The effectiveness of therapeutic exercise for joint hypermobility syndrome: A systematic review

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TITLE PAGE

Title

The effectiveness of therapeutic exercise for joint hypermobility syndrome: a systematic review

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ABSTRACT

Title: The effectiveness of therapeutic exercise for joint hypermobility syndrome: a systematic review

Background: Joint hypermobility syndrome (JHS) is a heritable connective tissue disorder characterised by excessive range of movement at multiple joints accompanied by pain. Exercise is the mainstay of management yet its effectiveness is unclear.

Objectives: To establish the effectiveness of therapeutic exercise for JHS.

Design: Systematic literature review.

Data sources: A search of nine online databases, supplemented by a hand search and snowballing.

Study eligibility criteria (participants and interventions): People diagnosed with JHS (rather than asymptomatic generalised joint laxity); therapeutic exercise (of any type) used as an intervention; primary data reported; English language; published research.

Study appraisal and synthesis methods: Methodological quality was appraised by each reviewer using Critical Appraisal Skills Programme checklists. Articles were then discussed collectively and disagreements resolved through debate.

Results: 2 001 titles were identified. Four articles met the inclusion criteria, comprising one controlled trial, one comparative trial and two cohort studies. All studies found clinical improvements over time. However there was no convincing evidence that exercise was better than control or that joint-specific and generalised exercise differed in effectiveness.

Limitations: The studies used heterogeneous outcome measures, preventing

pooling of results. Only one study was a true controlled trial which failed to report between-group statistical analyses post-treatment.

Conclusions and implications of key findings: There is some evidence that people with JHS improve with exercise but there is no convincing evidence for specific types of exercise or that exercise is better than control. Further high quality research is required to establish the effectiveness of exercise for JHS.

Keywords

Joint hypermobility; benign hypermobility syndrome; exercise; exercise therapy; systematic review

MANUSCRIPT

TITLE

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INTRODUCTION

Rationale

Joint hypermobility syndrome (JHS) has been defined as a *"heritable disorder of the connective tissues characterised by hypermobility, often affecting multiple joints, and musculoskeletal pains in the absence of systemic inflammatory joint disease such as rheumatoid arthritis"* [1]. Variation in diagnostic criteria makes interpretation of published literature difficult but the revised Brighton Criteria [2] are now widely used. JHS is generally accepted to be more prevalent in children, in females and in some ethnic groups. Approximately 5% of women and 0.6% of men experience symptomatic joint hypermobility [3].

Joint pain in JHS is thought to be caused by excessive movement increasing stress on joint surfaces, ligaments and neighbouring structures [3]. Pain may cause muscle inhibition, leading to atrophy and reduced joint control [4]. Proprioceptive acuity may also be adversely affected [5, 6], perhaps due to joint mechanoreceptor damage [7]. The inability to acknowledge extreme joint ranges may create an even more unstable joint by further stretching supporting structures. JHS can be accompanied by fatigue [8], anxiety and depression, impacting negatively on social

function [9] and thereby having a substantial impact on individuals.

Acute pain episodes may be managed using taping, bracing or splinting [4] or with non-steroidal anti-inflammatory drugs [3]. However education [10, 11] and therapeutic exercise [12] are the mainstays of long term management. Encouraging an active lifestyle may improve function and enhance quality of life [13].

Strengthening exercises targeting stabilising muscles around hypermobile joints might enhance joint support throughout movement and reduce pain [14, 15]. Closed chain exercises may reduce strain on injured ligaments [16], enhance proprioceptive feedback [4], and optimise muscle action [17]. Coordination and balance exercises such as wobble board training may improve proprioception [18, 19]. Neural pathways and movement patterns consisting of muscle pair co-contractions are reinforced [20]. This can encourage compensation reactions [21], preventing joints moving into extreme ranges and avoiding further injury [3].

