

Alteration in hypoplasia of the hindfoot structures during early growth in clubfeet treated using the Ponseti method

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Abstract

Purpose Previous reports have demonstrated diminished size of the hindfoot bones in patients with idiopathic clubfoot deformity. However, no study has quantified the percentage of hypoplasia as a function of early growth, during the brace phase of Ponseti treatment.

Methods We measured the dimensions of ossified structures on radiographs in patients with unilateral Ponseti-treated clubfeet to determine changes in the percentage of hypoplasia between two and four years of age.

Results The degree of hypoplasia varied among the osseous structures in Ponseti-treated clubfeet at age two years, with greater hypoplasia being observed in the talus (7.3%), followed by calcaneus (4.9%) and the cuboid (4.8%). Overall, the degree of hypoplasia diminished by four years, such that the degree of hypoplasia was greatest in the talus (4.2%) and the calcaneus (4.2%) followed by the cuboid (0.6%). At four years of age, the greatest degree of hypoplasia persisted in the talus and calcaneus.

Conclusions Changes occurred in the size of the ossification of hindfoot bones between two and four years of age, and the observed changes in the percentage of hypoplasia varied among the different structures. At four years of age, the greatest percentage of hypoplasia was observed in the talus and calcaneus at values similar to those previously reported in skeletally mature patients. The results suggested that

the relative difference in size of the feet may be expected to remain constant in a child with a unilateral clubfoot after this age.

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Introduction

A diminished length of the affected foot in patients with unilateral clubfoot deformity has been commonly observed. The hypoplasia of the foot is part of a regional hypoplasia found in the affected lower extremities of these patients which includes the tibia and femur.¹⁻³

Segev et al⁴ studied the length of several osseous structures on plain radiographs taken at a mean of 15.2 months in 22 patients with unilateral clubfeet managed with the Ponseti method.⁵ They observed that the size of the ossification centres of the posterior and medial hindfoot structures appeared significantly smaller on radiographs of patients with unilateral clubfeet compared with their uninvolved side. However, they did not provide data on the influence of subsequent growth on these structures during the first several years following treatment.

Following initial correction of clubfeet in infancy using the Ponseti method, a foot abduction orthosis is worn for three to four years to help obviate a relapse of the deformity.⁶ The child's foot grows rapidly during the first four years, reaching approximately 50% of its adult size by 2.5 years of age, and approximately 62% of the adult foot size in males and 67% in females by four years of age.⁷ Despite these observations, to our knowledge, there are no studies evaluating alterations of the growth of the ossification centres of Ponseti-treated clubfeet during this critical period of rapid growth.

We compared the dimensions of the osteocartilaginous structures of affected and unaffected feet in children with unilateral clubfoot treated using the Ponseti method on radiographs taken at two and four years of age. We

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sought to answer the following questions: 1) what are the dimensional differences between the osteocartilaginous structures of the hindfoot in unilateral clubfoot patients treated using the Ponseti method at two and four years of age?; 2) are the ratios of growth deficiency (expressed as percentage of hypoplasia) similar among all bones of the hindfoot? and; 3) how does the ratio of growth deficiency change between two and four years of age?

Patients and methods

Patient population

All patients treated at our Clubfoot Clinic over the past decade have been prospectively invited to participate in an Institutional Review Board-approved long-term clinical outcome database study. All idiopathic, unilateral clubfoot patients treated using the Ponseti method, with a minimum of two years of clinical follow-up, who began treatment prior to six months of age and who received no previous outside surgical treatment were eligible for inclusion. Following correction, all patients were placed in a Mitchell-Ponseti brace (Wayland, Iowa).^{8,9}

There were 66 unilateral clubfoot patients who met the inclusion criteria and had radiographs taken at two years of age. Of those 66 patients, 49 also had radiographs taken at four years of age. In total, 48 of the patients were male and 18 were female. The mean age at the start of treatment was 7.0 weeks (0.9 to 23.3). A mean of 5.2 pre-tenotomy casts were applied (3 to 11) and 61 of 66 patients required a heel cord tenotomy (92.4%). Using the Dimeglio scheme¹⁰, the mean score was 13 (6 to 16), three were moderate, 56 were severe and seven were very severe. During the study period, 32 patients experienced a relapse that necessitated further cast treatment. Of those, only nine patients received an anterior tibial tendon transfer prior to four years of age.

