

Review Article

Cite this article: Cafarella P, Effing T, Chur-Hansen A (2024) Identifying the active content of interventions targeting the psychological well-being of carers of people with motor neuron disease: A systematic review. *Palliative and Supportive Care* 22(5), 1469–1488. <https://doi.org/10.1017/S1478951524000877>

Received: 30 October 2023

Revised: 15 March 2024

Accepted: 30 April 2024




Keywords:

Motor neuron disease; carers; psychological well-being

Corresponding author: Paul Cafarella;

Email: paul.cafarella@adelaide.edu.au

Identifying the active content of interventions targeting the psychological well-being of carers of people with motor neuron disease: A systematic review

Paul Cafarella, B.A.^{1,2,3,4} , Tanja Effing, PH.D.^{1,4}  and Anna Chur-Hansen, B.A., PH.D.¹ 

¹School of Psychology, Faculty of Health and Medical Sciences, University of Adelaide, Adelaide, SA, Australia;

²Department of Respiratory Sleep Medicine and Ventilation, Flinders Medical Centre, Southern Adelaide Local Health Network, Bedford Park, SA, Australia; ³College of Nursing and Health Sciences, Flinders University of South Australia, Bedford Park, SA, Australia and ⁴College of Medicine and Public Health, Flinders University of South Australia, Bedford Park, SA, Australia

Abstract

Objectives. The primary aim of this research was to use a taxonomy of behavior change techniques (BCTTv1) to identify, map, and describe the active components of intervention and comparator groups in studies evaluating the psychological well-being (PWB) of motor neuron disease (MND) carers. Secondary aims were to (a) identify absent active ingredients and (b) explore whether variability in the effectiveness of interventions targeting the PWB of MND carers could be better explained through improved characterization of the active content of these interventions.

Methods. Mixed-methods systematic review based on Joanna Briggs Institute methodology for quantitative, qualitative, and mixed-methods reviews and Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. Content-coding of interventions targeting the PWB of MND carers using BCTTv1 was conducted.

Results. Sixteen manuscripts describing 14 studies were included. Forty-one of the possible 93 behavior change techniques (BCTs, 44%) were identified as active ingredients, while 52 BCTs (56%) were absent. BCTs were identified in all 14 intervention groups and 4 control groups. Four of the 16 overall BCTTv1 categories were absent. Eleven of the 14 studies demonstrated PWB benefits from their interventions.

Significance of results. Identified and absent BCTs and BCTTv1 categories were mapped for all study groups, enabling a transparent characterization of active intervention content associated with positive PWB outcomes. Directions to improve interventions in this nascent field of research included the investigation of relevant untested BCTs in this population and the management of reporting and methodological quality issues.

The negative impacts of caring for a person with motor neuron disease (MND) include depression, anxiety, prolonged grief, increased burden, decreased social contacts, and reduced quality of life (Aoun et al. 2020; Gluyas et al. 2017; Harris et al. 2017). There is growing recognition of the need for interventions that improve the psychological well-being (PWB) of individuals caring for people with MND. Our recent systematic review on this topic identified 12 studies using non-pharmacological interventions (NPIs) specifically designed to improve hedonic (subjective) and/or eudaimonic (functioning) aspects of carer PWB. The hedonic approach includes strategies to improve the emotional state of MND carers by encouraging positive emotions and reducing psychological distress, while eudaimonic methods emphasize the functioning aspects of PWB, such as self-acceptance, environmental mastery, positive relationships, personal growth, purpose in life, and autonomy (Cafarella et al. 2022; Ryan and Deci 2001). NPIs are usually complex, involve numerous interacting components, and incorporate behavioral and psychosocial processes involving specific actions or behaviors required for prevention, care, or cure (Craig et al. 2013; Ninot 2021). Commonly, NPIs are poorly reported compared with pharmacological interventions, with less detail on the intervention content (Hoffmann et al. 2013; McCleary et al. 2013).

Recently, steps have been made to progress the evidence base for developing behavior change interventions (Leventhal et al. 2008; Michie et al. 2011a). A taxonomy of behavior change techniques (BCTTv1), listing and describing 93 intervention components from 16 categories, has been developed to aggregate active ingredients that may be included in NPIs, providing a framework to categorize intervention content. Behavior change techniques (BCTs) refer to

replicable intervention components designed to alter or redirect causal processes regulating behavior (Michie et al. 2013).

In our recent systematic review, in which we evaluated methods and outcomes of NPIs targeting the PWB of MND carers, we noted that the active content of NPIs was not or insufficiently described (Cafarella et al. 2022). This can be an encumbrance to replication and understanding of the critical content factors contributing to the effectiveness of the NPIs. More transparency of the intervention components leading to positive effects is necessary. Applying the BCTTv1 (Michie et al. 2013) framework to PWB interventions for MND carers may facilitate a shared understanding of the specific strategies used within and across studies of interventions targeting the PWB of MND carers and may advance research and practice in this domain. In addition, it may enable theoretical links and provide further important methodological detail to promote accurate replication and practice, potentially enhancing effectiveness. It also allows the evidence derived from NPIs to be appropriately synthesized.

In alignment with the strategy of the Iterative Protocol for Evidence Base Accumulation (IPEBA), the content of all BCTs used in each study arm should be assessed independently, enabling more accurate isolation of the active ingredients and greater insight regarding the relationship between outcomes and intervention content (Peters et al. 2015). Without this insight, intervention replication and implementation are impeded, and the reporting of effect sizes for new interventions may remain uninterpretable (Michie et al. 2011b). Our recent review also revealed that information regarding the support provided to control group participants of PWB interventions for MND carers lacked detail (Cafarella et al. 2022).

A more explicit theory and evidence-based approach to designing PWB interventions may identify the need to include BCTs that have not previously been used with MND carers. BCTs that have been proven effective in interventions with similar populations (e.g., carer of chronic neurological conditions, Fakolade et al. 2020) could possibly be used to optimize existing or future PWB interventions for MND carers.

For this study, we conducted detailed content-coding of interventions targeting the PWB of MND carers using BCTTv1. The primary aim of this research was to use the BCTTv1 to identify, map, and describe the active components of intervention and comparator groups of studies evaluating the PWB of MND carers. Secondary aims were to (a) identify which active ingredients were not being utilized and (b) to explore whether variability in the effectiveness of interventions targeting the PWB of MND carers (conducted in our previous systematic review) could be better explained through improved characterization of the active content of these interventions.

Method

This study was conducted following the Joanna Briggs Institute (JBI) methodology for mixed-methods systematic reviews (MMSRs) (Aromataris and Munn 2020) and Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher et al. 2009).

Search strategy and study selection

The search strategy was constructed to capture a broad range of intervention studies designed to improve the PWB of MND carers. The search strategy involved 3 distinct steps and used the keywords

listed in Text Box 1. The first step included a search of CINAHL, PubMed, ProQuest, Scopus, and PsycINFO databases, followed by an analysis of the text words in the title, abstract, and index terms describing the article. Identified keywords and index terms were then included in the second search across the databases. Stage 3 involved a search for further studies by analyzing the reference lists of the included manuscripts. Studies were limited to those published in English from 1990 to December 2022. Manuscripts published before 1990 were excluded as our previous systematic review of interventions designed to improve the PWB of MND carers did not locate any studies before 2013. Included studies could be quantitative, qualitative, or mixed-methods. Interventions needed to be carer-based and designed to improve carer PWB. Interventions could be delivered to individuals or groups, and any delivery mode, duration or dose was acceptable. Spousal, family, or other informal carers of people with MND were included, but professional or paid carers were not. Carers of individuals from all MND disease stages and care settings were included.

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 behavio*r 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.

Outcomes of interest were any associated with the PWB (inclusive of hedonic and eudaimonic outcomes) of MND carers. In accordance with the JBI methodology for MMSRs, recommended screening protocols, design-appropriate standardized quantitative and qualitative critical appraisal forms to assess inclusion and methodological quality, and standardized data extraction tools were employed (Stern et al. 2020). Further evaluation of selective reporting bias was conducted by comparing measurements and outcomes in studies with published protocols (where available). In studies without published protocols, methods and results were compared. All studies were evaluated by each of the 3 reviewers (PC, TE, ACH) to determine inclusion, and disagreement was resolved by discussion. The study selection is displayed in Figure 1.

Data collection

The first author (PC) contacted the authors of the included articles to provide a detailed description of the intervention and any comparison groups. In addition, searches were performed to locate any published materials, including descriptive information about the study groups. Descriptive materials could include protocols (published or unpublished), intervention or study manuals, training resources or any other materials detailing the content of study groups. Authors were contacted via email and permitted 8 weeks (with 3 reminders) to provide the requested information.

