

Development, validation, and psychometric analysis of Foot and Ankle Flexibility Index (FAFI)[☆]

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ABSTRACT

Aim: To develop a new tool for identifying joint hypermobility of the paediatric foot and ankle, based on a dichotomous scoring system utilising the Lower Limb Assessment Score (LLAS), to separate the foot and ankle items.

Material and methods: A total of 205 children, aged between 5 and 10 years, participated in a cross-sectional study. The new tool Foot and Ankle Flexibility Index (FAFI) was predicated upon the last 7 items of LLAS, which are specific to the foot and ankle. The internal consistency was measured with Cronbach's test. Kappa statistics with 95% CI were calculated to verify the level of inter-rater and intra-rater agreement for the FAFI. **Results:** Cronbach's alpha returned 0.82. The correlations between items returned a mean of 0.59 (range: 0.43–0.74). The discrimination score on the ROC curve (4 points) showed that the model can be used to identify children with joint hypermobility of the foot and ankle. Inter-rater reliability was largely good (ICC = 0.89). Excellent intra-rater reliability was found (ICC = 0.96)

Conclusions: This study identified high reliability between evaluators, and high sensitivity and specificity, for a new reliable and valid tool for the identification of foot and ankle joint hypermobility.

1. Introduction

Joint hypermobility (JH) is considered to exist when the joint exceeds the range of normal limits within its physiological span, due to increased elasticity of the capsular and ligamentous tissues [1]. The physiologic joint range is dependent on age, sex, and ethnicity. JH has been shown to be inversely proportional to age; children are more flexible than adolescents, and adolescents are more flexible than adults [2]. The female sex is considered more hypermobile with a 2:1 or 3:1 ratio compared to the male sex [3]. Regarding ethnicity, it has been shown that Asian and African populations have more hypermobility than Caucasian populations [4,5].

This increase in the range of motion can be localized, affecting a particular joint, or it can affect multiple areas of the body. The term generalized joint hypermobility (GJH) is used when 5 or more joints are

involved [6].

The identification of GJH through the Beighton score has great international acceptance. The Beighton score consists of 5 tests with a predominance in the upper limb, and only one lower limb test, for knee extension [7]. The cut-off point in children is considered to be from 6/9 up to 4/9, it depends on the sex and age of the children adults [8]. The accuracy of the Beighton score to detect GJH is debated, given that the scoring focuses on the joints of the upper limb and does not encompass many major joints of the body, making direct identification of GJH difficult. The absence of assessment of the hip, ankle, and foot limits the Beighton score for assessment of joint hypermobility in the lower limbs.

Ferrari et al. [9] validated a lower extremity laxity test, the Lower Limb Assessment Score (LLAS). The LLAS provides a dichotomous scoring system where lower extremity hypermobility is assessed in detail across ranges of motion of the hip, knee, ankle, hindfoot, midfoot, and

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forefoot. It has a validated cut-off score of $\geq 7/12$ to detect lower limb hypermobility in children [9].

Ligament and capsule injuries such as strains, sprains, and subluxations are frequent in patients with GJH [10]. The contradiction of studies that analyse these lower limb injuries using Beighton score has directed our consideration of a more specific test to evaluate the paediatric foot and ankle [11]. The LLAS is a valid tool for identifying joint hypermobility of the lower limb, but as with the BS, it raises doubts as to whether there is a predominance of hypermobility in some joints more than in others, ie proximal versus distal.

Therefore, the objective of this study was to separate the LLAS foot and ankle items and obtain a new identification tool, through a short-form of the LLAS, specifically for joint hypermobility of the foot and ankle, using a dichotomous scoring system. In addition to obtaining psychometric values, the cut-off point, and validation of this new tool were sought.

2. Material and methods

2.1. Ethical approval

This study was carried out in full accordance with the provisions of the Declaration of Helsinki regarding ethical principles for medical research involving human subjects and was approved by the Ethics Committee. It was approved by the Ethics Committee of the San Antonio de Murcia Catholic University CE112104.

