Exploring the Experiences of Informal Carers of People Living with Motor Neurone Disease (MND).

A thesis submitted in partial fulfilment of the requirements for the Doctorate in Clinical Psychology.

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Declaration

I declare that this thesis has been submitted for the award of Doctor in Clinical Psychology at The University of Sheffield. It has not been nor will be submitted to any other institution or university for the award of any other qualifications. I confirm this is my original work and referencing has been used for sources of information.

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Lay Summary

Literature Review

Caring for a person living with Motor Neurone Disease (plwMND) presents challenges for informal carers, who are often family members or close relatives. Carers have shared their experiences of caregiving however there has been little investigation into the recent literature. The current review focused on research published after 2011 that explores the experiences and needs of informal carers, informed by a previous review. Three databases were searched and 23 qualitative studies were included. Studies were analysed using a qualitative approach and produced four themes: "the caregiving spectrum", "the unique needs of informal carers", "end-of-life care experiences", and "experiences of healthcare professionals and dealing with services". Findings suggested most informal carers experienced the burden of caregiving, combined with several unmet needs and a lack of faith in services and healthcare professionals (HCPs). Peer and formal counselling support, alongside training for carers and HCPs, were identified as necessary.

Empirical Project

Informal carers of plwMND experience psychological distress and challenges due to the burden of caregiving and face a loss of identity. However, few seek help for their difficulties, or experience barriers, highlighting an under-researched area. This study used a qualitative design to investigate help-seeking behaviours of informal carers, aiming to understand what facilitates help-seeking and what acts as a barrier. Twelve carers engaged in semi-structured interviews and transcripts were analysed to produce four themes: "physical, practical, mental and emotional impact on carers", "dominance and prioritisation of the needs of plwMND", "where, when and how carers *do* seek support", and "lack of knowledge and understanding from HCPs". Findings highlighted informal carers prioritise the plwMND's

needs over their own, resulting in reduced help-seeking. Previous experiences with services can encourage or prevent carers from seeking help. Carers emphasised the importance of services "making the first move" to promote engagement.

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Firstly, I would like to express my gratitude to my research supervisors, Dr Charlotte Wright and Dr Emily Mayberry. Your advice, reliability, compassion and support throughout this process has been invaluable and I want to thank you for continuing to believe in me and my abilities even when I didn't believe in myself. Learning from you, your skills and qualities has shaped who I want to be as a Clinical Psychologist.

Secondly, I want to thank the informal carers who volunteered to take part in my study, without whom this project would not have been possible. It was not an easy topic to speak about so thank you for your openness, engagement and honesty. I hope I have been able to honour your voices and I hope this research will do you and your experiences justice.

Thank you to my family; my parents, grandparents, siblings, aunties, uncles, and my husband's family for your constant emotional support and reassurance. You have comforted me and guided me throughout my Clinical Psychology journey and I cannot express in words how much I appreciate you. A special thank you to my fellow Trainee, Faiza, in whom I've found a lifelong friend and my 'doctorate bestie'. I've valued your friendship, care and humour and cannot wait to see you cross the finish line!

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Section One: Systematic Review

A Thematic Synthesis of the Recent Evidence Base Exploring Informal Carers' Experiences

Abstract

Objectives

The current systematic review aimed to synthesise qualitative research that explored the caregiving experiences of informal carers of people living with Motor Neurone Disease (plwMND). The review focused on literature published between 2011 and 2024, following on from a previous review. The following research question was applied: "What does the recent evidence base exploring the experiences of informal carers of plwMND highlight in terms of needs, experiences and barriers?"

Methods

Systematic searches were conducted across three databases (PsycInfo, Scopus and MEDLINE). Search terms were developed using the PICo search strategy. Twenty-three studies were included and the CASP appraisal tool was used for quality appraisal. Thematic synthesis was utilised to analyse the studies.

Results

Analysis identified four superordinate themes and nine subthemes: "the caregiving spectrum", "the unique needs of informal carers", "end-of-life care experiences", and "experiences of healthcare professionals and dealing with services".

Conclusions

Participants reported that caregiving was challenging and burdensome, however some shared that positive aspects can mitigate burden. Carers voiced many unmet needs such as lack of training or information about MND, as well as a lack of faith in services and healthcare professionals (HCPs).

Practitioner Points

- The findings highlight the need for training for informal carers in order to feel confident when caregiving, and for HCPs in order to develop concrete awareness and knowledge of MND.

- Peer and formal counselling support were identified as necessary.

- Future research should consider a 'shift' from identifying the needs and experiences of informal carers to working with HCPs and services to develop and implement tailored support for carers.

Keywords

Informal carers, experiences, Motor Neurone Disease, thematic synthesis, qualitative, systematic review.

Introduction

Motor neurone disease (MND), or amyotrophic lateral sclerosis (ALS), is a rapidly-progressing neurodegenerative disease that causes muscle wasting, difficulties with breathing and swallowing, and in some, behavioural and cognitive impairments (Andersen et al., 2012; Kiernan et al., 2011). There is limited treatment for MND and no known cure or cause (Mockford et al., 2006). Prognosis following a diagnosis of MND can vary; most people living with MND (plwMND) are given an average life expectancy of 2 – 4 years (Andersen et al., 2012). The impact of MND on plwMND has been well documented in the literature due to the significant changes and losses in physical and mental state experienced by plwMND. The progression of the disease is rapid and changes in the plwMND are often seen over days and weeks rather than months and years (Aoun et al., 2011). Research has highlighted the psychological effects that plwMND experience including depression (Clarke et al., 2005; Goldstein et al., 1998; Hogg et al., 1994), anxiety around diagnosis and as the disease progresses (O'Brien et al., 2011; Pavey et al., 2013), feelings of hopelessness (Averill et al., 2007) and demoralisation (Clarke et al., 2005).

When it comes to care for plwMND, healthcare professionals (HCPs) provide a level of support and input. However, the primary caregiver for plwMND is usually a family member or close relative (Mockford et al., 2006), often referred to as 'informal carers' due to the nature of their caregiving role which is unpaid, within the home of the plwMND, and moulded around the needs of the plwMND. More recently, research has provided insight into the experiences of informal carers, how they provide care, and their experiences of caregiving, which includes challenges they face and any positive aspects of caregiving. In most cases, informal carers describe the role as 'unrelenting' and a cause of distress, burden and reduced quality of life (Kaub-Wittemer et al., 2003; Trail et al., 2003).

Research has emphasised the demands placed on informal carers of plwMND; Krivickas et al. (1997) found informal carers, on average, spend up to 11 hours per day caring for their loved one. Caregiving activities consume the majority of informal carers' time, which can range from physical care to housekeeping (Chio et al., 2006). It is no wonder that the experience of informal carers is often referred to as 'caregiver burden' in the literature (de Wit et al., 2018) due to the nature of the MND caregiving role. Research also acknowledges the psychological impact of caregiving on informal carers; as the disease progresses and care demands increase, informal carers may experience depression, emotional distress and a reduced quality of life (Gauthier et al., 2007; Goldstein et al., 2006). The literature has demonstrated the impact of changes in roles and identity for informal carers of plwMND; Aoun et al. (2012) described the experiences of spousal informal carers who felt a shift in their role from spouse to caregiver.

The emotional, social and professional support needs of informal carers are unique and change over time as the disease progresses and care demands increase (Galvin et al., 2018). O'Brien et al. (2011) emphasised a gap in knowledge remains about what the specific needs and support needs of informal MND carers are and explored this with 28 current and bereaved carers. The authors heard from informal carers that they felt their *information needs* were not met and concerns about the disease and its progression were not addressed. Furthermore, they were unaware about available support services for their own mental health or felt they were missing, such as counselling for bereaved carers, highlighting an unmet psychological need. In relation to professional services, some informal carers were dissatisfied with 'paid-for in-home care', which could be a form of respite, providing a break from caregiving, however HCPs were viewed as unreliable and lacking in vital knowledge about MND.

Qualitative research in the area has taken place in various countries to explore differences in the caregiving experiences of informal carers and has provided deeper insight into individual experiences and situations. Lerum et al. (2016) conducted semi-structured interviews with 25

informal carers of plwMND from Norway; 17 were current carers and 8 were bereaved carers. Their findings highlighted 5 different 'lines of work' informal carers may engage in when caring for a plwMND. One example was *'immediate care work'*; informal carers provided physical assistance to the plwMND to engage in activities by replacing lost functions, such as turning a page in a book or moving their arms and legs. The authors emphasised the complexity of the caregiving role and the competing demands that informal carers face. Pinto et al. (2021) explored the emotional impact of caregiving for informal carers living in the UK and what might cause emotional distress. The authors conducted semi-structured interviews with 10 informal carers and analysed their findings using reflexive thematic analysis. The authors developed 8 core themes and 20 sub-themes relating to triggers of emotional distress in informal carers, such as '*keeping up with constant and multiple changes'*, and strategies to improve emotional wellbeing, such as '*finding hope and positivity'*. The study provided a voice to informal carers and generated an understanding of what can trigger emotional distress in informal carers and what strategies may be useful to improve their wellbeing.

Recently, there is research to suggest that informal carers may experience positive aspects of caregiving which can moderate and balance caregiving difficulties and burden (Galvin et al., 2016). As part of their mixed-methods study, Conroy et al. (2023) asked informal carers the open-ended question, "for you, what are the good things about caregiving?", and thematic analysis of the responses developed two main themes: *meaning in life* and *personal satisfaction.* Informal carers shared the importance of spending time together; opportunities for fun, humour and enjoyment provided *meaning in life* and caring for their loved one was seen as 'fulfilling' and 'rewarding'. This resulted in a sense of *personal satisfaction.* This novel research highlighted that caregiving experiences will vary between carers, and whilst it is widely considered that the majority of informal carers' experiences are related to distress and

burden, there are indications that these negative effects can be mitigated by the positive aspects of caregiving.

Existing research emphasises consideration of informal carers' needs and their relationship with professional services and HCPs. Evidence highlights the experiences of informal carers, but more importantly, emphasis is placed on their unmet needs and barriers they face which can lead to exacerbations in psychological and emotional distress.

A previous literature review by Aoun et al. (2012) investigated the qualitative evidence base on informal carers of plwMND and their experiences of caregiving and providing palliative/end-of-life care whilst highlighting their unique needs. The authors reviewed a 10 year period of research published between January 2000 and April 2011. A total of 59 studies were included. The findings of the review were consistent with previous research and demonstrated the level of burden and distress experienced by informal carers. Additionally, challenging points within the caregiving trajectory were emphasised such as the diagnosis experience, cognitive changes within the plwMND and end-of-life decision making. The authors emphasised the need for improvements in supporting informal carers, developing tailored interventions to meet their needs, and aiding access to palliative care services.

To the author's knowledge, there has not been a comprehensive synthesis of the recent evidence base from April 2011 onwards. The current review was informed by Aoun et al.'s (2012) review, using search terms that were further developed from those used by Aoun et al (2012) with an added focus on literature published between 2011 and 2024. The rationale for the current review was to improve the quality of the review using a systematic search approach and appropriate quality appraisal methods, highlighting any developments and advancements in the field. The author will take a qualitative approach to analysing the findings from numerous qualitative studies and integrating these into a qualitative synthesis. More specifically, thematic

synthesis, developed by Thomas and Harden (2008) was considered appropriate for this review as it encompasses many qualitative methods and explores participant experiences across contexts (Leeuwen et al., 2019). Thematic synthesis will allow for the analysis and integration of findings of relevant studies in order to develop salient themes reflecting the experiences of informal carers of plwMND.

Aims

The current systematic review aims to synthesise qualitative research published between 2011 and 2024 surrounding the experiences of informal carers of plwMND and answer the following research question: "What does the recent evidence base exploring the experiences of informal carers of plwMND highlight in terms of needs, experiences and barriers?"

Method

The protocol for this systematic review (Appendix A) was registered on PROSPERO (see https://www.crd.york.ac.uk/PROSPERO/display_record.php?RecordID=555943).

Systematic Review

Literature searches were conducted across three established databases (PsycInfo, Scopus and MEDLINE) in November 2024 to identify literature published between 2011 and 2024 that explored the caregiving experiences of informal carers of plwMND. The review was expanding upon a previous literature review by Aoun et al. (2012) that focused on caregiving experiences of informal carers between 2000 and 2011; the current review adopted a systematic approach to assess whether there have been any new developments in the field. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidance (Page et al., 2021) was adhered to when screening, identifying, selecting and critically appraising studies

identified for inclusion in this review (see Figure 1 for PRISMA flow diagram). Inclusion and

exclusion criteria is outlined in Table 1. Thematic synthesis (Thomas & Harden, 2008) was

selected to analyse the retrieved articles as it incorporates many epistemological perspectives

and qualitative methods, enabling the review to fully explore the entire scope of the literature.

Figure 1

PRISMA flow diagram

Identification	Records identified through database searching n=1,156 (PsycInfo n=195, MEDLINE n=295, Scopus n=666)

Total records (n=1,156)
Duplicates removed (n=430)

Records screened (n=726)

Records excluded following title and abstract screening (n=669)

Full-text articles assessed for eligibility (n=57)

Full-text articles excluded (n=34)

Quantitative (n=6)

Grey literature, case studies, surveys, theses, dissertation, non-peer reviewed articles, reviews (n=8)

Not relating explicitly to experiences of [informal] carers (n=13)

Focus only on post-death of plwMND (n=2)

Not in English (n=1)

*Unavailable (n=2)

Participants were paid carers (n=1)

Published before 2011 (n=1)

Studies included in synthesis (n=23)

*The lead researcher made attempts to contact authors of unavailable papers.

Table 1

Inclusion and exclusion criteria

Inclusion Criteria	Exclusion Criteria
Qualitative research	Quantitative research
Mixed-methods research with a distinct focus on qualitative aspect	Articles published in any language other than English
Articles focusing on experiences and needs of adult informal carers (18 years of over) of plwMND (bereaved and current)	Studies focusing solely on the bereavement period post death of the plwMND
Published in English	Studies which do not specifically refer to carers of plwMND or include paid carers
Articles published between 2011 and 2024	
	Where the carers' experience relates only to a medical procedure
	Grey literature, case studies, surveys, theses, dissertation, non-peer reviewed articles, reviews and any other unpublished studies

Search Strategy

The search strategy was produced using the PICo framework (Population, Phenomenon of Interest, and Context), an adapted version of the PICO tool for qualitative reviews (Stern et al., 2014). Table 2 outlines the developed search strategy. PICo is particularly suitable for qualitative synthesis because it does not rely on predefined outcomes (Stern et al., 2014). Although grey literature is often used to widen the range of studies identified, there is no consensus on the best approach to searching it (Mahood et al., 2014). As sufficient relevant papers were found, grey literature was not included in the review.

Scopus, MEDLINE and PsycInfo were systematically searched in November 2024. As the current review is expanding upon a previous review, searches were limited by date of publication, focusing on literature published between 2011 and 2024. Boolean operators were employed with search terms and the symbol * was used for truncation (Table 2). Titles, abstract and keywords were searched. The previous review by Aoun et al. (2012) was consulted for search terms and keywords. Preceding the initial searches being conducted by the lead researcher (SH), search terms were discussed with supervisors and checked by an external librarian at the university.

Table 2

Research strategy developed by PICo framework and search terms employed

Criteria	Description	Search Terms
Participant	Current or bereaved [informal] carers of a person living with MND.	Carer* OR caregiver* OR care-giver* OR caring OR caregiving* OR family caregiver* OR family carer* **AND**
Phenomenon of Interest	Qualitative experience of caregiving	Qualitative OR experience* OR interview* OR burden OR palliative care OR end of life care **AND**
Context	Motor Neurone Disease (MND), Amyotrophic Lateral Sclerosis (ALS)	Motor Neuron* Disease OR MND OR Amyotrophic Lateral Sclerosis

Screening

Following the searches, articles from each database were exported to EndNote 21, a reference management tool. Duplicates were identified and removed from the reference list. The titles and abstracts of all articles were screened against the inclusion and exclusion criteria, and articles meeting the exclusion criteria were excluded. Full-text screening was conducted on the remaining articles against the inclusion and exclusion criteria. The remaining eligible articles formed the final dataset for the synthesis. To confirm reliability, a random sample of articles that were included and excluded were reviewed by an independent researcher (YC). There were no discrepancies.

Data Extraction

Combined data extracted from the studies was organised into an Excel spreadsheet which included the author, year of publication, participant characteristics, study location, data collection methodology, inclusion of reflexivity and the main themes (findings) identified (Table 4).

Quality Appraisal

Included articles were critically appraised using the Critical Appraisal Skills Programme (CASP) qualitative 10-item checklist (2018; Appendix B), which is the most utilised tool for evaluating the quality of qualitative papers as it assesses validity and rigour. CASP does not employ a formal scoring system, therefore a similar review conducted with informal carers was consulted (Francis & Hanna, 2020) to determine the scoring system for this review. Studies were rated as low quality if they met 4 or less criteria, moderate quality if they met 5-7, and high quality if they met 8-10. The lead researcher (SH) carried out the ratings and 20% of the included articles were rated by an independent researcher (YC) to ensure reliability. Any discrepancies around individual CASP checklist categories were resolved via

discussion between the two researchers however both the independent researcher and the lead researcher rated the quality of all 23 papers as 'high'.

Thematic Synthesis

Data from the included papers was synthesised using the three-step thematic synthesis approach outlined by Thomas and Harden (2008). The first step consists of reading and re-reading the results and findings sections of the papers. Line-by-line coding was then undertaken by the lead researcher (SH) to identify meaningful areas of interest within the findings of each paper (Appendix C). The identified codes were compared across papers and categorised descriptively to develop themes; a thematic map was created to assist with theme development (Appendix D). Themes were characterised as superordinate themes and sub-themes relating to the experiences of informal carers of plwMND. Regular supervision with research supervisors helped guide this process and reflect on each step of the synthesis.

Reflexivity

Qualitative research is deeply influenced by the relationship between the researcher and the participant. Research has highlighted the importance of transparency regarding this relationship which is crucial for ensuring the integrity of qualitative findings (Dodgson, 2019). The lead researcher (SH) is a British Trainee Clinical Psychologist from a Pakistani ethnic minority background in her late twenties. She has no professional experience working with plwMND or their informal carers, and no personal experience.

Results

Quality Appraisal

A total of 23 studies were included as the final sample for synthesis. The CASP (2018) checklist was used to critically appraise the included studies and the results are documented in

Table 3. As the current review followed the guidance of Thomas and Harden (2008), no studies were excluded based on quality. All of the 23 included studies were found to be of high quality however, specific limitations of the included studies will be outlined. All but 5 studies failed to address reflexivity within their method. Two studies did not clearly detail their data analysis procedures. One study lacked reporting of ethical consideration and approval.

Table 3

Quality appraisal of included studies

Author and Date	Q1 – Aims	Q2 – Methodology	Q3 – Design	Q4 – Recruitment	Q5 – Data Collection	Q6 – Reflexivity	Q7 – Ethics	Q8 – Analysis	Q9 – Findings	Q10 – Research Value	Overall Rating
Anderson et al. (2019)	Y	Y	Y	Y	Y	?	Y	Y	Y	Y	High
Aoun et al. (2012)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Bentley & O'Connor (2016)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Cipolletta & Amicucci (2015)	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High

Conroy et al. (2023)	Y	Y	Y	Y	Y	N	Y	?	Y	Y	High
De Wit et al. (2019)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Larsson et al. (2015)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Lerum et al. (2016)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
O'Brien & Preston (2015)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
O'Brien et al. (2012)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Oh et al. (2021)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High

Olesen et al. (2022)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Oyebode et al. (2013)	Y	Y	Y	Y	Y	N	Y	?	Y	Y	High
Ozanne et al. (2015)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Poppe et al. (2022)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Poppe et al. (2022)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Trucco et al. (2023)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	High
Volpato et al. (2024)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	High

Warrier et al. (2020)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	High
Warrier et al. (2019)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Weisser at al. (2015)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	High
Whitehead et al. (2012)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	High
Yang et al. (2024)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	High

The researcher implemented a scoring template. Total score ratings: <4 = poor quality; 5-7 = moderate quality; 8-10 = high quality.