In contrast to specific muscle training, a generalised exercise approach can also be taken, addressing cardio-respiratory, musculoskeletal and neurological aspects of movement [22] and reducing general deconditioning [23]. Hydrotherapy can be a successful medium in which to perform such exercises [22], challenging balance and core strength within a supportive environment, with water resistance and buoyancy increasing exercise variability [24].

Although exercise is widely regarded as a core component of JHS management [12, 4], there is no clear consensus about its effectiveness. There is generally a lack of high quality research which might contribute to the prescription of inappropriate interventions [25] and negative experiences of physiotherapy [1]. It is timely that the available evidence for exercise should be systematically evaluated.

Objectives

This systematic review aimed to establish the effectiveness of therapeutic exercise for JHS. Due to the small number of studies identified in initial scoping work, it was decided not to prescribe the specific type of exercise or the clinical outcomes.

METHODS

This review has been reported in accordance with PRISMA recommendations [26].

Protocol and registration

No prior protocol was published.

Eligibility criteria

The following inclusion criteria were applied to retrieved records: 1. people with joint hypermobility syndrome (rather than asymptomatic generalised joint laxity); 2. therapeutic exercise (of any type) used as an intervention; 3. human participants; 4. primary data reported; 5. English language; 6. published research. The criteria were applied in turn to the titles, abstracts and full texts. No date restrictions were used to maximise record retrieval. All study designs were included.

Information sources

Following discussion and advice from a University librarian, nine online databases were searched. These were Allied & Complementary Medicine (AMED); British Nursing Index (BNI); Cumulative Index to Nursing & Allied Health Literature (CINAHL); Cochrane Library; Embase; Healthcare Management Information

Consortium (HMIC); Medline; Physiotherapy Evidence Database (PEDro); and SportDiscus. The OVID platform was used to search Embase and HMIC; EBSCO for AMED, CINAHL, Medline and SPORTDiscus; and ProQuest for BNI. The electronic search was supplemented by a manual hand search of relevant journals (Supplemental Information, Table A) and by snowballing of full articles retrieved.

Search

Key search concepts were identified as 'joint hypermobility syndrome' and 'therapeutic exercise'. Team discussion and an online thesaurus were used to identify alternative terms for the search key words. The final search terms are presented in Supplemental Information, Table B. The search strategy for EBSCO, OVID, ProQuest and the Cochrane Library were identical. PEDro required an adapted search strategy, where each search term for the ‘joint hypermobility syndrome’ concept was searched individually. This was felt to be sensitive enough for this physiotherapy-specific database. The search was conducted on 23rd November 2012.

Study selection

Duplicates were removed and the inclusion criteria applied to the titles of retrieved records. The abstracts of all remaining records were then obtained and the criteria applied again. Finally the full texts of remaining articles were obtained and the process repeated. Snowballing from the reference lists of the full articles maximised identification of relevant literature [27]. All decisions were discussed and agreed as a group, ensuring robust application of the inclusion criteria.

Data collection process and data items

Key data was extracted from the final articles, including study design, participant characteristics, sample size, diagnostic criteria, outcome measures, main findings and detailed information about the exercise interventions.

Risk of bias in individual studies

Risk of bias was assessed using Critical Appraisal Skills Programme (CASP) checklists [28]. CASP was selected because different checklists are available to assess the quality of different research designs. Each group member independently applied the appropriate checklist to each of the final articles. Individual critiques were discussed as a group with any disagreements resolved by group consensus.

Additional analyses

Where available, data on pain intensity from pre-treatment to immediately post-treatment was used to calculate standardised effect sizes. Due to heterogeneity in study design and outcomes, there was no other formal supplementary analysis or attempt to summarise or synthesise results across the included studies. Consistent patterns in the risk of bias across studies were identified following individual study assessment.

RESULTS

Study selection

The process of study selection is summarised in Supplemental Information, Figure A. After duplicates were removed a total of 2 001 potentially relevant articles were

identified (1 967 from the electronic search, two from the hand search and 32 from snowballing). Successive application of the inclusion criteria to the titles, abstracts and full texts left four articles for inclusion within the review (three from the electronic and hand search and one from snowballing).

