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SYSTEMATIC REVIEW

Behaviour change interventions targeting physical activity in adults with fibromyalgia: a systematic review

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Abstract

Physical activity (PA) and exercise programmes are recommended for the management of fibromyalgia. Despite positive effects on symptoms and function, PA promotion remains a significant clinical challenge. Behaviour change theories and techniques are recommended as part of complex health interventions; their integration into interventions aimed at PA behaviour in people with fibromyalgia is not known. This review explored behaviour change interventions targeting PA in adults with fibromyalgia. A systematic review was conducted; randomized and quasi-randomized controlled trials with at least one behaviour change intervention targeting PA were included. MEDLINE/OVID, EMBASE, PEDro, PsychINFO, CINAHL, Scopus, Web of Science, the Cochrane Central Register of Controlled Trials and relevant conference abstracts were searched. Two authors independently screened studies for inclusion and performed risk of bias assessments. Articles were reviewed for their use of behaviour change theory and behaviour change techniques (BCTs). The search identified 2491 records, from which eight studies (1416 participants) were included. PA and exercise behaviours were the primary focus of four interventions and were components of broader interventions in four studies. Behaviour change theories informed four interventions. Thirty-two different BCTs were used across studies. Five studies reported improvements in PA either post-intervention or at follow-up. Two studies used objective PA measures and seven studies used self-report measures. Short-term benefits in pain, quality of life, and physical fitness were also observed. Behaviour change interventions targeting PA in people with fibromyalgia have had limited success to date. With significant variations in intervention designs, the optimal intervention remains unknown.

Keywords Physical activity · Exercise · Fibromyalgia · Health behaviour · Behaviour change

Introduction

Fibromyalgia is characterised by chronic widespread pain, generally accompanied by one or more symptoms of fatigue, sleep disturbances, cognitive difficulties, and depression [1, 2]. The prevalence among the general population is approximately 3–5%. Fibromyalgia is three times more common in women [1, 3], and although poorly understood, it is generally accepted to be a disorder of central pain processing [1, 4].

In recently published guidance, EULAR recommended that the initial management of fibromyalgia should focus on non-pharmacological therapies, in the context of an individualised care plan, tailored through shared decision-making [5]. Therapeutic exercise programmes are core components of such an approach. In the last decade, a number of systematic reviews and meta-analyses have synthesised the outcomes of clinical studies exploring the effects of exercise. Their findings underpin a strong recommendation for exercise [5, 6], with aerobic exercise [7], resistance programmes [8], and aquatic training [9] demonstrating positive effects on symptoms and physical function; this is despite an initial increase in symptoms thought to be related to deconditioning and the effects of chronic pain.

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Such condition-specific exercises are just one component of the broader concept of 'physical activity' (PA), which includes "any bodily movement produced by skeletal muscles that results in energy expenditure" [10]. Work, transport, leisure time, and domestic activities all contribute to free-living energy expenditure. The health-related benefits of PA are numerous and well documented in the general population. PA has been found to reduce the risk of cardiovascular disease, obesity, colon and breast cancers, type 2 diabetes, and osteoporosis [11, 12]. It also improves musculoskeletal health and reduces the symptoms of depression [13].

Despite these benefits, PA and exercise promotion remains a significant clinical challenge in adults with fibromyalgia [14]. Objectively measured free-living PA suggests that individuals with fibromyalgia are less physically active than healthy controls [15, 16], spending significantly less time in moderate/vigorous physical activity [17] (MVPA) with just ~ 20% of women with fibromyalgia meeting recommendations for weekly PA [18]. Numerous factors influence PA behaviour, by mediating, moderating, or confounding the behaviour [19]. Few studies have explored correlates of maintenance of exercise following interventions in fibromyalgia, although it appears to be contingent on being able to deal with stress, pain, barriers to exercise, and disability [16, 20].

To maximise the potential benefits of PA and exercise-based interventions, it is necessary to understand behaviour and behaviour change. Over 80 theories of behaviour and behaviour change exist in the social and behavioural sciences [21], but their application to health-enhancing PA in fibromyalgia is largely unreported. A behaviour change technique (BCT) is "an observable, replicable, and irreducible component of an intervention designed to alter or redirect causal processes that regulate behaviour" [22]. The development of the BCT taxonomy [22] allows complex and diverse behaviour interventions to be systematically described, evaluated, and replicated. To date, BCTs targeting PA in people with fibromyalgia have not been explored.

This systematic review aims to appraise the effects of behaviour change interventions targeting PA in adults with fibromyalgia. In addition, this review will synthesise the approaches to behaviour change interventions, including use of underlying theories and BCTs, reported in the literature.

Methods

The reporting of this study was guided by the 'Preferred Reporting Items for Systematic Reviews and Meta-analyses' (PRISMA) recommendations [23]. The study protocol for this review was prospectively registered online with a registry of systematic reviews (http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42017060646).

