

Interventions targeting psychological well-being for motor neuron disease carers: A systematic review

Review Article

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Abstract

Objectives. This systematic review considers interventions designed to improve the psychological well-being (PWB) of carers of people with motor neuron disease (MND) using quantitative, qualitative, or mixed-methods studies, and aimed to (1) summarize current research, (2) assess the quality of evidence, and (3) evaluate the effectiveness of interventions.

Method. Mixed-methods systematic review (MMSR) was conducted based on Joanna Briggs Institute methodology for quantitative, qualitative, and mixed-methods reviews and Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines.

Results. Thirteen papers met the inclusion criteria, including 12 studies (six mixed-methods, four quantitative, and two qualitative). Four studies described randomized controlled trials, seven detailed uncontrolled longitudinal studies with a single treatment group and a pre-post design, and one was an observational survey. Critical appraisal of the studies revealed a wide range of weaknesses in the quantitative and/or qualitative methodologies. Due to the heterogeneity of interventions, outcomes, and measurements, a narrative and convergent approach to data synthesis was employed. While a minority of studies demonstrated some benefits to hedonic and eudaimonic aspects of PWB, the interpretability of these data was limited by methodological problems.

Significance of results. This MMSR highlighted a paucity of quality research regarding interventions for the PWB of MND carers. Although some benefits to PWB were demonstrated, most studies suffered from substantial methodological problems, rendering the overall evidence base low. High-quality and carefully designed studies are a priority to enable effective development and testing of much-needed interventions targeting the PWB for MND carers.

The psychological toll of caring for a person with motor neuron disease (MND) is well established (Harris et al., 2018) with the experience described as unrelenting (Aoun et al., 2012). Carers may assist the individual with MND in managing physical, psychological, and neuro-behavioral changes (de Almeida et al., 2021). Most people with MND live at home where their physical and psychological functioning can be significantly influenced by the support received in that environment. Consequently, carer psychological well-being (PWB) can be salient for the functioning of MND patients. Due to the demands of their role, numerous studies consistently demonstrate that MND carers experience substantial burden, depression, anxiety, poor quality of life, strain, fatigue, and reduced social contacts (Gluyas et al., 2017; Harris et al., 2018).

There is increasing recognition that the PWB of carers of people with MND needs to be addressed. Descriptive studies have dominated MND carer research, with a recent increase in research directly focusing on interventions for MND carers (Gluyas et al., 2017; Harris et al., 2018). While the focus has turned to carer PWB, the concept itself has not been well defined.

Psychological well-being

The term “psychological well-being” has evolved and is increasingly used as an outcome measure in health intervention studies. However, conceptualizing and defining the term, as well as providing a rigorous approach to research, has proven to be challenging (Dodge et al., 2012). This is an evolving theoretical and research domain in psychology, with open debate about the merits of the superiority of the concurrent hedonic, eudaimonic, and integrative approaches. The hedonic approach focuses on the degree to which people experience positive emotions of feelings and happiness and has sometimes been referred to as “subjective well-being” (Diener, 2000). It comprises both affective (mood, happiness) and cognitive components, as well as satisfaction (Fava and Ruini, 2003). In contrast, the eudaimonic approach emphasizes the

functioning components of PWB and is inclusive of a six-factor structure of purposeful behavior: self-acceptance, environmental mastery, positive relationships, personal growth, purpose in life, and autonomy (Ryff and Keyes, 1995). The variety of approaches, concepts, and measures can make it difficult to compare studies, and given the potential for theoretical overlap, many researchers have taken an integrative approach, realizing the value of both approaches in defining and measuring PWB.

This systematic review takes an integrative approach, encompassing both perspectives since numerous studies have noted higher levels of hedonic or eudaimonic PWB are associated with positive outcomes (Vazquez et al., 2009). In addition, the field of interventions designed to improve the PWB of MND carers is relatively new and, as such, an integrative approach to this review allows the potential for greater inclusiveness of studies. While there has been an increase in ideas and studies focusing on the PWB of MND carers, a systematic review of interventions has not yet been conducted. This review aims to (1) summarize the interventions designed to improve the PWB of MND carers; (2) assess the quality of the studies designed to improve the PWB of MND carers; and (3) evaluate the effectiveness of interventions designed to improve the PWB of MND carer.

Method

Conducting and reporting of this systematic review was based on the Joanna Briggs Institute (JBI) methodology for quantitative, qualitative, and mixed-methods reviews (Aromataris and Munn, 2020) and Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher et al., 2009).

Data sources and search strategy

The search strategy process intended to cover a broad range of intervention studies designed to improve the PWB of MND carers. Keywords (and their variations) used in the searches are detailed in [Box 1](#). A three-stage search strategy was employed. An initial limited search of CINAHL, PubMed, ProQuest, Scopus, and PsychINFO using pre-specified keywords was undertaken, followed by an analysis of the text words contained in the title and abstract and of the index terms used to describe the article. The second search used all identified keywords and index terms in a thorough search across all included databases. The third step involved searching for additional studies through the reference lists of all included studies. Only reviews published in English were included. Studies published before 1990 were excluded because MND care has significantly changed over the

Box 1. Search terms

Keywords (and their variations) used in the searches included those related to Condition: ("motor neuron* disease" OR MND or "amyotrophic lateral sclerosis" OR ALS), Population (famil* OR informal OR spous*) AND (carer* OR caregiv*) and Intervention ("behavio*r therapy" OR "cognitive behavior therapy" OR CBT OR "acceptance and commitment therapy" OR ACT OR mindfulness OR relaxation OR meditation OR counsel*ing OR grief OR bereavement OR "social support" OR "family therapy" OR "art therapy" OR "dance therapy" OR "music therapy" OR "complementary therap*" OR *Exercise OR Yoga OR "person centered*" OR "dignity therapy" OR psychotherap*OR psychosocial OR psychological OR treatment OR training OR education* OR program.

last 30 years and we were not expecting to find any relevant papers before 1990 (the "earliest" relevant paper we were aware of before doing this review was published in 2013). All articles published from 1990 to June 2021 were considered. Inclusion criteria comprised studies that were quantitative or qualitative or both (mixed-methods) if the quantitative or qualitative component could be extracted separately. Interventions needed to be carer-based and designed specifically for the purpose of improving carer PWB. Interventions could be delivered to individuals or groups and be inclusive of any mode of delivery, duration, and dose. The review included studies targeting spousal, family, and informal carers of people with MND and excluded professional or paid carers from government or private providers. Furthermore, the review included carers of people with MND independent of the disease stage and care setting.

Study selection

Outcomes of interest were any related to the PWB of carers, inclusive of any psychologically related (hedonic or eudaimonic) outcomes. Outcomes could be measured using any validated instrument, via observation or by self-report and measured before, during, or immediately after the intervention or after a follow-up period. The recommended JBI approach to a mixed-methods systematic review (MMSR) was followed. This process included JBI recommended screening protocols, design-appropriate standardized quantitative and qualitative critical appraisal forms to assess inclusion and methodological quality, and standardized data extraction tools as advocated by JBI (Stern et al., 2020). Selective reporting bias was further evaluated by comparing measurements and outcomes in papers with published protocols (where available), and methods and results were compared in papers without published protocols. The three reviewers separately assessed all studies to decide on in- or exclusion and disagreement was resolved by discussion. Data were subsequently extracted by two reviewers from the 13 selected papers with standardized JBI tools appropriate to each research design (Aromataris and Munn, 2020) (PC and TE: quantitative data; PC and ACH: qualitative data) using agreed narrative evidence descriptors. Study selection is displayed in the PRISMA flowchart ([Figure 1](#)).

