Caregiving in Multiple Sclerosis and Quality of Life: A Meta-synthesis of Qualitative Research

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Objective: The lack of adequate conceptualisation and operationalisation of quality of life (QoL) limits the ability to have a consistent body of evidence to improve QoL research and practice in informal caregiving for people with Multiple Sclerosis (MS). Thus, we conducted a meta-synthesis of qualitative research to improve the conceptual understanding of the experiences of MS carers and to identify factors that affect carers' QoL.

Design: Systematic searches of five electronic databases yielded 17 qualitative studies which were synthesised using the principles of meta-ethnography.

Results: The synthesis resulted in 9 inter-linking themes: Changes and losses; challenges revolving around MS; caregiving demands; burden of care; future concerns; external stressors; experiences of support; strategies used in managing the caregiving role; and motivating factors. Our findings suggest that MS carers can have both positive and negative experiences which may bring challenges and rewards to the carers.

Conclusion: We present a proposed QoL model for MS caregiving which can be used to inform the development of interventions for MS carers to improve their QoL. However, further empirical research is needed to examine the utility of this model and to explore the concept of QoL in MS carers in more detail.

Keywords: informal carers; meta-ethnography; meta-synthesis; multiple sclerosis; quality of life

Multiple Sclerosis (MS) is one of the most common chronic neurodegenerative diseases among adults, globally affecting approximately 2.3 million individuals (Multiple Sclerosis International Federation [MSIF], 2013). MS has an extremely variable progression, with some patients showing a steady rapid deterioration (i.e. primary progressive course), and most experiencing unpredictable exacerbations and remissions of symptoms (i.e. relapsing-remitting course; Mohr et al., 1999).

In the later stages of MS and at times of exacerbations, people with MS (pwMS) cannot manage daily living activities by themselves and can become functionally dependent on others. Generally, it is the immediate family members (e.g. spouse) or friends who take on the responsibility of caring for pwMS (Forbes, While, & Mathes, 2007), and are commonly referred to as 'carers' or 'caregivers'. A carer is someone who provides a significant amount of voluntary unpaid support and help to a friend, relative, or a neighbour, who suffers from an illness, disability, frailty, mental health problem, or addiction (Carers Trust, 2012; Carers UK, 2015; Department of Health [DoH], 2014). The word 'informal' is commonly used to distinguish voluntary unpaid carers from health professionals or care workers who get paid for providing care.

As a result of the progressive, unstable and unpredictable nature of MS, caring for pwMS causes high levels of carer burden and is recognised as a potential threat to caregivers' health and well-being (Corry & While, 2009; Devins, Seland, Klein, Edworthy, & Saary, 1993). Indeed, there is consistent evidence in the current literature that overall quality of life (QoL) is low among informal carers of pwMS (see Corry & While, 2009). High levels of emotional distress and carer burden, certain caregiver characteristics (e.g. gender, being a spouse) and patient characteristics (e.g. patient QoL) were found to be significant contributors to the MS carer QoL (Corry & While, 2009). However, there is conflicting evidence from quantitative studies on the relationship between clinical characteristics of pwMS and carer QoL. For instance, Aronson (1997) reported that decreased carer QoL was associated with increased severity of care-recipients' symptoms and physical disability, whereas Argyriou et al. (2011) have failed to find a significant association between clinical deficits of patients and carer QoL. Similarly, Figved, Myhr, Larsen, and Aarsland (2007) reported that physical disabilities of pwMS were not a significant predictor of carer QoL, whereas they found support for the association between neuropsychiatric symptoms of pwMS

and carer QoL. This discrepancy among the findings may be attributable to the use of different generic instruments to measure QoL (e.g. SF-12, WHOQoL), all of which have different domains that fail to consider the MS-specific factors.

There is also a lack of adequate conceptualisation and operationalisation of QoL which makes comparison across studies very difficult and limits the ability to have a consistent body of evidence to improve MS carer QoL research and practice (Bakas et al., 2012). Several researchers have argued that the concept of QoL is difficult to operationalise (Cummins, 1997; Rapley, 2003). Despite the growing utilisation of QoL as a primary outcome measure in health research, there are several concerns and criticisms on its definition and conceptualisation, as well as how it should be measured. QoL is a highly complex, multi-level and amorphous concept and there is no agreed definition of QoL or standard form of measure (Brown, Bowling, & Flynn, 2004; Cummins, 1997). Therefore, Aubeeluck, Buchanan, and Stupple (2012) suggested that researchers need first to operationalise their definition of QoL to clarify what specifically is being measured.

