How Reliable Are Standard Radiographic Measures of the Foot and Ankle in Children With Achondroplasia?

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Abstract

Background Radiographic measurements are typically used in achondroplasia (ACH) during correction of lower limb alignment. However, reliabilities for the measurements on weightbearing radiographs of the foot and ankle in patients with ACH have not been described, and the differences between the ACH population and subjects without ACH likewise have not been well characterized; these issues limit the use of studies on this subject.

Questions/purposes We proposed (1) to measure the inter- and intraobserver reliability of a number of

radiographic measures of ankle and foot alignment in an achondroplastic cohort of patients; and (2) to compare our radiographic measurement values with age-matched literature-based normative values.

Methods Ten radiographic measurements were applied to foot and ankle radiographs of 20 children (40 feet) with ACH (mean age, 10 years; range, 8–16 years). Interobserver and intraobserver reliabilities of these radiographic measurement methods were obtained and expressed by intraclass correlation coefficients (ICCs). The mean values were calculated and compared with the literature-based values.

Results The interobserver reliability was excellent for eight measurements with ICCs ranging from 0.801 to 0.962, except for lateral talo-first metatarsal angle and mediolateral column ratio, which were much lower. The intraobserver reliability was excellent for all 10 radiographic measurements with ICCs ranging from 0.812 to 0.998. Compared with existing literature-based values, all 10 measurements had a significant difference ($p < 0.01$).

Conclusions We suggest tibiotalar angle, calcaneal pitch angle, tibiocalcaneal angle, talocalcaneal angle, naviculocuboid overlap, talonavicular coverage angle, metatarsal stacking angle, and AP talo-first metatarsal angle with excellent interobserver and intraobserver reliabilities should be considered preferentially in analysis of foot and ankle alignment in children with ACH.

Level of Evidence Level II, diagnostic study. See Guidelines for Authors for a complete description of levels of evidence.

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Introduction

Achondroplasia (ACH), the most common skeletal dysplasia and the most frequent cause of short-limbed

Fig. 1 Measurement of the tibiotalar angle. A = anatomic axis of the tibia, created by connecting the midpoints of lines ab (proximal medial and lateral margins of the diaphysis of the tibia) and cd (distal medial and lateral margins of the diaphysis of the tibia); B = superior margin of the articular surface of the talus. The tibiotalar angle is the angle created by the intersection of B and the perpendicular to A. Valgus alignment is positive; varus alignment is negative.

Fig. 2 Measurement of the calcaneal pitch angle. A = plantar prominence of the calcaneus proximal to the calcaneocuboid articular surface; B = plantar aspect of the calcaneal apophysis or most plantar prominence of the tuberosity of the calcaneus; C = line along the plantar aspect of the soft tissue shadow of the hindfoot when weight bearing; Calcaneal pitch = angle created by the intersection of AB and C.

Fig. 3 Measurement of the tibiocalcaneal angle. Line AB is as described in Figure 2. C = anatomic axis of the tibia created by connecting the midpoints of lines ab and cd. Tibiocalcaneal angle is created by the intersection of AB and C.

Fig. 4 Measurement of the talocalcaneal angle. Line AB is as described in Figure 2. C = long axis of the talus, created by connecting the midpoints of lines ab (margins of the articular surface of the talus) and cd (margins of the neck of the talus). Talocalcaneal angle is created by the intersection of AB and C.

dwarfism, is characterized by inappropriate differentiation of the growth plate and a deficiency of endochondral growth, which occur because of mutations in the gene that encodes for fibroblast growth factor receptor-3 [11, 13, 16, 21]. In addition to short stature, achondroplastic patients often have spinal anomalies, neurologic and craniofacial abnormalities as well as limb deformities [6, 11]. The most common lower limb anomalies seen in this patient population are genu varum and ankle varus. A study of genu varum in ACH reported that foot and ankle alignment plays a significant part in changing the loading axis of the lower

Fig. 5 Measurement of the naviculocuboid overlap. Points a, b = superior and inferior margins (respectively) of the cuboid; Point c = inferior margin of the navicular; Naviculocuboid overlap = $ac \cdot 100/ab$.

