The evaluation of flexible flat feet in children aged eight to fifteen years old

Dissertation submitted to the University of Oxford in partial
fulfilment of the requirements for the degree of

Doctor of Philosophy

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Trinity Term 2015

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by Alpesh Kothari, St Hugh's College, DPhil, Trinity 2015

Abstract

Paediatric flexible flat feet (PFF) are a very common presentation, but considerable debate continues about its aetiology, diagnosis and management. In this thesis a cohort of children with PFF (n=48) and neutral feet (NF)(n=47) have been investigated to test the theory that flat foot posture is related to the presence of symptoms and biomechanical dysfunction. Aetiological and clinical factors that may result in functional impairments have also been investigated. The ultimate aim of this thesis was to better define treatment indications for PFF. Firstly the question of what defines PFF was addressed and appropriate categorical and continuous measures of foot posture were proposed. Subsequently the clinical importance of foot posture was contextualized by demonstrating that children with PFF had significant impairment of health-related quality of life (HRQOL) compared to NF children. Investigation into the aetiology of PFF demonstrated that an absent anterior articulation of the subtalar joint, increased lower limb flexibility and knee valgus were factors associated with this condition. Inabilities to perform a single heel raise and to attain plantigrade were found to be sensitive tests to identify children with more symptomatic PFF. Differences in navicular motion of PFF compared to NF children were noted, hinting at midfoot dysfunction, and formal gait analysis demonstrated a number of deviations in gait parameters associated with a flat foot posture. Of these, walking speed and increased forefoot supination were related to impaired HRQOL. There was also evidence of potential instability in the PFF children, and foot ankle joint moments in this group were suggestive of impaired transition of the foot from a supple compliant structure to a rigid lever during stance. Whilst children with PFF were more likely to have proximal joint symptoms, kinematic and kinetic analysis did not identify the mechanism by which this could occur. The findings of this thesis have been utilized to define an evidence-based algorithm for the treatment of PFF.

Acknowledgements

I would firstly like to thank Mr Tim Theologis, Dr Amy Zavatsky and Dr Julie Stebbins for their advice, expertise and encouragement throughout this project. I would also like to thank my fellow DPhil student Philippe Dixon for showing me the art in computer programming. Thanks for your patience and instruction.

This work would not have been possible without the research training fellowship awarded to me by Action Medical Research.

Finally I would like to dedicate this thesis to my wife Sarah, and our boys Farris and Joseph.

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Chapter 1. Introduction

1.1. Purpose and aim

There are few orthopaedic conditions with the notoriety of flat feet. One could question a member of the public, and not only would they know someone with flat feet, they would also have an opinion about this ‘affliction’. A common opinion that continues to be voiced by many is, ‘you can’t go into the army with them’. Unbeknownst to these people, they are actually referring to the 19th century work by Ilfeld [136]. He demonstrated that 30-40 percent of army recruits were dismissed from the army because of foot pain and that there was an association between foot pain and flatfootedness. This led to the prevailing view among the general public and some clinicians that flat feet are a problem. After Ilfeld’s publication probably the most cited paper in the field of flat feet research was published by Harris and Beath [120]. They investigated the feet of Canadian army recruits and found that flat feet were not necessarily a cause of disability. It was only in certain circumstances that the army recruit developed problems related to his flat feet. “It may appear strange that there is any necessity in discussing such a common and apparently simple entity as flat-foot” is the opening line from this seminal 1948 text. If this was the view in 1948, then one would assume that with over sixty more years of research on the topic, it would be even stranger to need to discuss flat feet today. This, however, is not the case, and even in 2015 significant controversies still exist over the flat foot, not least as to whether it is a pathology or just a normal variant. Further controversies also exist with regard to aetiology, classification and treatment of flat feet. Not only do these issues warrant discussion, they also warrant further investigation and resolution. Flat feet take various guises, but the focus of this thesis, and the cause of the most controversy, is flexible flat feet in children (paediatric flexible flat feet - PFF).

