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REVIEW

Experiences of people with multiple sclerosis and clinicians in using cognitive behavioural therapies for hidden symptoms: a systematic review and meta-aggregation

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Abstract

Purpose Cognitive behavioural therapies (CBTs) are a standard of care for treatment of many 'hidden symptoms' in people with MS (PwMS), such as stress, depression, and fatigue. However, these interventions can vary widely in formatting and may not be tailored for PwMS. To optimize CBTs for MS, understanding the experiences of PwMS and clinicians is essential. This systematic review and meta-aggregation synthesizes existing qualitative data on stakeholder perspectives of CBTs for PwMS.

Methods Systematic searches across five major electronic databases were conducted. Studies reporting qualitative data were identified. Two reviewers performed screening, quality assessment, data extraction, and certainty of evidence assessments. Meta-aggregation was performed as per the Joanna Briggs Institute approach, entailing qualitative data extraction, developing categories, and synthesizing overall findings.

Results Twenty-eight studies were included in this review, comprising data from 653 PwMS and 47 clinicians. In the meta-aggregation, 122 qualitative results were extracted and grouped into nine categories. Categories were then combined into six synthesized findings: (1) setting the context–life with MS, (2) reasons for participating in CBTs, (3) acceptability of and experiences with participating in CBTs, (4) perceived benefits of CBTs, (5) perceived challenges with CBTs, and (6) suggestions to improve CBTs for PwMS.

Conclusions A range of benefits including psychological, social, and lifestyle improvements were reported, but varied based on the design of the CBT intervention. Future CBT interventions should be tailored to participant needs, delivered in group settings, offer online options, and be delivered by a trained facilitator familiar with MS. Further exploration of the ideal CBT design for PwMS, as well as engagement with caregivers and clinicians treating MS, is warranted.

Keywords Multiple sclerosis · Cognitive behaviour therapies · Patient preferences · Affective symptoms

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Introduction

Multiple sclerosis (MS) is a chronic neurodegenerative condition characterized by inflammation and demyelination of the central nervous system [1]. Global estimates suggest that over 2.8 million people are currently living with MS and this continues to increase each year [1]. Although MS has been found in a variety of populations, females and those of European descent tend to be at highest risk [1]. Although advances in the treatment modalities have expanded the life expectancy of people with MS (PwMS), living longer does not always translate into living well [2]. For instance, symptoms of anxiety, depression, and fatigue are highly prevalent among PwMS [3–5]; with prevalence rates of 30.5% for depression and 22.1% for anxiety reported in a recent meta-analysis [6]. "Hidden symptoms" such as these can have profound effects on physical, emotional, cognitive, and social functioning [1, 7, 8]. Nevertheless, the effective interventions for hidden symptoms and their impacts on daily life are lacking [7].

Cognitive behavioural therapy (CBT) refers to a family of psychological interventions and is widely considered to be the "gold standard" of psychotherapy [9, 10]. In the context of MS, CBTs can also be used in the treatment of 'physical' hidden symptoms such as fatigue, pain, and sleep disorders [9, 10]. By focusing on the links between thoughts, emotions, and behaviours, CBT-based therapies enable participants to identify and alter maladaptive thought patterns [10]. This is achieved through activities such as thought tracking, setting goals, and engaging in behavioural experiments [10]. Notably, in the recent decades, 'third-wave' CBTs have gained momentum [11]. In this paradigm, the core principles of CBT are applied with an emphasis on acceptance, mindfulness, and compassion [11, 12]. This approach is thought to be of greater relevance to people living with chronic conditions, but the evidence to support this assumption is still evolving [12]. Moreover, these interventions have not often been designed for PwMS. Knowing that treatment adherence for psychological therapies can be low in PwMS (especially when hidden symptoms are present) [13, 14], it is important to understand how PwMS engage with, experience, and use CBTs.

Meta-aggregation is a systematic review method used to synthesize the results from qualitative research studies [15]. This approach is founded in pragmatism, with the objective of producing practical recommendations ("lines of action") to inform policy, implementation, and practice [16]. Through the aggregation and descriptive summary of qualitative results, meta-aggregations can reveal and identify insights not otherwise discernible from quantitative data [15, 16]. To date, no prior qualitative systematic review on the experiences of PwMS participating in CBTs could be identified in the literature. Nevertheless, CBTs continue to be recommended in clinical care guidelines [17]. Therefore, it is necessary to understand how CBTs are experienced by PwMS, and to identify ways in which these interventions could be tailored to make them more accessible, acceptable, and potentially effective. The aim of this review was to systematically review and synthesize the existing qualitative research evidence on the experiences of PwMS, clinicians, and other relevant knowledge users with CBT interventions in the treatment of hidden symptoms associated with MS.

Methods

Protocol and registration

The protocol for this review was prospectively registered in 2022 with PROSPERO, Centre for Reviews and Dissemination, University of York: CRD42022337034.

Information sources

A comprehensive search strategy combining MeSH terms with keywords relating to CBT and MS was developed for five major databases (Medline, Embase, PsycINFO, AMED, and CINAHL). Additional searches involving grey literature, reference lists of reviews and published trials, and the Science Citation Index were also performed. The search was performed on June 16, 2022, for articles published prior to 2022, in English, with human research participants. Duplicates were removed as per the Bramer method [18]. The full search strategy is detailed in Supplementary Appendix 1.

Selection process

All search results were imported into Covidence. Initial title and abstract screenings were performed, with subsequent full-text screening to determine eligibility. Discrepancies were resolved through group discussion until a consensus was reached.

Qualitative studies including mixed- or multi-methods studies were included in this review. Any studies reporting on the experiences of PwMS, caregivers, clinicians, or other relevant knowledge users with CBT interventions were included. Any form of CBT (e.g., bibliotherapy, in-person, online, asynchronous, etc.) or intervention incorporating CBT was considered eligible. Interventions that did not explicitly contain a cognitive – behavioural aspect (e.g., mindfulness-based stress reduction rather than mindfulness-based cognitive therapy) were excluded.

Data collection

A standardized data extraction template was used by two reviewers (GF and SP). Variables pertaining to study design, participant demographics, intervention characteristics, and key results were collected for each included study. The extracted data were then used to produce evidence tables.

Quality appraisal

A generic quality appraisal tool, the Critical Appraisal Skills Programme (CASP) for Qualitative Studies, was used given the broad inclusion criteria for this review. The CASP tool is a 10-item questionnaire used to assess the risk of bias and overall quality of a given study. Items can be rated as "Yes", "No", or "Can't tell", and the tool concludes with a rating on the overall value of the study. Two reviewers appraised each included study using the CASP tool and discrepancies were resolved through group discussion until a consensus was reached. Studies with fewer than six "Yes" ratings were considered to have insufficient methodological quality and were excluded from this review.

Evidence synthesis and reporting

The meta-aggregation approach outlined in the Joanna Briggs Institute (JBI) manual was used to inform the synthesis of results [19]. After data extraction, a list of extracted results was produced with the themes reported, or main qualitative results, from each study. These results were then grouped into preliminary categories with similar and overlapping findings. Finally, synthesized findings encompassing overlapping categories were formed. This process was conducted over multiple consensus-building meetings among the study team. The 21-item Enhancing Transparency in Reporting the synthesis of Qualitative research (ENTREQ) framework and Preferred Reporting Items for Systematic Reviews (PRISMA) guidelines were used to guide reporting in this review [20, 21].

Certainty of evidence

As per the JBI manual, the credibility of individual results presented in each study was rated as either "unequivocal", "credible", or "not supported" by two reviewers. Discrepancies were resolved through consensus-building study team meetings. After compiling the results, an assessment of the studies included in each synthesized finding was performed. This entailed checking the number of unequivocal, credible, or not supported results underlying each synthesized finding.

