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# The experience of informal caregivers of patients with motor neurone disease: A thematic synthesis

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## ABSTRACT

*Objective:* Research is required in order to illustrate and detail the experiences of informal caregivers of patients with motor neurone disease (pwMND) to further advance the research base and to inform the development of future support structures and services. Due to the heterogeneous nature of caregiving for pwMND, one way in which this can be achieved is through a qualitative review. A qualitative thematic analysis of existing qualitative studies has not, to the best of the authors' knowledge, been previously undertaken. Thus, the present synthesis aims to identify caregivers' experiences and to suggest factors that contribute to these experiences in order to fulfill the required research needs.

*Method:* A thematic synthesis of qualitative literature was conducted. AMED, Medline, SPORTDiscus, CINAHL, and PubMed were electronically searched from inception until September of 2015. Studies were eligible if they included qualitative literature reporting on firsthand experience of informal caregivers of patients with MND, were published in English, and contained verbatim quotations. Critical appraisal was undertaken using a 13-item consolidated criteria for reporting qualitative studies (COREQ) checklist.

*Results:* A total of 10 studies met the inclusion criteria, with 148 (50 male) current or previous informal caregivers of pwMND identified. Critical appraisal demonstrated that study design and reflexivity were underreported. The synthesis derived three themes: (1) loss of control, (2) inability to choose, and (3) isolation.

*Significance of results:* The synthesis highlighted the factors that contribute to both positive and negative caregiving experiences. Through these experiences, such suggestions for service provision as improving communication with healthcare professionals and having a single point of contact emerged. However, the outcome of such suggestions on the experience of caregivers is beyond the scope of our synthesis, so that further research is required.

**KEYWORDS:** Caregivers, Motor neurone disease, Qualitative research, Experience

## INTRODUCTION

Motor neurone disease (MND) is an adult-onset neurodegenerative disorder represented by four subtypes, the most common being amyotrophic lateral sclerosis (ALS) (Aoun et al., 2013; Nageshwaran, 2014). The terms ALS and MND are utilized interchangeably and inconsistently: the United Kingdom (UK) uses MND as an umbrella term, whereas other countries employ ALS (Mitchell & Borasio, 2007; Aoun et al., 2013). Therefore, they will be used synonymously in our study. Around 4,500 people in the UK are living with MND at any point in time, and peak onset is between the ages of 50 and 70 years (MND Association, 2015; 2016). Although symptoms and rate of progression vary between the four subtypes (Herz et al., 2006; Nageshwaran, 2014), the most common symptoms include muscle wasting, weakness, fasciculations, and dysarthria (Mitchell & Borasio, 2007; Nageshwaran, 2014). Currently there is no cure, and 50% of patients do not live beyond 3 years after the onset of symptoms (Mitchell & Borasio, 2007).

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In the period between symptom onset and death, care of the patient often falls upon family members or friends (Mockford et al., 2006). The time spent caregiving is variable and depends on patients' disease progression, with Chio et al. (2006) reporting for patients with a mild disability (ALS Functional Rating Scale [ALS-FRS] score $>30$) a mean of 5 hours/day and for patients with severe impairments (ALS-FRS score $\leq 10$) a mean of 15 hours/day. Caregiver quality of life (QoL) is significantly negatively impacted by undertaking this role (Peters et al., 2013), and many caregivers experience burden associated with their role, attributed to physical, emotional, social, financial, and psychological factors (Sorrell, 2014).

Although research focusing on the caregiver experience is expanding, it remains sparse (Mockford et al., 2006). Two relevant reviews have been published. Mockford et al. (2006) focused on published literature from 1994 to 2004, and the main findings discussed caregivers experiencing ill heath when unsupported and the positive impact of maintaining social support and activities in terms of reducing the chances of ill health. Aoun et al. (2013) conducted a more recent review for the period 2000–2011 which indicated that, although research has highlighted such factors as diagnostic process and access to information and services that contribute to burden and negatively impact QoL, there is a need to develop literature focusing on improving the caregiver experience. As further literature has been published since 2011, our study aims to update the existing reviews.

Researchers have called for further consideration of caregivers' experiences, particularly qualitative-based reviews (Goldstein et al., 2006; Murphy et al., 2009; Atkins et al., 2010). Due to the expanding literature base and the risk of single qualitative studies being overlooked in practice if not synthesized (Malpass et al., 2009), it was considered important to utilize a qualitative review-based approach to address these calls. Both Mockford et al. (2006) and Aoun et al. (2013) included quantitative and qualitative literature, and hence, as our study aims to focus solely on a qualitative approach, it has the potential to offer an alternative interpretation, and the results of the caring experience can be considered as heterogeneous and subject to personal or unique interpretations (Crellin et al., 2014). Thus, our study aimed to provide a thematic synthesis of existing qualitative literature regarding the experiences of informal caregivers of pwMND. We aimed to encompass the spectrum of experiences through not focusing on a specific stage of disease progression. Additionally, we also aimed to highlight the areas of service development to reduce caregiver burden in the future through discussing positive caregiver experiences and by heeding the suggestions made in the existing literature.

## METHODS

The methods section herein is reported in accordance with enhanced transparency in reporting the synthesis of qualitative research (ENTREQ) guidelines (Tong et al., 2012).

