Gait in 5-year-old children with idiopathic clubfoot

A cohort study of 59 children, focusing on foot involvement and the contralateral foot

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Background and purpose — Idiopathic clubfoot can be bilateral or unilateral; however, most studies of gait have assessed clubfoot cases as one uniform group. The contralateral foot in children with unilateral clubfoot has shown deviations in pedobarographic measurements, but it is seldom included in studies of gait. We evaluated gait in children with idiopathic clubfoot, concentrating on foot involvement.

Patients and methods — Three-dimensional gait analyses of 59 children, mean age 5.4 years, with bilateral (n = 30) or unilateral (n = 29) idiopathic clubfoot were stratified into groups of bilateral, unilateral, or contralateral feet. Age-matched controls (n = 28) were evaluated for comparison. Gait assessment included: (1) discrete kinematic and kinetic parameters, and (2) gait deviation index for kinematics (GDI) and kinetics (GDI-k).

Results — No differences in gait were found between bilateral and unilateral idiopathic clubfoot, but both groups deviated when compared to controls. Compared to control feet, contralateral feet showed no deviations in discrete gait parameters, but discrepancies were evident in relation to unilateral clubfoot, causing gait asymmetries in children with unilateral involvement. However, all groups deviated significantly from control feet according to GDI and GDI-k.

Interpretation — Bilateral and unilateral idiopathic clubfoot cases show the same persistent deviations in gait, mainly regarding reduced plantarflexion. Nevertheless, knowledge of foot involvement is important as children with unilateral clubfoot show gait asymmetries, which might give an impression of poorer deviations. The results of GDI/GDI-k indicate global gait adaptations of the contralateral foot, so the foot should preferably not be used as a reference for gait.

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Half of all children with idiopathic clubfoot have bilateral involvement (Wallander et al. 2006). The etiology of clubfoot is believed to be multifactorial (Dobbs and Gurnett 2009). However, no theories have been put forward to explain the possible differences between bilateral and unilateral idiopathic clubfoot. Regardless of foot involvement, clubfoot is treated the same way—most often with serial casting, Achilles tendon lengthening, and bracing according to the Ponseti method (Jowett et al. 2011, Ponseti 1992). The goal of treatment is to obtain a well-functioning foot with good mobility (Ponseti 1992).

Gait analysis is an objective tool for evaluating treatment outcomes, and it has been considered useful in patients with clubfoot (Graf et al. 2012). Previous studies of gait in children with idiopathic clubfoot have found persistent gait deviations (Church et al. 2012, Duffy et al. 2013, Mindler et al. 2014, Jeans et al. 2015). To date, bilateral and unilateral clubfoot have been evaluated as one homogeneous group that usually includes a mixture of unilateral and bilateral cases. This can be problematic, since Gray et al. (2014a) found that bilateral clubfoot tends to be more severe at birth than unilateral involvement alone. Moreover, in the absence of using appropriate statistical methods, both feet of a child should not be regarded as independent observations (Gray et al. 2014b).

Studies of foot pressures and forces of the contralateral foot in children with unilateral idiopathic clubfoot have proposed that the contralateral side should not be referred to as normal or used as a control (Favre et al. 2007, Cooper et al. 2014). To our knowledge, there have been no gait analysis studies of the contralateral foot or of potential differences between the feet of children with unilateral idiopathic clubfoot that has been treated nonoperatively. We therefore evaluated gait in children with idiopathic clubfoot, focusing on foot involvement. Specific research questions were as follows. Do bilateral idio-

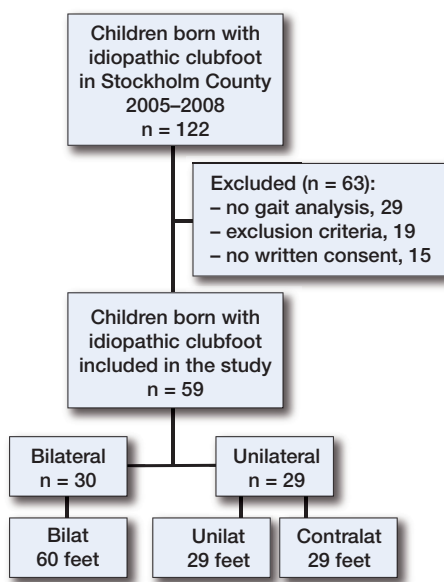Figure 1. Flowchart of children with idiopathic clubfoot who were included and group division within the study.

