

Development of Foot Length in Children with Congenital Clubfoot up to 7 Years of Age: A Prospective Follow-Up Study

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Research article

Keywords: Clubfoot, Foot length, Foot growth, Relapse

Posted Date: January 11th, 2021

DOI: <https://doi.org/10.21203/rs.3.rs-142033/v1>

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Abstract

Background: Clubfeet are typically shorter than normal feet. This study aimed to describe the development of foot length in a consecutive series of children with congenital clubfoot and to relate foot length to development of relapse and motion quality.

Methods: Foot length was measured every 6 months in 72 consecutive children with congenital clubfoot (29 bilateral) aged from 2 to 7 years. The initial treatment was nonsurgical followed by standardized orthotic treatment. Foot length growth rate was calculated every half year. In children with unilateral clubfeet, the difference in foot length between the clubfoot and the contralateral foot was calculated. Motion quality was evaluated by the Clubfoot Assessment Protocol (CAP). Student's *t* test, the Mann–Whitney *U* test and Spearman's correlation were used for group comparisons. Bonferroni correction was used when multiple comparisons were performed.

Results: Clubfeet were smaller ($P < 0.001$) than reference feet at all ages but had a similar growth rate up to age 7. Unilateral clubfeet with greater difference in size compared with the contralateral foot at the first measurement, relapsed more frequently ($P = 0.016$) and correlated with poorer motion quality ($r = 0.4$; $P = 0.011$).

Conclusions: Clubfeet were smaller than reference feet at all ages but had a similar growth rate. Children with unilateral clubfeet and greater foot length difference at 2 years of age had a higher tendency to relapse and poorer motion quality at 7 years of age.

Background

Congenital clubfoot includes hindfoot equinus and varus, midfoot cavus, and forefoot adduction [1, 2]. The characteristic abnormal foot positioning is a result of altered alignment, orientation and shape of bones and joints of the lower limb associated with soft-tissue abnormalities, such as muscle atrophy and a high proportion of adipose tissue [1, 2]. Even well-treated clubfeet are usually smaller due to bone hypoplasia and soft-tissue contractures [1–4].

In our clinical settings, we have observed irregular foot growth in relapsed clubfeet and poorer motion quality in children with smaller clubfeet compared with children with larger clubfeet. Foot size at birth can affect treatment results as it is sometimes more difficult to correct the initial deformity in smaller feet [4–7]. Hemo et al. found that severity score at baseline and number of casts needed for correction correlated with foot length before the start of treatment [7]. Foot size at end of growth can also depend on the treatment method [8–10]. Wallace et al. showed that in unilateral cases, nonsurgically treated clubfeet were 1.3 shoe sizes larger than surgically treated clubfeet, and that both surgically and nonsurgically treated clubfeet were significantly shorter than the contralateral feet at ages 10 to 12 years [8]. Kesemenli et al. showed that unilateral clubfeet were 9–15 mm shorter than the contralateral feet depending on the age and treatment method, with the surgically treated clubfeet showing a greater difference [10]. Additionally, Cooper and Dietz showed that unilateral clubfeet at maturity were, on average, 10 mm

shorter than the contralateral feet [11]. While foot length measurements have been used during initial treatment and follow-up [12], to our knowledge there are no longitudinal studies evaluating clubfoot growth or the usability of such measurements.

The aims of this study were to (1) describe the development of foot length from 2 to 7 years of age in children treated for congenital clubfoot and (2) analyze the relationship between foot length and (a) relapse and (b) motion quality.

Methods

Seventy-eight children were born with idiopathic congenital clubfoot within our catchment area from 1995 to 2007 and were invited to participate in this prospective longitudinal study. Children with eight examinations or more with at least 6 months between each visit were included. The median age at the first foot length measurement was 2 years (range, 2–3 years). The median number of measurements for each child was 10 (range, 8–11).

Initial treatment and follow-up

Clubfeet were initially corrected using either the Copenhagen stretching method or the Ponseti casting technique (Fig. 1) [13, 14]. Percutaneous Achilles tenotomy was performed on 26 feet and posteromedial release on 22 feet. In both groups, all feet were fully corrected before starting the orthotic treatment. Until the age of 4, all children had the same orthotic treatment with individually made dynamic orthoses [15]. These types of orthoses have shown good results, comparable with the results obtained with Foot Abduction Orthosis [15]. The children were followed according to a standardized protocol [16]. They were treated once every 1–3 weeks in the initial months, once every 3 months to the age of 2 years and then every 6 months from ages 2 to 7 years. The assessment included foot length measurement and physical examination according to the Clubfoot Assessment Protocol (CAP) (Fig. 2) [17–19].