Study characteristics

Table 1 provides a synopsis of each of the four included studies and Table 2 describes the exercise interventions more fully. The final four studies comprised a randomised comparative trial [29], a randomised controlled trial [30], and two cohort studies [1, 31]. Barton and Bird [31] failed to report their diagnostic criteria whilst the others used the Brighton criteria. The study by Kemp et al [29] was in a paediatric population, whilst the other studies were in adults. Sample sizes in the exercise intervention groups ranged from n=15 [30] to n=30 [29]. The studies by Sahin et al [30] and Ferrell et al [1] were specific to the knee joint, whilst the other two studies incorporated whole body exercise interventions.

Risk of bias within studies

The CASP tool for randomised controlled trials was used to assess the trials by Kemp et al [29] and Sahin et al [30]. The CASP tool for cohort studies was applied to Ferrell et al [1] and Barton and Bird [31]. Key findings from this quality appraisal are detailed below.

The randomised comparative trial by Kemp et al [29] compared generalised exercise against targeted (joint-specific) exercise. The assessing therapist was reported to be blind to treatment allocation and the treating therapist was blind to assessment data, although the success of blinding was not reported. Randomisation

was via a computer-generated list sequence contained in opaque envelopes but it was not clear who opened these and made the treatment allocations. The prospective sample size calculation of n=48 in each group was not reached and attrition was high (28% at 2 months and 44% at 5 months). The authors did not find statistically significant differences in baseline characteristics between those who did and did not complete the final assessment, although such analysis could be subject to type two errors. Closer inspection suggests a trend towards those dropping out having: less back pain, joint swelling, pain with exercise and medications; lower CHAQ scores; higher shuttle test performance; and higher parent's assessment of child's pain and parent's global assessment. Issues related to exercise adherence were not explicitly assessed. Other aspects of the trial seemed rigorous.

The randomised controlled trial by Sahin et al [30] compared the effectiveness of knee proprioception exercises against a control group. The process of allocating JHS patients to exercise and control conditions was inadequately reported and there was no reference to blinding patients, assessors or doctors delivering the exercise intervention. As highlighted in Table 1, there is some confusion in the study report related to sample sizes and there was no prospective sample size calculation. Exercise adherence and participant attrition are not reported. Statistical analyses of between-group differences after treatment are not reported and conclusions are instead based upon analysis of changes over time.

The cohort study by Ferrell et al [1] evaluated knee exercises. Analysis was limited to those who completed the exercise intervention, with 10% attrition due to relocation (n=2). It is not known whether there was any attempt to blind assessors or patients to the aims of the study or outcome scores. The wording used for the assessment of pain by visual analogue scale (VAS) was not clearly described. Other

aspects of the study are reported well. Adherence was monitored using an exercise diary and was found to be generally very positive.

The cohort study by Barton and Bird [31] investigated a general exercise programme. There was a lack of detail concerning outcome assessment. The study used a questionnaire that seems to have been developed by the authors but the method of development or psychometric properties are not reported. The same assessor was used throughout to enhance reliability, although attempts to blind patients or assessors are not reported. Exercise adherence was recorded but not reported.

Results of individual studies

Kemp et al [29] found no differences between groups in childrens' pain, parents' pain, CHAQ scores or the six-minute shuttle test. The only difference between groups was for parental global assessment which was better with targeted exercise at ~5 months (but not at ~2 months). When groups were combined, childrens' pain, parents' pain, and CHAQ scores improved over time (at both ~2 and ~5 months); parental global assessment improved only at ~2 months; but shuttle test performance did not change.

Sahin et al [30] found that exercise reduced participants' pain (at rest and on movement) and increased knee joint proprioception. This conclusion is based upon significant improvements observed over time in the exercise group which were absent in the control group. However there is no specific between-group statistical analysis reported and therefore a question mark remains about the true effectiveness of exercise. The AIMS-2 data demonstrated a statistically significant improvement over time in the exercise group for the occupational activity subscale

(but not for physical status, emotional status, symptoms or social activity status).