Summary of Characteristics of Included Studies

A summary of the characteristics of each study are presented in Table 4. As per the inclusion criteria, publication dates ranged from 2012 to 2024. Six studies took place in the UK whilst the rest were conducted in Australia, India, Italy, Ireland, the Netherlands, Norway, Sweden, Switzerland, Denmark, Korea and China. The majority conducted semi-structured interviews (n=19) and the remainder used narrative/other qualitative interview approaches. Across the papers, there was a total of 379 informal carers (male=133; female=229); one study failed to report the gender of their participants (Poppe et al., 2022). Participants across the studies were a mix of spouses, partners, children, siblings and parents of plwMND. Ten studies included current carers only, 7 included bereaved carers only and 6 included both. Several qualitative analysis methods were used; most common was thematic analysis (n=12). Other methods used included interpretative phenomenological analysis (n=4), descriptive phenomenological approaches (n=2), content analysis (n=2), theoretical reading (n=1), interpretative description (n=1), and grounded theory (n=1).

Table 4

Study Characteristics

Author and Date	Location	Recruitment	Sample (n)	Participant Characteristics	Current or Bereaved Informal Carers?	Data Collection	Data Analysis	Findings/Main Themes
Anderson et al. (2019)	Australia	Progressive Neurological Diseases Clinic	15	F = 8; M = 7, 14 spouses, 1 adult child	Current	Semi-structured interview	Thematic Analysis	Three key themes related to caring experiences: *The Thief, The Labyrinth, Defying Fate.*
Aoun et al. (2011)	Australia	Motor Neurone Disease Association (MNDA)	16	F = 13; M = 3, All spouses; Age range 53 – 81 years	Bereaved	Semi-structured interview	Thematic Analysis	Five key themes related to bereaved carers' experiences: *The work of MND family carers, The change in relationship from spouse to carer, Family caring as a series of losses,*

								Coping mechanisms of family carers, Supportive and palliative care experiences of family carers.
Bentley & O'Connor (2016)	Australia	MNDA	12	F = 7; M = 5, 11 spouses, 1 adult child	Bereaved	Semi-structured interview	Thematic Analysis	Three main themes related to end-of-life carer experiences: *Accessing appropriate supports, Accessing information, Feeling prepared*
Cipolletta & Amicucci (2015)	Italy	Website (forum)	13	F=10; M=3, 2 sons, 6 daughters, 1 husband, 2 wives, 1 girlfriend, 1 sister; Age range 24 – 64 years	Bereaved	Semi-structured interview	Interpretative Phenomenological Analysis	Three main themes related to caring for a plwMND: *Meaning of ALS, Family relationships, Health care context.*

Conroy et al. (2023)	Ireland	National ALS/MND Centre	17	F=13, M=4; 14 spouses, 3 daughters; Age range 32 to 82 years	Current	Semi-structured interview	Mixed-methods; Thematic Analysis	Two main themes related to positive aspects of caregiving: *Meaning in Life, Personal Satisfaction.*
De Wit et al. (2019)	Netherlands	6 ALS care teams (hospitals/rehabilitation centres)	21	F=15, M=6; 20 current carers, 1 bereaved; 15 partners, 5 adult children, 1 sibling; Age range 20 – 79	Both	Semi-structured interview	Thematic Analysis	Four major themes related to support needs of informal carers: *More personal time, Assistance in applying for resources, Counselling, Peer contact.*

Larsson et al. (2015)	Sweden	Neurology clinics	15	F=11, M=4; 11 spouses, 4 daughters; Age range 26-79 years	Bereaved	Semi-structured interview	Content Analysis	Two main themes related to carers' experience of care and support: *Patient-centred care inspired a feeling of security, Support was available but different factors influenced its use by the Relatives.*
Lerum et al. (2016)	Norway	MND clinics	25	F=18, M=12; 17 current carers, 8 bereaved; Age range 20 – 79	Both	Semi-structured interview	Theoretical Reading (Corbin & Strauss, 1988)	Five main lines of work that carers engage in: *Immediate care work, Seeking information and clarity, Managing competing obligations, Maintaining normality,*

								Managing external resources and assistance.
O'Brien & Preston (2015)	Northwest England	MND care and research centre	21	F=10, M=11; 18 bereaved carers, 3 current	Both	Narrative interview	Thematic Analysis	Four main themes related to carer perspectives of acute hospital care: *Lack of knowledge, Basic care, Reluctance for admission, Final memories.*
O'Brien et al. (2012)	Northwest England	MND care and research centre	28	F=14, M=14; 18 current carers, 10 bereaved	Both	Narrative interview	Thematic Analysis	Six main themes relating to carers' support service needs: *Impact on carers, Information, Paid-for-in-home care, Respite care, Counselling, Carers' training needs.*

Oh et al. (2021)	Korea	Tertiary hospital ALS clinic	10	F=4, M=6; 5 spouses, 2 parents, 1 adult child, 2 siblings	Current	Qualitative interview	Descriptive Phenomenological Approach (Colaizzi, 1978)	Three main themes related to caregiver suffering: *Frustration with seeing a patient suffering, Burnout at the cost of a life of dedication, Desperate need for help.*
Olesen et al. (2022)	Denmark	Websites	7 *(16 including HCPs)*	F=6, M=1; 2 adult children, 5 partners or spouses; Age range <22 - >66 years.	Bereaved	Semi-structured interviews with carers	Interpretative Description	Three main themes related to carer experiences of everyday challenges of MND and cognitive impairments: *Adjusting to new roles while balancing, Accepting that nothing else matters, Realising*

							different values in relationships.
Oyebode et al. (2013)	UK	MND clinic	8	F=6, M=2; Age range 40-70 years; 7 spouses, 1 partner.	Current	Semi-structured interview	Interpretative Phenomenological Analysis	Two main themes related to partner experiences of caregiving: *Impact on life, Adjusting to the situation.*
Ozanne et al. (2015)	Sweden	ALS team at hospital	13	F=5, M=8; Age range 38-87 years; All spouses.	Current	Semi-structured interview	Content Analysis	Two main themes related to spouses finding meaning whilst caregiving: *Feeling limited and isolated in the proximity of death, Finding meaning despite the proximity of death.*

Poppe et al. (2022)	Switzerland	ALS clinics	23 (36 including HCPs)	9 current carers, 14 bereaved; Age range 28 – 74 years;	Both	Semi-structured interview	Thematic Analysis	Five main themes related to carer support needs: *Administrative demand, Healthcare providers, Home support, End-of-life support, Social and other support.*
Poppe et al. (2022)	Switzerland	ALS centres	23	F=18, M=5; 16 spouses, 6 daughters, 1 friend; Age range 28-74 years;	Current	Semi-structured interview	Constructivist Grounded Theory	Core category: *Holding the balance.* Secondary categories: *organising support, being present, managing everyday life, keeping up with ALS.*

Trucco et al. (2023)	UK	MND carer support groups	14	F=10, M=4; All spouses; Age range 48-81 years.	Current	Semi-structured interview	Thematic Analysis	Three main themes related to carer experiences: *Destabilisation of diagnosis, Adapting to new circumstances and identifying coping strategies, Maintaining emotional coping.*
Volpato et al. (2024)	Italy	Rehabilitation Unit	40	F=25, M=16; 17 spouses, 16 children, 7 relatives; Age range 34-82 years	Bereaved	Semi-structured interview	Interpretative Phenomenological Approach	Five main themes relating to caregivers 'experiences of loss: *Caregiver's perception of his/her life, Caregiver's feelings, Caregiver's life after patient's death, Caregiver's*

							disease description, Caregiver's help resources.
Warrier et al. (2020)	India	Tertiary referral centre for neurological disorders	2	F=2; Both spouses; Ages 33 and 36 years	Current	Semi-structured interview	Interpretative Phenomenological Approach	Four main themes related to spouse experiences of caregiving: *Meaning of motor neuron disease, Relationship, Adaptation, Life without the loved one.*
Warrier et al. (2019)	India	Tertiary care centre	7	F=5, M=2; 6 spouses, 1 adult child; Age range 32 – 56 years	Bereaved	Semi-structured interview	Thematic Analysis	Four major themes relating to bereaved carers' experiences: *Transition from person to patient, Support, Death, Impact on the caregivers.*

Weisser at al. (2015)	UK	Websites; Neurology Clinics	10	F=7, M=3; Age range 35 – 62 years; 8 spouses, 2 partners.	Current	Semi-structured interview	Thematic Analysis	Four main themes related to caregiver experiences: *Resilience, Being rewarded, Carrying a burden, Having needs.*
Whitehead et al. (2012)	Northwest England	MND care and research centre	28 (52 including plwMND)	F=14, M=14; 18 current carers, 10 bereaved.	Both	Narrative interview	Thematic Analysis	Four main themes relating to carer experiences of end-of-life stages: *Anxieties, End-of-life decision-making and advance care planning, Services at the end-of-life stage, Impact on carers, Euthanasia.*

Yang et al. (2024)	China	Neurology department (tertiary hospital)	11	F=8, M=3; Age range 26 – 70 years; 6 spouses, 4 adult children, 1 parent.	Current	Semi-structured interview	Descriptive Phenomenological Approach (Colaizzi, 1978)	Three main themes relating to caregivers experiences: *Helplessness and adaptation to life changes, Hopelessness and compassion for the patient's prognosis, Gratitude and expectation for diverse support.*

Thematic Synthesis

Thematic synthesis of the 23 included papers produced 4 superordinate themes and 9 subordinate themes (Table 5). The 4 superordinate themes were:

1. "The caregiving spectrum"

2. The unique needs of informal carers

3. End-of-life care experiences

4. Experiences of healthcare professionals and dealing with services.

The first superordinate theme relates to the caregiving experiences of informal carers of plwMND which highlighted how carers may experience both *positive aspects* and *negative aspects* of caregiving. The second superordinate theme highlighting the unique needs of carers was further explored and three subordinate themes were identified: *need for information, training and knowledge, emotional and psychological needs,* and *support needs at different timepoints*. The third superordinate theme explored *end-of-life care experiences* as the disease progressed to the advanced stages, encompassed further by two subordinate themes: *changes in the caregiving role* and *feeling unprepared for death.* The fourth and final superordinate theme conveyed informal carers' *experiences of healthcare professionals and dealing with services*. Within this theme, the *importance of consistency and continuity* and *staff need to be experienced, knowledgeable and compassionate* were the two identified subordinate themes.

Table 5

Superordinate and subordinate themes

Superordinate Themes	Subordinate Themes
"The Caregiving Spectrum"	Negative aspects of caring Positive aspects of caring
The Unique Needs of Informal Carers	Need for information, training and knowledge Emotional and psychological needs Support needs at different timepoints
End-of-Life Care Experiences	Changes in the caregiving role Feeling unprepared for death
Experiences of Healthcare Professionals and Dealing with Services	Importance of consistency and continuity Staff need to be experienced, knowledgeable and compassionate

"The Caregiving Spectrum"

This superordinate theme illustrates the overall caregiving experience for informal carers of plwMND, highlighting that the role falls within a 'spectrum' with some carers experiencing both positive and negative aspects of caring.

Negative aspects of caring

Throughout the included studies that focused on the caregiving experience of informal carers of plwMND, the negative aspects of caregiving were made explicit by participants. Informal carers experienced feelings of helplessness, hopelessness and burden as their lives altered to focus on the plwMND.

"I used to have my hobbies, although my life is gone since he became ill…I suffer from frequent insomnia…and no longer know the meaning of my life." (Yang et al., 2024).

Many participants found it difficult to accept the disease and the diagnosis of MND which continued to be the case for some throughout the disease trajectory.

"I feel powerless. Then I think 'why did he get ALS? Why?' There's no answer." (de Wit et al., 2019).

Within the majority of the studies, participants were often spouses or partners of a plwMND who had adopted the informal carer role. They discussed the difficulty with adapting to this change in role and identity, feeling as though they were now a carer first and spouse/partner second.

"The wife role was gone and I was a nurse and that's what I regret." (Aoun et al., 2011).

Due to this change in role, informal carers experienced losses within their life and environment, feeling restricted and no longer able to engage in desired activities or prioritise other important roles such as being a parent.

"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, Smith & Morrison, 2013).

"…I have a daughter in [city] with three grandchildren…used to go over every six weeks…wouldn't leave [patient] now…I miss that." (Trucco et al., 2023).

However, this experience of losses also pertained to their loved one; carers felt grief and pity for their loved one as their physical condition deteriorated.

"He must be feeling frustrated. Not being able to move, that's the worst part. He had lots of friends. They all left him once he got sick." (Oh, Kim & Chu, 2021).

Loss of function and ability in their loved one resulted in some carers taking on additional duties that were once the responsibility of the plwMND.

> "*He always made the major decisions [so] I had to learn the finances.*" (Aoun et al., 2011).

Positive aspects of caring

Although the primary focus of most of the papers was on the burdensome aspects of caregiving, some participants shared the rewarding and positive aspects. Most common was that caring for a loved one provided purpose and meaning in life for carers.

> "*Connection, closeness, positivity that we both found…he's been brave for me and I've been brave for him.*" (Conroy et al., 2023).

Participants experienced fulfilment and personal satisfaction from caring for their loved one, particularly when they received positive feedback and felt appreciated.

> "*Seeing how much she appreciates it.*" (Conroy et al., 2023).

Informal carers highlighted how caring for a loved one with MND encouraged them to 'live in the moment' and make the most of the present time they had together rather than worrying about what the future holds.

> "*Your whole view of life changes…your priorities change…you don't look too far ahead. You just get on with each day.*" (Oyebode, Smith & Morrison, 2013).

Studies that included spouses demonstrated how caregiving had led to a deeper connection with their other half and strengthened the marital relationship.

> "*When me and [patient] became more open, we shared a lot more.*" (Weisser, Bristowe & Jackson, 2015).

Various sources of support allowed carers to cope with the challenges of caregiving and participate in activities that they value, improving their quality of life. Examples included engaging with nature and connecting with faith and religion.

"*So the forest and nature...still very important for me...getting out into nature, preferably alone.*" (Ozanne, Graneheim & Strang, 2015).

"*Religion is very comforting. I go to church once a week...I express my gratitude...and ask for another good week.*" (Oh, Kim & Chu, 2021).

Participants valued meaningful relationships with family members, friends and HCPs; significant individuals and groups in a carer's life formed a support system for them and helped them cope with the difficult aspects of caregiving.

"*We have a care co-ordinator...she is simply worth her weight in gold. I love her more than anything else in the world...stood by us right from day 1...she took care of everything.*" (Olesen et al., 2022).

The Unique Needs of Informal Carers

The second superordinate theme demonstrates how informal carers of plwMND have distinct emotional, psychological, support and information needs that often remain unmet.

Need for information, training and knowledge

Within the included studies, informal carers voiced that their information needs about the disease were not met and they were lacking in sufficient knowledge about MND and its trajectory.

"Nobody has actually told us what to really expect from the disease…I have had little guidance on what to expect." (O'Brien et al., 2012).

Informal carers highlighted a gap in practical training and education around manual handling as they delivered physical care for their loved one without sufficient knowledge.

"If you as a relative could be taught some tricks…how to turn a person over in bed, a person who can't move…I should have some knowledge that I didn't. This would have made it easier for me." (Larsson et al., 2015).

Participants also expressed a need for advice and training around managing behavioural changes in the plwMND which would have been a valued resource.

"I'd like to receive tips on how I can deal with [patient's apathy]. Do I need to push him? Or just leave things as they are?" (de Wit et al., 2019).

Emotional and psychological needs

It was evident within several of the papers that informal carers have unique emotional and psychological needs related to their caregiving role. However, these needs can be overlooked by carers as they focus on their loved one and deprioritise themselves.

"You don't think about it. You don't think about yourself at the time, you just think about your sick loved one who needs help." (Larsson et al., 2015).

Some participants shared how tailored emotional support was needed but missing, communicating a desire to speak with someone openly about their concerns for the future.

"…perhaps a counselling service, a talking service, talking to somebody who knew what you were on about, at my stage…probably be a good idea…I wish I had access to that…I don't have anybody to talk to." (O'Brien et al., 2012).

Others voiced that speaking with relatives in similar situations to their own could offer a form of peer support to help carers cope with the loss of their loved one and bereavement.

> "*I would like to speak to someone who had also lost their mum or dad to ALS and like me, is struggling with it.*" (de Wit et al., 2019).

Support needs at different timepoints

As the disease progressed, informal carers shared how their support needs changed at different timepoints in the disease trajectory, often highlighting an increased need for support. For some carers, help was required with communication with the plwMND during the later stages of the disease as the ability to communicate verbally was lost.

> "*...because my mother couldn't speak anymore and things didn't always go well with this computer...and then my mother just had to blink...actually works a lot through the eyes, we also made a board with the most important things that you can ask.*" (Poppe, Phil & Wangmo, 2022).

As the plwMND continued to lose vital functions and abilities, informal carers required support from professionals to apply for resources and aids to manage care provision. However, they shared frustrations that the responsibility was often left with them, increasing burden and mental fatigue.

> "*It would be so helpful if the team [rehabilitation team] would apply for the devices for me. It would really help my frustration. I put a lot of time and effort in it [applying for devices] but I'm just not getting anywhere.*" (de Wit et al., 2019).

Throughout the disease trajectory, informal carers expressed needing support with requesting respite care to allow them to have a break from caring. Many carers were unaware of the

process for applying for respite care and would have valued being signposted to support and resources.

> "*Respite care is too complicated in our family. I have been in contact about this topic several times.*" (de Wit et al., 2019).

Whilst others shared that respite support was not always readily available to them and was often impracticable.

> "*Well, respite is a really big problem…I couldn't book a holiday on the off chance there might be a bed…hopeless…respite is an area which needs to be looked at*". (O'Brien et al., 2012).

End-of-Life Care Experiences

The third superordinate theme derived from the studies relates to end-of-life care experiences of informal carers, particularly the changes in their caregiving role and duties, and facing the fatality of the disease.

Changes in the caregiving role

Informal carers expressed changes in their caregiving duties as their loved one entered the later stages of the disease. Carers highlighted how they were involved in advance care planning and end-of-life decision making, advocating for their loved one.

> "*We went to see the GP and because he wants to die at home, he doesn't want any intervention or anything…GP was very supportive…that was a big weight off my shoulders.*" (Whitehead et al., 2012).

Carers found there was a significant increase in level of care they were providing; additional care duties which the plwMND was no longer able to carry out clearly took a toll on carers.

> "*He was bedridden for the last seven to eight months. Initially, I was…doing everything, even household chores. After a month, I became totally tired.*" (Warrier et al., 2019).

For some participants, deterioration of their loved one during the end stages was slow whereas for others, it was much quicker. Rapid deterioration often meant carers had to problem-solve constantly as support solutions would only have limited and temporary usefulness.

> "*If you look to the end stage then…you know, sooner or later you're going to need a wheelchair and sooner or later the wheelchair's not really going to be useful at all.*" (Anderson et al., 2019).

Feeling unprepared for death

Many carers across the included studies found it difficult to accept their loved one was dying or in the final stages of their life. Informal carers voiced anxieties around the anticipation of death and in what ways their loved one dying would affect them.

> "*I don't know how it is going to affect me when something happens, you can't lose somebody after thirty years and not feel it, I don't quite know how badly I am going to feel it yet, I know that's in the future.*" (Whitehead et al., 2012).

Participants shared how they did not receive enough information about what the end stages of MND would entail and death had not been discussed with them by services and HCPs.

> "*[Nobody talked to me about it]. I wasn't aware of what to expect in those final stages. I did start to read more [to find out on my own].*" (Bentley & Connor, 2016).

Understandably, there was a lack of preparedness for death amongst informal carers. The death of the plwMND was often unexpected and traumatic, with some carers feeling shocked that 'death had come so soon', especially if the plwMND was still functioning to an extent.

> "*If he could not communicate or if he was bed-bound and totally dependent, we would not have felt so bad.*" (Warrier et al., 2019).

For some carers, the shock of the sudden death of their loved one was intensified if HCPs had recently visited the plwMND and not indicated that they are in the final stages of their life.

> "*We had a visit from the nurse on the Friday before…she died…she didn't say to me she hasn't got long to go or we ought to be making other arrangements, which is the information I would have liked to have.*" (Bentley & Connor, 2016).