Results

After removing duplicates, the literature search process returned a total of 1456 citations which were screened for inclusion in this review. Through the title and abstract screening, 1285 articles were found to be ineligible, leaving 171 articles for full-text screening. In circumstances where full-text articles could not be retrieved through institutional access privileges, attempts were made to contact corresponding authors. The full-text screening process revealed 143 of the articles to be ineligible; 88 used a non-qualitative study design, 32 did not study a CBT intervention, 10 were not published in English, eight did not have a full-text article available, and five did not meet quality threshold criteria. In total, 28 studies were found to be eligible for inclusion in this review. The search and screening process is outlined in Fig. 1.

Study characteristics

Full details on study characteristics and results are outlined in Table 1. Out of the 28 studies included in this review, 21 used mixed- or multi-methods and seven adopted a purely qualitative design. Of the 21 mixed- or multi-methods studies, 12 involved randomized controlled trials (RCTs) [22–32], and 10 involved non-randomized studies (e.g., quasi-experiments) [33–42]. In total, 12 studies were conducted in the UK [22, 24, 27, 29–32, 34, 37, 41–43], 10 studies in Europe [25, 26, 33, 35, 36, 39, 44–47], two in Oceania [23, 38], three in North America [28, 40, 48], and one in the UK and Oceania [49]. The studies sampled both PwMS (26 studies) [22–35, 37–46, 48, 49] and relevant knowledge users (three studies) [32, 36, 47]. Sample sizes in the included studies ranged from $n = 3$ to 175, with a combined total of 653 people living with MS and 47 other knowledge users (psychologists, psychotherapists, and physiotherapists) included in this review.

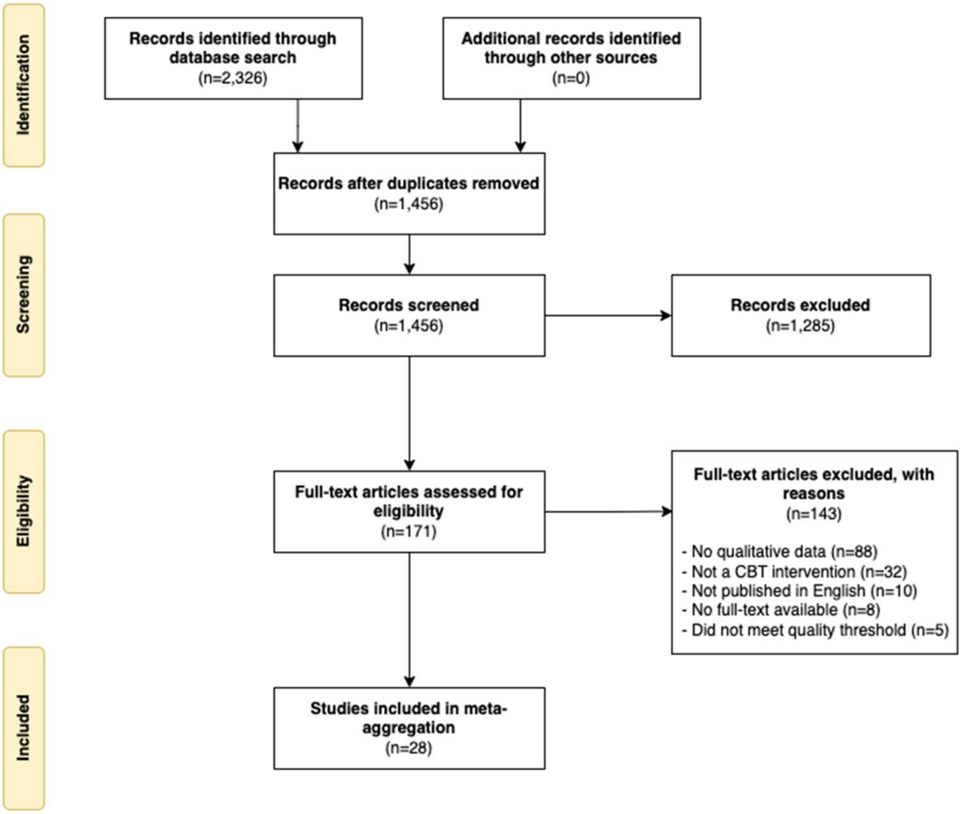
Quality appraisal

In total, six studies were rated "Yes" across all nine criteria included in the CASP qualitative checklist [24, 34, 40, 41, 45, 48]. Eight studies were assigned "Yes" on eight criteria [22, 23, 25, 28, 29, 42, 47, 49], and another eight studies were assigned "Yes" on seven criteria [30, 32, 35, 37, 39, 43, 44, 46]. Finally, six studies were assigned "Yes" on six criteria [26, 27, 31, 33, 36, 38]. Full details of the quality appraisal are reported in Table 2.

Methods used in included studies

A variety of data sources and analytic methods were used. In total, 19 studies utilized thematic or framework analyses (data-driven, inductive, and deductive) [22, 24, 26, 28–32,

Fig. 1 PRISMA flow diagram detailing the systematic search and screening process



34, 37–43, 45, 46, 49]. Eight studies used content analysis (inductive and deductive) [23, 25, 27, 33, 35, 36, 44, 47], while one study reported the use of interpretive description [48]. These analyses used a range of data sources including interviews, focus groups, participant journals, and questionnaires.

Participant characteristics

All studies reported participant characteristics, but with varied levels of detail. All studies reported age, with participants ranging from 20 to 71 years of age [22–49]. Across all studies [22–49], over half of participants were female, with three studies containing an entirely female sample [28, 47, 48]. When ethnicity was reported, participants were mainly or entirely White [22, 24, 28, 29, 32, 34, 37, 38, 40, 48, 49]. Most studies did not report socioeconomic status or comorbidities. Out of the 11 studies that reported level of education, most participants had at least high-school or post-secondary education [25, 26, 30, 33, 35, 38, 40, 41, 44, 46]. Most studies that included PwMS included multiple disease phenotypes including primary-progressive, secondary-progressive, relapsing–remitting, or unknown/other [22, 24–35, 37–44, 48]. Across these studies, relapsing–remitting MS was often the most common phenotype. A range of Expanded Disability Status Scale (EDSS) [50] scores were also reported, ranging from 0 (“normal neurological function”) to 8 (“restricted to bed or wheelchair”) [22, 25, 26, 28, 29, 31–33, 35, 37, 43–46].

CBT intervention characteristics

Interventions all drew upon the principles of CBT in some capacity (i.e., incorporating psychoeducation, cognitive – behavioural strategies, etc.) [22–49]. Among the studies exploring the use of an intervention for PwMS, a range of traditional psychotherapy programs (e.g., CBT, acceptance and commitment therapy [ACT], mindfulness-based cognitive therapy [MBCT]) and composite interventions with cognitive therapy components (e.g., CBT plus exercise, education) were used [22–49]. These interventions were delivered both in-person [23–27, 32–36, 38–41, 44, 46, 47] and remotely [28, 29, 31, 34, 37, 45], in group settings [22, 23, 25–27, 33, 35, 38–41, 45, 46] and one-on-one [24, 26, 28, 29, 32, 44]. In addition to these programs, self-administered interventions involving the use of a mobile app, directed readings, or video modules were also used [30, 31, 37, 42, 43, 49].

