**Family-based interventions benefit individuals with musculoskeletal pain in the short-term but not in the long term: A systematic review and meta-analysis**

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**Abstract**

**Introduction:** The benefits of family-based interventions for patients with musculoskeletal pain have been previously shown in individual randomised controlled trials (RCTs), but no systematic review has summarised their effects.

**Materials and methods:** A systematic review was conducted to assess the effectiveness of family-based interventions on clinical and biopsychosocial outcomes in people with musculoskeletal pain (PROSPERO CRD42018118442). Meta-analyses were performed for the outcomes of pain intensity, disability, mood, self-efficacy, and marital adjustment.

**Results:** Of 1,223 records identified, 18 reports representing 15 RCTs were included in the qualitative review and 10 in the meta-analyses. Family-based interventions were more effective to reduce pain [Mean difference (MD): -3.55/100; 95%CI -4.03, -3.06], and disability (MD: -1.51/100; 95%CI -1.98, -1.05) than individual-focused interventions at short-term, but not at mid or long-term. There were no effects on other outcomes. Family-based interventions were more effective to reduce pain (MD: -6.05/100; 95%CI -6.78, -5.33) compared to usual care only at short-term. No effects were found on disability and other outcomes.

**Discussion:** There is moderate-quality evidence that family-based interventions result in small, significantly better pain and disability outcomes in the short-term compared to individual-focused interventions in patients with musculoskeletal pain. Based on low-quality evidence, family-based interventions result in small improvements on pain in the short-term compared to usual care. Future studies should review the content and optimise the mechanisms underpinning family-based interventions in musculoskeletal pain so that the approach could be further tested in adequately powered RCTs.

**Keywords:** musculoskeletal pain, family, systematic review

**Introduction**

Musculoskeletal pain conditions are best framed within the biopsychosocial model, which considers the disease and its complex interaction between biological, psychological and societal factors [1-3]. As an example, pain reported by patients with symptomatic osteoarthritis is understood to be an interaction between structural pathology (such as effusions and bone marrow lesions) and its impacts on the peripheral nervous system, spinal cord pain signaling, and cortical processing; and psychosocial factors, such as psychosocial comorbidities, socioeconomic status, and coping behaviour [1]. The familial environment is embedded within the societal factors of the biopsychosocial model and can have a significant influence on health and behaviour [4-6].

Research has shown an increasing interest in the influence of family members (e.g. partners, parents) on healthy lifestyle behaviours, particularly in the management of long-term conditions [7-9]. Studies show that family members can influence the health and behaviour of other family members, both positively and negatively [10-12]. While providing support and autonomy appears to have positive effects, being overprotective, controlling, or providing a hostile unhealthy environment can impact negatively on health behaviours and outcomes [10-12].

The overall evidence assessing the effects of including family members in the management of long-term conditions is growing. For example, involving family members in educational programs for poorly controlled diabetes has been shown to lead to better control of glucose markers (HbA1c) in comparison to usual care [13]. Positive effects of involving family members have also been demonstrated on caloric and fat intake in people with high blood lipoprotein density [14]. Furthermore, greater engagement in physical activity has been observed in patients with coronary artery disease [15] when participating in family-based programs compared to individual-focused interventions alone.

Musculoskeletal pain conditions such as low back pain and knee osteoarthritis are among the most common long-term conditions [16]. They are among the top causes of days lived with disability globally [16] and increasing burden to healthcare systems [17]. Clinical management commonly includes interventions that attempt to change the patient’s lifestyle (e.g. through exercise, physical activity and psychological approaches) [1, 18] and addresses common co-morbidities (e.g. obesity) [1]. There have been several previous studies investigating the benefit of family-based (i.e. involving partners or other family members) compared to individual-focused interventions, for patients with musculoskeletal pain. Results of individual studies have shown greater improvements in measures of partner support [19], marital function [20], communication skills [19], and fear of movement (kinesiophobia) [21] in patients with chronic low back pain [20, 21] or osteoarthritis [19]. Previous systematic reviews have also assessed the influence of including partners in the management of chronic physical illnesses (e.g. cancer, diabetes, cardiovascular diseases, musculoskeletal, and chronic pain) and have reported positive effects on pain, pain behaviour and catastrophising, self-efficacy, illness appraisal, spouse relationship and quality of life [22, 23]. The vast majority, however, were conducted on patients with cancer and other chronic diseases, such as cardiovascular diseases, diabetes and HIV [22, 23] and none has provided specific results for patients with musculoskeletal pain. This is a clear omission within the literature, especially as there is now a growing body of evidence in this field [24-26]. Therefore, the aim of this systematic review was to investigate the effect of family-based interventions on pain intensity and biopsychosocial outcomes compared to individual-focused interventions for patients with musculoskeletal pain.

**Materials and Methods**

**Search Strategy**

The review protocol was registered on the International Prospective Register of Systematic Reviews (CRD42018118442) and followed the PRISMA 2009 statement [27]. The following databases were searched from their respective inception dates to January 2020: MEDLINE (via Ovid), AMED (via OVID), EMBASE (via OVID), PsycINFO (via OVID), CENTRAL (via OVID), CINAHL (via EBSCO), Web of Science and PEDro. Keywords (including exploded terms and MESH terms) concerning musculoskeletal pain (and specific disease terms – e.g. low back pain) AND family therapy (and specific terms – e.g. mother, father, partner) AND randomised controlled trial were combined in the search strategy (Appendix 1).

**Inclusion and exclusion criteria**

Randomised controlled trials (RCTs) published in peer-reviewed journals which investigated the effectiveness of family-based interventions as treatment for musculoskeletal pain, such as low back pain, neck pain, hip pain, shoulder pain, knee pain, osteoarthritis (knee, hand or hip), fibromyalgia, or rheumatoid arthritis, compared with individual-focused interventions or usual care were included. The search was not restricted by age, gender, race, ethnicity, language, or year of publication.

**Population**

We included RCTs concerning family-based interventions in adults (>18 years of age), adolescents, or children with musculoskeletal pain. Studies of patients with general bodily chronic pain (including patients with musculoskeletal pain, headache or abdominal pain) were only included if more than 50% of the sample reported musculoskeletal pain. Studies focusing on individuals with other types of pain conditions (i.e. cancer, neurologic diseases or pain arising from nerve root compromise) were excluded.