Study eligibility criteria

Randomised controlled trials (RCTs) and quasi-RCTs in which at least one of the groups received a behaviour change intervention targeting PA behaviour, using at least one recognised BCT, were considered for inclusion. Review articles, observational studies, case reports, commentaries, and studies with ≤ 5 participants were excluded.

Studies comparing behaviour change interventions to no intervention or standard care controls, or other specified interventions were eligible. PA could be one of a number of target behaviours within an intervention aiming to change multiple health behaviours, or could be the sole focus of an intervention. Studies not directly targeting PA behaviour were excluded.

Studies involving adults with a reported diagnosis of fibromyalgia according to established diagnostic criteria were eligible. Studies including participants younger than 18 years of age were excluded.

The primary outcome of interest was PA level measured over at least 24 h. This included both objective measures (e.g., by accelerometer, pedometer) and self-report measures (e.g., questionnaires, diaries). Outputs expressed as continuous variables (e.g., steps per day, minutes of MVPA per day) or categorical variables (e.g., high/moderate/low PA level) were eligible. The primary outcome was a prerequisite for study inclusion; if present, secondary outcomes were considered, including physical fitness outcomes (e.g., BMI, VO_{2MAX}), condition-related outcomes (e.g., Fibromyalgia Impact Questionnaire, Widespread Pain Index), quality of life (e.g., SF-36), and reports adverse events (e.g., injury).

Sources and study selection

Studies were retrieved by searching MEDLINE (OVID), EMBASE, PEDro, PsychINFO, CINAHL, Scopus, Web of Science, and The Cochrane Central Register of Controlled Trials from their inception to July 2018 (date of final search). Search terms were adapted for use within each database, and consisted of common keywords and medical subject headings related to fibromyalgia, PA and exercise, and behaviour change. No search restrictions were imposed. See Supplementary File 1 for search strategy.

The electronic database search was supplemented by searching abstracts from the annual congresses of the European League Against Rheumatism (2002–2018), the American College of Rheumatology (2006–2017) and the World Confederation of Physical Therapy (2003–2017). When only abstracts were available in the published literature, authors were contacted seeking full-text manuscripts of relevant studies. Finally, a hand search of the reference lists of included studies was conducted.

Two reviewers (TOD and FW) independently screened titles, abstracts, and keywords to identify studies that potentially met the review eligibility criteria. Full texts of these reports were retrieved and assessed for eligibility (TOD and FW). Disagreements between reviewers on inclusion were resolved by discussion to achieve a consensus, and in the absence of agreement, a third reviewer (FW) was consulted. Covidence systematic review software (Veritas Health Innovation, Melbourne, Australia) was used during the study selection process.

Data extraction and analysis

A standardised data extraction template based on Cochrane recommendations was piloted on two randomly selected studies, and modified accordingly for this review [24]. The review team recorded the following: (1) study characteristics; (2) participant characteristics; (3) features of the behaviour change intervention and comparator intervention groups; and (4) relevant primary and secondary outcome data. In cases where elaboration on published material was needed, or further data were required, study authors were contacted to request additional detail.

The Behaviour Change Technique Taxonomy Version 1 (BCTTv1) [22] was used to code BCTs. The BCTTv1 is an extensive, integrated, hierarchical classification system for reliably specifying intervention components. It includes 93 BCTs, in 16 groupings [22]. A BCT was only coded when there was clear evidence of inclusion in the published manuscript or in additional material provided by study authors.

For continuous data, the mean difference (MD) between groups with 95% CI was calculated. For continuous data reported using different scoring systems, standardised mean difference was used. Dichotomous data were reported as risk ratios with 95% confidence intervals (95% CI). Where studies reported outcomes at multiple timepoints, data were extracted at clinically relevant timepoints, e.g., short term (0–12 weeks following the intervention), medium term (3–9 months), and longer term (longer than 9 months).

Data synthesis

When appropriate, results of comparable groups of trials were pooled; in such cases, a random-effects model was used. This decision to pool and the choice of model were guided by the extent of heterogeneity of effect size across the pooled studies. Clinical heterogeneity was assessed subjectively based on data about the participants, interventions, and outcome measurements of each study. Statistical heterogeneity was further assessed by visual inspection of effect estimate similarities, the overlap of confidence intervals on forest plots, and consideration of the statistical test output of the Chi-square test ($P < 0.01$ was interpreted as significant heterogeneity) and the I^2 statistic (I^2 of 75–100% may be interpreted as considerable heterogeneity, as suggested by Higgins et al. [24]). Due to heterogeneity of interventions and outcome measures, planned meta-analyses and sub-group analyses were not appropriate for primary outcomes; these data were presented in tables and synthesised qualitatively.

Risk of bias and levels of evidence

A risk of bias appraisal of included studies was performed independently by two reviewers (FW and SM). Disagreements between the reviewers were resolved through discussion to achieve consensus. Failing agreement, a third reviewer (TOD) arbitrated. The Cochrane collaboration's risk of bias tool rated risk of bias across six domains as low, high, or unclear [25]; the domains included selection bias, performance bias, detection bias, attrition bias, reporting bias, and other sources of bias.

