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Special Issue Reprint

Orthopaedics and Biomechanics in Children

Edited by
Pieter Bas de Witte and Jaap J. Tolk

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About the Editors

Pieter Bas de Witte

Dr. Pieter Bas de Witte MD PhD is a fellowship-trained pediatric orthopedic surgeon with a clinical focus on pediatric orthopedics, joint reconstruction, and traumatology. He has a broad pediatric orthopedic research line with a focus on pediatric hip disorders, hip dysplasia, and the role of artificial intelligence in improving diagnostics, surgical planning, and clinical decision making.

Jaap J. Tolk

Dr. Jaap J. Tolk MD PhD is a dual fellowship-trained pediatric orthopedic surgeon with a clinical interest in pediatric and adolescent hip disorders and limb reconstruction for congenital and acquired pediatric deformities. His research aims to improve growth and outcome predictions, reduce treatment morbidity, advance guided growth techniques, and enhance patient-oriented care.

Preface

This Special Issue of *Children* presents a collection of research articles focused on the critical role of biomechanics in the development, progression, and treatment of musculoskeletal diseases in children. The papers included in this volume illustrate the complex interplay between mechanical factors, growth, and physical function, highlighting the importance of understanding these biomechanical principles in optimizing diagnostic and treatment strategies for pediatric orthopedic conditions.

This comprehensive collection of research articles aims to enhance our understanding of the complex relationship between biomechanics, skeletal growth, and musculoskeletal pathology in children, ultimately leading to improved diagnostic and treatment approaches for this patient population. We hope that the findings presented in this Special Issue will serve as a valuable resource for clinicians, researchers, and trainees working in the field of pediatric orthopedics.

Pieter Bas de Witte and Jaap J. Tolk
Guest Editors

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Editorial

Friend or Foe? Biomechanics and Its Key Role in Paediatric Orthopaedics

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1. Introduction

Biomechanics play a key role in the development, progression and treatment of musculoskeletal disease in children. These biomechanics can be either friend or foe. On the one hand, there is excellent remodeling potential; on the other hand, as just one example, growth plate injuries and consequent growth disturbances can lead to significant problems.

There is a complex interaction between mechanical factors, growth and development, movement and physical function. Understanding these biomechanical factors is crucial in optimizing diagnostic and treatment strategies and improving outcomes for children with orthopaedic conditions.

In this Special Issue of *Children*, we address biomechanics in paediatric orthopaedics. A wide range of quality papers regarding this subject are included, increasing our knowledge and understanding of the complex relationship between pathology, skeletal growth and mechanical factors during childhood.

2. Biomechanics as a Friend

On the one hand, biomechanical aspects of the growing skeleton can often be used as an advantage during treatment, for example, in clubfoot correction with stepwise casting and/or tendon transfers or the treatment of limb length and/or axis discrepancies.

A strong example of the beneficial use of growth and biomechanics in the treatment of children with orthopaedic conditions is the application of guided growth techniques. These techniques involve surgical procedures around the growth plates, for example, by drilling (epiphysiodesis) or with the use of temporary implants, such as tension plates, screws or staples, to modulate the growth of a specific bone or limb. These implants alter the mechanical forces acting on the growth plate, allowing for the correction of angular deformities (hemi-epiphysiodesis) or limb length discrepancies.

The above methods are well known and widely applied for leg length differences or leg varus/valgus correction procedures around the knee [1–3]. However, several papers in this Special Issue discuss novel applications of guided growth techniques.

Lebe et al. show that the guided growth of the proximal femur has great potential as a low-invasive treatment method for hips at risk of dislocation in cerebral palsy patients. They conclude that the technique is effective and predictable with an overall low complication rate; however, further work is required to identify the best candidates and surgical timing, as well as choice of technique and implant [Contribution 1].

Paley and Shannon explore the application of guided growth beyond angular and longitudinal deformities, focusing on the correction of rotational deformities in the lower extremities using a flexible tether device. In their preliminary study, they concluded that rotational guided growth can successfully correct torsional malalignment without invasive osteotomy surgery [Contribution 2].

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Though less invasive than corrective osteotomies, guided growth techniques are not without morbidity. Braun et al. show that in the treatment of valgus deformities with hemi-epiphysiodesis, a relevant number of patients sustain prolonged pain and limited mobility. Patients with simultaneous plate implantation at the femur and tibia, as well as metaphyseal plate positioning, experienced resulting prolonged pain and a delay in functional recovery. These findings highlight the importance of carefully considering the specific indications and techniques used in guided growth procedures to minimize complications and optimize patient outcomes [Contribution 3].

Additionally, for guided growth, biomechanics can be a friend in other ways. For example, Segal et al. analysed a cohort of patients with cerebral palsy and assessed the potential beneficial effect of using a Functional electrical stimulation of the ankle dorsiflexor (DF-FES). They found limited benefit of the DF-FES on gait parameters; postural control seemed to be improved at the cost of a slower but more controlled gait [Contribution 4]. Furthermore, Gangaram and colleagues studied pre-operative traction before closed reduction in children with developmental dysplasia with a dislocated hip. In a retrospective pair-matched study, they analysed whether maintenance of hip reduction was influenced by the application of pre-operative longitudinal traction. They conclude that traction treatment does not significantly improve the short-term or mid-term outcomes for closed reduction and, therefore, should not be used as standard care for dislocated hips in Developmental Dysplasia of the Hip (DDH) [Contribution 5].

3. Biomechanics as a Foe

Disadvantageous effects of biomechanics in the growing skeleton have to be considered as well; examples include physeal injuries leading to growth disturbance or the progression of idiopathic scoliosis during adolescence [3,4].

In a large longitudinal cohort of hip dysplasia patients, Merchant et al. show that leg length differences in children treated for DDH are common. In two-thirds of the patients, the affected DDH leg was longer, mainly arising from the subtrochanteric segment. On the other hand, patients with a higher grade of AVN were often found to have shortening of the DDH leg. Interventions to correct leg-length differences were performed in 27.5% of their patients. They recommend careful monitoring of LLD in the follow-up of patients with DDH [Contribution 6].

Jansen et al. reviewed windswept deformities in children. These deformities can be a tell-tale sign of underlying disorders and can significantly impact daily functioning. The authors present a literature overview with a step-by-step guide for clinicians who encounter a child with windswept deformity [Contribution 7].

4. The Role of Biomechanics in Research and Diagnostics

Lastly, biomechanics can be utilized in the analysis of the consequences of structural skeletal abnormalities on joint mechanics and function for research or diagnostic purposes or for the development of innovative surgical techniques [5].

The influence of structural orthopaedic disorders on gait and physical function is an example of the strong interaction with biomechanical factors. A series of papers assessing this interaction, using instrumented gait analysis, could be included in this Special Issue.

Liu et al. summarize the available literature on gait characteristics in typically developing toddlers, providing a reference for clinical assessment and further clinical research [Contribution 8]. And in a systematic literature review, Grin et al. provide an in-depth analysis of the range of kinematic gait differences that can be expected in children with clubfoot treated with the Ponseti method, as compared to healthy controls. They provide strong recommendations for future research, with the implementation of multi-segmental foot models and a focus on the relationship between gait impairments and functional problems [Contribution 9].

5. Conclusions

This Special Issue provides a comprehensive overview of advancements in orthopaedics and biomechanics in children, highlighting topics such as guided growth, structural skeletal alignment and length and the impact of orthopaedic disorders on gait and physical function. The studies described above, combined with the contributions of Moerman et al., Hollyer et al. and Fernandez-Perze et al. [Contributions 10–12], suggest that future research should concentrate on analyzing the application of guided growth beyond longitudinal and angular abnormalities, with specific attention to appropriate patient selection and the assessment of long-term outcomes. Additionally, further investigation into the relationship between gait deviation, physical function and the impact of treatment modalities is needed to enhance our understanding in this area. Research papers presented in this Special Issue add to the scientific basis for further research and the improvement of patient care for children with orthopaedic disorders in the future.

Conflicts of Interest: The authors declare no conflicts of interest.

List of Contributions:

1. Lebe, M.; van Stralen, R.A.; Buddhdev, P. Guided Growth of the Proximal Femur for the Management of the 'Hip at Risk' in Children with Cerebral Palsy-A Systematic Review. *Children* **2022**, *9*, 609.
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Systematic Review

Guided Growth of the Proximal Femur for the Management of the 'Hip at Risk' in Children with Cerebral Palsy—A Systematic Review

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Abstract: Background: Guided growth is frequently used to modify lower-limb alignment in children, and recently temporary medial hemiepiphysiodesis of the proximal femur (TMH-PF) has been used for the management of hips at risk of subluxation in cerebral palsy (CP) patients. The aim of our study was to evaluate the efficacy of TMH-PF in the management of neuromuscular hip dysplasia in children with cerebral palsy. Methods: A systematic search of the literature was performed by using PubMed, EMBASE, CINAHL, MEDLINE, Scopus and Cochrane databases. Pre- and postoperative radiographic changes of the migration percentage (MP), head-shaft angle (HSA) and acetabular index (AI) were included in a meta-analysis. Secondary outcomes were treatment complication rates, technical considerations and the limitations of this novel technique. Results: Four studies (93 patients; 178 hips) met the eligibility criteria for inclusion in the meta-analysis. All three radiographic measurements showed significant changes at a minimum of 2 years of follow-up. Mean changes for MP were 8.48% (95% CI 3.81–13.14), HSA 12.28° (95% CI 11.17–13.39) and AI 3.41° (95% CI 0.72–6.10), with I^2 of 75.74%, 0% and 87.68%, respectively. The serious complication rate was overall low; however, physeal 'growing off' of the screw was reported in up to 43% of hips treated. Conclusion: TMH-PF is an effective and predictable method to treat CP patients with 'hips at risk', and the overall complication rate is low; however, further work is required to identify the best candidates and surgical timing, as well as choice of technique and implant.

Keywords: guided growth; DDH; cerebral palsy; temporary medial hemiepiphysiodesis for the proximal femur (TMH-PF)

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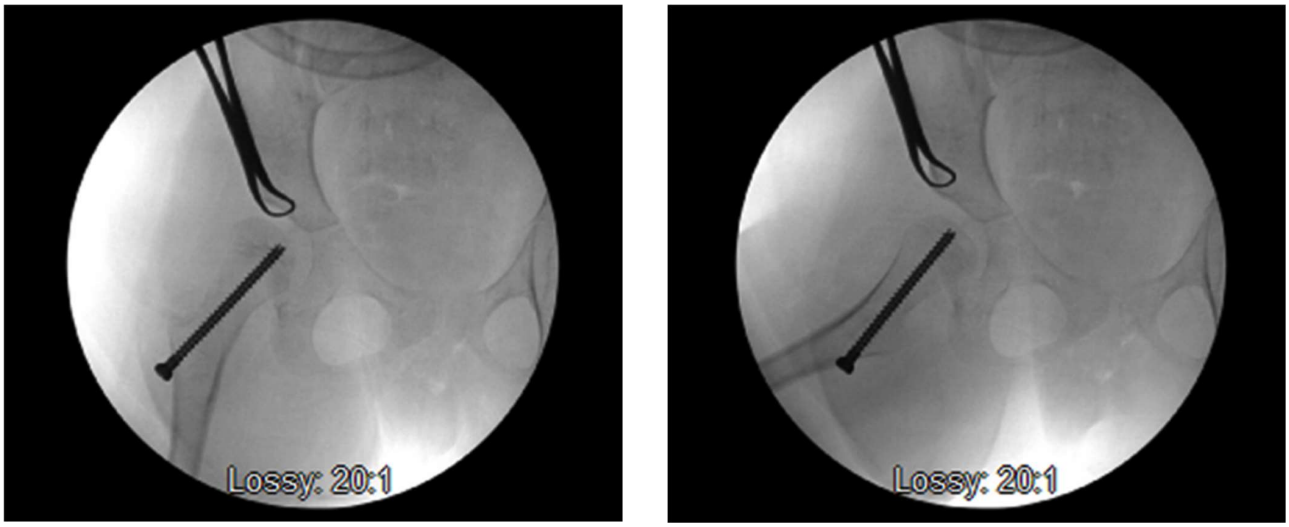
1. Introduction

Coxa valga is a complex three-dimensional deformity of the proximal femur, caused by altered growth of the proximal femoral physis [1–3]. Its cause can be idiopathic; projectional on plain radiographs, due to femoral anteversion; or in association with a variety of conditions, including developmental dysplasia of the hip (DDH), Charcot–Marie–Tooth (CMT) disease or cerebral palsy (CP) [4,5]. In CP patients, coxa valga, which is related to excessive anteversion, is commonly related to the functional status of the patient, in combination with muscle spasticity and weakness, and subsequent contractures can lead to symptomatic, progressive hip joint subluxation and dislocation, causing disturbed seated balance or standing abilities, difficulty with perineal care, the development of decubitus ulcers and poor quality of life [6–11]. Up to one-third of children with CP have hip instability, with an increasing incidence associated with GMFCS level—>60% of GMFCS IV/V [12–15], as measured using the Reimers migration percentage [16].

Traditional surgical management, typically reserved for hips with a migration percentage of 40% or more, includes hip reconstruction involving soft tissue releases, femoral

and pelvic osteotomies [17]. These procedures are associated with significant perioperative morbidity, including pain; increased blood loss; and lengthy anesthetic and inpatient recovery times, often complicated with peri-operative infections [18,19]. With improved surgical techniques, orthopedic implants and enhanced postoperative pathways, weight-bearing can be resumed shortly after surgery; however, traditional treatment commonly included a period of non-weight-bearing, with some surgeons preferring to augment their reconstruction with a hip spica or abduction brace [20,21].

Guided growth procedures are well established in the treatment for the gradual correction of angular and rotational limb deformities in children [22–25]. Anterior hemiepiphysiodesis of the distal femur has been shown to be effective in the treatment of fixed flexion deformity of the knee when compared to traditional osteotomies [26–30]. Figure 1 shows intra-operative radiographs of this minimally invasive technique, which has been recently applied to the proximal femoral physis for various conditions [23,31–35]. By placing a screw over the physis on the medial side, the tethering that occurs on the medial side will result in progressive varus of the proximal femur. It is understood that this manipulation of the proximal femoral anatomy can alter the course of secondary acetabular dysplasia [36,37]. Furthermore, it is recognized that guided growth procedures of the proximal femoral physis can be carried out as day case procedures, require a shorter operating time and allow for immediate weight bearing/standing when performed in non-ambulatory patients.



(**a**) (**b**)

Figure 1. Final AP (**a**) and lateral (**b**) fluoroscopy image showing the desired screw placement across the proximal femoral physis (with copyright permission from Jon Davids [35]).

This systematic review reports a quantitative summary of postoperative radiological outcome measures of temporary medial hemiepiphysiodesis for the proximal femur (TMH-PF) in children with CP. We also summarize the technical considerations, reported treatment complications and limitations of this novel intervention.

2. Materials and Methods

This systematic review was performed in accordance with the guidelines of the Cochrane Handbook for Systematic Reviews and the PRISMA-P statements [38]. The protocol followed was registered with and accepted by the International Prospective Register of Systematic Reviews (PROSPERO) on 15.01.2021 (CRD42021226864).

2.1. Information Sources and Search Terms

A comprehensive search of the literature was performed by using PubMed, MEDLINE, Cochrane, Embase and Scopus databases, and Level IV or higher original articles were

selected for this review. Search terms, including Boolean operators suitable for each database, were ("guided growth" OR "hemiepiphysiodesis" OR "TMH") AND ("coxa valga" OR "hip" OR "DDH" OR "pelvis" OR "prox* femur"). Cross-reference search results of the included studies and gray literature were included when available. The literature search was performed in January 2021.

Our inclusion criteria were pediatric, skeletally immature patients with cerebral palsy, as described and updated by Bax et al. [39,40]; and a "hip at risk" of progressive subluxation, as described by Davids et al. and others [12,35].

Exclusion criteria were previous proximal femur or pelvis operations, case reports, technical notes, and published abstracts.

2.2. Selection Process

The PRISMA flow chart is illustrated in Figure 2. Two independent reviewers (ML and PB) separately, and blinded to each other, conducted the screening of search results against the in/exclusion criteria based on title, abstract and keywords. Disagreements were resolved by an independent third author (RvS). After the removal of duplicates, 9 titles were selected for full-text review, of which 5 were excluded with reasons. Four articles were included for quantitative analysis.

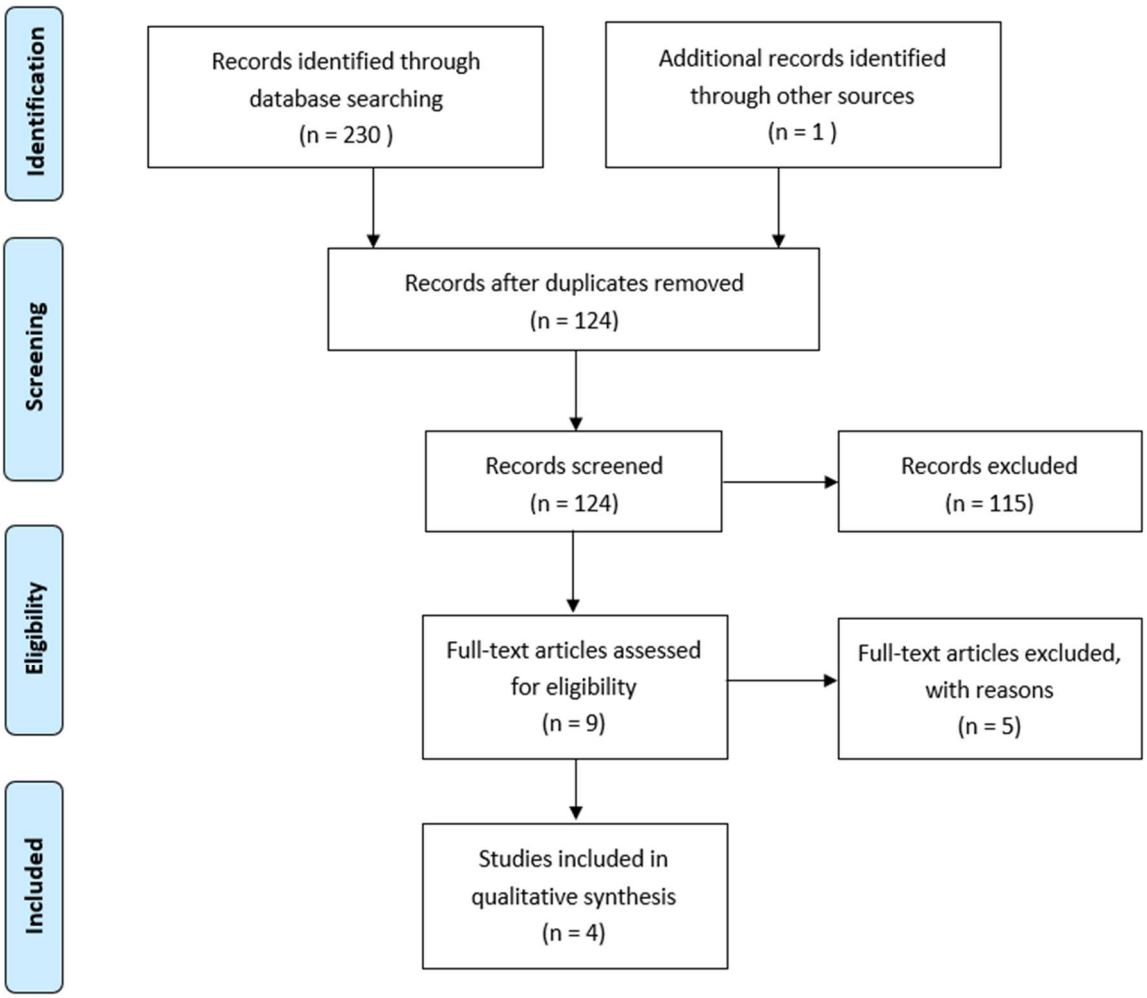


Figure 2. PRISMA flowchart demonstrating the results from the literature search and exclusions of papers.

2.3. Assessment of Quality and Bias

Risk of bias was assessed for all studies, using the ROBIN-I checklist [41], as recommended in the Cochrane Handbook for Systematic Reviews [42]. Findings are summarized in Table 1 and show the overall risk for bias as critical. All four studies had a small sample and were retrospective, with level IV case series and risk of bias due to confounding and selection of patients; moreover, the measurement of outcomes data remains a concern.

2.4. Outcome Measures and Statistics

The primary outcome was a change of radiographic angles after at least 2 years of follow-up. Secondary outcomes were complication rates, as graded by the Clavien–Dindo System, as well as a qualitative analysis of technical considerations based on the included papers [43–45]. Meta-analysis was performed by using Stata (StataCorp. 2019. Stata Statistical Software: Release 16. StataCorp LLC, College Station, TX, USA). Changes in pre- and postoperative radiographic angles were evaluated by means and standard deviations (SDs), and heterogeneity tests were performed; the random-effect model was applied if heterogeneity existed.

Table 1. ROBINS-I: Risk of bias assessment of non-randomized trials.

Reference	Bias Due to Confounding	Bias in Selection of Participants	Bias in Classification of Intervention	Bias due to Deviations from Intended Intervention	Bias Due to Missing Data	Bias in Measurement of Outcomes	Bias in Selection of the Reported Results	Overall Risk of Bias
[32]	Critical	Critical	Low	Low	No information	Serious	Moderate	Critical
[34]	Critical	Critical	Low	Low	No information	Serious	Moderate	Critical
[46]	Critical	Critical	Low	Low	Low	Critical	Moderate	Critical
[47]	Critical	Critical	Moderate	Low	Low	Critical	Moderate	Critical

3. Results

Our literature search has identified n = 231 titles. After the removal of duplicates, n = 124 titles, abstracts and keywords were screened for inclusion. Nine articles underwent full-text review, of which n = 4 met our in-/exclusion criteria and were subsequently selected for quantitative analysis. All studies included were level IV retrospective case series; the study characteristics and outcomes are summarized in Table 2.

3.1. Primary Outcomes

Postoperative changes of radiographic measures after $\geq$2 years of follow-up are presented in Table 2, which describes patient characteristics, methods and outcome measures used by all studies [32,34,46,47] included in our analysis. Most commonly, changes in the migration percentage (MP), head/neck-shaft angle (HSA/NSA) and acetabular index (AI) were reported at one year, as well as at two years or last follow-up review and compared with preoperative measurements. Some authors have performed additional soft tissue releases, and in >95% of patients included (178 hips in 93 patients), guided growth procedures were performed bilaterally during one theater attendance. One author [47] has, in addition, performed a subgroup analysis to assess the influence of transphyseal screw position on femoral remodeling and physis growing off the screw, as well as relevant predictive factors for a postoperative decrease in HSA.

Table 2. Study characteristics and reported outcomes of CP patients undergoing TMH-PF surgery.

a. Study Characteristics.

Reference	Study Design	Time Frame for Inclusion	Number of Hips in Number of Patients	Age at Surgery	GMFCS Level		Mean Follow-Up	Method of Fixation	Concomitant Soft Tissue Releases	Concomitant Botox Injections
[32]	Retrospective case series	January 2004–May 2012	13 hips in 9 patients	Mean 6.2 years (range 4.1–9.3 years)	IV V	6 patients 3 patients	45.6 months (range 24–96 months)	7.0 mm partially threaded Synthes screw	9/9 patients (common locations were psoas, adductor longus, gracilis and hamstrings)	0/9 patients
[34]	Retrospective case series	January 2007–December 2010	56 hips in 28 patients	Mean 7.5 years (range 4–11 years)	III IV V	7 patients 9 patients 12 patients	Not mentioned	4.5 mm partially threaded titanium Synthes screw	22/28 patients (bilateral distal hamstring lengthening	3/28 patients (medial hamstrings and adductors)
[46]	Retrospective case series	January 2012–December 2016	48 hips in 24 patients	Mean 8 years (range 5–12 years)	I II III IV V	3 patients 4 patients 7 patients 7 patients 3 patients	Mean 50 months (range 25–72 months)	6.0 mm fully threaded Acutrak, Acumed screw / 7.0 stainless steel, partially threaded, Synthes screw	24/48 hips 12/24 patients (adductor tenotomy)	0/24 patients
[47]	Retrospective case series	July 2012–September 2017	61 hips in 32 patients	Group 1–Median age 7 years (interquartile range 6.5–9.0) Group 2–Median age 7.5 years (interquartile range 6.0–9.0)	I II III IV V	4 patients 6 patients 10 patients 9 patients 3 patients		6.0 mm fully threaded Acutrak, Acumed screw / 7.0 stainless steel, partially threaded, Synthes screw	Not described	Not described

b. Reported Outcomes.

Reference		Preoperative Radiographic Measurements	Radiographic Measurements at 3 Months	Radiographic Measurements at 6 Months	Radiographic Measurements at 1 Year	Radiographic Measurements at 2 Years	Radiographic Measurements at Final Follow-Up	Number of Hips Growing Off Screws	Revision Surgery
[32]							Final follow-up at 5.8 years	13/13 hips (100%)	
	MP	52.2% (range 36–83%)	45.8% (p = 0.012)		40.3% (p = 0.016 *)	37.1% (p = 0.021 *)			
	HSA		173.3°		166.4° (p < 0.001 *)	162.7° (p = 0.15 *)	157.2°		
[34]							Final follow-up at 5 years	9/56 hips 6/28 patients	6 screw revisions 3 subsequent VDROs
	MP	33.5% (±11.29%)		29.23% (p < 0.001)	25.96% (p < 0.001 †)		23.16% (p < 0.001†)		
	NSA	156° (±10°)		150° (p < 0.001)	146° (p < 0.001 †)		142° (p < 0.001 †)		
	AI	23° (±6°)		20° (p < 0.001)	18° (p < 0.001 †)		17° (p < 0.001 †)		

Table 2. *Cont.*

			Final follow-up at a mean of 50 months	
[46]	HSA	163° (±6°)	150° ($p < 0.001$ †)	
	HEL	10° (±4°)	25° ($p < 0.001$ †)	
	AI	22° (±6°)	19° ($p < 0.001$ †)	
	MP	39% (±10%)	29% ($p < 0.001$ †)	
[47]	HSA	Group 1–163.6° Group 2–161.8°	Group 1–149.7° ($p < 0.001$ †) Group 2–153.1° ($p < 0.001$ †)	Group I–16/37 hips Group 2–4/24 hips
	MP	Group 1–28.7% Group 2–29.0%	Group 1–23.8% ($p < 0.001$ †) Group 2–27.5% ($p < 0.265$ †)	
	AI	Group 1–21.0° Group 2–21.2°	Group 1–19.4° ($p < 0.001$ †) Group 2–19.8° ($p < 0.010$ †)	
	FAVA	Group 1–32.0° Group 2–31.2°	Group 1–24.3° ($p < 0.001$ †) Group 2–24.9° ($p < 0.001$ †)	

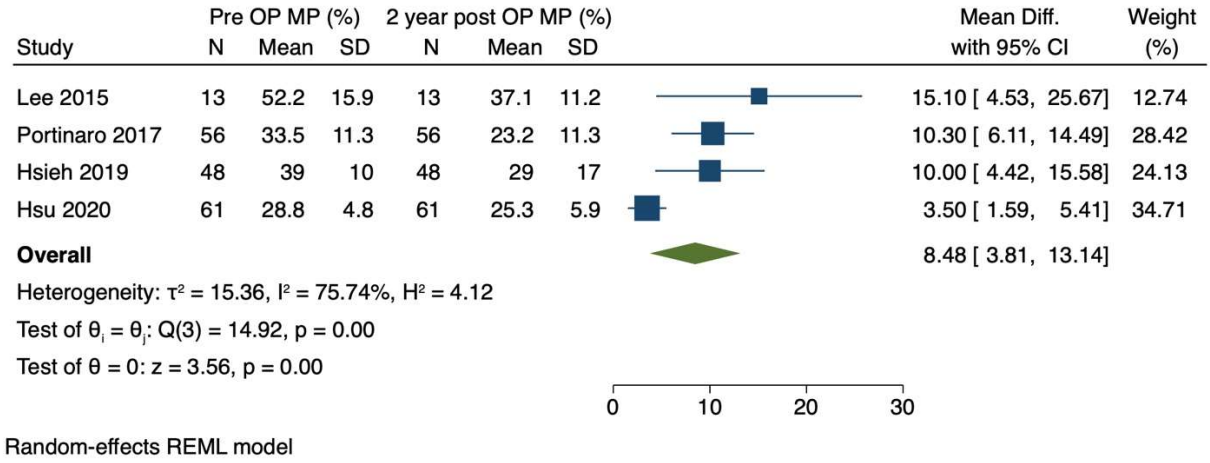
HEL–Hilgenreiner's epiphyseal angle; * p-value calculated by comparison to previous radiographic measurement; † p-value calculated by comparison to preoperative radiographic measurement.

3.2. Quantitative Analysis

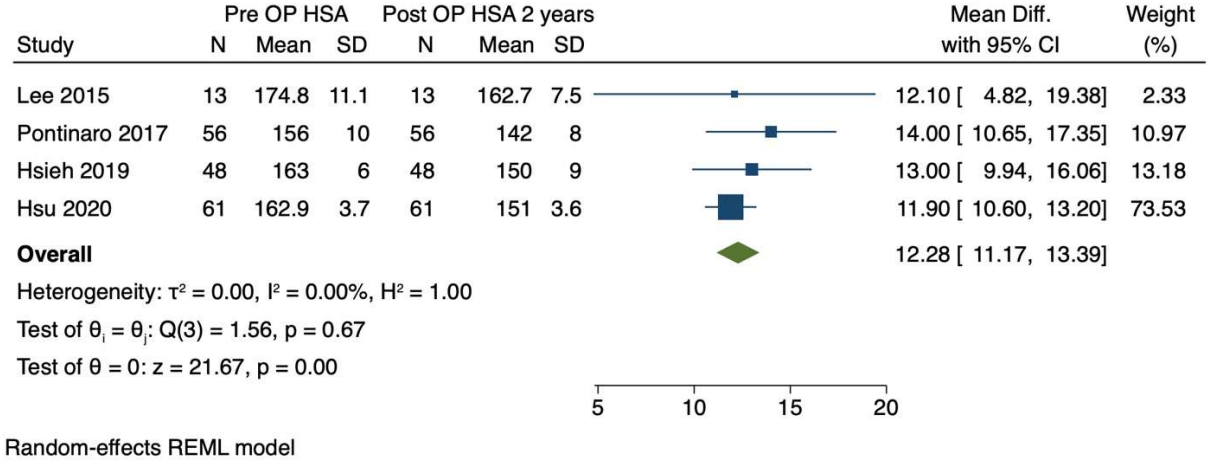
We have performed three random-effect meta-analyses to reflect the most commonly reported changes of mean radiographic angles after TMH-PF surgery (MP, HSA and AI). Hsu et al. [48] and Lee et al. [32] published 2 years of post-operative data, Portinaro et al. [34] reported 5 years of post-operative data, and Hsieh et al. [46] reported data with a mean follow-up of 50 months and a minimum of 2 years. To allow statistical analysis, we combined 2 years or more of published follow-up data in our analysis.

3.2.1. Migration Percentage

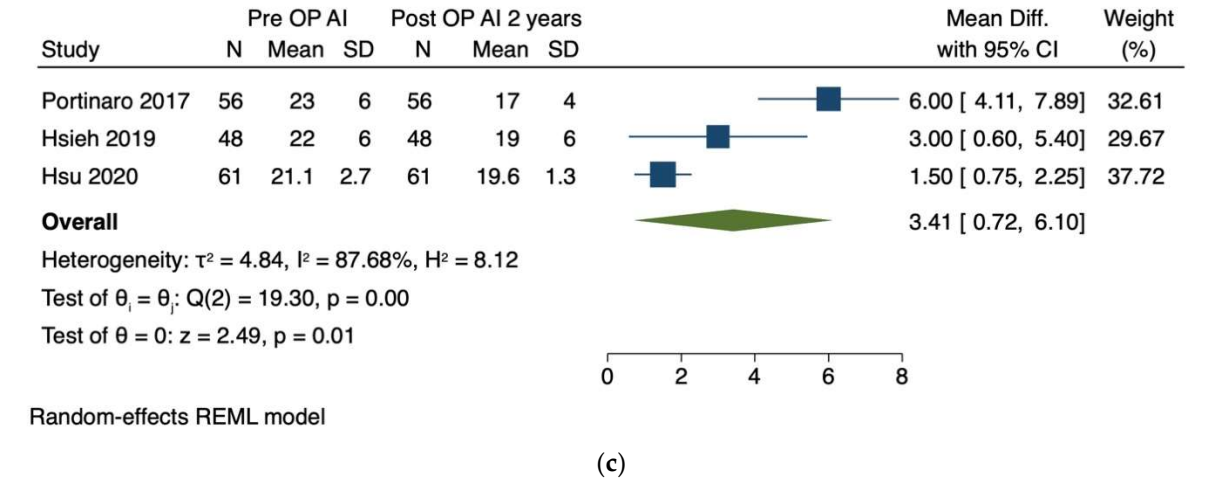
The mean migration percentage was reported in $n = 178$ hips by four authors [32,34,46,47] preoperatively and at least 2 years post-operatively as 34.74% (SD 11.45) and 26.50% (SD 12.27), respectively. We identified a significant ($p < 0.01$) weighted mean difference of 8.49% (95% CI 3.81–13.14, Figure 3a), with an I^2 of 75.7% and an average Hedges's g effect size of 0.77 (95% CI 0.66–0.99).



(**a**)



(**b**)



(**c**)

Figure 3. Forest plot demonstrating changes of primary outcomes measures after 2 and more years of follow-up. (**a**) Changes of migration percentage (MP). (**b**) Changes of head-shaft angle (HSA). (**c**) Change of acetabular index (AI).

3.2.2. Head-Shaft Angle

The mean head-shaft angle was also reported in $n = 178$ hips by four authors [32,34,46,47], with a mean preoperative HSA of 161.63° (SD 8.80) and 148.75° (SD 8.97) after at least 2 years of post-operative follow-up. We identified a significant ($p < 0.01$) weighted mean difference

of 12.28° (95% CI 11.17–13.39, Figure 3b), an I^2 heterogeneity of 0% and an average Hedges's g effect size of 1.94 (95% CI 1.07–2.81).

3.2.3. Acetabular Index

The mean acetabular indices were also reported in $n = 165$ hips by three authors [34,46,47], with a mean preoperative AI of 22.0° (SD 5.07) and 18.54° (SD 4.19) after at least 2 years of postoperative follow-up. We identified a significant ($p < 0.01$) weighted mean difference of 3.41° (95% CI 0.72–6.10, Figure 3c), I^2 heterogeneity was 87.68% and the average Hedges's g effect size was 0.79 (95% CI 0.41–1.17).

3.3. Secondary Outcome-Reported Technical Considerations, Complications and Limitations of TMH-PF

Lee et al. [32] described the surgical technique used for TMH-PF. They placed the screw two or three threads across the physis and assessed for protrusion of the screw into the joint. In their cohort, there were no complications, including infection, femoral neck fractures, implant failure, chondrolysis or osteonecrosis. All screws, however, backed out of the femoral epiphysis between 1 and 2 years postoperatively as the child grew. They reported no significant changes of the HSA between 1 and 2 years, and in some cases, continuing varus deformation after backing out of the screw, indicating a potential premature partial physeal closure.

While describing their surgical technique, Portinaro et al. [34] emphasized placing the cannulated screw guidewire in the inferomedial quadrant of the proximal femoral growth plate in order for the tip of the screw to reach 2 to 3 mm under the bony contour of the femoral head. They highlighted the importance of preventing the protrusion of the k-wire and screw into the joint and utilized a 4.5 mm cannulated screw instead of a 6.5 or 7.0 mm cannulated screw. In 9/56 hips, the physis grew off the screw, of which two hips underwent screw replacement surgery; however, in one of these cases, the screw head of the initial screw broke. The subsequent screw revisions ($n = 4$) were performed by adding a second screw, rather than exchanging the initial screw. The remaining three cases required VDROs, and there were no other complications, such as AVN, chondrolysis, fractures or wound infections.

Hsieh et al. [46] described the ideal position of the screw, as aimed at the medial one-third of the capital epiphysis on the coronal plane and centered along the axis of the femoral neck on the lateral plane. Depending on the femoral size, 6.0 mm fully threaded or 7.0 mm partially threaded screws were used, aiming to pass at least three threads across the physis. They found that the physis grew off the screw in 21 of 48 hips (43%), 15 of 48 hips underwent a replacement with a longer screw, and 8 hips in 5 patients underwent subsequent reconstructive surgery, such as VDROs. They concluded that this technique offers predictable results if the migration percentage is under 50% and there is enough growth remaining. They recommended restricting its use in patients with a migration percentage over 50%.

With respect to technical considerations, Hsu et al. [47] mention that the optimal position of the screw remains unclear. In order to prevent iatrogenic injury to the growth plate, repositioning was not attempted once a transphyseal position was achieved through the medial physis. They described 16 cases of the physis growing off the screw, and younger age at the time of surgery was identified as a significant risk factor (mean age of 7 years comparted to 9 years). Furthermore, it was suggested that medial positioning of the screw increases the risk of physeal growing off; this might only be appropriate for older children with less remaining growths. Describing only a limited improvement in the acetabular index at the 2-year follow-up, they concluded that the effect of guided growth on the acetabular development might be limited.

4. Discussion

Temporary medial hemiepiphysiodesis of the proximal femoral physis (TMH-PF) is a relatively novel surgical technique that was first reported in an animal model [36,37]. It has since been successfully performed in pediatric patients with coxa valga, due to type II AVN in DDH [49], and in patients with cerebral palsy with 'hips at risk' [47]. This study is the first systematic review and meta-analysis to summarize postoperative radiographic changes, complications and revision rates. We found significant changes in the migration percentage, head-shaft angle and acetabular index after at least 2 years of follow-up, with a mean difference of 8.48%, 12.28 degrees and 3.41 degrees, respectively. Growing off of the screw can be classified as a grade IIIb complication according to the modified Clavien–Dindo System and occurs in 15–50% of cases, whereas progressive hip subluxation (failure of treatment) needing invasive osteotomies, was reported in 5 to 21% of cases [34,45]. Several factors, including age, screw position, growth potential of the capital physis and level of gross motor function, are understood to influence the individual amount and velocity of anatomic changes of the proximal femur, and it remains unclear to what extent guided growth moderates those changes [50,51]. The possibility of coxa vara overcorrection due to physeal injury also requires further investigation [37].

Davids [35] has published a detailed technical summary of TMH-PF and has identified guided growths as a minimally invasive, safe and effective treatment options for CP patients with hip dysplasia. Most patients in the reported studies [32,34,47] were between 4 and 12 years of age and had a GMFCS of III–V; however, TMH-PF was performed in GMFCS I and II children by others [46]. It therefore remains controversial to apply guided growths procedures to ambulating patients, since their natural history of hip migration differs from GMFCS IV and V patients, and MP in GMFCS I patients commonly resolves spontaneously [52,53]. However, the progression of MP and late hip dislocation was reported in ambulating CP patients, with leg-length discrepancy, scoliosis, pelvic obliquity or deteriorating gait patterns being risk factors for poor outcome [54]. This can justify extended hip surveillance into adulthood and surgical intervention in selected cases [55]. Apart from GMFCS, Davids recommends an MP of 25 to 50% and an age between 4 and 10 years as indications for this guided growth procedure, and it was hypothesized that early surgical treatment is associated with greater potential for improvement of hip valgus; however, the likelihood of screw revision surgery due to the physis growing off the screw (whereby the screw no longer crosses the physis) is also increased [35]. A recent publication from the Cerebral Palsy Integrated Pathway Scotland (CPIPS) database concluded that the 'point of no return' for hip subluxation in this population was a MP > 46%, making spontaneous improvement unlikely [56]; however, others advocated a lower threshold for surgical intervention [12]. Furthermore, it was suggested that TMH-PF might be less effective in patients with an excessive (>50%) MP [45].

Implant choice varied considerably amongst all authors, ranging from 4.5 to 7.0 mm screw, both fully or partially threaded; however, it remains unclear if complications, including the screw backing out, are associated with the implant choice. Furthermore, it was recommended to pass two or three screw threads past the physis into the epiphysis [57], and Hsu et al. [48] have concluded that a centered screw position within the physis is associated with a reduced risk for physis growing off. The authors have suggested a centred screw position in young children, where early re-operation surgery i.e. due to growing off the physis is undesirable. In contrast, a more eccentrically placed screw near the medial physeal border is advised in older children, nearing skeletal maturity. Furthermore, the cox analysis revealed that an increased preoperative HSA was associated with higher rates of the screw growing off the physis. Hsieh et al. reported a combination of guided growth with simultaneous adductor tendon release [46].

The results of this meta-analysis are comparable to guided growth procedures performed for dysplasia of the hip (DDH) in smaller case series reported by other authors [33,49,58], where improvements of the femoral alignment and center edge angle (CEA) were reported after 2 years of treatment. Agus et al. [31] have trialed hip hemiepiphysiodesis procedures

in DDH without the use of an implant; in order to avoid the need for screw revisions, the authors drilled the proximal medial physis in a small case series and found significantly improved physeal inclinations during follow-up.

Our study has several limitations. Firstly, our meta-analysis is based on four small case series with high heterogeneity of the included patient characteristics (including age, GMFCS levels and length of follow-up). Guided growths of the proximal femur are, however, a relatively novel treatment, especially within the subgroup of cerebral palsy patients. Secondly, we noticed the poor quality of all studies included during the ROBIN-1 assessment. Thirdly, outcome measures and follow-up intervals were varied, and we pooled some outcomes to allow for further assessment during the meta-analysis, which can lead to an overestimation of the effect sizes reported. Hsieh et al. [46] included the outcomes of patients up to 12 years of age at the time of surgery and with a mean follow-up of 50 months (range 25–72 months); however, in the absence of long-term follow-up studies, it remains unclear how guided growths alter hip anatomy in the long term, including beyond skeletal maturity.

In conclusion, we have performed the first systematic review on the guided growth of the proximal physis in children with cerebral palsy. This novel and minimally invasive procedure has been shown to be safe and effective in the modulation of the proximal physis to correct coxa valga deformities, which can prevent progressive subluxation of the hip joint and may prevent the requirement for complex open-hip reconstruction surgery in this vulnerable cohort. Depending on the treatment duration and patient age, physis growing off the screw is a common complication, and patients and caregivers need to be counselled that screw revision is needed in about 50% of cases. Invasive pelvis reconstructions and femur osteotomies may be needed in only 5–21% of patients initially treated with guided growth, as reported in small cohort studies with short-term follow-ups, and long-term studies are needed to investigate appropriate indications and limitations of TMH-PF, including in ambulating patients or when combined with soft tissue releases [34,46].

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Rotational Guided Growth: A Preliminary Study of Its Use in Children

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Abstract: Torsional malalignment of the legs is common in children, and those that do not remodel may benefit from surgical correction. Traditionally, this is corrected with an open osteotomy. Guided growth is the gold standard for minimally invasive angular correction and has been investigated for use in torsional deformities. This study presents our preliminary results of rotationally guided growth in the femur and tibia using a novel technique of peripheral flexible tethers. A total of 8 bones in 5 patients were treated with flexible tethers consisting of separated halves of a hinge plate (Orthopediatrics Pega Medical, Montreal, QC, Canada), which were fixed to the epiphysis and metaphysis at 45° angles to the physis and connected with Fibertape (Arthrex, Naples, FL, USA). The implants are placed medially and laterally in the opposite 45° inclination, determined by the desired direction of rotation. Additionally, the average treatment time was 12 months. All patients corrected the rotational malalignment by clinical evaluation. The average rotational change was 30° in the femurs and 9.5° in the tibias. Further, the average follow-up was 18 months, with no recurrence of the rotational deformity. There was no change in longitudinal growth in the patients who underwent bilateral treatment. Rotational guided growth with flexible tether devices is a novel technique that successfully corrects torsional malalignment without invasive osteotomy surgery.

Keywords: guided growth; femoral anteversion; tibial torsion; rotational malalignment; miserable malalignment; growth modulation; hemiepiphysiodesis; growth tether

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1. Introduction

Torsional malalignment of the lower extremities is common in children. Most patients present because their parents complain that their child is in-toeing or out-toeing while walking. They may also complain that they fall more often because of this. At older ages, they may complain of hip, knee, or ankle pain associated with in- or out-toeing. Those who do not remodel with growth may benefit from surgical correction. Traditionally, the only surgical technique available for treating rotational deformities of the femur or tibia was osteotomy, requiring open surgery, a period of non-weightbearing, and frequently, an inpatient hospital stay. The use of guided growth has been of great interest in the treatment of torsional deformities due to the decreased morbidity and limited recovery time that have been demonstrated in the treatment of frontal plane deformities [1]. A minimally invasive technique was recently reported as a method to correct torsional deformities in the femur in children [2] using a circumferential cable. The purpose of this study is to report the preliminary results of a novel technique to treat rotationally guided growth in the femur and tibia using counter-opposed, crossed, inclined peripheral flexible tethers.

2. Materials and Methods

The institutional review board's approval was obtained. Torsional deformity was defined as a positive (external) or negative (internal) foot progression angle outside the normal physiologic parameters (+5 to +15 degrees) [3,4]. Once a torsional deformity was identified and determined to be symptomatic by history, a physical examination was performed to

quantify the rotational profile. The tibial rotation was measured using a goniometer to measure the prone thigh-foot axis. Additionally, femoral rotation was measured using a goniometer in the prone hip internal and external rotation profile according to Staheli [3] (Figure 1).

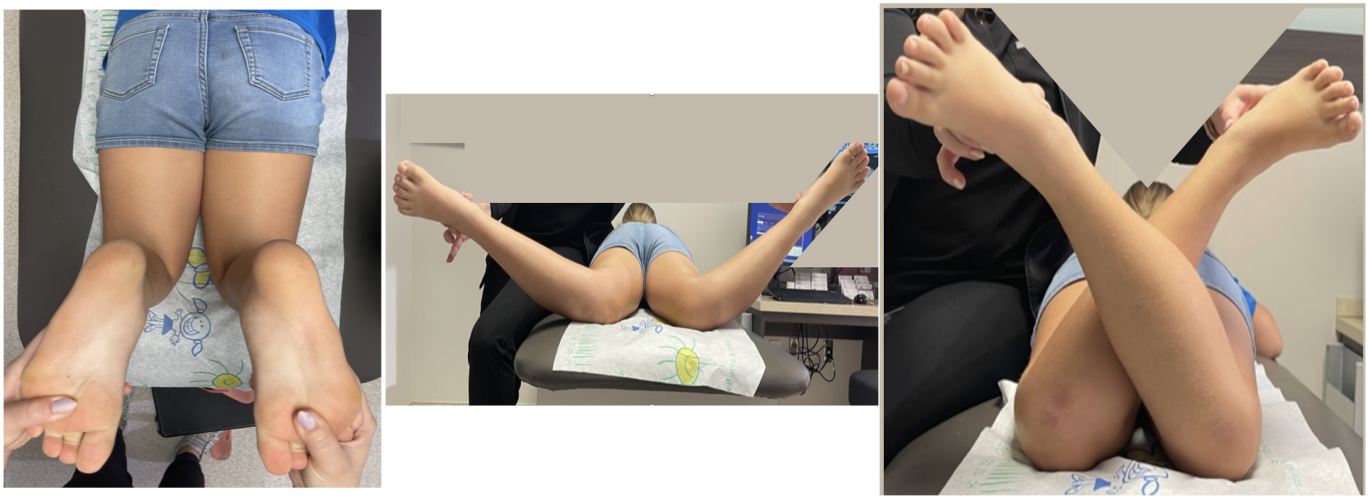

Figure 1. Prone clinical evaluation of the thigh-foot axis (**left**). Prone measurement of internal femoral rotation (**middle**) and external femoral rotation (**right**) (Patient #1 preoperatively).

A total of five patients with torsional deformities in eight bone segments (femur 5, tibia 3) were treated with flexible counter-opposed crossing tethers using the surgical method described. Two of the patients underwent bilateral distal femoral rotationally guided growth for idiopathic bilateral femoral anteversion. One patient underwent two ipsilateral rotational guided growth surgeries, first on the femur and then on the tibia, for internal torsional deformity. Additionally, the other two patients underwent unilateral rotationally guided growth to correct tibial rotation, one with internal torsion and one with external torsion. The demographics, preoperative, and postoperative rotational profiles for all included patients are listed in Table 1.

Surgical Technique

The Hinge Plate (Orthopediatrics, Pega Medical, Montreal, QC, Canada) is a hemi-epiphysiodesis screw-plate device that consists of two plate halves connected by a hinge. The hinge rivet is removed, and the two plate halves are separated (Figure 2). Only the male half of the plate is used. The separated halves are fixed into the epiphysis and metaphysis based on the desired rotational orientation and secured with a proprietary screw. In addition, the sections of plate are connected with Fibertape (Arthrex, Naples, FL, USA) to create a flexible tether. The fibertape is looped twice between the ends of the hinge plate halves, going through the hole where the hinge rivet was located. The fibertape ends are tied taut with five knots. Further, the lines created by the fibertape on each side of the bone should be at 45° to the long axis of the bone and perpendicular (90°) to each other (Figure 3. The two incisions are then closed. The patients are allowed to resume full weight-bearing and return to all activities as tolerated without restrictions.

Table 1. Demographics and rotation profiles before and after rotationally guided growth.

				Before Treatment						After Treatment						Amount of Correction (IR/ER/TFA)		
				Hip Rotation Internal (IR)		Hip Rotation External (ER)		Thig Foot Axis (TFA)		Hip Rotation Internal		Hip Rotation External		Thig Foot Axis				
Patient	**Age at Insertion (Years)**	**Diagnosis**	**Site**	**R**	**L**	**R**	**L**	**R**	**L**	**R**	**L**	**R**	**L**	**R**	**L**	**R**	**L**	**Time to Correct (Months)**
1	10.5	Femur anteversion	Bilateral Femur	60	65	20	20			40	40	50	45			−20/30/-	−25/25/-	15
2	5.4	Congenital pseudarthrosis of the tibia with external tibial torsion	Right Tibia					30						19		-/-/−11		18
3	7.7	Femur anteversion	Right Tibia					−7						10		-/-/17		11
3	8.6	Internal tibial torsion	Right Femur	60		40				50		60				−10/20/-		8
4	15.7	Congenital Femoral Deficiency with internal tibial torsion	Left Tibia						−15						−10		-/-/5	
5	2.6	Femur anteversion	Bilateral Femur	90	90	0	0			45	45	35	35			−45/35/-	−45/35/-	7

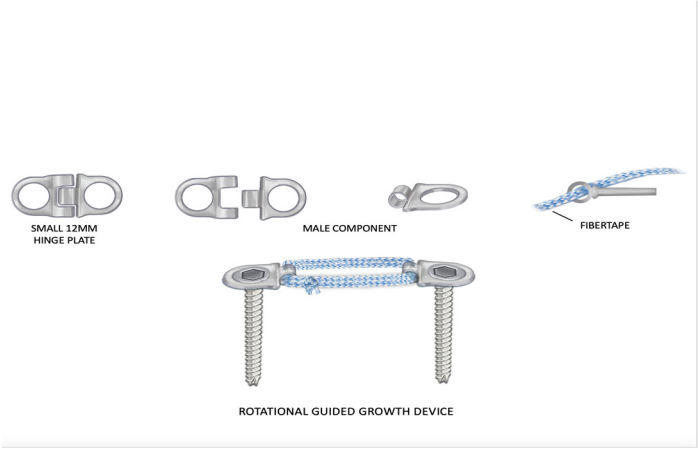

Figure 2. Illustration of the components of the rotationally guided growth implant used. Two of the same halves of the hinge plate were used. They were tethered together with fibertape. They were fixed to the bone with screws. Illustrations copyrighted to the Paley Foundation; reprinted with permission.

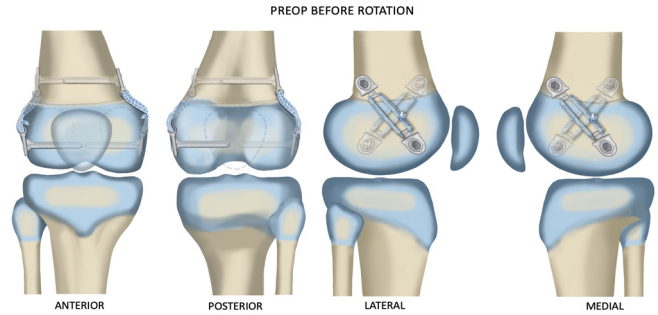

Figure 3. One implant was placed medially and the other laterally inclined, as shown, so that the crossing angle was as close to 90° as possible. Illustrations copyrighted to the Paley Foundation; reprinted with permission.

Marks are made on the skin in line with the planned placement of the fixation on the medial and lateral sides of the distal femur or proximal tibia using the image intensifier. The two lines are made at opposite 45° angles to the long axis of the bone. In addition, when superimposed, the lines should cross each other at an angle of 90°.

The orientation of the implant angle on each side of the bone depends on the direction of rotational correction desired. Therefore, on the right distal femur, to correct excessive internal rotation of the knee relative to the hip, the medial epiphyseal screw is placed posterior to the metaphyseal screw. The lateral epiphyseal screw is placed anterior to the metaphyseal screw (Figure 4). In addition, the medial epiphyseal screw is placed anterior to the metaphyseal screw, and the lateral epiphyseal screw is placed posterior to the metaphyseal screw for the right proximal tibia, to externally rotate the foot relative to the knee. Furthermore, to correct excessive external rotation of both the right distal femur and proximal tibia, the above screw orientations would be reversed. The medial epiphyseal screw is placed posterior to the metaphyseal screw, and the lateral epiphyseal screw is placed anterior to the metaphyseal screw in order to correct internal rotation of the left distal femur. Additionally, in the left internal tibial torsion, the medial epiphyseal screw is anterior and the lateral epiphyseal screw is posterior to their metaphyseal screws. There is no fibular fixation used for rotational correction of the tibia in either direction. The previous pattern is reversed for the left external tibial torsion.

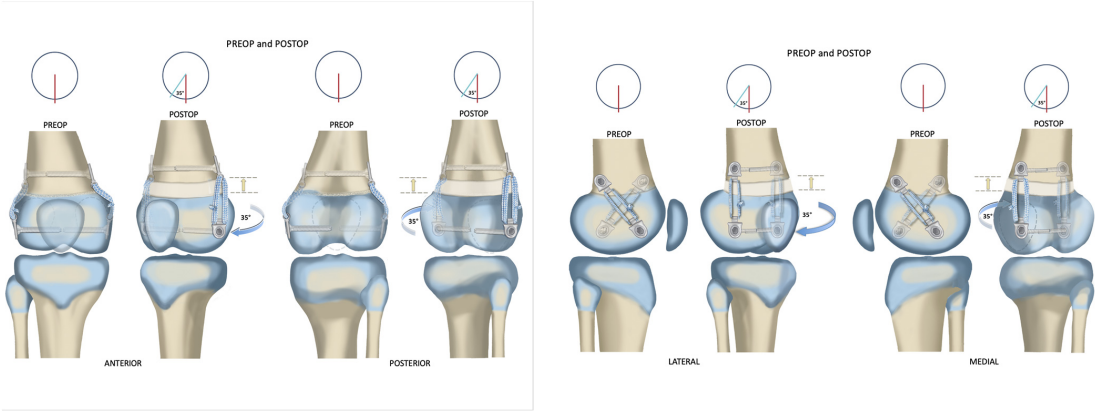

Figure 4. Anterior, posterior, lateral, and medial views of correction before and after rotationally guided growth correction. lllustrations copyright to Paley Foundation; reprinted with permission.

3. Results

The average age at the time of rotational plate insertion was 8 years and 5 months (range: 2 years and 6 months–15 years and 7 months). The total number of patients had open growth plates on preoperative radiographs. The underlying diagnoses included idiopathic femoral anteversion (3), internal tibial torsion (1), healed congenital pseudarthrosis of the tibia with NF-1 (1), and congenital femoral deficiency (1).

The post-operative rotational change was observed in all 8 bones treated. The average change in the femoral rotation patients was 30° (range 10°–45°). Additionally, the average change in the tibial rotation patients was 9.5° (ranging 5° to 17°). Further, the average time to correction was 11.8 months (range 7–18 months). Two of the patients underwent staged removal of the devices, removing the lateral plate tethers and leaving the medial plate tethers in place to correct residual genu valgum deformity. These plates have been subsequently removed. The average follow-up after plate removal was 18 months (range 2–33 months). Loss of rotational correction was not observed during this follow-up time. Moreover, the longitudinal growth during the time of rotational correction was evident but could not be compared to a contralateral normal side in the 3 patients (4 rotation guided growths) who underwent unilateral correction due to pre-existing leg length discrepancy (LLD). Two patients who did not have a LLD underwent bilateral treatment, and longitudinal growth remained the same on both sides. We were unable to determine if any slowing of longitudinal growth occurred as a result of the bilateral physeal tethering. There was no evidence of alteration of the posterior proximal tibial angle (PPTA) or posterior distal femoral angle (PDFA). The total number of patients returned to their preoperative level of activity after plate insertion and final plate removal (Figure 5).

Figure 5. Postoperative internal (**left**) and external (**right**) rotation of both hips after rotationally guided growth correction. Compare this to the preoperative photos of the same patient in Figure 1 (Patient #1).

4. Discussion

Hemiepiphysiodesis has evolved from the use of staples with perpendicular fixed legs to the use of a plate with pivoting screws [1]. The principle in both is to create a peripheral tether outside the physis, causing the physis to grow at its normal rate at the point farthest from the tether while limiting or temporarily stopping the growth at the point closest to the implant. This process has been renamed "guided growth" [5]. Until recently, the application of guided growth was to create an angular change in the frontal, sagittal, or oblique planes. Guided growth plates have also been used to create growth stoppage by epiphysiodesis [6]. This requires the placement of implants on opposite sides of the physis. It has been posited that the placement of plates on opposite sides of the growth plate at an inclined angle would lead to rotational tethering of growth before epiphysiodesis. This was corroborated in small animals (rabbits) by Arami et al., Sevil-Kilimici et al., and Lazarus et al. [7–9]. It was also corroborated in large animals (calves) by Martel et al. [10]. Most recently, it was also demonstrated to work in humans by Metaizeau et al. [2]. On the basis of these reports as well as this preliminary study, it is evident that rotationally guided growth can be achieved by two counter-opposed, obliquely oriented tethers on either side of the physis. The previously reported method used in animals and humans is a cable going through the bone and around the sides [2,7,10]. An alternative method was reported in rabbit studies using two inclined plates on opposite sides of the bone with no fixed structure passing through the bone from one side to the other [9]. The concern with the first method is that it can cause direct damage to the physis as the pressure of the cable cuts into the side of the bone between the epiphysis and metaphysis. The only human study reported knee stiffness as a common complication [2]. The disadvantage of the second method using inclined plates is that the plates are very stiff and nonmalleable, which could be prominent or restrictive as rotation occurs [9]. In this study, we used a tether without passing from one side of the bone to the other. The anchorage points to the bone were tethered using a flexible, soft material (fibertape), which lay on the surface of the physis. This may be safer and less abrasive to the physis than a taut metal cable.