Results

Demographic summary of the studies

In total, 13 papers were included ([Figure 1](#)), describing 12 studies as one study published quantitative and qualitative components of their research in separate papers (de Wit et al., 2019, 2020). Of the 12 studies, 6 included mixed methods, 4 used only quantitative methods (Creemers et al., 2014; van Groenestijn et al., 2015; Kavanaugh et al., 2020; Pagnini et al., 2021), while another 2 employed solely a qualitative approach (Marconi et al., 2016; Cipolletta et al., 2018). The included papers were all published between 2013 and 2021, with 8 of the 13 papers published since 2017. Of the 12 studies, 4 were conducted in Australia (Bentley et al., 2014; Aoun et al., 2017, 2018; Ugalde et al., 2018), 3 in the USA (Creemers et al., 2014; Steinhauer et al., 2016; Kavanaugh et al., 2020), 3 in Italy (Marconi et al., 2016; Cipolletta et al., 2018; Pagnini et al., 2021), and 2 in the Netherlands (van Groenestijn et al., 2015; de Wit et al., 2019,

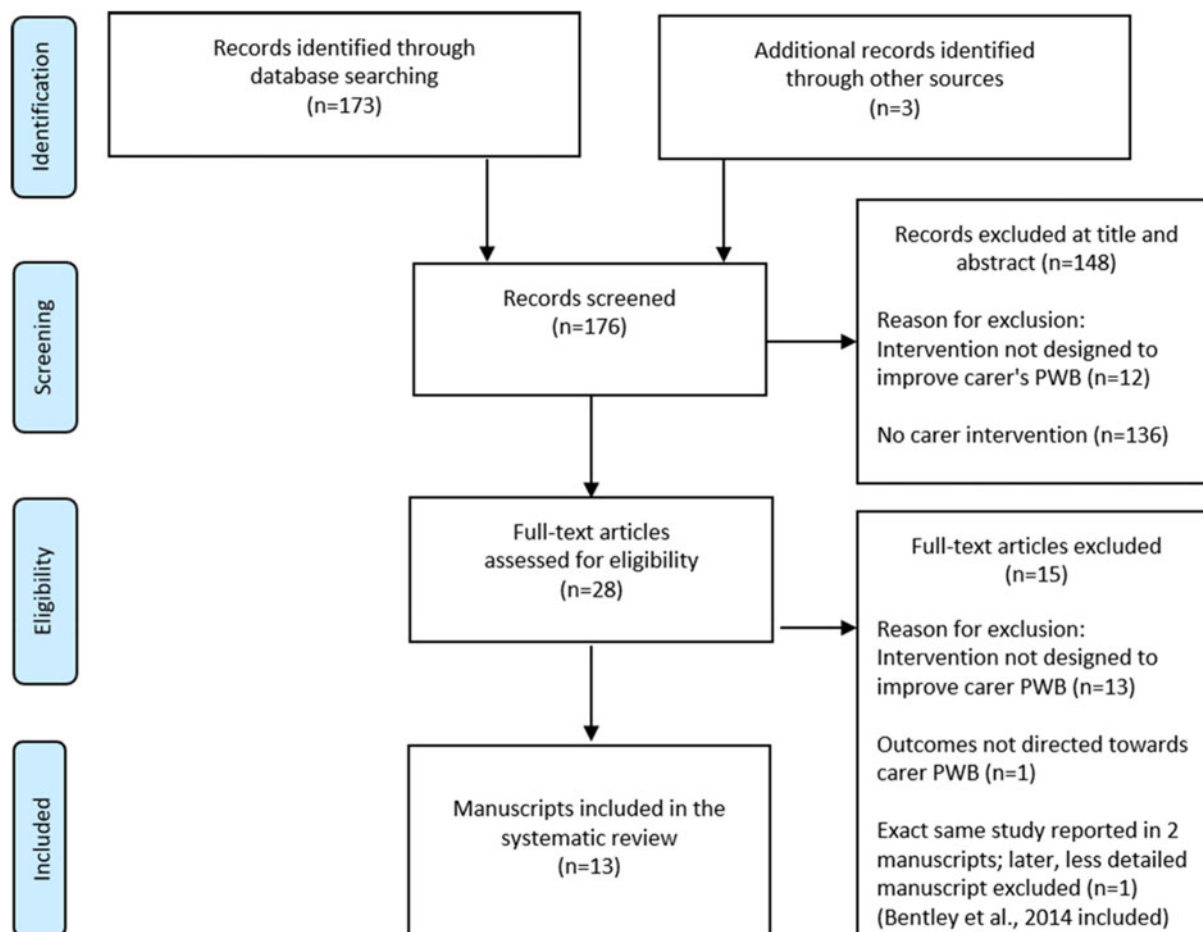


Fig. 1. PRISMA flowchart of paper selection.

2020). A total of 530 MND carers were studied in the quantitative component of the review, and 239 in the qualitative element.

Studies were based on applications of psychological treatments (e.g., Cognitive-Behavioral Therapy: van Groenestijn et al., 2015; Mindfulness and/or Meditation: Marconi et al., 2016; Ugalde et al., 2018; Pagnini et al., 2021; Dignity Therapy: Bentley et al., 2014; Acceptance Commitment Therapy: de Wit et al., 2019, 2020), and processes (e.g., Self-disclosure: Steinhauser et al., 2016), while others were founded in individualized case management or needs (Creemers et al., 2014; Aoun et al., 2017, 2018), support groups (Cipolletta et al., 2018), or carer skill for youth (Kavanaugh et al., 2020). One study used an online intervention (Pagnini et al., 2021).

Of the 12 studies, 6 were specifically designed for MND carers (Steinhauser et al., 2016; Aoun et al., 2017; Cipolletta et al., 2018; Ugalde et al., 2018; De Wit et al., 2019, 2020; Kavanaugh et al., 2020). Four studies detailed interventions designed for both MND patients and carers, including services or programs tailored to the needs of both (van Groenestijn et al., 2015; Aoun et al., 2018; Pagnini et al., 2021) or treatment with different protocols (Marconi et al., 2016). Two papers described patient-based interventions and primary outcomes, with carer PWD only a secondary focus (Bentley et al., 2014; Creemers et al., 2014).

The intensity of the intervention varied from a single 2.5-h group period (Ugalde et al., 2018) to eight, 90-min sessions over eight weeks (Marconi et al., 2016).

Critical appraisal

Methodological characteristics of studies with a quantitative component (10 studies)

Four of the 10 quantitative papers described randomized controlled trials (RCTs) with sample sizes of 15 (van Groenestijn et al., 2015), 27 (Pagnini et al., 2021), 126 (Creemers et al., 2014), and 148 (de Wit et al., 2020) and mean participant ages ranging between 53 and 62. Five papers described uncontrolled longitudinal studies with a single treatment group and a pretest, post-test design (n range = 13–31) (Bentley et al., 2014; Steinhauser et al., 2016; Aoun et al., 2017; Ugalde et al., 2018; Kavanaugh et al., 2020). Four studies involved adult carers (Bentley et al., 2014; Steinhauser et al., 2016; Aoun et al., 2017; Ugalde et al., 2018) (mean ages 57–64 years; median 61; Bentley et al., 2014), while one included 19 youth carers (aged 9–19) (Kavanaugh et al., 2020). One paper described an observational study using a survey design ($n = 117$, median age 64) (Aoun et al., 2018). Eight papers included a higher percentage of females (60–75%), while two studies involved more males (Creemers et al., 2014; Kavanaugh et al., 2020).

Table 1 summarizes the methodological characteristics of the quantitative studies using the JBI critical appraisal tools with a summary quality score (median 9, IQR 5.5, range 0–21) in the final row. Only the four RCTs achieved summary quality scores above 50% (score range between 57% and 78%).