Experiences of Healthcare Professionals and Dealing with Services

The final superordinate theme provides insight into how informal carers have experienced HCPs and services, both in relation to themselves and the plwMND, stressing the importance of consistent, continued support and staff being sufficiently experienced and trained.

Importance of consistency and continuity

Within the included studies, participants shared that they valued consistent support and input from HCPs when caring for their loved one. Carers experienced a sense of security if care was delivered by the same staff at each appointment.

> "*Not having to say the same thing or worry about forgetting to say something of importance at the visits. That it was the same staff who knew the patient at each appointment. It was a great source of security.*" (Larsson et al., 2015).

When there was a high turnover of staff, informal carers found it challenging to accept care within the privacy of their home and care was often disrupted due to changes in staffing.

> "*There are constantly people in our home; I don't have a private life anymore.*" (de Wit et al., 2019).

> "*I think at one stage…she had twenty different carers…that is an awful lot of people to say 'no actually I can't do this'… 'that's kept in there'.*" (O'Brien et al., 2012).

Some participants voiced that when the plwMND died, services and HCPs were quick to withdraw support. Carers felt alone during the crucial period of bereavement where continued support would have been valued to cope with loss and grief.

> "*You go from having a whole army of people; then it's just you.*" (Aoun et al., 2011).

> "*When (patient) died, (specialist nurse) never got in touch with me…devastated about that…you are just cut off then…the notes go in a drawer.*" (Whitehead et al., 2012).

Staff need to be experienced, knowledgeable and compassionate

Interviews with informal carers of plwMND revealed experiences of dealing with staff and HCPs who were lacking in relevant experience and training. In relation to paid care, carers found themselves adopting a 'teacher' role and reiterating key information about the care needs of their loved one, which proved to be burdensome.

> "*They didn't know much about the disease. It felt like they were lacking in experience. The nurses who came to the home didn't really know. Sometimes you had to tell them that they couldn't do some things.*" (Larsson et al., 2015).

Some participants encountered HCPs who lacked knowledge about MND during acute hospital stays which frustrated informal carers.

"The staff need an awful lot more training in how to deal with people suffering MND...there are so many little incidences which show lack of understanding." (O'Brien & Preston, 2015).

At the time of diagnosis, some carers experienced HCPs who lacked compassion and empathy when conveying the MND diagnosis and were left feeling shocked and bewildered by the encounter.

"The neurologist told us that he knew someone with MND and that they shot themselves right away. Hearing that was just terrible 'cause here I am thinking we will beat this...we didn't go to that neurologist again." (Aoun et al., 2011).

Discussion

The current review aimed to systematically synthesise qualitative research published between 2011 and 2024 that focused on the caregiving experiences of informal carers of plwMND, highlighting needs and potential barriers. Thematic synthesis of the 23 included studies resulted in four superordinate themes: *'The Caregiving Spectrum', The Unique Needs of Informal Carers, End-of-Life Care Experiences* and *Experiences of Healthcare Professionals and Dealing with Services.*

'The Caregiving Spectrum'

All of the papers contributed to this superordinate theme which demonstrated how caregiving experiences are unique and individual to each informal carer. The papers highlighted how caregiving can fall on a 'spectrum', with carers experiencing negative and positive aspects of caring for their loved one.

The negative features of caregiving are well documented in the evidence base. This includes informal carers experiencing the 'burden' of caregiving, feeling hopeless and helpless, and neglecting their own needs. In line with the findings of this review, Rabkin et al. (2009) found informal carers of plwMND will often 'let go' of their own self-care needs and as a result, may experience physical exhaustion and fatigue. The 'burden' of caregiving is increased when informal carers lead restricted lives and lack social connection as was found across the papers in this review. Love et al. (2005) demonstrated how social restriction resulted in carers experiencing social isolation and a reduced quality of life (QoL). The findings of this review around the negative aspects of caregiving are similar to Aoun et al.'s (2012) review, demonstrating the importance of investigating the negative effects of caregiving to allow for a deeper understanding of how informal carers can be supported.

Less documented are the positive aspects of caregiving, which were found across some of the included papers. Studies highlighted how informal carers experience positive effects of caring, including deeper connections and bonds, finding meaning in life and solace in sources of support. Whilst caregiving was a difficult and demanding task, the challenges were somewhat mitigated by positive factors that helped them cope and view caregiving from a different perspective. Previous research has explored mitigating factors that could reduce caregiver burden and increase QoL; Calvo et al. (2011) found religiousness was a valued source of support that improved the QoL of informal carers. Finding meaning in life was found to be a protective factor for carers against psychological distress (Farran et al., 1997). As the evidence base is still limited, future research would benefit from further exploring positive aspects that could mitigate the challenges informal carers face.

The Unique Needs of Informal Carers

This superordinate theme described the individual and unique needs that informal MND carers have, including information needs, emotional and psychological needs and support needs which alter across the disease trajectory. For most, these needs were unmet and carers expressed gaps in support.

Carers described feeling left to their own devices to learn about MND and were not provided with sufficient information about the disease. Consequently, informal carers experienced uncertainty about the disease and did not have answers about its progression or trajectory. This need for information and knowledge about MND has been presented across various past studies as carers want to know more about MND outcomes (Akiyama et al., 2006; Hughes et al., 2005). Lackey & Gates (2001) highlighted the need for carers to receive training for practical aspects of care such as physical handling and utilising technological devices.

The emotional and psychological needs of informal carers were demonstrated across several of the included papers. Previous research and reviews, including Aoun et al. (2012), illustrated the extent of the psychological impact of caregiving on informal carers; Jenkinson et al. (2000) found 70% of informal carers scored below the population norm on mental health. Limited research has explored how to practically assist or support carers experiencing psychological distress. This gap was voiced by carers in the current review as they expressed a need for tailored emotional support in the form of either peer support or formal counselling.

The progression and deterioration of MND can vary from person to person. Across the included papers, informal carers shared the need for support at different timepoints to help them cope; in particular, access to and availability of respite care was seen as crucial during the final stages. This finding is supported by previous research which demonstrated the value of respite care; it was "singled out as the most important service" to aid carers with continued caregiving

(McCabe, Roberts & Firth, 2008). Barriers to accessing respite care were revealed by carers, such as lack of signposting, which future research would benefit from exploring.

End-of-Life Care Experiences

This theme was encompassed by informal carers' experiences of delivering care as their loved one entered the end-of-life stages. Across the included papers, informal carers felt a shift in the caregiving role as their duties increased and they learned new skills such as legal work and advance care planning.

Understandably, an increase in caregiving duties took a toll on carers who experienced exhaustion and fatigue as the plwMND deteriorated. This is in line with previous research by Pagnini et al. (2010) who found carers showed signs of somatic depression such as lack of sleep and appetite as their loved one lost ability and function.

Despite the existence of palliative care guidelines (Oliver & Webb, 2000), and an emphasis within the literature that plwMND and their family carers should receive additional support during end-of-life stages (Kristjanson, Toye & Dawson, 2003), barriers to accessing palliative care prevent informal carers from utilising these services. Specific barriers such as services being reluctant to provide care to plwMND who have high-dependency needs (O'Leary & Tiernan, 2008) are in line with findings from the current review. Little research has focused on overcoming barriers to allow ease of access to palliative care services for informal carers.

Consequently, carers across the papers felt unprepared for the death of their loved one, highlighting lack of support from services. Carers shared concerns about the future and worrying about how the death of their loved one would impact them. Many stressed the importance of bereavement support and counselling for carers as they cope with emotions such as grief and anger. This remains a gap in care and clearer referral procedures are required (Aoun et al., 2012; Aoun et al., 2011).

Experiences of Healthcare Professionals and Dealing with Services

The fourth superordinate theme addressed the experiences informal carers had of engaging with HCPs and various services. Key findings included carers receiving inconsistent support from HCPs, feeling alone when support was taken away, and encountering HCPs who lacked empathy, relevant experience and knowledge.

Carers highlighted the burden they carried adopting a 'teacher' role for HCPs who lacked awareness of how to care for the plwMND. This lack of awareness was coupled with a lack of knowledge and understanding of what MND was and carers often felt anxious about their loved one receiving care from staff who lacked specific MND experience. Previous studies have elaborated on the poor understanding many HCPs (who are not MND specialists) have of MND (Hugel et al., 2006) and its impact on patient care (Hughes et al., 2005). However, there is limited investigation of the direct impact on caregivers of plwMND. Carers in the current review stressed the necessity of HCPs being adequately trained and educated around MND. This has been supported by the existing evidence base, including Aoun et al.'s (2012) review, which encourages professional training programmes for HCPs and services to improve understanding of MND (O'Brien et al., 2012).

Consistent with Aoun et al. (2012), the current review demonstrated the importance of the delivery of the MND diagnosis and the longstanding impact of lack of compassion on carers and plwMND. An unempathetic approach to communicating the MND diagnosis eliminated hope and resilience in carers. Previous studies have supported this finding in relation to poorly relayed diagnoses; Aoun et al. (2006) reported distress in carers and plwMND when healthcare specialists lacked empathy during the diagnostic process. An area of concern that is still being highlighted, one suggestion is that further training for HCPs in delivering and communicating

a diagnosis of MND to carers and their loved ones could ease distress (McDermott & Shaw, 2008).

Critique of Included Studies

Whilst all of the included studies were rated as 'high' quality following critical appraisal using the CASP (2018) checklist, it is worth noting the limitations of specific papers.

The most common limitation within the methodology of the 23 included studies was that only 5 studies (Trucco et al., 2023; Volpato et al., 2024; Warrier et al., 2020; Weisser, Bristowe & Jackson, 2015; Yang et al., 2024) mentioned reflexivity in detail, addressing the researcher-participant relationship. Within qualitative research, reflexivity is a crucial process that demonstrates transparency and considers the context within which researcher-participant interactions take place (Dodgson, 2019). It can be argued that including and appropriately considering reflexivity in qualitative research can enhance the credibility of findings.

One study (Cipolletta & Amicucci, 2015) failed to report any ethical considerations or ethical approval within their publication thus questioning the integrity of their research as it was unclear whether appropriate ethical standards were upheld.

Two studies (Conroy et al., 2023; Oyebode, Smith & Morrison, 2013) did not make clear their approach to rigorous analysis or provide sufficient detail about the steps that were taken. Without this level of detail, it could be difficult to accurately replicate the investigation.

A strength of all of the included studies was their explicit reporting of research aims and clear justification of their qualitative methodology and data collection techniques, demonstrating a high level of face validity and integrity of research.

Strengths, Limitations and Future Research Directions

To the author's knowledge, this is the first review adopting a systematic approach to synthesising the recent evidence base (2011 onwards) exploring the caregiving experiences of informal carers of plwMND. The review provided a useful extension to Aoun et al.'s (2012) existing review and has highlighted key developments such as the increase in published literature within the field.

A strength of the review is the inclusion of 23 qualitative studies that all received 'high' quality appraisal ratings. The high quality of the studies demonstrated strong reliability of the current review which was reinforced by the ratifying assessments made by the independent reviewer using the CASP (2018) checklist.

The CASP (2018) checklist is an accessible critical appraisal tool that is widely employed to assess research quality. However, it has been criticised regarding its sensitivity and weaker assessment of methodological quality (Majid & Vanstone, 2018). In the current review, despite a 'high' rating overall, one study completely failed to acknowledge ethical approval (Cipolletta & Amicucci, 2015). Furthermore, variation in rater assessments is more likely due to the 'yes', 'can't tell' or 'no' response categories. It is worth acknowledging that interpretation and ratings may have differed using other tools with more reliable sensitivity and the quality of studies may have been more accurately appraised.

The current reviews used broad search terms to include as many studies as possible whilst adhering to the inclusion/exclusion criteria. However, some important studies may have been missed due to the lack of specific terms, for example, Aoun et al. (2012) included 'neurodegenerative disease' in their search terms which may have captured a higher number of studies. Furthermore, specific terms related to relationships of informal carers to plwMND were not included in the current review such as 'wife', 'son', 'daughter', which may have

depicted a wider group of family caregivers. Future research would benefit from including both broad search terms and specific terms related to carers and MND to portray a more holistic representation of caregiver experiences.

The exclusion of grey literature may have resulted in important, unpublished research being missed. In particular, third-sector associations and charities such as the MND Association may have carried out relevant research exploring the experiences of informal carers of plwMND but lack the resources for publication. Future reviews and research should consider reviewing the grey literature within this field to complement the existing review and highlight similar findings.

A strength of the current review is the breadth of included studies from a range of countries such as the UK, India, Switzerland, Italy, Australia, Korea and China, allowing for many cultural perspectives being captured and strengthening the generalisation of the findings as they could be applied to informal carers from diverse backgrounds.

Clinical Implications

The current review has implications for clinical settings and practice as well as informing future research. The previous review advocated for tailored support and intervention for informal carers, particularly highlighting the need for more integration of palliative care support for carers (Aoun et al., 2012). The included studies within the current review have suggested there is still this need to provide appropriate support for carers, and research should 'shift' from just understanding the needs of informal carers to practically informing and tailoring interventions and services to meet the specific needs of carers. These advancements within the evidence base could guide services and professionals to developing appropriate support resources for carers which could help mitigate the challenges of caregiving.

Furthermore, to ensure systemic working and that all those involved in the care of plwMND and their carers are providing adequate support, an understanding of how specific interventions can be delivered to MND carers is needed. This includes interventions provided by psychologists, other HCPs, and by charities/third-sector organisations as the review has highlighted a number of services are usually involved. For example, charities may facilitate carer support groups at crucial times i.e. bereavement/post-death of the plwMND, whilst mental health services may provide individual counselling.

Another clinical implication highlighted by the findings of the current review is the importance of training and support for HCPs to ensure those caring for plwMND understand the disease and the needs of plwMND and MND carers. This is required to improve the way a diagnosis of MND is delivered and to ensure plwMND and MND carers feel confident that they can trust the professionals looking after them rather than feeling responsible for educating them. This would also be an opportunity to educate staff on the importance of offering MND carers access to palliative care services and respite care. Additionally, professionals should ensure MND carers have access to appropriate information and training to facilitate their role in providing care, and support that meets their unique and changing emotional support needs.

Conclusion

The current review has provided valuable findings that are consistent with and build on the findings of the previous review by Aoun et al. (2012). The findings demonstrate the unique caregiving experiences and needs of MND carers that are not currently being met by services. The importance of HCPs having appropriate training and MND experience has been emphasised, and the potential value of linking MND carers in with palliative care services. Research since the previous review has shown the possibility of MND carers experiencing positive aspects of caregiving and factors that can mitigate burden, and these are areas that

warrant further exploration. The review also highlights the importance of developing support

tailored to the unique needs of MND carers.

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Appendices

Appendix A – PROSPERO protocol

A systematic review and thematic synthesis of studies exploring informal carers' experiences of caring for people living with Motor Neurone Disease (MND)

Saba Hussain, Charlotte Wright

Review methods were amended after registration. Please see the revision notes and previous versions for detail.

Citation

Saba Hussain, Charlotte Wright. A systematic review and thematic synthesis of studies exploring informal carers' experiences of caring for people living with Motor Neurone Disease (MND). PROSPERO 2024 CRD42024555943 Available from: https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42024555943

Review question

What does the recent evidence base exploring the experiences of informal carers of people living with MND highlight in terms of needs, experiences and barriers?

Searches [1 change]

Searches will be conducted on a number of relevant health and social care electronic databases including Scopus, PsycINFO and PubMed. Reference lists of previous literature searches will be sourced for relevant studies. Searches will be conducted from October to November 2024. Articles and studies printed in English language will be included, published between 2011-2024 as the current review is exploring the current evidence base, expanding upon Aoun et al's 2013 literature review that reviewed publications between 2000 and 2011. It is anticipated that searches will be re-run just before the final analyses. Search terms related to MND, ALS, palliative care and [informal] carers will be implemented.

Types of study to be included

Studies that employ qualitative methods to explore the caregiving experiences of current or bereaved informal caregivers of adults with MND; are published in English. Studies which use both quantitative and qualitative methods of data collection will be included if the qualitative data are reported separately and can be clearly extracted.

Condition or domain being studied

Motor neurone disease (MND) is a neurodegenerative disease which has a sudden onset and progresses rapidly (Aoun et al 2013). It is currently without cure, and the progression of symptoms and subsequent onset of disability are likely to occur over weeks and months rather than years (Aoun et al 2013). Informal carers of people living with MND (plwMND) experience challenges when caring for their loved ones, usually family members, with a physical, practical, mental and emotional impact on their wellbeing (Aoun et al 2013). It is therefore important that the needs, experiences, and views of informal carers are explored and

understood to ensure their needs are being met and supported by services; research has found current support for informal carers to be 'inadequate and unsatisfactory' (Love et al 2005).

Participants/population

Inclusion criteria:

Anyone above the age of 18 who is a current or bereaved informal carer for a person living with MND.

Exclusion criteria:

Formal carers or paid carers of a person or people living with MND such as MND nurses.

Intervention(s), exposure(s) [1 change]

Inclusion:

All articles focusing on the experiences of informal carers of plwMND, both current carers and bereaved carers. Articles that explore the relationship informal carers have with professional services and the impact of this on their wellbeing.

Qualitative or mixed-method studies where there is a distinct focus on the qualitative aspect.

All relevant studies which meet the critical appraisal checklist (CASP).

Studies published in English language.

Exclusion:

Studies which do not specifically refer to carers of plwMND.

Studies focusing only on the bereavement period post death of the person with MND.

Where the carers' experience relates only to a medical procedure.

Grey literature, case studies, surveys, theses, dissertation, non-peer reviewed articles, reviews and any other unpublished studies.

Studies not published in English language.

Comparator(s)/control

Not applicable.

Main outcome(s)

To identify and explore the experiences and needs of informal carers of plwMND, with a focus on the recent literature from 2011 onwards.

Additional outcome(s)

None.

Data extraction (selection and coding)

Dependent on the database, databases will be searched using appropriate search terms and Boolean words. The relevant inclusion and exclusion criteria will be used to filter databases.

Initially, titles will be screened for eligibility, followed by abstracts and full-text articles for those included after the title screening. Backwards and forwards citation searches will be carried out on included papers to identify any additional papers, which will then be subject to the same screening process.

For included papers, data extraction will include:

- Author and publication date

- Study design, data collection and methodology

- Study aims and objectives

- Participant demographics

- Main themes related to the systematic review question:

What does the recent evidence base exploring the experiences of informal carers of people living with MND highlight in terms of needs, experiences and barriers?

Researcher and research supervisor will cross-check inclusion and screening process and data extraction.

Risk of bias (quality) assessment

https://casp-uk.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf

Risk of bias or quality of papers included will be assessed using the CASP Qualitative quality checklist tool. The characteristics assessed by this tool are:

- Clear aims

- Appropriateness of qualitative methodology and design to meet the aims

- Appropriateness of recruitment strategy

- Collection of data in a way which addressed the research question

- Reflexivity

- Consideration of ethical issues

- Rigour of data analysis

- Clarity of presentation of findings and statement of value

Quality assessment will take place at the individual study level. Papers will not be excluded on the basis of quality, but this assessment will be considered alongside the synthesis to inform the overall interpretation of the results.

Quality will be assessed primarily by the researcher. An independent researcher will assess the quality of a random 20% of included studies to ensure inter-rater reliability. Any discrepancies will be discussed and if these cannot be resolved it will be taken to the research supervisor for further discussion.

Strategy for data synthesis

Thematic synthesis methodology, defined by Thomas and Harden (2008), will be used to synthesise data extracted from relevant and included studies. This will involve coding, developing descriptive themes and finally, generating analytic themes. This information will be taken from the results and discussion sections of all included papers. It will then be reported in the results and discussion section of this review.

Discussion of themes between the researcher and research supervisor will take place to ensure reliability.

Analysis of subgroups or subsets

Not applicable.

Contact details for further information

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Review team members and their organisational affiliations

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Dr Charlotte Wright. University of Sheffield

Type and method of review

Systematic review

Anticipated or actual start date [2 changes]

05 November 2024

Anticipated completion date [1 change]

06 December 2024

Funding sources/sponsors

University of Sheffield

Conflicts of interest

Language

English

Country

England

Stage of review

Review Ongoing

Subject index terms status

Subject indexing assigned by CRD

Subject index terms

MeSH headings have not been applied to this record

Date of registration in PROSPERO

23 October 2024

Date of first submission

18 October 2024

Stage of review at time of this submission [2 changes]

Revision note

Updated progress with review which has now started.