Table 1. Demographic characteristics [a]

| | Clubfoot population | | |
	Bilateral n = 30	Unilateral n = 29	Control n = 28
Age in years, mean (SD)	5.3 (0.3)	5.5 (0.6)	5.5 (0.6)
Boys, n (%)	20 (67)	21 (72)	18 (64)
Body length in cm, mean (SD)	112 (6)	115 (6)	115 (6)
Feet with clubfoot, n	60	29	na
Treatment			
Number of casts, mean (SD)	11.0 (3.4)	10.6 (2.6)	na
Weeks in cast, mean (SD)	10.6 (2.5)	10.2 (1.7)	na
Achilles lengthening, n	58	29	na
Ongoing bracing, n	28	12	na
Dimeglio classification (88 feet)			
Moderate (5–10 p), n	12	4	na
Severe (11–15 p), n	31	17	na
Very severe (16–20 p), n	17	7	na
Secondary treatment			
Tibialis anterior transfer, n	2	1	na
Casting only, n	0	2	na

na: not applicable.
[a] No significant differences were found between the groups.

pathic clubfoot and unilateral idiopathic clubfoot differ from each other regarding gait characteristics; and if so, do they differ similarly in relations to typically developed feet? Do contralateral feet in children with unilateral idiopathic clubfoot differ in gait from unilateral clubfeet and typically developed feet?

Material and methods

Subjects and design

We performed a retrospective cross-sectional study of children with idiopathic clubfoot in Stockholm County, who were born in the period 2005–2008. As part of the standard treatment program in Stockholm, children with idiopathic clubfoot are routinely referred for three-dimensional (3D) gait analysis around the age of 5. In order to include children at the same stage of gait development, the inclusion criterion for age was set to 4.5–6.5 years in this study. Children initially treated by professions other than orthopedic surgeons or those with additional neurological or orthopedic conditions were excluded. 59 children with idiopathic clubfoot (30 bilateral and 29 unilateral) were included (Figure 1).

All clubfeet were initially treated with above-the-knee casting, followed by a percutaneous Achilles tenotomy in 87 out of the 89 cases. Thereafter, treatment continued with a custom-made knee-ankle-foot orthosis (KAFO) with an open knee joint. The KAFO was manufactured to hold the foot in a maximally corrected position. Caregivers were instructed that the child should wear the KAFO for 23 hours a day in the first months after serial casting. The usage time was gradually reduced to only night and nap-times from the age of 1

year. The Dimeglio classification score was used to evaluate the severity of clubfoot at birth (Dimeglio et al. 1995). Severity and treatment information were taken from the patient medical record. An age-matched control group of 28 typically developed children was included for comparison (Table 1).

Three-dimensional (3D) gait analysis

The body motions and forces during gait were captured with a 3D 8-camera system (Vicon, Oxford Metrics, Oxford, UK) and 2 force plates (Kistler, Winterthur, Switzerland), in order to analyze joint and segment angles (kinematics), joint moments, and power (kinetics) and speed. The Vicon Plug-in-Gait lower body marker placement was used (Davis et al. 1991). To assess gait, children were asked to walk barefoot at a self-selected speed. 3 representative trials were chosen from observing the videos and gait curves in order to verify representative trials. A mean for every foot was calculated from the selected trials. Due to difficulties of clean force plate strikes, only 2 trials containing kinetic data were collected. Despite this, not all children had sufficient kinetic data. Prior to gait analysis, an experienced physiotherapist from the gait laboratory assessed the passive range of motion according to a standardized procedure with a goniometer. Dorsiflexion (with extended and flexed knee), plantarflexion, and knee extension were assessed in supine position. Tibial torsion (thigh-foot angle) and hip rotations were assessed in prone position with flexed knee.

