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**The experience of informal caregivers of patients with Motor Neurone Disease:
A thematic synthesis**

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Abstract

Objectives: 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 heterogenous 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, this synthesis aims to identify caregivers' experiences and to suggest factors that contribute to these experiences in order to fulfil the required research need.

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

Results: Ten studies met the inclusion criteria, with 148 (50 male) current or previous informal caregivers of pWMND identified. Critical appraisal demonstrated study design and reflexivity were under-reported. The synthesis derived three themes: 1) loss of control, 2) inability to choose, 3) isolation.

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

Key words 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 used inconsistently; the UK uses MND as the umbrella term whereas other countries use ALS (Mitchell and Borasio, 2007; Aoun *et al.*, 2013). Therefore, they will be used synonymously in this study. Around 4,500 people in the UK are living with MND at any time and peak onset is between 50-70 years (MND Association, 2015). 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 and Borasio, 2007; Nageshwaran, 2014). Currently there is no cure, and 50% of patients do not live beyond three years from onset of symptoms (Mitchell and Borasio, 2007).

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 on the patients' disease progression with Chio *et al.*, (2006) reporting for patients with a mild disability (ALS-FRS score >30) a mean of 5hours/day and for patients with severe impairments (ALS-FRS score ≤10) a mean of 15hours/day. Caregivers' quality of life (QoL) is significantly negatively impacted by undertaking the 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 upon caregivers' 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 1994-2004; the main findings discussed caregivers experiencing ill health when unsupported and the positive impact of maintaining social support and activities on 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 factors such as diagnosis process and access to information and services which contribute to burden and negatively impact QoL, there is a need to develop literature focusing on improving caregivers' experience. As further literature has been published since 2011, this study aims to update existing reviews.

Researchers have called for further consideration of caregivers' experiences and in particular 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 synthesised (Malpass *et al.*, 2009) it was considered important to utilise 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 this study aims to focus solely on a qualitative approach, it has the potential to offer alternative interpretation and results as the caring experience can be considered as heterogeneous and subject to personal or unique interpretations (Crellin *et al.*, 2014). Thus, this study aims to provide a thematic synthesis of existing qualitative literature regarding experiences of informal caregivers of pwMND. The study aims to encompass the spectrum of experiences through not focusing on a specific stage of the disease progression.

Additionally, the study aims to highlight areas for service development to reduce caregiver burden in the future through discussing positive caregiver experiences and suggestions made in existing literature.

Methods

The methods section has been reported in accordance with the enhancing transparency in reporting the synthesis of qualitative research guidelines (ENTREQ) (Tong *et al.*, 2012a).

Search strategy

Electronic databases were searched from inception until September 2015; Allied and Complementary Medicine Database (AMED), Medline, SPORTDiscus, Cumulative Index to Nursing and Allied Health Literature (CINAHL), and PubMed. Multiple databases were utilised to maximise search yield (Wu *et al.*, 2012). The selected databases were chosen as they focus on 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 and Condon, 2015).

Combinations of the following key terms were utilised: “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 utilised as they retrieved large volumes of irrelevant studies during scoping search. Additional search strategies included citation chasing, reviewing reference lists, and contacting researchers (n=1).

Eligibility criteria

Included studies needed to fulfil the eligibility criteria which utilised the SPIDER (sample, phenomenon of interest, design, evaluation, research type) search tool (Cooke *et al.*, 2012);

- *Sample*: Informal primary caregivers were defined as individuals who previously or currently provide the majority of support and assistance to a pwMND at any disease stage, without financial reward.
- *Phenomenon of interest*: Studies reporting informal caregivers' first-hand experience with MND were included.
- *Design*: Any type of qualitative design was considered. Excluded designs: were quantitative research, mixed-method research not containing clear qualitative data, grey literature; unpublished or protocol studies, thesis, 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

This review used an adapted 13-item version of Tong *et al.*, (2007) Consolidated Criteria for Reporting Qualitative Studies (COREQ), which has three domains:

- (i) research team and reflexivity which increases transparency of personal characteristics of the research team as well as their relationship with participants which could influence bias within data collection and interpretation of results,

(ii) study design which appraises participant retention and the data collection process,

(iii) data analysis and reporting which appraises the process of coding and derivation of themes.

