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Review

Treatment of knee hyperextension in post-stroke gait. A systematic review



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ABSTRACT

Background: Post-stroke, patients exhibit considerable variations in gait patterns. One of the variations that can be present in post-stroke gait is knee hyperextension in the stance phase.

Research question: What is the current evidence for the effectiveness of the treatment of knee hyperextension in post-stroke gait?

Methods: MEDLINE, EMBASE, PEDro, CINAHL, and the Cochrane library were searched for relevant controlled trials. Two researchers independently extracted the data and assessed the methological quality. A best evidence synthesis was conducted to summarize the results.

Results: Eight controlled trials (5 RCTs, 3 CCTs) were included. Three types of interventions were identified: proprioceptive training, orthotic treatment, and functional electrostimulation (FES). In the included studies, the time since the stroke occurrence varied from the (sub)acute phase to the chronic phase. Only short-term effects were investigated. The adjustment from a form of proprioceptive training to physiotherapy training programs seems to be effective (moderate evidence) for treating knee hyperextension in gait, as applied in the subacute phase post-stroke. Neither evidence for effects on gait speed nor gait symmetry were found as a result of proprioceptive training. Orthoses that cover the knee have some effects (limited evidence) on knee hyperextension and gait speed. No evidence was found for FES.

Significance: This is the first systematic literature review on the effectiveness of interventions on knee hyper-extension in post-stroke gait. We found promising results (moderate evidence) for some "proprioceptive approaches" as an add-on therapy to physiotherapy training programs for treating knee hyperextension during the subacute phase post-stroke, in the short-term. Therefore, initially, clinicians should implement a training program with a proprioceptive approach in order to restore knee control in these patients. Because only studies reporting short-term results were found, more high-quality RCTs and CCTs are needed that also study mid- and long-term effects.

1. Introduction

Stroke is a global health problem. In 2016, approximately 13.7 million strokes occurred worldwide. There are estimated to be 80.1 million stroke survivors globally [1].

Over the past two decades, the number of stroke survivors has increased [2]. For everyone who suffers a stroke, rehabilitation is an important aspect of recovery [3]. Of all the possible rehabilitation goals, being able to walk again is high on the list for most stroke survivors [4]. Post-stroke, patients exhibit considerable variations in gait patterns,

depending on the residual function and the severity of sensorimotor impairment [5]. One of the variations that can be present in post-stroke gait is hyperextension of the knee in the paretic limb during the stance phase [6]. Knee hyperextension is also known as genu recurvatum, which is characterized by a ground reaction force vector, which passes in front of the knee resulting in full knee extension (0°) or more [7]. Knee hyperextension is a progressive, disabling deformity that impairs walking speed, decreases gait efficiency, increases use of energy during walking, and can result in or might be associated with knee pain [8–10]. Moreover, knee hyperextension can reduce gait symmetry and thus,

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affect cosmetic appearance. The asymmetric gait pattern generally results in the increased duration of the swing phase of the paretic limb [11]. Post-stroke, 20–68 % of the walking population reports hyperextension of the knee [7,8,12,13]. The knee hyperextension can be caused by either a single symptom or a combination of symptoms such as weakness, spasticity or retraction of the paretic limb muscles, limited ankle mobility, proprioceptive disorders, and diminished velocity properties of the distal limb muscles [14].

In scientific literature, several interventions have been reported for the treatment of knee hyperextension in post-stroke gait. In 2010, Bleyenheuft et al. [7] conducted a systematic review that studied interventions to treat knee hyperextension in hemiparetic adults. They reported on the effectiveness of retraining methods (functional electrostimulation (FES) and electro goniometric feedback), surgical treatment, and orthotic treatment.

Since 2010, studies reporting on other treatments such as proprioceptive training and interventions to influence spasticity in the lower limb muscles on knee hyperextension post-stroke have also been published [12,15,16]. However, until now, healthcare professionals have not had a clear treatment preference. Therefore, the aim of the current study was to systematically review the scientific literature in order to provide more clarity on the preferred method to address knee hyperextension in post-stroke gait.

2. Methods

This systematic review was performed according to the Preferred Reporting Items for Systematic review and Meta-Analysis Protocols (PRISMA-P) 2015 statement [17].

2.1. Literature search

A computer-aided literature search was carried out in MEDLINE, EMBASE, PEDro, CINAHL, and the Cochrane library up to May 2021. The search strategy, which was composed with the assistance of an experienced librarian, is shown in Appendix 1. In addition, references of the included studies were checked by hand to identify additional relevant studies.

2.2. Inclusion criteria

Trials carried out following a comparative study design, where a test group and a control group were compared, were used in this current review. This included control groups consisting of people post-stroke such as randomized controlled trials (RCT) and clinical controlled trials (CCT), including cross-over and paired sample designs. Studies were considered eligible for inclusion if they met all of the following criteria: (1) the study population consisted of stroke survivors with knee hyperextension in gait, (2) the studies had a minimum of 10 participants, (3) the studies reported on the effect of interventions in the treatment of knee hyperextension, (4) the outcome measures that were assessed included: measurements on knee hyperextension in stance phase of gait, gait velocity, gait symmetry, or energy cost, and (5) the full texts of the articles were available, in Roman alphabet. Studies were included when at least 75 % of the participants had been diagnosed with a stroke and the results of these participants were reported separately.

2.3. Study selection

Two reviewers (MG and NM) independently applied the inclusion criteria to select relevant trials based on titles, abstracts, and full texts respectively. A consensus method was used in the cases where there was a disagreement. A third reviewer (BH) was consulted if no consensus could be reached.

2.4. Methodological quality

Two reviewers (MG and NM) independently assessed the methodological quality of the included trials. A consensus method was used in the cases where there were any disagreements.

The Downs and Black checklist [18] was used to assess the methodological quality of the included trials (Appendix 2). This checklist is considered relevant for both randomized and nonrandomized studies. It consists of 27 questions. Each question was scored as "yes" (1 or 2), "no" (0) or "unable to determine" [19–21].