Data extraction and analysis

Quality assessments and data extraction from the selected studies using design-appropriate JBI tools were conducted by two

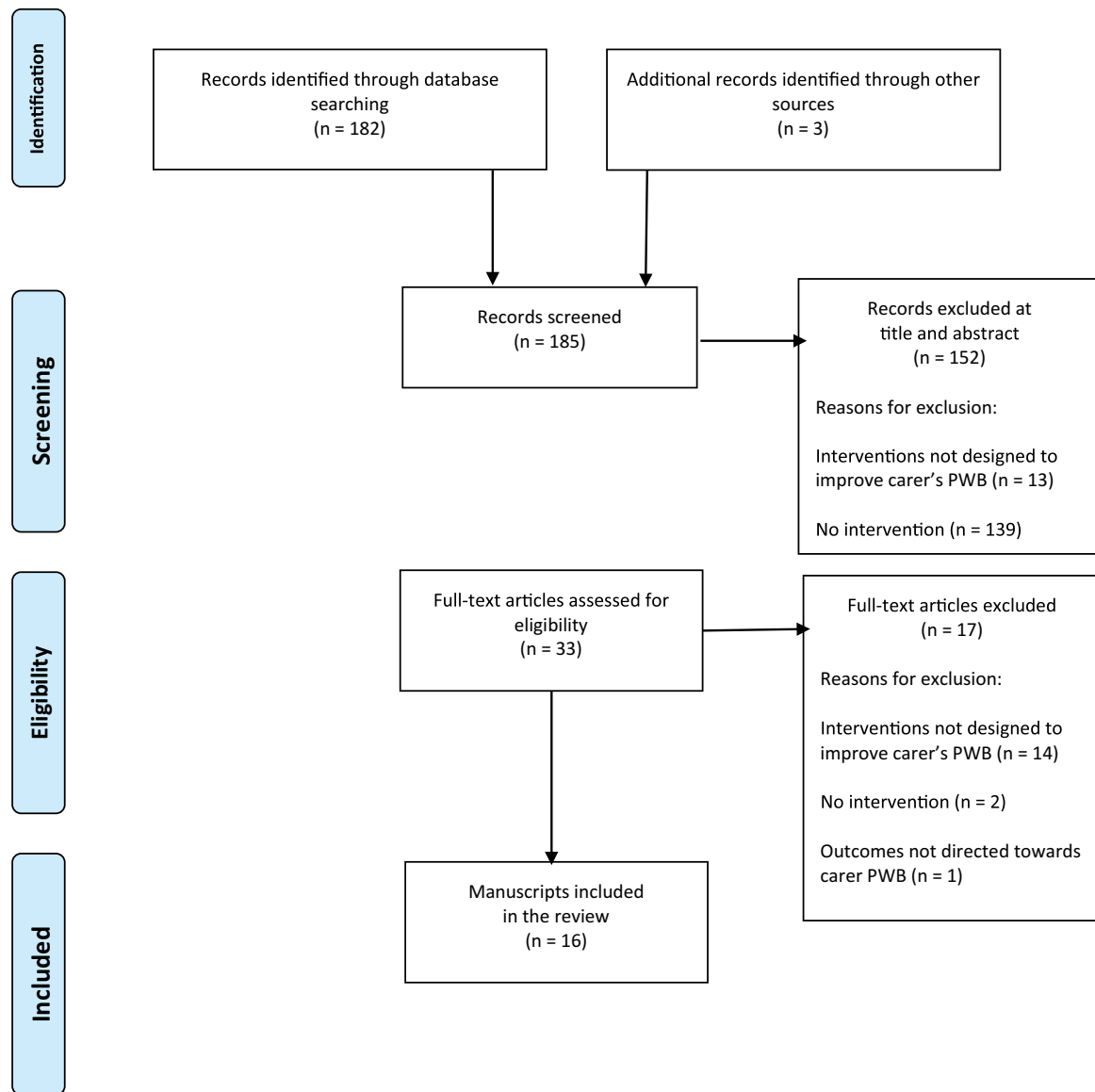


Figure 1. PRISMA flow diagram of manuscript selection.

reviewers (PC and TE: quantitative data; PC and ACH: qualitative data) using agreed narrative evidence descriptors (Aromataris and Munn 2020). Data were presented narratively and synthesized descriptively as recommended by the JBI guidelines for MMSRs (Aromataris and Munn 2020). From our previous study results (Cafarella et al. 2022), we knew a priori that data pooling for a meta-analysis would not be possible due to the heterogeneity of the interventions, outcomes and measurements, and the small number of studies.

Content for all available study groups of the included studies was coded independently by two trained reviewers (PC and research assistant SW – see acknowledgments) using BCTTv1, a valid and reliable method of retrospectively specifying the active content of interventions, which lists and describes 93 intervention components from 16 categories (Michie et al. 2013, 2015) (listed in Table 1). Coder training in using BCTTv1 may increase the validity of coding BCTs and coder competence (Michie et al. 2015). The coders (PC, SW) completed the interactive online training

(Bct-taxonomy.com 2023). In accordance with IPEBA, the BCT content of each study group was assessed independently to increase awareness of the potential bias associated with BCTs used with the control groups, enable more accurate isolation of the active ingredients and greater insight regarding the relationship between outcomes and intervention content (Peters et al. 2015). Both coders discussed any differences in the identification of BCTs until a consensus was reached. A third trained coder (TE) was available for resolution in cases where consensus could not be reached.

Results

Sixteen manuscripts were included (Figure 1), describing 14 unique studies. One study published quantitative and qualitative research components in separate papers (de Wit et al. 2020, 2019), and another study was described in 2 manuscripts (Aoun et al. 2015; Bentley et al. 2014). Six of the 14 studies included mixed-methods, while 4 used solely quantitative methods

Table 1. BCTTv1 categories and BCTs identified per study

BCTTv1 categories	BCTs	Studies														Total No. of studies (/14) where the BCT was identified, (%)	
		Aoun et al. 2017	Aoun et al. 2018	Aoun Bentley et al. 2014	Cipolletta et al. 2018	Creemers et al. 2014	de Wit et al. 2020	de Wit et al. 2019,	Kavanaugh et al. 2020	Marconi et al. 2016	Olesen et al. 2022	Pagnini et al. 2021	Steinhauser et al. 2016	Thomas et al. 2022	Ugalde et al. 2018		van Groenestijn et al. 2015
1. Goals and planning	1.1 Goal setting (behavior)						x		x				x				3 (21.4%)
	1.2 Problem solving	x	x			x	x		x		x	x	x	x	x		11 (78.6%)
	1.3 Goal setting (outcome)								x								1 (7.1%)
	1.4 Action planning	x	x				x				x		x				5 (35.7%)
	1.5 Review behavior goal(s)	x															1 (7.1%)
	1.6. Discrepancy between current behavior and goal																0 (0%)
2. Feedback and monitoring	1.7. Review outcome goal(s)																0 (0%)
	1.8. Behavioral contract																0 (0%)
	1.9 Commitment						x										1 (7.1%)
	2.1 Monitoring of behavior by others without feedback														x		1 (7.1%)
	2.2 Feedback on behavior	x				x	x		x				x				5 (35.7%)
	2.3 Self-monitoring of behavior						x				x						2 (14.3%)
	2.4 Self-monitoring of outcome(s) of behavior																0 (0%)
	2.5 Monitoring of outcome(s) of behavior without feedback	x		x	x	x	x		x		x	x	x		x	x	11 (78.6%)
																	(Continued)

Table 1. (Continued.)

BCTTv1 categories	Studies															Total No. of studies (/14) where the BCT was identified, (%)
	Aoun et al. 2017	Aoun et al. 2018	Aoun Bentley et al. 2014	Cipolletta et al. 2018	Creemers et al. 2014	de Wit et al. 2020	Kavanaugh et al. 2020	Marconi et al. 2016	Olesen et al. 2022	Pagnini et al. 2021	Steinhauser et al. 2016	Thomas et al. 2022	Ugalde et al. 2018	van Groenestijn et al. 2015		
BCTs																0 (0%)
2.6. Biofeedback																
2.7 Feedback on outcome(s) of behavior																
3. Social support	3.1 Social support (unspecified)	x	x	x	x	x	x	x	x	x	x	x	x	x	x	14 (100%)
	3.2 Social support (practical)	x	x		x	x	x	x	x			x				8 (57.1%)
	3.3 Social support (emotional)	x	x	x	x	x	x	x	x	x		x	x			9 (64.3%)
4. Shaping knowledge	4.1 Instruction on how to perform the behavior						x	x	x	x	x	x	x	x	x	8 (57.1%)
	4.2 Information about antecedents															0 (0%)
	4.3 Reattribution									x	x					2 (14.3%)
4.4 Behavioral experiments																
5. Natural consequences	5.1 Information about health consequences	x	x				x									3 (21.4%)
	5.2 Salience of consequences															0 (0%)
	5.3 Information about social and environmental consequences	x	x					x	x							4 (28.6%)
5.4 Monitoring of emotional consequences																
5.5 Anticipated regret																
							x		x	x	x					4 (28.6%)
																0 (0%)

(Continued)

Table 1. (Continued.)

BCTTv1 categories	Studies														Total No. of studies (/14) where the BCT was identified, (%)	
	Aoun et al. 2017	Aoun et al. 2018	Aoun Bentley et al. 2014	Aoun Bentley et al. 2015/2019	de Wit et al. 2020	de Wit et al. 2019, 2020	Creemers et al. 2014	Cipolletta et al. 2018	Kavanaugh et al. 2020	Marconi et al. 2016	Olesen et al. 2022	Pagnini et al. 2021	Steinhauser et al. 2016	Thomas et al. 2022		Ugalde et al. 2018
BCTs																
	x				x						x					3 (21.4%)
6. Comparison of behavior	6.1 Demonstration of the behavior				x				x	x	x	x		x	x	7 (50%)
	6.2 Social comparison				x			x	x					x	x	6 (42.9%)
	6.3 Information about others' approval															0 (0%)
7. Associations	7.1 Prompts/cues															0 (0%)
	7.2 Cue signaling reward															0 (0%)
	7.3 Reduce prompts/cues															0 (0%)
	7.4 Remove access to the reward															0 (0%)
	7.5 Remove aversive stimulus															0 (0%)
	7.6 Satiation															0 (0%)
	7.7 Exposure															0 (0%)
8. Repetition and substitution	7.8 Associative															0 (0%)
	8.1 Behavioral practice/rehearsal				x			x	x	x	x	x				5 (35.7%)
	8.2 Behavior substitution									x						1 (7.1%)
	8.3 Habit formation								x							1 (7.1%)

Table 1. (Continued.)