Qualitative synthesis

Qualitative results and credibility, as well as categories and synthesized findings, are displayed in Fig. 2. Overall, 122 qualitative results were extracted and grouped into

Table 1 Summary of key characteristics of included studies

Authors	Methods	Participants	Intervention	Main Results
Babbage et al. (2019) [49]	• Multi-methods • Convenience sampling • Qualitative interviews 5–6 weeks after installing the app • Inductive and deductive thematic analysis	• *N* = 11 people with MS • Age = 41–59 years, 54.5% female, 54.5% NZ European, 27.3% White British	MS Energize app, self-administered, 5–6 weeks	1. Validation ■ Relating to real people ■ Someone doing something 2. Personal cost ■ Using the app could be fatiguing ■ Focusing on fatigue could prompt negative thoughts ■ Challenging existing strategies 3. Reframing my experience and adding to my knowledge 4. A good idea – for someone ■ Hitting the mark ■ A good idea if you're new
Bogosian et al. (2016) [22]	• Mixed-methods • Criterion sampling • Semi-structured interviews after completion of intervention • Deductive thematic analysis	• *N* = 15 people with MS • Mean age = 53.4 years, 52.6% female, 89.5% White, 68.4% college or higher education, 26.3% PPMS, Mean EDSS = 6.8	Adaptation of mindfulness-based cognitive therapy, virtual, group setting, 8 weeks	1. Acceptance and experiential avoidance 2. Decentring 3. Self-compassion 4. Self-efficacy 5. Group processes 6. Beliefs prior to the course
Borghi et al. (2018) [33]	• Mixed-methods • Analysis of transcripts from each session • Deductive content analysis	• *N* = 41 people with MS • Mean age = 42.3 years, 55% female, 22% college or higher education, 95% RRMS, EDSS range = 1–5.5	Cognitive – behavioural intervention, in person, group setting, 5 sessions over 6 months	1. Resistance and Openness to Change ■ Aggression toward the group ■ Aggression toward the external world ■ Aggression toward the setting ■ Avoidance ■ Denial ■ Displacement ■ Freezing
Brown et al. (2016) [48]	• Qualitative study • Convenience, snowball, and random sampling • Focus groups • Interpretive description	• *N* = 10 people with MS • Age range = 23–62 years, 100% female, 100% White, "Majority" with RRMS	N/A (focus groups with potential participants of CBT)	1. Coming to know yourself with cognitive changes 2. Learning to cope with cognitive changes 3. Living a changed life

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Dennison et al. (2013) [34]	• Qualitative study nested within experimental trial • Opportunistic and maximum variation sampling • Qualitative interviews ~ 9 weeks after completion of intervention • Inductive thematic analysis	• $N = 30$ people with MS • Mean age = 43.5 years, 76.6% female, 80% White, 86.7% RRMS, Mean EDSS = 5.0	CBT and supportive learning, in person and via telephone, 8 sessions	1. Tuning in and sharing thoughts and feelings 2. Learning and enacting explicit strategies for living with MS 3. Buying into therapy ■ Being motivated to engage ■ Being the right person at the right time ■ Experiencing a high-quality interaction ■ Having a tailored approach ■ Overcoming practical barriers to participation 4. Achieving personally valued changes 5. Experiencing ongoing benefits
Dunne et al. (2021) [23]	• RCT with qualitative strand • Voluntary response sample • Practice diaries and reflective journals during intervention • Content analysis	• $N = 55$ people with MS • Mean age = 48.4 years, 83.3% female, 38.2% with anxiety and/or depression	Mindfulness for MS program vs chair yoga, in person, group setting, 8 weeks	1. Acceptability ■ Online program engagement ■ Valued social interactions ■ Supportive learning 2. Perceived benefits of mindfulness practice ■ Reducing stress ■ Managing pain ■ Improving cognition functioning ■ Positive sleep effects 3. Perceived barriers to mindfulness practice ■ Finding time ■ Feeling fatigued ■ Feeling pain
Fortune et al. (2020) [24]	• RCT with qualitative strand • Purposive sampling • Semi-structured interviews after completion of intervention • Framework analysis	• $N = 15$ people with MS • Age range = 39–71, 66% female, 100% White, 46.7% RRMS,	Physical activity behaviour change intervention, in person, individual, 12 weeks	1. "I can do this": developing competence in physical activity 2. "I felt valued": the nurturing culture 3. "What can I do": empowered enactment

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Giovannetti et al. (2022) [36]	• Multi-methods • Convenience sampling • Open-ended questionnaires after phase 2 and 3 • Content analysis	• $N = 44$ psychologists • Mean age = 42.2 years, 93.2% female	Training program for psychologists to deliver ACT to people with MS, in person, group setting, 3 phases	1. The Training ■ Satisfaction with the training ■ Training impacts on participants ■ Training active elements 2. Trainees' view of the READY program ■ READY strengths, weaknesses, and suggested changes ■ READY impacts on PwMS ■ READY active elements
Giovannetti et al. (2022) [35]	• Multi-methods • Voluntary response sampling • Open-ended questionnaire after completion of intervention • Inductive content analysis	• $N = 175$ people with MS • Mean age = 48.0 years, 76.6% women, 34.4% college or higher education, 78.8% RRMS, Median EDSS = 3	ACT-informed resilience training, in person, group setting, 7 weekly sessions + booster	1. Perceptions of program composition ■ Satisfaction with the program ■ Satisfaction with its formats ■ Satisfaction with materials 2. READY impacts on life domains 3. READY active elements 4. Suggested improvements
Giovannetti et al. (2020) [25]	• RCT with nested qualitative strand • Voluntary response sample • Interviews within 3 months of completing intervention • Inductive content analysis	• $N = 30$ people with MS • Age range = 20–59 years; 60% female, 59.3% college or higher education, 80% RRMS, EDSS range = 0–6.5	ACT-informed resilience training, in person, group setting, 7 weekly sessions + booster	1. Attitudes towards participation 2. Perceptions of program composition 3. Program impacts on life domains 4. Program active elements 5. Program improvement trajectories 6. Program differences and similarities 7. Suggested READY improvements
Gottberg et al. (2016) [44]	• Qualitative and descriptive study • Convenience sample • Semi-structured interviews after completion of intervention • Content analysis	• $N = 12$ people with MS • Mean age = 38 years, 75% female, 50% university education, 84% RRMS, 84% with EDSS score between 0 and 3.5	CBT, in person, individual, Weekly sessions for ~ 25 weeks	1. CBT as a demanding process ■ Meaningful sessions with a guiding therapist ■ Laborious process with a purpose that emerged slowly ■ CBT required planning and space in everyday life 2. Confronting everyday life after CBT with self-knowledge and well-being ■ Increased self-knowledge and acceptance of one's limitations ■ Increased well-being when learning to cope with anxiety and depression
Harrison et al. (2017) [37]	• Mixed-methods • Purposive sampling • Semi-structured interviews 12 weeks from baseline • Inductive thematic analysis	• $N = 7$ people with MS • Median age = 45 years, 57% female, 85.7% White, 42.8% RRMS, EDSS range = 4.5–8	Guided cognitive-behavioural self-management, telephone/self-administered, 8 self-administered sessions + 3 h of telephone sessions	1. Goals tapped motivation 2. Human contact kept up motivation 3. Create an online version please!

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Hersche et al. (2019) [26]	• RCT with qualitative strand • Purposive sampling • Semi-structured interview 10 weeks from baseline • Thematic analysis	• $N=6$ people with MS • Age range = 39–57 years, 50% female, 16.6% more than 16 years of schooling, Median EDSS = 5	Energy conservation intervention with CBT techniques, in person, individual and group setting, 3 weeks	1. All the interviewed participants judged the IEME approach as very interesting and enriching 2. They had positive memories of exchanges between group members, the discussions on the different topics and the non-judgmental, supportive atmosphere 3. Although most participants stated that the education provided little new knowledge, they appreciated the time spent on in-depth reflection and on the practical application of fatigue management strategies 4. All participants achieved some behavioural change; however, this takes time and is not finished yet 5. Workload reduction and ergonomic behaviour were easier to implement, whereas the redesign of daily structure, roles and responsibilities appeared to be more challenging because it was easy to fall back into old patterns 6. The participants thought that the behavioural changes were their own responsibility, but that a local group or OT could help to improve their self-confidence during the implementation of the changes
Hind et al. (2010) [43]	• Qualitative study • Voluntary response sample • Written feedback and semi-structured interviews after first session and after completion or withdrawal from study • Framework analysis	• $N=17$ people with MS • Median age = 46 years, 76.5% female, 53% diagnosed with MDD, 88.2% RRMS, Median EDSS = 4.5	Allocation to one of two computerized CBT packages, self-administered, 5–8 weekly sessions in total	1. The burden of CCBT for people with physical and cognitive morbidity 2. Lack of human input ■ Social isolation ■ Problem identification and definition ■ Goal setting ■ Failure to distinguish between events, thoughts and beliefs 3. Acknowledgement of physical illness ■ Grieving ■ Depression symptom inventories ■ Inappropriate material