Growth retardation was reported in rabbits by Lazarus et al. (mean 4.2%) [9] and Arami et al. [7] (mean 7%) in rabbits. Young rabbits grow exceptionally fast, and such tether-related retardation is not surprising. In contrast, in a large animal model using calves, Martel et al. found no evidence of significant growth retardation, although the tethers were only left in for three months [10]. In the only human study, Metaizeau et al. suggested that in their 20 cases there was a mean of 12 mm growth retardation over the course of 2 years of rotationally guided growth [2]. In this study, we did not find any leg length difference between sides in the two bilateral femoral rotationally guided growth cases. The other three patients all had leg length discrepancies to start, so it would be difficult to know if any additional growth inhibition occurred. There were no secondary angular deformities in the frontal or sagittal planes after correction. Two pre-existing angular deformities were corrected by converting the bilaterally guided growth into a unilateral hemiepiphysiodesis (Figure 6). The total time for correction ranged from 7–18 months, which is very similar to the time taken for angular-guided growth in the frontal or sagittal planes. The degree of correction was judged clinically and not radiographically. The correction achieved in the femur ranged from 20° to 35°. Additionally, the correction achieved in the tibia ranged from 5° to 17° (Figure 7). The foot progression angle returned to normal, and all parents and patients were satisfied with the improvement. There were no patients needed to be considered for torsional correction by osteotomy.

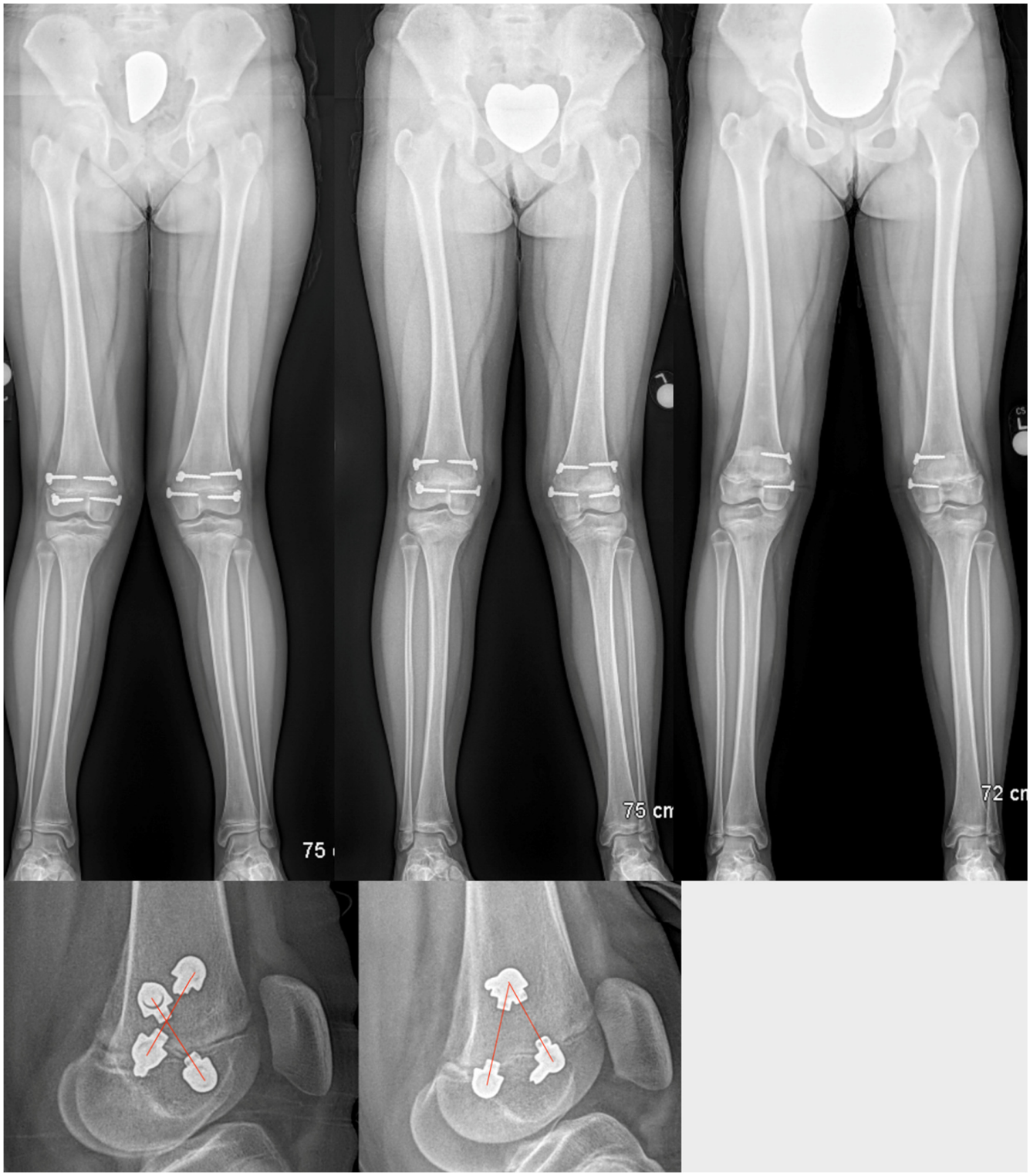

Figure 6. Standing radiographs of both lower limbs of the same patient as in Figures 1 and 5 (Patient #1) at the time of insertion of rotational guided growth implants (**top left**), end of rotational guided growth (**top middle**), and medial hemiepiphysiodesis with the medial implant to correct valgus (**top right**). The crossing angle of the plates is seen at the beginning (**lower left**) vs. at the end (**lower right**) of the correction. Note the change in crossing angle.

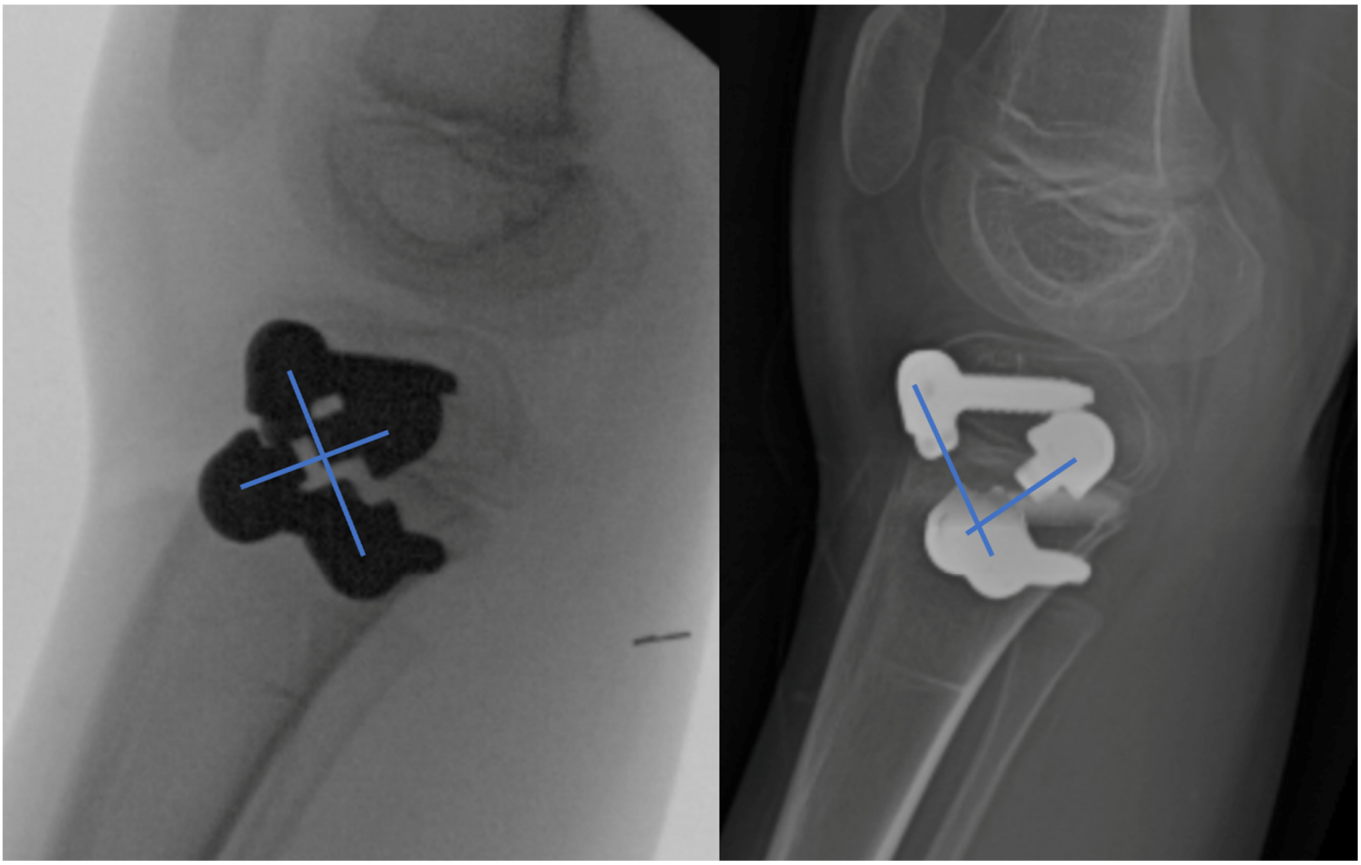

Figure 7. Rotationally guided growth of the tibia (patient #3). Lateral radiographs at the beginning (**left**) and end (**right**) of correction. Note the change in crossing angle. The tibial correction in this case was 17°.

5. Conclusions

Counter-opposed, inclined peripheral flexible tethers are an effective method to treat rotational malalignment in growing children. Further follow-up and a larger patient cohort will be needed to study the longer-term results and risk for rebound effects. While this study did not show evidence of growth retardation, we cannot rule out that some growth retardation may occur, as was seen in small animals.

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Institutional Review Board Statement: The study was conducted in accordance with the Declaration of Helsinki and approved by the Institutional Review Board of St. Mary's Medical Center.

Informed Consent Statement: Informed consent was obtained from all subjects involved in the study. Written informed consent has been obtained from the patient(s) to publish this paper.

Data Availability Statement: Not applicable.

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Conflicts of Interest: The authors declare no conflict of interest.

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Factors for Prolonged Pain and Restriction of Movement Following Hemiepiphysiodesis Plating for the Correction of Lower Limb Malalignment in the Frontal Plane: An Explorative Analysis

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Abstract: The correction of valgus leg malalignment in children using implant-mediated growth guidance is widely used and effective. Despite the minimal invasive character of the procedure, a relevant number of patients sustain prolonged pain and limited mobility after temporary hemiepiphysiodesis. Our aim was to investigate implant-associated risk factors (such as implant position and screw angulation), surgical- or anesthesia-related risk factors (such as type of anesthesia, use, and duration), and pressure of tourniquet or duration of surgery for these complications. Thirty-four skeletally immature patients with idiopathic valgus deformities undergoing hemiepiphysiodesis plating from October 2018–July 2022 were enrolled in this retrospective study. Participants were divided into groups with and without prolonged complications (persistent pain, limited mobility of the operated knee between five weeks and six months) after surgery. Twenty-two patients (65%) had no notable complications, while twelve patients (35%) had prolonged complications. Both groups differed significantly in plate position relative to physis ($p = 0.049$). In addition, both groups showed significant differences in the distribution of implant location ($p = 0.016$). Group 1 had a shorter duration of surgery than group 2 (32 min vs. 38 min, $p = 0.032$) and a lower tourniquet pressure (250 mmHg vs. 270 mmHg, $p = 0.019$). In conclusion, simultaneous plate implantation at the femur and tibia and metaphyseal plate positioning resulted in prolonged pain and a delay of function. In addition, the amplitude of tourniquet pressure or duration of surgery could play a factor.

Keywords: lower limb deformities; leg axis malalignment; postoperative pain; postoperative complications; pediatric orthopedic; implant-mediated growth guidance; tension band plate; hemiepiphysiodesis plate

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1. Introduction

The correction of axial lower limb malalignment in the frontal plane in children and adolescents by implant-mediated growth guidance with hemiepiphysiodesis plates for temporary hemiepiphysiodesis is a common and effective pediatric orthopedic procedure [1,2]. In contrast to the higher initial compression force of staples, hemiepiphysiodesis plates have a lower risk of physis fusion compared to these rigid staples [3–5]. The risk of extrusion or dislocation is lower in hemiepiphysiodesis plates because of their screws, and with their longer moment arm, faster correction rate is postulated [6]. The surgical procedure itself is minimally invasive compared to corrective osteotomies for angular deformities and has very low approach morbidity [7]. Nevertheless, the procedure appears to be associated with prolonged postoperative pain and reduced mobility and activity [8,9].

The current literature on hemiepiphysiodesis plating focuses on clinical outcomes, particularly its effectiveness as a guided growth system for correcting deformities, the speed of correction, and the incidence of rebound compared with other procedures [3,10,11]. However, Gregoire et al. [8] showed that 38% of patients still needed to take pain medication four weeks after temporary hemiepiphysiodesis and 65% did not return to previous activities during that time. Another study also showed a delay in postoperative return to full function [9]. There have been few reports to date with regard to surgical and implant-related risk factors for prolonged recovery with increased pain and limited postoperative knee range of motion. To the best of our knowledge, no study has investigated the cause of such complications. Therefore, the aim of this explorative study was to investigate implant-associated, surgery- or anesthesia-related, and other risk factors for postoperative complications. Our main hypothesis was that implant position is a factor in functional delay and prolonged pain after hemiepiphysiodesis plating. In particular, we hypothesized that the implantation angle of the screws has an influence on the incidence and duration of postoperative pain. We assumed that a divergent angle would result in more initial pain than parallel screws due to increased pressure and compression force on the physis.

2. Materials and Methods

2.1. Patients

Children and adolescents with idiopathic valgus deformities without other comorbidities were prospectively enrolled at our institution between October 2018 and July 2022. The indication for implant-mediated growth guidance with hemiepiphysiodesis plating was set for skeletally immature patients with a pathological idiopathic valgus alignment deformity (mechanical axis deviation (MAD) of >10 mm and/or mechanical femorotibial angle (MFA) of >3°) of one or both lower extremities [12]. To decide whether the angular deformity originated in the femur or tibia, the mechanical lateral distal femur angle (mLDFA) and mechanical medial proximal tibia angle (mMPTA) were determined, and the indication for surgery on the femur, tibia, or both segments was made according to the pathological joint surface angles (physiological values for mLDFA 88° +/− 2.5° and for mMPTA 87° +/− 2.5°) [13]. Eight-Plates (Orthofix, Lewisville, TX, USA) or Pedi-Plates (Orthopediatrics Inc., Warsaw, IN, USA) were used in this study. The same surgical technique was performed in all patients. The plates were inserted through a minimally invasive technique in an open procedure under fluoroscopic control. Local anesthesia was not applied to any patient.

Immediately after surgery, patients were allowed to resume full weight bearing, but sports and high impact activities were not permitted until four weeks postoperatively. Patients were discharged from the hospital after achieving 90° knee flexion and returned for follow-up visits at four weeks, three months, and then at three-month intervals until the leg axis was corrected (successful growth guidance was determined by an MFA of 0° +/− 2° or an MAD of 0 mm +/− 6 mm) and the plates were removed. Physical therapy was performed daily during the inpatient hospital stay (usually 1–3 days). Braces were not applied.

Patients were included in this study only if they completed a postoperative questionnaire asking about the presence of pain (yes or no) and limitation of motion in the operated knees during routine postoperative appointments at one, three, and six months after implantation of the plates. In addition to the questionnaire, a clinical examination was performed in each case to determine if there was any limitation of the knee joint movement and range of motion (see Figure 1).

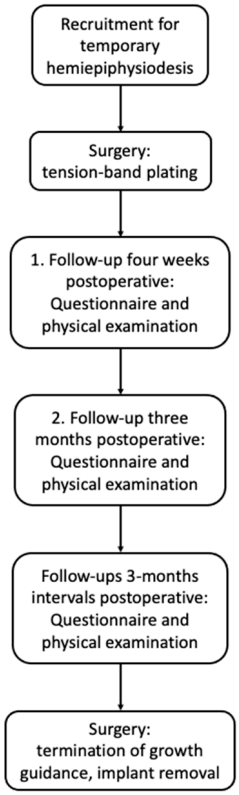

Figure 1. Flow chart of the study design.

Exclusion criteria were: rheumatoid arthritis, anterior cruciate ligament deficiency, neuromuscular disorders, achondroplasia or hypochondroplasia, sagittal plane deformities (genu pro- and recurvatum), flexion contractures in the hip or knee joint, leg length discrepancy of >10 mm, avascular necrosis of the femoral head or knee condyles, history of severe trauma or sport injury to the lower extremities, knee surgery within 12 months before enrollment in this study, chronic joint infections, or prior intraarticular corticosteroid injections.

Patients were divided into two groups: one with no particular complications after surgery (no complications group) and the other with marked complications persisting over a period of at least five weeks after surgery. Marked complications were defined as persistent pain and limited mobility of the operated knee after hemiepiphysiodesis plating between five weeks and six months. Participants and their parents provided written informed consent to participate in this study, which was approved by the local ethics committee (182/16) and in accordance with the Helsinki Declaration (date of approval: 30 December 2015). This study was registered with DRKS (German Clinical Trials Register) under the number DRKS00010296.

2.2. Radiographic Measurements

Implant position was analyzed on postoperative lateral and anterior–posterior X-rays of the knee. Data were obtained on the insertion site (femur and/or tibia), implant size, angulation of the plate in relation to the shaft axis, angulation of the screws at time of implantation (parallel, divergent, convergent), ratio between screw length and epiphyseal width (Figure 2), plate position in relation to the center of the shaft axis, and plate position in relation to the physis (Figure 3). A 25.4 mm diameter metal ball was placed adjacent to the knee and served as a reference for determining the individual magnification factor. Radiographic measurements were performed by the same orthopedic surgeon (SB) with a commercially available templating program, mediCAD® (version 5.98; Hectec, Niederviehbach, Germany).

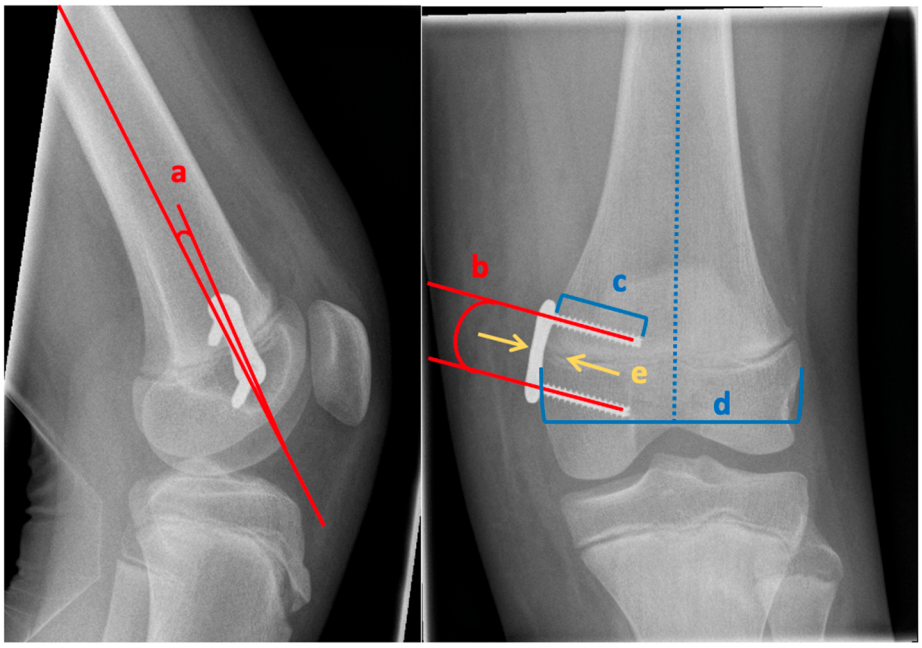

Figure 2. Radiological assessment: a—angle between the implant and femur axis, b—angle between the two screws, c—length of the screw, d—width of the epiphysis (greatest width of the epiphysis perpendicular to the axis of the femur), e—relation of the center of the plate to the physis (left arrow pointing at the center of the plate and right arrow pointing at the center of the physis).

Each patient's medical record was reviewed for information on age, sex, diagnosis with relevant leg malalignment and joint angle parameters (MAD, MFA, joint line convergence angle), as well as the date of implantation and surgery to remove the hemiepiphysiodesis plates. In addition, data were extracted from the surgical report: type of anesthesia, use, duration, and pressure amplitude of tourniquet, and duration of surgery. All patients received general anesthesia combined with regional anesthesia (peripheral neuraxial blocks: single-shot adductor canal block, femoralis block, or psoas compartment block), followed by a standardized analgetic postoperative treatment with a dose of ibuprofen or acetaminophen adjusted to the patients' body weight.

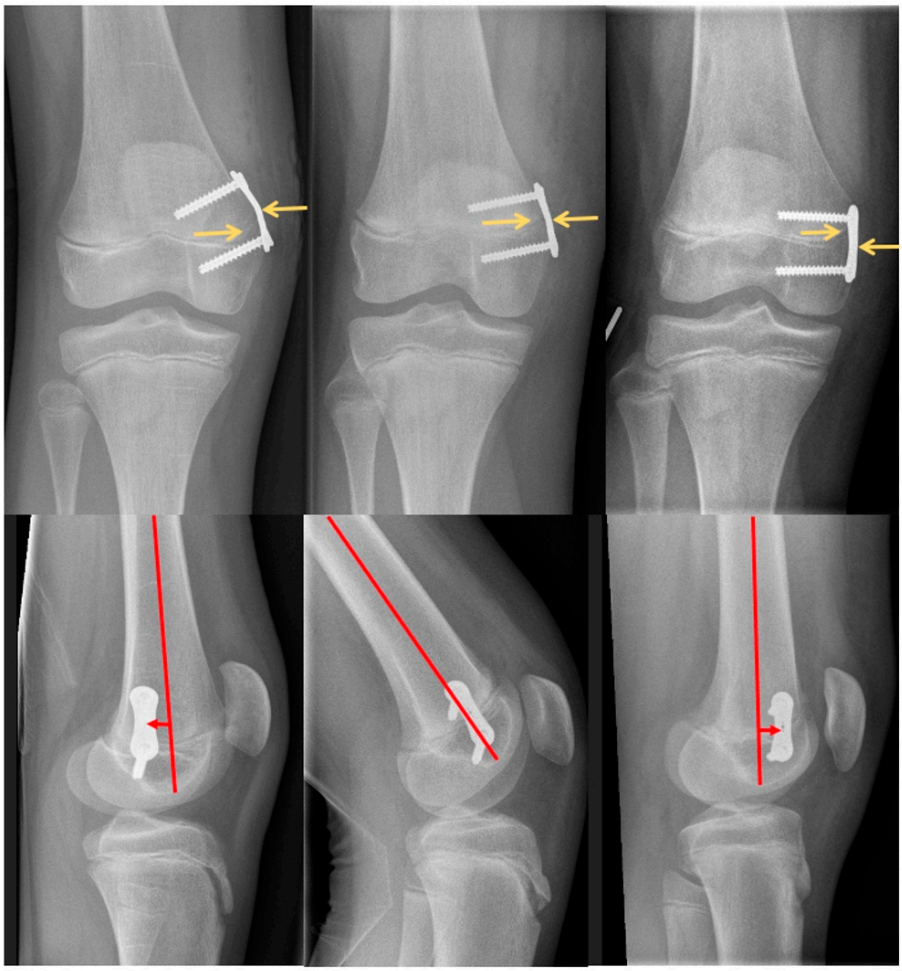

Figure 3. Radiological evaluation of the plate position: **top** row—plate position in relation to the physis, left arrow pointing at the center of the physis and right arrow pointing at the center of the plate) (**left** image: metaphyseal position, **center** image: centered position, **right** image: epiphyseal position); **bottom** row—plate position in relation to the center of the shaft axis, arrow pointing in the direction of insertion (**left** image: posterior position, **center** image: centered position, **right** image: anterior position).

2.3. Statistical Analysis

The Shapiro–Wilk test was used to test the normal distribution of the parameters analyzed. Non-parametric independent variables were compared with the Mann–Whitney U test. For normally distributed data, unpaired two-sided Student's *t*-tests were used to assess statistical significance between two sample means. Differences between nominally distributed variables were analyzed using the chi-square test or Fisher's exact test when the expected count was less than 5. For this explorative analysis, no confirmatory primary endpoint/hierarchical test approach was selected. The significance level for all statistical tests was set at $p \leq 0.05$. No α-adjustments for multiple testing were applied. Statistical data analysis was performed using SPSS (version 29, IBM Corporation, New York, NY, USA).

3. Results

Thirty-four patients met the criteria for evaluation. Group 1 (no complications) consisted of 22 patients, 55% of whom were female and had a mean age at surgery of 12.5 years (range 11.1–15.7). Group 2 (complications) included 12 patients (25% females) with a mean age at surgery of 13.2 years (range 11.5–14.3). Accordingly, 35% of patients had persistent pain and limited mobility of the operated knee for more than four weeks after hemiepiphys-

iodesis plating. Table 1 shows the anthropometric data and radiological extent of angular deformity before surgery. Patients had idiopathic genu valgum deformity with a mean MFA of 5.7° (1.8) and MAD of −19.4 (5.7) mm in group 1 and 5.3° (2.1) and −19.2 (7.9) mm in group 2. There were no statistical differences in anthropometric data, extent of the initial deformity, and duration of guided growth between the two groups. All patients experienced no complications other than the aforementioned mobility limitations and prolonged pain in the operated knee joint.

Table 1. Group differences.

Patients (n = 34)			
Parameter	**Group 1 No Complications**	**Group 2 Complications**	***p*-Value**
Patient characteristics			
Number of subjects	22 (65%)	12 (35%)	
Sex (female/male)	12 (55%)/10 (45%)	3 (25%)/9 (75%)	0.09
Age at surgery (years, months)	12.5 (1.1)	13.2 (1.0)	0.11
Height (m)	1.61 (0.10)	1.68 (0.13)	0.06
Body mass index (kg/m^2)	22.1 (3.8)	22.5 (3.6)	0.78
Extent of deformity (X-ray)			
Mechanical axis deviation (MAD) (mm)	−19.4 (5.7)	−19.2 (7.9)	0.90
Mechanical femorotibial angle (MFA) (°)	−5.7 (1.8)	−5.3 (2.1)	0.45
Joint-line conversion angle (°) *	1.0 (1.0–2.0)	1.0 (1.0–2.0)	0.32
Duration of guided growth (weeks) *	40.1 (34.3–51.7)	41.4 (41.0–46.9)	0.61

Parametric data: mean with standard deviation in parenthesis. * Non-parametric data: median with interquartile range in parenthesis. Mechanical axis deviation: negative values describe a valgus alignment. Mechanical femorotibial angle: negative values describe a valgus alignment.

Both groups differed significantly in plate positioning relative to the physis ($p = 0.049$). In group 1, 77.7% of plates were placed centered, 16.7% closer to the epiphysis, and 5.6% closer to the metaphysis. In group 2, 77.8% of plates were placed centered, 22.2% closer to metaphysis, and no plate was placed closer to epiphysis (Table 2). There was no difference in plate positioning in relation to the shaft axis (parallel, in flexion, in extension) ($p = 0.312$), plate position in relation to the center of the shaft axis ($p = 0.388$), angulation of the screws at the time of implantation (parallel, convergent, or divergent) ($p = 0.264$), and screw length/epiphysis width ratio ($p = 0.797$).

Table 2. Radiographic evaluation of implant positioning/characteristics.

Parameter	**Group 1 No Complications**	**Group 2 Complications**	***p*-Value**
Plate position in relation to the physis			0.049
centered	28 (77.7%)	14 (77.8%)	
epiphyseal	6 (16.7%)	0 (0.0%)	
metaphyseal	2 (5.6%)	4 (22.2%)	
Plate position in relation to the shaft axis of femur/tibia (°)	2.1 (11.2)	5.9 (13.5)	0.31
Plate position in relation to the center of the shaft axis of femur/tibia			0.39
centered	18 (50.0%)	10 (71.4%)	
posterior	12 (33.3%)	2 (14.3%)	
anterior	6 (16.7%)	2 (14.3%)	
Angulation of the two screws (°)	0.9 (5.3)	2.8 (6.3)	0.26
Epiphyseal width/diameter (cm)	8.2 (1.0)	8.4 (0.9)	0.44
Length of the screw (cm)	2.7 (0.4)	2.8 (0.4)	0.60
Ratio screw length/epiphyseal width	0.34 (0.03)	0.33 (0.03)	0.80

Parametric data: mean with standard deviation in parenthesis. Positive values of plate position in relation to the shaft axis of femur/tibia mean plate insertion in flexion, negative values mean plate insertion in extension. Positive values of angulation of the screws mean divergent screws, negative values mean convergent screws.

In group 1, 18 patients (82%) underwent bilateral surgery, compared to 10 patients (83%) in group 2. Four patients (18%) in group 1 and two patients (17%) in group 2 underwent unilateral surgery. There were significant differences in the distribution of implant localization between these two groups ($p = 0.016$). In 90.2% of patients in group 1, plates were inserted at the distal femur, in 4.9% of patients at the proximal tibia, and 4.9% of patients at the distal femur and proximal tibia. In group 2, plates were inserted at the distal femur in 69.2% of patients, and at the distal femur and proximal tibia in 30.8% of patients. In no patients were they placed only at the proximal tibia (Table 3).

Table 3. Types of implant and implant localization.

Parameter	Group 1 Complications	Group 2 Complications	*p*-Value
Procedures/Types of implant			
Unilateral surgery (number of patients)	4 (18%)	2 (17%)	
Bilateral surgery (number of patients)	18 (82%)	10 (83%)	
Number of implants	41	26	
Pedi-Plate (number)	37 (90%)	26 (100%)	
Eight-Plate™ (number)	4 (10%)	0 (0%)	
Implant localization			0.016
Medial distal femur	37 (90.2%)	18 (69.2%)	
Medial proximal tibia	2 (4.9%)	0 (0.0%)	
Medial distal femur and medial proximal tibia	2 (4.9%)	8 (30.8%)	

Parametric data: mean with standard deviation in parenthesis. There was no difference in the type of additive regional anesthesia ($p = 0.060$). All patients had general anesthesia and most also had additional regional anesthesia (see Table 4). Group 1 had a shorter duration of surgery (32 min vs. 38 min, $p = 0.032$) and a lower pressure of tourniquet (250 mmHg vs. 270 mmHg, $p = 0.019$) compared with group 2. The duration of tourniquet inflation showed no significant difference ($p = 0.162$).

Table 4. Group differences.

Parameter	Group 1 Complications	Group 2 Complications	*p*-Value
Type of anesthesia			0.06
Adductor canal block + general anesthesia	27 (69.3%)	17 (77.3%)	
Femoralis block + general anesthesia	8 (20.5%)	0 (0.0%)	
Psoas compartment block + general anesthesia	2 (5.1%)	3 (13.6%)	
General anesthesia	2 (5.1%)	2 (9.1%)	
Surgery characteristics			
Tourniquet pressure (mmHg) *	250 (250–265)	270 (250–280)	0.019
Tourniquet inflation (minutes)	34 (9)	37 (15)	0.16
Duration surgery (minutes)	32 (10)	38 (13)	0.032

Parametric data: mean with standard deviation in parenthesis. * Non-parametric data: median with interquartile range in parenthesis.

4. Discussion

With this study, we aimed to investigate implant-associated, surgery- or anesthesia-related, and other risk factors for complications regarding prolonged pain and limited range of motion postoperatively in children and adolescents with an idiopathic valgus deformity treated with temporary hemiepiphysiodesis. We hypothesized that implant insertion and position are related to function and pain after hemiepiphysiodesis plating and that screw implantation angle is associated with postoperative complications.

The results of the present study indicate that neither the initial extent of lower limb malalignment nor the timing of implant-mediated growth guidance (time from implant placement to implant removal) appear to be associated with a high number of prolonged postoperative complications. Anthropometric characteristics such as body mass index and age also had no statistical effect on the rate of prolonged pain after surgery.

Fillingham et al. [9] showed that patients older than 11 years of age at the time of implantation tended to have a greater delay in function. The results of the present study could not exactly confirm this finding because all our patients were older than 11 years and there was no difference in the age distribution of the two groups. However, because 35% of our patients had prolonged pain and functional delay, a comparison with a younger cohort of patients would be useful to support their findings.

In the present study, we demonstrated that the precise positioning of the plates in relation to the physis may have an impact on the postoperative complication rate. In this context, a metaphyseal position of the plate could lead to increased and prolonged postoperative pain and functional limitations. We suspect that periosteal preparation further proximally (femoral) or distally (tibial) leads to increased muscle dissection, which could result in a prolonged healing process with pain and limited range of motion. In addition, 25% of patients from group 2 (complications) had a protruding plate (Figure 4). A metaphyseal positioned plate does not necessarily have to protrude, as care should be taken to ensure contour-fit insertion onto the corticalis of the femur during implantation. Nevertheless, a metaphyseally placed plate may protrude more easily. In this case, soft tissue could interpose between the bone and the plate and cause pain symptoms. Therefore, surgeons must be careful to position the implant correctly to avoid such complications.

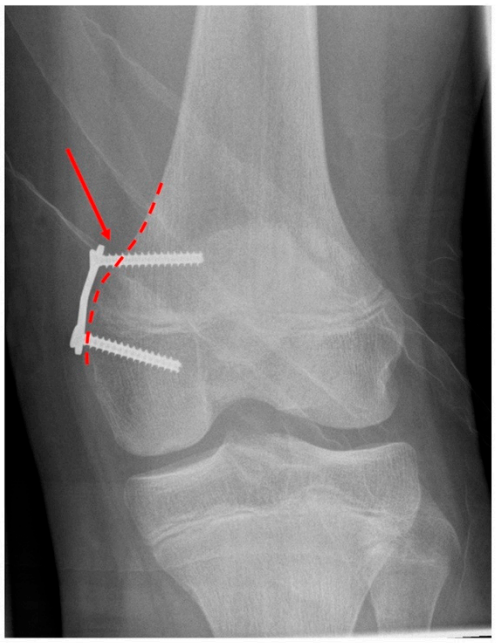

Figure 4. Metaphyseal plate position with pull-out and loss of initial contouring of the metaphyseal screw. Dashed line outlines the bone structure and cortical contour. Arrow pointing at the pull-out and loss of initial contouring resulting in lift-off of the plate at the metaphyseal end.

We hypothesized that the implantation angle of the screws has an influence on postoperative pain due to the compression force exerted on the physis [14]. Here, the existing literature is divided as to whether a divergent angle of the screws increases the correction rate and exerts more pressure on the epiphyseal joint. Burghardt et al. [15] suggested that the rate of correction is slower when the screws are initially inserted in parallel, and the correction accelerates when the screws become more divergent as growth progresses. In contrast, Schoenleber et al. [16] demonstrated in a biomechanical study that initial parallel screw positioning results in faster correction compared to divergent screws. Eltayeby et al. [17] showed that the initial screw angle ranging from parallel to 30° in divergence had no significant effect on the speed of correction during hemiepiphysiodesis. Assuming that screw angle does indeed result in an altered correction velocity due to pressure and compression force changes, these altered pressures could also result in greater pain

and limitation of knee range of motion. To our knowledge, no previous study has investigated the effect of screw angle on postoperative knee pain and range of motion after hemiepiphysiodesis plating. In our study, we were able to demonstrate that the initial screw implantation angle did not make a difference between the two groups and, therefore, was unlikely to be responsible for persistent pain symptoms or limitation of knee range of motion. Consequently, our hypothesis regarding the angulation of the screws has to be rejected.

Fillingham et al. [9] showed that plating bilaterally versus unilaterally, femur versus tibia, or the number of implants used for implant-mediated growth guidance conferred a greater risk of functional delay. Given the predominance of plates in the distal femur (only 2 of 66 implants were placed at the proximal tibia) in our patients, our study is not sufficiently powered to detect a difference based on location. Nevertheless, in accordance with the study by Fillingham et al. [9], the results of the present study showed that patients with simultaneous tibial and femoral implantation had higher complication rates ($p = 0.016$).

Regional anesthesia with peripheral neuraxial blocks is commonly performed in pediatric orthopedic surgery [18]. They are associated with a very low risk of complications [19,20] and have the same effect regardless of the different types of regional anesthesia [21]. In the present study, we demonstrated that the different types of regional anesthesia (adductor canal block, femoralis block, psoas compartment block) combined with general anesthesia showed no difference in postoperative pain or a delay of function after surgery. However, in the present study, patients tended to have a higher complication rate and pain after implantation of hemiepiphysiodesis plates at higher tourniquet pressures. Accordingly, Hanna et al. [22] found a reduction of opioid consumption in the postoperative period by avoiding the use of tourniquet in pediatric patients with lower limb surgery. Another study from Kamath et al. [23] found that the incidence of tourniquet pain was directly proportional to the duration of tourniquet use but not to the amplitude of tourniquet inflation pressure. In their study, 7.7% of patients experienced tourniquet pain after a surgery that lasted less than 60 min, compared with 35.8% after a procedure that lasted longer than 60 min. Tourniquet pain is described as a poorly localized, dull, tight, aching sensation at the site where the tourniquet is applied [24]. In the present study, the duration of the procedure did not exceed 60 min in either group (group 1: mean 32 min: group 2: mean 38 min). Therefore, it is difficult to distinguish between surgery-related pain and tourniquet-related pain in our patient group. Lieberman et al. [25] investigated tourniquet pressures with values of 50 mmHg above the occlusion pressure measured by Doppler. According to their work, lower tourniquet pressures (176.7 +/− 28.7 mmHg, range 140 to 250 mmHg) can maintain adequate hemostasis in a lower extremity surgery in pediatric patients [25]. Consistent with our data, we can recommend tourniquet pressures of no more than 250 mmHg to preclude persistent complications and impairments after surgery.

In children and adolescents, symptoms such as persistent pain, restricted mobility, and range of motion after surgery may also be caused by the complex regional pain syndrome (CRPS). In affected children, the peak of CRPS type I, the cause of which has not yet been conclusively identified but which can occur after surgery, appears to be at the age of 13 years. Chronic pain, generally unilateral and limited to the extremities, autonomic and motor dysfunction, and trophic disturbances are the main symptoms of CRPS type 1 [26]. The diagnosis of CRPS type 1 is typically made clinically and is based on the Budapest diagnostic criteria [27]. However, the diagnosis remains difficult given the lack of validated diagnostic tests and the difficulties in differential diagnosis [26]. The children in this study were not explicitly screened for this diagnosis.

From an anatomical point of view, the cause of prolonged postoperative pain can be assumed to be the injury of small cutaneous nerves due to the open procedure itself. The medial and lateral part of the knee are innervated by different nerves. The saphenous nerve is the primary cutaneous nerve that supplies sensation to the skin over the medial knee. The superior medial genicular nerve also supplies sensation to the skin over the medial knee,

both are branches of the femoral nerve. The superior lateral genicular nerve is a branch of the common peroneal nerve (a branch of the sciatic nerve) which supplies sensation to the lateral aspect of the knee joint capsule and the skin over the lateral knee [28]. Even during such a minimally invasive procedure as the implantation of temporary hemiepiphysiodesis, these small nerve branches can be injured. One solution can be the percutaneous insertion of the plates and screws through two 6 mm incisions, which, according to a large amount of experience in percutaneous insertion, reduces this frequently observed, undesirable complication after surgery [29].

When interpreting the results of the present study, its limitations should be considered. The influence of the surgeon on the success of surgery cannot be demonstrated with certainty because of the large number of surgeons involved. In addition, it should be noted as a limitation that the postoperative assessment and questionnaires did not explicitly differentiate between pain at the knee or at the insertion site of the plate and the application of the tourniquet to the proximal thigh. Lastly, no adjustment for multiple testing was applied, thus overall, our results and their interpretation have an exploratory character and should be treated with some caution as they can be due to coincidence.

5. Conclusions

In conclusion, 35% of patients had more postoperative limitations than expected, with persistent pain and limited mobility of the operated knee after hemiepiphysiodesis plating between five weeks and six months. In implant-mediated growth guidance with hemiepiphysiodesis plating for temporary hemiepiphysiodesis, the simultaneous implantation of the plates at the femur and tibia and the metaphyseal positioning of the plates may result in prolonged pain symptoms and a delay of function. In addition, surgery-related factors such as the amplitude of tourniquet inflation pressure or the duration of surgery, could play a role in the development of a poor outcome. In contrast, neither the initial extent of lower limb malalignment nor the timing of implant-mediated growth guidance (time from implantation to implant removal) appear to be associated with longer-lasting postoperative complications. Body mass index and age also had no effect on the complication rate after surgery.

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Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.

Data Availability Statement: The data presented in this study are available upon request from the corresponding author.

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children

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Article

Functional Benefit and Orthotic Effect of Dorsiflexion-FES in Children with Hemiplegic Cerebral Palsy

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Abstract: Functional electrical stimulation of the ankle dorsiflexor (DF-FES) may have advantages over ankle foot orthoses (AFOs) in managing pediatric cerebral palsy (CP). This study assessed the functional benefit and orthotic effect of DF-FES in children with hemiplegic CP. We conducted an open-label prospective study on children with hemiplegic CP $\geq$ 6 years who used DF-FES for five months. The functional benefit was assessed by repeated motor function tests and the measurement of ankle biomechanical parameters. Kinematic and spatiotemporal parameters were assessed by gait analysis after one and five months. The orthotic effect was defined by dorsiflexion $\geq 0^{\circ}$ with DF-FES at either the mid or terminal swing. Among 26 eligible patients, 15 (median age 8.2 years, range 6–15.6) completed the study. After five months of DF-FES use, the results on the Community Balance and Mobility Scale improved, and the distance in the Six-Minute Walk Test decreased (six-point median difference, 95% CI (1.89, 8.1), –30 m, 95% CI (−83.67, −2.6), respectively, $p < 0.05$) compared to baseline. No significant changes were seen in biomechanical and kinematic parameters. Twelve patients (80%) who showed an orthotic effect at the final gait analysis experienced more supported walking over time, with a trend toward slower walking. We conclude that the continuous use of DF–FES increases postural control and may cause slower but more controlled gait.

Keywords: hemiplegia; cerebral palsy; FES; functional benefit

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1. Introduction

Cerebral palsy (CP) defines a group of permanent disorders of movement and posture that are attributed to non-progressive disturbances in the developing brain [1]. Hemiplegic CP accounts for 21–40% of all cases of CP [2,3]. Children with hemiplegic CP are typically ambulant with high motor functioning (Gross Motor Function Classification Scale (GMFCS) I/II), but with asymmetry of gait and a greater risk of instability and falling [4]. The ankle joint is affected in virtually all patients, causing insufficient clearance of the foot during the swing phase ("foot drop") and abnormal foot contact during the stance phase of gait [5].

Children with hemiplegia show deviations in their spatiotemporal gait parameters. Their gait is asymmetric, as manifested by a shorter stance phase on the affected side compared to the unaffected side. In addition, these children have slower walking speeds and a more supported gait, with a longer double support phase (when both feet are in contact with the ground) than typically developing children. [6,7].

Ankle–foot orthoses (AFOs) are usually prescribed to improve foot positioning and prevent foot drop in these children [8]. However, while AFOs have known benefits, compliance deteriorates as children get older [9].

Functional electrical stimulation (FES) is a well-known neuroprosthesis that delivers electrical stimulation to the motor nerve and activates the desired muscle group. Dorsiflexion FES (DF-FES) stimulates the common peroneal nerve and activates the ankle dorsiflexors, correcting upper motor neuron foot drop [10]. Theoretically, the repetitive stimulation of the DF muscle may give DF-FES an advantage over AFOs, which passively support the ankle and may increase muscle atrophy [8,11,12]. Indeed, studies have shown improvement in ankle kinematic parameters with DF-FES (this is typically referred to as an orthotic effect). However, the orthotic effects of DF-FES may vary between children [13,14]. There is no consensus on what clinical features can predict appropriate candidates for DF-FES, although an adequate ankle range of motion (ROM) was suggested as a prerequisite [15].

In addition, studies in adults and children have pointed to a "therapeutic effect" after the continuous use of DF-FES, meaning improvement in any aspect of gait, including biomechanical or other functional parameters, which continues when the patient is not using the DF-FES device. Some studies have shown peripheral improvement in ankle biomechanical parameters, e.g., ankle ROM, muscle strength and size, and spasticity [6–18]. Studies have also shown improved balance scores [8,18–22]. However, studies examining a kinematic therapeutic effect have reached conflicting conclusions [11,18,19]. In addition, the findings on the effects of continuous DF-FES use on spatiotemporal parameters and walking speed are scarce and mixed [19,21,23]. Finally, the long-term therapeutic effect of DF-FES—namely, whether it has a prolonged carry-over effect (without the device) or whether improvements are only temporary—remains a major open question [15].

The aim of this study was to evaluate the functional and therapeutic effects of DF-FES use over five months in children with hemiplegic CP, including effects on postural control; walking endurance and speed; and ankle biomechanical, spatiotemporal, and kinematic parameters. In the rest of this paper, the effects observed with DF-FES turned on will be called functional effects or benefits, while the effects observed with DF-FES turned off will be called therapeutic effects.

In addition, we looked for clinical, kinematic, and biomechanical parameters that may serve as predictor(s) for the achievement of an orthotic effect. Previous studies [13,18,19,24–26] used improvement in any ankle swing kinematic parameters (e.g., peak dorsiflexion angle) to indicate an orthotic effect. We introduce a more precise measure for the orthotic effect of DF-FES. In this study, an orthotic effect was defined by whether it prevents excessive swing plantarflexion (>1 SD from the mean)—i.e., foot drop—and achieves swing dorsiflexion $\geq 0^{\circ}$ at the mid or terminal swing phase.

2. Materials and Methods

2.1. Study Design

This was a prospective open-label study on hemiplegic CP children with foot drop who used DF-FES (WalkAide; Innovative Neurotronics, Austin, TX, USA) for five months. Motor function tests were conducted at baseline, after one month, and after five months of device use. Motor tests included the Community Balance and Mobility Scale (CB&M), the Six-Minute Walk Test (6MWT), and the Timed Up and Down Stairs Test (TUDS) (see details below).

Falling questionaries were filled in by parents at baseline and at the end of the study. In addition, at the end of the study, children filled in satisfaction questionaries. Biomechanical ankle parameters (see below) were taken at baseline and at the end of the study.

To test for the presence of an orthotic effect, we conducted gait analysis with DF-FES switched off and switched on. On the assumption that the device requires adjustment, in order to assess the true orthotic effect, the first gait analysis was conducted after one month.

Gait analysis was repeated after five months of device use, allowing us to assess changes in kinematic and spatiotemporal parameters (Figure 1).

To assess predictors for failure or success in the achievement of an orthotic effect (namely, swing dorsiflexion $\geq 0^{\circ}$ at the mid or terminal swing phase), the clinical kinematic and biomechanical parameters of children who showed vs. did not show an orthotic effect were compared (see below).

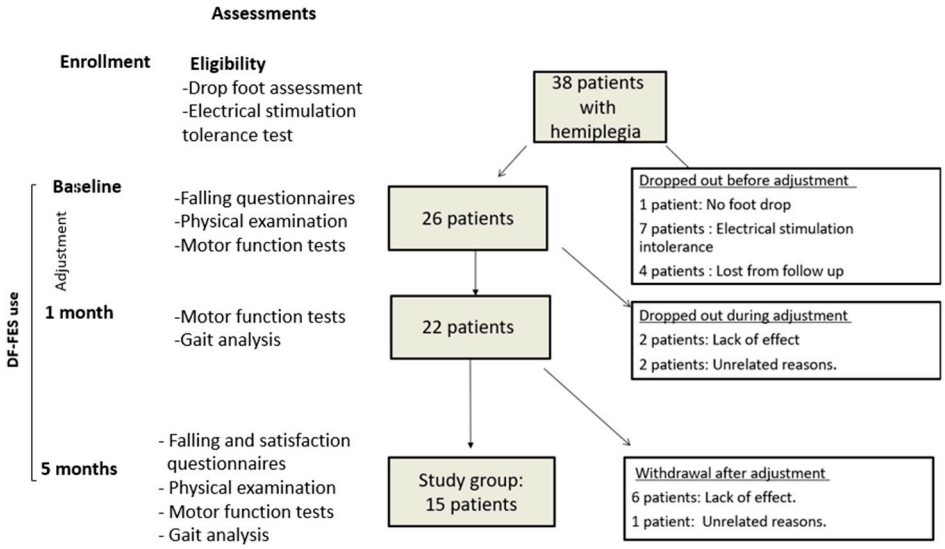


Figure 1. Flow chart and study design.

2.2. Study Population

The inclusion criteria comprised children ($\geq$6 years) with hemiplegic CP, GMFCS I/II, and foot drop. The exclusion criteria were fixed ankle joint contracture (passive ROM $< 0^{\circ}$ with knee extended); an inability to tolerate the electrical stimulation of DF-FES; orthopedic surgery or botulinum toxin injection to the lower limbs within six months of enrollment; moderate to severe intellectual disabilities; or uncontrolled epilepsy. For the power calculation, we used Shieh's [27] method. For a power of 0.8 with alpha = 0.05, based on previous studies [18,19], and assuming a median-difference-to-standard-deviation ratio of 0.8 in motor function tests, the required sample size was calculated as between 11 (Laplace distribution) and 14 (uniform distribution). The final sample in our study comprised 15 patients (see Results Section 3 and Figure 1).

2.3. Enrollment

The passive ankle range of motion (to ensure passive ROM $\geq 0^{\circ}$ with knee extended), tolerance of DF-FES, and foot drop were assessed at enrollment to determine inclusion (Figure 1). Foot drop was defined by excessive swing plantarflexion during the mid or terminal swing (>1 standard deviation (SD) from the mean). During enrollment, patients were asked to walk barefoot for 15 m using the RehaGait mobile gait analysis system (RehaGait, HASOMED GmbH Service, Magdeburg, Germany). This is a mobile clinical tool equipped with motion sensors which utilize the inertia of the mass to detect movement changes. The sensors include a three-axis accelerometer for recording linear acceleration, a gyroscope for recording angular velocity, and a magnetometer for recording orientation in relation to the earth's magnetic field; the system measures kinematics and angles while producing a graphical presentation of the gait cycle. In previous studies [28,29], measures of sagittal plane joint kinematics produced by this system were comparable to those from a camera-based system. A graphical presentation of the gait cycle (with upper and lower borders $+/-1$ SD) was used to assess excessive plantarflexion (>1 SD) during the mid/terminal swing.

2.4. Motor Function Tests

Motor function tests were conducted at baseline, after one month, and after five months. All tests were carried out by the same physiotherapist, in the same order, for all patients, and at each visit. Each test was conducted with the device attached to the patient's leg. At baseline (before adjustment), tests were carried out with the device switched off, and at one and five months, each test was carried out first with the device switched off and then with it switched on. Motor tests included the Community Balance and Mobility Scale (CB&M) [30], the Six-Minute Walk Test (6MWT) [31], and the Timed Up and Down Stairs Test (TUDS) [32].

2.4.1. Community Balance and Mobility Scale (CB&M)

The CB&M is a clinical tool used to assess postural stability and dynamic balance. It includes tasks that are representative of the motor skills thought to be necessary for everyday functioning in community settings. It has been used in adults and children with acquired brain injury and also in high-functioning children with hemiplegic CP who reach a ceiling effect on other objective measures such as the Gross Motor Function Measure (GMFM). The CB&M comprises a total of 13 tasks, with 6 items measured bilaterally. Each task is rated on a six-point scale (0–5), with one item allowing for a bonus point. The highest possible score is 96. A change of five points is considered clinically meaningful [18].

2.4.2. Timed up and down Stairs Test (TUDS)

The TUDS is a functional mobility test used to assess postural control [32]. The patients were asked to walk up and then down an 11-step flight of stairs. The steps were 18.5 cm high and 31 cm deep; no stickers or other means were used to help patients determine the depth or height. At the start of the test, the patients were asked to stand 30 cm from the bottom step. They were then instructed to go up quickly but safely, to turn around on the top step, and to descend until both feet were on the bottom step. The patients were allowed to choose any method of traversing the stairs, including using a handrail. The TUDS score was the time in seconds from the start cue until the second foot returned to the bottom step [30]. To the best of our knowledge, no minimal meaningful change for this test has been published for the cerebral palsy population.

2.4.3. Six-Minute Walk Test (6MWT)

The 6MWT has been reported to reflect functional capacity in terms of activities of daily living in the cerebral palsy population [31]. The patients were instructed to walk as far as possible in a straight line for six minutes along a 10 m course, without running or jogging. They were permitted to slow down or stop to rest but were instructed to resume walking as soon as they could. Masking tape was placed at two-meter intervals along the course, and the distance covered in six minutes was recorded to the nearest meter [31]. Thompson et al. [33] reported minimal meaningful changes for school-aged children with cerebral palsy of 61.9 m and 64.0 m for GMFCS Levels I and II, respectively.

Overall, we hypothesized that patients using DF-FES continuously would show a functional benefit manifested by an improvement in stability and balance, a greater distance in the 6MWT due to increased walking endurance and/or speed, and faster speed walking up and down stairs.

2.5. Falling and Satisfaction Questionnaires

The parents filled out a questionnaire in which they scored their child's falling frequency as daily, weekly, monthly, or less at baseline and at the end of the study. At the end of the study, the children were asked to rate their satisfaction with the device on a scale of 1–5 (1 = very much, 5 = not at all).

2.6. Ankle Biomechanical Assessments

At baseline and after five months of device use, four assessments—plantarflexor muscle spasticity, dorsiflexor muscle strength, muscle selectivity, and a precise measurement of passive ankle ROM (which was tested at enrollment only for exclusion criteria)—were carried out by a physiotherapist, as follows:

1. Passive ankle range of motion with knee flexion and extension (with the leg supported on a bed), with the subtalar joint maintained in a neutral position. The measurement was conducted using a goniometer aligned at one end with the fibula and at the other end with the fifth metatarsal bone [34]. Foot deformities such as midfoot break were accounted for by accurately measuring calcaneal dorsiflexion with the foot held in supination.
2. Plantar-flexor muscle spasticity with knee flexion and extension (for soleus and gastrocnemius muscle assessment, respectively). The measurement was conducted by dorsiflexion of the foot from maximum possible plantarflexion to maximum possible dorsiflexion. Spasticity was scored using the modified Ashworth scale [35].
3. Muscle selectivity. Ankle joint selectivity was measured using the Selective Control Assessment of the Lower Extremity (SCALE), with patients in a sitting position with the knee extended. Patients were asked to move their foot up, down, and up again. Selectivity was scored based on a three-point scale (zero points = unable, one point = impaired, two points = normal) [36].
4. Dorsiflexor muscle strength. Strength was evaluated in side-lying and seated positions using Kendall's manual muscle testing scale [37].

2.7. Dorsiflexion-Functional Electrical Stimulation Device (DF-FES)

The WalkAide device (WalkAide; Innovative Neurotronics, Austin, TX, USA) is a small device which is attached to the patient's leg by a cuff that sits just under the knee on the affected side. One electrode is placed on the belly of the tibialis anterior muscle, and the other on the common peroneal nerve. Electrical stimulation is triggered by a tilt sensor which senses the change in the tibia angle during the swing phase [24].

For each patient, at baseline setup, the pulse width was adjusted to between 25 and 50 microseconds, and the frequency range was adjusted to between 16.7 and 33 pulses/s, in order to achieve ankle dorsiflexion with tolerable stimulus. During the one-month adjustment period, families were instructed to (a) gradually increase the intensity of electrical stimulation according to the child's tolerance in order to maximize ankle dorsiflexion; and (b)to reach minimal requirements for constant DF-FES use of at least, on average, 5 days/week, 4 h/day and 1500 steps/day (75% of the average steps per day in our preliminary tests). Compliance with these requirements was confirmed via the device's internal log, which recorded stimulations ("stims") and hours of "device on" per day.

The patients were instructed to maintain good hygiene in order to avoid skin irritation and burns beneath the electrodes.

2.8. Gait Analysis

Gait analysis was conducted at the end of the first month of device use (allowing for adjustment) and was repeated at the end of the study after five months of use. Gait analysis was performed in the laboratory using a three-dimensional motion analysis system (the Plug-in Gait model (PGM) by Vicon®, Oxford Metrics, UK) with a sampling rate of 120 Hz. The Plug-in model provides full upper or lower body joint kinematics and kinetics modeling using a pre-defined Plug-in Gait marker set. These retro-reflective markers were applied to anatomical landmarks to capture gait performance [38]. Where a midfoot break was identified, the forefoot marker was placed proximally along the foot axis toward the hindfoot in order to avoid measuring midfoot dorsiflexion (as opposed to ankle dorsiflexion).

The children were asked to walk barefoot at a self-selected speed along a 14 m walkway. Ten representative gait cycles were captured from five gait trials conducted with FES turned

off, and another ten cycles were captured from five trials conducted with FES turned on. Ankle kinematic parameters were analyzed at defined points in each cycle: initial contact angle; maximal and minimal dorsiflexion angle at the mid and terminal swing; and maximal ankle dorsiflexion at mid-stance as an indication of ankle ROM during the weight-bearing state [39]. Video recordings of the trials were used to evaluate the heel strike. We hypothesized that DF-FES would improve kinematic parameters and correct swing foot drop (i.e., produce an orthotic effect) in those with a better ankle ROM.

In addition, spatiotemporal parameters were analyzed, including stance time (in sec) and percentage of gait cycle; walking speed; cadence (steps/min); step time (in sec) and length (in cm); and single and double support time (in sec). Although data in the literature are scarce, we hypothesized that if DF-FES has a beneficial effect on the spatiotemporal parameters of gait, we should expect a trend toward the normalization of gait deviations when comparing the final gait analysis to the first one, including reduced supported gait (with a decrease in the double/single support time ratio), an increased walking speed, and an increase in the percentage of the stance of the gait cycle.

