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CHILDREN'S ORTHOPAEDICS

A systematic review of the non-surgical treatment of Perthes' disease

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Aims

Perthes' disease is a condition leading to necrosis of the femoral head. It is most common in children aged four to nine years, affecting around one per 1,200 children in the UK. Management typically includes non-surgical treatment options, such as physiotherapy with/without surgical intervention. However, there is significant variation in care with no consensus on the most effective treatment option.

Methods

This systematic review aims to evaluate the effectiveness of non-surgical interventions for the treatment of Perthes' disease. Comparative studies (experimental or observational) of any non-surgical intervention compared directly with any alternative intervention (surgical, non-surgical or no intervention) were identified from: Cochrane Central Register of Controlled Trials, MEDLINE, EMBASE, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), EMcare, Allied and Complementary Medicine Database (AMED), and the Physiotherapy Evidence Database (PEDro). Data were extracted on interventions compared and methodological quality. For post-intervention primary outcome of radiological scores (Stulberg and/or Mose), event rates for poor scores were calculated with significance values. Secondary outcomes included functional measures, such as range of movement, and patient-reported outcomes such as health-related quality of life.

Results

In all, 15 studies (1,745 participants) were eligible for inclusion: eight prospective cohort studies, seven retrospective cohort studies, and no randomized controlled trials were identified. Non-surgical interventions largely focused on orthotic management (14/15 studies) and physical interventions such as muscle strengthening or stretching (5/15 studies). Most studies were of high/unknown risk of bias, and the range of patient outcomes was very limited, as was reporting of treatment protocols. Similar proportions of children achieving poor radiological outcomes were found for orthotic management and physical interventions, such as physiotherapy or weightbearing alteration, compared with surgical interventions or no intervention.

Conclusion

Evidence from non-randomized studies found no robust evidence regarding the most effective non-surgical interventions for the treatment of children with Perthes' disease. Future research, employing randomized trial designs, and reporting a wider range of patient outcomes is urgently needed to inform clinical practice.

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Introduction

Perthes' disease is a condition of unknown aetiology that causes hip pain and disability in children.[1] It is most common in those aged four to nine years, and boys are four-times more likely to be affected than girls.[2] Overall, this disease affects around one per 1,200 children in the UK, but children from parents of low socioeconomic status may be disproportionately affected.[3]

The first stage of the disease is characterized by a temporary disruption in blood supply causing the femoral head to become necrotic.[4] Over time, the damaged bone is

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reabsorbed and new bone is generated.[5] The femoral head eventually heals, but during the disease process, deformity can develop, typically leading to gait disturbance, restricted mobility, pain, and reduced physical activity.[6] Occasionally the hip deformity is so severe that the child may require a total hip arthroplasty,[7] although surgery is generally only considered once skeletal maturity has been reached in late adolescence.[8]

Treatments for Perthes' disease aim to maintain the optimum local environment in and around the hip joint for self-healing to occur with minimal deformity of the femoral head.[9,10] Traditionally, non-surgical treatment options include orthotic management (e.g. braces and callipers), physical interventions such as strengthening and stretching regimes, walking aids, activity modification, or watchful waiting.[11,12] In recent decades, surgery has also often been considered.[7] In the absence of clinical guidelines, there is currently no standardized approach to treatment selection.

Given the life-long impact of Perthes' disease, the British Society for Children's Orthopaedic Surgery (BSCOS) consensus exercise, and a separate James Lind Alliance Priority Setting Partnership, have identified Perthes' disease management as one of the highest priorities for research.[13-15]

The aim of this review was to evaluate the use of any non-surgical treatment for Perthes' disease, seeking comparisons to other surgical or non-surgical interventions.

Methods

A protocol for this systematic review was registered with the international prospective register of systematic reviews (PROSPERO).[16]

Search strategy. The following electronic databases were searched from inception: Cochrane Central Register of Controlled Trials (The Cochrane Library 2019, Issue 7, July 2019); MEDLINE (1946 to July 2019) using ProQuest via the NICE HDAS interface; EMBASE (1974 to July 2019) using 'disease' and 'physical therapy'. Reference lists of potentially eligible studies were reviewed, and citation tracking was used to identify additional studies.