Radiographic measurements

As a routine part of clinical care by the senior author (LEZ), anteroposterior (AP) and lateral standing radiographs of the affected and unaffected feet are taken at two and four years of age. The unaffected contralateral foot in unilateral patients is considered to provide the best possible information by which to assess the affected foot in young growing children. The radiographs were stored in an electronic archiving system (Picture Archiving and Communication System). Standardised methods to correct for errors from magnification have been established and were followed at our institution. Similarly, standardised protocols for positioning of the feet were used to obtain true lateral radiographs.

The following measurements were taken for each pair of AP and lateral radiographs. On each AP radiograph,

measurements included: the length of the first metatarsal excluding non-ossified epiphyses and length of the fifth metatarsal excluding the non-ossified epiphyses (Fig. 1a). On each lateral radiograph, measurements included: lengths of the ossification centres of the calcaneus, talus and cuboid, and the width of the tibia at the level of the distal tibial physis (Fig. 1b and c).

Measurements of all available radiographs were taken by three independent observers (LEZ, JJB, MHJ) who were blinded as to which was the affected foot. The mean differences among the three observers were below 1.0 mm for

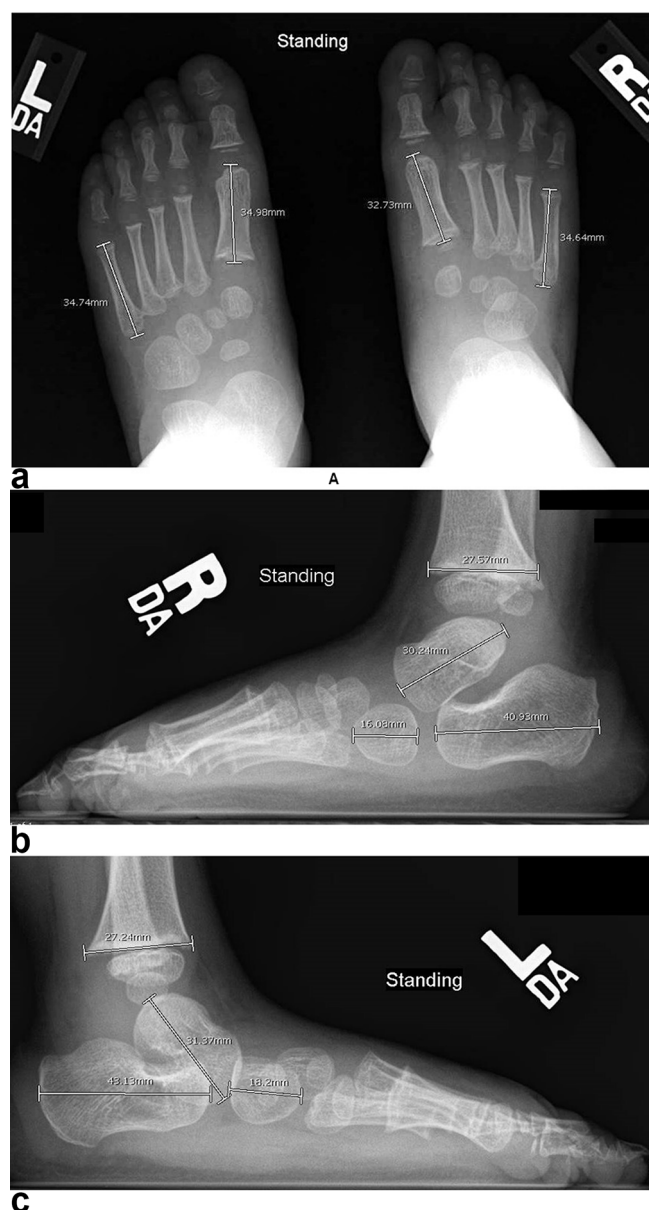


Fig. 1 (a) Depiction of anteroposterior radiographic measurements of a patient with a right idiopathic clubfoot; (b) lateral measurements of the affected foot; (c) lateral measurements of the unaffected foot.