Fig. 6 Measurement of the lateral talo-first metatarsal angle. Line C is as described in Figure 4. A = long axis of the first metatarsal, created by connecting the midpoints of lines ab (distal dorsal and plantar margins of the diaphysis of the first metatarsal) and cd (proximal dorsal and plantar margins of the diaphysis of the first metatarsal). Lateral talo-first metatarsal angle is created by the intersection of A and C. When the first metatarsal is dorsiflexed relative to the talus, the angle is positive.

limb [14] and for that reason, surgical correction of the angular deformities should be considered [1].

The goal of surgery is to improve limb alignment and function long term [11, 21]. Lower extremity radiographs including weightbearing radiographs of the feet and ankles are crucial for preoperative planning [5, 7, 8, 20], but the reliability of radiographic measures in this patient population is unclear [5], and differences between the ACH

population and children without ACH remain poorly characterized.

Thus, we proposed (1) to measure the inter- and intra-observer reliability of a number of radiographic measures of ankle and foot alignment in an achondroplastic cohort of patients; and (2) to compare our radiographic measurement values with age-matched healthy literature-based normative values.

Patients and Methods

We retrospectively reviewed 136 patients with ACH who visited our orthopaedics clinic between April 2004 and July 2012 and underwent a radiographic examination. The radiographic evaluation included a weightbearing lateral foot radiograph, weightbearing AP foot radiograph, and weightbearing AP ankle radiograph. We excluded 116 patients for the following reasons: age younger than 8 years with incomplete ossification of cartilage, age older than 16 years because of previous lower extremity surgery, lack of one or two of the three required radiographs, history of foot and ankle fractures, or any congenital malformation, neuromuscular disease, tumor, or infections that could cause foot and ankle deformities. These exclusions left 20 patients (eight boys and 12 girls). In each of these patients, both sides of lower extremities were symmetrical and had no difference on the radiographic reading. The mean age of patients was 10 years (range, 8–16 years). This retrospective study was approved by the institutional review board at our hospital.

We conducted the sample size analysis to determine the minimum number of patients required according to reliability testing. In this study, minimum sample size for reliability was calculated as 36 feet by setting the intraclass correlation coefficient (ICC) target as 0.8, 95% confidence interval as 0.2, and number of raters as three with a Bonett's approximation [2]. Thus, we selected both sides of 20 patients (40 feet).

The foot and ankle radiographs were taken using a UT 2000 X-ray machine (Philips Research, Eindhoven, The Netherlands) at a source-to-image distance of 90 cm at 50 kVp and 5 mAs with patients standing. Radiographic images were retrieved using a STAR PACS (INFINITT, Seoul, Korea), and radiographic measurements were performed using Pi View STAR software Version 5.0.6.1 (INFINITT). The radiographic imaging and measuring system was digital.

Selection of radiographic measurements was based on a review of the literature. We described a quantitative technique that used 10 radiographic measurements to assess the segmental alignment of the foot based on qualitative techniques derived from the foot model developed by

Fig. 7 Measurement of the talonavicular coverage angle. Points a, b = medial and lateral margins, respectively, of the articular surface of the talus; Points c, d = medial and lateral margins, respectively, of the articular surface of the navicular. Line A is perpendicular to ab, and line B is perpendicular to cd. Talonavicular coverage is the angle created by the intersection of lines A and B.

Fig. 8 Measurement of the metatarsal stacking angle. A = plantar aspect of the head of the great toe metatarsal; B = plantar aspect of the base of the fifth metatarsal; C = plantar aspect of the head of the fifth metatarsal; Metatarsal stacking angle = angle created by the intersection of AB and BC.