### Search Strategy

The following electronic databases were searched from inception until September of 2015: the Allied and Complementary Medicine Database (AMED), Medline, SPORTDiscus, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), and PubMed. Multiple databases were utilized to maximize search yield (Wu et al., 2012). The selected databases were chosen because they focus on the medical, allied health, and social science disciplines (Shaw et al., 2004; Hewitt, 2007), and all but PubMed have been used in published reviews and synthesis concerning MND (Aoun et al., 2013; Soundy & Condon, 2015).

Combinations of the following key terms were utilized: "care*" OR "caregiv*" AND "motor neuron* disease" OR "amyotrophic lateral sclerosis" OR "MND" AND "experience" AND "qualitative" OR "mixed method" AND "interview" OR "focus group." Although highlighted as potential keywords, "family" and "perceptions" were not utilized, as they retrieved large volumes of irrelevant studies during the scoping search. Additional search strategies included citation chasing, reviewing reference lists, and contacting researchers ($n = 1$).

### Eligibility Criteria

Included studies needed to fulfill eligibility criteria that utilized the SPIDER (sample, phenomenon of interest, design, evaluation, research type) search tool (Cooke et al., 2012):

- **Sample**: Informal primary caregivers are defined as individuals who previously or currently provide the majority of support and assistance to an MND patient at any disease stage, without financial reward.
- **Phenomenon of interest**: Studies reporting informal caregivers' firsthand experiences with MND are included.
- **Design**: Any type of qualitative design was considered. The excluded designs were quantitative research, mixed-method research not containing clear qualitative data, gray literature,

unpublished or protocol studies, theses, conference proceedings, and case studies.

- **Evaluation**: Any qualitative method of data collection—interview, focus group, or mixed-method articles containing clear qualitative research—were included.
- **Research type**: Studies including clear qualitative data from a qualitative or mixed-method study. Studies published in languages other than English were excluded.

### Critical Appraisal

Our review used an adapted 13-item version of Tong et al.'s (2007) consolidated criteria for reporting qualitative studies (COREQ) checklist, which has three domains:

1. Research team and reflexivity, which increase the transparency of the personal characteristics of the research team as well as their relationship with participants, which could influence bias in data collection and interpretation of results.
2. A study design that appraises participant retention and the data collection process.
3. Data analysis and reporting that appraise the process of coding and derivation of themes.

The adapted version of COREQ was devised by Soundy et al. (2016) following a critique of the larger 32-item checklist (Tong et al., 2007), when items that were not relevant were removed.

Studies were independently appraised scoring 1 for reporting a checklist item and 0 if unclear or unreported (Tong et al., 2007). A total score of 13 was possible and, based on Soundy et al.'s (2016) previous research, a score of 4 was identified as a threshold for further consideration. Any studies scoring less than 4 were discussed to identify if there was a methodological weakness that compromised the results and warranted exclusion of the article. No articles were excluded following this process.

### Synthesis

An adapted four-stage approach was taken for our synthesis (Thomas & Harden, 2008; Aria et al., 2007; Rodgers et al., 2009): Stage 1 involved textual description, extraction, and tabulation of results, following which a summary of individual extracted quotations via line-by-line coding was used to facilitate comparison. Stage 2 involved idea webbing to visualize and connect concepts between studies (Rodgers et al., 2009). Stage 3 involved translating, interpreting, and converting similar concepts between studies (Aria et al., 2007). Stage 4 involved synthesis of translations to combine the analyzed themes and form a new interpretation of the studies involved. See the supplementary file for the audit trail (see Supplementary Materials).

## RESULTS

### Systematic Search

The full search process is depicted via a PRISMA (Moher et al., 2009) flow diagram (Figure 1). A total of 10 articles (Brown, 2003; Akiyama et al., 2006; Herz et al., 2006; Ray & Street, 2007; Aoun et al., 2012; O'Brien et al., 2012; Whitehead et al., 2012; Hyunjin & Schepp, 2013; Oyebode et al., 2013; Weisser et al., 2015) met the inclusion criteria and were analyzed within the thematic synthesis. The characteristics of the included participants are presented in Table 1.

### Critical Appraisal

The results of the 13-item COREQ evaluation ranged from 4 (Akiyama et al., 2006; Whitehead et al., 2012) to 8 (Ray & Street, 2007), with a mean total score of 6. Hence, no studies were excluded nor discussed for exclusion. For full COREQ results, see the supplementary file (see Supplementary Materials).

Although Akiyama et al. (2006) and Whitehead et al. (2012) scored the minimum of 4, they were the only included studies to score 0/5 within domain 1 of COREQ. This impacts the credibility of these studies' results, as there is a lack of transparency with regard to researchers' credentials and their relationship with participants, so that there is an inability to distinguish the degree to which personal bias may have influenced interpretation of the findings (Tong et al., 2007). Both Akiyama et al. (2006) and Whitehead et al. (2012) failed to report on the number of, or reason for, participant dropout, resulting in the possibility of selection bias (Tong et al., 2007; Cote & Turgeon, 2009). Furthermore, although both studies described a coding process, Akiyama et al. (2006) failed to report on the derivation of themes. Therefore, there is a lack of transparency regarding study design and the researchers' rationale for their interpretation and analysis of the data (Fossey et al., 2002). Hence, due to the lack of transparency limiting an ability to interpret results, novel data from Akiyama et al. (2006) and Whitehead et al. (2012) were interpreted with caution and with consideration of the findings from the other included studies.