Two questions were modified: Question 15 (blinding measurement of main outcome) was also given a score of 1 if measurements were done with cameras, graphic recorders or potentiometers, which were considered to minimize the risk of bias. In this way, we could make a distinction between trials that measured the occurrence or grade of knee hyperextension by personal perception or using an objective measuring tool. Question 27 (power/sample-size calculation) was modified from a 5-point score to a 0/1-point score as was done in other studies [19,20,22,23]: A score of 1 was given if a power or sample-size calculation was reported and a 0 was given where there was no power or sample-size calculation or a study lacked an explanation as to whether the sample-size was appropriate to detect a clinically important effect. Each paper was considered to have an "excellent" (24–28 points), "good" (19–23 points), "fair" (14–18 points) or "poor" (<14 points) methodological quality.

2.5. Data extraction

Two reviewers (MG and NM) independently extracted the data, using a data extraction form. Disagreements were addressed by means of discussions. Information was collected on study design, study population, aetiology of knee hyperextension, interventions, and outcome measures. Follow-up periods were categorized as short-term (0-3 months), mid-term (4-6 months), and long-term (>6 months).

2.6. Data Analysis/Synthesis

We considered pooling the results of the included trials in case patient characteristics, interventions, and outcome measures were similar. In case a quantitative analysis was not possible, the results were summarized by using a best-evidence synthesis. A trial was included in the best-evidence synthesis if a level of significance was reported. The rating system was based on van Tulder et al. [24] and adapted from Hombergen et al. [25] (Table 1).

Table 1 Levels of evidence for differences between the groups.

- Strong evidence: consistently significant findings* among multiple high-quality RCT's or other controlled intervention studies (rated "excellent" or "good" on the Downs and Black checklist¹⁸)
- Moderate evidence: consistently significant findings" among multiple low quality RCTs or other controlled intervention studies, (rated "fair" or "low" on the Downs and Black checklist) and/or one high quality RCT or other controlled intervention study (rated "excellent" or "good" on the Downs and Black checklist)
- Limited evidence: significant findings* within one low-quality RCT or other controlled intervention study (rated "fair" or "low" on the Downs and Black checklist)
- Conflicting evidence: provided by conflicting (significant) findings in RCTs or other controlled intervention studies (<75 % of the studies reported consistent findings)
- No evidence: no significant findings were found

 $^{^*}$ The results were consistent when at least 75 % of the studies showed results in the same direction, which was defined according to significance (P < 0.05).

3. Results

3.1. Characteristics of the included studies

The computerized search distinguished 1545 potentially eligible studies. Of these 1506 were excluded based on titles and abstract. Two articles [26,27] were excluded because full text could not be obtained, neither through contacting the authors of otherwise. Another 29 studies were excluded after screening the full texts. Finally, a total of eight trials were included in this review: five RCTs [15,28–31] and three (non-randomized) CCTs [6,32,33] (Fig. 1).

3.2. Data extraction

Characteristics of the included studies are presented in Table 2.

3.3. Methodological quality

Table 3 presents the results of the quality assessment. Three trials [15,28,29] were determined to be of excellent quality, two trials [31,33] of good quality, and the others of fair quality. The most prevalent methodological flaws were not mentioning adverse effects (100 %), not blinding the participants to the intervention (88 %), and not providing a power calculation to detect a clinically important effect (88 %).

3.4. Effectiveness of interventions

Various treatment options for knee hyperextension are available. The interventions studied in the included trials could be divided in three categories: 1) proprioceptive training [15,28,29,31,32], 2) orthotic treatment [6,30,33] and 3) functional electro stimulation (FES) [33]. Aman and colleagues [34] classified "active movement/balance training, passive movement training, somatosensory stimulation training, somatosensory discrimination training, and combined/multiple system training" as proprioceptive interventions. In case the intervention of a study included in this systematic review matched one of these categories as established by Aman et al. [34], we called it proprioceptive training. Due to the heterogeneity of the included trials and the differences in the content of the treatments, statistical pooling of the data was impossible. Therefore, we used a best-evidence synthesis (BES) to summarize the results. The evidence for the effectiveness of the interventions of the included trials is presented in Table 4.

3.4.1. Proprioceptive training

Five trials [15,28,29,31,32], in which the interventions were classified as proprioceptive training according to the classification of Aman et al. [34], investigated the influence of retraining knee control in the stance phase of gait by proprioceptive training. In the proprioceptive approach, participants were encouraged to avoid hyperextension by keeping the paretic knee in a slightly flexed position during gait, by

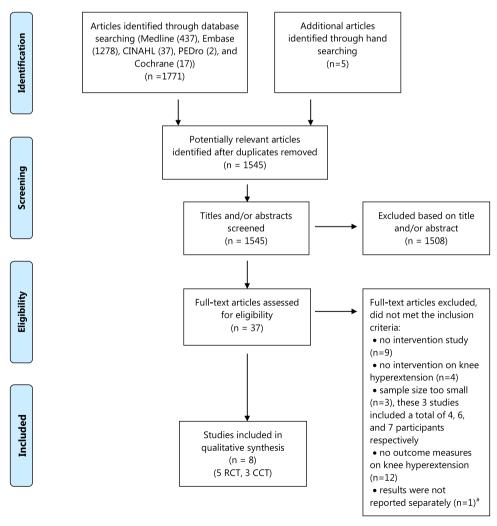


Fig. 1. Flowchart of the literature search.

Studies were included when at least 75 % of the participants had been diagnosed with a stroke and the results of these participants were reported separately.

