J Child Orthop (2010) 4:277–290
DOI 10.1007/s11832-010-0268-4

ORIGINAL CLINICAL ARTICLE

Surgical correction of equinus deformity in children with cerebral palsy: a systematic review

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Received: 3 February 2010 / Accepted: 22 May 2010 / Published online: 1 July 2010

Abstract
Purpose Equinus is the most common deformity in cerebral palsy. However, despite the large volume of published studies, there are poor levels of evidence to support surgical intervention. This study was undertaken to examine the current evidence base for the surgical management of equinus deformity in cerebral palsy.
Methods A systematic review of the literature using "equinus deformity", "cerebral palsy" and "orthopaedic surgery" generated 49 articles. After applying inclusion and exclusion criteria, 35 articles remained. The Oxford Centre for Evidence-Based Medicine (CEBM) levels of evidence and the Methodological Index for Non-Randomized Studies (MINORS) were used to grade the articles.
Results Studies ranged in sample size from 9 to 156 subjects, with an average of 38 subjects. The mean age of subjects at index surgery ranged from 5 to 19 years. Nineteen studies used instrumented gait analysis, with an average follow-up of 2.8 years. Seven studies reported that a younger age at index surgery was associated with an increased risk of recurrent equinus. The average rate of calcaneus in hemiplegic children was 1% and it was 15% in those with spastic diplegia. Most studies were level 4 quality of evidence, leading to, at best, only grade C recommendation.
Conclusions Cerebral palsy subtype (hemiplegia versus diplegia) and age at index surgery were the two most important variables for determining the outcome of surgery for equinus deformity in cerebral palsy. Despite the great emphasis on differences in surgical procedures, there was less evidence to support the type of operation in relation to outcome.

Keywords Cerebral palsy · Equinus · Systematic review · Calcaneus

Introduction

Equinus is a very common deformity in children with cerebral palsy [1]. Equinus is defined as the inability to dorsiflex the foot above plantigrade, with the hindfoot in neutral and the knee extended [2]. However, in the literature, there exist many definitions for both equinus deformity and equinus gait, which contribute to the difficulty in interpreting the results on this deformity.

There is extensive literature on the management of equinus deformity in cerebral palsy by both operative and non-operative measures. Apart from the large number of original papers, there are a number of review articles, of which two are noteworthy. Goldstein and Harper [3] published the results of a multi-disciplinary workshop convened to explore the current state of knowledge, best clinical practice and research needs for the management of equinus gait associated with cerebral palsy. This report compared the evidence base for physical therapy, orthoses, casting, botulinum toxin A and surgery across the domains of treatment, which had been suggested by Condie and

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Meadows and reported on by Stuberg [4]. Condie and Meadows suggested that the aims of the management of equinus in cerebral palsy were the prevention of deformity, correction of deformity, the promotion of a base of support, the facilitation of training skills and an improvement in the efficiency of gait. When these aims of treatment were considered in the context of evidence-based medicine, botulinum toxin A was noted to have level 1 or level 2 evidence, whereas surgery had consistently level 4 and level 5 evidence according to the Oxford Centre of Evidence-Based Medicine (CEBM) levels of evidence [5].

In a more recent and traditional review of surgical studies, Koman et al. [6] reviewed the recurrence of equinus deformity in cerebral palsy patients following surgery. This useful review of 31 published articles from 1938 to 2001 focused mainly on the issue of recurrent equinus deformity. The primary conclusion from this review was that age at first surgery is the greatest predictor of recurrent equinus. For this reason, temporising measures such as the use of botulinum toxin A, casting and orthoses should be considered in younger children to postpone the age at first surgery for as long as possible.

Central to discussion of the surgical management of equinus is the issue of recurrent deformity, the need for additional surgery, as well as the opposite outcome, over-correction, calcaneus deformity and subsequent crouch gait. Detection of these dichotomous outcomes requires studies of adequate power and appropriate duration, with sensitive and objective outcome measures. It is now recognised that the cerebral palsy type is considered to be of great importance in determining the outcome of equinus surgery in children. Cerebral palsy can be classified in the domains of movement disorder, topographical distribution and gross motor function [7]. Historically, studies on equinus surgery have grouped heterogenous patients together, thus, compromising the conclusions of equinus surgery. Single-event multi-level gait improvement surgery (SEMLS) has become the standard of care in many institutions for the treatment of walking children with cerebral palsy [8–11]. In this era of SEMLS surgery, equinus surgery is rarely performed in isolation and more commonly performed in concert with other surgical procedures designed to correct lever arm dysfunction, improve the biomechanics of the plantar flexion/knee extension couple and correct fixed musculoskeletal contractures.

However, despite the large volume of published studies, the review by Koman et al. [6] and the annotation by Goldstein and Harper [3] both found very poor levels of evidence to support surgical intervention for equinus deformity. The complexity and evolving nature of cerebral palsy in children, coupled with the difficulties of designing and conducting randomised clinical trials, have led to a paucity of robust evidence to guide surgical practice. This study was undertaken to systematically examine the current evidence base for the surgical management of equinus deformity in children with cerebral palsy.

Methods

Search strategy

In March 2008, an electronic search was performed of MEDLINE (1966–March 2008), EMBASE (1980–2006) and the Cochrane Database. Keywords used in the search strategy included ‘cerebral palsy’, ‘equinus’, ‘ankle’, ‘achilles’, ‘gastrocnemius’, ‘gait analysis’, ‘contracture’ and ‘calf’. Key terms were matched to Medical Subject Headings (MeSH) index and exploded or searched as keywords. Surgical procedures identified as being used in the correction of equinus deformity were also searched as keywords. Targeted searching was undertaken from the reference lists of the articles included for review and other key articles concerning the topic. In addition, to improve our search yield, the tables of contents from key paediatric orthopaedic journals were hand-searched over the last 10 years.