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

Studies were independently appraised scoring one for reporting a checklist item and zero if unclear or unreported (Tong *et al.*, 2007). A total score of 13 was possible and, based on Soundy *et al.*, (2016) previous research, a score of 4 was identified as a threshold where further consideration of the study was made. Any studies scoring less than 4 were discussed to identify if there was a methodological weakness which 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 this synthesis (Thomas and Harden, 2008; Aria *et al.*, 2007; Rodgers *et al.*, 2009): Stage one 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 two involved idea webbing to visualise and connect concepts between studies (Rodgers *et al.*, 2009). Stage three involved translation; interpreting and converting similar concepts between studies (Aria *et al.*, 2007). Stage four involved synthesis of

translations to combine the analysed themes and form a new interpretation of the studies involved. See supplementary file for the audit trail.

Results

Systematic search

The full search process is demonstrated via Prisma (Moher *et al.*, 2009) flow diagram (Figure 1). Ten articles (Brown, 2003; Akiyama *et al.*, 2006; Herz *et al.*, 2006; Ray and Street, 2007; Aoun *et al.*, 2012; O'Brien *et al.*, 2012; Whitehead *et al.*, 2012; Hyunjin and Schepp, 2013; Oyebode *et al.*, 2013; Weisser *et al.*, 2015) met the inclusion criteria and were analysed within the thematic synthesis. Characteristics of included participants are presented in table 1.

INSERT FIGURE 1 HERE

INSERT TABLE 1 HERE

Critical Appraisal

The results of the 13-item COREQ ranged from four (Akiyama *et al.*, 2006; Whitehead *et al.*, 2012) to eight (Ray and Street, 2007) with a mean total score of six. Hence, no studies were excluded nor discussed for exclusion. For full COREQ results see the supplementary file.

Although Akiyama *et al.*, (2006) and Whitehead *et al.*, (2012) scored the minimum score of four, they were the only included studies to score 0/5 within domain one of the COREQ. This impacts credibility of these studies' results as there is a lack of transparency of the researchers' credentials and relationship with participants thus there is an inability to distinguish the degree to which personal bias may be influencing the 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 dropouts resulting in a possibility of selective reporting (Tong *et al.*, 2007; Cote and 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 the study design and the researchers' rationale for their interpretation and analysis of the data (Fossey *et al.*, 2002). Hence, due to lack of transparency limiting the ability to interpret results, novel data from Akiyama *et al.*, (2006) and Whitehead *et al.*, (2012) was interpreted with caution and with consideration of findings from the other included studies.

Thematic synthesis

The thematic synthesis highlighted that, although the caregivers experience is non-homologous, shared key themes exist. These themes are explored below and are grouped as follows: 1) factors contributing to the experience of loss of control, 2) factors contributing to the sense of choice, 3) factors contributing to the experience of isolation. Studies supporting each theme are demonstrated in table 2.

INSERT TABLE 2 HERE

Loss of control

Progressive nature of MND

The progression of the cared for patients' MND 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 alongside the progression of the patients' MND (Aoun *et al.*, 2012; Brown, 2003; O'Brien *et al.*, 2012; Ray and Street, 2007; Whitehead *et al.*, 2012). Caregivers were aware symptoms would progress but were unable to highlight how symptoms would change and how this would impact their caregiving role (O'Brien *et al.*, 2012). Furthermore, caregivers were uncertain of the rate of the patients' MND progression (Aoun *et al.*, 2012; O'Brien *et al.*, 2012; Oyebode *et al.*, 2013; Ray and Street, 2007; Whitehead *et al.*, 2010). 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 and Street, 2007; Whitehead *et al.*, 2012). One caregiver epitomises this through the quote; "This is so undetermined you just don't know it from 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 and Street, 2007). Furthermore, this inability to prepare for continual progression resulted in a sense of progressive overwhelming responsibility and a struggle to stay in control (Herz *et al.*, 2007). For some, uncertainty resulted in fear and anxiety (Aoun *et al.*, 2012; O'Brien *et al.*, 2012), conversely other caregivers could embrace uncertainty and 'live in the moment' as a coping mechanism (Weisser *et al.*, 2015; Oyebode *et al.*, 2013).