BCTTv1 categories	Studies														Total No. of studies (/14) where the BCT was identified, (%)
	Aoun et al. 2017	Aoun et al. 2018	Bentley et al. 2014	Cipolletta et al. 2018	Creemers et al. 2014	de Wit et al. 2019, 2020	Kavanaugh et al. 2020	Marconi et al. 2016	Olesen et al. 2022	Pagnini et al. 2021	Steinhauser et al. 2016	Thomas et al. 2022	Ugalde et al. 2018	van Groenestijn et al. 2015	
BCTs															
8.4 Habit reversal								x						1 (7.1%)	
8.5 Overcorrection														0 (0%)	
8.6 Generalization of target behavior														0 (0%)	
8.7 Graded tasks														0 (0%)	
9. Comparison of outcomes	9.1 Credible source	x		x	x	x	x	x			x	x	x	11 (78.6%)	
	9.2 Pros and cons								x					1 (7.1%)	
	9.3 Comparative imagining of future outcomes								x					1 (7.1%)	
10. Reward and threat	10.1 Material incentive (behavior)													0 (0%)	
	10.2 Material reward (behavior)													0 (0%)	
	10.3 Non-specific reward													0 (0%)	
	10.4 Social reward													0 (0%)	
	10.5 Social incentive													0 (0%)	
	10.6 Non-specific incentive													0 (0%)	
	10.7 Self-incentive													0 (0%)	
	10.8 Incentive (outcome)													0 (0%)	
	10.9 Self-reward													0 (0%)	
(Continued)															

(Continued)

Table 1. (Continued.)

BCTTv1 categories	Studies															Total No. of studies (/14) where the BCT was identified, (%)	
	Aoun et al. 2017	Aoun et al. 2018	Aoun et al. 2014	Bentley et al. 2015/2014	Cipolletta et al. 2018	Creemers et al. 2014	de Wit et al. 2019, 2020	Kavanaugh et al. 2020	Marconi et al. 2016	Olesen et al. 2022	Pagnini et al. 2021	Steinhauser et al. 2016	Thomas et al. 2022	Ugalde et al. 2018	van Groenestijn et al. 2015		
11. Regulation	BCTs																0 (0%)
	10.10 Reward (outcome)																0 (0%)
	10.11 Future punishment																0 (0%)
	11.1 Pharmacological support																0 (0%)
	11.2 Reduce negative emotions	x		x	x		x	x	x	x	x	x		x	x		11 (78.6%)
	11.3 Conserving mental resources	x							x								2 (14.3)
	11.4 Paradoxical instructions																0 (0%)
	12. Antecedents																0 (0%)
	12.1 Restructuring the physical environment																0 (0%)
	12.2 Restructuring the social environment	x								x							2 (14.3%)
	12.3 Avoidance/reducing exposure to cues for the behavior																0 (0%)
	12.4 Distraction																0 (0%)
	12.5 Adding objects to the environment	x	x														2 (14.3%)
	12.6 Body changes								x								1 (7.1%)
	13. Identity																2 (14.3%)
	13.1 Identification of self as role model			x						x							2 (14.3%)

Table 1. (Continued.)

BCTTv1 categories	BCTs	Studies														Total No. of studies (/14) where the BCT was identified, (%)
		Aoun et al. 2017	Aoun et al. 2018	Bentley et al. 2014	Cipolletta et al. 2018	Creemers et al. 2014	de Wit et al. 2020	Kavanaugh et al. 2020	Marconi et al. 2016	Olesen et al. 2022	Pagnini et al. 2021	Steinhauser et al. 2016	Thomas et al. 2022	Ugalde et al. 2018	van Groenestijn et al. 2015	
14. Scheduled consequences	13.2 Framing/reframing			x	x		x		x		x				x	7 (50%)
	13.3 Incompatible beliefs								x							1 (7.1%)
	13.4 Valued self-identify	x					x		x		x					4 (28.6%)
	13.5 Identity associated with changed behavior				x				x		x					3 (21.4%)
	14.1 Behavior cost															0 (0%)
	14.2 Punishment															0 (0%)
	14.3 Remove reward															0 (0%)
	14.4 Reward approximation															0 (0%)
	14.5 Rewarding completion															0 (0%)
	14.6 Situation-specific reward															0 (0%)
14.7 Reward incompatible behavior															0 (0%)	
14.8 Reward alternative behavior															0 (0%)	
14.9 Reduce reward frequency															0 (0%)	
14.10 Remove punishment															0 (0%)	
(Continued)																

(Continued)

Table 1. (Continued.)

BCTTv1 categories	Studies														Total No. of studies (/14) where the BCT was identified, (%)
	Aoun et al. 2017	Aoun et al. 2018	Aoun Bentley et al. 2014	Cipolletta et al. 2018	Creemers et al. 2014	de Wit et al. 2019, de Wit et al. 2020	Kavanaugh et al. 2020	Marconi et al. 2016	Olesen et al. 2022	Pagnini et al. 2021	Steinhauser et al. 2016	Thomas et al. 2022	Ugalde et al. 2018	van Groenestijn et al. 2015	
15. Self-belief	BCTs														
	15.1 Verbal persuasion about capability						x								1 (7.1%)
	15.2 Mental rehearsal of successful performance														0 (0%)
	15.3 Focus on past success										x				1 (7.1%)
	15.4 Self-talk									x					2 (14.3%)
16. Covert learning	16.1 Imaginary punishment														0 (0%)
	16.2. Imaginary reward														0 (0%)
	16.3. Vicarious consequences														0 (0%)
Total BCTs identified	17	8	5	5	10	7	23	12	17	23	12	8	10	10	7
Total categories identified	8	4	5	5	6	4	11	8	9	11	10	7	6	7	7

Key: x = BCT identified

(Creemers et al. 2014; Kavanaugh et al. 2020; Pagnini et al. 2021; van Groenestijn et al. 2015), and another 4 employed a qualitative approach (Cipolletta et al. 2018; Marconi et al. 2016; Olesen et al. 2022; Thomas et al. 2022). Four randomized controlled trials (RCTs) were included (Creemers et al. 2014; de Wit et al. 2020; Pagnini et al. 2021; van Groenestijn et al. 2015). All papers were published between 2013 and 2022 (10 of the 16 in the last 5 years) (Tables 2 and 3).

Ten of the 14 studies were conducted in 3 countries. Four studies originated in Australia (Aoun et al. 2015, 2017, 2018; Bentley et al. 2014; Ugalde et al. 2018), 3 in the U.S.A. (Creemers et al. 2014; Kavanaugh et al. 2020; Steinhauser et al. 2016), and 3 in Italy (Cipolletta et al. 2018; Marconi et al. 2016; Pagnini et al. 2021). Two studies were conducted in the Netherlands (de Wit et al. 2020, 2019; van Groenestijn et al. 2015), 1 in Denmark (Olesen et al. 2022), and another in India (Thomas et al. 2022). Overall, 530 MND carers were studied in the quantitative component of this review and 256 in the qualitative component (Tables 1 and 2).

Eleven of the 14 studies either had separately published protocols ($n = 6$) or the authors provided additional materials (including 1 separate unpublished protocol). The remaining studies ($n = 7$) described the protocol within the text of the original manuscript. An email response with additional information was received from the authors of 10 of the 14 included studies.

Forty-one of the possible 93 BCTs (44%), as defined by BCTTv1 (Michie et al. 2013), were identified as active ingredients. We identified BCTs in all 14 intervention groups (Figure 2) and the 4 control groups. The number of identifiable BCTs ranged from 5 to 23 (mean: 12.1, SD: 5.78) in the intervention groups and 1 to 3 (mean: 1.8, SD: 0.96) in the control groups.

Among the BCTs identified for the 14 intervention groups, the most prevalent were 3.1 Social support (unspecified) (100%), 1.2 Problem-solving, 2.5 Monitoring of outcome(s) of behavior without feedback, 9.1 Credible source (85.7%), and 11.2 Reduce negative emotions (78.6%) (Table 1 and Figure 2). BCTs were identified in 4 of the control groups: 2.5 Monitoring of outcome(s) of behavior without feedback ($n = 4$), 3.1 Social support (unspecified) ($n = 2$), and 9.1 Credible source ($n = 1$) (Table 1). Fifty-two of the possible 93 BCTs were not identified in either the intervention or control group descriptions.

The coding exercise identified 12 of the 16 overall BCT categories described in BCTTv1 (Michie et al. 2013). The 4 BCT categories not described in the studies were Associations, Reward and threat, Scheduled consequences, and Covert learning (Table 1). The most frequently deployed BCTTv1 categories were Social support (100%), Feedback and monitoring, Comparison of outcomes (85.7%), Goals and Planning, and Regulation (78.6%). The other categories used included Identity (64.3%), Shaping knowledge, Comparisons of behavior (57.1%), Natural consequences (42.9%), Repetition and substitution (35.7%), Antecedents, and Self-belief (28.6%) (Table 1).

Seven of the 10 studies with a quantitative component and 9 of the 10 studies with a qualitative component reported PWB benefit(s) from their interventions. Overall, 11 of the 14 studies demonstrated some benefits. The identified BCTs and BCT categories associated with these studies are detailed in Tables 2 and 3, enabling characterization of the active content of the interventions associated with hedonic and eudaimonic PWB outcomes. Among the 11 studies reporting intervention benefits, the most prevalent identified BCTs were 3.1 Social support (unspecified) (100%), 1.2 Problem-solving (81.8%), 11.2 Reduce negative emotions (81.8%), 2.5 Monitoring of outcome(s) of behavior without

feedback (72.7%), 4.1 Instruction on how to perform a behavior (72.7%), 3.3 Social support (emotional) (72.7%), and 9.1 Credible source (72.7%) (Tables 2 and 3). A higher number of BCT categories was identified in studies with intervention benefits (mean: 7.9, SD: 2.21) compared to those not demonstrating PWB gains (mean: 5.3, SD: 1.53) (Tables 2 and 3).

