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University of Southampton

Faculty of Environmental and Life Sciences

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Supporting Carers of People with Motor Neuron Disease: A Systematic Review of Psychological Interventions for Terminally Ill Patients, Understanding How Spousal Carers of People with Motor Neuron Disease Experience Palliative Care in the UK

by

Georgia Steed

Thesis for the degree of Doctorate of Clinical Psychology

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Abstract

Faculty of Environmental and Life Sciences
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Doctorate in Clinical Psychology

Supporting Carers of People with Motor Neuron Disease

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Chapter 1 of this thesis is a systematic review of the literature in psychological interventions for carers of people with terminal illnesses. This paper aimed to understand the current evidence base for psychological interventions for depression and caregiver burden and compare effectiveness of treatments for this client group. The paper explored and compared the findings of 18 studies on outcomes for depression and caregiver burden, using Cognitive Behavioural Therapy, Existential Behavioural Therapy, Mindfulness-Based Cognitive Therapy, Psychoeducation and Counselling. Overall CBT approaches were most effective for depression and caregiver burden, however there is an overall lack of transparency in the current research. Quality and methodological considerations are explored and evaluated.

Chapter 2 of this thesis is a qualitative paper which aimed to understand how spousal carers of people with Motor Neuron Disease experience palliative care in the UK. This paper used interpretative phenomenological analysis and identified 3 group experiential themes: ‘Battle,’ ‘Coping’ and ‘Feelings brought on by receiving palliative care.’ These were divided into subthemes and explored in depth. Clinical considerations, recommendations and considerations for future research are explored.

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Research Thesis: Declaration of Authorship

Print name: Georgia Steed

Title of thesis: Carer Support in Motor Neuron Disease

I declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

I confirm that:

1. This work was done wholly or mainly while in candidature for a research degree at this University;
2. Where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
3. Where I have consulted the published work of others, this is always clearly attributed;
4. Where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
5. I have acknowledged all main sources of help;
6. Where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;
7. None of this work has been published before submission

Signature: .... Date: 19/05/23

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This research is dedicated to all the families I spoke to and all of those I did not speak to, who are battling the impacts of MND. It is also dedicated to my own family, to my stepdad Rich who we lost to MND in 2018 and to my amazing mum Paula. Without your dedication, love and insights throughout this process, this research would not have been possible.

Finally, to my loving husband, thank you for your support and patience throughout this journey.

Chapter 1 A Systematic Review of Psychological Interventions for Carers of Terminally Ill Patients

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1.1 Abstract

Carers of people with terminal illnesses have many needs which impact their psychological wellbeing and functioning. They can be vulnerable to developing difficulties with depression and caregiver burden, however few reviews have explored which psychological interventions are beneficial for carers of people with terminal illnesses. This review explored and compared the findings of 18 studies on outcomes for depression and caregiver burden, using Cognitive Behavioural Therapy, Existential Behavioural Therapy, Mindfulness-Based Cognitive Therapy, Psychoeducation and Counselling. Overall CBT approaches were most effective for depression and caregiver burden, however there is an overall lack of transparency in the current research. Quality and methodological considerations are explored and evaluated.

Keywords: Carers, Terminal Illness, Psychological Interventions, Depression, Caregiver Burden.

1.2 Introduction

1.2.1 Terminal Illness and the Role of Services

Terminal illness is an incurable illness which will likely lead to death. Conditions considered terminal include advanced cancer, dementia, motor neuron disease (MND), lung disease, Parkinson’s disease, Multiple Sclerosis, and advanced heart disease (Lyn et al, 1997).

Varying perspectives with regards to what constitutes a good death and therefore, good end-of-life care, poses a challenge for services supporting the terminally ill and their families (Meier et al, 2016; Borgstorm, 2020). NICE guidelines for supporting people at end-of-life recommend multi-practitioner care to meet complex needs with good communication when coordinating care between health and social care organisations (NICE guidelines, 2019).

End-of-life care can include involvement with palliative care services, hospice care and home support. Palliative care emphasises holistic care which provides support in social, psychological, existential, and spiritual areas of an individual’s life (Radbruch & Payne, 2010). The concept of end-of-life care is broad and complex and therefore, so is research in this area.

NICE guidelines recommend that services consider emotional and practical support for carers (NICE guidelines, 2019).

Relationships between patient and carer outcomes are bidirectional in nature, which highlights the importance of supporting carers within services to facilitate good outcomes (Bidwell et al, 2017; Götze et al, 2014). In line with these findings, supporting carers at the end-of-life stage is an essential part of the UK end-of-life care strategy (Department of Health, 2008). Similarly, adequate support for carers has also been associated with decreased likelihood of placement breakdown within at home end-of-life care (Sarmento et al, 2017).

The impact of experience with services can continue after the death of their loved one. Satisfaction in palliative care received is associated with reduced risk of developing a complex grief response (Mason, et al 2020). In addition, positive relationships with the care team have been associated with better experience of end-of-life care and the perception of a greater quality of death (An et al, 2020).

1.2.2 Challenges for Carers

Informal caregivers are defined as individuals who provide voluntary unpaid care to a relative or friend (Schultz & Tompkinson, 2010). Informal carers face social, emotional, health and financial consequences associated with caregiving (Eifert et al, 2015). They are vulnerable to developing depression, anxiety and stress associated with the challenges of caring, including the significant time spent providing care (Diggory & Reeves, 2022). Carers may experience ambiguous loss (Balintona, 2018), challenges adapting to the ever-changing needs of the care recipient (Anoun et al, 2021), and guilt associated with a sense of responsibility (Midtbust et al, 2021). Carers may also experience pre-loss grief, which increases risk of prolonged grief longitudinally (Luk, 2021).

There are individual differences in the degree to which carers experience distress. This can be predicted by attitudes to and communication about death, which is mediated by acceptance (Davis, et al 2015). The individual difference in carers' needs and experiences may be associated with the variability in individual carer experience (Wang et al, 2019). For example, emotion-focused coping and positive reappraisal were more common in caregivers of cognitively intact patients (Livermore, 2014). These differences can impact how services support carers.

Caregiver burden has been a key focus in the literature due to high prevalence rates and strong association with caregiver burnout (Gérain, & Zech, 2019). In addition, caregiver burden has been associated with increased risk of depression (del-Pino-Casado et al, 2019). Depression has been found to be the most common mental health difficulty for carers of people with multiple difficulties including advanced cancer (Oechsle et al, 2019), dementia (Bussè, et al, 2022) and those in intensive care (Johnson et al, 2018). In addition, depression has been associated with poorer physical and mental quality-of-life for carers (Petruzzo et al, 2019).

Caregiver Burden

There are many factors associated with increased carer burden including increased time spent providing unpaid care (Guerriere et al, 2016), hours of care provided (Galvin et al, 2016), poor relationship quality (Luth & Pristavec, 2020), symptom severity or aggressive behaviours towards the carer (Hiyoshi-Taniguchi, et al 2018; de Wit et al, 2018), negative perception of quality of care provided (Lethin et al, 2020), caregivers mental and physical health (van den Kieboom et al, 2020), caregiver perspective on patient quality of life (Macchi et al, 2020) and relationship to care recipient (Pennin and Wu, 2016). Research has indicated possible gender differences in terms of experienced caregiver burden, however a systematic review noted that research has failed to account for possible covariates which may explain this difference (Xiong et al, 2020). Additionally, self-efficacy has been found to mediate caregiver burden and improve wellbeing (Park et al, 2019).

Depression

Depression is another widely researched area. There are many factors associated with increased prevalence and severity of depressive symptoms including care recipient's symptom distress and functional impairment (Wen et al, 2019), duration of caregiving and caregiver employment status (Zyada, et al 2013), care recipients using emotional support coping strategies (Nipp et al, 2016), being a partner or spouse of a terminally ill care recipient (Nielsen et al, 2019), higher levels or pre-loss grief (Nielson et al, 2017), carers living with the care recipient (Govina et al, 2019), caregiving hours (Grande et al, 2021) and caregiver being unemployed, having poor physical health, and poor sleep quality (Geng et al, 2018).

Therefore, carers experience a wide range of needs associated with their caring roles. This can result in significant psychological distress which may be further impacted by difficulties accessing services (Bentley & O'Connor, 2016). Carers may need support from services to manage their wellbeing, and the wellbeing and quality of care for the care recipient.

1.2.3 Interventions for Carers

Due to the impact of caring and bidirectional impact on care recipients, it is important to deliver high quality psychological interventions to support carers. However, this is conceptually challenging given the breath of challenges faced by caregivers, variation in care recipients' presentation, and the associated demands of caring. As such, there have been numerous recommendations for approaches and treatment targets. Luce (2020) found that carers felt ill prepared for the loss of their loved one and the uncertainty around this. This paper recommended using an existential framework to improve psychological interventions for carers of terminally ill people. In addition, Mirsoleymani et al (2021) recommended interventions based around family coherence which targeted family resilience and acceptance through mindfulness, which may support adjustment to multiple sclerosis (Pakenham and Samios, 2013).

When considering unmet needs of carers, 70% of these were psychoeducational in nature (Ducharme et al, 2014). Similarly, Henriksson et al (2013) recommended that psychoeducational interventions aim to increase caregivers' preparedness to care through emotional, practical and communication support. Park et al (2019) also recommended psychoeducation to support caregivers to manage emotional distress and improve their capacity to manage the care recipient's illness. In addition, Hsieh et al (2016) suggested that psychoeducational programs provide strategies for coping with challenging behaviours to reduce caregiver burden. De Wit et al (2020) however, recommended monitoring in addition to psychoeducation to target caregivers' perceived control.

Although there are numerous systematic reviews regarding palliative care interventions, there are limited reviews looking specifically at psychological interventions for carers of terminally ill people. This may reflect reported gaps in interventional research to identify what is effective in supporting carers (Poppe et al., 2020).

Schildmann and Higginson (2011) conducted a systematic review evaluating the strengths and limitations of study designs in psychoeducational interventions for informal carers of patients receiving palliative or cancer care. This paper found that randomised control trials (RCTs) were the gold standard however there were limitations in terms of biased recruitment and attrition. This paper recommended well designed randomised control trials (RCT's) to mitigate methodological issues. However, research in cancer and palliative care were grouped, when participants educational needs, and effectiveness of interventions may vary. In addition, they failed to compare effectiveness of interventions.

Jaffray et al (2016) conducted a systematic review on mindfulness-based interventions for informal palliative caregivers. This research found evidence of feasibility and potential benefit of such interventions, however attributed small effects to incorrect outcome measuring and recommended qualitative research to explore outcomes in more depth. This review did not explore effect sizes, included uncontrolled trials, did not include grey literature or compare mindfulness-based interventions with other interventions. This research failed to consider factors such as how 'informal carer' is defined for example partner or friend, or how much time the caregivers spent caring.

There have been two systematic reviews which have compared a range of psychological interventions specifically for family carers of people with terminal illnesses. Hudson et al (2010) examined psychosocial interventions for family carers of palliative care patients. This review identified interventions focussed on education, support, coping, symptom management, sleep and family meetings. It found that psychoeducation could be beneficial, highlighted the importance of tailored support to meet needs in areas such as sleep and noted an improvement in study quality. It also highlighted however, that health professionals were operating without a suitable evidence-base. This review focused on family carers, therefore mitigated some of the difficulties which exist in the literature of grouping carers under the umbrella of “informal” which poses an issue for homogeneity and impacts which interventions may be beneficial given the increased demand on family carers. However, there are methodological issues regarding the search strategy. The paper grouped carers/family with grief/death/bereavement in their search strategy which combines different constructs. The search terms were not suitably concise to ensure a breadth of psychological interventions could have been detected. The search terms for caregivers missed key terms like “spouse” which detected many papers on scoping searches. In addition, the search terms were broad therefore yielded many unrelated studies, for example focussed on medical interventions. This may have resulted in the inclusion of research which was not based on evidence-based psychological principles. Carers in this review had to be receiving palliative care, which can be a broad concept and may miss carers of people with terminal conditions who were not yet with palliative care services. This is important given the reported delays in families being referred to palliative care services following a diagnosis of MND, despite guidelines for referral at diagnosis (Flemming et al, 2020). Therefore, this research may have missed suitable papers for review. This was also conducted in 2010 and therefore may warrant an updated review.

Chi et al (2016) explored behavioural and educational interventions for family carers in end-of-life care. This found that CBT resulted in better outcomes. However, it noted that more rigorous randomized controlled trials with larger and more diverse samples are needed. It recommended that future research should aim to find the most efficient intervention method. Although this review considered some important intervention types, it did not consider or compare existential or third wave CBT interventions, which can be commonly used within palliative care settings (von Blackenburg & Leppin, 2018).

To date, no reviews have compared the effectiveness of a range of evidence-based interventions including third-wave CBT interventions for family carers of people with terminal illnesses.

1.2.4 Aim of Current Review

The prospective review aimed to address the methodological issues with the previously discussed papers. Firstly, search terms focused on carers and relational roles are not grouped with other concepts (e.g., adding “care giver,” “partner”, “spouse”), as well as terms for specific evidence-based psychological interventions (e.g. “Acceptance and Commitment Therapy,” “ACT”) which included third-wave and existential approaches. In addition, this paper had no exclusion criteria regarding involvement in palliative care services.

This review aimed to understand the current evidence-base for psychological interventions for carers of terminally ill people, understand which psychological interventions have been found to be effective, and compare psychological interventions to determine which are the most effective for this group of carers. Scoping reviews identified that caregiver burden and depression were the most common targets for intervention. Due to this, along with the prevalence and impact of these difficulties, this review focused on intervention which targeted depression and/or caregiver burden.

The specific review questions were:

1. What is the evidence-base for psychological interventions for reducing depression and caregiver burden for family carers of people with a terminal condition compared to controls?
2. What is the effectiveness of psychological interventions in reducing depression and caregiver burden for family carers compared to controls?
3. What psychological interventions are most effective at reducing depression and caregiver burden for family carers of terminally ill people?

1.3 Methods

1.3.1 Search Strategy

Studies were identified through electronic databases, grey literature and hand searching. The electronic databases used were MEDLINE, PsychINFO and Web of Science. Grey literature was searched using the online platforms ProQuest, EThOS and SCIE. Papers published until the search date on 28th October 2022 were included.

Search terms were informed by previous studies (Hudson et al, 2010; Chi et al, 2016) with additional terms for psychological interventions and family carers (see Appendix A). Boolean, truncation, and adjacency were used. Adjacency was used with guidance from an experienced library team, which placed carer and intervention terms within three words to manage the difficulties regarding the breadth of unrelated studies identified during scoping searches.

In addition, key subject headings were also included in the search to increase the number of relevant papers. These were searched with key words in the categories of family carer AND palliative AND psychological intervention.

1.3.2 Inclusion and Exclusion Criteria

Inclusion criteria were: (1) An evaluation of a psychological intervention aimed at supporting family carers of adults with a terminal illness. Dementia was included as a terminal illness when there were details of a clear diagnosis of distinct dementia; (2) An intervention for adult carers; (3) Provides a psychological intervention operationalised for the purpose of this review as an evidence-based intervention including counselling, mindfulness, acceptance and commitment therapy, compassion focussed therapy, psychological intervention, all cognitive and/or behavioural therapies and psychoeducation; (4) Collected and reported at least one piece of quantitative data; (5) Has at least one control group; (6) English language; (7) Peer reviewed if published literature; (8) Had depression and/or caregiver burden as primary or secondary outcomes.

Exclusion criteria were: (1) Studies about 'informal carers' whereby the relationship to the care recipient was not a family member or was not reported in sufficient detail to ascertain relationship; (2) Studies which looked at care recipients who had illnesses which may not be terminal in nature (for example dementia without details to determine if this was part of a terminal illness, studies who failed to state a clear diagnosis of distinct dementia for example grouping "other dementia" or dementia associated with a stroke or early stage cancer); (3) Intervention focused on patient not carer; (4) Studies which did not provide a psychological intervention (for example study protocol); (5) Studies which collected only qualitative data; (6) Study designs which did not include a control group such as cross-sectional experimental designs (7) Studies which evaluated interventions which were specifically to support carers during bereavement; (8) Interventions which were not based on psychological theory (for example complimentary therapies). Studies were excluded if the psychological theory used was not obvious or clearly stated. Psychological theory included problem solving, coping skills, cognitive and/or behavioural theory, quality of life, caregiver burden, wellbeing and mindfulness; (9) Studies which do not include depression or caregiver burden as primary or secondary outcomes.

This paper was registered on PROSPERO on 14th November 2022 (CRD42022375088).

1.3.3 Study Selection

The process of selecting eligible papers for this review is demonstrated in Figure 1. The electronic database searching yielded 300 studies. Duplicates were removed, titles were screened for eligibility and abstracts were screened, removing any review papers or papers which were clearly unrelated to the research question. This left 92 papers to be screened for eligibility against the inclusion and exclusion criteria. Of these, 44 papers were screened in full (see figure 1).

An additional five papers were identified through citation searching, and one additional paper was identified through grey literature searching. In total of 18 papers were included in the review.

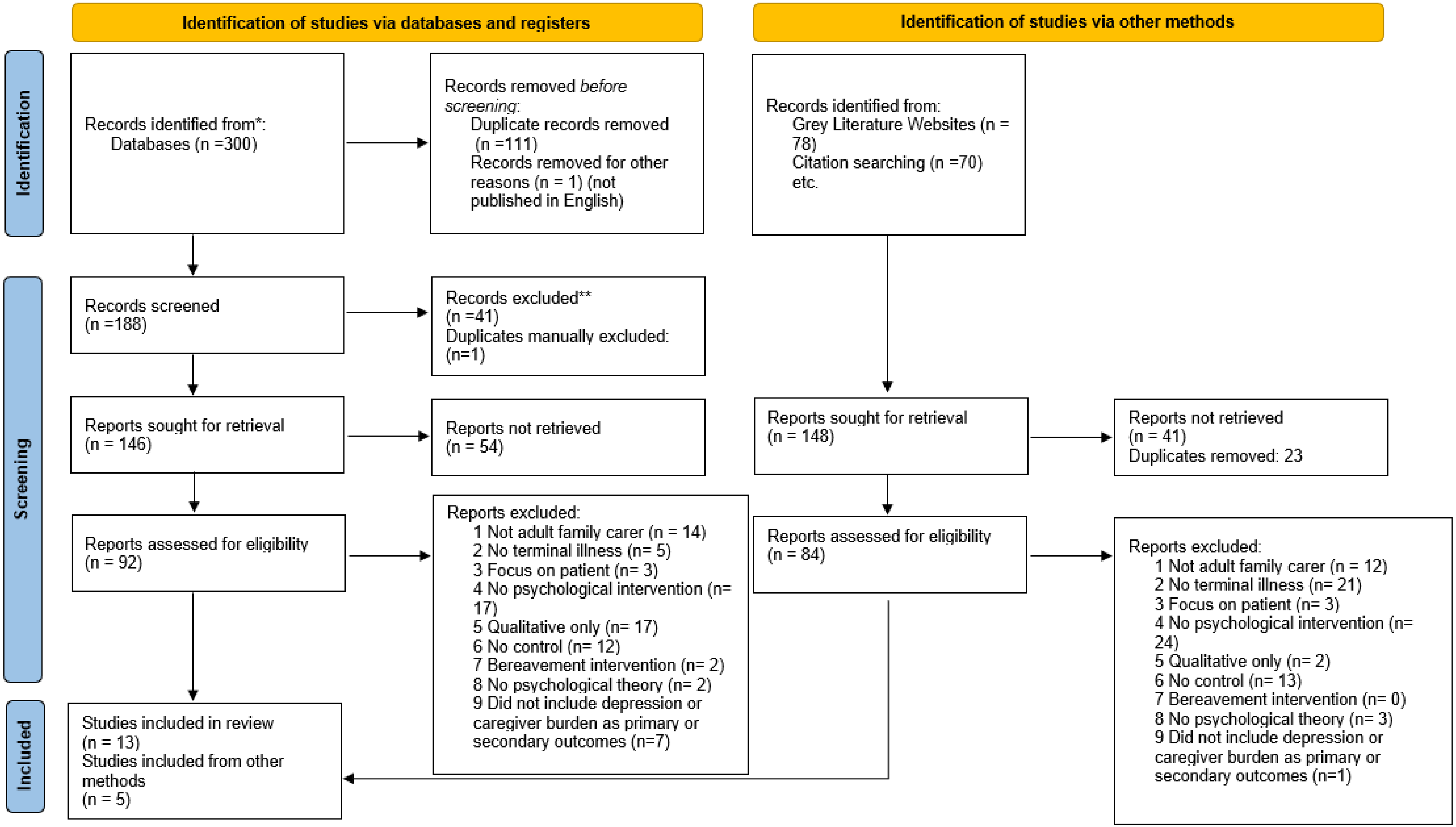

Figure 1: PRISMA flow diagram showing study selection

1.3.4 Quality Rating

The quality of the included papers was evaluated using the Effective Public Health Practice Project Quality Assessment Tool for Quantitative Studies (EPHPP) (Ciliska et al 1998). This tool was considered a suitable length and level of detail for this review. It was suitable to the range of papers identified in this review which included practice-based evidence with waitlist controls and larger scale RCT's. This tool has fair inter-rater reliability for individual component scores and excellent inter-rater agreement of the final grade (Armijo-Olivio et al, 2012). This tool has been used in a similar systematic review (Han, et al 2021).

This tool consists of 22 items examining a range of criteria. It includes component ratings on selection bias, study design, confounders, blinding, data collection methods, withdrawals and dropouts, intervention integrity and analyses. Components are rated strong (1), moderate (2) weak (3) or not applicable. Each paper is then given on overall rating of strong, moderate or weak.

1.3.5 Data Extraction and Synthesis

A data extraction tool was developed and trialled on three papers. The data from a total of 18 papers was extracted.

Effect sizes were reported and calculated where possible.

A narrative synthesis approach was used following guidelines by Popay et al (2006). This included considering four elements: the development of theory, preliminary synthesis, exploration of relationships and assessment of robustness. In the preliminary synthesis, study characteristics and findings were collated to explore similarities and differences between the papers. This included exploring characteristics associated with high rates of caregiver burden such as hours caring per week. Effect sizes were reported and compared between types of intervention and treatment target.

1.4 Results

In total, 18 studies were included in the final analysis (*n*=1850). Of these, 16 examined effects on caregiver's depression and 10 studies examined effects on caregiver's perceived burden.

Table 1: Data extraction table and summary of key findings of all papers in the present review

Authors (date)	**Sample**	**Relationship to patient**	**Study design**	**Intervention**	**Control**	**Treatment target and measure**	**Findings** **(P value, effect size)**	**Limitations**	**Study quality:**
Au et al. (2015)	Carers of people with Alzheimer's in Hong Kong. Intervention group (n=29, age M=55.21) vs. control group (n=30, age M=57.93). Male (n=10), Female (n= 49).	Spouse (n=18), child (n=35), other (n=6)	Longitudinal RCT with 3 timepoints.	Telephone psychoeducation followed by BA sessions (8x biweekly, 15-20 minutes each).	Telephone psychoeducation followed by 8x biweekly telephone sessions (15-20 minutes each).	CES-D (α =0.9)	Intervention group with BA sig. reduced depression (*p*<.001, *d*= 0.56). Control group found increase in depressive symptoms from time 2 to 3.	Sample size small, short duration psychoeducation sessions, demographic information not reported (e.g. ethnicity, employment), recruitment and randomisation process unclear, no reported power analysis, unclear paraprofessional's level of training, treatment adherence not monitored,	**Moderate**
Au, A., Li, S., Lee, K., Leung, P., Pan, P. C., Thompson, L., & Gallagher-Thompson, D (2010)	Chinese carers of people with Alzheimer's. 37- 10 drop out leaving 13 in experimental group, 14 in control. Age (M=54.15), All female.	Daughters: 54%, WLC: 50%. Spouses CWC: 39%, WLC: 29%.	Between subjects two-armed study. Randomly assigned to experimental or control group.	Cognitive behaviours psychoeducation group sessions (13x, weekly, 2 hour) stress coping strategies from CWC manual e.g., relaxation and behaviour modification.	Wait list controls: No group therapy, offered usual activities offered by the centres.	Centre for Epidemiologic studies Depression Scale (CES-D) α =0.85 (good). Both scales validated locally	Used pre scores as covariate Treatment effect on depression in the treatment group was greater when compared to the control group but not significant (F = 4.07, p = .06). Effect size post intervention *d*=0.56.	Small sample, longer term follow up is needed, no detail on recruitment or randomization, no power analysis, not reported number of employed in each condition, severity of illness not reported, higher attrition for experimental condition- no exploration or reason for drop out, how they measured adherence unclear.	**Weak**

Authors (date)	Sample	Relationship to patient	Study design	Intervention	Control	Treatment target and measure	Findings (P value, effect size)	Limitations	Study quality:
Campbell, T. L. (1997)	Spouses of patients with moderate-severe Alzheimer's. n= 205, 86 over age 60, white (90%), Over 40% high levels of depressive symptoms. >50% women, but 43% in treatment group.	All participants were spouses	Between subjects two-armed study. Randomly assigned to groups. Randomization not described.	Family counselling session (4x) and individual session (2x). Education about AD, communication skills and community resources was provided.	Normal service from NYU-ADRC: signposted to resources and agencies, offered support groups (40% of control group attended these) but no formal counselling.	Measured caregiver depression at baseline, and 4, 8 and 12 months later.	No difference between baseline depression after controlling for gender. At 8 months, treatment group significantly less depressed than control group. Improvement in depression was not related to specific measures of support, e.g visits, phone calls, and unpaid help.	No limitations discussed, demographics including hours caring, employment or if living with carer, or power analysis. No details on recruitment of randomization, no details on assessment of moderate dementia, assessors qualifications/training, measures used, attrition rate or adherence. Results not reported transparently unable to calculate effect size,	**Weak**
Cheng, S. T., Mak, E. P., Kwok, T., Fung, H., & Lam, L. C. (2020)	103 Hong Kong Chinese carers or people with mild-moderate Alzheimer's included. Final sample 96 Power analysis: 20 per arm for medium effect. Age (M- 56.16 years), 86% Female.	24% spouse, 67% children, 6% children in law, 3% grandchildren or nieces/nephews	Between subjects, three-armed study. Randomly assigned (double blind) to either experimental or two control groups 4 and 10 month follow up.	Individual benefit finding intervention (4x, biweekly, 3 hour) with_ psychoeducation, modelling, role play, self-monitoring using journals and cognitive reappraisal Three first-degree psychology graduates supervised by the first author facilitated. Adherence monitored.	Simplified psychoeducation (lectures only slower pace than standard and BFT), Standard psychoeducation (lectures and skills practice, slower pace than BFT)	Depressive symptoms (Chinese Hamilton depression rating scale) α=-.72 (acceptable). Global Burden (Chinese Zarit burden interview) α = 0.87 (Good)	No sig group difference on demographics. Diminishing BFT effects over time. Reduced depression in BFT at 4-month follow-up (SIM-PE d = −0.85, p<.001), STD-PE d= −0.75, p=.003). Moderate effect at 10-month follow-up comparing BFT with SIM-PE (d = −0.52, p=.039). No significant difference between BFT and STD-PE. Depression reduced in both control groups. No BFT effect on global burden.	Unclear how it would be applied in clinical practice. Small sample not powered to detect small effects. Convenience sample limiting generalizability. Informed they may be asked to keep diary- this may have given away the treatment condition, Adherence monitored by checklist completed by instructor not independent, no clear details of randomization procedure.	**Strong**
de Rotrou, J., Cantegreil, I., Faucounau, V., Wenisch, E., Chausson, C., Jegou, D., ... & Rigaud, A. S. (2011).	Carers of people with mild-moderate Alzheimer's (MMSE). 167 patient–caregiver dyads from 15 French memory clinics. 141 after dropouts. 6 months: 115 Power analysis: 60 needed. Age (M=65), Female (68%),	Spouse (57.3%), Child (28.7%), other relative (14%)	Multi-centre randomised, single blind trial using parallel groups design. Randomisation was centralised and stratified by independent statistician. Assessed at 0, 3 and 6 months.	Structured group psychoeducation sessions (12, 2 hour, weekly) by a trained psychologist and experienced health professionals. Debriefing: discussed events of the week and solutions were raised from the group. Ecological stimulation: taught how to stimulate relatives in daily activities.	Wait list controls offered intervention at the end of the study.	Depressive symptoms: The Montgomery and Asberg Depression Rating Scale. Caregiver Burden (Zarit Caregiver Burden Interview	Similar demographics between. No significant difference between groups for depression or burden at M0, M3 or M6. At M3 Burden *d=0.09,* Depression *d=0.22,* final follow up Burden *d*= 0.22, Depression: *d=0.28* Depression in carers increased significantly in the control group only suggesting	Difficulty recruiting, dropouts due to availability, lack of statistical power for subgroup, absence of tool to collect and measure effectiveness of stimulation at home. Monitoring adherence, high attrition, reliability of measure, recruitment and referral process not discussed. Employment caring responsibilities not reported. Delivered by a wide range of professionals.	**Moderate**

