Foot and Ankle Surgery 25 (2019) 378–382

Contents lists available at ScienceDirect

Foot and Ankle Surgery

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

Natural progression of radiographic indices in juvenile hallux valgus deformity

Ki Hyuk Sung[a,1], Soon-Sun Kwon[b,1], Moon Seok Park[a], Kyoung Min Lee[a], Jonghyun Ahn[c], Seung Yeol Lee[c,*]

[a] Department of Orthopaedic Surgery, Seoul National University Bundang Hospital, 82 Gumi-ro173beon-gil, Bundang-gu, Seongnam-si, Kyungki 13620, Republic of Korea

[b] Department of Mathematics, College of Natural Science, Ajou University, 206 Worldcup-ro, Yeongtong-gu, Suwon-si, Kyungki 16499, Republic of Korea

[c] Department of Orthopaedic Surgery, Ewha Womans University Mokdong Hospital, 1071 Anyangcheon-ro, Yangcheon-gu, Seoul 07985, Republic of Korea

A R T I C L E I N F O

Article history:
Received 11 October 2017
Received in revised form 4 December 2017
Accepted 2 February 2018

Keyword:
Juvenile hallux valgus
Linear mixed model
Hallux valgus angle
Radiographic index

A B S T R A C T

Background: This study aimed to estimate the annual change in radiographic indices for juvenile hallux valgus (JHV) and to analyze the factors that influence deformity progression.

Methods: Patients aged <15 years who had JHV and were followed up for at least 1 year were included. Hallux valgus angle (HVA), hallux interphalangeal angle, intermetatarsal angle, metatarsus adductus angle, distal metatarsal articular angle, anteroposterior talo-first metatarsal angle, and lateral talo-first metatarsal angle were evaluated. The progression rate of HVA was adjusted by multiple factors by using a linear mixed model.

Results: A total of 133 feet were included. The HVA and distal metatarsal articular angle both increased by 0.8° per year ($p < 0.001$ and $p = 0.003$, respectively). HVA increased by 1.5° per year ($p < 0.001$) at under the age of 10, and the HVA progression in the older patients was not statistically significant.

Conclusions: JHV deformity could progress with aging. Most deformity progression could occur before the age of 10 years.

1. Introduction

Juvenile hallux valgus (JHV) deformity is described as varus deviation of the first metatarsal and valgus deviation of the first proximal phalanx in a child who is skeletally immature [1]. Compared with the adult deformity, JHV deformity shows less valgus deformity at the first metatarsophalangeal (MTP) joint, a smaller medial eminence, less bursal thickening, and typically no joint degeneration at the first MTP joint [1].

Although a strong hereditary trait for JHV exist [2], the incidence, etiology, risk factors, and natural history of JHV have not been completely defined [3]. A research on first ray axis pathology is also still required [4]. Although controversies regarding JHV treatment were reported, operation is recommended to be performed after the completion of bone maturation because of high recurrence rate with early surgical treatment [2]. The probability of progression of deformity is assumed on the basis of the remaining skeletal growth, and currently, no study has documented the progression of JHV without treatment [3].

Patients with hallux valgus (HV) can be assessed with weightbearing foot radiographs, which have been widely used for preoperative assessment and postoperative follow-up [5–8]. Radiographic indices on weightbearing foot radiographs showed good to excellent reliability [7]. In addition, radiographic findings showed a good correlation with the clinical appearance of HV [9].

Therefore, this study aimed to estimate the annual change of radiographic indices for JHV and to analyze the factors that influence deformity progression.

2. Materials and methods

2.1. Patients selection

This retrospective study was approved by the institutional review board at our hospital. Informed consent was waived because of the retrospective nature of the study.