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Holmes et al. (2012) [27]	• RCT with qualitative strand • Random sampling from those enrolled in RCT • Structured phone interview after completion of intervention • Content analysis	• $N = 12$ people with MS*	CBT, in person, group setting, 6 sessions over 12 weeks	1. Out of 132 comments, 109 were positive; 14 were neutral and 9 were negative 2. All participants were satisfied with the information or support prior to starting the group 3. The helpful aspects included talking in a group with other people who have MS, hearing others' opinions and having support for the mental aspects of the disease 4. Confidence 5. Social element 6. Content (depression, mixed opinions on discussing physical aspects of MS) 7. Structure (discussion time vs psychoeducation, duration) 8. Experiencing benefits vs not 9. Formatting 10. Venue 11. Would recommend the groups to others
Learmonth et al. (2019) [28]	• RCT with nested qualitative study • Semi-structured interviews after completion of intervention • Inductive category and thematic analysis	• $N = 18$ people with MS • Mean age = 50 years, 100% female, 77.8% White, 100% RRMS, Median EDSS = 1	Exercise training intervention with behavioural strategies informed by SCT, virtual, individual, 8 sessions over 4 months	1. Enrolment and assessments 2. Suggestions for improvements in the design and delivery of the exercise programme components

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Meek et al. (2021) [29]	• RCT with qualitative strand • Voluntary response sample • Semi-structured interviews after completion of intervention • Framework analysis	• $N = 6$ people with MS • Age range = 47–64 years, 66% women	Brief ACT, first session in person and subsequent sessions via telephone, individual, weekly for 6 weeks	1. Social Barriers ■ Guilt about seeking partners support ■ Lack of societal understanding restricts openness ■ Protecting family ■ Support available 2. Healthcare Support ■ Feeling guilty and being dismissed ■ Availability ■ Professionals' time and being dismissed ■ Frustration and feeling selfish 3. Talking Therapy ■ Felt comfortable ■ Impartiality and avoiding medication ■ Impartiality and reliability ■ Not needed 4. Motivation to Help ■ Benefiting others with MS ■ Freeing up the GP ■ Purpose and being valued ■ Benefiting others selflessly 6. Telephone Sessions 7. The Workbook
Moss-Morris et al. (2012) [30]	• Mixed-methods pilot RCT • Voluntary response sample • Semi-structured interviews after completion of intervention • Data-driven thematic analysis	• $N = 15$ people with MS*	Self-management package for fatigue based on CBT programme and feedback from people with MS, virtual, self-administered, 8 weekly sessions (25–50 min per session)	Most participants perceived an improvement in their fatigue, felt they understood their fatigue better, were more in control of it and were trying to implement lifestyle changes suggested by the programme
Pakenham et al. (2018) [38]	• Multi-methods • Voluntary response sample • Open-ended questionnaire after completion of intervention • Thematic analysis	• $N = 25$ people with MS*	ACT-based resilience training, in person, group setting, 7 modules + booster session	1. Mindfulness, defusion, acceptance 2. Mindfulness, defusion, acceptance, values 3. Life enhancement, increased acceptance of MS, effective management of MS-related distressing thoughts and feelings 4. No changes, structural changes, more discussion time, different group composition and management, more information on MS

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Pöttgen et al. (2015) [39]	• Pilot study with qualitative strand • Focus groups after each module in the intervention • Framework analysis	• $N = 27$ people with MS in full study, 5–6 participants selected for each focus group • Mean age = 46.2 years, 85% female, 67% RRMS	Metacognitive training program, in person, group setting, 6 modules (90 min each)	1. Benefit and Problems of Group Sessions 2. Learning Gain 3. Active role/Involvement in the training 4. Presentation
Proctor et al. (2018) [31]	• RCT with nested qualitative strand • Maximum variation sampling • Interviews after completion of intervention • Deductive framework analysis	• $N = 10$ people with MS*	Telephone-supported ACT bibliotherapy, self-administered, individual, 8 weeks	1. Recruitment 2. Randomization 3. Measures 4. Support calls 5. Self-help text
Russell et al. (2022) [45]	• Qualitative study • Convenience sampling • Semi-structured interviews after completion of intervention • Inductive thematic analysis	• $N = 17$ people with MS • Mean age = 42.9 years, 82.4% female,	Social cognitive behaviour change physical activity intervention, in person, group setting, 10 weeks	1. Psychological and social factors 2. Intervention processes 3. MS Identity
Ryan et al. (2020) [32]	• RCT with nested qualitative strand • Voluntary response sample • Focus groups and interviews after completion of intervention • Framework analysis	• $N = 25$ people with MS* • $N = 4$ physiotherapists*	Physical activity intervention with brief cognitive – behavioural strategies, in person, individual, 4 sessions over 3 months	1. Feasibility and acceptability 2. Feasibility of definitive trial
Shevil and Finlayson (2009) [40]	• Multi-methods • Voluntary response sample • Focus groups during 5th session and open-ended questionnaires after completion of intervention • Question analysis and thematic analysis	• $N = 35$ people with MS • Mean age = 52.4 years, 74.3% female, 80% White, 54.3% more than 15 years of education, 51.4% stable MS status	Self-management cognitive program, in person, group setting, 5 sessions (once per week)	1. Course Content ■ Increasing knowledge and awareness about cognitive changes ■ Problem-solving through cognitive challenges ■ Practicing solutions and strategies ■ Emotional and social implications ■ Changing perceptions 2. Format and methods of delivery ■ Repetition ■ Group-based intervention ■ Length ■ Handouts ■ Homework ■ Between-session calls ■ Facilitator ■ Location 3. Recommended Changes ■ Incorporating caregivers ■ Additional content

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Thomas et al. (2021) [42]	• Mixed-methods • Convenience and voluntary response sample • Focus groups during development of intervention • Deductive thematic analysis	• $N = 3$ in focus group 1 and $N = 6$ in focus group 2 • 88.9% female	Cognitive behavioural approach to fatigue management program (digital toolkit), self-administered, individual, 6 weeks (accompanies in-person intervention)	Key toolkit requirements suggested by focus group participants: ■ Should be a “tool to help rather than a time-consuming task” ■ Suitable for those unfamiliar with FACETS ■ Important to include positive aspects ■ Include self-monitoring feedback (e.g., dashboard and g raphs) Preferences for the toolkit expressed by focus group participants: ■ App should be freely available at app stores ■ Reminder (e.g., reminders to take rests) and note functions would be useful

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Thomas et al. (2010) [41]	• Mixed-methods • Voluntary response sample • Focus groups during development and after completion of intervention • Thematic analysis	• $N=8$ in focus group 1, Mean age = 44 years, 75% female, 62.5% RRMS • $N=6$ in focus group 2*	Cognitive behavioural approach to fatigue management program, in person, group setting, 6 weeks	*Focus group 1* 1. Invisibility 2. Emotions 3. Practical issues ■ Activity diary ■ Involvement of supportive other ■ Group format ■ Is programme appropriate for newly diagnosed? 4. Variability and Impact 5. Fatigue management strategies ■ Learning to say no to others demands ■ Pleasant activity scheduling ■ Planning ■ Prioritising tasks and activities ■ Incorporating scheduled relaxation periods ■ Delegation *Focus group 2* 1. Content and structure 2. Venue 3. Group format 4. Group size 5. Facilitators 6. Involvement of supportive others 7. Homework 8. Questionnaire booklet 9. Accompanying materials 10. Recruitment 11. Strategies for managing fatigue 12. Barriers to change 13. Long term impact

Table 1 (continued)