**Family-based and individual-focused interventions**

All types of interventions with active participation of patients and family members [(i.e. spouse/partners, parents, siblings, offspring, or others (e.g. close friend, carer/caregiver)] were included in this review. Family-based interventions could be compared to usual care, waitlist control or individual-focused interventions. Two main analyses were performed to compare the effects of family-based interventions to individual-focused interventions (i.e. interventions that were similar to the family-based interventions but without the involvement of a family member) and to compare the effects of family-based interventions to usual care.

**Outcomes**

The primary outcome was pain intensity, which could be assessed by, but not limited to, Visual Analogue Scale, Numerical Rating Scale, McGill Pain Questionnaire, Western Ontario and McMaster Universities Arthritis Index. Secondary outcomes were other biopsychosocial outcomes, including disability, health-related quality of life, relationship with partners/family members, mood (i.e. anxiety and depression), pain coping, self-efficacy, and pain catastrophising.

**Study inclusion and data extraction**

Titles/abstracts and full text were screened by two independent reviewers (AKFS and CGF). Any disagreements over study eligibility were resolved through discussion with a third reviewer (PHF). In addition, all references from the selected RCTs were reviewed to supplement the search. Data from included RCTs were extracted by two independent reviewers (AKFS and CGF) and checked for potential errors. The following data were extracted: type of musculoskeletal pain condition, pain/disease duration, sample size, mean age (±SD) of overall and group sample, demographic characteristics, intervention type (including frequency, number, duration, mode and settings), family members and health professionals involved, time-points of follow-up, outcome measures at each time-point, and main RCT findings. We contacted five correspondent authors once via email and requested missing data or information about the protocol design, with one author replying.

We extracted data on the mean (±SD) of pain, disability, health-related quality of life, relationship with partners/family members, psychological disability (i.e. depression and anxiety), depressive symptoms, pain coping, self-efficacy, and pain catastrophising on post-intervention and follow-up time-points for the quantitative analyses. We classified the follow-up period according to time post-intervention, as short-term (10 weeks or less), mid-term (greater than 10 weeks and up to 6 months), and long-term follow-up (greater than 6 months and up to 12 months) for the meta-analyses, independently of the duration of the intervention, and in agreement with the Cochrane Back and Neck Group recommendations [28].

**Assessment of methodological quality and strength of the evidence**

The PEDro (Physiotherapy Evidence Database) scale [29] was used to evaluate RCT quality. RCTs were classified as being of excellent (9-10), good (6-8), fair (4-5), and poor (<4) methodological quality.

In addition, the overall quality of the evidence for each outcome was assessed using the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) [30]. We downgraded the evidence by one level for each of three domains considering: a) risk of bias (25% of patients from studies with PEDro score <6 points); b) inconsistency of results (I2>50 %); and c) imprecision (<400 patients in total for each outcome). Indirectness was not considered for this review due to the study focus on a specific population, comparator, and outcome measures. Inconsistency assesses the extent of heterogeneity of the studies’ results included in the meta-analyses and indicates poor overall evidence quality when there is uncertainty of where the variability in the results is coming from [31]. Imprecision refers to the uncertainty in the estimates of the treatment effect [32]. The quality of evidence was considered as high quality (i.e. further research is unlikely to change the confidence in the estimate of effect); moderate quality (i.e. further research is likely to have an important impact on confidence in the estimate of effect and might change the estimate); low quality (i.e. further research is likely to have an important impact on the confidence in the estimate of effect and is likely to change the estimate; and very low quality (i.e. any estimate of effect is uncertain) [33]. Assessments were performed by two independent reviewers (AKFS and CGF) and any disagreements were resolved through discussion with a third reviewer (PHF).

**Data synthesis and analysis**

A narrative synthesis of the findings was conducted regarding included RCTs’ populations, interventions, and outcomes. We performed meta-analyses of primary and secondary outcomes when sufficient data were available (i.e., at least two RCTs with data on a similar outcome and follow-up time-point). We performed a random-effects meta-analysis with mean differences for continues variables (e.g. pain) and calculated 95% confidence intervals and two-sided P values for each outcome for two comparisons: family-based interventions compared to individual-focused interventions, and family-based interventions compared to usual care. The Chi-squared test and the I-squared statistic were used to assess heterogeneity (with heterogeneity classified as I2>50%). Secondary analyses per musculoskeletal pain condition were performed according to data availability, and follow-up time-points (i.e., short, mid, and long-term). However, we could only perform separate, secondary analyses for osteoarthritis and low back pain, as these were the only conditions included in multiple trials. When RCTs included three or more comparator groups, we considered the interventions that were more similar within the studies’ groups, with the main difference being the inclusion or not of a family member. Thus, if a trial included two groups consisting of different family-based interventions, we selected the one that more closely resembled the individual-focused intervention. For example, if the study included two family-based interventions (group 1 included cognitive behaviour training whilst group 2 was educational only) and one individual-focused intervention (cognitive behaviour training without the involvement of the spouse), data from group 1 were included in the meta-analyses as the intervention was considered more similar to the individual-focused group. Scores from different outcome measures were converted to a common scale of 0-100. All analyses were performed with Review Manager version 5.3.5.

**Results**

The search strategy identified 1,634 articles (Figure 1). After removing duplicates, screening titles and abstracts, 33 full-text articles were assessed. A total of 18 articles representing 15 RCTs were included in the qualitative analysis [19-21, 24-26, 34-45], and 10 RCTs were eligible for combining in the meta-analyses [20, 21, 24, 35, 36, 38-40, 42-45]. Only one RCT recruiting children with musculoskeletal pain met our inclusion criteria [26] and was included in the review to comply with our registered protocol. The RCT was not included in the meta-analysis to reduce heterogeneity in the meta-analytical approach. In addition, four RCTs did not provide enough data [19, 25, 37, 41] to be included in the meta-analysis.

RCTs included patients with osteoarthritis (n=6) [19, 25, 34-36, 38, 39], chronic low back pain (n=5) [20, 21, 37, 43-45], chronic pain (>50% musculoskeletal pain) (n=2) [24, 40], rheumatoid arthritis (n=2) [41, 42], and juvenile idiopathic arthritis (n=1) [26]. The mean symptom duration of the overall sample ranged from 1.6 [20, 43, 44] to 18.0 years [38], and the follow-up duration in the studies ranged from 3 weeks [24] to 60 months [20, 43, 44]. Trials were conducted in the United States (n=9) [19, 25, 34-36, 38-41, 45], the Netherlands (n=2) [37, 42], Finland (n=1) [20, 43, 44], Iran (n=1) [21], Australia (n=1) [24], and Denmark (n=1) [26]. The overall characteristics of RCTs and follow-up time-points are summarised in Table 1 while intervention characteristics are summarised in Table 2.