Results

Study selection

A total of eight studies [26–33], reported across 11 articles published between 1998 and 2017, were included in this review. The search strategy is summarised in Fig. 1. When multiple articles reported different data from the same study, results were pooled under a primary study. As such, the primary study by Ang et al. [31] was pooled with reports of secondary analysis [34, 35], and Fontaine et al. [29] was pooled with follow-up data [36]. Two authors, when contacted, provided additional information relating to their study [30, 37].

Study characteristics: design and participants

Seven RCTs [27–33] and one quasi-RCT [26] were included in this review. Studies with two-armed designs compared behaviour change interventions to education [29–31] or usual care [28], or compared a combination of behaviour change and exercise interventions to relaxation [32, 33] or relaxation and a home exercise programme (HEP) [26]. Schachter et al. [27] compared three groups: one group performing twice daily, short bouts of aerobic exercise, one group performing long bouts of aerobic exercise once daily, and an attention-control group. The median study sample size was 163 (IQR 119–194). A total of 1416 participants

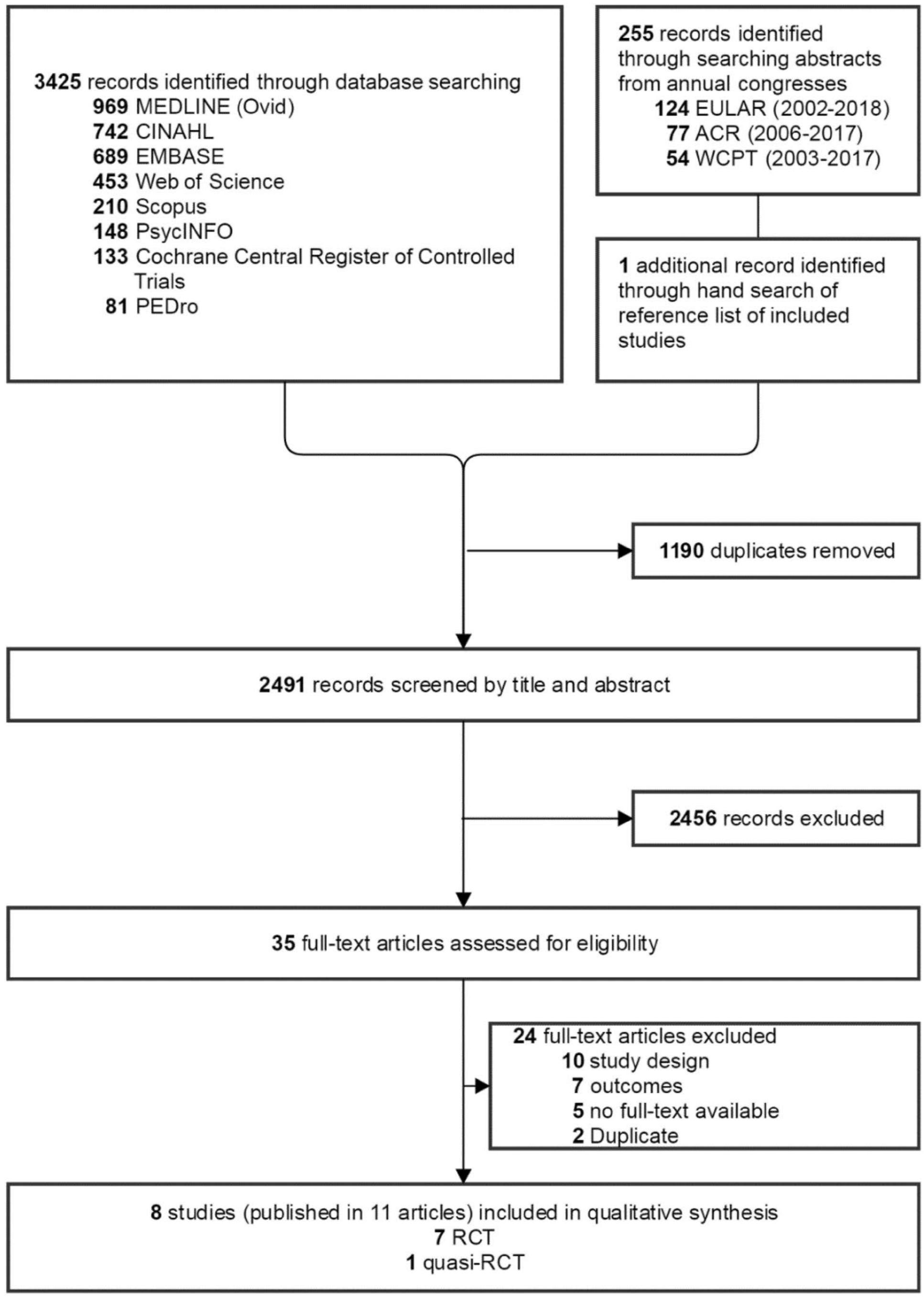

Fig. 1 Preferred reporting items for systematic reviews and meta-analyses flow diagram of search strategy and study selection process. *RCT* randomised controlled trial

with fibromyalgia were included. Participant characteristics are summarised in Table 1.