Authors	Methods	Participants	Intervention	Main Results
Wendebourg et al. (2016) [46]	• Qualitative study • Convenience sample • Interviews before intervention and focus group after intervention • Framework analysis	• $N = 4$ interviews, $N = 12$ in focus group • Median age = 47.5 years, 56.2% female, 81.2% with EDSS between 2.5 and 4	Fatigue management CBT intervention, in person, group, 6 weeks (3 sessions)	1. Experiences with fatigue ■ Fatigue burden and causes ■ Resting ■ Adverse effects on daily life ■ Psychological effects ■ Stressors 2. Applied coping strategies ■ Sports ■ Sleep ■ Balanced lifestyle ■ Acceptance 3. Social experiences ■ Lack of understanding 4. Vision of life without fatigue ■ Activities 5. Expectations and wishes concerning the program ■ Content ■ Treatment options ■ Contact with other patients ■ Type of program ■ Duration per session ■ Homework*
Ytterberg et al. (2017) [47]	• Qualitative study • Voluntary response sample • Semi-structured interviews after completion of intervention • Inductive content analysis	• $N = 3$ psychotherapists • Mean age = 55 years, 100% women	CBT intervention, in person, individual, 15–20 sessions	1. Trusting their expertise as psychotherapists while lacking MS-specific knowledge ■ Understanding the complexity of living with MS ■ Perceived lack of knowledge of MS giving rise to insecurity 2. The process of exploring the participants' readiness for CBT with modifications of content and delivery ■ Expectations as to the patients being more prepared for undergoing a demanding therapy ■ Motivated for CBT despite cancellations and problems with logistics ■ Modifications to make the CBT more concrete

ACT acceptance and commitment therapy, *CBT* cognitive behavioural therapy, *EDSS* Expanded Disability Status Scale, *MDD* major depressive disorder, *MS* multiple sclerosis, *PPMS* primary progressive multiple sclerosis, *PwMS* people living with multiple sclerosis, *RCT* randomized controlled trial, *RRMS* relapsing–remitting multiple sclerosis, *SCT* social-cognitive theory

* Demographic data on qualitative study participants not reported/could not be extracted

Table 2 Quality appraisal of included studies (using the Critical Appraisal Skills Programme [CASP] checklist for qualitative studies)

	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Babbage et al. (2019) [49]	Y	Y	Y	CT	Y	Y	Y	Y	Y	V
Bogosian et al. (2016) [22]	Y	Y	Y	Y	Y	CT	Y	Y	Y	V
Borghi et al. (2018) [33]	Y	Y	Y	CT	CT	CT	Y	Y	Y	V
Brown et al. (2016) [48]	Y	Y	Y	Y	Y	Y	Y	Y	Y	V
Dennison et al. (2013) [34]	Y	Y	Y	Y	Y	Y	Y	Y	Y	V
Dunne et al. (2021) [23]	Y	Y	Y	Y	Y	N	Y	Y	Y	V
Fortune et al. (2020) [24]	Y	Y	Y	Y	Y	Y	Y	Y	Y	V
Giovannetti et al. (2022) [36]	Y	Y	CT	Y	CT	CT	Y	Y	Y	V
Giovannetti et al. (2022) [35]	Y	Y	Y	Y	CT	CT	Y	Y	Y	V
Giovannetti et al. (2020) [25]	Y	Y	Y	Y	Y	CT	Y	Y	Y	V
Gottberg et al. (2016) [44]	Y	Y	Y	CT	Y	CT	Y	Y	Y	V
Harrison et al. (2017) [37]	Y	Y	Y	Y	Y	CT	Y	CT	Y	V
Hersche et al. (2019) [26]	Y	Y	Y	Y	Y	N	Y	CT	N	NV
Hind et al. (2010) [43]	Y	Y	Y	Y	Y	CT	Y	Y	Y	V
Holmes et al. (2012) [27]	Y	Y	Y	CT	Y	Y	Y	N	N	NV
Learmonth et al. (2019) [28]	Y	Y	Y	CT	Y	Y	Y	Y	Y	V
Meek et al. (2021) [29]	Y	Y	Y	Y	CT	Y	Y	Y	Y	V
Moss-Morris et al. (2012) [30]	Y	Y	Y	Y	Y	Y	Y	CT	CT	V
Pakenham et al. (2018) [38]	Y	Y	Y	Y	CT	N	Y	Y	CT	NV
Pöttgen et al. (2015) [39]	Y	Y	Y	CT	Y	CT	Y	Y	Y	V
Proctor et al. (2018) [31]	Y	Y	Y	Y	CT	Y	Y	N	N	NV
Russell et al. (2022) [45]	Y	Y	Y	Y	Y	Y	Y	Y	Y	V
Ryan et al. (2020) [32]	Y	Y	Y	Y	Y	Y	Y	CT	N	NV
Shevil and Finlayson (2009) [40]	Y	Y	Y	Y	Y	Y	Y	Y	Y	V
Thomas et al. (2021) [42]	Y	Y	Y	Y	Y	CT	Y	Y	Y	V
Thomas et al. (2010) [41]	Y	Y	Y	Y	Y	Y	Y	Y	Y	V
Wendebourg et al. (2016) [46]	Y	Y	Y	Y	Y	CT	Y	Y	CT	V
Ytterberg et al. (2017) [47]	Y	Y	Y	CT	Y	Y	Y	Y	Y	V

CT can't tell, *N* no, *NV* not valuable, *V* valuable, *Y* yes

Q1. Was there a clear statement of the aims of the research?

Q2. Is a qualitative methodology appropriate?

Q3. Was the research design appropriate to address the aims of the research?

Q4. Was the recruitment strategy appropriate to the aims of the research?

Q5. Was the data collected in a way which addressed the research issue?

Q6. Has the relationship between the researcher and participants been adequately considered?

Q7. Have ethical issues been taken into consideration?

Q8. Was the data analysis sufficiently rigorous?

Q9. Is there a clear statement of findings?

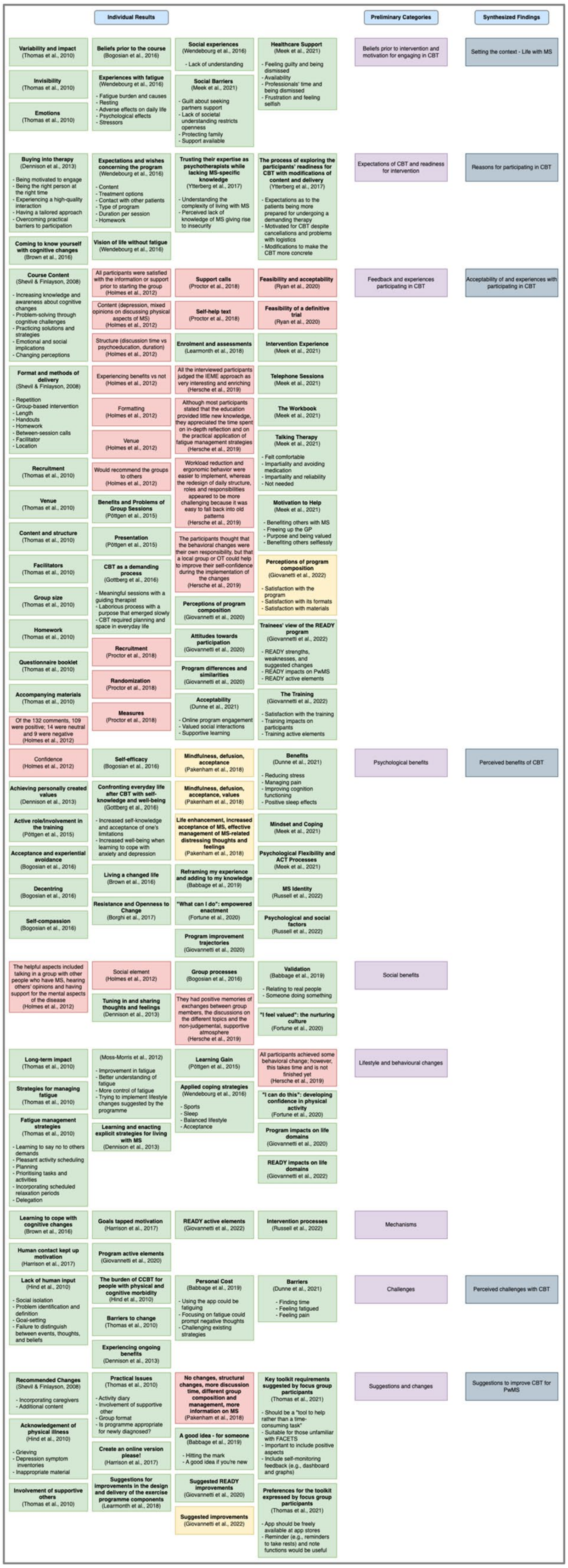
Q10. How valuable is the research?