The following discrete gait parameters were chosen, with consideration of clinical significance. At initial contact: foot flexion and hip rotation. In stance: maximal dorsiflexion, foot progression (foot position in relation to gait direction), maximal knee extension, and mean hip rotation. At terminal stance:

maximal plantarflexion. At terminal swing: mean flexion of the foot. Internal moments were chosen accordingly: maximal dorsiflexion and plantarflexion moments, maximal knee flexion moment, and also maximal hip abduction moment in early stance. Power parameters were chosen to reflect the ability to generate propulsive concentric actions of involved muscles: maximal ankle power generation and maximal ankle to hip power generation ratio. Furthermore, walking and nondimensional speed was added to evaluate potential speed differences between the groups, since diverse speed might affect both kinematics and kinetics (Hof 1996, Schwartz et al. 2008).

The gait deviation index (GDI) and gait deviation index-kinetic (GDI-k), 2 multivariate measures of overall gait pathology (Schwartz and Rozumalski 2008, Rozumalski and Schwartz 2011), were calculated to represent quantitative values of gait kinematics and kinetics. The GDI includes 9 kinematic gait curves from the pelvis and hip in all 3 planes, the knee and ankle in the sagittal plane, and finally the foot progression angle in the transversal plane. Correspondingly, the GDI-k includes 9 kinetic gait curves: the internal moments from the hip, knee, and ankle in the sagittal and frontal plane and the total joint power from the hip, knee, and ankle. Both indices are developed to quantify how far the curves are from the control mean and provide unique values for each individual leg, in which 100 points or more indicate gait without pathology.

Statistics

Children with idiopathic clubfoot were stratified into units of Bilateral or Unilateral according to foot involvement. From the 2 units, 3 groups were made: Bilateral clubfoot (Bilat), Unilateral clubfoot (Unilat), or Contralateral foot (Contralat) (Figure 1). Chi-square test, Mann-Whitney U-test, and 1-way ANOVA were used to compare the distribution of demographic data in the clubfoot units and the control group, which consisted of typically developed children (Table 1).

In order to be able to include all feet, a linear mixed-model was used for statistical comparison between the groups. The within-subject factor was side, that is: left and right foot for Bilateral and controls; or Unilat and Contralat for Unilateral, with covariance structure (compound symmetry). The between subject factor represented unit or group (i.e. Bilateral, Unilateral, or control). The linear mixed-model did not show any statistically significant differences between left and right foot in Bilat and control. However, a few parameters were not normally distributed. The Kruskal-Wallis test was therefore used for those parameters. To ensure statistical independence in the Kruskal-Wallis test, the analysis was performed between either Unilat or Contralat and a mean of left and right foot in the Bilat and control groups. The 95% confidence intervals (CIs) of the mean differences in parameters deviating from the control group were calculated in order to give indications of relationships and differences between the groups.

A power analysis of pilot data showed that a sample size of at least 25 children in each group would be required for a power of 0.8 and an alpha error of 0.05 with regard to GDI. The scores of GDI and GDI-k were calculated in MATLAB (MathWorks, Natick, MA). SPSS version 21 was used for all statistical calculations.

Ethics

This study was approved by the ethical review board in Stockholm (entry no. 2012/659-31/3, June 20, 2012) and all parents/caregivers provided informed consent for the children to participate.

Results

Demographic characteristics and speed

26 of the 59 children were still using KAFO at night when gait was evaluated. 1 child with bilateral clubfoot and 3 children with unilateral clubfoot (5 feet in total) had undergone a second casting period or a tibialis anterior transfer prior to gait evaluation. As illustrated in Tables 1 and 2, missing data for Dimeglio classification and the physical examination, respectively, were limited. No statistically significant differences were found in demographic characteristics between children with bilateral involvement and children with unilateral involvement, or compared to the control group (Table 1). Regarding walking speed, all groups were similar (Table 3).