Ferrell et al [1] found that therapeutic exercise enhanced proprioceptive acuity, balance and strength; reduced pain VAS scores; and improved the physical functioning and mental health components of the SF-36.

Barton and Bird [31] found significant improvements in the maximum distance walked and pain on movement (in both the most affected joint and in all joints in general). The other 11 (out of 14) questionnaire items were non-significant. Range of motion of both knee joints improved with exercise but the other 15 (out of 17) joints were unchanged. Mean Carter and Wilkinson scores [32], an earlier version of the Beighton score, were also non-significant.

Synthesis of results

Synthesis of results was not possible due to heterogeneity of study designs and outcome measures. Standardised effect sizes for pain ranged from 0.75 to 1.72.

Risk of bias across studies

A common risk of bias includes convenient sampling from single centres.

DISCUSSION

Summary of evidence

This review identified one randomised comparative trial in children [29], and one randomised controlled trial [30] and two cohort studies in adults [1, 31]. The evidence suggests that people with JHS who undertake exercise improve over time in a range of patient (and parent) reported outcomes (including pain, global assessment of the

impact of hypermobility, maximum distance walked and quality of life) and objective outcomes (including proprioception, balance, strength and range of movement). There was no convincing evidence that improvements were any better than comparator groups. No adverse effects were reported. The quality of the two randomised trials [29, 30] has previously been independently rated as 6/10 and 3/10 respectively [33].

Limitations

There were some issues evident with sampling, diagnostic criteria and sample sizes, increasing the likelihood of type two errors and reducing external validity. All four studies used convenience sampling and one study [29] was on a paediatric population. The Brighton Criteria [2] were used for diagnosis in three of the four studies [1, 29, 30], although application differed slightly (See Table 1). Barton and Bird [31] report using recruitment interviews but fail to explicitly outline their diagnostic criteria. Sample sizes were small, ranging from n=20 to 57 (with n=15 to 30 in the exercise intervention arms). Only Kemp et al [29] reported prospective sample size calculations, although they failed to recruit to those.

Randomisation and blinding issues were also evident. Of the two randomised studies, only Kemp et al [29] report a clear randomisation process. Sahin et al [30] failed to state their randomisation method so potential allocation bias is unknown. Three studies fail to report attempts to blind researchers [1, 30, 31]. Although Kemp et al [29] conducted a single-blind trial, the success of blinding was not reported.

Kemp et al [29] lost 44% of their participants to follow up and Ferrell et al [1] lost two of their 20 participants due to relocation (10%). Intention-to-treat analyses were not employed but may have helped to reduce potential attrition bias [34].

Attrition was not reported in the other studies [30, 31].

The exercise interventions demonstrated wide heterogeneity (Table 4). Two studies concentrated on the knee joint [1, 30], limiting generalisability. Barton and Bird [31] provided a ‘menu’ of available exercises, avoiding exercises known to exacerbate individuals’ symptoms. There is variable focus on proprioceptive, balance and strength exercises, depending on individual study aims. This means that observed improvements cannot easily be attributed to one type of exercise. The descriptions of specific exercises, repetitions and progression are often difficult to interpret and replicate. There were very different levels of exercise supervision between studies and the location of exercise (home versus clinic) also varied (see Table 2). The very close supervision implemented by Sahin et al [30] (three times per week for eight weeks, supervised by a doctor in clinic) seems unrealistic for most healthcare settings.

The only trial to include a no exercise control [30] failed to conduct direct between-group statistical analyses, basing their conclusions on differences over time. The lack of a no exercise control group [29] and complete lack of comparison groups [1, 31] in the other studies means that the true effectiveness of exercise in this condition remains unknown. The length of follow up varied from immediately following the end of the exercise intervention [1, 30] to six weeks [31] and approximately 3 months afterwards [29]. Barton and Bird [31] recommended abstention from exercise during follow up, which saw a reversal in training effects. It is not clear what advice patients in Kemp et al [29] received about maintaining exercise during the follow-up period but most improvements were maintained at 3 months. The long term effects of exercise remain unclear.