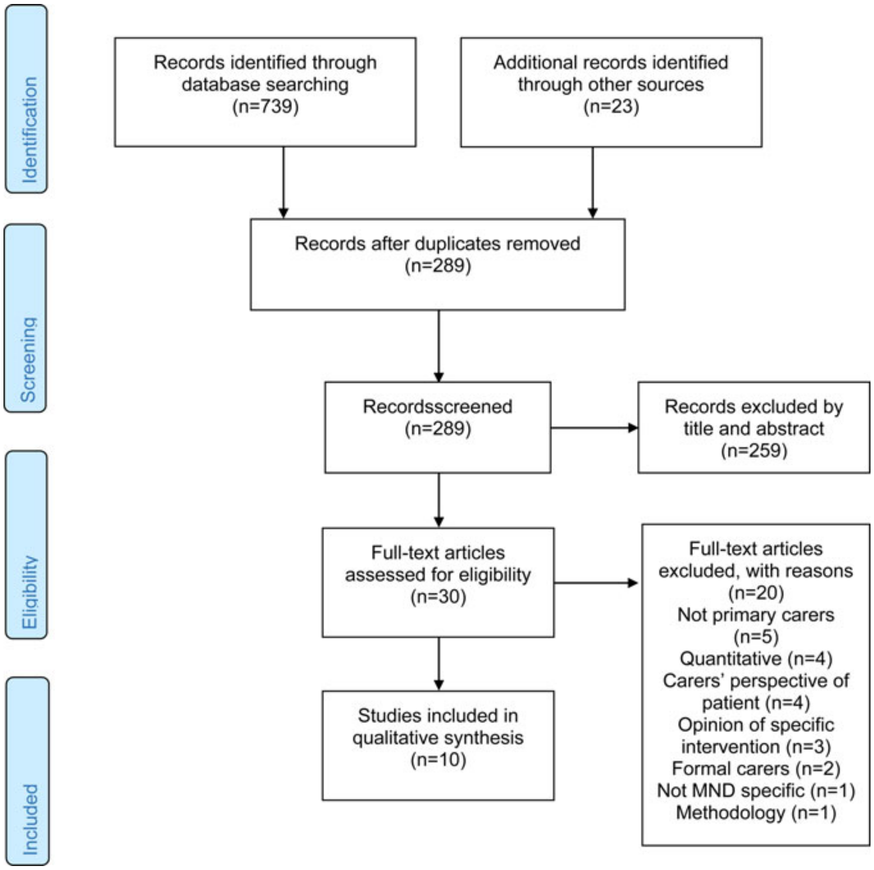

**Fig. 1.** PRISMA flow diagram.

## Thematic Synthesis

The thematic synthesis highlighted that, although the caregiver experience is nonhomologous, shared key themes do exist. These themes are explored below and are grouped as follows: (1) factors that contribute to the experience of loss of control, (2) factors that contribute to a sense of choice, and (3) factors that contribute to an experience of isolation. Studies that support each theme are presented in Table 2.

**Table 1.** *Characteristics of participants included in the synthesis from the 10 studies*

| Participant characteristics | $N = 148$ |
|---|---|
| Country of residence | |
| Australia | 45 |
| Japan | 12 |
| South Korea | 11 |
| United Kingdom | 80 |
| Caregiver type | |
| Family | 63 |
| Spouse only | 84 |
| Non-relational | 1 |
| Caregiver status | |
| Bereaved | 44 |
| Current | 104 |
| Gender | |
| Female | 98 |
| Male | 50 |

## Loss of Control

### *Progressive Nature of MND*

The progression of the MND of the cared-for patients was considered in relation to the idea of loss of control within the caregiving role. Several studies highlighted that caregivers were unsure of how their role would evolve along with the progression of the disease (Aoun et al., 2012; Brown, 2003; O'Brien et al., 2012; Ray & Street, 2007; Whitehead et al., 2012). They were aware that symptoms would progress but were unable to explain how the symptoms would change and how this would impact their caregiving role (O'Brien et al., 2012). Furthermore, caregivers were uncertain about the rate of progression (Aoun et al., 2012; O'Brien et al., 2012; Oyebode et al., 2013; Ray & Street, 2007; Whitehead et al., 2012). These factors contributed to caregivers' inability to plan and prepare for the future within their caregiving role (Aoun et al., 2012; Brown, 2003; O'Brien et al., 2010; Ray & Street, 2007; Whitehead et al., 2012). One caregiver epitomized this as follows: "This is so undetermined. You just don't know it from

**Table 2.** *Overview of themes and study support at the subtheme unit*

| Theme | Subtheme | Supporting studies |
|---|---|---|
| Loss of control | Due to the progressive nature of MND | 2, 3, 4, 6, 7, 8, 10 |
| | Over daily activities | 1, 2, 3, 4, 5, 6, 7, 8, 9 |
| Choice | Uptake of caregiver role | 1, 4, 6, 7, 10 |
| | Access to alternatives | 2, 3, 4, 6, 7, 10 |
| Isolation | Communication with healthcare professionals | 2, 3, 4, 6 |
| | Communication between family and friends | 1, 2, 4, 5, 6, 7, 8, 9 |
| | Changes to relationship dynamic | 2, 4, 5, 7, 8, 9 |
| | Loss of intimacy | 2, 5, 7, 8 |
| | Opportunity for interaction | 1, 2, 3, 4, 6, 8, 9 |

1 = Akiyama et al. (2006); 2 = Aoun et al. (2012); 3 = Brown (2003); 4 = Herz et al. (2006); 5 = Hyunjin & Schepp (2013); 6 = O'Brien et al. (2012); 7 = Oyebode et al. (2013); 8 = Ray & Street (2007); 9 = Weisser et al. (2015); 10 = Whitehead et al. (2012).

one moment to the other. Whether it's going to stop in its tracks or plummet downhill. You just don't know, and that's very difficult" (Ray & Street, 2007). Furthermore, this inability to prepare for continual progression resulted in a sense of progressively overwhelming responsibility and a constant struggle to stay in control (Herz et al., 2006). For some, uncertainty resulted in fear and anxiety (Aoun et al., 2012; O'Brien et al., 2012); conversely, other caregivers could cope by embracing uncertainty and "living in the moment" (Weisser et al., 2015; Oyebode et al., 2013).

