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REVIEW

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Systematic review: exercise training for equinus deformity in children with cerebral palsy

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Abstract

Background: Children with spastic cerebral palsy have motor deficits that can lead to joint contractures. Ankle equinus deformity is the most common foot deformity among children with CP. It is caused by spasticity and muscular imbalance in the gastrocnemius-soleus complex. Exercise enhances ankle function, improves gait in children with CP, and prevents permanent impairment. Therefore, there is a need to investigate the effectiveness of different types of exercise used in equine management. The aim of this review is to assess the evidence of the effectiveness of exercise training on equinus deformity in children with cerebral palsy.

Methodology: The American Academy for Cerebral Palsy and Developmental Medicine and Preferred Reporting Items for Systematic Reviews and Meta-Analyses methodology were used to conduct this systematic review. Four databases (PubMed, Cochrane Library, Physiotherapy Evidence Database (PEDro), and Google Scholar) were searched till January 2022 using predefined terms by two independent reviewers. Randomized controlled trials published in English were included. This review included seven studies with 203 participants ranging in age from 5 to 18 years. Methodological quality was assessed using AACPDM, PEDro scale; also, levels of evidence adopted from modified Sacket's scale were used for each study. Primary outcomes were dorsiflexion angle, plantar flexion angle, and plantar flexors strength.

Results: The quality of studies ranged from good (six studies) to fair (one study). The level of evidence was level 1 (six studies) and level 2 (one study) on modified Sacket's scale. There is a low risk of bias in the included studies. Meta-analysis revealed a non-significant difference in plantar flexor strength, plantar flexion angle, and dorsiflexion angle between the study and control group.

Conclusions: There is a need for high-quality studies to draw a clear conclusion as the current level of evidence supporting the effectiveness of various types of exercises on equinus deformity in children with cerebral palsy is still weak.

Keywords: Cerebral palsy, Exercise, Treatment, Foot deformity, Equines, Systematic review, Children

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Background

Systematic reviews use a structured and pre-defined procedure that requires the use of methodological approaches (comprehensive methods) to ensure that the results are both reliable and purposeful to the patients [1]. These reviews are widely used to inform the development of consistently reliable clinical guidelines [2–4], and they may be considered the best evidence-based healthcare [1]. A systematic review, according to the Cochrane handbook, uses explicit, systematic methods to minimize bias, thus providing more trustworthy findings from which conclusions can be reached and decisions can be made [5].

Cerebral palsy (CP) is defined as "a group of developmental disorders of movement and posture that cause functional limitation or disability associated with disturbances occurring in the foetal or infant brain". The motor impairment may include a seizure disorder and also affection of sensation, cognition, communication, and/or behaviour, in addition to secondary musculoskeletal disorders that may also exist [6]. It is commonly caused by a brain injury that occurs prior to or during birth, which is the major contributing factor. It can also happen between the ages of 3 and 5 years [7]. Cerebral palsy is a complex disease resulting from injury to the developing brain rather than a single illness [8]. The severity as well as the patterns of motor involvement and associated impairments such as those of communication, intellectual ability, and epilepsy varies greatly [7]. The mixture of postural and gait abnormalities is one of the most noticeable characteristics found in children with CP [9, 10]. Paresis and spasticity, which are caused by a lesion in the upper motor neuron, are significant contributors to the motor impairments and can lead to the development of joint contractures, which are dynamic at first but can become fixed joint deformities if left untreated. Equinus is often associated with spasticity in the gastrosoleus muscle complex (GSC) [11].

Ankle equinus deformity, which is the inability to dorsiflex the foot beyond the plantigrade level, or neutral [12, 13], is the most common foot deformity among children with CP [14, 15]. Patients with equinus have a lack of ankle dorsiflexion and toe-walking pattern, which causes ankle instability, the risk of falling, dysfunction, and consequent joint deformity [16–18]. Spasticity and muscle imbalance, with a strong dominance of the gastrocnemius-soleus complex, cause equinus deformity. Experimental evidence suggests that spastic muscles grow at a slower rate than muscles and bones in their normal state, supporting the development of contracture [19]. Since this deformity is initially dynamic, stretching and orthosis may be effective treatments.

Exercise and muscle tone management are both significant treatment strategies for improving ankle function [20]. Strength training is an effective method to reduce muscle weakness, which is the most common consequence of CP, while also improving functional ability [21, 22]. Strengthening programmes targeting lower extremities play a critical role in improving the functional abilities such as walking [21, 23, 24] and the Gross motor function classification system (GMFCS) scoring after strength training completion [21]. Stretching, on the other hand, improves the extensibility of the muscle and helps to avoid or postpone the need for orthopaedic interventions. Furthermore, stretching is important in preventing permanent muscle or joint shortening (muscle contractures, for example) as well as reducing joint stiffness and decreasing functional movement [25].

Therapeutic interventions such as stretching and active movement training goal are enhancing ankle function and improving gait in children with CP [26]. The aim of this study is to conduct a systematic review of the quality of evidence regarding the efficacy of several types of exercises used in the management of equine deformity in children with cerebral palsy.

Main text

Methods

Two reviewers conducted an electronic search up to January 2022. To find relevant published studies, the following databases were searched: the Cochrane Library, the Physiotherapy Evidence Database (PEDro), PubMed, and Google Scholar. The following keywords were used to search those databases: "Cerebral palsy", "equinus gait", " gastrocnemius tightness", "dorsiflexor weakness", "equinus deformity", "equinus contracture", "Talipes Equinus", "exercise therapy", "Resistance Training", " Physical Activity", "exercise fitness", "Exercise Movement Technique". The Cochrane Handbook for Systematic Reviews of Interventions [5], the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) methodology for developing systematic reviews of treatment interventions [27], and the Preferred Reporting Items for Systematic Reviews (PRISMA) guidelines [28] were used to conduct this systematic review. All potentially eligible articles were obtained in full text, and reference lists from eligible studies were also screened.

