■ CHILDREN'S ORTHOPAEDICS

An investigation into the aetiology of flexible flat feet

THE ROLE OF SUBTALAR JOINT MORPHOLOGY

A. Kothari,
S. Bhuva,
J. Stebbins,
A B. Zavatsky,
T. Theologis

From NDORMS, University of Oxford, Oxford, United Kingdom

Aims
There is increasing evidence that flexible flatfoot (FF) can lead to symptoms and impairment in health-related quality of life. As such we undertook an observational study investigating the aetiology of this condition, to help inform management. The hypothesis was that as well as increased body mass index (BMI) and increased flexibility of the lower limb, an absent anterior subtalar articulation would be associated with a flatter foot posture.

Patients and Methods
A total of 84 children aged between eight and 15 years old were prospectively recruited. The BMI for each child was calculated, flexibility was assessed using the lower limb assessment scale (LLAS) and foot posture was quantified using the arch height index (AHI). Each child underwent a sagittal T1-weighted MRI scan of at least one foot.

Results
An absent anterior subtalar articulation ($p < 0.001$) and increased LLAS ($p = 0.001$) predicted a low AHI. BMI was not a significant predictive factor ($p = 0.566$).

Conclusion
This is the first study to demonstrate the importance of the morphology of the subtalar joint on the underlying foot posture *in vivo*.

Take home message: Flexibility of the lower limb and absence of the anterior facet of the subtalar joint are associated with flexible FF and may influence management of this common condition.

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■ A. Kothari, MRCS, MSc, DPhil, Specialist Trainee, Trauma and Orthopaedic Surgery, NDORMS
■ T. Theologis, FRCS, MSc, PhD, Consultant Orthopaedic Surgeon, NDORMS
University of Oxford, Windmill Road, Oxford, OX3, UK.

■ S. Bhuva, FRCR, Consultant Radiologist
Oxford University Hospitals NHS Trust, Churchill Hospital, Old Road, Oxford, OX3 7LE, UK.

■ J. Stebbins, DPhil, Clinical Scientist, Oxford Gait Laboratory, Nuffield Orthopaedic Centre
Oxford University Hospitals NHS Trust, Windmill Road, Oxford OX3 7HE, UK.

■ A B. Zavatsky, MA, DPhil, Reader in Engineering Science, Tutor in Engineering, Department of Engineering Science
University of Oxford, Parks Road, Oxford OX1 3PJ, UK.

Correspondence should be sent to Mr A. Kothari; e-mail: alpesh.kothari@ndorms.ox.ac.uk

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Flexible flatfoot (FF) deformity is common, being present in up to 20% of children and adolescents.[1] A distinction needs to be made clinically between FF in older children, developmental FF in toddlers and young children, and rigid FF due to a structural abnormality. It is accepted[2] that developmental flat feet require no intervention, and the treatment for rigid flat feet commonly due to congenital vertical talus or tarsal coalition is well described.[2] Significant controversy exists, however, about the clinical significance, and thus the management of FF in older children. It has been suggested that it is a benign normal variant requiring no intervention.[3,4] However, there is increasing evidence that a FF posture can lead to significant symptoms and impairment of health-related quality of life.[5-8]

To understand how FF may cause symptoms and how they might be treated, it is important to understand the underlying aetiology. An association between increased body mass index (BMI) and a flatter foot has been demonstrated.[9,10] El et al[11] demonstrated that hypermobile children had double the prevalence of FF of non-hypermobile children. Gender and heritable factors may also play a role in determining foot posture.[12]

An aetiological factor, ignored in contemporary literature, was initially discussed in a seminal paper by Harris and Beath.[13] From osteological specimens they suggested that there are two types of subtalar joint; one 'firm' supporting the talus well, and the other 'weak' allowing the foot to adopt a FF posture. Morphological variations of the subtalar joint have subsequently been described by several authors, with the most striking variant being an absent anterior articulation (AA) (Fig. 1).[14,15] The anterior facet on the os calcis forms an integral part of the 'acetabulum pedis'[16] (AP) supporting the talar head. Absence of this articulation could lead to a FF posture,[14] however, this has not been demonstrated *in vivo*.