A wide range of outcome measures were used, with all four using a visual

analogue scale (VAS) for pain, albeit very differently. For example Kemp et al [29] used a VAS with anchors of 'no pain' to 'worst pain possible' for children aged eleven to sixteen but a faces pain scale for those aged seven to eleven. Barton and Bird [31] do not report the anchors used but four separate VASs assessed ‘the most affected joint at rest’, ‘the most affected joint on movement’, ‘the pain in all your joints in general at rest’ and ‘the pain in all your joints in general on movement’. Sahin et al [30] used anchors of ‘no pain’ and ‘severe pain’ for knee pain ‘during movement’ and ‘resting position’. Ferrell et al [1] do not report the anchors used. Such variations in methodology complicate accurate comparisons and pooling of study results.

Ferrell et al [1] established reliability of their outcome measures by retesting a subgroup of participants prior to implementing the exercise intervention. However, Barton and Bird [31] use a self-composed questionnaire with no evidence of psychometric properties and fail to report whether goniometry assessed active or passive movement.

It would be useful if **future research** addressed issues related to sampling bias and sample size through multi-centre recruitment. The Brighton (1998) criteria [2] should be used to standardise diagnosis and participant and researcher blinding should be enhanced. Longer-term follow up and more complete description of the exercise interventions would be helpful.

A limitation of this review is that it was restricted to published literature in the English language and it is therefore possible that relevant material may have been missed.

Conclusions

Overall, the available evidence suggests that patients who received an exercise

intervention improved over time and no adverse effects were reported. However, there was no convincing evidence that generalised exercise was any better than joint-specific exercise [29] or that knee exercises were any better than a control condition [30]. Clear cause-effect relationships for exercise have therefore not been demonstrated. The methodological quality of the included studies was generally lacking, particularly with regards statistical power and adequate control conditions. Further robust studies are required to determine the effectiveness of therapeutic exercise for the management of JHS.

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Conflict of interest: There are no conflicts of interest.

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Table 1. Synopsis of included studies.

Authors	Kemp et al (2010) [29]	Sahin et al (2008) [30]	Ferrell et al (2004) [1]	Barton & Bird (1996) [31]
Study Design	Randomised Comparative Trial	Randomised Controlled Trial	Cohort Study	Cohort Study
Participant Characteristics Mean age (range), gender, location of recruitment	10.9 years (7-16) 38 male, 19 female Children's Rheumatology Department, UK	26.9 years (20-45) 6 male, 29 female [a] Physical Medicine & Rehabilitation Department Outpatient Clinic, Turkey	27.3 years (16-49) 2 male, 16 female Hypermobility Clinic, UK	Age not reported 2 male, 23 female Hospital referral or Patient Support Group, UK
Sample Size	n=57 randomised (General Exercise n=27, Targeted Exercise n=30) n=41 completed intervention (n=18, n=23) n=32 completed follow up (n=15, n=17)	n=40 (Exercise n=15, Control n=25) [b]	n=20 at baseline n=18 completed intervention	n=25
Diagnostic Criteria	Revised (Brighton 1998) Criteria: Beighton Score 4/9 or above & one major criteria, one major and two minor, four minor criteria, or two minor criteria with first-degree relative with hypermoblity	Revised (Brighton 1998) Criteria: Beighton Score 4/9 or above & one major or two minor symptoms Knee Pain	Revised (Brighton 1998) Criteria: Beighton Score above 4/9 & one major criteria or one major & at least two minor criteria Knee Pain	Not stated
Exercise Intervention	Whole body exercises: *General Exercise:* aim to maximise muscle strength and fitness *Targeted Exercise:* aim to address functional stability of symptomatic joints	Knee exercises: Knee proprioception exercises	Knee exercises: Knee proprioception exercises Balance exercises Knee strength exercises	Whole body exercises: Warm up/mobility exercises Specific joint exercises Proprioception exercises
Outcomes (Assessment method)	*Primary Outcome:* Child's pain (VAS) *Secondary Outcomes:* Parent's assessment of child's pain (VAS) Parent's global assessment of impact of pain (VAS)	Knee pain (VAS) at rest and on movement Knee proprioception (active-active method, biodex system 3pro multijoint system isokinetic dynamometer) Functional Status (AIMS-2	Knee pain (VAS) Knee joint proprioception (threshold detection paradigm) Balance (instrumented balance board) Knee strength (Kin-Com isokinetic dynamometer)	Pain at rest (VAS) Pain on Movement (VAS) Beighton Score Joint ROM (Loebl hydrogoniometer) Function (non-validated questionnaire)