2.2. Participants

A total of 205 children, aged between 5 and 10 years, participated in this cross-sectional study. The measurements were made in 2022, between January and June. The participants were evaluated at the San Francisco de Asís school in Lorca, Murcia (Spain).

All participants met the following inclusion criteria.

- age between 5 and 10 years
- not experiencing any foot pain at the time of assessment.

The exclusion criteria were.

- congenital structural alterations of the ankle joint
- cerebral palsy
- surgical treatments in the foot or lower extremity
- genetic, neurological, inflammatory, or muscular conditions.

Informed consent was obtained from the parents/guardians, after explanation of the study.

2.3. Procedure

Before assessing the participating children, demographic data regarding, sex, age, weight, and height were collected. The children wore comfortable sports clothes, shorts, and no shoes. The children were instructed to be as relaxed as possible, and all joint range manoeuvres were performed without pain. To assess data reliability, two podiatrists with 1 year experience in a screening gait program for children were engaged as examiners. All participants attended two test sessions, the first, where both the LLAS [9]. and Beighton score [7] were evaluated independently and blind by the two examiners. At the second session, a further expert in paediatric podiatry with more than 15 years of clinical experience and researcher at university in the field of paediatric gait, measured the hypermobility of ankle and foot.

The two examiners carried out the tests with specified cut-off levels.

- Beighton score for males 5–7 years $\geq 5/9$ and 8–10 years $\geq 4/9$;
- Beighton score for females 5–7 years $\geq 5/9$ and 8–10 years $\geq 4/9$;

- LLAS $\geq 7/12$ for all.

The new Foot and Ankle Flexibility Index (FAFI) consisted of the last seven items of LLAS, which address the foot and ankle. Fig. 1 illustrates the test version, with item definition. The experienced clinical expert assessed 196 children on a separate day. In the absence of a 'gold standard' for lower limb hypermobility, LLAS was validated via expert opinion. This clinical judgment was based on clinical tests of foot and ankle range of motion, without using the LLAS. In addition, 25 children were evaluated to obtain intra-rater reliability, with both examiners assessing independently, and same day, for inter-rater reliability.

2.4. Statistical analysis

All statistical analyses were performed using SPSS version 29 [IBM SPSS Statistics SPSS Inc, 2022]. An exploratory data analysis was performed that included descriptive statistics for age, sex, and BMI. The internal consistency was measured with Cronbach's test (7 items) and considered satisfactory from 0.7 [12] Pearson's correlation was used for inter-item and item-total correlations.

Discriminant validity (i.e., cut-off score for the FAFI) was calculated using the receiver operating characteristic curve (ROC) [13], offering us the sensitivity and specificity for each level and the area under the curve (AUC). The cut-off point represents the strongest discrimination between children with and without joint hypermobility of the foot and ankle. The clinical opinion of an expert was used as the 'gold standard' for the analysis. Once the cut-off point was obtained, the positive and negative predictive values were evaluated. Kappa statistics with 95% CI were calculated to verify the level of inter-rater and intra-rater agreement for the FAFI [14]. Intraclass correlation statistics were used to assess reliability. According to Landis and Koch [15], ICC coefficients less than 0.20 indicated slight agreement, 0.20–0.40 indicated fair reliability, 0.41–0.60 indicated moderate reliability, 0.61–0.80 indicated substantial reliability, and 0.81–1.00 indicated near perfect reliability.

Pearson's correlation was used to test the relationship between age, and ANOVA of one factor for sex. A paired samples *t*-test was used to compare the FAFI scores between left and right sides. Finally, a percentage agreement between FAFI, BS, and LLAS was evaluated to compare the convergent validity of the FAFI in the identification of foot and ankle joint hypermobility. A Pearson correlation was performed between FAFI and the LLAS, and the BS. P-values less than 0.05 were considered statistically significant.