2.9. Orthotic Effect

In this study, the orthotic effect was defined by whether it prevents excessive swing plantarflexion (>1 SD from the mean)—i.e., foot drop—and achieves swing dorsiflexion $\geq 0^{\circ}$ at the mid or terminal swing phase. To test for an effect, we compared the minimal and maximal dorsiflexion angles at the mid and terminal swing using the medians of the 10 gait cycles captured with DF-FES turned off and on. An orthotic effect was considered present (OE+) when DF-FES produced dorsiflexion ($\geq 0^{\circ}$) at either the mid or terminal swing and absent (OE–) when no such change appeared.

2.10. Statistical Analyses

The results of repeated motor function tests, the kinematic and spatiotemporal parameters, and the ankle biomechanical parameters of the 15 patients who completed the study protocol were compared by the Wilcoxon signed-rank test. CB&M scores were compared only for 14 patients because of low cooperation by one girl (6.5 years old). In order to define the right candidates for DF-FES, demographic, clinical (e.g., AFO use and history of botulinum toxin injection), and physical parameters (e.g., ankle spasticity using the Modified Ashworth Scale (MAS) and ROM) were compared between the study group and patients who dropped out. In addition, demographic, clinical, physical, kinematic, and device-use parameters were compared between patients who did and did not experience an orthotic effect according to the first gait analysis, using Fisher's exact test for categorical variables (e.g., age, GMFCS, MAS score, muscle strength, and selectivity) and the Mann–Whitney test for continuous variables (e.g., ankle ROM, ankle kinematic parameters, and device use parameters (stims and hours/day)) (see Supplementary Table S2). Since most patients who did not experience OE dropped out during the study, this analysis could not be conducted at the final gait analysis. The diagnostic value of various parameters as predictors for an orthotic effect was assessed using the Pearson correlation coefficient. MedCalc Statistical Software version 20.115 (MedCalc Software, Ostend, Belgium; https://www.medcalc.org (accessed on 1 January 2023); 2020) was used for the statistical analyses.

3. Results

3.1. Clinical and Demographic Variables

Among 38 patients with hemiplegia who were tested for eligibility, 26 patients met the inclusion criteria and began to use DF-FES according to the adjustment instructions. Twenty-two patients completed the first month of use according to the minimal requirements and underwent gait analysis. Fifteen patients (the study group) completed five months of device use and repeated the motor function tests, biomechanical tests, and gait analyses. Those 15 patients showed good compliance, with an overall average of 3802 $\pm$ 790.8 stims/day,

21.6 ± 4.86 days/month, and 6.72 ± 1.55 h/day. Satisfaction questionnaires showed a high satisfaction (average 1.82 ± 1.25 on a scale from 1 (very much) to 5 (not at all)).

Of the 11 patients who dropped out between enrollment and the five-month mark, eight patients (72%) withdrew due to a lack of a positive effect of FES on gait (as perceived by the patient and family), and three (28%) withdrew for unrelated reasons (Figure 1). Comparing the clinical and demographic variables, a history of botulinum toxin injections was more prevalent in patients who dropped out (n = 11) than in the study group (73% vs. 20%, respectively; p = 0.01) (Table 1).

Table 1. Clinical and demographic parameters of the study group and patients who withdrew.

Parameters	Study Group (N = 15)	Withdrawn Patients (N = 11)
Age (y)	8.2 (7, 10.5)	8 (6.8, 8.8)
M:F	10:5	5:6
Term (≥37 w)	7 (47%)	6 (55%)
GMFCS I:II	14:1	8:3
Current AFO use	7 (46%)	7 (63%)
Botulinum toxin—LL		
No	12 (80%)	3 (27%)
Yes	3 (20%)	8 (73%) *
Surgery to LL (n)		
No	14 (93%)	9 (82%)
Yes	1(7%)	2 (18%)
MAS		
1	2 (13%)	3 (27%)
1+	4 (27%)	2 (18%)
2	9 (60%)	6 (55%)
Passive ankle ROM		
Knee flexion (degrees)	10° (5, 15)	10° (7.5, 15)
Knee extension (degrees)	5° (2, 9)	5° (1, 10)

* $p < 0.05$ (Fisher's exact test). M = Male; F = Female; AFO = ankle foot orthoses; LL = lower limb; MAS = Modified Ashworth Scale; ROM = range of motion.

3.2. Falling

According to the questionnaires filled out by the parents, 12 patients in the study group (60%) fell frequently (daily or weekly) at baseline vs. 3 (20%) after five months of FES use (p = 0.06).

3.3. Motor Function Tests

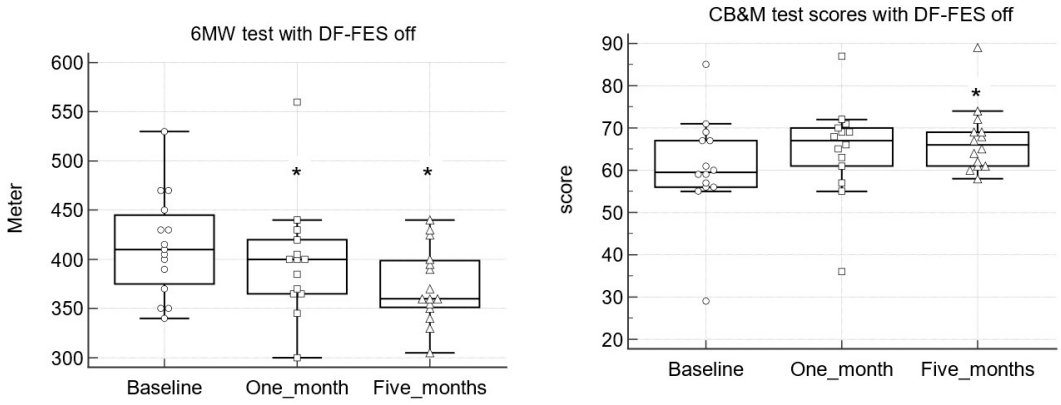
3.3.1. Community Balance and Mobility Scale (CB&M)

The CB&M measures postural stability and dynamic balance. Repeated CB&M tests at baseline, after one month, and after five months of FES use demonstrated improvement, with higher scores that reached statistical significance after both one month and five months. Comparing the baseline (DF-FES off) to the final test after five months (device on and off), we found median differences of 6.5 (95% CI (2.79, 10), $p < 0.01$) and 6 (95% CI (1.89, 8.1), $p < 0.01$) with DF-FES switched on and off, respectively. Between one and five months, there was a trend toward improvement, with increased median differences, although these findings were not significant (Table 2; Figure 2).

Table 2. Motor function scores—median difference.

Tests		Median Difference (95% CI) *		
		Baseline to 1 Month	**1 Month to 5 Months**	**Baseline to 5 Months**
CB&M (score) (N = 14)	**DF-FES off**	3 $^{\$}$ (1, 7.2)	3 (−3.2, 4.1)	6 $^{\$}$ (1.89, 8.1)
	DF-FES on	4.5 $^{\$}$ (0.89, 8.3)	1 (−4, 7.1)	6.5 $^{\$}$ (2.79, 10)
6 MWT (meter) (N = 15)	**DF-FES off**	−17.5 (−67.08, 15)	−17.5 (−73.12, 16.04)	−30 # (−83.67,−2.6)
	DF-FES on	−30 # (−55, −4.47)	−12.5 (−45.5, 11.04)	−35 # (−99.67, −3.97)
TUDS (sec) (N = 15)	**DF-FES off**	−0.19 (−2.34, 0.4)	−0.19 (−0.76, 0.7)	−0.83 (−2.28, 0.42)
	DF-FES on	−0.7 (−1.71, 1.19)	−0.68 (−1.71, 0.4)	−0.41 (−2.7, 0.28)

* Main comparisons are between baseline scores (DF-FES off) and one- or five-month scores (DF-FES off and on). CB&M = Community Balance and Mobility Scale; 6MWT = Six-Minute Walk Test; TUDS = Timed Up and Down Stairs Test. $^{\$}$ $p < 0.01$; # $p < 0.05$ (Wilcoxon test).



(**a**) (**b**)

Figure 2. Repeated motor function test (**a**) and scores (**b**) test scores with DF-FES off, at baseline and after one month and five months of device use. Error bars represent 95% CI of median. Dots represent individual data. CB&M = Community Balance and Mobility Scale; 6MWT = Six-Minute Walk Test; * $p < 0.05$ in comparison to baseline (Wilcoxon test).

3.3.2. Timed up and down Stairs Test (TUDS)

The TUDS assesses postural control, and improvement should be manifested by faster speeds in climbing up and down stairs. Our findings showed only a minor trend in this direction. The median differences in seconds between five months (FES on and off) and baseline (FES off) were −0.41 (95% CI (−0.27, 0.28), $p = 0.06$) and −0.83 (95% CI (−2.28, 0.42), $p = 0.09$) for FES on and off, respectively (Table 2).

3.3.3. Six-Minute Walk Test (6MWT)

The 6MWT is used to measure functional ability. According to previous results [17] and our hypothesis, improvement should be manifested by an increase in walking distance. However, our study showed the opposite trend. Repeated 6MW tests at baseline, after one month, and after five months of FES use revealed a decrease in walking distance over time, which reached statistical significance after one month with DF-FES switched on and after five months with the device both on and off (Table 2, Figure 2). The median differences in

distance between five months (DF-FES on and off) and baseline (DF-FES off) were −35 m (95% CI (−99.67, −3.97), $p < 0.05$) and −30 m (95% CI (−83.67, −2.6), $p < 0.05$) with DF-FES on and off, respectively. The trend toward reduced distance continued between one and five months but did not reach statistical significance (Table 2).

3.4. Kinematic, Spatiotemporal, and Biomechanical Parameters

3.4.1. Orthotic Effect

The study sample (N = 15) exhibited significant improvement in ankle kinematic parameters when DF-FES was turned on (vs. off) at both the first (after one month) and final (after five months) gait analysis, including the initial contact angle ($p < 0.01$) and minimal and maximal dorsiflexion during the mid and terminal swing ($p < 0.01$) (Table 3).

Table 3. Ankle kinematic and spatiotemporal parameters with DF-FES off and on at the first and final gait analyses.

	DF-FES off (N = 15)		**DF-FES on (N = 15)**	
FES	**First**	**Final**	**First**	**Final**
Maximal dorsiflexion—mid swing (degrees)	−4.57° (−9.1, 4.63)	−3.3° (−10.05, 4.06)	3.13° * (−5.97, 6.01)	3.2° # (−4.04, 6.64)
Maximal dorsiflexion—terminal swing (degrees)	−3.52° (−7.53, 2.05)	−3.86° (−7.77, 1.79)	3.97° * (−0.39, 6.49)	3.36° # (1.64, 7.62)
Minimal dorsiflexion—mid swing (degrees)	−11.68° (−15.07, 0.74)	−6.72° (−16.5, −0.99)	−0.97° * (−11.62, 1.63)	−0.58° # (−9.95, 3.28)
Minimal dorsiflexion—terminal swing (degrees)	−11.46° (−14.5, −4.74)	−11.24° (−15.59, −7.68)	−1.88° * (−6.28, 1.46)	−1.6° # (−6.82, 0.94)
Initial contact (degrees)	−7.08° (−9.1, −1.02)	−7.2° (−11.78, −3.79)	−0.49° * (−4.32, 2.99)	0.81° # (−2.33, 2.7)
Peak swing dorsiflexion (degrees)	−0.82° (−6.59, 4.89)	−2.18° (−4.3, 4.22)	4.51° * (0.08, 7.18)	4.27° # (1.94, 8.03)
Stance time (sec)	0.53 (0.46, 0.57)	0.56 (0.53, 0.63)	0.52 (0.49, 0.61)	0.56 (0.51, 0.59)
Stance—%gait cycle	56.88 (55.65, 57.59)	57.42 (56.59, 57.72)	56.29 (54.76, 56.96)	56.54 (55.61, 57.23)
Walking speed (meter/sec)	1.09 (0.955, 1.260)	1.04 (0.97, 1.13)	1.1 (0.95, 1.2)	1.07 (0.96, 1.17)
Cadence (steps/min)	127.9 (120.12, 147.16)	122.4 (111.42, 127.54)	126.32 (115.91, 134.91)	118.44 (112.69, 131.87)
Double/single support time ratio	0.42 (0.35 to 0.46)	0.47 (0.4 to 0.53)	0.42 (0.36 to 0.45)	0.4 (0.36 to 0.44)
Step length (cm)	0.49 (0.42, 0.52)	0.51 (0.46, 0.53)	0.51 (0.48, 0.57)	0.5 (0.46, 0.53)
Step time (sec)	0.49 (0.43, 0.54)	0.53 $ (0.5, 0.59)	0.52 (0.48, 0.54)	0.54 (0.48, 0.5)

Data are presented as median degrees (interquartile range (IQR)); * First gait analysis: FES off vs. FES on, $p < 0.01$; # Final gait analysis: FES off vs. FES on, $p < 0.01$. $ Final vs. first gait analysis, $p < 0.05$ (Wilcoxon test).

Analyzing the kinematic effect of the device at the first gait analysis (N = 22), this effect varied among the patients. A total of 11 of 22 patients (50%) showed an orthotic effect (OE+), with plantarflexion (<0°) prevented during the mid and/or terminal swing, while the other 11 patients showed no orthotic effect (OE–), with no correction of the foot drop (see Supplementary Table S1).

Six (85%) of the seven patients who dropped out after the initial gait analysis were OE–. At the final gait analysis, 12 (80%) of 15 patients demonstrated an orthotic effect, and 3 (20%) failed to achieve OE, with no difference in compliance ($p = 0.6$).

3.4.2. Predictors of OE+ and OE− at First Gait Analysis

Supplementary Table S2 shows different baseline variables for participants who did and did not gain an orthotic effect at the first gait analysis. There were no significant differences between the 11 patients who showed an orthotic effect and the 11 who did not, in terms of demographic, device use, clinical, kinematic, or biomechanical variables, including passive ankle ROM (knee extension/flexion). The only significant difference found between the subgroups was ankle dorsiflexion at mid-stance (MS-DF), which was significantly correlated with the presence of OE at gait analysis (r = 0.56, $p < 0.01$). Specifically, children in the OE+ group had greater ankle dorsiflexion at mid-stance compared to the OE– group (median 20.37° (15.87, 22.05) vs. 11.75° (10.1, 16.66), respectively, $p < 0.01$).

3.4.3. Kinematic Parameters

No statistical change was noted in the study group with respect to the first and final ankle kinematic parameters (off vs. off and on vs. on) (Table 3).

3.4.4. Spatiotemporal Parameters

Comparing the spatiotemporal parameters of the full study group (N = 15) between the first and final gait analysis, there was no evidence for the normalization of gait deviations, with no significant change in the proportions of the gait cycle comprising the stance phase nor a decrease in the double/single support ratio. In contrary to our primary hypothesis, there were indications of slower walking, including an increase in step time (0.49 s (0.43, 0.54) vs. 0.53 s (0.5, 0.59) for the first and final gait analysis, respectively, with DF-FES off, $p < 0.05$), and a trend toward a decrease in cadence (steps/min; $p = 0.06$) with no change in step length. Still, in direct measures of walking speed, the decrease was not statistically significant (Table 3).

The indications of slower walking grew stronger when comparing the spatiotemporal parameters between the first and final gait analysis only for the twelve patients who gained OE (at the final analysis). We found a significant increase in the stance time (sec), a decrease in cadence (steps/min), and an increase in the double/single support ratio (supported gait). Specifically, the stance times were 0.52 (0.46, 0.56) vs. 0.56 (0.53, 0.64) sec; the cadence was 128.4 (123.2, 146.1) vs. 120 (108.6, 127.4) steps/min; and the double/single support time ratio was 0.29 (0.25, 0.31) vs. 0.32 (0.29, 0.34), all for the first and final gait analyses, respectively (DF-FES off; $p < 0.05$).

3.4.5. Biomechanical Parameters

Neither the ankle MAS score, ankle passive ROM, nor muscle strength of the study group changed statistically between the baseline and final assessment. However, some patients did show an improvement in these parameters. For example, seven (46%) and nine (60%) improved their ankle ROM with the knee flexed and knee extended, respectively (see Supplementary Table S3).

4. Discussion

This prospective open-label study assessed the effects of five months of DF-FES use on aspects of daily motor functioning, such as stability and postural control, which are impaired in high-functioning (GMFCS I/II) children with hemiplegic CP. The findings demonstrated a significant change over time in the Community Balance and Mobility Scale, with a median difference of 6.5 points (95% CI (2.79, 10), $p < 0.01$) after five months of device use, where a change of five points is considered clinically meaningful [16]. The CB&M was developed to evaluate the balance and mobility of patients who may be ambulatory yet still have balance and mobility deficits. Wright and Bos [28] showed that even children with

typical development normally do not reach the maximal score, so this test may be used in high-functioning children with hemiplegia who reach a ceiling effect on other objective measures [18]. Similar to our results, Pool et al. [16] showed an improvement in this scale of 8.3 units (95% CI [3.2, 13.4]) compared to a control group after eight weeks of FES use in children with hemiplegic CP. It should be noted that significant improvement was noticed in the present study already after four weeks of device use. After the first month, improvement continued, although the difference between month one and month five was not significant. In addition, falling frequency questionnaires filled out by parents revealed a trend toward improvement in stability, although those results were not statistically significant ($p = 0.06$).

Pool et al. [16,18] found improvement in ankle biomechanical parameters, including spasticity, range of motion, and muscle strength, under continuous DF-FES use. These biomechanical improvements may account for better postural stability. In addition, Pool [18] hypothesized that the repetitive motion of the ankle leads to improved reciprocal inhibition, reduced muscle co-activation, and better coordinated muscle activation. Our study found no statistically significant changes in either ankle biomechanical parameters or kinematic parameters over time. Still, the absence of statistical change may have other causes, such as the limited sensitivity of methods such as manual muscle testing for detecting minor changes in muscle power [35] or reduced power due to our small study sample. Taking a close look at the particular biomechanical parameters of the patients at baseline and final assessments (Supplementary Table S3), we can see that many patients did show improvement in some of their biomechanical parameters. Larger studies are needed to define more precisely which patients may or may not improve their biomechanical parameters.

As noted in the introduction, studies examining the therapeutic effects of DF-FES (namely, any improvement after the continuous use of DF-FES that continues without the device) have been inconclusive [13]. Bailes [17] found evidence for a therapeutic effect in some but not all parameters of the SWOC (Standardized Walking Obstacle Course test) after four months of DF-FES use. Our findings, like those of Pool [18], showed improvement over time in the CB&M test with DF-FES turned off, implying at least an immediate therapeutic effect. Khamis [13] argued that the retention effect is probably temporary and dependent on the continuous use of FES, but Pool [18] showed that the effect lasts for at least six weeks post-treatment. More studies are needed to evaluate the persistence of this carry-over effect.

We used the TUDS test as another method to assess postural control in the population of children with CP. This test requires a certain strength in the lower extremities and trunk, ROM in the lower extremities, coordination during fast reciprocal movements, and postural control. The present findings show only a trend toward minor improvement in the TUDS. A lack of a significant change in biomechanical parameters such as ankle ROM may explain the lack of significant improvement in the TUDS test.

Our findings with respect to the 6MWT call for close scrutiny. Bailes [19] reported a mean increase of 52 m in the 6MWT after four months on DF-FES, which may imply increased physical endurance. Contrary to our primary hypothesis, in our study, the 6MWT revealed the opposite trend, with a statistically decreased walking distance of 30 m (DF-FES off) over time. This finding seems to reflect a change toward slower walking speeds. Previous work suggests a minimum detectable change of 61.9 m in the 6MWT for GMFCS Level I, [33], which is not met by our study. However, the changes in spatiotemporal parameters point to a real trend. While there was no improvement in spatiotemporal deviations towards the norm, there were several indications of slower walking, especially in the subgroup that gained an orthotic effect. In this subgroup, there was a decrease in cadence (steps/min) with no change in step length, an increase in step time, an increase in stance time, and an increase in the double/single support ratio. Taken together, the data point to slower walking, even though in a direct measure of walking speed across a 14 m walkway, this trend was not statistically significant. Walking for a longer distance, as in the 6MWT, may be needed to notice a significant change.

Indeed, the literature is characterized by conflicting results regarding the effect of FES on walking speed in children with CP, with some results showing an increase [17,23], others a decrease [25], and others no change [24]. The implications and causes of changes in walking speed, and, in particular, reduced walking speed, are also unclear. One possibility is that slower walking may reflect a negative effect of DF-FES. Bailes et al. [19] reported a deterioration over time in kinematic parameters (e.g., a decrease in peak swing dorsiflexion) during DF-FES use. They hypothesized that this deterioration may derive from a lack of voluntary muscle effort and weakness among patients who rely on the device [10,19]. Another explanation for slower walking could be a natural decline in functional capacity over time in the CP population. However, our study did not show a decrease in kinematic parameters over time or muscle weakness. In addition, the reduction in walking distance was noted very early, already after one month, which implies that the change should be attributed to the device rather than natural decline.

On the other hand, slower walking also has some advantages. Van der Linden et al. [25] suggested that since many children with CP have difficulty controlling their forward progression, a decrease in speed may reflect a more controlled gait pattern. Slower walking causes an increase in the double support phase, both in time and as a percentage of the gait cycle. During double support, stability is increased since patients have more control over their center of mass movement [39]. Children with hemiplegia have a higher baseline double/single support ratio than typically developing children [6]. It seems that DF-FES does not repair this deviation, but, by causing slower walking, it increases stability.

Patients did not experience the device as annoying or uncomfortable, so a slower gait is probably not attributable to adverse effects of the electrical stimulation. In addition, the decrease in distance appeared even when DF-FES was turned off. Damiano [10] showed improvement in max swing DF over time only at self-selected speeds, but not at the patient's fastest walking speed. It may be that inherent features of the device limit a full dorsiflexion effect during fast walking, leading patients to walk slower in order to obtain the full effect of the device. This may reflect a limitation of the device, as it lacks a closed loop control system that would allow for adaptation to different walking speeds [40]. On the other hand, it may encourage patients to adopt a slower and more controlled gait pattern, even in the DF-FES off state. Larger and directed studies are needed to test the orthotic effect at different walking speeds.

There are currently no clinical tools able to identify appropriate candidates for DF-FES in children with hemiplegic CP. In this study, only 58% of eligible patients completed the study protocol. Patients who dropped out had a significantly higher prevalence of previous botulinum toxin injections and a non-significant trend toward a higher prevalence of AFO use. These findings may imply a worse baseline condition in those patients. It is noteworthy that the high drop-out rate was related mostly to the inefficacy of the device in producing an orthotic effect. Six (85%) of seven patients who dropped out after the first gait analysis did not achieve swing ankle dorsiflexion $\geq 0^\circ$ at either the mid or terminal swing. Our findings emphasize that a precondition for gaining a functional benefit from the continuous use of DF-FES is an achievement of an orthotic effect; otherwise, compliance will be poor. Khamis [13] noted that while no absolute criteria were found to predict the suitability of FES devices, a prerequisite is the ability to maintain adequate passive dorsiflexion; however, they left open the precise meaning of "adequate." Previous studies have used different patient inclusion criteria [5]—e.g., passive ankle ROM with knee extension above 0° [19] or 5° [18].

With the aim of identifying predictors for the achievement of an orthotic effect, we compared the demographic, device use, and clinical parameters of those who did (OE+) or did not (OE–) achieve such an effect at the first gait analysis. We hypothesized that those with a better ankle ROM would gain an orthotic effect (OE+). Although passive ankle ROM angles were not significant predictors for the presence or absence of OE, we found that a larger dorsiflexion angle during the mid-stance is a significant predictor for OE+. We looked specifically at this parameter as another indication for ankle ROM during the

weight-bearing state [41]. This parameter may reflect the functional ankle ROM better than a passive ankle ROM test. Thus, adequate ankle ROM, as a prerequisite for using DF-FES, should not be measured passively but rather during gait or through other weight-bearing measures [42,43]. Among the 15 patients who completed the study, 3 failed to achieve OE, although no difference in compliance was noted. Two of those who did not gain OE had a limited mid-stance angle. Larger studies are needed to define other predictors for orthotic effect achievement and appropriate candidates for this intervention.

This study has limitations. Some of our borderline statistical results (e.g., falling frequency) may reflect limited power due to the small size of the study cohort. Methodologically, this was an open-label study with no control group. This limitation may raise the possibility that the improvement observed in balance scores relates to the effects of time and practice and not to the DF-FES intervention. However, the early change in balance scores, together with walking distance, implies the true influence of the device.

The timing of the first gait analysis is another potential limitation, since it was conducted after one month of device use and not at the true baseline. This decision was made a priori (at study protocol), based on previous work [12,19,24] suggesting that a period of adjustment to the device is necessary in order to assess the true orthotic effect at gait analysis. Due to budget limitations we chose to conduct two gait analyses, after one month and after five months of use, on the assumption—supported by previous studies [10,19]—that baseline kinematic and spatiotemporal parameters would not change significantly during the first month of intervention. If this assumption is incorrect, significant changes in kinematic parameters could have been missed.

It should also be noted that this study focused on changes in kinematic parameters on the affected side. Changes on the unaffected side should be tested further.

5. Conclusions

The continuous use of DF–FES produces an early functional benefit and immediate therapeutic effect with better stability and postural control. A precondition for gaining a functional benefit from the continuous use of DF-FES is the achievement of an orthotic effect; otherwise, compliance will be poor. Limited mid-stance dorsiflexion can serve as a predictor for a failure to achieve an orthotic effect. Results regarding the walking speed are still inconclusive; however, we found indications that, over time, walking becomes slower, especially in those who gain an orthotic effect. While this may imply a negative outcome, slower walking is more supported and controlled, which may contribute to improved stability.

Supplementary Materials: The following supporting information can be downloaded at https://www.mdpi.com/article/10.3390/children10030531/s1, Table S1: Kinematic parameters of patients showing (OE+) and not showing OE (OE–) at first gait analysis; Table S2: Clinical and demographic parameters of patients showing (OE+) and not showing OE (OE–) at first gait analysis; Table S3: Biomechanical parameters at baseline and final assessments of the study group.

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The Effect of Traction before Closed Reduction in Patients with Developmental Dysplasia of the Hip

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Abstract: Developmental dysplasia of the hip (DDH) with a dislocated hip can be treated with traction before closed reduction (CR). Currently, there is insufficient evidence supporting the use of preoperative traction treatment for a successful CR. The objective of this study was to determine the effect of preoperative traction on the success rate of primary CR in DDH patients with dislocated hips. A retrospective pair-matched study was performed in DDH patients with dislocated hips. Patients with preoperative traction treatment prior to primary CR were matched (based on age and the severity of DDH on the radiograph) to patients without preoperative traction treatment. The primary outcome was the presence or absence of maintained reduction after three weeks. A match was found for 37 hips, which resulted in the inclusion of 74 hips. No significant difference was found in the number of successful reductions after three weeks between the traction group and the control group (31 vs. 33 hips, $p = 0.496$). Traction treatment did not significantly improve the short-term or mid-term outcomes for closed reduction. Based on these results, we suggest that traction treatment should not be used as standard care for dislocated hips in DDH.

Keywords: traction; developmental dysplasia of the hip; closed reduction; avascular necrosis

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1. Introduction

Developmental dysplasia of the hip (DDH) is the most common musculoskeletal disorder in infants and young children [1]. DDH includes a wide spectrum of developmental disorders of the hip, varying from stable dysplastic hips to unstable or dislocated hips [2].

Currently, hip dislocation due to DDH is first treated with a Pavlik harness [3]. If the Pavlik harness fails, the next step is closed reduction (CR) and the application of a spica cast under general anesthesia [4]. CR is considered successful when the femoral head is correctly positioned in the acetabulum and remains reducedduring follow-up. If dislocation persists or redislocation occurs, an open reduction can be performed. Open reduction has more complications than CR, and should preferably be avoided [5].

As with all medical interventions, CR has a risk of complications, e.g., redislocation (8–40%) and avascular necrosis (AVN) of the femoral head (10%) [6–8].

When the hip has a limited range of motion, or when the femoral head has migrated proximally, traction treatment prior to CR can be used to improve the success rate and to reduce the incidence of AVN [6,8–11]. During this treatment, the hips are gradually reduced via traction and abduction. The range of motion of the hip is improved as the muscles and ligaments are stretched due to traction. There is a wide variation in traction methods and duration [12].

Whether or not traction treatment improves the success rate of CR and reduces the incidence of AVN, it remains controversial. Previous studies have shown no clinically relevant difference in AVN or success rate of CR [7,12–14]. Currently, there is no consensus in the literature for whether traction treatment should be used as part of standard care.

The primary objective of this study was to determine the effect of traction treatment on the success rate of a primary CR, defined as maintained reduction, in DDH patients

with dislocated hips. Secondary, the effect of traction on (1) long-term redislocation, (2) the number of adductor tendon tenotomies at primary CR, (3) the development of AVN, (4) residual dysplasia, and (5) the improvement of acetabular development is evaluated.

2. Materials and Methods

2.1. Study Design

A retrospective pair-matched study was performed in DDH patients with dislocated hips, treated with CR from 2010 to 2018 at the Department of Pediatric Orthopaedics at our hospital. The Medical Ethics Committee of our institution provided a waiver of approval for this study (MEC-2018-1525).

The inclusion criteria were (1) DDH with 1 or 2 dislocated hips, (2) primary CR with or without preoperative traction treatment, (3) spica cast for three months (range, 10–14 weeks) and (4) a follow-up of minimally three weeks. Patients were excluded in cases of (1) teratologic dislocation, (2) neuromuscular disease, (3) previous CR, (4) incomplete data, (5) incomplete traction treatment, or (6) a combination of CR with pelvic or femoral surgery.

Two patient groups were identified: (1) traction treatment prior to CR and (2) direct CR (control group). Whether or not traction treatment was started was the physician's choice, based on clinical and radiographic findings, such as the Ortolani test, limited abduction, and radiographic presence of a neoacetabulum.

Traction treatment consisted of two weeks of traction in a clinical setting. Vertical traction (90° hip flexion) was used for patients under six months of age, and horizontal traction (hip extension), for patients older than six months. Three age groups were defined, based on the type of traction (horizontal or vertical) and hip development: 0–6 months, 6–9 months, and older than 9 months. The initial anteroposterior (AP) pelvic radiograph was evaluated in each patient for the severity of the dislocation. This was categorized based on the presence of a neoacetabulum. Patients were matched by age group and severity of the dislocation. Bilateral dislocated hips were separately matched for both sides. The investigator who matched the cases was blinded to the outcome.

2.2. Outcome Measures

The primary outcome was a successful CR, which is defined as a maintained reduction at three weeks after the CR procedure. The position of the hip at three weeks was evaluated via transinguinal ultrasound [15]. A subgroup analysis was performed on the primary outcome in the three age groups.

The secondary outcome measures were (1) adductor tendon tenotomy during primary CR, (2) redislocation at six months after CR, (3) the presence of AVN, and (4) residual dysplasia at one year (a range of 9–18 months) and two years (a range of 21–30 months) of follow-up (5), and an improvement in acetabular development at one year and two years of follow-up. No subgroup analysis was performed on the secondary outcome measures.

AVN was defined using the Salter criteria and classified as a dichotomous outcome [16]. Residual dysplasia was defined as an acetabular index (AI) of 25 degrees or higher. Improvement of the acetabular development (the progression of AI) was calculated as the difference between the AI at baseline and the AI at the given times at follow-up. This measure can indicate the speed of improvement of the acetabulum.

2.3. Data Extraction

Baseline characteristics and outcome data were extracted from the medical charts, radiographic images, and surgery reports, using the hospital information system (PDMS, Picis Clinical Solutions, Wakefield, USA; Hix, ChipSoft B.V., Amsterdam, The Netherlands). When in doubt, a second opinion from a pediatric orthopedic surgeon was requested.

2.4. Statistical Analysis

Categorical variables are presented as numbers and percentages. Continuous variables were tested using the Shapiro–Wilk test for normality. If data showed a normal distribution, these were presented as mean and standard deviation (SD). Otherwise, data were presented as median and interquartile range (IQR). After the data were matched, the number of successful CR procedures and the number of adductor tendon tenotomies were analyzed using McNemar's test. Other secondary outcomes were tested using the chi-squared test or Fisher's exact test if the data were binary. Continuous variables were analyzed with either an unpaired *t*-test or the Mann–Whitney *U* test. Significance was set at a p value of <0.05. IBM SPSS Statistics 24 (IBM Corp., Armonk, NY, USA) was used for statistical analysis.

3. Results

3.1. Patient Inclusion

From July 2010 to October 2018, a total of 335 patients were treated with CR. One hundred and three patients met the inclusion and exclusion criteria, of which 37 had preoperative traction (Figure 1). For these 37 hips, 37 matching control hips were found, which resulted in a total of 74 hips.

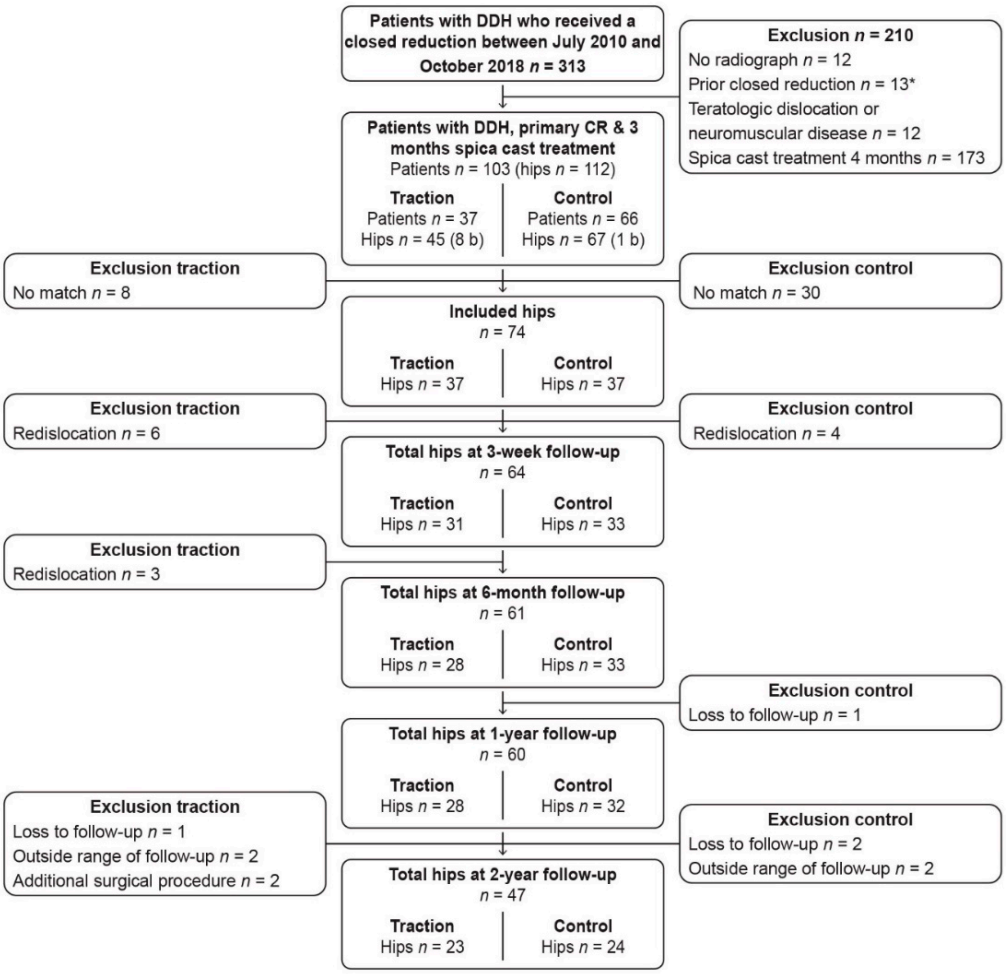

Figure 1. Flowchart of patient inclusion and follow-up. DDH = developmental dysplasia of the hip; CR = closed reduction; *n* = number; b = bilateral. * Patients with a closed reduction in the medical history were excluded.

3.2. Baseline Characteristics

Of the included patients, 89% were female and had a median (IQR) age of 31.8 (22.2–37.3) weeks at CR (Table 1). The presence of a neoacetabulum was seen on 56 AP

pelvic radiographs in both groups. Additional baseline characteristics can be found in Table 1.

Table 1. Baseline characteristics.

		Traction Group H37/P35	Control Group H37/P37	*p* Value
Age at intervention (weeks)		32.4 (23–36.7)	31.7 (20.9–37.3)	0.474
Sex	Female	32 (86.5)	34 (91.9)	0.687
Family history of DDH	+	13 (39.4) **	12 (32.4)	0.504
Breech position	+	7 (21.9) ^	12 (35.3) ^	0.287
Side	Left	23 (65.7)	28 (75.7)	0.667
	Right	5 (14.3)	8 (21.6)	
	Bilateral	7 (20)	1 (2.7)	0.031
AIF difference (degrees)		35 (15–45) *	20 (10–31.3) #	0.026
Ortolani	+	6 (17.6) ^	20 (64.5) #	<0.001
AI baseline (degrees)		39.1 (±4.4)	37.4 (±4.4)	0.148
Pavlik		25 (67.6)	21 (56.8)	0.454

DDH = developmental dysplasia of the hip; AIF = abduction in flexion; AI = acetabular index; H = hips; P = patients; Age and AIF differences presented as median (interquartile range); AI baseline presented as mean (±standard deviation); Categorical values are presented as number (%); AIF difference is observed in unilateral dislocated hips; Two bilateral dislocated hips in the traction group were matched for both sides, the remaining bilateral patients were matched for one side; Missing data * $n = 1$; ** $n = 2$; ^ $n = 3$; # $n = 6$.

3.3. Primary and Secondary Outcomes

A total of 64 (86.5%) hips were successfully reduced after three weeks, with 10 (13.5%) redislocations occurring (Table 2). There was no significant difference in the success rates between the traction group and the control group (84% and 89%, $p = 0.496$) at three weeks follow-up.

Table 2. Primary and secondary outcomes of redislocation.

		Traction ($n = 37$)	Control ($n = 37$)	*p* Value
Successful CR at 3 weeks		31 (83.8)	33 (89.2)	0.496
Redislocations at 3 weeks	Age group			
	0–6 m: $n = 26$	3 (8.1)	1 (2.7)	-
	6–9 m: $n = 34$	2 (5.4)	2 (5.4)	-
	9–21 m: $n = 14$	1 (2.7)	1 (2.7)	-
Redislocations at 6 months		9 (24)	4 (10.8)	0.127

CR = closed reduction; n = number; m = months; Data is presented as number (%).

In the age group 0–6 months, three (8%) redislocations were observed in the traction group, and one (3%) in the control group. For the age group 6–9 months, these numbers were two (5%) in the traction group and two (5%) in the control group. In the age group 9–21 months (the oldest match was 21 months), in both groups one (3%), redislocation was observed. Because of the small sample sizes, no statistical significance could be calculated for the subgroups.

The number of redislocations within 6 months after CR did not differ significantly between the two groups ($p = 0.127$) (Table 2).

The other secondary outcomes (number of adductor tendon tenotomies, AVN, residual dysplasia, and acetabular improvement) showed no significant differences between the traction group and the control group (Table 3).

Table 3. Secondary outcomes for adductor tendon tenotomy, AVN, and residual dysplasia.

	Total Hips	Traction	Control	*p* Value
Adductor tendon tenotomy	H74	13 (35.1)	11 (29.7)	0.824
AVN at 1 year	H60	6 (21.4)	7 (21.9)	1.000
AVN at 2 years	H49	4 (16.7)	3 (12)	0.702
Residual dysplasia (AI > 25°) at 1 year	H58	22 (78.6)	19 (63.3)	0.25
Residual dysplasia (AI > 25°) at 2 years	H49	16 (66.7)	13 (52)	0.296
Progression AI at 1 year	H58	10.9 (±5.1)	9.9 (±4.8)	0.432
Progression AI at 2 years	H49	13.5 (±7.4)	12.9 (±4.2)	0.771

AVN = avascular necrosis; AI = acetabular index; H = hips. At one year, 60 hips (28 traction, 32 control), and at two years, 49 hips (24 traction; 25 control) were in the follow-up. AI in degrees presented as mean (±SD); Categorical variables are presented as numbers (%).

At one year, 60 hips, and at two years, 49 hips, were in the follow-up. The total percentage of AVN was 22% at one year and 14% at two years of follow-up, no significant difference between the traction and control groups was found (Table 3).

4. Discussion

4.1. Redislocations

In this study, the additional value of preoperative traction treatment for stable closed reduction in DDH was evaluated. The main objective was to determine whether traction treatment improves the success rate of CR. No significant difference in maintained reduction was found between the traction and the control group at three weeks and at six months of follow-up.

In this study, we chose dislocation at three weeks after CR as the primary outcome, because most redislocations occur within the first three weeks after CR, based on our clinical experience. The literature suggests that the majority of redislocations can be expected within two weeks after CR [17]. We hypothesized that the effect of traction treatment does not last longer than three weeks. Stretched ligaments and tendons will adapt to their preferred lengths rapidly, but this is most likely within three weeks after the discontinuation of traction.

At six months follow-up, more dislocations were reported in the traction group when compared to the control group (not significant, $p = 0.127$). We have no clear explanation for this finding, but this might be due to baseline differences that could not be identified in this retrospective study.

4.2. Adductor Tendon Tenotomies, AVN, and Residual Dysplasia

We expected to find a decrease in adductor tendon tenotomies in the traction group, due to the gradual stretching of the tendons and muscles, including adductors. However, no significant difference was found between the two groups. This could be caused by hip rigidity in the traction group, requiring both traction and tenotomy. The effect of traction treatment could be limited, necessitating additional tenotomy.

One of the main reasons for commencing preoperative traction treatment is to reduce the risk of AVN. In our study, no difference between the groups was seen in AVN incidence at the one-year and two-year follow-ups. Previous research has shown that AVN at these young ages may not deteriorate any further, and can stay clinically insignificant [18]. To determine more clearly what type of AVN our patients will develop, and what the clinical importance will be, an evaluation at a later age will be needed [19]. The outcome of this study is similar to the results of two meta-analyses, in which no significant difference in AVN rate was found when preliminary traction treatment was applied [12,14].

No differences were reported in residual dysplasia and the improvement in AI between the traction and control groups; therefore, we can conclude that the AI improves at a similar pace.

4.3. Limitations

The main limitation of our study is selection bias, as the decision to give traction treatment was made by the primary physician. This decision was based on clinical findings (e.g., the range of motion and Ortolani) and the radiograph. There was a significant difference in range of motion and the Ortolani test at baseline between the two groups. This implies that hip rigidity was linked to traction treatment, causing these differences between the groups. These parameters could not be included in the matching procedure of the hips, due to a relatively high rate of missing data.

Secondly, we chose to classify and match the hips in the study population based on the presence of a neoacetabulum on pelvic radiographs. Although more measurements based on the radiograph and clinical assessment could be included in the matching method, these measurements provided insufficient information on the initial state of the hip. We believe that the presence or absence of a neoacetabulum provides most information on the duration of dislocation, stiffness, and chances of successful CR.

The effect of traction treatment on the number of open reductions was not investigated in this study. A recent study concluded that traction treatment does not reduce the cases of open reduction [20]. Future prospective randomized controlled trials (RCT) should include both closed and open reduction.

4.4. Interpretation of Findings

Currently, in most studies, successful CR is defined as a femoral head that is reduced in the acetabulum during the procedure and maintained in this position during follow-up [7,12,21]. We strongly believe that the effect of preoperative traction treatment is only present in the first days to weeks after the treatment; this is confirmed by our results. Therefore, the effect of traction treatment on the reduction can be determined in an early stage following CR. Inadequate acetabular remodeling can lead to instability and redislocations, but this occurs later during follow-up and is not affected by traction treatment.

The relation between traction treatment and the risk of AVN is difficult to investigate, as AVN is a multifactorial problem that can be provoked at all steps of treatment (e.g., Pavlik, traction, CR, and spica cast). Additionally, AVN can present multiple years after an intervention, making causality hard or impossible to prove, even with an RCT. As traction treatment is a challenging process for both child and parent, and the value is questionable, we do not advise traction treatment for this purpose anymore.

5. Conclusions

In this study, we did not identify any short-term or long-term benefits of traction treatment.

Previous comparative studies showed no benefit for traction treatment in achieving a higher success rate of CR, which is in line with our findings [7,12]. There are no studies comparing traction treatment to a control group that are in favor of traction treatment.

Based on these results, we suggest that traction treatment should not be used as standard care for dislocated hips in DDH. These results should be confirmed in prospective RCTs.

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Informed Consent Statement: Patient consent was waived as the Research involving Human Subjects Act (WMO) did not apply.

Data Availability Statement: The data presented in this study are available on request from the corresponding author.

Conflicts of Interest: The authors declare that there are no conflicts of interest. No funding was received for this study.

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children

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Article

The Importance of Monitoring and Factors That May Influence Leg Length Difference in Developmental Dysplasia of the Hip

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Abstract: In unilateral Developmental Dysplasia of the Hip (DDH), avascular necrosis (AVN), femoral or pelvic osteotomy, and residual dysplasia causing subluxation of the proximal femur may influence Leg Length Discrepancy (LLD). This can lead to gait compensation, pelvic obliquity, and spinal curvature. The aim of this study is to determine the prevalence of LLD, establish which limb segment contributes to the discrepancy, describe how AVN influences LLD, and ascertain variables that may influence the need for LLD corrective procedures. Methodology: This study assessed long-leg radiographs at skeletal maturity. Radiographs were assessed for the articulo-trochanteric distance (ATD) and femoral and tibial length. AVN was classified according to Kalamchi–MacEwen. Results: 109 patients were included. The affected/DDH leg was longer in 72/109 (66%) patients. The length difference was mainly in the subtrochanteric segment of the femur. AVN negatively influenced leg length. Older (≥three years) patients with multiple procedures were more likely to have AVN. LLD interventions were performed in 30 (27.5%) patients. AVN grade or type of DDH surgery did not influence the odds of needing a procedure to correct LLD. Conclusions: Procedures to correct LLD were performed irrespective of previous DDH surgery or AVN grades. In most patients, the affected/DDH leg was longer, mainly in the subtrochanteric segment of the femur, largely influenced by femoral osteotomy in patients with multiple operative procedures for DDH. We recommend careful monitoring of LLD in DDH.

Keywords: developmental dysplasia of the hip; limb length discrepancy; avascular necrosis

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1. Introduction

Management of developmental dysplasia of the hip (DDH) aims at establishing a stable, concentrically reduced hip that allows for normal remodelling of the acetabulum and femoral head [1,2]. This can be achieved by combining operative and non-operative modalities depending on age and severity of presentation [3–6]. While the focus of the clinician is primarily on the hip, leg length discrepancies can arise and should be appropriately monitored and treated where necessary [7,8].

Leg length discrepancy (LLD) has been found to be more common in the presence of DDH than in controls [9]. In unilateral DDH, an LLD may arise due to avascular necrosis (AVN), osteotomies of the femur, pelvis, or subluxation of the proximal femur caused by residual dysplasia [10]. The ipsilateral leg is usually longer and often attributed to femoral overgrowth after osteotomy or excessive growth driven by dysplasia (in the absence of a femoral osteotomy) [7–9]. History of a femoral osteotomy has been identified as an independent risk factor for ipsilateral limb overgrowth, despite the initial loss of length after a varus osteotomy was performed [7,11].

LLD in patients with unilateral DDH can be problematic for both patients and surgeons. It results in a pelvic tilt leading to gait asymmetry, spinal scoliosis, flexion

contracture knee (in the longer leg), or equinus contracture at the ankle (in the shorter leg) [12–17]. Surgical considerations include the timing of the guided growth interventions before skeletal maturity, the need for limb elongation procedures after skeletal maturity when the difference is large, and appropriate planning to ensure equal length after hip arthroplasty [8,13,18–21].

Leg length differences have previously been studied in patients with DDH. Yoon et al. [7] estimated length differences in 101 children with DDH, estimating the difference on standing ap pelvis radiographs. They reported limb overgrowth as common and mainly related to femoral osteotomy. Zhang et al. [20] studied long-leg radiographs of 67 skeletally mature patients with unilateral developmental hip dislocations and found that tibial length, lesser trochanter to tibial plafond length, and overall leg lengths on the affected leg were significantly longer, regardless of high or low dislocations. LLD is important to quantify since Tolk et al. [22] demonstrated a trend towards impaired acetabular development in unoperated DDH patients with greater limb length discrepancy. Other studies have shown an increased risk of total hip replacement on the longer side [23].

The aim of this study is to identify the prevalence of LLD, quantify which segment of the limb contributes to the discrepancy, and describe the influence of AVN on LLD in patients with unilateral DDH. This study also tries to ascertain variables that may influence the need for LLD interventions. This would forewarn clinicians and management and provide a better follow-up guide.

2. Materials and Methods

This study reports on a consecutive retrospective case series of all patients treated for DDH between January 2008 and December 2020 in our institution. Inclusion criteria were patients with unilateral DDH referred to our unit for managing primary or residual DDH prior to skeletal maturity with the availability of adequate long-leg radiographs. Exclusion criteria were patients with: (1) associated pathology affecting leg length (e.g., Neuromuscular conditions, congenital abnormalities, or skeletal dysplasia), (2) open triradiate cartilage at the time of review, and (3) bilateral cases. Patients with successful Pavlik harness treatment were not followed up after age of 5 unless they required further intervention, as per our published protocol and, therefore, were excluded from our cohort [23]. No ethical approval was required as this study was classed as an audit of historically treated patients.

2.1. Data Collection

Electronic patient files were reviewed for patient and treatment characteristics. Factors recorded were age at diagnosis, age at a final follow-up, side effects, and treatment. Along the course of treatment, some patients required multiple procedures. To reduce this ambiguity, patients were categorised into groups based on the last successful procedure. Groups included patients with the following results:

(1) Successful closed reduction;
(2) Failed (or successful) closed reduction proceeding to an open reduction without bone surgery;
(3) Reduction proceeding to a femoral osteotomy;
(4) Reduction proceeding to a pelvic osteotomy;
(5) Reduction proceeding to a femoral and pelvic osteotomy.

2.2. Radiographic Measurements

Our follow-up protocol recommends a clinical and radiological review, including leg-length assessments at prescribed time points [24].

Radiographic measurements were performed on the calibrated, standardised long-leg standing radiographs. Adequate radiographs were defined as patellae positioned forward; bony landmarks were visible with the presence of a templating ball or scale measure. All measurements were performed using TraumaCad (Brainlab, Petach-Tikva, Israel) software (see Figure 1) [25]. Measurements included the articulo-trochanteric distance (ATD),

femoral length, and tibial length. Measurements were performed using measurement tools in the software. ATD was measured by placing markers on the tip of the greater trochanter and superior aspect of the femoral head on a line oriented along the axis of the femur (Figure 1a). Femoral and tibial lengths were calculated by the software after the appropriate identification of bony landmarks by the investigator (Figure 1b,e). Proximal femoral growth disturbance was classified according to Kalamchi–MacEwen [26] for a grade of avascular necrosis. All three authors independently reviewed the measurements, and discrepancies were resolved with consensus [27].

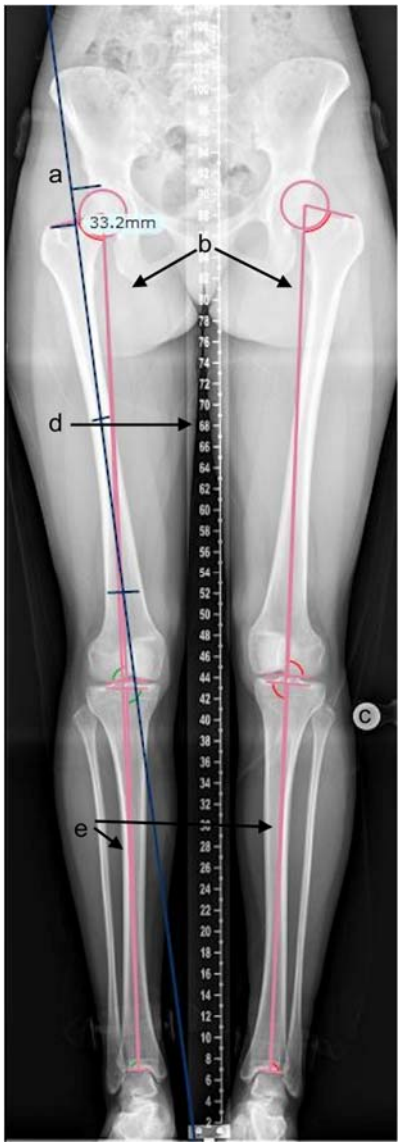

Figure 1. TraumaCad measurement of leg length parameters. (a) tool for articulo-trochanteric distance, (b) femoral length tool, (c) templating ball for calibration, (d) measuring ruler, (e) tibial length tool.

2.3. Statistical Analysis

For descriptive statistics of continuous variables, means were reported, and for discrete variables, counts and percentages were presented. Mean leg length differences between the longer leg vs. the shorter leg and DDH affected vs. unaffected legs were compared using *t*-test samples for each of the leg segments measured (total leg length, tibial length, total femoral length, ATD, and subtrochanteric femoral length) and reported as mean differences with 95% confidence intervals.

An analysis was made to assess whether having multiple procedures or procedures at a younger age ($\leq$3 years) influenced the final radiological outcome according to AVN graded by Kalamchi–MacEwen [26]. The statistical significance was calculated using chi-square and Mann–Whitney U test. An association between the various treatment modalities and the need for LLD intervention was also analysed, and the results were presented as odd ratios with 95% confidence intervals. A similar analysis was performed to assess the relationship between AVN grade and the need for LLD intervention.

All data were tabulated, and SPSS (IBM Corp. Released 2020. IBM SPSS Statistics for Macintosh, Version 27.0.) was used for statistical analysis.

3. Results

During the study period, 248 DDH patients were identified. 109 patients met all the criteria for the follow-up (Figure 2). Table 1 describes the patient characteristics and frequency of procedure types.

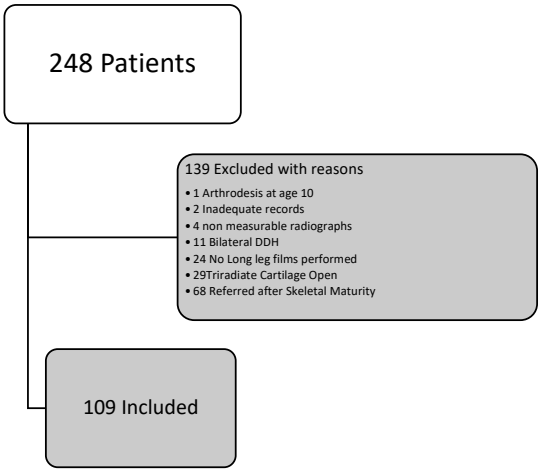

Figure 2. Patient inclusion and exclusion flow-chart.

Table 1. Patient demographics and frequency procedure types.

Patient Characteristics	***n* = 109**
Mean age at referral with range in years	2.7 (0–13)
Mean age at diagnosis with range in years	1.5 (0–10.5)
Mean age at follow-up with range in years	15.2 (10.6 to 49.9)
Side affected	Right 51/Left 58
Sex	Female 95/male 14
Surgical procedure Groups (Last surgical Procedure)	***n* = 109**
Closed reduction	17 (15.8%)
Open reduction only	17(15.8%)
Reduction + Femoral osteotomy	30 (27.5%)
Reduction + Pelvic osteotomy	15 (13.6%)
Reduction + Pelvic and femoral	30 (27.3%)
Total	109(100%)
Multiple Surgeries (bone and/or soft tissue)	25/109 (23%)
Values presented as mean with standard deviation between brackets for continuous variables and count with percentages between brackets for dichotomous variables.	

3.1. Avascular Necrosis

The AVN rate (type II, III, IV) was 52% (56/109). The most common grade of AVN encountered was Type II (Table 2).

Table 2. AVN distribution in patients who had their first operation at less than or equal to 3 years of age and greater than 3 years of age.

	Age of First Surgery		Total
	≤3 years	>3 years	
No AVN	26/95 (27.4%)	0	26 (23.9%)
AVN Type I	24/95 (25.3%)	3/14 (21.4%)	27 (24.8%)
AVN Type II	32/95 (33.7%)	8/14 (57.1%)	40 (36.7%)
AVN Type III	9/95 (9.5%)	2/14 (14.3%)	11 (10.1%)
AVN Type IV	4/95 (4.2%)	1/14 (7.1%)	5 (4.6%)
Total	95 (100.0%)	14 (100.0%)	109 (100.0%)

AVN rates were higher in patients that had multiple procedures (Figure 3), with the difference being statistically significant ($p < 0.001$ chi-square). There were fewer cases of 'poor' AVN (type III and IV) ($p = 0.029$, chi-square) in patients who had surgery at an earlier ($\leq$ three years) age (Table 2). The grade of AVN affected the ipsilateral leg length negatively, i.e., the ipsilateral leg transitioned from being longer to shorter with increasing AVN grade (Figure 4).

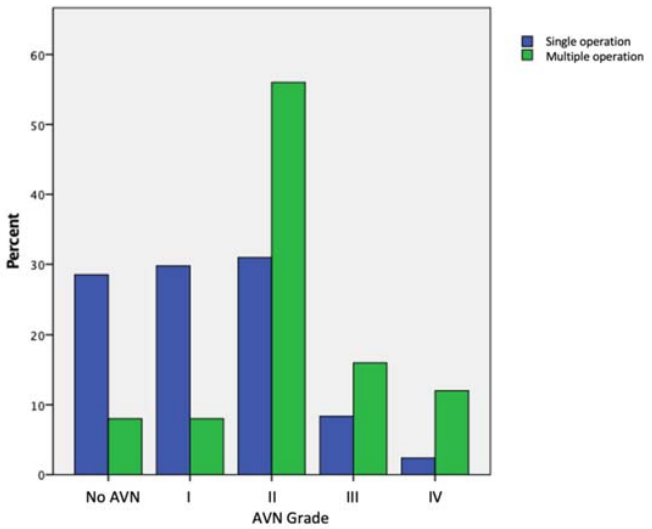

Figure 3. AVN grade frequency distribution for patients with single and multiple procedures.