Foot measurements

Foot length, including the contralateral nonaffected foot in children with unilateral clubfoot, was measured according to a standardized procedure. The child sat on a chair with ankles, knees, and hips in 90° flexion. A line was drawn around the foot with the pen kept vertical [20]. Parallel lines were drawn distally and proximally. To measure foot length from the drawings, the proximal line was drawn perpendicular to an imaginary line passing through the middle of the hindfoot. The distal line was drawn parallel to the proximal line, including the edge of the distal point of the foot. The distance between these lines was defined as foot length (Fig. 3). The same experienced physiotherapist (HA) made all the drawings. The first author (EM) later performed all foot length measurements.

The nonaffected foot in children with unilateral clubfoot served as the reference foot in the statistical analysis of foot length growth.

Relapse

Relapse was defined as presence of one or more of the following: dorsiflexion $< 0^\circ$ with extended knees; subtalar joint mobility in valgus $< 0^\circ$; foot outward rotation/abduction in relation to tibia $< 5^\circ$; forefoot adduction $> 10^\circ$; and/or in-toeing gait $> 10^\circ$. Relapse was treated by reintroduction of orthosis, serial casting, surgery, or a combination of these treatments where appropriate.

Motion quality

Motion quality was evaluated by the domain "Motion Quality I" (CAP_{MQI}) from the CAP by a single assessor (HA). CAP is a valid and reliable standardized multidimensional observer-administered test providing an overall profile of functional status for each side independently in children with clubfoot [18, 19]. CAP contains 19 items divided into subgroups (Fig. 2). The scoring for each item is graded from 0 (severe reduction/no capacity) to 4 (normal). Each grade is defined by specific criteria [16]. CAP_{MQI} includes four items: walking, running, toe walking, and heel walking (0–16 points). The last CAP_{MQI} registered between the ages of 6 to 7 years was used in the analysis. Scores ≤ 12 were considered as poor clinical outcomes, according to previously established cutoff points [21].

Data analysis and statistics

In children with bilateral involvement, the right or left foot was selected based on inclusion numbers, generating an equal number of left and right feet.

Foot length growth percentage (FLG%) was calculated by subtracting the previous foot length value from the current value. That value was then divided by the previous foot length value and multiplied by 100 to express foot length growth in percent. For example, the FLG% between ages 2 and 2.5 years equals (foot length at 2.5 years – foot length at 2 years) / foot length at 2 years $\cdot 100$.

Foot length difference percentage (uniFLD%) was calculated for every child with a unilateral clubfoot by subtracting the foot length of the clubfoot from the contralateral nonaffected foot. The difference was divided by the contralateral nonaffected foot length value and multiplied by 100 to express foot length difference in percentage.

Statistical analysis was performed using IBM SPSS Statistics software (version 25; IBM SPSS, Armonk, NY, USA). Student's t test was used to analyze the differences in foot length and FLG% between clubfeet and reference feet and between clubfeet with and without relapse. The paired Mann–Whitney Wilcoxon test was used to compare the distribution of uniFLD% at initial measurement between children with and without relapse before the age of 7. Spearman correlation test was used to analyze the correlation between uniFLD% at initial measurement and CAP_{MQI} at the age of 7. A *P* value of below 0.05 was considered statistically significant. For multiple comparisons, the Bonferroni correction was used. The α was set at 0.05 and the *P* value was adjusted to 0.005. Correlations were interpreted according to Cohen's method as low (0 to ± 0.29), moderate (± 0.30 to ± 0.49), and strong (± 0.5 to ± 1.0) [22].

Written consent was obtained from the caregivers of all participants and the study was approved by the local ethics committee (LU-667-03).

Results

Seventy-two children were included in this study (55 boys and 17 girls, 43 unilateral and 29 bilateral). Thirty children were initially treated with Copenhagen stretching and 42 with Ponseti casting. The median Dimeglio score was 10 (range 7–14). There were no statistically significant differences in relapse rate, baseline severity, gender, or laterality between the treatment groups. Children treated with the Copenhagen stretching method underwent significantly more posteromedial releases (17/30, 60%) at the end of the initial treatment than the children treated with Ponseti casting (5/42, 12%) ($P < 0.001$).

Development of foot length and foot growth

Clubfeet were smaller than reference feet at all ages ($P < 0.005$). Clubfeet grew from a mean and standard deviation (SD) of 134 ± 7 mm at the age of 2 years to a mean of 183 ± 12 mm at 7 years, whereas reference feet grew from a mean of 140 ± 8 mm to a mean of 193 ± 12 mm over the same period (Table 1). FLG% decreased from 5.4% at 2 years of age to 2.3% at 7 years of age (Table 2). Foot length and FLG% were similar among children treated with Copenhagen stretching and Ponseti casting.