Inclusion and exclusion criteria

Studies were shortlisted for inclusion if they pertained to the surgical management of equinus within a paediatric population with cerebral palsy and were available in English. Where doubt existed, the article was shortlisted. Additional inclusion and exclusion criteria were then applied to shortlisted articles. Articles were excluded when they examined a novel surgical intervention or had fewer than ten cases which were followed up. Articles included for review had a mean follow-up period of greater than 5 years, or used instrumented gait analysis, with a mean follow-up of greater than 12 months.

Data extraction

Included articles were examined in depth and key data extracted with the use of a custom data extraction form. This form focused on themes of sample characteristics, surgical intervention, outcome measures, data handling, follow-up and complication rates. The Oxford CEBM levels of evidence [5] was used to grade the selected studies. However, in surgical research, randomisation is not always possible or feasible [12], and in this review, all included articles were non-randomised. Few validated instruments are available to determine the methodological quality of observational or non-randomised studies, either from the reader's perspective or for the purpose of meta-analysis. As a result, the Methodological Index for Non-Randomized Studies (MINORS) [13] was applied to each article to further assess the quality of each study. This validated index involves 12 items, the first eight items specifically designed for non-comparative studies and the remaining four items applied to comparative studies. Items are scored as 0 (not reported), 1 (reported but inadequate) and 2 (reported and adequate). The maximum ideal score for non-comparative studies is 16 and for comparative studies, it is 24.

Results

Search strategy yield

The initial electronic search identified 6,905 articles for consideration, from which a shortlist was generated. From this, two authors identified 49 possible inclusions for review. These were examined in depth. After applying inclusion and exclusion criteria, 35 studies were included (Table 1) [2, 11, 14–46].

Descriptive aspects of the reviewed studies

The study sample size ranged from 9 to 156 subjects [31], with an average of 38. It should be noted that, in some papers, it was difficult to identify an accurate sample size. Occasionally, the number of "lower limbs" rather than the number of subjects were reported: given that hemiplegic children are often managed by unilateral surgery and diplegic children can be managed by either unilateral or bilateral surgery, it was difficult to establish an accurate sample size [16]. In these cases of ambiguity, it was left to the reviewer's discretion to estimate an accurate sample size. This ambiguity in sample size is another mark of the low quality of many of the studies. The mean age of subjects ranged from 4.8 to 19 (mean 8.7) years, with many studies reporting a wide age range at index surgery. Gender demographics were included sporadically within the reviewed articles, with several omitting this potentially important information.

Operative intervention

The surgical intervention was not always clearly identified by either a generic term or an eponym. Some studies reported on a single clearly described operative procedure, such as the White slide tendo-Achilles lengthening [25]. Others compared multiple procedures and in these studies, there was a tendency to refer to the original operative description [17]. Some articles specified modifications of earlier procedures [30], while other studies reported on combinations of surgical procedures, such as the Strayer (distal gastrocnemius recession) combined with slide lengthening of the Achilles tendon [18]. For clarification and consensus, we defined an "intramuscular lengthening" as a Baumann procedure, a "gastrocnemius recession" as a Strayer procedure and a "gastrocsoleus aponeurotic lengthening" as a Vulpius procedure (Fig. 1).

From proximal to distal, the procedures that were included in the reviewed articles were the Baumann procedure, the Strayer (distal gastrocnemius recession), the Baker and Vulpius (gastrocsoleus lengthening), the White and Hoke slide (tendo-Achilles) lengthenings (percutaneous and open), open Z tendo-Achilles lengthening (TAL) and heel cord advancement (HCA). The operative procedures are illustrated in relation to three specific zones of the triceps surae (Fig. 1). Zone 1 is from the gastrocnemius origin and ends at the most distal fibres of the medial belly of the gastrocnemius. The operative procedures in Zone 1 include the Baumann and the Strayer distal gastrocnemius recession. Zone 2 is from the distal gastrocnemius belly to the end of the soleus muscle fibres and includes the Baker and Vulpius gastrocsoleus aponeurotic lengthenings. Zone 3 is the Achilles tendon and includes all forms of lengthening of the Achilles tendon, as well as HCA.

Postoperative care

The immediate postoperative care was well described in most studies. The older literature reported above-knee casts for at least 6 weeks with long periods of restricted weight-bearing [16]. The more recent literature reports the use of below-knee casts and earlier weight-bearing [2, 14, 15, 20, 21, 24, 28, 29, 37]. The utilisation of ankle–foot orthoses (AFO) after cast removal, the type of AFO and the utilisation of night splints varied greatly and were poorly described in several studies.

Outcome assessment

The outcome measures reported included clinical examination and rating scales [2, 11, 14–46], gait rating scales [2, 14, 16–18, 20–22, 24, 25, 27, 29, 30, 32, 33, 36, 39, 42], goniometry [2, 14, 17, 21, 25, 36, 44], three-dimensional gait analysis [2, 11, 14, 15, 17, 21–24, 28, 29, 31, 32, 35, 37–39, 41, 44, 46], the modified Ashworth scale (spasticity) [2, 11, 36, 37], radiographs [17, 32, 45], electromyography [11, 15, 23, 24, 35, 38], the Gross Motor Function Measure (GMFM) [14, 21] and the need for floor reaction AFOs to manage crouch gait [20, 26]. Such measures were used to derive an outcome scale, which rated the overall result from good to poor. The duration of mean follow-up varied considerably between studies, from 1 to 14 (mean

Table 1 Description of the 35 articles fulfilling the criteria for systematic review