Flexible flat feet in children are common, present in up to 20% of the general population [85]. Whilst many children do not appear to suffer any adverse effects from PFF, it appears that some children can develop foot and ankle symptoms and functional limitation, as well as symptoms higher up the kinetic chain (p.25) [18, 129]. There continues to be uncertainty as to how, why and to what extent or severity these symptoms occur. A prominent theory proposes that excessive hindfoot eversion, and an unlocked midtarsal joint disrupt the normal re-supination process required for the foot to transition from a supple structure to a rigid structure used to “push off” against the ground [137, 316]. This in turn leads to abnormal midfoot and forefoot motion with stretching and attenuation of the capsuloligamentous structures. Pushing off with a supple foot may in turn lead to impaired propulsion, instability and abnormal foot and ankle loading [316]. Other factors that may also further impair the dynamic control of the arch include muscle fatigue [111, 188], increased flexibility [86, 180], increased body mass index [82, 330], plantarflexor contracture [120, 129] and altered lower limb alignment [291, 356]. Structural factors including the shape of the subtalar joint articulation may also have a bearing on foot posture [10, 36]. It is theorized that what starts as a purely flexible deformity may over time lead to secondary joint degeneration, although no longitudinal studies confirm this.

In terms of symptoms at joints proximal to the ankle it has been proposed that excessive hindfoot eversion, as observed in PFF, is coupled with increased internal rotation of the tibia and hip during stance [229, 296]. Increased internal rotation of the hip may in turn detrimentally alter the lever arm of the hip abductors, and increase the

Q-angle at the knee further overloading the lateral patellar facet. It has been demonstrated that arch height is related to the magnitude of accelerations at the lumbar spine during running [239], pelvis alignment [160] and gluteal and erector spinae muscle activity [21]. Whilst numerous theories exist as to how PFF may cause symptoms there is currently very little clinical evidence to corroborate them.

Treatment of PFF is routinely undertaken, and intervention is commonly in the form of orthotics and/or physiotherapy [221]. When symptoms are refractory to treatment by non-operative methods, surgical intervention may be considered [221]. Guidelines for the management of PFF have been produced by a number of authorities, although the underlying limitation to any guidance is that it remains unclear why some children have symptomatic PFF, and others do not [92, 119]. Without establishing the pathomechanics of PFF, treatment by any method, especially surgical, is difficult. This leads to the overall aim of this thesis: to discover if there are structural or functional factors associated with PFF that relate to the presence and severity of symptoms. By doing this, it is hoped that the treatment indications for children with PFF can be better defined.

1.2. Thesis Overview

Chapter 2 provides a complete description of the clinical entity of PFF as well as critical appraisal of the existing literature on this subject. The prevailing theories about how PFF may result in symptoms and functional impairment are presented, as well as the ongoing gaps in our knowledge. By the end of the chapter the motivation, rationale, and need for this study should be clear.

Chapter 3 begins with a discussion of the methods that could be used to achieve the specific aims of the study, and the justification for those which ultimately have been used. This is followed by a description of the study design, details of the subject recruitment process and general data collection protocols.

Before the main objectives of the thesis can be described, two important questions are addressed.

1. What constitutes a flat foot?

There is no consensus on the best method for classifying foot posture [261]. The overall purpose of **Chapter 4** is to define what constitutes a flat foot. It is imperative to establish this from the outset, as the findings and conclusions of the whole thesis will depend on this definition.

2. Is PFF actually a problem?

Whilst the thesis is based on the premise that some children with PFF do get significant foot and ankle symptoms and functional limitation, some authors disagree with this and state that PFF is a benign normal variant with a benign natural history [297]. To warrant further investigation into the structure and function of PFF it is vital to establish that PFF can cause problems. In **Chapter 5** the relationship between foot posture and health-related quality of life (HRQOL) is assessed, to confirm that further investigation into the pathomechanics of PFF is warranted.

Chapter 6 describes an investigation of two aspects of PFF. Firstly potential aetiological factors of PFF are considered, including subtalar joint morphology, with the view of guiding potential treatment strategies. Secondly the utility of examining for contracture of the ankle plantarflexors and ability to perform a single heel raise are evaluated as potential hallmarks of more symptomatic PFF.