One way of operationalizing such a complex concept is to learn from the views and experiences of informal carers of pwMS. Indeed, several qualitative studies have investigated the views of MS informal carers and their caregiving experiences (e.g. Hughes, Locock, & Ziebland, 2013; Mutch, 2010). Such studies are valuable for gaining a more detailed and nuanced view on the lived experiences of carers which might not be possible through quantitative studies. Although the value of qualitative research in guiding further research and practice has been recognised within healthcare (e.g. Kelly et al., 2009; Smith, 2011), the findings of individual qualitative studies risk being lost and relegated as distinct "little islands of knowledge" (Glaser & Strauss, 1971, p.181) unless they are synthesised with more well-established methods (Malpass et al., 2009; Sandelowski, Docherty, & Emden, 1997).

Synthesizing existing qualitative evidence can increase the generalizability of qualitative findings (Sandelowski et al., 1997) and can also enhance the understanding of the existing evidence (Borg Xuereb, Shaw, & Lane, 2012; Carlsen, Glenton, & Pope, 2007). Additionally, qualitative synthesis could "go beyond the detailed summarizing of the traditional narrative view by achieving fresh insights, conceptual development and theory" (Campbell et al., 2011, p. 2). Therefore, the aim of this meta-synthesis is to identify factors that may affect the QoL of MS carers positively and/or negatively, and to derive a new conceptual understanding of the views and experiences of carers of pwMS related to caregiving to help inform future research and practice.

Methods

The meta-synthesis was conducted in four phases using the principles of meta-ethnography (Noblit & Hare, 1988).

Phase 1 & 2: Search Strategy and Screening

To formulate the search strategy, the CHIP (Context, How, Issues of interest, Population) tool (Shaw, 2010) was used to break down the research aim into its constituent parts (Shaw, 2011). The final set of CHIP terms used in the search strategy is shown in supplementary material Table 1. A systematic search of five electronic databases (Medline, EMBASE, CINAHL, Web of Science and PsychInfo) was conducted in October 2013 and updated in July 2014. The search terms were combined using the Boolean logic terms "and" and "or". The MeSH explode was used in databases where this was possible. To maximise identification of suitable studies, we examined the reference lists of all potentially relevant articles that passed the inclusion criteria. In addition, we contacted key researchers in this area to identify any additional relevant articles they or their colleagues had published.

Retrieved studies were initially screened using three inclusion criteria: (a) published studies using qualitative techniques for the collection and analysis of data; (b) studies that explored views or experiences of adult informal carers of pwMS in relation to the carers' health and well-being; and (c) articles that were available in English. Initial assessment was undertaken by the first author by looking at the titles and abstracts, and if unsure full-text was obtained and screened. After duplicates and irrelevant articles were removed, eligibility screening was performed, record by record, by asking two questions: (a) is the research relevant to the synthesis topic (i.e. views or experiences of adult informal carers of pwMS in relation to the carers' health and well-being) and (b) does it use qualitative methods for the collection (e.g. interviews, focus groups, etc.) and analysis (e.g. thematic analysis, interpretative phenomenological analysis, etc.) of data? If eligibility was unclear this was discussed across the wider review team. We did not exclude articles that use mixed methods because these studies contained valuable qualitative data, provided the qualitative research met the above criteria and formed a substantial element of the study.

As the main focus of this review was specifically on understanding MS carers' views and experiences in more detail, articles focusing on caregiving in other conditions were excluded. Mixed-sample studies (i.e. including pwMS as well as MS carers, or MS carers as well as those from other disease groups) were also excluded from the study unless at least 75% of the sample comprised MS carers and we could specifically identify the views and experiences of MS carers, or subsample analysis was carried out on the relevant participants. The 75% cut-off point has been justified by a previous Cochrane Systematic Review of articles on pwMS (see das Nair, Ferguson, Stark, & Lincoln, 2012).

Phase 3: Critical Appraisal

The quality of studies was appraised using the Critical Appraisal Skills Programme [CASP],

(2010) tool for qualitative research. Three broad issues concerning rigour, trustworthiness, and relevance of the studies were considered by asking 10 questions about the research design, methodology, results and value of the research. The utility of the CASP tool for appraising qualitative studies has been demonstrated in previous meta-synthesis studies (e.g. Malpass et al., 2009; Garip & Yardley, 2011). Instead of using a numerical scoring system for appraising articles with the CASP tool (e.g. Feder, Hutson, Ramsay, & Taket, 2006), we adopted Malpass et al.'s (2009) approach in their synthesis where they evaluated each article based on the Traffic Light System (Dixon-Woods et al., 2007). Quality appraisal was undertaken independently by two research team members (XX, XX *[names deleted to maintain the integrity of the review process]*) who then met with the other two team members to agree on the quality of the studies.