Table 1
Foot length (mm) in reference feet* and in clubfeet at different ages

Age (years)	Reference		Clubfeet		P value
	n	mean (SD)	n	mean (SD)	
2	36	140 (8)	61	134 (7)	<i>0.001</i>
2.5	38	148 (8)	67	141 (8)	<i>0.000</i>
3	41	155 (9)	70	147 (9)	<i>0.000</i>
3.5	37	160 (9)	62	152 (10)	<i>0.000</i>
4	43	165 (9)	71	157 (9)	<i>0.000</i>
4.5	42	171 (9)	70	161 (10)	<i>0.000</i>
5	40	176 (10)	69	165 (10)	<i>0.000</i>
5.5	33	179 (10)	57	169 (10)	<i>0.000</i>
6	37	185 (9)	64	174 (10)	<i>0.000</i>
6.5	37	189 (9)	59	179 (9)	<i>0.000</i>
7	35	193 (11)	60	183 (12)	<i>0.000</i>
<i>n, number of measurements; SD, standard deviation. P values after Student's t test. Significance set up to P < 0.005 after Bonferroni correction. *Contralateral feet in children with unilateral clubfoot</i>					

Table 2
Foot length growth percentage in reference feet* and in clubfeet

<i>Age interval (years)</i>	<i>Reference</i>		<i>Clubfeet</i>		<i>P value</i>
	<i>n</i>	<i>mean (SD)</i>	<i>n</i>	<i>mean (SD)</i>	
2–2.5	36	5.4 (4)	61	5.4 (4)	0.971
2.5–3	36	5.0 (3)	65	4.4 (3)	0.300
3–3.5	37	3.5 (2)	61	3.9 (2)	0.362
3.5–4	38	3.3 (2)	62	2.9 (2)	0.329
4–4.5	42	3.4 (2)	69	2.8 (2)	0.077
4.5–5	39	2.9 (2)	67	3.0 (2)	0.900
5–5.5	32	2.8 (2)	55	2.5 (2)	0.419
5.5–6	30	2.4 (2)	52	2.7 (2)	0.464
6–6.5	34	2.4 (2)	55	2.9 (2)	0.203
6.5–7	31	2.6 (2)	49	2.3 (2)	0.688
Total (2–7)	28	40 (6)	49	38 (7)	0.216
<i>n, number of measurements; SD, standard deviation. P values after Student's t test. Significance set up to P < 0.005 after Bonferroni correction. *Contralateral feet in children with unilateral clubfoot</i>					

In children with unilateral clubfoot, the foot length difference percentage (uniFLD%) between the clubfoot and the contralateral nonaffected foot was approximately 3% at 2–2.5 years of age, and 5% from 2.5–7 years of age.

Foot length in relation to relapse

Before the age of 7 years (median 5 years), 23 of the 72 children (32%) were treated for relapse. Seventeen children had one relapse and six children had two relapses (Fig. 4). No differences in relapse rate were seen with respect to initial treatments, unilateral or bilateral involvement or gender. No statistically significant differences were found in foot length or in FLG% between children with and without relapse (Table 3, Fig. 5).

Table 3
Foot length development (mm) in children with clubfeet with and without relapse

Age (years)	Relapse		No relapse		P value
	n	mean (SD)	n	mean (SD)	
2	17	133 (7)	44	134 (8)	0.519
2.5	21	139 (9)	46	142 (8)	0.254
3	22	143 (10)	48	148 (8)	0.051
3.5	18	148 (11)	44	154 (8)	0.033
4	22	154 (10)	49	158 (9)	0.065
4.5	23	157 (10)	47	162 (9)	0.032
5	23	162 (9)	46	167 (9)	0.024
5.5	20	165 (9)	37	171 (9)	0.05
6	21	171 (9)	43	175 (10)	0.074
6.5	20	177 (9)	39	180 (9)	0.314
7	20	181(12)	40	184 (12)	0.354

n, number of measurements; *SD*, standard deviation, *P* values after Student's *t* test. Significance set up to $P < 0.005$ after Bonferroni correction.

Children with unilateral clubfoot that relapsed before 7 years of age had larger uniFLD% at their first measurement around 2 years of age (5%; interquartile range (IQR), 3–7%), compared with those without relapse (3%; IQR, 0.75–5%) ($P = 0.016$).

Foot length difference in relation to motion quality in children with unilateral clubfoot

The median CAP_{MQI} at the last measurement of all clubfeet (median age, 7 years; range, 5.5–7 years) was 13 (IQR, 12–15), with 76% of the children having scores above cutoff, indicating good motion quality [21]. In children with unilateral clubfoot, a moderate correlation was found between the uniFLD% at baseline and CAP_{MQI} at 7 years of age ($r = 0.4$; $P = 0.011$).