Bilat vs. Unilat

No statistically significant differences were found in any of the following parameters: physical examination, discrete kinematic and kinetic parameters, and GDI and GDI-k (Tables 2 and 3).

Bilat and Unilat vs. control

Statistically significant deviations were found in several parameters across the clubfoot groups compared with controls (Figure 2). Concerning the physical examination, reduced motion was found for dorsiflexion and plantarflexion of the foot and internal rotation of the hip, while an increase in knee extension was observed in the clubfeet (Table 2).

Both clubfoot groups showed several statistically significant deviations in discrete gait parameters compared to controls, demonstrating reduced dorsiplantar flexion range and plantarflexion, and less internal dorsiflexion and plantarflexion moments. Additionally, clubfeet presented a more pronounced internally-rotated foot progression. In ankle power and ankle/hip power ratio, clubfeet were found to have lower values of approximately one-quarter compared to controls. The Bilat group deviated from the controls in having a reduced internal hip abduction moment. Overall gait pathology was found in both the Bilat and the Unilat groups, with statistically significant deviations in GDI and GDI-k (Table 3).

Table 2. Physical examination. Data are presented as mean and 1 standard deviation in degrees (passive range of motion)

| | | | | | | | | | p-values | | | | |
| | Clubfoot population | | | | | | | | Unilat vs. Bilat | Unilat vs. Contralat | Control vs. Bilat | Control vs. Unilat | Control vs. Contralat |
	Bilat	n	Unilat	n	Contralat	n	Control	n					
Total no. of feet		60		29		29		56					
FOOT Dorsiflexion													
knee extended	11 (6.4)	58	8 (6.4)	29	18 (3.9)	29	20 (3.7)	56		< 0.001	< 0.001	< 0.001	
knee flexed	14 (7.7)	60	11 (7.6)	29	25 (4.8)	29	29 (4.4)	56		< 0.001	< 0.001	< 0.001	0.04
Plantarflexion	44 (12)	51	44 (11)	26	53 (9.0)	26	59 (7.2)	56		< 0.001	< 0.001	< 0.001	
KNEE Extension	11 (6.0)	58	11 (4.5)	29	9 (5.1)	29	7 (2.4)	56		0.01	0.001	0.006	
Tibia torsion	12 (4.8)	56	12 (5.4)	29	13 (3.9)	29	13 (3.4)	56					
HIP Internal rotation	52 (12)	36	51 (9.6)	20	49 (8.7)	20	59 (9.0)	56		0.002	0.04	0.04	0.005
External rotation	43 (10)	36	41 (7.1)	20	42 (5.6)	20	46 (5.1)	56					
LEG Length, cm	56 (3.6)	60	57 (3.5)	29	58 (3.5)	29	56 (3.9)	56		< 0.001			

Only p-values that reached statistical significance (p < 0.05) are presented.
Bilat: bilateral clubfoot; Unilat: unilateral clubfoot; Contralat: contralateral foot; n: number of feet.

Table 3. Gait parameters. All data presented as mean (SD)