Phase 4: The Synthesis

We followed the principles of meta-ethnography, which is an interpretive method of synthesizing qualitative evidence and is currently one of the most well-established methods for synthesizing qualitative research (Britten et al., 2002; Campbell et al., 2011; Malpass et al., 2009). Meta-ethnography focuses on the "translation of qualitative studies into one another" (Noblit & Hare, 1988, p. 25) through the first, second and third order constructs with an objective of developing new conceptual and theoretical insights (Malpass et al., 2009). Malpass et al. (2009) define first order constructs as "interpretations of experience" (i.e. participants' views and interpretations of their experiences); second order constructs as "interpretations of interpretations of experience" (i.e. views and interpretations of authors on participants' views expressed as themes); and third order constructs as "interpretations of interpretations of interpretations of experience" (i.e. synthesists' views and interpretations of first and second order constructs expressed in themes and key concepts).

First and second order constructs were identified and recorded verbatim on the data extraction form for each article, after reading them several times. A matrix was created to facilitate the translation of studies into one another. This matrix was then used to conduct the reciprocal synthesis (i.e. a form of synthesis in meta-ethnography; Noblit & Hare, 1988) where we compared and contrasted themes across articles to develop our third order constructs (i.e. higher order themes). We then created a line of argument through these higher order constructs to explore what the set of individual studies say as a whole (Malpass et al., 2009; Noblit & Hare, 1988; Shaw, 2011). XX *[name deleted to maintain the integrity of the review process]* led the synthesis with independent input from XX, XX and XX *[names deleted to maintain the integrity of the review process]* to verify the third order constructs.

Results

Electronic database searches yielded 4425 references, excluding duplicates. We excluded 4367 articles during the initial screening process. Of the remaining 58 full-text articles, 34 examined a mixed group of participants which did not achieve the 75% cut-off point so were excluded. Fourteen articles met the inclusion criteria. Three additional studies were identified during our database search update from November 2013 until July 2014, through contacting relevant authors and by looking at the publications lists of relevant authors (see Supplemental figure for the step-by-step study selection and screening process).

The Synthesis Articles

Seventeen articles met the inclusion criteria (see supplemental material Table 2 for study characteristics). Some articles drew on the same data (i.e. Articles #6 & #7, articles #10 & #15, articles #11 & #12; see reference list for assigned article numbers for each paper included in the synthesis). The total number of participants across all studies was 1023 adult informal carers of pwMS. The age range of participants across the studies was 17-84 years.

Nine articles specifically targeted carers who provided support to a spouse with MS. The remaining papers examined not only spouses but also other family members or friends who provided voluntary unpaid care to their loved ones with MS.

Critical Appraisal

Quality of the selected studies was assessed using the CASP tool in relation to the appropriateness of the research design, recruitment, analysis and ethical considerations (See supplemental material Table 3 for the ratings of each article). Only two mixed method studies were rated as 'unsure' because of a lack of clear description of the research design or qualitative data analysis procedures. However, it is acknowledged that restrictions in journal word limits might cause problems for authors in terms of providing methodological or analytical details (Garip & Yardley, 2011; Walsh & Downe, 2005). Appraisal was not undertaken to exclude articles, but rather to test their contributions to the synthesis and to identify their potential limitations (Bennion, Shaw, & Gibson, 2012; Malpass et al., 2009). Therefore, we have not excluded any articles on the basis of critical appraisal, but the findings of articles rated as 'unsure' were given less emphasis in the synthesis process (Dixon-Woods et al., 2006).

Synthesis Findings

The second order themes in each article were compared with those from other studies to form the *developing second order constructs* (i.e. broader categories developed to organise a large number of second order constructs), using the original words of authors or a paraphrase with little reinterpretation. Supplemental material Table 4 shows the list of 35 developing second order constructs from the 17 articles and also gives a narrative translation of each developing second order construct that has meaning for all the articles from which it is drawn. These definitions are either the authors' own words or a close paraphrase, combining key concepts

across the relevant articles that summarise the developing second order construct in the most meaningful way (Malpass et al., 2009). Finally, through an iterative process of review and discussion, we conducted a reciprocal synthesis to synthesise these translations and, as a result, nine inter-linking third order constructs were developed. Supplemental material Table 5 illustrates participants' quotes that are representative of the third order themes.

Third order Themes

Experiences of Change and Loss in a Shifting Context

Almost all of the articles found that informal carers of pwMS experience several changes and losses in their lives once they took on the carer role.-Eight out of seventeen studies found that MS carers experienced a change in their role and identity as they became the carer of their loved ones and gradually took on new roles which were often complex and difficult (e.g. caregiving, breadwinner, single parenting, etc.; Articles #4, #5, #6, #9, #10, #14, #15, & #16).