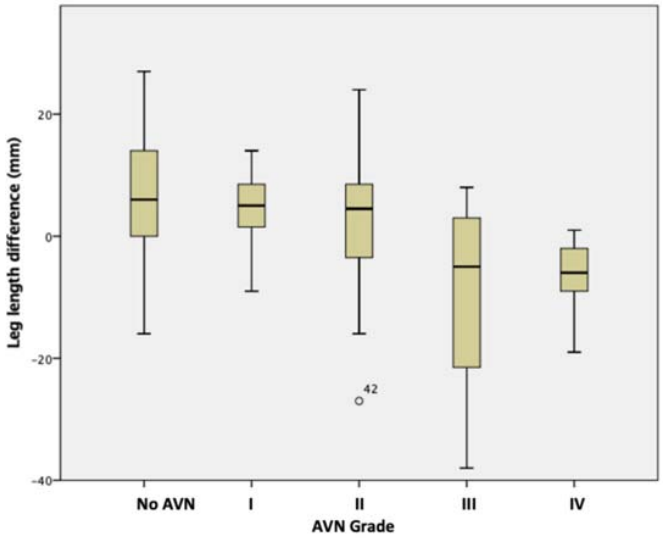

Figure 4. The average leg length difference between the DDH leg and the unaffected leg, grouped by AVN grade.

3.2. Leg Length and Segmental Differences

At skeletal maturity, the ipsilateral leg was longer than the unaffected leg in 72 (66%) patients and shorter in 34 (32%) patients. Three patients had symmetric leg lengths at the final follow-up. The difference was of no clinical significance (i.e., difference <1 cm) in 70/109 (64.2%) patients, leaving 36/109 (33%) patients with a significant difference of ≥1 cm.

When the ipsilateral leg was longer, most of the segmental difference was in the subtrochanteric length. Conversely, when it was shorter, most of the segmental difference was in the articulo-trochanteric distance, with a statistically significant difference (Table 3).

Table 3. Total leg length and segment lengths represented with median values and range.

Segmental Leg Length Difference	DDH Leg Longer (Mean Length Difference in mm, CI) (*n* = 72)		DDH Leg Shorter (Mean Length Difference in mm, CI) (*n* = 34)	
Total leg length discrepancy	8.25 (6.8, 9.6)	p < 0.001	−9.5 (−12, −6.4)	p < 0.001
Femoral length difference	7.5 (6.04, 8.8)	p < 0.001	−6.5 (−9.6, −3.2)	p < 0.001
Tibial difference in	0.5 (−0.5, 1.4)	p = 0.3	−1.9 (−4.2, 0.3)	p = 0.09
Articulo-trochanteric distance difference	0.28 (−0.9, 1.4))	p = 0.63	−5.2 (−9.2, −1.01)	p = 0.016
Diaphyseal length difference	7.1 (5.4, 8.9)	p < 0.001	−1.32 (−5.9, 3.3)	p = 0.565

3.3. Interventions for Leg Length Difference

In total, 30 (27.5%) patients underwent n LLD intervention. Of those, 9 (30%) patients had a residual leg length inequality of ≥1 cm after an LLD intervention. This latter group comprised 3 patients, each with no AVN, type II and III AVN. Surgically, the same group included 4/9 patients with revision femoral osteotomies; 2/9 had closed reduction only, and 3/9 had an open reduction, pelvic and femoral osteotomies.

Corrective procedures for LLD prior to skeletal maturity included 12 patients who underwent a permanent drill epiphysiodesis and 14 patients who underwent a temporary epiphysiodesis with medial and lateral tension band plates. One eleven-year-old patient had surgery for the hexapod external fixator frame for a 6 cm leg length difference, resulting from multiple DDH surgeries. Three patients underwent acute shortening of the longer leg after skeletal maturity. The majority (22/30) of procedures were carried out to shorten the DDH-affected limb. Of the remaining eight patients whose ipsilateral legs were lengthened, 4/8 patients had type IV AVN, 3/8 had type III AVN, and one patient had no AVN.

The odds of needing an LLD intervention for each surgical treatment group and AVN grade were calculated (Tables 4 and 5). The risk was greater for patients with a history of femoral osteotomy, combined pelvic and femoral osteotomy (not pelvic osteotomy alone), groups with multiple prior operations, and type IV AVN groups. However, the confidence interval in all groups overlapped the null value; therefore, no statistically significant conclusions could be drawn. LLD interventions were required across all surgical groups and AVN grades.

Table 4. The risk of the leg length corrective intervention stratified for the treatment group.

	Frequency (*n* [%])	OR	95% CI
Closed reduction (*n* = 17)	4/17 (23.5%)	0.78	(0.23–2.62)
Open reduction (*n* = 18)	2/18 (11.1%)	0.31	(0.07–1.42)
Femoral osteotomy (*n* = 30)	11/30 (36.7%)	1.83	(0.74–4.52)
Pelvic osteotomy (*n* = 15)	4/15 (26.7%)	0.95	(0.28–3.26)
Pelvic and femoral (*n* = 29)	9/29 (31.0%)	1.18	(0.47–2.99)
Multiple operations (*n* = 25)	9/25 (36.0%)	1.69	(0.65–4.38)

OR; odds ratio; CI; confidence interval.

Table 5. The risk of leg length corrective intervention, stratified for the AVN grade.

	Frequency (f/n [%])	OR	95% CI
No AVN/Type I (n = 53)	15/53 (28.3%)	1.08	(0.47–2.50)
Type II (n = 40)	8/40 (20.0 %)	0.53	(0.21–1.35)
Type III (n = 11)	3/11 (27.3%)	0.99	(0.24–4.00)
Type IV (n = 5)	4/5 (80%)	12.00	(1.28–112.25)

Risks stratified for avascular necrosis (AVN) grade according to Kalamchi–McEwan. OR; odds ratio; CI; confidence interval.

4. Discussion

This study shows that the leg length discrepancy is common in patients with unilateral DDH, with nearly twice the number of patients with a residual LLD $\geq$1 cm at skeletal maturity compared to the general population despite appropriate surveillance and treatment [28]. Leg length discrepancy can arise and would need treatment irrespective of the type of procedure or AVN grade. Higher AVN grades were more common in patients with multiple procedures and surgery performed after three years of age and negatively impacted the length of the ipsilateral leg. Conversely, in patients with low-grade or no AVN, the ipsilateral leg was, on average, longer than the unaffected leg.

4.1. Leg Length Discrepancy in DDH

The ipsilateral leg was longer than the unaffected leg in seventy-two (66%) patients, similar to other reports of 66-78% in the literature [8,20]. When considering an LLD of $\geq$1 cm, the prevalence in the general population is 15% [28]. This study identified 36 (33%) patients with a residual LLD of $\geq$1 cm after surveillance and treatment. This value is lower than that reported by Yoon et al. [7], who identified 44% of their 105 patients with a $\geq$1 cm LLD. Therefore, appropriate surveillance and timely interventions for LLD are recommended in DDH patients.

Unsurprisingly, this study identified the femur to significantly contribute to LLD with little to no contribution from the tibia. When the DDH leg was shorter, it was the ATD that contributed to the discrepancy. As the ATD is influenced by the severity of AVN, this explains why the DDH leg was, on average, shorter with worsening AVN grade. Femoral overgrowth and its influence on length after the femoral osteotomy is well documented [7–9]. Zhang et al. [20] reviewed 67 patients with a mean age of 25 years, assessing segmental leg lengths in Hartofilakidis' low and high dislocations of patients with DDH (excluding patients with femoral osteotomy). They reported the ipsilateral femoral shaft to be longer in 78% of cases, regardless of high or low dislocation. If we take this to be the natural history of femoral overgrowth with the femur untreated, this will justify our previous recommendation to shorten the femur at the time of osteotomy. This reduces the soft tissue tension protecting the femoral head from AVN and pre-empting the sequelae of overgrowth [11]. LLD in different lower limb segments is particularly important during hip arthroplasty as surgeons may focus on restoring the hip centre of rotation and not consider the difference in diaphyseal lengths, resulting in a length discrepancy [27].

4.2. Consequences of LLD in DDH

LLD can lead to a gait asymmetry, while there are no measurable kinematic changes in minor differences $\leq$1 cm, a pelvic obliquity compensation can arise above a 2 cm difference [12]. In the coronal plane, pelvic obliquity can cause dynamic acetabular dysplasia in the longer leg or over-coverage and potential impingement of the shorter leg, both of which have been shown to affect the long-term outcome of hip joint development [13]. Segmental LLD in DDH has implications on hip arthroplasty planning as subtrochanteric length differences can be easily missed [29]. The longer leg tends to do more mechanical work, and energy expenditure increases with increasing leg length discrepancy [30,31]. It is, therefore, essential to monitor leg length discrepancy in DDH patients and consider treatment where necessary to limit the impact on gait dysfunction.

4.3. Appropriate Monitoring of LLD in DDH

The literature suggests that clinical examination and tape measures are effective screening tools. However, imaging modalities are more accurate in measuring leg length differences [32]. Our protocol of long-leg standing radiographic follow-up of DDH from age of five allows early diagnosis and planning of appropriate intervention for the management of the leg length discrepancy [24]. Early detection should be managed with simple raises, and follow-up allows for surgical intervention based on bone age. A moderate LLD found in these patients can be treated safely and reliably with epiphysiodesis. The success rate in our case was nearly one in three patients [10,33].

The appropriate imaging modality for monitoring LLD should be chosen, taking into account the radiation exposure, patient movement artefact, and beam distortion [32]. One option for such monitoring is using standing anteroposterior pelvis radiographs and using femoral head height difference (FHHD) to assess leg lengths [7]. This modality does not allow asymmetries resulting from apparent limb length discrepancy due to adduction, abduction, and flexion contractures or fixed spinal deformities. We agree with the opinion of Zhang et al. [20] that using the lesser trochanter on standing AP pelvic radiographs to predict LLD is unreliable. The use of full-length standing anteroposterior radiographs for preoperative templating is advisable for this special group of patients, and its use in our follow-up protocol is justified [24].

4.4. Treatment of LLD in DDH

There is a paucity of literature on whether interventions are needed to address leg length discrepancies in DDH patients. This study found that 30 (27.5%) patients required an LLD intervention, similar to Yoon et al. [7], who reported 23.7%. Inan et al. [10] reported that 12 of their 398 patients required epiphysiodesis, but they did not report on other modalities used to correct LLD or leg length differences within the cohort of patients. In our study, LLD interventions were required across all procedural groups, and we were unable to show a strong correlation with any procedure (Table 4). There was a trend towards increased odds of the need for intervention in the femoral osteotomy group; however, this was not statistically significant. We attribute this to several factors, which include small numbers in each group, successful surveillance and routine shortening of the femur performed during osteotomy at our institution.

4.5. Influence of AVN on LLD in DDH

Fifty-two per cent of our patients were identified to have type II or higher grade of post-avascular necrosis-related proximal femoral growth disturbance (Table 2). More severe grades (III and IV) were more prevalent in the groups with multiple prior operations and older age groups (Table 2 and Figure 3). LLD Interventions were required across all AVN grades, and Type IV had the highest odds of requiring an intervention; however, there were only four patients in the subgroup (Table 5). All LLD procedures in patients with Type III and IV AVN relatively lengthened the shorter ipsilateral leg. Inan et al. [10] found AVN to be a common risk factor in their series of 12 patients that required epiphysiodesis. All their patients had type II or higher AVN. In our series, 15 (50%) patients that required a limb length discrepancy intervention had type II or higher-grade AVN. However, we were not able to find any statistical significance (Table 5).

4.6. Consequences of Untreated Leg Length Discrepancy

Untreated leg length discrepancy can cause pelvic obliquity, resulting in compensatory spinal curvature, also called functional scoliosis [34]. This can result in the wear of facet joints over a long period and a more structural or fixed curve [13,34]. There is currently no evidence on how much LLD contributes to developing a spinal deformity [13,34]. Other compensatory mechanisms include increased knee flexion, external ankle rotation on the longer side, and ankle equinus on the shorter side [13]. These can potentially transition from flexible gait adaptations to fixed contractures if LLD is left untreated [13].

The strengths of this study include a cohort of patients who were systematically followed up by an established protocol. To the best of our knowledge, this is the largest cohort of patients with DDH, followed up by standing radiographs. All radiographs were standardised and calibrated. Limitations include the retrospective nature of the study and the fact that we offered LLD intervention based on clinical necessity and patient choice. This may underestimate the true impact of the leg length discrepancy on our patients.

5. Conclusions

The present study showed that LLD is higher in patients treated for DDH than in the normal population. This knowledge led to the need for a leg length equalisation procedure in 27.5% of the patients. In most patients, the DDH leg was longer, mainly arising from the subtrochanteric region, with data showing a trend towards this femoral overgrowth after femoral osteotomy and in patients with multiple operative procedures to the affected hip. In patients with shorter DDH legs, this was associated with the occurrence of AVN. Our results underline the need for careful monitoring of LLD with long-leg standing films at appropriate stages in patients treated for DDH.

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children

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Review

Windswept Deformity a Disease or a Symptom? A Systematic Review on the Aetiologies and Hypotheses of Simultaneous Genu Valgum and Varum in Children

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Abstract: Objective: The objective of this study is to create an overview of the possible aetiologies of windswept deformity and to emphasize the points of attention when presented with a case. Methods: A systematic search according to the PRISMA statement was conducted using PubMed, African Journals Online, Cochrane, Embase, Google Scholar, and Web of Science. Articles investigating the aetiology of windswept deformity at the knee in children, and articles with windswept deformity as an ancillary finding were included. The bibliographic search was limited to English-language articles only. The level of evidence and methodological appraisal were assessed. Results: Forty-five articles discussing the aetiology of windswept deformity were included. A variety of aetiologies can be brought forward. These can be divided into the following groups: 'Rickets and other metabolic disorders', 'skeletal dysplasias and other genetic disorders', 'trauma' and 'descriptive articles without specific underlying disorder'. With rickets being the largest group. Interestingly, in the group without a specific underlying disorder, all patients were from African descent, being otherwise healthy and presented with windswept deformity between two and three years of age. Conclusion: We have presented an overview that may help identify the underlying disorder in children with windswept deformity. A step-by-step guide for clinicians who see a child with windswept deformity is provided. Even though, according to the Oxford level of evidence, most articles have a low level of evidence.

Keywords: windswept deformity; genu valgum; genu varum; children; rickets

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1. Introduction

In 1975, Oyemade made mention of windswept deformity (WSD), under the term "varovalga", characterised by the formation of a valgus deformity of one knee, and a varus deformity of the contralateral knee [1]. In 1976, the term windswept deformity was used by Fulford et al. to describe the general postural deformity acquired by children with cerebral palsy, in their first weeks of life [2]. Consequently, windswept deformity is used to describe the phenotypical presentation of a varus and valgus deformity, however, the location of this may vary, as well as the underlying pathology. In this article, we will focus on windswept deformity of the knee.

Although the long-term consequences of windswept deformity have not yet been described, it is presumable that it has a similar impact as other angular deformities of the knees. Untreated, it can lead to further deformity, gait abnormalities, limb shortening and osteoarthritis [3].

To date, the aetiology of windswept deformity remains unknown. Oyemade and Smyth described windswept deformity of the knee in previously healthy children, mostly of sub-Saharan descent, with normal developmental milestones [1,4]. The onset usually

occurs in the second or third year of life, shortly after the onset of walking. According to Smyth, the valgus deformity develops first, rapidly followed by a varus deformity on the contralateral side [4]. Suggested hypotheses can be divided into the following categories: metabolic or dietary [5–10], mechanical pressure [1,11], reactive to unilateral disease [12], genetic/race [1,4,13] and traumatic [14]. Windswept deformity can be treated either surgically (corrective osteotomies, stapling) or conservatively (plaster casting) [11].

Apart from varovalgum at the knee, different forms of windswept deformity exist, including hip deformity in children with cerebral palsy [15]. The aim of this study is to perform a systematic search according to the PRISMA statement, in order to analyse which aetiologies of windswept deformity have been published, and assess their level and quality of evidence, as well as any bias. Our aim is to create an overview of the possible aetiologies for windswept deformity and emphasize the points of attention when presented with a case.

2. Materials and Methods

This systematic review was written according to the PRISMA statement for reporting systematic reviews and meta-analyses [16].

2.1. Eligibility Criteria

Types of studies: all articles investigating the aetiology of windswept deformity at the knee and all articles with windswept deformity as an ancillary finding. The language was restricted to articles written in English. There was no restriction on the date of publication.

Types of participants: all patients with a presentation of windswept deformity during childhood (0–18 years) were included. There were no restrictions on gender or race.

2.2. Information Sources

The following databases were searched on 16/10/2020: PubMed, African Journals Online, Cochrane, Embase, Google Scholar, and Web of Science. Articles were screened for eligibility, based on title, abstract and the full text. Additionally, the reference lists of the included articles were screened for further identification of any relevant articles and they were included where applicable.

2.3. Search Strategies

The following search terms were used. The limit "English" was added as mentioned in the eligibility criteria.

1. (windswept deformity) OR (windswept);
2. (((genu valgum) AND (genu varum)) OR ((genu valgum[MeSH Terms]) AND (genu varum[MeSH Terms]))) OR (combined valgus and varus knee) AND ((humans[Filter]) AND (allchild[Filter])) AND (English[Filter]);
3. (((((varo-valgum) OR (varovalgum)) OR (genu varo-valgum)) OR (genu varovalgum)) OR (varo-valga)) OR (varovalga).

An overview of the search can be found in Appendix A.

2.4. Study Selection and Data Extraction

Studies were selected based on the eligibility criteria. First, duplicates were removed, followed by the selection based on title and abstract. The remaining articles were screened for their eligibility based on the full text. The included articles were screened upon a second survey and consensus amongst the authors was reached. The relevant data were extracted and reviewed by the authors.

2.5. Level of Evidence and Quality Assessment

The level of evidence was scored according to the Centre for Evidence-Based Medicine (CEBM) by three authors, followed by an assessment of methodological appraisal. The latter was performed according to the Joanna Briggs Institute (JBI) for case reports, case

series, cohort studies, case-control studies, and cross-sectional studies [17]. Cut-off values were obtained using a scoring system, where a "yes" answer scored 2 points, "unclear" 1 point, "no" 0 points, and "not applicable" was subtracted from the maximum obtainable score. For each article, the obtained score was divided by the maximum obtainable score which led to a percentage. An article was considered to be of high quality when the score was 75% or higher, moderate between 50% and 75%, and low when the score was lower than 50%. Articles classified as "literature review" did not meet the criteria for level of evidence scoring or critical appraisal. An overview of the methodological appraisal can be found in Appendix B.

3. Results

An overview of the results and the number of records retrieved from the final search can be found in Figure 1.

The three searches, performed in six databases, yielded a total of 773 records. After removing 193 duplicates, 580 records remained. Of these records, 424 were excluded based on title/abstract screening, leaving 156 records to be assessed based on the full text. From these 156 full-text records, 109 were excluded, as shown in Figure 1. This resulted in a total of 47 publications that were selected, of these, all the references (n = 1201) were screened from which four additional articles were selected. Upon second survey another six articles were excluded based on age > 18 years (n = 1), not being windswept deformity (n = 2) and no clear aetiology stated (n = 3). This resulted in a final number of 45 selected publications.

Although 45 articles is a substantial amount of included articles, only very few focus on the aetiology of windswept deformity, with most articles describing windswept deformity as an ancillary finding. The Oxford CEBM level of evidence regarding these publications is generally low, with most articles ranked as level IV. Additionally, the quality of the articles, assessed as described in the methods, varied greatly, ranging from a score of 50% to 100%.

In Table 1, general information can be found about the articles, including the country of study, study design, the aim of the study, and the main information related to windswept deformity, as well as the Oxford CEBM classification on the methodological quality score.

From the selected articles, a variety of aetiologies for windswept deformity can be brought forward. These can be divided into the following groups:

- Rickets and other metabolic disorders;
- Skeletal dysplasias and other genetic disorders;
- Trauma;
- Descriptive articles without the specific underlying disorder.

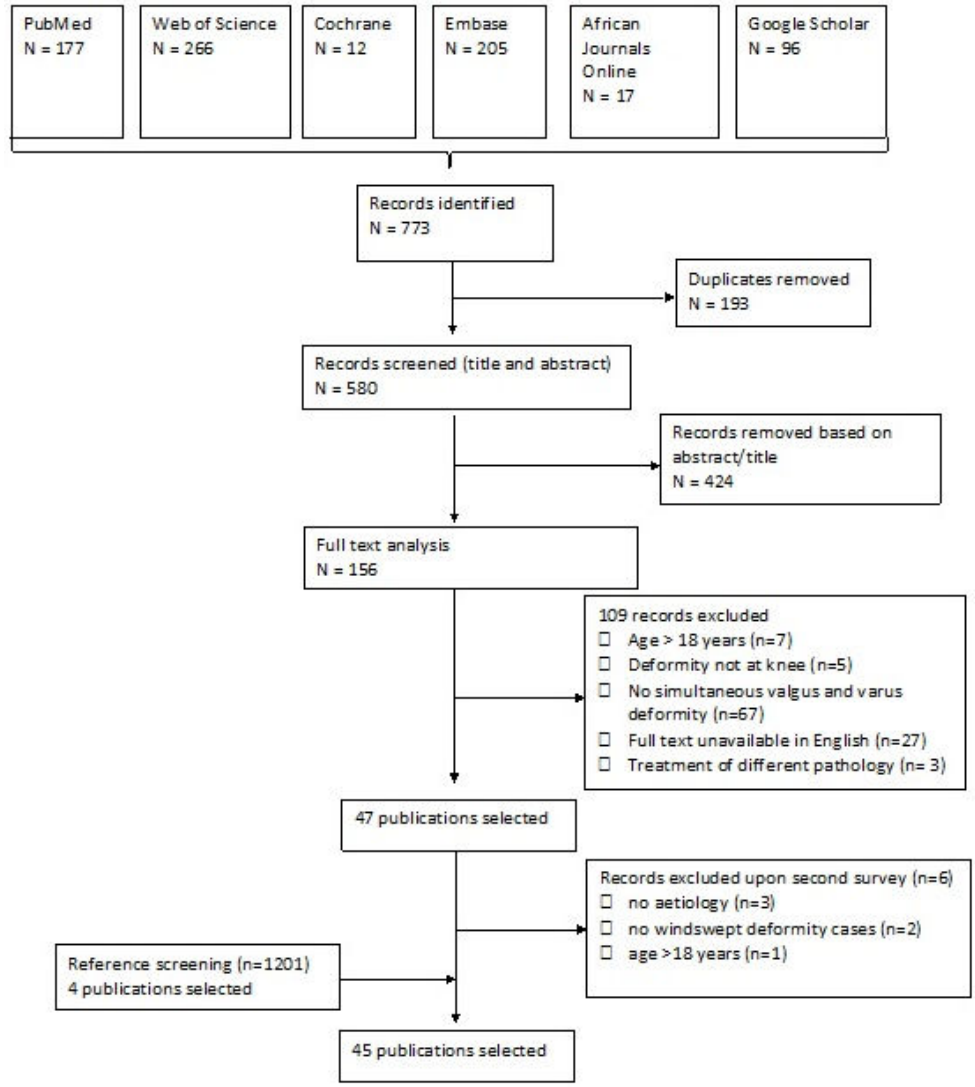

Figure 1. Flow diagram of studies screened and included in the review.

Table 1. Overview of the demographic data of all included studies regarding windswept deformity at the knees.

Article	Country of Study	Study Design	Aim of Study	Elaboration of WSD	WSD Aetiology	Level of Evidence (CEBM) and Methodological Quality
Akpede et al. [5]	Nigeria	Prospective cross-sectional	Determine the prevalence of clinical and biochemical rickets.	Two of ten patients who showed biochemical rickets, though not radiologically, did show WSD, suggesting a form of healed rickets.	Rickets, no radiographic evidence of active rickets	IV high
Al Kaissi et al. [18]	Austria	Case report	WSD in a patient with Schwartz-Jampel syndrome (SJS).	One patient with SJS, with WSD.	SJS	IV high
Al Kaissi et al. [19]	Austria	Case series	Record and discuss WSD in patients with X-linked hypophosphataemic rickets.	In seven patients with hypophosphataemic rickets, the most common angular deformity is WSD.	Hypophosphataemic rickets (from PHEX mutation)	IV moderate
Bar-On et al. [20]	Israel	Retrospective case series	Characterise deformities in patients with renal osteodystrophy (ROD).	One out of five patients showed WSD.	ROD	IV high
Bar-On et al. [14]	Israel	Retrospective case series	To investigate patients with insensitivity to pain.	One patient developed WSD as a consequence of growth disturbance due to untreated fractures of the growth plate.	Trauma	IV high
Bharani et al. [21]	India	Case report	To describe two siblings with sickle cell anaemia, presenting with bilateral lower limb deformities.	Two siblings, both male, 2 and 10 years old with progressive genu valgus on the right, genu varus on the left.	Distal renal tubular acidosis (dRTA)	IV High
Bhimma et al. [22]	Natal, South Africa	Case series	To determine the clinical spectrum of rickets among black children.	WSD was found in two patients with vitamin D deficiency and one patient with Ca deficiency in a total population of 37 patients.	Vitamin D and/or Ca deficiency rickets	IV high
Dudkiewicz et al. [23]	Israel	Case report	Describe the procedure of bone elongation in hypophosphataemic rickets.	One WSD with right genu valgum/left genu varum.	Hypophosphataemic rickets	IV moderate

Table 1. *Cont.*

Article	Country of Study	Study Design	Aim of Study	Elaboration of WSD	WSD Aetiology	Level of Evidence (CEBM) and Methodological Quality
Eralp et al. [24]	Turkey	Case report	Investigate the result of treatment with fixator-assisted intramedullary nailing in two cases with WSD.	Two patients with WSD, treated for vitamin-D-resistant rickets at younger age.	Vitamin-D-resistant rickets	IV high
Gigante et al. [25]	Italy	Case series	Evaluate temporary hemiepiphysiodesis in lower limb deformities in children with renal osteodystrophy (ROD).	One of the seven patients with ROD had WSD. Started as unilateral varus, developed valgus alignment in the contralateral knee.	ROD	IV high
Gupta et al. [26]	India	Literature review	Review the different types of nutritional vs. non-nutritional rickets.	Mention of WSD as a skeletal finding in nutritional rickets. It is not mentioned in non-nutritional rickets.	Nutritional rickets	n.a. *
Ikegawa [27]	Japan	Literature review	Review of the recent advances and current status of the genetic analysis of skeletal dysplasias.	Describes one WSD case, 17 years old with genu valgum on the left.	Skeletal dysplasia	n.a. *
Iyer and Diamond [28]	USA	Literature review	Review the effects of the resurgence of vitamin D deficiency and rickets.	Describes WSD as a possible clinical presentation of rickets.	Vitamin D deficiency rickets	n.a. *
Iyer and Diamond [29]	USA	Literature review	Review of the clinical, radiographic and biochemical manifestations of rickets.	Describes WSD as a possible clinical presentation of rickets.	Vitamin D deficiency rickets	n.a. *
Kenis et al. [30]	Austria	Case report	To describe the deformities in a patient with dysspondyloenchondromatosis (DSC).	One patient with WSD with genu valgum of 30° on the right, genu varum of 10° on the left side. Age at start walking: 3 years.	DSC	IV high
Kim et al. [31]	Korea	Case series	Investigate the mutation frequency in individuals with multiple epiphyseal dysplasia (MED) and identify radiographic predictors.	Two of the fifty-five patients that identified with a previously reported mutation pathogenic for MED presented with WSD at the knee. One MATN3 and one COMP mutation.	MED	IV moderate

Table 1. *Cont.*

Article	Country of Study	Study Design	Aim of Study	Elaboration of WSD	WSD Aetiology	Level of Evidence (CEBM) and Methodological Quality
Lambert and Linglart [6]	France	Literature review	To describe the different causes and therapies of genetic and nutritional rickets.	WSD in walking children is a clinical manifestation of rickets.	Rickets	n.a. *
McKeand et al. [32]	USA	Case-control study	Describe the natural history of pseudoachondroplasia (PSACH).	WSD in 11/67 cases (16.4%), 8/11 cases (72.7%) needed a corrective operation for WSD.	PSACH	IIIb moderate
Muensterer et al. [33]	USA	Literature review	Describe pseudoachondroplasia, and its radiographic features.	In patients with pseudoachondroplasia, WSD typically develops around puberty, when genu varum transforms into WSD due to the progressive joint laxity.	PSACH	n.a.*
Nayak et al. [34]	India	Case report	Describe a case of epidermolytic hyperkeratosis (EHK) with rickets.	Epidermolytic hyperkeratosis (EHK) with rickets in a 6-year-old boy showed progressive WSD since age 3.	EHK	V high
Nishimura et al. [35]	Germany	Case series	Describe TRPV4 mutations in patients with spondylo-epiphyseal dysplasia (SED) and parastremmatic dysplasia.	A 7-year-old patient with reported low birth weight and length. Onset of walking at age 4. At 7 years she had a short stature (-4SD) and WSD. A TRPV4 mutation was found.	Parastremmatic dysplasia (TRPV4 mutation)	IV moderate
Nishimura et al. [36]	Switzerland	Literature review	To describe the different skeletal dysplasia's related to TRPV4 mutations.	Patients with parastremmatic dysplasia have restricted joints and severe misalignment of the lower limbs (severe genu valgum, genu varum or WSD).	Parastremmatic dysplasia (TRPV4 mutation)	n.a. *
Oginni et al. [37]	Nigeria	Prospective case series	Response of oral calcium in Nigerian children with rickets.	Nine out of twenty-six children with underlying Ca-deficiency rickets presented with WSD, they were treated with calcium supplements, with good results.	Calcium-deficiency rickets	IV high

Table 1. *Cont.*

Article	Country of Study	Study Design	Aim of Study	Elaboration of WSD	WSD Aetiology	Level of Evidence (CEBM) and Methodological Quality
Oni and Keswani [38]	Nigeria	Case series	To describe the radiological findings of idiopathic or primary WSD.	Eight WSD patients were found, the onset of clinical and radiological alterations is abrupt, where the disease arises from a formerly normal epiphysis. The radiological features are similar to Blount, and therefore the etiological considerations that apply to Blount may also apply to primary WSD.	Hypotheses: similar to Blount, mechanical pressure, illness	IV moderate
Oni et al. [11]	Nigeria	Case series	To describe windswept deformity.	Eight patients with osteochondrosis with abrupt onset in previously healthy children, with formerly normal epiphyses.	Hypothesis: similar to Blount	IV moderate
Oyemade [13]	Nigeria	Case series	To describe the correction of primary knee deformities in children with and without rickets.	Rachitic patients WSD 12/47, Non-rachitic patients: WSD 15/67.	Rachitic or idiopathic (Blount)	IV moderate
Oyemade [1]	Nigeria	Case series	To clarify aetiological factors in primary deformities of the knee in children.	WSD: peak age male and female 2 years. Rachitic: WSD (12/47). Non-rachitic WSD (15/67).	Rachitic and non-rachitic Blount-like (weight-bearing)	IV moderate
Paruk et al. [39]	South Africa	Case report	Describe two cases of primary hyperparathyroidism (PHPT) in adolescence, mimicking rickets.	A 13-year-old male with progressive pain and WSD (right varus, left valgus). Caused by a parathyroid adenoma.	PHPT	IV high
Pavone et al. [40]	Italy	Literature review	Review hypophosphataemic rickets.	WSD described as a clinical feature of X-linked hypophosphataemic rickets.	X-linked hypophosphataemic rickets	n.a. *
Pettifor et al. [10]	South Africa	Literature review	Presentation of vitamin D deficiency and nutritional rickets in children.	In older children with vitamin D deficiency rickets, WSD may be present.	Vitamin D deficiency rickets and Calcium deficiency rickets	n.a. *

Table 1. *Cont.*

Article	Country of Study	Study Design	Aim of Study	Elaboration of WSD	WSD Aetiology	Level of Evidence (CEBM) and Methodological Quality
Pettifor et al. [41]	South Africa	Case series	Clinical, radiographic and biochemical findings in four children with severe bone deformities resembling rickets.	One out of four children had WSD.	Calcium-deficiency rickets	IV moderate
Prakash et al. [42]	India	Prospective cohort	To evaluate the behaviour of lower limb deformities due to rickets.	Five out of one-hundred and seventeen nutritional rickets patients had WSD. Varus deformity being the youngest, valgus and WSD being older.	Nutritional rickets	Iib high
Prentice et al. [43]	Gambia	Case-control study	Biochemical profile in Gambian children with rickets of unknown aetiology and normal 25OHD.	One out of thirty-seven patients had WSD.	Calcium-deficiency rickets	IIIb high
Shehzad and Shaheen [44]	Pakistan	Case report	Describe a case of epidermolytic hyperkeratosis (EHK) with rickets.	A 13-year old female with WSD, started around the age of 5. Scaling of the skin since birth.	Epidermolytic hyperkeratosis (EHK) with rickets	IV high
Simsek-Kiper et al. [45]	Turkey	Case series	Report on five patients from 2 unrelated families with SEMDFA (spondyloepimetaphyseal dysplasia Faden-Alkuraya type).	One patient presents with WSD (right genu varum, left genu valgum).	SEMDFA	IV High
Smyth [4]	Nigeria	Case report	Describe three cases of windswept deformity.	Three cases of WSD. Two cases with normal development, when suddenly WSD develops. In one case, there is a period of an acute febrile illness (possibly measles) preceding the development of WSD.	Period of epiphyseal instability + stress factor, geographical genetic dysplasia	IV moderate
Solagberu [12]	Nigeria	Prospective case series	Determine the varieties of angular deformities of the knee in children, in/around Ilorin, Nigeria.	Ten patients with WSD presented in one year. Age distribution between 2 and 5 years.	One bone diseased, while the other appears to be compensating	IV moderate

Table 1. *Cont.*

Article	Country of Study	Study Design	Aim of Study	Elaboration of WSD	WSD Aetiology	Level of Evidence (CEBM) and Methodological Quality
Teotia et al. [46]	India	Literature review	Report effects of endemic fluoride exposure on metabolic bone disease.	WSD as presentation of bony leg deformity due to high levels of fluoride exposure in drinking water.	Endemic chronic fluoride toxicity	n.a. *
Thacher et al. [47]	Nigeria/South Africa	Case report	Three cases of vitamin D-deficiency rickets associated with ichthyosis.	WSD in two patients with ichthyosis and rickets.	Ichthyosis with rickets	IV high
Thacher et al. [7]	Nigeria	Literature review	Describe the features of calcium-deficiency rickets.	Bowleg deformity is less specific for active rickets than knock-knee or WSD.	Calcium-deficiency rickets	n.a. *
Thacher et al. [9]	Nigeria	Case-control study	Determine whether low dietary calcium intake is associated with rickets in Nigerian children.	Of 123 Nigerian children with rickets: 16 (13%) had WSD.	Calcium-deficiency rickets	IIIb high
Thacher et al. [8]	Nigeria	Cohort study	Development of a clinical prediction model for active rickets.	The median age of onset of WSD was 24 months. WSD present in 39 of the 278 cases of active rickets (14%). Children presenting leg deformities over a span of 4 years, 95/736 had WSD (12.9%).	Rickets	IIb moderate
Vatanavicharn et al. [48]	USA	Case report	Radiographic patellar finding in a patient with pseudoachondroplasia (COMP mutation).	At age 5, the patient developed WSD (right genu varum, left valgum).	Pseudoachondroplasia (following COMP mutation)	IV high
Weiner et al. [49]	USA	Case series	Characterise the typical orthopaedic findings in pseudoachondroplasia.	Angular deformity of knees: genu valgum $n = 35$ (22%), genu varum $n = 89$ (56%), WSD $n = 35$ (22%). Laxity in all patients.	Pseudoachondroplasia (following COMP mutation)	IV moderate
Yilmaz et al. [50]	Croatia	Cohort study	Temporary hemi epiphysiodesis for correction of angular deformities in children with skeletal dysplasia.	One patient with metaphyseal dysplasia had WSD and underwent correction for both varus and valgus deformity.	Metaphyseal dysplasia	Iib moderate

* n.a.: not applicable, articles classified as "literature review" did not meet the criteria for level of evidence scoring or critical appraisal. Literature reviews did not provide the number of WSD patients included and therefore these numbers are not mentioned in the table for literature reviews.

In Table 2, a quantitative summary was made on the demographic data of windswept deformity. A total of 184 patients with windswept deformity were included in Table 2, in 69 cases the gender was described: 45 (65.2%) males and 24 (34.8%) females. Of the 170 patients where the race is described, 108 (63.5%) are of African descent, 11 (6.5%) Asian and 50 (29.4%) Caucasian (37 of the Caucasian patients were retrieved from the same article about pseudoachondroplasia) [49]. One person of Bedouin descent was classified as Middle Eastern (0.6%). Based on the articles found in this study, an overview of the identifying features of each aetiology of windswept deformity is presented in Table 3. Figure 2 shows a flowchart that can be used to find the possible cause of windswept deformity in a child.

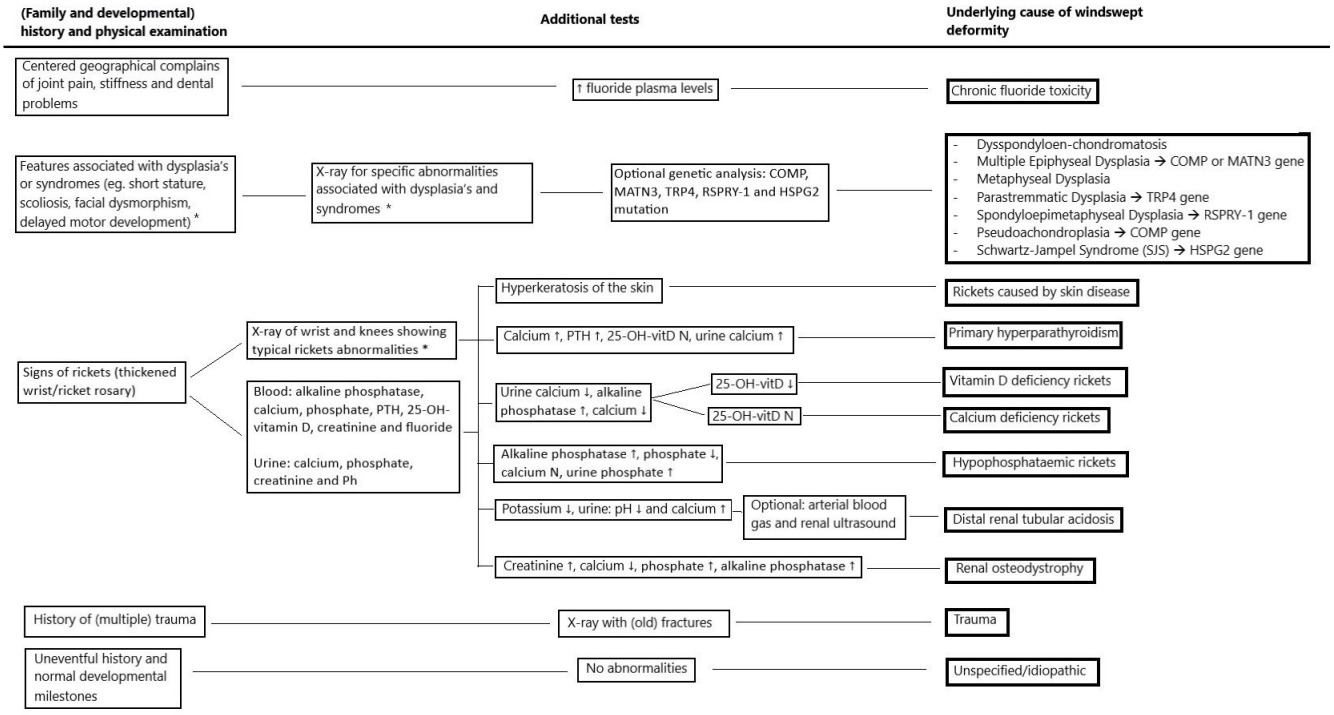

Figure 2. Flowchart to find the possible cause of windswept deformity in a child. * See Table 3 for more detailed descriptions.

Table 2. Quantitative overview of the demographic data from the studies describing the aetiology windswept deformity *.

Author	Country of Study	WSD Cases	Male	Female	Suggested Aetiology or Hypothesis	Ethnicity	Onset Age (Months)	Onset Valgus	Onset Varus	Valgus Right	Valgus Left
Akpede et al. [5]	Nigeria	2	-	-	Biochemical rickets	African	-	-	-	-	-
Al Kaissi et al. [49]	Austria	1	1	0	Schwarz-Jampel Syndrome (SJS)	-	-	-	-	-	1
Al Kaissi et al. [19]	Austria	7	7	0	X-linked hypophosphataemic rickets	-	14–18	-	-	-	-
Bar-On et al. [20]	Israel	1	1	0	Renal osteodystrophy (ROD)	-	-	1	-	1	-
Bar-On et al. [14]	Israel	1	0	1	Congenital insensitivity to pain (trauma)	Middle-East	-	-	-	1	-
Bharani et al. [21]	India	2	2	0	Distal renal tubular acidosis (dRTA)	Asian	36	-	-	2	-
Bhimma et al. [22]	South Africa	3	-	-	Vitamin D and/or Ca deficiency rickets	African	-	-	-	-	-
Dudkiewicz et al. [23]	Israel	1	1	0	Hypophosphataemic rickets	-	-	-	-	1	-
Eralp et al. [24]	Turkey	2	1	1	Vitamin D-resistant rickets	-	-	-	-	2	-
Gigante et al. [25]	Italy	1	1	0	Renal osteodystrophy (ROD)	-	-	-	1	1	-
Ikegawa [27]	Japan	1	1	0	Pseudoachondroplasia	Asian	-	-	-	-	1
Kenis et al. [30]	Austria	1	1	0	Dysspondyloenchondromatosis (DSC)	Caucasian	-	-	-	1	-
Kim et al. [31]	Korea	2	2	0	Multiple epiphyseal dysplasia (MED)	Asian	-	-	-	-	-
McKeand et al. [32]	USA	11	-	-	Pseudoachondroplasia	Caucasian	-	-	-	-	-
Nayak et al. [34]	India	1	1	0	Rickets (epidermolytic hyperkeratosis)	-	36	-	-	-	-
Nishimura et al. [35]	Germany	1	0	1	Parastremmatic dysplasia (with TRPV4 mutation)	African	-	-	-	-	1
Oginni et al. [37]	Nigeria	9	-	-	Ca deficiency rickets	African	-	-	-	-	-
Oni and Keswani [38]	Nigeria	8	3	5	Similar to Blount mechanical pressure + illness	African	6–24	1	1	2	-
Oyemade [1]	Nigeria	28	18	10	Rickets (12) or non-rachitic (15) (3 hypotheses: weight-bearing, dietetic, genetic)	African	12–108	-	-	-	-

Table 2. *Cont.*

Author	Country of Study	WSD Cases	Male	Female	Suggested Aetiology or Hypothesis	Ethnicity	Onset Age (Months)	Onset Valgus	Onset Varus	Valgus Right	Valgus Left
Paruk et al. [39]	South Africa	1	1	0	Primary hyperparathyroidism	African	150	-	-	-	1
Pettifor et al. [41]	South Africa	1	0	1	Ca deficiency rickets	African	-	-	-	1	-
Prakash et al. [42]	India	5	-	-	Nutritional rickets	Asian	48–120	-	-	-	-
Prentice et al. [43]	Gambia	1	-	-	Ca deficiency rickets	African	-	-	-	-	-
Shehzad and Shaheen [44]	Pakistan	1	0	1	Rickets (epidermolytic hyperkeratosis)	Asian	60	-	-	-	-
Simsek-Kiper et al. [45]	Turkey	1	1	0	Spondyloepimetaphyseal dysplasia Faden-Alkuraya type (SEMDFA)	Caucasian	36–48	-	-	-	1
Smyth [4]	Nigeria	3	2	1	Period of epiphyseal instability + stress factor, geographical genetics	African	24–54	3	-	1	1
Solagberu [12]	Nigeria	10	-	-	Compensation for 1 diseased bone	African	24–60	-	-	-	-
Thacher et al. [47]	Nigeria/South Africa	2	1	1	Ichthyosis with rickets	African	-	-	-	-	-
Thacher et al. [8]	Nigeria	39	-	-	Rickets	African	-	-	-	-	-
Vatanavicharn et al. [48]	USA	1	0	1	Pseudoachondroplasia	Caucasian	60	-	-	-	1
Weiner et al. [49]	USA	35	-	-	Pseudoachondroplasia	Caucasian	-	-	-	1	-
Yilmaz et al. [50]	Croatia	1	0	1	Metaphyseal dysplasia	Caucasian	-	-	-	-	1
Total		184	45	24		African n = 108 Asian n = 11 Caucasian n = 50 Middle-east n = 1 Missing ethnicity n = 14		5	2	14	8

* All literature reviews and other overlapping study populations were excluded from this quantitative overview.

Table 3. An overview of the clinical presentation of each specific aetiology.

Aetiology Group	Specific Aetiology	Family History	History	Physical Exam	Laboratory Findings	X-Ray	Articles
Rickets	Vitamin D deficiency rickets*	Generally uneventful	Decreased exposure to sunlight	Thickened wrists and ankles, rickety rosarymuscle weakness	Alkaline phosphatase ↑ Phosphate ↓/N/↑ Calcium ↓ 25-OH-vitD ↓ 1.25-di-OH-vit D ↓/N/↑ PTH ↑ Urine: Ca ↓	Widening of the growth plate and abnormal configuration of the metaphysis: – Fraying indistinct margins of the metaphysis – Splaying: widening of metaphyseal ends – Cupping: concavity of metaphysis Most seen in the wrist and knee Anterior rib ends: rachitic rosary	Bhimma et al. [22], Thacher [7], Iyer et al. [28,29], Gupta et al. [26], Prakash et al. [42]
	Calcium deficiency rickets*	Generally uneventful	Low-calcium diet	Thickened wrists and ankles, rickety rosary	Alkaline phosphatase ↑ Phosphate ↓ Calcium ↓ 25-OH-vitD N 1.25-di-OH-vit D ↑ PTH ↑ Urine: Ca ↓	See above	Bhimma et al. [22], Oginni et al. [37], Prentice et al. [43], Pettifor et al. [41], Gupta et al. [26]
	Hypophosphataemic rickets	X-linked (PHEX mutation) or autosomal dominant (FGF23 mutation) transmission	Delayed walking, muscular weakness, bone pain, failure to thrive, tooth abscesses	Thickened wrists and ankles, rickety rosary, dental abnormalities	Alkaline phosphatase ↑ Phosphate ↓ Calcium N 25-OH-vitD N 1.25-di-OH-vitD N/↓ PTH N/↑ FGF23 ↑Urine: phosphate ↑ Genetic testing: PHEX mutation	See above	Bhimma et al. [22], Al-Kaissi et al. [19], Dudkiewicz et al. [23], Pavone et al. [40], Eralp et al. [24], Gupta et al. [26], Prakash et al. [42]
	Skin disease (Epidermolytic hyperkeratosis or Ichthyosis)	Consanguinity and familial inheritance may occur	Bright red blisters after birth. Development of hyperkeratotic plaques	Generalised dry skin, hyperkeratotic and cobble-stone plaques. Rib beading, widening of wrists and ankles	Alkaline phosphatase ↑ Phosphate ↓ Calcium ↓ 25-OH-vitD ↓ PTH ↑ Skin biopsy: hyperkeratosis	See above	Shehzad and Shaheen [44], Nayak et al. [34], Thacher [47]
Other metabolic	Primary hyper-parathyroidism	Generally uneventful	Progressive pain, normal developmental milestones	No abnormalities	Alkaline phosphatase ↑ Phosphate ↓ Calcium ↑ PTH ↑ 25-OH-vitD N 1.25-di-OH-vitD ↑ Urine: Ca ↑	See above and a sestamibi scan: increased focal uptake of the parathyroid glands	Paruk et al. [39]

Table 3. *Cont.*

Aetiology Group	Specific Aetiology	Family History	History	Physical Exam	Laboratory Findings	X-Ray	Articles
	Chronic fluoride toxicity	Affected family members, centred geographic distribution of fluoride levels	Mild: generalised bone and joint painmoderate: stiffness and rigiditySevere: flexion deformities at hips and knees	Stiff and rigid spine and joints, flexion deformity hips, knees and elbows, hypo-mineralisation of tooth enamel	Alkaline phosphatase ↑ Phosphate N Calcium N 25-OHD N1-25(OH)2D ↑ PTH ↑ Plasma fluoride ↑	Osteosclerosis, periosteal bone formation, calcifications of interosseus membrane, rickets-like metaphyses	Teotia et al. [46]
	Distal renal tubular acidosis	Familial inheritance	Sickle cell disease, failure to thrive, polyuria, polydipsia	Low weight/height, frontal bossing, wrist widening (signs of rickets)	ABG: Metabolic acidosis Potassium ↓ Chloride ↑ Urine: pH↓, calcium ↑ Renal ultrasound: nephrocalcinosis	Osteopenia, angular deformities and signs of rickets	Bharani et al. [21]
	Renal Osteodystrophy	Familial inheritance may occur	Bone pain, muscle weakness	Significant growth retardation	Alkaline phosphatase ↑ Phosphate ↑ Calcium ↓ Creatinine ↑ PTH ↑	Widening and elongation of the growth plates and cupping of the metaphysis and signs of rickets	Bar-On et al. [20], Gigante et al. [25]
Dysplasia's and syn-dromes	Dysspondyloen-chondromatosis	Generally uneventful	Delays in motor development	Neonatal dwarfism, unequal limb length, flat midface with frontal prominence and progressive kyphoscoliosis	No abnormalities	Aniosospondyly and enchondroma-like lesions in the metaphyseal and diaphyseal portions of the long tubular bones	Kenis et al. [30]
	Multiple Epiphy-seal Dysplasia	Familial inheritance may occur	Joint pain, scoliosis, deformities hands, feet, knees and hips	Muscular hypotonia, ligamentous hyperlaxity, abnormal gait, angular deformities at hips and knees	Genetic testing: COMP or MATN3 mutations	COMP: small and round femoral head, MATN3: crescent-shaped femoral head	Kim et al. [31]
	Metaphyseal Dysplasia	Consanguinity and familial inheritance may occur	Mental, physical and height development are usually normal	Angular deformities of the knees, palpable widening of the distal femur and clavicles	no abnormalities	Erlenmeyer flask deformity	Yilmaz et al. [50]

Table 3. *Cont.*

Aetiology Group	Specific Aetiology	Family History	History	Physical Exam	Laboratory Findings	X-Ray	Articles
	Parastremmatic Dysplasia	Generally uneventful	Normal mental milestones, motor development may be slightly delayed, short stature	Windswept and flexural deformity of the legs, scoliosis, platyspondyly	Genetic testing: TRP4 mutation	Flaky metaphyses with wide zones of radiolucencies and flocky calcifications, disorganised epiphyseal ossifications, severe platyspondyly	Nishimura et al. [35,36]
	Spondyloepimetaphyseal Dysplasia Faden-Alkuraya type	Parental consanguinity, autosomal recessive inheritance	Difficulty walking, short stature, delayed motor and mental development	Short stature, hypertelorism, brachycephaly, short nose with depressed nasal bridge, tented upper lip, proptosis	Genetic testing: RSPRY-1 mutation	Mild spondylar dysplasia, epi-metaphyseal dysplasia of long bones (flat and irregular epi- and metaphyseal flaring)	Siimsek-kiper et al. [45]
	Pseudoachondroplasia	Autosomal dominant inheritance	Normal birthweight and length, At around 2-4yr of age short stature and disproportionately short limbs appear	Short stature, disproportionately short limbs, short and stubby fingers, increased joint laxity, waddling gait	Genetic testing: COMP mutation	Irregular or fragmented epiphyses, flaring, widening or trumpeting of the metaphyses, anterior beaking of vertebrae	Muensterer et al. [33], Vatanavicharn et al. [27], Weiner et al. [49], McKeand et al. [32]
	Schwartz-Jampel Syndrome (SJS)	Parental consanguinity and familial inheritance	Normal gestation with severe muscle stiffness at birth	Dysmorphic facial features, trismus	Genetic: HSPG2 gene mutation	Kyphoscoliosis, platyspondyly with coronal clefts in vertebrae, inferior femoral and superior tibial epiphyses look enlarged and distorted	Al-Kaissi et al. [48]
Trauma	Trauma	Generally uneventful	History of fractures	Abnormal gait, signs of bruises and evidence of (old) fractures	Non specific	Evidence of (old) fractures	Bar-On et al. [14]

* Lab findings in vitamin D deficiency and hypocalcemia rickets depend on the phase of rickets. Phase 1: hypocalcemia causes PTH rise, leading to bone resorption and hyperphosphatemia and rise in alkaline phosphatase. This phase has a relative resistance to PTH. Phase 2: PTH rises further and overcomes the resistance. Calcium rises to normal or slightly lower than normal range and phosphate decreases further (renal excretion due to PTH). Phase 3: hypocalcemia returns worse because of depleted reserves, hypophosphatemia persists and further rise of alkaline phosphatase. In phase 3 X-ray abnormalities become visible. ↓: lower compared to reference standard. ↑: higher compared to reference standard.

3.1. Rickets and Other Metabolic Disorders

About half of the articles included reported rickets in patients with windswept deformity (n = 23). In five articles, accounting for a total of 68 patients, the rickets type was unspecified [1,5,6,8,13]. However, in three articles, for a total of eight patients, rickets was found to be (X-linked) hypophosphataemic, due to diminished reabsorption of phosphate in the kidneys [19,23,40]. Nutritional rickets due to vitamin D or calcium deficiency was found in another 12 articles, accounting for another 36 patients with windswept deformity [7,9,10,22,24,26,28,29,37,41–43]. In four patients, from three articles, rickets was caused by a skin disorder, namely epidermolytic ichthyosis, which is present at birth [34,44,47]. This adds up to 116 patients with windswept deformity, likely due to rickets of varying types.

A further four types of metabolic disorders were found with windswept deformity at presentation, accounting for an additional six cases. These include primary hyperparathyroidism (PHPT) (n = 1) [39], chronic fluoride toxicity (n = 1) [46], distal renal tubular acidosis (dRTA) (n = 1) [21] and renal osteodystrophy (ROD) (n = 3) [20,25].

3.2. Skeletal Dysplasia and Other Genetic Disorders

Six cases of windswept deformity were found in patients with different types of dysplasia's: multiple epiphyseal dysplasia (MED) (n = 6) [31], dysspondyloenchondromatosis (DSC) (n = 1) [30], metaphyseal dysplasia (n = 1) [50], parastremmatic dysplasia (with TRPV4 mutation) (n = 1) [35,36] and spondyloepimetaphyseal dysplasia Faden-Alkuraya type (SEMDFA) (n = 1) [45]. These skeletal dysplasias are often characterised by a specific mutation. In MED cases, the mutations are either on the COMP or MATN3 gene, the former also being known to cause pseudoachondroplasia (PSACH), another disease in which windswept deformity can be found. We found 48 patients with PSACH presenting with windswept deformity, from five articles [27,32,33], 36 of which bring confirmed COMP mutations [48,49]. Schwartz-Jampel Syndrome (SJS) was another syndrome in which one patient with windswept deformity has been described [18]. Patients with skeletal dysplasia often present with scoliosis and consequently WSD towards the contralateral side.

3.3. Trauma

In one patient, trauma seemed to be the cause of the windswept deformity [14]. This patient was a young female with a history of fractures, often untreated due to congenital insensitivity to pain, presumably being the cause of the angular deformities of the legs.

3.4. Descriptive Articles without Specific Underlying Disorder

We grouped six articles together with a total of 36 patients, in which different hypotheses for the cause of windswept deformity were brought forward: a combination of mechanical pressure and a period of illness (similar cause as Blount disease) (n = 8) [11,38]; excessive or early weight-bearing, dietic or ethnicity (n = 15) [1,13]; a combination of the previously mentioned factors (n = 3) [4]; compensation, where only one side is diseased, while the other side compensates (n = 10) [12]. All of these hypotheses were (partly) based on mechanical loading or weight-bearing. The patients in these articles were all African and typically presented with windswept deformity between the age of 2 and 3 years, being otherwise healthy. None of these patients showed signs of (healed) rickets.

4. Discussion

This systematic review gives an overview of all previously published aetiology hypotheses for windswept deformity. Windswept deformity is generally limited to an individual with genu valgum on one side, and genu varum on the contralateral side. Further specific phenotypical descriptions vary depending on the aetiology.

Our results display that windswept deformity can be a manifestation of a broad variety of pathologies. However, in patients in whom no underlying illness was found, the deformity was deemed idiopathic, and the following hypotheses are brought forward:

weight-bearing (due to excessive weight or early walking) [1,11,13,38], epiphyseal instability [4], stress factors (illness) [4] and geographical genetic factors [1,4,13]. Though these hypotheses are comparable between articles, they lack supporting evidence.

Rickets of different types was found to be the most common pathology manifesting windswept deformity. Although it appears to be the most frequent cause of windswept deformity, windswept deformity is far from the most common presentation of rickets. Thacher [7], presents different clinical features and their utility in predicting radiologically active rickets; only 14% of the children with radiologically active rickets have windswept deformity and the probability that a child with windswept deformity has radiologically active rickets is 41%. Unfortunately, no further research has been conducted on factors influencing the development of windswept deformity, genu valgum or genu varum in children with rickets. Bhimma et al. [22] concluded that mainly vitamin D deficiency is responsible for rickets, however, this may be aggravated by calcium deficiency. These two deficiencies may explain the geographical and ethnic distribution of windswept deformity. As seen in Table 2, most of the patients with windswept deformity were African, and about half of the included studies were performed in sub-Saharan African countries. The diet of rural African children is often low in milk and other dairy products, hence leading to a reduced calcium intake from the diet, despite the fortification of products with calcium [51]. The vitamin D deficiency may be explained by the increased sunlight exposure required for black children [22], or by cultural and/or religious factors that limit the exposure to sunlight, such as covering garments or veils. Hypophosphataemic rickets is most commonly found in its X-linked inheritance form and causes rickets due to its defects in renal handling of phosphorus [52]. Furthermore, epidermolytic ichthyosis is described in windswept patients caused by rickets due to marked hyperkeratosis of the skin [53]. Consequently, there is a decreased synthesis of vitamin D in the epidermis stimulating parathyroid hormone secretion, and a higher risk of rickets [54].

The cases of windswept deformity occurring in patients with other metabolic disorders are often comparable to the pathophysiology behind the different forms of rickets. In calcium deficiency rickets, patients present with secondary hyperparathyroidism as the low calcium levels stimulate the increased production of PTH. Similarly, primary hyperparathyroidism, distal renal tubular acidosis (dRTA) and renal osteodystrophy cause abnormalities in calcium and phosphorus levels. Exposure to high levels of fluoride in drinking water may lead to a decrease in strength by altering the structural integrity of the bone microarchitecture, which possibly leads to skeletal deformities, such as windswept deformity. Teotia et al. describe the difficulty in differentiating calcium-deficiency rickets from fluoride toxicity, and believe that every child presenting with bone disease in areas endemic to fluorosis is a case of skeletal fluorosis until proven otherwise [46].