Authors (date)	Sample	Relationship to patient	Study design	Intervention	Control	Treatment target and measure	Findings (P value, effect size)	Limitations	Study quality:
							the intervention prevented the progression of depression.		
De Stefano, M., Esposito, S., Iavarone, A., Carpinelli Mazzi, M., Siciliano, M., Buonanno, D., ... & Tedeschi, G. (2022).	20 carers of people with early onset Alzheimer's disease. Age (M=53). Cared for >28 hours per week, samples matched by age, gender, and cognitive score.	Spouse (60%), Children (30%), Siblings (10%)	Not clearly defined- appears to be a between subjects, 2-armed design. Participants were randomly assigned o treatment or control group. Measures taken at baseline, at the fifth week and after 6 months.	4 (1 hour) non-directive counselling telephone sessions by one psychologist/psychotherapist.	Control group did not have any intervention however it is not stated whether they were wait list controls.	Depression: (The Zung self-rating Depression scale). Caregiver Burden (The Caregiver Burden Inventory).	No significant baseline difference. Significant decrease in burden at T1 in the treatment group only (p=.011). Depression scores were lower between T0 and T1, and T1 and T2 in the treatment group only, but this was not significant (p=.065, p=.065 respectively). Significant increase in caregiver burden between T1 and T2 and T0 and T2 in the experimental group only (p=.002, p=.025 respectively)	Small sample, no power analysis, drop out, adherence, hours caring, employment or cohabitation, range of MMSE scores, or reliability of measures. Covid-confounding, higher trauma scores in the treatment group at baseline- transfer effects. Table is mislabelled, gender not reported. Insufficient info to calculate effect size	**Moderate**

Authors (date)	Sample	Relationship to patient	Study design	Intervention	Control	Treatment target and measure	Findings (P value, effect size)	Limitations	Study quality:
Fegg, M. J., Brandstätter, M., Kögler, M., Hauke, G., Rechenberg-Winter, P., Fensterer, V., ... & Borasio, G. D. (2013)	133 carers of inpatient palliative care patients (cancer, neurological, other) in Germany. Power analysis (n=110 for power of 0.8). 13.6% inc. rate Age (M= 54.5), Female (69.9%).	Partner (61.7%), Parent (26.3%), Child (3%) other relative (9%)	Parallel-group design (with equal randomisation 1:1) externally created, computer generated. There were five assessments (baseline, pre-treatment, post-treatment, and follow-ups after 3 and 12 months)	6 Face to face existential behavioural therapy group sessions (3h 40). Included discussion of relatives' existential issues and home mindfulness practice. Six facilitators (behavioural therapists) received training and supervision. Treatment integrity evaluated via recording by six independent rates.	The control group had access to usual support (e.g., specialist palliative care staff and bereavement group). The use of other support used as covariates	Depression: (The Brief Symptom Inventory-depression subscale).	Controlled for age, gender, relationship to patient, time of death and other support. No difference between baseline outcomes. No significant effect but trend for effect on depression in the EBT group. 3 Months- multivariate model not significant. Pre/12-month comparison showed medium effect on depression. Depression at pose treatment *d=0.13,* 12 months between intervention and control d=0.27	Sampling: those who were particularly burdened may have consented to the trial. Expectancy effects in treatment group. Mix of relationships affecting heterogeneity, most relatives lost patient during intervention, mix of carers and bereaved. Did not report the intensity of care needs, number of hours caring, cohabitation status, reason for dropouts, or reliability of scale	**Moderate**
Ferrell, B., Kravits, K., Borneman, T., Pal, S. K., & Lee, J. (2019)	240 carers of people receiving palliative care for late-stage cancer. 183 carers left at follow up (24% attrition; 21% for control group; 26% intervention group). Age (M=55), Female (80%), White (48.8), White Hispanic/Latino (31.3%).	84% married/spouse, 16% other family	2 × 2 randomized longitudinal design. Survey at baseline and at 1 month.	Four part 1:1 psychoeducation session organised by QOL domains, assessment of current self-care strategies, and the formulation of a tailored self-care plan. Delivered by registered nurse. Telephone or in person (15-90 minutes)	The control group received usual care consisting of usual nursing care and referral to support services as needed. Received follow up phone call.	Caregiver Burden: Caregiver Burden Scale (Montgomery, Stull & Borgatte, 1985)	No demographic difference between groups. Objective burden was significantly lower at 1 month in control group (p = .04), but not significant for intervention (p = .51). Effect size 1 month: d=0.026. No interactions between burden and distress.	Higher attrition for experimental condition, no details on randomization, blinding, drop out reason or recruitment, reliability of measures, adherence, discussion on limitations, or exploration of the lack of difference in caregiver burden. Lack of conclusion about future directions for research. Intervention adapted to individuals so do not know what was effective and why, Sample-insufficient detail re demographics not split into groups.	**Moderate**
Holm, M., Årestedt, K., Carlander, I., Fürst, C. J., Wengström, Y., Öhlen, J., & Alvariza, A. (2016).	270 carers of palliative care patents from 10 services in Sweden. 98 received intervention, 96 received control. Follow up: n=119 Power analysis: 55 for medium effect Age (M=61.5), Females (66.45%).	Spouse= 48.45%, Adult child=34.54%, other relative= 12.89%	A 2x2 between groups randomized control trial (RCT) in accordance with the CONSORT statement. Computer generated stratified randomization. Data collection at baseline, post intervention, and 2 months after.	Manualised psycho-education group sessions (3x, 2 hour, weekly). Delivered by health professionals (physician, nurse, social worker/priest) at each care home. Topics: palliative diagnoses and symptom relief, daily care and nutrition and support and existential issues.	Treatment as usual: including practical and emotional support and opportunities for individual support, such as supportive talks and information.	Caregiver Burden Scale (Elmstahl Malmberg & Amner, 1996) α=0.7-0.87 (acceptable-good). Depression: The Hospital Anxiety and Depression Scale- α =0.82 (good).	No sig difference at baseline. No sig difference between groups for caregiver burden or depression short term (p= .597-.843, p= .316). No sig difference between groups for caregiver burden or depression long term(p= .376-.819, p= .290). Follow up 1 and 2 interventions had higher depression scores (d= 0.08, d=0.12). Burden- no overall score but general strain intervention slightly higher at follow up 2 (*d*= 0.14)	No information on adherence, training of facilitators, ethnicity, number of hours caring, reasons for drop out or exploration of this, severity of condition or needs, or final burden score. Almost half the sample not living with the care recipient, high attrition as could not continue if participant died Mainly cancer patients but no further details- lack of homogeneity. Delivered by wide range of professionals.	**Strong**

Authors (date)	Sample	Relationship to patient	Study design	Intervention	Control	Treatment target and measure	Findings (P value, effect size)	Limitations	Study quality:
							no difference at follow up 1 for general strain (*d=0)*		
Hudson, P. L., Aranda, S., & Hayman-White, K. (2005)	106 caregivers (358 eligible) of people receiving palliative care for advanced cancer in Australia. Power for medium/large effect. Age (M=60.78), Female (65.1%), Nationality (Australian= 74.3%), Time 3 n=45	Spouse: 66.7%, child: 16%, parent: 7.6%	2x2 between groups randomized controlled trial with piolet phase. Data collected at T1 bassline, T2 (5 weeks from baseline), T2 (8 weeks following patient death).	A psycho-educational intervention based on coping and self-care. Home visits (2x) by a nurse, follow up phone call between visits. Provided information and evaluated strategies. Given reflections from carers, self-care strategies and structured relaxation exercise	Usual care which included access to 24-hour phone advice and emergency visits from nurses in addition to pre-scheduled home visits from specialist nurses, doctors, and allied health professionals.	Depression: The hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983) not reported specifics for HADS but α for all measures in the study above .7 (adequate)	No significant difference between groups at baseline. 12 intervention and 15 control participants completed all 3 time points. Collected info from HADS but only reported anxiety not depression	No details on randomization, hours caring, reliability for HADS, demographics, training of nurses, adherence, or recruitment. High attrition particularly for intervention with no reasons for drop out.	**Weak**

Authors (date)	Sample	Relationship to patient	Study design	Intervention	Control	Treatment target and measure	Findings (P value, effect size)	Limitations	Study quality:
Kögler, M., Brandstätter, M., Borasio, G. D., Fensterer, V., Küchenhoff, H., & Fegg, M. J. (2015)	160 carers of palliative patients in Munich. 19 dropped out and 11 had missing data. Diagnosis: Cancer 88.8%, neurological= 13.8%, other= 5.4% Female (70.8%), Age (M=54.3),	Partner: 61.5%, Child= 26.9%, Parent= 3.1%, Other relative= 8.5%	2 arm between groups randomised control trial (1:1 randomization). Measures completed pre-intervention (T1), post-intervention and 6 weeks after T1 (T2), 3 months post-intervention (T3), 12 months after intervention (T4).	EBT group intervention (6x, 22-hour total) led by a psychotherapist. Included session and home mindfulness practice (2x daily, 15 minutes).. 10 groups were run with 7.3 +- 1.1 participants.	Treatment as usual- no further details regarding what this involved.	Depression: Brief Symptom Inventory (depression subscale)	Regression controlled for gender and age. No difference between mindfulness in control and intervention at T1. Mindfulness in informal caregivers of PC patients was significantly correlated with lower psychological distress. Grouped brief score so cannot compare depression between groups	Heterogeneity and varied relationships meaning not generalizable, no statistical analysis of baseline demographic differences and insufficient consideration of covariates. Did not discuss effectiveness or provide insufficient data for effect size to be calculated. Hours caring not reported. No power analysis, reliability of measures, discussion around adherence, randomization, recruitment, or dropouts.	**Moderate**
Leow, M. (2015)	Family caregivers from National Cancer Centre Singapore. Caring >20 hours per week. 80 participants, Power analysis: medium effect. Age (M=47.16, Female (67.5%), Nationality: Chinese (85%), Malay (10%), Indian (3.75%), Caucasian (1.25%).	Child: 57.5%, Spouse: 25%, Sibling: 3.75%, Parent: 2.5%	A piolet randomized controlled trial using computer randomization. Two group pre-test and post-tests. Data collected at baseline, 4 weeks post-intervention and 8 weeks post-intervention.	Caring for caregiver's program- 1:1 psychoeducation (1x, 1 hour), video clip, follow up phone call (2x, week 3 and 6, 15-30 minutes) and invite to online social support group. Delivered by one nurse researcher	Routine care from their respective home hospice organization, which included regular weekly to monthly visits (about 30 minutes) from a home hospice nurse.	Depression: Depression Anxiety Stress Scales (Lovibond & Lovibond, 1995). α =0.91 (Excellent). Burden: The Caregiver Quality of Life Index- Cancer (CQOLC) Burden subscale (Weitzner et al, 1999). α=0.91 (excellent)	Significantly higher retention for intervention group time1 & 2 (p<.001, p=.01) Significantly lower depression (p=.02) and burden subscale for intervention (p=.004). Depression baseline to week 8 increased for control but may be due to dropouts (d=.14), decreased for intervention d=.67. Time 1 control vs intervention scores were lower d=0.66, Time 2 control and intervention scores lower d-.68. Burden subscale: Time 1 intervention higher than control *d= 0.64* post 2 control v intervention burden scores higher (d=.75)	Possible sample bias due to convenience and English speaking. No blinding. Intervention process is unclear. Intervention individualised therefore unclear what was effective. High attrition. Failed to address increased burden scores after the intervention. Employment status, hours per week caregiving, cohabitation status not reported. Training of facilitator, adherence to protocol not discussed.	**Moderate**

Authors (date)	Sample	Relationship to patient	Study design	Intervention	Control	Treatment target and measure	Findings (P value, effect size)	Limitations	Study quality:
Pahlavanzadeh, S., Dalvi-Isfahani, F., Alimohammadi, N., & Chitsaz, A. (2015)	70 MS caregivers in Iran (power analysis needed 64). Evenly allocated. Age (M= 43.5), Female (42.9%) Employed: intervention 48.6%, control 45.7%. Disease severity: Intervention 3.2, Control: 3.1.	Parent: 34.3%, Spouse: 48.6%, Child: 2.9%, Immediate family: 14.3%	Between group 3x2 design. Randomly allocated by draw. Data collected before, immediately after and 1 month after intervention	Psycho-education program was administrated in study group (7x, weekly, 90 minute) by a master of psychiatric nursing. Included relaxation, and emotional control techniques.	The control group underwent no intervention but received the educational resources after the intervention.	Caregiver Burden: Zarit Burden Scale. Test-retest reliability calculated at 94%	Groups "almost homogenous" in terms of demographics. No sig difference between time points in control group. No significant difference before intervention in both groups. Significant reduction in family caregivers' burden immediately after and 1-month after intervention in the study group, compared to control (p<.001). Insufficient information to calculate effect size.	Short follow up. Did not explore meaning of "almost homogenous." Their table says control before was 82.61 but in the text, it is reported at 61.82. No details on drop out, training, adherence, randomization and recruitment. Hours caring not reported. Large variation in disease severity. On average lower end of disability.	**Moderate**
Marriott, A., Donaldson, C., Tarrier, N., & Burns, A. (2000).	42 patient carer dyads of 77 identified from NHS old age psychiatry service. Stratified for gender and health status score. 14 allocated to each group 13 in intervention after drop out. Age (M=63.6), Female: 69%. Carers GHQ score >5	Spouse- 52%, offspring: 40.5%, Sibling: 7%	Single-blind randomised controlled trial of family intervention compared with 2 control groups with blind and independent assessment including a 2 month and 12 month follow up. 3x3 design.	Psychoeducation, stress management and coping skills training using cognitive behavioural approach over 14 sessions by consultant clinical psychologist. 3 face to face education sessions on dementia, 6 session stress management with assessment. 5 coping sessions.	CFI interview control- received an audiotaped semi structured interview which derived expressed emotion ratings.	Depression: Beck Depression Inventory (Beck 1988)	No sig difference at baseline. Invention group depression dropped and significantly lower than no interview post treatment (P<.01) and interview control (p=.002). Effect size no-interview control vs intervention d= 0.56, interview control vs intervention d= 0.60. At follow up sig difference between intervention and no interview (p=.001) and interview control (p=.004). No sig difference between two control groups. Intervention v interview control at follow up: d=.79, intervention v no interview control at follow up: d=0.69.	Paper did not outline limitations. More spouse caregivers and less offspring care givers in intervention group, intervention group had cared for longer. Delivered by consultant psychologist- not be representative of clinical practice. Small sample. Relatively old paper- many social differences since this time in resources for carers. We know they were significantly distressed but not necessarily that this was due to caregiving demands. No details on randomisation, recruitment, reliability of measures, what the covariates were or adherence.	**Moderate**

Authors (date)	Sample	Relationship to patient	Study design	Intervention	Control	Treatment target and measure	Findings (P value, effect size)	Limitations	Study quality:
Oken, B. S., Fonareva, I., Haas, M., Wahbeh, H., Lane, J. B., Zajdel, D., & Amen, A. (2010).	Oregon USA (n=31) age (M=64.46) caring for family with progressive dementia >12 hours p/w, >9 on perceived stress scale. Ethnicity White (n= 28, 90.32%), African American (n=1, 2.91%), Asian (n=2, 6.45%). Female= 80.65%	Spouse (n=23, 74.19%), Child (n=8, 25.81%).	A between groups 3x2 pilot randomized trial using dynamic randomization. Researchers unblind, outcome assessments blind. Data collected at baseline and 8 weeks post-intervention.	Mindfulness based cognitive therapy group program. Two active group interventions: Both groups given 3 hours respite care and psycho education. MBCT intervention (6x, weekly, 90 minute). Directed by a clinical psychologist trained in MBCT. 2. Education: 6 weekly lectures by people trained in PTC. Covered self-care skills e. communicating skills, changing self-talk.	Respite only- respite for 3 hours once per week for 7 weeks. Time chosen by carer. Offered either MBCT or PCT after study.	Depression: Centre for Epidemiologic Studies Depression Scale (CESD)	Depression scores correlated with Mindful Attention Awareness Scale (p<.05). No significant effect of intervention on depression (p=.705). Depression score at baseline lower than both controls. Meditation v control post d= 0.30, education v control post d= 0.01	Outcome measures may not be well matched to potential benefits. Not explained how depression links conceptually to the goal of stress reduction. Did not explore individual predictors/effect modifiers due to lack of power. More women than men. Failed to address increased drop out in intervention. Did not explore whether difference between depression scores at start of research were significant. Hours caring, employment, reliability or adherence not reported. PSS score of 9 to participate but this is within the low range.	**Moderate**
Cheng, S. T., Chan, W. C., & Lam, L. C. (2019)	129 carers of people with mild-moderate Alzheimer's in Hong Kong, >14 hours per week. Baseline n=132, 2 months n=129, 6 months n=107, 12 months n=101. Female= 83.3-88.9%, age M= 55.22	Spouse/sibling= 27.01%, Child/child in law/ nephew/ niece = 72.93%	Cluster-randomized double-blind controlled trial. Assessments at baseline and post treatment. Randomized using true random number generator, 5 clusters per arm.	Benefit finding group intervention (8x, weekly, 120 minutes, 7-11 people M=8.6) delivered by people with undergraduate degree in psychology or related discipline trained by first author. Psychoeducation with positive reappraisal. Included videos of carers sharing their experiences.	Simplified psychoeducation (same content as BF without practice, lectures only slower pace), Standard psychoeducation (lectures and skills practice, slower pace). 8x weekly, 120 mins each.	Depression: Hamilton Depression Rating Scale. α=0.76 (Acceptable) Burden: Zarit Burden Interview α=0.9 (Excellent)	Covariates= age, sex, illness, care hours, BPSD and functional impairment. Trend for decrease in depressive symptoms and burden over time. Significant differences between BFT and SIM-PE at T3: d=-0.89, p<0.001; and T4: d=-0.70, p=0.001. STD-PE, d=-0.80, p<0.001 (T3) and d=-0.65, p=0.003 (T4). For global burden, as time progressed, the effect of BFT slightly increased. Less burden in BFT than SIM-PE and STD-PE participants at T3 d=-0.55, p=0.012, T3 d=-0.47, p=0.034) and T4 d=-0.56, p=0.010, d=-0.51, p=0.022).	Sample size not big enough for small effect sizes, did not add both previously identified mediators. Not reported if living with care recipient. Adherence monitored by facilitator using checklist with no objective measure. Not reported updated demographics after dropouts. Not reported ethnicity, very strongly female. Grouped spouse/sibling in demographics.	**Moderate**

Authors (date)	Sample	Relationship to patient	Study design	Intervention	Control	Treatment target and measure	Findings (P value, effect size)	Limitations	Study quality:
Cheng, S. T., Fung, H. H., Chan, W. C., & Lam, L. C. (2016)	129 carers of people with mild-moderate Alzheimer's in Hong Kong, >14 hours per week. Baseline n=132, 2 months n=129, 6 months n=107, 12 months n=101. Female= 83.3-88.9%, age M= 55.22	Spouse/sibling= 27.01%, Child/child in law/ nephew/ niece = 72.93%	Cluster-randomized double-blind controlled trial. Assessments at baseline and post treatment. Randomized using true random number generator, 5 clusters per arm.	Benefit finding group intervention (8x, weekly, 120 minutes, 7-11 people M=8.6) delivered by people with undergraduate degree in psychology or related discipline trained by first author. Psychoeducation with positive reappraisal. Included videos of carers sharing their experiences.	Simplified psychoeducation (same content as BF without practice, lectures only slower pace), Standard psychoeducation (lectures and skills practice, slower pace). 8x weekly, 120 mins each.	Depression: Hamilton Depression Rating Scale. α=0.78 (Acceptable) Burden: Zarit Burden Interview. α=0.9 (Excellent).	No sig group difference in baseline variable. BF treatment effects on all outcomes when compared with SIM-PE, and effects on depressive symptoms and Zarit burden when compared with STD-PE. Effect sizes were medium-to-large for depressive symptoms (SIM PE p=.002, d=.77, STD PE p=.019, d=.96), and medium burden (SIM PE p=.001, d=0.47, STD PE p=.001, d=0.65).	Sample not large enough for small effect size, did not add both mediators. Not reported cohabitation status or ethnicity. Adherence monitored by facilitator using checklist with no objective measure. Not reported updated demographics after dropouts. Very strongly female. Grouped spouse/sibling in demographics. Caregivers mildly depressed so dropping 3 points on the scale yielded a large effect size.	**Strong**
Cheng, S. T., Mak, E. P., Fung, H. H., Kwok, T., Lee, D. T., & Lam, L. C. (2017).	103 carers of people with mild-moderate Alzheimer's in Hong Kong. >14 hours caring per week. Baseline= 103, 2 months= 96 Age (M=56.18), total 81 female (84.38%),	Spouse: 23 (23.96). child/child in law/ niece/ nephew= 73 (76.04%)	3 arm between groups double blind randomised controlled trial. Assessments at baseline and 2 months post treatment.	Benefit finding 1:1 intervention (4x, biweekly, 3 hour) delivered by people with undergraduate degree in psychology, trained by first author. Psychoeducation on self-care and stress management with positive reappraisal. Included videos of carers sharing their experiences and encouraged to keep journals.	Simplified psychoeducation (same content as BF without practice, lectures only slower pace), Standard psychoeducation (lectures and skills practice, slower pace). 4x bi-weekly, 3 hours.	Depression: Hamilton Depression Rating Scale. α= 0.72 (Acceptable). Burden: Zarit Burden Interview. α= 0.81 (Good)	No sig difference at baseline for demographics or measures but severity of dementia approaching significance. Being married, education, chronic illness and BPSD were used as covariate. BF participants reported lower depressive symptoms after treatment, when compared with either SIM-PE (p=.001, d= 0.46) or STD-PE (p=.001, d= 0.50) participants.	No data to explain justification for low attrition. Being told they may use journals may have shown condition. Paid for home visits- risk of demand characteristics. Small sample re generalizability and power, difference in severity of dementia in groups, no follow up, participants minimally depressed so slight change led to moderate effect size. Reliability of depression measure just within the acceptable range	**Moderate**

1.4.1 Characteristics of Studies

As per the exclusion criteria, all studies in this review used a control group. As demonstrated in Appendix B, most studies used an active or treatment a usual control.

1.4.1.1 Types of Therapeutic Intervention

Table 2 shows how the studies were categorised based upon the psychological theory underpinning the intervention. The most common interventions across studies were CBT and psychoeducation.

Table 2: *Characteristics of interventions of studies*

Intervention Type	Description	Number of Studies
Cognitive Behavioural Therapy	Interventions focused on targeting caregivers' thinking and behaviour to change their emotional experience. This included behavioural activation, thought challenging and benefit finding	7
Existential behavioural therapy	An intervention developed for carers of palliative care patients which builds upon cognitive behavioural approaches to consider meaning, death and acceptance	2
Mindfulness based cognitive therapy	Intervention which combines cognitive behavioural strategies with mindfulness practice to increase awareness and understanding of thoughts to reduce emotional suffering	1
Psychoeducation	Interventions focused on learning and understanding about mental health and wellbeing	6
Counselling	Interventions which aim to support caregivers by offering the opportunity for them to talk through their feelings	2

1.4.1.2 Outcome Measures

Table 3 shows the variability in outcome measures used. One study did not report which measure they used. Out of the 16 studies which measured depression, 8 did not report a Cronbach's alpha. Of the 10 studies which measured caregiver burden, 4 did not report a Cronbach's alpha.

Table 3: Summary of outcome measures used

	Measure Used	Number of Studies	Cronbach's Alpha Range
Caregiver Burden	The Caregiver Burden Inventory	1	- *
	Caregiver Burden Scale	2	0.7-0.85*
	The Caregiver Quality of Life Index- Cancer	1	0.91
	Zarit Burden Interview	4	0.81-0.9 *
	Zarit Burden Interview (Chinese Version)	1	0.87
	Zarit Burden Scale	1	-*
Depression	Beck Depression Inventory	1	- *
	The Brief Symptom Inventory- Depression Subscale	2	- *
	Centre for Epidemiologic Studies Depressive Scare (CES-D)	3	0.85- 0.9*
	Chinese version of the Hamilton Depression Rating Scale	1	0.72
	The Depression Anxiety Stress Scale	1	0.91
	Hamilton Depression Rating Scale	3	0.72-0.78
	The Hospital Anxiety and Depression Scale	2	0.7-0.82
	The Montgomery and Asberg Depression Rating Scale	1	- *
	The Zung Self-Rating Depression Scale	1	- *

*Note: * Cronbach's Alpha not reported.*

1.4.1.3 Participant Demographics

The studies varied in terms of demographic information (Appendix B) including relationships to care recipient, gender, ethnicity, employment status, cohabitation status, hours per week caring and illness of care recipient. In 11 papers, most of the carers were spouses and in 4 studies, most were children of the care recipient. Three studies grouped relationships and 7 studies did not specifically report relationships. Most participants in the studies were female and one study had majority male participants. Ethnicity was reported in only four papers. Of these most participants were “white,” Australian and Chinese.

Between 29.2-50.5% of carers were in full-time employment and 11 studies did not report employment status. The number of carers who lived with the care recipient ranged between 24.72%-100% of the sample and 8 studies did not report cohabitation status. In total 10 studies did not report the number of hours per week spent caring.

Most studies focused on carers of individuals with Alzheimer’s disease. A total of 4 studies took place in palliative care services and therefore, care recipients had a mix of illnesses e.g., cancer, neurological illness or terminal illness categorised as ‘other.’

1.4.1.4 Attrition

Some studies had a large attrition rate (Table 4). Oken et al (2010), Pahlavanzade et al (2015), Kögler et al (2015), De Stefano et al (2022), Campbell et al (1997) and Au e al (2015) failed to provide information regarding dropouts. While Marriott et al (2000) had an attrition rate of 2.44% with a 2 month follow up, Hudson and Hayman-White (2005) had a high attrition rate particularly in the intervention group (63%), and they failed to explore this. The studies with the highest attrition rates (>30%) were psychoeducational interventions. Of the studies with the lowest attrition rates (<15%) the majority were CBT interventions, and 1 was a mindfulness-based CBT intervention.

Table 4: Summary of attrition rates in the control and intervention groups of the included studies

	Number of Studies		
Attrition Rate (%)	Intervention	Control	Total Only*
0-10	4	4	
11 -20	3	2	1
21- 30	3	5	
31- 40	1	2	
41- 50	2	1	
51- 60	0	2	
61- 70	1	0	

*Note * includes studies which only reported an overall attrition rate and did not include sufficient information for attrition rates for intervention and control group to be calculated*

1.4.2 Quality of Studies

The quality of the papers was assessed using the EPHPP (Ciliska, et al 1998) and can be found in appendix C. This assesses quality over six domains with an overall quality rating.

Most papers were rated as ‘moderate’ quality, with only 3 papers achieving a ‘strong’ rating (Cheng et al, 2020; Holm et al, 2016; and Cheng et al, 2016). Areas of strength were study design and cofounders. Only Campbell (1997) received a weak rating as it failed to provide details regarding its design. In total 11 papers were rated as ‘strong’ for cofounders. Cheng et al (2020), Marriott et al (2000), Cheng, Chan and Lam (2019) and Cheng et al (2017) used multiple regression analysis including cofounders. This may have managed some of the difficulties noted in sampling and improve the validity and reliability of their papers. Marriott et al (2000) however did not include important covariates, therefore this paper was rated ‘moderate’ for cofounders. None of the papers found significant difference between the groups at baseline for demographics. Pahlavanzade et al (2015) described the groups as “almost homogenous.”