One suggestion with regard to uncertainty is that the breadth of symptoms and life expectancy of pwMND is not fully understood and therefore cannot be accurately predicted (Ray & Street, 2007). However, the experience of other caregivers suggests a breakdown in communication between healthcare professionals and caregivers, where relevant information is not shared with caregivers (O'Brien et al., 2012; Ray & Street, 2007). A suggested reason for a lack of clear communication is that professionals do not have sufficient time for discussion with both the pwMND patients and their caregivers (Herz et al., 2006; O'Brien et al., 2012). Furthermore, insufficient time has been associated with professionals' lack of up-to-date MND-specific knowledge (Herz et al., 2006), which subsequently limits caregivers' knowledge and expectations about the intensity of future caregiving.

### *Loss of Control over Daily Activities*

The idea that caregivers have a sense of loss of control over their own lifestyle emerged in several studies (Akiyama et al., 2006; Aoun et al., 2012; Brown, 2003; Herz et al., 2006; Hyunjin & Schepp, 2013; O'Brien et al., 2012; Oyebode et al., 2013; Ray & Street, 2007). A key idea for the mechanism for this developed from the sense of "role captivity" (Akiyama et al., 2006; Aoun et al., 2012; Herz et al., 2006; Hyunjin & Schepp, 2013; O'Brien et al., 2012; Oyebode et al., 2013; Ray & Street, 2007): "You can't go out when you want to go out. You can't go where you wish to go or you used to be able to go" (Oyebode et al., 2013). Caregivers highlighted a sense of being solely responsible for daily care needs without the support of others to share that role and thus allow previous activities and social opportunities to be continued (Akiyama et al., 2006; Aoun et al., 2012; Herz et al., 2006; Hyunjin & Schepp, 2013; O'Brien et al., 2012; Oyebode et al., 2013; Ray & Street, 2007). Hence, the sense of captivity and subsequent loss of control could differ between caregivers depending on the level of support they received (Oyebode et al., 2013; Weisser et al., 2015). However, formal support may have a peak intensity that prevents a feeling of loss of control, and intensity above this threshold contributes to a caregiver's experience of loss of control due to loss of privacy and increased dependence on others (Weisser et al., 2015). However, this idea did not consistently emerge throughout the included studies, so it is plausible that the contribution of support to loss of control may be caregiver-specific.

Alternatively, one study explored the idea that the loss of ability to choose one's own daily activities arose from an inability to complete activities previously shared with the MND patient due to their functional decline (Ray & Street, 2007). This demonstrates how others influence the sense of control over decision making for one's own life.

For caregivers, one consequence of the experience of loss of control over activities was the sense of burden (Hyunjin & Schepp, 2013; O'Brien et al., 2012). Another was the sense of living for others, thus dehumanizing their own existence (Akiyama et al., 2006; Herz et al., 2006; O'Brien et al., 2012; Oyebode et al., 2013; Ray & Street, 2007). For some, this resulted in a change in an individual's self-perception and a sense of being a different version of themselves

(Herz et al., 2006; Ray & Street, 2007): "You're not yourself. You're certainly not yourself ... It's— You feel more of a carer than anything" (Ray & Street, 2007).

## Choice

### *Choice Regarding Uptake of the Caregiver Role*

Caregivers experienced a sense of obligation, resulting in an inability to choose whether or not to become a caregiver (Herz et al., 2006; O'Brien et al., 2012; Oyebode et al., 2013; Whitehead et al., 2012). One emerging idea was the sense that the wishes of the pwMND to be cared for by informal caregivers came before the caregivers' needs or desires (Akiyama et al., 2006; O'Brien et al., 2012; Whitehead et al., 2012) due to the sense of needing to alleviate suffering within a limited lifespan (Whitehead et al., 2012). Furthermore, to protect the MND patient from further suffering, there is a sense that caregivers conceal the difficulties and struggles from the patient, so that their own needs go unnoticed and neglected (Oyebode et al., 2013). However, for others the obligation was engrained within the expectation to care for loved ones (Herz et al., 2006) and to demonstrate a commitment to marriage (Oyebode et al., 2013).

### *Access to Alternatives*

Caregivers experienced a lack of alternative options outside of caring full-time for pwMND (Aoun et al., 2012; Brown, 2003; Herz et al., 2006; O'Brien et al., 2012; Oyebode et al., 2013; Weisser et al., 2015; Whitehead et al., 2012). Although unable to access support, some caregivers could identify the need for specialist support (Herz et al., 2006), while others suggested that they had insufficient time and that the captivity of the role did not allow for consideration of the need for support (Aoun et al., 2012).