3. Results

3.1. Participants

A total of 196 children were included in the final analysis, as 9 children were excluded for pain during testing. The average age of the participants was 7.6 (5–10) years; 60.2% were girls and 40.8% were boys. The mean body mass index (BMI) was 17.8 kg/cm².

3.2. Reliability analysis

Cronbach's alpha presented an $\alpha = 0.803$, eliminating the 7th item (excessive subtalar motion) due to the low correlation between items of $\alpha = 0.23$ (poor content validity). Therefore, the 7th item was removed from the final version of the new instrument. Once the invalid item was removed, Cronbach's alpha was re-calculated, as 0.82. The correlations between the final six items obtained a mean value of 0.59 (range: 0.43–0.74), shown in Table 1. The correlation matrix between items is detailed in Table 2.

FAFI	LEFT		RIGHT	
	YES	NO	YES	NO
Ankle dorsiflexion. The patient lies supine; the knee is flexed to 45°; with moderate to strong force the ankle is dorsiflexed. Does the ankle flex more than 15 degrees? Along with the increased movement there may be bulging of the skin and subcutaneous fat anterior to the ankle.	YES	NO	YES	NO
Ankle anterior drawer test. The patient lies supine; the knee is flexed to 45°; the examiner grasps the heel along the plantar and posterior surfaces with one hand and applied a stabilising force against the anterior of the tibia with the other hand. Using a strong anterior force, can the calcaneum and talus be brought forwards on the tibia? Any forward movement felt is a positive result.	YES	NO	YES	NO
Subtalar inversion. The patient is supine with their feet over the end of the couch; the examiner holds the posterior surface of the heel and moves the heel into inversion without moving the leg. Is excessive inversion of the subtalar joint seen using minimal force? The sole of the foot or visualisation of the neck of the talus should show movement of 45° inwards, the lateral head of the talus will be very prominent.	YES	NO	YES	NO
Midtarsal inversion. The patient is supine with their feet over the end of the couch; themidtarsal joint is isolated from the subtalar joint; the forefoot is grasped from lateral to medial along the metatarsals; only minimal - moderate force is applied to invert themidtarsal joint. Does themidtarsal joint invert beyond 45° so that the plantar surface of the metatarsal heads can be brought inwards by 45 degrees?	YES	NO	YES	NO
Midtarsal ab/adduction and dorsi/plantarflexion. The patient is supine with their feet over the end of the couch; the examiner grasps and stabilises the rearfoot; the forefoot is moved in the direction of ab/adduction and dorsi/plantarflexion. Normal movement should be 1cm in each direction. With minimal force, does the forefoot move easily, almost “wobbling”, in an increased amount? Excessive movement in either of the two planes is a positive result.	YES	NO	YES	NO
Metatarsophalangeal movement. The patient is supine with their feet over the end of the couch; the hallux is dorsiflexed using minimal – moderate force. Does the hallux dorsiflex easily beyond 90° relative to the metatarsal?	YES	NO	YES	NO
Excessive subtalar joint pronation (weight-bearing). The patient is to march on the spot and stop on command; the patient is asked to invert their foot and hold the position close to subtalar joint neutral; the patient is then asked to relax their foot; the movement is observed. Does the arch lower and flatten fully, excessively, and easily, with the talus bulging medially? The pronation noted should be at the end of range of the subtalar joint motion so that no further pronation is possible	YES	NO	YES	NO
To score, each limb is calculated separately giving a left score and right score. Each YES is given one mark. A total of score of 7 marks is available				

Fig. 1. Definition of the 7 tested FAFI items.

3.3. Identification of the cut-off point and predictive values

To clarify case identification with joint hypermobility of the foot and ankle using FAFI, a cut-off point was determined. All children were assessed by an expert clinician examiner to determine the presence of joint hypermobility of the foot and ankle (AUC 0.96, $p < 0.001$). The point on the ROC curve closest to both axes determined the appropriate score for sensitivity and specificity. The discrimination score on the ROC curve (4 points) showed that FAFI can identify children with joint hypermobility of the foot and ankle (Fig. 2).