Key issues were selection bias, data collection methods and withdrawals and dropouts. In total, 8 papers were rated ‘weak’ in selection bias. The papers used opportunity sampling possibly resulting in selection bias. Many papers failed to detail recruitment processes. Hudson Aranda and Hayman-White (2005), Marriott et al (2000) and Fegg et al (2013) were the only papers to provide overall inclusion rates relative to eligible participants (29.6%, 54.44% and 13.6% respectively). Although this method increased transparency in the paper, Hudson, Aranda and Hayman-White (2005) and Fegg et al’s (2013) low inclusion score resulted in a ‘weak’ rating.

Regarding data collection, 7 papers were rated ‘weak’ and 4 rated ‘strong.’ Papers lacked detail regarding management of missing data. De Rotrou et al (2011) and Holm et al, (2016) used mean imputation and only Chen et al (2019) described using intent to treat analysis. Kögler et al (2015) removed participants who presented with any missing data. Although this may increase validity with individual change, useful data may have been lost. This lack of transparency poses difficulties in assessing validity. Only 4 papers discussed the validity of the measures. Of the 10 papers who reported reliability of the measures, the majority were within the ‘good’ to ‘excellent’ range. Cheng et al (2017), Cheng et al (2015), Cheng et al (2019) and Cheng et al (2020) used the Hamilton Depression Rating Scale which had a Cronbach’s alpha within the ‘acceptable’ range. Hudson et al (2005) did not provide clear details regarding the reliability of their depression measures.

There was variation in dropout rates between the papers. In total, 6 papers were rated ‘weak’ however 6 were also rated ‘strong.’

1.4.3 Outcomes for Depression

In total, 16 papers looked at outcomes for depression. Of these, 7 used CBT approaches. Papers were categorised as CBT if they referenced CBT principles as their theoretical orientation and included skill development.

Of the 5 papers which analysed post-treatment scores, 3 found significant results: Marriott et al (2000), Cheng et al (2016) and Cheng et al (2017) with a medium to large effect size. However, for Cheng et al (2016) participants were only mildly depressed, therefore small changes to depression scores may have resulted in large effect sizes. Au et al (2010) found no evidence of significant effects, possibly due to the small sample (*n=27*) and ‘weak’ quality.

Table 5 Summary of significance rate and effect size for CBT interventions on depression scores

Paper	Sig post intervention?	Effect size	Sig follow up?	Effect size	Sig follow up 2?	Effect size
Au et al (2015)	No (p=0.26)	d= 0.21	Yes (p<.001)	d=.056	-	-
Au et al (2010)	No (p=.06)	*	-	-	-	-
Cheng et al (2020)	-	-	Yes- SIM PE (p<001), STD PE (p=.003)	SIM-PE d=0.85, STD PE d=0.75	Yes SIM PE (p=.039) No STD PE**	d= 0.52
Marriott et al (2000)	Yes NIC (p<.01), IC (p=.002)	NIC d=0.56, IC d=0.6	Yes NIC p=.001), IC (p=.004)	NIC (d=0.79) IC (d=0.69)	-	-
Cheng, Chan & Lan (2019)	-	-	Yes SIM PE (p=.001), STD PE (p=.003)	SIM PE (d=0.89), STD PE (d=0.80)	Yes SIM PE (p=.001), STD PE (p=.003)	SIM PE (d=0.7), STD-PE (d=0.65)
Cheng et al (2016)	Yes SIM PE (p=.002), STD PE (p=.019)	SIM PE (d=0.77), STD PE (d=0.96)	-	-	-	-
Cheng et al (2017)	Yes SIM PE (p=.001), STD PE (p=.001)	SIM PE (d=0.46), STD PE (d=0.50)	-	-	-	-

*Note *Significant difference between groups pre score. Pre score used as covariate therefore effect size end of treatment not possible to analyse. **Not reported. STD PE= Standard Psychoeducation, SIM PE= Simplified Psychoeducation, NIC= No Interview Control, IC= Interview Control*

Both papers which looked at follow-up found that the intervention significantly reduced symptoms of depression relative to controls, with medium-large effect sizes. Both papers which looked at follow-up only, found that effect sizes reduced between follow-up one and follow-up two. However, for Cheng et al (2019) these effect sizes remained large and for Cheng et al (2020) the effect size remained medium. Two papers looked at post-intervention and follow up (Au et al, 2015; Marriott et al, 2000) and both found the effect sizes increased at follow up, suggesting that impact on symptoms of depression increased with a delay. Au et al (2015) found no significant difference between depression scores for intervention vs controls post-intervention, however at follow-up controls increased and intervention decreased with a significant difference.

All papers which found significant results used regression models, therefore effect sizes were adjusted to consider covariates. Caution should therefore be taken when comparing these findings with other analysis methods. These findings suggest that CBT interventions are beneficial in reducing depression symptoms for carers of people with terminal illnesses in the short and long-term.

Table 6: Summary of significance rate and effect size for Existential Behavioural Therapy interventions on depression scores

Paper	Sig post intervention?	Effect size	Sig follow up?	Effect size	Sig follow up 2?	Effect size
Fegg et al (2013)	No (*p*=0.09)	*d*=0.13	No (*p*=.54)	*d*=0	Yes (*p*=.04)	*d*=.27

In total, 2 papers investigated the impact of existential behaviour therapy on depression scores (Table 6). Although Kögler et al (2015) collected data on the effectiveness of the intervention, this was not clearly reported. Fegg et al (2013) used multivariate analysis which was not significant post-treatment or at 3-month follow-up. However, this was significant at 12 months. The paper reported this was a ‘medium’ effect however did not report Cohen’s *d*. This was calculated for the purpose of this review as *d*= 0.27. This paper had some methodological issues including failure to explore covariates. Furthermore, the limited number of papers looking at existential behaviour therapy means it is difficult to ascertain its effectiveness on symptoms of depression for carers of terminally ill patients, particularly at post-treatment.

A single study examined mindfulness-based cognitive therapy: Oken et al (2010). This paper failed to find a significant effect on depression scores. However, it failed to monitor adherence to mindfulness practice and used a small sample which may have contributed to the null findings. It also failed to explore covariates which may have created error such as hours caring. Finally, the paper aimed to reduce stress for carers, however, it failed to adequately explore how this linked conceptually with depression. This review therefore has not found evidence that mindfulness based cognitive therapy is beneficial in reducing depression scores for carers of people with terminal illnesses.

Table 7: Significance rate and effect size for psychoeducation interventions on depression scores

Paper	Sig post intervention?	Effect size	Sig follow up?	Effect size	Sig follow up 2?	Effect size
de Rotrou et al (2011)	No (*p*=.21)	*d*=0.22	No (*p*=.21)	*d*=0.28	-	-
Holm et al (2016)	No (*p*=.32)	*d*=0.08	No (*p*=.29)	*d*=0.12	-	-
Leow (2015)	Yes (*p*=.02)	*d*=0.68	-	-	-	-

In total, 4 papers investigated the impact of psychoeducation on depression (table 7). Hudson et al (2005) collected depression ratings but failed to report them. De Rotrou et al (2011) and Holm et al (2016) failed to monitor adherence and interventions were delivered by multiple individuals which may contribute to the lack of significant effects. Only Leow (2015) found a significant reduction in depression scores in the intervention group relative to controls with a medium effect size. This suggests that this may be a beneficial treatment for carers of terminally ill people. However, there were significantly more dropouts in the control condition than the intervention and depression scores increased in the control group at follow up. Therefore, the sample may have been biased. In addition, the study adapted to the individual, therefore it is difficult to establish what element of support was beneficial. To conclude, there is limited evidence of the effectiveness of psychoeducation for carers of people with terminal illnesses.

Finally, 2 papers explored the impact of counselling on depression scores: Campbell et al (1997) and De Stefano et al (2022). Although Campbell et al (1997) claimed to find significant improvements in depression, this paper fail to provide specific scores to allow for comparison. De Stefano et al (2022) did not find a significant difference between intervention and control post intervention or at follow up (*p*=.16; *p*=.65). However, the intervention group had significantly higher trauma scores at each time point (p=.008, p=.05, p=.049). In addition, this paper did not report or control for gender and the sample size was small. Therefore, possible covariates and methodological issues may have impacted the findings and high trauma scores may have mitigated benefits which could be gained in the intervention group. There is insufficient evidence from this review to suggest that counselling is a beneficial treatment in reducing depression symptoms for carers of terminally ill people.

To conclude, CBT interventions appear to be the most effective treatment for carers of terminally ill people, however there is an absence of quality research in other therapeutic approaches, and some promising results have been identified in existential behaviour therapy.

1.4.4 Outcomes for Caregiver Burden

Table 8: Significance rate and effect size for CBT interventions on caregiver burden scores

Paper	Sig post intervention?	Effect size	Sig follow up?	Effect size	Sig follow up 2?	Effect size
Cheng et al (2016)	Yes SIM PE (*p*=.001), Yes STD PE (*p*<001)	SIM PE (*d*=0.47) STD PE (*d*=0.65)	-	-	-	-
Cheng et al (2017)	No SIM PE (*p*=.85) No STD PE (*p*=.42)	SIM PE (*d*=0.04) STD PE (*d*=0.22)	-	-	-	-
Cheng et al (2019)	-	-	Yes SIM PE (*p*=.012) Yes STD PE (*p*=.032)	SIM PE (*d*=0.55) STD PE (*d*=.47)	Yes SIM PE (*p*=.01) Yes STD PE (*p*=.022)	SIM PE (*d*=0.56) STD PE (*d*=0.89)
Cheng et al (2020)	No SIM PE (*p*=.67) No STD PE (*p*=.19) *	-	-	-	-	-

*Note: * Paper provided regression significance grouping time points and did not provide effect sizes for no-significant results.*

STD PE= Standard Psychoeducation, SIM PE= Simplified Psychoeducation

In total, 10 papers measured caregiver burden. Of these, 4 used CBT (table 8). Of the 3 papers which looked at effects post-intervention, 2 papers did not find significant effects on caregiver burden scores even though these papers were rated moderate-strong in terms of quality and used scales with good-moderate reliability. However, Cheng et al (2016) found that caregiver burden scores were significantly lower in the benefit-finding group compared with the simplified psychoeducation group and the standard psychoeducation group with low-medium effect sizes. In addition, Cheng et al (2019) found that the benefit-finding group had significantly lower caregiver burden scores compared with simplified psychoeducation and standard psychoeducation at follow-up one and follow-up two. Effect sizes were medium at the first follow-up and increased at the second follow-up. Therefore, CBT interventions can be effective in reducing caregiver burden for carers of people with terminal illnesses in the short-term, with a possible increase in effectiveness at long-term follow-up. However, there is variability between studies regarding the significance and consistency of the benefits of CBT interventions.

Table 9: Summary of significance rate and effect size for psychoeducation interventions on caregiver burden scores

Paper	Sig post intervention?	Effect size	Sig follow-up?	Effect size	Sig follow-up 2?	Effect size
Ferrell et al (2019)	No (*p*=.34)	*d*=0.03	-	-	-	-
de Routrou et al (2011)	No (*p*=.55)	*d*=0.09	No (*p*=.25)	*d*=0.22	-	-
Holm et al (2016)	No (*p*=.32)	*d*=0	No (*p*=.29)	*d*=0.12	-	-
Leow (2015)	Yes (*p*=.004)***	*d*=0.64**	Yes (*p*=.004)***	*d*=0.75**	-	-
Palavanzade et al (2015)	Yes (*p*<.001)	*	Yes (*p*<.001)	*	-	-

Note: *Insufficient information to calculate effect size, **Burden scores higher in the intervention group, ***Regression analysis grouped significance scores across time points.

In total, 5 papers explored the impact of psychoeducation on caregiver burden (table 9). Of these, only Palavanzade et al (2015) and Leow (2015) found a significant improvement in the intervention group relative to the control group. Palavanzade et al (2015) reported that the experimental condition had significantly lower caregiver burden scores at end of treatment and follow-up, however they provided insufficient data to calculate effect size. Leow (2015) identified significant differences between the experimental and control conditions at end of treatment and follow-up however, this paper failed to acknowledge that burden scores were higher in the intervention group at end-of- treatment and follow-up despite no difference between groups at baseline. As such, this review does not provide evidence that psychoeducation is beneficial in reducing caregiver burden for carers of terminally ill people.

Finally, De Stefano et al (2022) was the only study to explore the impact of counselling on caregiver burden. This found no significant difference in caregiver burden between intervention and control at any of the three time points. The only significant finding came from an increase in caregiver burden from post-treatment to follow-up 2 in the treatment group ($p=.025$). This may be associated with the increased trauma scores within the intervention group. To conclude, this review found no evidence of effectiveness of counselling approaches in reducing caregiver burden for carers of people with terminal illnesses.

To conclude, there is limited evidence of effectiveness of psychological therapies in reducing carer burden for carers of people with terminal illnesses, however of the treatment options available, CBT appears to be the most beneficial in this area.

1.5 Discussion

1.5.1 Summary of Main Findings

This study aimed to understand the evidence-base for psychological interventions for carers of terminally ill people, understand which psychological interventions have been found to be effective in reducing depressing and caregiver burden and compare effectiveness to determine the most effective intervention. This review found that most of the research examined CBT and psychoeducation. Overall, CBT was the most effective treatment for depression and caregiver burden with short and long-term effects. There was more variability in the findings for caregiver burden. CBT had the lowest attrition rates despite some studies including 12 month follow up which may suggest increased acceptability for participants and therefore suggest this may be an efficient approach for services to use given limited service resources.

Carers experience difficulties prioritising their needs, and express needs for opportunities to have personal time (Tatangelo et al, 2018). Given that most papers in this review reported carers spending 70-90 hours per week caring, carers may struggle to find the time to prioritise their needs. Individuals are more able to engage in self-care if they feel permission to prioritise their needs (Williams et al, 2022). Therefore, CBT interventions in this review may have been beneficial by allowing carers permission and space to prioritise their needs.

Cognitive reappraisal may be a key reason that CBT interventions were found to be most effective in the present review through increasing carers' resilience. A recent systematic review found that cognitive reappraisal moderated the relationship between stressors and negative outcomes and was a key factor in outcomes related to resilience (Riepenhaussen et al, 2022). The effectiveness of benefit-finding interventions in this review suggests that cognitive reappraisal is a key factor in the effectiveness of CBT interventions for burden and depression for carers of terminally ill people. This may have supported resilience in carers and allowed carers to become more connected to the positive aspects of caring, such as personal growth (Doris et al 2018). This research suggests shifting from reducing stress to optimising positive experiences. Research has also found that impacts of cognitive reappraisal are greater for women than men (Preston et al, 2022). As the sample from this review is predominantly women, this may explain why interventions focused on cognitive appraisal were particularly beneficial.

Previous research has suggested that information was key to support carers coping (Bressan et al, 2020). However, the limited effectiveness of psychoeducation alone in this review suggests this may not be enough. This may be due to having limited time to apply their learning due to the considerable time spent caring in the sample. This may explain why CBT was effective, as it provided structured space to implement strategies and participate in home practice (Senger et al, 2017).

Findings from existential behaviour therapy, mindfulness-based therapy, counselling, and psychoeducation were mixed, however there were also a lack of studies in these areas in this review. As such, it is difficult to explore in depth factors which may have contributed to the success or lack thereof. This may reflect some of the methodological issues discussed below.

Findings were less convincing for interventions effect on caregiver burden. There was variation in the findings and only 4 papers reduced caregiver burden. This may be associated with challenges or variations in defining and therefore measuring caregiver burden (van den Kieboom, 2020). In addition, De Stefano et al (2022) found that caregiver burden increased in the psychoeducation group, however this paper had some methodological issues. Despite this, there is still some promising evidence of effectiveness of CBT in reducing caregiver burden with gains which improve at follow-up, from high quality papers.

1.5.2 Methodological Considerations

Overall, the exclusion criteria in this review likely contributed to the quality of the papers in terms of study design, with 16 of the papers achieving ‘strong’ quality in study design as they utilised an RCT design.

Despite methodological strengths in some areas, the findings of the research were hindered by methodological issues of how attrition was reported and managed, selection bias and data collection methods. Many studies failed to explore or report the validity and reliability of the scales they used. There was a lack of transparency in the papers particularly in areas such as recruitment, drop-outs, and quality monitoring.

Carers supporting someone through terminal illness may be a difficult client group to access. The carers most in need may be less likely to participate, given that older age and lack of time were key obstacles for carers participating in research (Malm et al, 2021). Indeed, Holm et al (2015) reflected that carers only reported mild-moderate burden, therefore those most in need, or more likely to benefit from interventions may not have participated. This may be particularly relevant for papers which took place in palliative care services, as they are complex services with difficulties achieving inclusive research participation (Downey et al, 2023).

Although some papers controlled for a range of covariates, there was a lack of consideration on key areas such as hours per week caring. There is no information regarding range and many papers did not report hours caring. Hours caring have a mediating role in caregiver burden (Lin et al 2019), therefore, the lack of consideration to this factor may have explained the limited findings for improvement in caregiver burden scores.

The papers in this review covered a wide range of diagnoses, the most common of which was Alzheimer’s. It may therefore be difficult to apply and compare the findings in different groups as different disease profiles may lead to different carer needs (Bressan et al 2020). There was also variability in who delivered interventions, and overall, a lack of information regarding whether or how adherence to the model was monitored. In addition, some interventions were administered by consultant clinical psychologists which may not be reflective of the reality in services.

Several of the papers shared common authors. This suggests a lack of variety in researchers exploring this area using methods which would allow for inclusion to this review.

1.5.3 Limitations of this Review

There are several limitations to this review. Firstly, there were difficulties establishing a suitable search strategy. Although there is lots of research in the areas of carer support, terminal illnesses and psychological interventions, the pool of studies which include all three was relatively very small. Therefore, initial scoping searches yielded many papers which were vaguely related to these three broad concepts. Therefore, methods such as adjacency were needed to make the dataset manageable. This may have led to important papers being missed. This issue may have been mitigated using handsearching, however, of the papers which were identified from this, very few made it to the final review.

Only two papers provided information about overall inclusion rates, and as a result they received lower quality scores as inclusion rates were low. This may suggest weakness in the quality assessment tool, as papers which recruited participants through hospitals and are therefore reflective of the target population, but failing to discuss the inclusion rate may be rated ‘moderate’ in this area. This may be a problem more broadly within research as it encourages a lack of transparency which may mask selection bias.

This review applied exclusion criteria to increase the rigor of the papers which were selected, in line with previous findings around methodological issues within the research (Chi et al, 2016). This included limiting the studies to family carers of terminally ill patients. During screening, it was noted that many papers used ill-defined terms such as ‘informal’ carer or were not transparent about the relationships of the carers to the care recipients. In total, 7 papers were excluded as the relationships with the care recipient were poorly defined. Two papers also failed to clearly define the illness of the care recipient for example by labelling them with “other” or failing to state whether they had a progressive dementia. This issue of definitions may also be relevant to the research more broadly as many carers may not identify as carers and therefore may not be included in the literature (Staley et al, 2021). The lack of transparency in the papers meant it was difficult to fully assess the validity of the papers included in this review.

This review also had limitations with regards to the issues with homogeneity. For example, this review looked at family carers broadly, identified papers included many different relatives, and some papers failed to state if they lived together. As such, the extent of the carer burden across studies in this review is difficult to compare and evaluate change within. This hampered attempts to draw conclusions about which interventions are most beneficial.

The lack of homogeneity meant that it was not suitable for meta-analysis. Other options were explored, however there were not enough studies identified to allow for comparison between intervention type using a meta-regression. Therefore, comparisons between interventions are looked at using descriptive terms.

One of the goals of the review was to include third wave CBT treatments not included in Chi et al (2016) however few were identified. This may reflect the situation in the research currently, however it may reflect issues with the search strategy.

The papers in this review often failed to report the validity and reliability of measures. In addition, Hamilton depression rating scale which was used in 4 of the 7 studies looking at CBT interventions, only had acceptable validity. As such, the findings may not accurately represent the impact of intervention on depression, therefore findings using this scale should be interpreted with caution. The high attrition rates in this review may have also impacted the validity and reliability of the final sample and therefore, the validity of this review.

Finally, the impact of CBT interventions on depression may have been overestimated by relatively low baseline depression scores meaning small changes considerably impacted effect sizes.

1.5.4 Clinical Implications

Carers may benefit from CBT interventions such as behavioural activation, cognitive restructuring and benefit-finding through cognitive reappraisal if presenting with symptoms of depression or caregiver burden. Services should prioritise cognitive reappraisal as this review found it to be more effective than CBT interventions which did not include cognitive reappraisal. Existential behaviour therapy may be beneficial in the long-term for symptoms of depression, but more evidence is required. Services should offer psychoeducation to reduce depression and burden only in conjunction with other CBT approaches.

Although psychological support is central to models of support for carers of people with terminal illnesses (Oh & Kim, 2017), the use of evidence-based psychological interventions for carers of people with terminal illnesses may be less well understood (Hudson et al 2010). This review demonstrated that structured, evidenced-based interventions may be beneficial for both group and individual formats. This supports research which suggests that structured interventions can be beneficial and cost effective for carers of terminally ill people (Livingston et al, 2020). Accessing suitable support within end-of-life services may be even more difficult for those who care for patients with conditions which have an uncertain trajectory, or which involve complex decision making (Mayland et al, 2021). Therefore, these carers may benefit from additional consideration to ensure equity of care when offering evidence-based interventions. This review identified that face-to-face CBT support would be the most effective intervention, however as the telephone support in this review was either psychoeducation or counselling, more research may be beneficial to ascertain whether telephone CBT may be beneficial and meet the needs of carers who may struggle to access face-to-face support.

As this review highlighted a paper which found that burden scores increased significantly in the group with elevated trauma scores, screening for trauma symptoms may be beneficial prior to treatment to monitor burden scores longer-term and consider which intervention may be most appropriate.

Finally, the issues noted in accessing relevant research poses a significant challenge clinically. If few papers could be identified even with measures and specialist support, this may make it difficult for clinicians to work in an evidence-based way with carers of people with terminal illnesses.

1.5.5 Priorities for Future Research

Research has identified that mindfulness-based cognitive therapy can be as effective as CBT in general adult populations (Sverre, 2022). Future research should focus on exploring Existential Behavioural Therapy and Mindfulness Based Cognitive Therapy using methodologically sound approaches. This research should be transparent in reporting attrition rates, attempt to reduce bias in the sample and use scales with good validity and reliability. This may mitigate methodological issues found in this review and establish whether good effects can be identified. CBT research should also focus on factors which facilitate positive cognitive reappraisal for carers of terminally ill people. However, a recent systematic review identified that adverse events following meditation interventions were not uncommon even in adults with no history of mental illness (Farias et al, 2020), therefore this should be considered in future research with carers experiencing depression and burden.

Research should also focus on exploring psychoeducation by clearly defining such treatments and attempting to isolate which factors are beneficial to carers.

Overall, research should attempt to increase rigour by increasing transparency around inclusion rates and dropouts. It should also move towards using reliable and valid tools and ensuring the details of these are clearly and accurately reported.

Research should aim to build upon the promising findings noted in this review. This may be particularly important in the area of caregiver burden which has shown some good effects long-term; therefore, research should aim to replicate these findings and understand the effect in more detail.

Future research should aim to reduce the issues with heterogeneity noted by exploring or controlling for specific illnesses which may influence the needs of the carers. They should also control for key covariates such as number of hours caring, and measure severity of care needs to improve validity in the findings.

This review was biased towards female carers. It is unclear whether this reflects the patient population. Previous research has found that male carers ask for support only as a last resort which results in significant difficulties for them (Judd, 2015). Therefore, more research is needed to better understand the needs of male carers, and how to provide timely and effective interventions to support them.

In addition, very few studies reported the ethnicity of the participants. Therefore, future research should aim to explore the effectiveness of psychological therapies for carers from racially marginalised communities. This echoes findings from

Chi et al (2016) regarding the need for more diverse samples in this area of research.

Finally, the extent to which the use of CBT interventions fits within services for terminally ill people is unclear. One might anticipate an increased emphasis on acceptance-based third wave treatments like ACT which could be well suited for carers of people with terminal illnesses (Han et al 2021). No ACT studies were included in this review. This may reflect a theory-practice gap; therefore, clinicians should work towards practice-based evidence to ensure the evidence-base reflects the work best suited to services for carers of terminally ill people.

1.5.6 Conclusion

This review demonstrates that structured, evidence-based psychological therapies can be beneficial in reducing depression and caregiver burden in carers of terminally ill people. Future research should aim to increase the homogeneity in the research, control several covariates and improve transparency in the reporting of methods and dropouts.

Chapter 2 Understanding How Spousal Carers of People with Motor Neuron Disease Experience Palliative Care in the UK

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2.1 Abstract

Carers of people with MND can experience considerable challenges associated with caring. Previous research has identified palliative care support as key in supporting carers. However, there is limited understanding of the experiences and needs of UK carers of people with MND. This paper aimed to understand how spousal carers of people with MND experienced palliative care in the UK using interpretative phenomenological analysis (IPA). This identified 3 group experiential themes of battle, coping and feelings brought on by palliative care experience. These were split into subthemes and discussed in detail along with clinical implications and recommendations.

Keywords: Carers, Motor Neuron Disease, Palliative Care, Carer Support

2.2 Introduction

2.2.1 What is MND?

Motor Neuron Disease (MND) is a rare, terminal neurodegenerative condition with the average time between the onset of symptoms and death being between 2 and 4 years (Voltz et al, 2015). There are few disease modifying interventions to impact survival and prolong life (Harwood & McDermott, 2009).

2.2.2 Presenting Problems of MND

MND results in significant disability and care needs for individuals (Miller et al, 2012). MND causes cortical thinning and loss of motor neurons (Chang et al, 2005). This causes limb weakness, dysarthria, atrophy of the muscles and physical disability, difficulty swallowing and respiratory failure. Some also experience cognitive and behavioural changes associated with frontotemporal lobe degeneration including apathy and disinhibition (Cui et al, 2015).

2.2.3 Care needs of Individuals with MND

The progression of MND results in a broad range of ever-changing care needs (Cheng et al, 2018). Degeneration, muscle weakness and pain can lead to individuals using wheelchairs or other physical aids to engage with daily activities (Pavey et al 2015; Radunovic et al, 2017). Up to 75% of people with MND experience a decline in their speech so that intelligible speech is no longer possible therefore individuals with MND often require support with communication through support from those close to them to act as a “listener” and communicate their needs or use of technology such as voice banks (Ball et al 2004). Reduced mobility, dysarthria and loss of appetite can result in weight loss which is in turn a risk factor for death in people with MND, therefore they need support in terms of nutrition (Stavroulakis & McDermott, 2016).

People with MND have psychological and emotional needs associated with managing uncertainty about their future, feelings of being a burden and loss of hope (Veronese et al, 2015). Prevalence of depression and anxiety is varied in the literature (Carvalho et al, 2016; Grabler et al, 2015). Between 25-66% of people with MND experience depression (Wicks et al, 2007). This can lead to avoidant coping, which can impact engagement with services (Zarotti et al, 2019). Hopelessness and end-of-life concerns are common at different points in the disease progression (Averill et al 2007).

2.2.4 Impact on Carers

The needs of individuals with MND can place significant demands on carers (Ewing et al 2020). Carers are defined as “an adult who provides or intends to provide care for another” (Department of Health, 2014). Family carers face social, emotional, health and financial consequences associated with caregiving (Eifert et al, 2015). People who provide high levels of care are twice as likely to become permanently disabled and

72% of carers reported ill mental health because of caring (Carers UK, 2018).

Multiple studies regarding the needs of carers of individuals with multiple conditions (including dementia, cancer, schizophrenia, spinal cord injuries and MND) have highlighted limited training opportunities to support them to provide suitable care (Strommen et al, 2020; Given, Given & Sherwood, 2012; Outram et al, 2015; Jeyathevan et al, 2020; Bentley & O'Connor, 2016).

Caregiver burden is a negative response which occur because of the caregiving role (Lui et al 2020). Carers may experience loss of identity and role engulfment (Eifert et al 2015). There is a greater impact on carers if they felt they did not get enough help from services, if they were aged 60 or less, female or lost a spouse/partner (McNarma & Rosenwax, 2010).