Table 1. Methodological characteristics of studies with a quantitative component

Methodological characteristics	Paper (author, year, country)									
	Aoun et al., 2017, Australia	Aoun et al., 2018, Australia	Bentley et al., 2014, Australia	Creemers et al., 2014, USA	de Wit et al., 2020, Netherlands	Kavanaugh et al., 2020, USA	Pagnini et al., 2021, Italy	Steinhauser et al., 2016, USA	Ugalde et al., 2018, Australia	van Groenestijn et al., 2015, Netherlands
Cause/effect clear	?	–	?	+	+	?	+	–	?	+
Similar participants in comparisons	+	NA	+	+	+	+	+	+	+	+
Participants in comparisons receiving similar treatment, no other intervention	?	NA	?	+	+	?	?	?	?	?
Participants representative of target population	+	?	+	+	–	–	?	?	+	–
Multiple measurements of outcome pre- & post-intervention	+	–	+	+	+	–	+	+	+	+
Follow-up complete or appropriate strategies to address incomplete follow-up	–	NA	+	–	+	?	+	+	–	–
% completing ^a	80+	NA	80+	<60	<60	?	<60	80+	80+	<60
Outcomes of participants in comparisons measured same way	+	NA	+	+	+	+	+	+	+	+
Outcomes measured reliably	+	?	+	+	+	–	+	+	+	+
Results presented for all stated outcomes	+	+	+	–	+	–		+	+	–
Use of appropriate statistical analyses	–	–	+	+	+	?	+	+	?	+
Conclusions justified by data	+	+	–	+	+	–	–	+	+	+
Control group	–	–	–	+	+	–	+	–	–	+
True randomization for group assignment	NA	NA	NA	+	+	NA	+	NA	NA	+
Allocation concealed	NA	NA	NA	+	+	NA	–	NA	NA	+
Treatment groups similar at baseline	NA	NA	NA	+	–	NA	+	NA	NA	+
Participants blinded	NA	NA	NA	–	–	NA	–	NA	NA	–
Those delivering treatment blinded	NA	NA	NA	–	–	NA	–	NA	NA	–
Outcome assessors blinded	NA	NA	NA	+	?	NA	?	NA	NA	+
Participants analyzed in their randomized groups	NA	NA	NA	+	+	NA	+	NA	NA	+
Appropriate trial design for the topic	NA	NA	NA	+	+	NA	+	NA	NA	+
Quality score (/21)	8	2	9	16	15	2	12	9	8	14

Key: present (+), absent (–), unclear (?), not applicable (NA).

^aStudies reporting completion rates of 80% or above were assigned one point as recommended by the established guideline stating that up to 20% in-trial dropout is considered acceptable (Schulz and Grimes, 2002; Furlan et al., 2009).

The four studies using a control group were all RCTs (wait-list controls: de Wit *et al.*, 2020; Pagnini *et al.*, 2021; active controls: Creemers *et al.*, 2014; van Groenestijn *et al.*, 2015). The causal relationship between the intervention and carer PWB was demonstrated only in the four RCTs. In all RCTs, participants were aware of their group allocation and those delivering treatments were also not blind to participants' group allocation. Outcome assessors were blinded in two RCTs (Creemers *et al.*, 2014; van Groenestijn *et al.*, 2015). Whether or not outcome assessors were blind to treatment assignment was unclear in two studies (de Wit *et al.*, 2020; Pagnini *et al.*, 2021). Further, whether participants were all receiving similar treatment besides the studied intervention was only clearly defined in two quantitative studies (Creemers *et al.*, 2014; de Wit *et al.*, 2020).

Representativeness of the study population was problematic in six studies (van Groenestijn *et al.*, 2015; Steinhäuser *et al.*, 2016; Aoun *et al.*, 2018; De Wit *et al.*, 2020; Kavanaugh *et al.*, 2020; Pagnini *et al.*, 2021). In addition, there were sampling issues including small size and/or possible selection bias (Aoun *et al.*, 2018; de Wit *et al.*, 2020; Kavanaugh *et al.*, 2020; Pagnini *et al.*, 2021), disproportionately well-educated and financially stable participants (Steinhäuser *et al.*, 2016), rigid eligibility, and uneven group distribution (van Groenestijn *et al.*, 2015).

Follow-up was complete in six studies, incomplete in three studies (Bentley *et al.*, 2014; Steinhäuser *et al.*, 2016; de Wit *et al.*, 2020), and inapplicable in the cross-sectional survey (Aoun *et al.*, 2018). Only four studies reported intervention completion percentages of >80% (Bentley *et al.*, 2014; Steinhäuser *et al.*, 2016; Aoun *et al.*, 2017; Ugalde *et al.*, 2018), one study did not report completion rates (Kavanaugh *et al.*, 2020), while the four RCTs noted low completion rates (<60%) (Creemers *et al.*, 2014; van Groenestijn *et al.*, 2015; de Wit *et al.*, 2020; Pagnini *et al.*, 2021).

Appropriate statistical analyses were evident in the RCTs. Two RCTs were sufficiently powered (Creemers *et al.*, 2014, $n = 126$; de Wit *et al.*, 2020, $n = 148$). Only two non-RCT studies (Bentley *et al.*, 2014; Steinhäuser *et al.*, 2016) used appropriate statistical methods. The five uncontrolled longitudinal studies with a single treatment group and a pretest post-test design used small samples ($n = 13$ –24) with limited statistical power.

Methodological characteristics of studies with a qualitative component (eight studies)

Six of the eight studies described uncontrolled longitudinal studies with a single treatment group and a pre-test, post-test design (n ranging from 12 to 24 carers). One study reported on qualitative research embedded in an RCT ($n = 23$) (de Wit *et al.*, 2019), while another described a survey design ($n = 117$) (Aoun *et al.*, 2018). Six studies reported a higher percentage of female participants (58–100%); one did not present gender data (Marconi *et al.*, 2016). All 14 participants in one study (Steinhäuser *et al.*, 2016) were female and described as "white." The mean age of participants was reported in seven studies and ranged from 57 to 63 years.

Three qualitative studies met most criteria for good methodological practice as defined by the JBI qualitative critical appraisal tool (quality scores between 73% and 80%). The other five studies demonstrated weaknesses in qualitative methodological rigor (score range: 20–60%). The median quality score was 6.5 (IQR = 3.5, range 0–13, Table 2).

Methodological rigor was more evident in some domains. Seven papers clearly provided a statement of ethical approval (unclear in de Wit *et al.*, 2019). Seven studies presented illustrative quotes to support themes (no themes presented in Ugalde *et al.*,

2018), while six studies demonstrated congruity between the research methodology and both the research question and data collection methods. Aoun *et al.* (2018) used open-ended questions without data analysis in a cross-sectional survey to collect intervention data during the last year of the 3-year study. Ugalde *et al.* (2018) used responses to open-ended questions with a limited representation of participant voices or data analysis. The relationship of the conclusion to data analysis or interpretation was evident in six studies. In Marconi *et al.* (2016), carer voices were not represented in all the domain-based illustrative quotes and conclusions sometimes grouped MND patients and carers together, while Ugalde *et al.*'s (2018) paper made conclusions based on responses to open-ended questions with limited representation of participant voices or theme generation.

No qualitative studies presented a statement locating the researcher culturally and/or theoretically or commented on the influence of the researcher on the research, and vice versa. Additionally, congruity between the stated philosophical perspective and the research methodology was absent in all but one study (Steinhäuser *et al.*, 2016). These studies did not present a theoretical or philosophical perspective or if they did it was incompatible with the methodology (e.g., interpretive perspectives based on survey methodology, Aoun *et al.*, 2018; Ugalde *et al.*, 2018).