Author	Surgical procedures	SEMLS	No. of subjects	Age (years) mean (range)	Gender M, F	Cerebral palsy type: H, D, Q	Outcome measures	Follow-up (years/range)	OX LOE	MINORS score
Abel et al. [14]	Vulpius	Yes	30	8.7 (4–20)	–	30 D	3DGA, GMFM, goniometry	2 (1.6–3.1)	4	10
Adolfsen et al. [11]	Vulpius, TAL (NS)	Yes	31	8.5 (5–15)	–	10 H, 20 D, 1 Q	3DGA, EMG, clinical exam	1.9 (0.7–6.4)	4	6
Baddar et al. [15]	Vulpius	No	34	7.2 (2.6–15.2)	17 M, 17 F	34 D	3DGA/EMG, clinical exam	1.3 (0.8–1.9)	4	8
Banks [16]	White TAL	No	200 feet	–	–	–	OGA	10 (–)	4	3
Borton et al. [17]	Hoke TAL (Perc), Z TAL, Baker	No	134	7.6 (2–18)	78 M, 56 F	45 H, 65 D, 24 Q	3DGA, PRS, goniometry	6.9 (5–10)	4	11
Craig and van Vuren [18]	Strayer ± White TAL	No	59	6 (1–15)	–	–	Clinical exam	6 (–)	4	8
Damron et al. [19]	Z TAL, Baker, Vulpius	No	59	4.8 (2–11)	31 M, 28 F	2 H, 18 D, 28 Q	Goniometry, functional scale	7 (–)	3b	9 (14)
Dietz et al. [20]	Hoke TAL (O), Z TAL	Yes	79	7.7 (2–47)	–	23 H, 34 D, 15 Q	OGA, GRAFO	7 (–)	4	11
Engsberg et al. [21]	HCA, Vulpius, White TAL	No	32	7 (–)	19 M, 13 F	32 D (GMFCS I)	Goniometry, 3DGA, GMFM	1 (–)	4	11
Fabry et al. [22]	TAL (O) (NS), TAL (P) (NS)	No	15	15.7	8 M, 7 F	15 D	2DGA, clinical exam	9.5	4	4
Gough et al. [23]	Strayer	Yes	12	9.8	8 M, 4 F	12 D	3DGA, EMG, clinical exam, functional status	1.5 (–)	3b	9 (15)
Gough et al. [24]	Strayer	Yes	13	6.4 (5.9–6.9)	–	13 D	3DGA, GGI, EMG, clinical exam	2 (2–5)	3b	10 (16)
Graham and Fixsen [25]	White TAL	No	35	6.9 (3–15)	18 M, 17 F	35 H	Goniometry, VRS	13 (9–16)	4	9
Javors and Klaaren [26]	Vulpius	No	47	6.8 (2–14)	–	15 H, 23 D, 9 Q	Clinical exam	5.7 (1–12)	4	3
Kay et al. [2]	Strayer, TAL (NS)	Yes	54	9.8 (4–16)	28 M, 27 F	23 H, 26 D, 6 Q	3DGA, PRS, Ashworth	1.5 (1–2)	4	8
Lofterød and Terjesen [27]	Strayer, Z TAL	No	15	8.8 (6–14)	8 M, 7 F	6 H, 9 D (GMFCS I + II)	3DGA, clinical exam	3 (1.1–4.6)	4	6
Lofterød and Terjesen [28]	Strayer	Yes	47	11.5 (5–19)	29/18	13 H, 34 D (GMFCS I + II + III)	3DGA exam, GMFCS, FMS	1.2 (0.8–1.5)	4	8
Lyon et al. [29]	Z TAL, Hoke TAL	Yes	14	9.1 (4–17)	6 M, 8 F	14 D	3DGA	1.5 (0.5–2.5)	4	6
Olney et al. [30]	Baker	No	156	5.5 (2–14)	–	90 H, 108 D, 21 Q	Clinical exam, recurrence	7.5 (3–14)	4	9
Orendurff et al. [31]	TAL (NS)	No	9	–	–	–	3DGA, muscle length	1 (–)	4	5
Park et al. [32]	Vulpius, Z TAL (NS)	No	16	8 (3–16)	11 M, 5 F	16 H	3DGA	1.25 (0.75–2)	4	5
Pierrot and Murphy [33]	HCA, White TAL, Z TAL	No	41	7 (3–10)	23 M, 18 F	16 H, 9 D, 13 Q	VRS	6 (3–10)	4	9
Rattey et al. [34]	Z TAL	No	57	5.4 (1.4–14.7)	–	27 H, 30 D	Clinical exam, recurrence	9.6 (5–15)	4	9
Rose et al. [35]	Baker	Yes	20	6 (4–26)	–	5 H, 15 D	Clinical exam, 3DGA	1 (–)	4	7
Sala et al. [36]	Hoke TAL	No	27	5 (2–10)	15 M, 12 F	4 H, 17 D, 6 Q	Clinical exam, VRS, MAS	(2–9)	4	9
Saraph et al. [37]	Baumann	Yes	22	12.6 (7.4–16.6)	–	22 D	Clinical exam, 3DGA	2 (2.1–4)	4	5
Saraph et al. [38]	Baumann	Yes	32	11.1 (8.7–13.5)	–	32 D	Clinical exam, 3DGA, EMG	4.4 (1.0–5.4)	4	9

Table 1 continued

Author	Surgical procedures	SEMLS	No. of subjects	Age (years) mean (range)	Gender M, F	Cerebral palsy type: H, D, Q	Outcome measures	Follow-up (years/range)	OX LOE	MINORS score
Segal et al. [39]	Hoke TAL (P), White TAL, Z TAL, HCA, Vulpius	Yes	20	5.2 (2.7–8.2)	9 M, 11 F	20 D	Clinical exam, 3DGA	5.8 (1.1–11)	4	11
Sharrard and Bernstein [40]	Z TAL, Strayer	No	92	7 (–)3	–	45 H, 20 D, 27 Q	Clinical exam, OGA	8.9 (–)	4	7
Steinwender et al. [41]	Baumann	Yes	29	14.4 (–)	–	17 D	Clinical exam, 3DGA	3.9 (–)	3b	5 (13)
Truscelli et al. [42]	TAL (NS), Vulpius	No	58	–	–	–	Clinical exam	14 (2–27)	4	4
Uyttendaele et al. [43]	Z TAL, Scholder	No	32	19 (14–30)	–	13 H, 19 D/Q	Clinical exam, OGA	–	4	5
Yngve and Chambers [44]	Z TAL, Vulpius	Yes	33	9 (–)	–	14 H, 17 D, 2 Q	Clinical exam, 3DGA	1 (–)	4	8
Yoshimoto et al. [45]	HCA ± Vulpius	No	17	10 (5–17)	9 M, 8 F	11 H, 6 D	Clinical exam, OGA, X-ray	8 (5.7–9.11)	4	11
Zwick et al. [46]	Strayer	Yes	17	11.2 (5.7–16.4)	–	17 D	Clinical exam, 3DGA	3.8 (2.6–5.7)	4	9

OX LOE Oxford Levels of Evidence; *HCA* heel cord advancement; *G/S* gastrocsoleus; *3DGA* 3D gait analysis; *VRS* visual rating scale; *MAS* modified Ashworth scale; *OGA* observational gait analysis; *PRS* Physician Rating Scale

4.9) years. For the purposes of this review, studies with less than 5 years of clinical follow-up were only considered if they used instrumented gait analysis. Nineteen studies used instrumented gait analysis, with an average follow-up of 2.8 years. Many studies reported rates for recurrent equinus deformity or calcaneus gait/deformity. However, the definitions for both equinus/calcaneus gait and equinus/calcaneus deformity varied from study to study and were not always clearly stated.