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**Characteristics of family-based interventions**

The spouse was the most common family member included in intervention delivery in the included RCTs (13 of 15; 86.6%) [19-21, 24, 34-45], and the majority of the interventions included education models about the musculoskeletal pain condition for patients or patients and family members (e.g., how to best cope with pain and arthritis), and/or cognitive behaviour therapy (86.6%) [19, 21, 24, 26, 34-42, 45]. Four RCTs (26.7%) also included the provision of printed educational material [40, 42, 45] or handouts for exercise [25]. Four RCTs included exercise-based therapy as part of an intervention package [24, 36, 42, 45], while only one delivered an exercise-only intervention [25]. Some interventions also included health-related goals for the couple (n=4) [19, 21, 39, 40, 46] or joint home exercise practice (n=2) [26, 34]. Six RCTs (40%) also reported providing some training on relaxation techniques [21, 24, 34-36, 40, 41].

Family-based interventions were delivered by psychologists [20, 41, 43, 44], physiotherapists [24], nurses [24, 42], arthritis self-care course specialist [19, 38, 39] or yoga instructor [25], or by a multidisciplinary team [21, 24, 34-37, 40, 45]. Most of the RCTs (n=9) delivered educational sessions associated with self-management, coping skills training and/or setting health-related goals for the patients and their family members in group sessions with three to 10 patients and their relatives [19, 21, 26, 36, 39-42, 45]. The majority of the programs were delivered face-to-face in healthcare clinics, rehabilitation centres or hospitals, while some were delivered via telephone. Treatments ranged from four to 21 sessions delivered on a weekly (77.8%) [21, 25, 26, 34-38, 40-42, 45], monthly (16.6%) [20, 43, 44], or a mix of daily and weekly [24], or weekly and monthly [19, 39] basis. The duration of each intervention session varied from 20 minutes [38, 39] to 8 hours in an inpatient program [24].

**Risk of bias and quality of evidence**

The mean score on the PEDro quality scale for included RCTs was 4.7 out of 10 points (Table 3). Only two RCTs were of good methodological quality (i.e. 6-8/10 points). None of the included RCTs blinded participants (although this would be impossible given the nature of the interventions), and none clearly reported concealed allocation. Furthermore, only one RCT reported blinding of therapists [41], and three reported blinding of assessors [20, 37, 43-45]. The overall quality of evidence for musculoskeletal pain as the primary outcome was considered moderate for the short-term follow-up (10 weeks or less), and low for mid and long term follow-up periods assessed with GRADE scale [30] (Supplementary Table 1) when family-based were compared to individual-focused interventions. The overall quality of evidence for pain was considered low for the short-term follow-up and very-low for the mid-term follow-up when family-based interventions were compared to usual care (Supplementary Table 4).

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**Synthesis of results**

Ten RCTs were included in the meta-analyses. Data reported on the primary and secondary outcomes of the systematic review by RCTs that were not included in the meta-analyses are reported in Supplementary Table 2 and Supplementary Table 3.

**Effects of family-based interventions compared to individual-focused interventions**

Nine RCTs comparing family-based interventions to individual-focused interventions were included in the analyses, performed for the outcomes of pain (n=8) [21, 35, 36, 38-40, 42, 45], disability (n=6) [21, 24, 35, 38, 39, 42], psychological disability (n=3) [35, 36, 42], depressive symptoms (n=3) [24, 38, 45], self-efficacy (n=5) [24, 35, 36, 38, 39] or marital adjustment (n=4) [24, 35, 36, 40]. Only one RCT had a follow-up shorter than 6 weeks [24] with these follow-up data not to be included in the meta-analyses.

Based on overall moderate quality evidence, family-based interventions had a small, significant positive effect on pain (MD: -3.55/100; 95%CI -4.03, -3.06) at short-term follow-up (n=6) (Figure 2A). Based on very low or low quality evidence, there were no between-group differences at mid (MD: -0.60/100, 95% CI -4.92, 3.72) (n=6), and long-term (MD: 0.38/100, 95%CI -5.08 to 5.84) follow-ups (n=5) (Figure 2A).

Based on moderate quality evidence, family-based interventions had a very small, significant positive effect on disability (MD: -1.51/100; 95%CI -1.98, -1.05) at short-term follow-up (n=6) (Figure 2B). Based on very low or low quality evidence, there were no between-group differences at mid (MD: -0.97/100, 95% CI -4.31, 2.37) (n=3), and long-term (MD: 0.02/100, 95%CI -3.21 to 3.26) (n=3) follow-ups (Figure 2B).

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There were no differences between family-based versus individual-focused interventions on mood, self-efficacy, or marital adjustment at any follow-up time-points (Figures 3A, 3B, 3C).

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*Secondary-analyses*

We conducted secondary analyses on pain, disability, mood, self-efficacy, and marital adjustment for RCTs that compared family-based to individual-focused interventions and included patients with osteoarthritis (peripheral joint [35, 36, 39], or peripheral joint and spinal [38] osteoarthritis) or chronic low back pain [21, 45]. For osteoarthritis, family-based interventions resulted in small positive improvements in pain (MD -5.22/100; 95%CI -9.72, -0.72), and disability (MD -1.06/100; 95%CI -1.38, -0.74) only at short-term follow-up (Figures 4A and 4B) compared to individual-focused interventions. No differences were observed for other outcomes and follow-up time-points (Figures 4C, 4D, 4E).

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For chronic low back pain, it was possible to perform subgroup meta-analyses only for pain at short and long-term follow-ups. There were no differences between groups at both short term (MD: -1.29/100, 95%CI -8.67, 6.08), or long term (MD: 2.01/100, 95%CI -7.62, 11.64) follow-up time-points (Figure 5).

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**Effects of family-based interventions compared to usual care**

Six RCTs comparing family-based interventions to usual care [20, 21, 36, 39, 40, 43-45] were included in the meta-analyses performed for the outcomes of pain (n=6) [20, 21, 36, 39, 40, 43-45], disability (n=3) [20, 21, 43-45], self-efficacy (n=2) [36, 39] or marital-adjustment (n=2) [36, 40]. Only one RCT [20, 43, 44] had a follow-up greater than 12 months (60 months) and these data were not included in the meta-analyses.