A variety of skeletal dysplasias have been described in cases of windswept deformity. Often, these dysplasias are caused by a specific mutation, and a positive family history may therefore be present. Additionally, these patients often present additional symptoms, for example, brachydactyly and craniosynostosis for SEMDFA [45]. PSACH is caused by a mutation in the COMP gene and is usually inherited in an autosomal dominant manner. Although PSACH is not usually discovered until the age of 2-3 years, when disproportionate short stature, waddling gait and evidence of increased joint laxity starts to develop, these features should be used to distinguish PSACH as the cause of windswept deformity [49]. Most patients with a syndrome or dysplasia presenting with windswept deformity have other clinical features, such as malformations of the eyes and face, which can be used to identify the underlying syndrome.

Only a single article, by Bar-On et al. [14], describes trauma leading to windswept deformity. The precise location of the fractures is not described, and therefore, it is impossible to conclude whether this case of windswept deformity was due to compensation for one malformed leg, or if the trauma occurred in both legs, leading to the deformity. The child in this single case suffered from congenital insensitivity to pain, and therefore had a higher risk of multi-trauma leading to lower limb deformities.

The remaining articles, in which other hypotheses for windswept deformity were described, explained its development in patients where there is no underlying cause found. We found no explicit evidence of early walking or excessive weight. Although the beforementioned causes were poorly reported, all patients in these studies had comparable stories. All were of African descent, otherwise healthy, with no signs of healed rickets and the age of onset was between 2 and 3 years of age. There might be geographical or genetic factors that could explain the distribution of windswept deformity in the unspecified group.

When a child presents with windswept deformity, the number of possible underlying causes is extensive, and a complete overview is helpful to make the correct diagnosis. Table 3 shows an overview of the identifying features of each aetiology found in this review. To find a possible cause we advise extensive history taking, including family history (to exclude genetic dysplasias or syndromes) and developmental history; detailed physical examination, looking for clinical features suggestive of rickets (thickened wrists and ankles, and/or rickety rosary), or other identifying features associated with dysplasias or syndromes (facial dysmorphisms, ligamentous laxity, deformities at multiple joints, etc.); and additional testing, such as X-rays of the lower limb and wrist, looking for evidence of rickets or other abnormalities which might fit specific skeletal dysplasias, blood panel (alkaline phosphatase, calcium, phosphate, magnesium, PTH, 25-OH-vitamin D, albumin, creatinine and fluoride) and spot urine test (calcium, phosphate, creatinine and Ph) to exclude or identify rickets and/or other metabolic causes. If required, additional blood tests (e.g., chloride, potassium, 1.25-di-OH-vitamin D, arterial blood gas (ABG) and FGF23) can be performed. Genetic analysis may be indicated when a specific dysplasia or syndrome (COMP, MATN3, TRP4, RSPRY-1 and HSPG2 mutation), or hypophosphataemic rickets (PHEX mutation) is suspected. Additionally, a renal ultrasound can be performed in the case of distal renal tubular acidosis or a skin biopsy to confirm hyperkeratosis. However, when the cause of windswept deformity cannot be found it may be multifactorial, including mechanical loading or weight-bearing.

Despite the large number of articles included in this review, only very few focus on the aetiology of windswept deformity, and most articles have a different aim, describing the deformity as an ancillary finding. Additionally, most articles have old publishing dates. On the other hand, most articles occur in low and middle-income countries, which may result in an underestimation of the problem as less research is conducted in low and middle-income countries. Hence, the specific data available on patients with windswept deformity is often limited. Despite not being found in the literature as causes of windswept deformity, logically there are more underlying disorders that can cause windswept deformity, for instance, other skeletal dysplasias or metabolic disorders (e.g., hypomagnesemia or hypo-albuminemia).

5. Conclusions

Currently, the existing evidence on the aetiology of windswept deformity of the knee shows a broad spectrum of underlying causes that may lead to its development. However, when none of these specific causes can be identified, it appears that the aetiology is multifactorial, resting on the hypotheses of weight-bearing, epiphyseal instability, stress factors and geographical genetic factors. Presently, not enough evidence is available to confirm these hypotheses, and more research is necessary. Nevertheless, we have presented an overview, which helps guide clinicians presented with a case of windswept deformity. A thorough (family and developmental) history, followed by physical examination, and additional tests, such as X-rays, blood panels, urine tests, renal ultrasound and genetic analysis, may help identify the underlying disorder.

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Appendix A

Table A1. Search terms.

Database	Search 1	Search 2	Search 3
Pubmed	(windswept deformity) OR (windswept)	(((genu valgum) AND (genu varum)) OR ((genu valgum[MeSH Terms]) AND (genu varum[MeSH Terms]))) OR (combined valgus and varus knee) AND ((humans[Filter]) AND (allchild[Filter])) AND (english[Filter])	((((((varo-valgum) OR (varovalgum)) OR (genu varo-valgum)) OR (genu varovalgum)) OR (varo-valga)) OR (varovalga)
AJOL	(windswept deformity) OR (windswept)	((genu valgum) AND (genu varum)) OR (combined valgus and varus knee)	(((((varo-valgum) OR (varo valgum)) OR (genu varo-valgum)) OR (genu varo valgum)) OR (varo-valgo)) OR (varovalga)
Cochrane	windswept deformity in Title Abstract Keyword OR windswept in Title Abstract Keyword	((“genu valgum”):ti,ab,kw OR (“genu valgus”):ti,ab,kw) “AND” ((“genu varum”):ti,ab,kw OR (“genu varus”):ti,ab,kw)	(genu varovalgum) OR (genu varo-valgum) OR (varo-valgum) OR (varovalgum) OR (varovalga) OR (varo-valga)
Embase	windswept deformity.mp. OR windswept.mp.	valgus knee/AND varus knee/limit to (human and English language)	varo-valgum.mp. OR genu varo-valgum.mp.
G-scholar	knee or genu, “windswept deformity” or windswept, -cerebral, -palsy, -osteoarthritis	combined valgus and varus OR “simultaneous valgus and varus” OR “combined varus and valgus” OR “simultaneous varus and valgus” OR “simultaneous varus and valgus”	genu varovalgum
WoS	(windswept deformity OR windswept)	(TS = (genu valgum “and” genu varum) OR (TS = (combined valgus and varus knee “OR” simultaneous valgus and varus))) AND LANGUAGE: (English)	genu varovalgum OR genu varo-valgum OR varovalgum OR varo-valgum OR varo-valga

Appendix B

Table A2. Overview Table of Methodological Appraisal and Scoring.

Author	Study Design	1	2	3	4	5	6	7	8	9	10	11	Score	Maximum Score	Percentage	Low/Moderate/ High
Akpede et al. [5]	cross-sectional	Y	Y	Y	Y	N	N	Y	Y				12	16	75	high
Al Kaissi et al. [18]	case report	Y	Y	Y	Y	Y	Y	Y	N				14	16	87.5	high
Al Kaissi et al. [19]	case series	N	Y	Y	UC	UC	Y	N	UC	Y	NA		11	18	61.1	moderate
Bar-On et al. [20]	case series	Y	UC	UC	Y	UC	Y	Y	Y	Y	NA		15	18	83.3	high
Bar-On et al. [14]	case series	Y	Y	Y	UC	UC	Y	Y	Y	Y	NA		16	18	88.9	high
Bharani et al. [21]	case report	Y	Y	Y	Y	N	NA	N	N				12	14	85.7	high
Bhimma et al. [22]	case series	Y	Y	Y	UC	UC	Y	N	Y	Y	NA		14	18	77.8	high

Table A2. *Cont.*

Author	Study Design	1	2	3	4	5	6	7	8	9	10	11	Score	Maximum Score	Percentage	Low/Moderate/ High
Dudkiewicz et al. [23]	case report	N	Y	N	N	N	Y	Y	Y				8	16	50	moderate
Eralp et al. [24]	case report	Y	N	Y	Y	Y	Y	N	Y				12	16	75	high
Gigante et al. [25]	case series	Y	Y	UC	Y	UC	N	Y	Y	Y	NA		14	18	77,8	high
Kenis et al. [30]	case report	N	Y	Y	Y	Y	Y	N	Y				12	16	75	high
Kim et al. [31]	case series	Y	Y	Y	N	UC	Y	Y	NA	N	Y		13	18	72.2	moderate
McKeand et al. [32]	case-control study	Y	UC	Y	Y	Y	N	N	Y				11	16	68.75	moderate
Nayak et al. [34]	case report	N	Y	Y	Y	Y	Y	N	Y				12	16	75	high
Nishimura et al. [35]	case series	Y	Y	Y	N	NA	N	N	Y	Y	NA		10	16	62.5	moderate
Oginni et al. [37]	prospective case series	Y	Y	Y	Y	Y	N	N	Y	Y	Y		16	20	80	high
Oni and Keswani [38]	case series	Y	UC	UC	UC	UC	N	Y	NA	Y	NA		10	16	62.5	moderate
Oni et al. [11]	case series	Y	UC	UC	Y	UC	N	Y	NA	N	NA		9	16	56.3	moderate
Oyemade [13]	case series	N	Y	Y	Y	UC	N	Y	Y	Y	NA		13	18	72.2	moderate
Oyemade [1]	case series	Y	Y	Y	Y	UC	N	Y	N	Y	NA		13	18	72.2	moderate
Paruk [39]	case report	Y	Y	Y	Y	Y	Y	Y	Y				16	16	100	high
Pettifor et al. [41]	case series	N	Y	Y	UC	UC	Y	Y	Y	N	NA		12	18	66.7	moderate
Prakash [42]	prospective cohort	NA	NA	Y	NA	N	Y	Y	Y	Y	Y	Y	14	16	87.5	high
Prentice et al. [43]	case-control study	UC	UC	Y	Y	Y	N	Y	Y				12	16	75	high
Shehzad and Shaheen [44]	case report	Y	Y	Y	Y	Y	Y	N	N				12	16	75	high
Simsek-Kiper et al. [45]	case series	N	Y	Y	UC	UC	Y	Y	NA	Y	NA		12	16	75	high
Smyth [4]	case report	Y	N	Y	Y	N	N	N	Y				8	16	50	moderate
Solagberu [12]	prospective case series	Y	UC	Y	Y	UC	N	Y	N	Y	Y		14	20	70	moderate
Thacher et al. [47]	case report	Y	Y	Y	Y	Y	Y	N	Y				14	16	87.5	high
Thacher et al. [9]	case-control study	Y	Y	Y	Y	Y	N	N	Y				12	16	75	high
Thacher et al. [8]	cohort study	Y	Y	Y	N	N	N	Y	NA	NA	NA	Y	10	16	62.5	moderate
Vatanavicharn et al. [48]	case report	Y	Y	Y	Y	NA	NA	NA	Y				10	10	100	high
Weiner et al. [49]	case series	N	Y	UC	UC	UC	Y	Y	NA	Y	NA		11	16	68.8	moderate
Yilmaz et al. [50]	cohort study	Y	Y	Y	N	N	Y	Y	Y	Y	UC	Y	15	22	68.2	moderate

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Review

Biomechanical Characteristics of the Typically Developing Toddler Gait: A Narrative Review

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Simple Summary: This narrative review clarified the gait biomechanics of typically developing toddlers and revealed the changes of gait characteristics at different age stages till independent walking. The remarkable gait characteristics and developmental nature of toddlers indicate that the gait pattern of the junior independent walkers differs from the senior and experienced cohort. Gait patterns are associated with neuromuscular maturation. Changes in gait biomechanics are age-dependent. Therefore, it is necessary for pediatric clinicians to understand the characteristics and stages of normal or abnormal development. Developmental neuromotor control suggests that early identification and intervention may expedite treatment and optimize outcomes.

Abstract: Independent ambulation is one of the most important motor skills in typically developing toddlers. Gait analysis is a key evaluation method in basic and clinical research. A narrative review on the literature of toddler gait development was conducted following inclusion criteria, explicitly including the factors of English article, age range, no external intervention during the experimental process of studies involved, the non-symptomatic toddler, and no pathological gait. Studies about toddlers' morphological, physiological, and biomechanical aspects at this developmental stage were identified. Remarkable gait characteristics and specific development rules of toddlers at different ages were reported. Changes in gait biomechanics are age and walking experience-dependent. Gait patterns are related to the maturation of the neuro and musculoskeletal systems. This review thus provides critical and theoretical information and the nature of toddler walking development for clinicians and other scientific researchers. Future studies may systematically recruit subjects with more explicit criteria with larger samples for longitudinal studies. A particular design could be conducted to analyze empirically before practical application. Additionally, the influence of external interventions on the development of toddler gait may need consideration for gait development in the toddler cohort.

Keywords: toddler; foot; biomechanics; plantar pressure; gait development

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1. Introduction

Human gait is a motion state of the motor system during walking, which is the process of moving in a particular direction with a series of continuous and coordinative activities of hip, knee, foot, and ankle. In other words, gait refers to a movement pattern of limbs, especially lower limbs on a substrate, which can fulfill the primary need of locomotion and provide propulsion and support for the body [1]. Behind the gait lies several key indications of neuromusculoskeletal growth and development. Toddlers are

in a critical period of independent walking. Therefore, a systematic and comprehensive understanding of toddlers' gait is important. During this stage, toddlers experienced dynamic and progressive changes, such as rapid growth and development of anatomical, neuromuscular, and sensory systems [2], the ossification of bones [3,4], and the appearance of the foot arch structure [4]. Children are considered infants, if less than or equal to 12 months, as toddlers between 13 and 36 months [5].

Clinical examination and locomotor milestones are the mainstream means to identify potential motor impairment in toddlers. However, such assessments present non-specific characteristics, which challenge the evaluation of locomotor milestones. For example, regarding the onset of independent walking, it is reported that this process occurs at the age of 12 months, while 10% of the typically developing toddlers cannot walk independently until 14.4 months or later [6]. Not all alternative gaits are caused by diseases, toddlers also exhibit this feature during walk learning. The finding indicates that toddler gait is influenced by a variety of factors, such as age, walking experience [7], body dimensions [7–9], maturation of central nervous system [9,10], the muscle-fat ratio [11], development of musculoskeletal system [12,13], and head-trunk posture stability [14].

The research of gait has been relatively mature up till now. With the development and improvement of measurement technologies, researchers can carry out dynamic quantitative analyses on the characteristics of human locomotion. It is vital to understand or master the gait differences between typically developing toddlers and adults to describe, study and treat abnormal gaits in toddlers. Several cross-sectional and few longitudinal studies have analyzed toddler gait at the onset of independent ambulation, including spatiotemporal characteristics [15–25], plantar pressure, and kinematics [8,25–34]. Predecessors have done similar studies. For example, Price et al. summarized existing literature quantifying biomechanical characteristics in toddler cruising, supported, and independent walking [35]. The current review is more focused on the independent walking of toddlers since 12–36 months, which is a developmental stage after the transition from sitting to walking.

To describe, study and take immediate treatment of abnormal gait, researchers should master the differences between toddler and adult gait, the time when toddler gait is mature, and the factors influencing gait maturation [12]. Further research, especially more longitudinal studies, may investigate biomechanical characteristics of the typically developing toddler gait. The present review aims to clarify and reveal the changes of gait characteristics of toddlers at different age stages to independent walking. The remarkable gait characteristics (or parameters analyzed), such as plantar pressures, joint motion, moment, and specific developmental nature of toddlers are reported, guiding clinicians, scientific researchers, and even parents in gait evaluation, correction, and development.

2. Method

While collating and reviewing relevant literature, the authors found that the diversity and quantity of literature related to gait development in the typically developing toddlers were sufficient. However, due to individual differences in the growth and development of toddlers, there is no unified age range of toddlers. Meanwhile, the authors aimed to summarize and critique literature, so we chose a narrative literature review rather than a systematic review for this study.

2.1. Information Sources

A comprehensive and reproducible search strategy was performed in the following databases (PubMed, Web of Science, and Google Scholar) from January 2000 to December 2021.

2.2. Search Strategy

The search terms used in each database are as follows, (1) In PubMed, the search string is "(((infant*) OR (child*) OR (toddler*)) AND (gait)) [Title/Abstract]." (2) In Google Scholar, the search string is "(((infant) OR (child) OR (children) OR (toddler gait)) AND (gait)) [Title]." (3) In Web of Science, the search string is "(((infant*) OR (child*) OR (toddler*)) AND ((gait) OR (gait development) OR (developing gait))) [Title]."

2.3. Inclusion and Exclusion Criteria

Reference lists from identified literature were manually searched for completeness by the authors to confirm content relevant to the development of the toddler gait. Studies were included for this review if met the following eligibility criteria: (1) written in English, (2) age range (studies with subjects between 10–36 months were included), (3) no external intervention during the experiment of the study involved (For example, the effects of clothes, shoes and visual environmental distraction), (4) no pathological gait, (5) the non-symptomatic toddler.

Studies were excluded if met the following criteria: (1) citations and patents, (2) review articles, (3) written in non-English, (4) abstract papers without data, (5) other no considered gait characteristics (For example, muscle activation and energy expenditure were excluded).

2.4. Risk of Bias in Individual Studies

The Cochrane Collaboration Risk of Bias Assessment Tool was used to evaluate the risk of bias in individual studies. Two independent authors (Wei Liu and Qichang Mei) evaluated all the included studies, and any disagreements were discussed. An independent arbitrator (Bíró István) was invited when an agreement was not met.

3. Results

The search yielded 3839 titles and abstracts for initial screening. A total of 47 full texts were screened, and 25 were excluded. Twenty-two studies were included for the final analysis. The identification process was illustrated by a flow chart in Figure 1. Based on the information of all the full texts included, the results of data collection and summary measurement of each included study were presented in Tables 1 and 2. The details included authors, study site, participants, age, design, measurement frequency and timing, data collected, anatomic sites, walking speed and style, footwear/attire, instruments, variables.

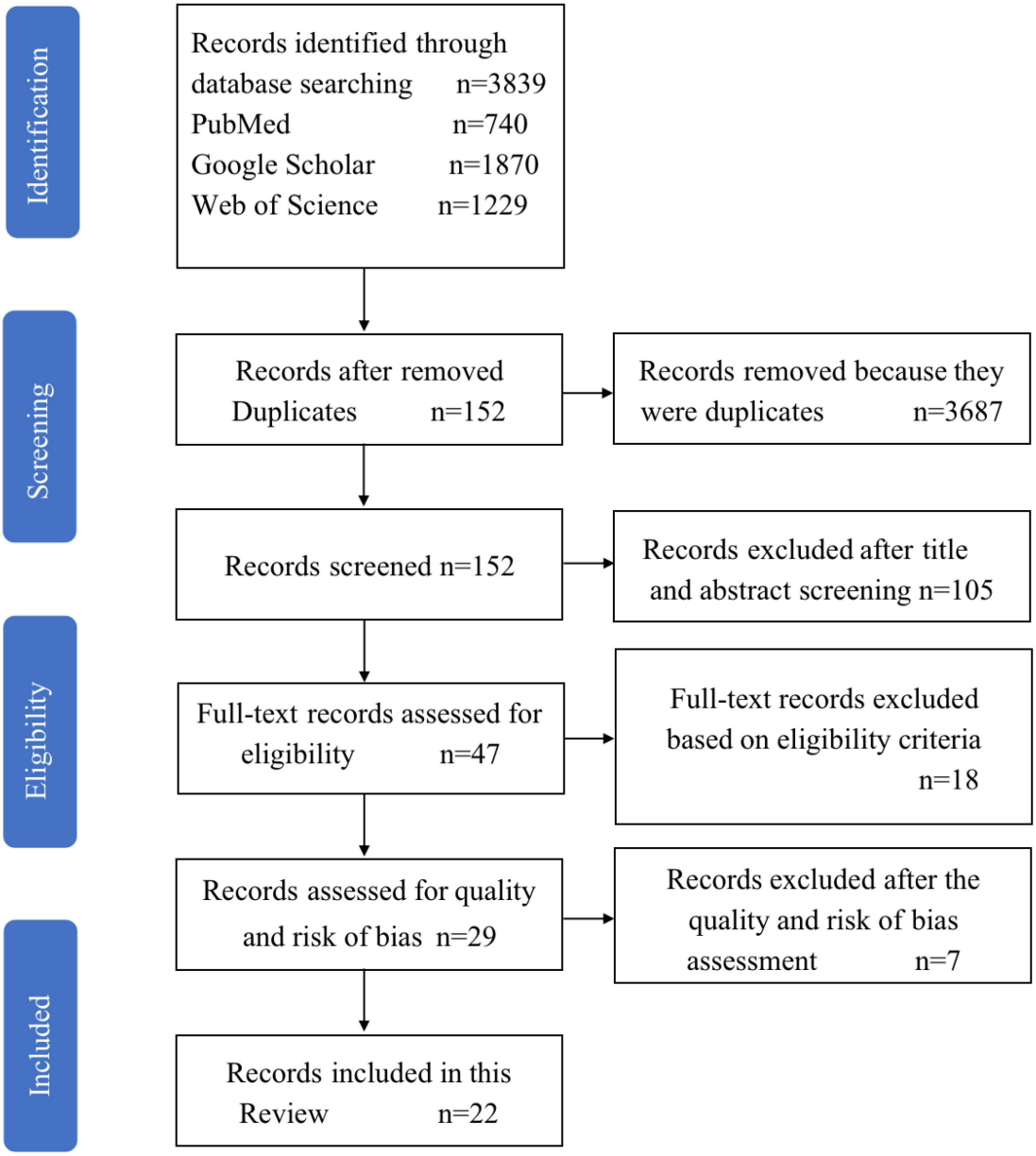

Figure 1. The flow chart of the literature inclusion in this review.

Table 1. Literature relating to spatiotemporal parameters of toddler gait.

	Lee et al. [15]	**Bisi et al. [16]**	**Hallemans et al. [17]**	**Hallemans et al. [18]**	**Van Dam et al. [19]**
Study site	America	Italy	Belgium	Belgium	Belgium
Participants	97	20	10	84	100
Age	10.75 to 19.99 months	10 to 16 months	12.6 ± 1.64 to 17.2 ± 1.64 months	1–10 years (10–36 months included)	15–36 months
Design	Cross-sectional	Longitudinal	Longitudinal	Cross-sectional	Cross-sectional
Measurement frequency and timing	Once: 67 infants. 2 or 3 times: 30 infants.	5 times: 0, 1, 2, 3, 6 months after walking onset.	Every 6 months for 9 measurements scheduled.	Once: tested age ranges from 1 to 10 years.	Once: tested age ranges from 15 to 36 months.
Data Collected	1 walking trial per participant with free play task, 6 consecutive steps per participant with straight-path task.	1 walking trial per participant along the corridor spontaneously.	336 trials of all individuals.	Clear foot strikes on the force plates and full marker visibility for at least 2 consecutive strides.	2 walking trials per participant.
Anatomic sites	Foot.	Trunk and leg.	Full body (focusing on the lower extremities).	Ankle and foot.	Foot.
Walking speed and Style	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.
Footwear/Attire	Not reported	Not reported	Not reported	Bare foot	Soft leather shoes
Instruments	Pressure-sensitive mat (4 sensors/in2). Three video cameras.	Tri-axial wireless inertial sensors. Video camera.	Instrumented walkway surrounded, infrared cameras (Vicon Motion Systems), force platforms (AMTI).	Optometric movement registration system. Force plate.	Strip of paper of 5 m long and 0.84 m wide. Leather shoes with stamps. Ink.
Variables	Speed, step length, and step width.	Stride time, swing time, stance time, cadence, acceleration, regularity.	Vertical ground reaction forces, cadence, walking speed, double support time, stride length and step width, joint kinematics and kinetics.	Spatial margin of stability, walking speed, stride length, step width, swing.	Step-time parameters: step length, step width, cadence and walking speed.

Table 1. *Cont.*

	Guffey et al. [20]	Looper et al. [21]	Marencakova et al. [22]	Adolph et al. [23]	Hamme et al. [24]	Hallemans et al. [25]
Study site	America	America	Czech	America	France	Belgium
Participants	84	8	20	151	106	10
Age	2–4.9 years	6–11 months	7–13 months	11.8–19.3 months	1 to 7 years (10–36 months included)	13.5 to 18.5 months
Design	Cross-sectional	Longitudinal	Cross-sectional	Cross-sectional	Cross-sectional	Cross-sectional
Measurement frequency and timing	Once: 6 age groups split every 6 months.	5 times: 1, 2, 3, 4, 5 months after walking onset.	Once: analyzed age ranges from 7 to 13 months.	Once: tested age ranges from 11.8 to 19.3 months.	several times	Once: tested age ranges from 13.5 to 18.5 months.
Data Collected	3 walking trials per participant.	4 trials of each individual.	1 walking bout consisting of a minimum of 5 complete gait cycles.	1 trials of each individual with free play task and straight-path task.	1 to 6 gait trials per gait analysis.	3–5 trials of each individual.
Anatomic sites	Foot.	Foot.	Leg.	Foot.	Full body (focusing on the lower extremities).	Full body (focusing on the lower extremities).
Walking speed and Style	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.
Footwear / Attire	Bare foot	Not reported	Not reported	Not reported	Not reported	Not reported
Instruments	GAITRite system.	GAITRite system.	Free accessible tool Tracker.	Video camera. GAITRite system.	Instrumented walkway surrounded, infrared cameras (Vicon Motion Systems), force platforms (AMTI).	Instrumented walkway surrounded, infrared cameras (Vicon Motion Systems), force platforms (AMTI).
Variables	Step and stride length, velocity, cadence, step time, cycle time, stance time, swing time, single support time and double support time.	Velocity, cadence, step length, step length and cadence normalized by leg length and single support.	Falls frequency, stops frequency, cadence, time of stance phase, swing phase and double support phase.	Step length, step width, time walking, steps/hour, distance/hour.	Vertical ground reaction forces, angle, moment, power.	Vertical ground reaction forces, cadence, stride time, single support time, double support time, stride length and step width.

Table 2. Literature relating to kinematic and kinetic parameters of toddler gait.

	Zeininger et al. [8]	**Hallemans et al. [17]**	**Hallemans et al. [25]**	**Samson et al. [26]**	**Hu et al. [27]**	**Dulai et al. [28]**
Study site	America	Belgium	Belgium	France	China	Canada
Participants	18	10	10	75	319	102
Age	11.5 to 43.1 months	12.6 ± 1.64 to 17.2 ± 1.64 months	13.5 to 18.5 months	1 to 6 years (10–36 months included)	2–6 years (10–36 months included)	2–17 years (10–36 months included)
Design	Cross-sectional	Longitudinal	Cross-sectional	Longitudinal	Cross-sectional	Cross-sectional
Measurement frequency and timing	Once: tested age ranges from 11.5 to 43.1 months.	Every 6 months for 9 measurements scheduled.	Once: tested age ranges from 13.5 to 18.5 months.	4 times, 2 times and 1 time per year after 1, 2 and more than 3 of independent walking.	Once: tested age ranges from 2 to 6 years.	Once: 5 age groups (2–3, 4–6, 7–10, 11–14, 15–17 years)
Data Collected	4 trials of each individual.	336 trials of all individuals.	3–5 trials of each individual.	6 trials of each foot on force plate.	3 walks for each foot.	3 trials of each individual.
Anatomic sites	Total foot.	Full body (focusing on the lower extremities).	Full body (focusing on the lower extremities).	Ankle, knee and hip joints	Hallux, toes, metatarsal heads, mid-foot, medial and lateral heel.	Hallux, heel, lesser toes, medial and lateral forefoot, midfoot.
Walking speed and Style	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.	Not reported.
Footwear/Attire	Bare feet	Not reported	Not reported	Not reported	Not reported	Bare feet
Instruments	Vicon MX motion analysis system synchronized with Bertec force plates.	Instrumented walkway surrounded, infrared cameras (Vicon Motion Systems), force platforms (AMTI).	Instrumented walkway surrounded, infrared cameras (Vicon Motion Systems), force platforms (AMTI).	Motion Analysiss system with Eagles cameras, force platform, Footscans plantar pressure platform.	RSscan (4 sensors cm^2).	emed-x platform (4 sensors cm^2).
Dynamic variables	Location of the center of pressure relative to the calcaneus, the orientation and magnitude of ground reaction forces during foot contact.	Vertical ground reaction forces, cadence, walking speed, double support time, stride length and step width, joint kinematics and kinetics.	Vertical ground reaction forces, cadence, stride time, single support time, double support time, stride length and step width.	Moment, speed.	Relative force-time integral (FTIrel) (%).	Foot impulse, regional percent impulses, impulse ratios.

Table 2. *Cont.*

	Bosch et al. [29]	**Cowgill et al. [30]**	**Samson et al. [31]**	**Hallemans et al. [32]**	**Ivanenko et al. [33]**	**Ivanenko et al. [34]**
Study site	Germany	America	France	Belgium	Saint Lucia	Saint Lucia
Participants	62	10	42	9	26	7
Age	15.1 ± 2.4 to 63.2 ± 2.4 months	1 to 3.9 years	1 to 6 years (10–36 months included)	12 to 18 months	11 to 153 months (10–36 months included)	12 to 15 months
Design	Longitudinal	Cross-sectional	Cross-sectional	Cross-sectional	Cross-sectional	Cross-sectional
Measurement frequency and timing	Every 6 months for 9 measurements scheduled.	Once: tested age ranges from 1 to 3.9 years.	Once: tested age ranges from 1 to 6 years.	Once: tested age ranges from 12 to 18 months.	Once: tested age ranges from 13.5 to 18.5 months.	Once: tested age ranges from 12 to 15 months.
Data Collected	5 trials of each individual.	1 trail of 1 foot on force plate and speed changed less than 20%.	1 trials of each foot on force plate.	5 trials of each individual.	10 trials of each individual.	10 trials of each individual.
Anatomic sites	Total foot.	Total foot, greater trochanter, lateral femoral condyle and lateral malleolus.	Total foot, metatarsophalangeal and ankle joint.	Full body.	Trunk, pelvis, thigh, shank, and foot.	Trunk, pelvis, thigh, shank, and foot.
Walking speed and Style	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.	Self-selected speed.
Footwear/Attire	Bare feet	Bare feet	Not reported	Not reported	Not reported	Not reported
Instruments	capacitive pressure distribution platform (4 sensors cm^2).	infrared cameras (Vicon MX4) with skin marker attachment and embedded force plate (AMTI).	Motion Analysiss system with Eagles cameras, force platform, Footscans plantar pressure platform.	Instrumented walkway surrounded, infrared cameras (Vicon Motion Systems), force platforms (AMTI).	ELITE or VICON motion analysis systems, force platform.	VICON motion analysis systems, force platform.
Dynamic variables	Contact area, peak pressure, force-time integral, relative maximum force, contact time and the force-time integral.	Peak ground reaction forces and ground reaction forces impulse.	Vertical ground reaction forces, moment.	Vertical ground reaction forces, speed, moment.	Vertical ground reaction forces, speed, moment.	Vertical ground reaction forces, speed, step length and width.

4. Discussion

4.1. Spatio-Temporal Characteristics of Typically Developing Toddler Gait

Toddlers develop rapidly in structure and function as growth. Expression of alternative gait patterns might require neuromuscular maturation and learning time during the period of independent walking [36]. As an essential supporting structure, the toddlers' foot has experienced dynamic and gradual changes, such as the bone and arch structures [4]. These alterations led to apparent differences in toddlers' locomotion strategies. Changes in gait biomechanics were age-dependent, and the gait parameters, such as stride length, step width, and duration of swing, vary with age [12,17,20,25,37–41]. The changes in these parameters were closely related to balance, coordination, and metabolic cost [3,5–7,9,16,42–47].

4.1.1. Temporal Parameters of the Toddler Gait

While walking independently for about two months, the temporal parameters showed developmental changes, i.e., stride time, stance time (stance time as a percentage of stride time), cadence, and normalized cadence. These parameters presented a turning point two months after toddlers walked independently (decreasing from the first week of independent walking (T0) to two-month after independent walking (T2) and increasing from T2 to six-month after independent walking (T6)) [16]. Time gait cycle parameters increased with age, while cadence decreased [12,37,38]. At the same time, swing time as a percentage of stride time increased from a value of 40–50% at T0–T2 and then remained constant [16]. Novice and experienced walkers spent 42.5% and 33.9% of the stance phase in the double support [22]. This finding was slightly lower than the result obtained by Clark et al. that double support time of junior independent walkers was from 20% (adult referral value) to 60% [47]. The study [22] observed toddlers aged 7–13 months in the non-laboratory environment (Inclusion criteria for the novice group, hands in high-guard position, uncertainty during walking, a lack of stability and control), and it could be the reason why the results were lower. The temporal characteristics of immature gait patterns were a broad base of support, prolonged stance duration [12,39], and increased double support [12,25,39]. The changing trend of the above parameters showed a gradual decline close to the adult standard, suggesting the transition process of toddler gait becoming gradually matured [25].

As the walking experience increased, toddlers acquired the balance strategy. During this stage, the walking cadence and speed increased [15], but faster-walking speed was due to the high cadence rather than step length [21]. However, Bisi et al. [16] and Bril et al. [48] considered that increase in walking speed was due to the long step length rather than cadence. Sutherland et al. thought that increased leg length without an increase in cadence would increase the walking speed with a stable walking pattern [12]. The differences in the above perspectives might lie in the results between different sample sizes and ages. A more extensive study might better understand the relationship between step cadence and speed in early walking development [21]. A large-scale biomechanical gait parameters database of healthy junior children (including toddlers aged 1 to 3) have been built by Hamme et al. The databased presented an original regression of parameters with age, walking speed, and the age–speed interaction and deduced the typical reference targets from regressions [24].In any case, the growth (faster and longer) of steps was a sign of a more mature gait in toddlers [17,19,49].

Different views on the relationship between gait cadence, age, and walking experience were reported, for example. Gait cadence increased significantly with age and walking experience [17,21,22]. Owen et al. denoted that cadence showed a slight downward trend with the increase of age [12,37,38]. Guffey et al. showed that standard cadence decreased by 15% from 2 to 4 years [20]. However, Bisi and Stagni presented that cadence increased first and decreased from novice walkers to stable walkers [16]. Current studies agreed that the changing nature of gait cadence could be mainly divided into several stages. At the beginning of walking (3–6 months after the onset of independent walking), toddlers concentrated on overcoming gravity to maintain postural gait requirements, gradually

to the fine-tune gait patterns [16,17,21,49]. In general, a relatively high gait cadence was observed at the beginning of walking. When toddlers slowly grasped the skill of gravity, the gait cadence decreased gradually and eventually remained stable after adulthood [22].

4.1.2. Spatial Parameters of the Toddler Gait

At the initiation of toddlers' independent walking, a toddler strategy was employed. According to the findings of Bisi and Stagni, toddlers have determined the different gait strategies in the first month of independent walking [16]. Whitall and Getchell believed that individual walking and running techniques appeared after 9.5 months of independent walking [50]. The spatiotemporal characteristics of gait strategies changed the loading mode of feet, mainly manifested in the increase of step frequency (or cadence) to keep balance [21,23,51]. At the onset of independent walking, toddlers who took relatively wider steps, longer steps, and a shorter swing duration had a large normalized spatial stability margin. However, with the increase of age, the spatial margin of stability would gradually decrease [18]. The mid-lateral distance was significant [15], and step width exceeded step length. Over the next few months, toddler walking improved significantly, and step width diminished while walking speed and step length increased gradually [12,15,17,21,23,51]. Smaller step width provided stability and narrowed as the balance improved [25,48,52–54], which showed the trend of mature gaits close to adulthood [51].

In addition, step and stride length increased with age, and there were significant differences between step and stride length and age groups [20]. Toddlers walked slowly between 10 and 15 months, with steps shorter than leg length [17,40]. A few months later, speed and step length dramatically increased [17], and occasionally, steps larger than leg length could be observed [40]. Dusing and Thorpe have also proved that normalized step length and velocity increased from 1 to 4 years old [37]. However, Moe-Nilssen et al. measured the movement of lower limbs through a triaxial accelerometer and demonstrated that stride regularity and step regularity had a low frequency in the toddler gait [55]. Bisi et al. also found no significant increase in the regularity [16]. Perhaps a more comprehensive longitudinal study should focus on how these parameters would change during gait maturation in the future. The data obtained by Guffey et al. also demonstrated that the difference of normalized spatial-temporal parameters in different age groups was not statistically significant [20]. Other normalization methods might cause the difference mentioned above. Dusing et al. utilized the height [37], and Guffey et al. believed that leg length was more suitable for normalizing the spatial dimensions [20], causing the proportion between the leg and torso of toddlers to change gradually. With the continuous maturity of gait, toddlers' independent walking stability increased continuously. However, normalized gait parameters were mature after three years old [18].

To adapt to the maturation of locomotor patterns, i.e., walking and running, toddlers needed to constantly alter the interlimb coordination [42–46,56,57]. Hallemans et al. suggested an early gait maturation after four months of independent walking [25]. Bril and Breniere confirmed that toddlers showed developmental changes with 5–6 months of independent walking experience [48]. Van Dam et al. indicated a relation between morphology (the head and pelvis) and the step-time parameters of gait in toddlers between 15 and 36 months [19]. Unlike standard laboratory environments, non-laboratory investigations (an open-sourced toddler gait video analysis) also described how toddlers walk in familiar settings. With the increase of walking experience, the frequency of toddler falling gradually decreased [22]. A study compared the traditional straight-path task with spontaneous walking in 97 toddler gait characteristics, which correlated highly with each other. The free-play task benefited understanding improvements in walking to control balance and forces. Meanwhile, gait characteristics during spontaneous walking had implications for studying the development of walking in toddlers with impairments [15].

4.2. Kinematic Characteristics of Typically Developing Toddler Gait

Special attention was paid to the foot kinematics while collating and reviewing literature related to the development of gait in toddlers. Whether in mechanical energy or kinematics, the ability to walk slowly developed from initial independent steps to about seven years old [33,34,49,58,59]. Toddlers needed to constantly change the interlimb coordination, which was a slow development process [36]. The gait of toddlers aged 10–15 months was characterized by high variability and low stability [60]. Walking independently for about one month, toddlers started to show the characteristics of the pendulum mechanism [16,33].

Zeininger et al. reported that the initial heel contact steppers went through lower vertical forces at impact, indicating the absorption of peak force by knee yielding or the transition to the heel-toe contact pattern [8]. Knee yielding followed heel strike [17], similar to adults [61], further illustrated that the initial heel contact mode was the transition to adult gait. Flat foot contact steppers bore higher vertical forces and appeared the rapid downward trajectory of the foot and leg, showing a less yielding gait [8]. A conclusion was supported by Hallemans et al., suggesting that the knee did not yield or absorb energy between two weeks and five months of walking experience for toddlers [25]. There were no significant differences in ankle angle between the initial heel contact and flat foot contact [8].

In addition, age had a significant influence during the pre-swing phase, according to the study of Samson et al. [31]. With an increase in age, knee flexion decreased [26]. The maximum flexion of the metatarsophalangeal joint increased with age. However, there was no significant difference between different age groups, indicating that metatarsophalangeal joints were more passive and matured faster. There was also no significant difference in dorsiflexion/plantarflexion and inversion/eversion between different groups [31]. Hallemans et al. observed slight plantar flexion movement of the ankle was likely to be a passive movement, possibly coming from gravity on foot segment in toddlers (aged 13.5 to 18.5 months) [25]. Another follow-up longitudinal study by Hallemans et al. observed that ankle plantarflexion at foot contact and maximal hip extension in stance increased with increasing walking experience [17]. By comparing kinematics and EMGs in toddlers (aged 12 to 15 months) at the onset of independent walking with or without hand or trunk support, Ivanenko et al. [34] indicated that immaturity of global gait parameters did not depend on postural stability. Even with or without support, toddlers still exhibited a characteristic gait pattern until the occurrence of the first unsupported steps and rapidly matured thereafter.

4.3. Kinetic Characteristics of Typically Developing Toddler Gait

4.3.1. Ground Reaction Force Related Parameters of the Toddler Gait

Previous studies reported that toddlers transited from the early flat foot contact to the initial heel contact and then developed towards adult gait patterns [17,62–66]. This alteration was a typical mature contact pattern in the process of human gait. Consistent initial heel contact usually occurred at 12 months after independent walking [39]. This view was also supported by Sutherland et al. [12] and Hu et al. [27], suggesting that the heel-strike model did not appear until two years old.

Under a laboratory setting, the ground reaction force of toddler gait during walking was examined by analyzing the kinetic data of 18 toddlers (aged 11.5–43.1 months) [8]. The initial heel contact and flat foot contact steps differed significantly in the location of central pressure relative to the calcaneus. Toddlers with flat feet at touchdown showed the characteristic of wide heel in the morphology of feet and less walking experience. The morphological changes would assist in reducing the heel loading at the onset of toddler independent walking and frequently shift the center of pressure in front of the heel [8,67,68], showing an evenly distributed plantar loading. Compared with the pattern of flat foot contact, the toddlers who landed with initial heel contact were more dominant in age, weight, leg length, and walking experience. The ratio of pressure under the heel was higher, and the periods of heel loading lasted longer than in adults [8,62–65]. Therefore, the

load was concentrated on the anterior calcaneus and a narrower heel, suggesting the need for increased calcaneal robusticity during development to mitigate injury. The load was mainly focused on the anterior calcaneus and narrow heels [8], illustrating the distribution characteristics of the foot loading of toddlers.

Meanwhile, Samson et al. compared different age groups of the ground reaction force, ankle joint, and the metatarsophalangeal joint [31]. As observed in the research of Hallemans et al. [17] and Sutherland [12], the second peak was almost non-existent in vertical force at pre-swing [31] and increased with age [31,69,70]. With increasing walking experience, ground reaction force patterns evolved towards a double-peak (impact and active peak) [17]. Toddlers' data about the maxima of resistance and propulsion configurations were statistically smaller than adults [31], which indicated that toddlers had an alternative strategy of mainly ankle stabilization and hip propulsion [71]. Biomechanical maturation of joint dynamics occurred approximately age four for the ankle [26].

A large number of studies have shown that in addition to shock absorption as a cushion, the arch also played a vital role in the dispersion and transmission of force in the development process of heel-toe contact mode [4,27,72–75]. A study found through force transfer algorithm [27] that the pressure and contact area in the middle forefoot increased with toddler development. Strong forefoot support was also found [27,73] and showed the performance of the transverse forefoot arch. Moreover, it was proved that the transverse arch was formed in the early stage (2 years old) by the transfer rule of the load on the forefoot between 2 to 5 years old toddlers. The maximum medial/lateral force decreased with age at early stance, which significantly differed from adults [31]. It was probably due to a decrease in stride width [17]. The windlass mechanism was mature after three years old, causing the load transfer from the middle forefoot to the medial and lateral forefoot. Relevant research proved that the foot can support or transfer loads in the anterior-posterior and media-lateral directions before 6 years old [27]. Thus, the transverse and longitudinal arch appeared in early toddlerhood. The arch promoted force transfer and played an aspirator's role in the windlass mechanism during walking.

4.3.2. Plantar Pressures of the Toddler Gait

The middle foot area was the main loading area for toddlers to bear body mass [74]. Dulai et al. reported that the impulse in the middle foot area was the highest in the toddler group (2–3 years old) and through the medial forefoot was correspondingly lower [28]. From the perspective of toddler foot morphology and anatomical nature at the onset of independent walking, several studies indicated that the midfoot was full of the fat pad, which was a structure that can effectively relieve the pressure increase caused by weight gain before foot arch maturity [72,75–78]. Although the arch of toddlers was in the process of continuous development, the middle foot of toddler-aged two has been the transition area connecting the fore and hindfoot [27]. The mid-foot impulse decreased gradually as age advances [27,28], corresponding to the clinical observation of flat feet in toddlers [79].

A longitudinal study on the gait symmetry in typically developing toddlers existed, reporting that the total foot contact area presented symmetry [29]. However, spatiotemporal gait parameters had a certain degree of asymmetry [29,80]. One of the leading causes of foot deformity was caused by asymmetric contact area. In typically developing toddlers aged up to three years old, foot loading patterns might show asymmetric characteristics and become symmetrical with the increase of age and walking experience [29,81]. Joint dynamics were influenced by age during the early childhood [26]. The metatarsophalangeal and ankle joints showed that the maximum eversion moment decreased with age. It was also probable that favoring stability in toddlers [31]. The inverted pendulum mechanism started to mature after three months of walking experience. To minimize energy expenditure, toddlers (at least partially) may use the inverted pendulum mechanism of the energy exchange [32,33], which may not be perfect because of slow walking speed, tossing gait, and smaller kinetic energy fluctuations than potential energy fluctuations. The percentage of mechanical energy recovery increased with walking experience and decreased the

gradual variability of kinematic and kinetic parameters [33]. Since each toddler's speed at which gait matures would be different. Thus, a longitudinal study may be more appropriate to investigate subtle changes and growth [32]. Hallemans et al. [25] reported the kinetics feature of immature gait in toddlers. The dominance of the hip and knee extending moments throughout stance, together with a sustained power production observed around these joints. These findings were supported by the previous reporting [54,82]. Another one was the reduced complexity of the joints (hip, knee, ankle) moment profiles in toddlers, likely caused by immature walking control. The relative rounder shafts of toddlers may be viewed as an early functional adaptation to the unusual demands of the "waddling" locomotion [30].

5. Limitations and Future Research

Several other limitations should be considered. The selected kinetic data were limited to the foot and ankle sections. The rotational profile of the lower limbs was not documented. The potential impact of knee joint and hip joint during walking on foot loading symmetry was not included. The laboratory environment could not represent real-world toddler activity. Pressure data collected by walking in a straight line had limitations [35], which reduced gait variability, therefore masking the differences between developmental stages of natural gait [22]. The present results have a particular value, primarily as normative foot loading data, and provide information on the development of foot loading symmetry four years after independent walking. Future studies concerning the typically developing toddler gait may need attention to address a few issues. A particular design may be conducted to analyze empirically before practical application. Research shall systematically recruit subjects with larger samples for a longitudinal study. Anthropometrically and anatomically matched musculoskeletal models may be developed to further decipher the neuro-musculoskeletal biomechanics in the toddler gait [83]. Current techniques, such as the wearables [84] and advanced statistical analysis [85], may also be employed to reveal gait biomechanics. Additionally, the influence of various external environmental interventions on the development of toddler gait also has research value, for example, clothing, footwear, weight-bearing, visual environment interference.

6. Conclusions

The study reviewed the biomechanical characteristics of toddler gait at different age stages to independent walking, covering the spatiotemporal parameters, kinematics, and kinetics. The remarkable gait characteristics and typical development nature were reported, indicating that the gait patterns of junior independent walkers differed from senior and experienced cohorts. Gait patterns were associated with the maturation of the neuro and musculoskeletal systems. Changes in gait biomechanics were age and walking experience-dependent. The longitudinal arch played a vital role in the dispersion and transmission of force in developing heel-toe contact. Therefore, it is necessary for pediatric clinicians to understand the characteristics and stages of normal or abnormal development. Knowledge may provide practical implications and healthy references for the diagnosis of gait disorders.

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children

MDPI

Review

Kinematic Gait Impairments in Children with Clubfeet Treated by the Ponseti Method: A Systematic Review and Meta-Analysis

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Abstract: Background: Being aware of possible gait impairments in Ponseti-treated clubfoot children might be useful for optimizing initial and additional treatment. Therefore, this systematic review and meta-analysis aimed to identify kinematic gait abnormalities in children with clubfoot treated with the Ponseti method (with and without relapse). Methods: A systematic search was conducted. Studies comparing kinematic gait parameters of Ponseti-treated clubfoot children to healthy controls were included. Meta-analyses and qualitative analyses were conducted on the extracted data. Results: Twenty studies were identified. Twelve of the 153 reported kinematic outcome measures could be included in the meta-analysis. Plantarflexion at push-off, maximum ankle dorsiflexion during the swing, maximal plantarflexion, and ankle range of motion was significantly lower in Ponseti-treated clubfoot children. Ponseti-treated clubfoot children showed more internal foot progression. Qualitative analysis revealed 51 parameters in which pre-treatment relapse clubfeet deviated from healthy controls. Conclusions: Ponseti-treated clubfoot children showed several kinematic gait differences from healthy controls. In future studies, homogeneity in measured variables and study population and implementation of multi-segmental foot models will aid in comparing studies and understanding clubfoot complexity and treatment outcomes. The question remains as to what functional problems gait impairments lead to and whether additional treatment could address these problems.

Keywords: congenital talipes equinovarus; gait analysis; functional evaluation; relapse; multi-segment foot model

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1. Introduction

Worldwide approximately 100,000 children are born with unilateral or bilateral clubfoot (*talipes equinovarus*) yearly [1–3]. This deformity of the foot involves the *equinus*, *varus*, *cavus*, and *adductus* [4]. Left untreated, clubfoot leads to deformity, functional disability, and pain [5]. The treatment of this condition aims to achieve a normal-appearing, functional, and painless foot [6]. Nowadays, the Ponseti method is the gold standard for the initial treatment [5,7]. The Ponseti method consists of serial manipulations and casting combined with an Achilles tenotomy. The casting phase is followed by a brace period up to the age of 4 years to prevent relapses during early life [4,5].

Despite the effects of good initial treatment, reported relapse percentages following treatment with the Ponseti method range from 1.9% up to 67.3% [8–10]. The prevention

and treatment of a relapse clubfoot are one of the great challenges in clubfoot care. Strict adherence to the Ponseti method, good brace compliance, and frequent clinical follow-up visits are important aspects of preventing relapse [11]. Although a clear definition is lacking, the common consensus is that a relapsed clubfoot requires additional treatment following initial correction [8]. This treatment may vary from repeated Ponseti casting to Tibialis Anterior Tendon Transfer (TATT) and a la carte salvage procedures such as anterior distal tibial epiphysiodesis [11,12].

Besides the occurrence of relapse also, the functional status of the patient is of interest. Functioning in children can be captured using the International Classification of Functioning, Disability, and Health for Children and Youth (ICF-CY) [13]. The ICF-CY contains three main aspects which affect a child's functioning: (1) body structures and function, (2) activities, and (3) participation. Although these aspects together are considered to give a complete overview of the functioning of children, most research on outcomes of treatment in clubfoot patients focuses on body structures and function [13,14]. Extensive 3D gait analysis is a frequently applied tool to evaluate body structures and function, as part of the ICF, in the treatment outcomes [15] and to detect early signs of relapse [11].

With 3D gait analyses, objective kinematic and kinetic parameters of clubfoot patients can be derived [16–18]. Ponseti-treated clubfoot patients previously showed impairments in kinetic outcome measures, such as ankle plantar flexor moment and ankle power [19]. These kinetic outcomes depend on a child's movement pattern, including joint angles. Hence, in order to establish whether a fully functional foot is achieved after initial treatment with the Ponseti method, kinematic parameters are also of interest. In the past few years, an increasing number of studies regarding gait kinematics in children with Ponseti-treated clubfeet have been published. A systematic overview of the reported gait deviations in various clubfoot populations provides insights into the functional outcome of the Ponseti method. Being aware of possible gait impairment is potentially useful for optimizing the Ponseti method, the detection of relapse clubfoot, and developing additional (physio)therapy or surgical treatment [20]. Therefore, this systematic review and meta-analysis aimed to identify kinematic gait abnormalities in children with clubfoot treated with the Ponseti method (with and without relapse).

2. Materials and Methods

2.1. Protocol and Registration

The protocol for this review was registered in the prospective international register of systematic reviews: PROSPERO number CRD42022375837. The Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRIMA) guidelines 2020 were applied while conducting and reporting this systematic review [21–23].

2.2. Eligibility Criteria

Articles should be published in peer-reviewed journals in English, Dutch, or German. Studies comparing kinematic gait parameters of children with clubfoot treated with Ponseti to healthy controls were included. Studies describing the result of 3D gait analyses as an outcome of the Ponseti treatment as well as 3D gait analyses pre-relapse treatment, were considered. A minimum of 5 participants per group was set, and a 3D recording system for gait analysis was required. Cross-sectional, retrospective, and prospective follow-up studies were eligible, and book chapters, conference abstracts, and reviews were excluded. Furthermore, studies using only pedobarography or electromyography to determine gait parameters were excluded.

2.3. Literature Search

A literature search was conducted in the Embase, Medline Ovid, Web of Science, Scopus, Cochrane, Cinahl Ebsco, and Google Scholar databases by an experienced information specialist on 3 October 2022. Search terms included synonyms of clubfoot, gait analysis, and

specific clubfoot treatments, such as Ponseti (Appendix A). Duplicates were removed. In addition, reference lists of related articles were checked for additional relevant references.

2.4. Study Selection Procedure

A systematical selection of articles was made independently by two of the three researchers involved in this phase (MS, LO, and LG). Titles and abstracts of the obtained articles were screened on relevance with a focus on gait analysis in children with club feet. After this first selection, full texts were examined on content and relevance by two researchers (MS, LO, and LG). The absence of consensus on eligibility was resolved by a discussion between the researchers.

2.5. Data Extraction

Data were extracted by one researcher (LO or LG) with the use of a data extraction form. The accuracy of the data extraction was verified by a second researcher (LG or MS). Study characteristics and kinematic outcome measures were extracted with respect to the segment (foot, ankle, etc.), the moment during the gait cycle (stance, gait, terminal stance, etc.), the actual outcome, and whether there was a significant difference between clubfoot patients and healthy controls and the type of clubfoot population (clubfoot without relapse, clubfoot with relapse for which additional treatment was planned or overcorrected clubfoot). In case of lack of clarity, authors were contacted via email for additional information.

2.6. Risk of Bias Assessment

Individual examination of the risk of bias was performed for each study separately and performed by two researchers (MR and BV or MS and LG). The Dutch checklist for prognosis (Cochrane Netherlands) was applied with modifications to the items set to the relevance of the current study objectives (Appendix B). Items focused on the selection of participants, comparability of groups, description of groups, and a validated and blinded measurement of outcome. Items could be scored with 'low risk' (+), 'high risk' (−), or 'unclear' (?). The individual forms were compared and discussed for final consensus.

2.7. Data Synthesis and Analysis

Meta-analyses were performed for outcome measures that were reported with mean and standard deviation by at least three studies and gathered in the same clubfoot population (clubfoot without relapse, pre-treatment relapse, or overcorrected clubfoot). All meta-analyses were performed using Review Manager (RevMan 5.4.1) (Copenhagen, The Cochrane Collaboration, 2020). Kinematic outcome measures, which were presented separately for unilateral clubfoot and bilateral clubfoot, were merged using the RevMan Calculator and were considered as one group in this review and meta-analyses. The consistency of results was estimated with I^2 statistics. In cases of no significant statistical heterogeneity, the fixed effects model was used. The random effects model was used in statistical heterogeneity cases ($I^2 > 50\%$ and $p < 0.05$). If outcome measures were discussed in two or fewer studies, they were compared in a descriptive manner.

3. Results

3.1. Study Selection and Characteristics

Initially, the search strategy provided 1194 unique articles. After screening articles for inclusion and exclusion criteria, 20 studies met the criteria [24–43]. Articles were mainly excluded since the described clubfoot cohort was not treated with the Ponseti method, and no kinematic outcomes were reported (Figure 1).

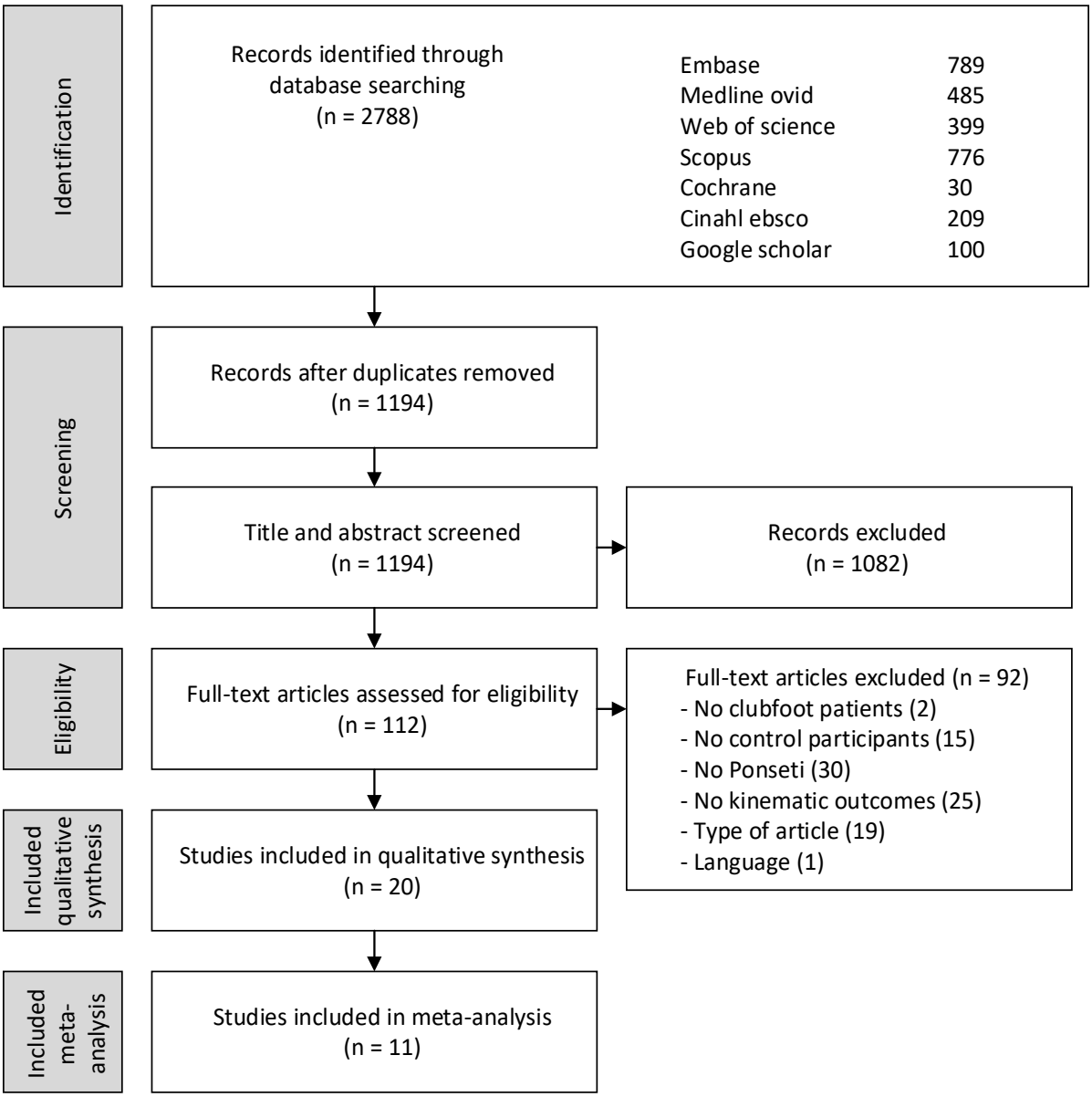


Figure 1. Flowchart selection procedure.