Recurrent equinus deformity and type of surgery

Twelve of the 35 studies stated explicit criteria for the definition of equinus deformity and/or recurrent equinus [2, 15, 17–19, 25, 27, 34, 36, 39, 40, 44]. Definitions for equinus deformity were either based on clinical examination and/or sagittal ankle kinematic data. In an attempt to increase the relevance of this heterogenous body of evidence, the results were grouped according to the surgical zone (Fig. 1). Where results by sub-group were not specifically stated and could not be confidently derived from the presented data, they were indicated to be not available. The rate of recurrent equinus deformity for procedures involving lengthening of the Achilles tendon was given in 13 studies [2, 16, 17, 19, 25, 27, 29, 33, 34, 36, 40, 42, 43] (Table 2). Seven studies reported on the rates of recurrent equinus deformity following surgical lengthening in Zone 2 (the Baker and Vulpius procedures) [14, 17, 19, 26, 30, 42, 45] (Table 3). Eleven studies reported a Zone 1 procedure, comprising three Baumann and eight Strayers. Six studies reported on the rates of recurrent equinus following surgery [2, 18, 24, 27, 37, 40] (Table 4). No recurrent equinus was found at a mean follow-up of just over 2 years. The only other Zone 1 procedure, the Strayer distal gastrocnemius recession, was reported in combination with Achilles tendon lengthening or in sub-groups of studies reporting multiple surgical procedures.

The overall recurrence rates varied from 0 to 43%, but this increased up to 62% when specific sub-groups were considered (hemiplegia compared to diplegia). The majority of studies reporting three-dimensional gait analyses had short-term follow-up and reported much lower rates for recurrent equinus deformity than the clinical studies, which all included a mean follow-up of at least 5 years [27, 29, 31, 37, 41, 44].

Calcaneus deformity, calcaneus gait and crouch gait by topographical distribution

Nine of the 35 studies reported explicit criteria for calcaneal gait, calcaneal deformity or crouch gait [2, 17, 19, 20, 22, 27, 38, 39, 44]. These definitions included the inability to passively plantarflex the foot to neutral, greater than 14°

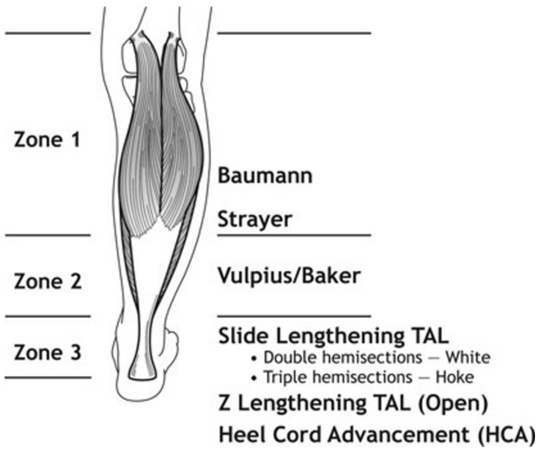

Fig. 1 Anatomic zones of the gastrocsoleus complex. The gastrocsoleus complex has three discrete anatomic zones, which are illustrated here with the corresponding surgical procedures associated with each zone

of ankle dorsiflexion in stance or 10° of ankle dorsiflexion greater than the normal, as a definition of calcaneus deformity and the need for a floor reaction orthosis for crouch gait. In terms of kinematic criteria, peak dorsiflexion in stance more than two standard deviations above the reference range was quoted in several studies and in kinetic terms, reduced push-off or A_2 power generation was also quoted. Using these variable terms, the rates of calcaneus varied from 0 to 36% overall, but it was up to 80% in specific sub-groups.

Calcaneal deformity was seen postoperatively in all three surgical zones, with slightly differing rates of occurrence. Twelve studies reported the rate of calcaneus for operative procedures in Zone 3 [2, 17, 19, 20, 25, 27, 29, 33, 34, 39, 40, 42]. One study reported on the development of a spastic flat foot and knee flexion deformity as a postoperative complication [43] (Table 2). Nine studies reported the rate of calcaneus following surgical procedures in Zone 2 [14, 15, 17, 19, 26, 30, 39, 42, 45] (Table 3). Five studies reported the rate of calcaneus following Zone 1 surgical procedures [2, 18, 27, 37, 40] (Table 4). The report by Saraph et al. [37] on the Baumann procedure reported no calcaneus in 22 children and adolescents with spastic diplegia who had multi-level surgery, including a Baumann gastrocsoleus lengthening.

Varying rates of calcaneus were seen between hemiplegic and diplegic children. Twelve studies reported the rates of calcaneal gait in diplegic children [17, 19–21, 26, 29, 30, 34, 38, 39, 44, 45]. Kay et al. [2] reported no significant difference in the rates of calcaneus gait in children with spastic diplegia who had either a gastrocnemius recession or an Achilles tendon lengthening (Table 4). Thirteen studies reported the rates of calcaneus gait in hemiplegic children [14, 15, 17, 19, 20, 26, 29, 30, 34, 38–40, 45]. Again, Kay et al. [2] found no significant difference in calcaneus gait in hemiplegic children who had either a gastrocnemius recession or a lengthening of the Achilles tendon.