Based on low quality evidence, family-based interventions had a small positive effect on pain (MD: -6.05/100; 95%CI -6.78, -5.33) at the short term (n=3). Based on very low-quality evidence, there was no between-group difference (MD: -2.27/100; 95%CI -10.61, 6.07) at mid-term follow-up (n=4) (Figure 6A). There was no between-group difference on disability at short-term (MD: 1.73/100, 95%CI -13.84, 17.30) (n=2) and mid-term (MD: -2.22, 95%CI -6.49, 2.06) (n=2) follow-ups (Figure 6B).

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Family-based interventions had a small, significant positive effect on self-efficacy (MD: -6.06, 95%CI -6.69, -5.44) at mid-term follow-up (n=2) (Figure 7A). Nonetheless, there was no between-group difference on marital adjustment (MD: -0.96, 95%CI -9.24, 7.31) at mid-term follow-up (n=2) (Figure 7B).

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One RCT that included children with juvenile idiopathic arthritis compared a family-based intervention to usual care and was not included in the pooling due to heterogeneity of clinical populations [26]. Results from this RCT showed no differences between groups on pain intensity, disability and quality of life and secondary outcomes (i.e. pain catastrophising, pain-specific beliefs, self-efficacy, and disease activity) (Supplementary Table 3).

**Discussion**

Fifteen RCTs were included in the qualitative review and ten RCTs were included in the meta-analyses of this systematic review. Pooled data from primary analyses including all musculoskeletal conditions showed that family-based interventions had small positive effects on pain and disability at the short-term compared to individual-focused interventions, but not at mid and long-term follow-ups, nor on mood, self-efficacy and marital adjustment. Results from the secondary analyses including patients with low back pain or osteoarthritis evidenced small positive effects on pain intensity and disability for patients with osteoarthritis at the short-term, but no effects were observed on pain intensity for patients with low back pain. When compared to usual care, family-based interventions resulted in small greater effects on pain and self-efficacy at the short term, but not on pain at mid-term follow-up, nor disability or marital adjustment.

To the best of our knowledge, this was the first systematic review to investigate the effect of family-based interventions in the treatment of musculoskeletal pain specifically. Strengths of this review include a comprehensive search strategy, inclusion of patients with various musculoskeletal conditions and analyses of the effects of family-based interventions on different health-related and relationship outcomes. However, included RCTs were of fair methodological quality and most included patients and their spouses, whilst only one RCT included children and their parents. Few studies were included in each meta-analysis (especially in the secondary analysis for low back pain and osteoarthritis), most of them had a small sample size and did not have a long follow-up resulting in a certain degree of caution when interpreting the precision related to the estimates of the meta-analysis. The low quality of evidence due to serious inconsistency and imprecision at mid and long-term follow-ups in the outcome of pain should also be considered. Moreover, there were design differences in the original studies regarding the delivery of family-based interventions, including the duration of the intervention and the degree of involvement of family members, which should also be considered as possible limitations of the study.

Various factors may be considered to explain the possible small and inconsistent effects of family-based interventions observed in this review. These factors may include: i) a few RCTs with overall small sample sizes, with one RCT [39] explaining most of the positive results; ii) the presence of treatment contamination – i.e. some RCTs [34-36] encouraged patients randomised to individual-focused interventions to communicate more effectively with their family members and share the knowledge gained from the treatment sessions; iii) the fact that individual-focused interventions were evidence-based and shown to be effective in previous studies (e.g. standard cognitive behavioural pain management programme [47]), perhaps the most important factor. Alternatively, it is possible that family-based interventions might add only small advantages over individual-focused and usual care interventions, and therefore trials in the field would require much larger sample sizes to detect small between-group differences [24]. This might be even more relevant to RCTs comparing family-based to usual care interventions in children with musculoskeletal pain since parents are usually included in the usual care offered to children.

The characteristics of the family-based interventions could also contribute to the results observed in the review. When comparing RCTs included in the meta-analysis of our primary outcome (pain), there are no clear differences in the characteristics of family-based interventions which had positive effects [35, 38, 39] and no effect [36, 39, 40, 42, 45] on pain. Interventions were delivered within a similar number of sessions (6-10 [35, 38, 39] vs 5-12 [21, 36, 40, 42, 45] weekly sessions) of similar duration (approximately 2 hours) with spouses being the family members involved [21, 35, 36, 38-40, 42, 45]. In addition, interventions had similar components, including education, coping skills training, communication and goal-setting [21, 35, 36, 38-40, 42, 45]. Moreover, RCTs were of varied sample sizes and there were also no clear differences between the studies with positive and no effects [21, 35, 36, 38-40, 42, 45]. However, RCTs reporting a positive effect of family-based interventions were conducted with slightly older patients (61 to 71 years old) with osteoarthritis [35, 38, 39], while RCTs which did not find differences between groups were conducted with younger patients (44 to 60 years old) with rheumatoid arthritis [42], musculoskeletal pain [40], low back pain [21, 45] and osteoarthritis [26]. This might indicate that older patients could benefit more from family-based interventions than younger counterparts.

At this stage, we can only speculate why the results from studies conducted with patients with osteoarthritis and low back pain presented different results. We would not go as far as attributing the differences in effects to types or features of interventions delivered for these two conditions, as they were fairly similar (6-10 weekly, 2 hours groups sessions with spouses or partners involving pain education, coping skills training, combined goal setting). Possible explanations for the differences in results across conditions could include different levels of adherence to treatment. However, only two RCTs reported data on intervention adherence [21, 39]. Attendance rate by patients to family-based interventions was also similar among the two RCTs (89% in both studies), but patients with osteoarthritis presented a lower attendance rate to individual-focused intervention (58%) [39] when compared to patients with chronic low back pain (89%) [21]. The methodological quality of the two groups of trials was also similar (PEDro score = 5/10 points). Therefore, it is possible that these differences might reflect differences in response to treatment as a result of specific disease characteristics. It is also possible that age might have influenced patients’ response to treatment, as the studies that included patients with osteoarthritis reported a higher mean age (mean age range: 54 to 71 years) compared to those with low back pain (mean age range: 39 to 46 years). Again, it is possible that older patients might benefit more from having their family members and/or peers involved in their disease management process than younger people. This should be investigated in future clinical trials.