We reviewed the medical records of consecutive patients with JHV (age <15 years) who were followed up for at least 1 year and who underwent weightbearing foot radiographs (anteroposterior

* Corresponding author.
E-mail addresses: 89412@snubh.org (K.H. Sung), qrio1010@ajou.ac.kr (S.-S. Kwon), 65407@snubh.org (M.S. Park), oasis100@snubh.org (K.M. Lee), 21344@eumc.ac.kr (J. Ahn), syleemd@ewha.ac.kr (S.Y. Lee).

[1] These authors contributed equally to the writing of this article.

https://doi.org/10.1016/j.fas.2018.02.001
1268-7731/

[AP] and lateral view) more than twice between May 2003 and December 2016. JHV was deemed to be present if the hallux valgus angle (HVA) was ≥15° [10]. Moreover, patients with metatarsus primus varus and who have an intermetatarsal angle (IMA) >9° regardless of HVA were also included because most of the patients with JHV have increased IMA [11], and some authors argued that increased IMA is the primary deformity that could result in a secondary deformity at the first MTP joint [12]. Information obtained from the medical records included patient's age at examination, sex, laterality of foot, and underlying disease. The exclusion criteria were as follows: (1) patients who underwent prior foot surgery that might affect foot appearance, e.g., fracture surgery, (2) patients with neuromuscular disease, including cerebral palsy, and (3) inadequate radiographs available for review. If surgery for JHV was performed during follow-up, only preoperative radiographs were included. Foot radiographs (anteroposterior (AP) and lateral views) were obtained with a UT 2000 X-ray machine (Philips Research, Eindhoven, The Netherlands) at a source-to-image distance of approximately 110 cm; the beam was inclined vertically at 15° and centered on the midtarsal joint with the patient standing barefoot. The radiograph settings were 60 kVp and 10 mAs. All conventional radiographic images were digitally acquired using a picture archiving and communication system (PACS; Infinitt, Seoul, Korea), and measurements were subsequently performed using PACS software.

2.2. Radiographic measurements

As radiographic measurements for HV is reliable and widely used [7,13], eight measurements were selected for assessing JHV. Seven items were measured on AP radiograph: HVA, hallux interphalangeal angle (HIA), IMA, metatarsus adductus angle (MAA), distal metatarsal articular angle (DMAA), AP talo-first metatarsal angle, and AP talo-second metatarsal angle (Fig. 1). Tibial sesamoid position was not evaluated because of the diverse timing of its ossification in the patients. Lateral talo-first metatarsal angle was assessed on lateral radiograph (Fig. 1).

2.3. Reliability test

Reliability testing was conducted before recording the main measurements. Interobserver reliability was determined using intraclass correlation coefficients (ICCs) for three orthopedic surgeons. These three surgeons measured the radiographs independently without knowledge of the other surgeons' measurements. Three weeks after measurement by all three surgeons, one surgeon repeated the radiographic measurements to assess intraobserver reliability.

2.4. Statistical analysis

In this study, reliability was assessed using ICCs and a 2-way random effect model, assuming a single measurement and absolute agreement [14]. Using an ICC target value of 0.8, Bonett's approximation was used setting 0.2 as the width of 95% confidence intervals. The minimal sample size was calculated to be 36 [15]. ICC values >0.8 were considered excellent reliability.

The Kolmogorov–Smirnov test was used to verify the normality of distribution of continuous variables. To summarize patient demographics, descriptive statistics, including mean and standard deviation, were used. Interaction between radiographic indices on the foot is possible; thus, correlations between two radiographic indices were analyzed using Pearson's correlation analysis as a parametric method. The progression rates of HVA, HIA, IMA, MAA, and DMAA were adjusted by multiple factors