all measurements. Although several of the differences were statistically significant ($p < 0.05$), the magnitudes of these differences were very small. For example, for tibial width, the mean differences among observers were below 0.3 mm ($p = 0.03$). There were no inter-observer differences exceeding 1.0 mm. Therefore, for the purpose of the present study, for each dimension, the mean of the measurements taken by the three independent observers was used. All measurements were recorded to the nearest tenth of a millimeter.

Statistical analysis

All statistical analysis was performed using SPSS (version 19; IBM Inc., Armonk, New York). Paired *t*-tests were used to compare measurements made by each pair of observers. The difference in each of the measurements between affected and unaffected foot for every individual patient were compared using paired *t*-tests. The percentage of hypoplasia for each osseous structure for each patient was calculated using the following formula: (Unaffected foot – Affected foot) / (Unaffected foot × 100). A general linear model was constructed to compare the ratio of hypoplasia among the various osseous structures. This was performed for each measurement at both two and four years, separately. Finally, hypoplasia at two and four years for patients with both sets of radiographs ($n = 49$) was compared for each dimension using paired *t*-tests.

Results

Dimensional differences between affected and unaffected structures in unilateral clubfeet

All the dimensions reported are based on clinical radiographic measurements, which include an approximate magnification of $< 5\%$. Nearly all measurements comparing the affected and unaffected feet at two years were significantly different, $p < 0.05$, indicating slightly larger dimensions on the unaffected side (Table 1). The only measurements that were not statistically different between affected and unaffected feet in the same patient, were the AP fifth metatarsal ($p = 0.73$) and lateral tibia width ($p = 0.37$) (Table 1). All dimensions that were statistically significant were larger on the unaffected side as compared with the affected side, by as much as 1.9 mm (lateral talus length, $p < 0.001$, Table 1).

Likewise, at four years, nearly all measurements comparing the affected and unaffected feet were significantly different, $p < 0.05$, indicating slightly larger dimensions on the unaffected side in most cases (Table 2). The only measurements that were not statistically different between affected and unaffected feet in the same patient at four years were the AP fifth metatarsal ($p = 0.15$) and lateral cuboid length ($p = 0.44$) (Table 2). Further, the lateral

Table 1. Comparison of length in millimeters for affected versus unaffected foot at two years

Osseous measurement	Affected (sd)	Unaffected (sd)	Significance (p-value)
AP first metatarsal length	29.2 (2.5)	30.1 (2.5)	< 0.001
AP fifth metatarsal length	28.5 (2.4)	28.5 (2.4)	0.73
Lateral calcaneus length	33.6 (2.8)	35.4 (3.0)	< 0.001
Lateral talus length	22.2 (2.6)	24.1 (2.7)	< 0.001
Lateral cuboid length	13.9 (1.2)	14.7 (1.5)	< 0.001
Lateral tibia width	24.4 (1.9)	24.6 (1.9)	0.37

AP, anteroposterior

Table 2. Comparison of length in millimeters for affected versus unaffected foot at four years

Osseous measurement	Affected (sd)	Unaffected (sd)	Significance (p-value)
AP first metatarsal length	37.6 (4.1)	38.3 (4.3)	0.01
AP fifth metatarsal length	36.6 (3.8)	36.4 (3.7)	0.15
Lateral calcaneus length	43.5 (3.1)	45.4 (3.5)	< 0.001
Lateral talus length	32.4 (3.5)	33.9 (3.7)	< 0.001
Lateral cuboid length	18.9 (1.9)	19.1 (1.9)	0.44
Lateral tibia width	28.1 (2.2)	27.8 (1.9)	0.03

tibial width was slightly larger on the affected side than the unaffected side by 0.3 mm ($p = 0.03$, Table 2).