Considering the influence of family members on each other’s physical activity levels [5, 48] and the importance of physical activity in the management of musculoskeletal disorders [49, 50], it is surprising that none of the RCTs included in the systematic review assessed the influence of family-based interventions on physical activity. Most of the RCTs included in the review delivered interventions based on pain education, pain management, and/or cognitive behaviour therapy [19, 21, 24, 26, 34-42, 45], while only four studies delivered interventions targeting other lifestyle factors, such as physical activity [24, 36, 42, 45]. Perhaps including physical activity components in family-based interventions could represent an effective strategy to optimise the benefits of the family-based approaches for patients with musculoskeletal pain.

Previous systematic reviews have shown positive and promising effects of including family members in the treatment of chronic illnesses on depressive symptoms [22], pain [22], marital functioning (satisfaction or partner support) [22, 23], self-efficacy [23], and quality of life [23]. Findings from our meta-analyses only partially support these positive findings. Interestingly, the family-based interventions delivered in the studies included in the previous reviews had similar components to those included in the current review, including disease education and management. However, most of the studies included in the previous reviews were conducted with patients with cancer [22, 23], which might have contributed to the overall larger effect sizes compared to the current review. Furthermore, studies included in the current review were of fair methodological quality while studies included in a previous systematic review were of moderate methodological quality [23].

In conclusion, there is moderate-quality evidence that family-based interventions result in small but significantly better pain and physical function in the short-term compared to individual-focused interventions in patients with musculoskeletal pain. There is low-quality evidence that family-based interventions have small greater effects on pain at the short-term compared to usual care. However, results might be too small to be considered worthwhile. No differences were found at mid and long terms and for the outcomes of mood, depressive symptoms, self-efficacy, and marital adjustment when family-based interventions were compared to individual-focused interventions. A small greater effect was found on self-efficacy but not on marital adjustment when family-based interventions were compared to usual care. Future research that further develops and tests family-based interventions on mid to long-term outcomes in musculoskeletal pain is needed. Future studies should review the content and optimise the mechanisms underpinning family-based interventions in musculoskeletal pain so that the approach could be further tested in adequately powered RCTs.

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**Figures’ legends**

**Figure 1.** Flow-chart of included RCTs

**Figure 2A.** Forest plot of the effect of family-based compared to individual-focused interventions on pain intensity

**Figure 2B.** Forest plot of the effect of family-based compared to individual-focused interventions on disability

**Figure 3A.** Forest plot of the effect of family-based compared to individual-focused interventions on mood

**Figure 3B.** Forest plot of the effect of family-based compared to individual-focused interventions on self-efficacy

**Figure 3C.** Forest plot of the effect of family-based compared to individual-focused interventions on marital adjustment

**Figure 4A.** Forest plot of the effect of family-based compared to individual-focused interventions on pain intensity in patients with osteoarthritis

**Figure 4B.** Forest plot of the effect of family-based compared to individual-focused interventions on disability in patients with osteoarthritis

**Figure 4C.** Forest plot of the effect of family-based compared to individual-focused interventions on mood in patients with osteoarthritis

**Figure 4D.** Forest plot of the effect of family-based compared to individual-focused interventions on self-efficacy in patients with osteoarthritis

**Figure 4E.** Forest plot of the effect of family-based compared to individual-focused interventions on marital adjustment in patients with osteoarthritis

**Figure 5.** Forest plot of the effect of family-based compared to individual-focused interventions on pain intensity in patients with chronic low back pain