Eligibility criteria

The following criteria were used to determine whether studies were eligible:

- *Study design*: randomized controlled trials (RCT)

- *Participants*: children with various forms of CP from both sexes; their age is up to 18 years old
- *Intervention*: exercise training (strengthening exercises, stretching exercises, or a combination of strengthening and stretching exercises)
- *Outcomes*: dorsiflexion angle, plantar flexion angle, plantar flexors strength
- *Language*: available English full-text studies

Criteria for exclusion

- Studies that have not yet been published
- Non-RCT study designs, such as narrative reviews, case reports or series, observational studies, and conference proceedings
- Research that investigated outcomes that were unrelated to our scope
- Studies that combined exercise training with other modalities

Data extraction

Two reviewers filled the data extraction sheet developed by the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) [27] to extract relevant information or data, such as the following: (a) the author and year of publication; (b) population information, including the number of children by diagnosis and age; (c) study design; (d) methodology; (e) measured outcomes; and (f) results. When compared to a similar tool, the inter-rater reliability and convergent validity of the AACPDM's evidence and study quality ratings for group design studies are acceptable [28].

Methodological quality assessment

The current systematic review's methodological quality was assessed using the AACPDM rating system of quality assessment [27] and the PEDro scale. PEDro is a reliable valid indicator of clinical trial methodological quality [29]. The internal validity of the included studies was assessed through PEDro scale criteria of adequate randomization; allocation concealment; blinding of participants, therapists, and assessor; incomplete outcome data; similar baseline; and use of intention-to-treat analysis. The items are scored as either present (1) or absent (0). To reach the final decision, the methodological quality of the included studies was independently assessed by two reviewers, and discrepancies were resolved by consulting with a third reviewer. After categorizing each item as "present" or "absent", the total score of each study was calculated as the sum of "present" responses. Scores range from 0 to 10, with higher scores indicating higher RCT methodological quality. The individual study's quality would be rated as excellent (score 9–10), good (score 6–8), fair (score 4–5), or poor (score 3) [30].

The AACPDM questions include clear eligibility criteria, reliable measures of outcome, blinding of assessors, power calculations, and controlling for other sources of bias (for example, unclear type of random sequence generation or allocation concealment, insufficient follow-up, lack of intention to treat). Each question should be answered with "yes" (criterion/criteria present) or "no" (criterion/criteria absent). Individual studies' quality would be assessed as strong ("yes" on 6–7 of the questions), moderate (scoring 4 or 5), or weak (score 3) for group studies [27].

Level of evidence

The modified Sackett scale [31] was used to assess the level of evidence in all included studies. The modified Sackett scale has five levels of evidence, which are as follows: level 1 if the study is RCT (PEDro score ≥ 6); level 2 if the study is RCT (PEDro score < 6), prospective controlled trial or cohort; level 3 if the study is case-control; level 4 if the study is one-group pretest-posttest or case series; and level 5 if the study is a case report. The strength of the evidence for the intervention was determined using this 5-level scale.

Assessment of risk of bias of included studies

Bias is defined as a systematic error in results or inferences. Biases can work in both ways: different biases can lead to an underestimation or overestimation of the true intervention effect. Biases can range in intensity. It is usually impossible to determine the extent to which biases influenced the findings of a particular study. The domains that represent the risk of bias in the current systematic review are random sequence generation, allocation concealment, selective reporting, other sources of bias, blinding (participants and personnel and outcome assessment), and incomplete outcome data [32].

Data analysis

If two or more published studies were similar in terms of intervention, patient demographics, outcome measures, and adequate quality, data were statistically summarized. Data from homogenous studies were analysed using Review Manager (RevMan – version 5.4.1, The Nordic Cochrane Center, The Cochrane Collaboration, Copenhagen, Denmark, 2021). A formal meta-analysis was conducted for all outcomes if the data were sufficient. Pooled data were expressed as the mean difference (MD) with 95% CI. Meta-analyses including studies with different scales were summarized using the standardized mean difference (SMD) with the 95% CI. For the studies reporting the mean and 95% CI, the SD of the paired difference was calculated in 2 steps: (1) dividing the confidence interval (CI) by the t constant according to the degree of freedom that depends on the sample size to get the standard error of the mean (SEM), then (2) applying the equation SD = SE × SQRT (n) to calculate the SD. The heterogeneity test (I^2) helps to assess whether there are differences between the studies included in the meta-analysis [33–35]. If the I^2 value is $<$ 50%, and the test P value is $>$ 0.05, so the studies can be considered homogenous. Between-study statistical heterogeneity was explored and quantified using the I^2 test. By default, the fixed-effect model was used in all analyses. The 2-sided statistical analysis testing was considered the α-error level at 0.05.

Results

Literature search

The search strategy revealed 5170 articles from the previously mentioned databases as follows: Cochrane Library (13), PEDro (1), PubMed (16), and Google Scholar (5140). There were duplicate articles (9). The remaining 5161 articles, titles, and abstracts were independently screened by the reviewers. Thirteen articles were filtered based on full text; as shown in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow chart (PRISMA) (Fig. 1) [36], 5148 articles were excluded because they were outside the scope, used different therapies, the children's diagnosis was not CP, or the outcome of interest was absent. The remaining seven studies made the basis for the current systematic review.

This systematic review includes meta-analysis for five studies and descriptive analysis for two studies because of the difference in terms of intervention.

Study characteristics

In terms of general participant characteristics, outcome measures, and evaluation procedures, there was some homogeneity among the studies included (Table 1). As a result, these studies underwent meta-analysis.