nine categories. Categories with overlapping meanings were subsequently combined into six synthesized findings: (1) setting the context – life with MS, (2) reasons for participating in CBTs, (3) acceptability of and experiences with participating in CBTs, (4) perceived benefits of CBTs, (5) perceived challenges with CBTs, and (6) suggestions to improve CBTs for PwMS. Notably, two studies reported observations of low credibility (i.e., lack of supporting participant quotes), rather than cohesive themes, but were still included as they met the quality appraisal cut-off for inclusion [26, 27]. Key points from these studies were extracted in place of themes.

Setting the context—life with MS

In terms of health status prior to engaging in CBTs, PwMS commonly described hidden symptoms and the impact of these symptoms on their daily lives [22, 41, 46]. Examples included fatigue, stress, and cognitive changes, which interfered with their ability to engage in day-to-day tasks.

Fig. 2 Categorization and synthesis of results from included studies. ▸ The credibility of results in green were rated as “unequivocal”, results in yellow were rated as “credible”, and results in red were rated as “not supported”



> Well, my family forget … I think they genuinely forget coz I just walk into the house as I am. I haven’t got a label on my head, they just forget all the time & I just sit there & think ‘oh I’m so tired’. (Person with MS) [41]
>
> I feel worse, if I rest I feel worse. I feel guilty coz I haven’t done anything that day. (Person with MS) [41]

Often, when others did not validate these symptoms, PwMS experienced guilt and felt as if they were a burden to those around them [29, 46]. This included interactions with family members and healthcare providers.

> Not even my husband understands me. He thinks I’m just lazy. (Person with MS) [46]
>
> It feels like you’re wasting your GP’s time. (Person with MS) [29]

Reasons for participating in CBTs

Participants of CBTs similarly expected to gain self-management skills and learn more about their symptoms [46, 48]. However, studies of both participants and psychotherapists noted that these expectations could not be met if those living with MS did not feel ready to participate or unpack their symptom experiences [34, 47]. This emphasizes the potential importance of timing for CBT interventions.

> I would like to know the difference between fatigue and normal tiredness. (Person with MS) [46]
>
> I think it depends on the stage you’re at … and how accepting you are of it … and until people (with MS) are actually ready and willing to listen, you could talk to them all you want, it’s going to go through one ear out the other. (Person with MS) [48]

Psychotherapists expressed the desire to help PwMS through CBTs and believed it could help [47]. However, they also expressed a desire to have more disease- and treatment-specific training on MS prior to administering these interventions [47]. Although they were able to learn about MS symptomatology from patients directly, not understanding the medical aspects of MS made it difficult to set realistic treatment goals.

> What was difficult, I’d say, was to distinguish the sort of tiredness that’s depressive, that’s a symptom of depression. But after a time I did feel that there was something specific about this sort of tiredness – specific to MS, I believe, and I thought I could distinguish

> it, you might say. There was something I thought made it different from the depression I'd treated before. (Psychotherapist) [47]

Acceptability of and experiences with participating in CBTs

Participants in CBTs generally expressed acceptability and satisfaction with the intervention as a whole [23, 26, 27, 32, 35, 35, 39]. Constructive feedback regarding specific aspects of the CBT programming was also provided [25–29, 31, 35, 36, 39–41]. In terms of formatting, online delivery (i.e., live/synchronous) was accepted by participants and they expressed that it helped to reduce barriers to participation such as needing to travel to a physical location [23].

> I really enjoyed the online version of mindfulness program as I didn't have to travel anywhere and could stay in the comfort of my own home. (Person with MS) [23]

In terms of length, participants in one study expressed the desire for the program to extend beyond five weeks [40].

> We really thought there should be more hours per session and more sessions overall. The whole group agreed. (Person with MS) [40]

The content in certain CBT interventions was also accepted by participants [29, 40, 41]. When the material was tailored towards PwMS, participants stated that this enabled them to meet their expectations of the intervention (e.g., increased awareness and understanding of cognitive processes).

> I really didn't know what cognitive meant other than finishing a task and being able to set off some kind of organized plan… I didn't realize all the other ramifications about things like trouble finding words or losing things … I learned a lot. (Person with MS) [40]
> Unhelpful thoughts, yes you suddenly realise that you do feel guilty a lot more often than you thought you did for sitting down doing nothing and then learning to address that. I thought that was quite useful. (Person with MS) [41]

Additional aspects of CBT including homework, workbooks, and interactions with facilitators were also discussed and generally accepted by participants [25, 27, 29, 31, 39–41]. In some interventions, participants also received support calls from CBT facilitators in which they could discuss their personal challenges and experiences with the program. Feedback on the duration of support calls with the CBT facilitator was mixed, as some participants found them to be tiring while others suggested a longer duration [29, 31, 40].

> [The facilitator] made us feel that what was happening to us wasn't because we weren't smart or educated or knew how to get through life. (Person with MS) [40]
> [Support calls] were just right; I just wish that they'd been a bit longer. (Person with MS) [29]

Importantly, engaging in CBT was described as a "demanding process" requiring meaningful interactions with facilitators, acting with intention, and allowing for the outcomes to emerge slowly over time [26, 44].

> I think that if you really want to sort of go all the way into it, really give yourself up to it and try to get something out of it, you've got to devote time- and that's terrifically demanding. (Person with MS) [44]

Perceived benefits of CBTs

Psychological benefits were the most commonly reported positive outcome of CBTs by participants. These benefits were predominantly related to changes in mindset (e.g., self-acceptance, self-efficacy, and self-compassion) and resulting improvements in coping skills [22, 25, 27, 29, 33, 34, 38, 39, 44, 45, 48, 49]. Additional psychosomatic benefits such as reductions in pain and improvements in sleep were also reported [23].

> I really truly think that I've moved on, quite a few steps towards acceptance of this god awful condition that we've all got. I can say it quite cheerfully and be quite pragmatic. (Person with MS) [22]
> I think about my MS every single day and that will never change but I don't … a thought will come into my head now and I can get rid of it as quickly as it came in. I'm not in that same dreadful thought process that I was before. (Person with MS) [34]

Social benefits were also cited, particularly among participants in group-based CBT interventions [22, 24, 26, 27, 34, 49]. For instance, being able to meet other PwMS was commonly described as validating. These interactions helped to establish a sense of belonging and comradery which increased participants' motivation to complete the program.

> It just makes me feel more of a human or something to relate to somebody else who's having the same kind of experiences. (Person with MS) [49]
> Commonality of the disease I found very helpful cause you're all going through and can share the same difficulties and often the same fears (Person with MS) [22]

Participation in CBTs also led to deliberate changes in lifestyle and behaviour [24–26, 30, 34, 35, 39, 41, 46]. Examples included applying coping skills to everyday situations, engaging in health-promoting behaviours such as exercise, and making time for relaxation.

> … And you do have those thoughts come flooding over but it's learning what to do with those thoughts. (Person with MS) [34]
> I am no more on automatic pilot. I notice what is around me (sounds, smell, etc.). When my resilience is low I stop, relax and breathe. Once per week I cultivate my social relationships with family and friends. This program has changed my life, the way I connect with people, with myself and the disease. (Person with MS) [35]

These benefits were attributed to various aspects of the program. For instance, interacting with other participants helped to increase motivation to attend CBT sessions, ultimately leading to social and psychological benefits [37, 45]. Meanwhile, the content of the CBT sessions encouraged activities such as goal setting, which led to psychological and lifestyle changes [25, 35, 37, 45, 48].