Fig. 9 Measurement of the AP talo-first metatarsal angle. A = long axis of the first metatarsal, created by connecting the midpoints of lines ab (distal medial and lateral margins of the diaphysis of the first metatarsal) and cd (proximal medial and lateral margins of the diaphysis of the first metatarsal); B = long axis of the talus, created by bisecting perpendicular to line ef (medial and lateral margins of the articular surface of the talus). The AP talo-first metatarsal angle is the angle created by the intersection of lines A and B.

Inman [9, 10] and Davids et al. [4]. We used the tibiotalar angle (Fig. 1) from the standing AP ankle radiograph and the calcaneal pitch angle (Fig. 2), tibiocalcaneal angle (Fig. 3), and talocalcaneal angle (Fig. 4) from the standing lateral foot radiographs to describe the alignment of the hindfoot. We selected the naviculocuboid overlap (Fig. 5), lateral talo-first metatarsal angle (Fig. 6) from the standing lateral foot radiograph, and the talonavicular coverage angle (Fig. 7) from the standing AP foot radiograph to describe midfoot alignment. We measured the metatarsal stacking angle (Fig. 8) from the standing lateral foot radiograph and the AP talo-first metatarsal angle (Fig. 9) from the standing AP foot radiograph to describe the alignment of the forefoot. The mediolateral column ratio, measured from the standing lateral radiograph, was used to describe the relative length of two columns of the foot (Fig. 10).

During the consensus-building session, measurement techniques and radiographic landmarks were reviewed and clarified with five pediatric orthopaedic surgeons (HRS,

Fig. 10 Measurement of the mediolateral column ratio. A = the distal-most margin of the head of the great toe metatarsal; B = the most proximal margin of the posterior dome of the talus. The distance between A and B represents the length of the medial column. C = the distal-most margin of the head of the fifth toe metatarsal; D = the most proximal margin of the posterior aspect of the calcaneus. The distance between C and D represents the length of the lateral column. The mediolateral column ratio is the length of AB divided by the length of CD.

QLK, KWP, YJJ, JX) with orthopaedic experience of 33, 22, 10, 4, and 3 years, respectively [4]. We accessed the interobserver reliability by comparing the measurements of the 10 angles or ratios for 40 feet among three examiners (KWP, YJJ, JX). Intraobserver reliability sessions were determined by repeated measurements of the 10 radiographs performed by three examiners at separate sessions 4 weeks apart. All examiners were blinded to the measurements made by other examiners and were unaware of the information of the patients' radiographs they measured.

The interobserver and intraobserver reliabilities were determined by using ICCs and their 95% confidence intervals, which were calculated in the setting of a two-way random effect model assuming a single measurement and absolute agreement. An ICC of 1 indicates perfect reliability and an ICC of > 0.8 indicates excellent reliability [17]. Analysis of the values of the entire sample was descriptive with mean values and SDs. Age-matched healthy literature-based normative values of foot and ankle were selected according to a review of the literature. Along with specialty literature book review, we conducted a PubMed search using the key words "normal foot alignment", "normal ankle alignment", and "children AND radiography". One of the orthopaedic surgeons (JX) reviewed the abstracts and articles. The newest age-matched normative values of foot and ankle alignment were summarized in David et al.'s study [4]. Statistical comparison was done using the one-sample t-test.

Statistical analyses were calculated using SPSS for Windows (Version 15.0; SPSS, Chicago, IL, USA).

Results

In terms of intraobserver reliability, all 10 radiographic parameters had excellent ICC values ranging from 0.812 to 0.998 (Examiner 1), 0.814 to 0.996 (Examiner 2), and 0.873 to 0.991 (Examiner 3) (Table 1). Eight of 10 parameters showed excellent interobserver reliability with ICC values ranging from 0.801 to 0.962 (Session 1) and 0.804 to 0.922 (Session 2); the lateral tibio-first metatarsal angle and mediolateral column ratio were not nearly as good with ICC values ranging from 0.504 to 0.791 (Session 1) and 0.473 to 0.674 (Session 2) (Table 2).

