Gait & Posture 90 (2021) 415–421

Contents lists available at ScienceDirect

Gait & Posture

journal homepage: www.elsevier.com/locate/gaitpost

Full length article

Forefoot adduction and forefoot supination as kinematic indicators of relapse clubfoot

L. Grin[a,b,*], M.C. van der Steen[c,d], S.D.N. Wijnands[a,c], L. van Oorschot[b], A.T. Besselaar[c,d], B. Vanwanseele[a,b]

[a] KU Leuven, Department of Movement Sciences, Tervuursevest 101, 3001, Heverlee, Belgium
[b] Fontys University of Applied Sciences, Dominee Theodoor Fliednerstraat 2, 5361 BN, Eindhoven, the Netherlands
[c] Department of Orthopaedic Surgery & Trauma, Máxima Medical Center, Postbus 90052, 5600 PD, Eindhoven, the Netherlands
[d] Department of Orthopaedic Surgery & Trauma, Catharina Hospital Eindhoven, Postbus 1350, 5602 ZA, Eindhoven, the Netherlands

ARTICLE INFO

Keywords:
Relapse
Clubfoot
Gait analysis
Ponseti
Kinematics
Multi segment foot model

ABSTRACT

Background: Understanding the kinematic characteristics of relapse clubfoot compared to successfully treated clubfoot could aid early identification of a relapse and improve treatment planning. The usage of a multi segment foot model is essential in order to grasp the full complexity of the multi-planar and multi-joint deformity of the clubfoot.

Research question: The purpose of this study was to identify differences in foot kinematics, using a multi-segment foot model, during gait between patients with Ponseti treated clubfoot with and without a relapse and age-matched healthy controls.

Methods: A cross-sectional study was carried out including 11 patients with relapse clubfoot, 11 patients with clubfoot and 15 controls. Gait analysis was performed using an extended Helen Hayes model combined with the Oxford Foot Model. Statistical analysis included statistical parametric mapping and discrete analysis of kinematic gait parameters of the pelvis, hip, knee, ankle, hindfoot and forefoot in the sagittal, frontal and transversal plane.

Results: The relapse group showed significantly increased forefoot adduction in relation with the hindfoot and the tibia. Furthermore, this group showed increased forefoot supination in relation with the tibia during stance, whereas during swing increased forefoot supination in relation with the hindfoot was found in patients with relapse clubfoot compared with non-relapse clubfoot.

Significance: Forefoot adduction and forefoot supination could be kinematic indicators of relapse clubfoot, which might be useful in early identification of a relapse clubfoot. Subsequently, this could aid the optimization of clinical decision making and treatment planning for children with clubfoot.

1. Introduction

Idiopathic clubfoot (talipes equinovarus) is a three dimensional foot deformity characterised by equinus, varus of the hindfoot, cavus and forefoot adductus [1]. The incidence of clubfoot in Europe ranges between 1.09 and 1.52 per 1000 children [2–4]. Nowadays, the Ponseti method is considered as the golden standard for the initial treatment of clubfoot [1,5]. This treatment initially achieves a normal looking, functional and painless foot in approximately 90 % of the children [6–8]. Despite good clinical outcomes of the Ponseti method, 20–41 % of the children with initially successfully corrected clubfoot will face reoccurrence of one or more aspects of the initial clubfoot [2]. This reoccurrence, also known as a relapse, causes functional and pain-related problems. Thereby, possible additional surgical interventions could negatively affect pain, functionality, and cosmetics [9]. Early identification, and therefore objective characterization of the specific foot impairments, of a relapse is essential to prevent the need for additional –surgical-interventions and improve treatment [10].

Gait analysis is frequently used to identify gait characteristics of children with clubfoot as an objective evaluation of treatment outcome. Literature shows multiple deviations in functional gait of children with a clubfoot compared to typically developing children. Observed differences are increased dorsiflexion during stance [11], less maximum dorsiflexion during swing [12–14], decreased ankle range of motion [11,

* Corresponding author at: Fontys University of Applied Sciences, Dominee Theodoor Fliednerstraat 2, 5361 BN, Eindhoven, the Netherlands.
E-mail address: l.grin@fontys.nl (L. Grin).

https://doi.org/10.1016/j.gaitpost.2021.09.185
Received 24 March 2021; Received in revised form 7 September 2021; Accepted 16 September 2021
Available online 20 September 2021

12,14,15] and increased internal rotated foot progression angle [11–16]. Furthermore, previous studies have shown that the gait pattern of children with clubfoot is affected as a result of compensation mechanisms. Such compensations are knee hyperextension [13,14] and increased external hip rotation [11,12,14–16]. However, there is only limited research available reporting gait analysis in cases of relapse clubfoot [17–20]. These studies reported impaired ankle joint kinematics, compensatory external hip rotation and increased pelvic transversal plane range of motion, but only one study used a multi-segment foot model [20]. Consequently, little is known about detailed foot kinematics of children with (relapse) clubfoot.

The usage of multi-segment foot models is, although of importance considering the characteristics of the clubfoot, rare. From a clinical point of view, applying a multi-segment foot model besides a standard lower extremity model during the 3D gait analysis is essential in order to grasp the full complexity of this multi-planar and multi-joint deformity. By dividing the foot in multiple segments based on anatomical references, foot models allow for detailed analyses of hindfoot and forefoot motion. This is especially important in complex movements such as intoeing [14]. Since intoeing [19] is a known problem in patients with relapse clubfoot, a multi-segment foot model is required to provide complete insight in the kinematics within –relapse–clubfeet. Especially when one considers more complex foot deformities that often occur in case of a relapse over the age of 5 [10]. More detailed information of the deformity could aid clinical decision-making. Previous research already showed that the added information from gait analyses led to adapted treatment protocols in previously surgically treated clubfoot [17,21]. Moreover, as far as we are aware, no study focused on the characteristics of relapse clubfoot compared to successfully treated clubfoot. This comparison could possibly lead to kinematic indicators of a relapse, which could aid early identification.