Changes in ratio of growth deficiency from two to four years of age

At two years of age ($n = 66$), the structures with the greatest percentage of hypoplasia were the talus (7.3%), cuboid (4.9%) and calcaneus (4.8%), followed by the first metatarsal (3.0%), distal tibial metaphysis (0.5%) and fifth metatarsal (-0.2%) (Fig. 2). At four years of age ($n = 49$), the structures with the greatest percent of hypoplasia were: talus (4.2%), calcaneus (4.2%) and first metatarsal (1.8%), followed by the cuboid (0.6%). Two structures did not show hypoplasia: the fifth metatarsal (-0.6%) and distal tibial metaphysis (-1.4%) (Fig. 3). The percentage of cuboid hypoplasia diminished, while hypoplasia of the talus and calcaneus remained largely unchanged between two and four years of age.

Of the 66 patients who were included in the two-year radiographic analysis, 49 also had four-year radiographs available for analysis. The results of the paired analysis for these 49 patients indicated a decrease in percentage of hypoplasia of the cuboid length, from 5.1% at two years to 1.5% at four years ($p < 0.001$, Table 3). While other dimensions also decreased from two to four years, none were statistically significant ($p > 0.10$, Table 3).

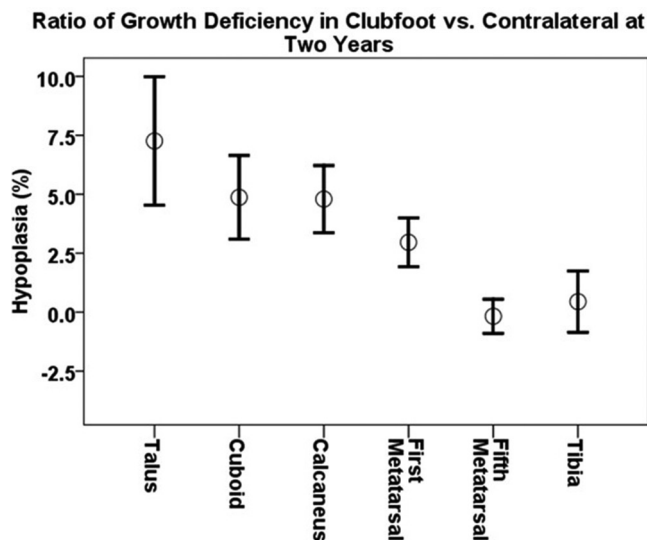


Fig. 2 Ratio of the growth deficiency (percentage hypoplasia) in affected versus unaffected, contralateral feet of unilateral clubfoot patients at two years of age.

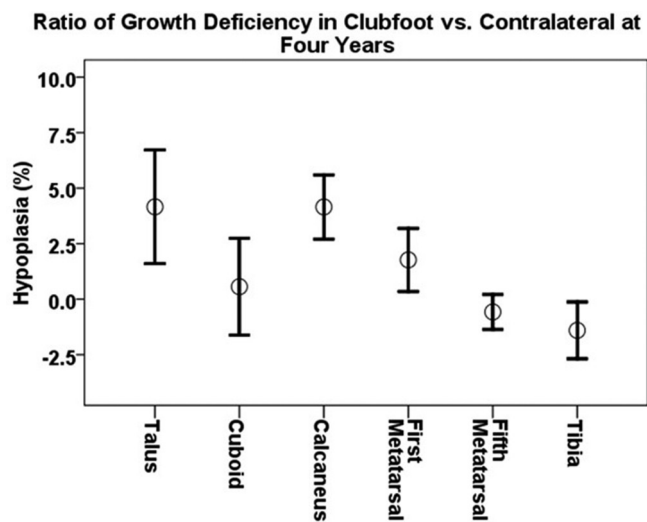


Fig. 3 Ratio of the growth deficiency (percentage hypoplasia) in affected versus unaffected, contralateral feet of unilateral clubfoot patients at four years of age.