Compared with existing literature-based values [4], the mean values of all 10 measurements had a significant difference ($p < 0.01$; Table 3).

Discussion

ACH is a genetic disorder that causes angular limb deformities in children such as genu varum and ankle varus [1]. Usually, the condition requires realignment surgery with a goal of limb alignment correction or limb lengthening [11, 18, 20]. The hip and knee alignment in achondroplastic children has been analyzed by Inan et al. [7, 8]; however, the foot and ankle alignment in achondroplastic children is less well understood and easy to be overlooked [5]. Guichet et al. [5] proposed that planning surgery on limbs requires a good understanding of limb mechanics with precise evaluation of any malalignment problems, including the ground reaction applied to the foot and ankle. Thus, thorough evaluation of foot and ankle alignment before operation is necessary and important. Furthermore, the measurement methods should be relatively easy to apply and should have acceptable reproducibility and reliability. To our knowledge, no previous article has mentioned the reliabilities of these measurements in achondroplastic children. We therefore investigated the inter- and intraobserver reliabilities of radiographic measurements and compared mean values of our results with age-matched literature-based normative values to determine which measurement should be considered preferentially in analysis of foot and ankle alignment in children with ACH. As a result, eight of 10 radiographic measurements had excellent both inter- and intraobserver reliabilities; all 10 values had significant differences from age-matched normative values [4].

Before discussing the findings of our study, some limitations should be present. First, we studied only the

Table 1. Intraobserver reliabilities for 10 radiographic parameters

Measurement	Examiner 1		Examiner 2		Examiner 3	
	ICC	95% CI	ICC	95% CI	ICC	95% CI
Hindfoot						
Tibiotalar angle	0.96	0.915–0.981	0.814	0.610–0.912	0.944	0.882–0.973
Calcaneal pitch angle	0.988	0.976–0.994	0.953	0.901–0.978	0.977	0.951–0.989
Tibiocalcaneal angle	0.992	0.984–0.996	0.974	0.946–0.988	0.991	0.980–0.996
Talocalcaneal angle	0.976	0.950–0.989	0.98	0.958–0.990	0.965	0.926–0.983
Midfoot						
Naviculocuboid overlap	0.978	0.955–0.990	0.951	0.897–0.977	0.982	0.962–0.991
Talonavicular coverage angle	0.812	0.606–0.911	0.992	0.983–0.996	0.873	0.733–0.940
Lateral talo-first metatarsal angle	0.998	0.996–0.999	0.994	0.988–0.997	0.957	0.910–0.980
Forefoot						
Metatarsal stacking angle	0.975	0.948–0.988	0.989	0.976–0.995	0.991	0.981–0.996
AP talo-first metatarsal angle	0.997	0.993–0.998	0.996	0.992–0.998	0.926	0.844–0.965
Columns						
Mediolateral column ratio	0.921	0.833–0.962	0.916	0.823–0.960	0.962	0.920–0.982

ICC = intraclass correlation coefficient; CI = confidence interval.

Table 2. Interobserver reliabilities for 10 radiographic parameters

Measurement	Session 1		Session 2	
	ICC	95% CI	ICC	95% CI
Hindfoot				
Tibiotalar angle	0.857	0.738–0.927	0.895	0.808–0.947
Calcaneal pitch angle	0.927	0.866–0.963	0.917	0.847–0.958
Tibiocalcaneal angle	0.921	0.855–0.960	0.904	0.825–0.951
Talocalcaneal angle	0.884	0.787–0.941	0.848	0.721–0.923
Midfoot				
Naviculocuboid overlap	0.962	0.931–0.981	0.922	0.856–0.960
Talonavicular coverage angle	0.806	0.644–0.901	0.804	0.673–0.898
Lateral talo-first metatarsal angle	0.504	0.090–0.747	0.473	0.033–0.732
Forefoot				
Metatarsal stacking angle	0.84	0.706–0.918	0.821	0.671–0.909
AP talo-first metatarsal angle	0.801	0.636–0.899	0.821	0.672–0.909
Columns				
Mediolateral column ratio	0.791	0.616–0.893	0.674	0.402–0.834