It was apparent in most articles that family members and friends of pwMS adopted the identity of carer to different degrees. Some embraced the identity of carer, others felt that it was forced on them by changing circumstances and that it was conflicting with their ideal identity (e.g. Articles #15, #16). Some participants drifted into the carer role and gradually took on more responsibilities without noticing as the MS progressed (e.g. Articles #4, #5, #10, & #16), whereas others rejected the carer identity even though they acknowledged doing some caring tasks (Articles #10 & #16). Hughes et al. (2013; Article #16) suggested that this variability in acceptance might be related to other people's expectations about whether one should assume the carer role. In particular, they reported that those who embraced the caring role and identity of carer were more likely to have a more tangential relationship with the person with MS (e.g. sibling, ex-partner), whereas those who are resisting both the role and

identity were the ones who are expected to assume the caregiving role (e.g. spouses; Article #16).

The progression of MS and accumulated caring tasks caused certain interruptions in carers' employment and lifestyle (Articles #2, #4, #6, #9 & #10). Most carers either reduced their working hours or stopped working altogether to accommodate patients' MS, whereas others had to start working more to help the household finances (e.g. Article #9). Lifestyle changes occurred unavoidably because of loss of income (Articles #6 & #10), restricted time and reduced opportunity to engage in social activities (Article #10). Carers who started working felt the pressure of an additional identity imposed on them and experienced certain identity conflict, whereas others who gave up their jobs were reported as experiencing a sense of identity loss and decreased self-esteem.

MS and the caring role caused strained family relationship dynamics with the loss of companionship (Articles #6 & #11), significant disagreements and conflicts (Articles #9 & #11), the pressure of constant proximity to the care-recipient (Articles #3 & #4) and unbalanced interpersonal relationship between the carers and pwMS (Articles #1 & #3). Some carers attributed their changed relationships to the loss of intimacy between themselves and their care-recipients (Articles #3, #4, #6, #8, #10 & #17). For instance, many carers felt grief over the loss of shared activities (Article #3, #10 & #17) and loss of a partner as an equal companion (Articles #4 & #6).

Challenges Revolving Around MS and Patient Issues

A large proportion of studies revealed that carers found it difficult and stressful to deal with the care-recipients' physical (e.g. lack of mobility), cognitive (e.g. poor memory), behavioural (e.g. demanding behaviour) and emotional (e.g. mood swings) problems which were all symptoms of MS. In particular, emotional and behavioural problems were described

as the most stressful symptoms of MS (Articles #3, #4, #7, #11, #13). Care-recipients'
demanding and self-centred behaviours, low moods, suicidal thoughts and outbursts of
despair were identified as very difficult situations for carers to manage (Article #7) and, at
times, could have a strong demotivating influence for continuing to care (Article #4).

The unpredictable nature of MS was another challenge that MS carers found hard to
deal with during their caring experiences. Four studies found that carers often worried about
the uncertainty of the progression of the disease (Articles #6, #7, #13 & #14). This made
them feel out of control, provoking anxious thoughts, anger and feelings of inadequacy and
helplessness (e.g. Article #14).

Negative and non-cooperative attitudes of pwMS were also reported as challenges
faced by carers (Articles #3, #4, #5 & #14). Some patients experienced difficulties in
accepting their disease and adjusting to their disabilities. Consequently, they did not wish to
have other family members' or formal carers' help other than their own carers, which made it
harder for these carers to manage the caregiving role (Articles #4, #5 & #14). Likewise,
Mutch (2010; Article #14) reported that some pwMS refused to go into respite care which
forced their carers to provide constant care without any holiday or break. Care-recipients'
lack of appreciation and respect was another difficulty faced by carers which made it harder
for them to remain patient, and led to feelings of disappointment and frustration (Article #4,
#7 & #16).

On a more positive note, some carers found that the positive attitude of the care-
recipient had helped them cope with the demands of MS and caregiving, and was an
important aspect contributing positively to their QoL (Article #14). For instance, Mutch
(2010; Article #14) found that when the care-recipients were happy and not acting bitterly
about their illness, spousal carers could have quality time with their MS affected partners and
have fun together, providing an opportunity for them to find mutual emotional support. In

addition, care-recipients who adjusted better to their illness could also adopt a more cooperative attitude and accept formal assistance from others (e.g. going into respite care, etc.) relieving some of the burden from carers (Articles #3, #4, #5, & #14).