Chapter 7 is the first of four chapters utilising gait analysis to quantitatively investigate function in PFF. It is hypothesized that there would be evidence of altered midfoot function in PFF compared to children with neutral feet (NF). This is tested by tracking the pattern of navicular motion in the sagittal and transverse planes, as well as the coupled relationship between motions in both planes.

In **Chapters 8 and 9** the relationship between foot posture and foot and ankle biomechanics is investigated. It is hypothesized that a flat posture will result in foot and ankle joint motion consistent with impaired re-supination. It is also hypothesized that impaired locking of the midtarsal joint in PFF leads to evidence of impaired propulsion, instability and abnormal foot and ankle loading. The relationship between positive findings and HRQOL has also been assessed to identify which factors are likely to be most contributory to foot and ankle symptoms.

In **Chapter 10** the relationship between PFF and the proximal joints is assessed. The first stage is to test the hypothesis that PFF are associated with proximal joint symptoms, by using logistic regression to predict the presence of knee and hip or back symptoms. Then kinematic and kinetic parameters that relate to foot posture are identified, with particular focus on hip and tibia rotation. The association between positive kinematic and kinetic findings and proximal joint symptoms is tested.

Chapter 11 is the final chapter, and here findings from all the individual chapters are summarized and discussed. The focus is on the ultimate aim of the thesis, which is to define treatment indications for children with PFF.

Chapter 2. Background and literature review

2.1. Introduction

The aim of this chapter is to fully describe the clinical entity of PFF and to undertake a critical appraisal of the existing literature on this subject. The prevailing theories about how PFF may result in symptoms and functional impairment as well as the problems that have arisen in the field are presented. By the end of the chapter the motivation, rationale, and need for this study should be clear. Before these objectives can be understood, a working knowledge of the terminology used in this thesis, as well as the functional anatomy and biomechanics of the foot and ankle complex, is required.

2.2. Terminology

Flat feet are a clinical presentation seen by a number of different healthcare professionals including orthopaedic surgeons, sports medicine doctors, physiotherapists, orthotists and podiatrists. Terminology and jargon varies between specialities, and even similar terms may be interpreted differently. As such this can lead to confusion both in clinical and research settings. To minimise confusion and ensure consistency throughout this thesis, commonly used terms for foot and ankle motion are defined in this section. Figure 2.1 defines the planes of reference used in this thesis.

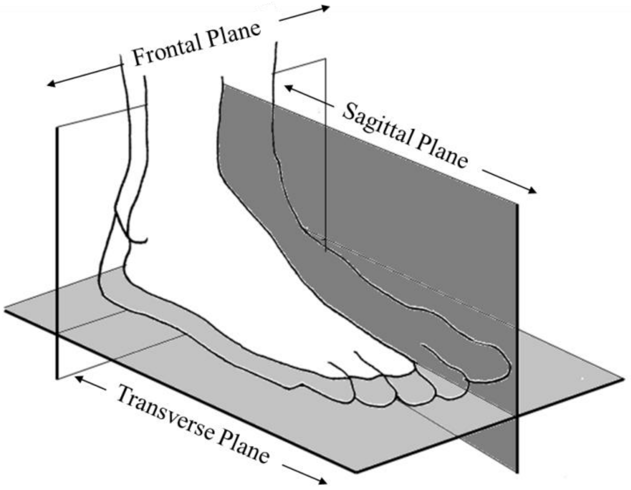


Figure 2.1 Reference planes of the foot. Frontal plane also referred to as coronal plane.

Foot motions

Dorsiflexion/Plantarflexion: Dorsiflexion refers to sagittal plane movement of the foot or foot segment such that the dorsal surface of the foot/segment is moving closer to the shin. Plantarflexion is when the dorsal surface of the foot/segment moves away from the shin (Figure 2.2). In the context of toe movement dorsiflexion is interchangeable with extension and plantarflexion is interchangeable with flexion.