Discussion

This prospective longitudinal study included a consecutive cohort and aimed to evaluate the development of foot length and foot growth in children with clubfoot from 2 to 7 years of age. The study cohort is representative with respect to unilateral and bilateral involvement, gender distribution, and relapse rate [24–29]. We found that clubfeet were shorter than reference feet at all ages. However,

clubfeet growth after the age of 2 years was similar to the growth of reference feet. Children with unilateral clubfeet, with a greater difference in foot length at initial measurement, relapsed more frequently and had poorer motion quality at 7 years of age.

Muller et al. analyzed foot length development in 10,000 typically developed children and found an average increase in foot length of 51 mm between ages 2 and 7 years [23]. These results are consistent with our findings where reference feet grew at an average of 53 mm between ages 2 and 7 years and clubfeet grew an average of 49 mm. Even though clubfeet were smaller compared with reference feet, the percentage foot length growth of reference feet and clubfeet was similar between 2 and 7 years of age. This could partly be explained by decreased bone hypoplasia as suggested by Beck et al. [3]. They found that bone hypoplasia decreased with age when evaluating children with clubfeet between the ages 2 to 4. In our study, a minor growth slowdown was observed in all clubfeet at ages 3.5 to 4.5 years, possibly caused by the absence of daily stretching when the orthosis treatment ended (Table 2).

The relapse rate in this study was 32%. Previous studies have reported relapse rates ranging from 3.7% up to 53% depending on the initial treatment method, bracing protocol, follow-up time, and relapse criteria [24–28]. Most (87%) of the relapses in this study were observed after completion of orthosis treatment. This coincides with increased variation in foot length and foot growth, indicating the importance of daily stretching to prevent relapse. Thus, careful follow-up of clubfoot development after ending orthosis treatment is imperative to detect early relapse.

Small clubfeet have been associated with difficulties at initial correction and increased risk of relapse [4, 6, 7]. We found that a greater difference in foot length at baseline in children with unilateral clubfeet was related to an increased number of relapses and worse motion quality score at the age of 7. These findings indicate that foot length at the age of 2 years could be used as a prognostic tool. Estimating the exact relationships between foot length at an early age and risk of relapse and poor motion quality later in life could be of value for clinical treatment and follow-up planning. In addition to the predictive value of systematically measuring foot length, foot drawings are an easy and inexpensive method to monitor foot growth and shape, providing informative visual feedback on clubfoot development. Furthermore, it is easily understood by both patients and parents.

As multiple comparisons were made, the Bonferroni correction was applied. Without this correction, the differences in length and growth between clubfeet with and without relapse were significant at around the ages of 3 to 5 years, when most of the relapses occurred (Tables 3 and 4). This finding is consistent with our clinical observation that clubfeet growth occasionally slows down during relapse and normalizes after appropriate intervention. However, we cannot exclude the possibility that a type 2 error occurred when foot length and FLG% were compared [30].

In this study, the contralateral feet were used as reference feet, which could be considered a limitation [31]. However, the reference feet in our study did not differ in size from typically developing feet as described in the literature [23]. The intra- and interrater reliability have not yet been established for

measurements made using the foot drawing method. In our study, the same assessors performed all drawing (HA) and length measurements (EM) to minimize operator error.

Another limitation is that the treatment methods used in our cohort are not gold standard and the results need to be confirmed in children treated within the strict Ponseti protocol. On the other hand, clubfeet in our study showed the same growth rate as reference feet, indicating generalizable results.

Conclusions

In conclusion, clubfeet were smaller than reference feet at all ages, but had, in general, a similar growth rate up to 7 years. Children with unilateral clubfeet that had greater foot length difference at 2 years of age had a higher tendency to relapse and poorer motion quality at 7 years of age.

Abbreviations

CAP
Clubfoot Assessment Protocol
CAP_{MQI}
Clubfoot Assessment Protocol Motion Quality I
FLG%
Foot length growth percentage
uniFLD%
Unilateral foot length difference percentage
SD
Standard deviation

Declarations

Ethics approval and consent to participate

The study was approved by the Ethics Board at Lund University (LU-667-03). Written consent was obtained from the caregivers of all participants.

Consent for publication

All authors read the final version of this manuscript and gave their consent for publication.

Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author on request.

Competing interests

The authors declare that they have no competing interests.

Funding

The study was supported by grants from: the Faculty of Medicine, Lund University, Region Skåne, Erik and Angelica Sparre Foundation, Promobilia Foundation, Kockska Foundation and "Stiftelsen för bistånd åt rörelsehindrade i Skåne".