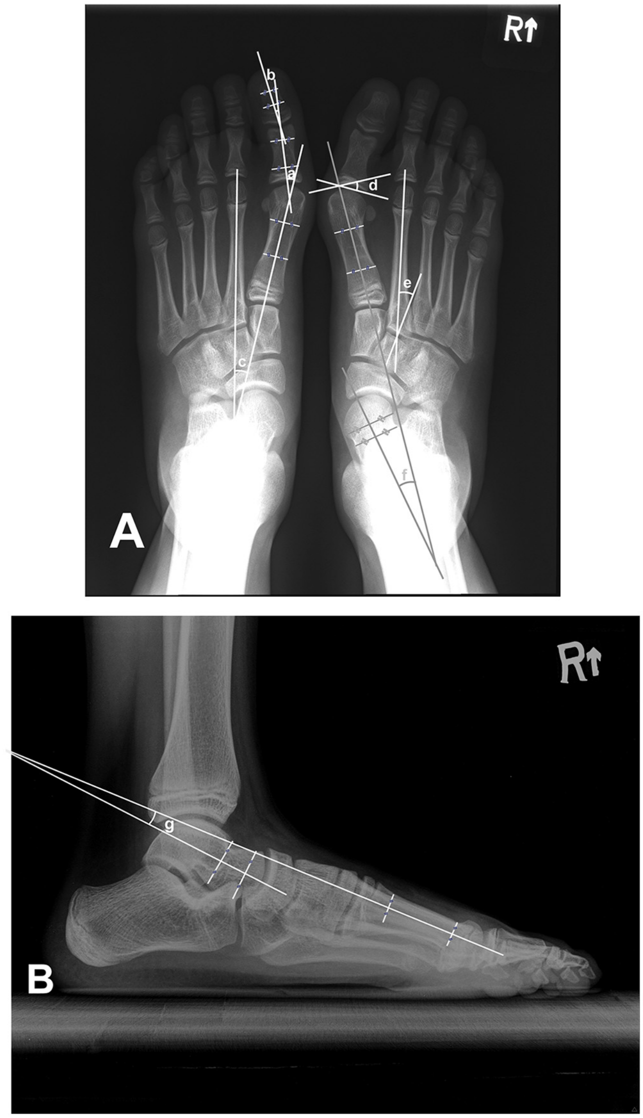

Fig. 1. The illustration shows (A) hallux valgus angle (a), hallux interphalangeal angle (b), intermetatarsal angle (c), distal metatarsal articular angle (d), metatarsal adductus angle (e), and AP talo-first metatarsal angle (f); and (B): lateral talo-first metatarsal angle metatarsal adductus angle (g).

using a linear mixed model (LMM), with gender and radiographic measurements as the fixed effects and laterality and each subject as the random effects. The covariance structure was assumed as the variance components. The restricted maximum likelihood estimation was employed to produce unbiased estimators. By examining the individual pattern of the annual changes in HVA and IMA and evaluating the follow-up duration, an LMM with a random slope and a random intercept was suggested. The slope was the progression rate of HVA and IMA per year. The follow-up duration effect was evaluated to estimate the measurements. The LMMs were compared using the Akaike Information Criterion (AIC) and the Bayesian Information Criterion (BIC). A smaller AIC or BIC value is preferred in terms of model selection. The models had low AIC/BIC scores and, thus, were accepted as valid to estimate HV progression. Data were analyzed using SAS version 9.4 (SAS Institute, Cary, NC). All statistical tests were two-tailed, and p values <0.05 were considered statistically significant. Confidence intervals were considered significant when they did not include zero.

3. Results

A total of 133 feet from 69 patients were included in this study. The mean age during the initial examination was 10.2 ± 4.0 years. Five patients showed unilateral involvement, while 64 patients showed bilateral involvement. The mean follow-up duration was 2.8 ± 2.4 years (range, 1.0–9.6 years). The mean HVA and IMA were 18.4 ± 8.0° and 14.9 ± 6.7°, respectively, at the initial examination (Table 1). Radiographic measurements for JHV showed good to excellent reliability. Intraobserver reliability was highest for the HVA (ICC, 0.930), whereas interobserver reliability was highest for the AP talo-first metatarsal angle (ICC, 0.875). Both intraobserver and interobserver reliabilities were lowest for the metatarsus adductus angle (Table 2). HVA was correlated with HIA (p = 0.003), IMA (p < 0.001), MAA (p = 0.037), DMAA (p < 0.001), and AP talo-first metatarsal angle (p = 0.009) (Table 3).