ICC = intraclass correlation coefficient; CI = confidence interval.

relationships of the preoperative radiographic measurements, not the symptomatic correlation of the radiographic measurements or postoperative responsiveness as a result of the retrospective nature of a study. However, most of achondroplastic children came to our clinic because of lower limb deformities without pain or other uncomfortable symptoms. So preoperative radiographic evaluation is necessary. A longitudinal study can also be performed to investigate the clinical relevance of the radiographic measurements. Second, not all the diagnoses in our study

were confirmed by genetic testing. We believe the patients with typical clinical manifestations of ACH do not need genetic analyses for proper diagnosis. Third, the angular deformities are first noticed at the standing age in 40% of all achondroplastic children and progress rapidly at the age of 3 to 4 years, again at 6 to 7 years [12], and the final progression of the deformity takes place during the pubertal growth spurt [14]. The age of our patients ranged from 8 to 16 years with an average age of 10 years. The results might be more representative if the age range was

Table 3. The results of mean values for 10 radiographic parameters and comparison with the age-matched literature-based values

Measurement	Value (range)	Literature value (range) [4]	p value
Hind foot			
Tibiotalar angle (°)	-5.8 ± 5.8 (−20 to 7.5)	1.1 ± 3.75 (−9 to 12)	< 0.001
Calcaneal pitch angle (°)	12.3 ± 5.0 (0.6–22.5)	17 ± 6.0 (5–32)	< 0.001
Tibiocalcaneal angle (°)	71.1 ± 10.3 (50.3–101.2)	69 ± 8.4 (44–86)	0.008
Talocalcaneal angle (°)	39.2 ± 7.9 (13.0–61.5)	49 ± 6.0 (36–61)	< 0.001
Midfoot			
Naviculocuboid overlap (%)	39.6 ± 20.1 (−7.9 to 103.3)	47 ± 13.8 (22–85)	< 0.001
Talonavicular coverage angle (°)	24.5 ± 11.0 (2.0–50.6)	20 ± 9.8 (5–39)	< 0.001
Lateral talo-first metatarsal angle (°)	3.4 ± 10.8 (−32.7 to 28.6)	13 ± 7.5 (1–35)	< 0.001
Forefoot			
Metatarsal stacking angle (°)	1.0 ± 8.4 (−30.2 to 25.0)	8 ± 2.9 (1–13)	< 0.001
AP talo-first metatarsal angle (°)	15.9 ± 10.4 (−4.5 to 48.6)	10 ± 7.0 (−3 to 28)	< 0.001
Columns			
Mediolateral column ratio	1.0 ± 0.1 (0.9–1.2)	0.9 ± 0.1 (0.8–1.1)	< 0.001

wider. However, almost all of our patients older than 16 years underwent previous surgery of the foot, ankle, or lower extremities. Although we avoided the two rapid progressions, the result could not account for significant variability. Fourth, the rotational element in the deformity of foot and ankle in patients of ACH was not assessed in the two-dimensional radiographic methods but it is a major factor in treatment planning. However, the measurement methods introduced in our study can be used to assess almost every achondroplastic patient older than 8 years who comes to the clinic the first time or can be performed as initial assessments before operation. Further three-dimensional examination and dynamic assessment can be performed after this first step.