Fifteen studies focused on kinematic outcomes after treatment with the Ponseti method [24,25,27,30,32–34,36–43], seven studies presented data from clubfoot patients prior to additional treatment for relapse [28–31,33–35], and one study described 3D gait analysis performed on overcorrected clubfoot [26]. Since the overcorrected clubfoot is a single specific group, the results of this study are presented in the Appendix C (Table A2). In 16 studies, children walked at a self-selected speed [24,26–31,33–36,38,40–43]. In the other four included studies, no information on walking speed was provided [25,32,37,39]. An overview of the study and participant characteristics of the included studies is shown in Table 1.

Table 1. Study characteristics for each included study (only characteristics concerning Ponseti vs. controls are provided).

Study	Treatment	N (feet)	Gender	Mean Age in Years (Range) or ± SD	N TATT	N Additional Treatment	Marker Position	Dimeglio Scale [1]
Karol 2009 [24]	Ponseti	- (34 feet)	-	5	[2]		-	12.8 (10–15) [a]
	Control	- (17)	-	5				
Church 2012 [25]	Ponseti	22 (35 feet) [3]	9M	6.3 ± 1.4 (5.0–10.0)	1 subject		Multi-segment foot model and single-segment marker set	4.0 (3.0) [b]
	Control	34	-	- (4.0–17.0)				
Duffy 2012 [36]	Ponseti	29 (42 feet)	20M	6.5 (5.0–8.0)	14 feet	4 subjects	-	-
	Control	26 (50 feet)	17M	7.9 (5.2–10.8)				
Smith 2014 [37]	Ponseti	18 (29 feet)	9M	29.2 ± 5.6	10 feet	6 feet	Milwaukee foot model	-
	Control	48	29M	23.2 ± 2.4				
Mindler 2014 [38]	Ponseti	32 (50 feet)	22M	6.0 (3.0–8.0)	5 feet		Cleveland model and Oxford foot model	-
	Control	15 (30 feet)	9M	6.0 (3.0–9.0)				
Manousaki 2016 [39]	Ponseti	20 (30 feet)	17M	7 ± 3.4 months	3 feet	3 feet	Plug in gait model including seven markers on the torso	11 (9–13) [c]
	Control	16		8.5 (6.1–12) [4]				
Lööf 2016 [40]	Ponseti	59 (89 feet)	41M	5.4 ± 0.5	3 feet		Plug in gait model	16 moderated, 48 severed, 24 very severed [d]
	Control	28 (56 feet)	18M	5.5 ± 0.6				
Jeans 2018 [41]	Ponseti	50 (75 feet)	-	10			Plug in gait model	13.4 ± 1.9
	Control	20 (40 feet)	-	10				
Manousaki 2019 [42]	Ponseti	20 (20 feet)	17M	7 ± 3.4 months		3 feet	Plug in gait model	-
	Control	16 (32 feet)		8.5 (6.1–12.0) [4]				
Loof 2019 [43]	Ponseti	47 (69 feet)	35M	5.4 (0.5)	3 feet		Plug in gait model	15 moderated, 36 severed 17 very severed [d]
	Control	28 (56 feet)	18M	5.5 (0.6)				
Dussa 2020 [26]	Ponseti overcorrected	14	-	9.9 (1.5)			Plug in gait model and Oxford foot model	-
	Control	25	-	9.9 (2.7)				
Ferrando 2020 [27]	Ponseti	22 (34 feet)	14M	8 ± 1	11 feet	5 feet	-	-
	Control	25 (50 feet)	18M	9 ± 2				
McCahill 2020 [28]	Ponseti relapse	31	24M	8.3 (5–16)	10 subjects		Oxford foot model	-
	Control	30	21M	10.7 (5–16)				
Mindler 2020 [29]	Ponseti relapse	17 (25 feet)	11M	6.8 (5.1–9.1)			Cleveland model and Oxford foot model	-
	Control	18 (36 feet)	6M	6 (4–9)				

Table 1. *Cont.*

Study	Treatment	N (feet)	Gender	Mean Age in Years (Range) or ± SD	N TATT	N Additional Treatment	Marker Position	Dimeglio Scale [1]
Grin 2021 [30]	Ponseti	11	9M	5.6 ± 1.6			Extended Helen Hayes and Oxford foot model	-
	Ponseti relapse	11	8M	5.7 ± 1.5				
	Control	15	8M	5.7 ± 1.4				
Li 2021 [31]	Ponseti relapse	17 (24 feet)	12M	6.34 ± 1.65 (4.47–10.2)			Helen Hayes model	-
	Control	16	M:F = 1.14:1	7.12 ± 2.23				
Recordon 2021 [32]	Ponseti	16 (23 feet)	-	15 (13–17)	5	5	-	5.8 ± 1.7
	Control	39 (78 feet)	-	Age-matched				
Brierty 2022 [35]	Ponseti relapse	16 (23 feet)	13M	5.58 (3.27–8.57)			Plug in gait model and Oxford foot model	-
	Control	9	-	6.31 (4.47–7.96)				
Grin 2022 [33]	Ponseti	18	18M	5.39 ± 1.46			Extended Helen Hayes and Oxford foot model	-
	Ponseti relapse	13	8M	5.46 ± 1.51				
	Control	21	12M	6 ± 1.57				
Wijnands 2022 [34]	Ponseti	15	12M	5.13 ± 1.25			Extended Helen Hayes and Oxford foot model	-
	Ponseti relapse	10	6M	5.70 ± 1.57				
	Control	19	11M	5.79 ± 1.40				

[1] Dimeglio scale: classification on a scale of 0–20 based on eight items, divided into four grades (benign, moderate, severe, very severe) [44], [2] included children with a tibialis anterior tendon transfer (TATT) as part of the Ponseti method but did not report the number of feet included. [3] multi-segment foot model data are only available for 23 of 35 involved feet. [4] median instead of mean. [5] included eight subjects with additional treatment but did not report which treatment. [a] mean (range). [b] median (interquartile distance). [c] medial (range). [d] number of feet with a moderate (5–10), severe (11–15), or very severe (16–20) score on the Dimeglio scale, a total of 88 feet has been scaled in Lööf 2016 and 68 feet in Lööf 2019. - no information was provided.

A large diversity of outcome measures was presented in the different studies (addressed in Sections 3.3–3.5). Twelve parameters described in eleven studies could be included in the meta-analyses. Lööf et al. (2016) made a clear distinction between unilateral clubfoot and bilateral clubfoot and compared them to the same group of healthy controls. This violates the assumptions of independence of observation that underpin the meta-analyses. Therefore, kinematic outcomes presented in Lööf et al. (2016) for uni- and bilateral clubfoot were merged using the RevMan Calculator and were considered as one group in this review and meta-analyses.

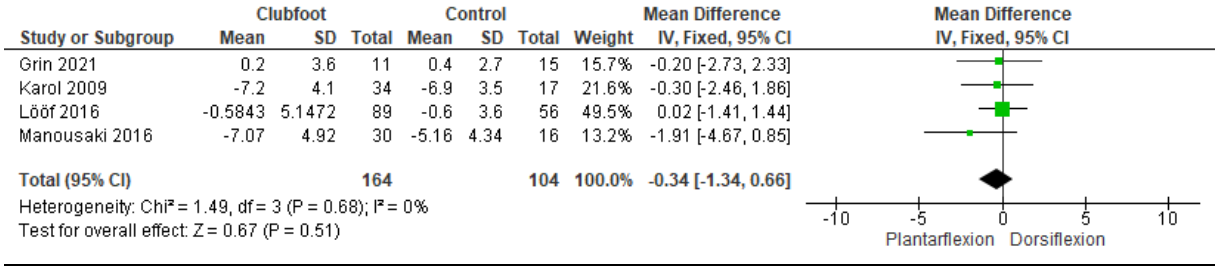
3.2. Risk of Bias Assessment

The risk of bias assessment for each study separately showed the unclear or high risk of bias for one or more items (Appendix B, Table A1). This was mostly due to a lack of information or no information at all presented in the included articles

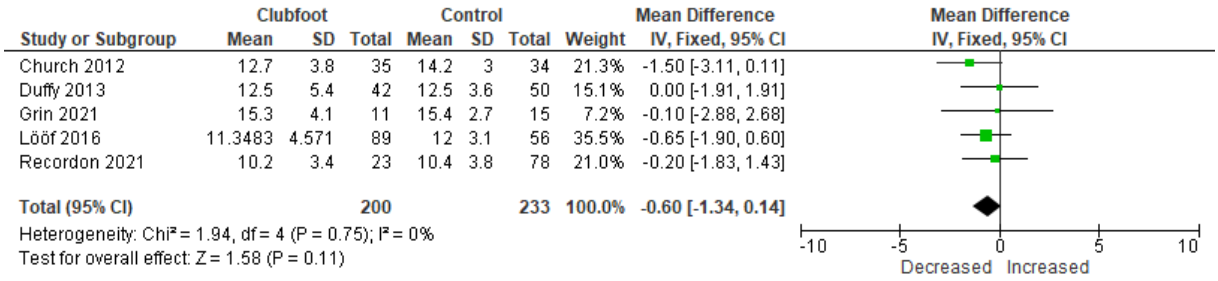
3.3. Meta-Analysis Clubfoot Treated with the Ponseti Method versus Controls

A total of twelve outcome measures could be included in the meta-analyses. Eight of these measures involved the movements of the ankle and knee joints in the sagittal plane during different phases of the gait cycle. Results showed no overall significant differences between children with Ponseti-treated clubfeet and healthy controls at initial contact and during the stance phase (Figure 2A–C).

(**A**) Ankle dorsiflexion/plantarflexion at initial contact.



(**B**) Ankle maximum dorsiflexion during stance



(**C**) Maximum knee extension during stance

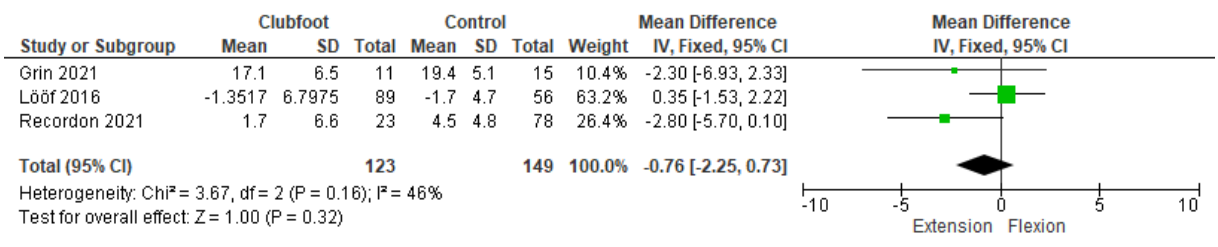
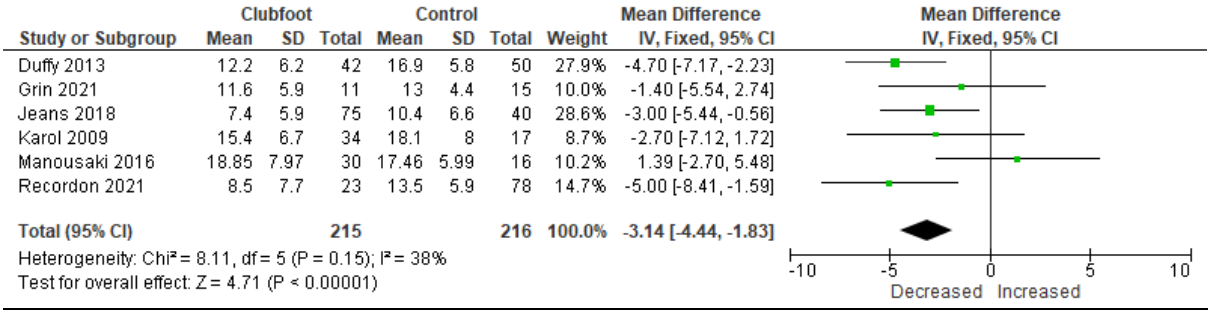
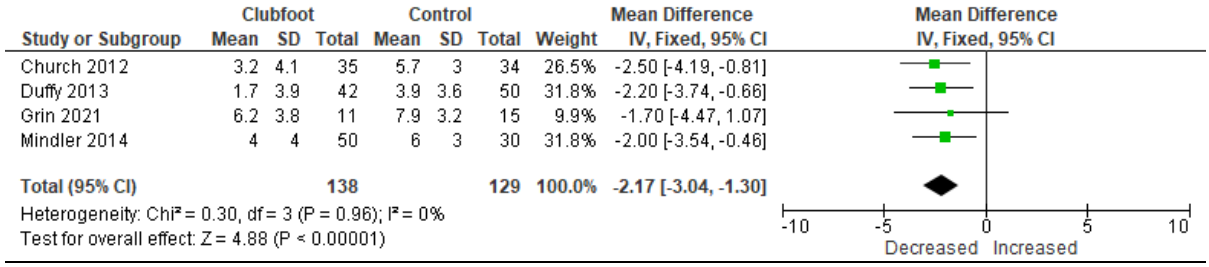


Figure 2. *Cont.*

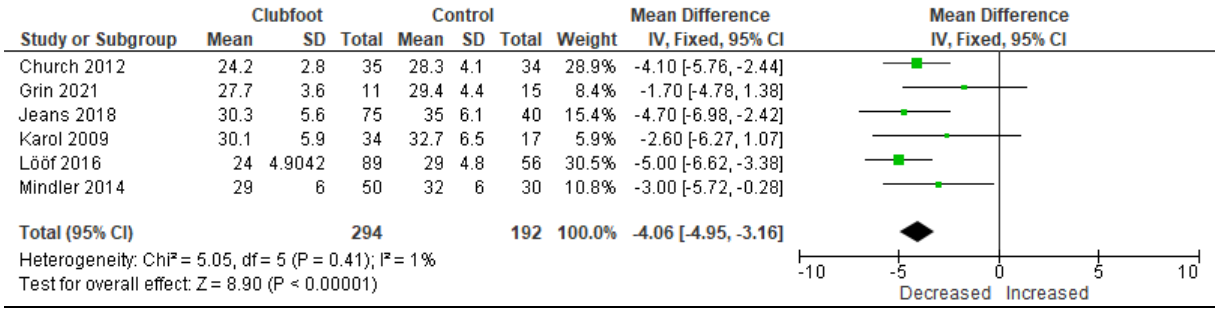
(**D**) Ankle plantarflexion at push-off



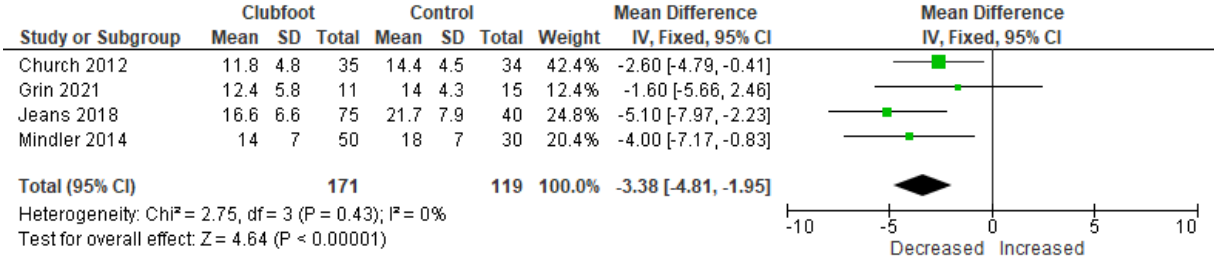
(**E**) Ankle maximum dorsiflexion during swing



(**F**) Ankle range of motion



(**G**) Ankle maximum plantarflexion during the gait cycle



(**H**) Ankle maximum dorsiflexion during gait cycle

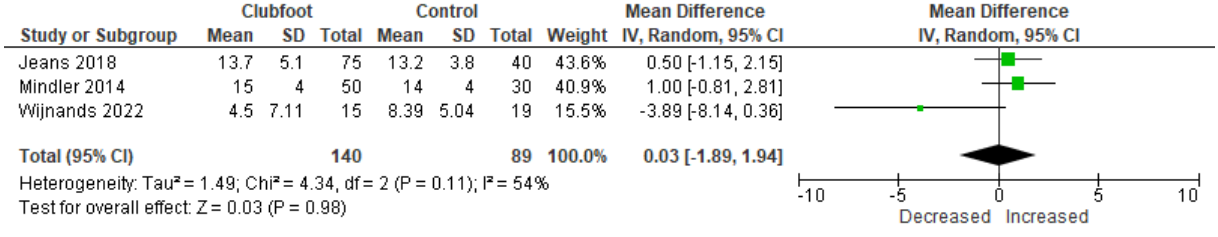


Figure 2. Meta-analysis parameters sagittal plane comparing clubfoot treated with the Ponseti method versus healthy controls [24,25,30,32,34,36,38–41].

At push-off, Ponseti-treated clubfeet showed a decreased plantarflexion [−3.14° (95% CI, −4.44–−1.83; $p < 0.\ 001$)] (Figure 2D). During the swing, maximum dorsiflexion in the ankle for Ponseti-treated clubfoot was significantly lower compared to healthy controls [−2.17° (95% CI, −3.04–−1.30; $p < 0.001$)] (Figure 2E). Over the whole gait cycle, Ponseti-treated clubfeet had a decreased range of motion in the ankle compared to healthy controls [−4.06° (95% CI, −4.95–−3.16; $p < 0.001$)] (Figure 2F) and a decreased maximal plantarflexion [−3.38° (95% CI, −4.81–−1.95; $p < 0.001$)] (Figure 2G). No overall significant difference was seen in maximum dorsiflexion (Figure 2H).

The four other included measures that could be included in the meta-analyses involved movements in the transversal plane and the frontal plane (Figure 3).

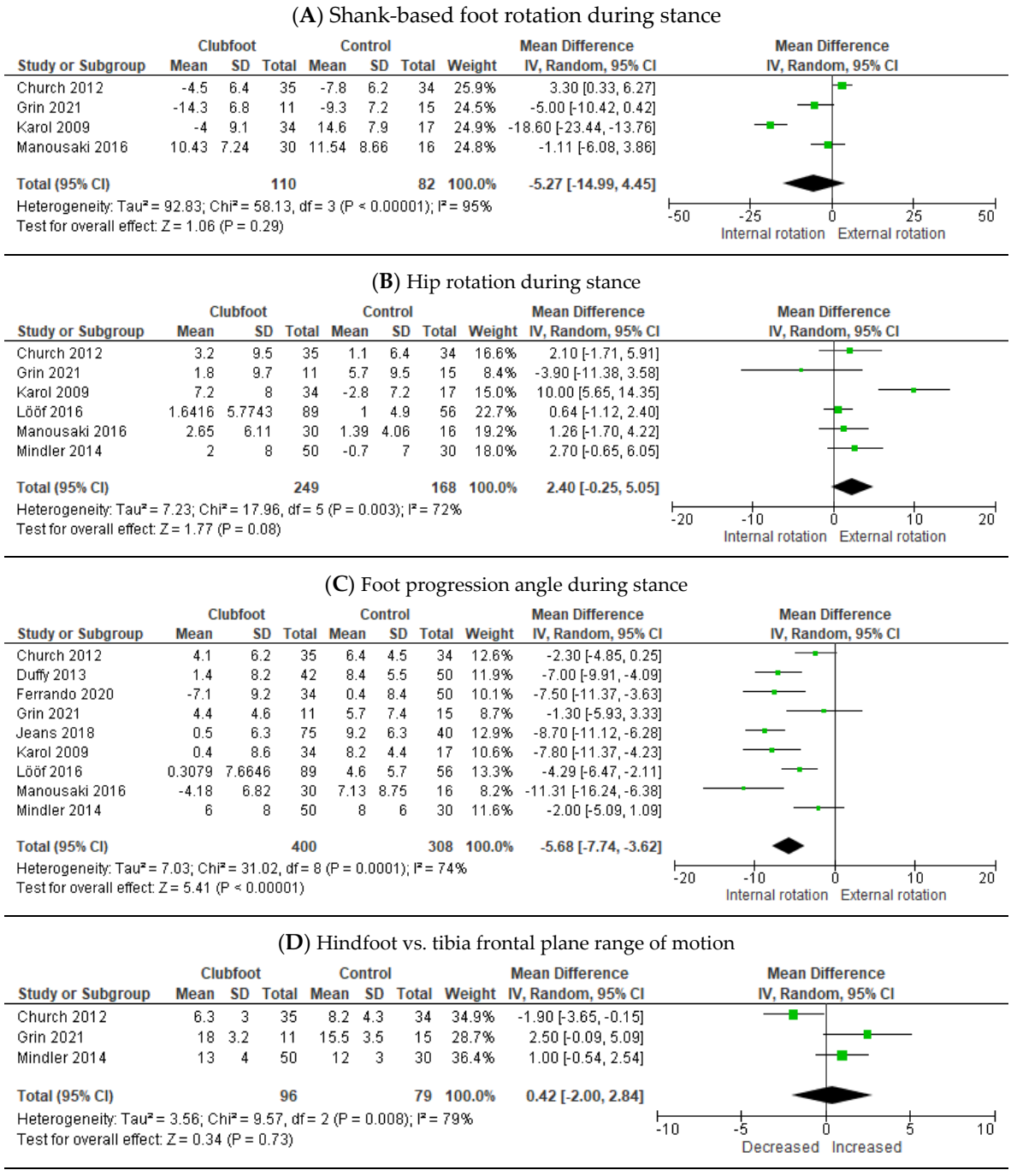


Figure 3. Meta-analysis parameters transversal (**A**–**C**) and frontal (**D**) plane comparing clubfoot treated with the Ponseti method versus healthy controls [24,25,27,30,36,38–41].

No overall difference was seen in shank-based foot rotation (Figure 3A) and hip rotation (Figure 3B) during stance. Compared to healthy controls, children with Ponseti-treated clubfeet showed overall a more inward-oriented foot progression angle during stance [-5.68° (95% CI, -7.74–-3.62; $p \leq 0.001$] (Figure 3C). Furthermore, no overall difference was seen in the frontal plane range of motion of the hindfoot in relation to the *tibia* (Figure 3D).

3.4. Qualitative Analysis Clubfoot Treated with the Ponseti Method versus Controls

An overview of outcome measures not eligible for inclusion (<3 articles or no standard deviation presented [43]) in the meta-analysis but reported in the different articles is displayed in Table 2 and Appendix D.

Table 2. Clubfoot versus controls—Outcome measures included the qualitative analysis presenting significant differences. Parameters without significant differences are presented in Table A3.

Outcome Measure		Moment in Gait Cycle	Studies	Significance
Foot	Mean tibial torsion (EXT)	Stance	[25]	Clubfoot < controls
	Foot progression (EXT)	Preswing	[37]	Clubfoot > controls
Forefoot vs. hindfoot	ROM sagittal (DF/PF)	Gait cycle	[30,38]	Conflicting outcome [2]
	Plantarflexion	20% gait cycle [1]	[37]	Clubfoot < controls
Forefoot vs. *tibia*	ROM sagittal (DF/PF)	Gait cycle	[30,38]	Conflicting outcome [2]
	ROM frontal (PRO/SUP)	Gait cycle	[30,38]	Conflicting outcome [2]
	ROM transversal (AB/AD)	Gait cycle	[30,38]	Conflicting outcome [2]
Ankle	Dorsiflexion	Mid-stance	[24,39]	Conflicting outcome [2]
	Max. plantarflexion	Terminal stance	[40]	Clubfoot < controls
	Dorsiflexion	Swing [1]	[30,37]	Conflicting outcome [2]
Knee	Max. extension	Mid-stance	[36]	Clubfoot < controls
	Max. extension	2nd half of stance	[38]	Clubfoot < controls
	Max. flexion	Swing	[30,38]	Conflicting outcome [2]
Hip	Mean abduction	Gait cycle	[38]	Clubfoot > controls
	Max. rotation (EXT)	Gait cycle	[30,38]	Conflicting outcome [2]
	Mean rotation (EXT)	Mid-stance	[36]	Clubfoot > controls
Total gait scores	GDI	Gait cycle	[36,39,43]	Clubfoot < controls

Abbreviations: ROM = range of motion/PF = plantarflexion/DF = dorsiflexion/INT = internal rotation/EXT = external rotation/AB = abduction/AD = adduction/PRO = pronation/SUP = supination Max. = maximum/GDI = gait deviation index. [1] information gained from figure. [2] in case of conflicting outcomes, additional information is provided in the text.

When comparing children with clubfeet and healthy controls, no significant difference was found for 67 outcomes (Appendix D). A significant difference was found for nine outcome measures, and conflicting results were found for eight outcome measures (Table 2). The outcome measures with a significant difference between the groups and variables with contradicting results are described below.

3.4.1. Stance Phase

From initial contact to mid-stance, no significant differences were reported. At mid-stance, one study mentioned a significantly smaller dorsiflexion in the ankle in Ponseti-treated clubfeet compared to the healthy controls [39], which is in conflict with another study where no significant difference was found [24]. Furthermore, Ponseti-treated clubfeet showed less forefoot plantarflexion in relation to the hindfoot compared to healthy controls [37]. During mid-stance, mean external hip rotation was increased in the clubfoot group, whereas maximum knee extension was decreased in this group compared to healthy controls [36]. Another study mentioned less maximum knee extension in children with Ponseti-treated clubfeet compared to healthy controls during the second half of the stance phase [38]. Subsequently, maximum plantarflexion in the ankle was decreased at a terminal stance in children with Ponseti-treated clubfeet compared to the healthy controls [40]. Furthermore, less external *tibial* torsion during stance was found in children with Ponseti-

treated clubfoot compared to the healthy controls [25]. The foot progression angle during pre-swing was higher in the clubfoot group compared to healthy controls [37].

3.4.2. Swing phase

During the swing phase, decreased maximum knee flexion and decreased dorsiflexion in the ankle were found in children with Ponseti-treated clubfeet compared to healthy controls [37,38], which is in conflict with another study where, although similar trend, no significant difference was found for both parameters [30].

3.4.3. Gait Cycle

When considering the entire gait cycle, mean hip abduction was increased in children with Ponseti-treated clubfeet compared to controls [38], whereas a conflicting result was found looking at maximum external hip rotation [30,38]. In one study, Ponseti-treated clubfeet showed increased external hip rotation [38], whereas the other study showed no significant differences [30]. Furthermore, using a multi-segment foot model, several conflicting results regarding the range of motion (ROM) were observed in the different foot segments [30,38]. One study showed a decreased sagittal range of motion for the forefoot in relation to the hindfoot as well as in relation to the *tibia*, a decreased transversal range of motion for the forefoot in relation to the *tibia*, and an increased range of motion in the frontal plane for the forefoot in relation to the *tibia* in Ponseti treated clubfeet compared to healthy controls [38]. Another study showed no significant differences for the previously mentioned range of motions [30]. When looking at the total gait pattern using the Gait Deviation Index (GDI), children with Ponseti-treated clubfoot showed a decreased GDI score compared to healthy controls [36,39,43].

3.5. Qualitative Analysis Pre-Treatment Relapsed Clubfoot versus Controls

Despite a large number of kinematic outcome measures, there were no outcome measures eligible for inclusion (<3 articles or no standard deviation presented [28]) in the meta-analysis. An overview of all outcome measures that are reported in the different articles is displayed in Table 3 and Appendix E.

Table 3. Pre-treatment relapsed clubfoot vs. Controls—Outcome measures included the qualitative analysis presenting significant differences. Parameters without significant differences are presented in Table A4.

Outcome Measure		Moment in Gait Cycle	Studies	Significance
Foot	Shank-based foot rotation (INT)	Stance	[30]	Relapse > controls
	Foot progression angle (EXT)	Stance	[29]	Relapse < controls
	Foot progression angle (EXT)	70% gait cycle [1]	[35]	Relapse < controls
	Shank-based foot rotation (INT)	Swing	[30]	Relapse > controls
Hindfoot vs. *tibia*	Mean adduction	Gait cycle	[28]	Relapse > controls
	ROM sagittal (DF/PF)	Gait cycle	[28–30]	Conflicting outcome [2]
	ROM transversal (INT/EXT)	Gait cycle	[28–30]	Conflicting outcome [2]
	Inversion	Gait cycle [1]	[35]	Relapse > controls
	Adduction	Gait cycle [1]	[35]	Relapse > controls
	Dorsiflexion	Initial contact	[29,30]	Conflicting outcome [2]
	Max. dorsiflexion	Stance	[29,30]	Conflicting outcome [2]
	Mean adduction	Stance	[29,30]	Conflicting outcome [2]

Table 3. *Cont.*

Outcome Measure		Moment in Gait Cycle	Studies	Significance
Forefoot vs. hindfoot	ROM sagittal (DF/PF)	Gait cycle	[28–30]	Conflicting outcome [2]
	ROM frontal (PRO/SUP)	Gait cycle	[28–30]	Conflicting outcome [2]
	Max. plantarflexion	Gait cycle	[29,30]	Conflicting outcome [2]
	Adduction	Gait cycle[1]	[35]	Relapse > controls
	Dorsiflexion	Initial contact	[29,30]	Conflicting outcome [2]
	Max. dorsiflexion	Stance	[29,30]	Conflicting outcome [2]
	Mean adduction	Stance	[29,30]	Conflicting outcome [2]
	Adduction	Toe-off	[30]	Relapse > controls
	Max. dorsiflexion	Swing	[29,30]	Conflicting outcome [2]
	Supination	80% gait cycle	[29]	Relapse > controls
Forefoot vs. *tibia*	ROM sagittal (DF/PF)	Gait cycle	[29,30]	Conflicting outcome [2]
	ROM transversal (AB/AD)	Gait cycle	[29,30]	Conflicting outcome [2]
	Max. plantarflexion	Gait cycle	[29,30]	Conflicting outcome [2]
	Mean adduction	Gait cycle	[28]	Relapse > controls
	Adduction	Initial contact	[29]	Relapse > controls
	Supination	Initial contact	[29]	Relapse > controls
	Mean adduction	Stance	[29,30]	Relapse > controls
	Plantarflexion	Toe-off	[30]	Relapse < controls
	Mean adduction	Swing	[29,30]	Conflicting outcome [2]
	Mean supination/pronation	Swing	[29,30]	Conflicting outcome [2]
	Dorsiflexion	80% gait cycle	[29]	Relapse > controls
	Adduction	80% gait cycle	[29]	Relapse > controls
Ankle	ROM sagittal (PF/DF)	Gait cycle	[29,30]	Relapse < controls
	Max. dorsiflexion	Gait cycle	[29,34]	Conflicting outcome [2]
	Plantarflexion	Toe-off	[30]	Relapse < controls
Knee	Mean rotation (EXT)	Stance	[29]	Relapse < controls
	Flexion	End of swing [1]	[35]	Relapse > control
Hip	Mean rotation (INT)	Stance	[29,30]	Conflicting outcome [2]
	External rotation	30–60% gait cycle [1]	[35]	Relapse > controls
	Abduction	50–90% gait cycle [1]	[35]	Relapse > controls
Total gait scores	GDI	Gait cycle	[31]	Deviated from normal [3]
	GDI*	Gait cycle	[33]	Deviated from normal [3]
	cFDI*	Gait cycle	[33]	Deviated from normal [3]
	Foot profile score	Gait cycle	[28]	Relapse > controls
	FVS hindfoot sagittal	Gait cycle	[28]	Relapse > controls
	FVS hindfoot frontal	Gait cycle	[28]	Relapse > controls
	FVS hindfoot transversal	Gait cycle	[28]	Relapse > controls
	FVS forefoot sagittal	Gait cycle	[28]	Relapse > controls
	FVS forefoot transversal	Gait cycle	[28]	Relapse > controls

Abbreviations: ROM = range of motion/PF = plantarflexion/DF = dorsiflexion/INT = internal rotation/EXT = external rotation/AB = abduction/AD = adduction/PRO = pronation/SUP = supination Max. = maximum/GDI = gait deviation index/GDI* = scaled gait deviation index/cFDI* = clubfoot deviation index/FVS = foot variable score. [1] information gained from figure. [2] in case of conflicting outcomes, additional information is provided in the text. [3] a score below 90 means a deviated gait pattern compared to controls [42]. Only significant results are included in this table.

Of the total of 106 outcome measures for 55 outcomes, no significant difference was found (Appendix E); for 32 outcome measures, a significant difference was found between children with pre-treatment relapsed clubfeet and healthy controls, and 19 outcome measures from different studies showed conflicting results (Table 3). The outcome measures with a significant difference between the groups and variables with contradicting results are described below.

3.5.1. Multi-Segment Foot Model

Most significant differences between children with pre-treatment relapsed clubfeet and healthy controls are found at foot level, analyzed using a multi-segment foot model. These differences were present in all three planes and multiple phases of gait. In the sagittal plane, children with a relapse showed a significantly decreased forefoot plantarflexion in relation to the *tibia* at toe-off and increased forefoot dorsiflexion in relation to the *tibia* at 80% of the gait cycle [29]. In the frontal plane, children with a relapse showed increased forefoot supination in relation to the *tibia* at initial contact and in relation to the hindfoot at 80% of the gait [29]. Furthermore, increased hindfoot inversion in relation to the *tibia* was seen during the entire gait cycle [35]. In the transversal plane, children with a relapse walked with a more internally shank-based foot rotation [30], a smaller foot progression angle [29,35], and increased forefoot and hindfoot adduction during all phases of gait [28–30,35]. In relation to the *tibia*, increased forefoot adduction was found during initial contact [29], during stance [29,30], at 80% of the gait cycle [29], and over the full gait cycle [28]. Increased forefoot adduction in relation to the *tibia* was found at the toe-off [30] and over the full gait cycle [35]. For the hindfoot, increased adduction was found in relation to the *tibia* during the full gait cycle [28,35].

3.5.2. Conventional Gait Model

When looking at the ankle, a decreased plantar flexion at the toe-off and a smaller sagittal range of motion is seen in children with a relapse [29,30]. Furthermore, children with a relapse showed less external knee rotation and more external hip rotation during stance [29,35]. During the swing, increased knee flexion and increased hip abduction were seen [35]. Additionally, when looking at the total gait pattern using several total gait scores, children with a relapse showed a deviated walking pattern compared to healthy controls [28,31,33].

3.5.3. Conflicting Results

A close look at the conflicting results revealed that one of the nineteen conflicts is also a contradicting result. Two studies presented a decreased transversal range of motion for the hindfoot in relation to the *tibia* [29,30], while one other study showed an increased range of motion in children with relapsed clubfeet [28]. The eighteen remaining conflicting outcomes showed a difference in significance. However, no difference in the direction of deviation in joint angles was seen.

4. Discussion

This systematic review identified a total of 153 different kinematic outcome measures, presented in 20 studies on gait analyses in clubfeet patients treated with the Ponseti method with and without relapse compared to healthy controls. Twelve parameters could be included in a meta-analysis. These meta-analyses comparing Ponseti-treated clubfoot children without relapse to healthy controls showed overall significant differences in ankle plantarflexion at push-off and maximal ankle plantarflexion during the gait cycle, maximum ankle dorsiflexion during the swing, ankle range of motion, and the foot progression angle during stance. Furthermore, on 17 and 51 different kinematic outcomes, one or more studies reported deviating results in respectively clubfoot patients without relapse and pre-treatment relapsed clubfeet compared to healthy controls.

Children with clubfoot have significantly decreased ankle plantar flexion angle at push-off, which is probably caused by a weakness or insufficiency of the plantar flexor muscles [36,45]. Smith et al. (2014), as well as Jeans et al. (2018), reported a decreased plantar flexor strength in children with Ponseti-treated clubfoot compared to healthy controls [37,41]. This finding is also in line with previous findings regarding decreased ankle power in children with clubfeet [19].

Significantly less maximum dorsiflexion during swing was seen in the Ponseti group, which can indicate a drop foot [38], and can consequently lead to insufficient floor clearance

and forefoot landing [46]. Lack of dorsiflexion during the swing can lead to compensations which are mostly seen in an increased hip flexion to lift the foot [46]. Brierty et al. (2022) and Grin et al. (2021) found no significant difference in the hip flexion angle during the full gait cycle using statistical parametric mapping (SPM) [30,35]. However, the results of the meta-analysis on hip rotation did show, although not significant, a tendency for increased external hip rotation. Additionally, one study presented increased hip abduction in children with club feet [38]. Hip rotation and hip abduction are part of a circumduction movement that could also be used to compensate for a decreased foot clearance due to a lack of dorsiflexion. Furthermore, from a clinical point of view, more knee flexion during the initial swing and mid-swing could also be expected to compensate for less dorsiflexion. However, in the two studies that reported knee flexion during swing, a decreased maximum knee flexion was found [30,38].

In addition, it should be noted that three out of the four studies included in the meta-analysis that reported less maximum dorsiflexion during swing also included children with a *tibialis anterior tendon transfer* (TATT) as part of the Ponseti protocol in their study population [25,36,38]. This early TATT was previously associated postoperatively with impaired passive dorsiflexion in a randomized controlled trial comparing the Ponseti method with early TATT (without Ponseti casting) [47]. However, it needs to be questioned whether this small (approximately 2 degrees) but significant difference in maximum dorsiflexion during gait will lead to functional problems in the clubfoot group and, as such, should be addressed in additional treatment.

As a result of a significantly decreased maximum ankle plantar flexion angle over the full gait cycle and a tendency to a decreased maximum ankle dorsiflexion angle during stance, children with a clubfoot showed a significantly decreased ankle range of motion in the sagittal plane. A limited range of motion can negatively affect a child's second ankle rocker and the ability to push off, which are needed for a normal translation of the center of mass during stance. From a clinical point of view, either decreased plantar flexion or decreased dorsiflexion can be treated clinically; however, it requires differentiation in the treatment approach.

A more internally rotated foot progression angle may lead to more compensatory external hip rotation in the transversal plane [48]. Correspondingly, a significantly more internally rotated foot progression and a tendency of increased external hip rotation during stance were found in clubfoot children compared to healthy controls. Additionally, one study reported an increased external hip rotation during mid-stance [36]. However, another study looked specifically at external hip rotation at initial contact and did not find a significant difference between clubfoot children and healthy control children [40]. Further, any torsional or foot deformations contributing to in-toeing could be compensated by external hip rotation during gait. These compensatory mechanisms highlight the importance of considering the entire kinematic chain for the clinical evaluation of gait analysis [49].

The clubfoot deformity has multi-segmental and multiplane characteristics. However, the majority of studies focused on the entire foot instead of separating the foot into different segments [24,27,31,32,35,36,39–43]. Notably, in recent studies, more frequently, a multi-segment foot model, such as the Oxford Foot Model, was used during the 3D gait analyses [25,26,28–30,33,34,37,38]. Although this resulted in an increased number of investigated kinematic parameters, combining a traditional model with a multi-segmental foot model does aid in fully grasping the complexity of the clubfoot deformity and treatment outcome [25,30,33,38,48]. A traditional single-segmental foot model is limited in representing foot motion in the frontal and transversal plane while considering the characteristics of the clubfoot foot motions, such as supination and adduction, are clinically highly relevant. Using a multi-segmental foot model allowed for a detailed analysis of hindfoot and forefoot motion [50], which resulted in the large number of differences at the foot level shown in the results.

In order to assist with the interpretation of the numerous gait- and foot-specific kinematic parameters that are included in the traditional and multi-segmental models, gait and

foot indices are used. Although the numerous kinematic parameters give detailed information regarding a child's gait pattern, all these parameters can be difficult to interpret. Therefore, it could be preferred to use gait or foot indices, in which multiple kinematic parameters are combined into a single score, to assess the overall gait and foot quality in clinical practice [51–53]. These gait indices were implemented in several studies and showed that the overall gait and foot quality is different in clubfoot patients [28,31,33,36,39,43].

In ten of the twelve included studies that compare clubfoot without a relapse to healthy controls, one or more patients had received additional surgical treatment besides the initial casting and bracing phase of the Ponseti treatment, most likely because of a former relapse [25,27,32,36,38–40,42,43]. This could affect the kinematic results due to an increased variability among clubfoot patients within a study population since previous studies showed that surgical treatment, for example, can affect the ankle range of motion [45,54]. To better understand the occurrence of relapse and to evaluate the effect of relapse treatment, it is—from a clinical point of view—necessary to investigate successfully treated clubfeet without a relapse or additional surgical treatment and relapsed clubfeet separately.

Seven studies, including data from relapse patients prior to additional treatment [28–31,33–35], revealed multiple additional kinematic parameters on which relapse clubfoot patients differ from healthy controls. As such, gait analyses might play an important role in the early identification of relapse and determining the necessity of additional treatment, which could prevent the need for major surgical interventions [49,55–57]. In the future, the comparison of clubfoot with and without relapse will be necessary in order to optimize the Ponseti treatment and the detection of relapsed clubfoot. Furthermore, gait analyses can be used to evaluate the outcome of additional treatment for a relapse [11,45,58]. Recent studies investigating the effect of TATT and repeated Ponseti treatment already gave the first insight into kinematic changes after treatment [29,31,59]. Future studies should continue investigating the effect of treatment to aid in optimizing and developing additional (physio)therapy or surgical treatment.

The lack of a clear definition for a relapsed clubfoot was also apparent in the literature describing gait analyses [8]. Some authors used specific relapse treatment as an inclusion criterion for the relapse group, while others based this on planned treatment or an aberrant gait pattern [28–31,33–35]. Considering the heterogeneous nature of a relapse [52,55] and different purposes for applying gait analyses, composing a homogeneous relapse group will be challenging but is important for the comparison and interpretation of results.

Besides the lack of a clear definition for a relapsed clubfoot, this review has a few other limitations. First of all, the quality of a systematic review depends highly on the number and the quality of the included studies. Of the presented kinematic parameters, only twelve could be included in a meta-analysis because of the diverse and numerous reported outcome measures. More homogeneity in measured kinematic variables should be taken into account in order to improve the comparison between separate studies. Secondly, all included studies compared children treated with the Ponseti method and healthy control children, but often the selection of participants and current status of the included patients was unclear, which could have led to selection bias. Thirdly, it seems that data from the same patients has been included in multiple studies. Furthermore, since bilateral club feet are highly correlated [60], future studies should show analyses of both sides if bilateral affected clubfoot patients are measured, especially if these are combined with data from unilateral affected clubfoot patients. However, we do believe that, as a strength of this review, the included studies describe a general population of clubfeet patients treated with the Ponseti method, and as such, the presented results are informative for the clinic. Moreover, the combination of meta-analyses and qualitative analyses led to a comprehensive overview of all studied kinematic characteristics.

5. Conclusions

In conclusion, this systematic review showed that there are several differences in joint angles during gait in children with Ponseti-treated clubfoot with and without relapse compared to healthy controls. When comparing Ponseti-treated clubfoot children without relapse to healthy controls, deviations are mainly found in the sagittal and frontal plane ankle joint kinematics. When comparing children with pre-treatment relapsed clubfeet and healthy controls, deviations are found at foot level in all three planes and multiple phases of gait. We, therefore, emphasize the importance of evaluating the gait pattern of children with clubfoot during clinical follow-up. Being aware of gait impairments in treated clubfoot patients is useful for optimizing the Ponseti method, the detection of relapsed clubfoot, and developing additional (physio) therapy or surgery. However, the question remains as to what functional and/or long-term problems these gait impairments lead to and whether or not these problems could be addressed with additional treatment. Hence, from a clinical point of view, future studies should shift their focus to comparing clubfoot with and without relapse, evaluating the impact of gait impairments, for example, in terms of participation with peers, and investigating the effect of (additional) treatment.

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Conflicts of Interest: The authors declare no conflict of interest.

Appendix A. Example Literature Search Embase.com

Embase.com

(clubfoot/de OR 'pes equinovarus'/exp OR (clubfoot OR clubfeet OR club-foot OR club-feet OR talipes OR equinovarus OR equino-varus):ab,ti) AND (therapy/exp OR 'treatment outcome'/exp OR surgery/exp OR therapy:lnk OR surgery:lnk OR 'clinical trial'/exp OR relapse/exp OR 'follow up'/exp OR 'evaluation study'/exp OR rehabilitation/exp OR rehabilitation:lnk OR 'single blind procedure'/exp OR 'double blind procedure'/exp OR 'triple blind procedure'/exp OR (surg* OR therap* OR treat* OR ponseti OR cast* OR outcome* OR nonoperat* OR nonsurg* OR comprehensive* OR release* OR interven* OR management* OR conservative* OR trial* OR random* OR correct* OR relaps* OR recur* OR (follow* NEXT/1 up*) OR followup* OR evaluat* OR rehabilitat* OR ((double OR single OR triple) NEXT/1 (blind* OR mask*)) OR Physiotherap*):ab,ti) AND (gait/exp OR 'gait disorder'/exp OR electromyogram/exp OR biomechanics/exp OR 'pressure measurement'/de OR (gait OR ((force OR forces OR pressure*) NEAR/3 (distribut* OR peak OR foot OR measur* OR plantar*)) OR EMG OR pedobarograph* OR electromyogr* OR Biomechanic*)).

Appendix B. Risk of Bias

Table A1. Risk of bias for each included study.

Study	Selection	Groups	Measurement	Blinded	Prognostic Factors
Karol 2009 [24]	−	+	+	?	+
Church 2012 [25]	+	?	+	?	?
Duffy 2012 [36]	?	+	+	?	?
Smith 2014 [37]	−	−	+	−	−
Mindler 2014 [38]	?	+	+	?	?
Manousaki 2016 [39]	+	?	+	?	?
Lööf 2016 [40]	?	+	+	?	?
Jeans 2018 [41]	−	?	+	?	+
Manousaki 2019 [42]	+	?	+	?	−
Lööf 2019 [43]	?	+	+	?	+
Dussa 2020 [26]	+	+	+	?	?
Ferrando 2020 [27]	+	+	+	?	?
McCahill 2020 [28]	?	+	+	?	?
Mindler 2020 [29]	+	+	+	?	?
Grin 2021 [30]	−	+	+	?	?
Li 2021 [31]	+	+	+	?	+
Recordon 2021 [32]	?	?	+	?	+
Brierty 2022 [35]	?	+	+	?	?
Grin 2022 [33]	−	+	+	?	?
Wijnands 2022 [34]	−	+	+	?	?

Selection: if stated "all patients in period . . . " or "consecutive patients"; and thus no selection has been made +. **Groups:** if the different groups are clearly defined and comparable with each other (e.g., based on age). **Measurement:** if a valid measurement system/gait analysis system was used. **Blinded:** if outcome measurements were independently/blindly determined. **Prognostic factors:** if clubfoot initial and current classification of clubfeet patients have been described, and the description of the control group states healthy controls. + Low risk/− High risk/? Unclear.

Appendix C. Results Dussa et al. (Overcorrected Clubfoot vs. Controls)

Table A2. Results overcorrected Ponseti clubfoot vs. controls (Dussa et al) [24].

Outcome Measure		Moment in Gait Cycle	Significance
Hindfoot vs. *tibia*	Peak dorsiflexion	Stance	Overcorrect < controls
	Peak eversion	Stance	Overcorrect > controls
	Peak internal rotation	Stance	No significance
	ROM sagittal (DF/PF)	Stance	Overcorrect < controls
	ROM frontal (INV/EV)	Stance	Overcorrect < controls
	ROM transversal (INT/EXT)	Stance	Overcorrect > controls
	Plantar flexion	Toe-off	No significance
	Inversion	Toe-off	Overcorrect < controls
	Mean supination		
Forefoot vs. hindfoot	Peak dorsiflexion	Gait cycle	Overcorrect > controls
	Peak pronation	Stance	Overcorrect > controls
	Peak adduction	Stance	Overcorrect > controls
	Sagittal ROM (PF/DF)	Stance	No significance
	Frontal ROM (PRO/SUP)	Stance	No significance
	Transversal ROM (AB/AD)	Stance	No significance
	Sagittal ROM (FLEX/EXT)	Stance	No significance
Hallux vs. forefoot	Flexion	Toe-off	Overcorrect < controls
	Mean flexion	Swing	Overcorrect < controls
	Sagittal ROM (FLEX/EXT)	Swing	No significance

Appendix D. Ponseti vs. Controls—Additional Outcome Measures Presented in Different Studies

Table A3. Outcome measures included the qualitative analysis with no significant differences.

Outcome Measure		Moment in Gait Cycle	Studies	Significance
Foot	Foot progression	Mid-stance	[32]	No significant difference
	Shank-based foot rotation (INT)	Swing	[30]	No significant difference
Hindfoot vs. *tibia*	ROM sagittal (DF/PF)	Gait cycle	[30,38]	No significant difference
	ROM transversal (INT/EXT)	Gait cycle	[30,38]	No significant difference
	Max. plantar flexion	Gait cycle	[30]	No significant difference
	Dorsiflexion	Initial contact	[30]	No significant difference
	Max. dorsiflexion	Stance	[30]	No significant difference
	Mean dorsiflexion	Stance	[30]	No significant difference
	Mean adduction	Stance	[25,30]	No significant difference
	Mean inversion/eversion	Stance	[30]	No significant difference
	Plantar flexion	Toe-off	[30]	No significant difference
	Adduction	Toe-off	[30]	No significant difference
	Mean adduction	Swing	[30]	No significant difference
	Mean plantar/dorsiflexion	Swing	[30]	No significant difference
	Max. dorsiflexion	Swing	[30]	No significant difference
	Mean inversion/eversion	Swing	[30]	No significant difference
Forefoot vs. hindfoot	ROM frontal (PRO/SUP)	Gait cycle	[30,38]	No significant difference
	ROM transversal (AB/AD)	Gait cycle	[30,38]	No significant difference
	Max. plantar flexion	Gait cycle	[30]	No significant difference
	Dorsiflexion	Initial contact	[30]	No significant difference
	Max. dorsiflexion	Stance	[30]	No significant difference
	Mean dorsiflexion	Stance	[30]	No significant difference
	Mean adduction	Stance	[30]	No significant difference
	Mean supination/pronation	Stance	[25,30]	No significant difference
	Plantar flexion	Toe-off	[30]	No significant difference
	Adduction	Toe-off	[30]	No significant difference
	Mean adduction	Swing	[30]	No significant difference
	Mean plantar/dorsiflexion	Swing	[30]	No significant difference
	Max. dorsiflexion	Swing	[30]	No significant difference
	Mean supiation/pronation	Swing	[30]	No significant difference
Forefoot vs. *tibia*	Peak plantar flexion	Gait cycle	[30]	No significant difference
	Dorsiflexion	Initial contact	[30]	No significant difference
	Max. dorsiflexion	Stance	[30]	No significant difference
	Mean dorsiflexion	Stance	[30]	No significant difference
	Mean adduction	Stance	[30]	No significant difference
	Mean supination/pronation	Stance	[30]	No significant difference
	Plantar flexion	Toe-off	[30]	No significant difference
	Adduction	Toe-off	[30]	No significant difference
	Mean adduction	Swing	[30]	No significant difference
	Mean plantar/dorsiflexion	Swing	[30]	No significant difference
	Max. dorsiflexion	Swing	[30]	No significant difference
	Mean supination/pronation	Swing	[30]	No significant difference
Ankle	Mean dorsiflexion	Stance	[24,30]	No significant difference
	ROM PF/DF	Stance	[32]	No significant difference
	Dorsiflexion	End of swing	[24,39]	No significant difference
	Mean dorsiflexion	Terminal swing	[40]	No significant difference
Knee	Mean rotation	Gait cycle	[38]	No significant difference
	ROM sagittal	Gait cycle	[30]	No significant difference
Hip	External rotation	Initial contact	[40]	No significant difference
Pelvis	Mean tilt	Gait cycle	[30]	No significant difference
	ROM transversal	Gait cycle	[30]	No significant difference
	Max. rotation (EXT)	Gait cycle	[30]	No significant difference
	Max. rotation (INT)	Gait cycle	[30]	No significant difference

Table A3. *Cont.*

Outcome Measure		Moment in Gait Cycle	Studies	Significance
Total gait scores	GPS overall	Gait cycle	[42]	-
	GPS affected side	Gait cycle	[42]	-
	GVS pelvis anterior/posterior	Gait cycle	[42]	-
	GVS pelvis int/ext rotation	Gait cycle	[42]	-
	GVS pelvis up/down	Gait cycle	[42]	-
	GVS hip flexion/extension	Gait cycle	[42]	-
	GVS hip adduction/abduction	Gait cycle	[42]	-
	GVS hip int/ext rotation	Gait cycle	[42]	-
	GVS knee flexion/extension	Gait cycle	[42]	-
	GVS ankle dorsal/plantar flexion	Gait cycle	[42]	-
	GVS foot int/ext rotation	Gait cycle	[42]	-
	GDI *	Gait cycle	[33]	No deviation [1]
	FDI *	Gait cycle	[33]	No deviation [1]
	cFDI *	Gait cycle	[33]	No deviation [1]

Abbreviations: ROM = range of motion/PF = plantarflexion/DF = dorsiflexion/INT = internal rotation/EXT = external rotation/AB = abduction/AD = adduction/PRO = pronation/SUP = supination Max. = maximum/GDI = gait deviation index/GDI * = scaled gait deviation index/ FDI* = scaled foot deviation index/cFDI * = clubfoot deviation index/ [1] a score below 90 means a deviated gait pattern compared to controls [42], '-' outcome compared with controls, but no statistical information was provided. All parameters: Clubfoot > controls.

Appendix E. Relapsed Clubfoot Pre-Treatment vs. Controls—Additional Outcome Measures Presented in Different Studies

Table A4. Outcome measures included qualitative analysis with no significant differences.

Outcome Measure		Moment in Gait Cycle	Studies	Significance
Foot	Foot progression angle	Gait cycle	[28]	Foot progression angle
Hindfoot vs. *tibia*	ROM frontal (INV/EV)	Gait cycle	[26–28]	No significant difference
	Max. plantarflexion	Gait cycle	[27,28]	No significant difference
	Mean dorsiflexion	Gait cycle	[26]	No significant difference
	Mean inversion	Gait cycle	[26]	No significant difference
	Inversion/eversion	Initial contact	[27]	No significant difference
	Adduction	Initial contact	[27]	No significant difference
	Mean dorsiflexion	Stance	[28]	No significant difference
	Mean inversion/eversion	Stance	[28]	No significant difference
	Plantarflexion	Toe-off	[28]	No significant difference
	Adduction	Toe-off	[28]	No significant difference
	Mean adduction	Swing	[27,28]	No significant difference
	Mean plantar/dorsiflexion	Swing	[28]	No significant difference
	Max. dorsiflexion	Swing	[28]	No significant difference
	Mean inversion/eversion	Swing	[28]	No significant difference
	Varus	80% gait cycle	[27]	No significant difference
Forefoot vs. hindfoot	ROM transversal (AB/AD)	Gait cycle	[26–28]	No significant difference
	Mean dorsiflexion	Gait cycle	[26]	No significant difference
	Mean supination	Gait cycle	[26]	No significant difference
	Mean adduction	Gait cycle	[26]	No significant difference
	Supination	Initial contact	[27]	No significant difference
	Adduction	Initial contact	[27]	No significant difference
	Mean dorsiflexion	Stance	[28]	No significant difference
	Mean supination/pronation	Stance	[28]	No significant difference
	Plantarflexion	Toe-off	[28]	No significant difference
	Mean adduction	Swing	[27,28]	No significant difference
	Mean plantar/dorsiflexion	Swing	[28]	No significant difference
	Mean supination/pronation	Swing	[28]	No significant difference
	Supination	80% gait cycle	[27]	No significant difference

Table A4. *Cont.*

Outcome Measure		Moment in Gait Cycle	Studies	Significance
Forefoot vs. *tibia*	ROM frontal (PRO/SUP)	Gait cycle	[28]	No significant difference
	Mean dorsiflexion	Gait cycle	[26]	No significant difference
	Mean supination	Gait cycle	[26]	No significant difference
	Dorsiflexion	Initial contact	[27,28]	No significant difference
	Max. dorsiflexion	Stance	[27,28]	No significant difference
	Mean dorsiflexion	Stance	[28]	No significant difference
	Mean supination/pronation	Stance	[28]	No significant difference
	Adduction	Toe-off	[28]	No significant difference
	Mean plantar/dorsiflexion	Swing	[28]	No significant difference
	Max. dorsiflexion	Swing	[28]	No significant difference
Ankle	Max. plantarflexion	Gait cycle	[28]	No significant difference
	Dorsiflexion	Initial contact	[28]	No significant difference
	Mean dorsiflexion	Stance	[28]	No significant difference
	Max. dorsiflexion	Stance	[28]	No significant difference
	Max. dorsiflexion	Swing	[28]	No significant difference
	Mean plantar/dorsiflexion	Swing	[28]	No significant difference
Knee	ROM sagittal (PF/DF)	Gait cycle	[28]	No significant difference
	Max. extension	Stance	[28]	No significant difference
	Max. flexion	Swing	[28]	No significant difference
Hip	Max. rotation (EXT)	Gait cycle	[28]	Max. rotation (EXT)
Pelvis	Mean tilt	Gait cycle	[28]	No significant difference
	ROM transversal	Gait cycle	[28]	No significant difference
	Max. rotation (EXT)	Gait cycle	[28]	No significant difference
	Max. rotation (INT)	Gait cycle	[28]	No significant difference
Total gait scores	FDI *	Gait cycle	[31]	No deviation [1]
	FVS forefoot frontal	Gait cycle	[26]	No significant difference

Abbreviations: ROM = range of motion/PF = plantarflexion/DF = dorsiflexion/INT = internal rotation/EXT = external rotation/AB = abduction/AD = adduction/PRO = pronation/SUP = supination/Max. = maximum/* = scaled foot deviation index/FVS = foot variable score. [1] a score below 90 means a deviated gait pattern compared to controls [42].

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children

MDPI

Article

Introduction of a Novel Sequential Approach to the Ponte Osteotomy to Minimize Spinal Canal Exposure

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Abstract: Ponte osteotomy is an increasingly popular technique for multiplanar correction of adolescent idiopathic scoliosis. Prior cadaveric studies have suggested that sequential posterior spinal releases increase spinal flexibility. Here we introduce a novel technique involving a sequential approach to the Ponte osteotomy that minimizes spinal canal exposure. One fresh-frozen adult human cadaveric thoracic spine specimen with 4 cm of ribs was divided into three sections (T1–T5, T6–T9, T10–L1) and mounted for biomechanical testing. Each segment was loaded with five Newton meters under four conditions: baseline inferior facetectomy with supra/interspinous ligament release, superior articular process (SAP) osteotomy in situ, spinous process (SP) osteotomy in situ, and complete posterior column osteotomy with SP/SAP excision and ligamentum flavum release (PCO). Compared to baseline, in situ SAP osteotomy alone provided 3.5%, 7.6%, and 7.2% increase in flexion/extension, lateral bending, and axial rotation, respectively. In situ SP osteotomy increased flexion/extension, lateral bending, and axial rotation by 15%, 18%, and 10.3%, respectively. PCO increased flexion/extension, lateral bending, and axial rotation by 19.6%, 28.3%, and 12.2%, respectively. Our report introduces a novel approach where incremental increases in range of motion can be achieved with minimal spinal canal exposure and demonstrates feasibility in a cadaveric model.

