


Joint hypermobility and preschool-age flexible flatfoot

Chia-Chun Tsai, PhD^a, Yu-Chia Chih, PhD^b, Chia-Lung Shih, PhD^c, Shu-Jung Chen MD^{d,e}, Po-Chih Shen, MD, PhD^{d,e}, Yin-Chun Tien, MD, PhD^{d,e} 

Abstract

Flexible flatfoot is the most common condition seen in pediatric orthopedic practice and generalized joint hypermobility is widely regarded as one of the predisposing factors. However, in previous studies, the flatfoot was defined by observers' subjective evaluation of the eversion of the bare foot in the standing position; and the joint hypermobility was defined by the Beighton score. The objective of this study is to evaluate the correlation between preschool-age flexible flatfoot and joint hypermobility in preschool-age children objectively.

Footprints were measured on a Harris and Beath footprint mat. Flatfoot flexibility was assessed by Staheli Plantar Arch Index (PAI). Other than the Beighton score, 2 new measurement methods, the thumb-to-forearm test and the thumb-thrust test were developed to evaluate joint hypermobility.

Of the 291 preschool children from 4 different kindergarten schools included in this study, 156 were boys and 135 were girls. The mean age was 64.18 ± 9.33 months (range 35–88 months). Pearson correlation analysis demonstrated PAI was not associated with the Beighton score ($R = 0.020$, $P = .735$), thumb-to-forearm grade ($R = 0.109$, $P = .066$), and thumb-thrust grade ($R = 0.027$, $P = .642$). Two-sample t-test results showed that the normal and flatfoot groups did not differ significantly in the Beighton score ($P = .404$), thumb-to-forearm grade ($P = .063$), and thumb-thrust grade ($P = .449$).

The results demonstrated no correlation between joint hypermobility and preschool-age flexible flatfoot when flatfoot was defined with Staheli PAI and joint hypermobility with the Beighton score. Even with 2 new methods, the thumb-to-forearm test and thumb-thrust test, to define joint hypermobility, we still found no correlation between preschool-age flexible flatfoot and joint hypermobility.

Abbreviations: HOF = hands on floor, PAI = plantar arch index.

Keywords: flexible flatfoot, joint hypermobility, orthopedics, pediatrics, Staheli plantar arch index

1. Introduction

Flexible flatfoot, which manifests as a lowered medial longitudinal arch with or without rearfoot eversion,^[1,2] is the most common condition seen in pediatric orthopedic practice.^[3] Generalized joint hypermobility is widely regarded as one of the predisposing factors of pediatric flexible flatfoot. According to previous studies, the Beighton score is related to joint laxity and hypermobility. Lin et al^[4] found that the joint laxity score of preschool children (36.7 ± 6.0 , $P = .0001$) was related to flatfoot. The results showed that age, height, weight, foot progression angle, occurrence of knock-knee, and joint laxity score were correlated with flexible flatfoot. In El et al's study^[2] of 579 primary school children (280 girls and 299 boys) with an age range of 6 to 12 years, the Beighton hypermobility score (4.09 ± 2.64) was also related to flatfoot ($P = .001$). However,

in this study, a score of >5 was defined as indicating joint hypermobility instead of the original cutoff point of >4 . Some researchers have claimed that a cutoff of >4 for the Beighton score is too low to define joint hypermobility and is not appropriate for children.

In addition, the inclusion of the “hands on floor (HOF)” maneuver in the Beighton score has been questioned. Corten et al^[5] found that the HOF maneuver does not add additional value to the Beighton score of children of Black African and mixed ancestry. They tested 460 children (median age 8.58 years [interquartile range, 7.33–9.50]), of whom 34.57% were hypermobile. However, only 8.91% of all children had a positive score for HOF. Questions included in several screening documents for the Beighton score to assess joint hypermobility are not necessarily accurate. Therefore, in this study, we created 2 new scoring methods—the thumb-to-forearm test and thumb-thrust test—to

The authors have no funding and conflicts of interest to disclose.