Study selection criteria. Studies designed to compare the effects of a non-surgical intervention with a comparator group were eligible for inclusion. This could include experimental designs - i.e. controlled clinical trials (randomized, quasi-randomized, or non-randomized allocation) or longitudinal observational studies (cohort studies). Systematic reviews, cross-sectional studies with reporting restricted to post-surgical outcomes, case-control studies, and 'before and after' observational studies were excluded.

Eligible studies recruited children aged 16 years and under with a radiologically-confirmed diagnosis of Perthes' disease. Participants were treated with a non-surgical intervention, including physical interventions such as physiotherapy or weightbearing modification, or management with an orthotic device. Studies were excluded if an English full-text version was not available.

Screening. One author (AG) screened all titles and selected an initial 'long list' of potentially eligible studies. These abstracts were independently reviewed by two authors (AG, TVH) to confirm potential eligibility, with any discrepancies adjudicated by a third reviewer (CC). Full-text articles were obtained for all short-listed studies and reviewed for eligibility by two authors (AG, TVH).

Data extraction. A standardized data extraction proforma was used to extract data (AG) from eligible studies, including year of publication, country of origin, study design, duration of follow-up period, and sample size available for analysis. Participant details that were extracted include the number of children recruited and completing each follow-up, number of hips (to account for cases of bilateral Perthes' disease), and age (range) at onset or diagnosis of Perthes' disease.

Radiological, functional, and patient-reported outcomes were extracted when reported. The primary outcome was the post-intervention radiological shape of the hip when the participant had reached skeletal maturity. Radiological shape was categorized using the Stulberg[6] and/or Mose[17] classification methods for Perthes' disease that are commonly used in practice. Event rate data were extracted, with an event defined as a Stulberg rating of '4 or 5' (indicating a poor outcome) and/or categorized by the authors as 'poor' using the Mose method of classification, foor which outcomes are 'poor' when there is a variation of more than 2 mm when assessing the sphericity of the femoral head.[17] To standardize data for comparison across studies, event rates were calculated as the proportion of children with a poor outcome, over the total number of children in the intervention group. Using the frequency data extracted from the papers, two-way tests for differences in proportions for independent groups were calculated ($\alpha = 0.05$) using the immediate commands in Stata v15 (StataCorp, College Station, Texas, USA).

Secondary outcome data included objective measures of function, such as goniometer measures of range of movement (ROM) at the hip joint, lower limb muscle strength measured on the Oxford scale,[18] and gait quality scores (e.g. the "12-minute walk" or the presence of a Trendelenburg sign). For these functional measures, the differences between the two limbs (affected and unaffected) were analyzed. We also extracted any available patient-reported health related quality of life outcomes.

Methodological quality assessment. Although randomized controlled trials were eligible for inclusion, no such studies were identified, so the Cochrane Risk of Bias criteria tool was not used.[19] Instead, the Newcastle