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

Loss of control over daily activities

The idea that caregivers had 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 and Schepp, 2013; O'Brien *et al.*, 2012; Oyebode *et al.*, 2013; Ray and 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 and Schepp, 2013; O'Brien *et al.*, 2012; Oyebode *et al.*, 2013; Ray and 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 the sense of being solely responsible for the pwMND daily care and needs without the support of others to share the role and allow previous activities and social opportunities to be continued (Akiyama *et al.*, 2006; Aoun *et al.*, 2012; Herz *et al.*, 2006; Hyunjin and Schepp, 2013; O'Brien *et al.*, 2012; Oyebode *et al.*, 2013; Ray and Street, 2007). Hence, the sense of captivity and subsequent loss of control could differ between caregivers depending on the level of support they receive (Oyebode *et al.*, 2013; Weisser *et al.*, 2015). However, formal support may have a peak intensity which prevents the feeling of loss of control with intensity above this threshold contributing to the caregivers' 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 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 daily activities arose from the inability to complete activities previously shared with the pwMND due to their functional decline (Ray and 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 and Schepp, 2013; O'Brien *et al.*, 2012). Another was the sense of living for others dehumanising their own existence (Akiyama *et al.*,

2006; Herz *et al.*, 2006; O'Brien *et al.*, 2012; Oyebode *et al.*, 2013; Ray and Street, 2007). For some, this resulted in a change in the individuals' self-perception and a sense of being a different version of themselves (Herz *et al.*, 2006; Ray and Street, 2007) "You're not yourself, you're certainly not yourself...It's, you feel more of a carer than anything" (Ray and Street, 2007).

Choice

Choice regarding uptake of the caregiver role

Caregivers experienced a sense of obligation resulting in an inability to choose whether 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 desire (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 pwMND from further suffering, there is a sense of caregivers concealing difficulties and struggles from the pwMND and hence their own needs going unnoticed and neglected (Oyebode *et al.*, 2013). However, for others the obligation was engrained in the expectation to care for loved ones (Herz *et al.*, 2006) and demonstrate commitment to marriage (Oyebode *et al.*, 2013).

Access to alternatives

Caregivers experienced a lack of alternative options to caring fulltime for the 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) whereas others suggested insufficient time and captivity of the role did not allow for the consideration of the need for support (Aoun *et al.*, 2012).

The synthesis highlighted the financial implications of accessing support for example equipment or paid for caregivers being a luxury not all could access (Herz *et al.*, 2006; Oyeboode *et al.*, 2013; Whitehead *et al.*, 2012). Furthermore, a lack of haste in processing applications for financial support prevented eligible pwMND receiving funding due to passing away prior to the reviewers coming to a decision (Whitehead *et al.*, 2012). Although this may relate to the difficulty in predicting progression of MND, it 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; Oyeboode *et al.*, 2013; Whitehead *et al.*, 2012). This resulted in caregivers not being confident that the needs of the pwMND would be met and subsequent unwillingness to release the caregiving role to unspecialised 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

An idea which emerged from the synthesis was that communication between professionals and caregivers contributed to the caregivers' sense of being unsupported and isolated (Aoun *et al.*, 2012; O'Brien *et al.*, 2012). One factor highlighted was the idea of high volumes of involved healthcare professions without clarity on each of their roles or a key member to contact (O'Brien *et al.*, 2012). Hence it could be suggested that potential sources of support are not utilised due to poor clarity of communication which leads to caregivers being overwhelmed. Furthermore, for some, discussion with professionals lacked depth and personalisation with failure to attend to emotional aspects of caregiving for a pwMND (Brown, 2003) resulting in 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 which attend to the caregivers' needs and a requirement for professionals to shift their focus when communicating with caregivers. However, others used counselling services which allowed cathartic release (Herz *et al.*, 2006; O'Brien *et al.*, 2012) and for some caregivers, depth and effectiveness of communication was accessible however this appears to be profession dependent and may highlight a greater need for access to counselling services to meet caregivers' needs.