| | | | | | p-values | | | | |
| | Clubfoot population | | | | Unilat vs. Bilat | Unilat vs. Contralat | Control vs. Bilat | Control vs. Unilat | Control vs. Contralat |
	Bilat	Unilat	Contralat	Control					
KINEMATICS	n = 60	n = 29	n = 29	n = 56					
GDI	92 (8.0)	88 (11)	91 (7.2)	100 (8.5)			< 0.001	< 0.001	< 0.001
FOOT									
Max. dorsiflexion (DF)	12 (4.0)	10 (5.4)	13 (3.4)	12 (3.1)	< 0.001				
Max. plantarflexion (PF)	11 (6.4)	14 (8.4)	16 (6.6)	18 (4.8)		< 0.001		0.04	
Dorsiplantar flexion range	24 (4.8)	24 (5.2)	29 (5.3)	29 (4.8)	< 0.001	< 0.001	< 0.001		
Flexion at initial contact	0.1 (4.7) DF	2.0 (5.8) PF	0.3 (2.9) DF	0.6 (3.6) PF	0.006				
Mean DF in terminal swing	4.1 (4.5)	1.3 (6.5)	4.1 (2.7)	3.8 (3.1)	0.002				
Mean foot progression in stance	0.8 (6.4) ext	2.6 (9.5) int	2.8 (7.6) ext	4.6 (5.7) ext	< 0.001	0.03	< 0.001		
KNEE									
Max. extension in stance	0.7 (7.4)	2.7 (5.2)	0.1 (4.4)	1.7 (4.7)	0.001				
HIP									
External rotation at initial contact	9.5 (7.9)	8.8 (7.6)	12 (8.2)	11 (5.8)					
Mean external rotation in stance	2.0 (6.0)	0.9 (5.3)	3.3 (6.3)	1.0 (4.9)					
KINETICS	n = 51	n = 24	n = 20	n = 46					
GDI-k	90 (9.8)	94 (7.2)	93 (8.5)	100 (8.6)			< 0.001	0.04	0.03
MOMENTS									
Foot max. DF	0.12 (0.08)	0.10 (0.05)	0.21 (0.08)	0.17 (0.06)	< 0.001	0.002	0.002		
Foot max. PF	0.87 (0.19)	0.90 (0.11)	1.05 (0.16)	1.10 (0.13)	< 0.001	< 0.001	< 0.001		
Knee max. flexion in stance	0.20 (0.14)	0.21 (0.11)	0.19 (0.13)	0.25 (0.10)					
Hip max. abduction in early stance	0.56 (0.14)	0.55 (0.10)	0.63 (0.17)	0.63 (0.14)	0.03	< 0.001			
POWER									
Ankle max. generating (W/kg)	2.8 (0.78)	2.8 (0.72)	3.6 (0.87)	3.6 (0.75)	< 0.001	< 0.001	0.001		
Ratio ankle/hip generating power	1.9 (0.69)	1.9 (0.54)	2.8 (1.10)	2.8 (0.98)	< 0.001	< 0.001	0.003		
SPEED									
Walking speed (m/s)	1.1 (0.17)	1.1 (0.13)	1.1 (0.13)	1.2 (0.17)					
Non-dimensional speed	0.48 (0.07)	0.47 (0.06)	0.48 (0.04)	0.51 (0.07)					

Kinematic data presented in degrees and moments in internal moments (Nm/kg).
Only p-values reaching statistical significance (p < 0.05) are presented.
Bilat, Unilat Contralat, and n, see Table 2. GDI: Gait Deviation Index, GDI-k: Gait Deviation Index Kinetic.