Four studies did not demonstrate that more than one researcher agreed on themes (unclear: Steinhäuser *et al.*, 2016; Aoun *et al.*, 2018; presented quotes without any evidence of analysis: Bentley *et al.*, 2014; Ugalde *et al.*, 2018). Carer voices were represented in only four studies. The study by Steinhäuser *et al.* (2016) presented only selective data, no data were included to represent deviant cases, and a clear method of representing the data based on the presented methodology was not apparent. Quotes in Bentley *et al.* (2014) and Ugalde *et al.* (2018) are presented without significant description in the results, evidence of representation, or data analysis. In Marconi *et al.* (2016), carer voices were not always present in illustrative quotes, and patient voices were used instead. Incongruity between the research methodology and results interpretation was present in four studies. This was exemplified by interpretive perspectives based on open-ended questions (Ugalde *et al.*, 2018); open-ended questions from a survey (Aoun *et al.*, 2018), mixed carer and patient data (Marconi *et al.*, 2016), and both limited and mixed (MND and cancer carer responses were mixed together) representation of the data (Steinhäuser *et al.*, 2016). Congruity between the research methodology and the representation and analysis of the data was not evident in the three studies. In Aoun *et al.* (2018), this congruity was unclear as the methodology was described as mixed-methods; however, selective responses to open-ended questions on a survey design were listed without any presentation of information regarding the data analysis methods or epistemological stance. Ugalde *et al.* (2018) demonstrated limited evidence of representation of participant voices and no analysis of responses to open-ended questions. In Steinhäuser *et al.* (2016), a clear method of data representation based on the presented methodology was not apparent. MND and cancer carers' responses were not separated.

Data extraction

Due to the low number of studies as well as the heterogeneity of the interventions, outcomes, and measurements, data pooling for meta-analysis was not possible. Results from studies with quantitative and qualitative components are presented in Tables 3 and 4 respectively. These results are presented in a narrative manner and

Table 2. Methodological characteristics of studies with a qualitative component

Methodological characteristics	Paper (author, year, country)							
	Aoun et al., 2017, Australia	Aoun et al., 2018, Australia	Bentley et al., 2014, Australia	Cipolletta et al., 2018, Italy	de Wit et al., 2019, Netherlands	Marconi et al., 2016, Italy	Steinhauser et al., 2016, USA	Ugalde et al., 2018, Australia
Congruity — stated philosophical perspective & research method	NA	—	NA	NA	—	—	+	NA
Congruity — research method & research question/objectives	+	—	+	+	+	+	+	—
Congruity — research method & data collection methods	+	—	+	+	+	+	+	—
Congruity — research method & representation & analysis of data	+	?	+	+	+	+	—	—
Congruity — research method & result interpretation	+	—	+	+	+	—	—	—
Statement locating researcher culturally or theoretically	—	—	—	—	—	—	—	—
Influence of researcher on the research & vice versa addressed	—	—	—	—	—	—	—	—
Participants, and their voices, adequately represented	+	+	—	+	+	—	?	—
Participants likely to be representative of the target population	+	—	—	—	+	?	—	—
Research ethical/evidence of ethical approval	+	+	+	+	?	+	+	+
>1 researcher agreed on themes	+	?	—	+	+	+	?	NA
Illustrative quotes to support themes	+	+	+	+	+	+	+	NA
Conclusions flow from the analysis, or interpretation, of the data	+	+	+	+	+	—	+	—
Quality score (/13)	10	4	7	9	9	6	6	1

Key: present (+), absent (—), unclear (?), not applicable (NA).

synthesised descriptively as recommended by the JBI guidelines for MMSRs (Aromataris and Munn, 2020).

Quantitative findings

The RCTs predominantly used standardized outcome measures focusing on hedonic aspects of PWB (Table 3). No effect was demonstrated using these measures in two RCTs (Creemers et al., 2014; de Wit et al., 2020). Two RCTs (van Groenestijn et al., 2015; Pagnini et al., 2021) reported a significant increase in the quality of life and a reduction in psychological comorbidities, although methodological concerns were present. One RCT (de Wit et al., 2020) demonstrated a significant hedonic PWB benefit for controlling upsetting thoughts about caregiving. Results were largely unresponsive for PWB benefits in the

quantitative components of the five uncontrolled longitudinal studies with a single treatment group and a pre-test, post-test design (Table 3). The one exception was the study by Aoun et al. (2017) (appraised as low methodological quality) where hedonic and eudaimonic PWB benefits were reported as reductions in carer support needs. Three (Bentley et al., 2014; Steinhauser et al., 2016; Ugalde et al., 2018: all appraised as low quality) of the other four uncontrolled studies did not demonstrate any benefits with standard PWB questionnaires. Hedonic and eudaimonic PWB benefits were seen from the acceptability questionnaire in Ugalde et al.'s (2018) study. The remaining uncontrolled study (Kavanaugh et al., 2020: appraised as very low methodological quality) reported a significant increase in confidence with several care tasks. The observational study

Table 3. Synthesized narrative findings for studies with a quantitative component

Intervention (author and year of publication)	Design (n) and intervention	Study outcome(s) and assessment tool(s)	Outcome summary	Intervention benefits: PWB categories (hedonic & eudaimonic)	Critical appraisal of quality score (/21)	Narrative synthesis
Carer Support Needs Assessment Tool (Aoun et al., 2017)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test (n = 24) Two visits (face-to-face or phone) from care advisors (6–8 weeks apart) to identify and address support needs using the CSNAT (median 10 min to complete)	* <i>Carer Support Needs Assessment Tool (CSNAT)</i>	Reductions in the percentage of participants with support needs in each of the domains except “your beliefs and spiritual concerns”	Hedonic: subjective well-being, emotions (feelings and thoughts) Eudemonic — environmental mastery, positive relations with others, autonomy	8	* Reductions in the percentage of participants with support needs were demonstrated, translating to hedonic and eudaimonic PWB benefits. * Outcomes were limited to CSNAT percentages; no further statistical analyses were performed. * Methodological quality of the study was appraised as low.
Person-Centred Model of care — MND Advisory Service (Aoun et al., 2018)	Observational Survey (n = 117) Person-centered model of care provided by the MND Advisory Service of the MND Association of Western Australia providing services associated with provision of information and support to pwMND and carers. No data regarding specific number and duration of contacts	* <i>Extent carers felt cared for and supported in the community to maintain the best quality of life throughout disease progression;</i> * <i>Extent carers felt supported to make informed and better decisions to manage their health and well-being throughout disease progression through accessible, understandable, and timely information;</i> * <i>Satisfaction with provision of information;</i> * <i>Satisfaction with provision of support</i> * Subscales on the Satisfaction with MND CARE-Family Carers questionnaire	High percentage (>80%) of participants felt: cared for and supported to maintain the best quality of life; supported to make informed and better decisions to manage their health and well-being; satisfaction with services received from MND Advisory Service; satisfaction with MND Advisory Service information	Hedonic: subjective well-being, emotions (feelings and thoughts), mental health status Eudemonic — environmental mastery	2	* High percentages of respondents reported hedonic and eudaimonic PWB benefits. * Quantitative outcomes were limited to percentages and no further statistical analyses were performed. Intervention variability was unclear and “dose”/ number of contacts received by the MND Advisory service was not stated. The design was a cross-sectional survey with no baseline data. Methodological quality of the study was appraised as very low.
Dignity Therapy (DT) (Bentley et al., 2014)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test (n = 18) Three patient-focused DT sessions, spaced between two and three days apart	* <i>Burden: Zarit Burden Inventory (ZBI)</i> * <i>Psychological distress: Hospital Anxiety Depression Scale (HADS)</i> * <i>Hopefulness: Herth Hope Index (HHI)</i> * Family feedback questionnaire	No effect (anxiety, depression, hopefulness); Mixed results from the family feedback questionnaire regarding the benefits of DT	No clear hedonic or eudaimonic benefits demonstrated	9	* No significant hedonic PWB benefits were demonstrated in caregiver anxiety, depression, burden, and hopefulness. * Mixed results from the family feedback questionnaire did not clearly support hedonic or eudaimonic PWB benefits. * Methodological quality of the study was appraised as low.