> It was the motivation and seeing other people doing it and saying well they can do it, so can I and that was the encouragement to me then to stay doing it. (Person with MS) [45]
> When I am overwhelmed, I learned to stop, notice and let thoughts and emotions go, I can recognise what really matters to me and chose how to behave… (Person with MS) [35]

Perceived challenges with CBTs

CBT interventions lacking therapist contact were identified as challenging by participants [43]. For instance, self-administered interventions using an app or computer were described by some participants as lacking in empathy, isolating, fatiguing, and inaccessible.

> With MS you can become very isolated because of your disability … So, I think when working with something that is a computer programme it makes you feel even more like you're not speaking to someone face to face. You don't get empathy there. (Person with MS) [43]
> Typing increases discomfort in my dominant right hand. … It's a bit tiring sitting there clicking away … because I have a bit of a problem with my right hand and I sort of, you know you're click, click, click. (Person with MS) [43]

Personal factors including physical symptoms and mental health were also identified as potential barriers to participation [23, 34, 41, 43, 49]. For example, symptoms of fatigue and pain could make it difficult to concentrate or fully engage in the intervention.

> Pain is making it difficult to focus on mindfulness. (Person with MS) [23]
> … The reason I wouldn't keep using the app is my experience of fatigue while I was putting so much thought into it was actually worse. (Person with MS) [49]

Suggestions to improve CBTs for PwMS

Tailoring the content/material presented in CBT interventions to be relevant for PwMS was crucial for many participants [25, 40, 43]. For instance, ensuring that the activities presented to participants are appropriate for their range of motion. In a study using a pre-existing CBT intervention, participants felt that the recommendations being made were insensitive and potentially triggering to those with physical disabilities [43].

> … And it says things like … playing baseball … The topics that it is suggesting are not MS-appropriate … It gave me a score and advised me that if I wanted to be happier, I ought to do more … and I'll be honest with you, were I sort of, more depressed with my situation than I possible am, I think I would be suicidal by the time I finished this. Because it has basically spent a vast amount of time telling me that I'm not happy because I can't do what I might otherwise do. (Person with MS) [43]

The importance of selecting an appropriate target audience for CBTs was discussed in a few studies [41, 49]. While some participants indicated that CBTs could be helpful for newly diagnosed patients (particularly the education components), others noted that the right time to engage in CBTs is dependent on the individual. The option of incorporating caregivers into CBT programming was also suggested (i.e., to educate and allow for caregivers to discuss with one another) [25, 40, 41].

> Newly diagnosed should be aware about the symptoms and understand why they're tired … but it's got to be timed at the right time for that individual person. (Person with MS) [41]
> Sometimes significant others need to hear what other significant others are going through … and then maybe at the end the two groups come together as sort of a wrap up. (Person with MS) [40]

Improving the accessibility of CBT interventions was also important to participants [37, 42]. This included making interventions freely available, offering online options, and ensuring that the intervention was not overly time-consuming.

> If the whole thing was on a website, you could just click onto the website and choose what you wanted to do. (Person with MS) [37]
>
> You have to be careful not to over … put so much information that it becomes overwhelming, that you can analyze down to so much and you think I can't think about this anymore and you put too much into it. (Person with MS) [42]

Modifying the formatting of CBT interventions was also discussed by some participants [25, 27, 28, 35, 38]. For instance, allowing more time for participants to socialize/connect with each other and spending less time on didactics.

> Too many Powerpoint's, just want to chat and didn't want a formal agenda. (Person with MS) [27]

Certainty of evidence

The eight individual results included in the first synthesized finding (setting the context – life with MS) were rated as "unequivocal" (i.e., unequivocally supported) [22, 29, 41, 46]. The six individual results included in the second synthesized finding (reasons for participating in CBTs) were also all rated as "unequivocal" [34, 46–48]. In the third synthesized finding (acceptability of and experiences with participating in CBTs), 25 of the results were rated as "unequivocal" [23, 25, 28, 29, 35, 36, 39–41, 44], one was rated "credible" [35], and 19 were rated as "unsupported" [26, 27, 31, 32]. In the fourth synthesized finding (perceived benefits of CBTs), 37 of the results were rated as "unequivocal" [22–25, 29, 30, 33–37, 39, 41, 44–46, 48, 49], three as "credible" [38], and five as "unsupported" [26, 27]. All six results in the fifth synthesized finding (perceived challenges with CBTs) were rated as "unequivocal" [22, 33, 40, 42, 48]. Lastly, 10 of the results included in the final synthesized finding (suggestions to improve CBTs for PwMS) were rated as "unequivocal" [25, 28, 37, 40–43, 49], one as "credible" [35], and one as "unsupported" [38]. Overall, most of the synthesized findings were based upon unequivocal results from the included studies. These ratings are illustrated in Fig. 2.

Discussion

Summary of main findings

A total of 28 studies with qualitative results were synthesized in this meta-aggregation. These studies explored the experiences of both PwMS and clinicians in using CBT-based interventions to address hidden symptoms associated with MS. Nine preliminary categories were formed using the 122 qualitative results from the included studies, eventually leading to six synthesized findings. These synthesized findings were: (1) setting the context–life with MS, (2) reasons for participating in CBTs, (3) acceptability of and experiences with participating in CBTs, (4) perceived benefits of CBTs, (5) perceived challenges with CBTs, and (6) suggestions to improve CBTs for PwMS.

Setting the context–life with MS

Recognition of the prevalence and impact of hidden symptoms among PwMS is critical in informing CBT programming for PwMS. For many PwMS, hidden symptoms such as fatigue and stress can be prominent and cause disruptions to everyday life. Unfortunately, these experiences are often misunderstood or invalidated by others. PwMS described both invalidating social experiences with members of their social circle (e.g., spouses) as well as with healthcare providers. In this regard, feelings of guilt and invalidation were recurrent in interviews with PwMS. Thus, CBT interventions could provide an opportunity to address these challenges (e.g., through building communication strategies and delivering the material with compassionate language).

Reasons for participating in CBTs

The notions of preparation and readiness among both PwMS and CBT facilitators were recurring. Participants in CBT interventions similarly expressed the desire to gain self-management skills and learn more about their MS symptoms. However, these expectations could not be met if participants were not ready to participate or unpack their symptom experiences. Moreover, psychotherapists administering CBTs expressed the desire to have more specialized training on MS in order to provide an intervention suitable for PwMS. As goal consensus has been identified as a key mediator in psychotherapy outcomes, this further highlights the importance of tailoring CBTs towards the needs and preferences of PwMS [51]. Efforts to explore PwMS' readiness for CBT, as well as to prepare facilitators to deliver appropriate treatment, are warranted.

Acceptability of and experiences with participating in CBTs

PwMS that participated in a CBT intervention generally expressed acceptability and satisfaction. This extended to specific aspects of the intervention such as the content and activities. PwMS noted the importance of repetition, practical steps in managing symptoms, workbooks, and group-based interventions. Nevertheless, there was also variability among participants. For instance, some participants preferred longer sessions while others did not. This heterogeneity among PwMS suggests that a "one size fits all" approach to CBT for PwMS may not be appropriate. Finally, CBT was described as a “demanding process” requiring PwMS to make deliberate efforts to engage with the program. The implications for the design of future CBT programs include some key content areas, the role of facilitators/providers, and offering CBTs in an accessible format (i.e., online).