Communication between family and friends

Changes in communication within pre-existing relationships with family and friends emerged in several studies (Herz *et al.*, 2006; Hyunjin and Schepp, 2013; O'Brien *et al.*, 2012; Oyebode *et al.*, 2013; Weisser *et al.*, 2015). A sense of inability to share was noted to avoid burdening others (Herz *et al.*, 2006; Hyunjin and 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 non-involved others due to their lack of understanding of the caregivers’ situation and burden (Hyunjin and Schepp, 2013) suggesting a lack of empathy and an alienating experience. However, other caregivers appreciated a sense of normality when communicating with friends (Akiyama *et al.*, 2006; Ray and Street, 2007) indicating that communication within previous relationships was not impacted by the caregiving role. However, it is plausible that this sense of ‘normality’ is a denial 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 topics which can be openly discussed when communicating with others.

Caregivers also experienced a decreased frequency of verbal communication with the pwMND, another pre-existing relationship (Hyunjin and Schepp, 2013; Weisser *et al.*, 2015). One contributing factor which emerged was that MND symptoms were attributed to decreased quality of communication due to a need to simplify language (Oyebode *et al.*, 2013). For some the frustration of the caregiving role led to tension and avoiding communication (Hyunjin and Schepp, 2013; O’ Brien *et al.*, 2012). Alternatively, others noted an increase in non-verbal communication through activities involved with the caregiving role (Weisser *et al.*, 2015), demonstrating a potential shift away from verbal language and hence altering the method of communication. Additionally, some caregivers found the ability to share more readily with the pwMND, increasing verbal communication (Weisser *et al.*, 2015), however,

the motive is unclear, and could be due to differing factors such as the terminal prognosis rather than directly due to the caregiving role.

Changes to relationship dynamics

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

Loss of intimacy

A 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 and Schepp, 2013; Oyebode *et al.*, 2013; Ray and Street, 2007). Aoun *et al.*, (2012) and Oyebode *et al.*, (2013) discussed physical barriers due to MND symptoms which limited intimacy and resulted in the experience of isolation due to absence of physical contact and sense of connection with their spouse. Additionally, for some caregivers the requirement to attend to the pwMND personal care limited the desire for physical intimacy (Hyunjin and Schepp, 2013; Ray and Street, 2007). Caregivers' decreased desire for physical intimacy resulted in

increased tension between spousal couples subsequently leading to a sense of isolation (Hyunjin and Schepp, 2013). However, as well as the direct relationship between attending to the pwMND care needs and loss of desire for intimacy, caregivers highlighted how attending to the pwMND personal care altered the relationship identity due to the loss of reciprocity in the relationship and it was the change in relationship identity which limited intimacy (Aoun *et al.*, 2012; Ray and Street, 2007). This demonstrates that relationship identity can be the cause of loss of intimacy and a result of loss of intimacy.

Opportunities for interaction

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 and Street, 2007; Weisser *et al.*, 2015). Decreased opportunity for interaction related to role captivity of caregiving and subsequent insufficient time. One caregiver reported "I used to go weeks and sometimes months before I saw anybody." (Brown, 2003) highlighting physical isolation from others limiting interaction. As well as limited time, fatigue associated with the caregiving role was associated with decreased interaction (Ray and Street, 2007). However, another caregiver highlighted the sense of isolation and loneliness despite maintaining work alongside the caregiving role (O'Brien *et al.*, 2012) suggesting physical isolation is not the only factor contributing to limitations in 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 both during the caregiving and bereavement phase (Ray and Street, 2007). Conversely Aoun *et al.*, (2012) suggest

that isolation was only experienced in the bereavement phase as caregivers were supported in the caregiving phase “You go from having a whole army of people 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 impact the sense of isolation and loss of interaction. Hence, opportunity for interaction maybe caregiver specific and level of support may impact the caregivers’ experience of isolation.

Discussion

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

A key idea highlighted across themes was the sense of being solely responsible for the pwMND without help or support from others. This responsibility can stem from the sense of obligation to fulfil the caregiver role either to satisfy the patient's wishes, demonstrate love and commitment, or due to the absence of alternatives or support. Access to support differed between participants included in the thematic synthesis with those individuals who were financially able in an advantageous position.