Participant characteristics

Two hundred and three children ranged from 5 to 18 years of age were diagnosed with spastic cerebral palsy with [GMFCS] levels I–III [37, 38, 40–42] and [GMFCS] levels I and II [39, 42, 43]. One study specifies CP type which is diplegic [42].

Intervention

All studies investigated the effect of different types of exercise training on equine deformity in children with cerebral palsy (strengthening exercise [37, 39, 41–43], stretching exercise [39, 40], combined strengthening and stretching exercise [38]).

Types of outcomes measured

Dorsiflexion angle [39, 43], plantar flexion angle [41, 43], and plantar flexors strength [37, 41–43].

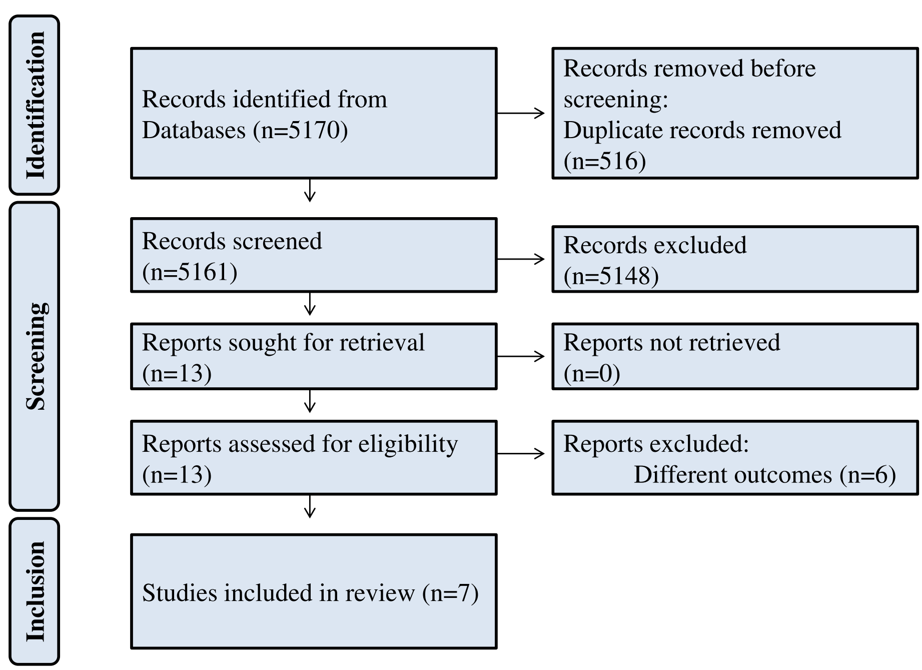

Fig. 1 PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flowchart

Table 1 Participant, diagnosis, intervention, duration, outcome measures, and results

Study	Participant	Diagnosis	Intervention	Duration	Outcome measure	Results
Rayn et al. [37]	$N = 64$ (Exp.gp = 33, Con.gp = 31) Age (yrs:m) mean (±SD): Exp.gp= 13:6 (2:7), Con.gp= 13:11 (2:6)	Spastic CP, ([GMFCS] levels I–III)	**Resistance group:** ankle plantar flexors (primarily gastrocnemius) exercises Over a 10-week period, the intensity of exercise increased progressively according to a standardized programme, with sets increasing from 4 to 8. Each session's resistance was increased until the fatigue level was reached, indicating that the exercise was being performed at the recommended intensity.	Completed 10 supervised sessions and 20 home sessions of resistance training over 10 weeks	Plantar flexors strength [Nm]	NS
					GMFM-66 dimension E, %	NS
					GMFM-66 dimension D, %	NS
					Gait speed [m s^{-1}]	NS
Kalkman et al. [38]	$N = 22$ (Exp.gp=12, Con.gp=10) Age (years) 7–14	Spastic CP, ([GMFCS] levels I–III)	Exercises were performed 4 times a week, of which the principal investigator supervised one session and 3 were performed at home. **Intervention group:** 4 weeks of calf muscle strengthening exercises, followed by 6 weeks of calf muscle stretching and strengthening exercises. 3 sets of 12 repetitions, reaching volitional fatigue at the end of each set	10 weeks	Plantar flexors strength	$P = 0.009$*
					Dorsiflexion angle	NS
					Ankle angle at resting length	NS
					Ankle angle at lengthening position shifted towards plantar flexion	P=0.03*
					Resting gastrocnemius muscle length	NS
					Resting gastrocnemius fascicle length	$P = 0.02$*
					Gastrocnemius muscle and tendon length after intervention	NS
					Achilles Tendon stiffness	$P = 0.04$*

Table 1 (continued)