During the interval between the two and four-year radiographic measurements, nine patients included in this study experienced a relapsed deformity and their families opted to have their child undergo an anterior tibial tendon transfer (ATTT) procedure. The percentages of hypoplasia in all six osseous structures at four years for these patients was compared with those of the patients who did not receive an ATTT. Overall, patients who received an ATTT had a greater percentage of hypoplasia than those who did not have surgery, however, none of the differences were statistically significant ($p > 0.05$).

Table 3. Comparison of ratios of growth deficiency between two-year and four-year measurements*

Osseous measurement	Two-year % (sd)	Four-year % (sd)	Significance (p-value)
AP 1st Metatarsal Length	2.5 (4.3)	2.4 (3.8)	0.79
AP 5th Metatarsal Length	0.0 (3.0) [†]	-0.7 (2.8) [†]	0.12
Lateral Calcaneus Length	4.8 (6.4)	4.4 (4.9)	0.55
Lateral Talus Length	6.1 (10.9)	4.3 (7.1)	0.11
Lateral Cuboid Length	5.1 (7.2)	1.5 (6.3) [†]	0.00
Lateral Tibia Width	0.0 (4.3) [†]	-1.4 (4.4)	0.11

*the ratios presented in this table include only those patients for whom both two-year and four-year follow-up radiographs were analysed and therefore could be included in the paired analysis ($n = 49$ patients). The p-value in the last column is calculated using a paired *t*-test

[†]indicates that the difference between affected and unaffected foot for this measurement was not significant (Table 2)

Discussion

In the present study, the degree of hypoplasia varied among the osseous structures in Ponseti-treated clubfeet at age two years; greater hypoplasia was observed in the talus (7.3%), followed by the cuboid (4.9%) and the calcaneus (4.8%). Overall, the degree of hypoplasia diminished between age two and four years, such that by age four years the degree of hypoplasia in the hindfoot structures was greatest in the calcaneus (4.4%) and the talus (4.4%) followed by the cuboid (0.6%). The degree of hypoplasia diminished most notably in the talus and cuboid. The greatest degree of hypoplasia was present in the talus and calcaneus at four years of age.

Segev et al⁴ studied the length of several osseous structures on radiographs taken at a mean of 15.2 months in 22 patients with unilateral clubfeet managed with the Ponseti method. These investigators noted that the size of the posterior and medial osteocartilaginous hindfoot structures appeared significantly smaller on radiographs of patients with unilateral clubfeet compared to their uninvolved side. However, they did not provide data on the influence of subsequent growth of these structures in Ponseti-treated feet. They reported that the percentage of hypoplasia was 17% in the talus and 4% in the calcaneus. While the percentage of calcaneal hypoplasia noted by these authors was similar to that found in our study, we observed the percentage of hypoplasia in the talus was smaller, declining from 7.3% at age two years to 4.3% at four years. In comparing the results, it is important to note that we measured length of the talus on the lateral view, while Segev and colleagues⁴ measured the talus on the AP view, which may have resulted in a diminished length of the talus in their study given the normal sagittal inclination of this bone.