	Functional Impairment (CHAQ) Six minute shuttle run	Questionnaire)	Quality of Life (Short Form 36 Questionnaire)	
Duration of intervention (Assessment points)	6 weeks (Baseline, ~2 months, ~5 months)	8 weeks (Baseline, 8 weeks)	8 weeks (Baseline, 8 weeks) [c] subgroup re-test between 2-8 weeks, of intervention, follow up)	6 weeks (Baseline, 6 weeks, 12 weeks)
Main Statistically Significant Findings (at end of treatment)	*Targeted Exercise only:* Reduced parent's global assessment (p=0.017) Reduced CHAQ (p=0.045) *Combined groups:* Reduced child's pain (p<0.001) Reduced parent's pain (p<0.001) Reduced parent's global assessment (p=0.005) Reduced CHAQ (p=0.024) [Maintained at ~5 months with the exception of parents' global assessment]	*Exercise Group:* Reduced pain (p<0.05) Increased knee proprioception (p<0.001)	Reduced pain (p=0.003) Increased proprioceptive acuity (p<0.001) Increased balance (p<0.001) Increased quadriceps and hamstrings muscle strength (p<0.05) Improved quality of life (physical functioning p=0.029, mental health p=0.008)	Reduced pain on movement (p<0.001) Increased maximum distance walked (p<0.006) Reduced Knee ROM (Left knee, p=0.003, Right knee, p=0.022) [Reversal of changes towards baseline at 12 weeks]
Standardised Effect Size for Pain (at end of treatment)	Child's pain (VAS) = 1.37	VAS at rest = 0.75 VAS on movement = 1.72	VAS = 1.12	Unable to calculate

[a] Note that the total number of males and females reported in the paper (n=6+29=35) varies from the reported total sample size (n=40).
[b] Note that the sample size is variably reported in the paper as n=30 (n=15 exercise, n=15 control), n=35 (n=15 exercise, n=20 control) and n=40 (n=15 exercise, n=25 control). The latter is most frequently reported in the paper and has therefore been used for the purposes of this review.
[c] Note that a subgroup of n=10 patients had repeat assessment 2-8 weeks after baseline (to test reproducibility) before receiving the exercise intervention.
Abbreviations: CHAQ = Childhood Health Assessment Questionnaire, HEP = Home Exercise Programme, ROM = Range of Movement, VAS = Visual Analogue Scale

Table 2. Description of the exercise interventions employed in each study.