Hosl et al. [39]	$N = 10$ (BDTT.gp=5, Stretch.gp=5) Age (yrs) mean (±SD): 12 (±4)	Spastic CP, ([GMFCS] levels I–II)	Two treatments and two crossover designs were used. Each one required a 9-week treatment period (3 sessions per week), which was preceded by a 5-week passive run-in and wash-out period. **Static calf stretching:** 7 exercises referring to Morrel and Lau. Exercises were alternately ended with 5 repetitions per leg and end-range positions were held for 20 s. **Backward Downhill Treadmill Training (BDTT):** Eccentric exercise backward-downhill treadmill training (BDTT). From the second week, the load was gradually increased by using a belt speed, a decline slope, and a weight belt. Participants were instructed to take large steps, maintain an erect posture, and limit their use of the handrail.	9 weeks	**The outcomes measured at comfortable walking speed and the fastest possible walking speed. The table will mention only the significant value of either the comfortable or the fastest possible walking speed if present.**	
					Peak plantar flexion moment [Nm/kg]	NS
					Peak plantar flexion power [W/kg]	NS
					Peak dorsiflexion stance [°]	NS
					Peak dorsiflexion swing [°]	NS
					Comfortable walking speed • Knee flexion angle in swing phase [°] • Mean dorsiflexion single stance [°]	• P=0.037* • P=0.041*
					Fastest possible walking speed • Knee flexion angle in swing phase [°] • Velocity [m/s] • Cadence [steps/min] • Peak gastrocnemius muscle-tendon unit velocity (GAS vel.) swing [cm/s]	• P=0.003* • P=0.017* • P=0.004* • P=0.023*
					Mean knee flexion single stance [°], step length [cm], mean toe clearance [cm]	NS
					Functional ambulatory mobility tests • GMFM D, E [Score 0–100] • Timed-Up-and-Go [sec] • Clinical spasticity assessment	NS
					Active ankle-joint assessments Peak moment [Nm]	NS
					Passive ankle-joint assessments • . Peak dorsiflexion [°] • Dorsiflexion at fixed moment [°] • Peak moment [Nm] • Joint stiffness [Nm/°]	NS
					Muscle-tendon properties medial gastrocnemius morphometrics [at rest] • Fascicle angle [°] • Thickness [cm] • Fascicle length [cm] • Muscle length [cm] • Tendon length [cm]	NS
					Total strain in medial gastrocnemius [resting length to max. length] • Fascicle [%] • Muscle belly [%] • Tendon [%]	NS
					Passive resistive ankle stiffness [between 20 and 80% common passive joint moment] • Tendon [Nm/cm] • Muscle belly [Nm/cm]	NS

Table 1 (continued)

Kruse et al. [40]	$N = 27$ (PRT.gp = 12, HICT.gp = 15) Age (yrs:m) mean (±SD): 12:8 (±2:6)	Spastic CP, ([GMFCS] levels I–II)	**Progressive resistance training (PRT):** For 8 weeks, perform 3 times a week. Warm up for 10 min and cool down for 5 min. In between, 5 functional lower extremity exercises (sit-to-stand, heel raises, forward lunges, lateral step-up, and bridging) are performed in 3 circuits with 10–12 repetitions. The exercises were performed in a controlled manner at a slow to moderate speed for 2 min. Take a break between sets. The training load was gradually increased using a weight vest and a 10 repetitions maximum test. **High-Intensity Circuit Training (HICT):** The warm-up, cool-down, and functional exercises were the same as in PRT, but the exercise load was defined by the number of repetitions that could be completed within a 30-s interval. A break period of 30 s was provided.	8 weeks	Plantar flexor strength	P=0.01*
					Ankle joint ROM [°]	NS
					Maximum dorsiflexion [°]	NS
					Maximum plantar flexion [°]	S*
					Resting angle [°]	NS
					Absolute plantar flexor muscle-tendon unit length	NS
					Gastrocnemius muscle belly length	NS
					Gastrocnemius fascicle length	NS
					Pennation angle	NS
					Gastrocnemius thickness	NS
					Vastus lateralis thickness	$P = 0.01$*
					Rectus femoris thickness	$P = 0.004$*
					Achilles tendon length	NS
					Achilles tendon cross-sectional area, mm^2	NS
					Peak passive ankle torque, N·m	NS
					Peak active ankle torque	NS
					Achilles tendon elongation, mm	NS
					Achilles tendon stiffness, N/mm	NS
					Achilles tendon strain, %	NS
					Achilles tendon stress, N/mm^2	NS
					Achilles tendon moment arm	NS
					Young modulus N/mm^2	NS
Bohm et al. [40]	N=8 (Exp.gp=4, Con.gp=4) Age (yrs:m) mean (±SD): 13:0 (±4:3)	Spastic CP, ([GMFCS] levels I–III), manual ability classification MACS (I - II).	The training protocol designed for the beginners over the 6-week training. 1st week: wand and safety gear 2nd week: bring the centre of gravity towards the wall 3rd week: stabilize at the wall 4th week: strength training 5th week: climb down the wall 6th week: all techniques practised A wash-out period of 4 weeks without therapy before the start of the study and between the two interventions	6 weeks	Ankle dorsiflexion angle [°]	NS
					Knee flexion angle [°]	P=0.039*
					Hip internal rotation angle [°]	NS
					Speed	NS
					Step length	NS
					Step time	NS
					Gait profile score (GPS) [°]	NS

Table 1 (continued)

Scholtes et al. [41]	$N = 51$ (Exp.gp=26, Con.gp=25) Age (yrs:m) mean (±SD): Exp.gp: 10:4 (1:10) Con.gp: 10:3 (2:3)	Spastic CP, ([GMFCS] levels I–III)	Functional Progressive Resistance Exercise training (PRE-training) exercises for 12 weeks. Every session consists of: • One exercise on a child-adapted leg press • Three functional exercises (sit-to-stand, lateral step-up, half knee-rise), loaded with a weighted vest The intensity was gradually increased. 3 times a week, the children do 3 sets of 8 repetitions. Each session lasted 60 min and is held in small groups of 4–5 children at school.	12 weeks	**All outcomes assessed at [after 6 weeks of training (T1)] were not significant.** **The table represents all outcomes assessed [at the 6-week follow-up (T3)] except if the outcome assessed was significant [after 12 weeks of training (T2)]**	
					Ankle plantar flexors strength (N/kg)	NS
					Knee extensors strength (N/kg)	NS IN (T2) $P = 0.01$*
					Knee flexors strength (N/kg)	NS
					Total strength (hip extensors, flexors, and abductors, knee extensors and flexors, and ankle plantar flexors) (N/kg)	NS In (T2) = 0.04*
					Anaerobic muscle power (W/kg)	NS
					ROM of gastrocnemuis (°)	NS
					ROM of soleus (°)	NS
					Step length (m)	NS
					Cadence (steps/min)	NS
					Comfortable walking speed (m/s)	NS
					Fast walking speed (m/s)	NS
					Timed stair test (m)	NS
Dodd et al. [42]	N=21 (Exp.gp=11, Con.gp=10) Age (yrs:m) mean (±SD): 13 :1 (±3:1)	Spastic diplegic CP	**Strength training exercise:** 3 exercises that target ankle plantar flexors, knee extensors, hip extensors. 1) Bilateral heel rises 2) Bilateral half squats 3) Step-ups After adjusting the initial load, the participants were instructed to perform 3 sets of 8 to 10 repetitions of each exercise 3 times a week for 6 weeks. Each session lasted between 20 and 30 min. The training load was adjusted by adding free weights to the child's backpack.	6 weeks	Ankle plantar flexors	NS
					Knee extensors strength	NS
					Combined ankle plantar flexors, knee extensors strength	S *
					Total extensors strength (ankle plantar flexors, knee extensors, hip extensors)	NS
					GMFM D, %	NS
					GMFM E, %	NS
					GMFM total, %	NS
					Walking speed, m/min	NS
					Timed stair, s	NS