Effect of age at surgery on the rates of recurrent equinus and calcaneus

The effect of age at index surgery on the rates of recurrent equinus or calcaneus was reported in 10 of the 35 studies [17–19, 25, 30, 34, 36, 39, 40, 43]. Damron et al. [19] found no clear relationship between age and outcome. Seven studies reported that a younger age at index surgery was associated with an increased risk of recurrent equinus, but one study found that age was not related to recurrent equinus [36]. One study [39] utilising gait analysis found that velocity and stride length improved most in younger patients. Twenty-five of the 35 studies made no comment on the effect of age on either the rate of recurrent equinus or calcaneus.

Outcome and topographical distribution of cerebral palsy

The type of cerebral palsy was, in general, poorly described. Very few studies reported the type of movement disorder, not all included such basic information as the topographical distribution and only five referred to the Gross Motor Function Classification System (GMFCS) [14, 21, 24, 27, 28]. Studies that gave a breakdown of either movement disorder or topographical distribution frequently utilised out-of-date classifications.

Thirteen studies reported on the rate of recurrent equinus deformity in children with spastic diplegia [14, 17, 24, 26, 27, 29, 30, 34, 36, 37, 40, 43, 45]. Additionally, the study by Kay et al. [2], whilst not providing an overall recurrence rate in spastic diplegia, reported no significant difference in recurrence rates when children with diplegia who had gastrocnemius recession were compared to those who had tendo-Achilles lengthening (Table 5).

Ten studies reported on the rates of recurrent equinus deformity in children with hemiplegia [2, 17, 25–27, 30, 34, 36, 43, 45] and this information is summarised in Table 6. Again, Kay et al. [2] reported no difference in the rate of recurrent equinus deformity in children with hemiplegic cerebral palsy who had either gastrocnemius recession or lengthening of the Achilles tendon. However, in this study, the choice of surgical intervention (TAL or GR) was determined by the patient's preoperative degree of equinus. This study bias may explain the observed results.

Fifteen studies recorded results of equinus surgery in the setting of SEMLS (Table 7). On average, seven surgical procedures were performed on each patient, including calf

Table 2 Zone 3: Achilles tendon lengthening: recurrent equinus and calcaneus

Author	Procedure	Feet	Hemis	Dis	Quads	Recurrent equinus (%)	Calcaneus (%)	Follow-up <5 years	3DGA
Adolfsen et al. [11]	TAL (NS)	6	–	–	–	–	–	x	y
Banks [16]	White TAL	200	–	–	–	27	–		
Borton et al. [17]	Z TAL	40	–	–	–	23	35		y
	Hoke TAL (P)	98				20	39		
Damron et al. [19]	Z TAL	80	–	–	–	2	2		
Dietz et al. [20]	Hoke TAL (O)	56	–	–	–	–	43		
	Z TAL	56				–	53		
Engsberg et al. [21]	White TAL	10	0	10	0	–	–	x	y
Fabry et al. [22]	TAL (O) (NS)	11	0	15	0	–	–		y
	TAL (P) (NS)	6							
Graham and Fixsen [25]	White TAL	35	35	0	0	43	0		
Kay et al. [2]	TAL (NS)	17	11	4	2	29	36	x	y
Lofterød and Terjesen [27]	Z TAL	10	2	8	0	0	0	x	y
Lyon et al. [29]	Z TAL	21	0	12	0	35	Most*	x	y
	Hoke TAL	2		2					
Orendurff et al. [31]	TAL (NS)	12	–	–	–	–	–	x	y
Park et al. [32]	Z TAL (NS)	13	13	0	0	–	–	x	y
Pierrot and Murphy [33]	White TAL	9	7	4	5	46	0		
	Z TAL	13							
Rattey et al. [34]	Z TAL	77	29	34	0	26	5		
Sala et al. [36]	Hoke TAL	27	4	17	6	22	–		
Segal et al. [39]	Hoke TAL (P)	12	0	6	0	–	25		y
	White TAL	6		3			25		
	Z TAL	12		7			25		
Sharrard and Bernstein [40]	Z TAL	77	37	12	24	23	0		
Truscelli et al. [42]	TAL (NS)	72	–	–	–	35	3		
Uyttendaele et al. [43]	Z TAL	25	10	15	0	20	–		
Yngve and Chambers [44]	Z TAL	27	11	16	0	–	–	x	y

*Avg difference in stance 24.1

x = <5 years average follow-up

y = 3D gait analysis examination

lengthening. The overall trends demonstrated an increase in ankle dorsiflexion through initial contact and a decrease in the degree of knee flexion at initial contact.

Quality of evidence

The majority of studies were level 4 evidence according to the Oxford CEBM [5] (Table 1). Only four studies met the criteria for level 3b evidence by demonstrating a comparable control group for comparison. Because the majority of the studies in this review were of poor methodological quality, the MINORS index was applied to each study. The MINORS index is a valid instrument designed to assess the methodological quality of non-randomised surgical studies, whether comparative or non-comparative. In this review, MINORS scores ranged from 3 to 16, demonstrating a significant amount of methodological heterogeneity amongst studies.

Discussion

Cerebral palsy type and outcome

Given that the rate of calcaneal deformity and calcaneal gait in children with spastic hemiplegia averaged only 1% (range 0–7%) compared to 15% (range 0 to 41%) in children with spastic diplegia, it would appear that the topographical distribution of cerebral palsy may well be the strongest determinant of outcome. When considering rates of over-correction and calcaneal deformity, the length of follow-up is paramount to the discussion, as the highest