(Continued)

Table 3. (Continued.)

Intervention (author and year of publication)	Design (n) and intervention	Study outcome(s) and assessment tool(s)	Outcome summary	Intervention benefits: PWB categories (hedonic & eudaimonic)	Critical appraisal of quality score (/21)	Narrative synthesis
Case Management (Creemers et al., 2014)	RCT (n = 126) Baseline and quarterly visits (ranging from 60–180 min) over 12 months by occupational therapists	* <i>Burden: Caregiver Strain Index (CSI)</i> * <i>Rating of the quality of care provided to the caregiver: Rating scale (1–10)</i>	No effect: burden or quality of care	No clear hedonic or eudaimonic benefits demonstrated	16	* No significant hedonic or eudaimonic PWB benefits demonstrated for caregiver burden or level of satisfaction. * Authors noted a lack of focus on caregiver issues in comparison with patient problems in the intervention. * Appraised as methodologically sound.
Blended psychosocial support program (based on Acceptance Commitment Therapy) (de Wit et al., 2020)	RCT (wait-list) (n = 148) 1 × 1 h face-to-face initial contact with a psychologist, 6 × 90 min online guided modules and 1 × 30 min closing telephone contact (total of 8–12 weeks; all guided by psychologists)	* <i>Psychological distress: Hospital Anxiety Depression Scale (HADS)</i> * <i>Burden: Zarit Burden Interview (ZBI)</i> * <i>Caregivers' quality of life: Care Related- Quality of Life (CarerQoL) + CarerQoL Visual Analog Scale (VAS)</i> * <i>Caregivers' beliefs about their capacity to obtain respite from caregiving: Respite care subscale of the Revised Scale for Caregiving Self-Efficacy (RSCSEResp)</i> * <i>Caregivers' beliefs about their ability to control distressing thoughts about caregiving: "Controlling upsetting thoughts about caregiving" subscale of RSCSE (RSCSE-Contr)</i> * <i>Perceived control over fulfilling caregiver tasks: Job Content Questionnaire (JCQ)</i>	No effect: distress, burden, quality of life, caregiver beliefs about their capacity to obtain respite, perceived control over fulfilling caregiver tasks Significant intervention × time interaction ($b = -5.39$, $se = 1.99$, $p < 0.01$) for "Controlling upsetting thoughts about caregiving"	Hedonic: emotions (feelings and thoughts)	15	* No significant hedonic PWB benefits demonstrated for distress although baseline levels were low. * No significant hedonic PWB reduction in burden, eudaimonic PWB improvement in caregiver beliefs about their capacity to obtain respite or perceived control over fulfilling caregiver tasks. * Significant hedonic PWB benefit regarding controlling distressing thoughts about caregiving. * Caregivers main reported motivation for RCT was to take part in ALS research rather than a need for support. * Appraised as methodologically sound.
YCare Training Protocol for youth carers (based on the tenets the Individual and Family Self-Management Theory) (Kavanaugh et al., 2020)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test (n = 19) 4 × 50 min modules in groups (youth of similar ages) over one training day	* <i>Caregiving self-efficacy: For each task identified in the Multidimensional Assessment of Caring Activities (MACA-YC18), youth were asked to rate their level of confidence in completing the task.</i>	Effect: significant increase in confidence scores with several care tasks: use of respiratory equipment (mean improvement = 1, SE = 0.45, $p = 0.039$), communication systems (mean improvement = 0.94, 0.32, $p = 0.009$), power chair use (mean improvement = 0.88, SE = 0.37, $p = 0.031$)	Eudaimonic: environmental mastery	2	* Eudaimonic PWB (environmental mastery) effect demonstrated with significant increase in confidence with several care tasks. * Numerous methodological problems. * Methodological quality appraised as very low.

Online non-meditative mindfulness (based on Langerian mindfulness) (Pagnini et al., 2021)	RCT (wait-list) ($n = 27$) Online website including video and written mindfulness content and two daily exercises, (2–10 min to complete). A specific mindfulness topic (attention to variability, positive and negative events, unpredictability, sense-making, novelty seeking and producing) covered each of the five weeks of the program	* <i>Quality of life</i> : Short-Form 36 * <i>Anxiety</i> : Hospital Anxiety Depression Scale ** <i>Depression</i> : Hospital Anxiety Depression Scale <i>Caregiver burden</i> : Zarit Burden Inventory	Time × group effects in favor of mindfulness intervention: care burden ($F = 4.347$, $df = 3$, 38.23 , $p = 0.01$), depression ($F = 4.868$, $df = 3$, 27.65 , $p < 0.01$), anxiety ($F = 3.03$, $df = 3$, 29.92 , $p < 0.05$), role limitations due to personal or emotional problems ($F = 3.96$, $df = 3$, 33.67 , $p < 0.05$), energy/fatigue ($F = 2.91$, $df = 3$, 32.14 , $p < 0.05$), emotional well-being ($F = 2.88$, $df = 3$, 34.05 , $p < 0.05$)	Hedonic: subjective well-being, mental health status, emotions (feelings and thoughts)	12	* Hedonic PWB time × group effects demonstrated in favor of the mindfulness intervention. * Due to the small sample, the study focused on effect sizes rather than significance between group differences. There was a lack of clarity regarding the exact effect sizes and significance at each time point. * Methodological quality of the study was appraised as low to moderate
Chaplain-led self-disclosure (manualized chaplain-led intervention (“Caregiver Outlook” — based on the human development and self-disclosure literature) to improve well-being by exploring role-related meaning) (Steinhauser et al., 2016)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test ($n = 23$) Three-weekly 1-h sessions focusing on: (1) a relationship review, (2) forgiveness and (3) legacy	* <i>Burden</i> : Caregiver Reaction Assessment (CRA) * <i>Anxiety</i> : anxiety subscale from the modified Brief Profile of Mood States (POMS) * <i>Depression</i> : Center for Epidemiology Studies Depression Scale (CES-D) * <i>Preparation and completion of caregiving role</i> : preparation and completion subscales of the Quality of Life at the End-of-Life scale (QUAL-E — fam) (family members) * <i>Meaning, peace, and faith</i> : The Functional Assessment of Chronic Illness Therapy–Spiritual Well-Being Scale * <i>Grief</i> : Prolonged Grief Scale (PG-12) * <i>Religious coping</i> : Brief Religious Coping Activity Scales (RCOPE)	No effect (burden, anxiety, depression, grief, religious coping, preparation and completion of carer role, meaning, peace and faith)	No hedonic or eudaimonic PWBbenefits demonstrated	9	* No significant benefits demonstrated for any of the outcome measures related to hedonic or eudaimonic PWB. * No statistically significant changes on any of the outcome measures nor any comment on clinically significant changes (MID or MCID). * At baseline, participants did not demonstrate clinical threshold levels of anxiety, depression, or other indicators of distress, so improvement was unlikely. * Methodological quality of the study was appraised as low.
Mindfulness (Ugalde et al., 2018)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test ($n = 13$) Single, 2.5-h group session including mindfulness, self-care and problem-solving	* <i>Burden</i> : Caregiver Reaction Assessment (CRA) * <i>Psychological distress</i> : <i>Depression, anxiety, and somatization</i> : Brief Symptom Inventory (BSI-18) * <i>Preparedness</i> : Preparedness for Caregiving Scale * <i>Problem-solving confidence, approach-avoidance style, and personal control</i> : Problem-Solving Inventory * <i>Mindfulness</i> : Cognitive and Affective Mindfulness Scale–Revised * <i>Acceptance of intervention</i> : Acceptance questionnaire	No significant effects (distress, burden, problem-solving, mindfulness, preparedness) Acceptance questionnaire results indicated that most respondents believed they benefitted in terms of their PWB	No clear hedonic or eudaimonic benefits shown from standardized questionnaires Acceptance questionnaire benefits: subjective well-being (hedonic), positive relations with others (eudaimonic)	8	* No significant change on any pre-post hedonic or eudaimonic PWB measures post-intervention. * Hedonic (subjective well-being) and eudaimonic (positive relations with others) benefits were seen from the acceptability questionnaire although only descriptive statistics were presented. * Methodological quality of the study was appraised as low.