Authors' contributions

EM: design of the study, collection of data, statistical analysis, interpretation of data, drafting of the manuscript.

ACE: design of the study, interpretation of data, critical review of the manuscript.

GH: design of the study, interpretation of data, critical review of the manuscript.

HA: concept and design of the study, and interpretation of data and critical review of the manuscript.

Acknowledgments

The authors want to thank Axel Ström (Medical statistician) for assistance with the statistical analysis.

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Figures

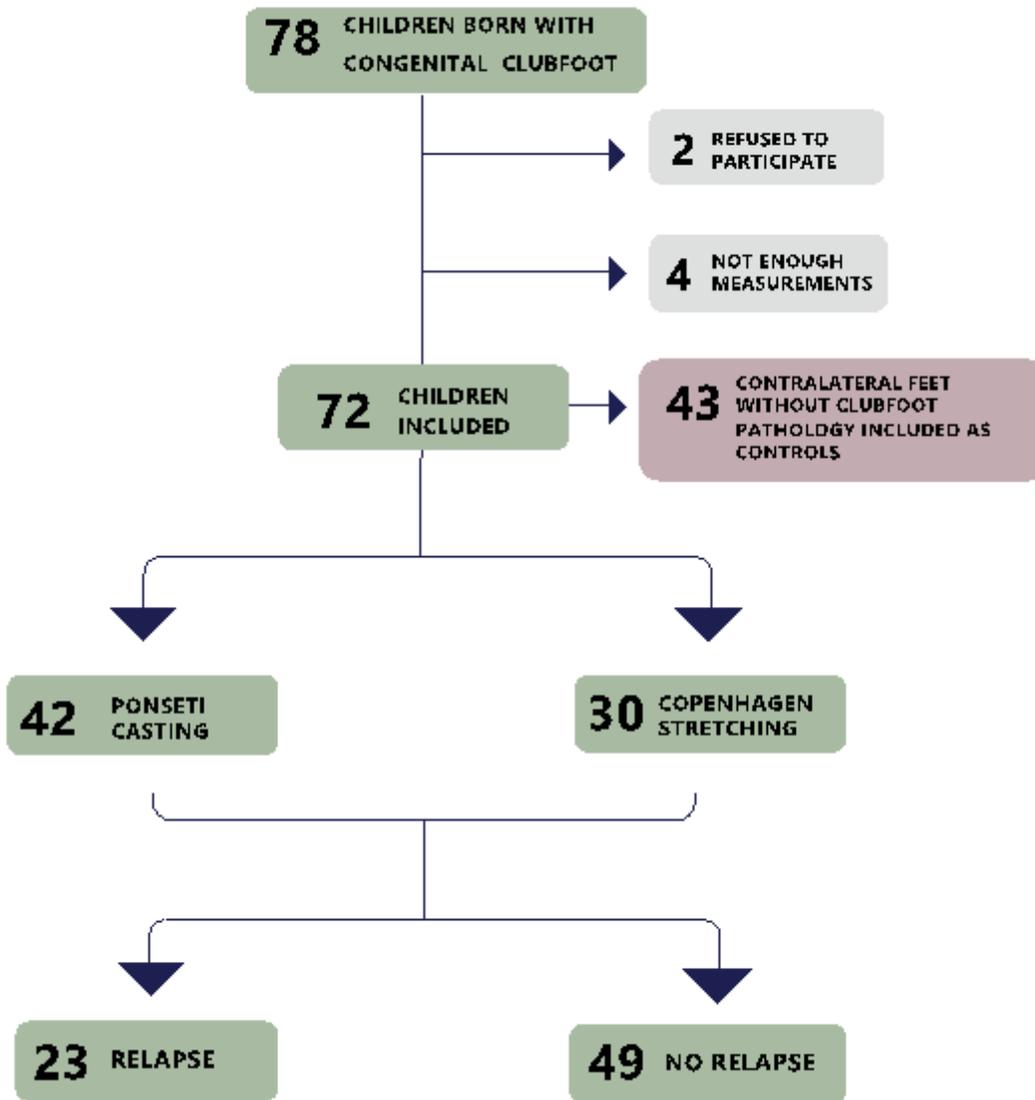


Figure 1

Flowchart of included children with clubfoot.