Table 3 Zone 2: mid-calf gastrocsoleus recession

Author	Procedure	Feet	Hemis	Dis	Quads	Recurrent equinus (%)	Calcaneus (%)	Follow-up <5 years	Gait analysis
Abel et al. [14]	Vulpius	42	0	21	0	0	0	x	y
Adolfsen [11]	Vulpius	33	–	–	–	–	–	x	y
Baddar [15]	Vulpius	22	0	11	0	–	0	x	y
Borton et al. [17]	Baker	57	–	–	–	26	32		y
Damron et al. [19]	Baker, Vulpius	38	–	–	–	12	0		
Engsberg et al. [21]	Vulpius	10	0	10	0	–	–	x	y
Javors and Klaaren [26]	Vulpius	79	15	46	18	4	4		
Olney et al. [30]	Baker	219	80	108	21	48	0		
Park et al. [32]	Vulpius	3	3	0	0	–	–	x	y
Rose et al. [35]	Baker	24	5	15	0	–	–	x	y
Segal et al. [39]	Vulpius	2	0	2	0	–	0		
Truscelli et al. [42]	Vulpius	22	–	–	–	23	36		
Yngve and Chambers [44]	Vulpius	22	3	16	3	–	–	x	y
Yoshimoto et al. [45]	Vulpius (+HCA)	20	11	6	0	0	0		

x = <5 years average follow-up

y = 3D gait analysis examination

Table 4 Zone 1: proximal calf gastrocnemius recession

Author	Procedure	Feet	Hemis	Dis	Quads	Recurrent equinus (%)	Calcaneus (%)	Follow-up <5 years	Gait analysis
Craig and van Vuren [18]	Strayer	100	–	–	–	9	3		
Gough et al. [23]	Strayer	22	0	12	0	–	–	x	y
Gough et al. [24]	Strayer	21	0	12	0	25	–	x	
Kay et al. [2]	Strayer	38	12	22	4	19	22	x	y
Lofterød and Terjesen [27]	Strayer	10	4	6	0	10	10	x	y
Lofterød and Terjesen [28]	Strayer	42	–	–	–	–	–	x	y
Saraph et al. [37]	Baumann	28	0	22	0	0	0	x	y
Saraph et al. [38]	Baumann	23	0	32	0	–	–	x	y
Sharrard and Bernstein [40]	Strayer	53	5	25	23	15	2		
Steinwender et al. [41]	Baumann	34	0	17	0	–	–	x	y
Zwick et al. [46]	Strayer	11	0	6	0	–	–	x	y

x = <5 years average follow-up

y = 3D gait analysis examination

rates of calcaneus gait were seen in studies with longer clinical follow-up [17, 20, 34, 39, 42]. Several of the studies utilising instrumented gait analysis also reported high rates of calcaneus gait [2, 17, 39]. The outcomes in terms of recurrent equinus versus calcaneus in children with hemiplegia versus children with diplegia are so different as to merit separate studies.

This review demonstrates that there is very little information on the type of movement disorder affecting children with cerebral palsy. It is very likely that a number of children with dystonia or mixed movement disorders have been included in these studies, but who have not been clearly identified. The outcome of surgery, including surgery for equinus deformity, in children with dystonia and mixed movement disorders is usually considered to be more unpredictable but no information from the reviewed studies could corroborate this hypothesis [47].

The choice of studies for this review in two main groups, those with a mean follow-up of more than 5 years and those with instrumented gait analysis, give two quite different perspectives on both recurrent equinus deformity and calcaneus gait. Gait analysis is expensive and the majority of studies report a preoperative and a single postoperative study, usually within 1–2 years after surgery. Although the kinematic parameters are objective and sensitive in the definition of under-correction and over-

Table 5 Equinus surgery outcomes in spastic diplegia

Author	Procedure	Diplegic feet	Recurrent equinus (%)	Calcaneus (%)	Follow-up <5 years	Gait analysis
Abel et al. [14]	Vulpius	42	0	0	x	y
Adolfsen et al. [11]	Vulpius, TAL (NS)	20	–	–	x	y
Baddar et al. [15]	Vulpius	22	–	0	x	y
Borton et al. [17]	Baker, Z TAL, Hoke TAL (P)	110	16	40		y
Damron et al. [19]	Baker, Vulpius, Z TAL	37	–	0		
Dietz et al. [20]	Hoke TAL (O), Z TAL	34 pts	–	41		
Engsberg et al. [21]	HCA	12	–	–	x	y
	Vulpius	10				
	White TAL	10				
Fabry et al. [22]	TAL (O) (NS)	11	–	–		y
	Z TAL (P) (NS)	6				
Gough et al. [23]	Strayer	22	–	–	x	y
Gough et al. [24]	Strayer	22	25	–	x	y
Javors and Klaaren [26]	Vulpius	46	0	0		
Kay et al. [2]	Strayer	22	–	–	x	y
	TAL (NS)	4				
Lofterød Terjesen [27]	Strayer	14	7	–	x	y
	Z TAL					
Lofterød Terjesen [28]	Strayer	34	–	–	x	y
Lyon et al. [29]	Hoke TAL, Z TAL	23	35	High*	x	y
Olney et al. [30]	Baker	108	44	0		
Rattey et al. [34]	Z TAL	56	18	2		
Rose et al. [35]	Baker	19	–	–	x	y
Sala et al. [36]	Hoke TAL	17	24	–		
Saraph et al. [37]	Baumann	28	0	0	x	y
Saraph et al. [38]	Baumann	23	–	–	x	y
Segal et al. [39]	Vulpius	2	–	30		y
	Hoke TAL (P)	12				
	Z TAL	12				
	White TAL	6				
	HCA	6				
Sharrard and Bernstein [40]	Z TAL	12	33	0		
	Strayer	25	4	4		
Steinwender et al. [41]	Baumann	34	–	–	x	y
Uyttendaele et al. [43]	Z TAL	15	33	–		
Yngve and Chambers [44]	Vulpius	16	Infrequent	Infrequent	x	y
	Z TAL	16				
Yoshimoto et al. [45]	Vulpius ± HCA	6	0	0		
Zwick et al. [46]	Strayer	11	–	–	x	y

* Average difference in stance 24.1

y = 3D gait analysis examination

correction, the brevity of follow-up dramatically weakens the value of these studies. The one study which combines instrumented gait analysis and a medium-term follow-up is that by Segal et al. [39], who reported a 30% prevalence of calcaneal gait, but there were only 20 children in their study and there were five different combinations of surgical procedures [39].