The record owner confirms that the information they have supplied for this submission is accurate and complete and they understand that deliberate provision of inaccurate information or omission of data may be construed as scientific misconduct.

The record owner confirms that they will update the status of the review when it is completed and will add publication details in due course.

Versions

23 October 2024

07 November 2024

07 November 2024

Appendix B – CASP Checklist

CASP
Critical Appraisal
Skills Programme

Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
• what was the goal of the research
• why it was thought important
• its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
• If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
• Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
• if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

Comments:

4. Was the recruitment strategy appropriate to the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the researcher has explained how the participants were selected
- If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study
- If there are any discussions around recruitment (e.g. why some people chose not to take part)

Comments:

5. Was the data collected in a way that addressed the research issue?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the setting for the data collection was justified
- If it is clear how data were collected (e.g. focus group, semi-structured interview etc.)
- If the researcher has justified the methods chosen
- If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide)
- If methods were modified during the study. If so, has the researcher explained how and why
- If the form of data is clear (e.g. tape recordings, video material, notes etc.)
- If the researcher has discussed saturation of data

Comments:

6. Has the relationship between researcher and participants been adequately considered?

Yes ☐
Can't Tell ☐
No ☐

- If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location
- How the researcher responded to events during the study and whether they considered the implications of any changes in the research design

Comments:

Section B: What are the results?

7. Have ethical issues been taken into consideration?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained
- If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the study on the participants during and after the study)
- If approval has been sought from the ethics committee

Comments:

8. Was the data analysis
sufficiently rigorous?

Yes ☐

Can't Tell ☐

No ☐

HINT: Consider
- If there is an in-depth description of the analysis process
- If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
- Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
- If sufficient data are presented to support the findings
- To what extent contradictory data are taken into account
- Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

Comments:

9. Is there a clear statement
of findings?

Yes ☐

Can't Tell ☐

No ☐

HINT: Consider whether
- If the findings are explicit
- If there is adequate discussion of the evidence both for and against the researcher's arguments
- If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
- If the findings are discussed in relation to the original research question

Comments:

10. How valuable is the research?

HINT: Consider
- If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature
- If they identify new areas where research is necessary
- If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Comments:

Appendix C – Line-by-Line Coding Examples

with MND, and which could inform the development of support interventions, are reported.

Ethical considerations

Calvary Health Care Bethlehem Research Ethics and Ethics Committee approved this study (Number 14051501 dated 20 May 2014). All participants indicated interest in receiving a summary of the research findings, which were provided with a thank you letter.

RESULTS

Participants

From 23 eligible persons approached, the recruitment strategy generated 15 participants (65% consent rate). Participants refused because of difficulty in discussing the caregiving experience, with participants citing feeling overwhelmed or upset thinking about their experience. Participants' demographic details are presented in table 1. Interviews lasted between 30–70 min, with a mean of 45 min.

Themes

Three key themes were identified: (1). *The Thief* entailed the experience of loss and grief across varied facets of life; (2). *The Labyrinth* related to the experience of finding ways to address the ever-changing challenges as the disease progressed; and (3). *Defying fate* reflected the experience of resilience and hope as caregivers tried to make the most of the time remaining with their loved one. Themes are supported by participant quotes and accompanied by a participant number.

Table 1 Participant characteristics

Caregiver (N=15)	
Gender	
Female	8
Male	7
Age (years)	
40–50	2
50–60	1
60–70	7
70–80	4
80–90	1
Care duration (months)	
1–6	6
6–12	1
12–24	5
24–36	3
Living situation	
Together	14
Separate	1
Relationship	
Spouse	14
Adult child	1

The Thief—lost aspects of life

The impact of losses was personal; many expressed shock, grief and helplessness associated with the first few months following diagnosis and as facets of their life and the life of the person with MND changed. One stated "it was a real shock to begin with and so it kind of turned my life upside down" (06) and another stated "we felt… almost paralysed I guess by the enormity of what we were facing" (01). Another significant area of loss was an individual's aspirations. Participants described how the condition had devastated their anticipated life experiences and future plans, particularly for retirement. As one participant stated "The next stage of our lives, for some time it felt like that got taken away from us" (01).

Participants also described the experience of loss in regard to the physical capacity and identity of the person with MND. For instance, caregivers shared in the loss of the person with MND's vocation, personal interests, social activities, motivations and independence, demonstrated as follows "… all he wanted to do was just stay at home, not see anyone, not talk to anyone" (04); "she can't drive any more… that's probably upset her" (05). The progression of the condition was also associated with loss of certain features in the spousal relationship. Some shared experiences about loss of communication and intimacy: "You lose that connection of a soul mate. You lose that, you know, sitting down over a glass of wine and debriefing the day or talking about deeper things" (06). Further, some participants' experienced anticipatory losses, including deteriorating health and eventual death of their loved one; a participant stated, "I'm really scared of what''s going to happen when he''s bedridden and when it gets worse…" (07) and "I have been thinking about that, you have to do it. Because you know death''s going to come" (10).

The progressive nature of MND was associated with the experience of a profound sense of loss and unfolding changes. Participants described the experience of loss and grief across varied facets of life, gradually changed by the patient's condition. One area of loss was in regard to individual identity and self-concept. For instance, as the condition progressed and caregiving duties increased, some caregivers experienced loss in regard to their vocation. One participant stated "I've gone from.. a full on job.. to nothingness in many ways" (01). Further, some had safety concerns about leaving the person with MND and experienced loss related to their personal interests, demonstrated as follows: "you lose an awful lot of freedom" (02); "I'm a reader…knit and sew but I haven't been doing any of those kind of things lately" (03).

The Labyrinth—difficulty finding a way through disease progression

Participants described their perception of the uncertain nature of the progression of MND, and how it

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emotional concerns for which they were responsible, such as making appointments, arranging medical needs of their spouse and attending to other daily tasks of caring (e.g. preparing food, daily cleaning duties, helping with showering and shaving). The participants also noted their responsibility for transferring and repositioning the person with MND, often throughout the night, which resulted in very little sleep for the family carer. These duties were taken up by each carer without personal regard to their own needs for sleep, recreation or exercise and were maintained even when the carer was unwell or exhausted. One participant described:

> I think you're traumatised, quite frankly, I think you're traumatised. All the symptoms of trauma – numb[ness] and shock and all of those things. (F3)

The change in relationship from spouse to family carer

The participants described the impact that being a family carer had on their intimate relationship. Some participants indicated a sense of regret when they reflected on their change of role as going from a wife/ husband to a 'nurse' or 'carer', whilst others spoke about the role change in a factual tone, suggesting that is was just 'what you had to do'. One participant stated, 'The wife role was gone and I was a nurse and that's what I regret' (F1) while another declared, 'It ruined our marriage; you're no longer a wife, you're a carer' (F5). This change was often a sudden shift, with one participant noting, 'One minute I was working, the next I was a full-time carer' (F9).

The change in role also brought the opportunity to learn new tasks and duties that previously were the realm of the care recipient. For instance, one carer proclaimed, 'I've learnt some new skills, like washing up!'(M1) and another stated, 'He always made the major decisions [so] I had to learn the finances' (F10). The participants also described how their days became busier and longer as they undertook more of the responsibilities that were once their partners'. All of the participants described how the change from spouse to carer had a negative impact on the level of relationship intimacy. For instance, one participant recalled the following incident:

> I turned around and I said, 'Give me a kiss' and I thought at the time he was denying me a kiss, but I realise now he had no muscles in his face to kiss me with. I backed off and thought, oh hello, it's not that you've lost feelings for me; it's just that you can't. (F1)

This decrease in intimacy was often attributed to not only being a carer but also feeling exhausted from all the duties this new role required. One participant stated:

> I don't feel guilty about it but I do think maybe I could have shown him a little bit more love. But maybe my way of attending to him was my love. (F6)

Family caring as a series of losses

There was a clear sense that participants thought MND presented a constant series of losses for the care recipient, with changes occurring on a daily basis. One carer described the progression as, 'it was just all bang, bang, bang, because every day [it was] something else' (F10) and another stated her husband 'was in a wheelchair within about four, five weeks, then he was in an electric wheelchair [and then] his breathing went' (F2). As a result, the carers recounted that they did not have the time to recognise the increasing amount of time they were dedicating to their role.

Feelings of hopelessness were reported by 11 carers, including all who met the criteria for prolonged grief. The hopelessness was described by one participant as 'the most defeating thing'. The sense of hopelessness that participants felt seemed to be compounded further when they contrasted MND to cancer. For example, one stated, 'With cancer there is hope; with MND there is nothing' (F4) while another declared, 'if it had to be a choice between cancer and MND, I'd say cancer any day' (F7).

Coping mechanisms of family carers

The prospect of a dying partner was frightening for all of the participants and the coping mechanisms they reported differed according to their PG-13 scores. Those participants who did not meet the criteria for prolonged grief generally indicated in their interviews that they accepted the notion of their partner dying, right from their diagnosis. For instance, one stated, 'From the start, definitely, yes. Had no problem with that in the sense of accepting, you know, we'd had a really good life, we were both in our sixties' (F2) while another commented, 'It's a death sentence, and it was how long it was going to be, and what sort of course it was going to take' (M1).

In contrast, five out of the six participants with prolonged grief described difficulty in accepting their partner's illness as terminal. These participants tended to reject all notions of their spouse dying, with some describing it as their only way of coping. For instance one recalled, 'I didn't accept that. I just kept thinking,
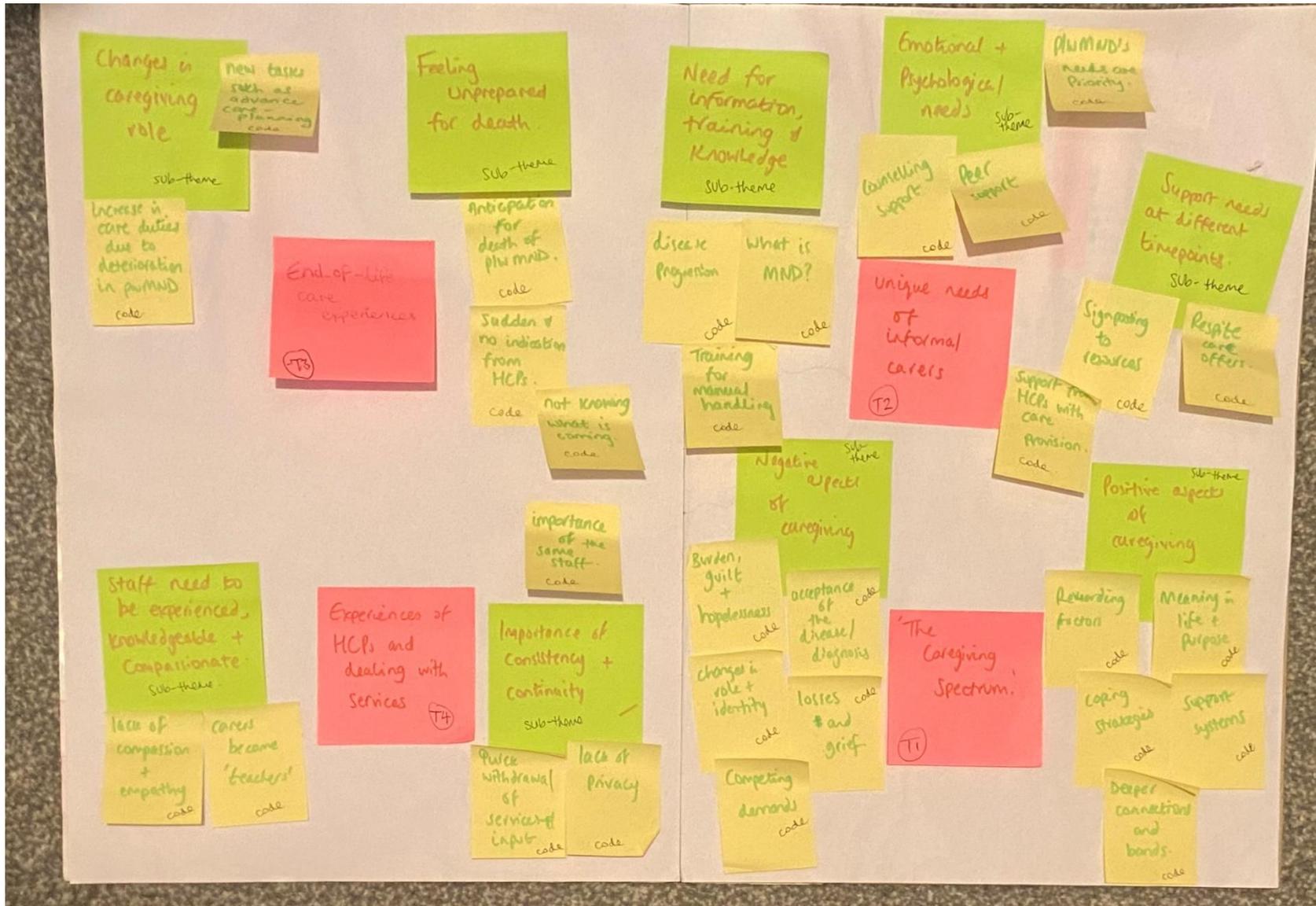
Section Two: Empirical Project

Help-Seeking Behaviour in Informal Carers of People Living with Motor Neurone Disease

(MND).

Abstract

Objectives

Research has highlighted that informal carers of people living with Motor Neurone Disease (plwMND) experience psychological distress and difficulties related to their caregiving role and experiences. However, little is known about the help-seeking behaviours of informal carers. The current study aimed to ascertain factors that may contribute to informal carers seeking help for their own difficulties whilst highlighting any potential barriers.

Methods

The study employed a qualitative design using thematic analysis. The participant sample consisted of twelve informal carers who took part in one-off semi-structured interviews.

Results

Analysis identified four main themes and five subthemes: "physical, practical, mental and emotional impact on carers", "dominance and prioritisation of the needs of plwMND", "where, when and how carers *do* seek support", and "lack of knowledge and understanding from healthcare professionals (HCPs)".

Conclusions

Participants reported prioritising the needs of their loved one over their own which often prevented them from seeking help for their own difficulties. Most informal carers had poor previous experiences with services and HCPs, discouraging them from future help-seeking. However, carers voiced the importance of services "making the first move" to ease mental load and burden and promote engagement.

Practitioner Points

- Findings highlight HCPs and service providers need to develop a deeper understanding of the unique needs of informal carers.

- Further training should be considered for HCPs as carers require practical and respite support from HCPs they trust are experienced and understand MND.

- Increased input is needed during early phases i.e. when the disease is first diagnosed.

- Services should consider ways of encouraging carers to recognise their own needs and seek help using methods such as Motivational Interviewing.

Keywords

Informal carers, experiences, help-seeking, barriers, Motor Neurone Disease, thematic analysis, qualitative research.

Introduction

Motor Neurone Disease (MND) is a rapidly progressing and fatal condition that affects the nerves in the spinal cord and brain that control muscle movement (MND Association, 2022). At any one time, MND can affect up to 5,000 people in the UK and there is a 1 in 300 risk of getting MND across a lifetime (MND Association, 2022). Due to its impact on the brain and spinal cord, MND causes a range of physical difficulties such as muscle weakness, difficulty swallowing and breathing, speech and communication difficulties and pain or discomfort (MND Association, 2022).

The physical symptoms of MND are well documented within the literature (Merrilees et al., 2010; Mitsumoto & Rabkin, 2007) and more recently, the psychological impact of MND on people living with MND (plwMND) is being extensively researched. Studies have found plwMND experience depression and anxiety (Clarke et al., 2005; Goldstein et al., 1998), particularly when they are facing the "uncertain path" of diagnosis (Mistry & Simpson, 2013). Coping strategies such as avoidance and denial are often developed (Zarotti et al., 2019), leading to a declined quality of life (Lee et al., 2001). Averill et al. (2007) found plwMND experience hopelessness and demoralisation which can be a strong indicator of suicidal ideation (McLeod & Clarke, 2007).

Alongside plwMND experiencing the psychological impact of MND, family members and friends of plwMND also unsurprisingly face difficulties when caring for plwMND (Goldstein & Leigh, 1999). They encounter other challenges as they adjust to the losses and changes in their family member and in their own lives, including "letting go of time for themselves" (Goldstein & Leigh, 1999). Although the research in this area is limited, the research that has been conducted with informal carers of plwMND has provided insight into the emotional

distress carers face such as burden, guilt and struggling with acceptance, isolation and prolonged grief disorder (Weisser et al., 2015; Aoun et al., 2011).

Although some awareness has been raised of the psychological impact on informal carers, gaps remain in affirming what the needs of informal carers are and how best to support them (Thomas et al., 2010). Love et al. (2005) recognised that the available informal support for MND carers was often inadequate or unsatisfactory as the disease progressed. Focus has been given to peer and social support for informal carers rather than specific support for carers provided by health and social care agencies (O'Brien et al., 2012).

Studies have highlighted the burden of caregiving in MND and the need for tailored psychological intervention and support for informal carers (Aoun et al., 2013; Baxter et al., 2013). There has been little investigation into how to tailor approaches for informal carers, how informal carers can access this support, and most importantly, what barriers informal carers face when attempting to access support. Cafarella et al. (2022) conducted a systematic review of thirteen studies exploring interventions designed to improve the psychological wellbeing of carers of plwMND. High attrition rates were described; only four out of the thirteen studies reported percentages of above 80% for intervention completion. Furthermore, the authors found the degree to which the specific needs of the carer were addressed within the interventions varied across the studies. Two of the studies focused primarily on the needs of the patient, with the psychological wellbeing of carers as a secondary focus. It was unsurprising that these studies described no or less improvement in the carers' psychological wellbeing compared to that of the patient's (Bentley et al., 2014; Creemers et al., 2014).

The research has demonstrated that informal carers of plwMND have specific needs that are longstanding and often unmet, and it is important to consider factors that may contribute to the perception carers have of available psychological support. Factors that have been under-

researched but are important to note are help-seeking attitudes and behaviour, and barriers carers may face in the context of help-seeking. Despite limited research with informal carers, a small number of studies have explored factors that may improve help-seeking behaviour more generally. Tomczyk et al. (2020) investigated the intention-behaviour gap in help-seeking for currently untreated depressive symptoms. This research indicated that if individuals perceived professional help as positive and it was recommended by those in their close environment, there was a higher likelihood of them seeking help. Challacombe and Halpin (2021) explored whether encounters of carers with mental health services affected their help-seeking attitudes and influenced their stigma towards mental health difficulties. Positive carer experiences of mental health services were associated with positive help-seeking attitudes whilst poor experiences suggested carers were less likely to seek help in the future, due to increased stigma and/or negative affect encountered within the environment. It is evident that further explorations into the help-seeking behaviour and attitudes of informal carers of plwMND are required to ensure accessible, specific support is available.