Our synthesis highlighted the financial implications of accessing support (e.g., equipment or paid-for caregivers) as being a luxury that not everyone can access (Herz et al., 2006; Oyebode et al., 2013; Whitehead et al., 2012). Furthermore, a lack of timely haste in processing applications for financial support prevented eligible pwMND from receiving funding due to their passing away prior to the reviewers coming to a decision (Whitehead et al., 2012). Although this may relate to the difficulty of predicting the progression of the MND, it also highlights the need for more rapid consideration of applications.

Also highlighted was a lack of specialist caregiving services for the needs of pwMND (Aoun et al., 2012; Herz et al., 2006; Oyebode et al., 2013; Whitehead et al., 2012). This resulted in caregivers not being confident that the needs of pwMND would be met and a subsequent unwillingness to release the caregiving role to unspecialized support services, such as paid-for caregivers (Herz et al., 2006; Whitehead et al., 2012). However, this is not conclusive, as other caregivers were able to access specialist support from services such as the MND Association and hospice care (Herz et al., 2006). Hence, care quality may be inconsistent between providers.

## Isolation

### *Communication with Healthcare Professionals*

Another idea that emerged from our synthesis was that communication between professionals and caregivers contributed to caregivers' sense of being unsupported and isolated (Aoun et al., 2012; O'Brien et al., 2012). One factor highlighted was the idea of the high volumes of involved healthcare professionals without clarity about each of their roles and without a key member to contact (O'Brien et al., 2012). It could thus be suggested that potential sources of support are not utilized due to poor clarity of communication, which leads to caregivers being overwhelmed. Furthermore, discussions with professionals for some lacked depth or personalization and was not focused on the emotional aspects of caregiving for an MND patient (Brown, 2003), resulting in the caregivers feeling unsupported (Brown, 2003; Herz et al., 2006): "I think it's all talked about mainly in a business-like manner rather than in an emotional way" (Brown, 2003). This suggests a lack of services that attend to caregivers' needs and a requirement for professionals to shift their focus when communicating with caregivers. However, others used counseling services that allowed cathartic release (Herz et al., 2006; O'Brien et al., 2012), so that depth and effectiveness of communication were accessible for some caregivers, but this appears to be profession-dependent and may highlight a greater need for access to counseling services in order to meet caregivers' needs.

### *Communication between Family and Friends*

Changes in communication within preexisting relationships with family and friends emerged in several studies (Herz et al., 2006; Hyunjin & Schepp, 2013; O'Brien et al., 2012; Oyebode et al., 2013; Weisser et al., 2015). A sense of an inability to share was noted as a way to avoid burdening others (Herz et al., 2006; Hyunjin & Schepp, 2013; O'Brien et al., 2012), which resulted in concealing emotions from others and a sense of isolation: "I'd cry. I used to cry in the shower. The shower was my friend. I loved my shower. Because they don't notice your tears in the shower"

(Herz et al., 2006). Additionally, caregivers struggled to communicate with uninvolved others due to their lack of understanding of the caregivers' situation and burden (Hyunjin & Schepp, 2013), suggesting a lack of empathy, certainly an alienating experience. However, other caregivers appreciated a sense of normality when communicating with friends (Akiyama et al., 2006; Ray & Street, 2007), indicating that communication within previous relationships was not necessarily impacted by the caregiving role. However, it is plausible that this sense of "normality" is a denial-related coping mechanism where the caregiver avoids sharing their current experience or concerns. Thus, it is arguable that communication is impacted, as there may be a shift in the topics that may be openly discussed when communicating with others.

Caregivers also experienced a decreased frequency of verbal communication with pwMND, another preexisting relationship (Hyunjin & Schepp, 2013; Weisser et al., 2015). One contributing factor that emerged was that MND symptoms were attributed to a decreased quality of communication caused by the need to simplify language (Oyebode et al., 2013). For some, the frustration of the caregiving role led to interpersonal tension and avoidance of communication (Hyunjin & Schepp, 2013; O'Brien et al., 2012). Alternatively, others noted an increase in nonverbal communication through activities involved with the caregiving role (Weisser et al., 2015), demonstrating a potential shift away from verbal discussion and hence altering the method of communication. Additionally, some caregivers found an ability to share more readily with pwMND, increasing the amount of verbal communication (Weisser et al., 2015); however, the motive for this is unclear and could be due to differing factors, such as the terminal prognosis, as opposed to being directly due to the caregiving role.

### *Changes to Relationship Dynamics*

Caregivers experienced a change in the dynamics of their relationship with pwMND (Aoun et al., 2012; Herz et al., 2006; Hyunjin & Schepp, 2013; Oyebode et al., 2013; Ray & Street, 2007; Weisser et al., 2015). Former spousal relationship identities became a nurse–patient or parent–child relationship due to the dependence of the MND patient and a lack of reciprocity within the relationship (Aoun et al., 2012; Hyunjin & Schepp, 2013; Ray & Street, 2007). Caregivers explored the sense of disconnection from their partner and the sense of now being alone in their marriage (Hyunjin & Schepp, 2013; Oyebode et al., 2013). For some, this escalated to questioning their commitment to the marriage due to the dependence of the MND patient and the intensity of the caregiving role (Herz et al., 2006; Hyunjin & Schepp, 2013).