(Continued)

Table 3. (Continued.)

Intervention (author and year of publication)	Design (n) and intervention	Study outcome(s) and assessment tool(s)	Outcome summary	Intervention benefits: PWB categories (hedonic & eudaimonic)	Critical appraisal of quality score (/21)	Narrative synthesis
Cognitive-Behavior Therapy (CBT) (based on the stress-coping model) (van Groenestijn et al., 2015)	RCT (n = 15) 5–10 tailored CBT sessions (1 h) delivered to pwMND-carer pairs (individually or together) by a psychologist over 16 weeks. Sessions comprised of six modules: coming to terms with the diagnosis; coping with emotional instability; maintaining autonomy; mobilizing social support; coping with fear of the future; maintaining activity levels	* <i>Mental Quality of Life</i> : Short-Form 36 (SF36) * Mental Component Score (MCS) * <i>Burden</i> : Caregiver Strain Index (CSI) * <i>Distress</i> : Hospital Anxiety Depression Scale (HADS) total	Significant increase in mental QoL in the CBT group by 7.5 points (clinically relevant) Significant between time × group effect regarding burden (CBT group lower) No effect on distress	Hedonic: subjective well-being, mental health status, emotions (feelings and thoughts)	14	* Some benefits to hedonic PWB (mental QoL and caregiver burden) * Authors acknowledged methodological limitations, limiting conclusions about the effectiveness of the CBT intervention, and the trial was stopped prematurely due to slow recruitment. Small, under-powered sample as only 4/10 patient-carer pairs completed the minimum of five CBT sessions. * The inclusion criteria were based on patient rather than caregiver level of psychological distress. * Appraised as methodologically sound

PWB, psychological well-being; n, sample size; PwMND, person(s) with MND; QoL, quality of life; RCT, randomized controlled trial; SE, standard error.

Table 4. Synthesized narrative findings for studies with a qualitative component

Author & Intervention	Design (n) and intervention	Data collection and analysis	Outcomes summary	Intervention benefits: PWB categories (hedonic and eudaimonic)	Critical appraisal of quality score (/13)	Narrative synthesis
Carer Support Needs Assessment Tool (Aoun et al., 2017)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test (n = 24) For intervention information, see Table 3.	Semi-structured telephone interviews within two weeks of intervention completion. Thematic analysis supported by NVivo 10 software.	Theme 1: The Overwhelming Caregiver Journey with MND Theme 2: The CSNAT Practicality and Usefulness Theme 3: Validation of the Caregiver Role and Empowerment Theme 4: Reassurance of Support	Hedonic: Emotions (feelings & thoughts) Eudaimonic: self-acceptance, environmental mastery, purpose in life, personal growth	10	* Benefits to hedonic and eudaimonic aspects of PWB were evident through the data associated with each of the four themes. * Appraised as methodologically sound.
Person-Centered Model of care — MND Advisory Service (Aoun et al., 2018)	Observational Survey (n = 117) For intervention information, see Table 3.	Open-ended questions as part of an anonymous postal survey. The process of analyzing qualitative data was not described apart from open-ended responses being categorized to two support domains: practical or emotional.	Finding 1: Practical support Finding 2: Emotional support	Hedonic: mental health status, subjective well-being, emotions (feelings & thoughts) Eudaimonic: environmental mastery, positive relationships, personal growth	4	* The data associated with each of the findings were indicative of hedonic and eudaimonic PWB improvements, but significant methodological issues were identified. * Appraised as low methodological quality.
Dignity Therapy (DT) (Bentley et al., 2014)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test (n = 18) For intervention information, see Table 3.	A family feedback questionnaire collected family carers' opinions and experiences of DT. The questions contained space for brief explanation. The data analysis process was not described, but a sample of carers' responses to each item in the open-ended questions from the family feedback questionnaire was presented in tabular format. No themes were derived.	Mixed results regarding the helpfulness of DT for carer PWB was evident from the open-ended responses to the carer feedback questions.	No clearly demonstrated hedonic or eudaimonic benefits	7	* Benefits to both hedonic and eudaimonic aspects of PWB were unclear. * No thematic analysis and only select comments were included for each acceptability item. * Appraised as low to moderate methodological quality.
Mutual support groups for ALS family caregivers (Cipolletta et al., 2018)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test (n = 12) Two support groups for carers (facilitated by two psychologists) were conducted, one	Post-intervention semi-structured interviews. Content analysis of the transcripts of the interviews and the 20 group sessions.	Themes identified: (1) Group experience: Partners group: No benefits noted. Adult children's group: Benefits were acceptance of the disease, helped prepare for the future,	(1) Group experience (adult children): Hedonic: emotions (feelings and thoughts) Eudaimonic: purpose in life, positive relationships,	9	* Benefits to hedonic and eudaimonic aspects of PWB were evident from the data. * The benefits conferred were profoundly influenced by group composition and dynamics.

(Continued)

Table 4. (Continued.)

Author & Intervention	Design (n) and intervention	Data collection and analysis	Outcomes summary	Intervention benefits: PWB categories (hedonic and eudaimonic)	Critical appraisal of quality score (/13)	Narrative synthesis
	<p>for partners and one for adult children (total of 10 × 30 min sessions).</p>		<p>useful to re-think the meaning of their experience, ongoing, and reliable support. (2) Group evaluation benefits: improvement in emotional and mental states; personal space to share with people undergoing similar experiences; psychologist facilitator infused security and encouraged reflection; less alone; confirmation of performing caring roles well; better understand patients' point of view; rethinking of role leading to attempts to maintain own space and engage in hobbies without feeling guilty.</p>	<p>environmental mastery (2) Group evaluation: Hedonic: mental health status, subjective well-being, emotions (feelings & thoughts) Eudaimonic: environmental mastery, autonomy, positive relations with others</p>		<p>* Qualitative component of the study was appraised as methodologically sound.</p>
<p>Blended psychosocial support program based on Acceptance and Commitment Therapy (de Wit et al., 2019)</p>	<p>RCT (wait-list) interviews were conducted with purposively sampled 23 of the 148 ALS/PMA caregivers enrolled in the RCT 1 × 1 h face-to-face initial contact with a psychologist, 6 × 90 min online guided modules and 1 × 30 min closing telephone contact (total of 8–12 weeks; all guided by psychologists).</p>	<p>Individual in-depth interviews Thematic analysis supported by NVIVO 10 software</p>	<p>Caregivers more aware of their own situation, perceived more control over caregiving, accepted negative emotions and thoughts, increased attention to their partner relationship, felt acknowledged, empowered caregivers to make choices according to their own needs which they perceived as a positive change. Mixed evaluations regarding mindfulness and peer contact components — most participants did not find them helpful</p>	<p>Hedonic: emotions (feelings and thoughts), subjective well-being Eudaimonic: self-acceptance, environmental mastery, positive relationships, autonomy</p>	<p>9</p>	<p>* The program evaluation demonstrated clear benefits to hedonic and eudaimonic aspects of PWB. * The mixed evaluations regarding mindfulness and peer contact components emphasize the need for program personalization. * Appraised as methodologically sound.</p>