**Figure 6A.** Forest plot of the effect of family-based interventions compared to usual care on pain intensity

**Figure 6B.** Forest plot of the effect of family-based interventions compared to usual care on disability

**Figure 7A.** Forest plot of the effect of family-based interventions compared to usual care on self-efficacy

**Figure 7B.** Forest plot of the effect of family-based interventions compared to usual care on marital adjustment

**Table 1.** Characteristics of included RCTs

|  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Author (year)****Country** | **Condition** | **Symptom duration****(years; mean±SD)** | **Sample size (% females)** | **Age (mean±SD)** | **Outcome measures reported** | **Interventions arms** | **Duration of the intervention** | **Follow-up time-points** |
| Abbasi (2012) Iran | CLBP | 6.16(0.5-23.0) | G1: 10 (%NR)G2: 12 (%NR)G3: 11 (%NR) | Not reported | Pain (VAS)Disability (RMDQ)Kinesiophobia (TSK)Pain Catastrophising (PCS) | G1: Spouse-assisted multidisciplinary pain management interventionG2: Individual-focused multidisciplinary pain management interventionG3: Usual care | 7 weeks | 7 weeks and 12 months |
| Kole-Snijders (1999) Netherlands | CLBP | G1:8.2±8.6G2:10.7±8.9G3:11.3±8.6 | G1: 59(63%)G2: 58(66%)G3: 31(61%) | G1:39.7±8.8G2:39.2±9.2G3:41.1±9.6 | Pain (VAS; MPQ)Pain coping (PCL; CSQ)Pain Behavior (CHIP; PaBS)Pain Catastrophising (MLPC)Anxiety (NHQ)Aerobic fitness (BAT) | G1: Spouse-assisted operant behavioural intervention with cognitive coping skills training G2: Spouse-assisted operant behavioural intervention with discussion group G3: Waitlist control and Individual-focused operant behavioural intervention  | 2 months | 2, 6 and 12 months |
| Saarijarvi (1991a,1991b, 1992) Finland | CLBP | G1:1.6±1.4G2:2.1±1.9 | G1: 33(42%)G2: 30(53%) | G1:46.5±9.6G2:46.4±8.0 | Pain (Pain Index from SNQ)Disability (Impairment Index from SNQ; ADL; FCI)Relationship with partners (MMQ)Psychological distress (BSI)Health attitudes (AS) | G1: Spouse-assisted interventionG2: Usual care | 5 months | 5, 12 and 60 months |
| Turner (1990)USA | CLBP | 12.9(0.59 – 4.0) | *Overall*N: 96 (48%)*Groups:*G1: 24 (%NR)G2: 25(%NR)G3: 24(%NR)G4: 23(%NR) | *Overall*44.0(25.0-64.0)\* | Pain (MPQ)Physical and psychosocial dysfunction (SIP)Pain behavior (PBC; observer rating of pain behavior)Depression (CES-D)Physical fitness (physical work capacity; strength; flexibility) | G1: Spouse-assisted behaviour and exercise intervention G2: Spouse-assisted behaviour interventionG3: Individual-focused exercise interventionG4: Usual care | 2 months | 6 and 12 months |
| Buchanan (2017)USA | OA (73% knee, 54% hip)§ | Not reported | *Overall*N: 17 (47%)*Groups:*G1: 10(%NR)G2: 07(%NR) | G1:54.0(50.0-68.0)\*G2:56.0(50.0-72.0)\* | Pain (WOMAC)Disability (WOMAC)Stiffness (WOMAC)Depression (PHQ-8)Sleep (Actigraph; PROMIS; ISI) | G1: Partner-assisted yogaG2: Individual-focused yoga | 3 months | 3 months |
| Keefe (1996,1999)USA | knee OA | 10.7±7.83 | G1: 30(60%)G2: 28(68%)G3: 29(52%) | G1: 63.5G2: 62.8G3: 61.4 | Pain (AIMS)Disability (AIMS)Marital adjustment (DAS)Pain coping (CSQ)Pain Behavior (observer-rated)Psychological disability (AIMS)Self-efficacy (ASES) | G1: Spouse-assisted coping skills interventionG2: Spouse-assisted arthritis education support control G3: Individual-focused coping skills intervention | 10 weeks | 10 weeks, 6 and 12 months |
| Keefe (2004)USA | knee OA | Not reported | G1: 18 (50%)G2: 20 (65%)G3: 16 (38%)G4: 18 (61%) | G1:60.0±12.1G2:60.2±9.0G3:60.2±8.7G4:57.6±14.2 | Pain (AIMS)Marital adjustment (DAS)Pain coping (CSQ)Psychological disability (AIMS)Self-efficacy (ASES)Aerobic fitness (ergometry and muscle strength) | G1: Spouse-assisted coping skills interventionG2: Spouse-assisted coping skills intervention plus exercise trainingG3: Individual-focused exercise training G4: Usual care | *Intervention:*3 months*Follow-up:*3 months |  |
| Martire (2003)USA | OA (92% knee, 67% back)§ | 18.0±14.5 | G1: 13 (100%)G2: 11 (100%) | *Overall*71.8±7.8 | Pain (AIMS)Disability (HAS)Relationship with partners (satisfaction with spousal assistance; spousal emotional support and insensitive responses)Depression (CES-D)Self-efficacy (ASES) | G1: Spouse-assisted arthritis self-help course interventionG2: Individual-focused education intervention | 6 weeks | 6 weeks |
| Martire (2007)USA | hip or knee OA | G1:14.3±9.4G2:15.3±11.8G3:16.1±12.0 | G1: 89 (72%)G2: 99 (73%)G3: 54 (72%) | G1:68.0±8.0G2:69.2±7.2 G3:68.4±7.5 | Pain (WOMAC)Disability (WOMAC)Stiffness (WOMAC)Depression (CES-D)Self-efficacy (ASES) | G1: Spouse-assisted oriented education and support interventionG2: Individual-focused oriented education and support interventionG3: Usual care | 6 weeks | 6 weeks and 6 months |
| Martire (2008)USA | hip or knee OA | G1:14.1±8.9G2:14.5±12.2 | G1:64 (75%)G2: 62 (68%) | G1:69.0±7.2G2:68.7±8.4 | Spousal support and responses (WHYMPI) | G1: Spouse-assisted oriented education and support interventionG2: Individual-focused oriented education and support intervention | 6 weeks | 6 weeks and 6 months |
| Moore (1985)USA | CP(>65% MSK pain)  | 16.5±12.6 | *Overall:*N: 43 (2%)*Groups:*G1: 17(%NR)G2: 14(%NR)G3: 12(%NR) | *Overall*49.3±13.2 | Pain (VAS)Physical and psychological dysfunction (SIP)Marital adjustment (MAT)Mood/emotion (MMPI; SIP)Pain Behavior (spouse-rated - VAS)Other outcomes (care seeking; spouse-rated personal and role skill)  | G1: Spouse-assisted interventionG2: Individual-focused interventionG3: Usual care | 3 months | 3 and 7 months |
| Ramke (2016)Australia | CP(89% MSK pain) | G1:6.2±3.5G2:8.4±4.9 | G1: 19 (68%)G2: 26 (35%) | G1:45.3±8.4G2:47.4±13.2 | Disability (RMDQ)Marital adjustment (DAS)Family impact of pain (FIPS)Self-efficacy (PSEQ)Kinesiophobia (TSK)Depression, anxiety and stress (DASS) | G1: Spouse-assisted interventionG2: Individual-focused intervention | 3 weeks | 3 and 7 weeks |
| Radojevic (1992)USA | RA | 11.8 (1-33)\* | *Overall*N: 59(76%) | *Overall*54.4 | Pain (AIMS)Disability (AIMS)Psychological disability (AIMS)Depression (CES-D)Disease activity (assessed by a rheumatologist) | G1: Family-based education supportG2: Family-based behaviour therapy supportG3: Individual-focused behaviour therapyG4: Usual Care | 6 weeks | 6 weeks and 2 months |
| Riemsma (2003) Netherlands | RA | Not reported | G1: 71 (58%)G2: 71 (66%)G3: 76 (62%) | G1:57.2±10.3G2:55.1±10.3G3:57.0±8.3 | Pain (AIMS)Disability (AIMS)Relationship with partners (social interactions assessment)Psychological disability (AIMS)Self-efficacy (ASES)Disease activity (DAS-28) Health Behavior (self-reported)Fatigue (VAS) | G1: Partner-based self-management education interventionG2: Individual-focused self-management education interventionG3: Individual-focused education  | 2 months | 2, 6 and 12 months |
| Lomholt (2015) Denmark | JIA | G1:5.1±4.0G2:7.6±4.0 | G1: 9(89%)G2: 10 (70%) | G1:11.4±2.0G2:12.0±1.4 | Pain (VAS)Disability (FDI)Quality of life (PedsQL)Pain catastrophising (PCQ)Pain specific beliefs (SOPA)Self-efficacy (CASE)Satisfaction with treatment (ESQ)Credibility of treatment (Likert scale) | G1: Family-based cognitive behaviour interventionG2: Usual care | 2 months | 2 months |

**Legend:** CLBP = Chronic Low Back Pain; OA = Osteoarthritis; CP = Chronic Pain; MSK = Musculoskeletal; RA = Rheumatoid Arthritis; JIA = Juvenile idiopathic arthritis; yrs = years; hs = hours; min = minutes; \*median(min-max). G1: Group 1; G2: Group 2; G3: Group 3; G4: Group 4; §Participants with pain in various body areas