Analysis between sex found no difference in the cut-off point. The

related maximum Youden Index was 0.81 and the cut-off point was 4, hence a score $\geq 4/6$ indicates foot and ankle joint hypermobility (Table 3).

3.4. Intra-rater and inter-rater reliability

Excellent intra-rater reliability was found using intraclass correlations, (ICC2,1 = 0.96, 95% CI = 0.91 to 0.98, $p < 0.001$), and excellent inter-rater reliability was also observed (ICC2,1 = 0.89, 95% CI = 0.82 to 0.93, $p < 0.001$).

Table 1
Descriptive scores and reliability of items.

	Mean (SD)	Corrected item-total correlation	Cronbach's alpha (7th item deleted)
Ankle dorsiflexión	0.62 (0.48)	0.437	0.831
Ankle anterior drawer test	0.25 (0.43)	0.443	0.827
Subtalar inversión	0.51 (0.5)	0.645	0.788
Midtarsal inversión	0.67 (0.47)	0.654	0.787
Midtarsal ab/adduction and dorsi/ plantarflexion	0.58 (0.49)	0.746	0.766
Metatarsophalangeal movement.	0.53 (0.50)	0.656	0.786

SD: Standard Deviation.

3.5. Effects of age, sex, and laterality (left versus right)

The correlation between FAFI and age was significant and inverse ($r = -0.25$; $p < 0.01$).

With respect to sex, significant differences were found, girls were more hypermobile compared to boys, $F = 8.59$ $p < 0.005$.

Concerning laterality, there were no significant differences between the scores of the left and right sides for FAFI.

A correlation was found between sex and different groups of ages with FAFI, showing statistical significance differences ($p < 0.01$). Higher hypermobility was shown in the female younger groups. On the other hand, less hypermobility was shown in male older groups (Fig. 3).

3.6. Percentage agreement and correlation between the different joint hypermobility tests

After obtaining a cut-off score of 4/6 for the identification of foot and ankle joint hypermobility, FAFI scores were compared and contrasted with both the LLAS $\geq 7/12$, and Beighton score $\geq 5/9$.

There was case concordance between FAFI and the Beighton score of 53%. Comparing with BS, 47% of cases with FAFI $\geq 4/6$, were also positively identified with BS. In addition, 19% of cases with a positive BS, had a negative FAFI test result.

There was case concordance between FAFI and LLAS in 84%. Comparison with LLAS, found that 73% of cases where FAFI was $\geq 4/6$, LLAS

was also positive. In addition, 8% case discordance was found, with positive LLAS but negative FAFI.

A significant correlation was found between FAFI, LLAS, and Beighton score (Table 4).

4. Discussion

The principal objective of this study was the development of a tool to identify JH specific to the paediatric foot and ankle, with demonstrated test reliability and internal consistency, a defined cut-off point, and demonstrated validity.

We consider that this new specific tool is necessary to obtain the JH of the foot and ankle, based on a short form of the LLAS test, reducing it to 6 foot and ankle items, by eliminating one item, due to poor content validity, obtaining a good internal consistency (Annex 1). Excessive subtalar movement component was the item removed, possibly because it was the only item that was carried in relaxed loading. The rest of the FAFI items were measured with passive movements, with the patient in the supine position. In contrast, the subtalar movement component, was carried out by assessing the passive bone and ligamentous stability and active muscular and tendon stability with the patient standing. We theorized that this difference in the motor control of subtalar stability could have generated values that were not in accordance with the rest of the items.