Table 2 provides a summary methodological quality score (range: 0–21) for the quantitative studies, calculated using the JBI critical appraisal tools (median: 9, IQR: 7.75). The 4 RCTs were the only studies that achieved summary quality scores above 50% (score range between 57.1 and 76.2%). Table 3 presents a summary methodological quality score (range: 0–13) for the qualitative studies, calculated using the JBI critical appraisal tools (median: 8, IQR: 3.75). Four studies achieved summary quality scores below 50% (score range: 7.7–46.2%). Six studies scored between 53.9 and 100%.

Discussion

This is the first study to comprehensively identify and map the active components of interventions (and comparator groups) targeting the PWB of MND carers using the BCTTv1 taxonomy. We identified 14 studies reporting interventions targeting MND carer PWB, of which 4 were RCTs. Fifty-two of the 93 BCTs listed in BCTTv1 were not identified in the reviewed studies, nor were 4 of the 16 BCT categories, highlighting the untested intervention content in this field. The results enabled a more transparent comprehension and characterization of the active content of the interventions demonstrating hedonic and eudaimonic PWB benefits for MND carers. This review also provides directions to improve interventions in this developing field of research.

The BCT category of “Social support” was identified as an intervention component in each study. Among the BCTs in this category, Social support (unspecified) (100%) was the most frequently deployed, with Social support (emotional) (64.3%) and Social support (practical) (57.1%) regularly used. In studies demonstrating PWB benefits for carers, Social support (emotional) (72.7%) and Social support (practical) (63.6%) were commonplace. However, BCTs associated with the social support category were not among the most frequently identified in a systematic review of BCTs used in health interventions for persons with chronic neurological conditions and their caregivers (Fakolade et al. 2020). Nevertheless, the need for social support for MND carers has been demonstrated in several studies, with practical aspects such as equipment provision, demonstration and support identified as pragmatic needs (Aoun et al. 2021, 2017). A lack of emotional support has also been expressed as a notable problem for MND carers (Aoun et al. 2021, 2020). The form in which social support is manifested appears relevant, as professional support is not commonly evaluated as helpful by MND carers (Aoun et al. 2020). Peer support groups have demonstrated PWB benefits among carers of people with progressive neurological illnesses (Pasquini et al. 2022), while peer support groups for MND carers improved the PWB of one cohort but not another (Marconi et al. 2016). Although peer support may facilitate the development of coping strategies mediating hedonic PWB variables such as carer stress (Pearlin et al. 1990), some authors have argued a “goodness-of-fit” approach to carer support needs, incorporating the risk of mental health problems (Aoun et al. 2020) and MND patient stage (Poppe et al. 2020) may be more effective than a “one-size-fits-all” method (Aoun et al. 2021). While the need for social support is evident, the impact of this active ingredient on MND carer PWB outcomes is difficult to determine from our study

Table 2. Synthesized narrative findings and identified BCTs in studies with a quantitative component

Intervention, author, design	Q	Narrative synthesis of PWB benefits (hedonic & eudaimonic categories)	Identified BCTs, total BCTs, total No. of BCT categories
Carer Support Needs Assessment Tool (Aoun et al. 2017) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 24)	8	*Reductions in the percentage of participants with support needs Hedonic: subjective well-being, emotions (feelings and thoughts) Eudaimonic – environmental mastery, positive relations with others, autonomy *Methodological quality of the study was appraised as low.	1.2 Problem solving, 1.4 Action planning, 1.5 Review behavior goals, 2.2 Feedback on behavior, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 5.1 Information about health consequences, 5.3 Information about social and environmental consequences, 5.6 Information about emotional consequences, 9.1 Credible source, 11.2 Reduce negative emotions, 11.3 Conserving mental resources, 12.2 Restructuring the social environment, 12.5 Adding objects to the environment, 13.4 Valued self-identity (Total BCTs = 17, Categories = 8)
Person-centered model of care – MND Advisory Service (Aoun et al. 2018) Observational survey (n = 117)	2	* >80% of respondents reported hedonic and eudaimonic PWB benefits (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 Eudaimonic – environmental mastery *Methodological quality of the study was appraised as very low.	1.2 Problem solving, 1.4 Action planning, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 5.1 Information about health consequences, 5.3 Information about social and environmental consequences, 12.5 Adding objects to the environment (Total BCTs = 8, Categories = 4)
Dignity therapy (DT) (Bentley et al. 2014; Aoun et al. 2015) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 18)	9	*No significant hedonic PWB benefits were demonstrated in caregiver anxiety, depression, burden, and hopefulness. *No clear hedonic or eudaimonic benefits demonstrated from the family feedback questionnaire. *Methodological quality of the study was appraised as low.	2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 9.1 Credible source, 11.2 Reduce negative emotions, 13.2 Framing/reframing (Total BCTs = 5, Categories = 5)
Case management (Creemers et al. 2014) RCT (n = 126)	16	*No significant hedonic or eudaimonic PWB benefits demonstrated for caregiver burden or level of satisfaction. *Appraised as methodologically sound.	1.2 Problem solving, 2.2 Feedback on behavior, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 9.1 Credible source (Total BCTs = 7, Categories = 4)
Blended psychosocial support program (based on acceptance commitment therapy) (de Wit et al. 2020) RCT (wait-list) (n = 148)	15	*No significant hedonic benefits demonstrated for distress, burden, or quality of life. *No significant eudaimonic benefits demonstrated for caregiver beliefs about their capacity to obtain respite, perceived control over fulfilling caregiver tasks. *Significant hedonic PWB benefit (Emotions: feelings and thoughts) regarding controlling distressing thoughts about caregiving. *Appraised as methodologically sound.	1.1 Goal-setting (behavior), 1.2 Problem solving, 1.4 Action planning, 1.9 Commitment, 2.2 Feedback on behavior, 2.3 Self-monitoring of behavior, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 4.1 Instruction on how to perform the behavior, 5.1 Information about health consequences, 5.4 Monitoring of emotional consequences, 5.6 Information about emotional consequences, 6.1 Demonstration of behavior, 6.2 Social comparison, 8.1 Behavioral practice/rehearsal, 9.1 Credible source, 11.2 Reduce negative emotions, 13.2 Framing/reframing, 13.4 Valued self-identity, 15.1 Verbal persuasion about capability, 15.4 Self-talk (Total BCTs = 23, Categories = 11)

(Continued)

Table 2. (Continued.)

Intervention, author, design	Q	Narrative synthesis of PWB benefits (hedonic & eudaimonic categories)	Identified BCTs, total BCTs, total No. of BCT categories
YCare training protocol for youth carers (based on the tenets of the Individual and Family Self-Management Theory) (Kavanaugh et al. 2020) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 19)	2	*Eudaimonic PWB (environmental mastery) effect demonstrated with significant increase in confidence with several care tasks (use of respiratory equipment, communication systems, power chair use). *Methodological quality appraised as very low.	1.1 Goal-setting (behavior), 1.2 Problem solving, 1.3 Goal setting (outcome), 2.2 Feedback on behavior, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 4.1 Instruction on how to perform the behavior, 6.1 Demonstration of behavior, 6.2 Social comparison, 8.1 Behavioral practice/rehearsal, 9.1 Credible source, 11.2 Reduce negative emotions (Total BCTs = 12, Categories = 8)
Online non-meditative mindfulness (based on Langerian mindfulness) (Pagnini et al. 2021) RCT (wait-list) (n = 27)	12	*PWB time x group effects demonstrated in favor of the mindfulness intervention (care burden, depression, anxiety, role limitations due to personal or emotional problems, energy/fatigue, emotional well-being) corresponding to hedonic PWB categories of subjective well-being, mental health status, emotions (feelings and thoughts) *Methodological quality of the study was appraised as low to moderate	1.2 Problem solving, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 4.1 Instruction on how to perform the behavior, 4.3 Reattribution, 5.4 Monitoring of emotional consequences, 6.1 Demonstration of behavior, 8.1 Behavioral practice/rehearsal, 11.2 Reduce negative emotions, 13.2 Framing/Reframing, 13.5 Identity associated with changed behavior, 15.4 Self-talk (Total BCTs = 12, Categories = 10)
Chaplain-led, manualized self-disclosure exploring role-related meaning ("Caregiver Outlook") (Steinhauser et al. 2016) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 23)	9	*No significant benefits demonstrated for any of the outcome measures (burden, anxiety, depression, grief, religious coping, preparation and completion of carer role, meaning, peace, and faith) related to hedonic or eudaimonic PWB. *Methodological quality of the study was appraised as low.	1.1 Goal-setting (behavior), 1.2 Problem solving, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 9.1 Credible source, 11.2 Reduce negative emotions, 13.4 Valued self-identity, 15.3 Focus on past success (Total BCTs = 8, Categories = 7)
Mindfulness (Ugalde et al. 2018) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 13)	8	*No significant change on any pre-post hedonic or eudaimonic PWB measures (distress, burden, problem-solving, mindfulness, preparedness) post intervention. *Hedonic (subjective well-being) and eudaimonic (positive relations with others) benefits were seen from the acceptability questionnaire. *Methodological quality of the study was appraised as low.	1.2 Problem solving, 2.1 Monitoring of behavior by others without feedback, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.3 Social support (emotional), 4.1 Instruction on how to perform the behavior, 6.1 Demonstration of behavior, 6.2 Social comparison, 9.1 Credible source, 11.2 Reduce negative emotions (Total BCTs = 10, Categories = 7)
Cognitive-behavior therapy (CBT) (based on the stress-coping model) (van Groenestijn et al. 2015) RCT (n = 15)	14	*Significant benefits to hedonic aspects (subjective well-being, mental health status, emotions (feelings and thoughts) of PWB (mental QoL and caregiver burden) but no effect on distress. *Appraised as methodologically sound	1.2 Problem solving, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 4.1 Instruction on how to perform the behavior, 9.1 Credible source, 11.2 Reduce negative emotions, 13.2 Reframing (Total BCTs = 7, Categories = 7)

Q = Critical appraisal of quality score (21); PWB = psychological well-being; n = sample size.