 Table 2

 Characteristics and outcomes of the selected studies

Authors (year) Design, total number of patients, phase post-stroke	Study population	Etiology knee hyper extension	Intervention	Control/comparison	Outcome measures	Results
Proprioceptive training						
Guo et al. [28] (2015) Pilot RCT n = 30 subacute phase post-stroke	Stroke survivors Age 47-61y. Time since stroke (days) (mean (SD)) (control/ experimental group) 59.4(61.4)/66.9 (42.9) Participants had standing balance score ≥ 2.	Not specified	Whole body vibration training (30–60 s of vibration and 10 s rest intervals, 10 rounds per set, 8 sets per day) while standing on a platform in semi-squatting position, with even weight distribution and single leg standing with knee bending within 0–15°, for 8 weeks. In addition: regular exercises: Range of motion exercises of lower limb, PNF exercise, climbing stairs, walking with brace and electrical stimulation.	Same program with vibration switched off.	Number of knee hyperextension occurrence in 10 m walking, at 8 weeks follow up	Between groups: (number of) d = 1.749*, 95 %CI [2.915, 7.285] p < 0.001*. in favor of the experimental group. Within groups: (number of) before treatment/after treatment Mean(SD) Control group 26.2 (5.4)/10.5(3.2)* p < 0.05 Experimental group 24.8(4.7)/5.4(2.6)*, p < 0.05*.
			n = 15	n = 15	Maximum gait speed over 10 m.(sec), At 8 weeks follow up	Between groups: (sec) d = 1.345, 95 %CI [1.896, 6.704], p = 0.001* in favor of the experimental group. Within groups: Mean(SD) (sec) Control group 24.6 (4.5)/16.3(3.7)* p < 0.05* Experimental group 24.7(5.5)/12.0(2.6)*, p < 0.05*.
Dalal et al. [15] (2018) RCT n = 32 acute and subacute phase post-stroke	Stroke survivors Age 40–80y. Time since stroke: n.r Participants were able to walk 5 m with or without assistance or a walking aid.	Not specified	Prowling along with proprioceptive training (i.e. squats, single limb stance/ squats) for 15–20 min as an adjunct to 6 sessions of routine physiotherapy for 45–60 min in 6–10 consecutive days.	6 sessions routine physiotherapy for 45–60 min in 6–10 consecutive days.	Mean knee hyperextension (degrees) change score 0–6 sessions	Between groups: Mean change (Median (IQR)) knee hyperextension degrees Control vs experimental group -0.35(-1.18, 1.75) vs -4.52(-5.72, -2.82) p
			n = 16	n = 16		< 0.001*. Within groups: before treatment/after treatment (Median (IQR)) degrees Control group: 8(5.25, 10.29)/7.87 (5.12, 9.96) NS Experimental group 8.67(7.17,10.40)/3.87 (3.26, 5.00) p = 0.001*
					Gait speed over 5 m (sec) change score 0–6 sessions	Between groups: Mean change Median (IQR) Control vs experimental group -2.5(-8.00, 0.00) vs -5.00(-8.00, - 0.50) NS Within groups: Mean change Median (IQR) sec before/after treatment, Control group:

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Table 2 (continued)

Authors (year) Design, total number of patients, phase post-stroke	Study population	Etiology knee hyper extension	Intervention	Control/comparison	Outcome measures	Results
						26(16.75, 31.50)/22.1 (13.75, 28.75) p = 0.041* Experimental group: 23(15, 29.75)/18 (11.25, 22.50), p = 0.002*.
Morris et al. [29] (1992) RCT n = 26 subacute phase post-stroke	Stroke survivors Age (y)(Mean(SD)) 64.2(11.9) Time since stroke (days) (mean (SD)) 62 (37) Participants were able to walk safe for 4 × 10 m without aids or orthoses.	Not specified	In phase 1 (4 weeks): Conventional therapy with electro goniometric feedback during standing and gait training. (auditory signal at a certain knee angle). In phase 2 (4 weeks): conventional therapy without electro goniometric feedback. Treatment duration in both phases: 5 times per week for (mean(sd) 27.0(11.6) min. The conventional therapy was a Motor Relearning Programme: gait training in which stereotyped and abnormal synergistic activity was discouraged.	Conventional therapy in both phases (8 weeks) 5 times per week for (mean(SD) 27.0 (11.6) min.	Peak Knee Extension (PKE) i.e. maximum angle of knee extension during stance phase (degrees); Change score 0-4-8 weeks	Mean reduction PKE After Phase1: Between groups Mann-Whitney test p 0.173 NS Mean reduction (SD) degrees Control vs experimental group 2.4(4.2) vs 3.1(3.0) After Phase2: Between groups Mann-Whitney test, p = 0.011* Mean reduction (SD) degrees Control vs experimental group 0,4(3.1) vs 1.7(1.8)
			n = 13	n = 13	Gait symmetry measured by single limb support. Change score 0-4-8 weeks	After Phase1 the symmetry improved: Between groups: Mean change Asymmetry (SD) % of gait cycle Control vs experimental group: 6.1(11.9) % vs 2.7 (11.7) % NS After Phase2 the symmetry regressed: Between groups: Mean change Asymmetry (SD) % of gait cycle Control vs experimental group: 2.4(12.1) % vs 0.4 (16.5) % NS
					Gait speed (m/min), at start, improvement after phase1 and phase2. Change score 0-4-8 weeks	Mean speed improvement (SD) (n min): After Phase1 Between groups: Control group vs experimental: 0.8 (12.5) m/min vs 6.3 (10.2) m/min NS After Phase2: Between groups: Control group vs experimental 5.8(11.2) m/min vs 8 (9.9) m/min
cee et al. [31] (2017) Cross-over RCT n = 18 (and 15 healthy	Stroke survivors Age y(Mean(SD) 39.2 (16.8), Time since stroke (months) (mean (SD))	Not specified	Treadmill gait with assistive guidance force using tubing (GTG) to improve knee stabilization during midstance.	Conventional treadmill gait (CTG)	Knee hyperextension in midstance, (degrees) measured during a set of 5 min CTG and 5 min GTG.	Mean knee hyperextension(SD) degrees CTG(control) vs GTG (experimental) in the (continued on next page

Table 2 (continued)