Study characteristics: interventions

Study intervention durations ranged from 6 weeks [28] to 16 weeks [27], with seven studies including follow-up periods ranging from 3 months [26, 30] to ~12 months [28, 29, 31, 33]. PA and exercise behaviours were the primary focus of four interventions [27, 29, 31, 33], and were components of multiple health behaviour targets in four studies [26, 28, 30, 32]. Four studies included practical exercise sessions as part of their behaviour change intervention [26, 31–33].

Interventions were primarily delivered in group settings ($n=6$), online [28], or by phone [27]. A variety of health-professionals delivered, moderated, or facilitated. Group sessions were supplemented with DVDs or printed materials for home use; in one study participants were provided with pedometers [29]. Half of the interventions ($n=4$) used email or telephone follow-up. All interventions included

Table 1 Description of study design, participant characteristics, and outline of intervention

Study	Design, country	Participants information Age, years Symptom duration, years Sex (% female) FM activity, FIQ score	Duration of intervention; follow-up	Intervention description	Group (*n*)	Retention at end of study, % (*N*)
Ang et al. [31]	RCT Indiana, USA	Age 45.8 (11.2) Symptoms 9.0 (7.0) 96% female FIQ 67.0 (12.8)	12 weeks F/up 3 months + 6 months	Motivational interviewing: Aerobic exercise prescription and two individualised supervised exercise sessions from a qualified instructor + 6 sessions of telephone-delivered MI to encourage the adoption and/or maintenance of exercise sessions. Focus on enhancing patient motivation to exercise, strategies to strengthen commitment to exercise (develop plan for change and review positive consequences of exercise), and follow-through strategies to prevent relapse of inactivity	107	93.5% (100)
				Education control: Aerobic exercise as a/a + 6 sessions of telephone-delivered education sessions delivering didactic health information: overview of FM; pain; fatigue; sleep; stress; living well with FM	109	93.6% (102)
Fontaine et al. [29]	RCT Maryland, USA	Age 47.6 (11.0) Time since Dx 7.6 (6.2) 97% female FIQ 68.5 (12.6)	12 weeks F/up 6 months + 12 months	Lifestyle Physical Activity (LPA): 6 × 60-min group sessions over 12 weeks. Protocol addressed FM-specific challenges to becoming more physically active. At the first session, participants were taught how to perform LPA at moderate intensity. They were prescribed 15 min, above usual level, of accumulated moderate-intensity LPA 5–7 days a week, and asked to increase the daily duration of LPA by 5 min each week. Subsequent sessions: self-monitoring, goal setting, dealing with symptom flares, problem solving strategies to overcome barriers to being more physically active, and instruction in finding new ways to integrate short bouts of LPA into their daily lives	46	65.2% (30)
				Fibromyalgia Education: 3 × 90–120 min group sessions, conducted by experienced FM support group facilitator, monthly meeting for 3 months. This minimal intervention was divided into three components: education, questions and answers, social support. Topics included symptoms, diagnosis, and treatment of FM	38	60.5% (23)

Table 1 (continued)

Study	Design, country	Participants information Age, years Symptom duration, years Sex (% female) FM activity, FIQ score	Duration of intervention; follow-up	Intervention description	Group (*n*)	Retention at end of study, % (*N*)
Hammond et al. [32]	RCT Derbyshire, UK	Age: 48.5 (10.9) Symptoms 6.5 (4.9) 90.2% female FIQ 54.9 (12.6)	10 weeks F/up 4 months + 8 months	Cognitive-behavioural patient education–exercise programme. 10 week, 1 × weekly 2-h classes. Education included: theoretical causes of FM, physiological basis of symptoms, the cycle of FM symptoms perpetuation, and how self-management approaches can improve symptoms. Exercise included: postural training, stretching, strengthening exercises using light weights, Tai Chi for Arthritis programme. Other interventions included: activity pacing, sleep hygiene, relaxation, problem solving, pain, fatigue and stress management	71	73.2% (52)
				Relaxation programme. 10 weeks, 1 × weekly 1-h classes. FM information booklet	62	79.0% (49)
Keel et al. [26]	qRCT Basel, Switzerland	Age 49 (NR) Symptoms 13.4 (NR) 88.9% female FIQ NR	15 weeks F/up 3 months	IG: 15 × once weekly 2-h sessions discussing information, instruction in self-control strategies, exercise, relaxation and group discussion. Pain control strategies, stress inoculation and cognitive restructuring techniques taught. A home exercise plan was developed to include stretching and aerobic exercises (details not available) + Autogenic Training: daily, home relaxation and exercise, with application of pain control techniques. Handouts of home exercise programme were provided	16	87.7% (14)
				CG: 15 × once weekly 45–60 min supervised relaxation + home relaxation in the same manner as the IG	16	81.3% (13)
Larsson et al. [33]	RCT Gothenburg, Stockholm, and Linköping, Sweden	Age: 51.4 (9.4) Symptoms 10.3 (8.0) 100% female FIQ 60.8 (15.8)	15 weeks F/up 13–18 months	Resistance exercise: 15 weeks, 2 × weekly, supervised group exercise. Introductory meeting to discuss past experience with exercise, attitudes towards exercise, potential barriers to exercise, exercise instruction, individualisation of exercise loads leading to a written protocol to be used as the exercise programme. Exercises were progressed every 3–4 weeks if appropriate	67	71.6% (48)
				Active control group: the relaxation therapy consisted of 5–8 participants. Individual introductory meeting for individualisation of mattress and pillows. Relaxation therapy included mental exercise, and was followed by stretching exercises	63	68.3% (43)