Considering multiple influencing factors, including radiographic indices and the use of an LMM, we found that, based on the radiographs, HVA progressed as the patients grew older (Fig. 2). Both HVA and DMAA increased by 0.8° per year (p < 0.001 and p = 0.003, respectively). By contrast, HIA decreased by 0.3° per year (p = 0.019). Additionally, progression of the IMA and MAA with aging was not statistically significant. HVA in female patients was greater (7.4°) than HVA in male patients. HIA was greater (5.9°) in male patients than in female patients (Table 4). Increased lateral talo-first metatarsal angle toward flatfoot deformity was associated with increased IMA and MAA (Table 4).

Progression of the radiographic indices between older (≥10 years) and younger (<10 years) patients differs. HVA increased by 1.5° per year (p < 0.001) in younger patients, whereas HVA progression with

Table 1
Patient demographics.

Parameter	Value
No. subjects (male/female)	69 (23/46)
No. of radiographs	594
Laterality (right/left)	68/67
Age at first visit (years)	10.2 ± 4.0
Follow-up duration (years)	2.8 ± 2.4 (1.0–9.6)
No. of follow-up	2 (2–12)
Radiographic parameter	**Value**
Hallux valgus angle	18.4 ± 8.0
Hallux interphalangeal angle	14.9 ± 6.6
Intermetatarsal angle	10.1 ± 3.2
Metatarsus adductus angle	17.6 ± 5.4
Distal metatarsal articular angle	14.4 ± 12.8
AP talo-1st metatarsal angle	8.7 ± 6.9
AP talo-2nd metatarsal angle	12.8 ± 8.3
Lateral talo-1st metatarsal angle	4.6 ± 3.7

Age at first visit, follow-up duration, follow-up interval, and radiographic parameters; mean ± standard deviation (range).
No. of follow-up; mode (range).
Age = decimal years.
AP = anteroposterior.

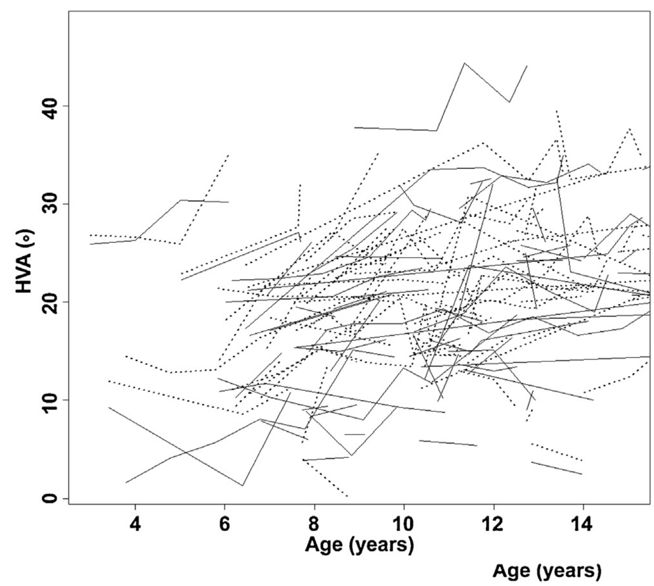

Fig. 2. The reference values of hallux valgus angle are given in the graph. A solid line represents estimation of the progression of hallux valgus angle by a linear age effect.

Table 2
Intra- and interobserver reliabilities of the radiographic measurements.

Measurements	Intraobserver reliability		Interobserver reliability	
	ICC	95% CI	ICC	95% CI
Hallux valgus angle	0.930	0.868–0.964	0.851	0.758–0.915
Hallux valgus interphalangeal angle	0.836	0.702–0.913	0.866	0.781–0.824
Intermetatarsal angle	0.819	0.634–0.909	0.810	0.691–0.892
Metatarsus adductus angle	0.609	0.347–0.781	0.644	0.474–0.783
Distal metatarsal articular angle	0.904	0.822–0.950	0.684	0.479–0.822
AP talo-1st metatarsal angle	0.858	0.739–0.925	0.845	0.749–0.911
Lateral talo-1st metatarsal angle	0.788	0.622–0.886	0.664	0.490–0.799

ICC = intraclass correlation coefficient, CI = confidence interval, AP = anteroposterior.