The Beighton score cut-off score of 5/9 has previously shown sensitivity of 72% and specificity of 78%¹⁶. Similarly, the LLAS cut-off point has previously shown sensitivity of 68% and specificity of 86% [16]. The current study has determined $\geq 4/6$ as the optimal FAFI cut-off point, with high sensitivity (85%) and specificity (95%). The cut-off point was investigated for each sex, and both returned a cut-off point of 4. The present cut-off point has been calculated in a European ethnicity group, and it is expected that there may be differences in other participants with different ethnicity, who are reportedly more hypermobile [3,4,17].

Previous reliability investigation [18], has also provided psychometric data, and found inter-rater Beighton score reliability of 0.73 (0.42–0.88) and LLAS inter-rater reliability of 0.78 (0.41–0.93), indicative of excellent inter-rater reliability. The same study [18] obtained intra-rater reliability values for Beighton score (ICC = 0.96–0.98) and LLAS (ICC = 0.90–0.98). In our study, we obtained equal or higher values in both inter- and intra-observer reliability.

This research found similarities to other studies when hypermobility was compared with sex, age, and differences between left and right limbs. We found significant differences regarding sex, where girls were

Table 2
Inter-item correlation matrix.

	Ankle dorsiflexion	Ankle anterior drawer test	Subtalar inversion	Midtarsal inversion	Midtarsal ab/adduction and dorsi/plantarflexion	Metatarsophalangeal movement.
Ankle dorsiflexión	1					
Ankle anterior drawer test	196 ,252 ^a ,000	1				
Subtalar inversión	196 ,291 ^a ,000	196 ,471 ^a ,000	1			
Midtarsal inversión	196 ,414 ^a ,000	196 ,264 ^a ,000	196 ,508 ^a ,000	1		
Midtarsal ab/adduction and dorsi/ plantarflexion	196 ,345 ^a ,000	196 ,391 ^a ,000	196 ,597 ^a ,000	196 ,680 ^a ,000	1	
Metatarsophalangeal movement.	196 ,370 ^a ,000	196 ,313 ^a ,000	196 ,542 ^a ,000	196 ,516 ^a ,000	196 ,661 ^a ,000	1
	196	196	196	196	196	1

^a The correlation is significant at the 0.01 level (bilateral).

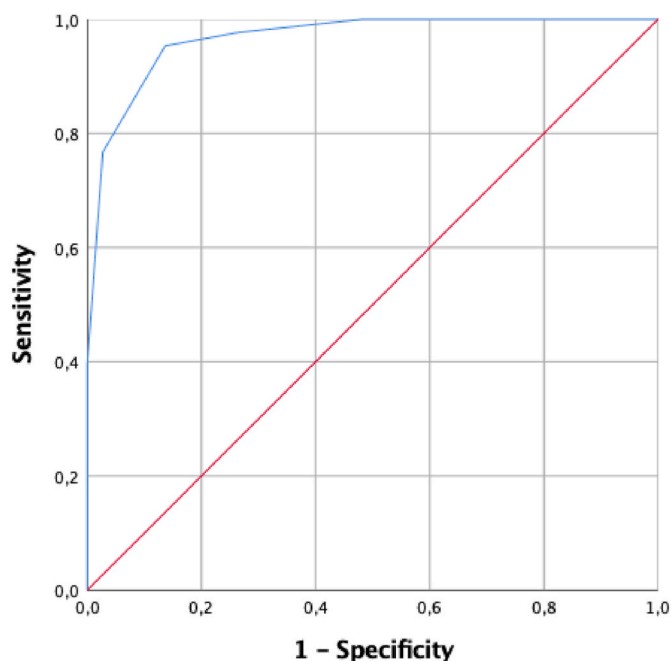


Fig. 2. ROC curve for the FAFI.

more flexible than boys at the foot and ankle level. These results agree with previous studies when compared with LLAS [19] and Beighton score [7,20,21]. When comparing left and right sides, no differences were found, in common with a previous study [19]. The current study found that joint hypermobility of the foot and ankle was inversely proportional to age ($r = -0.23$; $p < 0.01$), this may be due to the relationship between the loss of flexibility in the tissues and the aging of the joints and the decrease in physical activity [22]. Similarly, other studies have found the same relationship in lower limbs [19], and GJH [20,21,

Table 3
Sensitivity and 1-specificity for items of the FAFI.