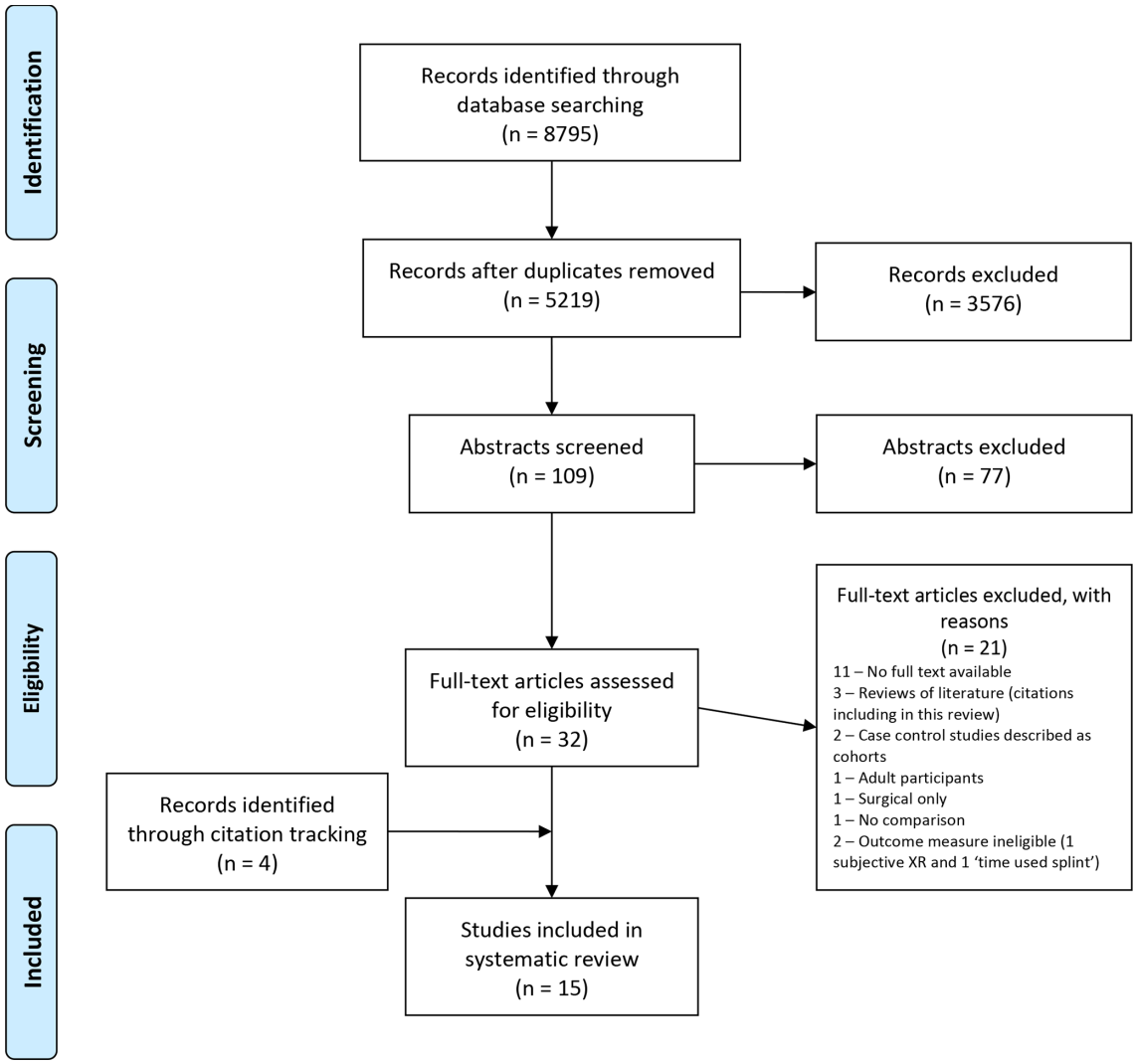

Fig. 1
PRISMA flow chart showing literature search process.

Ottawa Scale (NOS), designed to assess quality and risk of bias in non-randomized studies,[20,21] was applied. The NOS uses a points system to judge three domains: selection, comparability, and outcome, with a maximum score of eight points. A risk of bias score is then allocated according to the overall number of points, categorized as 'high' (0 to 3 points), 'moderate' (4 or 5 points), or 'low' (6 to 8 points).[22] One author (AG) scored each study, while a second author (TVH) independently verified the scoring.

To assess the quality of reporting of the study interventions, the Template for Intervention Description and Replication (TIDieR) checklist was used, with data extracted by one author (AG). This 12-point scale has a maximum of two points available for each category, with an overall potential score of 24. Higher scores indicate that the quality of reporting is more likely to aid implementation or replication of the interventions.[20] The TIDieR tool is becoming a widely recognized measure of the completeness of intervention reporting within studies.[23,24]

Analysis. A narrative synthesis was undertaken because studies were deemed too clinically and/or methodological heterogeneous for statistical pooling of data. The first stage of analysis was to develop a typology of the non-surgical interventions evaluated, with each intervention classified as either a physical intervention or orthotic management. The results were then stratified by the type of interventions being compared. For example, we grouped studies comparing orthotic management with another non-surgical intervention separately from studies comparing orthotic management to a surgical intervention comparator. Data with similar outcomes were then synthesized within each of these groups.

Results

Study selection. Electronic database searches identified 8,795 records (including duplicates). A PRISMA diagram of the review process is provided in Figure 1.[25] After screening titles and abstracts, 32 full-texts were considered, and ultimately 15 studies were included in the review (Table I). Orthotic management includes any orthoses, calliper, or casting used. Comparators could include another non-surgical intervention or a surgical intervention (i.e. femoral or pelvic osteotomies, or surgery to the soft tissue or muscular/tendons). Physical interventions include treatments such as stretching, both active and passive, strengthening, and also treatments such as weightbearing modification.