Table 3. Synthesized narrative findings and identified BCTs in studies with a qualitative component

Intervention, author, design	Q	Narrative synthesis of PWB benefits (hedonic & eudaimonic categories)	Identified BCTs, total BCTs, total No. of BCT categories
Carer Support Needs Assessment Tool (Aoun et al. 2017) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 24)	10	Benefits associated with acknowledgement of the overwhelming caregiving journey, CSNAT practicality and usefulness, validation of caregiver role and empowerment, and reassurance of support. Hedonic: Emotions (feelings & thoughts); Eudaimonic: self-acceptance, environmental mastery, purpose in life, personal growth *Appraised as methodologically sound.	1.2 Problem solving, 1.4 Action planning, 1.5 Review behavior goals, 2.2 Feedback on behavior, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 5.1 Information about health consequences, 5.3 Information about social and environmental consequences, 5.6 Information about emotional consequences, 9.1 Credible source, 11.2 Reduce negative emotions, 11.3 Conserving mental resources, 12.2 Restructuring the social environment, 12.5 Adding objects to the environment, 13.4 Valued self-identity (Total BCTs = 17, Categories = 8)
Person-centered model of care – MND Advisory Service (Aoun et al. 2018) Observational survey (n = 117)	4	Benefits associated with practical and emotional support. Hedonic: mental health status, subjective well-being, emotions (feelings & thoughts); Eudaimonic: environmental mastery, positive relationships, personal growth *Appraised as low methodological quality.	1.2 Problem solving, 1.4 Action planning, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 5.1 Information about health consequences, 5.3 Information about social and environmental consequences, 12.5 Adding objects to the environment (Total BCTs = 8, Categories = 4)
Dignity therapy (DT) (Bentley et al. 2014; Aoun et al. 2015); Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 18)	7	No clearly demonstrated hedonic or eudaimonic benefits *Appraised as low to moderate methodological quality.	2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 9.1 Credible source, 11.2 Reduce negative emotions, 13.2 Framing/reframing (Total BCTs = 5, Categories = 5)
Mutual support groups for ALS family caregivers (partners group & adult children group) (Cipolletta et al. 2018) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 12)	9	Adult children group benefits: acceptance, preparation, rethinking the meaning, support, emotional, and mental improvement; peer sharing; security and reflection; less alone; role competence confirmation; understanding patients' viewpoint; role rethink led to attempts to maintain own space and hobby engagement without guilt. Hedonic: emotions (feelings and thoughts), subjective well-being, mental health status; Eudaimonic: purpose in life, positive relationships, environmental mastery, autonomy *Appraised as methodologically sound.	2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 6.2 Social comparison, 9.1 Credible source, 11.2 Reduce negative emotions, 13.1 Identification of self as a role model, 13.2 Framing/reframing, 13.5 Identity associated with changed behavior (Total BCTs = 10, Categories = 6)
Blended psychosocial support program based on acceptance and commitment therapy (de Wit et al. 2019) RCT (wait-list) Interviews were conducted with purposively sampled 23 of the 148 ALS/PMA caregivers enrolled in the RCT	9	Benefits: 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. Hedonic: Emotions (feelings and thoughts), subjective well-being; Eudaimonic: self-acceptance, environmental mastery, positive relationships, autonomy *Appraised as methodologically sound.	1.1 Goal-setting (behavior), 1.2 Problem solving, 1.4 Action planning, 1.9 Commitment, 2.2 Feedback on behavior, 2.3 Self-monitoring of behavior, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 4.1 Instruction on how to perform the behavior, 5.1 Information about health consequences, 5.4 Monitoring of emotional consequences, 5.6 Information about emotional consequences, 6.1 Demonstration of behavior, 6.2 Social comparison, 8.1 Behavioral practice/rehearsal, 9.1 Credible source, 11.2 Reduce negative emotions, 13.2 Framing/reframing, 13.4 Valued self-identity, 15.1 Verbal persuasion about capability, 15.4 Self-talk (Total BCTs = 23, Categories = 11)

(Continued)

Table 3. (Continued.)

Intervention, author, design	Q	Narrative synthesis of PWB benefits (hedonic & eudaimonic categories)	Identified BCTs, total BCTs, total No. of BCT categories
Meditation based on an adapted mindfulness-based stress reduction protocol (Marconi et al. 2016) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 18)	6	Benefits: Improvements in well-being, relaxation, emotional self-regulation, acceptance, consciousness, breathing issues, sleep cycle, relationships, effectiveness of a group setting with peers Hedonic: Emotions (feelings and thoughts) *Appraised as low methodological quality.	3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 4.1 Instruction on how to perform the behavior, 5.3 Information about social and environmental consequences, 5.4 Monitoring of emotional consequences, 6.1 Demonstration of behavior, 6.2 Social comparison, 8.1 Behavioral practice/rehearsal, 8.2 Behavior substitution, 8.3 Habit formation, 8.4 Habit reversal, 9.1 Credible source, 11.2 Reduce negative emotions, 11.3 Conserving mental resources, 12.6 Body changes, 13.2 Framing/reframing (Total BCTs = 17, Categories = 9)
EMBRACE: A 4-month online palliative rehabilitation program (EMBRACE) to support the ability of carers to handle daily challenges of their role (Olesen et al. 2022) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 10)	13	Benefits: Hedonic: Emotions (feelings and thoughts), subjective well-being Eudaimonic: self-acceptance, environmental mastery, positive relationships, autonomy, personal growth *Appraised as methodologically sound	1.2 Problem solving, 1.4 Action planning, 2.3 Self-monitoring of behavior, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 4.1 Instruction on how to perform the behavior, 4.3 Reattribution, 5.3 Information about social and environmental consequences, 5.4 Monitoring of emotional consequences, 5.6 Information about emotional consequences, 6.1 Demonstration of behavior, 8.1 Behavioral practice/rehearsal, 9.2 Pros and cons, 9.3 Comparative imagining of future outcomes, 11.2 Reduce negative emotions, 12.2 Restructuring the social environment, 13.1 Identification of self as a role model, 13.2 Framing/reframing, 13.3 Incompatible beliefs, 13.4 Valued self-identity, 13.5 Identity associated with changed behavior (Total BCTs = 23, Categories = 11)
Chaplain-led, manualized self-disclosure exploring role-related meaning ("Caregiver Outlook") (Steinhauser et al. 2016) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 14)	6	Benefits: Reflection on role and role change; processing emotions opportunity; stimulating communication; anonymity of conversation Hedonic: Emotions (feelings and thoughts); Eudaimonic: self-acceptance, positive relationships *Appraised as low methodological quality.	1.1 Goal-setting (behavior), 1.2 Problem solving, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 9.1 Credible source, 11.2 Reduce negative emotions, 13.4 Valued self-identity, 15.3 Focus on past success (Total BCTs = 8, Categories = 7)
Psychosocial needs assessment and intervention in low-resource settings based on Rolland's theory of psychosocial typology for chronic illness (Thomas et al. 2022); Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 7)	9	Benefits: confidence to perform the caregiving role Eudaimonic: Environmental mastery, autonomy *Appraised as methodologically sound	1.2 Problem solving, 1.4 Action planning, 2.2 Feedback on behavior, 3.1 Social support (unspecified), 3.2 Social support (practical), 3.3 Social support (emotional), 4.1 Instruction on how to perform the behavior, 6.1 Demonstration of behavior, 6.2 Social comparison, 9.1 Credible source (Total BCTs = 10, Categories = 6)
Mindfulness (Ugalde et al. 2018) Longitudinal, uncontrolled, single treatment group, pretest, posttest (n = 13)	1	Benefits: valued 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 *Appraised as very low methodological quality.	1.2 Problem solving, 2.1 Monitoring of behavior by others without feedback, 2.5 Monitoring of outcome(s) of behavior without feedback, 3.1 Social support (unspecified), 3.3 Social support (emotional), 4.1 Instruction on how to perform the behavior, 6.1 Demonstration of behavior, 6.2 Social comparison, 9.1 Credible source, 11.2 Reduce negative emotions

Q = Critical appraisal of quality score (13); PWB = psychological well-being; n = sample size.