Table 1 (continued)

Study	Design, country	Participants information Age, years Symptom duration, years Sex (% female) FM activity, FIQ score	Duration of intervention; follow-up	Intervention description	Group (n)	Retention at end of study, % (N)
Lorig et al. [28]	RCT California, USA	Pooled participant information reported for RA, OA and FM. Subgroup details not available	6 weeks F/up 6 months + 1 year	Internet-based Arthritis Self-Management Programme: Web-based instruction, discussion board, and tools (exercise logs, medication diaries, and tailored exercise programmes); Arthritis Helpbook containing all of the programme content. Participants logged on at least 3 times a week for 1–2 h at a time to participate in weekly activities, tasks and self-tests + Email reminders to participate and tailored exercises suggested via automated algorithms + Trained peer moderators led workshops, assisting participants with the programme by reminding them to log on, modelling action planning and problem solving, offering encouragement and posting to the bulletin boards	40	NR
				CG: usual care	46	NR
Schachter et al. [27]	RCT Saskatchewan, Canada	Age 42.5 (7.6) Symptom 8.7 (5.3) 100% female FIQ 5.6 (1.5)	16 weeks	Long-bout exercise group: low-impact aerobics programme: 1 × daily, 20 min at 40–50%HRR increasing to 40 min by week 9 and 65–75% HRR by week 12. With supporting videotape, accompanying instruction booklet, daily exercise and symptom log book + monthly small group meetings on the topics of: familiarisation with exercise programme and self-monitoring exercise intensity; problem solving difficulties with exercise programme; ways to enhance post-study exercise level and adherence + monthly supportive telephone from group leader	51	70.6% (36)
				Short-bout exercise group: low-impact aerobics programme: 2 x daily, 15 min at 40–50%HRR increasing to 25 min by week 9 and 65–75% HRR by week 12. Group meetings and supportive telephone calls as in Long-bout exercise group	56	62.5% (35)
				CG: maintained sedentary lifestyle + monthly small group meetings to discuss experiences with FM, without additional education from researchers + monthly telephone from group leader to inquire as to status	36	86.1% (31)

Table 1 (continued)

Study	Design, country	Participants information Age, years Symptom duration, years Sex (% female) FM activity, FIQ score	Duration of intervention; follow-up	Intervention description	Group (*n*)	Retention at end of study, % (*N*)
Stuifbergen et al. [30]	RCT Texas, USA	Age 53.1 (9.9) Time since Dx 9.0 (5.7) 100% female FIQ 61.6 (17.4)	8 weeks group + 3 months phone support F/up 3 months	Lifestyle Counts intervention to promote the health of women with FM: 8 weeks, 1 × weekly 2 h group sessions + Bimonthly telephone conversations using motivational interviewing techniques to encourage progress towards goals and problem solve barriers	98	85.7% (84)
				Attention-control. 8 × group education on topics related to disease management + follow-up phone conversations scripted to answer any participant questions on class content	89	91.0% (81)

CG control group, *Dx* diagnosis, *F/up* follow-up, *FIQ* Fibromyalgia Impact Questionnaire, *FM* fibromyalgia, *HRR* heart rate reserve, *IG* intervention group, *min* minutes, *NR* not reported, *OA* osteoarthritis, *RA* rheumatoid arthritis, *RCT* randomized controlled trial

discussion or advice on such topics as problem solving, or integrating PA into daily life. Education was included in the majority of programmes ($n=5$), addressing topics such as the physiological basis of fibromyalgia symptoms, or providing an overview of medication (see Tables 1 and 2 for descriptions of interventions).

Study characteristics: behaviour change

The study by Stuifbergen et al. [30] reported integrating concepts from the eHealth belief model, Pender's model of health promotion and self-efficacy theory. Ang et al. [31] employed a method-based intervention focused on Motivational Interviewing, and Fontaine et al. [29] based their intervention on a cognitive-behavioural PA promotion programme, Active Living Every Day. The patient education programme in the study by Hammond et al. [32] was based on social cognitive theory and the self-management cognitive-behavioural therapy approach. The other studies did not report the intervention being underpinned by a behaviour change theory.