Authors (year) Design, total number of patients, phase post-stroke	Study population	Etiology knee hyper extension	Intervention	Control/comparison	Outcome measures	Results
adults, not included in this review) subacute to chronic phase post-stroke	6.7(2.9) Participants had a Berg Balance Score >40.		2–5 sessions of 10 min CTG and 10 min GTG were conducted for each individual for modification of the tubing tension.			hemiparetic group: 189(3.9) vs 178(7.7)* p < 0.01*
post-stroke			n=18	n=18		
Ceceli et al. [32] (1996) CCT n = 41 acute to chronic phase post-stroke	Stroke survivors Age experimental group 13–67y (median 49.5), age control group 12–65y (median 54). Time since stroke 0- >1y. Participants had a normal passive range of motion in lower extremity, were able to walk unassisted for 50 steps.	Not specified	Joint-position biofeedback training (acoustic signal when knee extension exceeded 180° (+/-3°) for 30 min per day, for 10 days in addition to conventional physiotherapy.	conventional physiotherapy (i.e., exercises for pelvis, hip control and weight shifting)	Number of steps with knee hyperextension in 50 steps In advance, after 10 days of training and after 6 months follow up	Between groups: significant difference after 10 days of training in favor of the experimental group, p < 0.05* Within groups: Mean number of steps with knee hyperextension before/after training Control group: 48.67/39.38. Experimental group 45.19/7.538. After 6 months: In control group 2/9 walked without knee hyperextension vs 4/1 in the experimental group. No statistical analysis available on six months.
			n = 26	n = 15	Gait speed (number of steps per minute) in advance and after 10 days of training.	Gait speed: NS
Orthotic treatment						
Portnoy et al. [30] (2015) Cross-over RCT n = 31 subacute to chronic phase post-stroke	Stroke survivors Age (y) (mean(SD)): 59.9 (15,1) Time since stroke (y) (mean (SD)) 6.1(6,7), range 3 months-25y Participants walked independently with or without walking aid.	spasticity, paresis lower limb muscles	Use of a hinged soft knee orthosis, set on 10° of knee flexion, for 4 weeks. Two groups A and B. After initial measurement Group A used orthosis for 4 weeks, group B no intervention. After second measurement group B used orthosis for 4 weeks and group A no intervention. Measurement 3. n = 31 (n = 17 group A, n = 14	No intervention $n = 31$ $(n = 17 \text{ group A}, n =$	Peak Knee Extension in stance phase (PKE) (degrees) Mean score at baseline and after 4 weeks wearing orthosis. Gait speed over 10 m (sec) Mean score at baseline and after 4 weeks wearing orthosis.	Mean PKE (SD) degree without orthosis vs with orthosis: -8.2(7.2) vs 10.1(9.5 p < 0.001* Mean time over 10 m (SD) sec. without orthosis vs with orthosis 22.9(16.6) vs 21.3(16.6), p = 0.011
			(n = 17 group A, n = 14 group B)	(n = 17 group A, n = 14 group B)	Gait symmetry indices without and with knee orthosis. Mean score at baseline and after 4 weeks wearing orthosis.	NS
Boudarham et al. [6] (2012) CCT (Cross-over trial) n = 11 chronic phase post-stroke	Stroke survivors Age (y) (mean (SD)): 51 (15). Time since stroke 72–672 months. Participants were able to walk 10 m without walking aids.	spasticity quadriceps (n = 6), spasticity of m. triceps surae (n = 3), weakness of quadriceps (n = 2)	Use of a Knee-ankle-foot orthosis (KAFO) Two gait conditions were measured in a fixed order: without KAFO and with KAFO n = 11	No intervention $n = 11$	Peak Knee Extension in stance phase (degrees) Score in one session Gait speed over 10 m (m/sec) Score in one session	Mean PKE(SD) degree without KAFO vs with KAFO -16.2 (11.9) vs -7.6 (7.4) p = 0.029*. Mean speed(SD) m/se without KAFO vs with KAFO 0.57(0.36) vs 0.73 (0.34), p = 0.025*,
					Swing phase asymmetry duration ratio Score in one session	Mean swing phase asymmetry duration ratio(SD) without KAFO vs with KAFO: $1.93(0.77)$ vs 1.27 (0.10), $p=0.014*$

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Table 2 (continued)

Authors (year) Design, total number of patients, phase post-stroke	Study population	Etiology knee hyper extension	Intervention	Control/comparison	Outcome measures	Results
					Stance phase asymmetry duration ratio; Score in one session	(meaning symmetry significantly increased Mean stance phase asymmetry duration ratio(SD) without KAFO vs with KAFO 0.82(0.11) vs 0.86 (0.08), NS
					Step length (non) paretic limb without and with KAFO(m) Score in one session	Mean step length(SD) m. without KAFO vs with KAFO: non- paretic limb 0.35(0.18) vs 0.40 (0.20) p < 0.05*; paretic limb: 0.42(0.16) vs 0.48 (0.15), NS.
FES						
Bae et al. [33] (2019) CCT (Cross-over trial) n = 12 chronic phase post-stroke	Stroke survivors Age (y) (mean(SD)): 54.41(19.29), Time since stroke (months) (mean(SD)): 13.16(9.73)	Excessive plantar flexion ankle due to weakness dorsiflexor or spasticity plantar flexor	Comparison of three walking conditions: consecutively barefoot, AFO and FES FES Foot drop stimulator on peroneal nerve (back of fibula head) and anterior tibial muscle (5 cm under fibula head)	Barefoot walking and AFO walking	Knee angle at mid stance (degrees) Score in one session	Between groups: N.S. Within groups: Mean angle (SD) degrees Barefoot 8.40(11.22) FES 9.53(11.66) AFO 8.20(11.25)
			$n=12$ AFO walking AFO featuring 5° of ankle dorsiflexion	$\begin{aligned} n &= 12 \\ \text{Barefoot walking} \end{aligned}$	Gait speed over 6 m (cm/sec) Score in one session	Gait speed: N.S.
			n = 12 Each walking modality was repeated three times (six meters, three minutes break between each time). Between each modality ten minutes break was mandatory.	n = 12		

Abbreviations: RCT: randomized controlled trial; CCT: controlled clinical trial; NS: Not Significant; PKE: Peak Knee Extension; FES: functional electrical stimulation; AFO: Ankle foot orthosis; *: statistically significant; m: meters; CI: Confidence Interval; SD: Standard Deviation; d:difference; IQR: Inter Quartile Range; n:number.

instruction or biofeedback, and the quadricep muscles were trained in a flexed position.