Quality assessment. NOS risk of bias scores ranged from 3 to 8 out of 8 (Table I) with 1/15 assessed as high risk, 8/15 assessed as moderate risk, and 6/15 as low risk. The most common quality issues identified were inadequate description of follow-up and failure to consider or control for potential confounders. Interventions were generally poorly described, with TIDieR scores ranging from 6 to 14 out of 24 (Table I). Study scores were reduced because they failed to report intervention procedures including materials, dosage and who carried out the intervention. A more detailed description of how the NOS and TIDieR scores are categorized is presented in supplementary Tables 1 and 2.

Characteristics of included studies. All 15 included studies recruited children with a radiological diagnosis of Perthes' disease, providing a total of 1745 participants (Table I).[26-40] Sample sizes ranged from 17 children[28] to 337 children,[38] and the age of children at onset of symptoms ranged from one to 15 years old. One study followed children up for 12 weeks.[28] Otherwise, children were followed up for a minimum of one year[33] and a maximum of 33 years.[26]

In terms of study design, there were two multicentre prospective cohort studies,[34,40] six single-centre prospective cohort studies,[28,31,33,35,36,39] one multicentre retrospective cohort study[30] and six retrospective cohort studies.[26,27,29,32,37,38]

Interventions compared. A range of non-surgical interventions were evaluated within the 15 studies and 13 included a surgical comparison group.[27,29-40] With regards to non-surgical interventions, four studies included an intervention group that consisted of 'active observation.'[26,28,34,37]

Orthotic management (including callipers, braces, casts and any other orthoses) was evaluated in 14/15 studies.[26,27,29-40] Physical interventions (including strengthening exercises, stretching exercises, and/or mobility adaptation such as altered weightbearing statuses and balance work) were reported in 5/15 studies.[28,30,38-40] One study reported that a group was managed 'conservatively with theatre if at risk', but the conservative management protocol was not described.[35]

Four studies included at least one comparator intervention comprising multiple treatment components.[26,27,33,36] Two of these studies combined either a brace[26] or a cast after surgical intervention (tenotomy)[27] with physiotherapy if clinically indicated. A third study compared two groups with traction and either calliper application or surgery.[33] A fourth included two multicomponent groups: traction and orthosis; and surgery followed by physiotherapy.[36]

Primary outcomes. Six studies applied the Stulberg classification[26,27,29,34,39] and four studies used the Mose classification,[31,32,36,37] with two studies measuring both these radiological outcomes.[36,37] Two of the studies reported a 'modified' Stulberg score which combined Stulberg categories 1 and 2 to create a new category for a favourable outcome, and combined Stulberg categories 4 and 5 to create a new category for a poor outcome. Category 3 remained unchanged, equating to a 'fair/moderate' outcome.[26,38] Five other studies reported relevant interventions but without using objective radiological outcome measures that could be synthesized (data not reported).[28,33,35,36,38] Two papers did not report the primary outcomes; however, given the relevance of the study to this review they were included.[36,38]

Secondary outcomes. Functional outcomes were reported in four studies: two reported ROM outcomes,[28,32] one described a muscle strength outcome (Oxford scale),[28] and one study had used the 12-minute walk test to assess gait quality in children pre- and post-intervention.[33] Only one study reported patient health-related quality of life, but the authors did not state what questionnaire was used or previously validated in this population.[29] The authors also failed to give details on when it was used, stating 'at clinical assessment each patient answered a questionnaire', and they provided no more information as to how frequently these clinical assessments took place.

Studies comparing orthotic management with no intervention or surgery. The primary outcomes (Stulberg and/or Mose score) for studies comparing orthotic management with the comparator of no intervention (three studies[26,34,37]) or surgical repair (nine studies[27,29,30,32,34,37-40]) are presented in Tables II and III. Although the treatment protocol in all of these studies mandated orthotic management, two studies also provided supplementary physiotherapy input 'if needed'; 47%[26] and 40%[27] of children received supplementary physiotherapy input respectively. Comparing orthotic management with no intervention, all studies reported similar proportions of children with poor radiological outcomes between groups.[26,34,37] When orthotic management was compared with surgical intervention, findings across the nine studies included in our review were inconsistent. One study that compared surgery with two different types of orthosis treatments[39] reported a greater proportion of children with favourable outcomes for children managed with a Petrie cast orthosis

Table I. Summary of studies included in review.

