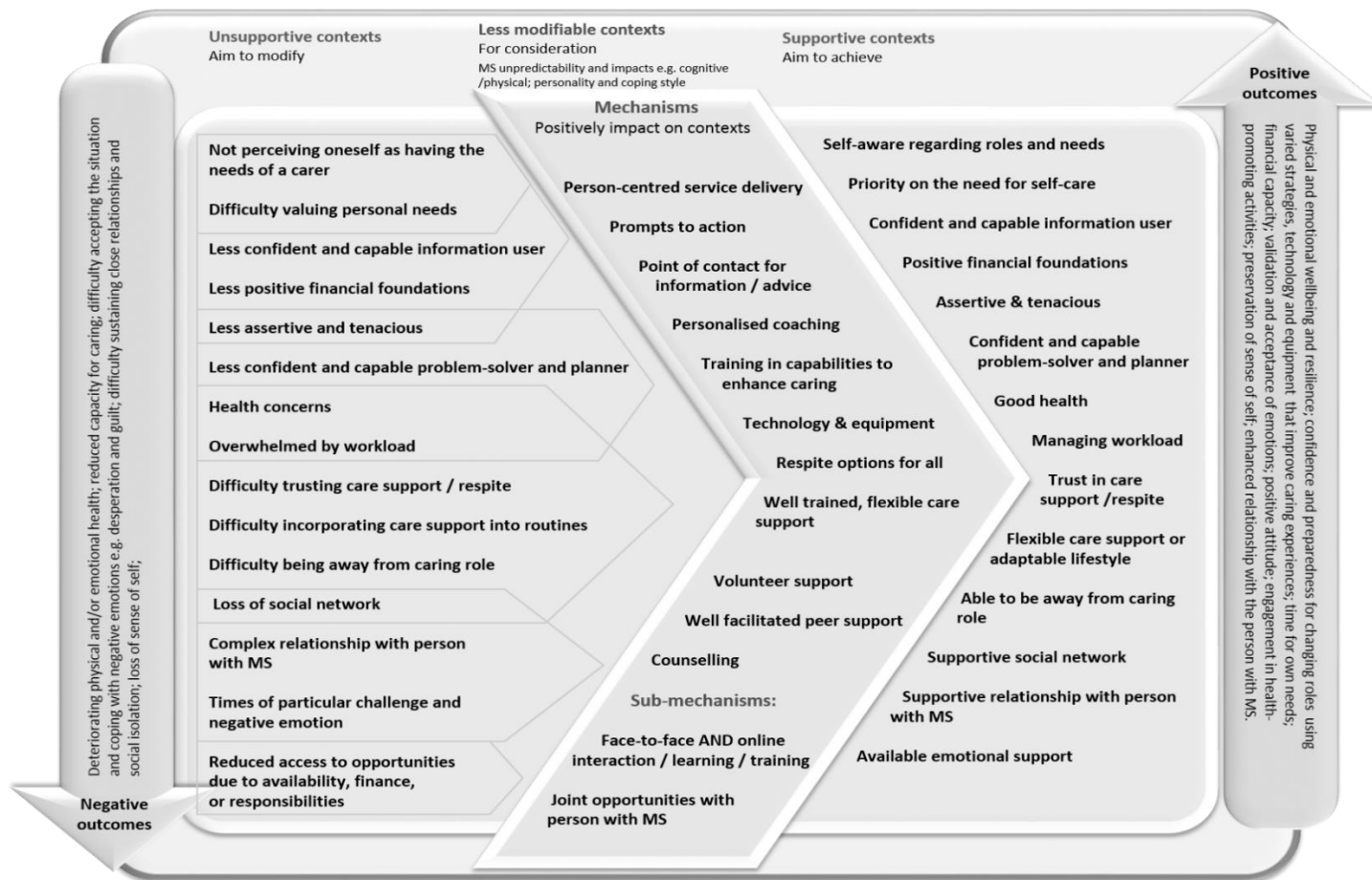


Figure 2. Model of mechanisms to modify contexts for family and carers to improve outcomes

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

Stage	Process (took place in 2016)	Outcomes (see Figure 1 for process and results)
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 (10.6084/m9.figshare.12996683).</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>	2037 hits → seven articles selected 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; OALster; 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 (10.6084/m9.figshare.12996683).</p> <p><u>Inclusion criteria:</u> as above</p> <p><u>Exclusion criteria:</u> as above</p>	3203 hits → 21 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 (10.6084/m9.figshare.12996683), 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>	Five additional papers identified 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