3.4.1.1. Adjustment of Whole-Body Vibration to a training program versus a training program. Guo et al. [28] (RCT, n = 30, excellent quality) included participants who were 0-3 months post-stroke with a standing balance score ≥ 2. The eight-week training program contained exercises in a semi-squatted position, with even weight distribution, and single leg standing with knee flexion within 0-15° as well as regular exercises. During the training, participants were positioned on the platform of a vibration machine. The experimental group received whole body vibration during the training. The control group received the same training on the same platform, but the vibration machine was turned off. Short-term training effects (after 8 weeks) were measured by counting the times that the knee hyperextended during a 10 m-walk and measuring gait speed. Both groups reported significant benefits based on the times that the knee hyperextended during the 10 m-walk. The experimental group improved significantly more based on knee hyperextension (degrees) as compared to the control group: 1.749, (2.951, 7.285), (mean difference, (95 % CI)), p < 0.001 and gait speed in m/s: 1.345, (1.896–6.704), p = 0.001.

In conclusion, we found moderate evidence in favour of whole-body vibration being included in a training program versus the training program on its own for the effectiveness on knee hyperextension and gait speed in the short-term.

3.4.1.2. Prowling and proprioceptive training adjusted to routine physiotherapy versus routine physiotherapy. Dalal et al. [15] (RCT, n = 32, excellent quality) included persons in the acute and subacute phases post-stroke. The participants had a Brunnstrom Recovery Stage (BRS) of the affected lower extremity ≥ 3 (i.e. synergy dependent voluntary movement). Both the experimental and control groups received 6 sessions of routine physiotherapy lasting 45–60 min for 6–10 consecutive days. As an adjunct, the experimental group needed to perform 15-20 min of prowling (walking with bilateral knee flexion with trunk in mild forward flexion) and proprioceptive training of the affected limb (partial squats, single limb stance, single limb partial dynamic squats). In addition, the participants of the experimental group were instructed to walk with their knees flexed during routine daily activities. The content of the routine physiotherapy was not described. Short-term training effects (after 6 sessions) were measured on the degree of knee hyperextension and gait speed over a five-meter distance. Group comparisons showed significant benefit to knee hyperextension (degrees) in favour of the experimental group: -4.52 (-5.72, -2.82) (mean change Median (IQR)) in the experimental group versus -0.35 (-1.18, 1.75) in the control group, p < 0.001. Both groups significantly improved gait speed,

Table 3Risk of bias table and methodologic quality scores of the studies included.

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Trial (ref)	rep	orting										ernal va		inte	rnal va	lidity	- bias				in	ternal val	idity - co	nfour	ding			power	TOTAL/28		
Guo ²⁸	1	1	1	1	2	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1)	26		
Dalal ¹⁵	1	1	1	1	2	1	1	0	1	1	1	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1)	25		
Morris ²⁹	1	1	1	1	2	1	1	0	1	1	1	1	1	0	1	1	1	1	1	1	1	1	1	0	1	1)	24		
Bae ³³	1	1	1	1	1	1	1	0	1	0	1	?	1	0	1	1	1	1	1	1	1	?	0	0	1	1)	19		
Lee ³¹	1	1	1	1	0	1	1	0	1	1	1	0	1	0	1	1	1	1	1	1	1	?	1	?	0	1			19		
Portnoy ³⁰	1	1	1	1	2	1	1	0	1	1	?	?	1	0	0	1	1	1	1	1	1	?	- 1	2	1	1		1	18		
Boudarham ⁶	1	1	1	1	1	1	1	0	1	1	?	?	1	0	1	1	1	1	1	1		2		0		1			17		
Ceceli ³²	1	1	1	1	1	1	0	0	1	0	?	?	1	0	1	1	1	1	1	1	1	?	0	0	0	1)	16		

although no significant difference between the groups was found.

According to the BES, moderate evidence was found for the effectiveness, on degree of knee hyperextension, of prowling and proprioceptive training adjusted to routine physiotherapy as compared to routine physiotherapy in the short-term. We found no evidence for the extra benefits of prowling and proprioceptive training on gait speed.

3.4.1.3. Biofeedback training adjusted to a Motor Relearning Programme [35] (MRP) versus MRP. Morris et al. [29] (RCT, n = 26, excellent quality) included participants 1-3 months post-stroke. The effects of biofeedback training as an add-on treatment to MRP were investigated. Participants were included if they were able to safely walk 10 m without aids or orthotics. Both the experimental and control groups received a MRP [35] for 8 weeks. In this program, gait training was given in which stereotyped and abnormal synergistic activity was discouraged. As an add-on treatment to the MRP, the experimental group received biofeedback training: standing and gait training with auditory feedback from an electro-goniometer during the first 4 weeks of therapy. For the biofeedback training an electro-goniometric device mounted on the knee of the participant, provided feedback on the joint position of the knee. An acoustic signal was given when the knee extension exceeded a certain angle. Short-term training effects (after 4 and 8 weeks) were measured on peak knee hyperextension, gait speed and gait symmetry. After 8 weeks of training, a significant benefit of biofeedback training was found to reduce the peak knee hyperextension (Mann-Whitney test, p = 0.011). No significant differences between groups were found on peak knee hyperextension after 4 weeks of training or on gait speed and gait symmetry after 4 and 8 weeks of training.

According to the BES, moderate evidence was found in favour of biofeedback treatment adjusted to a MRP versus MRP alone [35] on knee hyperextension in the short-term after 8 weeks, although no evidence was found after 4 weeks. We found no evidence for an extra benefit of biofeedback treatment on gait speed.

3.4.1.4. Guidance Tubing Gait on the treadmill versus a conventional treadmill gait. Lee et al. [31] (cross-over RCT, n=18 stroke survivors and 15 healthy controls, good quality) compared a guided tubing gait (GTG) on a treadmill with a conventional treadmill gait (CTG) in stroke survivors 4-10 months post-stroke, having a Berg Balance Score > 40, with healthy controls. For the purpose of this review, we only used the data in which both treatments were compared in stroke survivors. An assistive guidance force using tubing was given to improve knee joint stabilization during mid-stance and restore knee joint muscle imbalances and kinematics in gait. Tubing resistance was applied on the popliteal fossa and above the medial malleolus. Previous to the measurements, each participant underwent 2-5 sessions of 10 min of CTG and 10 min of GTG to modify the tubing tension: the applied force was

individually adjusted for each participant to achieve a normal walking pattern. The short-term effects on knee hyperextension in midstance and the electromyographic activation in the quadriceps and hamstrings were measured in one session which entailed 5 min of CTG and 5 min of GTG. In the post-stroke group, a significant difference in favour of GTG was found on knee hyperextension in mid-stance (degrees) pre- and post-testing: 189.1(3.9) (mean(SD)) and 178.1(7.7) p < 0.01.