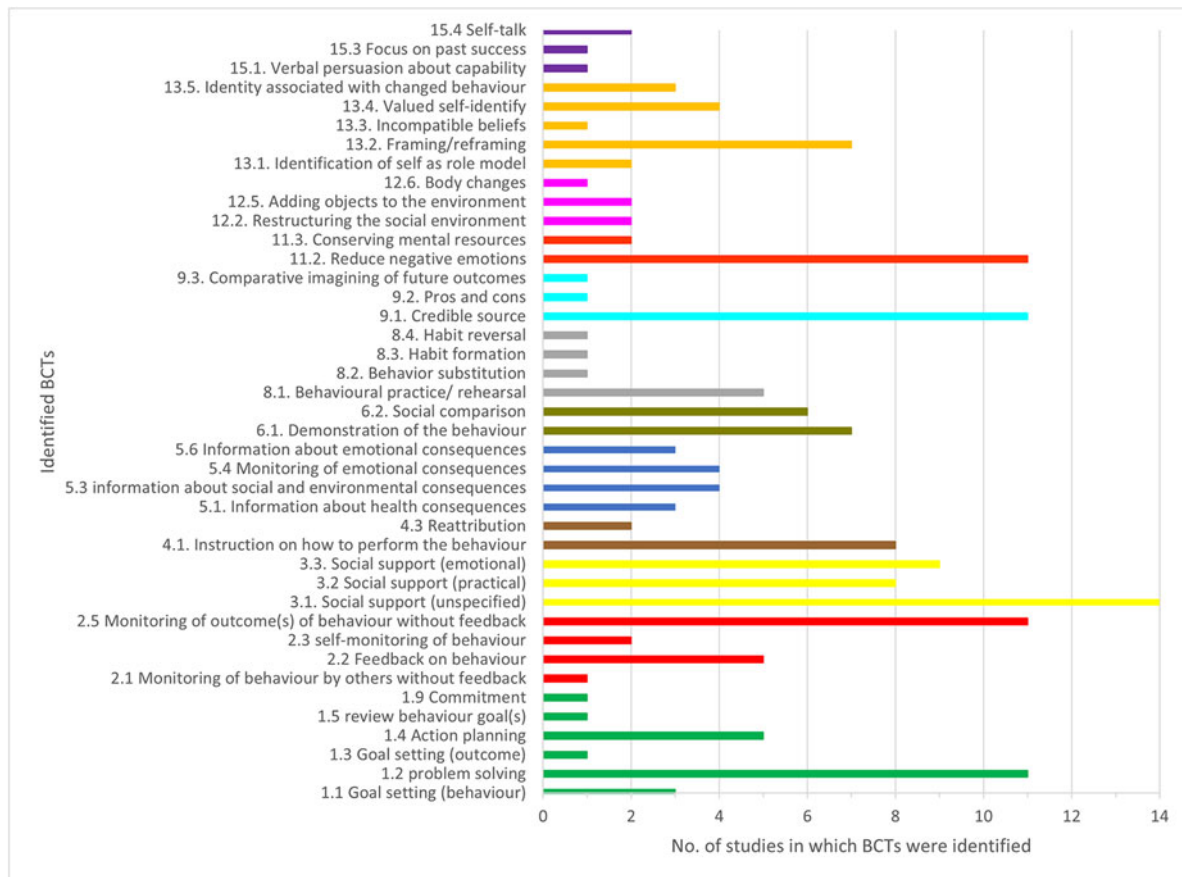


Figure 2. Number of studies in which specific BCTs were identified in the intervention groups.

since this BCT category was identified in all the studies (including those not demonstrating PWB benefits), and 2 of the 4 control groups.

Our results elucidate the potential of exposure of the control group to active intervention ingredients, which may affect study outcomes. While the BCT categories of Social support and Feedback and monitoring were the most frequently deployed, they were also prevalent in the control groups, reinforcing the importance of specifying the active ingredients in all study groups. Further, a closer look at the data indicates that the high frequency of the Feedback and monitoring category is due to the prevalence of the specific ingredient “Monitoring of outcome(s) of behavior without feedback” (85.7%), essentially tracking outcomes for participants in all study groups. Therefore, our results support the IPEBA approach, where the BCT content of each study arm is independently assessed, enabling greater insight into the relationship between outcomes and intervention content (Peters et al. 2015). Results may be rendered uninterpretable in the absence of this insight casting doubt over the accuracy of reported intervention effect sizes (Michie et al. 2011b; Peters et al. 2015). This imprecision can be further exacerbated by poor reporting of the control conditions, as the description may not provide all the included BCTs, potentially biasing the results and interpretation (de Bruin et al. 2016).

“Reduce negative emotions” was another prevalent (81.8%) BCT identified in the intervention studies demonstrating PWB benefits to MND carers. The rationale for including this component is evident from the high rates of mental health issues linked with the

role (Aoun et al. 2020; Gluyas et al. 2017; Harris et al. 2017). Despite the high frequency of this BCT, the results of this study and our previous systematic review (Cafarella et al. 2022) found that quantitative studies using traditional, hedonic PWB outcome measures (e.g., psychological distress scales) rarely demonstrated intervention benefits on these questionnaires. It remains unclear if the mode of measurement or lack of sensitivity associated with specific quantitative, hedonic outcome measures contributed to the lack of positive effect. However, more carer PWB benefits were evident when studies used qualitative data and eudaimonic measures of PWB, demonstrating the value of including these approaches (e.g., sensitivity, responsiveness, and the importance of non-hedonic aspects of PWB) (Cafarella et al. 2022). Eudaimonic outcomes incorporate and value PWB components such as skill development, competency, self-efficacy, and a sense of control, which can help to reduce the guilt associated with their perceived caring ability and burden (Keating et al. 2021; Teahan et al. 2020). An example is Kavanaugh et al.’s (2020) study, which focussed on upskilling young MND carers and demonstrated gains in eudaimonic PWB. The eudaimonic variable of growth is a significant mediator of resilience among carers of people with dementia (O’Dwyer et al. 2016). The BCTTv1 category of Identity was prevalent in our review (64.3%) and relates to the eudaimonic aspects of PWB, demonstrated to contribute positive aspects of caring and positive emotions (Beach et al. 2022; Teahan et al. 2020). The category of Self-belief was less prevalent (28.6%), although perceived competency in caring among those looking after people with dementia has been associated with reduced psychological distress (Bennett 2018;

Moore et al. 2017) and aligns with capabilities perspectives of eudaimonic PWB (Keating et al. 2021). Such perspectives emphasize the importance of a carer's perceived ability and resources to conduct valued caring tasks in their overall PWB (Keating et al. 2021), yet this remains an under-explored area in PWB research with MND carers.

Another frequently deployed BCT identified in positive intervention studies was "Problem-solving" (81.8%), the most prevalent component of the BCTTv1 Goals and Planning category. While the value of goals and planning in initiating and maintaining PWB has been demonstrated in chronic health populations such as spinal cord injuries, older adults, cancer, and arthritis, a recent systematic review found this BCTTv1 category was less prevalent and likely under-utilized in dyadic health interventions for persons with chronic neurological conditions and their carers (Fakolade et al. 2020). Problem-solving seems inherent in the role of an MND carer and appears to be logically linked with PWB and the prevalence of this BCT in positive intervention studies provides tentative support for this plausible connection.

The BCT labelled "Credible source" was one of the most prevalent identified among the studies and was coded frequently in those reporting intervention benefits. Qualified health professionals typically delivered interventions in the included studies. A recent systematic review of BCTs used in health interventions for persons with chronic neurological conditions and their caregivers, also found this BCT to be among the most frequently identified (Fakolade et al. 2020). Characteristically, dyadic psychosocial interventions for neurodegenerative diseases have been delivered by qualified health professionals (Fakolade et al. 2020). However, being a "credible source" may not guarantee that professionals delivering the BCTs were sufficiently trained, as some health professionals did not believe they were adequately trained to deliver the BCTs during the intervention effectively (Fakolade et al. 2020; Hardeman et al. 2008; Keogh et al. 2018).

This study also identified BCTTv1 categories and individual BCTs that were absent or infrequently utilized. Fifty-two of the possible 93 individual BCTs were not identified in either the intervention or control group descriptions. Some of the absent BCTs may not be applicable for implementation with MND carers; however, the majority (55.9%) of these techniques remain untested with this population. The knowledge void regarding the value of absent BCT categories is further exacerbated by a lack of direct comparison with similar populations because there is currently only 1 review of the BCTs in caregivers of chronic neurological conditions (Fakolade et al. 2020). That review included 27 studies and identified 10 of 16 BCTTv1 categories. Twelve of the 16 BCTTv1 categories were identified in our study, while those absent were Associations, Reward and threat, Scheduled consequences, and Covert learning. The potential value of these absent categories and individual BCTs in fostering MND carer PWB is yet to be determined. For example, the Associations category was present in a systematic review of BCTs used in health interventions for persons with chronic neurological conditions and their caregivers (Fakolade et al. 2020). Prompts, cues, and associative learning, individual BCTs from the "Associations" category, have been shown to support the development of habit formation in critical areas such as medication delivery among carers of ageing people (Stawarz 2017). Similarly, in relation to the "Reward and threat" category, various forms of reward have been associated with facilitating the formation of routine-based strategies essential for perceived competence in caregiving, such as medication monitoring (Stawarz 2017). Consistent with our study, the absent

categories in the systematic review of BCTs used in health interventions for persons with chronic neurological conditions and their caregivers, were Reward and threat, Scheduled consequences, and Covert learning. Categories identified in our study, but absent in their research, were Regulation, Identity, and Self-belief (Fakolade et al. 2020). Additionally, the categories of Repetition and substitution, Antecedents, and Self-belief were less prevalent in our study. Without more studies enabling direct comparison with similar reference populations, it is unknown if absent BCT categories and/or absent individual BCTs represent under-utilized intervention components, potential novel interventions, or whether they are less relevant or effective. An alternative explanation may be that the BCTs reported may not accurately represent the intervention content. While 11 of the 14 studies either had separately published protocols or the authors provided additional materials, a known problem in coding exercises is that BCTs are often poorly described or under-reported in published materials (Chakraborty et al. 2022). BCT coding remains dependent upon and therefore limited by the detail provided in the reported content (Fakolade et al. 2020). The reporting of NPIs frequently lacks precision and detail even after authors have been contacted (Hoffmann et al. 2013). Published descriptions may report less than half the BCTs evident from a protocol (Lorenzatto et al. 2013). Hence, the possibility remains that some BCTs may remain uncoded.