Author(s)	Study design (length of follow-up)	Setting	Children, hips (n)	Age at onset (range)	Intervention(s)and treating clinician*	Comparator	Primary outcome(s)	Secondary outcomes	NOS score	TIDieR score
Askoy 2004 Turkey[26]	Retrospective cohort (10 to 33 years)	Not reported	48 (51) Int: 23 (23) Comp: 25 (28)	6 to 9 years	Braced† (mean time 14 months; range 12 to 18 months)	Non-braced	Stulberg	N/A	4	11
Arkader 2008 Brazil[27]	Retrospective cohort (5 to 22 years)	Not reported	43 (43]) Int: 22 (22) Comp: 22 (22)	9 to 14 years	Multi-component (Orthotic (brace) with physiotherapy if needed) Treating clinician - physical therapist for input.	Surgical (FVO or Chiari osteotomy)	Stulberg	N/A	4	6
Brech 2006 Brazil[28]	Prospective cohort (12 weeks)	Physiotherapy clinic	17 (17) Int: 8 (8) Comp: 9 (9)	3 to 8 years	Physiotherapy (Stretches, strengthening and balance - 12 weeks) Treating clinician - Physiotherapist	Active observation	N/A	ROM	7	13
Citlak 2012 Turkey[29]	Retrospective cohort (8 to 25 years)	Not reported	25 (27) Int: 14 (16) Comp: 11 (11)	6 to 8 years	Orthotic (Thomas splint) (mean time 16.3 months; range 4 to 24 months).	Surgical (FO)	Stulberg	IOWA Scale questionnaire	4	8
Cooperman 1984 Sweden, USA, Yugoslavia, Israel[30]	Multicentre retrospective cohort (3 to 25 years)	Not reported (hospital names provided)	248 (248) Int1: 48 (48) Int2: 58 (58) Int3: 72 (72) Comp: 70 (70)	2 to 15 years	Int1: Orthotic (NAO) Int2: Orthotic (SRO) Int3: Crutches (no data on treatment time)	Surgical (FO)	Subjective radiograph review	N/A	4	9
Edvarson 1981 Norway[31]	Prospective cohort (3 to 13 years)	Not reported (hospital)	58 (63) Int: 28 (32) Comp: 30 (31)	2 to 11 years	Bed rest/sling (Bed rest mean 9 months, sling mean 18 months, no ranges reported)	Surgical (VO)	Mose	Gait assessment (Trendel-enburg)	3	13
Evans 1988 USA[32]	Retrospective cohort (4 to 10 years)	Not reported (hospital)	36 (36] Int: 17 (17) Comp: 19 (19)	5 to 12 years	Orthotic (NAO) (range 8 to 42 months)	Surgical (VDO)	Mose	ROM	4	11
Fulford 1993 Scotland[33]	Prospective cohort (3 to 6 years)	Paediatric surgical unit	94 (99) Int: 42 (43) Comp: 52 (56)	4 to 10 years	Traction (6 weeks) followed by orthotic (abduction calliper) (Mean 20 months, range 8 to 32 months)	Traction followed by surgery (osteotomy and plate insertion)	No Stulberg or Most (used Caterall)	12 min walk	4	14
Herring 2004 USA[34]	Multicentre prospective cohort (1 to 15 years)	Not reported	337 (345) Int1: (129) Int2: (27) Int3: (19) Comp: 120 (no. of children not given for each intervention group)	6 to 12 years	Int1: Orthotic (SRO) (no timeframes reported but instruction to wear until radiological re-ossification) Int2: ROM exercises (standing abduction stretch and active ROM once a day, no duration of regime reported) Int3: No treatment (symptom relief only)	Surgery (FO or IO; 52 and 68 respectively)	Stulberg	Modified Pillar Classi-fication	7	12