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

KMUH/IRB/AF/08E-03/000 (2017/10/17-2020-07-31). Assessment and Influential Factors Analysis of Motor Development for Preschool Age Children.

^a Department of Nursing, Fooyin University, Kaohsiung City, Taiwan, ^b Department of Medical Laboratory Science and Biotechnology, Fooyin University, Kaohsiung City, Taiwan, ^c Department of Orthopedics, Ditmanson Medical, Foundation Chia-Yi Christian Hospital, Chiayi City, Taiwan, ^d Department of Orthopedics, Kaohsiung Medical University Hospital, Kaohsiung City, Taiwan, ^e College of Medicine, Kaohsiung Medical University, Kaohsiung City, Taiwan.

*Correspondence: Yin-Chun Tien, PhD, 100, Shih-Chun 1st Road, San-Ming District, Kaohsiung City 80708, Taiwan (e-mail: d740113@kmu.edu.tw).

Copyright © 2022 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and buildup the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

How to cite this article: Tsai C-C, Chih Y-C, Shih C-L, Chen S-J, Shen P-C, Tien Y-C. Joint hypermobility and preschool-age flexible flatfoot. *Medicine* 2022;101:31(e29608).

Received: 1 December 2021 / Received in final form: 18 March 2022 / Accepted: 5 May 2022

<http://dx.doi.org/10.1097/MD.0000000000029608>

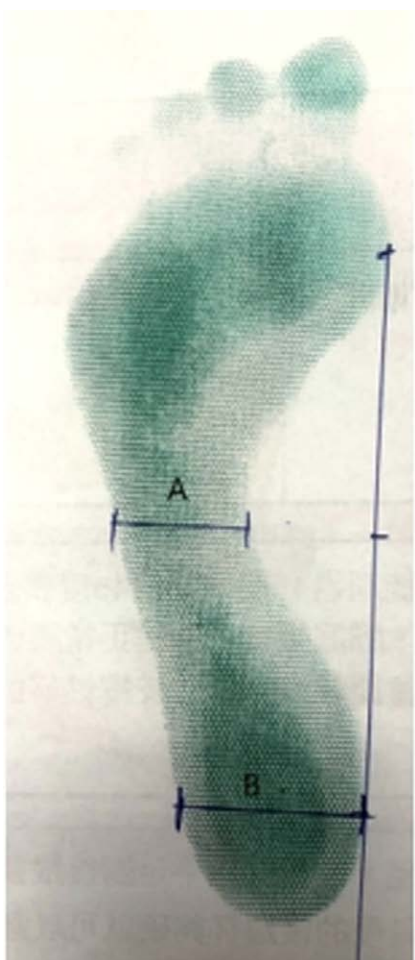


Figure 1. Calculation of the Staheli plantar arch index.

evaluate joint laxity. These tests classify joint laxity into 4 grades: normal, mild, moderate, and severe. The Beighton score is the sum of responses to dichotomous questions, with yes or no answers, on 5 evaluation items, but the new methods score joint laxity based on the 4 grades of severity of a single indicator.

Flatfoot is diagnosed through a variety of measures, including plain film radiographs (e.g., X-ray), static foot posture measures, and footprint analysis (3). Of these, footprint analysis is more widely accepted. However, Lin et al^[4] and El et al^[2] diagnosed flatfoot by observers' subjective evaluation of the eversion of the bare foot in the standing position and determination of the longitudinal arch in the dynamic (weight-bearing) position. In this method, feet are grouped by the appearance of the longitudinal arch on weight bearing. The foot is graded as normal if the medial arch looks normal. If the arch is only slightly impressed but still visible, the foot is graded as mild flexible flatfoot. In moderate flexible flatfoot, the longitudinal arch is not visible in stance. If the medial border of the foot is convex, with the head of the talus presenting on the plantar aspect of the foot immediately below and anterior to the medial malleolus, the foot is graded as severe flexible flatfoot.^[6] This method defines severity based on observation of the appearance of the longitudinal arch in children, and is not as objective as the footprint method.