The present study will investigate help-seeking behaviours using components of the Theory of Planned Behaviour (TPB; Ajzen, 1991) and the Adapted Hope Enablement Model (HEM; Soundy et al., 2014; Soundy & Condon, 2015) as the underpinning theoretical frameworks and to guide the project. The TPB (Ajzen, 1991, 2002) identifies three core components, "*subjective norms, personal attitudes towards health behaviour and perceived behavioural control*" (Ajzen, 1991; figure 1) that can help predict intention, attitudes and lead to effective help-seeking behaviour (Tomczyk et al., 2020). The Adapted HEM (figure 2) identified factors that may influence hope within plwMND, such as initiating in social interaction, and responses that disable hope, such as disengaging from others and isolation (Soundy & Condon, 2015). The current project seeks to understand such factors in order to further understand help-seeking

behaviour in informal MND carers, examining behaviour and intentions, as well as understanding whether hope influences help seeking.
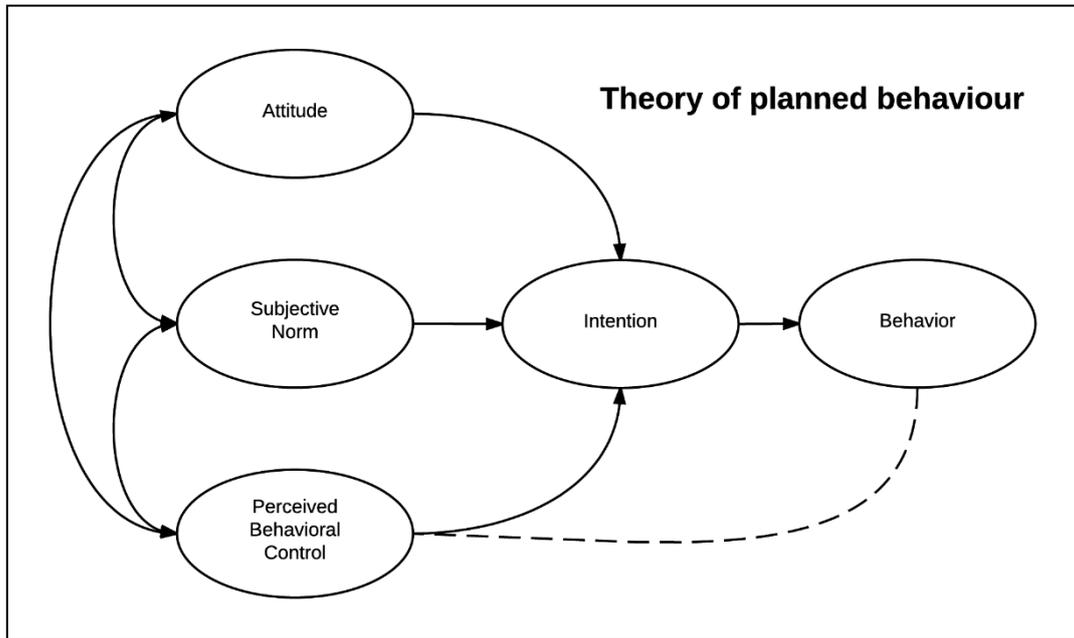

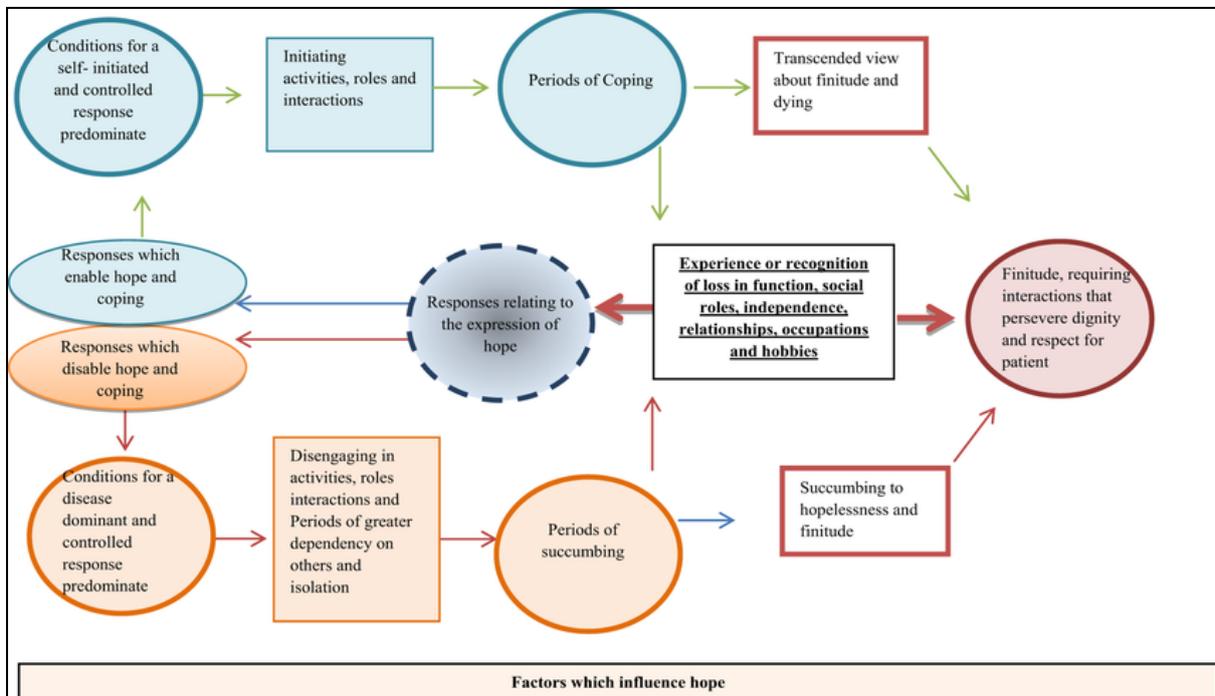***Figure 1****: The Theory of Planned Behaviour (Ajzen, 1991)*

Figure 2*: The Adapted Hope Enablement Model (Soundy & Condon, 2015)*

The use of qualitative methods of analysis and data collection can provide rich, meaningful data and are commonly employed within the research of informal carers of plwMND (e.g. Aoun et al., 2011). Thematic analysis (TA; Braun & Clarke, 2006) is a qualitative method of describing and analysing data that involves the interpretation of processes and constructing themes (Kiger & Varpio, 2020). The current project will utilise TA to identify the gap in the literature concerning informal carers of plwMND. Further research is required to understand factors that contribute to informal carers of plwMND seeking help for their own psychological difficulties, and to ensure support is accessible and meaningful.

Aims

The present research aims to explore and ascertain factors that contribute to informal carers of plwMND seeking help for their own psychological and emotional difficulties and the barriers they face when seeking help.

Method

Research Philosophy

Due to the qualitative nature of this research, extracting and making sense of the experiences of informal carers of plwMND, the researcher adopted a '*critical realist*' approach, acknowledging that participants' experiences of the interviews are situated within their own social framework and locating the researcher within this context (Braun & Clarke, 2022; Pilgrim, 2014). A '*critical realist*' approach effectively captures participants' experiences of their own narratives, recognising that their personal "reality" may not represent the experiences of others. It acknowledges the way this "reality" is expressed can be influenced and altered by the interview process and the researcher's interpretations (Pilgrim, 2014; Sims-Schouten et al., 2007). In this context, language is seen as intentional (Hall, 1997); we have a unique perception of the world and we use language as a means to communicate our stories.

Design

TA was selected as an appropriate qualitative approach in the current study to evoke meaningful data relating to the aims of the research and pinpoint recurring themes from participant responses.

The rationale for TA as the chosen design relates to its ability to allow us to take a pragmatic approach, bringing in theory from previous research to guide the types of questions that are asked and to inform analysis, while still keeping the focus on the reports of MND carers. TA allows flexibility and the drawing out of themes that are raised across individual participants to try to understand help-seeking in this under-researched group. Previous studies involving informal carers of MND have also utilised TA; Anderson et al. (2016) explored the experiences of informal carers of plwMND and identified three key themes following one-off semi-structured interviews: "*The Thief*" defined losses in carers' lives*, "The Labyrinth"* defined how carers navigate disease progression, and *"Defying Fate"* defined ways in which carers find hope and resilience. This is in keeping with the aims of the current study as one-off semi-structured interviews were conducted however the current study differed from Anderson et al's (2016) study as there was an added focus on establishing the help-seeking behaviours of informal carers of plwMND.

Interview questions used within the interview schedule were informed by the psychological models presented in the introduction – the Theory of Planned Behaviour (Ajzen, 1991) and the Adapted Hope Enablement Model (Soundy et al., 2014; Soundy & Condon, 2015). However, the models were not used to inform the whole design of the current study and they primarily informed the development of the interview questions.

Participants

The current project recruited adult participants who are a current or former informal

carer of a person living with MND. Inclusion and exclusion criteria (outlined in Table 1) were explicitly outlined in the study advert (Appendix A) and information sheet (Appendix B).

Inclusion Criteria	Exclusion Criteria
Over 18 years of age	Former (informal) carer of a person or plwMND over 3 years ago
Former (informal) carer of a person or plwMND – must be within the last 3 years (*part-time or full-time*)	Any individual who cares for plwMND professionally, for example, a MND nurse
Current (informal) carer of a person or plwMND (*part-time or full-time*)	Not fluent in English

Table 1

Inclusion and exclusion criteria applied when recruiting participants

There is no defined sample size in the literature for TA. The majority suggests smaller sample sizes are sufficient for data collection in qualitative studies using TA, with an emphasis on 10-12 participants (e.g. Ando et al., 2014; Guest et al., 2006). Guest et al. (2006) found data saturation (Glaser & Strauss, 1999) had occurred by the 12th interview. However, the concept of data saturation has been criticised as it demonstrates a low level of transparency regarding sample sizes, with little specification of how saturation was actually assessed (Carlsen & Glenton, 2011; Mason, 2010). Therefore, in the current study, the concept of information power (Malterud et al., 2016) was used to assess qualitative data throughout the research process and

whether more participants should be recruited. A study is deemed to have sufficient information power if study aims are narrow, the sample is specific to the aims, established theory has been applied, there is strong quality of dialogue, and a stringent strategy for analysis is used (Malterud et al., 2016). The current project was deemed to have sufficient information power after 12 interviews.

During the recruitment phase, fifteen participants initially volunteered to take part in the project. Due to drop out and 1 interview not being audible enough to be transcribed, 12 participants formed the final sample (see Table 2 for participant characteristics). Four participants were male and 8 were female. Participants were between the ages of 35 and 76 years. Five participants were former carers and 7 were current carers of a plwMND.

Recruitment Procedures

Participants were recruited in a number of ways to ensure individuals from a variety of backgrounds take part.

Due to their interest in MND research and previous advertisement of projects, the UK charity, MND Association (MNDA), was approached to advertise the project in their newsletter and on their website. The MNDA consists of over 10,000 members including informal carers of plwMND. The MNDA consented to advertising the project and a copy of the project advert was sent the via email. They were asked to include the advert in their regular newsletter to distribute to their members and it was also advertised as a webpage on their website. This method of recruiting participants helped recruit a representative sample as the newsletter and website reached members of the MNDA across the UK.

The project was also advertised on social media platforms (LinkedIN and Twitter/X) of the lead researchers, allowing carers who are not involved with the MNDA to be recruited.

Table 2

Participant demographic information

Participant	Age	Gender	Ethnicity	Country of Residence	Religion	Socioeconomic Status	Carer Status	Time as a Carer	Relationship to plwMND
P1	64	Male	White British	England, UK	Christianity	Retired	Former	7 years	Father
P2	68	Female	White British	England, UK	None	Retired	Current	7 years	Wife
P3	58	Female	White British	Scotland, UK	None	Employed	Current	5 years	Wife
P4	42	Male	White British	England, UK	Atheism	Employed	Current	10-11 years	Son-in-law
P5	47	Female	White British	England, UK	None	Employed	Current	6 months	Wife
P6	37	Female	White British	England, UK	Spiritual	Employed	Former	2 years	Daughter
P7	59	Female	White British	England, UK	Christianity	Retired	Current	3-4 years	Wife
P8	76	Female	White British	England, UK	Christianity	Retired	Former	3 years	Mother
P9	35	Male	White British	England, UK	Atheism	Employed	Current	2-3 years	Husband
P10	53	Female	White British	Scotland, UK	Christianity	Employed	Former	2 years	Wife
P11	56	Female	White British	England, UK	None	Unemployed	Current	10 years	Wife
P12	42	Male	White British	Northern Ireland, UK	Christianity	Employed	Former	4 years	Son

Service User Involvement

The Sheffield MND Research Advisory Group (SMND RAG) were contacted for their involvement in the project as they have previously supported MND research. The SMND RAG consists of plwMND and current and bereaved carers whose feedback was invaluable in the development and adaptation of the interview schedule and project. Two plwMND and two carers, as well as research panel members, reviewed and provided written feedback about the interview schedule (Appendix C), the study advert and the participant information sheet. Changes were made to the aforementioned accordingly.

Procedure

Despite most qualitative studies involving TA using face-to-face interviews (e.g. Anderson et al., 2016), it was expected that most informal carers recruited in this study will have current caring responsibilities and may find it difficult to meet face-to-face at a designated location. Therefore, semi-structured interviews were conducted via Google Meet which is licensed by the University of Sheffield. If participants were unable to use Google Meet, a telephone conversation was offered. A total of 11 interviews took place via Google Meet and 2 interviews took place over telephone.

Once participants came forward to take part in the project, they were sent a link to complete a demographic form (Appendix D). Participants who met the inclusion criteria and had completed the demographic form were emailed the participant information sheet and consent form (Appendix E) to complete and return, which included details about the interviews being recorded and shared with a transcriber. Following the completion of consent forms, an interview time/date was scheduled.

Each interview lasted between 45 minutes to one hour. The author confirmed with participants at the beginning of each interview that they were happy for the interview to be recorded. Each

interview adhered to the interview schedule and following completion, participants were offered a debrief of the project (Appendix F) and the option for further support and resources was available.

Data Collection

The interviews were recorded using Google Meet's recording option, including the recording of telephone interviews. Recorded interviews were saved on the researcher's personal and secure Google Drive which can only be accessed by designated and approved professionals i.e. the researchers involved in the project.

A university-approved transcriber was contacted via email and agreed to transcribe the 12 interviews. The transcriber read through instructions and signed the confidentiality agreement (Appendix G). A secure folder was created on Google Drive that could only be accessed by the researchers and the transcriber.

Data obtained from the interviews was stored in accordance with the University of Sheffield's data security policies and GDPR guidelines.

The transcriber emailed completed transcripts to the author, ensuring all documents were password-protected and encrypted to maintain confidentiality. To reduce the likelihood of human error, completed transcriptions were checked against corresponding audio files by the author.

Data Management Plan

Audio files and transcribed interviews were stored on the author's secure Google Drive. Transcript data was stored according to the University's data security policy, however audio files were deleted from Google Drive once they had been transcribed. All data will remain

anonymous without any identifying participant information as participants were assigned a participant number.

A digital data management plan (DMP) was created on "DMP Online" (Appendix H) and was shared with supervisors. This was regularly reviewed and updated during supervision.

Ethical Approval

Ethical approval for the study was obtained from the University of Sheffield Research Ethics Committee (Reference Number 051051; Appendix I).

Quality and Rigour

To ensure rigour, quality control and credibility of the research, the researcher adhered to the quality standard guidelines outlined by Elliot et al. (1999), as shown in Table 3.

Table 3

Elliot et al.'s (1999) quality standard guidelines

Guideline	Application to current study
Owning one's perspective	The lead researcher addressed their own position, thoughts and biases within a reflexive statement (Appendix J) and a reflective log was kept throughout the research process (Appendix K).
Situating the sample	Demographic information for the participant sample is presented (Table 2) to allow the findings to be understood within the context of participants' individual situations.
Grounding in examples	Direct quotes ("raw data") from participant interviews are provided throughout the results section of the report to support themes.

Credibility checks	Service user involvement allowed for feedback on the interview schedule and study documents such as the study advert and participant information sheet. Changes were made to relevant documents. The first interview recording was discussed in research supervision with supervisors who provided feedback about interviewing style. Initial ideas, codes and themes were discussed and reflected on with research supervisors. Member checking was not conducted as it does not align with the 'critical-realist' stance taken by the researcher however the author acknowledges this is a limitation of the study.
Coherence	Analysis methods and steps are clearly detailed in the report and evidence of theme development is provided (Appendices L, M, N). Results are reported in a succinct and concise manner. Themes and subthemes are outlined in a table for ease of access.
Accomplishing general vs. specific research tasks	The findings of this study are limited to the experiences of the carers who participated which represent the perspectives and help-seeking behaviours of informal carers from a White British background. The researcher acknowledges generalisation of the findings would not be appropriate. Broader implications and limitations of this are discussed in the 'discussion' section of the report.
Resonating with readers	Clinical practice implications and considerations for future research are discussed in the 'discussion' section.

Analysis

Coding and Analysis

Following the completion of interviewing and transcribing, the author analysed the findings using the six TA steps outlined in Table 4. Earlier phases were revisited to enable enrichment and development of later stages. Despite Braun and Clarke (2006) presenting the phases as distinct and in a linear style, the researcher treated the analysis process as cyclical and reflective, shifting between steps and phases as required within the process.

Table 4

Thematic analysis steps undertaken by the researcher

Thematic Analysis Phase	Steps taken at this phase
Phase 1 - familiarisation	The researcher immersed themselves in the data, reading and re-reading transcripts, to understand its depth and nuances. Familiarisation notes were taken at this point to note down areas of interest within the interview transcripts.
Phase 2 – generating initial codes	After familiarisation with the data, the researcher systematically coded the data by identifying key information that stood out to the researcher and providing a meaningful, interpretative label. The researcher adopted a "bottom-up" and "data-driven" approach, allowing the data to drive themes and interpretations.
Phase 3 – searching for themes	Generated codes were categorised into potential themes, or patterns of meaning, which consisted of relevant data extracts. A visual representation in the form of a thematic map was used to assist theme generation which included the production of four main themes and five sub-themes.
Phase 4 – reviewing themes	Generated themes and sub-themes were then refined and checked against the dataset to ensure they accurately represent the data.
Phase 5 – defining and naming themes	The final themes were then named with labels that clearly defined the essence of each theme and determined how they fit into the overall narrative. For each theme, a detailed analysis was conducted, considering how it fits into the broader "story" being told about the data in regards to research aims/questions.
Phase 6 – producing the report	The final part of interpretation included the development of a report of a full set of refined themes. The write-up used an 'analytic narrative' that described the data and supported the research aims (Braun & Clarke, 2006).

Reflexivity, defined as "*a process of self-examination, revealing ourselves as individuals and researchers whilst understanding how our personal biases may influence the research process*" (Berger, 2015; Creswell, 2014), is an important aspect of qualitative research that must be considered when conducting TA. There is potential for data to be influenced by the researcher's views, what is asked and how, what is found, and how this information is interpreted. It is recommended for the researcher to be up-front about views and opinions to ensure a degree of openness and integrity, demonstrating transparency which can increase rigour (Anderson, 2008). Therefore, the lead researcher is a British Trainee Clinical Psychologist from a South Asian background with no previous experience of MND or working with informal carers (see Appendix J for reflexive statement). Within the current project, the author utilised a reflective log to enable transparency and reduce researcher bias, recording preconceptions and personal influences and finding opportunity to discuss these during confidential research supervision meetings (see Appendix K for excerpts from the reflective log).

Results

TA was conducted on the entire dataset of 12 interview transcripts by the researcher. Four themes and five sub-themes were identified, as shown in Table 5. Participant contribution is illustrated in Table 6.

The core themes that related to the help-seeking behaviours of carers of plwMND were identified as:

- "physical, practical, mental and emotional impact on carers",

- carers emphasising the "dominance and prioritisation of the needs of plwMND",

- demonstration of "where, when and how carers *do* seek support",

- highlighting a "lack of knowledge and understanding from healthcare professionals (HCPs)".

The physical, practical, mental and emotional impact of caring on carers was explored at a deeper level through identifying the "emotional and mental effects of caring" and the "physical impact and practicality of caring". Demonstrating where, when and how carers seek support was achieved by understanding carers' "perception and experiences of services and offers of support", identifying the "suitability of available support and barriers to access", whilst acknowledging that carers may seek support from different sources such as "support networks formed with other carers/non-professionals".

Table 5

Themes derived from thematic analysis of dataset

Theme	Sub-themes
Physical, practical, mental and emotional impact on carers	Emotional and mental effects of caring Physical impact and practicality of caring
Dominance and prioritisation of the needs of plwMND	
Where, when and how carers *do* seek support	Perception and experiences of services and offers of support Suitability of available support and barriers to access Support networks formed with other carers/non-professionals
Lack of knowledge and understanding from HCPs	

Table 6

Participant contribution to main themes and sub-themes

	P1	P2	P3	P4	P5	P6	P7	P8	P9	P10	P11	P12
Physical, practical, mental and emotional impact on carers	X	X	X	X	X	X	X	X	X	X	X	X
Emotional and mental effects of caring	X	X	X	X	X	X	X	X	X	X	X	X
Physical impact and practicality of caring		X		X	X		X	X	X	X		
Dominance and prioritisation of the needs of plwMND	X	X	X	X	X	X	X	X	X	X	X	X
Where, when and how carers *do* seek support	X	X	X	X	X	X	X	X	X	X	X	X
Perception and experiences of services and offers of support	X	X	X	X	X	X	X		X	X	X	X
Suitability of available support and barriers to access	X	X		X	X	X	X	X	X	X	X	X
Support networks formed with other carers/non-professionals		X	X		X	X	X	X	X	X	X	X
Lack of knowledge and understanding from HCPs	X	X	X		X	X	X	X		X	X	X

1. **Physical, practical, mental and emotional impact on carers**

Participants acknowledged the significant impact of caring for a loved one with MND and what they initially perceived caring would be like.