We found moderate evidence for effectiveness in favour of GTG as compared to CTG on knee hyperextension in the short-term (as applied in one session) for the post-stroke group.

3.4.1.5. Biofeedback training adjusted to conventional physiotherapy versus conventional physiotherapy. Ceceli et al. [32] (CCT, n = 41, fair quality) included persons 0 to >1 year post-stroke. Participants had a normal passive range of motion in the affected lower extremity and were able to walk unassisted for 50 steps. The experimental group received gait training with the instruction not to hyperextend the knee. Auditory feedback on knee hyperextension was provided by using an electromechanical rotational goniometer for ten days, 30 min per day, which was added to the conventional physiotherapy. The control group only received conventional physiotherapy i.e. exercises for the pelvis, hip control and weight shifting. Researchers measured the number of steps with knee hyperextension within 50 steps and gait speed after 10 days and 6 months of training. Between groups, results showed a significant decrease in the number of steps with knee hyperextension in favour of the experimental group in the short-term (10 days): the experimental group went from 45.19 to 7.538 steps (mean (SD: not reported)) versus the control group that went from 48.67 to 39.38 steps (p < 0.05). No short-term differences between the groups were found on gait speed.

The long-term effects (after 6 months) were not statistically analysed because the number of participants that showed up at the check-up was too low.

Therefore, we found limited evidence in favour of the biofeedback treatment adjusted to conventional therapy versus conventional therapy on knee hyperextension in the short-term. No evidence was found for the effectiveness of the interventions on gait speed and gait symmetry in the short-term.

3.4.2. Orthotic treatment

Three trials [6,30,33] investigated the benefits of the use of an orthosis compared to no orthosis to prevent knee hyperextension. Bae et al. [33] compared FES and orthotic treatments (see 3. **FES**)

3.4.2.1. Hinged soft knee orthosis versus no orthosis. Portnoy et al. [30] (cross-over RCT, n=31, fair quality) investigated the added value of a hinged soft knee orthosis in persons, 3 months to 25 years post-stroke. All participants walked independently at the time of recruitment and

Table 4 Evidence for effectiveness of treatment on knee hyperextension post-stroke

Treatment Phase post-stroke	Evidence for Grade of knee hyperextension in stance phase	Gait speed	Gait symmetry
Proprioceptive training			
Training program (a.o.in semi			
squatting position) plus			
Whole-Body Vibration* vs			
training program			
Subacute phase Short-term effects (8 weeks)	++	1.1	
Prowling and proprioceptive	++	++	
training (both with knee			
flexion) adjusted to routine			
physiotherapy* vs routine			
physiotherapy			
Acute/subacute phase	++	NE	
Short-term effects (6 sessions)			
Biofeedback training (during walking with attention to a			
flexed knee) plus Motor			
Relearning Program (MRP)*			
vs MRP			
Subacute phase	++	NE	NE
Short-term effects (8 weeks)			
Guidance Tubing Gait			
(assistive guidance force to knee flexion using tubing) on			
treadmill* vs conventional			
treadmill gait			
Subacute-chronic phase	++		
Short-term effects (1 session,			
during tubing gait)			
Biofeedback training with			
attention to a flexed knee* plus conventional			
physiotherapy vs			
conventional physiotherapy.			
Acute-chronic phase			
Short-term effects (10 days)	+	NE	NE
Orthotic treatment			
Hinged soft knee orthosis* vs			
no orthosis Subacute-chronic phase	_	_	NE
Short-term effects (4 weeks)	+	+	INE
Knee ankle foot orthosis			
(KAFO)* vs no orthosis			
Chronic phase			
Short-term effects (1 session)	+	+	+ (in swin
			phase)
			NE (in stance
			phase)
AFO (5° ankle dorsiflexion)* vs			,
no orthosis (barefoot			
walking)			
Chronic phase	NE	NE	
Short-term effects (1 session) Functional Electrical	NE	NE	
Stimulation (FES)			
Dorsiflexor FES vs barefoot			
walking			
Chronic phase			
Short-term effects (in one	NE	NE	
session)			
Dorsiflexor FES vs AFO (5°			
ankle dorsiflexion) Chronic phase			
Short-term effects (in one	NE	NE	
session)			

[,] in favor of.

had a paresis or spasticity in the lower limb muscles. An 8-week training program was conducted in which the participants wore the orthosis during the day for 4 weeks; the other 4 weeks, they did not wear the orthosis. Measurements on knee hyperextension, gait speed and gait symmetry were conducted without orthosis at baseline, after 4 weeks of wearing the orthosis, and after 4 weeks of not wearing the orthosis. When wearing the orthosis for 4 weeks, knee hyperextension was completely prevented: hyperextension (degrees) with versus without orthosis: 10.1 (9.5) (mean(SD)) versus -8.2 (7.2) p < 0.001. On gait speed, the authors reported no significant improvement in velocity (measured in step length × (number of steps/min)) and a statistically significant improvement on the 10 m-walk-test (10MWT) in favour of the orthosis group (21.3(16.6) (mean(SD)) versus 22.9(16.6) for the group without orthosis, p = 0.011). We contacted the authors to get a better understanding of the results concerning gait speed. We decided to only include the results of the 10MWT because the authors pointed out that gait velocity was not measured validly due to the small laboratory. No significant changes in gait symmetry were found in the short-term.

So, according to the BES, in the short-term, limited evidence was found for the effectiveness of wearing a soft-hinged orthosis on knee hyperextension and gait speed compared to not wearing the orthosis.