Authors Exercise type, duration	**Details of exercise intervention and progression**	**Location: frequency, duration, supervision**
Kemp et al (2010) [29] Whole body exercises, 6 weeks	*General Exercise:* shuttle-runs; bunny-hops; squat-thrusts; sitting-to-standing; step-ups; star-jumps. Progression: Start at 30 seconds (or 10 repetitions) and add 15 seconds (or 5 or 10 repetitions) at a time *Targeted Exercise:* Control neutral joint position (facilitate optimal joint alignment in a resting position); Re-train dynamic control (maintain optimal joint alignment while moving adjacent joints); Motion control (improve control of the joint through its full range); Specific tissue lengthening (stretch short mobiliser muscles). Progression: reduce support, increase repetition, speed and duration. All exercises should be pain free.	Clinic: x1 per week, 30 minutes, supervised by physiotherapist Home: daily, duration not stated, no supervision
Sahin et al (2008) [30] Knee exercises, 8 weeks	Week 1: walking backwards, heel walking, walking on fingertips, walking with eyes closed, single leg balance, forward-backward bends on one leg - eyes open & closed (all 30 seconds duration), sit to stand from high chair (20 reps) Week 2: added exercise with rocker bottom wood (2-3 mins), slow sit-to-stand from low chair (10 reps), plyometric exercises (jumping over 15cm height, 10 reps), walking exercises (slow walk-broad circle, fast walk-broad circle, slow walk-narrow circle, fast walk-narrow circle, 5 reps each) Week 3: added biomechanical ankle platform system (BAPS) board balance wood (2-3 minutes), mini-trampoline jumping (30 reps)	Clinic: x3 per week, duration not stated, supervised by doctor
Ferrell et al (2004) [1] Knee exercises, 8 weeks	Week 1: squats, pliés, bridging (5 reps, 1 set) Week 2: doubled sets Week 3: added front lunges Week 4: doubled sets Week 5: increased to 10 reps but 1 set, added static hamstring exercises & balance board (2 mins x 3 sets). Week 6: doubled sets, balance board (4 sets) Week 7: increased to 15 reps but 1 set, added side lunges Week 8: doubled sets, balance board remained at 4 sets	Home: x4 per week, duration not stated, no supervision
Barton & Bird (1996) [31] Whole body exercises, 6 weeks	Individual exercise programmes with a number of the following: Warm up/mobility exercises: shoulder rolls, arm circles, neck rotations, neck lateral flexions, wrist circles, side flexions of spine, thoracic rotations in sitting Specific joint exercises: hamstring curls in standing/prone, static hamstring in sitting, hip extensions in prone (knee extended/flexed), pelvic tilts, sit ups, chest press in supine, arm elevations in supine, resisted bicep curls, resisted bicep curls at 90degrees shoulder abduction, finger opposition, wrist flexion/extension, pronation/ supination, heel raises, alternate tiptoe-heel walking, ankle plantar/dorsiflexion, resisted ankle inversion/eversion Proprioception exercises: single leg ball rolling, single leg balance Progression: None	Home: frequency not stated, duration not stated, no supervision Assessments of outcome measures every 2 weeks

TABLE AND FIGURE LEGENDS

Table 1. Synopsis of included studies.

Table 2. Description of the exercise interventions employed in each study.

SUPPLEMENTAL INFORMATION

Table A. Journals included in the hand search.

Journal	Years
Australian Journal of Physiotherapy	1988-2009
British Journal of Sports Medicine	1992-2010
Clinical Rehabilitation	1995-2004
Physical Therapy	1984-2010
Physical Therapy Reviews	1997-2010
Physiotherapy	1985-1994
Physiotherapy Canada	1987-2010
Physiotherapy in Sport	1987-2006
Physiotherapy Practice	1985-1988
Physiotherapy Research International	1996-2006
Physiotherapy Theory & Practice	1986-1998
The Physician & Sports Medicine	1990-2005

Table B. Search strategy key word list. Terms within each key search concept were combined with the Boolean operator 'OR' and the two key search concepts were then combined with 'AND'. Truncations were used to gain pleural terms and various suffixes (for example 'physical therap*' will include 'physical therapy', 'physical therapist' and 'physical therapies').

Key search concept 1	Key search concept 2
Joint hypermob*	Therapeutic exercis*
Benign joint hypermobility syndrome	Exercise therap*
JHM*	Physical exercis*
JHS	Physical therap*
HMS	Aerobic*
BJH*	Balanc*
	Hydrotherap*
	Activit*
	Strength training
	Physical intervention*

Figure A. Flow chart of study selection, following PRISMA recommendations [28].