Spousal carers face specific challenges with associated needs. They are particularly vulnerable to the negative impacts of caring (McNarma & Rosenwax, 2010) and experience increased difficulties managing a change in identity when transitioning from feeling part of a couple to feeling alone (Hennings & Froggatt, 2019).

Carers of people with MND also have specific support needs compared to other carers. They experience greater problems with mood and quality of life than other degenerative neurological conditions (McCabe et al, 2009) which increased as the disease progressed (Peters et al, 2012). This may be related to the speed of decline for individuals with MND and subsequent unrelenting uncertainty and pressure on carers to adapt emotionally and practically to the changing needs of their loved one (Flemming et al, 2020). The breadth and severity of impairments in people with MND can also result in increased burden for carers (Galvin et al, 2016). The relative rarity of MND may result in fewer opportunities for professionals to develop skills and knowledge in MND to support families, which may negatively impact carer experience (Bentley & O'Connor, 2016). This may also impact access to peer support, increasing the experience of isolation for carers (Rose et al, 2023). There is a need for more practical, informational, psychological, physical, emotional, and spiritual support for carers of people with MND (Oh & Kim, 2017).

2.2.5 Carers and Palliative Care

A 10-year literature review identified that increased access to palliative care services was needed to meet the specific needs of carers of people with MND (Aoun et al, 2013). Despite these recommendations, a recent systematic review identified that palliative care was rarely discussed (Flemming et al, 2018). Additionally, early access to palliative care is poorly implemented (Harris et al, 2018). People with MND have been found to receive palliative support at later stages of the disease, despite this support being beneficial earlier in the disease progression (Flemming et al 2020).

Guidelines have been developed in Australia to support carers within palliative care services. These recommend family caregiver support, assessment of need and planning, preparing for death, and bereavement support (Hudson et al, 2012). However, there is a tenuous link with these guidelines and the experience of carers, particularly regarding pre-bereavement assessment and support (Auon et al, 2017).

End-of-life support is vital for better subsequent adaptation to bereavement, regardless of the time that had passed since death (Akiyama, Numata & Mikami, 2010). Despite recommendations for tailored bereavement support based on carers' needs and risk (Aoun et al, 2012), this has not been found to fit the experience of carers. Aoun et al (2017) found that half of carers felt they had enough support from palliative care services and did not receive follow-up contact from services. Carers experienced processes as "blanket" approaches which were unhelpful (Aoun et al, 2017).

UK carers experienced grief and a "void" left post-bereavement when support was withdrawn (Harrop et al, 2016). Dynamic models of palliative care for patients with neurological conditions are recommended, which includes palliative support at all stages. Care should increase as the disease progresses and increase at trigger points in progression (Bužgová, Kozáková & Bar, 2020).

It is unclear whether services are meeting the needs of carers of people with MND in the UK as much of the research has been conducted in Australia (Aoun et al, 2017). Australia has less ‘death avoidance’ as a culture, relative to other western countries (Ray et al, 2014). UK service users, professionals and their families experience discomfort in discussing death and dying (Graham-Wisener et al, 2022). The individualistic culture in the UK may also result in additional pressure on carers and less support from the extended family (Wang, Abbot & Butcher, 2017). The UK has a specific health and benefit system, which impacts the experiences of carers (Petrie & Kirkup, 2018). The importance of the system in carer experience, and possible differences in the UK, mean that we may not fully understand the palliative care experience of carers of people with MND in the UK. This is particularly important when considering the specific needs of spousal caregivers.

2.2.6 Research Question

The present study aimed to fill the gap in current literature by understanding how spousal carers of people with MND experience palliative care in the UK. The lack of research meant the present study needed to be exploratory which informed the use of qualitative methods. It aimed to answer the question, how do spousal carers of people with MND experience palliative care in the UK?

2.3 Methods

2.3.1 Design

The current study aimed to explore the experiences of spouses who cared for a person with MND in the UK using interpretive phenomenological analysis (IPA) and is based upon a social constructionist framework (Larkin, Shaw & Flowers, 2019). An IPA approach allowed thorough exploration of how participants make sense of their experience of being a carer of someone with MND and receiving support from palliative care. This aimed to build understanding of the meaning which was made from these experiences. An important aspect of an IPA approach is to allow the researcher to explore these meanings in depth and reflect on their role within this meaning making process, whilst considering social and cultural factors related to the experience.

The primary researcher adopted a social constructionist perspective and had personal experience with MND. This was reflected upon throughout the research process using a reflective log. IPA principles were used to consider the impact of this on the double hermeneutic process.

2.3.2 Ethics

This study received ethical approval from the University of Southampton ethics committee (ERGO ID: 72086).

2.3.3 Recruitment

Participants were recruited through social media and online advertisements through the Motor Neuron Disease Association between September 2022 to March 2023.

2.3.4 Participants

Overall, 5 individuals were interviewed. Participants were all white British women with a range of involvement from palliative care (Appendix D). Inclusion criteria (Appendix E) stated that participants must have cared for a spouse with MND through to death in the past 6 years. To increase homogeneity of experience in line with IPA principles (Larkin et al, 2021), inclusion criteria stated that participants must have received palliative care input after 2016 as this was when guidelines for supporting people with MND and their carers were updated (NICE, 2016). Carers needed to be >6 months post-bereavement in line with DSM-V definitions of persistent complex bereavement disorder (American Psychiatric Association, 2013).

Participant must have had some experience of palliative care during this time however there was no exclusion criteria regarding the amount of support. This aimed to gain information on all experiences.

2.3.5 Exclusion Criteria

Anyone who was currently caring for their spouse or were unable to provide informed consent to participate were excluded from this research. Participants who lost their partner <6 months before the interview were excluded from the study. Full inclusion and exclusion criteria can be found in Appendix E.

2.3.6 Data Collection

Prospective participants viewed details of the research through the MND Association website and/or social media. They contacted the researcher, were given the information sheet and screened for suitability.

In line with recommendations, participants were given agency over where and how interviews took place (Eatough & Smith, 2017). For example, online using Microsoft teams or in person. All participants chose to be interviewed online. They were sent consent forms which they returned prior to the interview. Participants were sent a confirmation email which included a recommendation to bring someone with them for emotional support following the interview. Participants were sent a link to this meeting and a reminder email one week prior to, and one day the interview.

Participants were able to ask questions and gave verbal consent before the interview. All interviews were recorded using a Dictaphone. Participants were informed that they were in control of what they chose to share. All participants planned how they would communicate if the interview became too distressing or they did not want to talk about something.

Interviews lasted 60-73 minutes (*M=64.6)*. All participants were asked “tell me what palliative care means to you?” Follow up questions were dependant on their responses. However, an interview schedule (Appendix F) was used as a structure and designed under supervision of a Consultant Clinical Psychologist in Palliative Care. This was used as a prompt when needed in line with IPA recommendations (Eatough & Smith, 2017).

Participants were offered a grounding exercise and signposting to support services to manage any distress following the interview as part of the debrief.

2.3.7 Analysis

Interviews were transcribed using an independent transcribing service to increase the reliability of the transcripts. Transcripts were repeatedly read and replayed. Descriptive, linguistic and conceptual notes were made which were used to identify experiential statements following the IPA approach as detailed by Larkin et al (2021). These were then clustered into Personal Experiential Themes (PETs) for each participant. Analysis for each interview was completed before the next interview was conducted to guide future interviews and ascertain the quality of each interview as recommended by Larkin et al (2021). In line with the iterative process of IPA, individual transcripts were revisited, and themes re-clustered during analysis. When all interviews were analysed, patterns were identified across all interviews to develop Group Experiential Themes (GETs) which formed the final analysis. A reflective log was maintained from study design, throughout the research to ensure transparency and reflexivity (Appendix G). Reflections can be found in Appendix H. Themes were discussed with research supervisors throughout development to support the Hermeneutic process of meaning making, interpretation and reflection (Larkins et al, 2021).

2.4 Results

The analysis resulted in three Group Experiential Themes (GETs); battle, coping and feelings brought on by palliative care experience (see Table 1). Each GET included subthemes. See Appendix H for master table.

Table 1: Group Experiential Themes and Subthemes developed from the final analysis

Group Experiential Themes (GETs)	Subthemes
Battle	
	Searching for allies
	Conflict
	Identity
Coping	
	Searching for validation
	Seeking safety
Feelings brought on by receiving palliative care	
	Loss
	Loneliness
	Vulnerability

2.4.1 Battle

This GET discusses themes of struggle which were experienced within and between the individuals and services. Three subthemes were identified under this GET which are expanded upon below. Participants experienced battle for their identity, experienced conflict and therefore sought allies to support them in their battle.

2.4.1.1 Identity

Participants battled to hold onto the identity of themselves and their partner. This battle was an internal struggle with reorganisation of their sense of self, and external, whereby participants experienced services as a threat to their identity that must be fought.

Most participants experienced a distressing shift in role. They fought to protect the identity of their partner, experiencing feelings of disrespect and anger if this was challenged.

> “No, no, this is a man that a few weeks ago was writing reports for an expert witness to the courts.” (Lisa, 29, 904).

As such, their engagement with services was centred around holding onto identity. For some this meant avoiding services, which allowed them to resist transitioning to carer and patient roles. Others fought to feel seen by services for who they were.

> "We probably both resisted that uh because it very much marks the end of being a couple." (Debbie, 29, 815)

Part of this battle for identity seemed to support their role as a carer and part seemed to survive the emotional impact of palliative care.

Along with resisting role transition, carers fought support from services to hold onto their new identity.

> "And that was my role, um which I wanted to-to perform." (Lizzie, 10, 270)

Only Rachel received support from hospice care, and she did not experience the same battle for identity. Palliative care services were allies in holding her identity and took on practical tasks which removed this battle therefore mitigating its impact.

> "Um so I was just sort of relinquishing my role for a while, and I was able just to-to relax a little." (Rachel, 23, 650).

Rachel experienced her identity being seen by services and toyed with a perspective of being a patient, however this shift was not painful or frightening to her. This may be because she was seen and understood by the team and given permission to be herself. A reduction in her need to battle identity was associated with a reduction in the need to search for allies.

Only Cath did not experience any change in her sense of identity. She fiercely held onto this and did not subscribe to the concept that she was his carer not his wife. Her fight for identity was centred around wanting professionals to see her identity.

> "No, I'm not his carer, I'm his wife, I'm always his wife, I'm not his carer, I just happen to look after him." (Cath, 11, 327).

Carers experienced oscillating battle between identity and accepting care. This explains some of the struggle faced in carers experiences of seeking allies.

2.4.1.2 Conflict

Participants experienced conflict with professionals their husband and internal conflict through palliative care.

> “He would never ever talk about dying or death. Whereas I’m quite open about all of that stuff.” (Lisa, 35, 1073).

Lisa’s coping was blocked by her husband’s avoidant coping, leading to a conflict in understanding her needs while prioritising his.

All individuals spoke of duty to prioritise their husband’s needs resulting in internal conflict. The internal conflict came from different coping strategies and values in their role as a wife.

> "From a purely selfish point of view, it would have been good to have somebody stay with him overnight so that you could get a full night’s sleep." (Debbie, 27, 771).

Therefore, having basic physical needs was experienced as selfish and divergent from her values as a wife.

One individual spoke of conflict in behaviour whilst trying to balance prioritising her and her husband’s needs together.

> “So, I knew support was there, but I wasn’t able to access it.” (Lizzie, 2, 39)

This sheds light on some of the processes around safety seeking from services though permission seeking.

Prioritising their husbands needs put their own physical health in decline, which mirrored the physical decline of their husband. They experienced extreme physical consequences particularly around a loss of sleep which impacted their functioning.

> “I’d lost a lot of weight, the weight just mel-melted away, uh day by day, week by week.” (Lizzie, 4, 96).

The physical demand prevented them from providing the care they wanted to, resulting in internal conflict of values and subsequent guilt.

> “That's probably my only regret, really, that I just couldn’t cope with him, (Laughter) through lack of sleep.” (Cath, 34, 1036).

Some experiences were impacted by the Covid-19 pandemic whereby they were unable to have the support they needed.

> "My husband uh had to was offered a hospice bed a week later, because I had dislocated my shoulder." (Rachel, 18, 508)

Rachel fought to keep her husband at home, however an injury from caring resulted in hospice involvement. Although this conflicted with her values, it resulted in her receiving excellent care showing that avoidance of conflict may be more problematic than the conflict itself.

Carers were conflicted in holding onto their independence versus getting help from services.

> “I felt very isolated. Because I'd said at the outset, “I want to do things myself,” and right up to the end I did want to do things myself and I did do that.” (Cath, 4, 98).

This shows that Cath took pride in independence, however, she then felt very isolated, and struggled to balance this conflict.

Participants highlighted conflict within the intentions and actions of services. Services provided too much of the wrong care, made the situation more difficult and tried to help but this was blocked by poor understanding.

> “Everybody was really trying to help, I'm not saying they weren’t, but it just well it just- it just got too much.” (Cath, 8, 214)

Cath particularly highlighted conflict with professionals. Support services made things more difficult by contributing to a feeling of isolation, and violation by invading their home, and blocked the coping strategies of carers and their husbands. For example, the use of avoidance, humour and connecting as a couple.

> “We needed time to get our heads around it as well, without it becoming medicalised and everybody coming in and talking about it all the time.” (Cath, 10, 281)

The absence of conflict in Rachel’s journey shows the impact of understanding care which provided safety. She was the only participant whose husband wanted to talk about death. Perhaps this absence of conflict is associated with acceptance, so she could communicate her needs more easily and services could hear this and respond in a way which fitted her needs.

The more conflict experienced, the more individuals needed to search for allies, which resulted in further conflict.

2.4.1.3 Searching for Allies

Participants experienced a process of searching for allies. This means finding ways to identify people within services to support them in their battle.

Carers fought for allies at early stages of diagnosis.

> “In the first few days, I-I went online to see who these specialist nurses were because I knew we were going to need some support.” (Cath, 17, 492).

This demonstrates they could not fight this battle alone and searching for allies supported coping. There was a shared experience around searching for allies who understood their needs.

> "Everything was a bit of a battle for when any agencies were involved." (Lisa, page 17, line 507).

This showed that when services did not understand; involvement from services contributed to the experience of a battle rather than alleviating it through allyship. Some sought allies who were more skilled to provide a sense of safety, Cath sought a shift in power in searching for members of a team which she was leading.

> "Because I'd always worked as part of a team, it was important to me to have somebody that I could work alongside." (Cath, 18, 517).

This shows the variation in the form of allyship, but shared experience of the function.

Carers sought allies to fight processes which did not fit their needs. When they were unable to find this, they sought alternatives. Some held onto their independence when managing the difficulties in finding allies, others sought allyship from non-professionals.

> "And thankfully my two kids, my daughter moved back in with me to help me look after her dad." (Lisa, page 3, line 77).

This shows that carers first looked to services for allyship due to the expected expertise. When this was not met, it was vital to fill the need for allyship in any other way, regardless of expertise or skill. Therefore, battle for allyship seemed centred around understanding and community. They also fought for different allies to meet different needs.

> "Um friends helped. I had- I had one good friend. Well, lots of good friends, but uh one friend in particular that I was able to talk to" (Debbie, 19, 538).

This shows that the extent to which they experienced an individual as an ally depended on their relationship and what aspect of caring they needed support with.

At times Lizzie gave up on her search for allies which was distressing and demonstrates the importance of this battle.

> "It became apparent, I think, that we just need to, almost, probably try and cope on our own" (Lizzie, 14, 406).

Rachel experienced understanding and trust from services which supported a sense of safety which meant that she did not have to search for allies in the same way as others.

> "His thoughts and feelings and my thoughts and feelings were-were taken into consideration" (Rachel, 11, 312).

This resulted in even power distribution between her and services. She was given choices and space to be heard which developed a sense that they were on the same side of the battle. This shows why battling was so important for the other participants.

Professionals were deemed as allies or not which meant carers experienced services in extreme terms. This led to further uncertainty, therefore a lack of safety which reinforced a pattern of searching for allies.

2.4.2 Coping

This GET described how participants receiving palliative care sought to manage and survive this experience and identified themes of searching for validation and seeking safety. This included coping with the impact of MND through services and coping with the impact of the services themselves. This was achieved though ‘searching for validation’ and ‘seeking safety.’

2.4.2.1 Searching for Validation

Carers sought to cope by searching for validation from services. The degree to which they experienced this varied. A key factor in this experience was their understanding of MND and whether services demonstrated commitment in understanding them as a family, as individuals and adapted their approach.

> “They have no idea how you’re feeling, what you’re going through.” (Lisa, 19, 586)

Approaches from services aimed at helping carers made things worse. This was routed in the absence of validation which led to experiencing care as violating for example when services came into their home when they did not want them there, attempted to make decisions about personal aspects of their life and home and violated their sense of safety within their home through failing to provide choice. Palliative care services were experienced as overly task-focused which was experienced as invalidating.

> “It is a checklist, whatever way you look at it, it's a checklist.” (Cath, 27, 802)

As a result, some carers had to seek validation from family or people with shared experiences.

> “All the other people were saying, “You’re doing a fantastic job”, “You’re doing really, really well. Keeping going”, blah, blah, blah.” (Lisa, 13, 395).

Therefore, receiving validation from non-professionals did not always provide what they needed and there was something different in this coming from professionals.

Validation was a key feature of the people that carers identified as allies. When validation was received, carers named individuals to celebrate the impact they had on their experience.

> “She can look at me and know how I’m feeling you know. I’ve not slept more than an hour for three days now (laugh) you know and she knows she knows how I’m feeling whereas some of the others they don’t know.” (Lisa, 37, 1152)

The language used included war-type rhetoric, suggesting that seeking validation was so important because the realities of their experiences could not be understood by anyone who had not seen it.

> “But nobody understood what we were facing, day in and day out. Why would they? But an-but the association visitor did because she had lived-lived through it.” (Lizzie, 3, 63)

> “With-with people, who-who have seen you at your worst and seen the worst possible things.” (Debbie, 655, 23).

Searching for validation supported carers to cope with strong feelings of shame and the uncertainty they experienced, regarding the future, the skill of professionals, and their abilities as a carer.

Professionals involved in their palliative care often had no or little experience or expertise in MND, which may have motivated task focus within MND palliative care services, providing less space to provide validation.

> "Probably, 75% of them were caring and compassionate about what they did, um but nevertheless, still had-didn’t have that understanding of MND." (Debbie, 7, 188)

Only Rachel did not have to search for validation. Validation was provided in a holistic, consistent, and timely manner.

> “And so we had another we a listening ear to the palliative care team to actually express some of our, um or my husband’s, sort of wishes.” (Rachel, 13, 354).

Rachel experienced this as a sense of power in powerless situations resulting in unity and reassurance for her. The significance of the impact of validation demonstrated why this means of coping was utilised for carers.

Through this search for validation, some individuals experienced extremes in care. The stark difference from when they were validated and when they were not seemed to underpin this. They used this to forge the support which was right for them and find allies who offered this validation. This helped to manage the emotion impact of the journey.

2.4.2.2 Seeking Safety

Carers also coped with palliative care by seeking safety. Services provided this through expertise, understanding, relationships and emotional containment.

Carers sought safety by avoiding the reality of the horrors they were experiencing.

> “Every month, there was another bit of equipment in the house, you know another demonstration of how- “(Rachel, 6, 166).

Although Rachel accepted the equipment, she used cognitive avoidance to manage the unwanted reminders of his decline.

Avoidance was present linguistically, through humour when discussing distressing events.

> "But then the whole thing was (laugh) you know you didn’t want to be there anyway. " (Debbie, 10, 270).

> “He liked her, but he hated her coming because he always felt like she was the Grim Reaper. (Laughter)” (Cath, 16, 481).

This shows they held onto moments of humour with their partner and palliative staff to cope.

> “She hoisted Paul up and he fell on the floor (laugh) um- not in a bad way you know - but we just laughed, and we were just all in a heap on the floor you know having a laugh. That was just what we needed, somebody that was real and got it.” (Lisa, 19,556)

This allowed avoidance of the reality of the situation and provided safety through human connection.

Safety came through avoiding palliative care services cognitively or behaviourally. Some escaped the idea of needing services and avoided contact until they could not cope, which provided safety through control over the inevitability of the disease.

> “Because by having it, I was almost forced to accept what was happening (tearful).” (Lizzie, 23, 668).

Avoidance reinforced a need to seek safety by inadvertently contributing to experiences of isolation. Carers feared the impact of their emotions on themselves and others, resulting emotional suppression and avoidance.

> “Then shut the front door and the mask comes off, and the flood gates open.” (Lizzie, 18, 504)

This may have driven task focus for some of the carers. Avoidance helped carers to manage the feelings of hopelessness and helplessness and allowed them to move from the deficit-focus which permeated their experience of palliative services.

> “There was- it just, basically, said, “Well, you can’t do this and you can’t do that.” Um s- and i- if you felt that there was something that you could be doing to improve things, then it wouldn’t- wouldn’t have been so bad (Debbie, 14, 372).

Therefore, a deficit-focus interfered with carers attempts to cope by holding onto their life.

Carers sought safety through approach-based coping, task focus and searching for points of control.

> "Um partly, personality. Um I’ve always been quite an outcome-focused, positive person, so solution based." (Debbie, 9, 515).

Services mirrored this task focus suggesting a parallel coping process. This was particularly seen for participants who received limited or unspecialised palliative support.

> “You won’t be able to do…you can’t have that” “You’ve got to have a wet room built.” I was like, “Aargh” and oh, it was just awful.” (Lisa, 18, 533)

Rachel however experienced services meeting her needs, so she did not have to rely on so many of her own internal or external coping strategies relative to the other carers.

> “It was like a blanket of love, being in there.” (Rachel, 22, 618)

> “Out of a very, very bad, bad thing, was a very gentle and kind and comforting experience.” (Rachel, 11, 294)

The love Rachel experiences supported relational safety. This removed responsibility from her, allowing her to be present and process her journey.

Safety through relationship building was also key for others. When relationships were established through reliability and validation from services, some of the emotional impacts of the palliative care journey reduced. Genuine human connection, consistency, understanding, validation and love, fostered relationship building.

> “A real person, somebody that just you know they didn’t expect you to go flying round with the hoover before

> you visited and (laugh) you know just normal." (Lisa, 21, 703)

For Lisa, safety and closeness came from not feeling judged.

> "They were interested in us as a couple, as people, I suppose, which is probably something we didn't really get particularly from the carers." (Debbie, 11, 306)

Like Lisa, Debbie highlighted authenticity which fostered relational safety. This connection helped her to be seen as a person and them as a couple.

Relational safety supported carers to cope with the isolation which occurred when experiencing palliative care. When individuals did not feel a human connection or felt judged by professionals, repairing relationships was difficult and they withdrew and relied on more avoidant coping strategies to re-establish safety.

> "I went, "Fair enough." Um so, we sort of bowed out in that respect." (Cath, 27, 815)

Some escaped services to forge their own path where they could both feel confident, and therefore, achieve a sense of safety.

For Lizzie, seeking relational safety supported post-traumatic growth.

> "They taught me that you know you don't have to put the mask on, you know, don't-don't protect other people

> from the emotions that you're feeling. Um and over time, I-I guess, I've learnt to do that." (Lizzie, 18, 507).

Lizzie's relationships with professionals provided safety for her to show her emotions. This changed her relationships with her emotions demonstrating the profound impact relational safety had on her.

To cope with the impact of their palliative care journey carers sought validation and safety. What constituted feelings of safety was different for carers, however, this was impacted by responses from palliative care services, and external factors such as their husband's coping.

2.4.3 Feelings Brought on by Receiving Palliative Care

This GET explores the feelings brought on for carers through their palliative care experiences. This included feelings of 'vulnerability', 'loneliness' and 'loss.'

2.4.3.1 Vulnerability

Carers experienced vulnerability related to the fear and uncertainty from the disease progression, and the care they might receive.

> "And and who you know where do you define 'last, last days'? You know, you just don't know what's coming, so um you don't know what the last days are." (Debbie, 2,29).

Debbie experienced uncertainty of when dying started. The presence of services signified her husband's imminent death causing fear, which led to emotional vulnerability.

Carers experienced vulnerability from overwhelming responsibility, which impacted their engagement with services.

> "Even though I'd been an intensive care nurse, frightened the life out of me, dealing with that at home." (Cath, 16, 468).

Cath felt responsibility for holding her husband's life in her hands. Her fear did not come from a lack of skill due to her nursing background, but from a vulnerability in holding this alone.

The responsibility they held resulted in perceived inadequacy in their skill, which left them vulnerable to feeling criticised by services. For some, the process of accepting services into their homes left them feeling exposed and vulnerable.

> "She made me feel absolute rubbish and um she got she said, "Well, the bed guard needs checking, I'm going to get the Red Cross round" and all of this. She said, "They're coming out tomorrow."" (Lisa, 13, 379)

Lisa felt that her home was being violated, and this intrusion served purely to inspect her skills as a carer.

> “Some of them they come in, they are so stuck up. And like I said, they looked down their nose at you, but they have no idea.” (Lisa, 19, 583).

Lisa worried about not having the skills she needed to care for him. When palliative care services came to her home, these fears were exposed leaving her vulnerable and invalidated.

For others, when services held the responsibility, their vulnerability reduced.

> "They sort of took that out of our hands and said, “You don’t need to worry about that. You know you will not be attending that interview. And it you know it will all be sorted out." (Debbie, 14, 395)

For Debbie, services anticipated the vulnerability she might feel and took that from her by holding responsibility, therefore reducing her feelings of vulnerability.

When services placed expectations and pressure on carers, this seemed to increase feelings of vulnerability.

> “You get given this ventilator, and they go, “Off you go,” you're out of here.” (Cath, 31, 916).

Cath felt overwhelmed and frightened by the responsibility placed on her. The nature in which staff offered support was experienced as flippant. It is worded as an instruction showing that she felt she had no choice in being left to manage this, which contributed to her feelings of vulnerability.

As Lizzie’s feelings of vulnerability reduced with time, her perspective on palliative care changed. She moved to a place of gratitude which she could not connect with at the time due to the intensity of grief and her vulnerability.

> “My approach to using them, perhaps, would be very different now.” (Lizzie, 23, 652).

Therefore, the role of palliative services in supporting carers through this period of vulnerability may not be seen until later.

2.4.3.2 Loneliness

Carers experienced loneliness during palliative care. For some, services contributed to this loneliness through a lack of understanding and validation.

> “That was awful because it just reinforced that I didn't really have anyone.” (Cath, 17, 490).

Cath perceived a lack of understanding of MND as a sign that she could not trust them to help her as an ally, leading to loneliness.

For others, knowing services were contactable on their terms, helped to reduce this feeling despite no change to services’ physical presence.

> “She was always at the end of the phone. She made it very clear she was there.” (Lisa, 22, 660)

Lisa was afforded connection to trusted members of the team. This was emphasised by this individual, which allowed her to feel less alone. Therefore, when carers trusted services to be there when and how they needed them, their loneliness reduced.

Some carers experienced loneliness with many professionals present, therefore the quality of the relationship determined the experience of loneliness.

> “We weren't getting anywhere. There were so many people in that room, and nobody was speaking.” (Cath, 28, 824).

Cath was comforted by the number of professionals in the room however she experienced their silence as a statement that she could not trust them, and she was alone.

Even in the presence of close relationships, carers still experienced loneliness.

> “Despite having people around me and-and lots of good and loving people around me, still feeling um (tearful) very isolated and alone. That’s through no-no-no fault of-of anybody, at all you know. Marie Curie wer-were-were lovely. Um It was just my (tearful) feeling, my-my mental state.” (Lizzie, 7, 177).

Therefore, for some, loneliness was a grief response which could not be resolved by actions of services.

There was a difference between being alone and feeling lonely.

> “In some ways, almost, would feel (pause) relief when they’d gone. that we were just on our own again,” (Lizzie, 11, 301).

Carers craved time alone and experienced relief when they regained this. Therefore, loneliness may be associated with absence of control and choice.

Some carers identified COVID-19 as a contributing factor to loneliness, however, their acceptance of this seemed to lead to less distress associated with loneliness.

> “I know they would have been there if I needed them, that was the thing, but they couldn’t.” (Cath, 34, 1034)

Although this was distressing for Cath, knowing they would have been there helped manage some of her retrospective experiences of loneliness.