3.4.2.2. Knee Ankle Foot Orthosis (KAFO) versus no orthosis. Boudarham et al. [6] (CCT, n = 11, fair quality) investigated the use of a KAFO in people 6-56 years post-stroke. All participants were able to walk 10 m without walking aids and exhibited spasticity or weakness in the quadriceps. Each participant performed two gait cycles: without KAFO (control condition), and - after a 10-min rest - with their own KAFO. In one session, the degree of knee hyperextension, gait speed and gait symmetry were measured. In the stance phase, the knee hyperextension (degrees) was significantly reduced with a KAFO as compared to no KAFO, although no effect size was reported: with KAFO versus without KAFO: -7.6 (7.4), (mean(SD)) versus -16.2(11.9), p = 0.029. The knee hyperextension was not totally resolved: the mean decrease of hyperextension with KAFO was around 8°. Gait speed (m/sec) increased significantly in the KAFO condition: with a KAFO versus without a KAFO 0.73(0.34) (mean(SD)) versus 0.57(0.36), p = 0.025. Gait symmetry was measured in both the swing phase and stance phase. In the KAFO condition, the symmetry between the paretic and the non-paretic limb increased significantly during the swing phase: asymmetry ratio with a KAFO versus without a KAFO 1.27 (0.10) (mean(SD)) versus 1.93 (0.77), $p\,=\,0.014.$ No significant difference between the two conditions was found in the stance phase.

In conclusion, we found limited evidence for short-term effectiveness in favour of a KAFO on knee hyperextension, gait speed and gait symmetry in the swing phase, although not in the stance phase, as measured immediately after putting the KAFO on.

3.4.3. Functional Electrical Stimulation (FES)

In FES, the electrical stimulation of muscles with poor nerve control is conducted to evoke a contraction and obtain functional movement.

3.4.3.1. Dorsiflexor FES versus 5° ankle-foot orthosis (AFO) and barefoot walking. Bae et al. [33] (CCT, n = 12, good quality) compared three walking conditions, consecutively barefoot walking, ankle-foot orthosis (AFO), and dorsiflexor FES in a cross-over trial in participants who were at least six months post-stroke. Comparing the three walking conditions in all participants, neither significant differences on the knee angle in midstance nor gait speed were found.

So, according to the BES, in the short-term (one session), we found no evidence for the effectiveness of dorsiflexor FES or 5° ankle dorsiflexor AFO on knee hyperextension in the stance phase or the gait speed as compared to barefoot walking.

^{+++,} strong evidence found; ++, moderate evidence found; +, limited evidence found.

^{+/-,} conflicting evidence found; NE, no evidence found for the effect of treatment; KAFO: knee ankle foot orthosis: AFO; Ankle foot orthosis.

4. Discussion

This systematic review studied the evidence for the effectiveness of interventions on knee hyperextension in post-stroke gait. Three types of interventions were identified: proprioceptive training, orthotic treatment, and FES. The eight included studies investigated the effectiveness of an intervention on knee hyperextension in the stance phase of gait. Seven studies also investigated the influence of a treatment on gait speed and three investigated gait symmetry. Only short-term effects were reported (from one day to eight weeks).

The adjustment of a form of proprioceptive training to physiotherapy training programs with the aim of keeping the knee flexed during exercises and walking seems to be effective (moderate evidence) on knee hyperextension in gait, in the short-term, as applied during the subacute phase post-stroke. Neither evidence for the effects on gait speed nor gait symmetry were found for proprioceptive training, although the adjustment of Whole-Body Vibration had some effect on gait speed (moderate evidence). Nevertheless, we would expect that the other studies on proprioceptive training included in this review would have found similar results on gait speed if gait speed was related to the results on knee hyperextension.

In the three studies we found on orthotic treatment, three different orthoses were studied. The highest level of evidence that could be reached was 'limited'. It seems that only orthoses that cover the knee had some effect on knee hyperextension and gait speed. On symmetry, no evidence was found, except for a KAFO in the swing phase.

No evidence was found for FES, although this conclusion was based only on one single study.

The benefits of proprioceptive training on knee hyperextension, as found in this review, were confirmed by the findings of two trials [36, 37] that we had to exclude: Basaglia et al. [36] included both people with stroke and intracranial pressure and found long-term benefits (one year) on knee hyperextension using biofeedback. McCain et al. [37] found better gait kinematics, gait speed and symmetry as well as an absence of knee hyperextension after gait training with a partial weight bearing system as applied before walking over ground was trained, in the sub-acute phase post-stroke.

Within this review, the study of Bae et al. [33] found no evidence for the benefit of a 5° dorsiflexor AFO on knee hyperextension. However, from a clinical point of view, we know that an AFO is an orthosis that is easy to put on with one hand. Positives signs for findings on other types of AFO in clinical practice can also be found in two other small studies: Kobayashi et al. [21] (n = 6) and Sutdet [38] (n = 4). These studies were excluded from our systematic review because of the small sample-size (n<10). Kobayashi et al. [21] (n = 6) found a significant reduction of knee hyperextension using an articulated AFO with adjustable plantar flexion resistance when the plantar flexion resistance increased. Further, Sutdet et al. [38] (n = 4) found a significant reduction of knee hyperextension using a Rigid Tuned AFO inclining the tibia angle from 0° to 15°. Therefore, in our opinion, it could be worthwhile to investigate the benefits of AFO types on a larger scale.

On FES, we could only include the study of Bae et al. [33]. Another study on FES from Springer et al. [39] also found no benefits of dorsiflexor FES on knee hyperextension. However, the latter study was excluded because only seven people with hemiparesis (of which six post stroke) were included. Nevertheless, and maybe interesting to study in future research, Springer et al. [39] found that dual channel FES, activating both the peroneal and hamstring muscles decreased knee hyperextension in this group.

Although many researchers [7,15,28,29,32] from the trials included in the current review described the problem 'increased energy cost while walking with knee hyperextension', none of them investigated the effect of the intervention on energy costs. Nevertheless, from a patient's perspective, energy cost in gait is an important factor in life after a stroke [40]. Higher energy costs of walking is a functional burden and might limit walking and daily activities related to walking post-stroke [41–43].

So, interventions on knee hyperextension that are beneficial to energy cost in gait might improve walking participation and the performance of daily activities.

Bleyenheuft et al. [7] concluded that none of the selected articles they included in their literature review (2010) considered the various causes of knee hyperextension. They described a proposal for a specific treatment strategy for each cause of knee hyperextension based on their clinical experience, in order to help identify the most appropriate treatment. From our point of view, a clinician should start with proprioceptive training, keeping in mind that in some cases, additional treatment might also be necessary to create better motoric conditions for preventing knee hyperextension. These additional interventions could include reducing spasticity or stimulating muscles using FES or even compensating for the deficits such as in orthotic treatment.