Fig. 1a Fig. 1b Fig. 1c Fig. 1d

Diagram showing the variation in the articulating surface morphology of the os calcis as described by Bruckner.[14] A has three separate articular surfaces (anterior/middle/posterior). The anterior and middle articular surfaces joined in B and C. The anterior articular surface is missing in D.

The treatment of symptomatic FF usually involves orthotics, physiotherapy or both, and when these fail surgery may be performed. The aim of treatment is usually to make the foot appear more ‘normal’, with the assumption that the flatter the foot, the worse the symptoms. However, without fully understanding the aetiology, it is unclear whether this approach is appropriate. If the morphology of the subtalar joint is a significant determinant of FF, it may affect the efficacy of any intervention.

The aim of this study was to assess the role of potential aetiological factors in determining foot posture, with emphasis on the importance of the subtalar joint. The hypothesis was that the morphology of the subtalar joint would be an important determinant of foot posture, in conjunction with the flexibility of the lower limb and BMI.

Patients and Methods

This study had ethical approval (ref:-12/SC/0334). A total of 84 children were recruited prospectively from orthotic or orthopaedic clinics or from the community. Inclusion criteria were: neutral or flexible FF posture and aged between eight and 15 years. Exclusion criteria were: any bone, joint or neurological disease, evidence of rigid FF, previous surgery to the spine or lower limbs or concurrent use of orthotics. The children were part of a cohort from which health-related quality of life data were presented previously.[5]

The assessment of foot posture and clinical examination. The classification of foot posture on the basis of visual inspection is known to be subjective with poor reliability.[17] It was therefore quantified using the arch height index (AHI), described by Williams and McClay.[18] The AHI is the ratio of standing dorsal arch height as measured at 50% foot length divided by the truncated foot length (foot without toes). This measure has excellent inter- and intra-observer reliability.[18,19] Ranges of AHI can be applied to define low (< 0.31), neutral (0.31 to 0.37) and high arches (> 0.37),[20] but in this study AHI was used as a continuous variable alone. Where there was a reduced medial longitudinal arch (MLA) in the standing position, a double heel raise test was performed to ensure that the MLA was reconstituted, in order to exclude rigid FF deformity. All children had the same type of foot bilaterally, consistent with the findings of Mosca et al.[12]

Age and gender were recorded and BMI was calculated. Flexibility was assessed using the lower limb assessment scale (LLAS),[21] which is scored out of 12 for each limb and is based on the range of movement of the hip, knee and foot and ankle joints. As the LLAS focuses on the lower limb and foot, it was favoured over other commonly used measures.

MRI. Each child underwent an MRI scan (Philips Achieva 3.0 Tesla, Philips Medical Systems, Da Best, Amsterdam, The Netherlands). Scans were obtained from 127 feet of the 84 children, as a proportion of the children could not tolerate imaging of both feet. Bilateral studies were obtained for 43 children (51%) and unilateral for 41 children (49%). A SENSE Flex L coil was used to maximise spatial resolution. The imaging protocol included a sagittal T1-weighted spin-echo (T1-W) sequence. The T1-W sequence had a pixel size of 0.27 mm, slice thickness 1.5 mm to 2.5 mm, and slice increment 2.2 mm to 3.1 mm. The sagittal MRI images were inspected using Mimics software (Materialise; Leauven, Belgium). A protocol based on that recommended by Shahabpour et al[22] was used to identify the articular facets of the subtalar joint. The subtalar joint was then formally classified using Bruckner's classification.[14] (Fig. 1). An example of the visualisation of the subtalar joint is shown in Figure 2.

Statistical analysis. Continuous data were assessed for skewness and kurtosis; all variables were normally distributed. The inter- and intra-observer reliability of the classification of the subtalar joint was assessed in 20 feet by an orthopaedic surgeon (AK) and a radiologist (SB). Cohen's kappa (κ) was used to quantify agreement.[23] Multiple linear regression was used to assess whether age, gender, BMI,