Keywords: adolescent idiopathic scoliosis; Ponte osteotomy; pediatric

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1. Introduction

Adolescent idiopathic scoliosis (AIS) affects approximately 2–4% of adolescents and is a complex three-dimensional deformity of the spine characterized by abnormalities in the coronal, sagittal, and axial planes [1,2]. Treatment depends on curve magnitude and the patient's skeletal maturity, ranging from observation or bracing to surgery [3,4]. In general, patients with major Cobb angles greater than 50 are indicated for surgery in the form of spinal fusion [5]. Surgery aims to halt curvature progression, improve sagittal and coronal balance, reduce short-term and long-term complications associated with AIS, and improve patient appearance [4,6].

Historically, surgeons used a combination of anterior and posterior releases of the spine to increase spinal flexibility and improve deformity correction before fusion, as anterior discectomy was considered necessary in patients with stiff curves [6]. In recent years, however, biomechanical advances in pedicle-based instrumentation have dramatically increased the correction force that surgeons can apply to the spinal column for deformity correction and have lessened the need for combined releases [7]. Due to the morbidity associated with anterior approaches, many surgeons now advocate for posterior-only approaches for spinal fusion, even for patients with large curves [8,9].

Despite advances in powerful instrumentation, osteotomies of the spine are still sometimes necessary to correct more significant deformities [6]. One such osteotomy is

the Ponte-type osteotomy, a posterior-column-based technique that can be performed immediately prior to posterior spinal fusion with pedicle screw implants [10–14]. Although the Ponte osteotomy was initially described to treat sagittal deformities, it has since been modified and is now a widespread technique for coronal, rotational, and sagittal plane correction in major thoracic curves. The Ponte osteotomy has also been described for use in thoracolumbar and lumbar curve correction [15]. In this technical note, we will focus on thoracic correction.

Ponte first described what is now known as the Ponte osteotomy procedure for correcting the sagittal-plane deformity associated with Scheuermann kyphosis in 1987 [16]. The traditional Ponte osteotomy, as described by Ponte, is as follows (see Figure 1).

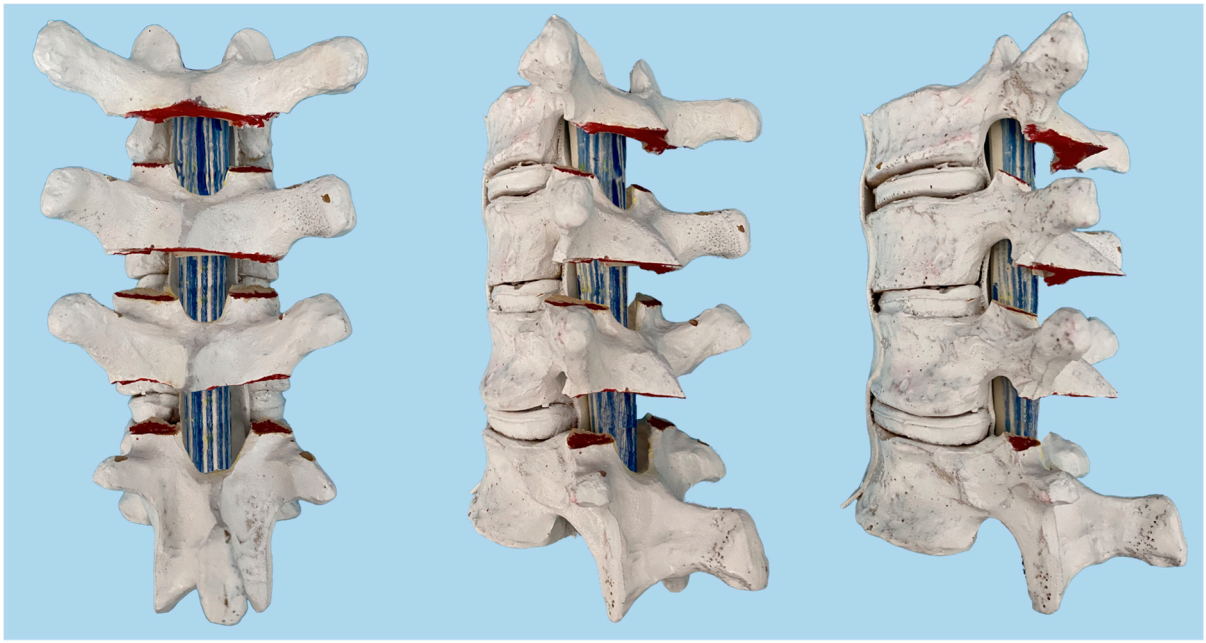


Figure 1. The Ponte osteotomy, as described by Ponte in the thoracic spine.

"Spinous processes are resected at their base to allow better visualization of the bony parts to be removed . . . An angled double-action rongeur and/or a Kerrison is used to perform the bony resections. Complete facetectomies and wide inferior and superior laminectomies are performed at every intersegmental level . . . A generous resection of facet joints and laminae, in severe deformities as far as the pedicles, is an essential step of the osteotomy and the technique . . . The ligamentum flavum is removed entirely at all levels." [17].

As seen from Ponte's description of the Ponte osteotomy, the complete removal of the spinous process and a wide lamina resection fully expose the spinal canal in the surgical field. This leaves the spinal cord at risk within the surgical field and may not be fully covered in bone even after compression of the posterior elements. Using rongeurs in the spinal canal also often results in epidural bleeding and theoretically increases the risk of dural tears or neurologic injury. Recent studies have also shown that Ponte osteotomies increase rates of intraoperative neuromonitoring alerts and blood loss during posterior spinal fusion [18–21]. Recently, ultrasonic bone-cutting devices have facilitated changes in osteotomy techniques to minimize exposure of the spinal canal in the surgical field.

Here we describe a novel sequential approach and modification of the Ponte osteotomy that aims at keeping the spinal cord covered by bone in the surgical field if possible. The method only progresses to a full Ponte osteotomy with exposure of the spinal canal when the prior sequential osteotomy steps cannot achieve appropriate spinal flexibility and curve correction. Ultrasonic bone cutters allow in situ osteotomies of the superior articular

process (SAP) and spinous process (SP), releasing the ligamentous tethers posteriorly with minimal bony resection. This approach allows surgeons to achieve the desired amount of spinal flexibility to facilitate deformity correction while minimizing exposure to the spinal canal. We investigated this aim using a human cadaveric non-scoliotic thoracic spinal specimen, an established biomechanical model, to study AIS surgical techniques.

2. Materials and Methods

2.1. Surgical Procedure

The Ponte osteotomy, as described above, involves the removal of the spinous process, facet joints, lamina, and ligamentum flavum, and by definition, exposes the spinal canal in the surgical field. Our novel technique involves sequential step-wise osteotomies, which we typically combine at multiple vertebral levels in vivo. If more flexibility is needed after the first round of osteotomies, then the next sequential osteotomy step is performed. It should be noted that an oscillating saw and osteotome were used to perform osteotomies on the cadaver specimen for this technical report, while in vivo, an ultrasonic scalpel was used. The sequential osteotomies, which were subjected to mechanical testing, were as follows (Figure 2).

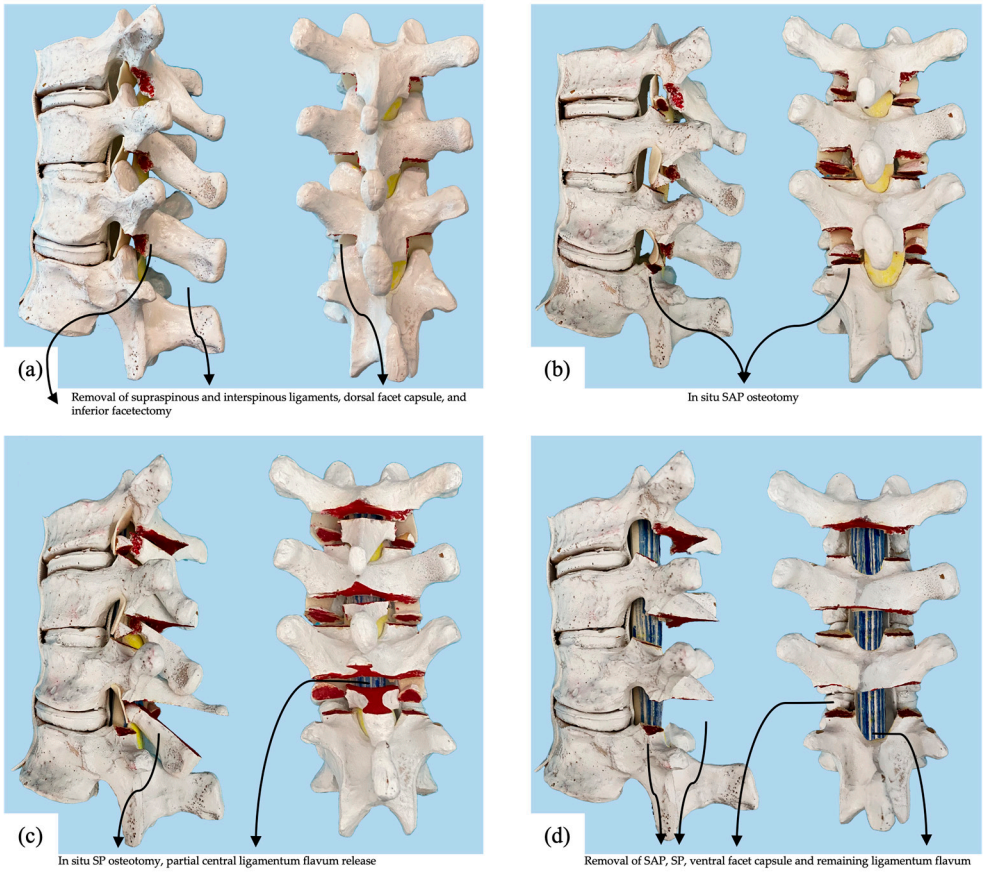


Figure 2. Sawbone thoracic spine models illustrating the experimental conditions of (**a**) baseline, (**b**) SAP osteotomies in situ, (**c**) SP osteotomy in situ, and (**d**) complete posterior column osteotomy.

Baseline: Supraspinous and interspinous ligaments were cut and partially excised using a rongeur. Inferior facetectomy was performed using an osteotome. The inferior articular processes were removed along with all the visible dorsal facet joint capsule.

O1: The in situ SAP osteotomy was performed using an oscillating saw. The cut began in the exposed SAP cartilage surface, and the saw was directed ventrally and cranially to avoid entering the pedicle at that level. Once this osteotomy was complete, the SAP

fragment was confirmed to be detached from its vertebra but remained tethered cranially by the joint capsule and ligamentum flavum.

O2: The SP osteotomy in situ was performed using an oscillating saw to connect the left and right facetectomy cuts across the lamina. The saw was directed ventrally and cranially to terminate the cut ventrally near the superior aspect of the attachment of the ligamentum flavum at that level, resulting in partial central ligamentum flavum release. After this step, the epidural space was typically visible through a small gap in the lamina. The lamina/spinous process fragment was free from the level above but remained tethered to the level below by the ligamentum flavum. The fragment was left in place to protect the canal and act as a bone graft.

O3: A complete posterior column osteotomy was performed using a rongeur to remove the SP/lamina fragment. A Kerrison rongeur was then used to release any remaining ligamentum flavum laterally and remove the SAP fragment. After this step, the spinal canal was open, and dura and epidural fat were visible. The exposure was now the same as if a Ponte osteotomy had been initially performed.

The primary outcomes were average degree change and percent change from baseline range of motion (ROM) under load for each sequential condition for the three cadaveric sections.

Biomechanical testing was first performed on the three initial spinal specimen sections from the single cadaver and repeated after each successive procedure. A board-certified pediatric orthopaedic surgeon performed osteotomies to ensure appropriate technique.

2.2. Specimen Preparation

One fresh-frozen human cadaveric thoracic spine specimen (age 57, BMI of 18) from T1-L1 with 4 cm of ribs was divided into three sections (T1–T5, T6–T9, T10–L1). All specimens were dissected free of paraspinal musculature to include only stabilizing ligaments, bones, intervertebral discs, and 4 cm sections of ribs. All specimens were studied using radiographs to ensure no history of fractures, osseous abnormalities, osteoporosis, bridging osteophytes, or previous spine surgery. Specimens were thawed for 24 h at room temperature before testing.

2.3. Biomechanical Testing and Analysis

A simVITRO robotic testing system (Cleveland Clinic, Cleveland, OH, USA) with a KR300 robot (Kuka, Ausburg, Germany) and an Omega160 6-axis load cell with an SI-2500–400 calibration (ATI, Apex, NC, USA) was used to apply the single-plane ranges of motion to the spine (Figure 3). Potting involved drilling a 3-inch wood screw placed anterior to posterior through the most inferior and superior vertebral body into the spinous process, which was confirmed with fluoroscopy. This wood screw then sat within the trough of the pot. Poly-methyl-methacrylate (PMMA) cement was added into the mold parallel to the vertebral end plate of the interior and superior vertebra. In order to mount each spine to the robot, the superior and inferior vertebrae of each specimen in PMMA blocks were rigidly fixed to custom clamps to prevent spine movement relative to the base and robot arm. Testing was conducted in all three modes of bending (flexion-extension, lateral bending, and axial rotation). Phosphate buffered saline solution was used to keep the soft tissue structures hydrated and preserve the mechanical integrity of the specimen throughout testing. Throughout the testing process, there was no evidence of loosening. Spines were mounted onto the robotic platform and initialized by determining the spatial relationships between the robot, load cell, and each vertebral segment using a Romer Absolute Arm Digitizer (Hexagon, RI, USA). Vertebral body coordinate systems definitions were established according to International Society of Biomechanics standards [22]. Joint motion and load were controlled by establishing geometric relationships to a coordinate system, and any changes in biomechanical responses were recorded. Additional information regarding the programming of the robot and coordinate system analysis has been described by Mageswaran et al. [23].

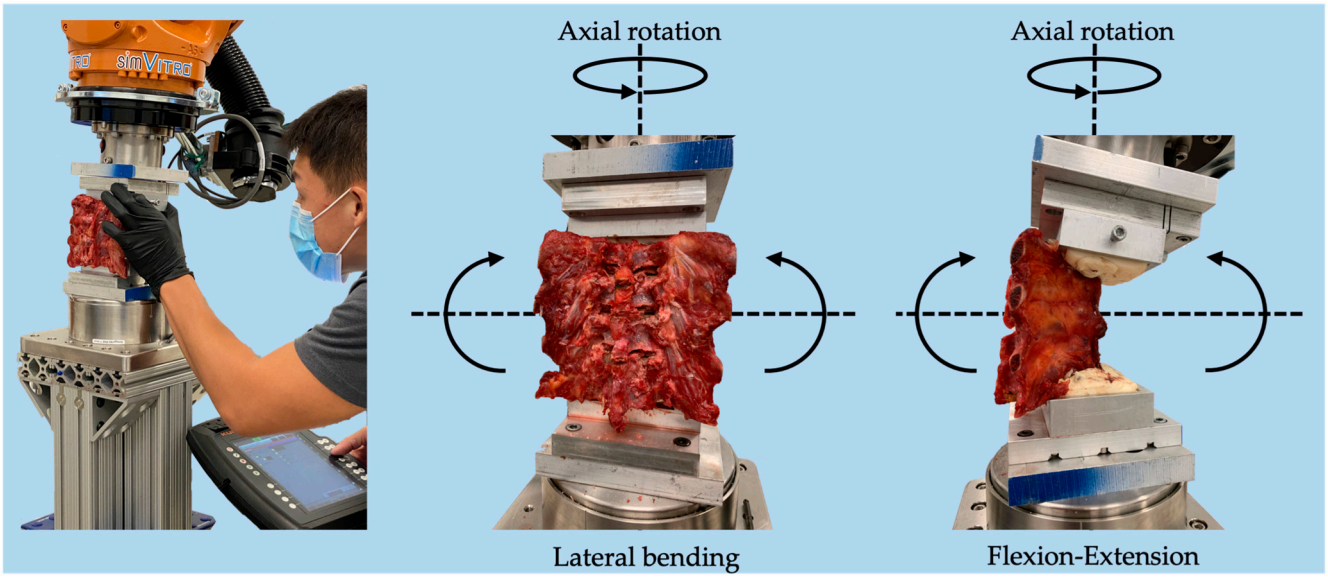


Figure 3. Illustration of experimental setup and directional loading of thoracic sections.

Loading conditions were performed along three primary single-plane axes (±5 Nm moment in flexion-extension, lateral bending, and axial rotation) while minimizing the load along the translational axes (Figure 3). To eliminate viscoelastic effects, the specimens were preconditioned for four cycles before measuring the fifth cycle.

3. Results

3.1. Average of the Entire Single Specimen Spine T1–T12

Under 5 Nm of torque, the T1–T12 percent change in flexion-extension, lateral bending, and axial rotation steadily increased in a stepwise fashion following initial SAP osteotomy (O1), a subsequent SP osteotomy (O2), and finally completion PCO with excision of the ligamentum flavum (O3). Final flexion-extension increased from the baseline specimen by 3.5% after O1, 15% following O2, and 19.6% following O3. The greatest percent change was in lateral bending, in which sequential osteotomies increased final ROM from the baseline specimen by 7.6% after O1, 18.3% following O2, and 28.4% following O3. Lastly, axial rotation increased by 7.2% after O1, 10.3% following O2, and 12.2% following O3 relative to the baseline specimen. In all testing conditions, the most significant percentage increase occurred following SP osteotomies (O2); flexion-extension increased by 11.5% from O1 to O2, whereas lateral bending increased by 10.7%, and axial rotation increased by 3.1% (Table 1, Figure 4).

Table 1. T1–T12 percent change from baseline in response to 5 Nm torque following stepwise osteotomies (% + SD).

T1–T12	SAP Alone (O1)	SAP + SP (O2)	SAP +SP + PC + LF (O3)
Flexion-Extension	3.5 ± 2.6	15.0 ± 2.0	19.6 ± 4.4
Lateral Bending	7.6 ± 5.9	18.3 ± 17.1	28.4 ± 31.0
Axial Rotation	7.2 ± 3.0	10.3 ± 3.9	12.2 ± 5.3

SAP, Superior Articular Process; SP, Spinous Process; PC, Posterior Column; LF, Ligamentum Flavum; O1, 1st osteotomy; O2, 2nd osteotomy; O3, 3rd Osteotomy.

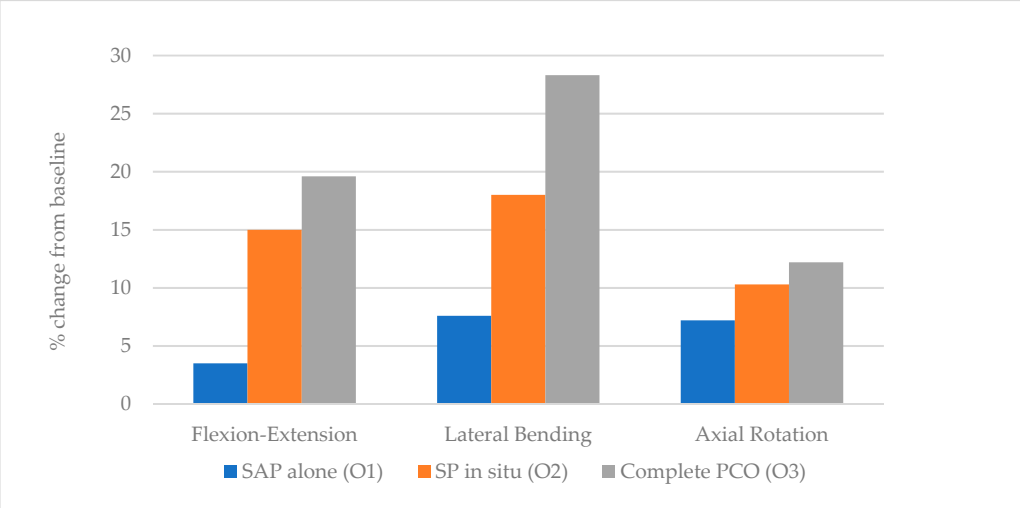
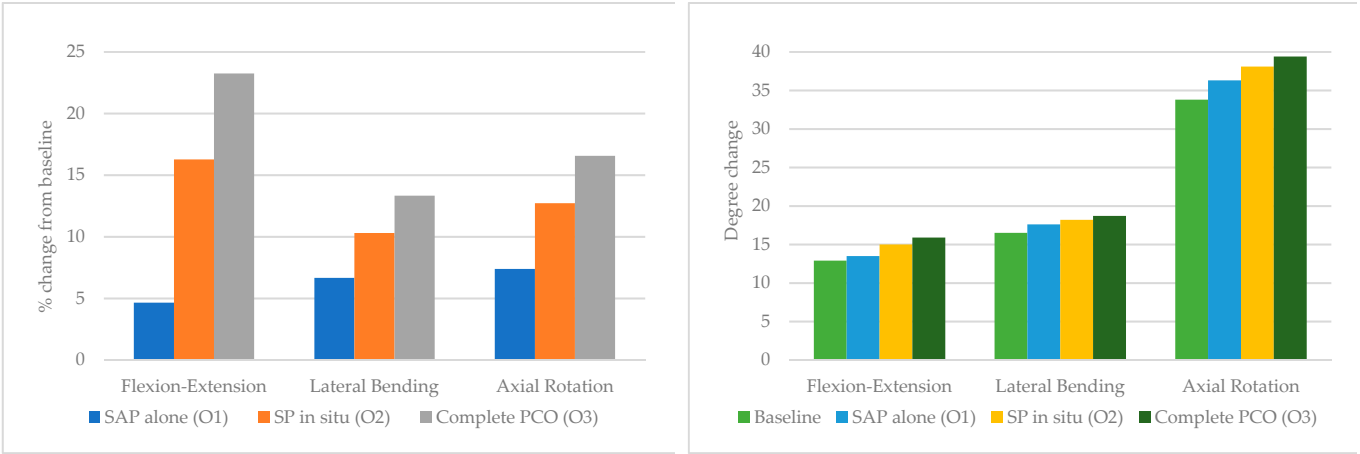


Figure 4. T1–T12 percent change from baseline in response to 5 Nm torque following stepwise osteotomies.

3.2. Upper Thoracic Segment T1–T4 from the Single Thoracic Specimen

Flexion-extension increased from 12.9° in the baseline condition to 13.5° following O1. This subsequently increased to 15.0° following O2 and 15.9° following O3. Lateral bending increased from 16.5° to 17.6° after O1, 18.2° after O2, and 18.7° after O3, Axial rotation increased from 33.8° to 36.3° after O1, 38.1° after O2, and 39.4° after O3.

This increase in ROM translated to a total increase of 4.7% in flexion-extension after O1, 16.3% after O2, and 23.3% after O3 relative to baseline. Lateral bending increased by 6.7%, 10.3%, and 13.3%, after O1, O2 and O3, respectively. Axial rotation increased by 7.4%, 12.7%, and 16.6%, after O1, O2, and O3, respectively (Tables 2 and 3, Figure 5).



(**a**) (**b**)

Figure 5. T1–T4 (**a**) percent from baseline and (**b**) degree change in response to 5 Nm torque following stepwise osteotomies.

3.3. Middle Thoracic Segment T5–T8 from the Single Thoracic Specimen

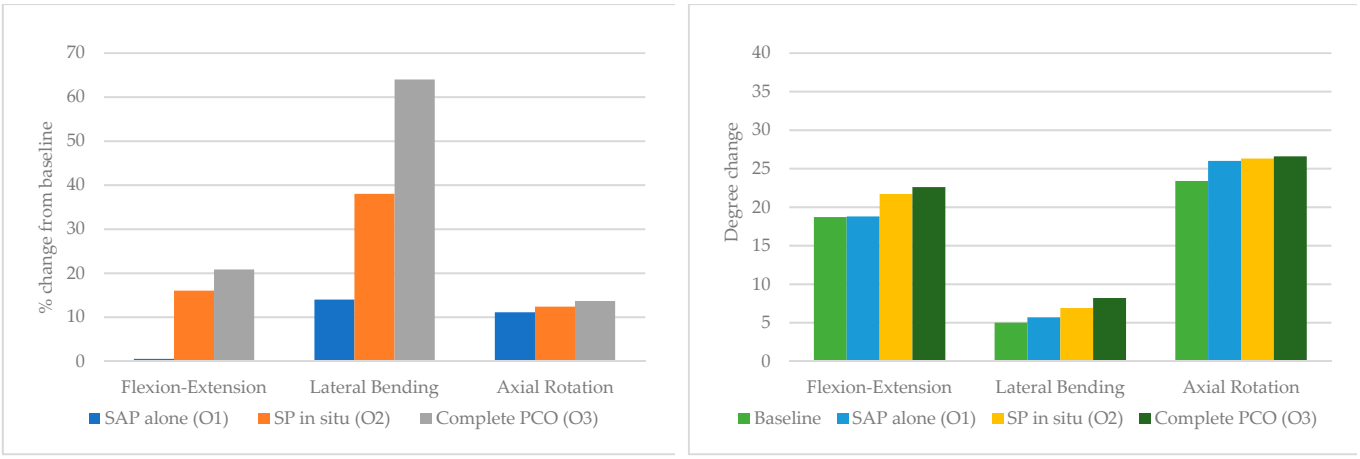
Flexion-extension increased from 18.7° in the baseline condition to 18.8° following O1. This increased to 21.7° following O2 and 22.6° following O3. Lateral bending increased from 5° to 5.7° after O1, 6.9° after O2, and 8.2° after O3. Axial rotation increased from 23.4° to 26° after O1, 26.3° after O2, and 26.6° after O3 (Tables 2 and 3, Figure 6).

Table 2. Percent change from baseline in response to 5 Nm torque following stepwise osteotomies in each thoracic segment (%).

		SAP Alone (O1)	SAP + SP (O2)	SAP + SP + PC + LF (O3)
T1–T4	Flexion-Extension	4.7	16.3	23.3
	Lateral Bending	6.7	10.3	13.3
	Axial Rotation	7.4	12.7	16.6
T5–T8	Flexion-Extension	0.5	16	20.9
	Lateral Bending	14	38	64
	Axial Rotation	11.1	12.4	13.7
T9–T12	Flexion-Extension	5.3	12.6	14.7
	Lateral Bending	2.2	6.7	7.8
	Axial Rotation	3.2	5.8	6.3

Table 3. Degree change in range of motion in response to 5 Nm torque following stepwise osteotomies (°).

		Baseline	SAP Alone (O1)	SAP + SP (O2)	SAP + SP + PC + LF (O3)
T1–T4	Flexion-Extension	12.9	13.5	15	15.9
	Lateral Bending	16.5	17.6	18.2	18.7
	Axial Rotation	33.8	36.3	38.1	39.4
T5–T8	Flexion-Extension	18.7	18.8	21.7	22.6
	Lateral Bending	5	5.7	6.9	8.2
	Axial Rotation	23.4	26	26.3	26.6
T9–T12	Flexion-Extension	9.5	10	10.7	10.9
	Lateral Bending	9	9.2	9.6	9.7
	Axial Rotation	19	19.6	20.1	20.2



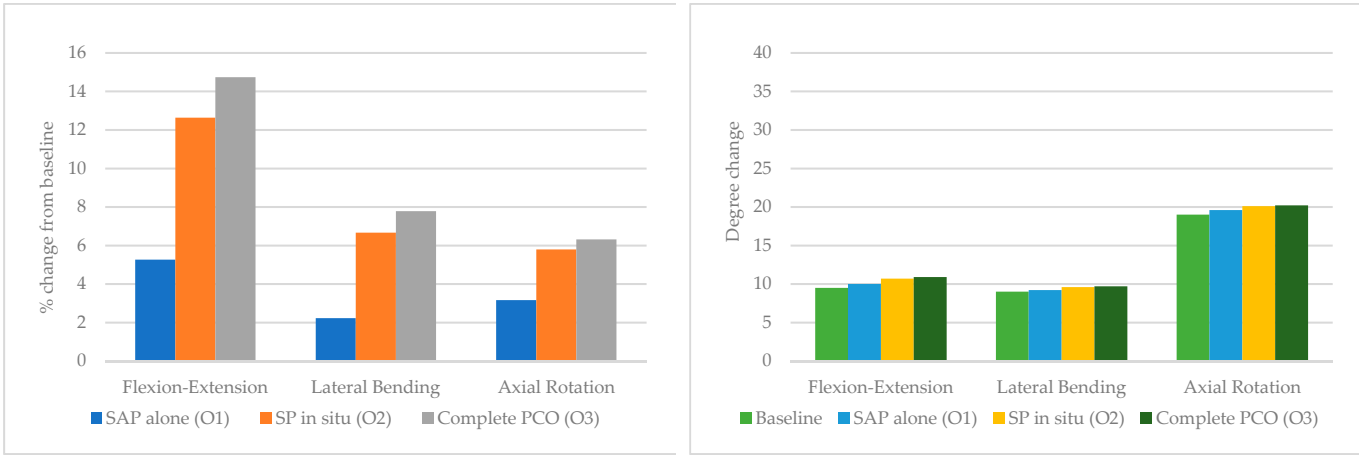
(**a**) (**b**)

Figure 6. T5–T8 (**a**) percent from baseline and (**b**) degree change in response to 5 Nm torque following stepwise osteotomies.

The highest percent change in ROM was seen in the middle thoracic segment (T5–T8); although flexion-extension initially increased by only 0.5% after O1, this subsequently increased by 16% and 20.9% after O2 and O3, respectively. Lateral bending increased by 14% after O1, 38% after O2, and 64% after O3. Axial rotation increased by 11.1%, 12.4%, and 13.7 after O1, O2 and O3, respectively.

3.4. Lower Thoracic Segment T9–T12 from the Single Thoracic Specimen

Flexion-extension increased from 9.5° at baseline to 10° following O1. This increased to 10.7° following O2 and 10.9° following O3. Lateral bending increased from 9° to 9.2° after O1, 9.6° after O2, and 9.7° after O3. Axial rotation increased from 19° to 19.6° after O1, 20.1° after O2, and 20.2° after O3 (Tables 2 and 3, Figure 7).



(**a**) (**b**)

Figure 7. T9–T12 (**a**) percent from baseline and (**b**) degree change in response to 5 Nm torque following stepwise osteotomies.

Lastly, the lower thoracic segment (T9–T12) saw the smallest increase in flexion-extension, lateral bending, and axial rotation. From baseline to O1, to O2, and to O3, flexion-extension increased by 5.3%, 12.6%, and 14.7%, respectively. Lateral bending increased by 2.2%, 6.7%, and 7.8%. Axial rotation increased by 3.2%, 5.8%, and 6.3% after O1, O2 and O3, respectively.

4. Discussion

Modern surgical treatment of AIS involves deformity correction in multiple planes through translation, derotation, and lengthening of the posterior column to restore kyphosis. The release of posterior elements increases spinal flexibility to allow better deformity correction in multiple planes [24,25].

With a single cadaveric thoracic spine, we demonstrated the feasibility of a novel sequential approach to the Ponte osteotomy, which resulted in an incrementally increasing range of motion in spinal flexion/extension, axial rotation, and lateral bending. SAP osteotomy alone provided a 3.5%, 7.6%, and 7.2% increase in flexion/extension, lateral bending, and axial rotation, respectively, while in situ SP osteotomy provided a 15%, 18%, and 10.3% increase. After these two in situ osteotomies, the spinal canal was still largely protected by the posterior elements. Adding a complete posterior column osteotomy improved flexion/extension, lateral bending, and axial rotation by 19.6%, 28.3%, and 12.2%, respectively.

The first two osteotomies described (O1, O2) could be performed with an ultrasonic bone-cutting device safely without passing rongeurs through the epidural space. According to our data, they provided roughly 75% of the flexibility gained by a formal Ponte osteotomy (70% in flexion-extension, 77% in lateral bending, and 77% in axial rotation). This biomechanical data supports a stepwise approach to the Ponte osteotomy and demonstrates that stepwise gains in mobility can be achieved while limiting spinal cord exposure.

When comparing the results from our single cadaveric specimen to the literature, our results are generally comparable in terms of flexion-extension and axial rotation. With lateral bending, however, we found a greater change from baseline than in several other comparable studies.

Holewijn et al. performed a stepwise posterior osteotomy study involving resection of the supra/interspinous ligament (SIL), inferior facet, flaval ligament, superior facet, and rib heads. The authors found an incremental increase in spinal flexibility with diminishing returns after each step [24]. In their study, SIL resection, flaval ligament, and complete facetectomies increased ROM by 29.6% in flexion, 12.1% in extension, 5.5% in lateral bending, and 15.3% in axial rotation [24]. Compared to Holewijn et al., we observed greater lateral bending and axial rotation with an in situ SP osteotomy, which effectively detached the ligamentum flavum without uncovering the underlying spinal canal posteriorly.

A study by Sangiorgio et al. found that a complete Ponte osteotomy increased flexion by 69%, extension by 56%, and axial rotation by 34%, but only minimally increased lateral bending by 2% [25]. A report by Wang et al. found that a Ponte osteotomy increased flexion by 23%, extension by 15%, and axial rotation by 21%, but only minimally increased lateral bending by 2% [26]. While our single thoracic cadaver showed similar flexion/extension and axial rotation improvements to Wang et al. and less than Sangiorgio et al., we saw a six-times greater lateral bending motion with the complete Ponte osteotomy. In another analogous study, Borkowski et al. used a two-step modification of the Ponte osteotomy in 10 thoracic cadaveric specimens mounted as a large unit from T1–T12 [27]. The authors used a biomechanical testing setup that recorded single plane motion (flexion-extension, lateral bending, axial rotation) after bilateral total facetectomies and increasing numbers of Ponte osteotomies up to four levels. Like Sangiorgio et al. and Wang et al., Borkowski et al. found lateral bending changed less than flexion-extension and axial rotation with a 9% increase from baseline after four-level Ponte osteotomy [27].

One issue in comparing our report to the previous studies is that each study used an intact spine as its baseline reference. We chose to begin from a baseline after SIL resection and inferior facetectomy because these are generally accepted as standard steps of the exposure for posterior spinal fusion. It is unclear why we saw greater lateral bending and coronal flexibility changes, but this highlights the potential variability between cadaveric specimen stiffness, which will be lessened with a greater sample size.

A significant limitation of our technical note was our use of a single adult cadaver specimen. This report is a conceptual demonstration, and greater statistical power was needed to support the proposed technique. Because of our experimental setup, we could not isolate the effect of each osteotomy on flexion versus extension range of motion. However, it was reasonable to assume that the bony resection associated with a formal Ponte osteotomy would better facilitate segmental extension through posterior column shortening. Further study is warranted to investigate this hypothesis. Another limitation of this report was that it was performed on a spinal specimen without deformity. Stiffness varied across patients with scoliosis, and the thoracic hypokyphosis or lordosis that is commonly seen in the thoracic spine of patients AIS may result in posterior spinal ligaments that are more stiff and contracted [28]. Posterior releases in different types and severities of scoliotic curves thus may have variable efficacy depending on rib cage deformation, Cobb angle, sagittal and coronal alignment, and vertebral axial rotation. We designed our experimental setup and loading parameters based on existing literature, but it is possible that the range of motion achieved by applying forces to the end vertebra was not directly correlated to the deformity correction that can be achieved when force is applied during surgery through segmental pedicle screws.

Another limitation of our technical note is that the range of motion increases seen from our cadaveric spine may not directly correlate with greater deformity correction achievable in vivo. Few studies have reported the forces necessary to achieve deformity correction during spinal surgery, and thus it is difficult to know how our results translate to clinical practice [29,30]. However, this limitation exists in all biomechanical cadaveric studies on spinal destabilization procedures with simulated loads. The load applied in this report of 5 Nm in each plane falls within the range of 2–6 Nm used in other related studies [24–27,31,32].

It should also be noted that the cadaveric model was stripped of stabilizing paraspinal musculature and separated from the anterior chest wall. A study by Mannen et al. using thoracic cadaver specimens with full rib cages found a significant yet small (<1°/Ponte osteotomy) correction in flexion but no significant axial rotation or lateral bending [31]. In our report, specimen preparation was in line with other studies in the literature but may have demonstrated a greater range of motion increases compared to specimens with a full rib cage or the in vivo setting [27,28,32,33].

Despite these known limitations, our experimental setup represents the best established and feasible cadaveric method to study posterior releases in an in vitro setting. The results from this technical report align with available retrospective clinical studies [12–14].

In vivo, our osteotomy techniques are currently performed freehand without any assistance from advanced technology to facilitate bony resections. However, advances in robotics, computer navigation (NAV), and virtual reality (VR) may one day further improve the execution of spinal corrective osteotomies in AIS [34,35]. VR and NAV have been commonly employed to aid in pedicle screw placement, but less has been published about the use of this technology in performing corrective osteotomies. In one report by Kosterhon et al., they preoperatively created a virtual resection plan for a pedicle subtraction osteotomy. They exported the 3-D plan into a navigation system that could display the planned resection intraoperatively via the surgical microscope's head-up display [36]. While the authors found the intraoperative visualization helpful, they noted that it might be more relevant in patients undergoing large complex osteotomies, such as a pedicle subtraction osteotomy for hemivertebrae. Our sequential osteotomy technique uses smaller bony resections, and we performed intraoperative manual spinal flexibility testing periodically to titrate the number of vertebral levels included and the degree of posterior release; thus, VR and NAV osteotomy planning appear less applicable for our proposed method at this time. However, it is reasonable to expect that these technologies will continue to offer new opportunities to improve the surgical treatment of AIS.

5. Conclusions

Posterior column osteotomies are safe and effective for the multiplanar correction of adolescent idiopathic scoliosis [21]. This report demonstrates a novel posterior spinal osteotomy sequence to progressively improve flexibility while protecting the spinal canal. Our results suggest that this stepwise risk-minimizing approach of the Ponte osteotomy may be adequate to achieve desired deformity correction in many scenarios and align with our clinical experience using this technique. Complete formal Ponte osteotomy can thus be reserved for severe cases or cases in which posterior column compression is necessary for deformity correction. Further cadaveric and clinical studies are needed to confirm our results.

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children

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Article

Musculoskeletal and Gait Characteristics in Patients with Stickler Syndrome: A Cross-Sectional Study

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Abstract: Background: Stickler syndrome (SS) is a connective tissue disorder of fibrillary collagen with very variable clinical manifestations, including premature osteoarthritis and osteopenia. This musculoskeletal alteration may affect gait maturity or produce strength difficulties. Objective: Our aim was to describe the musculoskeletal characteristics, bone stiffness, gait kinematics, and kinetics of SS patients. Methods: This is a cross-sectional study of children and youngsters with SS recruited by telephone calls through the Spanish SS Association. All participants underwent an analysis of musculoskeletal characteristics, including a 3D gait analysis. Results: The sample included 26 SS patients, mainly boys (65.4%) with a median age of 11 (IQR 5–14). The manual muscle testing was normal in 88.5% of patients. The median distance covered in the 6-min walking test was 560.1 ± 113.4 m. Bone stiffness index scores were 70.9 ± 19.7 for children under 10 years and 88.3 ± 17.5 for children older than 10 years. The gait indicators GPS and GDI were: 7.4 ± 1.9 and 95.3 ± 9.7, respectively, for the left side and 6.8 ± 2.0 and 97.7 ± 9.5 for the right side, respectively. Conclusions: In our series of patients with SS, we found muscle-articular involvement does not have a high impact on strength or gait problems. More work is needed to understand the effect of SS on the musculoskeletal system.

Keywords: Stickler syndrome; collagenopathy; 3D gait analysis; quantitative ultrasound stiffness index

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1. Introduction

Stickler syndrome (SS) was described in 1965 by Stickler et al. [1]. It is a dominantly inherited connective tissue disorder of fibrillary collagen with high variability in the manifestation of phenotypes [2,3]. It has an estimated incidence of 1 case per 10,000 births. Collagen is an extracellular fibrous protein that forms part of the connective tissue and is especially abundant in weight-bearing tissues such as cartilage, bone, tendons, fascia, and dermis. It is also the framework for all organs and tissues. There are 40 different genes that encode at least 27 different types of collagen [4].

SS is produced by heterogeneous mutation in four genes that control the synthesis of collagen 2, 9, and 11, so it has a very variable phenotypic expression. The responsible mutations are in COL2A1, COL11A1, COL11A2, and COL9A1 procollagen genes, leading to various degrees of abnormal synthesis collagen types II, XI, or IX [5]. Collagen 2 is found in the greatest proportion in the vitreous humor, cartilage and intervertebral discs. Collagen 9 is associated with type 2 collagen fibrils in mature articular cartilage, the cornea, and vitreous humor. Collagen 11 has a distribution similar to that of type 2. The three types of collagen are found in the cochlea. It is characterized by congenital conditions (i.e.,

megalophthalmos, retinal detachment, deafness, cleft palate, Pierre Robin sequence, joint hypermobility, and premature arthritis) [4].

SS is classified into different types according to the mutated gene, which explains the ophthalmological phenotype and, specifically, the anomalies in the architecture of the vitreous. Based on the vitreous abnormalities, Stickler syndrome is classified as type 1 ("membranous", which is characterized by a persistence of vestigial vitreous gel in the retrolental space) and type 2 ("beaded", which is characterized by sparse and irregularly thickened bundles throughout the vitreous cavity) [6,7].

The clinical manifestations are very variable, generally distributed in four large groups: (A) craniofacial findings: may include a flat facial profile, telecanthus and epicanthal folds, micrognathia, and cleft palate; (B) eye alterations: early cataracts and nonprogressive myopia are common; (C) hearing impairment, especially sensorineural deafness for high tones, is common but overall sensorineural hearing loss in type I Stickler syndrome is typically mild and not significantly progressive [8,9]; and (D) musculoskeletal features, specifically early onset arthropathy, short stature, and mild spondyloepiphyseal dysplasia. In children and adolescents, joint hypermobility is seen and usually becomes less prominent with age. Other manifestations in these patients are skeletal alterations related to orthopedic problems. They also experience frequent spinal abnormalities such as scoliosis, Scheuermann-like kyphosis deformities and spondylolisthesis [10,11]. Premature osteoarthritis and osteopenia are also frequent in these patients. There also appears to be a predisposition to femoral head complications such as Legg–Perthes disease or slipped epiphysis [3,11,12].

The presence of musculoskeletal disorders in children may affect gait maturity or may result in strength difficulties. The aim of this study was to analyze the musculoskeletal characteristics, gait kinematics, and kinetics of SS patients.

2. Materials and Methods

A cross-sectional study was conducted. The study protocol and materials were approved by the ethics committee of the Centro Superior de Estudios Universitarios LaSalle (CSEULS) in Madrid, Spain. The outcomes measured were taken on two days: one for musculoskeletal characteristics and the other for gait analysis and a walking test.

Patients were eligible for the study if they had a medical diagnosis of SS, their age was between 4 and 18 years, and they had the ability to walk at least eight meters. Eligible participants were recruited from December 2017 to March 2018 by telephone call via the Spanish Stickler Syndrome Association roll.

We recruited 26 participants for this study. Patients and their family were received into La Salle M-Lab, signed the informed consent, and were interviewed by one of the researchers. Information was collected on the following three groups of variables:

2.1. Musculoskeletal Characteristics, Muscular Strength, and Functional Tests

Information on the clinical characteristics of the musculoskeletal system (osteoarthritis, joint hiperlaxitude, marfanoid habit, spinal dysplasia, muscular atrophy, etc.) was collected. Muscular strength was evaluated with manual muscle testing (MMT) following the Medical Research Council muscle strength scoring system [13]. MMT is used in rehabilitation and recovery to evaluate contractile units, including muscles and tendons, and their ability to generate forces (score range 0–5; minimum 0, maximum 5/5). A score of 3 or higher is considered normal. In addition, the following functional tests were performed: (a) In the Duncan–Ely test (assessment of rectus femoris spasticity or tightness), the patient lies prone in a relaxed state. The test is positive when the heel cannot touch the gluteus maximus or the hip of the tested side rises from the table [14]. (b) The Galeazzi test or Allis' sign (exploration of hip dislocation or dysplasia) is performed by flexing the infant's knees when they are lying down so that the feet touch the surface and the ankles touch the buttocks. If the knees are not level, the test is positive [15]. (c) Thomas test (assessment of the flexibility of the hip flexors). A test is positive (if the iliopsoas muscle is shortened, or a contracture

is present) when the lower extremity on the involved side is unable to fully extend at the hip [16]. (d) The Silfverskiöld test differentiates gastrocnemius tightness from an Achilles tendon contracture by evaluating ankle dorsiflexion with the knee extended and then flexed [17]. We counted how many of the included patients presented a pathological result on the different tests.

2.2. Calcaneus Quantitative Ultrasound (QUS)

QUS is a quick, cost-efficient, and radiation-free method used to evaluate bone stiffness and indicates the density, structure, and composition of the bone [18,19]. Specifically, the calcaneus was found to be a reliable location to assess bone status. The calcaneus consists of 90% trabecular bone, which shows a high metabolic rate. The bone microarchitecture is similar to that of the lumbar spine and femoral neck, which are major body sites for diagnosing osteoporosis [18]. The results of the QUS of calcaneus are expressed as the stiffness index (SI), a composite of speed of sound (SOS) and broadband ultrasound attenuation (BUA). The stiffness index (SI) is calculated from the BUA and SOS in the Achilles system according to the following equation: $SI = [(0.67 \times BUA + 0.28 \times SOS) - 420]$ [20]. Previously, QUS calcaneal SI showed moderate correlation (r = 0.69) with total body bone mineral density measured by dual-energy X-ray absorptiometry [21]. QUS was measured using an Achilles EXP II system (Getz Healthcare, Bangkok, Thailand).

2.3. Gait and Walking Test

For 3D gait analysis, three-dimensional gait analysis was used for obtaining information on the kinetic (power) and kinematic (joint motion) parameters in the three planes: sagittal, frontal, and transverse. Eight optoelectronic cameras (BTS BioEngeneering SMART-DX 6000) and two BTS P-6000 dynamometric platforms were used to collect kinematic data, sampling at 250 Hz, and the components of forces in the three coordinate axes. The modified Helen–Hayes marker set was used to quantify the kinetics and kinematics of the lower limbs joints [22]. Patients were instructed to: (1) stand on the platform in anatomical position; (2) walk barefoot along an 8-m walkway, with 3 trials collected. Image capture (Smart-Capture BTS BioEngeneering) and Visual3D (Smart-Clinic BTS BioEngeneering) programs were used to track, process, and compare the results with normal values of the kinetics and kinematic data. To confirm the absence or presence of gait pathology, we used the gait profile score (GPS) and gait deviation index (GDI) [23,24]. The GDI is a global measure that provides a numerical value that expresses gait pathology (ranging from 0 to 100, where 100 indicates the absence of gait pathology). The GDI is more complete, less ambiguous, show better statistical performance, and is easier to use than the Gillette gait index [23,25]. In addition, the data obtained during 3D gait analysis were compared with those from a sample of healthy children and adolescents (n = 25) from the Hospital Infantil Universitario Niño Jesús, with the aim of describing the alterations in gait by comparing them with reference values in children and adolescents of similar ages.

A 6-min walking test (6MWT) was also performed. It is a submaximal exercise test that entails measurement of distance walked over a period of 6 min.

2.4. Statistical Analysis

The statistical analysis was carried out using IBM Corp. (Released 2020. IBM SPSS Statistics for Windows, Version 27.0. Armonk, NY, USA: IBM Corp.) Non-parametric tests were used due to the small sample size. Summary measures (median and interquartile range (IQR) for sample characteristics; 95% confidence interval (CI) for results obtained in quantitative measures and percentages for dichotomous outcomes) are used to describe the sample. Possible differences between the groups were explored by the Mann–Whitney U and chi-square tests for quantitative and qualitative variables, respectively. No imputation was performed for missing data.

3. Results

The sample consisted of 26 subjects, of whom 9 (34.6%) were girls, with a median age of 11.0 years (IQR 5–14). The median weight was 40.8 kg (IQR 21.4–55.9) and the median height was 1.5 m (IQR 1.2–1.6). The MMT (muscle examination) showed that almost all patients had normal strength. Of all the muscle groups explored, 88.5% of patients had a value of three or more points (which could be considered normal or almost normal). Only two patients (7.7%) had a muscle group with altered strength, and one patient (3.8%) had two muscle groups with a score of less than three over five in manual muscle testing.

The sample lost was n = 3 for musculoskeletal features, n = 1 for functional test, and n = 4 for 6MWT due to missing data and the fact that some children refused to participate in these measurements.

3.1. Musculoskeletal Characteristics and Physical Examination

Table 1 presents a summary of the main musculoskeletal features of these patients with SS. The most common alteration was ligament hyperlaxity syndrome (30%), followed by osteoarthritis, marfanoid habit, and muscular atrophy (17% each one). In the section of functional tests, the number and percentage of patients with altered tests are shown.

Table 1. Stickler Syndrome's features, functional tests.

Musculoskeletal SS's Features (n = 23)			
Variables	n	Value n (%)	95% confidence interval
Osteoarthritis	23	4 (17)	0.6 to 34.1
Ligament hyperlaxity syndrome	23	7 (30)	10.1 to 50.7
Marfanoid habit	23	4 (17)	0.6 to 34.1
Spinal dysplasia	23	2 (9)	−3.8 to 21.1
Muscular atrophy	23	4 (17)	0.6 to 34.1
Functional Tests (n = 25)			
Variables	n	Value (%)	
Duncan–Ely test	25	3 (12)	
Galeazzi or Allis test	25	2 (8)	
Thomas test	25	4 (16)	
Silfverskiold test	25	1 (4)	
Alteration in any test	25	7 (28)	

Abbreviations: SS = Stickler syndrome.

Twenty-two patients completed the 6MWT. The mean distance covered was 560.1 m (±113.4), with minimum and maximum values of 360 and 729 m, receptively (the median was 575, IQR 458–653). Table 2 shows the results by sex (without significant differences) and by age (the distance travelled by the oldest children being significantly greater; *p* value = 0.02).

Table 2. Bone and gait characteristics of children with Stickler syndrome.

Bone and Gait Characteristics			
Outcomes	Sample (n)	95% CI	Difference
Calcaneus SI	Total (26)	71.8 to 87.4	-
	Male (9) Female (17)	61.7 to 85.1 72.8 to 93.0	−24.9 to 5.9
	Under 10 years (13) Over 10 years (13)	60.2 to 81.6 78.8 to 97.8	**−31.7 to −3.1** *

Table 2. *Cont.*

Bone and Gait Characteristics			
6MWT distance (m)	Total (22)	512.7 to 607.5	-
	Male (7) Female (15)	445.7 to 646.7 512.8 to 620.2	−134.3 to 93.7
	Under 10 years (11) Over 10 years (11)	453.1 to 571.4 543.3 to 672.6	**−183.329 to −8.071** *
GPS	Left side (26) Right side (26)	6.7 to 8.1 6.0 to 7.6	-
GDI	Left side (26) Right side (26)	91.6 to 99.0 94.0 to 101.4	-

Bold values denote statistical significance: (*) $p < 0.05$ level. Abbreviations: CI = confidence interval; GDI = gait deviation index; GPS = gait profile score; MWT = meter walking test; SI = stiffness index; y = years old.

3.2. Calcaneus Quantitative Ultrasound, QUS

QUS was performed in 26 patients. The QUS results were as follows: the median values were 1555.3 (IQR 1545.5–1606.5) for SOS value, 91.9 (IQR 74.8–106.2) for BUA, and 77.5 (IQR 61–99) for SI. Likewise, a subanalysis was carried out by sex (no differences were found) and by age (with significant differences between those younger and older than 10 years; p value = 0.02) (Table 2). Previously, the QUS SI showed a moderate correlation (r = 0.69) with total body bone mineral density measured by dual-energy X-ray absorptiometry [21].

3.3. Gait Analysis (3D Gait Analysis and 6MWT)

Twenty-two children completed the 6MWT. The 95% ICs for total distance covered and difference between sex and age are shown in Table 2. We found significant differences between ages (the distance travelled by the oldest children being significantly greater; p value = 0.02), while the results between sexes showed no significant differences.

Twenty-six children completed 3D gait analysis. The results showed that the average gait pattern of the sagittal and frontal planes was normal for all joints (pelvis, hip, knee, ankle, and foot).

The kinematic graphics in the transversal plane showed torsional alterations in the knee and hip joints. Both tibias were kept in external rotation during all gait cycles (oscillating between -18° and -8° on the right and -12° and -3° on the left), with a difference of 6° between the left (less external rotation) and right. In the hip, the graphics show the opposite of that in the tibia; the internal rotation of the right hip was greater than in the left hip (with a difference of 5° between right and left) (Figure 1).

The power graphs showed a decreased peak during the preswing and take-off phases in the sagittal plane of the ankle. In the knee, the preswing and take-off phases had positive values, whereas in the initial contact, these values were negative. On the hip, there was an absence of peak power during the initial swing phase (Figure 1).

The sagittal moment graphic showed a lowered peak during the end of terminal stance and preswing gait cycles in both ankles. In the moments of the sagittal plane of the right knee, it is important to highlight the increased moment at the beginning of the cycle (initial contact and loading response). In addition, during the end of pre-swing and initial swing, the moment was negative when it should have been positive. Considering knee kinematics, the initial contact and loading response were normal, but during the terminal stance and preswing, the moments stayed positive. The moment on the right hip in the sagittal plane remained stable during all gait cycles, without reaching any peak in the initial contact, final stance, preswing. or terminal swing (Figure 2).

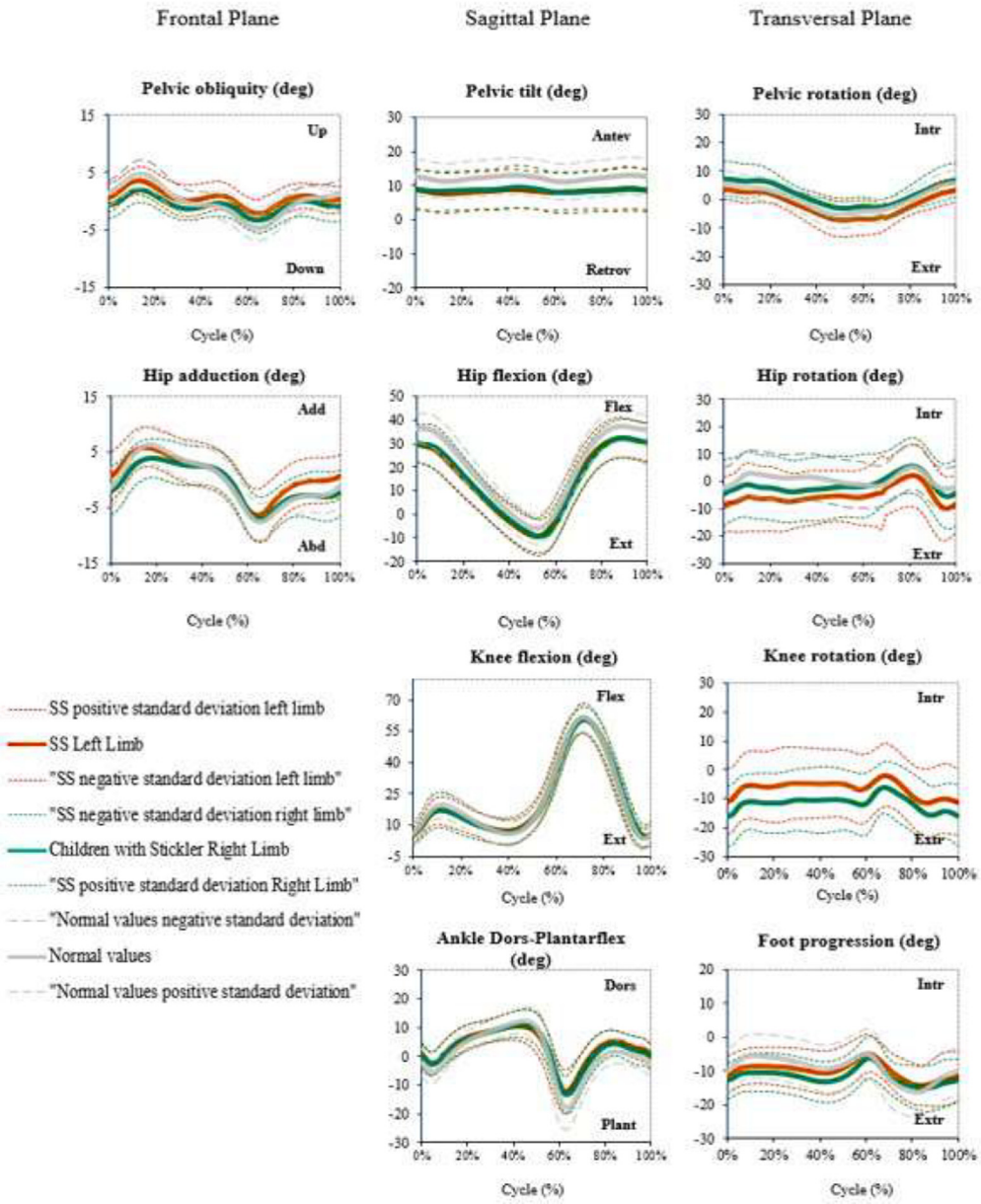

Figure 1. Kinematic 3D gait analysis of patients with SS compared with healthy children and adolescents. Abbreviations: Dors = dorsiflexion, SS = Stickler syndrome.