It is striking that, until now, in scientific literature no consensus can be found as to the different causes of knee hyperextension in people recovering from a stroke described. In 2012, researchers came to an agreement about the role of the posterior muscles of the lower leg (mm. triceps surae) [7,14,44,45] and weak hip extensor muscles [7,45] in causing knee hyperextension. The influence of over-activity or weakness of the knee extensors (m. rectus femoris) as stated by Blevenheuft [7] and Fish [45] was contradicted by the findings of Gross [12] and Cooper [44]. Gross [12] found that a reduction in the overactivity of the rectus femoris by a rectus femoris nerve blocker had no influence on knee hyperextension. Bleyenheuft [7] described the role of weak knee flexors (mm. hamstrings) failing to control knee flexion in the stance phase, causing knee hyperextension. This was not supported by the findings of Cooper [46]. Cooper [44] evaluated the association between muscle weakness and knee hyperextension. Weak hamstrings and quadriceps were found to play no role in knee hyperextension in gait. However, in the study of Lee [31] included in our review, researchers saw that the tested intervention had a significant effect of the quadriceps/hamstring muscle imbalance ratio. This may suggest that addressing the coordination between quadriceps and hamstring muscles could play a role in the normalization of the neuromuscular imbalance of gait.

Whether all cases of knee hyperextension should be treated in people recovering from a stroke remains a point of discussion. For functional recovery of the ability to walk, recovery of joint kinematics to support a normal walking pattern is not necessary [47]. Several authors described knee hyperextension as a useful adjustment developed to achieve limb stability while walking [13,45,47]. Different points of view may be caused by lack of mid- and long-term evidence for the occurrence of secondary knee pain. In general, in an active individual, the possible consequence of knee hyperextension after a stroke may be instability of the knee, which is caused by stretching the posterior ligaments and capsule, degenerative joint disease and chronic knee pain [10]. Younger or less severely impaired people with a more favourable walking ability might be more at risk and should definitely be treated, as well as people who are overweight and those who must maintain a standing position for a long time, due to the risk of overloading the knee joint [32,36]. However, treatment of knee hyperextension in individuals with a serious neurological outcome, poor walking autonomy, a reduced stance phase or a low physical activity level, preventing knee hyperextension to avoid knee pain might be less necessary, because there is a lower risk of overloading the knee joint in the case of incidental use. Tani et al. [13] suggest secondary knee pain might not be more common in people recovering from a stroke who suffer from knee hyperextension as compared to the general population. This supports the assumption that the need for treatment of the knee hyperextension depends on the individual stroke survivor.

The results of this review give us some indications that, initially, the clinician should attempt to restore knee control of the hyperextended knee by implementing a training program with a proprioceptive approach. Whole body vibration is an adjustment that seems beneficial to gait speed as well. It is unclear if the benefits are maintained in the long term. Proprioceptive training can be performed in both the (sub)

acute and chronic phases post stroke. In our opinion, in the cases where this retraining method is not successful, treatments might compensate for knee control by using and orthotic treatment, although it is not clear which orthosis is best. It should be taken into account that wearing an orthosis is not well-accepted by patients and often refused, for cosmetic reasons or due to discomfort [6,48]. In addition, putting on an orthosis is often difficult for a person recovering from a stroke due to a paretic arm. It should be taken into account that the adaptation of gait to the modifications (i.e. decreased hyperextension, increased range of motion of the ankle) is a long process [49] and thus, a period of training is required. FES in the case of muscle weakness or treatment aiming to reduce spasticity might be beneficial in individual cases, although we found no evidence for it in the present review.

To draw firm conclusions, more studies with larger sample sizes are needed. Future studies should also report mid- and long-term results in order to determine if the effects of the intervention are sustainable. The influence of these interventions on energy costs should be investigated. (Retrospective) research of mid-, and long-term effects of not treating knee hyperextension or non-adherence to orthosis would underline or deny the need for treating knee hyperextension in people experiencing secondary knee pain.

4.1. Study limitations

The current review is the first systematic literature review on the effectiveness of interventions on knee hyperextension in post-stroke gait. The review was conducted following the steps of the PRISMA-P 2015 statement [17]. However, some limitations should be addressed. Only five out of eight selected trials had a randomized study-design. Therefore, the results needed to be treated with caution because of the risk of bias in the analysis. In most studies, the sample sizes were low. Selection bias may have occurred due to omitting two full-text publications [26,27] that we had to exclude because the full text was not available [26], or was only available in Chinese [27]. A modified Downs and Black checklist [18] was used to assess the methodological quality. The ratings "excellent", "good", "fair" or "poor" are arbitrary, but nevertheless, frequently used in similar reviews [19,20,22,23]. A conflict of interest must be considered in the trial of Portnoy [30] as it was funded by an orthotic company.

5. Conclusion

Studies reporting on the effectiveness of proprioceptive training, orthotic treatment, and FES to treat knee hyperextension in post-stroke gait were found. Promising results (moderate evidence) were found for some "proprioceptive approaches" as an add-on therapy to physiotherapy training programs on knee hyperextension in the subacute phase post-stroke in the short-term. The effects of proprioceptive training on gait speed need further investigation; no evidence for benefits on gait-symmetry was found. For the effectiveness of orthotics and FES, only limited and no evidence in the short-term was found. None of the included studies reported on mid- and long-term effects. More high-quality studies concentrating on treating knee hyperextension in post-stroke gait, that not only report short-term, but also mid- and long-term effects are definitely needed.

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CRediT authorship contribution statement

Marieke Geerars: Conceptualization, Methodology, Validation, Formal analysis, Investigation, Writing - original draft. Nympha Minnaar-van der Feen: Formal analysis, Investigation, Writing - review

& editing. **Bionka M.A Huisstede:** Conceptualization, Methodology, Validation, Formal analysis, Writing - review & editing.

Declaration of Competing Interest

The authors report no declarations of interest.

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Appendix A. Supplementary data

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