The median number of BCTs included was 13 (IQR 11–17). In total, 32 different BCTs were identified within the eight studies. All studies ($n=8$) incorporated 'instruction on how to perform the behaviour', 'demonstration of the behaviour', 'behavioural practice/rehearsal', and 'credible source'. Of the 16 hierarchies in the BCTTv1, only 'Scheduled consequences' was not identified.

Risk of bias within studies

The risk of bias of included studies is summarised by for all studies in Fig. 2. Frequently assessments included an 'unclear' judgement; this often reflected a lack of information upon which to judge the item and could not be clarified by attempts to contact the authors.

Studies that were at 'low risk' of selection bias from sequence generation reported randomisation either by a computer-generated randomisation list or similar method [27, 29–33]; however, only two studies were also judged to be at low risk of selection bias from allocation concealment [32, 33]. Keel et al. [26] employed sequential allocation upon presenting to the study and were judged as being as 'high risk' of selection bias.

Five studies were judged as having a 'high' risk of performance bias, primarily due to the fact that it was not possible to blind participants because of the nature of the intervention [26, 28–30, 33]. Four studies were judged as 'low risk' of detection bias as outcome assessment was performed by a researcher blinded to group allocation [27, 28, 30, 33]. One study was judged as 'high risk' of detection bias as outcome assessment was performed by personnel who were not blinded to group allocation [26].

Table 2 Characteristics of behaviour change interventions

Study	Target behaviour		Delivery			Intervention content			Support			
	Multiple	PA	Group (facilitator)	1:1	Online	Exercise class or practical	HEP	Education about condition	Tel	Email	A/V or printed materials	Pedometer
Ang et al. [31]		√[a]		√(FI)		√	√		√(MI)			
Fontaine et al. [29]		√[a]	√				√	√				√
Hammond et al. [32]	√[a]		√(PT/OT)			√	√	√			√	
Keel et al. [26]	√		√(PT/Psy/Psych)			√	√	√			√	
Larsson et al. [33]		√	√(PT)			√						
Lorig et al. [28]	√				√		√	√		√	√	
Schachter et al. [27]		√	√(PT)				√		√		√	
Stuifbergen et al. [30]	√[a]		√(CNS)					√	√(MI)		√	

A/V audio visual, *BC* behaviour change, *CNS* clinical nurse specialist, *FI* fitness instructor, *Ex* exercise, *HEP* home exercise programme, *MI* motivational interviewing, *PA* physical activity, *PT* physiotherapist, *Psy* psychiatrist, *Psych* psychologist

[a]Based on behaviour change theory

Six studies were judged as 'low risk' of attrition bias [27, 28, 30–33]. Two studies were judged as 'high risk' of attrition bias due to incomplete reporting with loss to follow-up or inconsistencies across timepoints [26, 29]. Four studies published an a priori study protocol and were judged as 'low risk' of reporting bias [28, 29, 31, 33].

Physical activity outcomes

The PA outcomes are synthesised here, focusing on three comparisons. A meta-analysis was not undertaken due to the heterogeneity of study interventions and reported outcome measures.

Comparison 1: behaviour change interventions compared to usual care or education

Four studies compared behaviour change interventions to usual care or education, with two studies objectively measuring PA. Fontaine et al. [29], using pedometry to measure daily step count, reported that following a 12-week intervention, PA was increased significantly more in the behaviour change group than in the control group [MD (95% CI) 1896 (404, 3388) daily steps]; this was not maintained at 6- and 12-month follow-up. In contrast, Ang et al. [31] found that changes in time spent in MVPA were not significantly different between groups immediately after the 12-week intervention phase, or at 3- or 6-month follow-up, measured by accelerometers (GT1M ActiGraph) worn for 7 days. However, the authors also measured PA subjectively, using the CHAMPS measure, and found that immediately following the intervention phase, the behaviour change group significantly increased MVPA by 1.5 h/week (0.6) more than the education group; as with the objective findings, these short-term differences were not retained at 3- or 6-month follow-up. Stuifbergen et al. [30] reported that from baseline to 8-month follow-up, change scores on the PA subscale of the Health Promoting Lifestyle Profile II were significantly different, favouring the behaviour change intervention. This contrasts with the findings from Lorig et al. [28], who reported no significant differences in weekly time spent performing aerobic exercise, or stretching and strength exercise, at 1-year follow-up.