"*I think the thought of it you know was like (SIGH), this is gonna be awful, I don't mean from my point of view but just him as a person, I knew it was gonna be difficult.*" P3

Some participants stated that they did not realise when they fell into a 'caring role' for their loved one as it was a quick transition.

"*there wasn't like a single point when I became aware that I was caring for him apart from when he went into hospital and then like suddenly I was going to and from the hospital as well.*" P5

Participants addressed the various ways in which caregiving has impacted them, including their physical, emotional and mental wellbeing, as well as highlighting practical issues. These are discussed in the sub-themes below.

Emotional and mental effects of caring

Many participants shared that seeing their loved one decline due to MND affected their emotional wellbeing, witnessing the progression of the disease firsthand.

"*very hard emotionally…seeing who you love…deteriorating in front of your eyes, watching their struggle…absolutely horrendous.*" P6

Uncertainty about the trajectory of the disease caused emotional and mental distress as carers were unsure about the disease itself and could not anticipate its progression.

"scary, I would say fear more than anything with not really knowing what was coming, knowing it was going to be horrendous…not understanding, yeah lack of knowledge really." P7

Some participants described the emotional load of caring for their loved one and ensuring the wellbeing of other family members such as children and spouses.

"I guess we kind of wanted to not be too panicky because my son, it will be upsetting for him." P5

"I worry about my wife and whether, how she's coping mentally…she's not one that shows her emotions a lot, I think she bottles a lot up." P1

Since beginning caring for their loved one, participants shared that they had lost a part of their identity, and positive outlets such as coping mechanisms and self-care were no longer a part of their lifestyle, resulting in reduced wellbeing.

"I've more often not done things for myself and it's a way of either saving time or energy." P9

"to part-time work and then you know to obviously not working…I used to go to the gym a lot and…I trained to do some triathlons…I did love that…when he was diagnosed…I felt tremendously guilty about going for a run." P11

"you lose who you are, you do." P2

Physical impact and practicality of caring

Participants spoke about the practical elements of caring and the physical changes they noticed had developed or exacerbated health conditions.

"I got very sore hands, like my wrists and my thumbs and my fingers, and even though he passed away in July, they're still sore and they've not got back to normal." P10

"my running, athletic fitness has dropped and I've picked up quite a lot of injury, chronic injury through handling." P8

Participants emphasised how much time they spent caring, with most stating they offered care for up to 24 hours a day. Practically, this meant many carers did not have time for themselves or meeting their own needs.

"I feel like I don't have any time, like he's gone out with his mum to do something and it's like "oh he's gone out for like an hour, so that's like an hour to myself", but I don't get much time, like that's what I struggle with." P5

2. Dominance and prioritisation of the needs of the plwMND

Carers expressed that the needs of their loved one were more important than their own. This led to a prioritisation of their loved one's needs, and a lack of prioritisation of their own. A phrase that often came up was to *"just get on with it"*; a mindset that many of the participants had adopted to encourage them to keep going.

"taking one day at a time and not trying to think too much about what might happen, just trying to get on with it because there is so much to do." P5

"I just got on with it, there wasn't time to stop and think about…what was going on because my time was taken up with looking after him." P11

Participants highlighted that their loved one being diagnosed with MND was not a straightforward process and dominated the lives and wellbeing of both the plwMND and their carer.

"when he was first diagnosed, they said it was Parkinson's and I said "no, I know it's more than that" so I pushed for them to do some tests…they sent him to do the test for MND…I heard one just say 'MND' that's all I heard." P2

Initially, participants struggled with accepting their loved one's diagnosis and expressed disbelief in hearing the diagnosis and prognosis made by HCPs.

"I've worked in MND for a very long time, I thought…can't possibly be MND…what would the odds be, it seemed impossible." P4

"you can never accept it…you don't expect your children to go before their parents." P1

"it was just a freight train…saying something about life expectancy and I remember him saying two years…most awful thing." P7

Following their loved one's diagnosis, participants frequently worried about the plwMND's physical and mental health, which became a priority for carers.

"you were doing everything you can to try and make her life as comfortable as you could to make sure that she doesn't have to stress on stuff." P1

"I wanted to be with him and help him as much as I could, I just felt so sorry for him." P8

3. Where, when and how carers *do* seek support

"there's probably a lot of things that can help us, but if we don't know that they are there…not really going to ask for them…hard to understand which direction to go." P9

Participants expressed a sense of uncertainty about seeking support and how to do this. They highlighted challenges they faced when attempting to seek support for their own wellbeing, which often took the backseat. This felt reinforced by professionals as carers shared that they were not asked about how they are managing, resulting in them deprioritising their own needs as they were not prioritised by professionals.

> *"in terms of services for carers…the person that you're generally speaking to is there for the person that you're caring for… very rarely ask you how you are or how you are doing." P11*

Perception and experiences of services and offers of support

Participants spoke about the lack of clarity they encountered around the support available to them and what the process was for them to seek help.

> *"I don't know exactly what happens or like how to do it." P5*

Many participants felt uncertain about whether support could help with the specific and unique difficulties they were facing.

> *"never had it really offered through the NHS, like my GP…the only thing they could offer was CBT…not necessarily talking therapy, which I would have preferred…being able to pick a therapist who had experience in like coping and grief…tailored to my needs." P9*

> *"I do suffer mentally…I sit and think about the way he died…hits me more than anything…I don't know how he died…can't see how a counsellor would be able to help me with that." P8*

There was a sense that participants wanted services to 'make the first move' and offer support to help alleviate mental load and psychological burden.

"I know everybody's busy and the health service is under pressure…kind of like you want somebody to physically say "come in and see us"…"we want to see you and let's have a chat"." P1

"if someone at the hospital where [son] was would have said "would you like to have a chat?", I would definitely have said yes." P8

Help-seeking behaviours in terms of seeking support for their own difficulties was influenced by previous experiences of support services and HCPs. Participants shared whether previous experiences encouraged or discouraged them from seeking help in the future.

" I did try and talk to the Marie Curie on my own…she was no help at all…that's what put me off in a way." P8

"I tried to talk to them a couple of years ago about CBT but they didn't send me for it, they gave me a list of websites… felt like I was being fobbed off…discouraged." P12

For many participants, discouragement resulted from their first point of contact when they attempted to seek help, which was usually their GP.

"when I talked to the GP…tried to get help before with depression…it was completely unhelpful." P5

There were mixed impressions about the helpfulness and accessibility of support available to carers, provided by charities such as the MNDA. This influenced perceptions of support services.

"I've had counselling and it didn't help one single bit…I didn't think the counsellor understood at all and she was provided by MND Scotland…specifically for counselling for MND." P10

"much of the support that we've had…has been through…charitable organisations…through the MNDA…massive help from them…awful lot of the practical help has come from there as well." P7

Participants shared that if they were receiving valuable support from charitable organisations, they were less likely to seek support from professional services or NHS-funded organisations.

Suitability of available support and barriers to access

Interviews highlighted that the individual caring situation of carers impacts on their ability to engage with or access support. This also highlighted barriers to accessing support will vary, for example some carers preferred for support to be delivered online so it was more accessible and others wanted sessions to be face-to-face.

"the waiting lists around here are so long…it's not face to face, it's online which is no good because I'm already isolated, I don't want that." P2

Some common barriers were related to time and time management, organising alternative care provisions for their loved one, and feelings of guilt related to accessing support for themselves.

"I think time…there isn't time to fit everything in…making extra time for something like that would be difficult." P5

"you almost have…imposter syndrome on the extent to which you deserve help or deserve support…wouldn't it be strange if I was sitting around on a psychiatrist's couch talking about my father-in-law when my mother-in-law doesn't have that support." P4

Individual differences between participants demonstrated that carers have discrete needs and preferences for support, which can influence how accessible, available and acceptable carers perceive support to be.

Support networks formed with other carers and/or non-professionals

Whilst some carers were aware of the support available to them through professional services, they ultimately found more valuable support through connecting with other carers in similar situations to their own. Carers felt understood and heard by other carers, and emphasised the importance of relatability.

"*the carers in the campaign…I don't have to explain because…they'd know exactly what that meant and how frustrating that was…they would just know…it's a great level and people understand when you say "yeah I'm doing this all the time"…it is relatable." P11*

Participants highlighted how meaningful it was for them to engage with peer support and online forums used by other carers or non-professionals, exchanging advice and helpful tips.

"*one of the best things I found was a group on Facebook…an MND Support Group…over 10,000 members…giving medical advice…practical tips about mattresses, pillows…things that professionals couldn't tell you… really useful." P12*

Others expressed that connecting with family and friends was a valuable resource and an adequate form of support for them, allowing them to talk openly about how they are feeling and offering reminders to look after themselves.

"*my partner…he is very caring, very grounded…don't think I could have done it without him…reminding me of self-care and providing self-care…got some great friends…I talk to these people…because talking is really the best thing." P6*

4. **Lack of knowledge and understanding from healthcare professionals**

Some participants encountered HCPs with limited MND experience that left a lasting impression on carers about how understanding and supportive HCPs could be. A key

commonality between carers was their experience of the delivery of their loved one's MND diagnosis which was often perceived as a negative event.

> "*initially the way it was diagnosed…was very much "you have MND…you've had the symptoms for two years…the prognosis is serious…we think maybe you might have eighteen months"…that's how it was put to us…absolutely shock horror.*" P3

> "*we left that room with no information, all I had was he's got what Rob Burrow has got and he's got two years to live…both of those things were actually inaccurate.*" P7

Elaborating on the lack of understanding and knowledge from HCPs, which included MND nurses, neurologists, occupational therapists etc., participants felt that there was an absence of individualised offers of support, reflecting a 'one size fits all' approach rather than tailoring support to meet the individual needs of carers or their loved one.

> "*They went "leave him in bed until 10" and I went "he works from home…I can't leave him in bed until 10, he starts work at 9"…they went "we don't care, this is what we're giving you"…like it's one size fits all and one size doesn't fit all with MND.*" P10

Discussion

The current study aimed to explore and highlight factors that contribute to how informal carers of plwMND seek help for their own psychological and emotional difficulties. A key aspect of the research was to identify barriers to accessing support and seeking help for informal carers. The ultimate goal of the research was to provide recommendations and guidance for future research involving informal carers and clinical recommendations about how to offer accessible psychological support to this target group.

Physical, practical, mental and emotional impact on carers

This theme emphasises how the impact of caregiving can affect various aspects of a carer's life, including their emotional, physical and mental wellbeing. Carers faced physical and practical burdens to caregiving such as delivering physical care and handling, which sometimes resulted in the development or worsening of physical health conditions.

The prominence of the emotional and mental impact of caregiving was explored in the current study. The Adapted Hope Enablement Model (Soundy et al., 2014; Soundy & Condon, 2015) highlighted responses which can disable hope and coping for individuals, such as disengaging in activities, roles and interactions. This is consistent with findings of the current study whereby carers voiced that they felt a loss of identity as they adopted the informal carer role and "let go" of coping mechanisms and self-care strategies. Research within the field has shown that carers of plwMND have difficulty engaging with self-care due to a lack of time to meet their own needs (Rabkin et al., 2009).

The impact of caregiving for plwMND has been explored within the existing literature, however the evidence base is limited about the extent to which informal carers' mental, physical and emotional wellbeing are impacted. Research investigating how carers are impacted psychologically by caregiving found that depression tends to increase over time as the disease progresses and caregiving burden increases (Gauthier et al., 2007; Goldstein et al., 2006). Witnessing the progressive deterioration of their loved one has a lasting psychological impact on informal carers (Aneshensel et al., 1995) which was voiced by participants as they shared how challenging, long term and unrelenting the role can be.

Uncertainty about the disease itself was communicated by many participants as a contributing factor to the difficulties they faced as they felt left to their own devices to research and find out more information about MND, adding to their already elevated mental load. Carers shared that

adequate information sharing by HCPs about MND could help them manage and cope with caregiving; this has been supported by research which found that carers present a need from the initial diagnosis for more information about MND, and their loved one's diagnosis and prognosis (Hughes et al., 2005; Akiyama et al., 2006).

Dominance and prioritisation of the needs of the plwMND

This theme addresses how informal carers often prioritise the needs of their loved one over their own, emphasising how the disease can 'dominate' their lives. It was a shared belief amongst carers to 'just get on with it', particularly when asked about how they cope with the challenges of caregiving. Carers struggled with acceptance of the MND diagnosis, with many finding the diagnosis process unhelpful and time-consuming.

Existing literature has supported that caregiver time is dominated by the specific needs of the plwMND and how progressed their illness is; Chio et al. (2006) found that the amount of time spent caregiving for plwMND is highly related to overall disability due to the disease. Caregiving time ranged from 5 hours per day for those with mild impairment to 15 hours per day for those experiencing more severe impairment. This is in line with the findings of the current study where most participants expressed that they delivered care for their loved one 'almost 24 hours a day'.

Some carers shared that they had never contemplated or had the intention to seek help for their own difficulties as they were occupied with ensuring the wellbeing of their loved one. This is in keeping with the *'personal attitudes towards health behaviour'* component of the Theory of Planned Behaviour (Ajzen, 1991) as the personal attitude of carers towards seeking help for their own difficulties illustrated de-prioritisation as their own needs, physical and emotional, were not of great importance compared to those of their loved one.

Where, when and how carers *do* seek support

This major theme emphasises the different ways in which informal carers may seek help for the caregiving challenges they face. Key elements included how carers perceive the support available to them, addressing the suitability of available support and highlighting barriers, and the alternative forms of valuable support that carers engage with such as peer support or carer networks.

A number of carers shared that HCPs failed to ask them how they are managing and coping, which reinforced their belief that they need to focus on the needs of their loved one rather than engaging in support for themselves. Research has found that recognising the crucial role informal carers have in the lives of plwMND, and outlining the increased possibility of psychological distress, can help lower the boundaries for carers to accept offers of support (de Wit et al., 2018).

In line with previous literature, participants were more likely to access support if it was recommended by someone in their close environment, and was perceived as a positive experience. However, the opposite of this was also true as carers were discouraged from seeking help if they had either had a negative personal experience of services or a negative experience was shared by someone they know. Tomczyk et al. (2020) highlighted that poor experiences of mental health services were linked to reduced help-seeking behaviours in carers whilst Challacombe and Halpin (2021) found positive personal experiences of services was associated with increased help-seeking behaviours. This was a vital finding within the current study as previous experiences of services could impact future help-seeking behaviour. Future research would benefit from exploring this in depth, particularly ways to mitigate the effects of previous negative experiences.

Participants voiced a need for tailored support in order for them to be more willing to access available support as each carer has unique needs rather than a 'one size fits all' approach. This need has been expressed in previous studies (de Wit et al., 2018), suggesting a personalised intervention for carers would be beneficial. One adaptation that could be made is more frequent and timely carer assessments to be incorporated into available support to ensure carer needs are met. Ewing et al. (2020) determined the acceptability and suitability of a Carer Support Needs Assessment Tool (CSNAT) to identify the specific needs of carers and adapt intervention accordingly. The authors found that carers had extensive support needs which were appropriately assessed by the CSNAT domains and it was deemed a relevant tool to assess carer needs. This would be a useful addition when tailoring support for informal carers and could assist with the uptake of support available through professional services as it was important for carers that their voices were heard.

Barriers to accessing support or help-seeking for informal carers have been explored in the existing literature however the evidence base is limited. The current study ascertained several factors that can pose as a barrier to help-seeking for carers, such as time and time management, location and format such as online or face-to-face support, practical issues such as organising care for their loved one, and long waiting times. This coincides with the existing literature that found 50% of carers did not participate in a mindfulness intervention due to encountering barriers such as location, being unable to leave their loved one and their own health conditions (Ugalde et al., 2018). The barriers that carers faced were influenced by the individual caring situation and future research would benefit from exploring barriers at an individual level to promote access to support.

Lack of knowledge and understanding from healthcare professionals

This theme highlighted how experiences with HCPs, particularly negative experiences, either at the time of diagnosis or when delivering care for their loved one, influenced whether carers would be willing to seek help for their own difficulties from HCPs and services.

Carers shared that the delivery of their loved one's diagnosis of MND was often cold, blunt and with little understanding or compassion. The diagnosis process was often long and unrelenting, further impacting the emotional and mental wellbeing of informal carers. Previous research conducted has found that carers experienced high levels of distress when it took time for them to obtain a diagnosis and when HCPs and specialists lacked empathy and communication (Aoun et al., 2011; Aoun et al., 2006). Some carers felt left to their own devices and with little information or knowledge about the disease as it was their first experience of MND; the information needs of carers were not met, in line with previous literature which has emphasised a need from initial diagnosis to provide carers with more information about MND, diagnosis and prognosis, and about MND outcomes (Hughes et al., 2005; Akiyama et al., 2006). Carers stressed the cruciality of training for HCPs who had limited knowledge of MND, a finding that is supported by the existing evidence base that calls for professional healthcare providers to provide training to HCPs to improve their communication of MND to the plwMND and their family and to have useful information to hand (McDermott & Shaw, 2008; Chio et al., 2006).

Limitations and future research directions

The present study has provided valuable insight into the help-seeking behaviours of informal carers of plwMND including potential barriers carers may face, however the project is not without its limitations which must be addressed.

Firstly, the current study excluded former carers of plwMND who had cared for a loved one over 3 years ago. Whilst the rationale for this decision was clear, to ensure that caring experiences were recent enough for participants to recall in an interview, it is worth noting that a large population of carers may have been missed due to the constraints of the project. This impacts the generalisability of findings as they may not represent all informal carers of plwMND, particularly former or bereaved carers who cared for a loved one over three years ago. The authors are mindful about making overly generalised comments about the findings in relation to all informal carers, and firm conclusions for the entire caregiver population for plwMND would not be appropriate. Future research would benefit from exploring the help-seeking behaviours of former carers who had cared for a loved one over three years ago, to ensure a more holistic representation and depiction of carer needs is presented.

It was beyond the scope of the current project to include informal carers who were not fluent in English and they were therefore excluded. This may have limited the representation of the findings as it is not known how non-English speaking carers' experiences of help-seeking within this context would have differed or been similar to English-speaking carers. If viable, future research should consider including non-English speaking carers to ensure they are appropriately represented within this important field.

Additionally, the sample of informal carers within the current study were all from a White British background. This was surprising as the UK population encompasses individuals from a range of ethnic minority backgrounds; the 2021 CENSUS data from the Office for National Statistics (ONS; 2021) reported 81.7% of the UK population is from a White background whilst 18.3% belong to an ethnic minority group. The apparent lack of representation of these groups may suggest the findings are not wholly representative of the entire UK population. One reason for this potential underrepresentation may be lack of outreach or engagement; future research would benefit from exploring ways to reach out to ethnic minority groups for their involvement

in carer-related research to ensure their voices and perspectives are being heard. Adopting a purposive sampling technique could also allow for a more diverse participant sample.

Finally, a critique of qualitative research is that it can be influenced by the backgrounds of the researcher and participant, and the relationships and interactions between the two can impact findings (Pelias, 2011). To counteract this, a number of steps were taken by the lead researcher; most importantly, the researcher acknowledged their positionality within the research and utilised reflexive methods throughout to ensure transparency and a more authentic account of the research (Corlett & Mavin, 2018).

Implications for clinical practice

The current project raised several implications to consider in clinical practice pertaining to informal carers of plwMND.

Participants highlighted the lack of available tailored support and voiced concerns about their needs being missed or ignored by HCPs and services. At a service level, professionals should consider utilising tools such as the CSNAT to capture the unique needs of carers and develop interventions accordingly. Ensuring such an approach becomes regular practice would allow carer needs to be addressed regularly and appropriately and could encourage active help-seeking behaviours.