Continued

Table I. Continued

Author(s)	Study design (length of follow-up)	Setting	Children, hips (n)	Age at onset (range)	Intervention(s)and treating clinician*	Comparator	Primary outcome(s)	Secondary outcomes	NOS score	TIDieR score
Jani 1980 Switzerland[35]	Prospective cohort (follow-up data missing)‡	Not reported	83 (83) Int1: 19 (19) Int2: 40 (40) Comp: 24 (24)	Age not reported	Int1: Traction and orthosis (orthosis not described, or regime reported) Treating clinician – not specified Int2: Conservative (Int1) with surgery if symptoms develop.	Surgery (VO) with Physiotherapy post-op (regime not reported)	No Stulberg or most (used Caterall)	N/A	7	6
Marklund 1976 Sweden[36]	Prospective cohort (follow-up data missing)§	Not reported (hospital)	47 (49) Int: 22 (23) Comp: 25 (26)	2 to 11 years	Conservative management; bed rest ± traction and Thomas splint until radiological re-ossification was observed (range 1 to 3 years)	Surgical (sub-trochanteric osteotomy)	Subjective radiograph review	N/A	4	7
Osman 2009 Scotland[37]	Retrospective cohort (2 to 20 years)	Not reported (hospital)	44 (48) Int1: 12 Int2: 14 Comp: 22 (no. of children not given for each intervention group)	8 to 14 years	Int1: Orthotic (abduction cast) Int2: No treatment (no regimes reported)	Surgical (FVO or Shelf osteotomy; 4 and 18 respectively)	Stulberg Mose	N/A	5	9
Poussa 1993 Finland[38]	Retrospective cohort (2 to 16 years)	Not reported (hospital)	218 (232) Int: 96 (116) Comp: 112 (126)	5 to 13 years	Conservative; orthosis (Thomas splint) or crutches (no time frame specified)	Surgical (FO)	Subjective radiograph review	N/A	6	6
Wang 1995 USA[39]	Prospective cohort (2 to 27 years)	Not reported	124 (141) Int1: 38 (41) Int2: 38 (41) Int3: 23 (29) Comp: 25 (30)	2 to 12 years	Int1: Orthotic (SRO) (Mean time 8 months, range 1 to 20 months) Int2: NWB and exercises (Included bedrest, crutches, or callipers to prevent weightbearing on the affected hip, no regime reported) Int3: Orthotic (Petrie cast) (no regime reported)	Surgical (FVO or Salter osteotomy; 15 each)	Stulberg Mose	N/A	8	10

Continued

Table I. Continued

Author(s)	Study design (length of follow-up)	Setting	Children, hips (n)	Age at onset (range)	Intervention(s)and treating clinician*	Comparator	Primary outcome(s)	Secondary outcomes	NOS score	TIDieR score
Wiig 2008 Norway[40]	Multicentre prospective cohort (5 years)	Not reported	323 (323) Int1: 27 (27) Int2: 220 (220) Comp: 76 (76)	1 to 15 years	Int1: Orthotic (SRO) (No timeframes given) Int2: Physiotherapy (range of movement exercises with special emphasis on abduction, internal rotation and extension, in addition to muscle strengthening exercises)	Surgical (FVO)	Modified 2-group Stulberg	N/A	7	10

*Treating clinician not specified unless listed.
†Brace not specified other than 'trilateral socket hip abduction orthosis'.
‡rReported "until disease process complete".
§Reported as "primary end result".
¶Reported that "all were mature at follow-up".
AHI, acetabular head index; ATD, articulotrochanteric distance; Comp, comparison; FO, femoral osteotomy; Int, intervention; IO, innominate osteotomy; NAO, Newington abduction orthosis; NOS, Newcastle Ottawa Scale; ROM, range of movement; SAR, slope of acetabular roof; SRO, Scottish Rite orthosis; TIDieR, template for intervention description and replication; VDO, varus derotational osteotomy; VO, varus osteotomy.

Table II. Physical and orthotic interventions: Primary outcome assessed using Stulberg method.

Author	Children, hips (n)	Intervention	Intervention, n (%)*	Control†	Control, n (%)*	p-value‡
Studies testing orthotic and/or physical interventions as either a multi-component intervention, or in separate intervention groups						
Askoy 2004 Turkey[26]	48 (51)	Orthotics (brace), PT†	3/23 (13.0)	None, PT†	6/28 (21.4)	0.43
Arkader 2008 Brazil[27]	43 (43)	Orthotic (brace), PT (if needed)	6/21 (28.6)	Surgical	3/22 (13.6)	0.23
Herring 2004 USA[34]	337 (345)	Orthotics (brace)	22/129 (17.1)	None Surgical	3/19 (15.8) 12/120 (10.0)	0.89 0.53
		PT (ROM)	16/77 (20.8)	None Surgical		0.62 0.03
Wang 1995 USA[39]	124 (141)	Orthotics (SRO)	8/41 (19.5)	Surgical	6/30 (20.0)	0.96
		PT (NWB exercise)†	7/41 (17.1)	Surgical		0.76
		Petrie cast	1/41 (2.4)	Surgical		0.01
Wiig 2008 Norway[40]	323 (323)	Orthotics	13/13 (27.7)	Surgical	10/93 (10.8)	0.01
		PT	37/174 (21.3)	Surgical		0.03
Studies testing orthotic interventions only						
Citlak 2012 Turkey[29]	25 (27)	Orthotics (Thomas splint)	2/16 (12.5)	Surgical	0/11 (0.0)	0.22
Osman 2009 Scotland[37]	44 (48)	Orthotic (abduction cast)	3/12 (25.0)	None Surgical	8/14 (57.1) 2/22 (9.1)	0.10