However, as the synthesis included studies from any country, financial requirements may differ between countries and the support systems available (Sakellariou *et al.*, 2013). Also highlighted was the lack of communication about available support between healthcare professionals and caregivers. Reasons for lack of communication included insufficient time, inadequate knowledge of MND, involved healthcare professionals lack of awareness of caregivers' emotional difficulties, or having a multitude of involved healthcare professionals with lack of clarity over their roles. Highlighted was the need for healthcare professionals to have knowledge of, and ability to communicate, the likely future for the pwMND and how this subsequently impacts the caregiver. One idea which emerged was the need for a key worker to address caregivers' questions, provide practical and emotional support and highlight or signpost to relevant services to increase access to support.

However, this idea is not used in any of the included studies and thus the outcome cannot be determined within the scope of this thematic synthesis. However, this idea has been reflected in the UK through the NICE guidelines' recommendation to provide of a single point of contact within the MND-specific multidisciplinary team (NICE, 2016). This therefore supports the ideas emerging in the synthesis as they are recommended for implementation in clinical practice.

Caregivers' sense of being solely responsible for the pwMND resulted in insufficient time to maintain social interactions and previously completed activities due to being captive within the caregiver role. This was associated with a sense of burden, isolation, and living for others. This is supported by previous research regarding informal family caregivers for non-specific chronic conditions which highlights how insufficient time for activities which previously contributed to personal identity results in a sense of living for others and loss of self (Eifert *et al.*, 2015). Highlighted in this thematic synthesis was the need for support or respite to allow maintenance of activities however frequency, type, or feasibility of support was not discussed and hence it is beyond the scope of the synthesis to make conclusive recommendations. Despite this, the synthesis highlights the need for formal caregivers to have sufficient knowledge and skills to care for the pwMND to allow informal caregivers to readily accept support, as insufficient ability is a key identified reason for underutilisation of support. This is supported by Aoun *et al.*, (2013) review which suggested 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 avoiding burdening others, and lack of understanding or empathy from non-involved others limiting communication and interaction within previous relationships. Hence time for

social interactions through formal support alone would not address these issues. Further research is therefore indicated to understand the complexities in caregivers' social interaction to make conclusive recommendations to address the issues. Caregivers experienced a sense of isolation with one contributing factor being a shift in spousal relationship dynamics and identity due to the pwMND dependence. Previous research amongst stroke patients and their caregivers suggested that dependence can result in the experience of inequality in the relationship due to increased contribution compared to benefits received and is associated with caregivers' perception of burden (McPherson *et al.*, 2011). Furthermore, Ybema *et al.*, (2002) suggested receiving support from others maintained relationship equity and reduced caregivers' sense of burden. Hence, formal support could reduce the pwMNDs' dependence on the informal caregiver maintaining relationship equity and subsequently preserving relationship identity and dynamics. However, analysis of the outcome of this suggestion is beyond the scope of this review.

Limitations

Study limitations exist as, although systematic and thorough, it is possible that relevant studies may have been neglected due to variable indexing of qualitative research. The 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 symptoms of MND. Subsequently this could be explored in future research. Female caregivers were over-represented in this synthesis, although, as more men are affected by MND, this may not be problematic (Goldstein *et al.*, 2006). Demographical factors were not considered for example disposable income or culture; this may affect the ability of

caregivers to relate to the findings. Furthermore, the synthesis included studies from any country and thus consideration of different healthcare systems was not made.

Conclusion

This synthesis explores the experiences and challenges faced by non-formal caregivers. Although suggestions for service development have emerged from existing literature, there is a lack of information regarding the implementation and outcome of these suggestions upon the caregivers' experience.

Conflict of Interest

The authors declare there are no conflicts of interest.