Participants emphasised the high levels of psychological distress they experience during the initial stages of caring i.e. when the disease is first diagnosed, and a need for increased input. Services should consider increasing contact during these early phases to ensure adequate support is being provided to carers. Signposting (or facilitating) useful resources such as peer support groups could be helpful for carers, particularly as the majority voiced that they found comfort and solidarity amongst other carers in similar circumstances to their own.

Additionally, carers emphasised the need for services to 'make the first move' as they are already experiencing mental burden. Whilst HCPs and services would benefit from building awareness about this need and considering outreach approaches that could guide carers to engaging with services, most may feel already over-stretched and not have the capacity to 'make the first move'. It is worth considering how undertaking a more systemic approach to services could lead to thinking beyond individualistic approaches such as determining how to meet the needs of informal carers and over-stretched services and HCP's. Clinical psychologists have a vast amount to offer and could bridge a gap between carers and HCPs by delivering training, teaching, supervision, consultation, or community psychology approaches.

A key consideration from the present study is that carers appear to lack faith in HCPs if they believe they have little knowledge or experience of MND. Perhaps carers need more practical and respite support from HCPs they trust are experienced and understand MND, which in turn could relieve burden and provide them with time to participate in activities and to access support.

Finally, it was clear that carers do not attend to or even acknowledge their own needs, which take a backseat as they focus on the needs of their loved one. When looking at the help-seeking behaviours of carers through a theoretical lens, particularly the Transtheoretical Model (TTM; Prochaska & DiClemente, 1983), it was evident that most carers were at the 'precontemplation' stage of change and were reluctant to engage in support for themselves despite voicing a clear need. Knowing this, it would be essential for professionals and services to consider strategies that could assist carers with moving from 'precontemplation' to 'contemplation', allowing them to reflect on seeking help for their own difficulties. One way of implementing this could be to utilise Motivational Interviewing techniques (MI; Miller & Rollnick, 2013); research has shown that MI can successfully help individuals move from precontemplation to

contemplation, preparation and action stages of change (DiClemente & Velasquez, 2002). Utilising such methods with informal carers may promote help-seeking behaviours.

Conclusion

This study explored the help-seeking behaviours of informal carers of plwMND and highlighted factors that encourage or discourage carers from seeking help for their own difficulties. Informal carers are often uncertain about how to seek help, expressing a need for HCPs and services to 'make the first move'. Tailored support to the unique needs of informal carers was demonstrated as lacking and carers lack faith in HCPs with limited MND experience. The study emphasised how carers prioritise the needs of their loved one over their own and often do not consider seeking help which future research should explore further. Overall, this study provides valuable insight into the help-seeking behaviours of informal carers and has useful clinical implications and applications.

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Appendix A – Study Advert

PARTICIPANT INFORMATION SHEET

DATE: 09/08/2023

Project title

'Help-seeking behaviour in informal carers of people living with MND'

Project Aims and Purpose

To explore what helps informal carers seek help and what acts as a barrier. To tailor interventions to meet the specific needs of informal carers.

This study is for a thesis research project on the Doctorate of Clinical Psychology programme.

Your (the participant) role in the project

As an informal (non-professional) caregiver for someone with MND, we invite you to take part in this research project as we would like to hear about your experiences, things you find challenging, and how you would like to be supported by psychological services. We would need around **1 hour** of your time to talk to a researcher over **video call** or if you are not comfortable with this, we will complete the interview via telephone call. You may be asked to discuss things about your experience of caring for your affected friend/relative which you find upsetting or distressing. This is because we need to understand the challenges that informal carers face so that, in the future, we can deliver effective support by targeting these areas.

The interview will be audio-recorded, which you will have the opportunity to consent to beforehand. It will then be transcribed by an approved University of Sheffield transcriber and analysed using Thematic Analysis.

The research has been ethically approved by the University of Sheffield.

Your right to withdraw

You have the right to withdraw from the study at any time during the interview. You are not required to provide a reason for withdrawing, although you are welcome to do so. Simply inform the researcher if you wish to stop or withdraw and they will terminate the interview. Any anonymous data you have given until this point may still be used in the final analysis.

Confidentiality

We will **not** include any information which may be used to identify you in our analysis. This includes names, contact details, dates, or any information which is uniquely specific to you or the person you provide care to. Due to the remote format, you are encouraged to find a private setting for your interview; the researcher will ensure they are in a confidential environment, will be using headphones to ensure privacy, and will be contacting you via an approved and secure platform (instructions on how to connect to the interview will be provided). The researcher will contact you on an agreed date and time for your interview, after which your contact information will be deleted. However, you are still welcome to contact the researchers yourself after this if you have any queries or concerns.

There are some limits to the level of confidentiality that can be provided. If, during the interview, you suggest that you or someone else was at risk, the researcher may have to share this information so appropriate support can be obtained (for example emergency services). Only the information related to your safety or that of other people would be shared, everything else would remain private.

How your data will be used and stored

In a research project, data is gathered from each participant and then pooled together for analysis. Our interviews will collect qualitative (i.e., non-numerical, semantic) data to find common themes or topics which are shared by our participants. For example, if you and several other participants described feeling overwhelmed, that would suggest a theme in the data. Therefore, the information you provide will contribute to a greater anonymous dataset and will not be isolated for analysis, i.e., we will not scrutinise or single-out your specific personal thoughts and experiences in the final dataset. However, individual interviews will have to be transcribed (recorded) for us to extract the data later.

All data will be stored digitally on a secure, password-protected University drive. Data will be retained for at least 10 years before being destroyed, as per UK Research Innovation practices.

Debriefing

After your interview, the researcher will recap the details of the study and how your data will be used. You will be free to ask questions and leave the call when you are done. However, if you find that you would like to discuss something else or feel affected by the interview and would like support, the lead researcher, Saba Hussain (Trainee Clinical Psychologist) will be available to contact to provide you with a full debrief (details below).

Consent form and questions

Now that you have read the information sheet, if you are interested in taking part, you will be provided with a consent form to review and sign. This will be posted to you. The researcher will arrange a date and time (that suits you) to contact you, for you to discuss the form and ask any questions. After this, please send your completed consent form to the email address provided below (Research Lead).

If you have a query about the research or wish to raise a complaint, please contact the researcher lead and/or supervisors listed below. You can also contact the Head of the Psychology Department (see address below).

Research team details

RESEARCH LEAD

Saba Hussain (Trainee Clinical Psychologist)
Email: shussain26@sheffield.ac.uk

CO-SUPERVISORS

Dr Charlotte Wright (Senior Clinical Psychologist and Tutor)
Email: charlotte.wright@sheffield.ac.uk
Dr Emily Mayberry (Senior Clinical Psychologist)
Email: emily.mayberry@sheffield.ac.uk
Tel: 0114 222 2230 (ask for Emily Mayberry)

DATA CONTROLLER
The University of Sheffield

HEAD OF PSYCHOLOGY DEPARTMENT
Professor Milne
University of Sheffield
Department of Psychology

Interview Schedule

Introduction

Thank you for taking part in this research study. Before commencing, I wanted to let you know that I will be wearing headphones throughout our interview to keep the things we talk about confidential. I am sitting in a quiet room, free from distraction and interruption. Can I check you are alone in a quiet room, free from distraction or interruption? This interview will be recorded and saved to a secure location; are you still happy for that to go ahead?

As you will know from the participant information sheet, I am interested in exploring the experiences of informal carers of people living with MND, in particular, what helps carers seek help and what acts as a barrier. I will be asking you some questions about being a [*current/former*] carer for someone living with MND. Some information was sent to you about the study. Are there any questions you would like to ask?

[Complete participant consent form together – address any questions] Thank you for completing the consent form.

If you would like to stop the interview at any time, take a break, or discontinue, please let me know. If you have any questions or would prefer me to ask the questions in a different way, please let me know. Feel free to have something with you like a glass of water. The interview will take up to 1 hour.

Anything we discuss and talk about today is confidential and all of your information from the interview will be anonymised to ensure you can't be identified. There are

some limits to this which include if I felt concerned about you or someone you mention in our interview, that you could be a risk to yourself or someone else, in which case we could talk to a healthcare professional such as your GP to support you if you agreed to this. This would always be discussed with you first.

<u>Demographic Information (gathered online already)*</u>

What is your marital or relationship status?

Do you have any formal qualifications i.e. education, work-related?

How many hours a week would you say you provide or provided informal care?

(0 – 50, 50 – 100, 100+)

Have you received any diagnoses from healthcare professionals?

Would you say you are in good health?

<u>Main Interview Schedule</u>

1. When did you begin caring for someone living with MND? Can you talk me through how this felt for you?

   Prompt: How did you initially feel about the idea of becoming an informal carer? What concerns or worries did you have? What emotions were you experiencing at the time? If you were around at the time of diagnosis, what were you feeling at the time?

2. a. Have you noticed any changes in your life, in your mental and/or physical health since you began caring for someone living with MND?

Prompt: what did you find easy and what was a challenge? Was it different from what you were expecting, more or less difficult? What has changed in your lifestyle, hobbies, interests? Changes in social interactions, time for yourself and others? What has changed with your mental wellbeing? Physical wellbeing?

b. How soon into caring for someone did you notice these changes? How did you cope?

Prompt: was there anyone you could talk to for support? What coping strategies were you relying on? Did they help?

3. Have or had you ever thought about seeking (professional) help for the challenges you are facing/faced as an informal carer?

Prompt: (yes) – did you follow through with this? How was this for you? What prevented or prevents you from seeking help? What thoughts, emotions or feelings do/did you experience when thinking about seeking help? What did/do you think will get in the way or happen if you seek professional help? What barriers have you faced or do you face? Do you find psychological support to be accessible?

4. Have you ever engaged with mental health professionals in the past? What were these experiences like?

Prompt: would you describe them as positive/negative experiences? How did you feel during these experiences? What would you have liked to have been different? Did it encourage or discourage you from seeking help for the challenges you face?

5. If you were to seek help and engage with mental health professionals now, how do you think this would affect you?

Prompt: could they help you overcome the challenges you are facing? If no, why do you feel this is? What do you feel will happen if you engage in seeking help and working with a professional? What would you like mental health professionals to help you with? Do you believe they can support you appropriately? If yes, how do you think it could help?

6. Have others around you ever shared their experiences with mental health services or recommended them to you?
   Prompt: (yes) – did this prompt you to seek help or prevent you? Were you surprised by their accounts? Have others around you asked you to seek help? How did you react to this? Why was this your reaction?

7. Is there anything else that you would like me to know or talk about, that you feel would be important information relating to your experience of being an informal carer?

End of Interview

Thank you for taking part in this study today. This study has sought to understand what enables or prevents informal carers of people living with MND from seeking professional help for their own emotional and psychological distress, including any barriers they may face. Once I have completed all of the interviews, I will read through and analyse the information to see whether there are any shared experiences,

similarities or differences in the responses given by the participants. A report will be written up outlining the findings and I hope they will be useful when tailoring interventions to meet the needs of informal carers in an accessible and supportive way.

Discussing topics such as these can be difficult to talk about and share with others. Would you like to talk about anything further? Please contact your GP if you feel you need further support after this interview. Thank you again for your time. The recording will now stop.

A project exploring the experiences of informal carers of people living with MND and their help-seeking behaviours

Hello and thank you for volunteering to take part in my project! My name is Saba, I am a third year Trainee Clinical Psychologist at the University of Sheffield. For my doctorate thesis, I am exploring the experiences of informal carers of people living with MND and their help-seeking behaviours related to accessing help as well as any barriers they may face.

As stated on the study advert, recruitment numbers are limited for this project and not everyone who volunteers will be selected to take part.

To ensure we are recruiting a diverse and representative sample of the UK population, it would be extremely helpful if you could fill out this form which would provide us with demographic and background information about yourself.

The data will be stored securely and only accessible by the study researchers. All data will be anonymised and you will be assigned a participant number if you are selected to take part.

If you have any queries about this form or the study, please email myself at shussain26@sheffield.ac.uk.

Thank you once again!

1. Initials

[short-answer text box]

2. Age and Date of Birth

[short-answer text box]

3. Gender (assigned at birth)

[short-answer text box]

4. Ethnicity

[short-answer text box]

5. Religion

[short-answer text box]

6. Socio-Economic Status

[short-answer text box]

7. Years as a carer (please state whether former or current)

[short-answer text box]

8. Relationship to plwMND

[short-answer text box]

Appendix E – Consent Form

"Help-seeking behaviour in informal carers of people living with Motor Neurone Disease (MND)" Consent Form

Please tick the appropriate boxes	Yes	No
Taking Part in the Project		
I have read and understood the project information sheet or the project has been fully explained to me. (If you will answer No to this question please do not proceed with this consent form until you are fully aware of what your participation in the project will mean.)	☐	☐
I have been given the opportunity to ask questions about the project.	☐	☐
I agree to take part in the project. I understand that taking part in the project will include being interviewed using an online platform (Google Meet) and being audio-recorded for the duration of the interview.	☐	☐
I understand that my taking part is voluntary and that I can withdraw from the study at any time. I do not have to give any reasons for why I no longer want to take part and there will be no adverse consequences if I choose to withdraw.	☐	☐
How my information will be used during and after the project		
I understand my personal details such as name, phone number, address and email address etc. will not be revealed to people outside the project.	☐	☐
I understand and agree that my words may be quoted in publications, reports, web pages, and other research outputs. I understand that I will not be named in these outputs unless I specifically request this.	☐	☐
I understand and agree that other authorised researchers will have access to this data only if they agree to preserve the confidentiality of the information as requested in this form.	☐	☐
I understand and agree that other authorised researchers may use my data in publications, reports, web pages, and other research outputs, only if they agree to preserve the confidentiality of the information as requested in this form.	☐	☐
I give permission for the verbal information that I provide to be deposited so it can be used for future research and learning	☐	☐
So that the information you provide can be used legally by the researchers		
I agree to assign the copyright I hold in any materials generated as part of this project to The University of Sheffield.	☐	☐

Name of participant [printed]   Signature: Date:

Name of Researcher [printed]   Signature: Date:

Project contact details for further information:
Saba Hussain (shussain26@sheffield.ac.uk); Dr Charlotte Wright (lead researcher; charlotte.wright@sheffield.ac.uk); Dr Emily Mayberry (co-researcher, emily.mayberry@sheffield.ac.uk); Dr Gillian Hardy, Head of Department, University of Sheffield, Department of Psychology, Floor F, Cathedral Court, 1 Vicar Lane, Sheffield, S1 2LT.

Debrief Sheet

(Post-Interview)

Thank you for taking part in our study – your input is meaningful and appreciated.

We understand that this interview may have caused some distress for you due to the nature of the subject.

If it would be helpful, we can have a chat about how the interview went, any questions you might have, and where to seek further support if needed.

The interview questions were designed to obtain valuable information about your experiences as a carer of someone living with MND. We acknowledge that there is a shortage of tailored support and services available for carers and we hope research such as this could lead to the development of new services or support.

Do you have any questions about the interview or the study?

- *Questions about data anonymity and storage* – refer to storage of data up to 10 years and all identifying data will be anonymised. Data is only shared when required, and confidentiality agreements are signed.
- *Questions about what will happen next* – refer to data analysis, drawing up key themes, writing up the thesis and disseminating findings (this could potentially include MND Association).
- *Question about anticipated outcome of research – refer to aims of study. Anticipate findings will show carers of people living with MND (plwMND) require their own tailored support and healthcare services should offer wider support to families and carers of plwMND.*
- *Recommendations or feedback?*

[Talk through any anxiety/distress the participant is feeling, validating their experience and providing reassurance]

If you still feel like you could benefit from speaking to someone about taking part in this interview or the study, please do get in touch with support services available to you, such as:

- ***Samaritans:*** 24/7 support service if you are struggling and would like to speak to someone. Telephone: 116 123. Email: jo@samaritans.org
- ***Motor Neurone Disease Association (MND): MND Connect*** offers support and guidance on services and wider support. Telephone: 0800 802 6262. Email: mndconnect@mndassociation.org

If you would like to get in touch with researchers involved in the project at a later date, please email shussain26@sheffied.ac.uk.

Appendix G – Transcriber Confidentiality Agreement

Doctorate in Clinical Psychology, University of Sheffield

Transcribing Confidentiality Form & Guidance Notes

Type of project: Research thesis

Project title: **Help-seeking behaviour in informal carers of people living with Motor Neurone Disease (MND**

Researcher's name: Saba Hussain

The recording you are transcribing has been collected as part of a research project. Recordings may contain information of a very personal nature, which should be kept confidential and not disclosed to others. Maintaining this confidentiality is of utmost importance to the University.

We would like you to agree:

1. Not to disclose any information you may hear on the recording to others,

2. If transcribing digital recordings – only to accept files located on the secure X drive/UniDrive/Google Drive provided by the researcher via VPN.

3. When transcribing a recording ensure it cannot be heard by other people,

4. To adhere to the Guidelines for Transcribers (appended to this document) in relation to the use of computers and encrypted digital recorders.

5. To show your transcription only to the relevant individual who is involved in the research project.

6. If you find that anyone speaking on a recording is known to you, we would like you to stop transcription work on that recording immediately and inform the person who has commissioned the work.

Declaration

I have read the above information, as well as the Guidelines for Transcribers, and I understand that:

1. I will discuss the content of the recording only with the individual involved in the research project

2. If transcribing digital recordings – I will only accept files located on the secure X drive/UniDrive/Google Drive provided by the researcher via VPN.

3. I will not use external storage programmes or website, such as Dropbox, for transferring recordings as it does not meet any of the University's data security guidelines

4. When transcribing a recording I will ensure it cannot be heard by others

5. I will treat the transcription of the recording as confidential information

6. I will adhere to the requirements detailed in the Guidelines for transcribers in relation to transcribing recordings onto a computer and transcribing digital audio files

7. If the person being interviewed on the recordings is known to me I will undertake no further transcription work on the recording

I agree to act according to the above constraints

Your name SARAH Fox

Signature ███████████

Date ·13·2·24

Occasionally, the conversations on recordings can be distressing to hear. If you should find it upsetting, please stop the transcription and raise this with the researcher as soon as possible.

Appendix H – Data Management Plan

Help-seeking behaviour in informal carers of people living with Motor Neurone Disease (MND)

Defining your data

- What digital data (and physical data if applicable) will you collect or create during the project?
- How will the data be collected or created, and over what time period?
- What formats will your digital data be in? (E.g. .doc, .txt, .jpeg)
- Approximately how much digital data (in GB, MB, etc) will be generated during the project?
- Are you using pre-existing datasets? Give details if possible, including conditions of use.

The project will involve collecting digital data which is the audio files of recorded semi-structured interviews via Google Meet. The online interviews will be recorded on Google Meet using both video and audio, however only the audio will be sent to transcribers securely via a secure Google Meet drive. To match participants to their unique code/participant ID number, there will be one digital file that will be kept by the primary researcher in a locked document which will be deleted upon the commencement of data analysis.

There will be transcripts of data from the interviews but these will not contain any identifying information. These will be used for analysis. Thematic Analysis (TA) will be used to analyse the transcripts, and this would also count as data created during this project.

The data will be collected from January/February 2024 until the number of interviews required has occurred (10-12). It is hoped that the interviews will be completed by March 2024.

The audio files will be recorded or modified to ensure they are MP3 format. The locked document with the codes to match participants will be .doc. The transcriptions will be .doc until completed when they will be .pdf so that there is no accidental editing occurring or room for error once transcriptions are complete.

For up to 12 interviews for up to 30 minutes each using Google Meet, about 4 GB of audio data will be created. It is estimated this project could take up to 2MB of documental data (1000 pages).

Pre-existing datasets will not be used during this project.

Looking after data during your research

- Where will you store digital data during the project to ensure it is secure and backed up regularly? (E.g.University research data storage, or University Google drive)
- How will you name and organise your data files? (An example filename can help to illustrate this)
- If you collect or create physical data, where will you store these securely?
- How will you make data easier to understand and use? (E.g. include file structure and methodology in a README file)
- Will you use extra security precautions for any of your digital or physical data? (E.g. for sensitive and/or personal data)

Digital audio files of recorded interviews will be recorded on Google Meet and saved to a secure Google Drive that can only be accessed by the primary researcher and those that are allowed access i.e. other primary investigators.