Meditation based on an adapted Mindfulness-based Stress Reduction protocol (Marconi et al., 2016)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test ($n = 18$) 90-min meditation sessions were held weekly over eight weeks. Each session emphasized accepting the discomfort and physical limitations and were conducted by two trainers.	Semi-structured interview post-intervention Data were analyzed with a grounded theory approach.	Domains identified: Improvements in well-being, Relaxation, Emotional self-regulation, Acceptance, Consciousness, Breathing issues, Sleep cycle, Relationships, Effectiveness of a group setting with other people in a similar situation	Hedonic: emotions (feelings and thoughts) Unclear if further PWB benefits were obtained by caregivers	6	* Hedonic emotional benefit clearly linked to caregivers. Other benefits presented were based on data from “participants” where patients and caregivers were often bundled together. * Appraised as low methodological quality.
Chaplain-led self-disclosure (manualized chaplain-led intervention (“Caregiver Outlook” — based on the human development and self-disclosure literature) to improve well-being by exploring role-related meaning) (Steinhauser et al., 2016)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test ($n = 14$) For intervention information, see Table 3.	Evaluation interviews: Transcripts were reviewed for common and recurrent themes related to evaluation questions. Analyses followed a descriptive qualitative analytic approach.	Themes: Stepping back from day-to-day tasks to reflect on role and role change; opportunity to process emotions; stimulating communication with others; anonymity of phone conversation;	Hedonic: emotions (feelings and thoughts) Eudaimonic: self-acceptance, positive relationships	6	* Benefits to hedonic and eudaimonic PWB were identified, although numerous methodological concerns were present. * Appraised as low methodological quality.
Mindfulness (Ugalde et al., 2018)	Longitudinal, uncontrolled, single treatment group, pre-test, post-test ($n = 13$) For intervention information, see Table 3.	Open-ended questions as part of a post-intervention acceptability questionnaire. The data analysis process was not described. A sample of participants’ responses to the open-ended question were presented in narrative format. No themes were reported.	Benefits identified by participants: valued the opportunity to hear from others in similar situations, having time allocated specifically for them rather than the focus being on the patient, having professional input and finding out support is available	Eudaimonic: self-acceptance, positive relationships with others	1	* Eudaimonic aspects of PWB were identified, but significant methodological issues were present. * Appraised as very low methodological quality.

PWB, psychological well-being; n , sample size.

using a survey design (Aoun et al., 2018: appraised as low methodological quality) reported high percentages of respondents experiencing hedonic and eudaimonic PWB benefits. However, the lack of pre-post design in observational studies limits the interpretability of these results.

Qualitative findings

The three studies (Aoun et al., 2017; Cipolletta et al., 2018; de Wit et al., 2019) appraised as methodologically sound demonstrated benefits to a range of hedonic and eudaimonic aspects of PWB (Table 4). In the mutual support group intervention, the benefits conferred were influenced by group composition and dynamics (Cipolletta et al., 2018). Three (Marconi et al., 2016; Steinhauser et al., 2016; Aoun et al., 2018) studies appraised as low methodological quality reported benefits to both hedonic and eudaimonic aspects of PWB (Table 4). The other study (Bentley et al., 2014) reported mixed support for PWB from data derived from open-ended questions. While some eudaimonic PWB benefits were identified in the Mindfulness study (Ugalde et al., 2018), this was appraised as very low methodological quality.

Discussion

This is the first systematic review of interventions directly targeting the PWB of MND carers. The results indicate the dearth and recency of activity in this field as the number of included studies was low and papers describing interventions to improve carers' PWB were only found from 2013 (8 of the 13 papers were published from 2017). Heterogeneity was evident in the types of interventions ranging from psychological treatments to support groups.

This review identified future areas of methodological development, as the included studies did not meet all the JBI appraisal tools' criteria. Among the 10 studies with a quantitative component, only 4 described RCTs, considered as the gold standard for evaluating intervention effectiveness (Akobeng, 2005). The six non-RCT quantitative studies were evaluated as low or very low methodological quality, significantly limiting the interpretability and confidence in these findings. None of these studies were able to demonstrate a causal relationship or used a control group. Additionally, it was unclear in 8 of the 10 quantitative studies whether participants received any other treatments, potentially interfering with the attribution of "effect" to the intervention studied (Akobeng, 2005).

In future non-RCTs, researchers should carefully collect information regarding the most obvious potential confounders not only at baseline but also during the follow-up of the study. MND carer PWB has been linked to patient (e.g., symptom severity, emotional health, cognitive or behavioural change), carer (e.g., individual traits, problem-solving skills, coping styles), relationship (e.g., relationship satisfaction, nature of the relationship), and social factors (e.g., adequacy of social support, isolation) (Gluyas et al., 2017; Aoun et al., 2021). In addition, careful investigation, measuring, and reporting of participant exposure to treatments other than the intervention that may potentially influence carer PWB are important. The optimal design to minimise influence of confounders is an RCT (Akobeng, 2005). However, if using an RCT is not feasible, confounders can also be managed by using strategies such as restriction (e.g., inclusion and exclusion criteria), matching, and statistical control (Kahlert et al., 2017; Yan et al., 2020).

Whereas appropriate inclusion and exclusion criteria are essential to reduce confounding, the use of strict criteria may negatively impact on participant recruitment and generalisability of

data (e.g., van Groenestijn et al., 2015). Furthermore, when the inclusion of patient-carer dyads is only based on patient-related data, it may affect the relevance of carer-centric variables. For example, in cases where inclusion was based on patient rather than carer levels of psychological distress (e.g., van Groenestijn et al., 2015), carer-based variables associated with carer PWB may be missed and individual carer needs relevant to PWB may not be addressed.

Attrition rates were high in the included studies, with only four quantitative papers reporting intervention completion percentages of >80% (Bentley et al., 2014; Steinhauser et al., 2016; Aoun et al., 2017; Ugalde et al., 2018). Completion rates were below 60% in each of the RCTs, consequently reducing the possibility of answering the research question or detecting clinically meaningful effects (Akobeng, 2005). This is concerning as attrition rates greater than 20% involve serious threats to validity (Schulz and Grimes, 2002; Furlan et al., 2009). Managing carer attrition in longitudinal MND studies (RCTs and non-RCTs) is challenging because burden increases as MND progresses (Creemers et al., 2016) and patient deaths become common, which will directly impact carer attrition as it is closely linked to that of patients (Burke et al., 2018). Carer research burden minimisation plans are essential as the population is time-poor and often lacks access to essential services (Gluyas et al., 2017; Aoun et al., 2021). Research burden could be reduced by using: (1) less-time demanding interventions and (2) technological methods such as self-paced, web-based interventions, home-based data collection, and telehealth. Where intervention attendance is preferred, transport provision would be enabling. Furthermore, carers may be more available and less overwhelmed earlier in the MND trajectory (Aoun et al., 2013; Gluyas et al., 2017). Minimising exclusions post-randomisation and intention to treat analyses (Schulz and Grimes, 2002) will help to manage issues associated with participant noncompliance, protocol deviations, and withdrawal, while guarding against an over-estimation of treatment effect (Gupta, 2011).