Hence, the main goal of this study is to identify differences in foot kinematics, using a multi-segment foot model, during gait between patients with Ponseti treated clubfoot with and without a relapse and age-matched controls. In order to obtain further insights in possible compensation mechanisms for the impaired foot kinematics, kinematics of the entire lower extremity will be investigated. On one hand we aim to investigate the difference in kinematic pattern to specify changes in joint angles in the gait cycle timeline. In addition, we also want to study changes in peak and mean values to assess standard kinematic gait parameters.

2. Methods

2.1. Participants

A convenience sample of twenty-two children with unilateral or bilateral clubfoot was recruited at the Catharina Hospital and Máxima MC (Veldhoven, the Netherlands) by an orthopedic surgeon (AB) specialized in the treatment of clubfeet. Furthermore, a control group (n = 15) consisting of typically developing children was recruited via the researchers' network. All children met the inclusion criteria: they were between the age of 4 and 8 years and were able to follow instructions properly. For the patients with clubfoot additional inclusion criteria were: initial treatment with the Ponseti method and an idiopathic clubfoot. Furthermore, in case of bilateral clubfoot only the most affected foot was measured. The exclusion criteria for patients with clubfoot were prior treatment other than the Ponseti method and additional surgical treatment following initial clubfoot treatment. In this light, Achilles tendon tenotomy (AT) or renewed AT before the age of three were not considered as additional surgical treatment. Syndromic, neurogenic, or positional clubfoot were excluded.

Patients with clubfoot were divided in two groups by the treating orthopedic surgeon: patients with successfully treated clubfoot (corrected group, n = 11) and patients with relapse clubfoot (relapse group, n = 11). A relapse clubfoot was defined as a reoccurrence of one or more aspects of the initial clubfoot, needing additional treatment. Planned additional treatment could consist of repeated casting and/or bracing, additional physiotherapy or surgical treatment. Patients who received surgical relapse treatment prior to the measurement were excluded.

The study protocol was approved by the Medical research Ethics Committees United (METC NL53229.100.15/ 2014-69, MEC-U, Eindhoven, The Netherlands). Parents of the participants signed the informed consent form prior to data acquisition.

2.2. Data capture

Gait analysis was performed using a wireless active 3D-system (Codamotion Ltd., CX1, sampling rate: 100 Hz), including four tripod cameras and 25 infrared markers. Markers were placed according to an extended Helen Hayes model combined with the Oxford Foot Model (OFM) [22,23]. A recessed force plate (Advanced Mechanical Technology Inc. OR 6–7, sampling frequency: 500 Hz) measured the ground reaction force for one step per trial.

2.3. Measurement protocol

Two investigators (LG, LO) placed the markers on the right leg of the control children and on the most affected leg in patients with clubfoot. Participants walked barefoot in a straight line at a self-selected walking speed along an 8 m walkway. Measurements started when participants felt comfortable walking with all markers. Data from three to five consistent gait cycles (full foot contact on the force plate) were analyzed. Ground reaction force was used to determine initial contact and toe off (threshold: 10 N). In two children no proper force data was available due to small step length. In those cases the gait cycle was determined using velocity data. Kinematic modelling was done using the Helen Hayes model and the Oxford Foot Model (OFM) in Odin (Codamotion Ltd.). The Helen Hayes model was used to analyze the conventional gait kinematics of the lower extremity joints including the ankle angle, in which the movement of the foot - as one segment - in relation to the tibia was presented. Transversal movement of the foot was presented by the shank-based foot rotation, which we defined as the internal/external rotation of the foot segment in relation to the tibia, and by the foot progression angle which is the angle between the foot segment and the walking direction. In addition, the OFM was used to analyze the movement between segments within the foot, regarding the full complexity of the multi-planar and multi-joint deformity of clubfoot. Data was processed and analyzed in MATLAB R2019b (The MathWorks Inc). The data was interpolated with a 3rd order polynomial and filtered using a Butterworth filter (cut-off frequency: 6 Hz).

2.4. Statistical analysis

Statistical parametric mapping (SPM) was performed to analyze movement patterns (sagittal, frontal and transversal plane) of the foot segments: hindfoot in relation to the tibia, forefoot in relation to the hindfoot and forefoot in relation to the tibia, as well as, foot progression (angle between the foot segment and walking direction), ankle, knee, hip and pelvis angles. For the ankle angle, plantar/dorsiflexion and shank-based foot rotation were analyzed open-source SPM1d code (vM.01.0003; www.spm1D.org). For every subject, trials were normalized over time to represent a gait cycle from 0 to 100 %. Three to five trials were used to calculate mean kinematic patterns per subject. Comparison between study groups was done by SPM ANOVA over normalized time series. P-values of <0.05 were considered significant. In case of statistical significance, a post hoc t-tests (SPM{t}) was performed. Bonferroni correction was applied to adjust alpha (<0.017) for multiple comparisons.

As SPM analyses focus on kinematic pattern in gait cycle timelines but not on peak and mean values specifically, also discrete statistical analyses of standard kinematic gait parameters was performed. To compare the distribution of demographic data and standard kinematic gait parameters between groups, discrete statistical analysis (IBM SPSS Statistics 23) was used. For each subject kinematic parameters were determined based on individual gait cycles. Subject means were used in statistical analysis comparing three groups. A Shapiro-Wilk test was used to check normality of continuous parameters. An one-way ANOVA, including Bonferroni post hoc, was done for normally distributed data. Otherwise, non-parametric tests were used. To determine the strength of the results and aid interpretation, effect sizes according the partial eta squared (η_p^2) were calculated. Effect sizes of 0.01−0.05, 0.06−0.13 and higher than 0.14 were interpreted as small, medium and large respectively [24].

3. Results

3.1. Demographic characteristics

In total 37 children (11 relapse, 11 clubfeet and 15 healthy age-matched controls) were included for analysis. All patients underwent an Achilles tendon tenotomy as part of their initial Ponseti treatment. An overall significant difference in stride length was found, however Bonferroni post hoc analysis showed no differences between groups. No other significant differences in demographic characteristics were seen between the relapse group, corrected group and control group (Table 1).