The transcriber will have access to this drive for a limited time to transcribe the audio files of the interviews. Once these have been completed, access to the drive will be revoked. The recordings will be backed up on the primary researcher's University drive attached to their University e-mail. No identifying information will be in the interview recordings. Each participant will receive a code. The code key will be deleted once analysis begins - this is solely so participants can withdraw up until analysis. E.g. Participant 1, 2, 3.

It is not anticipated that any physical data will be collected or created as interviews will be completed online and participants will be sent documentation via email however if any physical data is collected, this will be stored in a locked file where the researcher has a key, in accordance with the University guidelines. This will include if the TA method requires creating physical data.

Data will be made easier to understand and use through transcription. A trained transcriber will be used to transcribe the data to PDF which will then be analysed by the primary researcher. Once finished, all documents will be secured in README files so that they are not altered. A locked document file will be used as a safety precaution with the first name of each participant and their last initial. Their consent forms will be kept in a secure file and destroyed at the appropriate time in the research.

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Storing data after your research

- Which parts of your data will be stored on a long-term basis after the end of the project?
- Where will the data be stored after the project? (E.g. University of Sheffield repository ORDA, or a subject-specific repository)
- How long will the data be stored for? (E.g. standard TUoS retention period of minimum 10 years after the project)
- Who will place the data in a repository or other long-term storage? (E.g. you, or your supervisor)
- If you plan to use long-term data storage other than a repository, who will be responsible for the data?

Following the completion of the project in September 2024 and after data has been analysed and written up for the main body of research, it will be securely stored for at least 10 years by The University of Sheffield before being destroyed, as per UK Research Innovation practices.

Sharing data after your research

- How will you make data available outside of the research group after the project? (E.g. openly available through a repository, or on request through your department)
- Will you make all of your data available, or are there reasons you can't do this? (E.g. personal data, commercial or legal restrictions, very large datasets)
- If there are reasons you can't share all of your data, how might you make as much of it available as possible? (E.g. anonymisation, participant consent, sharing analysed data only)
- How will you make your data as widely accessible as possible? (E.g. include a data availability statement in publications, ensure published data has a DOI)
- What licence will you apply to your data to say how it can be reused and shared? (E.g. one of the Creative Commons licences)

Data will not be made available to other parties. Transcription analysis will be available in the written report and will only be used for this project. Only analysed data will be shared.

Putting your plan into practice

- Who is responsible for making sure your data management plan is followed? (E.g. you with the support of your supervisor)
- How often will your data management plan be reviewed and updated? (E.g. yearly and if the project changes)
- Are there any actions you need to take in order to put your data management plan into practice? (E.g. requesting University research data storage)

The researcher and the primary research supervisor are responsible for ensuring the data management plan is followed. This will be reviewed and updated prior to the collection of data. The data management plan will be reviewed at research supervision (at least once every 3 months) and each time after new data is collected. If the project changes, the data management plan will be altered to reflect this. There are no further requirements at this time to put the data management plan into practice.

Appendix I – Ethical Approval Form

Saba Hussain
Registration number: 210154869
Psychology
Programme: Doctorate in Clinical Psychology

Dear Saba

PROJECT TITLE: Help-Seeking Behaviour in Informal Carers of People Living with Motor Neurone Disease (MND)
APPLICATION: Reference Number 051051

On behalf of the University ethics reviewers who reviewed your project, I am pleased to inform you that on 25/09/2023 the above-named project was **approved** on ethics grounds, on the basis that you will adhere to the following documentation that you submitted for ethics review:

- University research ethics application form 051051 (form submission date: 19/09/2023); (expected project end date: 15/08/2024).
- Participant information sheet 1126685 version 2 (19/09/2023).
- Participant consent form 1126686 version 2 (19/09/2023).

If during the course of the project you need to deviate significantly from the above-approved documentation please inform me since written approval will be required.

Your responsibilities in delivering this research project are set out at the end of this letter.

Yours sincerely

Department Of Psychology Research Ethics Committee
Ethics Admin
Psychology

Please note the following responsibilities of the researcher in delivering the research project:

- The project must abide by the University's Research Ethics Policy: https://www.sheffield.ac.uk/research-services/ethics-integrity/policy
- The project must abide by the University's Good Research & Innovation Practices Policy: https://www.sheffield.ac.uk/polopoly_fs/1.671066!/file/GRIPPolicy.pdf
- The researcher must inform their supervisor (in the case of a student) or Ethics Admin (in the case of a member of staff) of any significant changes to the project or the approved documentation.
- The researcher must comply with the requirements of the law and relevant guidelines relating to security and confidentiality of personal data.
- The researcher is responsible for effectively managing the data collected both during and after the end of the project in line with best practice, and any relevant legislative, regulatory or contractual requirements.

Appendix J – Reflexive Statement

Reflexive Statement

The lead researcher is a female, British Trainee Clinical Psychologist from a South Asian background who has no prior personal or professional experience of MND or working with informal carers. However, the lead researcher does have clinical experience of working with clients diagnosed with other neurodegenerative diseases, in particular Alzheimer's disease and other dementias, during her time on an Older Adult placement. It is worth noting that the experiences of plwMND will differ from those diagnosed with other neurodegenerative diseases, including the experiences of carers.

The lead researcher has personal experience of Alzheimer's disease as a relative was diagnosed with the disease and the researcher's mother delivered informal care which the researcher was, at times, involved in. Again, it is worth acknowledging that the caregiving experiences of informal carers of plwMND will vary from those caring for people diagnosed with other neurodegenerative diseases.

Appendix K – Reflective Log Excerpts

Reflective Log Excerpts

Excerpt A

I am starting to notice the level of empathy that I am feeling for the carers I have interviewed and the challenges they are experiencing now that I have completed a few interviews. I noticed it felt difficult for me to remain in the 'researcher' role with one particular participant who became upset after recalling the struggles she encountered whilst caring for her partner – at times, I felt like I wanted to slip into my 'therapist' role to provide support, 'sit' with certain feelings and offer solutions to problems she was facing. It made me think about how, within clinical psychology, the 'therapist' and 'researcher' role can often overlap or 'spill' into each other as it feels instinctive to be there for people. I noticed feeling a bit helpless after the interview was over and reflected on this with my research supervisor, who validated my feelings and told me that it was normal to feel this way as the research topic is very emotive. It almost feels unnatural for me to *not* offer support or 'therapy' but I will ensure I adopt a compassionate approach given the emotive subject. I received positive feedback on my questioning style and approach from supervisors which reassured me that I am still able to offer compassion to the carers whilst sticking to the restrictions of the interviewer role.

Excerpt B

Something that has really struck me is the lack of ethnic minority representation within my participant sample. I almost feel a bit disheartened about this as all of my interviews have taken place with White British carers, which is understandable given that White British represents the majority of the UK population. However, as someone from an ethnic minority herself, I was keen to hear from minoritised individuals and different cultural and religious perspectives of caregiving for plwMND. I notice these feelings are probably heightened because of my own background. I also wonder about how I was being perceived by the participants as someone who was visibly different to them – did my 'obvious' difference mean some participants held back or felt they could not share certain things in case I could not relate? Would there have been 'space' to talk about this with participants, or would this impact the research project in some way?

Appendix L – Analysis: Coding of Transcripts Example

Interview

Saba Hussain

Project title: Help-seeking behaviour in informal carers of people living with Motor Neurone Disease (MND)

[handwritten top right: Professionals 'signposting help', A]

1 P: When (NAME)-daughter had passed, you know, we had, we had the district nurses
2 contact us and then when we were talking to the hospice where NAME-(daughter)
3 went a couple of times just to see whether they could help with treatment, erm you
4 know they were all sort of like, "well you know, if you need any help pick up the
5 phone and there's people here etc", but that was the difficult thing (SIGH), being able *[handwritten: 'not needing help'?]*
6 to do that, and my wife was kind of like "no I don't need that", you know but it's very
7 erm, it's very hard because making that first step and maybe trying to realise, you *[handwritten: worrying about loved ones.]*
8 know, I worry about my wife and whether, how she's coping mentally, you know she's
9 not one that shows her emotions a lot, I think she bottles a lot up but its very difficult
10 to, to read her, erm you know, erm so yeah, you know, that's probably been one area *[handwritten: lack of involvement from services.]*
11 where post losing NAME-(daughter), erm there's not bee a lot of follow-up and I don't
12 know, I know everybody's busy and the health service is under pressure but its kind
13 of like you want somebody to actually physically say "come in and see us", you know, *[handwritten: Difficult to make first move?]*
14 "we want to see you and lets have a chat, lets", you know, do something like that but *[handwritten: -Want services to offer.]*
15 its, its, its hard because, you know, I've mentioned a couple of times, you know, "why
16 don't we go and talk to somebody" and my wife says "I don't need to, I don't need to",
17 so its just difficult.

18 T: Yeah.

19 P: The disease as they say changes things, not just for the person living with it but *[handwritten: impact of MND around.]*
20 everybody that it almost touches and, you know, there's a definite, erm, definite
21 change, you know, its changed, its changed things, you know it's one of them.

22 T: Thank you NAME-(Participant) and I think you're right about saying that it's a really
23 difficult time that you went through, and even after NAME-(daughter), kind of those
24 effects are still there, and you know we will kind of go into that a bit deeper in the

Saba Hussain

Project title: Help-seeking behaviour in informal carers of people living with Motor Neurone Disease (MND)

49   before we went over because they had their own lives to lead, you know, but it was

50   like, you know, "is it alright if we pop over", or "shall we pop over this evening", you

51   know and if they said "no, we'll be alright", that's fine, we left them alone, you know.

52   If they just wanted to chat we'd go over and, you know, so.

53 T: Yeah, and can you talk me through how that felt for you, so to be now an informal

54   carer for your daughter and any emotions you were experiencing at the time?

Putting on a strong face – conflicting feelings

55 P: Yeah, you know, its erm very hard because you want the best, you know, you want

56   the best for her and in the back of your mind you know the condition's terminal, erm

57   you know, in our daughters case, you know she had, she had an excellent career

life she could have had.

58   ahead of her mapped out via work before her diagnosis, so she was, you know

59   everybody that met her, you know if you met her out in Sheffield or whatever, you

60   know, you would just, you know she would just become a friend, she was one of

difficulty accepting diagnosis

61   those people. Erm and it was, it was, you know, it was hard when, hard when we'd

62   come back home sometimes, you know, erm to well you can't, you can never accept

death of child before parent.

63   it, you know you don't expect, you don't expect your children to go before their

64   parents, you know, particularly in her case, you know, she was just such a wonderful

65   girl and had so much ahead of her.

66 T: Yeah.

67 P: Yeah, yeah.

seeing advantages & positives.

68 T: So really, really difficult experience for you NAME-(Participant).

69 P: Yes very, you know we, the plus, the pluses were, you know, we know in our hearts

70   we did everything we could for her and we were there for her no matter what time of

impact on self was not priority.

71   day or night, how knackered you felt or, you know, sometimes there were arguments,

72   you know, I wont deny that because with the illness NAME-(daughter) would get

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Appendix M – Analysis: Thematic Map

'one size fits all' rather than individualised offers of support
CODE

Lack of knowledge/ understanding from HCPs (with less MND experience).
THEME 4

Delivery of diagnosis left lasting impression and impact.
CODE

Sub-theme
Physicality + practicality of caring.

Time management.
↓ lack of time for self/own needs.
CODE

Pre-existing or developed physical health needs.
CODE

Physical, practical, mental, emotional impacts on carers.
THEME 1

Sub-theme
Emotional + mental impact

Loss of identity, coping mechanisms, self-care + avoidance d Reduced wellbeing.
CODE

Uncertainty of disease trajectory.
CODE

Emotional load of caring for PlwMND and other loved ones.
CODE

Sub-Theme
Perception and experiences of services/offers.

Previous negative and/or positive experience of services influenced future help-seeking.
CODE

Wanting services to make the 'first move' to offer support.
CODE

Lack of clarity around what support could actually do/ help with/what's out there.
CODE

MND Association – mixed comments about helpful-ness and accessibility.
CODE

Informal carers of PlwMND feel their own wellbeing is not a priority.

"No-one asked me how I'm doing" – didn't get signposted to help/ support = de-prioritisation of own needs because not prioritised by professionals.

Worries about PlwMND → physical + mental health
CODE

Sub-theme
Suitability of available support and barriers to accessing.

Where, when and how carers do seek support.
THEME 3

Support networks formed with other carers/friends/etc.
Sub-theme

Dominance/ Prioritisation of needs of PlwMND.
THEME 2

Accessibility & availability & acceptability + individual variation in needs + preferences.
CODE

Individual caring situation impacts ability to extend or engage.
CODE

Valuable support from carers in similar positions
CODE

Importance of peer support forums (non-professionals/not directly involved in care).
CODE

Difficult to accept the MND diagnosis and prognosis.
CODE

"Just get on with it" → priority is to care for PlwMND
CODE

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Appendix N – Analysis: Theme Development

Physical, Practical, mental and emotional impact on carers (T1)

Dominance and Prioritisation of the needs of plwMND. (T2)

Sub-theme: Physicality & practicality of caring.

Sub-theme: Emotional + mental impact

P4: 10, he's so weak that I had the deadlift this 10/11 stone man off the floor and as I as I went into him, I went into a deep squat and lifted him up, and every joint in my body clicked, like my knees, my ankles, my elbows, my whole spine, so I put absolutely everything (LAUGH) I had into picking him up off the

P10: Err yeah absolutely, in everything, erm he had very swollen feet so with putting his socks and shoes on every day I got very sore hands, like my wrists and my thumbs and my fingers, and even though he passed away in July they're still sore and they've not got back to normal, his physio gave me wrist

P3: all now, so like I feel like I don't have any time, like he's gone out with his mum to do something and it's like "oh he's gone out for like an hour, so that's like an hour to myself", but I kind of don't get much time like that's what I struggle with, he enjoys just having a bit of time just to, especially with his

P5: wanted to not be too panicky because my son, it will be upsetting for him.

P9: stress, I also find myself like, I've more often not done things for myself, and it's a way of either saving time or energy, erm like but I like pretty much drive

P11: Yeah, no absolutely, I mean I did, I went erm from full time to part time work and then you know obviously to not working and I erm I used to go to the gym a lot and I used to do, I trained to do some triathlons and things like that, you probably cant tell if you look at me now (LAUGH) but I was, you know, I really enjoyed all of that and he did too you know previously so erm I did love that but then I had to say when he was diagnosed and when he was struggling to walk and things I felt tremendously guilty about going for a run or getting on

P6: chilled, so yes very hard emotionally because it's obviously seeing who you love, you know deteriorating in front of your eyes, watching their struggle, yeah horrendous, absolutely horrendous. The nurse that came from St

P7: Erm, scary, I would say fear more that anything with not really knowing what was coming, knowing it was going to be horrendous but not, not knowing, not understanding, yeah lack of knowledge really, and the you then got, knowing

69 P: Yes very, you know we, the plus, the pluses were, you know, we know in our hearts
70 P1: we did everything we could for her and we were there for her no matter what time of
71: day or night, how knackered you felt or, you know, sometimes there were arguments,
72: you know, I wont deny that because with the illness NAME-(daughter) would get

P2: back and sometimes we have weeks where its like one appointment after another, so I have had to cancel things to fit in and sometimes that's what I mean about time, you can't always have the time that you want because of all those things.

P8: I just know what I had to do and I just got on with it really, I wasn't thinking about me, I suppose I was thinking about him all the time, I can't really remember about my mental state really. Obviously I was upset but I just hated

P4: terms of how I was feeling, because I've worked in motor neurone disease for a very long time, I thought, well this can't possibly be motor neurone disease because what would the odds be, it seemed impossible and you know,

P1: come back home sometimes, you know, erm to well you can't, you can never accept it, you know you don't expect, you don't expect your children to go before their parents, you know, particularly in her case, you know, she was just such a wonderful girl and had so much ahead of her.

P7: It was. It was just a freight train, completely couldn't even put, I seem to remember saying something about life expectancy and I remember him saying two years. Yeah the most awful thing, and that was October 20 wasn't

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Sub-Theme
Perception and experiences of services / offers.

Sub-theme
Suitability of available support and barriers to accessing.

Support networks formed with other carers / friends / etc.
Sub-theme

terms of services for carer's again it's the person that you're generally speaking to is there for the person that you're caring for, you know, and so they very rarely ask you how you are or how you are doing. I've also got a ... **P11**

I do suffer mentally because I think he should be here and I, I sit and think about the way he died more than, that's the thing that hits me more than anything really, I don't know how he died and I can't see how a counsellor would be able to help me with that really, because I won't be able to tell me ... **P4**

know, I know everybody's busy and the health service is under pressure but its kind of like you want somebody to actually physically say "come in and see us", you know "we want to see you and lets have a chat, lets", you know, do something like that but ... **P1**

Yeah, if the opportunity had being there, I definitely would have done, but I did try and talk to the Marie Curie on my own, yeah when, NAME - (son), in his last couple of weeks, but she was no help at all, and I think that's what put me off in a way actually because I said to her "can I speak to you in private ... **P8**

I've had counselling and it didn't help one single bit, I didn't, I didn't think the counsellor understood at all and she was provided by MND Scotland so specifically for counselling for MND and I've tried it and (SIGH), I tried it to the point where I was just thinking 'oh I cant be doing with this session tonight, it's really getting in the way' (LAUGH), it was like an hour out of my life that I wasn't getting back, erm and I just felt it was absolutely pointless so ... **P10**

I think that's also a barrier that I, me and my partner sometimes have, its like there's probably a lot of things that can help us, but if we don't know that there would not really going to ask for them, so from that point of view it was hard to really understand which direction to go or who to ask or what we should be ... **P9**

I still count as a caregiver? Like you almost sort of have imposter syndrome on the extent to which you deserve help or deserve support, you know or as I say, you know wouldn't it be strange if I was sitting around on a psychiatrist couch talking about my father-in-law when my mother-in-law doesn't have that support, you know, if you have a couple of hours, why don't you go and sit ... **P4**

P: Because the waiting lists around here are so long and also like I say its not face to face, its online which is no good because I'm already isolated, I don't want that. **P12**

relationships, to seek that you have to go through a long process of giving all the information, filling in the questionnaires, like talking about the situation, and again those can feel like barriers in that space. **P9**

P: Yeah and I think were very lucky that were surrounded by very lovely neighbours which were lucky where we live and friends and family, so – **P2**

Yes, yeah and the carer's in the campaign, there is a couple of carers in the campaign that just like you know, they send me little things and I go (LAUGH) "oh you will never guess what's happened this week" (LAUGH), and they go "oh", and I you don't really have to, I don't have to explain because they also know what its, you know there is a shorthand between carer's so if I say "oh, you know, I, I spent four hours on the phone trying to get this sorted out", you know they'd know exactly what that meant and how frustrating that was and I wouldn't have to say, you know who I was trying to contact or how frustrating that it was, they would just know. **P11**

but I don't think (SIGH) its going to be a solution. One of the best things I found actually was a group on Facebook, there is an MND Support Group and its got over 10,000 members and even they were giving medical advice out there and it wasn't a 'change your tablet or change something', it was practical tips about mattresses, pillows, posture, things like that that professionals couldn't tell you, that was really useful. **P12**

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150 — them are just very much like that, 'this is what you've got, nothing much we can do, here's some booklet, contact the MNDA", you know "why", you know. Awful really. **P1**

and they went "leave him in bed until 10", and I went "he works from home, he is still working, I cant leave him in bed til 10, he starts work at 9", (LAUGH). They went, "we don't care, this is what we're giving you". So you know when somebody's doing that you think, (SIGH) you know, what was the point in this ... **P10**

It just sounds like its one size fits all and one size doesn't fit all with MND, it's a very, very different disease and I don't know another disease like it where ... **P11**

(husband) was very much "you have MND, you have got, maybe because you've had the symptoms for two years on and off pretty much, I think two years to that point, and the prognosis is serious so we think maybe you might have eighteen months". So that's how it was put to us; so absolutely shock horror. So we lived with that for a few days until our MND nurse came and ... **P3**

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