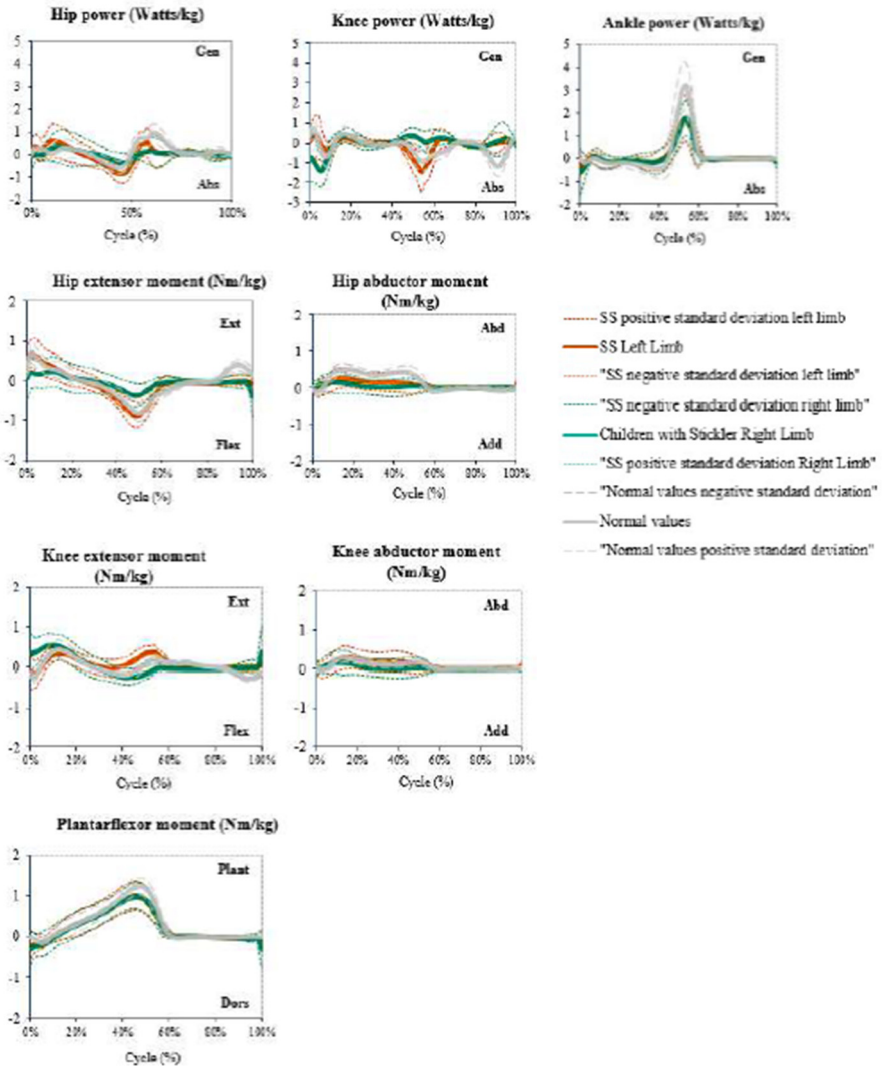

Figure 2. Kinetic analysis of patients with SS compared with healthy children and adolescents. Abbreviations: SS = Stickler syndrome.

The gait indicators GPS and GDI were 7.4 (±1.9) and 95.3 (±9.7), respectively, for the left side and 6.8 (±2.0) and 97.7 (±9.5), respectively, for the right side.

4. Discussion

This paper shows the results of a study of SS patients in whom little musculoskeletal involvement had been observed. Even though 30% of patients were found to have joint hyperlaxity, this finding did not seem to have repercussions on strength, functional tests, or gait alterations. This may have been due to several reasons, among which we would point to the low median age of the series [11] and because diagnosis of SS is perhaps more frequently performed by orofacial involvement and eye and hearing alterations.

The finding that there were significant differences in the distance travelled in the 6MWT according to age may have been due to several factors. Perhaps one of the most important is that younger children have more difficulty walking for this reason alone, regardless of their basal capacity.

Chronic illness, primary bone disease, or poor nutrition in children and adolescents may lead to impaired skeletal health. Approximately 90% of adult bone mass is gained in the first two decades of life, and many experts think that optimizing peak bone mass and bone strength early in life and stabilizing it during young adulthood play a significant role in preventing osteoporosis and fractures later in life. There are several known determinants

of skeletal mineralization and peak bone mass: genetics; race; gonadal status; sleep; and environmental factors such as nutrition and physical activity [21,26–28]. Some reports indicated that osteoporosis is common in SS, but this has not yet been systematically evaluated [11], and for these reasons, calcaneus QUS was measured in this study. In relation to bone mineral density, the SI is lower in children with SS than in European TD children (6 to 12 years (82.06 (12.43); >12 years (97.03 (16.09)) [26]. Similar findings were found in bone mineral density in children with juvenile idiopathic arthritis [29] and osteogenesis imperfecta [30], who presented an increased risk of fractures [30,31]. Moreover, the QUS calcaneal SI shoed moderate correlation (r = 0.69) with total body bone mineral density measured by dual-energy X-ray absorptiometry [21]. These findings probably corroborate the reduction in bone mineral density in SS, which could lead to an increased risk of fractures in childhood [32].

Children with SS showed slightly reduced values compared with TD children in GDI (TD 100 (SD 10) [25] and GPS (TD 5.3 (SD 1.4)) [24]. Moreover, the GPS difference was near to the minimal clinically important difference (MCID), which is 1.6°. We also found reduced ankle power during push-off and knee power at initial contact, while the rest of the joints exhibited results similar to those of TD children. The influence of reduced push-off force during gait has been reported in other studies. Huang T. W. et al. (2015) and Ong C. F. et al. (2019) applied gait simulator models to predict the adaptations to different type of deficits, finding an increase in work from other joints to maintain the gait velocity, while plantar flexor moments were minimally affected by plantar flexor weakness only [33,34]. Although children with SS have preserved plantar flexion strength, it appears that the propulsion strategy performed is not adequate. This means that the energy expenditure during gait will be higher and may be a factor for clinicians to consider when planning treatments.

Related to 6MWT results, we found significant differences in the distance traveled according to age due to several factors. Perhaps one of the most important is that younger children have more difficulty walking for this reason alone, regardless of their basal capacity.

The mean gait distance in 6MWT measured in meters in patients with SS at 95% IC (512 to 607) was lower than that reported in the literature for European TD children (619.8 (SD 58.3)) [24]. This difference was larger than the minimal clinically important difference, which ranges between 22.8 and 31 m [33–36]. Moreover, similar results were reported in children with juvenile idiopathic arthritis (478–602) [37], while this distance was lower in other pathologies that cause musculoskeletal disorders, such as hypophosphatasia (children: 350.4 (92.3); adolescents: 497.3 (98.8)) [33] or osteogenesis imperfecta type I (418 (175.0)) [38]. This finding suggests that children with SS have mild gait-related limitations, which could be a potential treatment target for SS patients. Therefore, this study provides new knowledge about the musculoskeletal characteristics of patients with SS. These findings may be useful to obtain general information on the functional status of these children, helping clinicians to better understand this pathology. However, a larger sample size and follow up are required to determine these bony misalignments as risk factors for joint pain in future studies.

Limitations

Despite being a relatively large series of patients with this rare disease, this study is not without limitations. MMT testing has several disadvantages, including that testing muscle groups is time consuming, patients often experience fatigue during the testing and occasionally experience muscle pain that makes muscle testing unpleasant and stressful, and children frequently are not able to cooperate for the entire muscle group test, resulting in incomplete results or inconsistent strength evaluation. The selection of matched age and sex pairs could be a bias for comparison purposes due to the impact of the disease on children's development. In addition, QUS is not the gold standard for bone mineral density measurement, so results should be interpreted with caution. Finally, the sample size was small, and no imputation was performed for missing data.

5. Conclusions

In this study, we described the musculoskeletal alterations in this sample of patients with SS. The impact of the disease on function is low, reporting less distance covered in the 6-min test and reduced bone mineral density compared with healthy children. Indeed, studies with follow-up periods are necessary to deepen our knowledge of what muscle-articular involvement of SS consists of in children, and the repercussions of this involvement on muscle strength and gait.

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children

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Article

The Predictive Value of Radiographs and the Pirani Score for Later Additional Surgery in Ponseti-Treated Idiopathic Clubfeet, an Observational Cohort Study

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Abstract: There are few validated predictors of the need for additional surgery in idiopathic clubfeet treated according to the Ponseti method. Our aim was to examine if physical examination (Pirani score) and radiographs at the age of three months (after initial correction of the clubfeet) can predict the future need for additional surgery. In this retrospective cohort study, radiographs of idiopathic clubfeet were made at the age of three months. The Pirani score was determined at the first cast, before tenotomy, and at the age of three months. Follow-up was at least five years. The correlation between the radiograph, Pirani score, and the need for additional surgery was explored with logistic regression analysis. Parent satisfaction was measured with a disease-specific instrument. The study included 72 clubfeet (50 children) treated according to the Ponseti method. Additional surgery was needed on 27 feet (38%). A larger lateral tibiocalcaneal angle (i.e., equinus) and a smaller lateral talocalcaneal angle (i.e., hindfoot varus) at the age of three months were correlated with the need for additional surgery. Higher Pirani scores before tenotomy and at the age of three months also correlated with additional surgery. Parent satisfaction was lower in patients who needed additional surgery. Both the Pirani scores and the lateral radiographs are predictive for future additional surgery.

Keywords: clubfoot; radiograph; Pirani score; additional surgery

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1. Introduction

Ponseti's method is internationally regarded as the gold standard for treating idiopathic clubfeet [1]. This method involves weekly manipulations and casting, followed by a tenotomy of the Achilles tendon. Correction is maintained with foot abduction orthoses until the child is four years old [1]. After initial Ponseti treatment, up to 67% of children need repeated casting and/or additional surgery due to relapse [2,3]. Most clubfeet require limited surgery, such as lengthening of the Achilles tendon or transposition of the anterior tibial tendon. Other clubfeet require more extensive surgery, such as partial or complete posteromedial release. In order to customize the follow-up protocol, identification of high-risk cases is needed [1].

Various factors have been indicated as predictors for the need for additional surgery after Ponseti treatment, such as poor evertor muscle activity and brace non-compliance [4]. The Pirani score system is an instrument for assessing the severity of the initial deformation of clubfoot via physical examination [5]. The Pirani score is based on clinical findings of midfoot and hindfoot deformity. Pirani scores can predict the amount of initial cast needed to correct the foot, but there is controversy as to whether this score can predict the need for additional surgery in the future [4,6–8].

Before the Ponseti method became the standard, preoperative radiographs of most children with clubfeet were made in order to optimize surgical planning [9]. Conflicting evidence exists for the ability of these radiographs to predict relapse in Ponseti-treated clubfeet. Kang and O'Halloran found that the angle between the tibia and the calcaneus, which represents equinus deformity, could predict the need for additional surgery [10,11]. However, Richards et al. did not find a relation between this angle and additional surgery rates [12].

Our aim was to examine if the Pirani score and radiographs at the age of three months (after initial correction of the clubfeet) can predict the future need for additional surgery. Furthermore, we assessed if parent satisfaction was related to the need for additional surgery.

2. Materials and Methods

2.1. Patients

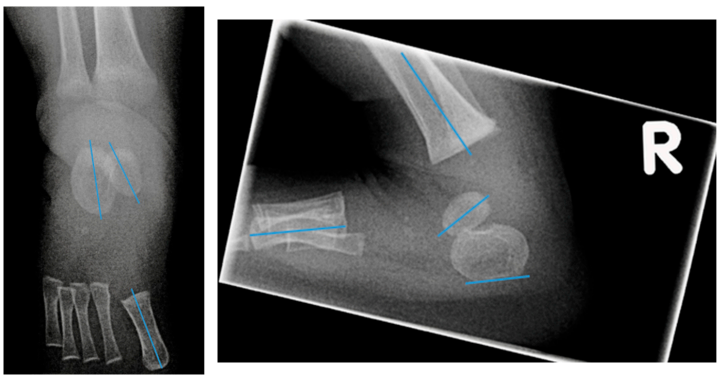
In this retrospective study, we used prospectively collected data to answer our research questions. The electronic database of our hospital was searched for eligible patients. All patients with clubfeet treated via the Ponseti method at Erasmus MC—Sophia Children's Hospital between March 2012 and June 2014 were eligible. Exclusion criteria were (1) children older than 3 months at presentation, (2) non-idiopathic clubfeet, (3) no radiographs at the age of three months available, and (4) follow-up of fewer than 5 years. Follow-up ended in April 2020. If patients moved or were treated elsewhere, parents were contacted. J.B. applied all casts and took Dimeglio and Pirani scores before the first cast, and Pirani scores before tenotomy and at the moment of the radiograph (age 3 months). Treatment and follow-up were performed according to Dutch guidelines [13].

2.2. Radiographs

Anteroposterior (AP) and lateral radiographs were obtained at the age of three months, following the protocol of Simons [9,14,15]. These radiographs were made as part of the standard treatment protocol. The angles were measured by two orthopedic surgeons independently in order to calculate interobserver reliability: (SM, NZK). The average of the two measurements was used to calculate the relation between the angles and the outcome of the clubfeet. The following angles were measured:

On an AP radiograph (Figure 1a):

1. Talocalcaneal angle (AP talocalcaneal): the angle between the long axes of the talus and calcaneus. This measurement describes the eversion of the calcaneus under the talus. A small value indicates hindfoot varus [9].
2. Talo first metatarsal angle (AP talo 1st MT): the angle between the long axis of the talus and 1st metatarsal. This measurement describes forefoot abduction or adduction [9].



(**a**) (**b**)

Figure 1. Radiograph obtained at the age of three months: (**a**) anteroposterior radiograph with lines through the longitudinal axis of the talus, calcaneus, and 1st metatarsal; (**b**) lateral radiograph with lines through longitudinal axis of the tibia, the talus, the 1st metatarsal, and the plantar aspect of the calcaneus.

On a lateral radiograph with maximum dorsiflexion (Figure 1b):

1. Lateral tibial calcaneal angle (lat tibiocalcaneal): the angle between longitudinal axis of the tibia and the plantar aspect of the calcaneus. This measurement describes equinus deformity.
2. Lateral talo calcaneal angle (lat talocalcaneal): the angle between the long axis of the talus and the plantar aspect of the calcaneus. Parallel lines indicate inversion between talus and calcaneus, thus hindfoot varus [9,16,17].
3. Lateral talo first metatarsal angle (lat talo 1st MT): the angle between the long axis of the talus and the first metatarsal. This measurement describes the presence of cavus deformity.
4. Lateral calcaneal 1st metatarsal angle (lat calcaneal 1st MT): the angle between the plantar aspect of the calcaneus and the long axis of the first metatarsal. This measurement also describes the presence of cavus deformity.
5. Foot dorsiflexion between the tibia and a radiolucent wooden board in maximum dorsiflexion (lat foot dorsiflexion). This measurement describes equinus.

2.3. Need for Additional Surgery

Outcomes were measured based on the protocol used by Richards et al. [12]. An excellent result was defined as no additional surgery. A good result was defined as the need for additional Achilles tendon lengthening. A fair result was defined as the need for one or more of the following surgeries: transfer of the tibialis anterior tendon, release of the posterior capsule, or plantar fascia release. These procedures could be combined with Achilles tendon lengthening. A poor result was defined as a full posteromedial release.

2.4. Parent Satisfaction

Parent satisfaction was measured via a disease-specific instrument (DSI), developed by Roye et al. [18] and translated and validated in Dutch by Wijnen et al. [19] The DSI is a ten-item questionnaire designed to measure satisfaction and functional outcome in patients with clubfeet and their parents (Table S1). The DSI was sent via mail to all parents of patients in the cohort in August 2019. If no reply was received, parents were contacted or asked to fill out the DSI during an outpatient visit.

2.5. Statistical Analysis

Categorical data are presented as the absolute number of subjects in each group, along with the percentages. Normally distributed, continuous data are shown as means with a 95% CI of mean and, in the case of a non-parametric distribution, as medians with the interquartile range (IQR). The interobserver reliability between the 2 raters was calculated by assessing the intraclass correlation coefficient (ICC), using a two-way random-effect model with absolute agreement. An ICC below 0.50 was classified as poor, between 0.50 and 0.75 as moderate, between 0.75 and 0.90 as good, and above 0.90 as excellent [20]. Continuous data were analyzed using an unpaired T-test in the case of a normal distribution, and a Mann–Whitney test in the case of a non-parametric distribution. The difference in angles measured on the radiograph and the Pirani scores between the group with additional surgery (good and fair outcomes) and without additional surgery (excellent outcomes) was measured with univariate logistic regression. Multivariate logistic regression with all univariate predictors with a p value > 0.2 was performed. A Bonferroni correction was applied to correct for multiple testing, setting the p-value at 0.0055.

3. Results

3.1. Patients

A total of 76 patients were treated for clubfeet during the study period. Ten of these patients were excluded because they had non-idiopathic clubfeet (two meningomyelocele, two arthrogryposis, one neuromuscular disease, one Kniest syndrome, and four other syndromes). Ten patients were excluded because no radiographs at the age of three months

were available. Two children presented in our hospital at an age older than three months. Four children were lost to follow-up at the ages of 6, 13, 21, and 36 months, respectively. This left a total of 50 children, with 72 clubfeet, available for analysis. Characteristics are described in Table 1.

Table 1. Characteristics of the children and feet of children treated according to Ponseti.

	Children (*n* = 50)	**Clubfoot (*n* = 72)**
Female, *n* (%)	14 (28%)	
Unilateral, *n* (%)	28 (56%)	
Dimeglio score before first cast, median (IQR) [a]		10 (7–12)
Pirani score before first cast		3.8 (3.4–4.1)
Pirani score before tenotomy [b]		1.4 (1.1–1.7)
Pirani score before radiograph [c]		0.3 (0.1–0.5)
Initial tenotomy performed, *n* (%)		55 (76%)
Age at initial tenotomy, weeks		8.3 (7.0–9.5)
Age at radiograph, weeks		14.0 (13.6–14.4)

[a] 35 feet missing data (Dimeglio registration started September 2013) [b] 25 feet missing data, [c] 5 feet missing data. Data are presented as means with 95% CI of mean in between parentheses.

3.2. Need for Additional Surgery

Overall, 45 feet (63%) had excellent results (no need for additional surgery), 9 feet (13%) had good results (8 feet had a single additional Achilles tendon lengthening, and 1 foot had two additional Achilles tendon lengthenings during follow-up); 18 feet (25%) had a fair result (9 of these feet had tibialis anterior transfers, 7 had a tibialis anterior transfer in combination with another procedure, 1 underwent a tibia rotation osteotomy, and 1 underwent a posterior capsule release); no clubfeet had a poor result (Table S2). The average age for additional Achilles tendon lengthening was 3.5 years (95% CI of mean 2.5–4.6). The average age at tibialis anterior transfer was 5.8 years (95% CI of mean 5.1–6.4 years).

3.3. Radiographs

For the angles measured on the radiographs, ICC scores between the two raters were moderate or good except for the AP talo first metatarsal angle (0.09 (95% CI −0.37 to −0.41)) (Table 2). Therefore, this parameter was excluded from further evaluation.

Table 2. Angles measured on radiograph and interrater reliability.

	Mean Angle	**95% CI of Mean**	**ICC Average Measure**
AP talocalcaneal	16.0	13.3–18.7	0.80 (0.68–0.88)
AP talo 1st MT	9.7	6.8–12.5	0.09 (−0.37–0.41)
Lat tibiocalcaneal	59.1	55.4–62.8	0.99 (0.98–0.99)
Lat talocalcaneal	31.2	28.5–33.9	0.94 (0.90–0.96)
Lat talo 1st MT	−26.9	−31.3–22.5	0.89 (0.80–0.94)
Lat calaneal 1st MT	13.4	11.0–15.7	0.85 (0.76–0.91)
Lat foot dorsiflexion	45.7	41.7–49.7	0.99 (0.99–1.00)

AP = anteriorposterior, Lat = lateral, MT = metatarsal, ICC = intraclass correlation coefficient.

We compared the angles measured on the radiographs between the patients with and without the need for additional surgery (Table 3). The lateral tibiocalcaneal angle was smaller in patients without additional surgery. The lateral talocalcaneal angle was larger in patients without additional surgery. The other values showed no differences between the groups. Pirani scores before tenotomy and at the time of radiograph were lower in patients without the need for additional surgery. The multivariate logistic regression did not yield any significant predictors for the need for additional surgery (Table S3).

Table 3. Angles measured on radiographs and Pirani scores in patients with and without need for additional surgery.

	No Additional Surgery		Additional Surgery						
	Excellent (n = 45)		Good (n = 9) Fair (n = 18)		t Test	Univariate Logistic Regression			
	Mean	SD	Mean	SD	p	B	S.E.	Exp (B)	p
AP talocalcaneal	16.4	11.0	15.4	12.2	0.75	−0.00	0.02	1.0	0.73
Lat tibiocalcaneal	54.7	12.0	66.5	18.9	<0.05	0.06	0.02	1.06	0.005
Lat talocalcaneal	33.6	11.3	27.1	10.5	<0.05	−0.06	0.03	0.95	0.025
Lat talo 1st MT	−27.3	20.1	−26.1	15.7	0.51	0.00	0.1	1.0	0.79
Lat calaneal 1st MT	13.7	8.4	12.8	11.8	0.75	0.0	0.03	0.99	0.72
Lat foot dorsiflexion	43.5	14.0	49.8	20.0	0.19	0.02	0.02	1.02	0.15
Pirani score before the 1st cast	3.6	1.5	4.1	1.4	0.09	0.30	0.18	1.35	0.099
Pirani score before tenotomy	1.0	0.6	1.9	1.2	<0.05	1.27	0.50	3.56	0.011
Pirani score before radiograph	0.1	0.3	0.7	1.1	<0.05	1.58	0.63	4.83	0.012

At the age of 3 months, 51 out of 67 clubfeet (76%) were fully corrected (i.e., Pirani score = 0) (Table 4). Fully corrected clubfeet (i.e., Pirani score = 0) had a lower risk of additional surgery (30% vs. 69%). In fully corrected clubfeet, the lateral tibiocalcaneal angle on the lateral maximum dorsiflexed radiograph was smaller (56 vs. 71 degrees, $p < 0.01$), and the talocalcaneal angle was larger (33 vs. 24 degrees, $p < 0.01$).

Table 4. Comparison between patients with fully corrected clubfeet at the age of three months (Pirani = 0) and not fully corrected clubfeet (Pirani > 0).

	Clubfeet Pirani = 0 n = 51	Clubfeet Pirani > 0 n = 16	
Lat talocalcaneal (radiograph)	32.9 (11.7)	24.3 (9.2)	$p < 0.01$
Lat tibiocalcaneal (radiograph)	55.8 (12.8)	71.3 (20.5)	$p < 0.01$
Result excellent	36 (71%)	5 (31%)	
Result good	4 (8%)	4 (25%)	
Result fair	11 (22%)	7 (44%)	

3.4. Parent Satisfaction

A total of 54 DSI scores were collected (74%). The median DSI score was 93 (IQR 79–97). Median DSI satisfaction was 93 (IQR 73–100), and median DSI function was 93 (IQR87–100). There were no differences between the group that filled in a DSI and the group without DSI in sex or laterality (uni- or bilateral). DSI satisfaction was higher in the excellent outcome group ($p < 0.05$) (Figure S1).

4. Discussion

We determined the relation between physical examination (Pirani score), radiographs at the age of three months, and the need for additional surgery in 72 clubfeet treated according to the Ponseti method. Additional surgery was needed for 27 feet (38%). A larger lateral tibiocalcaneal angle (equinus), and a smaller lateral talocalcaneal angle (parallelism, i.e., hindfoot varus) at the age of three months were correlated with additional surgery. A higher Pirani score before tenotomy and at three months was also correlated with additional surgery.

The lateral tibiocalcaneal angle describes the position of the calcaneus in relation to the tibia. Therefore, this is a marker for the length of the Achilles tendon and describes residual equinus deformity. The lateral tibiocalcaneal angle was higher (i.e., more equinus) in patients with additional surgery (67 degrees) compared to patients without additional surgery (55 degrees) $p < 0.05$. Previous studies have also demonstrated that the lateral tibiocalcaneal angle before tenotomy [10,11] and after the boots and bars treatment [16] positively correlate with relapse. Maximum foot dorsiflexion, measured on a lateral radiograph, did not correlate with additional surgery. When a midfoot break is present [10], the angle between the tibia and the radiolucent wooden board under the sole of the foot

can be small, while the 'real' equinus, measured with the tibiocalcaneal angle, is large. This phenomenon might have occurred in our data.

A smaller lateral talocalcaneal angle (parallelism) is an indication of the presence or persistence of inversion between the talus and calcaneus, and thus, hindfoot varus [17]. In our data, a smaller lateral talocalcaneal angle was associated with additional surgery (27.1 vs. 33.6, $p < 0.05$). In addition, a previous study by Shabtai et al. shows that parallelism measured after boots and bars treatment is associated with a higher relapse rate [16]. Richards et al. found that the lateral talocalcaneal angle was larger (i.e., less parallelism) in children with a good outcome, compared to a fair outcome, in 312 clubfeet between the ages of 18 and 24 months (during boots and bars treatment) [12]. Li showed that parallelism decreased after tenotomy, suggesting that a tenotomy can improve subtalar joint alignment [21].

Interrater reliability on the AP radiograph for AP talo first MT was extremely poor in our study [20]. The angle between the talus and the first metatarsal on the AP view was difficult to measure because of the circular shape of the talus on this view at this age. Interrater reliability for angles measured on the lateral radiograph was good or excellent. This is comparable to the recent literature [22].

The Pirani score before the first cast (age usually < 1 week) is known to predict the amount of initial cast needed, but other authors have stated that it cannot be used to predict a future need for additional surgery [4,6,23]. In our study, the Pirani score taken just before the first cast was not predictive of future surgery. A Pirani score taken before tenotomy (average age 8 weeks) and before radiographs (average age 3 months) was predictive for future surgery.

When we compare the fully corrected feet at the time of radiograph (Pirani score = 0, $n = 51$) to the not fully corrected feet (Pirani score > 0, $n = 16$), additional surgery rates are higher in not fully corrected feet (69% vs. 30%). Furthermore, lateral talocalcaneal angles were smaller (i.e., parallelism, thus hindfoot varus), and tibiocalcaneal angles were larger (i.e., more equinus) in the not fully corrected clubfeet. We found that radiographs at the age of three months show which feet were initially fully corrected. The definition of a residual deformity is a deformity that underwent primary treatment but was never fully corrected and needs additional treatment [24]. Clubfoot relapse is defined as any feature of the clubfoot reoccurring after initially successful treatment, which needs additional treatment [1]. We believe that part of the relapse and need for additional surgery we describe in this study is actually the consequence of residual clubfeet. More awareness of these residual clubfeet at an early age and early treatment with re-Ponseti casting might have lowered the number of cases in need of additional surgery [25].

We suggest that early identification of residual clubfeet can be carried out with a carefully performed Pirani score at three months or a lateral radiograph in maximum dorsiflexion at three months, along with measurement of the talocalcaneal and tibiocalcaneal angle. Sriharsha found high correlations between radiographs and the Pirani score [26]. The limited numbers included in this study do not allow us to prove that the Pirani score is better than the radiograph at predicting additional surgery, but the multivariate data indeed suggest it; moreover, radiation could be spared. More research should be performed to confirm this statement.

Parent satisfaction was very high (93 (IQR 79–97)) when we compared them to satisfaction rates other authors found (65–83%) [27–29]. Parents whose children needed additional surgery had a lower satisfaction score. This is of importance, since additional surgery in Ponseti treatment is not regarded as a failure of treatment. When we observe our data, a ceiling effect might have occurred, i.e., the instrument was not able to discriminate differences in mildly impaired individuals [28]. The DSI was developed in 2001 in a surgically treated cohort of clubfoot patients, which are known to have a lower satisfaction rate, while the current cohort was treated with Ponseti casting, and all surgeries were performed extra-articular [18,28,29].

Strengths of this study include the low number of missing data, the length of follow-up (5 to 8 years), and the measurement of parent satisfaction, along with rates of additional surgeries. A limitation is that three patients did not visit the outpatient clinic. Instead, their parents were contacted via telephone. We admit this could have led to an underestimation of the need for additional surgery, as a child could have dynamic supination that is not noticed by the patient or his parents. In 76% of patients, an initial tenotomy was performed, which is less than 85%, as published by Ponseti [1]. However, only three patients in the group that did not receive a tenotomy needed additional surgery (3/27, 11%). Data on brace compliance were not gathered prospectively, although brace compliance is known to be a large predictor of the need for additional surgery [4]. The length of follow-up is considerable, but the need for additional surgery might occur even after this follow-up, especially since the average age of the tibialis anterior transfer was 5.8 years [3]. Finally, the number of included feet is small (72), and the number of explored risk factors is large (9 factors), dictating a Bonferroni correction. With this adjustment, a significant difference was only found in the lateral tibia calcaneal angle and not in the lateral talocalcaneal angle or Pirani scores.

5. Conclusions

A careful physical examination at the age of three months, such as an examination using a Pirani score, is a good method for predicting the need for additional surgery in the future. Lateral radiographs of the foot at the age of three months can also be predictive for additional surgery, probably because they reveal residual deformity much the same as the physical examination. We suggest that a lateral radiograph of the foot might aid when the physical examination is inconclusive, but more research has to be performed.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/children9060865/s1, Table S1: Disease-specific instrument for patients with clubfeet, Table S2: Result according to Richard, Table S3: Multivariate regression analysis of all univariate predictors ($p > 0.2$) of additional surgery, Figure S1: Boxplot DSI scores in outcome groups.

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Data Availability Statement: The data presented in this study are available on request from the corresponding author. The data are not publicly available due to privacy.

Conflicts of Interest: The authors declare no conflict of interest.

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children

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Article

Automated Measurements of Long Leg Radiographs in Pediatric Patients: A Pilot Study to Evaluate an Artificial Intelligence-Based Algorithm

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Abstract: Background: Assessment of long leg radiographs (LLRs) in pediatric orthopedic patients is an important but time-consuming routine task for clinicians. The goal of this study was to evaluate the performance of artificial intelligence (AI)-based leg angle measurement assistant software (LAMA) in measuring LLRs in pediatric patients, compared to traditional manual measurements. Methods: Eligible patients, aged 11 to 18 years old, referred for LLR between January and March 2022 were included. The study comprised 29 patients (58 legs, 377 measurements). The femur length, tibia length, full leg length (FLL), leg length discrepancy (LLD), hip–knee–ankle angle (HKA), mechanical lateral distal femoral angle (mLDFA), and mechanical medial proximal tibial angle (mMPTA) were measured automatically using LAMA and compared to manual measurements of a senior pediatric orthopedic surgeon and an advanced practitioner in radiography. Results: Correct landmark placement with AI was achieved in 76% of the cases for LLD measurements, 88% for FLL and femur length, 91% for mLDFA, 97% for HKA, 98% for mMPTA, and 100% for tibia length. Intraclass correlation coefficients (ICCs) indicated moderate to excellent agreement between AI and manual measurements, ranging from 0.73 (95% confidence interval (CI): 0.54 to 0.84) to 1.00 (95%CI: 1.00 to 1.00). Conclusion: In cases of correct landmark placement, AI-based algorithm measurements on LLRs of pediatric patients showed high agreement with manual measurements.

Keywords: artificial intelligence; leg angle measurement assistant; LAMA; long leg radiographs; pediatric; orthopedics

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1. Introduction

Long leg radiographs (LLRs) serve as a crucial diagnostic tool for assessing bone length, lower limb alignment, and joint line orientation. Specifically in children, LLRs play an important role in the diagnosis and quantification of various limb malalignments and deformities, including genu varum (bow legs), genu valgum (knock knees), and leg length discrepancy (LLD) [1]. However, performing and interpreting length and angle measurements manually on LLRs of patients is very time-consuming for clinicians and prone to intra-and interobserver bias [2–4]. In this study, we evaluated the performance of an artificial intelligence (AI)-based software application for automatic assessment of LLRs in pediatric patients.

LLD and lower limb malalignment are common pediatric orthopedic issues that are associated with various musculoskeletal disorders including gait deviation, scoliosis, low back pain, osteoarthritis, and compromised postural control [5,6]. Although an LLD < 1 cm is often asymptomatic and present in up to 90% of the population, LLD in children can be progressive, and LLD > 2 cm may become symptomatic later in life [6–9]. Similarly, malalignment (e.g., valgus and varus leg angles) developed during childhood may increase

the risk of early osteoarthritis in adulthood [10]. Minimally invasive procedures (i.e., guided growth procedures) are available to manage LLD and lower limb malalignment to prevent future symptoms [8]. LLR measurements play a critical role in the clinical decision-making and follow-up for children treated with guided growth procedures [11–14]. Automation of such measurements with the use of artificial intelligence (AI) techniques, particularly deep learning (DL) algorithms, has the potential to improve the speed, accuracy, and efficiency of these evaluations. This could save time for clinicians and subsequently for patients while at the same time improving the consistency and accuracy of LLR measurements [15–17].

Recent studies have investigated the use, performance, and added value of AI-based algorithms in orthopedic radiology [15]. However, little is known about AI-based measurement programs for LLR in children. We set out to explore an AI-based leg angle measurement assistant (LAMA) that can automate length and angle measurements on LLRs. In previous studies, the performance of this AI-based algorithm has been studied in adults [16–20]. To our knowledge, there are no studies that have used the LAMA software to evaluate a comprehensive set of LLR measurements, including bone length and joint angle measurements, specifically in the pediatric population. The latter is actually one of the largest groups of patients undergoing these radiologic investigations. If the measurements of the AI-based algorithm are consistent with manual measurements, the software may be used in clinical practice as an adjunct or even substitute for the traditional manual measurements, thereby saving valuable time for both clinicians and patients.

Therefore, the aim of this study was to assess the accuracy and reliability of measurements performed by the LAMA software compared to manual measurements on LLRs in patients under the age of 18 years.

2. Materials and Methods

2.1. Patient Inclusion and Image Acquisition

For this observational cohort study, the study population consisted of pediatric patients referred for LLR between January and March 2022. Patients were included if they were aged 11 to 18 years at the time of LLR. The following exclusion criteria were used: visual artifacts or poor visibility on radiographs, incorrect positioning, non-weight-bearing or abnormal cropping. Abnormal cropping refers to the issue where an LLR does not capture the entire area of the leg(s) or where part of the leg(s) is unintentionally excluded from the radiograph. This can occur due to incorrect patient positioning, improper image capture settings, or technical errors during the imaging process. Artifacts were defined by abnormal or misleading image features that were not caused by the patient's anatomy. LLRs were acquired in a standardized, weight-bearing manner using a digital Aseco+ X-ray system with CXDI detectors (Canon Medical Systems Corporation, Otawara, Japan). The imaging parameters included a tube voltage of 85 kVp and a tube current of 450 mA. Three separate X-ray images were taken: (1) pelvis to mid-femur, (2) mid-femur to mid-tibia, and (3) mid-tibia to foot. These images were subsequently stitched together to create a single LLR image.

2.2. Measurements

All measurements on the LLRs were performed by two observers and LAMA. Firstly, a senior pediatric orthopedic surgeon (>5 years of experience) and an advanced practitioner in radiography (>15 years of experience) independently performed all measurements manually. Observers were blinded to each other's measurements. Secondly, automatic assessment of LLRs was performed using commercially available software based on DL technology (LAMA, version 1.03, ImageBiopsy Lab, Vienna, Austria). The LAMA application was trained on a dataset comprising over 15,000 radiographs sourced from various studies, including the Osteoarthritis Initiative, the Multicenter Osteoarthritis Study, the Cohort Hip and Cohort Knee study, and five sites in Austria [21–23]. The training cohort contained LLRs from adult patients of different ages and ethnic backgrounds and data acquired using different radiography systems. In order to perform the measurements, the

LAMA application identifies anatomical bony landmarks and provides measurements of angles and lengths. For additional details on the model training, readers are directed to the supplement provided by Simon et al. [17].

2.3. Image Analysis

Manual assessment of LLRs was performed using IDS7 software (Sectra AB, IDS7 version 25.2, Linköping, Sweden). With regard to automated measurements, results of the LAMA analysis were visually evaluated in the IDS7 software to assess the correct placement of landmarks, including the top of the femoral head, the medial femoral condyle, the mid-tibial roof, the mechanical axis of the tibia, the proximal tibial knee joint orientation line, the mechanical axis of the femur, and the distal femoral knee joint orientation line (Table 1). With these landmarks, the following measurements were obtained: femur length, tibia length, full leg length (FLL), leg length discrepancy (LLD), hip–knee–ankle angle (HKA), mechanical lateral distal femoral angle (mLDFA), and mechanical medial proximal tibial angle (mMPTA) (Figure 1). Cases with incorrect measurements due to the inability of LAMA to identify the correct landmarks were excluded.

Table 1. Overview of landmarks and measurement variables.

Landmarks	Description
1. Top of the femoral head	Most superior point of the femoral head
2. Medial femoral condyle	Most distal point of the medial femoral condyle
3. Mid-tibial roof	Middle of tibial plafond in the tibiotalar joint
4. Mechanical axis of the tibia	Axis passing through the center of the ankle joint and the midpoint of the knee joint
5. Proximal tibial knee joint orientation line	Line crossing the two lowest points of the tibia plateau
6. Mechanical axis of the femur	Axis passing through the center of the femoral head and the midpoint of the knee joint
7. Distal femoral knee joint orientation line	Line passing through the most distal points of the femoral condyles

Measurement Variables	Description	Landmarks Used
Femur length	Distance between the most superior point of the femoral head and the most distal point of the medial femur condyle	1 and 2
Tibia length	Distance between the most distal point of the medial femoral condyle and mid-tibial roof	2 and 3
Full leg length	Distance between the most superior point of the femoral head and mid-tibial roof	1 and 3
Leg length discrepancy	Difference between the full leg lengths of both legs within the same patient	1 and 3
mMPTA	Angle between the mechanical tibial axis and proximal tibial knee joint orientation line	4 and 5
mLDFA	Angle between the mechanical femoral axis and the distal femoral joint orientation line	6 and 7
HKA	Angle between the mechanical femoral and tibial axes	6 and 4

mMPTA, mechanical medial proximal tibial angle; mLDFA, mechanical lateral distal femoral angle; HKA, hip–knee–ankle angle.

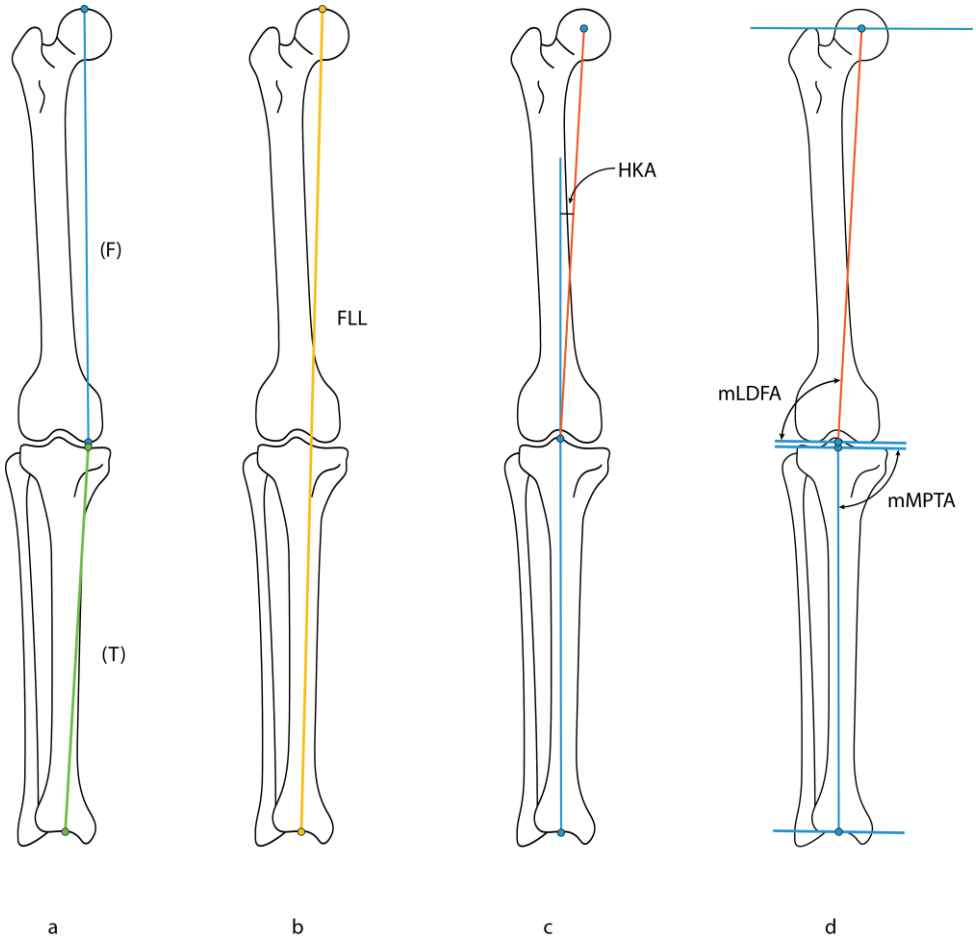

Figure 1. Schematic overview of measurements performed on long leg radiographs (LRRs), including (**a**) femur length (F) and tibia length (T), (**b**) full leg length (FLL), (**c**) hip–knee–ankle angle (HKA), (**d**) mechanical lateral distal femoral angle (mLDFA), and mechanical medial proximal tibial angle (mMPTA).

2.4. Statistical Analyses

Patient demographics and distributions of all measurements (2 observers) were evaluated using descriptive statistics: means with standard deviations (in case of normally distributed data) and medians with ranges (in case data were not normally distributed). The primary outcome was correct landmark placement by LAMA. The secondary outcome of this study was the comparison of quantitative analyses (i.e., agreement) between LAMA and manually performed measurements. The manually performed measurements of 2 observers were compared to each other and to the LAMA results.

Paired *t*-tests were used to compare the mean of manual measurements (2 observers) to the measurements obtained by using LAMA for each of the 29 patients (total of 58 legs). For analysis of LLD, left and right legs were compared. Analysis of interobserver agreement was performed by comparing manual measurements between the two observers. Agreement was determined by calculating the intraclass correlation coefficients (ICCs) [24]. We assessed the agreement between the observers using an absolute agreement ICC in a two-way random effects model [25]. Furthermore, agreement was determined between the mean values of the manual measurements (of the two observers) and measurements obtained by LAMA. The agreement between the manual and AI measurements was assessed using an absolute agreement ICC in a two-way mixed effects model [25]. The categorization used for interpreting the ICC values were as follows: values less than 0.50—poor reliability, between 0.50 and 0.75—moderate reliability, between 0.75 and 0.90—good reliability, and greater than 0.90—excellent reliability [25]. Furthermore, manual and LAMA measurements were

visually presented using Bland–Altman plots, including the lower and upper limits of the 95% confidence interval (CI) of agreement [26]. Analyses were performed using SPSS (version 29.0, IBM SPSS Statistics, Armonk, NY, USA).

3. Results

A total of 29 patients (58 legs) were included in this study. The median age of the patients was 13.7 years (range 12–16). Furthermore, a total of 12 patients were male (41%) and 17 were female (59%). LLRs were taken for (suspected) LLD (12 patients), tall stature (11 patients), screening for fibrous dysplasia (1 patient), genu valgum (3 patients), or genu varum (2 patients). In a total of eight LLRs, there was erroneous placement of the anatomical landmarks by LAMA. Figure 2 demonstrates a number of these erroneously placed landmarks.

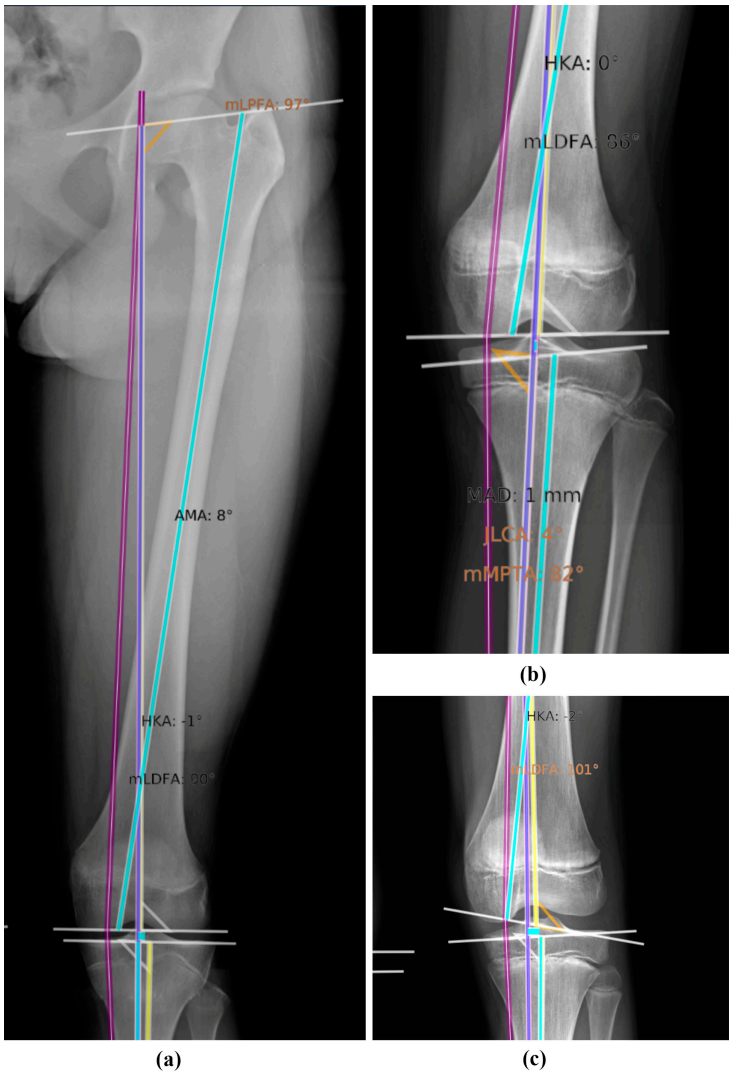

Figure 2. Examples of erroneously placed landmarks by the leg angle measurement assistant (LAMA) showing incorrect identification of (**a**) the top of the femoral head and femoral head center, (**b**) the proximal tibial knee joint orientation line, (**c**) the distal femoral knee joint orientation line.

The erroneous placements were due to failure in the identification of the top of the femoral head (for length measurements) in seven legs, the femoral head center (for angle measurements) in five legs, the placement of the distal femoral knee joint orientation line in one leg, and the proximal tibial knee joint orientation line in one leg. This led to the exclusion of the following LAMA angle measurements: two HKA, five mLDFA, and one

mMPTA measurement in six LLRs. With regard to length measurements by LAMA, seven femur lengths, seven FLLs, and seven LLDs (22 of 29 pairs of legs had correct landmark placements in both legs) were omitted. Landmark placement was correct in 91% to 98% of the cases with regard to angle measurements and in 76 to 100% with regard to length measurements (Table 2).

Table 2. Overview of correct landmark placements with AI software per measurement variable.

Measurement Variables	cLMP (%)	Legs Analyzed (n)
Femur length	88%	51
Tibia length	100%	58
FLL	88%	51
LLD	76%	44
mMPTA	98%	57
mLDFA	91%	53
HKA	97%	56

cLMP, correct landmark placement; FLL, full leg length; LLD, leg length discrepancy; mMPTA, mechanical medial proximal tibial angle; mLDFA, mechanical lateral distal femoral angle; HKA, hip–knee–ankle angle.

3.1. Comparison of Manual Observations

One observer (an advanced practitioner in radiography) reported slightly higher mean values for HKA and mLDFA, while the second observer (a pediatric orthopedic surgeon) measured higher means for LLD. However, the differences between both observers for all measurement variables were small (and considered clinically not to be relevant) and statistically non-significant. Moderate to excellent agreement between the observers for all measurement variables was observed (Table 3).

Table 3. Mean measurement and interobserver agreement of manual measurements on long leg radiographs (LLRs).

Measurements	Legs Analyzed (n)	Observer 1 (AP)	Observer 2 (OS)	Mean Difference	ICC
Femur length [mm]	51	512.2 (47.6)	512.2 (47.8)	0.0 (−0.4 to 0.5)	1.00 (1.00 to 1.00)
Tibia length [mm]	58	410.6 (43.3)	410.5 (43.3)	0.1 (−0.1 to 0.4)	1.00 (1.00 to 1.00)
FLL [mm]	51	923.5 (89.7)	923.5 (89.6)	0.0 (−0.5 to 0.4)	1.00 (1.00 to 1.00)
LLD [mm]	44	8.9 (9.2)	9.1 (9.2)	−0.2 (−0.8 to 0.3)	0.99 (0.98 to 1.00)
mMPTA [°]	57	88.6 (2.0)	88.5 (1.9)	0.1 (−0.2 to 0.4)	0.88 (0.79 to 0.93)
mLDFA [°]	53	87.0 (1.9)	86.4 (2.0)	0.8 (0.5 to 1.0)	0.91 (0.62 to 0.97)
HKA [°]	56	−0.2 (2.5)	−0.3 (2.5)	0.2 (0.0 to 0.3)	0.98 (0.97 to 0.99)

Measurements of both observers are presented as mean with standard deviation. Mean differences and intraclass correlation coefficients (ICCs) are presented with a 95% confidence interval. Lengths are presented in millimeters and angles in degrees. AP, advanced practitioner in radiography; OS, orthopedic surgeon; FLL, full leg length; LLD, leg length discrepancy; mMPTA, mechanical medial proximal tibial angle; mLDFA, mechanical lateral distal femoral angle; HKA, hip–knee–ankle angle.

3.2. Comparison of LAMA with Manual LLR Measurements

After the exclusion of measurements with erroneously placed landmarks, the LAMA software showed comparable mean values for all lengths and angles compared to the mean manual measurements. For length measurements, the agreement was excellent (ICC $\geq$ 0.99). For angle measurements, the ICC ranged from moderate to excellent agreement (ICC 0.73 (95%CI 0.54 to 0.84) to 0.97 (95%CI 0.70 to 0.99)) (Table 4). Based on inspection of the Bland–Altman plots, the difference in femur length measurements seemed to increase with larger femoral length (Bland–Altman Figure S1, see Supplementary Material). For the mMPTA, the difference between LAMA and manual measurements seemed to increase when the angle became smaller (Bland–Altman Figure S5, see Supplementary Material).

Table 4. Comparison of mean long leg radiograph (LLR) measurements performed by two observers with the measurements obtained by the leg angle measurement assistant (LAMA).

Measurements	Legs Analyzed (n)	LAMA	Manual	Mean Difference	ICC
Femur length [mm]	51	511.3 (47.3)	512.2 (47.7)	0.9 (0.6 to 1.2)	1.00 (1.00 to 1.00)
Tibia length [mm]	58	411.4 (43.6)	410.6 (43.3)	−0.9 (−1.1 to −0.6)	1.00 (1.00 to 1.00)
FLL [mm]	51	923.8 (89.8)	923.5 (89.7)	−0.3 (−0.7 to 0.1)	1.00 (1.00 to 1.00)
LLD [mm]	44	9.0 (9.4)	9.0 (9.1)	0.0 (−0.5 to 0.4)	0.99 (0.99 to 1.00)
mMPTA [°]	57	87.4 (2.8)	88.5 (1.8)	1.2 (0.7 to 1.6)	0.73 (0.54 to 0.84)
mLDFA [°]	53	87.1 (2.2)	86.8 (1.9)	−0.3 (−0.6 to 0.0)	0.91 (0.84 to 0.95)
HKA [°]	56	0.3 (2.5)	−0.3 (2.5)	−0.6 (−0.8 to −0.4)	0.97 (0.82 to 0.99)

Measurements obtained with LAMA or manually are presented as mean with standard deviation. Mean differences and ICCs are presented with a 95% confidence interval. Lengths are presented in millimeters and angles in degrees. The mean values of the measurements of the two observers are compared to the measurements obtained with the leg angle measurement assistant (LAMA). Lengths are presented in millimeters and angles in degrees. FLL, full leg length; LLD, leg length discrepancy; mMPTA, mechanical medial proximal tibial angle; mLDFA, mechanical lateral distal femoral angle; HKA, hip–knee–ankle angle.

4. Discussion

LLRs are frequently performed on children to assess and follow up on leg alignment and length differences, as well as for surgical planning. However, performing length and angle measurements on these radiographs is labor-intensive and time-consuming. To our knowledge, this was the first study in pediatric patients to assess the performance of automatically analyzed LLRs using a DL-based LAMA software application compared to manually annotated LLRs by two observers. We found that LAMA was able to accurately identify the anatomical landmarks that are needed for length and angle measurements in the vast majority of cases. However, because correct landmark placement ranged between 76% and 100% for different LLR measurements, LAMA should not be used in clinical practice to analyze the LLR of pediatric patients without oversight of landmark placement by a clinician. In cases where landmark placement was correct, the agreement between LLR measurements obtained with LAMA and manual measurements was high, as depicted in the ICCs for both length and angle measurements.

Considering the existing literature on LAMA, Schwarz et al. reported correct landmark placement in 92% and produced an output rate (angle measurements) of 96% [16]. Simon et al. found an overall accurate landmark placement in 89% of cases and a higher output rate (length and angle measurements) of 98.0% [17]. Although we obtained an output rate in all of the cases with LAMA, the percentage of correctly placed anatomical landmarks in our study was slightly lower compared with these studies in adult patients. One explanation could be that ossification is still ongoing in (younger) children or that children with the indication for LLR have anatomical abnormalities (i.e., LLD or varus/valgus due to an underlying disease), making it difficult for LAMA to identify the right landmarks and draw the correct lines. Based on some of the observed erroneous landmark placements in our study by LAMA on the proximal tibial knee joint orientation line, it may seem that it can be difficult to find the most distal point in the tibia plateau groove in children (as depicted in Figure 2b). Also, for the human eye, finding the exact most distal central point of a reasonably flat surface on a 2D image can be rather difficult. Thus, resulting in more variability between human observers and LAMA. The rate of correct landmark placement may be further improved by providing data for the retraining of skeletally immature patients.

Length measurements with LAMA resulted in high output rates and landmark placements, except for LLD. As shown, correct landmark placement was considerably lower in cases of LLD measurements. To obtain the LLD, two correctly measured FLLs of a patient are needed (i.e., from both legs), requiring four correct landmarks for each measurement. When a single landmark required for FLL in one leg is incorrectly placed, LLD cannot be determined, thus explaining the somewhat higher exclusion rate for LLD measurements in our study. Also, the top of the femur head is one of the landmarks that is needed to

calculate FLL and LLD. The presence of a deformed femoral head in children who have been identified as needing an LLR may hamper the correct identification of the top of the femur head landmark by LAMA.

With regard to angle measurements, specifically, mMPTA measurements appeared difficult in our dataset, as reflected by lower ICC compared to other measurements. This finding does not correspond with Archer et al., who evaluated the agreement in LLD and knee alignment measurements between LAMA AI software and two manual observers in adult patients [20]. The latter study reported an ICC of 0.89 (95%CI 0.85 to 0.92) for mMPTA when comparing the output of an AI model with manual observers. Another study by Erne et al. on adult patients, using an algorithm based on AI for automated leg measurements on LLR, also showed a higher ICC for mMPTA (ICC > 0.83) between the AI model and manual measurements compared with our study [27].

A recent study conducted by Zheng et al. investigated a different DL-based model on an LLR dataset of children and found a high consistency of LLD measurements between automated DL-based and manual measurements (Pearson correlation coefficient (r) 0.94) [28]. Although r and ICC evaluate validity and reliability, they highlight different aspects, hampering a direct comparison of their results to the findings of our study. Whereas r assesses a linear relationship, ICC provides an absolute and more robust agreement between the two methods. Lastly, de Villeneuve et al. compared an algorithm based on a machine learning process with 11 orthopedic surgeons and found mean differences for mLDFA of 2.1°, MPTA 1.6°, and HKA 1.3° [29]. In our study, similar differences between manual and LAMA measurements were found.

Our results suggest that AI applications like LAMA have the potential to enhance the time efficiency of LLR assessment in the pediatric orthopedic setting. This could be a significant gain in high-volume clinics, as it reduces the time and effort that is required for leg length and joint angle measurements. Furthermore, improved efficiency and reduced variability could potentially lead to better patient outcomes by enabling early and accurate detection of deviating growth in pediatric patients. As for imaging, the precision of measurements may be further improved by using Cone–Beam Computed Tomography (CBCT) or low-dose biplanar digital X-ray systems [30], hereby eliminating factors such as rotation and fan effect distortion [31].

There are some limitations of our study to take into account. Firstly, given that the measurements performed by the senior pediatric orthopedic surgeon and advanced practitioner in radiography are not deemed flawless, the question arises as to whether measurement discrepancies can be attributed to the flawed measurements of LAMA or variability and inaccuracy of the observers (i.e., there is no gold standard). To assess the reliability of LAMA fully, it is important to underline that both inter- and intraobserver variability exist in manual observations. Whereas automated software applications, such as LAMA, will always provide the exact same measurements (i.e., there is no intraobserver variability), intraobserver variability will be present for manual observers. We found moderate to excellent interobserver agreement for all length and angle measurements on LLRs in our cohort of children, but we did not assess the intraobserver variability within the observers. However, the possible intraobserver variability within the manual observers is expected to be very small [4,32]. Therefore, we do not expect that the latter would have significantly influenced our findings with regard to the agreement between LAMA and manually performed measurements. Another limitation is that the reason for landmark misplacement by LAMA is not always clear due to the opaque nature of the software application. In our study, a case was observed where the hip joint anatomy was abnormal, and LAMA completely missed this landmark. In the latter example, it may be clear why the placement of a specific landmark went wrong, but in other cases without obvious osseous or other structural deformations or image artifacts, it may not always be clear why the placement of specific landmarks was performed erroneously. The use of explainable AI methods could help to understand the reason for incorrect landmark placement and indicate what could be done to improve landmark placement by the addition of specific

data for retraining the model. Lastly, our sample size was relatively small, and we collected patients from a single center, which may limit the generalizability of our findings.

5. Conclusions

In conclusion, our findings suggest that the LAMA software is a reliable tool for LLR measurements in a pediatric setting that can potentially save valuable time for the treating physician. The LAMA software application demonstrated correct landmark placement in 91% to 98% of cases with regard to angle measurements and in 76 to 100% with regard to length measurements. The latter underlines that manual oversight of landmark placement by the LAMA software in LLRs of pediatric patients is important. If the landmark placement was correct, high agreement of LAMA with manually performed measurements on LLRs was observed.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/children11101182/s1, Figure S1: Bland-Altman plot of the artificial intelligence (AI) and manual femur length measurements. Figure S2: Bland-Altman plot of the artificial intelligence (AI) and manual tibia length measurements. Figure S3: Bland-Altman plot of the artificial intelligence (AI) and manual FLL measurements. Figure S4: Bland-Altman plot of the artificial intelligence (AI) and manual LLD measurements. Figure S5: Bland-Altman plot of the artificial intelligence (AI) and manual mMPTA measurements. Figure S6: Bland-Altman plot of the artificial intelligence (AI) and manual mLDFA measurements. Figure S7: Bland-Altman plot of the artificial intelligence (AI) and manual HKA measurements.

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