*Number achieving a Stulberg score of 4 or 5 indicating a poor radiological outcome.
†Two control group tested: no intervention (none) and surgery intervention.
‡p-values calculated by the review team from event rate data extracted from the paper.
NWB, non-weightbearing; PT, physiotherapy; ROM, range of movement; SRO, Scottish Rite orthosis.

Table III. Orthotic interventions: Primary outcome assessed using Mose method.

Author	Children, hips (n)	Intervention	Intervention, n (%)*	Control†	Control, n (%)*	p-value‡
Edvarson 1981 Norway[31]	58 (63)	Bed rest/sling	6/32 (18.8)	Surgical	7/31 (22.6)	0.71
Evans 1988 USA[32]	36 (36)	Orthotics (Newton Abduction Orthosis)	3/17 (17.6)	Surgical	5/19 (26.3)	0.53
Osman 2009 Scotland[37]	44 (48)	Orthotic (abduction cast)	5/12 (42.0)	None Surgical	11/14 (78.6) 13/22 (59.1)	0.06 0.34

*Number achieving a 'Poor' Mose score indicating a poor radiological outcome.
†Two control group tested: no intervention (none) and surgery intervention.
‡p-values calculated by the review team from data presented in the paper.

compared to those undergoing surgery ($p < 0.05$), but no difference between surgery and Scottish Rite orthosis management. In contrast, a later study[40] reported a greater proportion of children with poor radiological outcomes after orthotic management compared to surgical treatment ($p < 0.05$). The remaining six studies[27,29,31,32,34,37] found no between group differences.

There were no statistically significant between-group differences in any secondary outcomes (range of movement, gait disturbance, or quality of gait) reported in studies comparing orthotic management to surgery.[29,31-33]

Studies comparing physical intervention with no intervention or surgery. The primary outcome findings (Stulberg score) for studies comparing physical interventions (such as ROM exercises, or 'physiotherapy') with a comparator of no intervention[34] or surgical repair[39,40]) were also inconsistent (Table II). While reporting no between-group differences when comparing physical interventions with no intervention, Wiig et al[40] reported a higher proportion of children with poor radiological outcomes among those receiving physiotherapy intervention compared with those undergoing surgery ($p < 0.05$). In contrast, the proportions of children with poor outcomes were similar in the remaining two studies that provided ROM exercises[34] or a multicomponent intervention comprising weight-bearing modification and exercises.[39]

Regarding secondary outcomes, Brech et al[28] studied 17 children (eight physiotherapy, nine no intervention) treated with a physiotherapy regime consisting of stretching, strengthening and balance work compared with those receiving active observation (no details specified). Measures of ROM and strength were greater in the

physiotherapy group compared to those receiving no intervention (all $p < 0.05$).

Relationship between age and treatment effects. Several studies investigated the relationship between the age of intervening and clinical outcomes. In three studies, children treated under the age of 12 years were reported to have improved radiological outcomes when treated with non-surgical intervention (orthotic and physical interventions)[30] compared with those aged 12 years or over at the time of intervention (data not presented).[37,40] Cooperman et al[30] compared two orthotic interventions (Scottish rite orthosis and Newington abduction orthosis) with the use of crutches and surgery. No difference in outcome between the four methods was reported for children under the age of 12 years, although a higher proportion of children over 12 years using crutches had poor Stulberg outcomes when compared to other groups.[28] The lateral pillar classification, a radiological assessment of the hip joint used by Herring et al[34] indicated that there were no significant differences between children who had surgery and those who underwent physical interventions (ROM exercises) or orthotic management. However, these authors did report that a larger proportion of children were in the favourable Stulberg 1 or 2 category when compared with no treatment (40% overall compared with 26% for no treatment); when adjusted for patients under the age of 8 years old this proportion increased to 48% for ROM exercises.[37] Wiig et al[40] concluded that in children aged less than six years, physiotherapy resulted in the highest number of children in the favourable Stulberg 1 or 2 category compared to orthotic or surgical intervention. They also reported there was a higher proportion of children with femoral head necrosis (over 50%) aged six years or older.