Contralat vs. Unilat

Concerning the physical examination, the greatest discrepancies were found for dorsiflexion and plantarflexion, with less motion in the unilateral clubfoot (Table 2). For discrete gait parameters, unilateral clubfeet had a statistically significant decrease in dorsiflexion and dorsiplantar flexion range compared to the Contralat group. Further asymmetries observed were the finding of a plantarflexed foot at initial contact and
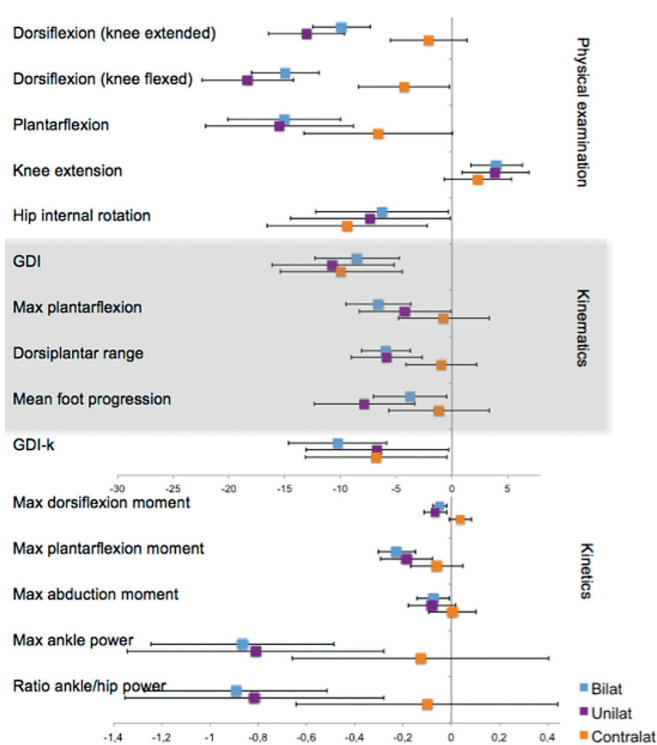
Figure 2. The 95% confidence intervals (CIs) of the mean differences in parameters deviating from the control group in either the Bilat, Unilat, or Contralat group. GDI: gait deviation index; GDI-k: gait deviation index-kinetic. Note that parameters included have different units (degrees, GDI, GDI-k, Nm/kg, and W/kg), so that the 95% CIs and mean differences are not comparable between different parameters, only between groups.

an increase knee extension in stance in Unilat compared to a dorsiflexed foot, and an almost straight knee in the Contralat group. Moreover, discrepancies were found for all discrete kinetic parameters, except for maximal internal knee flexion moment. In contrast, the findings from the GDI and GDI-k indicated similar gait pathology in the two groups, with no between group discrepancies (Table 3).

Contralat vs. control

No statistically significant differences were found in the physical examination, discrete kinematic and kinetic parameters of these groups except in passive dorsiflexion and internal rotation of the hip (Figure 2, Table 2). Nevertheless, the contralateral feet showed overall gait pathology in GDI and GDI-k (Figure 2 and Table 3).

Discussion

Our main findings in this cohort of children with idiopathic clubfoot were corresponding gait characteristics in bilateral and unilateral clubfoot. Likewise, similar gait deviations were found for both clubfoot groups, when compared to the group of children without clubfoot. Contralateral feet in children with unilateral clubfoot acted similarly to control feet when we observed discrete gait parameters, creating asymmetries in children with unilateral idiopathic clubfoot. Nevertheless, the results of the multivariate measurements of GDI and GDI-k indicate an overall global adaptation of contralateral feet in achieving gait symmetry.

Our findings indicate the same gait outcome in treated idiopathic clubfoot, independent of foot involvement. The only discrepancy we found when comparing to controls was in the hip abduction moment. However, as both bilateral and unilateral clubfoot had almost the same abduction moments, the statistical discrepancy was most likely due to different sample sizes. Overall, the mean values of the gait parameters were surprisingly similar between bilateral and unilateral clubfoot. Equally, the 95% CIs of the mean differences in the parameters deviating from control were observed adjacent to each other in the bilateral and unilateral clubfoot groups (Figure 2). Thus, we argue that bilateral and unilateral idiopathic clubfoot may be considered to be 1 homogeneous group in future gait studies. However, when assessing gait in children with idiopathic clubfoot, e.g. in a clinical setting without the use of a gait analysis system, awareness of foot involvement is important. This is evident in children with unilateral idiopathic clubfoot, as asymmetries were observed for discrete gait parameters, which might give the observer the notion of a poor treatment result. In children with bilateral clubfoot, no statistically significant differences were found between the right foot and the left foot, yet we must emphasize that asymmetries might still be present in an individual child.