*Significant values, *NS* non-significant, *Exp.gp* Experimental group, *con.gp* Control group, *yrs* Years, *m* Month, *s* Seconds

Table 2 PEDro score

Criteria	Rayn et al. [37]	Kalkman et al. [38]	Hosl et al. [39]	Kruse et al. [40]	Bohm et al. [41]	Scholtes et al. [42]	Dodd et al. [43]
1. Specified eligibility criteria	Yes	Yes	Yes	Yes	Yes	Yes	Yes
2. Random allocation of participants	Yes	Yes	Yes	Yes	Yes	Yes	Yes
3. Concealed allocation	Yes	Yes	No	Yes	Yes	Yes	Yes
4. Similar prognosis at baseline	Yes	Yes	Yes	Yes	Yes	No	Yes
5. Blinded participant	No	No	No	No	No	No	No
6. Blinded therapists	No	No	No	No	No	No	No
7. Blinded assessors	Yes	Yes	No	No	Yes	Yes	Yes
8. More than 85% follow-up for at least one key outcome	Yes	No	Yes	No	Yes	Yes	Yes
9. "Intention to treat" analysis	Yes	No	Yes	No	Yes	Yes	Yes
10. Between-group statistical analysis for at least one key outcome	Yes	Yes	Yes	Yes	Yes	Yes	Yes
11. Point estimates of variability for at least one key outcome	Yes	Yes	Yes	Yes	Yes	Yes	Yes
PEDro score	88/10	66/10	66/10	55/10	88/10	77/10	88/10

Table 3 AACPDM methodological quality

Study	Quality	1	2	3	4	5	6	7
Rayn et al. [37]	S 7/7	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Kalkman et al. [38]	S 6/7	Yes	Yes	Yes	Yes	Yes	No	Yes
Hosl et al. [39]	S 6/7	Yes	Yes	Yes	No	Yes	Yes	Yes
Kruse et al. [43]	M 5/7	Yes	Yes	Yes	No	Yes	No	Yes
Bohm et al. [40]	S 6/7	Yes	Yes	Yes	Yes	Yes	Yes	No
Scholtes et al. [41]	S 7/7	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Dodd et al. [42]	S 7/7	Yes	Yes	Yes	Yes	Yes	Yes	Yes

M moderate, *S* strong, *W* weak

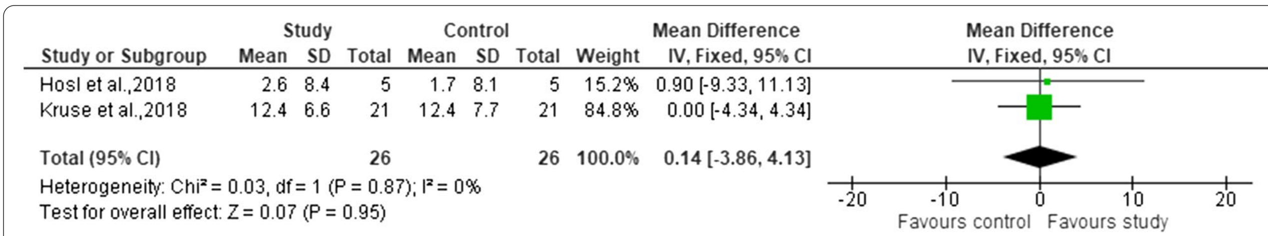

Fig. 2 Forest plot of comparison between study and control groups regarding dorsiflexion angle — post-intervention

Measurements of the outcomes

All studies used a dynamometer for measuring plantar flexors strength, goniometry, and markers for measuring plantar flexion angle and dorsiflexion angle.

Methodological quality level

Table 2 shows how each study scored on the PEDro scale. The mean score of the 7 studies was 6.9. Three studies were rated an 8 [37, 40, 42], one was rated a 7 [41],

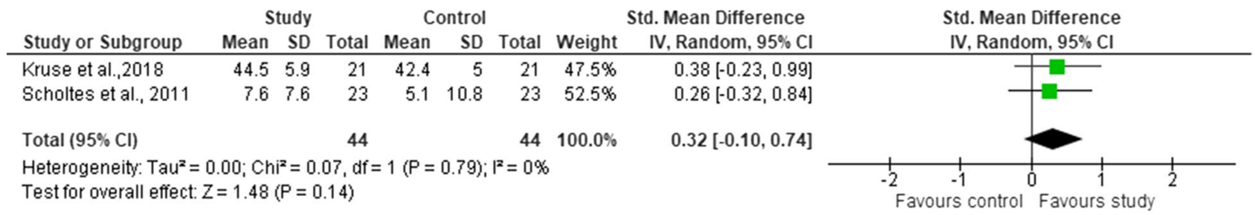

Fig. 3 Forest plot of comparison between study and control groups regarding plantar flexion angle — post-intervention