We hypothesized that the more detailed grading used in this study would improve the sensitivity of hypermobility diagnosis and the objectivity of footprint measures to define flatfoot. The aim of this study was to assess whether these new gradings of joint laxity were correlated with flexible flatfoot in preschool children.

2. Materials and Methods

Two hundred ninety-one children from 4 different kindergarten schools were enrolled in this study. Written consent was obtained from the children's parents. Demographic data, such as age, sex, weight, and height, were recorded. Each child's right and left footprints were obtained. The footprint was recorded during a sit-to-stand movement. Each child was instructed to

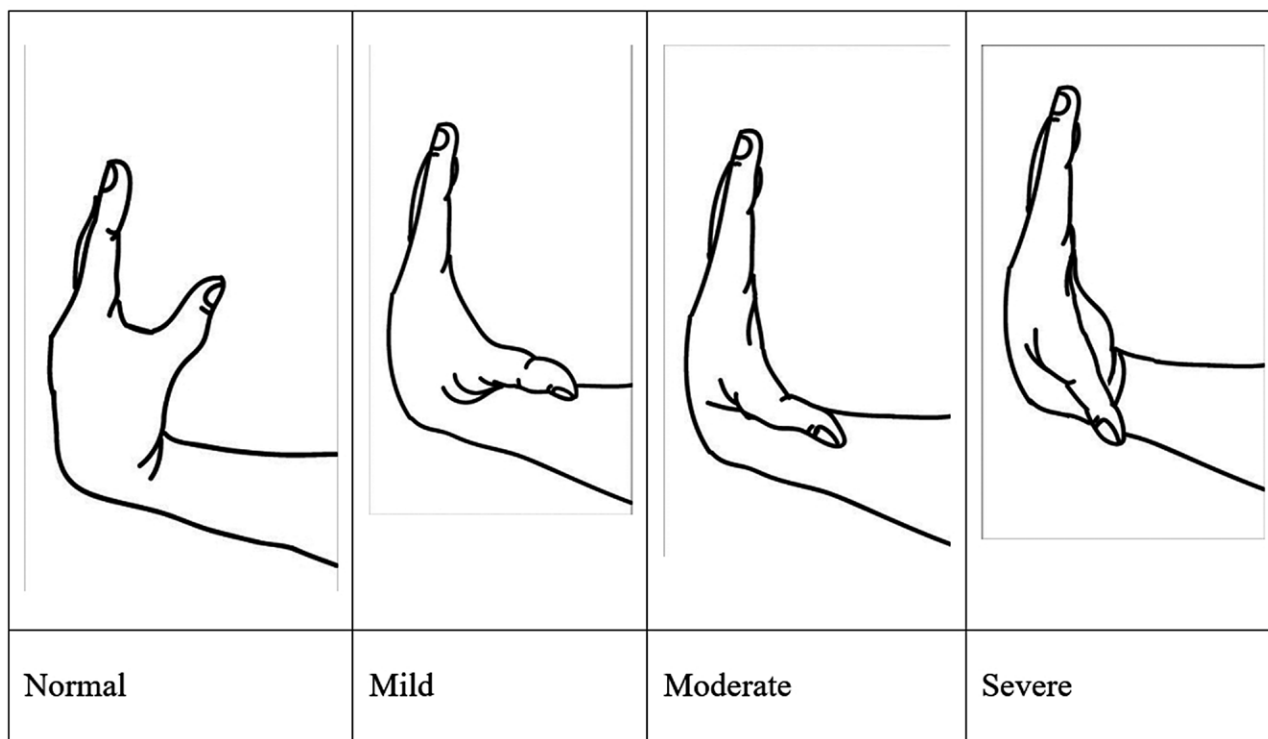


Figure 2. The thumb-to-forearm test for joint laxity.

place both feet on 2 Harris and Beath footprint mats while sitting on a chair. The child then stood up with even weight on both feet and returned to the sitting position to complete the footprint recording. The width of the arch and the width of the heel were measured to calculate the plantar arch index (PAI) for each foot, as described by Staheli et al^[3]: $PAI = A/B$, where A was the width of the central region of the foot and B was the width of the heel region in millimeters (Fig. 1).