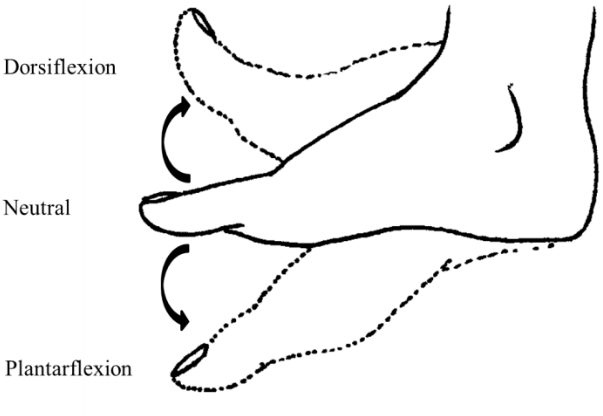


Figure 2.2 Dorsiflexion and plantarflexion of the foot.

Internal/External rotation: Only used in the context of transverse plane motion of the hindfoot. Internal rotation refers to the whole hindfoot rotating towards the midline of the body, in the anatomical position, with respect to the tibia. External rotation refers to rotation of the hindfoot away from the midline.

Eversion/Inversion: Coronal plane motion of the hindfoot. Inversion is movement of the hindfoot towards the midline and eversion is movement of the hindfoot away from the midline. Hindfoot varus is equivalent to an inverted hindfoot and hindfoot valgus is equivalent to an everted hindfoot (Figure 2.3).

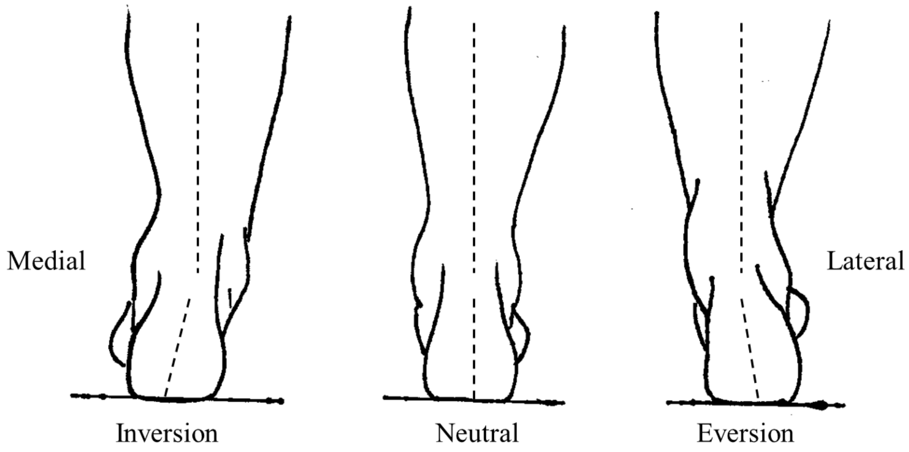


Figure 2.3 Eversion and inversion of hindfoot.

Abduction/Adduction: Transverse plane motion of the midfoot, forefoot and hallux. Abduction is movement of the segment away from the midline, and adduction is towards the midline.

Pronation/Supination: This term is responsible for the most confusion among clinicians. Classically pronation is defined as a combination of hindfoot eversion, foot plantarflexion and abduction. Supination is defined as hindfoot inversion, foot dorsiflexion and adduction [234]. Thus in the context of a non-weight bearing foot in pronation the sole faces laterally and in supination the sole faces medially (Figure 2.4a). The confusion arises in the weight-bearing setting where a foot can appear seemingly pronated, but in a non-weight-bearing setting, if the hindfoot is held in neutral, the forefoot may be actually supinated relative to the hindfoot with the sole facing medially. In pathological foot presentations the coordination between motions at different joints in different planes may be also disrupted. Thus the terms pronation and supination for this thesis will only refer to the position of the forefoot relative to the hindfoot. Pronation refers to the rotation of the forefoot relative to the hindfoot about the longitudinal axis of the foot such that the 1st ray moves plantarwards and 5th ray moves dorsally (clockwise for the right foot as viewed from the front). Supination is rotation of the forefoot in the opposite direction such that the 1st ray is elevated and the 5th ray lowered relative to the plantar surface of the foot (Figure 2.4b).