3.2. Gait analysis

The relapse group showed significant deviations in foot and ankle kinematics in the sagittal, frontal and transversal plane (Fig. 1, Fig. 2, Tables 2, Table 3). Furthermore, the relapse group showed deviated kinematic pattern in knee flexion and hip abduction. No other significant differences in pelvis, hip and knee kinematics were found between groups (Fig. 1, Table 3). Large effect sizes were found for forefoot/hindfoot mean adduction during swing, forefoot/tibia adduction at toe off, ankle and forefoot/tibia peak plantar flexion and all gait parameters that showed significant differences between groups (Supplementary data).

Table 1
Demographic characteristics of the relapse, corrected and control group (mean ± standard deviation, and count).

	Relapse (n = 11)	Corrected (n = 11)	Control (n = 15)
Male/Female	8/3	9/2	8/7
Age (yrs)	5.7 ± 1.5	5.6 ± 1.6	5.7 ± 1.4
Height (m)	1.12 ± 0.10	1.17 ± 0.09	1.19 ± 0.11
Weight (kg)	22.0 ± 4.2	22.1 ± 3.9	22.8 ± 5.2
Walking velocity (/s)[§]	1.65 ± 0.24	1.66 ± 0.20	1.75 ± 0.23
Stride length[§]	1.41 ± 0.12**	1.42 ± 0.13**	1.55 ± 0.12**
Affected side (uni/bi)	4/7	7/4	–
Initial treatment (n feet)			
Achilles tendon tenotomy	11	11	
renewed Achilles tendon tenotomy*	3	3	
Relapse characteristics°			
Equinus / Limited dorsiflexion	6		
Hindfoot varus	8		
Cavus	1		
Forefoot adduction	7		
Active supination	7		
Planned relapse treatment (n feet)			
Tibialis anterior tendon transfer	5	–	–
Anterior distal tibial epiphysiodesis	3	–	–
Additional bracing	3	–	–

N feet = number of feet. For each participant only one foot was measured *In all patients (feet) renewed Achilles tendon tenotomy was performed before the age of three. [§]Normalized for leg length. **$p < 0.05$ for general ANOVA, no significant difference between groups in Bonferroni post hoc analysis. °In 82 % of the cases a combination of relapse characteristics was present. An overview of relapse characteristics for each patient in the relapse group is presented in the supplementary data (Supplementary data - Table I – characteristics relapse group).

3.2.1. Conventional gait kinematics

SPM showed significant differences in the kinematic pattern of the knee flexion, hip adduction and the shank-based foot rotation angle. In general, the kinematic gait pattern of the knee is similar during the whole gait cycle. However, during the end of terminal swing there is less knee extension present in the relapse group compared to corrected group (Fig. 1.g). In the frontal plane, the relapse group showed a larger hip abduction during terminal stance compared to healthy controls (Fig. 1.e). In the transversal plane, increased internal shank-based foot rotation is seen during almost the entire stance phase and a part of mid-swing in the relapse group compared to healthy controls (Fig. 1.k).

Discrete statistics showed an increased mean shank-based foot rotation during the whole gait cycle (Table 3). Additionally, sagittal plane deviations were observed. The relapse group showed a reduced plantar/dorsiflexion range of motion and decreased plantar flexion at toe off (Table 3).

3.2.2. Hindfoot in relation to tibia

No significant differences were found in the gait cycle timelines using SPM (Fig. 2a–c). Discrete statistical analyses revealed reduced plantar/dorsiflexion range of motion. (Table 2).

3.2.3. Forefoot in relation to hindfoot

The relapse group showed increased forefoot supination in relation to the hindfoot during terminal swing compared to the corrected group (Fig. 2.e).

Additionally, discrete statistics show an increased mean supination angle during swing in the relapse group compared to corrected group. (Table 2). Furthermore, discrete statistics points out increased forefoot adduction in relation to the hindfoot in the relapse group compared to controls, as observed in increased mean adduction during the stance phase and more adduction at toe off (Table 2).

3.2.4. Forefoot in relation to tibia

In the frontal plane, deviated supination of the forefoot during mid stance was found. Bonferroni post hoc analysis did not show differences between groups (Fig. 2.h). Transversal plane kinematics showed increased forefoot adduction in the relapse group compared to the corrected group during terminal stance and compared healthy controls during almost the entire gait cycle (Fig. 2.i).

Discrete statistics showed increased mean forefoot supination in relation to the tibia during stance in the relapse group compared to the corrected group. Also, increased adduction in relation to the tibia was found during the stance phase and during the swing phase, compared to healthy controls (Table 2). Besides, in the sagittal plane, decreased plantar flexion at toe off was found in the relapse group (forefoot in relation to tibia) (Table 2).

4. Discussion

In this study, the use of a multi-segment foot model exposed kinematic differences within the foot of patients with relapse clubfoot. Gait analysis showed that patients with relapse clubfoot have deviated forefoot kinematics compared to patients with non-relapse clubfoot and healthy controls. Main differences were shown in the transversal and frontal plane, consisting of significantly increased forefoot adduction and forefoot supination in relation the tibia and the hindfoot.

Overall, our study showed that patients with relapse clubfoot walked with a higher mean forefoot adduction compared to patients with non-relapse clubfoot. Especially during the terminal stance, relapse