Table 3
Correlation coefficients among radiographic indices at the time of initial examination.

	HIA	IMA	MAA	DMAA	AP talo-1st MTA	Lat talo-1st MTA
HVA	−0.254 (p = 0.003)	0.410 (p < 0.001)	0.181 (p = 0.037)	0.543 (p < 0.001)	−0.225 (p = 0.009)	0.097 (p = 0.467)
HIA		−0.115 (p = 0.187)	−0.148 (p = 0.089)	−0.195 (p = 0.024)	0.087 (p = 0.321)	−0.017 (p = 0.899)
IMA			0.871 (p = 0.319)	0.153 (p = 0.079)	−0.120 (p = 0.170)	−0.471 (p < 0.001)
MAA				0.102 (p = 0.243)	−0.011 (p = 0.900)	−0.104 (p = 0.432)
DMAA					−0.104 (p = 0.236)	0.076 (p = 0.567)
AP talo-1st MTA						0.196 (p = 0.137)

HVA = hallux valgus angle; HIA = hallux interphalangeal angle; IMA = intermetatarsal angle; MAA = metatarsus adductus angle; DMAA = distal metatarsal articular angle; AP talo-1st (2nd) MTA = anteroposterior talo-first (second) metatarsal angle; Lat talo-1st MTA = lateral talo-first metatarsal angle.

Table 4
The estimation and fixed effects of radiographic indices using a linear mixed model.

	HVA			HIA			IMA			MAA			DMAA		
	Estimate	SE	p-Value	Estimate	SE	p-Value	Estimate	SE	p-Value	Estimate	SE	p-Value	Estimate	SE	p-Value
Age	0.8	0.1	<0.001	−0.3	0.1	0.019	0.0	0.1	0.543	0.0	0.1	0.791	0.8	0.3	0.003
Sex (male)	−7.4	1.7	<0.001	5.9	1.6	<0.001	−0.5	0.7	0.520	1.0	1.4	0.494	−2.6	3.5	0.449
Laterality	−0.398	1.7		−0.481	1.4		−0.3	0.6		0.7	1.2		−3.4	3.0	
AP talo-1st MTA	0.0	0.1	0.970	0.0	0.1	0.736	0.0	0.0	0.883	0.0	0.1	0.564	0.0	0.1	0.773
Lat talo-1st MTA	0.0	0.1	0.971	0.1	0.1	0.314	0.1	0.0	0.018	0.2	0.1	0.042	0.0	0.2	0.829

HVA = hallux valgus angle; HIA = hallux interphalangeal angle; IMA = intermetatarsal angle; MAA = metatarsus adductus angle; DMAA = distal metatarsal articular angle; AP talo-1st MTA = anteroposterior talo-first metatarsal angle; Lat talo-1st MTA = lateral talo-first metatarsal angle.
SE = standard error.

aging in older patients was not statistically significant. Moreover, HVA increased by 0.8° per year ($p < 0.001$) in patients who had both HVA <15° and IMA ≥10° at the initial examination. In patients who had HVA of ≥15° regardless of the IMA at the initial examination, HVA increased by 0.5° per year ($p = 0.001$).

4. Discussion

Information on the natural progression of JHV is lacking. Hence, we conducted this study to estimate the annual change of radiographic indices for JHV and to analyze the factors that influence the deformity progression. Our results showed that the HVA increased by 1.5° per year in patients age <10 years, whereas no significant change in the HVA was noted in patients age ≥10 years.