While BCTTv1 is a standardized, systematic approach to classifying the active content of interventions and has been described as the most comprehensive and frequently used of such taxonomies (Michie et al. 2013; Wood et al. 2015), there is always some degree of subjectivity and variability in the application of taxonomies (Chakraborty et al. 2022) even when coding intervention content independently to decrease bias. Another challenge is related to the uncertain intensity of individual BCTs and the unclear value of this knowledge (JaKa et al. 2019) as questions remain regarding the appropriate coding and reporting of BCT intensity and dose (Chakraborty et al. 2022).

Strengths and limitations

Although we have identified the active content of MND carer interventions for PWB and characterized those associated with positive outcomes, these inferences should be treated tentatively. The data pooling for a meta-analysis was not possible due to the heterogeneity of the interventions, outcomes and measurements, and the small number of studies. Additionally, many of the studies only achieved low scores on the JBI critical appraisal tools evaluating methodological quality, indicating that methodological development is needed in this nascent field of research. These methodological weaknesses are discussed in-depth in our systematic review of interventions targeting PWB for MND carers (Cafarella et al. 2022).

Our study included quantitative and qualitative methods and was undertaken with an integrative approach to PWB, incorporating hedonic and eudaimonic aspects of PWB, given that both have been linked with positive outcomes (Vazquez et al. 2009). In comprehensively mapping the active components of PWB interventions for MND carers using BCTTv1, we have identified the BCTs that are yet to be tested or potentially under-utilized and have facilitated a broader capture of potential intervention benefits. However, heterogeneity issues and the small number of studies precluded data-pooling, and methodological limitations were commonplace. There are known limitations associated with the use of taxonomies, such as subjectivity, lack of direction regarding the coding of

intensity, and dependency on the detail provided in the reported content (Chakraborty et al. 2022; Fakolade et al. 2020; JaKa et al. 2019).

Future recommendations

It is imperative that future research ensures that descriptions of the active content of study arms are explicit and sufficiently detailed to enable generalizability and replicability. Precision is essential in each study group, facilitating better comprehension of the relationship between results and intervention content, increasing confidence in effect sizes, and reducing the risk of bias in the outcomes and interpretation (de Bruin et al. 2016; Peters et al. 2015). Taxonomies such as BCTTv1 can assist in the design and description of the active content in study arms (Michie et al. 2011a, 2015) and facilitate the systematization of reporting standards in this field, as in others (Johnston 2021).

Although this study identified and characterized BCTs associated with positive PWB outcomes for MND carers such as 3.1 Social support (unspecified), 1.2 Problem-solving, and 11.2 Reduce negative emotions, the nature of these inferred connections requires further investigation due to reporting and methodological quality concerns. Moreover, most BCTs remain untested in this population. Absent BCTs, particularly those demonstrating PWB-related benefits among carers of other populations (e.g., prompts/cues, associative learning, rewards, Stawarz 2017) require closer scrutiny and potential investigation with well-designed intervention studies.

Future research would benefit from employing existing frameworks (e.g., JBI, PRISMA) to guide study design. Taxonomies of approaches to intervention development may inform future intervention design, piloting, evaluation, and implementation. This process emphasizes methodological rigor and supports theory development, which is particularly relevant for nascent fields such as interventions targeting the PWB of MND carers (O’Cathain et al. 2019).

Our recent systematic review concerning PWB interventions for MND carers revealed a need for theory identification and development (Cafarella et al. 2022). Applying a taxonomy of intervention components such as BCTTv1 facilitates comprehension of the relationships between intervention content and reported outcomes (Michie et al. 2015). This allows the potential introduction of theory to interventions since the BCTs are based on theoretical constructs and can add data to elucidate the “mechanisms of action” responsible for change (Carey et al. 2019). BCTs may be evident in interventions, but unfortunately, theory is often absent in the development of interventions (Prestwich et al. 2014).

Conclusions

This review provides a snapshot of the active content of the interventions (and comparator groups) targeting PWB for MND carers. Identified and absent BCTs and BCTTv1 categories were mapped, and those associated with positive outcomes were characterized. These inferences should be treated tentatively due to reporting and methodological quality issues. However, the results elucidate a more transparent comprehension of the active content of the interventions demonstrating hedonic and eudaimonic PWB benefits for MND carers and provide direction to improve interventions in this developing field of research.

Supplementary material. The supplementary material for this article can be found at <https://doi.org/10.1017/S1478951524000877>.

Acknowledgments. The authors thank Sharni Whitburn (SW) for her coding assistance.

Funding. This research received no specific grant from any funding agency, commercial or not-for-profit sectors. SW was a Research Assistant at the School of Psychology, University of Adelaide.

Competing interests. None.

References

- Aoun S, Cafarella P, Hogden A, et al. (2021) Why and how the work of Motor Neurone Disease Associations matters before and during bereavement: A consumer perspective. *Palliative Care and Social Practice* 15, 2632352421100953. doi:10.1177/26323524211009537
- Aoun SM, Chochinov HM and Kristjanson LJ (2015) Dignity therapy for people with motor neuron disease and their family caregivers: A feasibility study. *Journal of Palliative Medicine* 18(1), 31–37. doi:10.1089/jpm.2014.0213
- Aoun SM, Deas K, Kristjanson LJ, et al. (2017) Identifying and addressing the support needs of family caregivers of people with motor neurone disease using the Carer Support Needs Assessment Tool. *Palliative and Supportive Care* 15(1), 32–43. doi:10.1017/S1478951516000341
- Aoun S, Hogden A and Kho L (2018) “Until there is a cure, there is care”: A person-centered approach to supporting the wellbeing of people with Motor Neurone Disease and their family carers. *European Journal for Person Centered Healthcare* 6(2), 320–328. doi:10.5750/ejpc.v6i2.1488
- Aoun SM, Kissane DW, Cafarella PA, et al. (2020) Grief, depression, and anxiety in bereaved caregivers of people with motor neurone disease: A population-based national study. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration* 21(7–8), 593–605. doi:10.1080/21678421.2020.1790610
- Aromataris E and Munn Z (eds) (2020) *JBI Manual for Evidence Synthesis*. The Joanna Briggs Institute. <https://synthesismanual.jbi.global> (accessed 1 July 2023).
- Bct-taxonomy.com (2023) BCT taxonomy training. <http://www.bct-taxonomy.com/> (accessed 1 July 2023).
- Beach B, Bélanger-Hardy L, Harding S, et al. (2022) Caring for the caregiver: Why policy must shift from addressing needs to enabling caregivers to flourish. *Frontiers in Public Health* 10, 997981. doi:10.3389/fpubh.2022.997981
- Bennett M (2018) Blameless guilt: The case of carer guilt and chronic and terminal illness. *International Journal of Philosophical Studies* 26(1), 72–89. doi:10.1080/09672559.2017.1416422
- Bentley B, O’Connor M, Kane R, et al. (2014) Feasibility, acceptability, and potential effectiveness of dignity therapy for people with motor neurone disease. *PLoS One* 9(5), e96888. doi:10.1371/journal.pone.0096888
- Cafarella P, Effing T and Chur-Hansen A (2022) Interventions targeting psychological well-being for motor neuron disease carers: A systematic review. *Palliative and Supportive Care* 21(2), 321–336. doi:10.1017/s1478951522000311
- Carey RN, Connell LE, Johnston M, et al. (2019) Behavior change techniques and their mechanisms of action: A synthesis of links described in published intervention literature. *Annals of Behavioral Medicine* 53(8), 693–707. doi:10.1093/abm/kay078
- Chakraborty D, Bailey BA, Seidler AL, et al. (2022) Exploring the application of behaviour change technique taxonomies in childhood obesity prevention interventions: A systematic scoping review. *Preventive Medicine Reports* 29, 101928. doi:10.1016/j.pmedr.2022.101928
- Cipolletta S, Gammino GR, Francescon P, et al. (2018) Mutual support groups for family caregivers of people with amyotrophic lateral sclerosis in Italy: A pilot study. *Health and Social Care in the Community* 26(4), 556–563. doi:10.1111/hsc.12558
- Craig P, Dieppe P, Macintyre S, et al. (2013) Developing and evaluating complex interventions: The new Medical Research Council guidance.