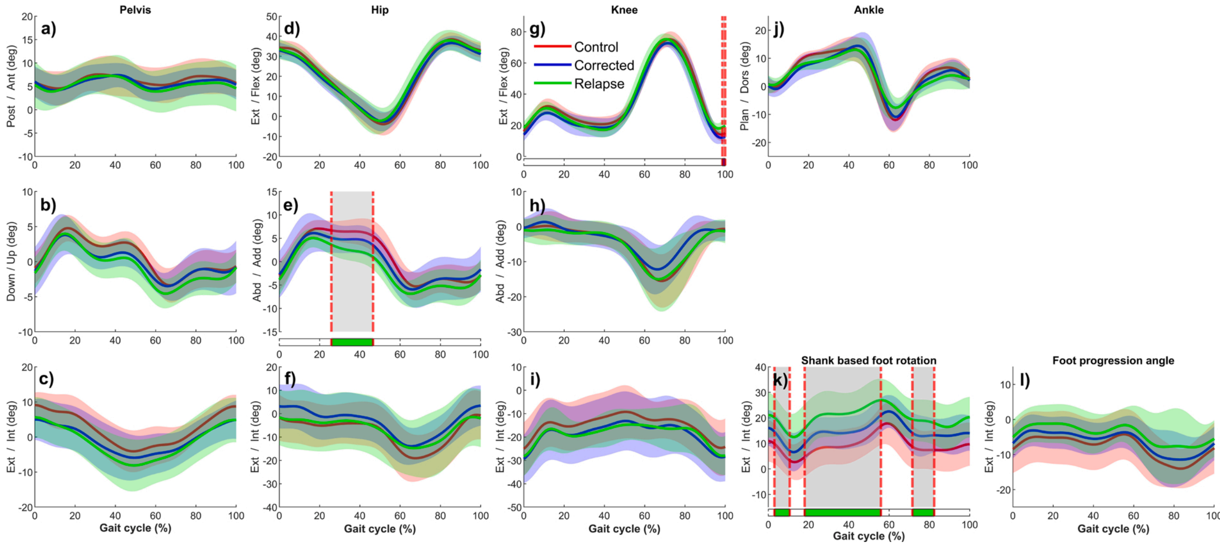

Fig. 1. Statistical parametric mapping - conventional gait kinematics. All angles are presented in degrees (deg). Solid lines indicate group means and the colored bands indicate the region of one standard deviation. Grey areas indicate significant differences between groups. Post-hoc results are shown in bars which indicate statistically significant results at the color-marked times of the gait cycle: relapse versus corrected (blue bar, knee flexion $p < 0.05$), relapse versus controls (green bars, hip ab/adduction $p < 0.001$, shank based foot rotation $p < 0.05$) (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.).

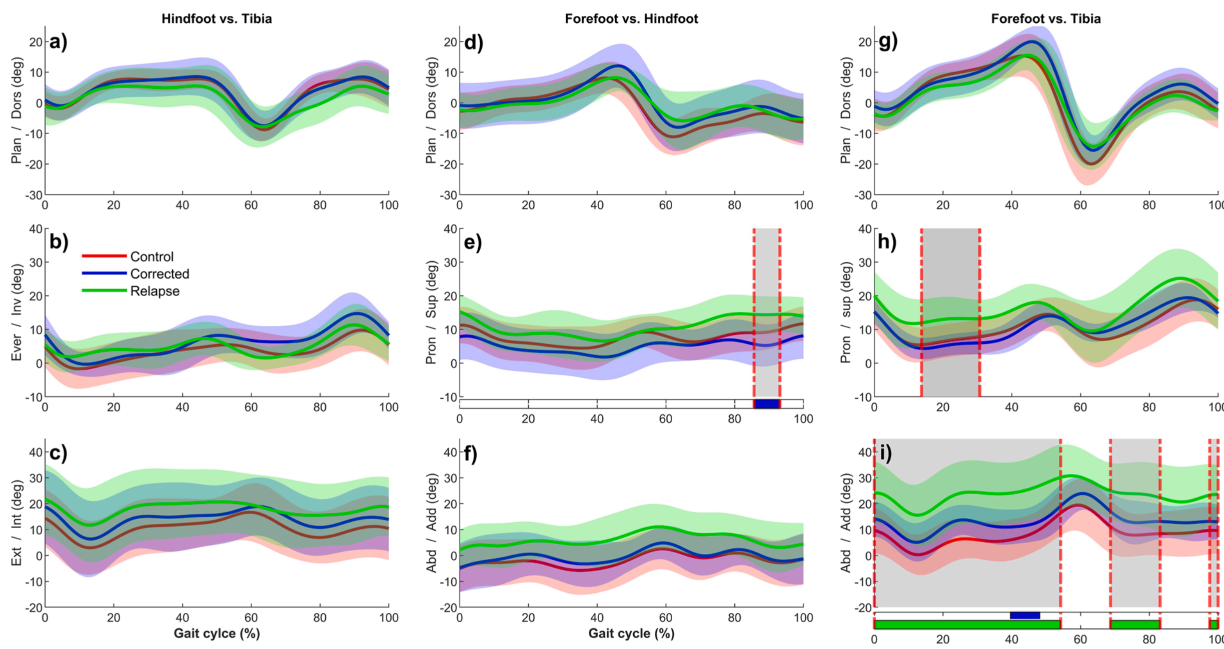

Fig. 2. Statistical parametric mapping – Oxford Foot Model foot kinematics. All angles are presented in degrees (deg). Solid lines indicate group means and the colored bands indicate the region of one standard deviation. Grey areas indicate significant differences between groups. Post-hoc results are shown in bars which indicate statistically significant results at the color-marked times in the gait cycle: relapse versus corrected (blue bar, forefoot vs. hindfoot frontal plane $p < 0.05$, forefoot vs. tibia transversal plane $p < 0.05$), relapse versus controls (green bars, forefoot vs. tibia transversal plane $p < 0.05$). Frontal plane forefoot versus tibia only an overall significance was found, so no post hoc results are presented (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.).

clubfoot had a higher forefoot adduction relative to the tibia. Besides forefoot adduction, the relapse clubfoot showed increased supination of the forefoot in relation to the tibia during stance. During swing, increased supination of the forefoot in relation to the hindfoot was found. Kinematic differences between relapse clubfeet and non-relapse clubfeet are, from a clinical point of view, highly interesting as they might serve as an objective indicator of relapse. As far as we are aware, this is the first study that compared quantitative gait parameters between a pre-treatment relapse group and a corrected group. There are, however, studies in which a descriptive clinical comparison is made between these two groups. In line with our results, these studies indicate that adduction and supination are typical characteristics for the relapse clubfoot [10,17,25]. Although a wide variety of involved components might be present in a relapse, often limited dorsiflexion is also linked to pre-treatment relapse clubfoot in a clinical setting [10]. However, this was not found in our study, possibly due to the variability in clinical

Table 2
Discrete statistics –Oxford Foot Model kinematics.