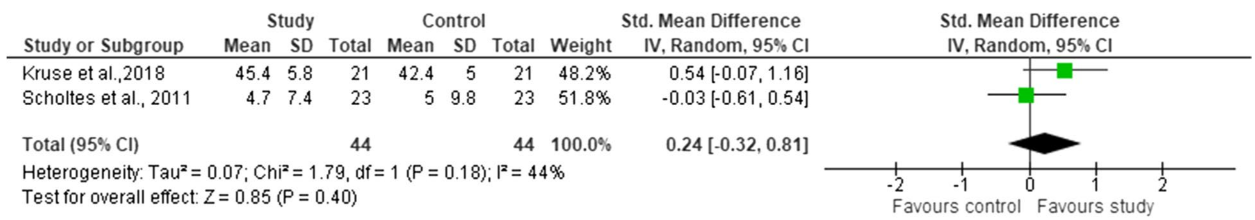

Fig. 4 Forest plot of comparison between study and control groups regarding plantar flexion angle — follow-up

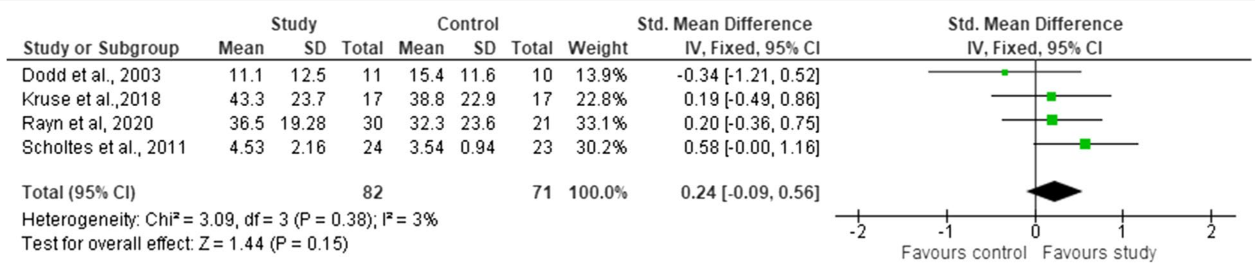

Fig. 5 Forest plot of comparison between study and control groups regarding plantar flexors strength — post-intervention

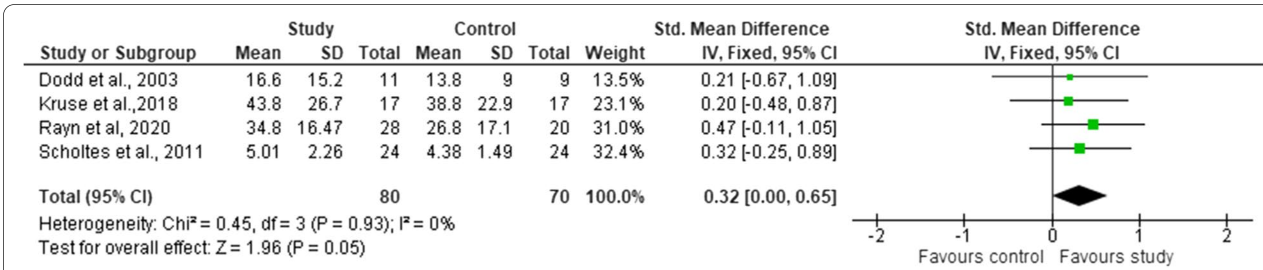

Fig. 6 Forest plot of comparison between study and control groups regarding plantar flexors strength — follow-up

two studies were given a 6 [38, 39], and one research was given a 5 [43] that represents "fair" quality.

The score of each study on the AACPDM conduct questions is also presented in Table 3. The mean score of the 7 studies was 6.3. Three studies had a score of 7 [37, 41, 42], and three studies received a score of 6 [38–40] which represents "strong" quality. A score of 5 was assigned to one study [43], indicating "moderate" quality.

The AACPDM conduct questions: (a) Had inclusion and exclusion criteria of the study participants well defined and followed? (b) Had the intervention well described and was there adherence to the intervention protocol? (For 2 group designs, had the control intervention also well described?) Both parts of the question must be met to score "yes." (c) Had the measures used clearly described, valid, and reliable for measuring the outcomes of interest? (d) Had the outcome assessor unaware of the intervention status of the participants (i.e. were the assessors masked)? (e) Did the researchers conduct and describe appropriate statistical measures including power calculations? Both parts of the question must be met to score "yes." (f) Had dropout/loss to follow-up reported and less than 20%? For 2 group designs, was dropout balanced? (g) Considering the potential within the study design, had the authors use appropriate methods for controlling confounding variables and limiting potential biases?

Level of evidence

According to the modified Sacket scale [31], five studies were ranked level 1 and two studies ranked level 2.

Descriptive synthesis of the risk of bias

All studies had random allocation of participants, had groups of similar baselines, reported results of between-group statistical comparisons, provided measures of variability for at least one outcome, and specified the eligibility criteria. There were five studies with concealed allocation [37, 38, 40–42], six studies with blinded assessors [37–42], and only one study with blinded participants and blinded therapists [42].

Meta-analysis

A meta-analysis was performed for three variables: dorsiflexion angle in the immediate assessment (Fig. 2), plantar flexion angle in the immediate assessment (Fig. 3) and the follow-up assessment (Fig. 4), and plantar flexion strength in the immediate assessment (Fig. 5) and the follow-up assessment (Fig. 6).