Avoidant coping and holding onto independence were used to manage loneliness, but this may have inadvertently reinforced it.

> “I just had to pick up the phone to a-to a friend (tearful) and-and they would be there, they-they would rally round but (tearful). In many ways, I couldn’t. Um (pause) and so, the loneliness, probably, was exacerba-exacerbated by that.” (Lizzie, 8, 209).

Therefore, Lizzie felt unable to contact people for support. She then avoided contact with others, to avoid becoming overwhelmed by emotion. This quote shows that she linked this to increasing her loneliness.

Carers craved connection to escape loneliness and vulnerability which came with it.

> "Um you felt abandoned. (Tearful) You had a lot of emotional stuff to deal with and nobody, really, to share it with. I mean, there was- there would have been counselling available and stuff like that, but that's not (pause)- that's not really what I felt the lack of (tearful)." (Debbie, 23, 646)

Debbie initially avoided services, then sought connection which managed her loneliness. When her husband died, she was left with loneliness which was magnified by the loss of the close relationships she developed.

Only Rachel did not discuss experiences of loneliness. The safety she felt from the outset from services may have prevented loneliness. Approaching her fears may have also reduced reliance on strategies which reinforced loneliness in the long-term.

2.4.3.3 Loss

Carers experienced unrelenting loss through palliative care. This included loss of their husband, self, and safety in their home.

> “And it just made me feel that my home was not my own.” (Cath, 8, 273)

Debbie experienced loss of her husband’s identity and dignity.

> "It takes people who are so fit and healthy and, within such a short space of time, just strips every shred of dignity and independence from them." (Debbie, 30, 850).

This shows the loss of his independence and speed of decline made the experience of loss more difficult, a sentiment shared by others.

For some, palliative care helped to manage these feelings of loss, whereas for others, palliative care contributed to it through lack of understanding and individualised care which contributed to increased loss of identity.

> “So you never got any time together, no quality time together.” (Cath, 8, 225)

Palliative care led to a loss of Cath’s time and connection with her husband which was so precious to her.

Carers experienced a loss of hope in palliative care. They lost their dreams and hopes for the future. They then sought hope, solutions or reassurance, which services did not or perhaps could not provide. This caused more loss of hope.

> “You’re on the internet again trying to find out what you can about that and none of it none of it’s any good,

> none of it’s reassuring because there is, there is nothing. There’s no drugs, you know.” (Lisa, 27, 830)

This showed Lisa’s process of resisting loss of hope. She sought information and reassurance and asked for medication which did not exist.

Carers also faced loss for the care they hoped they would receive. This was more prominent for carers who felt that the palliative care staff had also been failed.

> “So, I was disappointed for her and then not giving her the education to actually do the job.” (Cath, 20, 610)

Cath felt alignment to the professionals. She felt they could have helped her if they had been given the appropriate support to do their jobs, which exacerbated her sense of loss.

Some carers had experience with other family members with terminal illnesses. This shaped their expectations or retrospective understanding of care. When they did not receive the support they had hoped for, they experienced feelings of loss.

> "Carers came in and the carers were really good, um and they came to the funeral." (Debbie, 23, 629)

Debbie’s previous experience meant she expected bereavement support. She ignored her needs with a plan to return to them when her husband died. When care stopped suddenly when her husband died, she experienced a double grief. She grieved due to her expectations of care, but also the relationships she had built and did not expect to lose.

Positive relationships with palliative care services helped carers to manage their feelings of loss and contributed to it.

> "Um but yeah that was the other biggest void it was just the complete cessation of everything." (Debbie, 23, 640)

Debbie compared the loss of relationships with staff to the loss of her husband. This demonstrates the impossible position carers were in, whereby the things which helped them also hurt them.

Rachel had a different experience and did not speak of loss of relationships with palliative care teams.

> “Um so it was- it was a special, quite an uplifting, experience actually.” Rachel, 10, 271)

This shows she thought of professionals with gratitude and adoration and spoke in the past tense. Perhaps the supportive care she received meant she was able to process this loss during her palliative care journey.

Lizzie was the only person who received follow up after her husband died. Like Rachel, she did not experience loss of these relationships, due to the slow transition through bereavement.

> "They talked to me a lot about stages of grief and um, perhaps, to accept that it's okay to not be okay, sometimes, um and not have to put, necessarily, the mask on." (Lizzie, 18, 501)

Professionals supported her through loss, and she took comfort in their knowledge of grief processes which helped her to manage the loss of her husband and services.

The feelings brought on by experiencing palliative care were characterised by vulnerability, loneliness, and loss. These reinforced each other, with loss being associated with loneliness and vulnerability, and vulnerability and loneliness exacerbating feelings of loss.

2.5 Discussion

2.5.1 Summary of Key Findings

This research aimed to understand how spousal carers of people with MND experienced palliative care in the UK.

This analysis identified GETs of battle, coping and feelings brought on by receiving palliative care (Figure 1). Spouses seemed to experience palliative care as a battle which appeared to elicit strong emotions which may have reinforced the need to battle. They may have sought ways to cope with these feelings through seeking validation and safety from services. At time, these strategies may have led to further feelings of vulnerability, loss and isolation for example, through carers avoiding systems of support. At times, palliative care seemed to contribute to battle, and difficult emotional experiences, and at times it alleviated these feelings.

Spouses may have searched for allies to seek safety and validation to cope with vulnerability, loneliness and loss. This possibly caused conflict which may have reinforced the search for allies. Internal conflict in values may have impacted their ability to hold onto identity. Seeking safety may have led to further challenges with identity and conflict by challenging how they adapted to their change in role. Battle for identity may be associated with feelings of loss. This loss may have reenforced their experience of battle motivating them to seek safety, which may have led to further loss. This may have contributed to loneliness and vulnerability which drove carers to seek validation to reduce the emotional impact of isolation. These interpretations were made as there are several psychological factors identified which are assumed to hold linear and causal relationships.

Figure 1: Model of Understanding How Carers of People with MND Experience Palliative Care in the UK.

2.5.2 Review of Findings and Clinical Implications

This study appears to support Australian findings that family did not experience a tailored approach (Anoun et al, 2017) which impacted their experience of a battle. It may also support findings from Harrop et al (2016) that individuals grieved the loss of services post-bereavement and increased continuity of care was needed.

Participants may have wanted understanding and allyship to reduce loss, vulnerability and loneliness. Current literature recommends advanced care planning in palliative care. This involves services identifying values and preferences, and enabling communication with the individual, family and system which aims to increase collaboration (McMahan, Tellez & Sudore, 2021). This communication and collaboration may reduce feelings of vulnerability and loneliness. This is feasible for use with family members of people with MND however is underutilised for people with neurological conditions (Holland-Seebar et al, 2019). The findings of the present study may suggest an absence of advanced care planning including assessment of needs and bereavement support which might have mitigated the feelings of a battle and emotional difficulties for spouses. This may support findings that this approach is underutilised specifically in MND. This also supports Hudson et al's (2012) findings which recommended tailored, planned support in guidelines for supporting family members in palliative care services in Australia. Therefore, these guidelines may have links with the needs for spouses of people with MND in the UK.

The participant who received follow-up care benefited from psychoeducation in grief processes which supported her once her after care withdrew and reduced vulnerability to isolation and loss. This appears to support research that grief psychoeducation benefits family of terminally ill people by normalising grief processes (Eyre, 2019).

This study's findings supported previous research that individuals experience loss associated with identity (Eifert et al, 2015). However, one participant had a different experience to this. Although she experienced dissatisfaction with support from services which is a key risk factor for loss of identity (Eifert et al, 2015) she was able to hold their identity leading to feelings of pride. This may have been impacted by her identity as a nurse, possibly reflecting a gap in the literature around factors which protect identity. Therefore, having a 'deficit focus' may overshadow benefits of caring, supporting research that interventions may consider from reducing difficulties to optimise positive experiences (Doris et al, 2018).

This study supports previous research on difficulties managing the experience of loneliness. The experience of loneliness may have been so prominent as all participants were spouses, and as such were more vulnerable to difficulties in loss of identity, role engulfment and increased responsibility to provide care.

This research therefore supports the findings of Hennings and Froggatt (2019).

Participants may have experienced vulnerability associated with feeling underprepared and skilled to care for their husbands. They felt abandoned with equipment they were frightened to use with no formal training. This supported previous research on the lack of training for family carers of people with MND in Australia (Bentley & O'Connor, 2016). This study may also support Bentley and O'Connor's (2016) findings of difficulties accessing appropriate support. Participants in this study may have searched for allies to attempt to resolve this. Bentley and O'Connor (2016) found that family members struggled to access information and that services did not talk about death. However, in the present study, limited talk of death seemed to be associated with avoidant coping. When participants wanted to talk about death but couldn't because of possible avoidant coping from the care recipient, this seemed to be accepted by the participants in line with their values. This may reflect possible differences in openness to discussing death between the UK and therefore may suggest different needs for UK carers whereby they are more in need of a sensitive approach which offers choice in discussions around death and adapts to their coping styles as opposed to services assuming spouses' need or want to discuss death regularly.

Participants may have searched for validation to cope but were often met with a lack of understanding from services which may have fuelled excessive ‘task focus’. This may have then impeded services providing the validation individuals needed. This was linked to a lack of specialist MND knowledge in services. MND specific training was recommended in the review by Aoun et al (2013), and the present study found this was still an issue. This supports Bentley & O’Connor’s (2016) findings that family experience can be negatively impacted by limited MND knowledge in services. However, the present study also suggests parallel processes. Participants seemed to exhibit approach-based or avoidant coping before involvement with services. Professionals may mirror the coping styles of spouses which may underpin task focus and invalidation from professionals. This may then reinforce these coping styles, as opposed to spouses’ task focus being directly due to them mirroring professionals. Previous research has identified avoidance in palliative care staff to cope with the emotional impact of the work, particularly around displays of strong emotion (Brighton et al, 2019). Therefore, the more distressed participants were with greater need of validation, the more difficult this may have been for professionals which resulted in avoidance. When this need went unmet, family’s experience of a battle and loss for the care they expected seemed to increase. Gerhart et al (2016) found that mindfulness-based communication training was effective in supporting palliative care staff with avoidant coping strategies to manage the emotional impact of the work. Therefore, such training opportunities and support for staff may be beneficial in meeting the needs of families and staff.

Family members did not feel they had enough support across many areas possibly contributing to experience of battle and isolation. This supported the findings of Oh & Kim (2017). However, this was not the case for one family member who received hospice care. Therefore, availability of specialist services who may mitigate the experience of fighting a battle and isolation may meet their needs. However, some experienced support as violating. This may explain Oh & Kim’s (2017) findings in the challenges meeting the needs of family members. This highlights the importance of choice in their care.

Most participants were referred to palliative care at diagnosis. Some received limited palliative care support however, this was often their choice to prioritise the desires of their partners. When participants did want more support at points of change, the help was often not available which increased difficult emotions. This may explain the poor implementation of early access to palliative care noted in Harris et al (2018). Families may choose not to access palliative care at early stages, or services may prioritise offering it at early stages, and not revisit this in line with changing needs. Therefore, access to palliative care at points of change in MND may be important (Flemming et al, 2020).

2.5.3 Clinical Recommendations

Palliative care services may wish to consider implementing communication training for staff, particularly in the absence of support from specialist services such as hospices. This should focus on validation which may reduce experiences of vulnerability and isolation. Staff should be supported to understand and manage the emotional impact of their roles and consider how this may impact the experiences and engagement of family members. This may include the use of parallel formulations where appropriate (Lewis-Morton et al, 2015). Services may consider mindfulness-based communication training given its promising outcomes (Gerhart et al, 2016).

Services may wish to consider MND specific psychoeducation for staff and family. This may build trusting relationships which provide a basis for safety, reducing the experience of isolation, vulnerability and a battle. Services may also consider offering psychoeducation regarding grief processes to family members, ensuring this is adapted to their individual needs and coping.

Services may wish to reflect upon the language they use to describe family members. Some may experience the label of ‘carer’ as invalidating to their identity, which may increase their sense of battling to hold onto this. Services may wish to consider language which values the person they are, holding in mind how they want to be seen and how this interacts with their coping. For example, by referring to them based on their relationship to the care recipient or their names rather than referring to them as a carer. By services respecting and acknowledging the identity of family members, they may create safety for family member to consider their needs.

Services may wish to consider advanced care planning, which is reviewed regularly, to meet the changing needs of care recipients and family members. This should include post-bereavement planning and discussion around family expectations and service provision. Services may wish to value the coping styles of family and the individuals, and these should be communicated and respected at all stages. However, services should check in regularly and offer options for contact (e.g., telephone or visits to contain the vulnerability families feel). This may allow emotional, physical and cognitive resources to be used to support themselves and their loved one.

2.5.4 Limitations

This study explored the experiences of a small number of women who had cared for a spouse with MND and received palliative care in the UK. Given the limited literature in the UK, this was an appropriate method, and does not aim to be generalisable. The focus of this was to guide future thinking, provide considerations for services to explore and provide a direction for future research. There was homogeneity in participants in that they were all white British women of or approaching retirement age. These findings may not be transferable to individuals from other socio-cultural backgrounds. Only one participant experienced specialised palliative care. Therefore, there was heterogeneity in the sample in terms of the level of support and contact with services participants received and some received just district nursing. Some seemed unsure at times about who was involved from what service. Therefore, this research cannot be considered as an exploration of specialised palliative care, but what spouses experienced as palliative care and an analysis of the variability in experiences based on the level of support received. This heterogeneity and homogeneity observed in the sample may impact the transferability of the findings.

In addition, there was variability in terms of the amount of time which had passed since they loss of their partner. The research looked at retrospective accounts and perspectives of their experiences which may have changed as time passed. This may be particularly relevant when considering post-traumatic growth seen with Lizzie, who had the longest gap between losing her husband and participating in the research. It is possible that the experiences of the other participants may have been different had more time passed, and therefore our understanding of their experience should be considered in the context of this.

Two family members interviewed received support from palliative care during the Covid-19 pandemic. Therefore, this paper should be seen as an understanding at a specific time.

There may been learning within services from this time which changed the landscape of care.

An analysis of the quality of this research against Yardley's evaluation criteria was conducted (Appendix I). With regards to sensitivity to context, this paper does not explore in depth the context of care in the literature. For example, if participants received community or inpatient care. However, this is partly due to limited research to allow for these contexts to be explored thoroughly. The analysis lacked details and exploration of individuals socio-economic status, and therefore may have privileged some background characteristics to others and been insufficiently sensitive to aspects of context (e.g., culture). The goal of this research was to understand perspectives of UK carers and therefore was sensitive to some aspects of culture including age and gender. There was a lack of consideration of class and social economic background which may impact service experience. Additionally, culture is multi-faceted, and the UK is a multicultural country and therefore this may require some more targeted thinking in the literature which could be captured in interview schedules in future research. During data collection, some questions asked by the researcher were not in line with IPA processes outlined in Larkin et al (2021). For example, use of double or closed questions. This may have limited the quality of the data collected. Finally, the research does not make clear links with implications for policy, however, this may be more appropriate to be a focus for future research.

2.5.5 Future Research

All participants in this research were white British women. Future research should consider the experiences and needs of family from groups which are less represented in the current literature, including black family members offering care, male family members, and those with disabilities.

Research should also explore the experiences of professionals providing palliative care to individuals with MND and their families. This should aim to understand how they cope, experience, and respond to coping from family members. This will shape understanding of how to support members of people with MND through supporting staff.

Based on this study, future research may wish to explore the links between the themes identified in greater depth. This may include using quantitative means to understand any mediating or moderating relationships between GETs for example whether identity mediates the relationship between loss and seeking safety.

Research should develop the findings from this paper regarding spouses' experience of palliative services, and adapt the guidelines set out by Hudson et al (2012) to consider the cultural differences and specific needs of UK family members of people with MND. This could be achieved using focus groups with professionals and formal guideline development processes such as the modified Delphi processes used by Hudson et al (2012).

Finally, research should explore advanced care planning in MND in more detail, considering the impacts of this on family members and any possible barriers to implementing it in the UK.

2.5.6 Reflections

Researcher reflections were not placed in the results section to privilege participants voices. The researcher resonated with certain themes from personal family experience of MND. To explore whether the researcher was applying personal experience to the participants words, extracts were shared with others and decontextualized transcripts. Through this, the researcher noticed that fear of applying personal meaning blocked opportunities to analyse convergence.

When considering power, two participants were nurses. They may have felt more able to talk openly about their experience with the researcher as a professional. Participants with experience of feeling criticised by professionals could have struggled to openly discuss their actions through fear of criticism. The researcher managed this by building rapport and emphasising the goal of the research to understand participants’ experience.

All participants were white British women like the researcher. Perhaps this led to perceived shared experience which meant questions were missed through assumed shared understanding. Questioning meaning of words was used to manage this however there were times when this was not done seen the transcripts. All participants were older or retired woman. Greater vulnerability may have been perceived for them because of dominant discourses around gender and age.

The narrative account was used to break down convergence and divergence. Although prevalence was considered, the researcher identifying “gems” in transcripts (Smith, 2011). Therefore, a prevalence table was not included to prevent the reader privileging frequency. For further reflections see Appendix J.

2.5.7 Conclusion

This study has highlighted themes of battle, coping, and feelings brough on by receiving palliative care for five individuals who received palliative care while caring for a spouse with MND. Possible clinical implications and future research considerations have been explored.

Appendix A Search Strategy

Psych Info & Medline:

SI: DE "Medical Psychology" OR DE "Palliative Care" OR DE "Death and Dying" OR DE "Partner Death OR DE "Hospice" OR DE "Terminally Ill Patients"

S2: TI ("Palliative care" OR "End of Life" OR "Dying" OR "End-of-Life" OR "Terminal" OR "Hospice" OR "Motor Neuron* Disease*" OR “Amyotrophic Lateral Sclerosis” OR "Alzheimer*" OR "Multiple Sclerosis" OR "Huntington*") OR AB ("Palliative Care" OR "End of Life" OR "Dying" OR "End-of-Life" OR "Terminal" OR "Hospice" OR "Motor Neuron* Disease" OR “Amyotrophic Lateral Sclerosis” OR "Alzheimer*" OR "Multiple Sclerosis" OR "Huntington")

S3: *Search with OR*

S4: TI (("Carer*" OR "Partner*" OR "Spouse*" OR "Care giv*" OR "Caregiv*" OR "Family" OR "Families") N3 ("Psychological Intervention*" OR "Psychological Therap*" OR "Psychological Treatment*" OR "Psychoeducation*" OR "Psycho Education*" OR "Acceptance and Commitment Therap*" OR "Mindfulness" OR "Behavi* Therap*")) OR AB (("Carer*" OR "Partner*" OR "Spouse*" OR "Care giv*" OR "Caregiv*" OR "Family" OR "Families") N3 ("Psychological Intervention*" OR

"Psychological Therap*" OR "Psychological Treatment*" OR "Psychoeducation*" OR "Psycho Education*" OR "Acceptance and Commitment Therap*" OR "Mindfulness" OR "Behavi* Therap*"))

S5= S3 AND S4

Filters added: English language and peer reviewed.

Web of Science:

S1: TI ("Palliative care" OR "End of Life" OR "Dying" OR "End-of-Life" OR "Terminal" OR "Hospice" OR "Motor Neuron* Disease*" OR “Amyotrophic Lateral Sclerosis” OR "Alzheimer*" OR "Multiple Sclerosis" OR "Huntington*") OR AB ("Palliative Care" OR "End of Life" OR "Dying" OR "End-of-Life" OR "Terminal" OR "Hospice" OR "Motor Neuron* Disease" OR “Amyotrophic Lateral Sclerosis” OR "Alzheimer*" OR "Multiple Sclerosis" OR "Huntington")

S2: TI (("Carer*" OR "Partner*" OR "Spouse*" OR "Care giv*" OR "Caregiv*" OR "Family" OR "Families") NEAR/3 ("Psychological Intervention*" OR "Psychological Therap*" OR "Psychological Treatment*" OR "Psychoeducation*" OR "Psycho Education*" OR "Acceptance and Commitment

Therap*" OR "Mindfulness" OR "Behavi* Therap*")) OR AB (("Carer*" OR "Partner*" OR "Spouse*" OR "Care giv*" OR "Caregiv*" OR "Family" OR "Families") NEAR/3 ("Psychological Intervention*" OR "Psychological Therap*" OR "Psychological Treatment*" OR "Psychoeducation*" OR "Psycho Education*" OR "Acceptance and Commitment Therap*" OR "Mindfulness" OR "Behavi* Therap*"))

S3= S1 AND S2

Filters added: English language and peer reviewed.

<u>ProQuest</u>

S1 “Psychological intervention*” OR “Therap*” (Search in abstract)

S2 “Carer*” OR “Caregiver*” (Search in abstract)

S3 “Palliative Care” OR “Terminal” OR “Dying” (Search in abstract)

S4= S1 AND S2 AND S3

Filters added: English language, Doctoral dissertations only

<u>EThOS</u>

Therapy AND Carer AND Palliative

Social Care Institute for Excellence (SCIE)

Carer AND Psychological AND “Palliative Care”

Subject terms: Intervention

Content types: Research

Appendix B Demographic Information and Characteristics of Studies

Table summarising characteristics of studies

Characteristics of Studies	Number of Studies
Format of Delivery	
Individual Telephone	5*
Individual Face to Face	4*
Face to Face Group	10*
Face to Face Family	2
Duration	
3-5	7
6-9	7
10-14	3
Type of Control	
Active	7
Treatment as Usual	7
Wait List	2
No-Contact	2
Duration of Follow Up	
4-7 Weeks	2
2 Months	4
3-4 Months	6
6-8 Months	3
10-12 Months	6
Hours Caring Per Week	
30-39.9	1
60-69.9	1
70-79.9	2
80-89.9	3

Characteristics of Studies	Number of Studies
100+	1
Care Recipient Illness	
Alzheimer's Disease	10
Early Onset Alzheimer's Disease	1
Cancer	6
Multiple Sclerosis	1
Neurological	1
Other (Palliative Care)	2

*Note * indicated studies which included more than one format of delivery*

Appendix C Table to Show Study Quality

Summary of the quality ratings of included studies

Paper	**Overall Quality Rating**	**Selection Bias**	**Study Design**	**Cofounders**	**Blinding**	**Data Collection Methods**	**Withdrawals and Dropouts**
Au et al (2015)	*Moderate*	*Weak*	*Moderate*	*Strong*	*Weak*	*Moderate*	*Moderate*
Au et al (2010)	*Weak*	*Weak*	*Strong*	*Weak*	*Moderate*	*Strong*	*Moderate*
Campbell (1997)	*Weak*	*Weak*	*Weak*	*Weak*	*Weak*	*Weak*	*Weak*
Cheng et al (2020)	*Strong*	*Moderate*	*Strong*	*Strong*	*Strong*	*Moderate*	*Strong*
de Rotrou et al (2011)	*Moderate*	*Moderate*	*Strong*	*Strong*	*Moderate*	*Weak*	*Moderate*
De Stefano et al (2022)	*Moderate*	*Moderate*	*Strong*	*Strong*	*Weak*	*Moderate*	*Strong*
Fegg et al (2013)	*Moderate*	*Weak*	*Strong*	*Strong*	*Weak*	*Weak*	*Strong*
Ferrell et al (2019)	*Moderate*	*Weak*	*Strong*	*Strong*	*Moderate*	*Weak*	*Moderate*
Holm et al (2016)	*Strong*	*Strong*	*Strong*	*Strong*	*Moderate*	*Strong*	*Moderate*
Hudson et al. (2005)	*Weak*	*Weak*	*Strong*	*Weak*	*Moderate*	*Strong*	*Weak*
Kögler et al (2015)	*Moderate*	*Weak*	*Strong*	*Strong*	*Weak*	*Weak*	*Strong*
Leow, M (2015)	*Moderate*	*Moderate*	*Strong*	*Moderate*	*Moderate*	*Moderate*	*Weak*
Pahlavanzadeh et al (2015)	*Moderate*	*Moderate*	*Strong*	*Weak*	*Moderate*	*Moderate*	*Weak*
Marriott et al (2000)	*Moderate*	*Moderate*	*Strong*	*Moderate*	*Moderate*	*Weak*	*Moderate*
Oken et al (2010)	*Moderate*	*Moderate*	*Strong*	*Moderate*	*Moderate*	*Weak*	*Weak*
Cheng, Chan & Lam (2019)	*Moderate*	*Moderate*	*Strong*	*Strong*	*Moderate*	*Strong*	*Weak*
Cheng et al (2016)	*Strong*	*Moderate*	*Strong*	*Strong*	*Strong*	*Moderate*	*Strong*
Cheng et al (2017).	*Moderate*	*Weak*	*Strong*	*Strong*	*Strong*	*Moderate*	*Strong*

Appendix D Demographic Details of Participants

Table 1: Demographic details of participants

Name*	**Age**	**Ethnicity**	**Sexuality**	**Religion**	**Employed while caring?**	**Level of Palliative Care Input**	**Amount of Palliative care Involvement**	**Location of Death of Care Recipient**	**Length of Interview**
Lisa	61	White British	Heterosexual	Christian	No	District nursing, home palliative care nursing, outpatient hospice support for physical therapy	From diagnosis	Home	63
Debbie	56	White British	Heterosexual	None	Yes	District nursing, community OT and Physiotherapy, Marie Curie nursing, home carers	From diagnosis	Hospital	60
Rachel	65	White British	Heterosexual	None	No	Home support including carers and specialist palliative nursing, two hospice admissions, input from hospice psychology for her and husband	2 years post diagnosis	Hospice	73
Cath	66	White British	Heterosexual	Catholic	No	District nursing, home palliative care, community palliative care consultant	From diagnosis	Home	62
Lizzie	52	White British	Heterosexual	Christian	No	Marie Curie carers in last 2 weeks of life, monthly check ins with hospice key worker, 2 visits from specialist MND nurse, post-bereavement nursing contact	From diagnosis but most support was in the last 2 weeks of life	Home	65

*Note: * Participants names were changes to protect confidentiality. The names provided in this table are aliases.*

Appendix E Inclusion and Exclusion Criteria for Empirical Paper: Understanding How Spousal Carers of People with MND Experience Palliative Care in the UK

Inclusion Criteria

Cared for partner/spouse with MND in the UK

Cared for their partner through end of life to death

Cared for partner since 2016

Cared for partner for a minimum of 6 months from onset of symptoms

Received some experience of palliative care

Exclusion Criteria

People currently still caring for their partner

Carers who are unable to provide informed consent

Carer with cognitive impairment to the extent that they are unable to recall or describe their experience

Participants in mental health crisis

Participants under crisis services in the last 12 months

Participants who lost their partner <6months before the interview.

Appendix F Interview Schedule

1. Tell me what palliative care means to you
2. Tell me about what things were like for you before your partner started receiving palliative care
3. Tell me about your first experience of receiving palliative care?

   - Prompt if needed: What was this experience like? How did you feel about it at the time?
4. What was it like for you as a couple getting support from palliative care during their illness?
5. How did palliative care affect your experience of caring for your partner?
6. How did palliative care affect you personally?
7. Is there anything you would have liked to have been different in the palliative care you received?
8. What was your experience of the healthcare professionals involved/ how did you experience healthcare professionals involved?
9. I wanted to ask you some questions which may be difficult about experiences of your partner [*using their words for death, name of partner etc]* passing away- would this be ok with you? Tell me about your experience of palliative care when [*loved one passed away- using their words/name of loved one*] your loved one passed away?

10. Looking back on the whole experience, was it what you expected or was it different? how do you feel about the palliative care you received?
    Did it match up to expectations? Was it different?
11. Is there anything that we have not spoken about that you would like to share with me about your experience of palliative care when caring for your partner?

Prompts for depth:

Why?

How?

Can you tell me more about that?

Tell me what you were thinking?

How did you feel?