Caregiving Demands

Most studies emphasised the increasing daily hassles and caregiving demands in MS informal carers' lives. With the progression of MS and at the times of exacerbation, caregiving becomes more intensive and carers are forced to take on additional tasks and responsibilities which put them under a lot of pressure (Articles #1, #3, #4, #6, #10, #16 & #17). In addition to gaining responsibility for household duties and providing physical, personal, financial and emotional support, some studies showed that carers also gained the responsibility of decision making regarding all aspects of life (e.g. financial, health care, future) which could be highly stressful and could result in an increased burden for these carers (e.g. Articles #6 & #17).

Lack of time was another problem faced by MS carers (Articles #4, #11 & #14). Some carers emphasised the need to plan their day ahead of time to ensure it goes smoothly. However, this often led to feelings of frustration because they felt that their days centred upon the person with MS, thus spontaneity had been removed from their lives (Article #14). Carers also faced difficulties in planning their daily schedules because it mostly depended on how the MS progressed resulting in unpredictable additional tasks (Articles #4, #11 & #14).

Burden of Care

Most studies revealed that caregiving can place a tremendous strain on MS carers' emotional, social and physical health and well-being. Some studies found that carers feel physically exhausted because of their caring responsibilities (Articles #3, #4, #11, #13 & #14). In particular, fatigue and sleeping difficulties were commonly reported which negatively impacted on MS carers' health and well-being (Articles #3, #4, #11 & #14).

Studies also identified a variety of emotional problems among MS carers which were attributed to caring. Most carers reported feelings of grief, helplessness, depression, and loss of self-esteem as a result of their caregiving experiences. However, our synthesis revealed that carers often suppressed their own feelings and emotions, put their own needs and plans on hold and neglected their own health (e.g. Articles #4, #6, #14, #16 & #17). Some studies reported that carers found being constantly in the presence of the care-recipient very stressful (Articles #1, #6, #7, #14 & #16) and often emphasised the need for a temporary escape from the caregiving responsibilities to relieve this tension and to be able to continue caring (e.g. Articles #1 & #16).

Several studies also found that carers often felt isolated and alienated because of being restricted to home for caregiving responsibilities (Articles #6, #10, #1, #11 & #17). Most carers had to give up their hobbies and reduce the time spent on social activities (e.g. Articles #6 & #10). Some carers reported that they got worried and felt guilty about leaving the care-recipient alone at home because they feared the possible consequences, so they often chose staying at home rather than engaging in a social activity which, in turn, led to feelings of isolation (e.g. Articles #1, #5, #10, #14 & #17). Furthermore, Heward, Molineux, and Gough (2006; Article #10) found that carers yearned for the times before the diagnosis of MS where they had enjoyed the company of their loved ones and had less pressure on their shoulders. Similarly, Hainsworth (1996; Article #2) found that reminders of MS free times or seeing the lives of healthy people led to feelings of grief.

Future Concerns

MS carers often worried about their own future, the care-recipients' future or the future of their families in general. Concerns about further deterioration in patients were common among MS carers because this would mean increased demand and burden (Articles #7, #10 &

#14). Many carers were also worried about losing their own health which would limit their ability to care for their loved ones and would force them to put the pwMS in care homes in the future (Articles #5 & #7).

Heward et al. (2006; Article #10) reported that carers were unable to make plans for the future because of the unstable nature of MS and its unpredictable progression. Similarly, Cheung and Hocking (2004a; Article #6) found that patients felt difficulty in looking toward the future because they have no control over the illness. Therefore, carers tended to avoid making long-term plans and thinking about the future, and adopt a day-by-day approach (Articles #2, #6 & #14). However, there were also some carers who derived comfort from having a plan for end-of-life decision (Article #17).

External Stressors

Studies identified a variety of external stressors that might increase burden and affect carers' health and well-being negatively. Findings showed that many carers were faced with financial difficulties which had forced them to make changes to their lifestyles (e.g. no holidays; Article #6). Lack of financial resources also made it difficult for carers to make the necessary home modifications for the patients (Articles #3, #8 & #13) and to get formal help (Articles #1, #4 & #14) resulting in further burden. Another external stressor was the environmental barriers encountered during day-outs with the care-recipient, such as inaccessible or lack of facilities (Articles #1 & #6) and reduced mobility for the pwMS who relied on wheelchairs (Articles #1 & #9). Such environmental barriers forced many of these carers to stay at home resulting in more tension and alienation.