	Hindfoot vs. Tibia			Forefoot vs. Hindfoot			Forefoot vs. Tibia		
	Relapse	Corrected	Control	Relapse	Corrected	Control	Relapse	Corrected	Control
Sagittal plane									
Range of motion (deg)	**19.2 ± 3.3†**	20.2 ± 3.9	**22.9 ± 2.6**	20.6 ± 3.8	24.2 ± 3.1	23.4 ± 5.2	35.6 ± 5.5	39.1 ± 4.4	40.6 ± 6.3
Dorsiflexion at initial contact (deg)	−1.1 ± 6.5	0.9 ± 5.5	−0.4 ± 4.4	−2.3 ± 5.9	−0.9 ± 7.8	−2.7 ± 6.4	−3.6 ± 4.7	−1.1 ± 5.5	−3.9 ± 5.5
Peak dorsiflexion during stance (deg)	8.1 ± 6.0	10.2 ± 5.9	10.2 ± 2.8	10.5 ± 5.5	13.4 ± 8.2	9.9 ± 4.5	18.5 ± 5.8	21.5 ± 5.3	17.6 ± 5.5
Mean angle during stance (deg)	2.8 ± 5.9	4.3 ± 5.4	4.1 ± 2.8	1.7 ± 5.4	2.8 ± 7.6	1.3 ± 4.7	5.1 ± 4.8	7.0 ± 5.2	5.1 ± 4.7
Plantar flexion at toe off (deg)	−7.4 ± 7.1	−7.8 ± 6.1	−9.0 ± 3.6	−5.3 ± 8.4	−8.3 ± 7.9	−12.2 ± 6.7	**−13.6 ± 7.9†**	−16.1 ± 5.0	**−21.4 ± 8.3**
Peak plantar flexion (deg)	−9.7 ± 7.2	−8.7 ± 5.8	−10.8 ± 3.7	−10.0 ± 7.2	−10.9 ± 7.4	−13.5 ± 6.8	−17.1 ± 7.1	−17.5 ± 5.1	−23.0 ± 8.0
Mean angle during swing (deg)	−0.0 ± 6.5	2.5 ± 5.3	2.2 ± 3.8	−3.7 ± 7.4	−4.1 ± 8.1	−6.6 ± 5.9	−4.4 ± 4.8	−1.6 ± 4.9	−4.6 ± 5.6
Peak dorsiflexion during swing (deg)	7.1 ± 6.3	9.4 ± 5.5	10.1 ± 4.3	1.0 ± 7.8	0.8 ± 8.4	−1.1 ± 6.6	4.1 ± 5.0	7.0 ± 5.7	5.0 ± 5.7
Frontal plane									
Range of motion (deg)	15.8 ± 3.6	18.0 ± 3.2	15.5 ± 3.5	15.2 ± 3.3	12.3 ± 2.8	13.8 ± 3.0	21.0 ± 7.0	18.1 ± 4.4	19.3 ± 3.4
Mean angle during stance (deg)	4.3 ± 4.7	4.1 ± 5.2	2.4 ± 5.8	9.1 ± 5.7	4.4 ± 6.8	7.0 ± 5.3	**14.3 ± 6.5***	**8.7 ± 2.7***	9.5 ± 5.0
Mean angle during swing (deg)	6.0 ± 4.3	9.5 ± 5.2	5.4 ± 5.3	**13.2 ± 5.6***	**6.1 ± 5.8***	8.5 ± 5.1	18.1 ± 8.6	14.4 ± 2.9	12.7 ± 6.4
Transversal plane									
Range of motion (deg)	18.6 ± 3.5	19.1 ± 5.3	20.2 ± 4.9	17.7 ± 5.5	16.5 ± 3.5	16.1 ± 4.8	20.2 ± 6.7	21.5 ± 4.7	23.4 ± 3.4
Mean angle during stance (deg)	18.2 ± 13.8	14.0 ± 13.3	10.6 ± 10.4	**5.8 ± 8.4†**	−0.8 ± 8.3	**−2.7 ± 8.7**	**23.7 ± 12.2†**	12.7 ± 6.9	**7.7 ± 8.1**
Mean angle during swing (deg)	17.0 ± 12.3	14.0 ± 11.8	10.4 ± 11.0	6.6 ± 8.2	0.5 ± 7.2	−0.6 ± 8.1	**23.8 ± 12.2†**	14.7 ± 7.3	**10.2 ± 8.9**
Adduction at toe off (deg)	20.0 ± 12.8	19.3 ± 11.5	17.0 ± 12.2	**11.9 ± 8.6†**	5.0 ± 7.0	**3.1 ± 8.6**	31.8 ± 11.3	24.9 ± 7.0	20.2 ± 11.1

Kinematic data is presented in degrees (deg). Sagittal plane: positive values mean dorsiflexion, negative values mean plantar flexion. Frontal plane: for the hindfoot versus tibia positive values mean inversion, negative values mean eversion. Looking at the forefoot versus the hindfoot or tibia, positive values mean supination, negative values mean pronation. Transverse plane: for the hindfoot versus tibia positive values mean internal rotation, negative values mean external rotation. Looking at the forefoot versus the hindfoot or tibia, positive values mean adduction, negative values mean abduction. Significant $p < 0.05$: * indicates relapse vs. corrected, †indicates relapse vs. control.

indication of our patient groups (with and without a relapse). In our relapse group, limited dorsiflexion was not always observed, while in some of the patients without a relapse limited passive dorsiflexion was observed during the clinical exam although not to an extent that additional treatment was deemed necessary by the treating orthopedic surgeon. Moreover, limited passive dorsiflexion may not necessarily affect the range of motion during gait as the full range is not used. Thus, adduction and supination seem proven characteristics of a relapse clubfoot, whereas the role of limited dorsiflexion during gait of patients with relapse clubfoot in comparison with patients without relapsed clubfoot requires further investigation.

The current study observed a similar tendency of kinematic deviations in the corrected group compared to healthy controls as in previous studies, however no significance was reached [11–16]. In our study, the clubfoot group consisted of patients who were successfully treated with initial Ponseti method, excluding patients who received additional treatment such as a tibialis anterior tendon transfer (TATT), posterior release (PR) or posteromedial release (PMR). Other studies often did not differentiate between patients solely treated with initial Ponseti or a combination of Ponseti and extra-articular (e.g. TATT) or intra-articular (e.g. PMR) surgical interventions [11–16]. The results of our current study suggest the need to look at the relapse clubfoot separately from the non-relapse clubfoot in pre-treatment gait analysis.