Banwell et al's^[7] review suggested a PAI of > 1.07 for children aged 3 to 6 years and ≥ 1.28 for children aged 6 to 9 years. Chang^[8] suggested a PAI of ≥ 1.0 for children aged 6 to 9 years.^[9] In this study, a cutoff PAI of > 1.07 was used to define flexible flatfoot. Children were divided into 2 groups based on their results from the dynamic longitudinal arch evaluation. Children with a PAI of < 1.07 were in the normal group and those with a PAI of ≥ 1.07 were in the flatfoot group.

Joint laxity was evaluated by the following 3 methods. First, the *Beighton score* was based on examination and evaluation of 5 specific functions: passive extension above 90° of the fifth finger of the hand; the ability to passively adduct the thumb to the inner surface of the forearm; elbow joint hyperextension above 10°; hyperextension of the knee joint above 10°; and the ability to place palms on the ground while keeping legs straight.^[10] These 5 specific functions were assessed for both thumbs, both little fingers, both elbows, both knees, and the trunk, and used a score of 1 for “yes” or 0 for “no.”^[7] Joint hypermobility was diagnosed if the score was ≥ 4 points out of the possible 9. Second, the *thumb-to-forearm test* evaluated the extent of the flexion of the right thumb toward the volar surface of the forearm; 4 grades were defined according to the reach of the thumb tip. Laxity was “normal” (score = 0 points) if the tip could not touch the volar surface of the forearm. “Mild” laxity (score = 1 point) was defined as the thumb tip just touching the volar surface. Laxity was “moderate” (score = 2 points) if the tip reached between the volar and dorsal surface of the forearm, and “severe” (score = 3 points) if the tip reached beyond the dorsal surface (Fig. 2).

Third, the *thumb-thrust test* assessed the flexion of the right thumb toward the ulnar border of the palm; 4 grades were defined depending on the reach of the thumb. Laxity was “normal” (score = 0 point) if the tip could not reach the ulnar border of the palm. Laxity was “mild” (score = 1 point) if the tip just

touched the ulnar border, “moderate” (score = 2 points) if the thumb tip reached beyond ulnar border of palm, and “severe” (score = 3 points) if the inter phalanges flexion crease reached beyond the border (Fig. 3).

2.1. Statistical analysis

All statistical analyses were conducted using IBM SPSS Statistics, version 19 (IBM Corp., Armonk, NY). The relationships between PAI and age, sex, height, weight, the Beighton score, thumb-to-forearm grade, and thumb-thrust grade were assessed using Pearson correlation. The association between incidence of flexible flatfoot and age, sex, height, weight, Beighton score, thumb-to-forearm grade, or thumb-thrust grade was assessed using logistical regression. The differences in age, height, and weight between normal (PAI < 1.07) and flatfoot (PAI ≥ 1.07) groups were assessed using the 2-sample *t*-test, and the differences in the Beighton score and thumb-to-forearm and thumb-thrust grades between the 2 groups were assessed using the Mann–Whitney *U* test.

3. Results

Of the 291 preschool children from 4 different kindergarten schools included in this study, 156 were boys and 135 were girls. The mean age was 64.18 ± 9.33 months (range 35–88 months).

Pearson correlation analysis demonstrated a significant negative correlation between PAI and age ($R = -0.194, P = .001$) and PAI and weight ($R = -0.144, P = .020$). However, PAI was not associated with the Beighton score ($R = 0.020, P = .735$), thumb-to-forearm grade ($R = 0.109, P = .066$), and thumb-thrust grade ($R = 0.027, P = .642$; Table 1).

The associations between the incidence of flatfoot and gender, age, weight, height, Beighton score, thumb-to-forearm grade, and thumb-thrust grade are shown in Table 2. The results demonstrated a significant positive correlation between the incidence of flatfoot and gender (odds ratio [OR] = 2.091, $P = .004$) and a significant negative correlation between the incidence of flatfoot and age (OR = 0.952, $P = .001$), height (OR = 0.967, $P = .029$), and weight (OR = 0.922, $P = .031$).