Comparison 2: combined behaviour change and exercise compared to relaxation with or without home exercise

Three studies compared a combination of behaviour change intervention and exercise to relaxation with or without a HEP. In each study, PA was measured using subjective outcome measures. Hammond et al. [32] noted that at 8-month follow-up, the intervention group reported increased time

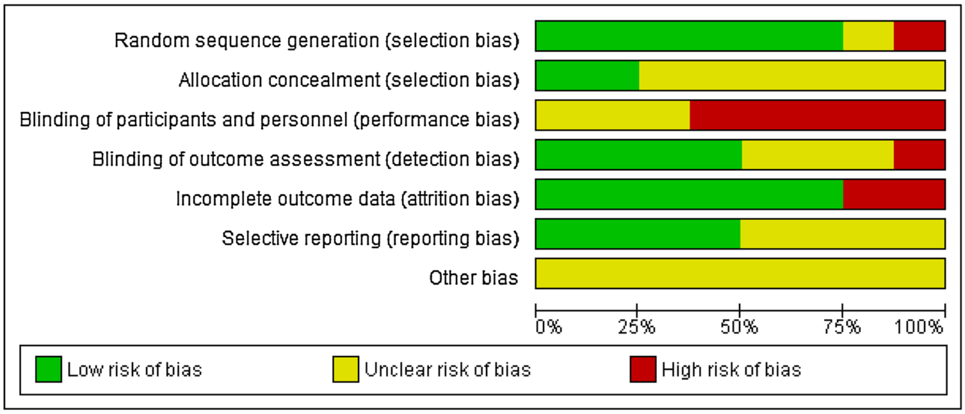

Fig. 2 Risk of bias of included studies

spent performing stretching/strengthening exercise [mean (SD) of 20.3 (48.7) min/week] compared to the control group [5.7 (34.8) min/week]. This difference was not observed at 4-month follow-up. No significant difference in weekly aerobic exercise was found at 4- or 8-month follow-up. Larsson et al. [33] recorded PA using the Leisure Time Physical Activity Instrument. During the intervention period, the level of PA increased significantly ($P<0.001$) in the resistance exercise group [2.3 (4.8) h] compared to the active control group [−0.1 (4.8) h]. MVPA increased significantly more in the resistance group [1.8 (3) h] compared to the active control group [0.4 (2.6) h] ($P=0.003$). These differences were not sustained at follow-up. In the study by Keel et al. [26] 'Active time', self-reported in a diary, was not significantly different between groups post-intervention, or at 3-month follow-up.

Comparison 3: two behaviour change and exercise interventions

The study by Schachter et al. [27] compared a behaviour change intervention combined with either a long-bout exercise programme or a short-bout exercise regime. For each, a 'duration index' was calculated by adding the minutes of exercise performed (according to exercise log) and dividing by the minimum number of minutes of exercise recommended. No significant difference was observed post-intervention (a third arm in this study, an attention-control group, was not part of this analysis).

Secondary outcomes

Adverse events

One participant reported a metatarsal fracture [27]. Increased pain, stiffness or fatigue were reported by a small number of participants across three studies as reasons for dropping out of studies, or for not participating in sessions ($n=16$ [32, 33], $n=$ not reported [27]).

Physical fitness

The intervention by Larsson et al. [33], which combined behaviour change and individual exercise programmes, resulted in improvements in isometric knee extension, elbow flexion and grip strength, as well as increased distance covered in a 6 min walk test, compared to relaxation therapy after 15 weeks. Schachter et al. [27] did not report significant differences in cardiorespiratory capacity after 16 weeks of aerobic exercise compared to an attention-control group. Results from studies comparing usual care or education to PA behaviour change interventions without exercise components were conflicting; Fontaine et al. [29] reported no significant between-group differences in BMI or 6MWT distance in the short- or long-term. Ang et al. [31] reported superior change scores in 6MWT distance in the intervention group compared to the control group.

Condition-related outcomes

The effects of behaviour change interventions on the impact of fibromyalgia [27, 29–33] and symptoms of fatigue [28] were not significantly different to control groups. Pain significantly improved in the intervention groups in the short term on VAS [29, 31, 33] but not on the Pain Disability Index [33]; no significant long-term differences were reported [28, 29, 31]. Pain self-efficacy improved significantly more following behaviour change interventions compared to controls in the short-term [27, 32].

Quality of life

Combined behaviour change and supervised exercise programmes resulted in short-term improvements in the physical component of the SF-36 compared to relaxation therapy [33]; these improvements were not sustained a follow-up. No significant group differences were reported in the mental component.

Secondary outcomes are summarised in Supplementary File 2.

Discussion

This review identified a small number of studies targeting PA behaviour in adults with fibromyalgia, with inconsistent findings reported. A number of interventions have demonstrated short-term improvements in PA compared to education or relaxation, without these improvements being maintained at follow-up [29, 31, 33]. Other studies did not note improvements at the end of the intervention phase, but reported increases in PA behaviour at follow-up [30, 32]. Finally, some studies did not report any significant effects on PA behaviour post-intervention or at follow-up [26, 28]. Such inconsistencies in findings may, in part, be attributed to differing behaviour change theories and techniques implemented across interventions, the mode of implementing the intervention, the variety of PA outcome measures used, and the varying risk of bias across the studies.