The two clinically based outcome studies with the longest follow-up and the largest number of cases of children with diplegia were the studies by Borton et al. [17] and

Table 6 Equinus surgery outcomes in spastic hemiplegia

Author	Procedure	Hemis	Recurrent equinus (%)	Calcaneus (%)	Follow-up <5 years	Gait analysis
Adolfsen et al. [11]	Strayer, TAL (NS)	10	–	–	x	y
Borton et al. [17]	Baker, Z TAL, Hoke TAL (P)	45	38	4		y
Damron et al. [19]	Baker, Vulpius, Z TAL	4	–	0		
Dietz et al. [20]	Hoke TAL (O), Z TAL	23	–	0		
Graham and Fixsen [25]	White TAL	35	43	0		
Javors and Klaaren [26]	Vulpius	15	4	0		
Kay et al. [2]	Strayer	12	–	–	x	y
	TAL (NS)	11				
Lofterød and Terjesen [27]	Strayer, Z TAL	6	0	–	x	y
Lofterød and Terjesen [28]	Strayer	13	–	–	x	y
Olney et al. [30]	Baker	90	62	0		
Park et al. [32]	Z TAL (NS)	13	–	–	x	y
	Vulpius	3				
Rattey et al. [34]	Z TAL	29	41	7		
Rose et al. [35]	Baker	5	–	–	x	y
Sala et al. [36]	Hoke TAL	4	50	–		
Sharrard and Bernstein [40]	Z TAL	37	27	0		
	Strayer	5	40	0		
Uyttendaele et al. [43]	Z TAL	10	20	–		
Yngve and Chambers [44]	Vulpius	3	–	–	x	y
	Z TAL	11				
Yoshimoto et al. [45]	Vulpius + HCA	11	0	0		

y = 3D gait analysis examination

Dietz et al. [20]. Borton et al. reported risk factors for both recurrent equinus and calcaneus in a large heterogenous population of children with cerebral palsy who were followed clinically for an average of 7 years (range 5–10 years). They found a very high incidence of calcaneus gait leading to crouch gait in both spastic diplegia and spastic quadriplegia, especially after percutaneous lengthening of the Achilles tendons. Dietz et al. confirmed these findings but added an objective measure of crouch gait: the need to prescribe a floor reaction brace [20]. Their study reports a single-surgeon case series with a highly standardised operative protocol, follow-up programme and consistency in the diagnosis and management of crouch gait. Again, the results were best in patients with either hemiplegia or subjects who required lengthening on only one side. They recommended the consideration of non-surgical treatment or more conservative surgical treatment, such as gastrocnemius recession fascial lengthening [20].

There is a clear consensus from this review that the type of cerebral palsy matters a great deal and should be carefully considered when deciding on the type of surgical procedure and overall management of the equinus deformity. Only five studies in this review classified children according to GMFCS levels [14]. In children with bilateral cerebral palsy (traditionally described as having spastic diplegia or spastic quadriplegia), the use of the GMFCS might well prove to be very valuable when deciding on appropriate surgical interventions. It is possible that outcomes vary considerably across GMFCS levels and this would be worthy of examination in future studies.

Effect of age at index surgery on the rates of recurrent equinus deformity and calcaneus gait

There was widespread agreement in the studies reviewed that early surgery was a major risk factor for recurrent equinus deformity and possibly for unpredictable outcomes in general [16, 17, 20, 30, 40, 45]. This led several authors to recommend delaying age at index surgery by the use of non-operative measures such as the injection of botulinum toxin A, serial casting and the use of orthoses [3, 6, 17, 20]. No study to date has reported on the outcome of sequential non-operative management of equinus using botulinum toxin injections followed by surgical lengthening at an appropriate age. However, several studies examining the use of botulinum toxin A have reported successful short-term outcomes and the ability to delay surgery until the age more than 6 years in the majority of children. A more

Table 7 Single-event multi-level surgery (SEMLS) articles

Author	Equinus procedures	Additional procedures	No. of subjects (avg)	Age (years) mean (range)	Preop DF IC	Postop DF IC	Preop max DF	Postop max DF	Preop KF IC	Postop KF IC	Outcome measures	Follow-up (years/range)
Abel et al. [14]	Vulpius	ADD R, REC T, HAM L, STF	27 (7)	8.7 (4–20)	0	0	3	10	40	25	3DGA, GMFM	2 (1.6–3.1)
Adolfsen et al. [11]	Strayer, TAL (NS)	REC T, HAM L, POTB	31 (6)	8.5 (5–15)	−5	−2	7	12	31	21	3DGA, EMG, clinical exam	1.9 (0.7–6.4)
Dietz et al. [20]	Hoke TAL (O), Z	POTB, HAM L, REC T, ADD R	79 (6)	7.7 (2–47)	–	–	–	–	–	–	OGA, GRAFO	7 (–)
Gough et al. [23]	Strayer	VDRO, HAM L, POTB, REC T, CO	12 (6.4)	9.8	–	–	−5	15	–	–	3DGA, EMG, clinical exam	1.5 (–)
Gough et al. [24]	Strayer	HAM L, POTB, CO, REC T	12 (5)	6.4 (5.9–6.9)	–	–	–	–	–	–	3DGA, GGI, EMG, clinical exam	2 (2–5)
Kay et al. [2]	Strayer, Z TAL (NS)	POTB, HAM L, CO, VDO, SMO, REC T, FTT	54 (5)	9.8 (4–16)	–	–	0.6	16	–	–	3DGA, PRS, Ashworth	1.5 (1–2)
Lofterød and Terjesen [27]	Strayer, Z TAL	POTB, REC T, VDO, HAM L	47 (3.9)	11.5 (5–19)	−21.9	−7	−6	10.9	23.9	18.4	3DGA exam, GMFCS, FMS	1.2 (0.8–1.5)
Lyon et al. [29]	Z TAL, Hoke TAL	HAM L, REC T, SMO, VDO, FTT, ADD R	14 (5.6)	9.1 (4–17)	−6.8	10.8	10.1	24.1	40.5	32.9	3DGA	1.5 (0.5–2.5)
Rose et al. [35]	Baker	VDO, SMO, POTB, ADD R, HAM L, REC T	20 (5.3)	6 (4–26)	−5	−3	4	12	25	16	Clinical exam, 3DGA	1 (–)
Saraph et al. [37]	Baumann	POTB, ADD R, HAM L, REC T, SMO, VDO	22 (8)	12.6 (7.4–16.6)	−16.8	2.2	11.8	7.1	27.2	18.9	Clinical exam, 3DGA	2 (2.1–4)
Saraph et al. [38]	Baumann	POTB, ADD R, HAM L, REC T, SMO, VDO, FF	32 (8)	11.1 (8.7–13.5)	−3.9	4.6	6.4	16	–	–	Clinical exam, 3DGA, EMG	4.4 (1.0–5.4)
Segal et al. [39]	Hoke (P) TAL White/Z TAL, Vulpius, HCA	POTB, ADD R, HAM L, VDO, SMO	20 (4)	5.2 (2.7–8.2)	–	1.45	–	20.3	–	10.7	Clinical exam, 3DGA	5.8 (1.1–11)
Steinwender et al. [41]	Baumann	POTB, ADD R, HAM L, REC T, SMO, VDO, FF	17 (13.5)	14.4 (–)	−11	0	−6	7.5	28	21	Clinical exam, 3DGA	3.9 (–)
Yngve and Chambers [44]	Z TAL, Vulpius	POTB, VDRO, ADD R, HAM L, REC T	33	9 (–)	−13	0	−5	11	29	21	Clinical exam, 3DGA	1 (–)
Zwick et al. [46]	Strayer	POTB, ADD R, HAM L, REC T, SMO, VDO, FF	17 (8)	11.2 (5.7–16.4)	−3.6	1.4	−0.9	5.1	31.6	23.6	Clinical exam, 3DGA	3.8 (2.6–5.7)