This study has some limitations. First, this was a retrospective study. As the natural progression of the deformity can be affected by various factors, a longitudinal prospective study, which can control potential influencing factors, is the best method to analyze deformity progression. However, longitudinal studies, e.g., a cohort study, need a long-term follow-up period. Thus, considering the prevalence of JHV and the potential for follow-up loss in longitudinal studies, we believe that analyzing existing data effectively would also be significant. To reduce the limitation of retrospective studies, we used an LMM to analyze the progression rate of radiographic indices for JHV. Second, JHV is a three-dimensional deformity of the first ray. Therefore, two-dimensional radiographic measurements might be insufficient to explain the complex nature of such deformity. Third, other clinical data, such as family history and genetic predisposition, were not included in the study. Although radiographic measurements are considered to be objective and the primary tool for evaluation, future studies considering genetic predisposition will be required. Fourth, this study failed to consider hypermobility of the first metatarsocuneiform joint articulation, which may be associated with the deformity.

Piggott reported that in a series of adult patients evaluated for HV deformity, 57% recalled having a bunion deformity in adolescence and only 5% recalled an occurrence after age 20 [16]. However, information on the progression rate of JHV deformity was lacking. Although a previous study showed that JHV is rarely seen before the age of 10 years [17], the deformity progression was greater in patients <10 years old than in patients ≥10 years old in our study. A study, which revealed that the age of JHV onset appears to be much earlier than that previously reported, supported our results [18]. Therefore, regular follow-ups are important, especially in children who have risk factors (e.g., family history).

JHV can be differentiated from HV deformity in adults by several characteristics. In JHV, IMA is often increased, whereas the HVA is typically lesser [5]. Thus, JHV deformities progressing to adult form of HV could be attributed to increased HVA rather than increased IMA. Moreover, the patients who had both HVA <15° and the IMA ≥10° at the initial examination showed increased HVA with aging in our study. These findings support that metatarsus primus varus is a significant factor in HV development [19]. Therefore, the initial IMA is crucial in JHV deformity progression regardless of increased IMA during childhood or adolescence. Furthermore, our results also showed that MAA progression with aging was not statistically significant. Metatarsus adductus has been cited as a cause of JHV [20]; however, controversies whether the MAA is associated with JHV development exist [18]. Although our study could not demonstrate whether metatarsus adductus is a predisposing factor for JHV, we showed that the MAA is correlated with HVA and IMA.

Most patients with JHV undergo regular X-ray during follow-up and are not treated. Surgeons routinely advise to postpone surgical correction until after skeletal maturity given that earlier surgical treatment has been associated with a high recurrence rate. However, a previous study introduced metatarsal hemiepiphysiodesis, which is a treatment strategy that uses growth potential to achieve correction of the deformity [3]. This surgical treatment seems to be a simple procedure compared with traditional surgical procedures for HV deformity, such as soft-tissue balancing and osteotomy. However, the timing should be carefully determined because, unlike temporary hemiepiphysiodesis that uses plates, screws, or physeal stapling, the procedure is permanent [21]. Although the procedure is less invasive than traditional treatment for HV, treatment strategies that use growth potential are challenging because of a lack of information of the natural history of JHV. Based on our study, we expect that further studies regarding the natural history of JHV can provide new treatment options for JHV.

5. Conclusions

In conclusion, JHV deformity could progress with aging. Our study indicated that increased HVA with aging primarily causes the progression of JHV deformity, most deformity progression could occur before the age of 10 years, and physicians can predict the degree of progression of each radiographic measurement in patients with JHV as the patients grow older. Moreover, our results could also help physicians choose the optimal surgical treatment strategy for JHV before skeletal maturation. Further studies regarding the natural history of JHV that include other factors, such as ethnicity, are also warranted to help physicians identify the best treatment strategy for JHV.

Funding sources

This work was supported by the new faculty research fund of Ajou University (S-2015-G0001-00092).

Conflict of interest

The authors declare that they have no conflict of interest.