From a clinical point of view, early detection of relapse characteristics, such as forefoot adduction and forefoot supination as shown in our study, could improve treatment and prevent the need for surgery. Compared to healthy controls, internal rotated foot progression angle and decreased dorsiflexion were reported in patients with relapse clubfoot who previously received additional extra- or intra-articular surgical treatment [19]. Furthermore, increased dorsiflexion and decreased plantarflexion was shown in patients with relapse clubfoot who received additional extra- or intra-articular surgical treatment, whereas no significant gait deviations were reported in patients with relapse clubfoot who received repeated Ponseti treatment compared to patients without relapsed clubfoot who only received the conservative Ponseti treatment [18,26]. Repeated Ponseti treatment is recommended for a relapse clubfoot which are recognized in the early stage [9,18]. Gait analysis is a widely accepted objective tool to quantify gait deviations, which also has proven its role in clinical decision-making [17]. However, due to its complexity this is in general not available in clinical setting. This underlines the need for easy quantitative measurement in a clinical setting to enable early identification of relapse clubfoot or relapse characteristics which indicate the need for three-dimensional gait analysis.

The usage of a multi-segment foot model in the current study revealed additional deviations that are in line with the multi-planar and multi-joint characteristics of a relapse deformity [10]. As far as we are aware, only two other studies previously investigated foot kinematics in pre-treatment relapse clubfoot using a multi-segment foot model [20,27]. In line with our results, multiple kinematic deviations at hindfoot and forefoot level were exposed in patients with relapse clubfoot compared to healthy controls [20,27]. In our study, we included patients with clubfoot who needed any additional treatment (e.g. bracing, different extra-articular surgical interventions) in addition to the initial Ponseti treatment, while Mindler and colleagues only included patients planned for a specific relapse treatment (TATT). Secondary analysis of the relapse group also shows considerable increased forefoot supination in the TATT group. However, looking at the patients planned for an anterior distal tibial epiphysiodesis or bracing the results show increased forefoot adduction in relation to the hindfoot plus decreased hindfoot dorsiflexion in relation to the tibia during swing and an increased internal rotation of the hindfoot in relation to the tibia, respectively (supplementary data). Although the general differences between successfully treated clubfoot and relapse clubfoot are in line with the initial clinical classification, these results imply that there could be specific kinematic characteristics indicating a different type of relapse clubfoot needing other treatment [9]. This insight might be useful in early identification of a relapse clubfoot and subsequently objective

Table 3
Discrete statistics – kinematics ankle, knee, hip and pelvis based on the extended Helen Hayes model.

	Relapse	Corrected	Control
Ankle			
Range of motion (deg)	**23.8 ± 3.0**†	27.7 ± 3.6	**29.4 ± 4.4**
Dorsiflexion at initial contact (deg)	0.6 ± 3.4	0.2 ± 3.6	0.4 ± 2.7
Peak dorsiflexion during stance (deg)	14.5 ± 4.2	15.3 ± 4.1	15.4 ± 2.7
Mean angle during stance (deg)	6.9 ± 2.9	6.2 ± 3.8	6.9 ± 2.0
Plantar flexion at toe off (deg)	**−7.5 ± 4.1**†	−11.6 ± 5.9	**−13.0 ± 4.4**
Peak plantar flexion (deg)	−9.3 ± 4.1	−12.4 ± 5.8	−14.0 ± 4.3
Mean angle during swing (deg)	−0.6 ± 3.8	0.1 ± 3.8	0.6 ± 3.1
Peak dorsiflexion during swing (deg)	5.7 ± 3.7	6.2 ± 3.8	7.9 ± 3.2
Shank based foot rotation during stance (deg)	**20.7 ± 8.7**†	14.3 ± 6.8	**9.3 ± 7.2**
Shank based foot rotation during swing (deg)	**19.4 ± 8.2**†	14.6 ± 7.4	**9.4 ± 8.3**
Foot progression angle (deg)	−2.3 ± 5.44	−4.4 ± 4.6	−5.7 ± 7.4
Knee			
Sagittal plane range of motion (deg)	63.2 ± 4.1	65.0 ± 3.7	65.6 ± 5.8
Minimum flexion during stance (deg)	16.4 ± 5.0	17.1 ± 6.5	19.4 ± 5.1
Maximum flexion during swing (deg)	76.8 ± 3.9	73.9 ± 2.6	76.6 ± 4.8
Hip			
Sagittal plan range of motion (deg)	42.2 ± 5.8	41.0 ± 6.0	44.1 ± 6.1
Minimum flexion (deg)	−3.3 ± 7.3	−3.2 ± 5.3	−4.6 ± 5.7
Maximum flexion (deg)	38.9 ± 5.1	37.8 ± 3.7	39.5 ± 3.5
Mean rotation during stance (deg)	−4.6 ± 12.9	−1.8 ± 9.7	−5.7 ± 9.4
Maximum external rotation (deg)	−17.1 ± 12.9	−15.0 ± 8.1	−20.7 ± 10.4
Pelvis			
Mean pelvic tilt (deg)	5.4 ± 4.4	5.7 ± 2.9	6.2 ± 3.5
Transversal plane range of motion (deg)	17.0 ± 2.4	14.4 ± 5.0	17.2 ± 6.5
Maximum external rotation (deg)	−7.5 ± 8.4	−6.0 ± 6.7	−5.7 ± 5.6
Maximum internal rotation (deg)	8.8 ± 6.0	7.8 ± 6.1	11.5 ± 3.2

Kinematic data is presented in degrees (deg).Sagittal plane: positive values mean flexion, negative values mean extension. Frontal plane: positive values mean adduction, negative values mean abduction. Transverse plane: positive values mean internal rotation, negative values mean external rotation. Significant $p < 0.05$: †indicates relapse vs. control.

three-dimensional gait analysis could aid to optimize clinical decision-making and planning of additional -if possible conservative- treatment. Based on these objective measures, it could be clinically interesting to define some threshold values to detect relapse and guide treatment based on the mean forefoot supination and adduction reported standard deviations. However, we need to take into account that groups in the current study were relatively small and showed large variability. Further research is necessary to determine thresholds that can be used in a clinical setting.