Applying theory to the design and evaluation of complex behaviour change interventions is viewed as good practice; however, to date, this has not been widely implemented [38]. The relationship between type and extent of theory used and the effectiveness is inconsistent [39]. In this review, four of the five studies that reported a positive change in PA behaviour were based on a behaviour change theory; the studies that did not report PA increases did not report incorporating theory into intervention design. A similar lack of theory driven interventions has been reported in rheumatoid arthritis [40], and osteoarthritis and chronic low back [41].

Half of the included studies implemented behaviour change interventions targeting PA, while the other studies examined interventions targeting multiple health behaviours. These two approaches differ in the number and types of BCTs typically used [42], and the best approach is unclear. Some studies have concluded that single health behaviour interventions are more effective at increasing PA [43, 44], while others report increased efficacy of interventions recommending multiple behaviour changes relative to those recommending single behaviour changes [45, 46].

The studies included in this review primarily used a group-based approach for intervention delivery. It is notable that eHealth has scarcely been explored in this cohort, with only the study by Lorig et al. [28] using a digital intervention. Interventions using smartphone applications to improve PA have been effective in the general population, and appear most effective as part of a multi-component programmes [47]. Connected health solutions represent an underexplored opportunity for delivering PA behaviour change programme to individuals with fibromyalgia.

Overall, the reporting of BCTs in study methods was poor, with insufficient details and description provided to allow replication of the intervention. In many cases, the rationale for including BCTs was not clear. To our knowledge, this is the first review of BCTs targeting PA in fibromyalgia. The most commonly used BCTs included instruction and demonstration of the behaviour, behavioural practice, goal setting, problem solving, and action planning. The programmes were delivered by ‘credible sources’. The studies which successfully improved PA used a variety of BCTs, which were similar to the studies which were ineffective at increasing PA behaviour. The most effective BCTs and the optimal combinations of BCTs remain unclear. Behaviour change interventions targeting PA behaviour in individuals with rheumatoid arthritis have had mixed success, with a similar heterogeneity in interventions [40].

Depending on the phase of the behaviour change process, the determinants of behaviour may vary. This may explain why some studies in this review demonstrated short-term effects without sustained change at follow-up, and why others showed no short-term benefits but at follow-up reported improvements. In a recent review of maintenance-relevant behaviour change theories, Kwasnicka et al. [48] reported five emergent themes: maintenance motives, self-regulation, resources, habits, and environmental and social influences. The BCTs that help people initiate change may have different effects on behaviour maintenance.

In evaluating the effects of the interventions on PA, the majority of studies used self-reported measures of PA. Such measures have known limitations. Self-reported PA measures can both under- and over-estimate PA compared with objective measures such accelerometry, with no clear trends in the degree to which PA estimates diverge across measures [49]. This is exemplified in the study by Ang et al. [31]; both accelerometry and the CHAMPS questionnaire were used to measure PA, and produced conflicting results. The consensus is that objective methods of measuring free-living PA are recommended for research trials; this is not currently reflected in the current fibromyalgia literature.

In a small number of cases, worsening symptoms or musculoskeletal injury were cited as reasons for dropping out of a study; participant retention rates in the intervention groups ranged from 65 to 94%. Overall, studies reported short-term improvements in pain, with no adverse effects on outcomes of fatigue. Therefore, while individuals may experience increased fatigue or pain, these symptomatic changes appear largely transient.

Limitations and future research

Due to the heterogeneity of interventions and outcomes, pooling of PA data was not deemed appropriate. A single author, rather than the suggested two [23], performed the data extraction task. Mapping of study BCTs to the BCTTv1 was based on the published materials and, when provided, on materials obtained from the study authors; due to sparsity of available data, there may have been minor misclassifications.

Future PA behaviour change interventions should seek to apply behaviour change theories to the design, implementation, and evaluation process of the intervention; this should be explicitly reported. Consideration should be given to employing different theories at different stages of the behaviour change journey. In exploring the effects of PA intervention, sufficient detail of the specific BCTs used, and the modes of delivering the intervention, should be reporting using standardised taxonomy. The question of whether PA should be the sole health behaviour target, or incorporated into a larger health behaviour interventions (e.g., with weight management, smoking cessation) is unanswered.

Conclusion

Despite numerous general health and condition-specific benefits, PA and exercise promotion is a significant clinical challenge in adults with fibromyalgia. A limited number of behaviour change interventions targeting PA have been examined, with mixed success. To determine the optimal approach to PA behaviour promotion, future trials should aim to integrate behaviour change theory into the design, implementation, and evaluation of interventions. Specified BCTs should be detailed to allow programmes to be evaluated and replicated.

Author contributions All authors were involved in the design of the study. TOD, DM, and FW conducted the literature search. SM, FW, and TOD conducted the risk of bias appraisal. TOD extracted the data. TOD, SM, LD, and FW contributed to data analysis and interpretation. All authors reviewed and approved the final version of the manuscript.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflicts of interest.

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