In our cohort, decreased plantarflexion appears to be the main concern in idiopathic clubfeet. This was evident for all parameters where plantarflexion was included (passive range of motion, kinematics, moment, and power). The observed decreases in plantarflexion and also ankle power are consistent with several other gait studies of children with idiopathic clubfoot (Karol et al. 2009, Church et al. 2012, Duffy et al. 2013, Mindler et al. 2014). The findings indicate weak plantarflexors, which is probably the reason for the observed power shift from the ankles towards the hips (ankle/hip power ratio). This shift has also been reported in children with juvenile idiopathic arthritis with foot involvement (Esbjornsson et al. 2015). Moreover, the 24% reduced ankle power found in the clubfeet compared to controls might be the reason for the 21% lower walking capacity in children with idiopathic clubfoot, as found by Lohle-Akkersdijk et al. (2015).

The results of the gait parameters of the contralateral foot in children with unilateral clubfoot indicate that the foot has similar gait development as feet in children who are born without the deformity. For GDI and GDI-k on the other hand, there were differences between the contralateral feet and the controls, these differences indicated a resembling gait pattern between the contralateral and the unilateral feet (Figure 2). Similar to our results, Davies et al. (2001) found no kinematic

deviations and few kinetic deviations of the contralateral foot in children with idiopathic clubfoot who were treated with surgical management. One could argue that the contralateral foot could therefore be used as a reference in gait. However, our findings of deviations in GDI and GDI-k of the contralateral foot reveal overall gait adaptations, and they are in line with several other studies (Maton and Wicart 2005, Wicart et al. 2006, Favre et al. 2007, Cooper et al. 2014). Because of this adaptation, it is not advisable to use this foot as a reference.

The children in our cohort performed the 3D gait analysis as part of the standard treatment program. As gait analysis has been considered useful in objectively evaluating the treatment outcome, it could also support secondary treatment planning such as surgery, brace adjustment, or physiotherapy. Thus, the gait analysis should be conducted as early as possible in order for the child to benefit from it. For practical reasons—and since gait kinematics and kinetics appear to stabilize around 4 years of age—our opinion is that the child should be about 5 years old at the time of gait analysis (Sutherland 1997). Moreover, we believe that both GDI and GDI-k could be useful in providing an overview of gait pathology in children with idiopathic clubfoot. We have found 1 previous study using GDI to evaluate gait in children with clubfoot, which found a GDI of 91 in children treated with the Ponseti method and a GDI of 84 in those treated with surgery (Duffy et al. 2013). Our findings were similar to those receiving the Ponseti treatment, indicating persistent impairments in gait despite treatment.

To our knowledge, GDI-k has not been used before in children with clubfoot. When introducing the index, Rozumalski and Schwartz (2011) found, as in our results, that the unaffected side in children with hemiplegia had a lower GDI-k than the hemiplegic side. Both GDI and GDI-k appear to capture the alteration in gait as seen in the contralateral foot, which is only evident when contemplating global measurements with a large amount of data. More research is needed for us to understand the relationship between gait analysis outcomes and the clinical and functional outcome in children with clubfoot.

The main strength of our study was the composition of the study population, which we believe is a representative sample, since the distributions between foot involvement and sex correspond to those in previous reports (Wallander et al. 2006). Moreover, demographic data and speed were found to be similar between the groups. In cases where children fail to undergo gait analysis, this is often due to organizational difficulties rather than the status of the clubfoot. A limitation was the use of the Plug-in Gait model, which might restrict the findings, since it models the foot as only 1 segment. Nowadays, we include the Oxford foot model, which provides additional information on the foot (Mindler et al. 2014), in our gait analyses. We will adopt this model in future gait studies, together with patient-reported outcome measures.

In summary, our findings indicate similar gait deviations in treated idiopathic clubfoot, independently of foot involvement, with the main concern being reduced plantarflexion.

The contralateral foot in children with unilateral idiopathic clubfoot showed no deviations in discrete gait parameters compared to controls. Thus, one should exercise caution when assessing gait in a clinical setting, as children with unilateral idiopathic clubfoot show asymmetry of gait. Global gait adaptations found in the contralateral foot indicate that it should not be used as a reference.

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