Discussion

Main findings. In this review, we found no high-quality evidence to suggest that specific types of orthotic management or physical interventions, either alone or in combination, were constituently associated with improved radiological outcomes when compared with alternative treatment strategies. Children achieving poor radiological outcomes ranged from 13% to 42% with orthotic interventions, 17% to 21% for those receiving physical interventions and 0% to 59% for surgical interventions. Although 15 studies met our inclusion criteria, none were randomized controlled trials. Furthermore, the quality of these studies was variable with most being at unknown or high risk of bias. The evidence available was difficult to synthesize due to heterogeneous designs and comparator interventions and limited reporting of treatment protocols for the interventions tested.

While conclusions are limited regarding effectiveness of interventions, a number of studies did reveal interesting differences in treatment response associated with the age of the child at time of diagnosis and intervention. Three studies reported better radiological outcomes associated with both surgical and non-surgical interventions in younger children.[28,34,40] Findings reported in the wider existing literature suggest that surgical interventions on the other hand may lead to better outcomes when performed in older children.[41,42] These differences in treatment response might be explained by the differing structural changes in those older children (aged > 5/6 years), such as loss of hip joint congruence leading to increased risk of femoral head deformation.[41]

Findings in context. This is the first systematic review to focus primarily on non-surgical treatments of Perthes' disease. A previous meta-analysis by Nguyen et al[43] analyzed the radiological outcomes of children with Perthes' disease following surgical intervention, compared with other surgical methods as well as some non-surgical approaches. This review concluded that for children aged under six years, there was no difference in radiological outcomes between different treatment approaches. Those older than six years in this review were all treated surgically, and outcomes appeared similar regardless of surgical technique. Since the completion of this meta-analysis, only one additional publication has been identified that assesses the effectiveness of non-surgical treatment.[29]

In the UK, a survey of members of BSCOS reported that 90% of clinicians refer children with Perthes' disease to physiotherapy services.[44] One of the aims of treatment delivered in physiotherapy services is to maintain mobility of the hip joint based on evidence, suggesting more favourable outcomes in children with preserved hip ROM.[45] Our review has highlighted the limited evidence base for this treatment approach; only four studies[27,28,34,40] tested a physical therapy intervention, and the findings were inconclusive.

Study strengths and limitations. This review includes the assessment of 1,805 hips in 1,745 children, who were followed-up for a range of 12 weeks to 33 years. All but two studies[26,28] reported follow-up until the point of skeletal maturity, which is important in the management of Perthes' disease to ensure disease process completion.[4]

While a major strength of this review is that it summarizes the available evidence on non-surgical treatment options for Perthes' disease, it is limited by the lack of robust evidence. Another limitation is that it was not possible to pool results for statistical analysis due to the heterogeneity in methodology and in non-surgical interventions evaluated within our broad categories of orthotic or physical management. To aid narrative synthesis, significance levels were calculated to aid the reader's interpretation of the radiological outcomes; however, as no adjustments for baseline characteristics could be made and these findings should be treated with caution. Finally, it was not possible to explore the impact

of interventions upon outcomes likely to be important to children and their families and carers, such as function and health-related quality of life, as the majority of studies limited their reporting to radiological and clinical outcome measures. A core outcome set (COS) for Perthes' disease has been created, which defines a much wider standardized set of outcomes that are important when measuring the success of interventions.[46] The use of this COS will allow standardization in clinical outcomes that can, in turn, support decision making for treatments in this patient population.

This review demonstrates a lack of evidence regarding the effectiveness of treatments for Perthes' disease, such that no recommendations can be made regarding the use of any non-surgical intervention compared to other non-surgical or surgical interventions. Future research must employ high-quality randomized trials to inform clinical practice. This research should not only include radiological outcomes, but should seek to include patient-important outcomes, such as pain and functional recovery, that make up the COS.

Take home message

- Evidence from non-randomized studies found no robust evidence regarding the most effective non-surgical interventions for the treatment of children with Perthes' disease.
- Future research, employing randomized trial designs, and reporting a wider range of patient outcomes, is urgently needed to inform clinical practice.

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Supplementary material

Tables showing the detailed Newcastle Ottawa Score and TIDieR checklist.

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