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References

- Akiyama.M., Kayama.M., Takamura.S., Ohbu.S., Fukuhara.S. (2006). 'A Study of the Burden of Caring for Patients with Amyotrophic Lateral Sclerosis (MND) in Japan.' *British Journal of Neuroscience Nursing*. 2(1):pp.38-43
- Aoun.S., Bentley.B., Funk.L., Toye.C., Grande.G., Stajduhar.K. (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):pp.437-446.
- Aoun.S., Connors.L., Priddis.L., Breen.L., Colyer.S. (2012). 'Motor Neurone Disease Family Carers' Experiences of Caring, Palliative Care and Bereavement: An Exploratory Qualitative Study.' *Palliative Medicine*.26(6):pp.842-850
- Aria.L., Britten.N., Popay.J., Roberts.H., Petticrew.M., Rodger.M., Sowden.A. (2007). 'Identify Themes Main or Recurrent which Can Group Concepts from Studies Together.' *Evidence & Policy*.3(3):pp.361-383
- Atkins.L., Brown.R., Leigh.N., Goldstein.L. (2010). 'Marital Relationships in Amyotrophic Lateral Sclerosis.' *Amyotrophic Lateral Sclerosis*. 11(4):pp.344-350
- Brown.J.B. (2003). 'User, Carer and Professional Experiences of Care in Motor Neurone Disease.' *Primary Health Care Research and Development*.4:pp.207-217
- Chio.A., Gauthier.A., Vignol.A., Calvo.A., Ghiglione.P., Cavallo.E., Terreni.A., Mtani.R. (2006). 'Caregiver Time Use in ALS'. *Neurology*.67:pp.902-904
- Cooke.A., Smith.D. and Booth.A. (2012). 'Beyond PICO: the SPIDER tool for Qualitative Evidence Synthesis.' *Qualitative Health Research*.22(10).pp.1435-1443
- Cote.L and Turgeon.J (2009). "Appraising qualitative research articles in medicine and medical education." *Medical Teacher*. 27(1):pp.71-75
- Crellin.N.E., Orrell.M., McDermott.O. Charlesworth.G. (2014). 'Self-efficacy and Health-related Quality of Life in Family Carers of People with Dementia: A Systematic Review.' *Aging & Mental Health*.18(8).pp.954-969
- Eifert.E., Adams,R., Dudley.W., Perko.M. (2015). 'Family Caregiver Identity: A Literature Review.' *American Journal of Health Education*.46(6):pp.357-367
- Fossey.E, Harvey.C, McDermott.F, and Davidson.L (2002). "Understanding and evaluating qualitative research." *Australian and New Zealand Journal of Psychiatry*. 36:pp.717-732
- Goldstein.L., Atkins.L., Landau.S., Brown.R., Leigh.P. (2006). 'Predictors of Psychological Distress in Carers of People with Amyotrophic Lateral Sclerosis: A Longitudinal Study.' *Psychological Medicine*.2006(06):pp.865-875

Herz.H., McKinnon.P. and 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):pp.209-214

Hewitt.M. (2007). 'How to Search and Critically Evaluate Research Literature. The NIHR RDS for the East Midlands/ Yorkshire & Humber.' Available at: http://www.worcester.ac.uk/documents/6_Critically_evaluate_research_literature_2009v2.pdf (Accessed 21 October 2015)

Hyunjin.O. and Schepp.K. (2013). 'Wives Becoming Caregivers: The Lived Experiences of Wives Caring for Amyotrophic Lateral Sclerosis Patients' *Journal of Hospice & Palliative Nursing*.15(4):pp.244-250

Malpass.A., Shaw.A., Sharp.D., Walter.F., Feder.G., Ridd.M., Kessler.D.(2009). 'Medication Career or Moral Career? The Two sides of Managing Antidepressants: A Met-ethnography of Patients' Experience of Antidepressants.' *Social Science & Medicine* 68(1):pp.154-168

McPherson.C., Wilson.K., Chyurlia.L., Leclerc.C. (2011). 'The Caregiving Relationship and Quality of Life Amongst Partners of Stroke Survivors: A cross-sectional Study.' *Health & Quality of life outcomes*.9(29):n.p.

Mitchell.J. and Borasio.G. (2007). 'Amyotrophic Lateral Sclerosis.' *The Lancet*.369(9578):pp.2031-2041

MND Association. (2015). '*Motor Neurone Disease: A Guide for GPs and Primary Care Teams*.' Northampton. MND Association.

MND Association. (2016). '*Your Rights as a Carer*.' Northampton. MND Association.

Mockford.C., Jenkinson.C and Fitzpatrick.R. (2006). 'A Review: Carers, MND and Service Provision.' *Amyotrophic Lateral Sclerosis*.2006(7):pp.132-141

Moher.D., Liberati.A., Tetzlaff.J., Altman.D., The PRISMA Group. (2009). 'Preferred Reporting Items for Systematic Reviews and MetaAnalyses: The PRISMA Statement.' *PLoS Med* 6(7).