### *Loss of Intimacy*

Another factor contributing to the changing dynamics of spousal relationships and subsequent experience of isolation was the loss of physical and emotional intimacy (Aoun et al., 2012; Hyunjin & Schepp, 2013; Oyebode et al., 2013; Ray & Street, 2007). Aoun et al. (2012) and Oyebode et al. (2013) discussed the physical barriers caused by MND symptoms that limited intimacy and resulted in an experience of isolation due to the absence of physical contact and the loss of a sense of connection with their spouse. Additionally, for some caregivers the requirement to attend to the personal care of pwMND limited their desire for physical intimacy (Hyunjin & Schepp, 2013; Ray & Street, 2007). Caregivers' decreased desire for physical intimacy resulted in increased tension between couples, subsequently leading to a heightened sense of isolation (Hyunjin & Schepp, 2013). However, as well as the direct relationship between attending to MND patient care needs and loss of desire for intimacy, caregivers highlighted how attending to the personal care of the patient altered the relationship due to the loss of reciprocity, and this also changed the relationship by limiting intimacy (Aoun et al., 2012; Ray & Street, 2007). This demonstrates that a change in relationship identity can be the cause of loss of intimacy and can result in a loss of intimacy.

### *Opportunities for Interaction*

A decreased opportunity for social interaction with others further contributes to caregivers' experience of isolation (Akiyama et al., 2006; Aoun et al., 2012; Brown, 2003; Herz et al., 2006; O'Brien et al., 2012; Ray & Street, 2007; Weisser et al., 2015). A decreased opportunity for interaction was also related to role captivity in caregiving and subsequent insufficient time. As one caregiver reported, "I used to go weeks and sometimes months before I saw anybody" (Brown, 2003), highlighting physical isolation from others and limiting interaction. As well as limited time, fatigue associated with the caregiving role was associated with decreased interaction (Ray & Street, 2007). However, another caregiver highlighted the sense of isolation and loneliness despite maintaining work along with the caregiving role (O'Brien et al., 2012), suggesting that physical isolation is not the only factor contributing to limitation of interactions. For other caregivers, the alteration in relationship dynamics and the inability to complete social activities previously completed as a couple limited social opportunities, leading to a sense of isolation during both the caregiving and bereavement phases (Ray & Street, 2007). Conversely, Aoun et al. (2012) suggested that isolation was only experienced during the bereavement phase, as caregivers were supported during the caregiving phase: “You go from having a whole army of people, [and] then it’s just you.” However, whether the caregiver received formal support in the form of paid caregivers or informal support from friends and family was not discussed, which may have impacted the sense of isolation and loss of interaction. Hence, an opportunity for interaction may be caregiver-specific, and the level of support may impact a caregiver’s experience of isolation.

## DISCUSSION

The aim of this thematic synthesis was to explore existing literature to increase the depth of our understanding of the experience of informal caregivers. Through synthesis of the existing qualitative literature, three themes were identified that demonstrate how loss of control, absence of choice, and isolation are experienced by caregivers, and we identified the factors that contribute to these experiences.

A key idea highlighted across themes was the sense of being solely responsible for the MND patient without help or support from others. This responsibility can stem from the sense of obligation to fulfill the caregiver role either to satisfy the patient’s wishes and demonstrate love and commitment, or it may be due to the absence of alternatives or support. Access to support differed between participants included in our thematic synthesis, with those individuals who were financially able being in an advantageous position. However, as the synthesis included studies from many nations, financial requirements and availability of support systems differed between countries (Sakellariou et al., 2013). Also highlighted was the lack of communication about available support between healthcare professionals and caregivers. The reasons for this lack of communication included insufficient time, inadequate knowledge about MND, a lack of awareness among healthcare professionals of caregivers’ emotional difficulties, or having a multitude of involved healthcare professionals with a lack of clarity about their specific roles. Also highlighted was the need for healthcare professionals to have sufficient knowledge and an ability to communicate about the likely future for pwMND and how this would subsequently impact caregivers. One idea that emerged was the need for a key worker to address caregivers’ questions, to provide practical and emotional support, and highlight or suggest relevant services to increase access to support. However, this idea was not employed in any of the included studies, so that this outcome cannot be determined within the scope of our thematic synthesis. However, this idea has been reflected in the UK through the NICE recommendation to provide a single point of contact within the MND-specific multidisciplinary team (NICE, 2016). This supports the ideas emerging from our synthesis, and it is strongly recommended for implementation in clinical practice.

Caregivers’ sense of being solely responsible for pwMND resulted in insufficient time to maintain social interactions and take part in previously completed activities due to being a captive within the caregiver role. This was associated with a sense of burden, a feeling of isolation, and the belief that one was living for others. This is supported by previous research regarding informal family caregivers for nonspecific chronic conditions that highlighted how insufficient time for activities that previously contributed to personal identity can result in a sense of living for others and a loss of self (Eifert et al., 2015). Our thematic synthesis highlighted the need for support or a respite to allow for maintenance of activities; however, the frequency, type, or feasibility of support was not discussed, so that it is beyond the scope of this synthesis to make conclusive recommendations. Despite this, our synthesis highlights the need for formal caregivers to have adequate knowledge and skills to care for pwMND and to allow informal caregivers to readily accept support, as insufficient ability is a key identified reason for underutilization of support. This is also suggested by Aoun et al. (2013), who proposed the need to train involved service providers to improve their understanding of working with pwMND; however, this was not discussed in the context of poor uptake of support services. Conversely, another barrier to social interaction is the sense of wanting to avoid burdening others, and a lack of understanding or empathy from uninvolved others limiting communication and interaction within previous relationships. Therefore, time for social interactions through formal support alone would not address these issues. Further research is indicated to understand the complexities of caregivers’ social interactions in order to make more conclusive recommendations to address these issues.