Previous studies showed several compensation mechanisms in children with Ponseti treated clubfoot [11–16,19]. External hip rotation is commonly seen to compensate for internal foot rotation [11,12,14–16]. However, we found increased and faster hip abduction during terminal stance instead, which could be a compensation for the increased internal shank based foot rotation. When taking a closer look at the compensation in the hip, it is clear that hip rotation and hip abduction are part of a circumduction movement that can be associated with the increased shank based foot rotation. As our results did not show a reduction in dorsiflexion, we also did not find the previously reported increased knee extension as a compensation for reduced dorsiflexion during stance [13,14]. Walking velocity could also play a role. Although walking velocity was not found significantly different between groups, there was a trend that patients chose a slightly lower walking velocity possibly avoiding kinematic compensation at knee, hip or pelvic level.

Even though walking provides important information as it is the most used movement in daily life, more challenging tasks also play a role in children's development. Since gross motor skills during these tasks are only poorly related to gait [28], future research should also focus on more challenging tasks, such as hopping and running. Previous studies reported increased motor impairment in children with clubfoot based on the Movement Assessment Battery for Children and the Clubfoot Assessment Protocol including more challenging tasks besides walking [28,29]. Patients with clubfoot may show more compensations during these challenging tasks, which are not exposed in our conventional gait analysis. Although our study provided detailed information about possible early identifiers in foot kinematics in patients with relapse clubfoot compared to patients with successfully treated clubfoot, further investigation of more challenging tasks could aid early identification of a relapse.

A limitation of this study that should be kept in mind is that, despite the fact that the OFM has a high repeatability in patients with clubfoot, hindfoot rotation is sensitive for variable heel marker placement [23]. This might affect the repeatability of the hindfoot kinematics. Marker placement variability was minimized through the use of a standardized marker placement protocol performed consistently by the same two researchers. Another limitation is our sample size which is relatively small. However, achieved effect sizes were large for all gait parameters that showed significant differences between groups.

In conclusion, gait analysis including a multi-segment foot model showed that patients with relapse clubfoot deviate from patients with successfully treated clubfoot in forefoot adduction and forefoot supination. This suggests that these parameters could be kinematic biomarkers for relapse. As previously described in literature deviations at ankle joint level, such as intoeing, may actually exist within the foot [14,30]. The usage of a multi-segment foot model in this study confirmed that multiple segments are involved in these kinematic gait deviations, especially highlighting forefoot adduction and forefoot supination as kinematic indicators of a relapse. Clinical assessment of possible relapse clubfoot should therefore particularly focus on the forefoot.

Author contributions

L. Grin: Investigation, Formal Analysis, Writing – Original Draft. **M. C. van der Steen:** Conceptualization, Supervision, Writing – Reviewing and Editing. **S.D.N. Wijnands:** Investigation, Formal Analysis, Writing – Reviewing and Editing. **L. van Oorschot:** Investigation, Writing – Reviewing and Editing. **A.T. Besselaar:** Conceptualization, Resources, Writing – Reviewing and Editing. **B. Vanwanseele:** Conceptualization, Resources, Supervision, Writing – Reviewing and Editing.

Declaration of Competing Interest

The authors report no declarations of interest.

Acknowledgement

This work was supported by SIA RAAK [Raak Publiek 2016; RAAK. PUB03.014] & Catharina Onderzoeksfonds 2017-4.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.gaitpost.2021.09.185.

References

[1] A.T. Besselaar, R.J.B. Sakkers, H.A. Schuppers, M.M.E.H. Witbreuk, E.V.C. M. Zeegers, J.D. Visser, R.A. Boekestijn, S.D. Margés, M.C.Marieke Van der Steen, K.N.J. Burger, Guideline on the diagnosis and treatment of primary idiopathic