Murphy.V., Felgoise.S., Walsh.S., Simmons.Z. (2009). 'Problem Solving Skills Predict Quality of Life and Psychological Morbidity in ALS Caregivers.' *Amyotrophic Lateral Sclerosis*.10(3):pp.147-153

Nageshwaran.S. (2014). 'Motor Neurone Disease'. *BMJ*. 2014;349:g4052

NICE. (2016). '*Motor Neurone Disease: Assessment and Management*'. Available at: <http://www.nice.org.uk/guidance/ng42/chapter/Recommendations> (Accessed: 3 March 2016).

- O'Brien.M., Whitehead.B., Jack.B., Mitchell.J.,(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 & Rehabilitation*.34(3):pp.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:pp.39-43
- Peters.M., Jenkinson.C., Doll.H., Playford.D., Fitzpatrick.R. (2013). 'Carer Quality of Life and Experiences of Health Services: A Cross-sectional Survey Across three Neurological Conditions.' *Health & Quality of Life Outcomes*.11(1):pp.103-111
- Ray.R. and Street.A. (2005). 'Who's There and Who Cares: Age as an Indicator of Social Support Networks for Caregivers Among People Living with Motor Neurone Disease.' *Health & Social Care in the Community*.13(6):pp.542-552
- Ray.R. and Street.A. (2007). 'Non-finite Loss and Emotional Labour: Family Caregivers' Experiences of Living with Motor Neurone Disease.' *Journal of Clinical Nursing*.16(3):pp.35-43
- Rodgers.M., Sowden.A., Petticrew.M., Arai.L., Roberts.H., Britten.N., Popay.J. (2009). 'Testing Methodological Guidance on the Conduct of Narrative Synthesis in Systematic Reviews.' *SAGE Publications*. 15(1):pp.47-71
- Sakellariou.D, Boniface.G, Brown.P. (2013). 'Experiences of living with motor neurone disease a review of qualitative research.' *Disability and Rehabilitation*. 35(21):pp.1765-1773.
- Shaw.R., Booth.A., Sutton.J., Miller.T., Smith.J., Young.B., Joones.D., Dixon-Woods.M. (2004). 'Finding Qualitative Research: an Evaluation of Search Strategies'. *BMC Medical Research Methodology*.4(5):n.p.
- Sorrell.J. (2014). 'Moving Beyond Caregiver Burden: Identifying Helpful Interventions for Family Caregivers.' *Journal of Psychosocial Nursing & Mental Health Services*.52(3):pp.15-18
- Soundy.A and Condon.A. (2015). 'Patients Experiences of Maintaining Mental Well-being and Hope within Motor Neuron Disease: A Thematic Synthesis.' *Frontiers in Psychology*.DOI=10.3389/fpsyg.2015.00606
- Soundy.A., Roskell.C., Elder.T., Collett.J., Dawes.H. (2016). 'The Psychological Process of Adaptation and Hope in Patients with Multiple Sclerosis: A Thematic Synthesis' *Open Journal of Therapy and Rehabilitation*.4:pp.22-47
- Thomas.J and Harden.A. (2008). 'Methods for the Thematic Synthesis of Qualitative Research in Systematic Reviews.' *BMC Medical Research Methodology*.8(45):n.p

Tong.A., Chapman.J., Wong.G., Kanellis.J., McCarthy.G., Craig.J. (2012). 'The Motivations and Experiences of Living Kidney Donors: A Thematic Synthesis.' *American Journal of Kidney Diseases*. 60(1):pp.15-26

Tong.A., Sainsbury.P. and 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):pp.349-357

Tong.A., Flemming.K., McInnes.E., Oliver.S., Craig.J. (2012a). 'Enhancing Transparency in Reporting the Synthesis of Qualitative Research: ENTREQ.' *BMC: Medical Research Methodology*.12:pp.181.