Fig. 2a

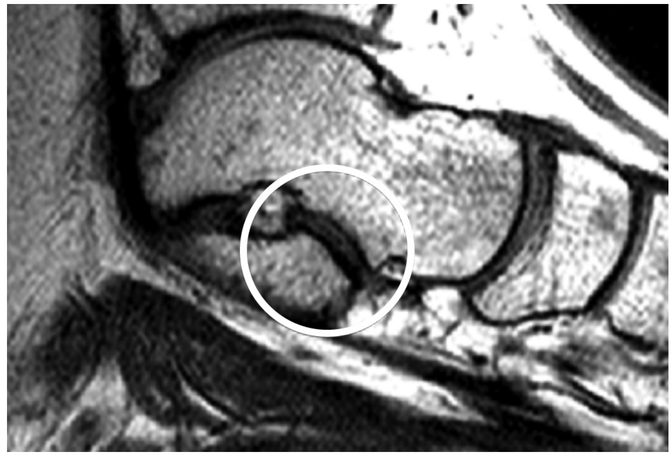

Fig. 2b

MRI showing the morphology of the subtalar joint. Discrete posterior, middle, and anterior articular facets of the os calcis can be seen clearly. As the articular facets were not joined, this foot was classified as Bruckner type A.[14]

Table I. Summary statistics for age, gender, lower limb assessment score (LLAS) and body mass index (BMI)

Parameter	All subjects (n = 84)
Age (yrs) mean (standard deviation (SD)), range	12.0 (2.2), 8.1 to 16.0
Gender (M:F)	46:38
Flexibility, LLAS median (interquartile range), range	4 (3), 0 to 11
BMI (kg/m^2) mean (SD), range	18.6 (3.3), 12.1 to 28.3

Table II. Subtalar joint morphology (supportive or unsupportive) in the children, with mean arch height index for those in each group. Pairs of feet had the same subtalar joint morphology

	Subtalar joint morphology	
	Supportive	Unsupportive
Number of feet (%)	85 (*67*)	42 *(33)*
Number of subjects (%)	56 (*67*)	28 *(33)*
Arch height index, mean (SD)	0.31 (0.03)	0.29 (0.03)

LLAS, and subtalar morphology could predict AHI. Regression standard errors were adjusted to account for paired foot data.[24] Regression diagnostics were used to ensure that model residuals were normally distributed and that the residual *versus* fitted values did not demonstrate any heteroscedasticity. The level of significance was set at 0.05. Effect size was reported using Cohen's f^2, calculated as $R^2/(1-R^2)$. The effect size gives an indication of the strength of the relationship between predictor variables and outcome (AHI), where $f^2 = 0.02$ is equivalent to a small effect size, $f^2 = 0.15$ medium effect size and $f^2 = 0.3$ a large effect size. For the multiple linear regression analysis of 84 children, the analysis had 80% power to detect an overall model effect size (f^2) of 0.12. Sample size and power calculations were made using GPower (v3.1.9.2 Universitat Kiel, Germany). Data analysis was undertaken using SPSS v22 (IBM) and Stata v13.0 (Statacorp, College Station, Texas).

Results

The age, gender, flexibility and BMI of the children are summarised in Table I. The mean AHI was 0.31 (SD 0.03; 0.22 to 0.37).

Reliability of classifying the morphology of the articular surface of the os calcis. The inter-observer reliability (κ) was 0.718, (95% confidence interval (CI) 0.600 to 0.836) and intra-observer reliability was 0.863 (95% CI 0.773 to 0.953). Whilst both were regarded as at least substantial agreement, difficulty was experienced in differentiating between Bruckner types B and C.[23] The classification of the articular morphology was therefore simplified to a binary outcome; supportive (Bruckner A, B, C) and unsupportive (Bruckner D). The AA was present in a supportive morphology and absent in the unsupportive type. Using this classification, inter- and intra-observer reliability both improved to κ = 0.886 (95% CI 0.776 to 0.996), represent-

Table III. Summary of results of multiple linear regression, including regression coefficient, standard error (std. error), significance (Sig.) and 95% confidence intervals of coefficient

Predictor	Coefficient	Robust std error	Sig. (p)	95% Confidence interval	
Flexibility (LLAS)	-0.0051	0.0014	0.001*	-0.0079	-0.0023
Body mass index	-0.0001	0.0010	0.566	-0.0003	0.0015
Subtalar morphology	0.0214	0.0060	< 0.001*	0.0095	0.0333
Gender	0.0105	0.0062	0.094	-0.0018	0.2276
Age	-0.0010	0.0014	0.464	-0.0038	0.0018

* Significant predictors
LLAS, lower limb assessment score

ing almost perfect agreement,[23] and this classification was used in the regression analysis.