VDO varus derotational osteotomy femur; *ADD R* adductor release; *HAM L* hamstring lengthening; *STF* sub-talar fusion; *FTT* foot tendon transfer; *REC T* rectus femoris transfer; *POTB* intramuscular psoas lengthening; *CO* calcaneal osteotomy; *SMO* supra malleolar osteotomy; *FF* foot fusion; *HCA* heel cord advancement; *TAL* tendo-Achilles lengthening widespread application of this policy might well improve surgical outcomes by delaying and, in some cases, preventing the need for surgical intervention, as well as reducing the risk of recurrent equinus and perhaps calcaneus.

Effect of surgical procedure on the rate of recurrent equinus deformity and calcaneus gait

A total of ten different operative procedures or combinations were reported in the articles in this review. Some studies focused on a single operative procedure [25, 30, 37], others on a comparison of multiple procedures [17, 33, 40], and some in the setting of SEMLS [2, 11, 14, 20, 23, 24, 27, 29, 35, 37, 38, 41, 45]. The reason for the choice of various procedures was often surgeon preference or frequently not clearly stated. The operative procedures range from very proximal procedures in Zone 1 of the gastrocsoleus to very distal procedures on the Achilles tendon. Despite a great emphasis on the importance of the differences in the surgical techniques, the evidence for differences in outcomes is weaker for surgical procedure than for cerebral palsy type or age at index surgery. In particular, several studies utilising three-dimensional gait analysis found little difference between Zone 2 surgical procedures (Baker and Vulpius) and surgical lengthening of the Achilles tendon. Within the Achilles tendon, a wide range of procedures was utilised, from open Z lengthening to percutaneous slide lengthening. There were no studies in which children were randomised to receive one surgical procedure versus another and the conclusions reported in most studies are questionable and highly susceptible to their inherent selection bias. Despite the paucity of evidence, opinions on the appropriate choice of surgical procedure are highly polarised. The lack of equipoise means that the design of randomised trials in which different procedures are compared is contentious and difficult.

Unfortunately, the majority of the studies included in this systematic review were of poor methodology. Only one of the included studies was prospectively designed and no studies were randomised with a blinded control group. The majority of studies were of level 4 quality of evidence according to the Oxford CEBM levels. Therefore, these results, at best, could lead to only a grade C treatment recommendation. Based on the general quality of these studies and their inherent weaknesses in the study design and methodology, further and more elegant statistical analyses applied to systematic reviews were not possible.

Given the well recognised difficulties of designing, recruiting, funding and implementing randomised surgical trials, consideration should be given to designing better cohort studies. Improvements in study design might include improved definition of equinus gait and associated functional problems using a combination of instrumented gait analysis, functional testing and quality of life measures. Surgical populations should be described much more rigorously in terms of cerebral palsy type (movement disorder, topographical distribution and GMFCS level). The surgery for equinus deformity should be clearly described avoiding eponymous terms. The need for a clear description of postoperative management, bracing and night splinting is also obvious. In addition, for children with spastic diplegia, it is essential to fully describe all additional procedures in multi-level surgical protocols.

Prospective studies with balanced outcome measures at prescribed intervals would greatly improve the evidence base for equinus surgery. A comparison of outcomes at baseline, 12 months and 5 years after surgery with appropriate outcome measures would be a major advance on the current poor evidence base.

Conclusion

Equinus deformity can be primary, secondary, either fixed or functional, and both impacts upon and is affected by the motor development of the child in question. A systematic review revealed the literature to be dominated by retrospective case series studies with no control groups and a lack of randomised controlled trials. The current literature is particularly weakened by inclusion of heterogenous patient groups and relatively short-term follow-up, which may be inadequate to detect the true rate of both recurrent equinus and calcaneus. There is insufficient evidence to state a clear preference for any single surgical procedure. However, the literature indicates greater incidence of recurrent equinus in children with hemiplegia regardless of procedure, and greater incidence of calcaneal gait and deformity in children with diplegia, particularly following procedures on the Achilles tendon.

Acknowledgments We would like to thank Mary Sheedy for her contributions to the organising and formatting of this manuscript. No external funding was received in support of this study. Kerr Graham is supported by a National Health and Medical Research Council of Australia Clinical Centre of Research Excellence (NH&MRC CCRE) Grant.

Conflict of interest statement None.

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