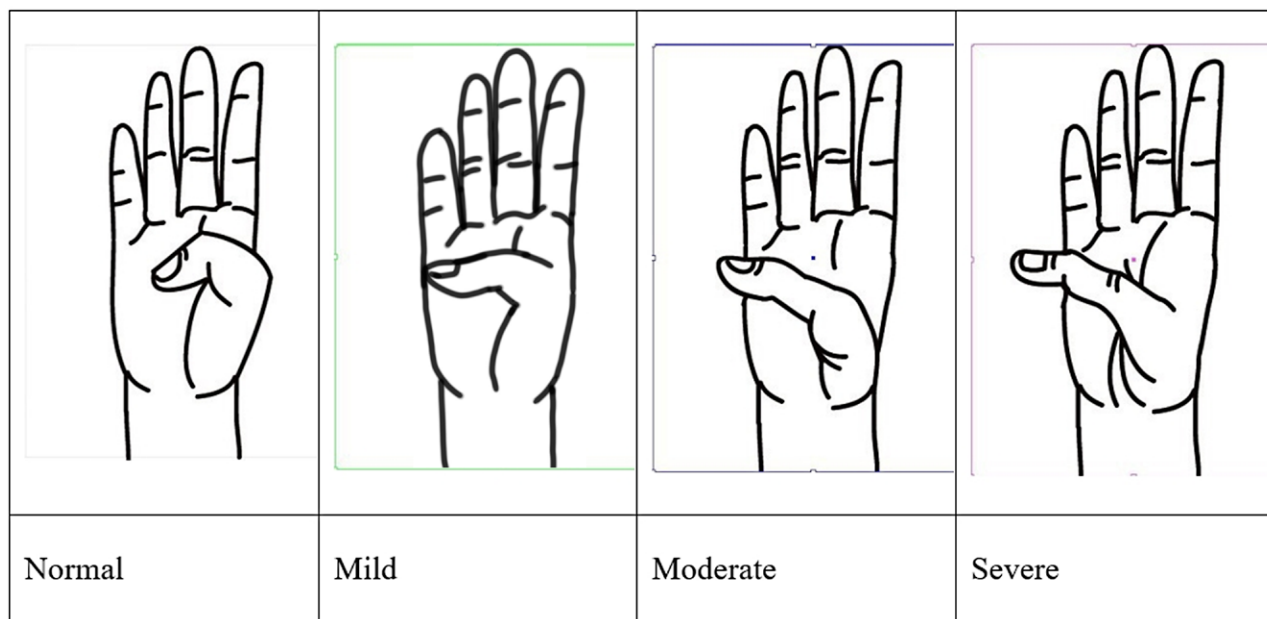


Figure 3. The thumb-thrust test for joint laxity.

Table 1

Pearson correlation between PAI and age, weight, height, the Beighton score, thumb-to-forearm grade, and thumb-thrust grade.

| Variable | R | P value |
|------------------------|--------|-------------|
| Age | -0.194 | .001 |
| Weight | -0.144 | .020 |
| Height | -0.089 | .162 |
| Beighton score | 0.020 | .735 |
| Thumb-to-forearm grade | 0.109 | .066 |
| Thumb-thrust grade | 0.027 | .642 |

Significant differences are indicated in bold format. R = Pearson correlation coefficient.

Table 2

Association between the incidence of flatfoot and variables.

| Variable | OR | 95% CI | DF | P value |
|------------------------|-------|-------------|----|---------|
| Male (ref: Female) | 2.091 | 1.274–3.433 | 1 | .004 |
| Age | 0.952 | 0.927–0.979 | 1 | <.001 |
| Height | 0.967 | 0.939–0.997 | 1 | .029 |
| Weight | 0.922 | 0.856–0.992 | 1 | .031 |
| Beighton score | 1.049 | 0.925–1.190 | 1 | .454 |
| Thumb-to-forearm grade | 1.300 | 0.966–1.750 | 1 | .083 |
| Thumb-thrust grade | 1.122 | 0.812–1.551 | 1 | .485 |

CI = confidence interval, DF = degrees of freedom, OR = odds ratio.