The predictors of AHI. Of the 84 children, about one third had unsupportive subtalar articulations. In children in whom MRI scans of both feet were obtained both had the same subtalar joint morphology (Table II).

In the multiple linear regression model, only two of the potential variables, flexibility (LLAS) and subtalar morphology, predicted AHI (Table III). The overall $R^2 = 0.32$ represents a large effect size ($f^2 = 0.47$). Of the overall variance in AHI, LLAS accounted for 22% ($f^2 = 0.28$) and subtalar morphology accounted for 9% ($f^2 = 0.10$). Thus the more flexible the lower limb, the flatter the foot, and feet were flatter in the group with no anterior subtalar facet. BMI, gender and age were not significant predictors of AHI. There was no significant interaction between predictive factors.

Discussion

The main purpose of this study was to investigate the determinants of FF posture. As age was not a significant predictor, we assume that our test sample was out of the age range of developmental FF. The finding that BMI was not a predictor of foot posture conflicts with previous literature and our initial hypothesis. Much of the literature describing the link between foot posture and BMI uses footprint measurements.[9,10] Increased BMI is associated with increased adiposity which affects the footprint indices[25] and the foot may appear flatter, when the underlying bony architecture is normal. The AHI may be less sensitive to adiposity than footprint parameters, and may be a better index for use in clinical practice. Alternatively, it may be because the mean BMI of the children in the study was relatively low at 18.6 kg/m^2. The results might have been different if more children with a high BMI had been recruited.

This is the first clinical study to demonstrate the importance of subtalar morphology in foot posture. The role of subtalar morphology has previously been hypothesised on the basis of observations in osteological specimens, but not confirmed *in vivo*.[13,14] The AA of the os calcis is an integral component of the talocalcaneonavicular joint, which is also known as the AP.[26] Dissection of foetal cadaveric specimens by Epeldegui and Delgado[16] demonstrated that the anterior facet of the os calcis normally forms part of the osseous floor of the AP, supporting the talar head. They suggested that variations in subtalar morphology would affect the shape of the foot. Barbaix et al[15] found that the articular surface area of the os calcis in the subtalar joint was significantly reduced when the anterior facet was missing, suggesting that this morphology provided less bony support to the talar head. Thus, an absent AA places greater reliance on the plantar ligaments to support the talar head. Over time these may stretch, allowing plantar-medial deviation of the talar head with reduction of the medial longitudinal arch, as seen in flexible FF deformity. Whilst no statistical interaction was found between flexibility and subtalar morphology, one might expect there to be an additive effect.

Increased flexibility may result in impaired static stabilisation of the joints by capsuloligamentous structures, thereby permitting increased and potentially pathological movement of the joint. This would be especially important at the subtalar and midtarsal articulations. Muscle strengthening regimes have been used, with a suggestion that these may improve foot posture.[27] Further work is required to evaluate dynamic stabilisation as a potential form of treatment.

With respect to subtalar morphology, there has been concern that lengthening of the lateral column may damage the subtalar articular surface,[28] and this may be less of a concern if there is no AA. If, however, the articular surface is Bruckner type B or C, no modification of the lateral column lengthening technique can prevent the surgeon from damaging the surface of the joint, and an alternative surgical approach may be preferred. Pre-operative imaging is recommended to assess the articular morphology if surgery is planned.

The main limitations of this study were the potential selection bias as children with flexible FF were recruited from orthotic and orthopaedic clinics, and that MRI may not be the best modality to assess bony morphology. Three-dimensional reconstructed CT would be better, however, the dose of ionising radiation precludes its use as a research tool in children.

In conclusion, we found that two of the most important factors for determining foot posture are flexibility of the lower limb and the morphology of the subtalar joint. These are new findings. Further work is required to clarify the relationship between these factors and the development of symptoms in children with flexible FF. This will have a bearing on monitoring and treatment, especially in asymptomatic children with a significant deformity.

Author contributions:
A. Kothari: Data collection and analysis, writing the paper.
S. Bhuva: Data analysis, writing the paper.
J. Stebbins: Data collection and analysis, writing the paper.
A. B. Zavatsky: Data collection and analysis, writing the paper.
T. Theologis: Data collection and analysis, writing the paper.

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