Lack of understanding from family, friends and public was another source of stress for carers. Cheung and Hocking (2004a; Article #6) found that carers felt inferior because of the negative attitudes of others' toward them (e.g. perceiving carers as having lower status or

accomplishment). They sometimes felt that their own needs and situation were considered to be of secondary importance by others (Article #4). Additionally, lack of knowledge about the MS resulted in ignorance and unawareness of how much help was required (Articles #5, #9 & #17). For instance, Courts, Newton, and McNeal (2005; Article #9) reported that carers had difficulties when parking in the spaces reserved for people with disabilities because members of public could not acknowledge the degree of decline in pwMS.

Obtaining support was another challenge faced by carers. For instance, McKeown, Porter-Armstrong, and Baxter (2004; Article #5) found that most carers were unable to request family members' or friends' support because they were unable to share their problems and thought it was unfair to ask for their support. In contrast, some carers reported that their families or friends did not want to get involved, therefore they chose not to offer any kind of support (Article #6 & #13). Finding and accessing formal support was also very challenging because of lack of services (Articles #6, #9, #13 & #17), restrictive admission criteria for support provision (Articles #5, #6, #7 & #13), inflexible and unresponsive services (Articles #5, #6, #7, #9, #13 & #17), or simply because of the lack of information about the available services (Article #5). A further challenge was that sometimes available information was unreliable, inaccurate or irrelevant which could miss some vital facts, or invoke fear and feelings of insecurities about MS and caregiving (Articles #9 & #15).

Experiences of Support

Studies identified a wide variety of useful support resources that aid carers to cope with the demands of MS and caregiving. Informal support (i.e. support from family, friends and support group members) were described as a major source of help (Articles #1, #2, #4, #5, #6, #15, & #16). Similarly, a large proportion of studies described how receiving formal support (i.e. support from health professionals, organisations, etc.) helped carers cope with

the demands of caregiving (Articles #1, #4, #5, #10, #14, #16, & #17).

However, the level of satisfaction with the support received, either formally or informally, varied among the carers (e.g. Article #2). For instance, McKeown et al. (2004; Article #5) reported that although some early- and middle-stage carers were resisting asking for support from their families and unwilling to receive help from them, they reported that they were disappointed and upset that their families did not offer their support or that they stopped offering as the time passed and the disease progressed.

Moreover, McKeown et al. (2004; Article #5) found that only few participants reported being satisfied with the help received from formal networks, whereas many of them reported being very unhappy with the formal support. These findings were supported with other studies and the common reasons for this dissatisfaction was: lack of flexibility (Articles #5 & #7), lack of understanding and caring (Articles #2, #7 & #14), long waiting periods (Articles #5 & #7), poor quality (Articles #2, #5 & #7), and inequity of the system (Article #14). Additionally, the unmet support needs reported by MS carers were mostly of an emotional and psychoeducational nature (Articles #2, #9, & #17). As well as needing informational and physical help from formal support networks (Articles #2, #9, & #17), carers expressed that they needed emotional support from health professionals. They reported that they needed more competent, compassionate and caring professionals who would listen and respond to their needs, in a respectful and non-judgmental manner, and help them cope with the demands of caregiving (Articles #2 & #9).

Strategies Used in Managing the Caregiving Role
A wide variety of coping strategies in managing the carer role were identified, including engaging in social activities (Articles #1, #2, #4, #10, #14, #15, #16 & #17), getting professional psychological help (Articles #2, #4, #9 & #14), embracing the current situation as it is (Articles #4, #5, #6, #10, #12 & #16) and seeking peace through religious explanations

(Articles #2 & #12). Many studies also reported that learning about MS and understanding it in more detail helped carers regain some control over the disease and their caring role (Articles #9, #13, #14 & #15) reducing the possible adverse effects of the unpredictability and uncertainty of MS. In contrast, some carers reported that in the early stages of MS, they did not want to know about the illness or its impact on patients and their families to protect themselves from the reality of the illness and to gain control as a way of maintaining normality (Article #5).

Motivating Factors for Caring

The commitment to the relationship and sense of duty appear to be an important source of motivation for continuing to care among the spousal MS carers (Articles #1, #2, #4, #5, #6, #9, #10, #12, #14 & #16). Whereas some carers fulfilled the caring role as a duty to be performed and felt obligated to do so because of the vows they made (Articles #4, #10, #12 & #14), others took the caregiving role on willingly as a way of expressing their love and commitment (Articles #1, #12, #14 & #16). Yet some spousal carers chose to care based on "virtual reciprocity" (Article #4, p. 248) because they believed that their spouse would do the same for them if they had become ill (Articles #4 & #16).

Another common motivating factor was the desire to maintain some degree of normality among their lives and to protect their family from the possible adverse effects of MS. Some carers either rejected the support offered by others or resisted asking for help with the intention of maintaining normality (Article #5). Some became the advocates for health care of their partners with the intention of protecting their loved ones (Articles #9 & #16). Others took on the caring tasks to maintain their usual way of living (Articles #4, #9 & #10). Additionally, two articles reported that some carers were motivated by the desire to prevent an admission to a care home (Articles #4 & #17).