- International Journal of Nursing Studies* 50(5), 587–592. doi:10.1016/j.ijnurstu.2012.09.010
- Creemers H, Veldink JH, Grupstra H, *et al.* (2014) Cluster RCT of case management on patients' quality of life and caregiver strain in ALS. *Neurology* 82(1), 23–31. doi:10.1212/01.wnl.0000438227.48470.62
- de Bruin M, Viechtbauer W, Eisma MC, *et al.* (2016) Identifying effective behavioural components of Intervention and Comparison group support provided in SMOKing cEssation (IC-SMOKE) interventions: A systematic review protocol. *Systematic Reviews* 5, 77. doi:10.1186/s13643-016-0253-1
- de Wit J, Beelen A, Drossaert C, *et al.* (2020) Blended psychosocial support for partners of patients with ALS and PMA: Results of a randomized controlled trial. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration* 21(5–6), 344–354. doi:10.1080/21678421.2020.1757114
- de Wit J, Vervoort S, van Eerden E, *et al.* (2019) User perspectives on a psychosocial blended support program for partners of patients with amyotrophic lateral sclerosis and progressive muscular atrophy: A qualitative study. *BMC Psychology* 7, 35. doi:10.1186/s40359-019-0308-x
- Fakolade A, Walters AJ, Cameron J, *et al.* (2020) Healthy together: A systematic review of theory and techniques used in health interventions for persons with chronic neurological conditions and their caregivers. *Patient Education and Counseling* 103(4), 788–803. doi:10.1016/j.pec.2019.10.022
- Gluyas C, Mathers S, Hennessy Anderson N, *et al.* (2017) Factors to consider for motor neurone disease carer intervention research: A narrative literature review. *Palliative and Supportive Care* 15(5), 600–608. doi:10.1017/S1478951516000912
- Hardeman W, Michie S, Fanshawe T, *et al.* (2008) Fidelity of delivery of a physical activity intervention: Predictors and consequences. *Psychology and Health* 23(1), 11–24. doi:10.1080/08870440701615948
- Harris M, Thomas G, Thomas M, *et al.* (2017) Supporting wellbeing in motor neurone disease for patients, carers, social networks, and health professionals: A scoping review and synthesis. *Palliative and Supportive Care* 16(2), 228–237. doi:10.1017/S1478951517000700
- Hoffmann TC, Erueti C and Glasziou PP (2013) Poor description of non-pharmacological interventions: Analysis of consecutive sample of randomised trials. *BMJ (Clinical Research Ed.)* 347, f3755. doi:10.1136/bmj.f3755
- JaKa MM, French SA, Wolfson J, *et al.* (2019) Understanding outcomes in behavior change interventions to prevent pediatric obesity: The role of dose and behavior change techniques. *Health Education and Behavior* 46(2), 312–321. doi:10.1177/1090198118798679
- Johnston M (2021) Progress in conducting and reporting behaviour change intervention studies: A prospective retrospection. *Health Psychology and Behavioral Medicine* 9(1), 567–581. doi:10.1080/21642850.2021.1939701
- Kavanaugh MS, Cho Y, Fee D, *et al.* (2020) Skill, confidence and support: Conceptual elements of a child/youth caregiver training program in amyotrophic lateral sclerosis – The YCare protocol. *Neurodegenerative Disease Management* 10(4), 231–241. doi:10.2217/nmt-2020-0004
- Keating N, McGregor JA and Yeandle S (2021) Sustainable care: Theorising the wellbeing of caregivers to older persons. *International Journal of Care and Caring* 5(4), 611–630. doi:10.1332/239788221X16208334299524
- Keogh A, Matthews J and Hurley DA (2018) An assessment of physiotherapist's delivery of behaviour change techniques within the SOLAS feasibility trial. *British Journal of Health Psychology* 23(4), 908–932. doi:10.1111/bjhp.12323
- Leventhal H, Weinman J, Leventhal E, *et al.* (2008) Health psychology: The search for pathways between behavior and health. *Annual Review of Psychology* 59(1), 477–505. doi:10.1146/annurev.psych.59.103006.093643
- Lorencatto F, West R, Christopherson C, *et al.* (2013) Assessing fidelity of delivery of smoking cessation behavioural support in practice. *Implementation Science* 8, 40. doi:10.1186/1748-5908-8-40
- Marconi A, Gragnano G, Lunetta C, *et al.* (2016) The experience of meditation for people with amyotrophic lateral sclerosis and their caregivers – A qualitative analysis. *Psychology, Health and Medicine* 21(6), 762–768. doi:10.1080/13548506.2015.1115110
- McCleary N, Duncan EM, Stewart F, *et al.* (2013) Active ingredients are reported more often for pharmacologic than non-pharmacologic interventions: An illustrative review of reporting practices in titles and abstracts. *Trials* 14, 146. doi:10.1186/1745-6215-14-146
- Michie S, Abraham C, Eccles MP, *et al.* (2011b) Strengthening evaluation and implementation by specifying components of behaviour change interventions: A study protocol. *Implementation Science* 6, 10. doi:10.1186/1748-5908-6-10
- Michie S, Richardson M, Johnston M, *et al.* (2013) The Behavior Change Technique Taxonomy (v1) of 93 hierarchically clustered techniques: Building an international consensus for the reporting of behavior change interventions. *Annals of Behavioral Medicine* 46(1), 81–95. doi:10.1007/s12160-013-9486-6
- Michie S, van Stralen MM and West R (2011a) The behaviour change wheel: A new method for characterising and designing behaviour change interventions. *Implementation Science* 6, 42. doi:10.1186/1748-5908-6-42
- Michie S, Wood C, Johnston M, *et al.* (2015) Behaviour change techniques: The development and evaluation of a taxonomic method for reporting and describing behaviour change interventions (a suite of five studies involving consensus methods, randomised controlled trials and analysis of qualitative data). *Health Technology Assessment* 19(99), 1–188. doi:10.3310/hta19990
- Moher D, Liberati A, Tetzlaff J, *et al.* and The PRISMA Group (2009) Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *PLoS Medicine* 6(7), e1000097. doi:10.1371/journal.pmed.1000097
- Moore KJ, Davis S, Gola A, *et al.* (2017) Experiences of end of life amongst family carers of people with advanced dementia: Longitudinal cohort study with mixed methods. *BMC Geriatrics* 17(1), 135. doi:10.1186/s12877-017-0523-3
- Ninot G (2021) *Non-Pharmacological Interventions*. Cham: Springer.
- O'Cathain A, Croot L, Sworn K, *et al.* (2019) Taxonomy of approaches to developing interventions to improve health: A systematic methods overview. *Pilot and Feasibility Studies* 5, 41. doi:10.1186/s40814-019-0425-6
- O'Dwyer ST, Moyle W, Zimmer-Gembeck M, *et al.* (2016) Suicidal ideation in family carers of people with dementia. *Aging and Mental Health* 20(2), 222–230. doi:10.1080/13607863.2015.1063109
- Olesen LK, la Cour K, With H, *et al.* (2022) A cross-sectional evaluation of acceptability of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis and cognitive and behavioral impairments. *BMC Health Services Research* 22(1), 697. doi:10.1186/s12913-022-07986-4
- Pagnini F, Phillips D, Haulman A, *et al.* (2021) An online non-meditative mindfulness intervention for people with ALS and their caregivers: A randomized controlled trial. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration* 23(1–2), 116–127. doi:10.1080/21678421.2021.1928707
- Pasquini S, Margaritini A, Gambella E, *et al.* (2022) A psychosocial intervention for supporting informal caregivers of older people with Alzheimer disease: Protocol for the InnFamiglia randomized controlled trial. *JMIR Research Protocols* 11(11), e37496. doi:10.2196/37496
- Pearlin LI, Mullan JT, Semple SJ, *et al.* (1990) Caregiving and the stress process: An overview of concepts and their measures. *The Gerontologist* 30(5), 583–594. doi:10.1093/geront/30.5.583
- Peters GJY, de Bruin M and Crutzen R (2015) Everything should be as simple as possible, but no simpler: Towards a protocol for accumulating evidence regarding the active content of health behaviour change interventions. *Health Psychology Review* 9(1), 1–14. doi:10.1080/17437199.2013.848409
- Poppe C, Koné I, Iseli L, *et al.* (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. doi:10.1080/21678421.2020.1771735
- Prestwich A, Sniehotta FF, Whittington C, *et al.* (2014) Does theory influence the effectiveness of health behavior interventions? Meta-analysis. *Health Psychology* 33(5), 465–474. doi:10.1037/a0032853
- Ryan RM and Deci EL (2001) On happiness and human potentials: A review of research on hedonic and eudaimonic well-being. *Annual Review of Psychology* 52, 141–166. doi:10.1146/annurev.psych.52.1.141
- Stawarz K (2017) Towards better medication adherence apps: Preventing forgetfulness by facilitating the formation of routine-based remembering strategies. PhD dissertation, Division of Psychology and Language Sciences,

- University of London, University College, London. <https://www.proquest.com/docview/2411496850>
- Steinhauser KE, Olsen A, Johnson KS, et al.** (2016) The feasibility and acceptability of a chaplain-led intervention for caregivers of seriously ill patients: A Caregiver Outlook pilot study. *Palliative and Supportive Care* **14**(5), 456–467. doi:10.1017/S1478951515001248
- Stern C, Lizarondo L, Carrier J, et al.** (2020) Methodological guidance for the conduct of mixed methods systematic reviews. *JBISIR-D-19-00169*
- Teahan Á, Lafferty A, McAuliffe E, et al.** (2020) Psychosocial interventions for family carers of people with dementia: A systematic review and meta-analysis. *Journal of Aging and Health* **32**(9), 1198–1213. doi:10.1177/0898264319899793
- Thomas PT, Warriar MG, Arun S, et al.** (2022) An individualised psychosocial intervention program for persons with MND/ALS and their families in low resource settings. *Chronic Illness*, 17423953221097076. Advance online publication. doi:10.1177/17423953221097076
- Ugalde A, Mathers S, Hennessy Anderson N, et al.** (2018) A self-care, problem-solving and mindfulness intervention for informal caregivers of people with motor neurone disease: A pilot study. *Palliative Medicine* **32**(4), 726–732. doi:10.1177/0269216317743434
- van Groenestijn A, Schröder C, Visser-Meily J, et al.** (2015) Cognitive behavioural therapy and quality of life in psychologically distressed patients with amyotrophic lateral sclerosis and their caregivers: Results of a prematurely stopped randomized controlled trial. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration* **16**(5-6), 309–315. doi:10.3109/21678421.2015.1038276
- Vazquez C, Hervas G, Rahona JJ, et al.** (2009) Psychological well-being and health: Contributions of positive psychology. *Annuary of Clinical and Health Psychology* **5**, 15–27.
- Wood CE, Richardson M, Johnston M, et al.** (2015) Applying the behaviour change technique (BCT) taxonomy v1: A study of coder training. *Translational Behavioral Medicine* **5**(2), 134–148. doi:10.1007/s13142-014-0290-z