Caregivers experienced a sense of isolation, with one contributing factor being a shift in the dynamics of and identity within the spousal relationship due to the dependence of the MND patient. Previous research among stroke patients and their caregivers suggested that dependence can result in the experience of inequality in the relationship due to increased contributions from the caregiver compared to loss of benefits received, which intensifies the sense of caregiver burden (McPherson et al., 2011). Furthermore, Ybema et al. (2002) suggested that receiving support from others maintained relationship equity and reduced the sense of caregiver burden. Hence, formal support could reduce the dependence of the MND patient on their informal caregiver, thus maintaining relationship equity and preserving relationship identity and dynamics. However, analysis of the outcome of this suggestion is beyond the scope of the present review.

## LIMITATIONS OF THE STUDY

One limitation of our study is that, although systematic and thorough, it is possible that relevant studies may have been neglected due to variable indexing of qualitative research. Our synthesis included caregivers of patients with any type of and at any stage of MND, thus the findings were not specific to any stage or symptom of MND. This could be explored in future research. Female caregivers were overrepresented, but as more men are affected by MND, this may not be problematic (Goldstein et al., 2006). Demographical factors were not considered (e.g., disposable income, culture), and this may affect the ability of caregivers to relate to our findings. Furthermore, we included studies from any country, so that consideration of different healthcare systems was not accounted for.

## CONCLUSIONS

Our synthesis explored the experiences and challenges faced by informal caregivers. Although suggestions for service development have emerged from the existing literature, there is a lack of information regarding the implementation and outcome of these suggestions upon the experience of caregivers.

## DISCLOSURES

The authors hereby declare that they have no conflicts of interest to disclose.

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## SUPPLEMENTARY MATERIALS

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## REFERENCES

Akiyama, M., Kayama, M., Takamura, S., et al. (2006). A study of the burden of caring for patients with amyotrophic lateral sclerosis (MND) in Japan. *British Journal of Neuroscience Nursing*, *2*(1), 38–43.

Aoun, S. M., Connors, L., Priddis, L., et al. (2012). Motor neurone disease family carers' experiences of caring, palliative care and bereavement: An exploratory qualitative study. *Palliative Medicine*, *26*(6), 842–850.

Aoun, S., Bentley, B., Funk, L., et al. (2013). A 10-year literature review of family caregiving for motor neurone disease: Moving from caregiver burden studies to palliative care interventions. *Palliative Medicine*, *27*(5), 437–446.

Aria, L., Britten, N., Popay, J., et al. (2007). Identifying themes main or recurrent which can group concepts from studies together. *Evidence & Policy*, *3*(3), 361–383.

Atkins, L., Brown, R. G., Leigh, P. N., et al. (2010). Marital relationships in amyotrophic lateral sclerosis. *Amyotrophic Lateral Sclerosis*, *11*(4), 344–350.

Brown, J. B. (2003). User, carer and professional experiences of care in motor neurone disease. *Primary Health Care Research & Development*, *4*(3), 207–217.

Chio, A., Gauthier, A., Vignol, A., et al. (2006). Caregiver time use in ALS. *Neurology*, *67*(5), 902–904.

Cooke, A., Smith, D. & Booth, A. (2012). Beyond PICO: The SPIDER tool for qualitative evidence synthesis. *Qualitative Health Research*, *22*(10), 1435–1443.

Cote, L. & Turgeon, J. (2009). Appraising qualitative research articles in medicine and medical education. *Medical Teacher*, *27*(1), 71–75.

Crellin, N. E., Orrell, M., McDermott, O., et al. (2014). Self-efficacy and health-related quality of life in family carers of people with dementia: A systematic review. *Aging & Mental Health*, *18*(8), 954–969.

Eifert, E., Adams, R., Dudley, W., et al. (2015). Family caregiver identity: A literature review. *American Journal of Health Education*, *46*(6), 357–367.

Fossey, E., Harvey, C., McDermott, F., et al. (2002). Understanding and evaluating qualitative research. *The Australian and New Zealand Journal of Psychiatry*, *36*(6), 717–732.

Goldstein, L. N., Atkins, L., Landau, S., et al. (2006). Predictors of psychological distress in carers of people with amyotrophic lateral sclerosis: A longitudinal study. *Psychological Medicine*, *36*(6), 865–875.

Herz, H., McKinnon, P. & Butow, P. (2006). Proof of love and other themes: A qualitative exploration of the experience of caring for people with motor neurone disease. *Progress in Palliative Care*, *14*(5), 209–214.

Hewitt, M. (2007). *How to Search and Critically Evaluate Research Literature*. Nottingham, UK: The NIHR RDS for the East Midlands/Yorkshire and the Humber. Available from http://www.worcester.ac.uk/documents/6_Critically_evaluate_research_literature_2009v2.pdf.

Hyunjin, O. & Schepp, K. (2013). Wives becoming caregivers: The lived experiences of wives caring for amyotrophic lateral sclerosis patients. *Journal of Hospice & Palliative Nursing*, *15*(4), 244–250.

Malpass, A., Shaw, A., Sharp, D., et al. (2009). "Medication career" or "moral career"? The two sides of managing antidepressants: A meta-ethnography of patients' experience of antidepressants. *Social Science & Medicine*, *68*(1), 154–168.