A variety of benefits of performing the caring role were also identified, including feeling proud of their achievements as a carer (Articles #5, #6, #12 & #16), personal growth and gaining strength (Articles #6, #8, #9, #10 & #12), learning new skills (Article #6), gaining personal satisfaction (Article #4 & #10), gaining insight and appreciating life (Articles #8 & #12). Some carers reported that gaining new skills and knowledge, and becoming involved in the MS community increased their self-esteem and boosted their confidence in their abilities (Articles #6 & #10). These gains also motivated carers to continue to care for prolonged periods, ameliorating the negative impact of caring on their lives.

The Line of Argument

Figure 1 summarises our line of argument and presents a proposed QoL model for MS carers explaining the relationships between the themes identified in this synthesis. Six of the nine higher order themes which were perceived by MS carers as problematic to their health and well-being were grouped together as stressors of caring. The remaining three themes were grouped as mediators because of their possible mediating effect on health and well-being, and QoL of MS carers.

[FIGURE 1 NEAR HERE]

Discussion

This synthesis has shown how MS carers' experiences of caregiving can be characterised by both positive and negative aspects, as providing care can bring both challenges and rewards to the carer. The line of argument resulting from this synthesis presents a proposed QoL model for MS caregiving which comprises of similar constructs to previously established generic (e.g. Lazarus & Folkman, 1984) and disease-specific (e.g. Haley, Levine, Brown, & Bartolucci, 1987; Pearlin, Mullan, Semple, & Skaff, 1990) caregiving models. For instance,

Haley et al.'s (1987) stress process model highlights caregiver stress as a relationship between the dementia caregiver and the environment and includes stressors, appraisal, coping responses, social support and adaptational outcomes (i.e. physical and psychological well-being). Similarly, our model describes stressors as physical, psychological, social and environmental influences (i.e. some of which are unique to the MS caregiving context) that might be problematic for the MS carer, and suggests that the use of coping resources and motivation might have a mediating effect on the relation between the stressors and the outcome.

In our proposed model, we adopted Cummins (1997) conceptualisation of QoL as the outcome. According to Cummins (1997), QoL is; "... objective and subjective, each axis being the aggregate of seven domains: material well-being, health, productivity, intimacy, safety, community, and emotional well-being. Objective domains comprise culturally-relevant measures of objective well-being. Subjective domains comprise domain satisfaction weighted by their importance to the individuals" (p. 6). This definition is one of the widely accepted generic QoL definitions because it attempts to define QoL as a global construct encompassing both positive and negative dimensions (Aubeeluck & Buchanan, 2006; Rapley, 2003).

The findings of our synthesis suggest that there is an inter-relationship between the seven domains of Cummins' (1997) QoL construct (ComQoL-A5) and the third order constructs that emerged from our synthesis (i.e. components of the proposed model). Eight out of nine themes could be related to at least one of the seven QoL domains of ComQoL-A5. However, the theme 'MS and patient related challenges' was unable to be related to the ComQoL-A5. This was not surprising, considering the fact that the ComQoL-A5 is a generic construct of QoL and lacks disease-specific factors. However, our findings revealed that MS-

specific factors are a prominent theme in MS carers' life. Therefore, it seems necessary to consider the MS-specific factors while examining MS carer's QoL.

For instance, our synthesis showed that unpredictability was a major concern for informal carers of pwMS. More specifically, the unpredictability of exacerbations and remissions were a great source of distress for carers. The caring tasks and the future might also become unpredictable and uncontrollable because of the unstable nature of MS. Studies with pwMS reveal similar findings suggesting that the unpredictability of MS can cause high levels of uncertainty which is associated with greater psychological distress, anxiety, pathological worry and depressive symptoms in pwMS (Bruce & Arnett, 2009; Lynch, Kroencke, & Denney, 2001; McCabe, Ebacioni, Simmons, McDonald, & Melton, 2015). The unpredictable nature of MS might also decrease the perceived control people can have on the disease, the caregiving role and the future. According to Armfield (2006), perceptions of uncontrollability and unpredictability might induce anxiety which could eventually have a negative effect on the QoL of the individuals. However, the relationship between unpredictability and MS carer QoL warrants further investigation as we are not aware of any research exploring this potential association.