Perceived benefits of CBTs

A range of benefits from participating in CBTs were reported by PwMS. Psychological benefits included changes in mindset, improved coping skills, and improvements in hidden symptoms. Social benefits included meeting and connecting with other PwMS which many described as validating. Changes in lifestyle and behaviour were also discussed (e.g., applying coping skills learned through CBT to everyday life, exercising, and making time for relaxation). Interestingly, participants of group-based CBTs reported that these settings increased their motivation to participate and adhere to the intervention, in addition to the social benefits. These findings suggest multiple benefits across multiple domains (e.g., enhanced well-being and acceptance of the condition, reframing the experience, and reductions in hidden symptoms), which were enhanced in group-based settings. Social desirability and social support (i.e., human connection) are both well-recognized ‘common factors’ in mediating group psychotherapy outcomes, and may be leveraged when designing CBT interventions for PwMS [51, 52].

Perceived challenges with CBTs

Participants of self-administered CBT interventions (with little to no therapist contact) commonly reported feelings of isolation and fatigue. This further suggests the importance of integrating social connection (i.e., with other PwMS and/or facilitators) into the design of CBT interventions. Moreover, PwMS also reported that their physical and mental health symptoms also made it challenging to engage in CBTs (particularly in the context of virtual CBT interventions). Although online delivery may improve some aspects of accessibility (e.g., by reducing travel barriers), it may also pose a different set of challenges. As such, the efforts to minimize physical demands such as typing and clicking or offering alternative ways to engage with the material should be explored to enhance accessibility and participation.

Suggestions to improve CBTs for PwMS

Numerous suggestions to improve CBTs for PwMS were made. Tailoring the content in CBT interventions to be relevant to MS was crucial, as inappropriate content (e.g., recommendations to engage in organized sports) was found to be both insensitive and upsetting to some participants. Identifying a target population was discussed, as some interventions seemed to be more useful for newly diagnosed PwMS, while others suggested that potential participants should not engage unless they are prepared to fully commit. Therefore, assessing the baseline health status and cognitive profile of incoming participants may be useful to tailor their CBT treatments accordingly. Finding ways to incorporate caregivers into CBT interventions was suggested and may be especially relevant in terms of addressing family dynamics/invalidating social experiences described by PwMS. Improving the accessibility and format of CBTs (e.g., cost, online delivery, length, allocation of time) were also discussed [52, 53].

Comparison with the existing literature

The results from this review overlap with much of the existing literature on lived experience with MS and with psychotherapy. In a recent study exploring the experiences of psychological therapies for PwMS, many participants described feelings of guilt and shame resulting from personal “choices” made before their diagnosis [54]. PwMS included in this review also described feelings of guilt, but in relation to interactions they had with loved ones and support figures. This suggests that feelings of guilt can persist across multiple points in time and arise from a variety of sources. In another study exploring PwMS’ preferences for psychological support, participants reported the desire to increase psychological well-being (37%) and learn self-management skills to apply in future situations (23%) [55]. These findings parallel the expectations and benefits of CBTs expressed by PwMS included in this review.

The wide-ranging benefits of CBTs found in this review have similarly been reported in a scoping review on third-wave CBTs for PwMS [12]. For instance, improvements in mental health symptoms, coping skills, and overall perspective were similarly described in their review [12]. Crucially, the importance of social connection has been reported across numerous studies and contexts. Studies on peer support programs for people living with chronic conditions [56] and MS [52] have reported significant improvements in self-efficacy, health-directed activity, and health status. Moreover, in a meta-aggregation specifically on Mindfulness-based interventions for PwMS, feelings of belonging and camaraderie were valued components of the intervention [57]. Finally, in a study exploring motivation to participate in digital data collection research, PwMS ranked the ability to exchange experiences with other PwMS as one of the top motivators [58]. Taken together, the importance of social connection has been well-documented, and is applicable to group-based CBTs as well.

The suggestions to improve CBTs for PwMS overlap with many of the key considerations included in a recent systematic review broadly covering psychological interventions for people with neurodegenerative diseases [59]. Similarities in recommendations included personalizing the intervention to meet the needs and preferences of recipients, improving the accessibility of interventions, and offering support at key points in the disease trajectory [59]. As many similarities in themes were discussed, this overlap speaks to the overarching experiences of people with neurodegenerative diseases and the need for comprehensive psychological therapies in this population.

Strengths and limitations of included studies

A fairly large number of studies reporting qualitative experiences engaging in CBTs were identified in the literature. Many of these studies used qualitative data as a means to support or explain the results from quantitative strands, as well as to independently explore experiences. In addition, studies including both PwMS and clinicians were identified in the literature. The inclusion of diverse knowledge user groups allows for a greater understanding of the implications of CBTs in the context of MS. Finally, the studies included in this review mostly had a low risk of bias and reported unequivocal or credible results.

Although studies involving psychotherapists' and physiotherapists' perspectives were included in this review, only three studies on these knowledge user groups were identified in the literature [32, 36, 47]. Moreover, studies exploring the perspectives of additional healthcare providers (e.g., primary care providers, neurologists, physiatrists, nurse practitioners, etc.) and caregivers of PwMS could not be found. As PwMS often rely on healthcare providers and caregivers for support, understanding how these knowledge user groups perceive CBTs is crucial. Few studies included in this review reported on participants' ethnicities and levels of education. Among those that did, participants were mainly reported to be White and to have a higher level of education. This raises issues around the generalizability of these findings to non-White populations and those with fewer years of education. Finally, most of the studies included in this review were conducted in the UK or Europe, further raising issues of generalizability.

Strengths and limitations of this review

This review explored the experiences of PwMS and clinicians with CBT interventions in the context of hidden symptoms associated with MS. A rigorous methodology was applied, with the literature search performed by an experienced health sciences librarian; and the screening, appraisal, extraction, and certainty of evidence assessments performed in duplicate by two reviewers. By aggregating the results from 28 studies, the experiences of knowledge users before, during, and after CBTs could be characterized. Moreover, the inclusion of other key stakeholder perspectives allowed for a more nuanced exploration of CBTs in PwMS. Nevertheless, this review is constrained by the limitations of the studies included in this review. Namely, the lack of feedback from caregivers and physicians, as well as generalizability to diverse populations. In addition, studies not published in English or without a full-text version could not be included, which may have limited the results of this review. Moreover, additional studies of relevance may not have been detected during the literature search or could have been mistakenly excluded.

Suggested "lines of action"

1. **Group setting**: To encourage the benefits of peer support, CBT interventions for PwMS may best be conducted in group settings. Dedicated time for participants to interact with each other and discuss their current challenges is key.
2. **Delivery**: Allow for online delivery of (synchronous) CBT interventions to reduce barriers to participation and increase accessibility, while exploring ways to incorporate and retain human connection. Exploring ways to minimize the burden and potential access barriers associated with computer use (e.g., clicking and other repetitive/straining motions) is warranted.
3. **Training**: Facilitators of CBTs should undergo specialized training to understand the nuances of MS symptoms, the impact of these symptoms, and ways to actively validate PwMS' symptom experiences
4. **Tailoring**: The content included in CBT interventions for PwMS should be tailored to ensure relevance and usability (i.e., taking into account disability and other MS-specific issues that can contradict the recommendations made in non-tailored CBTs)
5. **Design**: Co-design with PwMS and further research on participant preferences is warranted in order to optimize specific aspects of CBTs such as timing, length, format, setting, etc.
6. **Stakeholder engagement**: Involvement of caregivers and other healthcare providers in future studies exploring CBTs for PwMS to further understand its perceptions, as well as to address invalidating experiences that can result from these groups
7. **Timing**: Exploration of the ideal window for PwMS to engage in CBTs in future studies (e.g., newly diagnosed vs long-standing MS)

Conclusion

A range of benefits including psychological, social, and lifestyle changes are reported by PwMS following CBTs. Some of these benefits appear to be generic, but CBT interventions specifically tailored for PwMS were often linked to more positive benefits and fewer challenges. Thus, future CBT interventions should be tailored to PwMS, delivered in group settings to enable the benefits of peer support, offer online options to increase accessibility, and be delivered by a trained facilitator. Further exploration of the ideal CBT design for PwMS, as well as engagement with caregivers and clinicians treating MS, is warranted.

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Declarations

Conflicts of interest The authors have no conflicts of interest to declare.

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