Dorsiflexion angle

Two studies [39, 43] were included in a meta-analysis for dorsiflexion angle — post-intervention. As shown in Fig. 2, the total number of children included in the meta-analysis is 26 in study groups and 26 in control groups. The forest plot of mean difference across the two studies at 95% CI of mean difference is MD $= 0.14$ (95% CI of mean difference $= -3.86, 4.13$). Furthermore, the 95% confidence intervals of the overall effect estimate overlap the null effect value, so the meta-analysis level revealed a non-significant difference between the study groups and control groups (the overall effect P value is 0.95). The I^2 statistic ($I^2 = 0\%$, $P = 0.87$, random-effects model) is expressed as a percentage and represents the total variability in the studies' effect measure which is due to heterogeneity. The I^2 value is $<$ 50%, and the test P value is $>$ 0.05 so the studies can be considered homogenous.

Plantar flexion angle

Two studies [41, 43] were included in a meta-analysis for plantar flexion angle — post-intervention. As shown in Fig. 3, the total number of children included in the meta-analysis is 44 in study groups and 44 in control groups. The forest plot of mean difference across the two studies at 95% CI of mean difference is MD $= 0.32$ (95% CI of mean difference $= -0.10, 0.74$). Furthermore, the 95% confidence intervals of the overall effect estimate overlap the null effect value, so the meta-analysis level revealed a non-significant difference between the study groups and control groups (the overall effect P value is 0.14). The I^2 statistic ($I^2 = 0\%$, $P = 0.79$, random-effects model) is expressed as a percentage and represents the total variability in the studies' effect measure which is due to heterogeneity. The I^2 value is $<$ 50%, and the test P value is $>$ 0.05 so the studies can be considered homogenous.

Two studies [41, 43] were included in a meta-analysis for plantar flexion angle —follow-up. As shown in Fig. 4, the total number of children included in the meta-analysis is 44 in study groups and 44 in control groups. The forest plot of mean difference across the two studies at 95% CI of mean difference is MD $= 0.24$ (95% CI of mean difference $= -0.32, 0.81$). Furthermore, the 95% confidence intervals of the overall effect estimate overlap the null effect value, so the meta-analysis level revealed a non-significant difference between the study groups and control groups (the overall effect P value is 0.40). The I^2 statistic ($I^2 = 44\%$, $P = 0.18$, random-effects model) is expressed as a percentage and represents the total variability in the studies' effect measure which is due to heterogeneity. The I^2 value is $<$ 50%, and the test P value is $>$ 0.05 so the studies can be considered homogenous.

Plantar flexors strength

Four studies [37, 41–43] were included in a meta-analysis for plantar flexors strength — post-intervention. As shown in Fig. 5, the total number of children included in the meta-analysis is 82 in the study groups and 71 in the control groups. The forest plot of mean difference across the four studies at 95% CI of mean difference is MD $=$ 0.24 (95% CI of mean difference $= -0.09, 0.56$). Furthermore, the 95% confidence intervals of the overall effect estimate overlap the null effect value, so the meta-analysis level revealed a non-significant difference between the study groups and control groups (the overall effect

P value is 0.15). The I^2 statistic ($I^2 = 3\%$, $P = 0.38$, random-effects model) is expressed as a percentage and represents the total variability in the studies' effect measure which is due to heterogeneity. The I^2 value is ˂ 50%, and the test P value is ˃ 0.05 so the studies can be considered homogenous.

Four studies [37, 41–43] were included in a meta-analysis for plantar flexors strength — follow-up. As shown in Fig. 6, the total number of children included in the meta-analysis is 80 in the study groups and 70 in the control groups. The forest plot of mean difference across the four studies at 95% CI of mean difference is MD = 0.32 (95% CI of mean difference = 0.00, 0.65). Furthermore, the 95% confidence intervals of the overall effect estimate overlap the null effect value, so the meta-analysis level revealed a non-significant difference between the study groups and control groups (the overall effect P value is 0.05). The I^2 statistic ($I^2 = 0\%$, $P = 0.93$, random-effects model) is expressed as a percentage and represents the total variability in the studies' effect measure which is due to heterogeneity. The I^2 value is ˂ 50%, and the test P value is ˃ 0.05 so the studies can be considered homogenous.

Discussion

Best practice in rehabilitation requires sufficient evidence before an intervention can be considered appropriate in a defined population. The current systematic review used systematic methods for searching for and appraises available studies in order to assess the quality and strength of evidence of different forms of exercises for equines in children with cerebral palsy. In this review, the outcomes included are dorsiflexion angle, plantar flexion angle, and plantar flexion strength. Strengthening exercises promote bone growth, lower blood sugar, aid in weight control, improve balance and posture, and alleviate joint stress and pain [44]. Stretching the calf muscles has been shown to improve ankle range of motion in both young and old patients [45]. Calf stretching exercises are commonly prescribed and the most common clinical approach for treating ankle joint equines. It promotes and maintains flexibility, reduces pathological pressures and forces, and enhances dynamic gait [46].

This review included randomized controlled trials. This design allows for the estimation of the effect of exercise training on equinus deformity in children with cerebral palsy. Randomization reduces selection bias and provides a comprehensive tool for examining cause–effect relationships between an intervention and an outcome, which makes the RCT the gold standard for intervention and impossible to achieve with any other study design [47].

All studies in this systematic review had random allocation of participants and similar baselines, reported results of between-group statistical comparisons, and provided measures of variability for at least one outcome. The eligibility criteria were specified in all studies. Five studies [37, 38, 40–42] had blinded assessors and concealed allocation. Only one study [42] had blinded participants and a blinded therapist. All the included studies obtained at least one outcome from more than 85% of the participants initially allocated, and five studies [37, 39–42] used an intention-to-treat analysis.