McPherson, C., Wilson, K., Chyurlia, L., et al. (2011). The caregiving relationship and quality of life among partners of stroke survivors: A cross-sectional study. *Health & Quality of Life Outcomes*, *9*, 29. Available from https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3111340/.

Mitchell, J. & Borasio, G. (2007). Amyotrophic lateral sclerosis. *Lancet*, *369*(9578), 2031–2041.

MND Association (2015). *Motor Neurone Disease: A Guide for GPs and Primary Care Teams*. Northampton: MND Association. Available from https://www.mndassociation.org/wp-content/uploads/px016-motor-neurone-disease-a-guide-for-gps-and-primary-care-teams.pdf.

MND Association (2016). *Your Rights as a Carer*. Northampton. MND Association. Available from https://www.mndassociation.org/wp-content/uploads/3-your-rights-as-a-carer.pdf.

Mockford, C., Jenkinson, C. & Fitzpatrick, R. (2006). A review: Carers, MND and service provision. *Amyotrophic Lateral Sclerosis*, *7*(3), 132–141.

Moher, D., Liberati, A., Tetzlaff, J., et al. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *PLoS Medicine*, *6*(7), e1000097. Available from https://www.ncbi.nlm.nih.gov/pmc/articles/PMC2707599/.

Murphy, V., Felgoise, S., Walsh, S., et al. (2009). Problem solving skills predict quality of life and psychological morbidity in ALS caregivers. *Amyotrophic Lateral Sclerosis*, *10*(3), 147–153.

Nageshwaran, S. (2014). Motor Neurone Disease. *BMJ*, *349*, g4052.

NICE (2016). *Motor Neurone Disease: Assessment and Management*. London: NICE. Available from http://www.nice.org.uk/guidance/ng42/chapter/Recommendations.

O'Brien, M., Whitehead, B., Jack, B., et al. (2012). The need for support services for family carers of people with motor neurone disease: Views of current and former family caregivers a qualitative study. *Disability and Rehabilitation*, *4*(3), 247–256.

Oyebode, J., Smith, H. & Morrison, K. (2013). The personal experience of partners of individuals with motor neuron disease. *Amyotrophic Lateral Sclerosis & Frontotemporal Degeneration*, *14*(1), 39–43.

Peters, M., Jenkinson, C., Doll, H., et al. (2013). Carer quality of life and experiences of health services: A cross-sectional survey across three neurological conditions. *Health and Quality of Life Outcomes*, *11*(1), 103–111.

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

Rodgers, M., Sowden, A., Petticrew, M., et al. (2009). Testing methodological guidance on the conduct of narrative synthesis in systematic reviews. *Evaluation*, *15*(1), 47–71. Available from https://www.lancaster.ac.uk/shm/research/nssr/research/dissemination/publications/Rodgers_et_al.pdf.

Sakellariou, D, Boniface, G. & Brown, P. (2013). Experiences of living with motor neurone disease a review of qualitative research. *Disability and Rehabilitation*, *35*(21), 1765–1773.

Shaw, R., Booth, A., Sutton, J., et al. (2004). Finding qualitative research: An evaluation of search strategies. *BMC Medical Research Methodology*, *4*, 5. Available from https://www.ncbi.nlm.nih.gov/pmc/articles/PMC385230/.

Sorrell, J. (2014). Moving beyond caregiver burden: Identifying helpful interventions for family caregivers. *Journal of Psychosocial Nursing and Mental Health Services*, *52*(3), 15–18.

Soundy, A. & Condon, A. (2015). Patients' experiences of maintaining mental well-being and hope within motor neuron disease: A thematic synthesis. *Frontiers in Psychology*, *6*, 606. Available from https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4428059/.

Soundy, A., Roskell, C., Elder, T., et al. (2016). The psychological process of adaptation and hope in patients with multiple sclerosis: A thematic synthesis. *Open Journal of Therapy and Rehabilitation*, *4*(1), 22–47.

Thomas, J. & Harden, A. (2008). Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Medical Research Methodology*, *8*, 45. Available from https://www.ncbi.nlm.nih.gov/pmc/articles/PMC2478656/.

Tong, A., Sainsbury, P. & Craig, J. (2007). Consolidated Criteria for Reporting Qualitative Research (COREQ): A 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*, *19*(6), 349–357.

Tong, A., Flemming, K., McInnes, E., et al. (2012). Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. *BMC Medical Research Methodology*, *12*, 181. Available from https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3552766/.

Weisser, F., Bristowe, K. & Jackson, D. (2015). Experiences of burden, needs, rewards and resilience in family caregivers of people living with motor neurone disease/amyotrophic lateral sclerosis: A secondary thematic analysis of qualitative interviews. *Palliative Medicine*, *29*(8), 737–745.

Whitehead, B., O'Brien, M., Jack, B., et al. (2012). Experiences of dying, death and bereavement in motor neurone disease: A qualitative study. *Palliative Medicine*, *26*(4), 368–378.

Wu, Y., Aylward, B., Roberts, M., et al. (2012). Searching the scientific literature: Implications for quantitative and qualitative reviews. *Clinical Psychology Review*, *32*(6), 553–557.

Ybema, J., Kuijer, R., Hagedoorn, M., et al. (2002). Caregiver burnout amongst intimate partners of patients with a severe illness: An equity perspective. *Personal Relationships*, *9*(1), 73–88.