Clinical examination and motion quality assessment (CAP version 1.2)

Name: _____ Date of birth: _____
 Date of assessment: _____ Assessment number: _____
 Side: ○ Left ○ Right

Rating	0	1	2	3	4
Passive mobility					
I.					
1. Dorsiflexion	< -10°	-10°- < 0°	0°- < +10°	+10°- +20°	>+20°
2. Plantar flexion	0°- < 10°	10°- < 20°	20°- < 30°	30°- 40°	>40°
3. Varus/valgus	>20°var	20°- > 10°var	10°- > 0°var	0°- neutral	>0°vlg
4. Derotation	>20°inv	20°- > 10°inv	10°- > 0°inv	0°- 10°evr	>10°evr
5. Add/abd, ff	>20°add	20°- > 10°add	10°- > 0°add	0°- neutral	>0°abd
II.					
6. Flx.dig.long.	+ reduced		reduced		normal
7. Flx.dig.hall.	+ reduced		reduced		normal
Muscle function					
8. M. peroneus	absent/poor		reduced		normal
9.M. ext.dig.long	absent/poor		reduced		normal
Morphology					
10. Tib.rotation	+ inward		inward		normal
11. Calcaneus pos.	> 10° varus		10°- >0°varus		neutral/ vlg
12. Forefoot pos.	> 20°add		20°- 10°add		<10° add
13. Foot arch	+ cavus/planus		cavus/planus		normal

Motion quality					
I					
14. Running 2y	cannot	+deviant	deviant	± deviant	normal
15. Walking 2y	cannot	+deviant	deviant	± deviant	normal
16. Toe walking 3y	cannot	+deviant	deviant	± deviant	normal
17. Heel walking 3y	cannot	+deviant	deviant	± deviant	normal

II					
18. 1- leg stand 4y	cannot	+deviant	deviant	± deviant	normal
19. Hop 1 leg 4y	cannot	+deviant	deviant	± deviant	normal

Standard Questions

Pain with activities: Never Sometimes Regular Always
 Stiffness: Never Sometimes Regular Always
 Activity level of the child: Low Normal High
 Shoe problems: None Regular Always Orthopaedics shoes
 Leisure- time activities:
 Does your child experience specific problems in daily life activities
 such as in sports, cycling, playing and keeping up with peers:
 Never Sometimes Regular Always
 Specify problem(s): _____

Specification motion quality

Intoeing
 Lateral loading
 No IC
 Deviant knee motion
 Limp
 Decreased propulsion power
 Co-ordination problems

+ = pronounced / very, ± = slightly, var= varus, vlg= valgus, inver= inversion, evr= eversion, add= adduction, abd=abduction, inw= inward rotation, outw= outward rotation, flx= flexor, dig= digitorum, long= longus, hall= hallux, ext= extensor, tib= tibial, calc= calcaneus, pos= position, y = years

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Figure 2

The Clubfoot Assessment Protocol. Motion Quality I highlighted.