During normal gait, the coupling of ankle plantarflexion and knee extension moments during midstance promotes lower limb stability [48]. The soleus, an ankle plantarflexor muscle, restrains forward rotation of the tibia over the foot during stance and helps knee extension without the need for quadriceps activity [49, 50]. Equines are characterized by a reduction in ankle dorsiflexion range of motion and a limitation of the ankle joint complex in the sagittal plane associated with abnormal heel-to-toe loading during gait [49]. This limitation of ankle joint dorsiflexion is associated with impaired balance and functional ability [51, 52], as well as an increased risk of falling [53]. It also proposed that an equinus contributes to the development of gait pathologies by increasing the forefoot pressures, as the reduction in range of motion alters gait and forefoot loading patterns [51–54]. Increased stance phase knee extension in individuals with equinus is complicated by changes in the musculoskeletal system triggered directly or indirectly by the injury to the central nervous system. Knee hyperextension associated with equinus has been attributed to increased ankle plantar flexor stiffness from, for example, contractures [55], or changes in mechanical properties of sarcomeres secondary to the injury [56].

In this systematic review, strengthening exercises were used in five studies by Rayn et al. [37], Hosl et al. [39], Kruse et al. [43], Scholtes et al. [41], Dodd et al. [42]; combined stretching and strengthening exercises were used in a study by Kalkman et al. [38]; and eccentric stretching of the ankle joint was used in a study by Bohm et al. [40]. The duration of strengthening exercises ranged from 6 to 12 weeks. The strengthening exercises were performed three times per week, three to eight sets with eight to twelve repetitions.

The outcomes in the studies represent International Classification of Function (ICF) components of body structure and function [37–43] and activity and participation [37, 39–42].

Searle 2019 examined the effect of 8 weeks of stretching exercise of the calf muscle and found no significant impact, neither on the dorsiflexion angle nor on the plantar flexion pressure [57]. Continuous stretching can improve the range of motion [58]. Cui Zhang et al. found that resistance exercises for 16 weeks can improve ankle kinesthesia in the plantar flexion and dorsiflexion axis [59].

Ankle joint mobility, muscle strength, and walking performance can all be improved with exercise training that is suited to the subject's condition [60]. As fatigue was increased in gastrocnemius medialis during walking among CP children [61] and that isometric plantar flexor strength explained at least 50% of the variance measures of walking capacity in adults with CP [62], as a result, resistance training, which improves muscle strength in the ankle plantar flexion muscles, may improve walking performance. Muscular strength, mechanical power, and speed can all be improved with strength and power training [63].

A study by Katsura et al. found that aquatic exercise reveals a significant effect of progressive resistance exercise on plantar flexors strength as a result of increasing triceps surae strength [64], the muscle which the main function is to flex the ankle and maintain the upright posture [65]. Kruse et al. [43] is the only study that shows a significant result in favour of progressive resistance exercise in the plantar flexors strength. They assumed that the absolute value differences were related to the normal growth against their hypothesis as there is no increase in gastrocnemius thickness. The resistance exercise may be nonspecific enough to modify the gastrocnemius architecture.

McHugh and Cosgrave have suggested that stretching before sports activities should be avoided because it decreases muscle strength [66]. This decrease in muscle strength is related to stretch duration [67] and caused by decreasing in muscle-tendon unit stiffness and neuromuscular activity [68, 69]. The length-tension relationship may be affected by potential mechanical changes in muscle-tendon unit stiffness [70]. Aerobic exercise improves muscle strength by increasing neuromuscular activity [71], improving the muscle length-tension relationship [72], accelerating metabolism of the muscle [73], and also psychological preparation [74]. As a result, combining stretching and aerobic exercise may improve the muscle strength deficit caused by stretching alone due to an increase in neuromuscular activity by decreasing MTU stiffness and increasing muscle strength [75].

The limitations of this review were the small number of studies included in the meta-analysis and the variable methodology of these studies (the type, duration, and intensity of exercise training) and the very small number of participants in two of the studies [39, 40]. Further research into the effect of various types of exercise should be carried out. The results of this systematic review need to be interpreted cautiously in the context of the small number of available high-quality studies as the effect of specific types of exercise on equinus deformity in children with CP has rarely been reported. Therefore, there is a need for further research with adequately powered studies and a large sample to provide adequate evidence of different exercise training on equines in children with cerebral palsy.

However, this systematic review recommends an urgent need for further and specified research on using different types of exercises not only in the management of equines in children with CP but also in the prevention of the deformity, using diverse outcome measures, including activities and participation. It is also recommended for further research for the combination of exercise training with other treatment methods for equinus in children with cerebral palsy.

Conclusions

This systematic review aims to fill the knowledge gap that exists between research and clinical practice in the use of exercise in the management of equinus deformity. It can provide the best intervention for equinus deformity in children with cerebral palsy. The current level of evidence to support the effectiveness of exercise on equinus deformity in children with cerebral palsy is still weak, so there is a clear need for more high-quality randomized controlled trials to establish strong evidence.

Abbreviations

AACPDM: American Academy for Cerebral Palsy and Developmental Medicine; CI: Confidence interval; CP: Cerebral palsy; GMFCS: Gross motor function classification system; PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-analysis; RCT: Randomized controlled trial; GSC: Gastrosoleus muscle complex; NS: Non-significant; ICF: International Classification of Function; MD: Mean difference; PEDro: Physiotherapy Evidence Database; Exp.gp: Experimental group; con.gp: Control group; yrs: Years; m: Month.

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DA and WM performed an electronic search and data extraction independently and assessed the methodological quality of included studies where discrepancies between them were resolved by consultation with the third author FA to reach the final decision. The authors read and approved the final manuscript.

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Declarations

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