Recruiting difficulties could be addressed by broadening the range of eligibility criteria, including more recruitment sites (van Groenestijn et al., 2015) and minimising carer burden. The study with youth carers (Kavanaugh et al., 2020) also noted distinct recruitment issues associated with this population (e.g., parental protection and isolation) and the authors suggested methods of addressing these problems in future studies (e.g., parental reassurance and ease of access).

A lack of blinding of participants and those delivering and assessing treatments were evident in the RCTs. Blinding is not always possible due to the nature of the intervention but may introduce performance and detection bias. Future studies should thoroughly assess the possibilities for blinding issues at participant, deliverer, and assessor level to eliminate or account for potential bias (Boutron et al., 2007).

Representativeness was also uncommon in the quantitative studies and future research should be cognisant of reducing such sampling error as it may introduce selection and information bias, as well as limiting external validity and generalizability (Kukull and Ganguli, 2012).

While some positive results were obtained in the qualitative studies concerning interventions for the PWB of MND carers, our review also indicated that this emerging field of research requires methodological development. Only three of the eight qualitative studies met most of the criteria of the JBI critical appraisal tool and demonstrated a range of PWB benefits in both hedonic and eudaimonic domains from the use of the

Carer Support Needs Assessment Tool (Aoun et al., 2017), mutual support groups (Cipolletta et al., 2018), and a blended psychosocial support program based on Acceptance Commitment Therapy (de Wit et al., 2019). While the remaining five studies identified a qualitative component, the study characteristics often did not meet most criteria of the critical appraisal tool. For example, some of the studies (Bentley et al., 2014; Aoun et al., 2018; Ugalde et al., 2018) presented selective data obtained through open-ended questions. Although such responses qualify as qualitative data, free text (such as that obtained in surveys) does not constitute qualitative research (Hammarberg et al., 2016). While qualitative research may be inclusive of a wide range of epistemological and theoretical positions, best practice still requires that such frameworks are stated (Patton, 2015; Hammarberg et al., 2016). None of the qualitative studies provided this information, disallowing any meaningful assessment of the quality and credibility of the results in the absence of an understanding of the process and perspective the researcher brought to the study (Patton, 2015).

While some benefits to PWB were demonstrated in the included studies, it should be mentioned that the degree to which carer-specific needs were addressed in the designs of the interventions varied across studies. This has likely affected outcomes. For example, 2 of the 12 included studies (Bentley et al., 2014; Creemers et al., 2014) incorporated solely patient-based interventions with carer PWB only a secondary focus. Unsurprisingly, these studies reported no (Creemers et al., 2014) or less PWB improvements than patients (Bentley et al., 2014).

A broader range of PWB benefits were evident in the qualitative data when compared with quantitative outcomes. Quantitative studies largely used hedonic PWB outcome measures (e.g., mental health), with benefits suggested only in 2 (van Groenestijn et al., 2015; Pagnini et al., 2021) of 10 studies despite methodological concerns. Some qualitative studies reported benefits to a range of both hedonic and eudaimonic aspects of PWB although they did not specifically or clearly demonstrate systematic decreases in clinical symptoms of mental health problems such as depression, anxiety, and carer burden. Psychological morbidity remains a serious problem in the MND carer population (Aoun et al., 2020a), and well-designed, high-quality research demonstrating clear reductions in depression, anxiety, and burden are urgently required. Interestingly, some of the six studies using mixed-methods designs demonstrated variability in quantitative vs. qualitative outcomes. These studies reported no or limited PWB benefits in the quantitative measures yet demonstrated benefits or mixed results from the qualitative arm. This lack of concordance between quantitative and qualitative data is noteworthy in an MMSR (Stern et al., 2020), possibly demonstrating that the varying methodologies in these studies addressed different aspects of the phenomena of interest or sensitivity issues with the outcome measures selected. Lack of power in quantitative study arms (because of small study sample sizes) is also a very plausible explanation for the lack of concordance.

Strengths and limitations

This systematic review took an integrative approach, acknowledging the value of hedonic and eudaimonic aspects of PWB as both are associated with positive outcomes (Vazquez et al., 2009). This broad approach was advantageous given the early development of the field of inquiry, increased potential for greater inclusiveness of studies, enabled the capture of a wider range of potential intervention effects, and countered the limited view of simply defining

carer PWB in terms of responses to psychological morbidity questions. Furthermore, this process encompassed quantitative and qualitative approaches, and the discordance between these types of data highlighted the importance of considering different aspects of PWB. Data pooling for meta-analysis was not possible due to the heterogeneity of the interventions, outcomes, and measurements, as well as the low number of studies.

Future recommendations

Future studies should increase attention to the methodological features associated with high-quality research which can be facilitated by using existing frameworks and checklists (e.g., JBI). Researchers should be aware of the variety of approaches to the development of health interventions prior to designing treatments (O’Cathain et al., 2019). This knowledge can guide the development, piloting, evaluation, and implementation components of future research, while ensuring methodological rigor. Moreover, this process values what we know about MND carer needs (e.g., Aoun et al., 2017) and can facilitate the theory identification and development necessary in a new and rapidly developing field (O’Cathain et al., 2019).

Carefully designed mixed-methods studies can be informative as different forms of research may be better suited to capture diverse aspects of carer PWB. The inclusion of both hedonic and eudaimonic aspects of PWB may assist in obtaining a broader range of potential intervention effects and encapsulate benefits more relevant to the individual’s needs. The mixed results of some studies highlight the potential value of tailoring interventions. A generic approach may not address the PWB requirements of MND carers whose individual needs may change over the disease course. Differentiating carer needs have been identified at diagnosis, maintenance, terminal, and bereavement stages (Poppe et al., 2020). A “goodness-of-fit” approach, where individual risks and needs are assessed and matched to tailored treatment, has been proposed (Aoun et al., 2020b).

The heterogeneity among MND carers participating in the studies in this review further highlights the importance of assessing individual carer needs and tailoring interventions accordingly. Factors to consider when designing carer interventions include identifying those at higher risk of poorer PWB outcomes (Aoun et al., 2020a) and MND patient stage (Poppe et al., 2020). A narrative review of variables relevant to intervention design concluded that MND carers with limited problem-solving and coping skills, as well as those looking after patients with mental health issues, severe clinical symptoms, or neurobehavioral problems, were also at increased risk of poorer PWB (Gluyas et al., 2017). Poorer bereavement outcomes have also been noted in MND carers with mental health problems, lower family functioning, spousal relationships, and inadequate support (Aoun et al., 2020a). Moreover, as most MND carers in this review were spousal, future intervention designs may consider also promoting dyadic coping, which is defined as “partners’ coping responses to each other’s stress resulting from circumstances outside the relationship,” especially as this variable is linked with improved PWB (Falconier and Kuhn, 2019, p. 2).

Additionally, psychological research has recently focused on the recognition and treatment of trans-diagnostic treatment targeting shared mechanisms underlying different mental health problems (Sauer-Zavala et al., 2019). This could be particularly relevant for MND carers for whom symptoms of one mental health problem (e.g., depression) may exist with related issues

(e.g., burden) and treatment would involve a consideration of an individual's preexisting strengths and weaknesses.

Given the treatment access issues reported in MND research with carers (Aoun et al., 2021), e/telehealth interventions should be given more consideration. Finally, the range of countries (in this review: $n = 4$, developed, Western) participating in these interventions should be broadened to increase generalizability.

Conclusion

There is a significant body of research highlighting the negative PWB impacts of the MND caregiving role. However, there remains only a limited number of studies specifically designed to improve the PWB of carers. Benefits to carer PWB were only demonstrated in a small number of studies. Most studies suffered from substantial methodological problems, rendering the overall evidence base as low. Further attention and resources need to be applied to this domain, particularly as MND carers remain neglected, mental health consequences can be severe, and may directly impact the person living with MND (Aoun et al., 2020a).

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