In the present study, the cuboid showed a large decrease in the percentage of hypoplasia from 4.9% at

age two years to 0.6% at age four years. Gross anatomical studies of stillborn infants with clubfoot¹¹⁻¹³ as well as MRI studies in newborn clubfoot patients¹⁴ have demonstrated that the cuboid is medially positioned. In patients with clubfeet managed using the Ponseti method, the cuboid corrects to a more anterior and lateral position. Ippolito and Ponseti¹³ postulated that improvement of the skeletal deformities in clubfeet following manipulation and cast treatment may lead to more normal skeletal growth by restoring properly directed mechanical stimuli. While our study found the growth and ossification of the cuboid began to approximate the size of the structure in the unaffected foot by four years of age, future studies are needed to confirm that the changes in the cuboid are the result of an improved mechanical environment in a treated clubfoot.

Ponseti et al¹⁵ measured the size of the bones on radiographs taken in 32 patients with a unilateral clubfoot deformity after skeletal maturity. They found that the talus length was 5.2% smaller, and the calcaneal length was 3.8% smaller in the clubfeet compared to the bones of the unaffected feet. They reasoned that this hypoplasia was carried over from the congenital deformity present when treatment began, however, measurements of these osseous structures during early childhood were not available for their review. In the present study, patients who were four years of age showed the percentage of hypoplasia was 4.3% for talus and 4.4% for the calcaneus. Comparing our findings with those of Ponseti et al¹⁵ in skeletally mature patients, suggests that the changes in the degree of hypoplasia we observed in the talus and calcaneus in patients four years of age, may persist until adulthood.

Limitations

While this is the largest scale study to date on this topic, of the 66 unilateral clubfoot patients with available two-year radiographs, only 49 of those were old enough to have four year radiographs available for measurement. Further, although every effort was made to ensure uniform positioning of the feet, radiographs of young children can be difficult to consistently reproduce and positional alterations could have affected the data measurements.

We used plain radiographs to measure the length of the bones which were not completely ossified. However, data reported by Hubbard et al¹⁶ who quantitated the percentage of ossification of bones in the immature foot, found that the percentage of ossification in the talus was approximately 60% at 24 months and 70% at 48 months of age; the percentage of calcaneal ossification was 83% at 24 months and 88% at 48 months. As the rate of ossification may vary from one tarsal bone to another, we used as a control the percentage of hypoplasia for each bone, relative to the unaffected contralateral. Therefore,

we consider our findings accurate, despite the immaturity of the bones measured in this study. Although ideal, it would clearly not be possible to perform serial MRI studies on these young children due to the risks of sedation and anesthesia that would be unavoidable.

It is likely that the degree of hypoplasia is greater in feet with more severe deformity. Unfortunately, we could not assess this as the number of patients in each severity group was too small to detect a significant difference. Specifically, 56 of the 66 patients were classified as having severe, as opposed to moderate (n = 3) or very severe (n = 7), deformities at initial presentation. Lastly, 32 patients (48.4%) experienced a relapse that necessitated further cast treatment during the period of study, with nine patients undergoing an ATTT. This rate of relapse following initial deformity correction using the Ponseti method in our study patients is comparable with that reported by other investigators.^{5,17-19} No significant difference in the measurements of the osteocartilaginous structures was found in those patients who had relapsed, including those who had a tendon transfer, compared with those who did not.

Conclusion

Following correction of a clubfoot using the Ponseti method, those osteocartilaginous structures of the hindfoot showed the greatest percentage of hypoplasia on radiographs taken at two years of age. The results indicated an overall decrease in hypoplasia of the hindfoot structures in clubfeet between age two and four years. This decrease was most notable in the talus and cuboid. Although these changes may be related to alteration of stresses due to improved alignment of the bones, this hypothesis remains untested. Greater hypoplasia of talus and calcaneus persisted at four years of age, at values similar to those reported in skeletally mature patients, suggesting that the relative size of the feet may be expected to remain constant in a child with a unilateral clubfoot after this age.

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COMPLIANCE WITH ETHICAL STANDARDS

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OA LICENCE TEXT

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ETHICAL STATEMENT

Ethical Approval: Institutional review board approval was obtained for this study.

Informed Consent: All parents of patients included in this study signed an informed consent prior to inclusion in the Clubfoot Study Database.

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