Males were 2.09 times (95% confidence interval [CI] = 1.274–3.433) more likely to have flatfoot than females. The incidence of flatfoot was negatively correlated with age (OR = 0.952, 95% CI = 0.927–0.979), height (OR = 0.967, 95% CI = 0.939–0.997), and weight (OR = 0.922, 95% CI = 0.856–0.922).

We found that there was no association between the incidence of flatfoot and the Beighton score ($P = .454$), thumb-to-forearm grade ($P = .083$), and thumb-thrust grade ($P = .485$).

Two-sample t -tests showed that the normal and flatfoot groups did not differ significantly in the Beighton score ($P = .404$), thumb-to-forearm grade ($P = .063$), and thumb-thrust grade ($P = .449$; Table 3).

4. Discussion

Although previous studies have shown that there is a positive correlation between joint hypermobility and the incidence of flatfoot, our results did not find a correlation between the Beighton score and incidence of flexible flatfoot. However, our study differed from the previous 2 studies (i.e., those of Lin et al^[4] and El et al^[2]) in the methodology and criteria for judging flatfoot. In the previous studies, flatfoot was assessed by observing the degree of collapse of the longitudinal arch of the foot

Table 3

Differences between normal and flatfoot groups.

| Variable | Normal group (mean ± SD) | Flatfoot group (mean ± SD) | P value |
|------------------------|--------------------------|----------------------------|---------|
| Age (mo) | 65.63 ± 8.86 | 61.55 ± 9.64 | <.001* |
| Height (cm) | 111.45 ± 11.52 | 107.40 ± 12.91 | .011* |
| Weight (kg) | 20.10 ± 7.63 | 18.48 ± 3.57 | .054* |
| Beighton score | 5.153 ± 1.87 | 5.33 ± 2.026 | .404† |
| Thumb-to-forearm grade | 1.12 ± 0.82 | 1.30 ± 0.858 | .063† |
| Thumb-thrust grade | 1.11 ± 0.724 | 1.17 ± 0.785 | .449† |

*Two-sample t -test.

†Mann–Whitney U test.

when the child was standing barefoot. In our study, footprints were measured on a Harris and Beath footprint mat and the PAI was calculated using the Staheli method; a cutoff PAI of > 1.07 was used to define flexible flatfoot. In general, using the footprint for flatfoot assessment is more objective, and nowadays, this method is widely recognized and used.

There have been many controversies over the examination and evaluation of joint hypermobility. For calculating the Beighton score, 5 specific functions are assessed using dichotomous responses (yes or no scores), and the total score obtained is the criterion for evaluation. Because each maneuver is not graded along a severity scale, the degree of discrimination may be weak. Others, ligaments exist viscoelastic characteristics. Performance may be greatly influenced by temperature; therefore, the Beighton score for the same child could vary depending on whether or not the test was preceded by a warm-up exercise.

We used 2 different measurement methods, the thumb-to-forearm test and the thumb-thrust test, developed for this study to evaluate joint laxity. These tests classified the severity of joint laxity into 4 grades: normal, mild, moderate, and severe. We expected that the correlation between joint hypermobility and flatfoot may be more hierarchical. However, the statistical analysis, including Pearson correlation tests, logistical regression analysis, and 2-sample t -tests, demonstrated no significant association. These results suggest that joint hypermobility is not a key factor in flatfoot in preschool-age children.