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):pp.737-745

Whitehead.B., O'Brien.M., Jack.B., Mitchell.D., (2012). 'Experiences of Dying, Death and Bereavement in Motor Neurone Disease: A Qualitative Study.' *Palliative Medicine*.26(4):pp.368-378

Wu.Y., Aylward.B., Roberts.M., Evans.S. (2012). 'Searching the Scientific Literature; Implications for Quantitative and Qualitative Reviews.' *Clinical Psychology Review*.32(6):pp.553-557

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

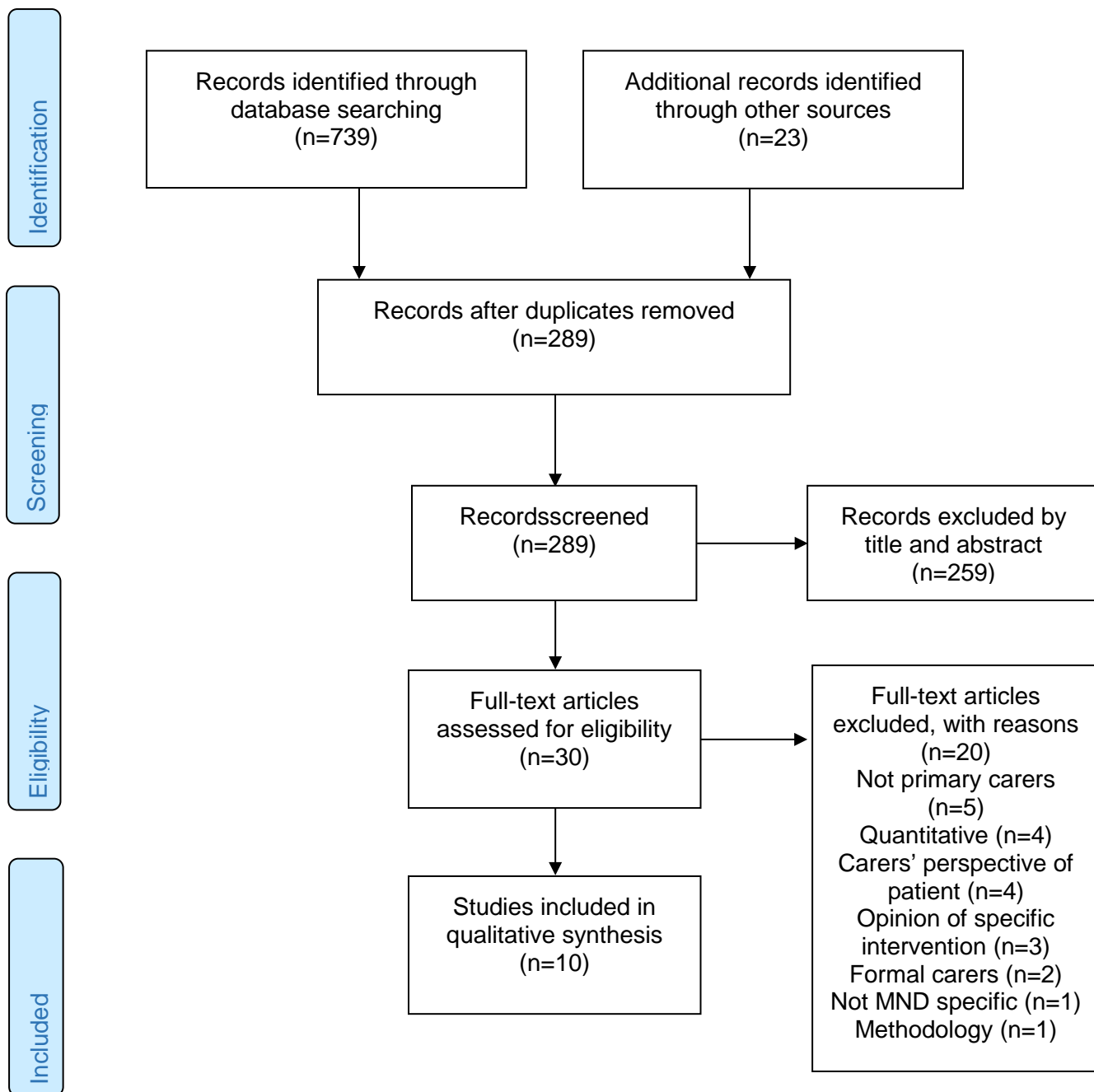


Figure 1. PRISMA flow diagram.

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

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

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