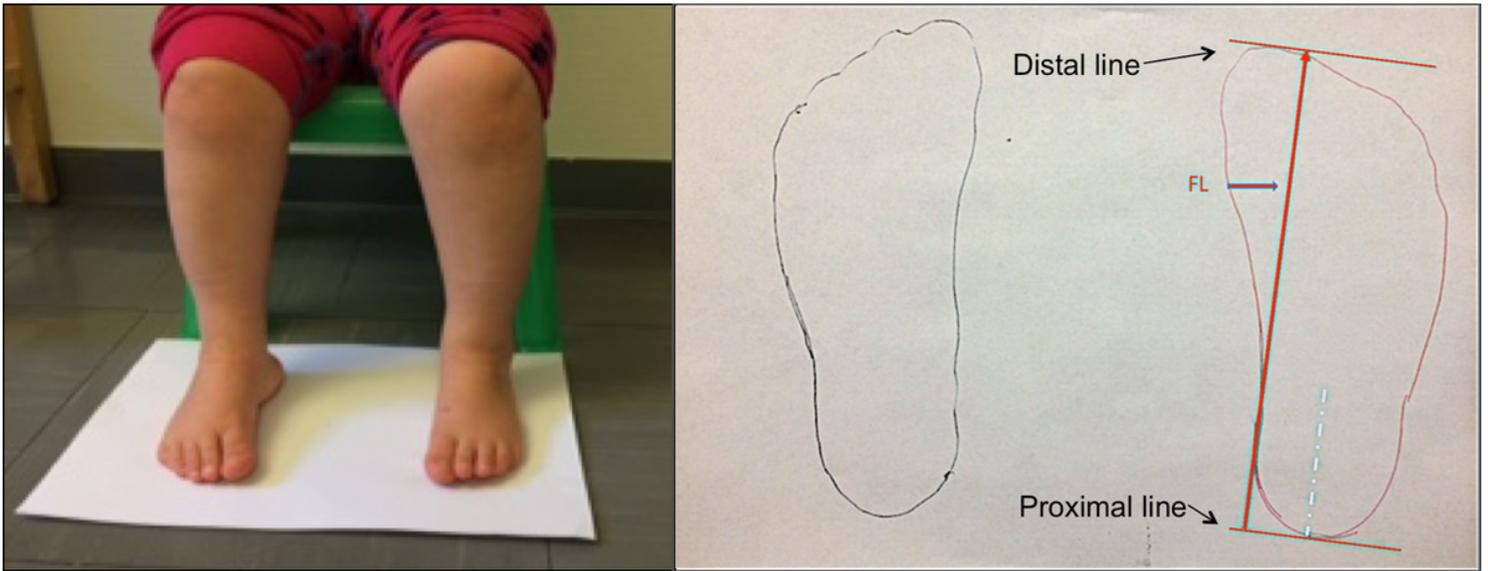


Figure 3

A and B. Foot length drawing and measurement. A: The child sits with the ankles, knees, and hips in 90° of flexion. A line is drawn around each foot with the pen kept vertical. B: Two parallel lines are drawn. The proximal line is drawn perpendicular to an imaginary line that passes through the middle of the hindfoot. The distal line is parallel to the proximal line. The distance between these lines is termed foot length.

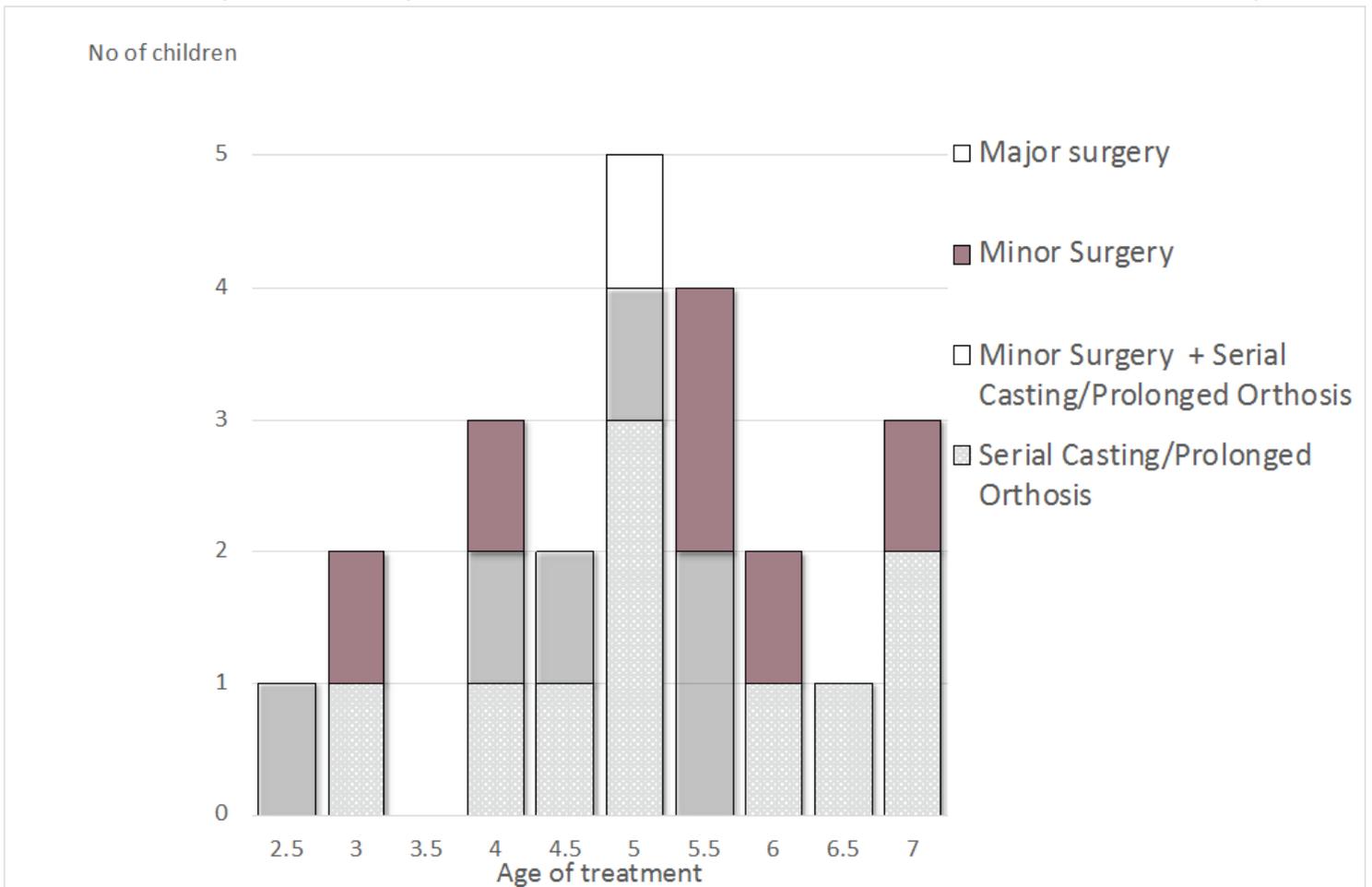


Figure 4

Treatment methods for relapse related to age.