References

[1] Coughlin MJ, Mann RA. The pathophysiology of the juvenile bunion. Instr Course Lect 1987;36:123–36.
[2] Groiso JA. Juvenile hallux valgus. A conservative approach to treatment. J Bone Joint Surg Am 1992;74(9):1367–74.
[3] Davids JR, McBrayer D, Blackhurst DW. Juvenile hallux valgus deformity: surgical management by lateral hemiepiphyseodesis of the great toe metatarsal. J Pediatr Orthop 2007;27(7):826–30.
[4] McCluney JG, Tinley P. Radiographic measurements of patients with juvenile hallux valgus compared with age-matched controls: a cohort investigation. J Foot Ankle Surg 2006;45(3):161–7.
[5] Coughlin M, Anderson R. Hallux valgus. In: Coughlin M, Saltzman C, Anderson R, editors. Mann's surgery of the foot and ankle. 9th ed. Philadelphia: Elsevier; 2014. p. 177–93.
[6] Coughlin MJ, Saltzman CL, Nunley 2nd JA. Angular measurements in the evaluation of hallux valgus deformities: a report of the ad hoc committee of the American Orthopaedic Foot & Ankle Society on angular measurements. Foot Ankle Int 2002;23(1):68–74.
[7] Lee KM, Ahn S, Chung CY, Sung KH, Park MS. Reliability and relationship of radiographic measurements in hallux valgus. Clin Orthop Relat Res 2012;470(9):2613–21.
[8] Oh IS, Choi SW, Kim MK, Lee SY, Lee JS. Clinical and radiological results after modified distal metatarsal osteotomy for hallux valgus. Foot Ankle Int 2008;29(5):473–7.
[9] D'Arcangelo PR, Landorf KB, Munteanu SE, Zammit GV, Menz HB. Radiographic correlates of hallux valgus severity in older people. J Foot Ankle Res 2010;3:20.
[10] Hardy RH, Clapham JC. Observations on hallux valgus; based on a controlled series. J Bone Joint Surg Br 1951;33-B(3):376–91.
[11] Vyas S, Conduah A, Vyas N, Otsuka NY. The role of the first metarsocuneiform joint in juvenile hallux valgus. J Pediatr Orthop B 2010;19(5):399–402.
[12] DuVries H. Surgery of the foot. 2nd ed. St Louis: Mosby; 1965.
[13] Lee KM, Chung CY, Park MS, Lee SH, Cho JH, Choi IH. Reliability and validity of radiographic measurements in hindfoot varus and valgus. J Bone Joint Surg Am 2010;92(13):2319–27.
[14] Shrout PE, Fleiss JL. Intraclass correlations: uses in assessing rater reliability. Psychol Bull 1979;86(2):420–8.
[15] Bonett DG. Sample size requirements for estimating intraclass correlations with desired precision. Stat Med 2002;21(9):1331–5.
[16] Piggott H. The natural history of hallux valgus in adolescence and early adult life. J Bone Joint Surg Br 1960;42B(4):749–60.
[17] Scranton Jr. PE. Adolescent bunions: diagnosis and management. Pediatr Ann 1982;11(6):518–20.
[18] Coughlin MJ, Roger A. Mann award. Juvenile hallux valgus: etiology and treatment. Foot Ankle Int 1995;16(11):682–97.
[19] Kilmartin TE, Barrington RL, Wallace WA. Metatarsus primus varus. A statistical study. J Bone Joint Surg Br 1991;73(6):937–40.
[20] Ferrari J, Malone-Lee J. A radiographic study of the relationship between metatarsus adductus and hallux valgus. J Foot Ankle Surg 2003;42(1):9–14.
[21] Jelinek EM, Bittersohl B, Martiny F, Scharfstadt A, Krauspe R, Westhoff B. The 8-plate versus physeal stapling for temporary hemiepiphyseodesis correcting genu valgum and genu varum: a retrospective analysis of thirty five patients. Int Orthop 2012;36(3):599–605.