Can you tell me what you mean by "…………

Appendix G Master Table of Group Experiential Themes, Subthemes and Quotes

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
Battle			
	Searching for allies	“Because initially when he was first diagnosed in the first few days, I-I went online to see who these specialist nurses were because I knew we were going to need some support.”	Cath (17, 492)
		“Um so everything was a bit of a battle for when any agencies were involved, yeah.”	Lisa (17, 507)
		“We found out sort of by trial and error a lot of the time, um but it- to me, it was because I'd always worked as part of a team, it was important to me to have somebody that I could work alongside.”	Cath (18, 517)
		“And thankfully my two kids, my daughter moved back in with me to help me look	Lisa (3, 77)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		after her dad. And my son also helped when my daughter had to go to work so, yeah.”	
		“Um friends helped. I had- I had one good friend. Well, lots of good friends, but uh one friend in particular that I was able to talk to.”	Debbie (19, 538)
		“So, um I think at that point we just lost confidence. The confidence was built up a little bit a couple of weeks later.”	Cath (15, 450)
		“And she understood what our needs were, and I think that made the difference. I think it was-a lot of it was about not really understanding what our particular needs were. We didn't fit the mould because of the way we wanted to operate, and that might be because I was a nurse, I don't know, but we didn't fit the mould.”	Cath (29, 869)
		“But, um yeah, the the actual the actual people that we I	Lisa (19,568)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		suppose ended up with (laugh) in the end were very, very good, yeah they were very good. Always there at the end of the phone if I needed them, including the specialist nurse at the hospital, she was amazing.”	
		“Then get him out at the hospital, at the other end, um it just-it became apparent, I think, that we just need to, almost, probably try and cope on our own.”	Lizzie (14, 406)
		"Um and we had I think that was probably about the time, which would have been about nine months to a year – no, about nine months – before uh he passed away that we started getting outside agencies involved"	Debbie (6, 147)
		“Um I almost sort of put the barriers up, I suppose, really, to protect Matthew, perhaps um.”	Lizzie (7, 193)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		"Um and yes, we didn't make that connection, so like when the carers stopped coming, I didn't feel any great loss. If anything, it was a bit of a relief not to have people coming into the house. Um however, I did feel at a loss, the contact with the other services (voice cracking- becoming tearful)."	Debbie (11, 297)
		"And during their sort of their MDTs, their meetings, that um it would be taken into consideration, that his thoughts and feelings and my thoughts and feelings were- were taken into consideration"	Rachel (11, 312)
	Conflict	"It was like you think you're prepared but I suppose we'd not even thought about- because Paul would not talk about before he was poorly ever he would never ever talk about dying or death. Whereas I'm quite open about all of that	Lisa (35, 1073)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		stuff, you know you're born, you live, you die."	
		"Everybody would say to me, "You're going to need support", "You're going to need help", "You're going to need care" and I was blowed if I was going to put him through that as well."	Lisa (3, 83)
		"Matthew, perhaps more so than myself, very extremely private person, and only wanted his immediate family uh around him."	Lizzie (2, 37)
		"Interviewer: What would go through your mind, when the nurse would say that, "Go back and get some sleep"? Lizzie: Um guilt. Yes, guilt um (tearful), when I knew (pause) I knew I had nothing to feel guilty about, but it doesn't stop you feeling those (tearful) those feelings."	Lizzie (10, 291)
		"From a purely selfish point of view, it would have been good to have	Debbie (27, 771)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		somebody stay with him overnight so that you could get a full night's sleep."	
		"And so, the support mechanisms that may have been available to us weren't taken up until almost, what ended up being the final two weeks. So, I knew support was there, but I wasn't able to access it."	Lizzie (2, 39)
		"Um and at that point, he uh, with much resistance, agreed to having uh carers come in and help with the-the washing, changing and stuff"	Debbie (6, 152)
		"Um and then, really, it was just the carers coming into the house, um and I guess they were probably viewed as a necessarily evil."	Debbie (10, 257)
		"Well, just very simple things like being able to have a shower because he needed care every minute, being able to go to the shops, being able to sleep at night,	Lisa (5, 114)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		being able to lie in a bed because I lay on the floor on cushions for the last few weeks when he was in the hospital bed.”	
		"Retrospectively, probably, an emotional exhaustion."	Debbie (18, 498)
		“And um (tearful) I needed help at that point because it was getting difficult physically to move him around, and I wasn’t getting any sleep, I was only getting two hours a night in that last week (tearful).”	Cath (33, 1004)
		“Um physically um crying, um physically uh I’d lost a lot of weight, the weight just mel-melted away, uh day by day, week by week. I think I’ve gone down to about eight stone.”	Lizzie (4, 96)
		Um so perhaps um perhaps that there’s a little bit maybe that- I-I actually was feeling a bit exhausted and things, and I did- I did, myself, um phone up a care agency, um to	Rachel (17, 487)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		see if they could come out.”	
		“So obviously I was aware that I was letting or feeling that I was letting him down.”	Lisa (4, 99)
		“That's probably my only regret, really, that I just couldn’t cope with him, (Laughter) through lack of sleep.”	Cath (34, 1036)
		“It was a night where we didn’t have a Marie Curie nurse. Um it was around 4 o’clock in the morning (tearful). I had drifted off to sleep in the chair, in-in the bedroom, despite best efforts to keep awake. I must have drifted off to sleep, come round and something just told me to go over to Matthew, lying in the bed. And it could, perhaps, only have been a few minutes prior. There was beads of sweat on his forehead, um eyes closed and I called his name.”	Lizzie (20, 508)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		“Then he was getting me up at 1:00 in the morning and then because he couldn't communicate other than through the eye gaze, it meant I had to have the light on all night. And then I couldn't go back to sleep because I was anxious about him and (crying). And then he got me up on the last night at about 1:00, and I just got really angry with him (crying). And then he died ten minutes later (crying).”	Cath (34, 1011)
		"Yes. Well the reason why my husband got admitted um was that I fell upstairs-, I slipped in the bedroom and I. That was in the June- May sorry May 2021. Um and my husband uh had to was offered a hospice bed a week later, because I had dislocated my shoulder."	Rachel (18, 508)
		“And I felt very isolated. Because I'd said at the outset, “I	Cath (4, 98)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		want to do things myself,” and right up to the end I did want to do things myself and I did do that.”	
		"Because you wouldn’t have wanted to need that. So you wouldn’t have wanted to be in the position that you were in, um that-that it was necessary to have that help."	Debbie (10, 279)
		“I mean they’re just coming in and they’re saying, “Well, you need a hospital bed,” “All that furniture’s got to go” and it’s like, “Well, where am I going to put it?” I’ve only got one other room, where am I going to put a three-piece suite and a table? And and and lift all of that on my own, you know (laugh) so it was just it was impossible.”	Lisa (20, 598)
		“in some ways, almost, would feel (pause) relief when they’d gone.”	Lizzie (11, 301)
		“And everybody was really trying to help, I'm not saying they	Cath (8, 214)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		weren't, but it just well it just- it just got too much, really. And it just made me feel that my home was not my own"	
		"We weren't getting anywhere. There were so many people in that room, and nobody was speaking."	Cath (20, 824)
		"It was just saying, "Well, yes, this has declined, this has declined, this is no longer..." you know. And again, there were no positives."	Debbie (14, 372)
		"We needed time to get our heads around it as well, without it becoming medicalised and everybody coming in and talking about it all the time."	Cath (10, 281)
		"And so then when there was a little bit of downtime, somebody would come in, so you never got any time together, no quality time together."	Cath (8, 229)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		"Um so, I would you know sleep in the other bedroom. Um the Marie Curie nurse would- would sit with Matthew, um and so we felt, with all due respect, I suppose, we felt that our privacy had gone."	Lizzie (9, 262)
	Identity	"No, no, this is a man that a few weeks ago was writing reports for an expert witness to the courts. You know he's not going to sit in the house with no pants and trousers on, that's not answer, that's not a solution."	Lisa (29, 904)
		"It was- I think, again, it was just the highlighting of the fact that he couldn't do these things anymore, for someone who was so independent."	Debbie (9, 231)
		"Um they were interested in us as a couple, as people, I suppose, which is probably something we didn't really get particularly from the carers. You did on a	Debbie (11, 309)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		superficial level, but you didn't feel that they knew you in the same way that particularly the district nurse."	
		"Um but that was another step. We probably both resisted that uh because it very much marks the end of being a couple (tearful)."	Debbie (29, 815)
		"That would actually treat me, um you know, be somebody with a bit of intelligence."	Cath (18, 521)
		"And that was my role, um which I wanted to-to perform."	Lizzie (10, 270)
		"Well it was it was just. Um it was just relinquishing (tearful) my role of being a carer, which I had been for a you know for a very long time, it seemed like a long time. Um so I was just sort of relinquishing my role for a while, and I was able just to-to relax a little. And you know we had a bit of quality time together you know where we."	Rachel (23, 650)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		“They- I think, when you're a nurse or anybody, your not you know- you keep this patient- and there’s a distance and you know. I know I would just be another person and David would just be another person coming through their system. But we did feel that they where- they treated us so individually, and so caring and um. Yes, it felt that you were a bit special (tearful).”	Rachel (22, 632)
		“And you also hear from people about how it changes your relationship. Um you become a carer and not a wife, and I've always bucked against that and said “No, I'm not his carer, I’m his wife, I'm always his wife, I'm not his carer, I just happen to look after him.”	Cath (11, 327)
Coping	Searching for Validation	“They have no idea how you’re feeling, what you’re going through.”	Lisa (19, 586)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		“They need training in or being around people with Motor Neurone Disease and their families, yeah. Otherwise, it becomes a textbook experience I think and that’s not good (laugh).”	Lisa (37, 1137)
		“Um they have the checklist I know they-, I mean, I'm on studies where they're looking at this checklist about… It is a checklist, whatever way you look at it, it's a checklist.”	Cath (27, 802)
		“Um friends helped. I had- I had one good friend. Well, lots of good friends, but uh one friend in particular that I was able to talk to because, about three years before that, she’d been diagnosed with cancer.”	Debbie (19, 538)
		“And I know I wasn’t because everybody said, all the other people were saying, “You’re doing a fantastic job”, “You’re doing really,	Lisa (13, 395)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		really well. Keeping going", blah, blah, blah "You're looking after him. You've done exactly what you should be doing."	
		"She can look at me and know how I'm feeling you know. I've not slept more than an hour for three days now (laugh) you know and she knows she knows how I'm feeling whereas some of the others they don't know."	Lisa (37, 1152)
		"And speech and language therapy particularly because I'd say something to Rodger, and then she'd come in and she'd say exactly the same thing, even though we hadn't discussed it."	Cath (29, 860)
		"I think, to talk to somebody who knew- You know, we are blessed with a very loving family and lots of good friends, but and were all sympathetic and empathic, but nobody understood what we were facing, day in and day	Lizzie (3, 63)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		out. Why would they? But an-but the association visitor did, because she had lived-lived through it with her husband.”	
		“Um yes, because you’ve spent 18 months building up a relationship with-with people, who-who have seen you at your worst and seen the worst possible things.”	Debbie (23, 655)
		“Um but the palliative nurse, as soon as she came, she said all the right things as she walked in the door. You know, “You need to spend some time getting out and about while you can, and I'm not going to be pestering you and being really sort of in-your-face, I will keep in touch, but, you know, I'll do things the way you want to do them, and how do you want to do things?” And it was that kind of thing.”	Cath (18, 528)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		“Uh but again that’s not their fault, that’s training and the opportunity to be around people because it is such a rare disease. Not everybody gets the opportunity to be around people with it but, yeah.”	Lisa (38, 1176)
		"But for the most part, I would say, probably, 75% of them were caring and compassionate about what they did, um but nevertheless, still had-didn’t have that understanding of MND."	Debbie (7, 188)
		“And so we had another we a listening ear to the palliative care team to actually express some of our, um or my husband’s, sort of wishes.”	Rachel (13, 354)
	Seeking Safety	“I wouldn’t necessarily say in denial (tearful), because you couldn’t fail to see what was going on, with my own eyes, what was in front of me, because the um the	Lizzie (6, 172)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		deterioration was so-so quick.”	
		“His physical condition was changing so much, sort of weekly or monthly, that every month, there was another bit of equipment in the house, you know another demonstration of how-“	Rachel (6, 166)
		"So um yes, it’s- it’s just something that we had to do and had to accept, um albeit reluctantly, but then the whole thing was (laugh) you know you didn’t want to be there anyway. "	Debbie (10, 270)
		“The hardest thing (laugh), I tell you the hardest thing and it’s the silliest thing really.”	Lisa (29, 884)
		“He liked her, but he hated her coming because he always felt like she was the Grim Reaper. (Laughter)”	Cath (16, 481)
		“"All that suddenly just disappeared um and like we,	Debbie (22, 619)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		literally, saw nobody ever again (laugh). They just completely stopped. "	
		“You know you want periods of time- When you're going through this, you want periods of time where you can actually do something, have a good time and forget about it.”	Cath (10, 285)
		“She hoisted Paul up and he fell on the floor (laugh) um- not in a bad way you know - but we just laughed, and we were just all in a heap on the floor you know having a laugh. That was just what we needed, somebody that was real and got it.”	Lisa (19, 556)
		“Um and as things got worse, that became harder because, because you know, as he became far more disabled, but we still had you know we still has a really good time when we were together on our own.”	Cath (10, 287)
		"We’d kind of resisted that as	Debbie (29, 834)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		long as possible. "	
		“Maybe we don’t want to accept- well, no, it’s not that you don’t want to accept because I think we did accept it. You just want that person to have as normal a time as possible.”	Lisa (8, 232)
		“Because by having it, I was almost forced to accept what was happening (tearful).”	Lizzie (23, 668)
		“You know, his condition could have gone on a lot longer than it did, but it did seem to be that we were in our- Once they were involved, that that it- that was going to be the final part of his MND journey.”	Rachel (6, 152)
		“Then shut the front door and the mask comes off, and the flood gates open (tearful), which is what I did, an awful lot.”	Lizzie (18, 504)
		"Yes, you were so wrapped up in just actually doing that you didn’t really have a lot of time to think about the emotional side."	Debbie (18, 488)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		"Um partly, personality. Um I've always been quite an outcome-focused, positive person, so solution based, always looking for ways to make things work."	Debbie (9, 515)
		"You have to have your porch knocked out." "You won't be able to have an electric wheelchair unless you have that knocked out." "You won't be able to do…you can't have that" "You've got to have a wet room built." I was like, "Aargh" and oh, it was just awful."	Lisa (18, 533)
		"There was- it just, basically, said, "Well, you can't do this and you can't do that." Um s- and i- if you felt that there was something that you could be doing to improve things, then it wouldn't- wouldn't have been so bad, but there was no point for it, just for them to mark the decline."	Debbie (14, 372)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		“So initially, particularly in the early stages, when you're actually trying to live your life.”	Cath (6, 171)
		“So we were in control really. Yes, we were in control. Um the teams were there, but we were in control when we accessed them, or to say yay or nay for a visit.”	Rachel (9, 248)
		“But after the month and a bit that he was in there- It was like a blanket of love, being in there.”	Rachel (22, 618)
		“I just felt, (tearful) out of a very, very bad, bad thing, was a very gentle and kind and comforting experience.”	Rachel (11, 294)
		“Um the right person um a real, as I said before, a real person, somebody that just you know they didn’t expect you to go flying round with the hoover before you visited and (laugh) you know just normal.”	Lisa (21, 703)
		“Interviewer: Okay. That	Debbie (11, 306)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		connection, them connecting with both of you, how did they do that? What did it look like?" Debbie: "Um (pause) that's a very hard thing to pin down because that's- it's kind of the key to what makes you like a person. Um and I don't know how you can define that, but um they were – I don't know – um they were interested in us as a couple, as people, I suppose, which is probably something we didn't really get particularly from the carers."	
		"And that they were sort of you know tucking my husband up at night and taking good care of him and loving him, like I would do."	Rachel (22, 641)
		"I mean I suppose I'm a bit awkward, I can tell how somebody is when I first see them (laugh), I get a feeling and I'm not usually wrong. Um and you'd think, "Oh,	Lisa (20, 592)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		crikey, here's one that doesn't really get it."	
		"So, um I think at that point we just lost confidence."	Cath (15, 450)
		"I went, "Fair enough." Um so, we sort of bowed out in that respect."	Cath (27, 815)
		"Um I suppose, they taught me that you know you don't have to put the mask on, you know, don't-don't protect other people from the emotions that you're feeling. Um and over time, I-I guess, I've learnt to do that."	Rachel (18, 507)
Feelings Brought on by Receiving Palliative Care	Vulnerability	"And and who you know where do you define 'last, last days'? You know, you just don't know what's coming, so um you don't know what the last days are."	Debbie (2, 29)
		"Which even though I'd been an intensive care nurse, frightened the life out of me, dealing with that at home."	Cath (16, 468)
		"And of course, I'm not medically trained so you're doing the best	Lisa (6, 155)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		you can with the little knowledge that you have and hoping that you're doing the right thing."	
		"And she made me feel absolute rubbish and um she got she said, "Well, the bed guard needs checking, I'm going to get the Red Cross round" and all of this. She said, "They're coming out tomorrow.""	Lisa (13, 379)
		"I mean I'm just normal, I'm just me, just a normal person but some of them they come in, they are so stuck up. And like I said, they looked down their nose at you, but they have no idea."	Lisa (19, 583)
		"Um and they sort of took that out of our hands and said, "You don't need to worry about that. You know you will not be attending that interview. And it you know it will all be sorted out," and they did sort all of that out. Uh so they made sure that we got- we were entitled to"	Debbie (14, 395)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		“But um the reality was, I-I-I was falling apart, bit by bit, day by day (tearful).”	Lizzie (4, 88)
		“He’d- we’d be together for about 25 years, I think, and he’d always been the one that had taken care of me.”	Debbie (19, 516)
		“Um and how you know-I still learnt it, it didn't take long, but it was having that confidence in the first place because you go, you get given this ventilator, and they go, “Off you go,” you're out of here.”	Cath (31, 916)
		“This is the benefit of time and hindsight, perhaps if ever I was faced with a-a need to access palliative care services, having gone through that experience, um and learnt a lot more, and have a lot more knowledge now than what I did then, my approach to using them, perhaps, would be very different now.”	Lizzie (23, 652)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
	Loneliness	“That was awful because it just reinforced that I didn't really have anyone.”	Cath (17, 490)
		“Um yeah, I think it was probably about three from the actual nurse um but she was there, she was always at the end of the phone. She made it very clear she was there.”	Lisa (22, 660)
		“Um but she was lovely and throughout she was brilliant. If ever I needed her, she was there. And a couple of times I did need her, and she was absolutely spot on.”	Cath (16, 471)
		“We weren't getting anywhere. There were so many people in that room, and nobody was speaking.”	Cath (28, 824)
		“and here I was, um hav-ha-having to access that support, and despite having people around me and-and lots of good and loving people around me, still feeling um (tearful) very	Lizzie (7, 177)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		isolated and alone. That's through no-no-no fault of-of anybody, at all you know. Marie Curie wer-were-were lovely. Um It was just my (tearful) feeling, my-my mental state, I suppose, (tearful) at the time."	
		"You know when we needed the stuff, fair enough, but when we didn't, we should have been left alone, really."	Cath (10, 273)
		"In some ways, almost, would feel (pause) relief when they'd gone. that we were just on our own again."	Lizzie (11, 301)
		"But it was COVID, it was nobody's fault. It's just the way it was."	Cath (37, 1119)
		"Um so, yeah, (tearful) they would have been there. I know they would have been there if I needed them, that was the thing, but they couldn't."	Cath (34, 1034)
		"But it's not something that he would have wanted. And therefore, it	Debbie (27, 773)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		wasn't something that we looked for."	
		"Um but I knew I-I-I just had to pick up the phone to a-to a friend (tearful) and-and they would be there, they-they would rally round but (tearful). In many ways, I couldn't. Um (pause) and so, the loneliness, probably, was exacerba- exacerbated by that."	Lizzie (8, 209)
		"Um you felt abandoned. (Tearful) You had a lot of emotional stuff to deal with and nobody, really, to share it with. I mean, there was- there would have been counselling available and stuff like that, but that's not (pause)- that's not really what I felt the lack of (tearful)."	Debbie (23, 646)
		"Well just that the- I suppose just that we had the experts with us, really, that was the-the pinnacle really."	Rachel (16, 441)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
	Loss	“And it just made me feel that my home was not my own.”	Cath (8, 273)
		"Everything has gone, everything has been stripped away, you’re almost back to being a baby where you can’t do anything, literally anything for yourself."	Lisa (26, 792)
		"And um but I think MND is particularly cruel because it takes people who are so fit and healthy and, within such a short space of time, just strips every shred of dignity and independence from them."	Debbie (30, 850)
		“And they didn't want to allow us to do that, for all the right reasons, but weren't listening. And I think just seeing me as difficult when I was trying to explain that to them. Um and that's something I would like to see change in lots of respects.”	Cath (37, 1150)
		"Um it very much marks the move from partner to patient. "	Debbie (28, 826)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		“So you never got any time together, no quality time together.”	Cath (8, 225)
		“So then of course (laugh) you’re on the internet again trying to find out what you can about that and none of it none of it’s any good, none of it’s reassuring because there is, there is nothing. There’s no drugs, you know there’s a drug that you can take to give you a few more weeks of life, well, of course he was on that.”	Lisa (27, 830)
		“Well, you can’t do this and you can’t do that.” Um s- and i- if you felt that there was something that you could be doing to improve things, then it wouldn’t- wouldn’t have been so bad."	Debbie (14, 375)
		“But it wasn't anybody’s fault, it was just, it was COVID, and that threw everything (crying). And it was the first COVID lockdown. So we	Cath (34, 1018)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		were only three weeks into it, and nobody knew what they could do, what they couldn't do (tearful)."	
		"So, I was disappointed for her and then not giving her the education to actually do the job."	Cath (20, 610)
		"Suppose it's become a little bit more obvious now because we've had experience of cancer palliative care and how different that is, and how insightful they seem to be, whereas we didn't have that."	Cath (24, 709)
		"Different council, but carers came in and the carers were really good, um and they came to the funeral and stuff. So you had that well, at least a couple of them did."	Debbie (23, 629)
		"Um but yeah that was the other biggest void it was just the complete cessation of everything, immediate cessation. You	Debbie (23, 640)

Group Experiential Theme (GETs)	Subtheme	Quote	Participant (page, line)
		know there was not any contact with anybody."	
		“Um so it was- it was a special, quite an uplifting, experience actually.”	Rachel (10, 271)
		“Um they talked to me a lot about stages of grief and um, perhaps, to accept that it’s okay to not be okay, sometimes, um and not have to put, necessarily, the mask on.”	Lizzie (18, 501)

Appendix H Yardley's Evaluation Criteria

Table Summarising Evidence and Concerns with Yardley's Evaluation Criteria

Yardley's Evaluation Criteria	Evidence	Concerns
Sensitivity to Context	Showed sensitivity to existing research across different disciplines considering nursing and medical research as well as psychological. Considering cultural factors to previous findings	Lack of homogeneity in terms of palliative care involvement
	Clear rationale for use of qualitative research methods and how this paper would address the gap in research	Lack of consideration for socio-economic status therefore privileging some socio-cultural factors over others
	Novelty in research question with consideration of UK specific culture	
	Described socio-cultural background of participants	
	Explored differences in socio-cultural background between participants and researcher and considered impacts of this	
	Sensitive to ethical considerations particularly around reduction of psychological harm to participants	
	Reflected on the impact of setting when collecting data	
	Privileged participants voice by not including my reflections in the results section	
Commitment and Rigour	Followed process set out in Larkin, Flowers and Smith (2001)	At times in interviews closed questions or double questions were used- associated with lack of experience in IPA

Yardley's Evaluation Criteria	Evidence	Concerns
	Attended IPA training	
	Engaged in further learning with IPA including reading and reflected on this in reflective log	
	Stated epistemological position in methods	
	Clearly stated why IPA was chosen over other methods	
	Suitable homogeneity of sample	
	Reflected on variations in sample, how tightly defined this is particularly considering the relationships carers had with care recipient and why	
	Followed interview process guidelines in Larkin, Flowers * Smith (2021)	
	Received feedback from supervisors of the suitability of interview schedule	
	Reflected on interview quality and depth in supervision	
	Very clear paper trail of whole analysis process alongside reflections in reflective log	
	Clear narrative of topic area- literature review mirrors my processes of getting to the research question	
	Clear research aim rationale stated in methods	
	Journal from start of study development and write up mirrors this	
	Analysis explores links to themes and accounts in detail	
	All interviews looked for "gems" and asked myself why these were important	

Yardley's Evaluation Criteria	Evidence	Concerns
	Completed perveance table for themes with a 50% cut off however this was not included in write up or appendices as I do not with the reader to privilege prevalence	
	Cross referenced prevalence table with "gems" to identify whether they were being missed	
	Paper trail- section of annotated transcript, example table of emergent themes/PET and final master table in appendix	
Transparency	Detailed description of design in methods with links to literature	Table of master quotes includes only quotes included in the write up
	Considerable evidence in appendix including data quotes, and reflective log	
	Thorough paper trail	
	Reflections on convergences and divergences	
	In analysis interpretations are grounded in quotes and detailed description of how interpretation was reached is present	
	The number of people who were included in each theme is explored and considered	
	Model of relationships between themes is available	
	Good number of quotes available for different themes	
Impact and Importance	Strong and thorough links to the impact of the literature on clinical practice	Lacking implications to policy
	Comment on the novelty of the study and the gap the literature fills	
	Clear links with findings, existing literature and gaps	

Yardley's Evaluation Criteria	Evidence	Concerns
	Clear recommendations for future literature Rationale for choosing not to include prevalence table presented in reflections in discussion	

Appendix I Reflections of Introduction, Methods, and Discussion

Introduction

During the process of developing this research I kept a comprehensive journal of all papers I had read and how my ideas formed to take me to the final research question. Therefore, the introduction section of this paper is a direct reflection of my thinking. When writing this I found it difficult to decide what papers were the most important to make it into the final draft. I felt wedded and stuck to my process as opposed to what would be readable and important for the reader and used supervision to bring clarity to some of my thoughts and the research out there currently. From considering the past research I was left thinking about how little is considered in the UK wit carers of people with MND. Perhaps the research itself mirrors the reality of services and therefore experience of carers in terms of isolation and lack of information.

Methods

When developing the methods for this research, I held the wellbeing of participants at the forefront of my mind. I noticed how much my career as a clinical psychologist shaped how I approached planning and delivering the interviews. At times, I felt unconfident and unsure given that this was the first full piece of qualitative research I have worked on, however I noticed a natural pull to search for meaning from my profession which supported this process. I also noticed that during interviews I had to work hard to not make interpretative statements like I would in therapy with individuals. There were points where the participants' next statements were different to interpretation in my mind as we were talking. Also, I noticed how different some of my final themes and experience of the interview was after full analysis. For example, for Lisa, after the interview I felt there was little emotion, however after analysis I saw the emotion came from linguistic comments as opposed to statements. This demonstrated to me the importance of not making interpretative comments during the interview. When writing the methods section of this paper I found it difficult to sculpt what might class as sufficient detail to replicate. Particularly given the iterative process of IPA analysis. I then noticed that getting pulled into an idea that there might be a checklist of what needed to be there went against IPA approached.

Discussion

In light of the findings from this study I changed my use of the term 'carers' to family members. Reflecting on the current research and links with my research I notice changes in how I think about how we support family members. I have clear ideas of what constitutes good care; however, this has shown me the importance of holding flexibility with this. This has changed my understanding of the field in moving away from an idea that there might be many shared things family members need and towards an idea of variety. This also changed how I think about how family members experience role transition and considering the western-centric nature of such values and how this can correspond with or vary comparative to research in other countries. When writing the discussion, I found it hard to establish where the results ended, and discussion began. My previous experience writing results and discussions is with quantitative research where all interpretations are for the discussion, whereas I was struck with the challenge in shifting to the results themselves being interpretative in nature.

Appendix J Journal of Death and Dying Instructions for Authors

Manuscripts can be submitted in APA style to **https://mc.manuscriptcentral.com/omega.**

Please refer to the latest Publication Manual of the American Psychological Association. A synopsis of this manual is available from the American Psychological Association. http://apa.org/

Originality Authors should note that only original articles are accepted for publication. Submission of a manuscript represents certification on the part of the author(s) that neither the article submitted, nor a version of it has been published, or is being considered for publication elsewhere.