clubfoot, Acta Orthop. 88 (2017) 305–309, https://doi.org/10.1080/17453674.2017.1294416.
[2] F. Bergerault, J. Fournier, C. Bonnard, Idiopathic congenital clubfoot: initial treatment, Orthop. Traumatol. Surg. Res. 99 (2013) S150–S159, https://doi.org/10.1016/j.otsr.2012.11.001.
[3] A.T. Besselaar, M.C. Kamp, M. Reijman, M. Van Der Steen, Incidence of congenital idiopathic clubfoot in the Netherlands, J. Pediatr. Orthop. Part B 27 (2018) 563–567, https://doi.org/10.1097/BPB.0000000000000525.
[4] H. Wallander, L. Hovelius, K. Michaelsson, Incidence of congenital clubfoot in Sweden, Acta Orthop. 77 (2006) 847–852, https://doi.org/10.1080/17453670610013123.
[5] L. Shabtai, S.C. Specht, J.E. Herzenberg, Worldwide spread of the Ponseti method for clubfoot, World J. Orthop. 5 (2014) 585–590, https://doi.org/10.5312/wjo.v5.i5.585.
[6] S.J. Laaveg, I.V. Ponseti, Long-term results of treatment of congenital club foot, J. Bone Jt. Surg. - Ser. A 62 (1980) 23–31, https://doi.org/10.2106/00004623-198062010-00004.
[7] M.B. Dobbs, R. Nunley, P.L. Schoenecker, Long-term follow-up of patients with clubfeet treated with extensive soft-tissue release, J. Bone Jt. Surg. - Ser. A 88 (2006) 986–996, https://doi.org/10.2106/JBJS.E.00114.
[8] J.A. Morcuende, L.A. Dolan, F.R. Dietz, I.V. Ponseti, Radical reduction in the rate of extensive corrective surgery for clubfoot using the ponseti method, Pediatrics 113 (2004) 376–380, https://doi.org/10.1542/peds.113.2.376.
[9] C. Radler, G.T. Mindler, Treatment of severe recurrent clubfoot, Foot Ankle Clin. 20 (2015) 563–586, https://doi.org/10.1016/j.fcl.2015.07.002.
[10] J.H. Stouten, A.T. Besselaar, M.C. Marieke Van Der Steen, Identification and treatment of residual and relapsed idiopathic clubfoot in 88 children, Acta Orthop. 89 (2018) 448–453, https://doi.org/10.1080/17453674.2018.1478570.
[11] L.A. Karol, K. Jeans, R. Elhawary, Gait analysis after initial nonoperative treatment for clubfeet: intermediate term followup at age 5, Clin. Orthop. Relat. Res. 467 (2009) 1206–1213, https://doi.org/10.1007/s11999-008-0702-9.
[12] C. Church, J.A. Coplan, D. Poljak, A.M. Thabet, D. Kowtharapu, N. Lennon, S. Marchesi, J. Henley, R. Starr, D. Mason, M.V. Belthur, J.E. Herzenberg, F. Miller, A comprehensive outcome comparison of surgical and Ponseti clubfoot treatments with reference to pediatric norms, J. Child. Orthop. 6 (2012) 51–59, https://doi.org/10.1007/s11832-012-0387-1.
[13] C.M. Duffy, J.J. Salazar, L. Humphreys, B.C. McDowell, Surgical versus ponseti approach for the management of CTEV: a comparative study, J. Pediatr. Orthop. 33 (2013) 326–332, https://doi.org/10.1097/BPO.0b013e31827d0b2c.
[14] G.T. Mindler, A. Kranzl, C.A.M. Lipkowski, R. Ganger, C. Radler, Results of gait analysis including the oxford foot model in children with clubfoot treated with the ponseti method, J. Bone Jt. Surg. - Am. Vol. 96 (2014) 1593–1599, https://doi.org/10.2106/JBJS.M.01603.
[15] E. Lööf, H. Andriesse, M. André, S. Böhm, E.W. Broström, Gait in 5-year-old children with idiopathic clubfoot: A cohort study of 59 children, focusing on foot involvement and the contralateral foot, Acta Orthop. 87 (2016) 522–528, https://doi.org/10.1080/17453674.2016.1202013.
[16] E. Manousaki, T. Czuba, G. Hägglund, L. Mattsson, H. Andriesse, Evaluation of gait, relapse and compliance in clubfoot treatment with custom-made orthoses, Gait Posture 50 (2016) 8–13, https://doi.org/10.1016/j.gaitpost.2016.08.005.
[17] W.N. Sankar, S.A. Rethlefsen, J. Weiss, R.M. Kay, The recurrent clubfoot: Can gait analysis help us make better preoperative decisions? Clin. Orthop. Relat. Res. 467 (2009) 1214–1222, https://doi.org/10.1007/s11999-008-0665-x.
[18] Y. Bin Liu, S.Y. Jiang, L. Zhao, Y. Yu, D.H. Zhao, Can repeated ponseti management for relapsed clubfeet produce the outcome comparable with the case without relapse? A clinical study in term of gait analysis, J. Pediatr. Orthop. 40 (2020) 29–35, https://doi.org/10.1097/BPO.0000000000001071.
[19] K.A. Pierz, J.R. Lloyd, M.J. Solomito, P. Mack, S. Õunpuu, Lower extremity characteristics in recurrent clubfoot: clinical and gait analysis findings that may influence decisions for additional surgery, Gait Posture 75 (2020) 85–92, https://doi.org/10.1016/j.gaitpost.2019.10.002.
[20] G.T. Mindler, A. Kranzl, C. Radler, Normalization of forefoot supination after tibialis anterior tendon transfer for dynamic clubfoot recurrence, J. Pediatr. Orthop. 40 (2020) 418–424, https://doi.org/10.1097/BPO.0000000000001542.
[21] M.S. Asperheim, C. Moore, N.C. Carroll, L. Dias, Evaluation of residual clubfoot deformities using gait analysis, J. Pediatr. Orthop. Part B 4 (1995) 49–54, https://doi.org/10.1097/01202412-199504010-00007.
[22] J. Stebbins, M. Harrington, N. Thompson, A. Zavatsky, T. Theologis, Repeatability of a model for measuring multi-segment foot kinematics in children, Gait Posture 23 (2006) 401–410, https://doi.org/10.1016/j.gaitpost.2005.03.002.
[23] J. McCahill, J. Stebbins, B. Koning, J. Harlaar, T. Theologis, Repeatability of the Oxford Foot Model in children with foot deformity, Gait Posture 61 (2018) 86–89, https://doi.org/10.1016/j.gaitpost.2017.12.023.
[24] J. Cohen, Statistical Power Analysis for the Behavioral Sciences, 2nd edition, Routledge, 2013, https://doi.org/10.4324/9780203771587.
[25] K. Gray, J. Burns, D. Little, M. Bellemore, P. Gibbons, Is tibialis anterior tendon transfer effective for recurrent clubfoot? Clin. Orthop. Relat. Res. 472 (2014) 750–758, https://doi.org/10.1007/s11999-013-3287-x.
[26] K.A. Jeans, L.A. Karol, A.L. Erdman, W.R. Stevens, Functional outcomes following treatment for clubfoot ten-year follow-up, J. Bone Jt. Surg. - Am. Vol. 100 (2018) 2015–2023, https://doi.org/10.2106/JBJS.18.00317.
[27] J.L. McCahill, J. Stebbins, J. Harlaar, R. Prescott, T. Theologis, C. Lavy, Foot function during gait and parental perceived outcome in older children with symptomatic club foot deformity, Bone Jt. Open 1 (2020) 384–391, https://doi.org/10.1302/2633-1462.17.bjo-2020-0046.r1.
[28] E. Lööf, H. Andriesse, M. André, S. Böhm, M.D. Iversen, E.W. Broström, Gross motor skills in children with idiopathic clubfoot and the association between gross motor skills, foot involvement, gait, and foot motion, J. Pediatr. Orthop. 39 (2019) 359–365, https://doi.org/10.1097/BPO.0000000000000964.
[29] H. Andriesse, L. Westbom, G. Hägglund, Motor ability in children treated for idiopathic clubfoot. A controlled pilot study, BMC Pediatr. 9 (2009) 78, https://doi.org/10.1186/1471-2431-9-78.
[30] T.N. Theologis, M.E. Harrington, N. Thompson, M.K.D. Benson, Dynamic foot movement in children treated for congenital talipes equinovarus, J. Bone Jt. Surg. - Ser. B 85 (2003) 572–577, https://doi.org/10.1302/0301-620X.85B4.13696.