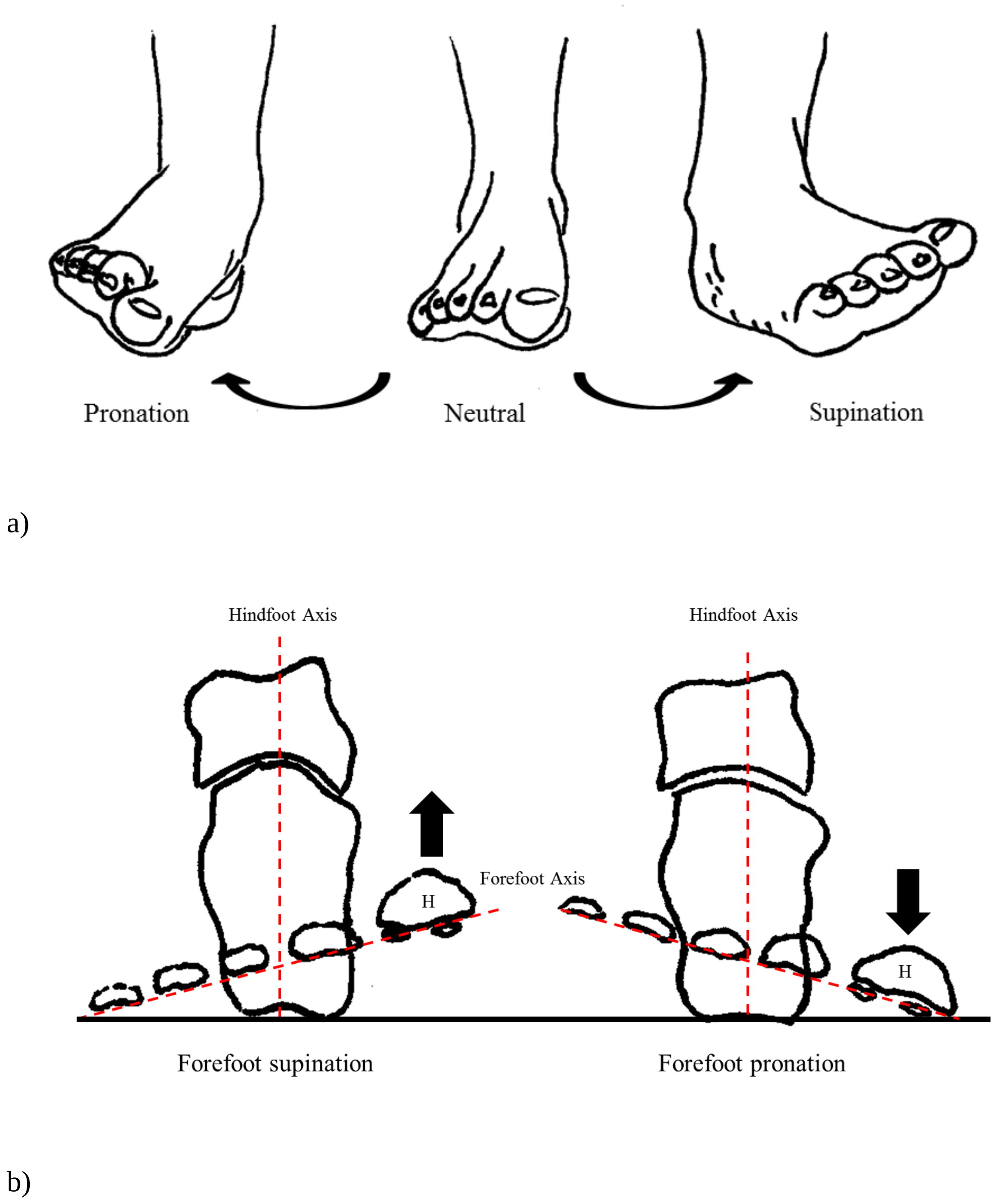


Figure 2.4 a) Non-weightbearing pronation and supination. b) Pronation and supination of the forefoot with respect to the hindfoot. Hallux (H). Right Foot.