Additionally, our review of the literature shows that none of the included studies explicitly focused on understanding the QoL of MS carers and that there is a lack of research exploring the QoL of MS carers from a qualitative perspective. Therefore, there is a need to conduct more qualitative studies with this population to provide triangulation of the synthesis findings in order to ensure their trustworthiness and credibility. Studies in this area will contribute to our understanding of QoL of MS carers and to operationalizing the definition of QoL for carers of pwMS for future research and practice in a more reliable way.

Strengths and Limitations

By synthesizing existing qualitative studies in MS caregiving literature, a more comprehensive overview of potentially relevant factors to MS carers' health and well-being were derived which could not be obtained through the findings from each of the individual articles. To our knowledge, this is the first systematic review and meta-synthesis of published qualitative studies of MS informal carers' needs and experiences. It is encouraging to note, however, that qualitative meta-syntheses of the impact of informal caregiving across conditions are beginning to be published. For example, Yong and Price (2014) have recently conducted a comprehensive meta-synthesis of qualitative studies on family caregiving in dementia exploring the occupational impact of dementia caregiving on carers. Although we cannot directly compare their findings to our own (as our review is focused specifically on MS and explored the impact of caregiving from a multidimensional perspective) we consider reviews such as Yong & Price's important for adding to our holistic understanding of the impact of caregiving and the multitude of ways this role can impact on the carer.

The critical appraisal of each article by two different reviewers and the discussion panel with two other independent reviewers enhanced the rigor of our synthesis. Similarly, third order constructs were cross-verified independently by all the members of the research team to ensure the trustworthiness of the findings.

This article highlights some important issues in informal MS caregiving; however, it is not without limitations. As with previous meta-syntheses, finding qualitative studies among electronic databases was challenging (Dixon-Woods et al., 2006; Dixon-Woods, Fitzpatrick, & Roberts, 2001; Shaw et al., 2004; Taylor, Shaw, Dale, & French, 2011). Although our search strategy was in line with the recommendations for identifying qualitative research articles within electronic databases (Shaw, 2011), it is possible that relevant qualitative studies were overlooked. We tried to overcome this limitation by asking key researchers in

this area to identify any additional relevant articles that they or their colleagues had published. For conducting a more robust synthesis in the future, there is still a need for a greater effort in indexing qualitative research in electronic databases and the use of explicit qualitative methodology descriptors from authors (Shaw et al., 2004).

Furthermore, it is unclear whether our findings are applicable to contexts other than those included in the synthesis. For instance, participants were mainly spousal carers and other family members or friends who provide voluntary unpaid care were underrepresented. It is clear that more research into other family members' or friends' caregiving experiences is needed. In addition, 15 of the included studies were undertaken in the UK, the USA and Australia, therefore the transferability of the findings beyond these contexts is unclear.

Only published articles were included in the synthesis. Therefore, relevant research reported in theses and book chapters or grey literature is not represented, which might have led us to omit some possibly important data. Although we included 17 articles, these represented only 14 studies. Synthesizing articles that use the same data might lead to overrepresentation of certain second order constructs (Malpass et al., 2009) and might limit the representativeness of the synthesis findings. It should also be noted that the process of meta-ethnography is interpretive in nature; it is possible that a different research team might have developed a different conceptual framework (Malpass et al., 2009).

Conclusion

Our findings indicate that caring for an individual with MS is a life-changing experience with many stressors and only some benefits, which may impact on the health and well-being of these informal carers. However, how carers cope with these challenges, how they use available resources and what motivates them to continue to care might also be important contributors to their well-being and can explain the variability in their health status.

Our synthesis presents a QoL model for informal MS caregiving and a preliminary list of operationalisations of MS carers' QoL. Our findings support McCullagh, Brigstocke, Donaldson, and Kalra's (2005) argument that caregiving is a complex, multidimensional and dynamic process where its nature and determinants evolve over time. In addition, our proposed model highlights the important role of the disease-specific factors (e.g. unpredictability of the disease, future and their carer role) in health and well-being of MS carers, which needs to be considered while designing, implementing and testing interventions. We believe that this synthesis presents novel conceptual and theoretical insights to MS informal carers' QoL which can be useful in designing interventions to improve carers' QoL and in developing disease-specific outcome measures to test the efficacy of such interventions. Our proposed framework can also guide the provision of services to MS carers in a more accurately targeted way at the appropriate stages of the caregiving process. For instance, during the initial stages of the caregiving process, interventions that are psychoeducational in nature would be more useful to increase the level of understanding of MS, its unpredictable symptoms and prognosis, and the level of adjustment to the caregiving role. However, further empirical research is needed to examine the utility of our model and to explore the concept of QoL in MS carers in more detail.

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Figure 1. Quality of life model for carers of people with Multiple Sclerosis representing the relationships between the themes identified from the synthesis.