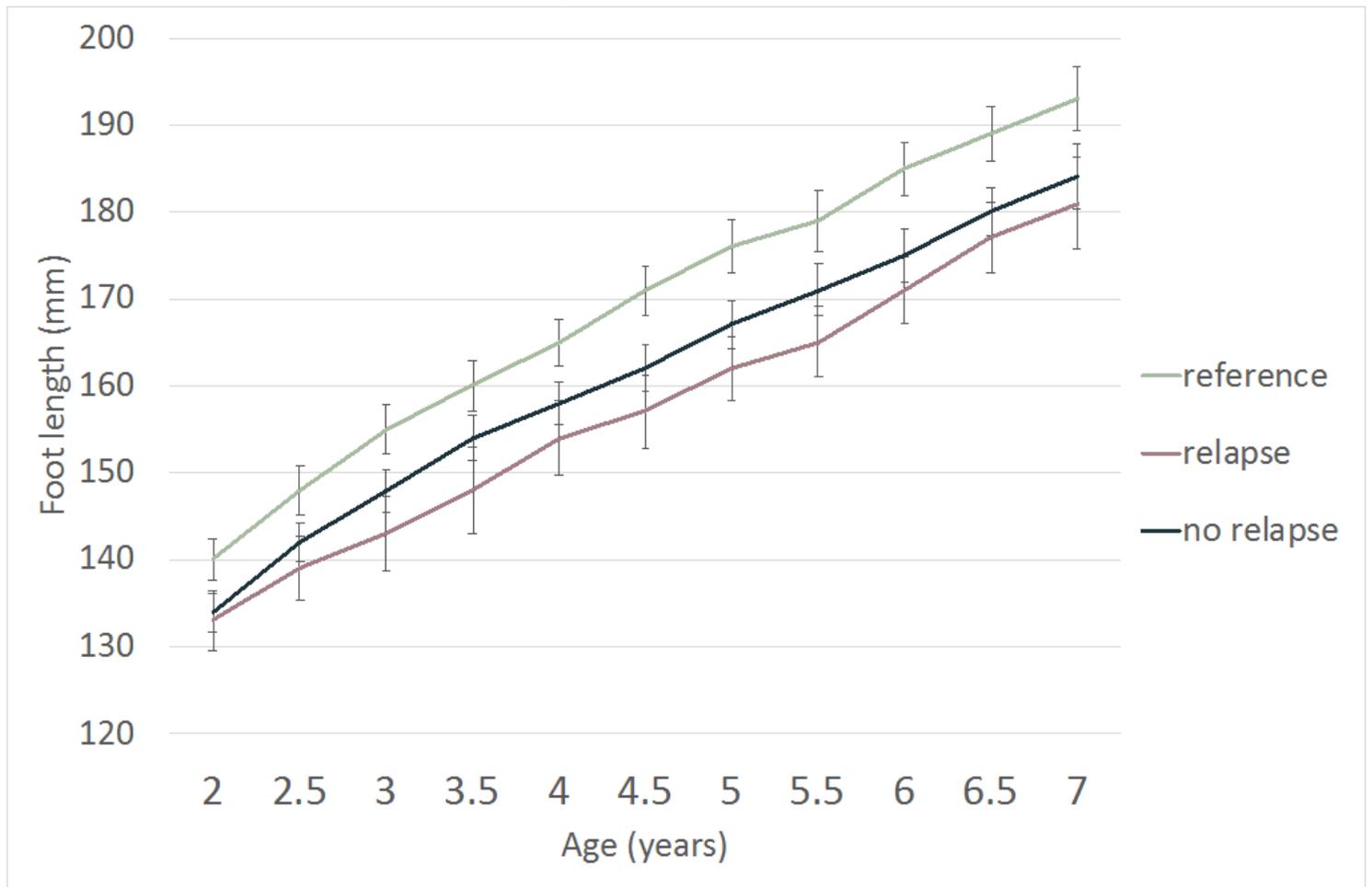


Figure 5

Foot length development in reference feet (green), clubfeet with relapse (pink), and clubfeet without relapse (dark blue). The gray lines show the confidence interval.