2.3. Functional anatomy and biomechanics of the foot and ankle

The foot and ankle complex is an incredibly intricate structure consisting of 28 bones (including sesamoids) (Figure 2.5), 35 articulations, over 100 ligaments, and 29 muscles employed to control its actions. To understand how flat feet may cause problems it is imperative to know how the foot and ankle function. In terms of motion, the foot is often divided into three functional units, hindfoot, midfoot and forefoot (Figure 2.5). The hindfoot consists of the os calcis and the talus, the midfoot consists of the cuboid, navicular and cuneiforms, and the forefoot consists of metatarsals and phalanges. The principal foot and ankle motions occur at ankle joint, subtalar joint and midtarsal joints. The functional anatomy and biomechanics of these joints are summarized in the following section.

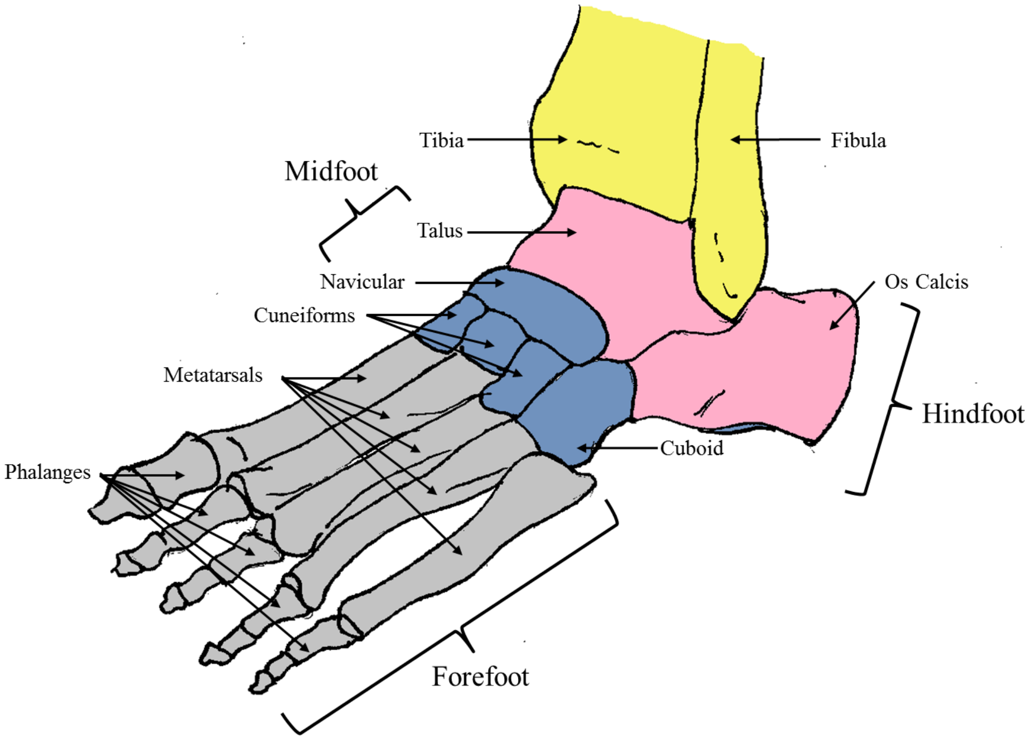


Figure 2.5 Bones of the foot with foot divisions.

2.3.1. The Ankle Joint/Talocrural joint

The ankle is a mortise joint formed by articulation of the fibula and distal tibia with the talus, with the main motions of dorsiflexion and plantarflexion. Inman [137] first defined the axis of the ankle joint as essentially passing through the distal tips of the malleoli. Hicks [123] and Barnett et al. [11] recognized that actually there were two distinct axes to the ankle. In plantarflexion this axis was orientated downward and medially, whereas in dorsiflexion it was orientated downward and laterally (Figure 2.6). Lundberg et al. [185] further investigated the ankle axis in vivo using Roentgen stereophotogrammetry of eight volunteers with tantalum beads embedded into their bones. Lundberg et al.'s findings, in part, corroborated the work of Hicks and Barnett, but also showed that the amount of medial or lateral inclination of the axis changed continuously, dependent on the degree of plantarflexion or dorsiflexion of the ankle joint. This moving instantaneous axis of rotation of the ankle joint has been demonstrated subsequently in vivo and in cadaveric studies [174, 284].