Format Prepare manuscripts according to the latest Publication Manual of the American Psychological Association. A synopsis of this manual is available from the American Psychological Association. http://apa.org

Manuscripts Manuscript must be word processed, double-spaced, with wide margins. Paginate consecutively starting with the title page, which should be uploaded as a separate file. The organization of the paper should be indicated by appropriate headings and subheadings. Please be sure to remove all self-identifying information from the manuscript file before submitting. Author information should only be included on the title page.

Style Technical terms specific to a particular discipline should be defined. Write for clear comprehension by readers from a broad spectrum of scholarly and professional backgrounds. Avoid acronyms and footnoting, except for acknowledgments.

Permissions Authors are responsible for all statements made in their manuscript and for obtaining from copyright owners to reprint or adapt a table or figures, or to reprint a quotation of 500 words or more. Authors should write to original author(s) and publisher to request nonexclusive world rights in all languages to use the material in the article and in future editions. Provide copies of all permission and credit lines obtained at the time of manuscript submission.

Manuscript Submission Guidelines:

Manuscript must be word processed using Word or Open Office Writer, double-spaced, with wide margins. Paginate consecutively, starting with the title page.

Title Pages should be uploaded as a separate file and include the follow as is applicable:

- Full article title
- Acknowledgements/credits
- Each author’s complete name and institutional affiliation(s)

- Grant numbers and/or funding information

- Corresponding author (name, address, phone/fax, e-mail)

- Up to five keywords as it should appear if it were to be published.

Abstracts of 100 to 150 words are required to introduce each article.

Most articles are between 5000-7500 words and while we accept long pieces that mandates additional evaluation because of space limitations.

Manuscripts should be saved in a Word .doc or .docx file type. The organization of the paper should be indicated by appropriate headings and subheadings.

Please be sure to remove all self-identifying information from the manuscript file before submitting.

When possible, all illustrations, figures, and tables are placed within the text at the appropriate points, rather than at the end. If this is not possible:

Figures should be referenced in text and appear in numerical sequence starting with Figure 1. Line art must be original "drawings" in black ink proportionate to our page size. Indicate top and bottom of figure where confusion may exist. Labeling should be 8 point type. Clearly identify all figures. Large figures should be drawn on separate pages and their placement within the text indicated by inserting:

Insert Figure 1 here

Tables must be cited in text in numerical sequence starting with Table 1. Each table must have a descriptive title. Any footnotes to tables are indicated by superior lower case letters. Large tables should be typed on separate pages and their approximate placement indicated within text by inserting:

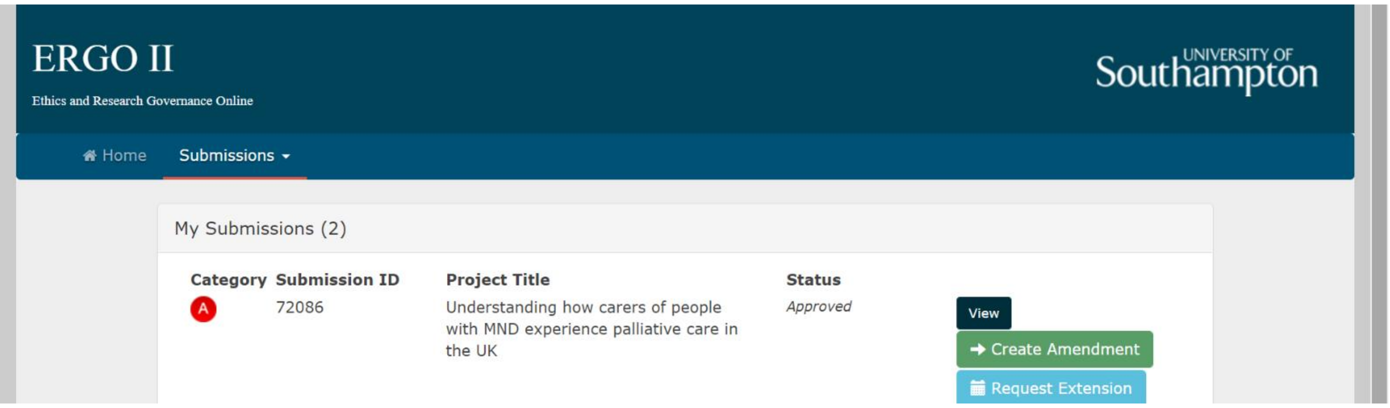
Insert Table 1 here

Direct questions to Dr. Kenneth J. Doka

E-mail: KnDok@aol.com

Manuscript Submission Guidelines: OMEGA - Journal of Death and Dying: SAGE Journals (sagepub.com)

Appendix K Ethical Approval



My Submissions (2)

Category	Submission ID	Project Title	Status
A	72086	Understanding how carers of people with MND experience palliative care in the UK	*Approved*

Appendix L Participant Information Sheet

Participant Information Sheet

Study Title: Understanding how carers of people with Motor Neuron Disease (MND) experience palliative care in the UK.

Researcher: Georgia Steed (Trainee Clinical Psychologist)

Supervisors: Dr Warren Dunger (Clinical Neuropsychologist), Dr Paul Beadon (Consultant Clinical Psychologist).

ERGO number: 72086, Version 2, 27/06/22

You are being invited to take part in the above research study. To help you decide whether you would like to take part or not, it is important that you understand why the research is being done and what it will involve. Please read the information below carefully and ask questions if anything is not clear or you would like more information before you decide to take part in this research. You may like to discuss it with others, but it is up to you to decide whether or not to take part. If you are happy to participate you will be asked to sign a consent form.

What is the research about?

My name is Georgia Steed, and I am a Trainee Clinical

Psychologists at the University of Southampton in the United Kingdom. This research is part of my academic qualification with the University of Southampton.

I am inviting you to participate in a study looking at how carers of people with Motor Neuron Disease (MND) experienced palliative care while caring for their loved one. I am particularly interested in understanding the lived experience of carers in the UK. There is limited research of carers of people with MND in the UK and therefore I hope to gain detailed information about carers' experiences within the unique culture and social and medical care offered in the UK. I hope that this research will help to better understand the needs of this group and whether any changes which may be beneficial in the palliative care delivery to this group.

The research question is: How do carers of people with MND experience palliative care in the UK?

This study was approved by the Faculty Research Ethics Committee (FREC) at the University of Southampton (Ethics/ERGO Number: 702086).

Why have I been asked to participate?

You have been asked to participate because you have at least 6 months experience in caring for a partner/spouse with MND (from when symptoms first started) in the past 8 years, before sadly losing your partner. To participate in this study, you must have experienced some palliative care input or support however there is no limit on the amount of input you and your partner received. A minimum of 6 month must have passed between the interview date and when you sadly lost your partner. I hope to recruit between 6-10 individuals to participate in this study.

What will happen to me if I take part?

This study involves taking part in an interview which will last for around 45-60 minutes. The audio of the interview will be recorded. This will allow them to be transcribed and analysed in detail after the interview is complete. Recording of the interview is a requirement for participation in this research. You will be able to choose whether you participate in the interview in person, or online using Microsoft Teams. If you feel this may be of interest to you, you will be contacted by the researcher for a brief discussion to establish whether you are eligible to participate. You will be given a consent form to read and sign if you are happy to participate. Together with the researcher, you will plan where and when the interview will take place. Therefore, this may involve some travel time if you wish to meet face to face.

Once the details of this meeting are agreed, a confirmation email will be sent to you. This will include a recommendation to bring someone with you for emotional support following the interview. If you choose to have an online interview, you will be sent a link for a meeting using Microsoft Teams when the consent form has been received. You will be sent a reminder email one week before the interview, and on the day before the interview.

If in person, on the day of the interview, if you choose to bring someone along for emotional support, the researcher will introduce themselves to them, provide them with the information sheet, ask if they have any questions and request that they wait outside of the room. If online, the researcher will do this online, and ask the elected support person to wait outside of the room the interview is conducted in.

You will have the opportunity to ask any questions, and if you are happy, you will give verbal consent to participate. The interview will be audio recorded when you give consent. You and the researcher will discuss the emotional distress which may arise from discussing such a sensitive topic. You will be in control of what you choose to share throughout the interview. You and the researcher will discuss and plan how you will let the researcher know if something is feeling too difficult to discuss, you would like a break, or would like to stop the interview.

The interview will last up to 60 minutes. You will be asked several questions about your experiences of palliative care when caring for your partner whilst they experienced MND. These might include things like “tell me about what experiences of palliative care you liked or found beneficial when supporting your partner with MND.” Participants would be expected to give detailed accounts of their personal experiences. You may also be asked questions to seek additional information which may be relevant. This may include things like “please could you tell me more about…..?”

You have the right to withdraw at any time during the interview. You can then withdraw your data up until the date that interviews are sent to be transcribed. The deadline date to withdraw your data will be provided on the day of your interview.

Following the interview, you will be thanked for your time and given a debriefing form which will include contact details. You will also be asked if they would like a copy of the research once it is complete. If the process of participating in the study causes distress for you, the researcher will allow time for the you to process the emotional impact of the interview and offered a grounding exercise for your wellbeing. You will be asked how you are feeling before leaving the room and if there is anything they would find helpful from the researcher.

You will then be sent an email confirming the deadline date to withdraw your data.

Following this, you will not be contacted again by the researcher unless you contact the researcher to ask a question, withdraw consent or have requested a copy of the research once complete.

Are there any benefits in my taking part?

If you choose to take part in the study, you will not receive any direct benefits, however, your participation will contribute to the understanding of the experiences carers of people with MND have in the UK. This may help us as a scientific community to better understand any areas which have been helpful or are missing from their palliative care experiences. This may support the development of recommendations for future support within palliative care services for partners of people with MND.

Are there any risks involved?

Discussing the illness and death of one’s partner/spouse may bring up the psychological distress caused by these experiences. This study aims to explore detailed accounts of personal experience. We recommend that you have someone available to offer emotional support after the interview. Your wellbeing will be monitored throughout the interview and options to manage any psychological distress will be discussed.

If you feel that your levels of distress require additional support after participating in this research, contact the following resources for support:

Your GP

Your local Community Mental Health Team or Crisis Resolution and Home Treatment Team (out of hours)

NHS 111

Samaritans – 116 123

You may experience inconvenience for yourself and elected emotional support individual, when getting to the interview of taking time out of your day to participate online.

Although physical discomfort is not expected during this research, you may experience physical discomfort whilst sitting for 45-60 minutes during the interview, or from looking at a computer screen for the duration of the interview. You will be offered breaks and any access needs will be discussed prior to the interview to help you to feel as comfortable as possible. Please bring any visual or hearing aids you may require to support your comfort if you choose to participate.

What data will be collected?

The interview will collect information about your personal experiences of palliative care when supporting your partner/spouse through the duration of their illness and death. This information is to help us to understand your experiences on a meaningful level. You may also be asked for demographic information including your age, gender and ethnicity. This will help us to understand some of the contextual factors of your experience.

The interview, including the questions asked and your responses will be audio recorded. These audio files will be sent to a professional transcribing service. They will be transcribed (converted into written form) verbatim, meaning that each word will be recorded exactly how it is said. The transcribing service will be confidential, and they will not share any information in the audio recordings with anyone else. Any identifiable information such as names, where people live, or work will be removed from the transcript. All files will be password protected and stored on a secure server.

Will my participation be confidential?

Your participation and the information we collect about you during the course of the research will be kept strictly confidential. Audio recordings of the interviews, transcripts, field notes and analysis of transcripts will be stored on a secure university server. They will be password protected and only accessible by the researcher and supervisors. After the research is complete, they will be stored in line with university policy.

A professional transcription service of accredited transcribers who are bound by a confidentiality agreement will be used. The audio recordings will be kept on secure, password protected servers until full data analysis is complete. This is to allow the researcher to listen back to parts of the interview and explore meaning which may be otherwise lost with the transcription for example tone of voice. Once full analysis is complete, the audio recordings will be destroyed.

Only members of the research team and responsible members of the University of Southampton may be given access to data about you for monitoring purposes and/or to carry out an audit of the study to ensure that the research is complying with applicable regulations. Individuals from regulatory authorities (people who check that we are carrying out the study correctly) may require access to your data. All of these people have a duty to keep your information, as a research participant, strictly confidential.

Once transcripts have been completed and identifiable information has been removed, you will be assigned a participant number which will be linked to all personal information. However, this study aims to gain detailed information about personal experiences of a rare disease. When this research is written up, it will include quotes form your interview which has the potential to make this person identifiable for example, if someone knew them and recognised their recollection of their experience. Therefore, it may not be possible to completely ensure that a person cannot be identified. You will be in control of what they choose to share including specific stories about your personal experience. This study will use an interpretive approach focussed on the meaning behind their experience. This may support the maintenance of confidentiality by not being focussed on description of events/experiences.

Do I have to take part?

No, it is entirely up to you to decide whether or not to take part. If you decide you want to take part, you will need to sign a consent form to show you have agreed to take part.

If you wish to take part in this research, please contact me on the details below.

What happens if I change my mind?

You have the right to change your mind and withdraw at any time without giving a reason and without your participant rights being affected.

You can withdraw at any point, up until the data is sent to be transcribed. You will be informed of date this is happening in advance. If you withdraw consent during the interview, the interview will be stopped, and the audio recording will be destroyed. If you have already participated in the interview and withdraw consent, your audio recording will be destroyed.

What will happen to the results of the research?

Your personal details will remain strictly confidential. Research findings made available in any reports or publications will not include information that can directly identify you without your specific consent.

It is hoped that the findings of this research will be published in a psychology journal for the purpose of sharing the findings with other healthcare professionals and researchers. This publication may include quotes of things you have said during the interview. Any details such as names/places will be removed so that you will not be directly identifiable. However, as this research explores personal accounts of experience of a rare disease which may mean you can be identified if others were aware of your experiences. This detail on personal experience is important to allow for a thorough and meaningful understanding.

Where can I get more information?

If you have any questions or would like more information about this study please contact the researcher, Georgia Steed, using the details below:

Email: g.steed@soton.ac.uk

Supervised by Dr Warren Dunger

Email: w.n.dunger@soton.ac.uk

What happens if there is a problem?

If you have a concern about any aspect of this study, you should speak to the researchers who will do their best to answer your questions.

If you remain unhappy or have a complaint about any aspect of this study, please contact the University of Southampton Research Integrity and Governance Manager (023 8059 5058, rgoinfo@soton.ac.uk).

Please quote the Ethics/ERGO number above. Please note that by making a complaint you might be no longer anonymous.

Data Protection Privacy Notice

The University of Southampton conducts research to the highest standards of research integrity. As a publicly-funded organisation, the University has to ensure that it is in the public interest when we use personally-identifiable information about people who have agreed to take part in research. This means that when you agree to take part in a research study, we will use information about you in the ways needed, and for the purposes specified, to conduct and complete the research project. Under data protection law, ‘Personal data’ means any information that relates to and is capable of identifying a living individual. The University’s data protection policy governing the use of personal data by the University can be found on its website (https://www.southampton.ac.uk/legalservices/what-we-do/data-protection-and-foi.page).

This Participant Information Sheet tells you what data will be collected for this project and whether this includes any personal data. Please ask the research team if you have any questions or are unclear what data is being collected about you.

Our privacy notice for research participants provides more information on how the University of Southampton collects and uses your personal data when you take part in one of our research projects and can be found at http://www.southampton.ac.uk/assets/sharepoint/intranet/ls/Public/Research%20and%20Integrity%20Privacy%20Notice/Privacy%20Notice%20for%20Research%20Participants.pdf

Any personal data we collect in this study will be used only for the purposes of carrying out our research and will be handled according to the University’s policies in line with data protection law. If any personal data is used from which you can be identified directly, it will not be disclosed to anyone else without your consent unless the University of Southampton is required by law to disclose it.

Data protection law requires us to have a valid legal reason (‘lawful basis’) to process and use your Personal data. The lawful basis for processing personal information in this research study is for the performance of a task carried out in the public interest. Personal data collected for research will not be used for any other purpose.

For the purposes of data protection law, the University of Southampton is the ‘Data Controller’ for this study, which means that we are responsible for looking after your information and using it properly. The University of Southampton will keep identifiable information about you for 10 years after the study has finished after which time any link between you and your information will be removed.

To safeguard your rights, we will use the minimum personal data necessary to achieve our research study objectives. Your data protection rights – such as to access, change, or transfer such information - may be limited, however, in order for the research output to be reliable and accurate. The University will not do anything with your personal data that you would not reasonably expect.

If you have any questions about how your personal data is used, or wish to exercise any of your rights, please consult the University’s data protection webpage (https://www.southampton.ac.uk/legalservices/what-we-do/data-protection-and-foi.page) where you can make a request using our online form. If you need further assistance, please contact the University’s Data Protection Officer (data.protection@soton.ac.uk).

A professional, accredited transcription service will be used. They will be bound by confidentiality and data management agreements.

It is not possible for data to be anonymised because of the nature of the personal accounts discussed in the interview. However, data will be pseudonymised using a code. This will be assigned to each participant at the start of data analysis. Only the researcher and supervisor at the University of Southampton will have access to these codes.

Thank you.

Thank the individual for taking the time to read the information sheet and considering taking part in the research.

Appendix M Consent From

Study title: Understanding how carers of people with Motor Neuron Disease experience palliative care in the UK.

Researcher name: Georgia Steed (Trainee Clinical Psychologist)

ERGO number: 702086, 27/05/22, Version 1

Please initial the box(es) if you agree with the statement(s):

I have read and understood the information sheet (27/05/22, Version 1) and have had the opportunity to ask questions about the study.	
I agree to take part in this research project and agree for my data to be used for the purpose of this study.	
I understand my participation is voluntary and I may withdraw at any point up until when data analysis begins for any reason without my participation rights being affected.	
I agree to take part in the interview for the purposes set out in the participation information sheet and understand that these will be recorded using audio.	

I understand that taking part in the study involves audio recording which will be transcribed and destroyed once data analysis is complete for the purposes set out in the participation information sheet.	
I agree for my interview to be sent to a professional, accredited transcription service	
I understand that I may be quoted directly in reports of the research but that I will not be directly identified (e.g. that my name will not be used).	
I understand that special category information will be collected about me including age, ethnicity, religion, gender identity and sexual orientation.	
I understand that special category information will be destroyed if I withdraw my consent.	

Name of participant (print name)……………………………………………………………………

Signature of participant……………………………………………………………………

Date………………………………………………………………
………

Name of researcher (print name)……………………………………………………………
……

Signature of researcher …………………………………………………………
………

Date………………………………………………………………
………

Appendix N Debrief Form

UNIVERSITY OF Southampton

Study Title: Understanding how carers of people with Motor Neuron Disease (MND) experience palliative care in the UK.

Debriefing Statement written

ERGO ID: 72086

Version 2, 27/06/22

The aim of this research was to develop a deep understanding of carers' experiences of palliative care in the UK. This will include understanding what people have been offered/received, how they make sense of their experience, understand any areas which may be missing, as well as what areas feel important and why this may be given the unique needs of this client group. Your data will help our understanding of your personal experience. This will allow us to draw themes with your experience and the experiences of other carers of people with MND. Once again results of this study will not include your name or any other identifying characteristics. The research did not use deception. You may have a copy of the summary of findings from this research once it is completed if you wish.

If you have any further questions, please contact me Georgia Steed at g.steed@soton.ac.uk.

You have the right to withdraw your data up until the date that interviews are sent to be transcribed. The deadline date to withdraw your data will be provided on the day of your interview. You will be sent an email with confirmation of this date.

Thank you for your participation in this research.

If you have questions about your rights as a participant in this research, or if you feel that you have been placed at risk, you may contact the University of Southampton Head of Research Integrity and Governance (023 8059 5058, rgoinfo@soton.ac.uk).

We understand that discussing such experiences can bring up lots of difficult emotions.

For specialist advice and support regarding MND please contact the MND association- 0808 802 6262

If you feel that your levels of distress require additional support after participating in this research, contact the following resources for support:

Your GP

Your local Community Mental Health Team or Crisis Resolution and Home Treatment Team (out of hours)

NHS 111

Samaritans – 116 123

References

Akiyama, A., Numata, K., & Mikami, H. (2010). Importance of end-of-life support to minimize caregiver's regret during bereavement of the elderly for better subsequent adaptation to bereavement. *Archives of Gerontology and Geriatrics*, *50*(2), 175-178.

Alonso, A., Logroscino, G., Jick, S. S., & Hernán, M. A. (2009). Incidence and lifetime risk of motor neuron disease in the United Kingdom: a population-based study. *European journal of neurology*, *16*(6), 745-751.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). https://doi.org/10.1176/appi.books.9780890425596

An, A. W., Ladwig, S., Epstein, R. M., Prigerson, H. G., & Duberstein, P. R. (2020). The impact of the caregiver-oncologist relationship on caregiver experiences of end-of-life care and bereavement outcomes. *Supportive Care in Cancer*, *28*(9), 4219-4225.

Anderson, N. H., Gluyas, C., Mathers, S., Hudson, P., & Ugalde, A. (2019). “A monster that lives in our lives”: experiences of caregivers of people with motor neuron disease and identifying avenues for support. *BMJ Supportive & Palliative Care*, *9*(4), e27-e27.

Aoun, S. M., Bentley, B., Funk, L., Toye, C., Grande, G., & Stajduhar, K. J. (2013). A 10-year literature review of family caregiving for motor neurone disease: moving from caregiver burden studies to palliative care interventions. *Palliative medicine*, *27*(5), 437-446.

Aoun, S. M., Connors, S. L., Priddis, L., Breen, L. J., & Colyer, S. (2012). Motor neurone disease family carers' experiences of caring, palliative care and bereavement: an exploratory qualitative study. *Palliative medicine*, *26*(6), 842-850.

Aoun, S. M., Rumbold, B., Howting, D., Bolleter, A., & Breen, L. J. (2017). Bereavement support for family caregivers: The gap between guidelines and practice in palliative care. *PloS one*, *12*(10), e0184750.

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

Averill, A. J., Kasarskis, E. J., & Segerstrom, S. C. (2007). Psychological health in patients with amyotrophic lateral sclerosis. *Amyotrophic Lateral Sclerosis*, *8*(4), 243-254.

Bakker C, de Vugt ME, Vernooij-Dassen M, *et al.* 2010. Needs in early onset dementia: a qualitative case from the needYD study. *Am J Alzheimers Dis Other Demen* **25**: 634–40.

Balintona, M. L. (2018). " It Gets Easier, Not Better": Ambiguous Loss and Resiliency in Adult Children Caring for Their Parent with Alzheimer's Disease.

Ball, L. J., Beukelman, D. R., & Pattee, G. L. (2004). Communication effectiveness of individuals with amyotrophic lateral sclerosis. *Journal of Communication Disorders*, *37*(3), 197-215.

Bentley, B., & O'Connor, M. (2016). The end-of-life experiences of people with motor neuron disease: family carers' perspectives. *Journal of Palliative Medicine*, *19*(8), 857-862.

Bentley, B., & O'Connor, M. (2016). The end-of-life experiences of people with motor neuron disease: family carers' perspectives. *Journal of Palliative Medicine*, *19*(8), 857-862.

Bidwell, J. T., Lyons, K. S., & Lee, C. S. (2017). Caregiver well-being and patient outcomes in heart failure: a meta-analysis. *The Journal of cardiovascular nursing*, *32*(4), 372.

Borgstrom, E. (2020). What is a good death? A critical discourse policy analysis. *BMJ Supportive & Palliative Care*.

Bressan, V., Visintini, C., & Palese, A. (2020). What do family caregivers of people with dementia need? A mixed-method systematic review. *Health & social care in the community*, *28*(6), 1942-1960.

Brighton, L. J., Selman, L. E., Bristowe, K., Edwards, B., Koffman, J., & Evans, C. J. (2019). Emotional labour in palliative and end-of-life care communication: A qualitative study with generalist palliative care providers. *Patient education and counseling*, *102*(3), 494-502.

Burgdorf, J. G., Wolff, J. L., Chase, J. A., & Arbaje, A. I. (2022). Barriers and facilitators to family caregiver training during home health care: A multisite qualitative analysis. Journal of the American Geriatrics Society, 70(5), 1325–1335. doi:10.1111/ jgs.17762

Bussè, C., Barnini, T., Zucca, M., Rainero, I., Mozzetta, S., Zangrossi, A., & Cagnin, A. (2022). Depression, anxiety and sleep alterations in caregivers of persons with dementia after 1-year of COVID-19 pandemic. *Frontiers in Psychiatry*, *13*, 826371.

Bužgová, R., Kozáková, R., & Bar, M. (2020). The effect of neuropalliative care on quality of life and satisfaction with quality of care in patients with progressive neurological disease and their family caregivers: an interventional control study. *BMC Palliative Care*, *19*(1), 1-10.

Carduff, E., Jarvis, A., Highet, G., Finucane, A., Kendall, M., Harrison, N., ... & Murray, S. A. (2016). Piloting a new approach in primary care to identify, assess and support carers of people with terminal illnesses: a feasibility study. *BMC Family Practice*, *17*(1), 1-9.

Carers UK. (2018). State of caring report. 2018.

Carers UK. (2021). State of Caring Report 2021.

Carvalho, T. L., Almeida, L. M. S. D., Lorega, C. M. A., Barata, M. F. O., Ferreira, M. L. B., Brito-Marques, P. R. D., & Correia, C. D.

C. (2016). Depression and anxiety in individuals with amyotrophic lateral sclerosis: a systematic review. *Trends in psychiatry and psychotherapy*, *38*, 01-05.

Chang JL, Lomen-Hoerth C, Murphy J, Henry RG, Kramer JH, Miller BL, Gorno-Tempini ML (2005): A voxel-based morphometry study of patterns of brain atrophy in ALS and ALS/ FTLD. Neurology 65:75–80.

Cheng, H. W. B., Chan, O. M. I., Chan, C. H. R., Chan, W. H., Fung, K. S., & Wong, K. Y. (2018). End-of-life characteristics and palliative care provision for patients with motor neuron disease. *American Journal of Hospice and Palliative Medicine®*, *35*(6), 847-851.

Chi, N. C., Demiris, G., Lewis, F. M., Walker, A. J., & Langer, S. L. (2016). Behavioral and educational interventions to support family caregivers in end-of-life care: a systematic review. *American Journal of Hospice and Palliative Medicine®*, *33*(9), 894-908.

Coyle, N. (2006). The hard work of living in the face of death. *Journal of pain and symptom management*, *32*(3), 266-274.

Cui, B., Cui, L. Y., Liu, M. S., Li, X. G., Ma, J. F., Fang, J., & Ding, Q. Y. (2015). Behavioral symptoms in motor neuron disease and their negative impact on caregiver burden. *Chinese Medical Journal*, *128*(17), 2295-2300.

Davis, E. L., Deane, F. P., & Lyons, G. C. (2015). Acceptance and valued living as critical appraisal and coping strengths for caregivers dealing with terminal illness and bereavement. *Palliative & Supportive Care*, *13*(2), 359-368.

del-Pino-Casado, R., Rodriguez Cardosa, M., López-Martínez, C., & Orgeta, V. (2019). The association between subjective caregiver burden and depressive symptoms in carers of older relatives: A systematic review and meta-analysis. *PloS one*, *14*(5), e0217648.

De Stefano, M., Esposito, S., Iavarone, A., Carpinelli Mazzi, M., Siciliano, M., Buonanno, D., ... & Tedeschi, G. (2022). Effects of Phone-Based Psychological Intervention on Caregivers of Patients with Early-Onset Alzheimer's Disease: A Six-Months Study during the COVID-19 Emergency in Italy. *Brain sciences*, *12*(3), 310.

de Wit, J., Bakker, L. A., van Groenestijn, A. C., van den Berg, L. H., Schröder, C. D., Visser-Meily, J. M., & Beelen, A. (2018). Caregiver burden in amyotrophic lateral sclerosis: a systematic review. *Palliative medicine*, *32*(1), 231-245.

de Wit, J., Beelen, A., van den Heerik, M. S., van den Berg, L. H., Visser-Meily, J. M., & Schröder, C. D. (2020). Psychological distress in partners of patients with amyotrophic lateral sclerosis and progressive muscular atrophy: what's the role of care demands and perceived control?. *Psychology, Health & Medicine*, *25*(3), 319-330.

Department of Health. (2014). *Carers strategy: Second National Action Plan 2014–2016*. London: Department of Health.