Of the 10 radiographic measurements of foot and ankle alignment we made, tibiotalar angle, calcaneal pitch angle, tibiocalcaneal angle, talocalcaneal angle, naviculocuboid overlap, talonavicular coverage angle, metatarsal stacking angle, and AP talo-first metatarsal angle had excellent interobserver reliability. All the measurements had excellent intraobserver reliability. Among these, tibiotalar angle, calcaneal pitch angle, tibiocalcaneal angle, and talocalcaneal angle were measured to describe the hindfoot alignment. Lee et al. [14] demonstrated that fibular overgrowth is present in achondroplastic children between 8 and 11 years of age, which leads to limitation of eversion of the hindfoot. Furthermore, the mean values of tibiotalar angle and talocalcaneal angle in our patients were less than those in the literature [4] ($p < 0.001$). That the mean tibiotalar angle value was negative indicated the varus alignment of the ankle. Also, the smaller the talocalcaneal angle, the more varus and adducted the hindfoot. These provided support for the data of Lee et al. [14]. In addition, the mean value of calcaneal pitch angle in our patients was

less than the mean literature value; the mean value of tibiocalcaneal angle was more than the literature value [4] ($p < 0.01$). The smaller the calcaneal pitch angle and the larger the tibiocalcaneal angle, the more plantarflexed the hindfoot. Because this technique of segmental analysis of the foot and ankle is built on combining the biomechanical modeling with the alignment and movement of the three segments [3, 4, 9, 10, 15], if the hindfoot moves toward inversion or varus malalignment, the midfoot will be locked toward supination. The forefoot segment follows the midfoot segment to become pronated. In our study, the naviculocuboid overlap, talonavicular coverage angle, and lateral tibio-first metatarsal angle were selected to describe midfoot alignment. Although only naviculocuboid overlap and talonavicular coverage angle had excellent interobserver and intraobserver reliabilities, the mean values of all three measurements had a significant difference compared with the literature values [4] ($p < 0.001$), which was characterized by supination, abduction, and cavus of the midfoot, respectively. The metatarsal stacking angle and AP talo-first metatarsal angle were measured to describe the forefoot alignment. Both had excellent interobserver and intraobserver reliabilities. Furthermore, the mean value of the former was less than that of the literature [4] ($p < 0.001$), whereas the latter value was more than the literature value [4] ($p < 0.001$), which indicated pronation and abduction of the forefoot. It has been opined that a high arch resulting from the short medial column relative to the lateral column is associated with hindfoot inversion or varus, midfoot supination, and forefoot pronation [4]. However, because the mean value of mediolateral column ratio in our study was more than that of the aforementioned study [4] ($p < 0.001$), the low arch occurred in our patients associated with the same other segmental malalignment

pattern. We suspect the reason for this may be the abduction of the midfoot and forefoot. Of the 10 measurements, the lateral talo-first metatarsal angle and mediolateral column ratio had low interobserver reliability. We believe the ICC values of these two measurements may be affected by a narrow measurement range [19] and unclear landmarks of the first and fifth metatarsals on the standing lateral foot radiographs.

According to the current study, it will be possible to quantitatively describe common patterns of foot and ankle segmental malalignment seen in children with ACH. These measurements are important in evaluating the hindfoot, midfoot, and forefoot deformities. Surgical planning should focus not only on the lower limb alignment [11, 20], but also on the complex deformities of the foot and ankle. Further research will apply these radiographic measurements to a series of affected feet in patients who have undergone operations for axial deviations to confirm whether these measurement methods of “virgin” feet are equally valid in postoperative patients with ACH. However, because the growth plate is still open in patients around 10 years of age, the malalignment of the lower limb might recur after axial deviation is corrected. Increasing followup times can make these measurements more representative.

In conclusion, the reliable measurement methods that are applicable for foot and ankle alignment evaluation in children with ACH include tibiotalar angle, calcaneal pitch angle, tibio-calcaneal angle, talocalcaneal angle, naviculocuboid overlap, talonavicular coverage angle, metatarsal stacking angle, and AP talo-first metatarsal angle with excellent interobserver and intraobserver reliabilities. Lateral talo-first metatarsal angle and mediolateral column ratio should be used as complementary measurement methods, because all values of 10 measurements had a significant difference from the age-matched literature-based normative values.

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