Cutpoint	Sensitivity (%)	Specificity (%)	PPV (%)	NPV (%)	Youden's index	AUC	Metric Score
0	1	0	43.88%	NaN%	0.000	0.966	1.00
1	1	0.27	51.81%	100%	0.273	0.966	1.27
2	1	0.51	61.87%	100%	0.518	0.966	1.52
3	0.97	0.73	74.34%	97.59%	0.713	0.966	1.71
4	0.95	0.86	84.54%	95.96%	0.817	0.966	1.82
5	0.76	0.97	95.65%	84.25%	0.740	0.966	1.74
6	0.39	1	100%	67.9%	0.395	0.966	1.40

23].

It is unlikely that the BS, with only knee range examined in lower limbs, and predominantly upper limb signs, would always indicate foot and ankle JH. In our study, half of the cases in which FAFI was positive, Beighton score did not identify JH. It is important to understand that for foot and ankle injuries, the Beighton score should not be a primary test, as recent meta-analysis supports [11], where GJH was used as a variable of JH for the relationship with lower limb injuries. Further investigations also suggest using JH tests located pertinent to the body area [24,25]. Malek et al. [24] have likewise considered integrating body region items in to a revised Beighton score.

In the literature, LLAS has been little investigated as an identifier of lower limb JH [18,26–28]. Normally the test chosen for investigations has been Beighton score, even when lower limbs have been investigated. It is of great importance to relate the variables of gait with specific lower limb tests. The same principle applies when evaluating foot and ankle variables, where it is important to use the most applicable test, ie FAFI. In our study, one of four cases in which FAFI was positive, LLAS did not identify JH. Clinicians should be careful when using the LLAS score as a tool that only assesses joint hypermobility of the foot and ankle. Given the distribution of anatomical items, LLAS should be used when general joint hypermobility of the lower limbs is studied.

Concerning JH, specificity relates to the ability to accurately distinguish people who do not have hypermobility. Neither Beighton score or LLAS, appear sufficiently specific, to differentiate hypermobility which is localized to the foot and ankle. Hence, the importance of incorporating the new FAFI, a demonstrably reliable and valid tool, for the identification of foot and ankle joint hypermobility, with a cut-off

Table 4
Correlation between FAFI, Beighton score and LLAS.

		BS	LLAS
FAFI (n = 196)	Pearson correlation	,437 ^a	,923 ^a
	P-value	,000	,000

^a The correlation is significant at the 0.01 level (bilateral).

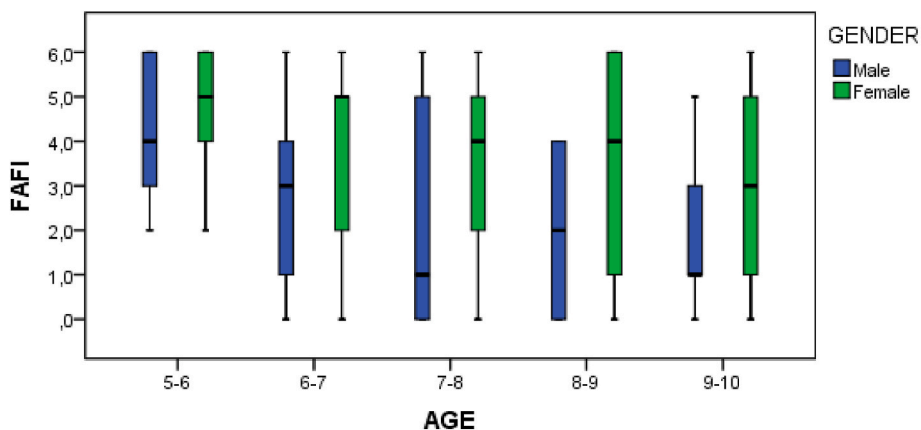


Fig. 3. FAFI relationship with age and sex.

point, for future foot and ankle research.