Department of Health. *End of Life Care Strategy: promoting high quality care for all adults at the end of life*. London: Department of Health, 2008.

Diggory, K., & Reeves, A. (2022). ‘Permission to be kind to myself’. The experiences of informal carers of those with a life-limiting or terminal illness of a brief self-compassion-based self-care intervention. *Progress in Palliative Care*, *30*(3), 149-157.

Doris, S. F., Cheng, S. T., & Wang, J. (2018). Unravelling positive aspects of caregiving in dementia: An integrative review of research literature. *International Journal of Nursing Studies*, *79*, 1-26.

Doris, S. F., Cheng, S. T., & Wang, J. (2018). Unravelling positive aspects of caregiving in dementia: An integrative review of research literature. *International Journal of Nursing Studies*, *79*, 1-26

Ducharme, F., Kergoat, M. J., Coulombe, R., Lévesque, L., Antoine, P., & Pasquier, F. (2014). Unmet support needs of early-onset dementia family caregivers: a mixed-design study. *BMC nursing*, *13*(1), 1-10.

Eatough, V., & Smith, J. A. (2008). Interpretative phenomenological analysis. *The Sage handbook of qualitative research in psychology*, *179*, 194.

Eatough, V., & Smith, J. A. (2017). Interpretative phenomenological analysis. *The Sage handbook of qualitative research in psychology*, 193-209.

Edge, R., Mills, R., Tennant, A., Diggle, P. J., & Young, C. A. (2020). Do pain, anxiety and depression influence quality of life for people with amyotrophic lateral sclerosis/motor neuron disease? A national study reconciling previous conflicting literature. *Journal of neurology*, *267*(3), 607-615.

Eifert, E. K., Adams, R., Dudley, W., & Perko, M. (2015). Family caregiver identity: A literature review. *American Journal of Health Education*, *46*(6), 357-367.

Ewing, G., Croke, S., Rowland, C., & Grande, G. (2020). Suitability and acceptability of the Carer Support Needs Assessment Tool (CSNAT) for the assessment of carers of people with MND: a qualitative study. *BMJ open*, *10*(12), e039031.

Eyre, V. E. M. (2019). *How to support caregivers and the bereaved: Psychoeducation and personal coping techniques* (Doctoral dissertation, The University of Western Ontario (Canada)).

Farias, M., Maraldi, E., Wallenkampf, K. C., & Lucchetti, G. (2020). Adverse events in meditation practices and meditation-based therapies: a systematic review. *Acta Psychiatrica Scandinavica*, *142*(5), 374-393.

Flemming, K., Turner, V., Bolsher, S., Hulme, B., McHugh, E., & Watt, I. (2020). The experiences of, and need for, palliative care for people with motor neurone disease and their informal caregivers: a qualitative systematic review. *Palliative medicine*, *34*(6), 708-730.

Flemming, K., Turner, V., Bolsher, S., Hulme, B., McHugh, E., & Watt, I. (2020). The experiences of, and need for, palliative care for people with motor neurone disease and their informal caregivers: a qualitative systematic review. *Palliative medicine*, *34*(6), 708-730.

Flemming, K., Turner, V., Watt, I., Hulme, B., & Bolsher, S. (2018). The Palliative Care Needs of Adult Patients and Their Informal Carers Living with Motor Neuron Disease (MND), or Bereaved Carers of People with MND: a Systematic Review of Qualitative Research. *Journal of Pain and Symptom Management*, *56*(6), e39-e40.

Foley, G., Timonen, V., & Hardiman, O. (2014). Understanding psycho-social processes underpinning engagement with services in motor neurone disease: a qualitative study. *Palliative Medicine*, *28*(4), 318-325.

Formagini, T., Poague, C., O'Neal, A., & Brooks, J. V. (2022). "When I Heard the Word Palliative": Obscuring and Clarifying Factors Affecting the Stigma Around Palliative Care Referral in Oncology. *JCO Oncology Practice*, *18*(1), e72-e79.

Galvin, M., Corr, B., Madden, C., Mays, I., McQuillan, R., Timonen, V., ... & Hardiman, O. (2016). Caregiving in ALS–a mixed methods approach to the study of burden. *BMC palliative care*, *15*(1), 1-12.

Galvin, M., Corr, B., Madden, C., Mays, I., McQuillan, R., Timonen, V., ... & Hardiman, O. (2016). Caregiving in ALS–a mixed methods

approach to the study of burden. *BMC palliative care*, *15*(1), 1-12.

Gaugler, J. E., Yu, F., Krichbaum, K., & Wyman, J. F. (2009). Predictors of nursing home admission for persons with dementia. *Medical Care*, *47*(2), 191–198.

Geng, H. M., Chuang, D. M., Yang, F., Yang, Y., Liu, W. M., Liu, L. H., & Tian, H. M. (2018). Prevalence and determinants of depression in caregivers of cancer patients: A systematic review and meta-analysis. *Medicine*, *97*(39).

Gérain, P., & Zech, E. (2019). Informal caregiver burnout? Development of a theoretical framework to understand the impact of caregiving. *Frontiers in psychology*, *10*, 466359

Gerhart, J., O'Mahony, S., Abrams, I., Grosse, J., Greene, M., & Levy, M. (2016). A pilot test of a mindfulness-based communication training to enhance resilience in palliative care professionals. *Journal of Contextual Behavioral Science*, *5*(2), 89-96.

Given, B. A., Given, C. W., & Sherwood, P. (2012, November). The challenge of quality cancer care for family caregivers. In *Seminars in oncology nursing* (Vol. 28, No. 4, pp. 205-212). WB Saunders.

Götze, H., Brähler, E., Gansera, L., Polze, N., & Köhler, N. (2014). Psychological distress and quality of life of palliative cancer patients and their caring relatives during home care. *Supportive Care in Cancer*, *22*(10), 2775-2782.

Govina, O., Vlachou, E., Kalemikerakis, I., Papageorgiou, D., Kavga, A., & Konstantinidis, T. (2019). Factors associated with anxiety and depression among family caregivers of patients undergoing palliative radiotherapy. *Asia-Pacific Journal of Oncology Nursing*, *6*(3), 283-291.

Graham-Wisener, L., Nelson, A., Byrne, A., Islam, I., Harrison, C., Geddis, J., & Berry, E. (2022). Understanding public attitudes to death talk and advance care planning in Northern Ireland using health behaviour change theory: a qualitative study. *BMC Public Health*, *22*(1), 1-19.

Grande, G., Rowland, C., Cotterill, S., Batistatou, E., & Hanratty, B. (2021). Factors associated with carer psychological and physical health during end-of-life caregiving: an observational analysis of a population-based post-bereavement survey of carers of people with cancer. *BMJ open*, *11*(10), e047275.

Guerriere, D., Husain, A., Zagorski, B., Marshall, D., Seow, H., Brazil, K., ... & Coyte, P. C. (2016). Predictors of caregiver burden across the home-based palliative care trajectory in O ntario, C anada. *Health & Social Care in the Community*, *24*(4), 428-438.

Guideline NG142, N. I. C. E. (2019). End of life care for adults: service delivery. *Methods*.

Harris, M., Thomas, G., Thomas, M., Cafarella, P., Stocks, A., Greig, J., & McEvoy, R. D. (2018). Supporting wellbeing in motor neurone disease for patients, carers, social networks, and health professionals: A scoping review and synthesis. *Palliative & supportive care*, *16*(2), 228-237.

Harrop, E., Morgan, F., Byrne, A., & Nelson, A. (2016). “It still haunts me whether we did the right thing”: a qualitative analysis of free text survey data on the bereavement experiences and support needs of family caregivers. *BMC palliative care*, *15*(1), 1-8.

Harwood, C. A., McDermott, C. J., & Shaw, P. J. (2009). Physical activity as an exogenous risk factor in motor neuron disease (MND): a review of the evidence. *Amyotrophic Lateral Sclerosis*, *10*(4), 191-204.

Hennings, J., & Froggatt, K. (2019). The experiences of family caregivers of people with advanced dementia living in nursing homes, with a specific focus on spouses: A narrative literature review. *Dementia*, *18*(1), 303-322.

Henriksson, A., & Årestedt, K. (2013). Exploring factors and caregiver outcomes associated with feelings of preparedness for caregiving in family caregivers in palliative care: a correlational, cross-sectional study. *Palliative medicine*, *27*(7), 639-646.

Hiyoshi-Taniguchi, K., Becker, C. B., & Kinoshita, A. (2018). What behavioral and psychological symptoms of dementia affect caregiver burnout?. *Clinical gerontologist*, *41*(3), 249-254.

Holland- Seeber, A. A., Pols, A. J., Hijdra, A., Grupstra, H. F., Willems, D. L., & de Visser, M. (2019). Advance care planning in progressive neurological diseases: lessons from ALS. *BMC palliative care*, *18*, 1-10. futher research needed to explore the imlimentation of this approach in the UK.

Hsieh, S., Leyton, C. E., Caga, J., Flanagan, E., Kaizik, C., O'Connor, C. M., ... & Mioshi, E. (2016). The evolution of caregiver burden in frontotemporal dementia with and without amyotrophic lateral sclerosis. *Journal of Alzheimer's Disease*, *49*(3), 875-885.

Hudson, P., Remedios, C., Zordan, R., Thomas, K., Clifton, D., Crewdson, M., ... & Bauld, C. (2012). Guidelines for the psychosocial and bereavement support of family caregivers of palliative care patients. *Journal of palliative medicine*, *15*(6), 696-702.

Isik, A. T., Soysal, P., Solmi, M., & Veronese, N. (2019). Bidirectional relationship between caregiver burden and neuropsychiatric symptoms in patients with Alzheimer's disease: A narrative review. *International journal of geriatric psychiatry*, *34*(9), 1326-1334.

Jeyathevan, G., Catharine Craven, B., Cameron, J. I., & Jaglal, S. B. (2020). Facilitators and barriers to supporting individuals with spinal cord injury in the community: experiences of family caregivers and care recipients. *Disability and rehabilitation*, *42*(13), 1844-1854.

Johnson, C. C., Suchyta, M. R., Darowski, E. S., Collar, E. M., Kiehl, A. L., Van, J., ... & Hopkins, R. O. (2019). Psychological sequelae in family caregivers of critically Ill intensive care unit patients. A systematic review. *Annals of the American Thoracic Society*, *16*(7), 894-909

Judd, R. (2015). *Meaning in palliative care: A literature review of psychological interventions and study of men's experience of caring* (Doctoral dissertation, University of Birmingham).

Lang G (1996) Motor neurone disease: mortality statistics in Australia 1986-94. Motor Neurone Disease Association of Victoria Inc., Caulfield South, Victoria.

Larkin, M., Flowers, P., & Smith, J. A. (2021). Interpretative phenomenological analysis: Theory, method and research. *Interpretative phenomenological analysis Second Edition*, 1-100.

Larkin, M., Shaw, R., & Flowers, P. (2019). Multiperspectival designs and processes in interpretative phenomenological analysis research. *Qualitative Research in Psychology*, *16*(2), 182-198.

Larson, D. G., & Hoyt, W. T. (2009). Grief counselling efficacy: What have we learned?. *Bereavement Care*, *28*(3), 14-19.

Leigh, P. N., & Ray-Chaudhuri, K. (1994). Motor neuron disease. *Journal of neurology, neurosurgery, and psychiatry*, *57*(8), 886.

Lethin, C., Leino-Kilpi, H., Bleijlevens, M. H., Stephan, A., Martin, M. S., Nilsson, K., ... & Karlsson, S. (2020). Predicting caregiver burden in informal caregivers caring for persons with dementia living at home–A follow-up cohort study. *Dementia*, *19*(3), 640-660.

Lewis-Morton, R., James, L., Brown, K., & Hider, A. (2015, November). Team formulation in a secure setting: Challenges, rewards and service user involvement–A joint collaboration between nursing and psychology. In *Clinical Psychology Forum* (Vol. 275, p. 65).

Litzelman, K., Kent, E. E., Mollica, M., & Rowland, J. H. (2016). How does caregiver well-being relate to perceived quality of care in patients with cancer? Exploring associations and pathways. *Journal of Clinical Oncology*, *34*(29), 3554.

Liu, Z., Heffernan, C., & Tan, J. (2020). Caregiver burden: A concept analysis. *International journal of nursing sciences*, *7*(4), 438-445.

Livermore, S. D. (2014). *Delirium rate and risk factors in palliative care outpatients and caregiver coping strategies*. University of San Diego.

Luce, C. D. (2020). *Exploring Adult Child Carers Existential Experience Caregiving Dying Parents* (Doctoral dissertation, Capella University).

Luk, T. C. (2021). *A Prospective Study on Grief-Related Cognitions and Grief Symptoms in the Caregivers of Terminal Cancer Patients* (Doctoral dissertation, The Chinese University of Hong Kong (Hong Kong)).

Luth, E. A., & Pristavec, T. (2020). Do caregiver experiences shape end-of-life care perceptions? Burden, benefits, and care quality assessment. *Journal of pain and symptom management*, *59*(1), 77-85.

Lynn, J., Harrell Jr, F. E., Cohn, F., & Hamel, M. B. (1996). Defining the terminally ill: insights from support. *Duq. L. Rev.*, *35*, 311, pg 307.

Macchi, Z. A., Koljack, C. E., Miyasaki, J. M., Katz, M., Galifianakis, N., Prizer, L. P., ... & Kluger, B. M. (2020). Patient and caregiver characteristics associated with caregiver burden in Parkinson's disease: A palliative care approach. *Ann Palliat Med*, *9*(Suppl 1), S24-33.

Marie Curie (2022). What is terminal illness? *Marie Curie*. Accesses 25/01/23. https://www.mariecurie.org.uk/who/terminal-illness-definition

Mason, T. M., Tofthagen, C. S., & Buck, H. G. (2020). Complicated grief: risk factors, protective factors, and interventions. *Journal of social work in end-of-life & palliative care*, *16*(2), 151-174.

McCabe, M. P., Firth, L., & O'Connor, E. (2009). A comparison of mood and quality of life among people with progressive neurological illnesses and their caregivers. *Journal of clinical psychology in medical settings*, *16*, 355-362.

McNamara, B., & Rosenwax, L. (2010). Which carers of family members at the end of life need more support from health services and why?. *Social science & medicine*, *70*(7), 1035-1041.

Meier, E. A., Gallegos, J. V., Thomas, L. P. M., Depp, C. A., Irwin, S. A., & Jeste, D. V. (2016). Defining a good death (successful dying): literature review and a call for research and public dialogue. *The American Journal of Geriatric Psychiatry*, *24*(4), 261-271.

Midtbust, M. H., Alnes, R. E., Gjengedal, E., & Lykkeslet, E. (2021). Separation characterized by responsibility and guilt: Family caregivers' experiences with palliative care for a close family member with severe dementia in long-term care facilities. *Dementia*, *20*(2), 518-533.

Miller, R. G., Mitchell, J. D., & Moore, D. H. (2012). Riluzole for amyotrophic lateral sclerosis (ALS)/motor neuron disease (MND). *Cochrane database of systematic reviews*, (3).

Mirsoleymani, S., Matbouei, M., Vasli, P., Marzaleh, M. A., & Rohani, C. (2021). The role of family caregiver's sense of coherence and family adaptation determinants in predicting distress and caregiver burden in families of cancer patients. *Indian Journal of Palliative Care*, *27*(1), 47.

Motor Neurone Disease Association. (2014). *An Introduction to Motor Neuron Disease (MND).* [Brochure]. MND Association. https://www.mndassociation.org/app/uploads/2019/01/Introduction-to-MND.pdf

Nielsen, M. K., Neergaard, M. A., Jensen, A. B., Bro, F., & Guldin, M. B. (2016). Psychological distress, health, and socio-economic factors in caregivers of terminally ill patients: a nationwide population-based cohort study. *Supportive Care in Cancer*, *24*(7), 3057-3067.

Nielsen, M. K., Neergaard, M. A., Jensen, A. B., Vedsted, P., Bro, F., & Guldin, M. B. (2017). Preloss grief in family caregivers during end-of-life cancer care: A nationwide population-based cohort study. *Psycho‐Oncology*, *26*(12), 2048-2056.

Nipp, R. D., El-Jawahri, A., Fishbein, J. N., Gallagher, E. R., Stagl, J. M., Park, E. R., ... & Temel, J. S. (2016). Factors associated with depression and anxiety symptoms in family caregivers of patients with incurable cancer. *Annals of Oncology*, *27*(8), 1607-1612.

Nogueira, M. M. L., Neto, J. P. S., Sousa, M. F. B., Santos, R. L., Rosa, R. D., Belfort, T., ... & Dourado, M. C. N. (2015). Spouse-caregivers’ quality of life in Alzheimer's disease. *International Psychogeriatrics*, *27*(5), 837-845.

Norinder, M., Årestedt, K., Lind, S., Axelsson, L., Grande, G., Ewing, G., ... & Alvariza, A. (2021). Higher levels of unmet support needs in spouses are associated with poorer quality of life–a descriptive cross-sectional study in the context of palliative home care. *BMC palliative care*, *20*(1), 1-11.

O’Brien, M. R., Whitehead, B., Jack, B. A., & Mitchell, J. D. (2012). The need for support services for family carers of people with motor neurone disease (MND): views of current and former family caregivers a qualitative study. *Disability and rehabilitation*, *34*(3), 247-256.

Oechsle, K., Ullrich, A., Marx, G., Benze, G., Wowretzko, F., Zhang, Y., ... & Bergelt, C. (2020). Prevalence and predictors of distress, anxiety, depression, and quality of life in bereaved family caregivers of patients with advanced cancer. *American Journal of Hospice and Palliative Medicine®*, *37*(3), 201-213.

Oh, J., & Kim, J. A. (2017). Supportive care needs of patients with amyotrophic lateral sclerosis/motor neuron disease and their caregivers: A scoping review. *Journal of clinical nursing*, *26*(23-24), 4129-4152.

Osse, B. H., Vernooij-Dassen, M. J., Schadé, E., & Grol, R. P. (2006). Problems experienced by the informal caregivers of cancer patients and their needs for support. *Cancer nursing*, *29*(5), 378-388.

Oudman, E., & Baert, J. C. (2022). Eye movement desensitization and reprocessing (EMDR) and mediative behavioral therapy for the treatment of suffocation related post-traumatic stress disorder (PTSD) in amyotrophic lateral sclerosis (ALS): A case report. *Palliative & Supportive Care*, 1-3.

Outram, S., Harris, G., Kelly, B., Bylund, C. L., Cohen, M., Landa, Y., ... & Loughland, C. (2015). ‘We didn’t have a clue’: family caregivers’ experiences of the communication of a diagnosis of schizophrenia. *International Journal of Social Psychiatry*, *61*(1), 10-16.

Pakenham, K. I., & Samios, C. (2013). Couples coping with multiple sclerosis: A dyadic perspective on the roles of mindfulness and acceptance. *Journal of behavioral medicine*, *36*(4), 389-400.

Pavey, A., Warren, N., & Allen-Collinson, J. (2015). “It gives me my freedom”: technology and responding to bodily limitations in motor neuron disease. *Medical Anthropology*, *34*(5), 442-455.

Penning, M. J., & Wu, Z. (2016). Caregiver stress and mental health: Impact of caregiving relationship and gender. *The gerontologist*, *56*(6), 1102-1113.

Peters, M., Fitzpatrick, R., Doll, H., Playford, E. D., & Jenkinson, C. (2012). The impact of perceived lack of support provided by health and social care services to caregivers of people with motor neuron disease. *Amyotrophic Lateral Sclerosis*, *13*(2), 223-228.

Petruzzo, A., Biagioli, V., Durante, A., Gialloreti, L. E., D’Agostino, F., Alvaro, R., & Vellone, E. (2019). Influence of preparedness on anxiety, depression, and quality of life in caregivers of heart failure patients: testing a model of path analysis. *Patient education and counseling*, *102*(5), 1021-1028.

Pop, R. S., Puia, A., & Mosoiu, D. (2022). Factors Influencing the Quality of Life of the Primary Caregiver of a Palliative Patient: Narrative Review. *Journal of palliative medicine*, *25*(5), 813-829.

Popay, J., Roberts, H., Sowden, A., Petticrew, M., Arai, L., Rodgers, M., Britten N., with Roen, K. & Duffy, S. (2006) *Guidance on the Conduct of Narrative Synthesis in Systematic Reviews*. A Product from the ESRC Methods Programme.

Poppe, C., Koné, I., Iseli, L. M., Schweikert, K., Elger, B. S., & Wangmo, T. (2020). Differentiating needs of informal caregivers of individuals with ALS across the caregiving course: a systematic review. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration*, *21*(7-8), 519-541.

Preston, T., Carr, D. C., Hajcak, G., Sheffler, J., & Sachs-Ericsson, N. (2022). Cognitive reappraisal, emotional suppression, and depressive and anxiety symptoms in later life: The moderating role of gender. *Aging & mental health*, *26*(12), 2390-2398.

Radbruch L, Payne S. (2010). White paper on standards and norms for hospice and palliative care in Europe: part 1. Recommendations from the European Association for Palliative Care. Eur J Palliat Care. 17:22–33.

Radunovic, A., Annane, D., Rafiq, M. K., Brassington, R., & Mustfa, N. (2017). Mechanical ventilation for amyotrophic lateral sclerosis/motor neuron disease. *Cochrane Database of Systematic Reviews*, (10).

Ray, R. A., Brown, J., & Street, A. F. (2014). Dying with motor neurone disease, what can we learn from family caregivers?. *Health Expectations*, *17*(4), 466-476.

Riepenhausen, A., Wackerhagen, C., Reppmann, Z. C., Deter, H. C., Kalisch, R., Veer, I. M., & Walter, H. (2022). Positive cognitive reappraisal in stress resilience, mental health, and well-being: A comprehensive systematic review. *Emotion Review*, *14*(4), 310-331.

Rose, L., Thaventhiran, T., Hobson, E., Rogers, R., James, K., Chu, P., ... & Ramsay, M. (2023). Digital peer-to-peer support programme for informal caregivers of people living with motor neuron disease: study protocol for a multi-centre parallel group, single blinded (outcome assessor) randomised controlled superiority trial.

Sarmento, V. P., Gysels, M., Higginson, I. J., & Gomes, B. (2017). Home palliative care works: but how? A meta-ethnography of the experiences of patients and family caregivers. *BMJ supportive & palliative care*, *7*(4), 00-00.

Schildmann, E. K., & Higginson, I. J. (2011). Evaluating psycho-educational interventions for informal carers of patients receiving cancer care or palliative care: strengths and limitations of different study designs. *Palliative medicine*, *25*(4), 345-356.

Schulz, R., and Tompkins, C. A. (2010). Informal caregivers in the United States: Prevalence, caregiver characteristics and ability to provide care in *The role of huma factors in home health care: Workshop summary.* Ed. (Washington, DC: National Academies Press), 322

Senger, K., Heider, J., Montini, R., & Schröder, A. (2017). Does Expectancy and Homework Compliance Predict Change in CBT. *Acta Psychopathol*, *3*(S2), 61.

Stavroulakis, T., Baird, W. O., Baxter, S. K., Walsh, T., Shaw, P. J., & McDermott, C. J. (2016). The impact of gastrostomy in motor neurone disease: Challenges and benefits from a patient and carer perspective. *BMJ Supportive and Palliative Care*, **6**(1), 52– 59. https://doi.org/10.1136/bmjspcare-2013-000609

Strommen, J., Fuller, H., Sanders, G. F., & Elliott, D. M. (2020). Challenges faced by family caregivers: Multiple perspectives on eldercare. *Journal of Applied Gerontology*, *39*(4), 347-356.

Sverre, K. T., Nissen, E. R., Farver-Vestergaard, I., Johannesen, M., & Zachariae, R. (2022). Comparing the efficacy of mindfulness-based therapy and cognitive-behavioral therapy for depression in head-to-head randomized controlled trials: A systematic review and meta-analysis of equivalence. *Clinical Psychology Review*, 102234.

Tatangelo, G., McCabe, M., Macleod, A., & You, E. (2018). “I just don’t focus on my needs.” The unmet health needs of partner and offspring caregivers of people with dementia: A qualitative study. *International Journal of Nursing Studies*, *77*, 8-14

van den Kieboom, R., Snaphaan, L., Mark, R., & Bongers, I. (2020). The trajectory of caregiver burden and risk factors in dementia progression: A systematic review. *Journal of Alzheimer's Disease*, *77*(3), 1107-1115.

van den Kieboom, R., Snaphaan, L., Mark, R., & Bongers, I. (2020). The trajectory of caregiver burden and risk factors in dementia progression: A systematic review. *Journal of Alzheimer's Disease*, *77*(3), 1107-1115.

Vasta, R., Chia, R., Traynor, B. J., & Chio, A. (2022). Unraveling the complex interplay between genes, environment, and climate in ALS. *EBioMedicine*, *75*, 103795.

Veronese, S., Gallo, G., Valle, A., Cugno, C., Chiò, A., Calvo, A., ... & Oliver, D. J. (2015). The palliative care needs of people severely affected by neurodegenerative disorders: a qualitative study. *Progress in Palliative Care*, *23*(6), 331-342.

von Blanckenburg, P., & Leppin, N. (2018). Psychological interventions in palliative care. *Current opinion in psychiatry*, *31*(5), 389-395.

Wang, H., O'Reilly, É. J., Weisskopf, M. G., Logroscino, G., McCullough, M. L., Thun, M. J., ... & Ascherio, A. (2011). Smoking and risk of amyotrophic lateral sclerosis: a pooled analysis of 5 prospective cohorts. *Archives of neurology*, *68*(2), 207-213.

Wang, X. R., Liu, S. X., Robinson, K. M., Shawler, C., & Zhou, L. (2019). The impact of dementia caregiving on self-care management of caregivers and facilitators: a qualitative study. *Psychogeriatrics*, *19*(1), 23-31.

Wang, Y., Abbott, A., & Butcher, C. (2017). Managing Eldercare in the United Kingdom and China. In *Managing the Ageing Workforce in the East and the West* (pp. 207-224). Emerald Publishing Limited.

Wen, F. H., Chen, J. S., Chou, W. C., Chang, W. C., Shen, W. C., Hsieh, C. H., & Tang, S. T. (2019). Family caregivers' subjective caregiving burden, quality of life, and depressive symptoms are associated with terminally ill cancer patients' distinct patterns of conjoint symptom distress and functional impairment in their last six months of life. *Journal of pain and symptom management*, *57*(1), 64-72.

Wicks, P., Abrahams, S., Masi, D., Hejda-Forde, S., Leigh, P. N., & Goldstein, L. H. (2007). Prevalence of depression in a 12-month consecutive sample of patients with ALS. *European journal of neurology*, *14*(9), 993-1001.

Williams, K., Hartley, S., Langer, S., Manandhar-Richardson, M., Sinha, M., & Taylor, P. (2022). A systematic review and meta-ethnographic synthesis of Mindfulness-based Cognitive Therapy for people with major depression. *Clinical Psychology & Psychotherapy*, *29*(5), 1494-1514.

Xiong, C., Biscardi, M., Astell, A., Nalder, E., Cameron, J. I., Mihailidis, A., & Colantonio, A. (2020). Sex and gender differences in caregiving burden experienced by family caregivers of persons with dementia: A systematic review. *PLoS One*, *15*(4), e0231848.

Yardley, L. (2000). "Dilemmas in qualitative health research." Psychology and Health. 15. pp. 215-228.

Young, C., Ealing, J., McDermott, C., Williams, T., Al-Chalabi, A., Majeed, T., ... & Tennant, A. (2022). Fatigue and anxiety mediate the effect of dyspnea on quality of life in amyotrophic lateral sclerosis. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration*, *23*(5-6), 390-398.

Zarotti N, Coates E, McGeachan A, Williams I, Beever D, Hackney G, HighCALS Study Group, et al. (2019) Health care professionals' views on psychological factors affecting nutritional behaviour in people with motor neuron disease: a thematic analysis. Br J Health Psychol;24:953–69. Available at: https://search.ebscohost.com/login.aspx?direct=true&db=psyh&AN=2019-51081-001&authtype=sso&custid=s8993828&site=ehost-live

Zyada, F., El Degwi, H., & Saad, R. (2013). Anxiety, depression, and strain among caregivers of terminally ill patients. *Egyptian Journal of Psychiatry*, *34*(2), 77.