VAS: Visual Analogue Scale; MAT: Marital Adjustment Test; RMDQ: Roland and Morris Disability Questionnaire; TSK: Tampa Scale of Kinesiophobia; PCS: Pain Catastrophising Scale; DASS Depression Anxiety Stress Scale; MPQ: McGill Pain Questionnaire; PCL: Pain Cognition List; CSQ: Coping Strategies Questionnaire; CHIP: Checklist Interpersonal for Pain Behavior; PaBS: Pain Behavior Scale; MLPC: Multidimensional Locus of Pain Control Questionnaire; NHQ: Nijmegen Hyperventilation Questionnaire; BAT: Behavioral Approach Tests; SNQ: Standardized Nordic Questionnaire; ADL: Activities of Daily Living; FCI: Functional Capacity Index; MMQ: Marital Questionnaire; BSI: Brief Symptom Inventory; AS: the Attitude Scale; SIP: Sickness Impact Profile; PBC: Pain Behavior Checklist; CES-D: Center for Epidemiologic Studies Depression Scale; WOMAC: The Western Ontario and McMaster Universities Osteoarthritis Index; PHQ-8: Patient Health Questionnaire-8; PROMIS: Patient-Reported Outcomes Measurement Information System; ISI: Insomnia Severity Index; AIMS: Arthritis Impact Measurement Scale; DAS: Dyadic Adjustment Scale; ASES: Arthritis Self-Efficacy Scale; HAS: Health Assessment Questionnaire; WHYMPI: West Haven-Yale Multidimensional Pain Inventory; MMPI: Minnesota Multiphasic Personality Inventory; FIPS: Family Impact of Pain Scale; PSEQ: Pain Self-Efficacy Questionnaire; PMI: Pain Management Inventory; DAS-28: Disease Activity Score; FDI: Functional Disability Inventory; PedsQL: Pediatric Quality of Life Inventory; SOPA: Survey of Pain Attitudes; PCQ: Pain Coping Questionnaire; CASE: Children’s Arthritis Self-Efficacy Scale

**Table 2.** Intervention characteristics of included RCTs

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Author (year)** | **Intervention type(s) and content(s)** | **Intervention characteristics (number, frequency and duration of sessions)** | **Family members involved** | **Health professionals involved, mode and setting** |
| Abbasi (2012) | G1: Group sessions (6 patients + spouses): Education about CLBP self-management, coping skills, influence of the spouse, health-related goal of the coupleG2: Same as G1 without information about influence of the spouse and health-related goal of the coupleG3: Usual care | 7 weekly sessions, 2h/ session  | Spouse | psychologist, orthopedic surgeon, psychiatrist,physiotherapist face-to-facepain clinic |
| Kole-Snijders (1999) | G1: Group sessions (number of participants not reported): operant behavior therapy including 5 weeks of inpatient and 3 weeks of outpatient (3x/week) treatment (physical activity goals, physiotherapy, occupational and psychological therapy, spouse group training including education and CBT) with cognitive coping skillsG2: Group sessions (number of participants not reported): operant behavior therapy (same as G1) and discussion group (reading of pain book for patients)G3: Waitlist group: received the operant behavioral therapy after the control period, but they were individualized and did not include spouse group training | Operant behavior therapy: 88h of physiotherapy; 38h of occupational therapy (frequency and number of sessions not reported); 8 weekly psychology sessions, 0.5h/ session; spouse group training: 7 weekly sessions, 1.5h/ sessionCognitive coping skills training and discussion group:12 sessions, 1.6h/ session, frequency not reported | Spouse | physiotherapists,occupational therapist, behavioral therapist, psychologist Face-to-faceNot reported |
| Saarijarvi (1991a,1991b, 1992)  | G1: Group sessions (number of participants not reported): couple therapy sessions based on the family systems approachG2: Usual care | 5 monthly sessions, 1-2h/ session | Spouse | psychologistface-to-facerehab. centre |
| Turner (1990)  | G1: Group sessions (5-10 patients): pain behavior, social reinforcement and communication training and behavioral goal-setting (spouses attended to 5/8 sessions) + communication booklet + aerobic exercise sessionsG2: Same as G1 without exercise interventionG3: Aerobic exercise sessions without involvement of the spouseG4: Usual care | 8 weekly sessions, 2h/ session | Spouse | psychologist,physiotherapistsface-to-facenot reported |
| Buchanan (2017) | G1: Group sessions (3-7 patients + partners): yoga sessions and handouts for combined home-practiceG2: Same as G1 without involvement of the partner | 12 weekly sessions, 1.25h/ session | Not reported | Yoga instructorface-to-facecommunity center |
| Keefe (1996,1999) | G1: Group sessions (4-6 patients + spouses): CBT to develop pain coping skills and couples’ skills (mutual goal-setting, communication, joint home exercise practice) G2: Group sessions (4-6 patients + spouses): education on arthritisG3: Same as G1 without involvement of the spouse and couples’ skills activities | 10 weekly sessions, 2h/ session | Spouse | psychologist, nursesface-to-facenot reported |
| Keefe (2004) | G1: Group sessions (3-5 patients + spouses): information about pain, coping skills education and importance of spouses’ involvementG2: Same educational sessions as G1 + strength and flexibility training sessionsG3: Group sessions (4-6 patients): cardiopulmonary, strength and flexibility training sessions without involvement of the spouseG4: Usual care | Coping skills training:12 weekly sessions, 2hs/ sessionsCardiopulmonary training: 36 sessions, 3x/week, 1.5h/ sessionStrength training:24 sessions, 2x/week, 1.5h/ session | Spouse | psychologist, exercise physiologists face-to-facenot reported |
| Martire (2003) | G1: Group sessions (4-6 patients): education on arthritis, pain, management and coping skills + spouse-assisted sessions covering emotional and communication aspects of managing arthritis as a coupleG2: Same educational intervention as G1 without spouse-assisted sessions | 6 weekly sessions, 2h/ educational sessions; 0.3h/ spouse-assisted sessions | Spouse | Arthritis Self-Help Course specialistface-to-facenot reported |
| Martire (2007) | G1: Group sessions (4-6 patients + spouses): education on arthritis, pain, management, coping skills, communication and support within the couple and combined health-related goal-settings (topics framed as couples’ issue when possible) + up to 5 booster telephone callsG2: Same intervention as G1 without involvement of the spouses and framing topics as couples’ issuesG3: Usual care | Group sessions:6 weekly sessions, 2h/ sessionBooster sessions:up to 5 monthly sessions, 0.3h/session | Spouse | Arthritis Self-Help Course specialistface-to-face + telephone booster sessionsnot reported |
| Martire (2008) |  G1: Group sessions (4-6 patients + spouses): education on arthritis, pain, management, coping skills, communication and support within the couple and combined health-related goal-settings (topics framed as couples’ issue when possible) + up to 5 booster telephone callsG2: Same as G1 without involvement of the spouses | Group sessions:6 weekly sessions, 2h/ sessionBooster sessions:up to 5 monthly sessions, duration not reported | Spouse | Arthritis Self-Help Course specialistface-to-face + telephone booster sessionsnot reported |
| Moore (1985) | G1: Group sessions (4-6 patients + spouses): pain education, goal-setting, problem-solving and pain coping strategies + homework assignmentsG2: Same as G1 without involvement of the spouses and framing topics as couples’ issuesG3: Usual care | 8 weekly sessions, 2h/ session | Spouse | psychologist, social worker, nurses face-to-facenot reported |
| Ramke (2016)  | G1: Group sessions (8-10 patients). education on pain, goal-setting, activity pacing, relaxation techniques, exercise and couple interaction and communication training + telephone intervention for spouses only (couple interaction and communication)G2: Same as G1 but without involvement of the spouses | Group sessions:21 daily sessions, 8 h/ sessionTelephone intervention:3 weekly sessions, 2h/ session | Spouse | pain specialist, psychologist, nurse, physiotherapistsface-to-face + telephone callshospital |
| Radojevic (1992)  | G1: Group sessions (3-6 patients + family members): education and discussion about RA G2: Group sessions (3-6 patients + family members): education on pain and family support, coping strategies and relaxation training + home practice of learned skillsG3: Same as G2 without involvement of family members and discussion of the role of family supportG4: Usual care | Group sessions:4 weekly sessions, 1.5h sessionHome practice: 2 weeks | Spouse, adult child, roommate or family member with daily contact | clinical psychologistface-to-facenot reported |
| Riemsma (2003) | G1: Group sessions (8 patients + partners): education on RA, pain, exercise, depression, goal-setting, self-management, relaxation and communication + information booklet and self-help guide + booster sessionsG2: Same as G1 without involvement of the partnersG3: self-help guide only | Educational sessions:5 weekly sessions, 2hs/ sessionBooster sessions:3 tri-monthly sessions, 2hs/ session | Spouse (88%), close relative (10%), friend (2%) | nurses face-to-facenot reported |
| Lomholt (2015) | G1: Group sessions (number of participants not reported): CBT with combined (parents and children) and separated parts + workbook, worksheet and guides for home practice and combined goalsG2: Usual care | 6 sessions, 4 weekly sessions, 2 fortnightly sessions, 2hs/ session | Parents | psychologistface-to-facePediatric Rheumatology Clinic |