Further, clinicians need to appreciate joint hypermobility as both a local or systemic condition, and the overlap with hypotonia [29]. It has been suggested that children with hypotonia, present joint hyperlaxity as adults [30]. Clinically, hypotonia is observed, and indicated by the pull-to-sit test, the frog-sit posture, vertical suspension floppiness, and the scarf sign. No test has been validated for the diagnosis of hypotonia [30].

A limitation of this study is that only Caucasian-European participants were included, given that greater flexibility in Asian and African populations is reported [4,5]. Therefore, the cut-off point is only applicable for this paediatric population ethnicity.

Further investigations utilising FAFI should consider both wider age range, and more diverse ethnicities.

Our future research will compare FAFI with the validated Foot Posture Index, for the study of joint hypermobility of the foot and ankle

and developing foot posture.

5. Conclusions

The findings of this study demonstrate the new FAFI as an instrument which has good inter-rater reliability, sensitivity, and specificity for the identification of paediatric foot and ankle joint hypermobility.

FAFI should be used for assessment of joint hypermobility in the foot and ankle for future research, discarding BS, unless used in tandem.

When investigating joint hypermobility, prioritizing FAFI will focus on foot-ankle, more specifically than LLAS.

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Annex 1. Definition of the 6-FAFI items

FAFI	LEFT		RIGHT	
Ankle dorsiflexion. The patient lies supine; the knee is flexed to 45°; with moderate to strong force the ankle is dorsiflexed. Does the ankle flex more than 15 degrees? Along with the increased movement there may be bulging of the skin and subcutaneous fat anterior to the ankle.	YES	NO	YES	NO
Ankle anterior drawer test. The patient lies supine; the knee is flexed to 45°; the examiner grasps the heel along the plantar and posterior surfaces with one hand and applied a stabilising force against the anterior of the tibia with the other hand. Using a strong anterior force, can the calcaneum and talus be brought forwards on the tibia? Any forward movement felt is a positive result.	YES	NO	YES	NO
Subtalar inversion. The patient is supine with their feet over the end of the couch; the examiner holds the posterior surface of the heel and moves the heel into inversion without moving the leg. Is excessive inversion of the subtalar joint seen using minimal force? The sole of the foot or visualisation of the neck of the talus should show movement of 45° inwards, the lateral head of the talus will be very prominent.	YES	NO	YES	NO
Midtarsal inversion. The patient is supine with their feet over the end of the couch; themidtarsal joint is isolated from the subtalar joint; the forefoot is grasped from lateral to medial along the metatarsals; only minimal - moderate force is applied to invert themidtarsal joint. Does themidtarsal joint invert beyond 45° so that the plantar surface of the metatarsal heads can be brought inwards by 45 degrees?	YES	NO	YES	NO
Midtarsal ab/adduction and dorsi/plantarflexion. The patient is supine with their feet over the end of the couch; the examiner grasps and stabilises the rearfoot; the forefoot is moved in the direction of ab/adduction and dorsi/plantarflexion. Normal movement should be 1cm in each direction. With minimal force, does the forefoot move easily, almost "wobbling", in an increased amount? Excessive movement in either of the two planes is a positive result.	YES	NO	YES	NO
Metatarsophalangeal movement. The patient is supine with their feet over the end of the couch; the hallux is dorsiflexed using minimal - moderate force. Does the hallux dorsiflex easily beyond 90° relative to the metatarsal?	YES	NO	YES	NO
To score, each limb is calculated separately giving a left score and right score. Each YES is given one mark. A total of score of 6 marks is available per each limb. Cut-off point $\geq 4/6$.				

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