Foot bones are supported by static connective tissue, which includes ligaments, tendons, and capsular structures, to form the normal medial longitudinal arch of the foot. Electromyographic studies have shown that neither intrinsic nor extrinsic muscles support and maintain the longitudinal arches in standing posture.^[11] However, during walking and other activities, both muscle groups maintain dynamic stabilization of the arch. In a study reinforcing this argument, Fiolkowski discovered the importance of the intrinsic muscles of the foot in providing support for the medial longitudinal arch.^[12] Flatfoot due to posterior tibial tendon insufficiency, which has been studied extensively, suggests the importance of this musculature. In flexible flatfoot, the medial longitudinal arch of the foot collapses at different degrees during weight-bearing. However, the foot arch appears again while the body is raised on tiptoe (Jack tiptoe test).^[13] Therefore, we believe that improved power of these muscles is crucial for the development of the medial foot arch. We propose that exercise programs for strengthening the posterior tibial muscle of 3- to 6-year-old children with flexible flatfoot would benefit foot arch development. On the other hand, a study that compared the prevalence of flatfoot in children with delayed motor development and normal children showed that the former's risk of developing flatfoot was 1.5 times that of the latter (OR = 1.511, $P = .005$); this further suggests that the motor development of muscles may play a critical role in the development of the foot arch.^[14]

The limitation of this study was that the sampling did not represent nationwide random samples, and relied exclusively on kindergartens located in an urban area of a single city. We are planning to perform an additional study which recruits samples from rural areas for further comparison.

5. Conclusion

We found no correlation between joint hypermobility and preschool-age flexible flatfoot when flatfoot was defined with Staheli PAI and joint hypermobility with the Beighton score. Even with 2 new methods, the thumb-to-forearm test and thumb-thrust test, to define joint hypermobility, we still found no correlation between preschool-age flexible flatfoot and joint hypermobility.

Author contributions

Chia-Chun Tsai: provision of data collection and manuscript writing. Yu-Chia Chih: provision of footprint recording. Chia-Lung Shih: provision of statistical analysis. Shu-Jung Chen: provision of PAI measurement. Po-Chih Shen: provision of PAI measurement. Yin-Chun Tien: provision of study design.

References

- [1] Evans AM. The paediatric flat foot and general anthropometry in 140 Australian school children aged 7 - 10 years. *J Foot Ankle Res.* 2011;4:12.
- [2] El O, Akcali O, Kosay C, et al. Flexible flatfoot and related factors in primary school children: a report of a screening study. *Rheumatol Int.* 2006;26:1050-3.
- [3] Staheli LT, Chew DE, Corbett M. The longitudinal arch. A survey of eight hundred and eighty-two feet in normal children and adults. *J Bone Joint Surg Am.* 1987;69:426-8.
- [4] Lin CJ, Lai KA, Kuan TS, et al. Correlating factors and clinical significance of flexible flatfoot in preschool children. *J Pediatr Orthop.* 2001;21:378-82.
- [5] Corten L, Ferguson G, Smits-Engelsman B. Does the item “hands on floor” add value to the Beighton score in identifying joint hypermobility? *Eur J Rheumatol.* 2020;7:79-83.
- [6] Barry RJ, Scranton PE Jr. Flat feet in children. *Clin Orthop Relat Res.* 1983;181:68-75.
- [7] Banwell HA, Paris ME, Mackintosh S, et al. Paediatric flexible flatfoot: how are we measuring it and are we getting it right? A systematic review. *J Foot Ankle Res.* 2018;11:21.
- [8] Chang CH, Chen YC, Yang WT, et al. Flatfoot diagnosis by a unique bimodal distribution of footprint index in children. *PLoS One.* 2014;9:e115808.
- [9] Chen KC, Yeh CJ, Tung LC, et al. Relevant factors influencing flatfoot in preschool-aged children. *Eur J Pediatr.* 2011;170:931-6.
- [10] Beighton P, Solomon L, Soskolne CL. Articular mobility in an African population. *Ann Rheum Dis.* 1973;32:413-8.
- [11] Basmajian JV, Stecko G. The role of muscles in arch support of the foot. *J Bone Joint Surg Am.* 1963;45:1184-90.
- [12] Cowan DN, Robinson JR, Jones BH, et al. Consistency of visual assessments of arch height among clinicians. *Foot Ankle Int.* 1994;15:213-7.
- [13] Atik A, Ozyurek S. Flexible flatfoot. *North Clin Istanbul.* 2014;1:57-64.
- [14] Chen KC, Tung LC, Tung CH, et al. An investigation of the factors affecting flatfoot in children with delayed motor development. *Res Dev Disabil.* 2014;35:639-45.