**Legend:** CLBP: Chronic Low Back Pain; CBT: Cognitive Behaviour Therapy, G: Group

**Table 3.** Risk of bias of included RCTs

|  |  |  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Author (year)** | **Eligibility criteria (no scored)** | **Random allocation** | **Concealed** **allocation**  | **Baseline comparability** | **Blind subjects** | **Blind therapists**  | **Blind assessors**  | **Adequate follow-up** | **Intention-to-treat analysis** | **Between-group comparisons**  | **Point estimates and variability** | **Total Score**  **(0-10)** |
| \*Abbasi (2012) | Marca de seleção | Marca de seleção | Fechar | Fechar | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | Marca de seleção | Marca de seleção | 5 |
| \*Buchanan (2017) | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Marca de seleção | Fechar | Marca de seleção | Marca de seleção | 5 |
| \*Keefe (1996, 1999) | Fechar | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Marca de seleção | Fechar | Marca de seleção | Marca de seleção | 5 |
| \*Keefe (2004) | Marca de seleção | Marca de seleção | Fechar | Fechar | Fechar | Fechar | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | 3 |
| \*Kole-Snijders (1999) | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Marca de seleção | Fechar | Marca de seleção | Marca de seleção | Marca de seleção | 6 |
| Lomholt (2015) | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | Marca de seleção | Marca de seleção | 6 |
| \*Martire (2003) | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Marca de seleção | Fechar | Marca de seleção | Marca de seleção | 5 |
| \*Martire (2007) | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | Marca de seleção | 5 |
| \*Martire (2008) | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | 4 |
| Moore (1985) | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | 4 |
| \*Radojevic (1992)  | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | 5 |
| Ramke (2016)  | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | 4 |
| \*Riemsma (2003) | Fechar | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | 4 |
| \*Saarijarvi (1991, 1991, 1992)  | Fechar | Marca de seleção | Fechar | Fechar | Fechar | Fechar | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Marca de seleção | 5 |
| \*Turner (1990)  | Marca de seleção | Marca de seleção | Fechar | Marca de seleção | Fechar | Fechar | Marca de seleção | Fechar | Fechar | Marca de seleção | Marca de seleção | 5 |

\* PEDro score provided from the PEDro database.  = Yes;  = No. Total score: 9-10 excellent, 6-8 good, 4-5 fair, and <4 poor.

**Figure 1**

Records identified through database searching

MEDLINE = 149

EMBASE = 690

PSYCINFO = 41

AMED = 10

WOS = 326

PEDRO = 31

CENTRAL = 200

CINAHL = 187

**n = 1634**

## **Identification**

## **Eligibility**

## **Screening**

Additional records identified through other sources
**n = 8**

Records after duplicates removed
**(n = 1223)**

Records excluded
**n = 1198**

Records screened
**n = 1231**

Full-text articles excluded, with reasons
**n = 15**

- Not RCT design (n=3)

- Not family-based intervention (n=5)

- Not musculoskeletal conditions (n=6)

- Family-based intervention given to both groups (n=1)

Full-text articles assessed for eligibility
**n = 33**

Articles included in qualitative synthesis
**n = 18**

RCTs included in qualitative synthesis

**n = 15**

RCTs excluded from quantitative synthesis, with reasons
**n = 6**

- Children (n=1)

- Data not reported (n=3)

- Outcomes for quantitative analyses were not measured (n=1)

## **Included**

RCTs included in quantitative synthesis

(meta-analysis)
**n = 10**