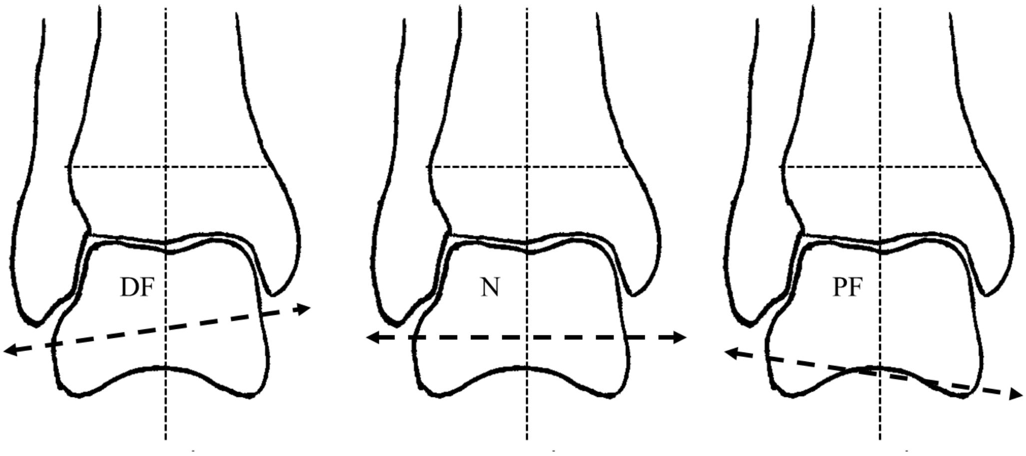


Figure 2.6 Changing orientation of the ankle axis (dashed line) dependant on whether in dorsiflexion (DF), neutral (N) or plantarflexion (PF) (adapted from [11]).

Reported normal ranges of ankle sagittal plane motion vary between authors ranging from 0° to 23° of dorsiflexion and up to 56.2° of plantarflexion [26, 273, 338]. The American Academy of Orthopaedic Surgeons has provided consensus values of 18° for dorsiflexion and 48° of plantarflexion [112].

Transverse and coronal plane motion of the ankle was historically felt to be negligible, but contemporary evidence has demonstrated that the ankle actually has marked freedom of movement. Using bone mounted pins Lundgren et al. [186] found considerable movement in these planes, with a mean range of motion of 8.1° in the coronal and 7.9° in the transverse planes. An even greater range of motion in these planes was reported by Nester et al. [230] using a dynamic cadaveric model (15.3° and 10.0°).

Thus the ankle cannot be regarded as a simple hinge joint restricted to sagittal motion, but as multiaxial joint with triplanar movement.

2.3.2. The Subtalar Joint/Talocalcaneal Joint

The subtalar joint is situated below the ankle joint and formed by the articulations between the inferior surface of the talus and the superior surface of the os calcis. Articulation is at three sites: the posterior, middle and anterior talocalcaneal articulations. Cadaveric studies by Manter [190] and Hicks [123] among others demonstrated an axis of motion orientated upward, anteriorly and medially (Figure 2.7). The precise inclination of the subtalar axis was very variable between individuals and between researchers. It was theorized that the orientation of this axis would dictate movement patterns at the subtalar joint. Manter noted that movement about this single axis also led to longitudinal displacement of the os calcis. Further study of the morphology of the joint articular facets led him to the conclusion that subtalar motion was actually helical, likening the shape of the articulation to the surface of a screw [190]. Van Langelaan et al. [324] took this a step further in their fluoroscopic study of 10 cadaveric specimens with embedded markers, demonstrating not just a single helical axis, but a discrete bundle of helical axes. This has been confirmed in more contemporary work [284].

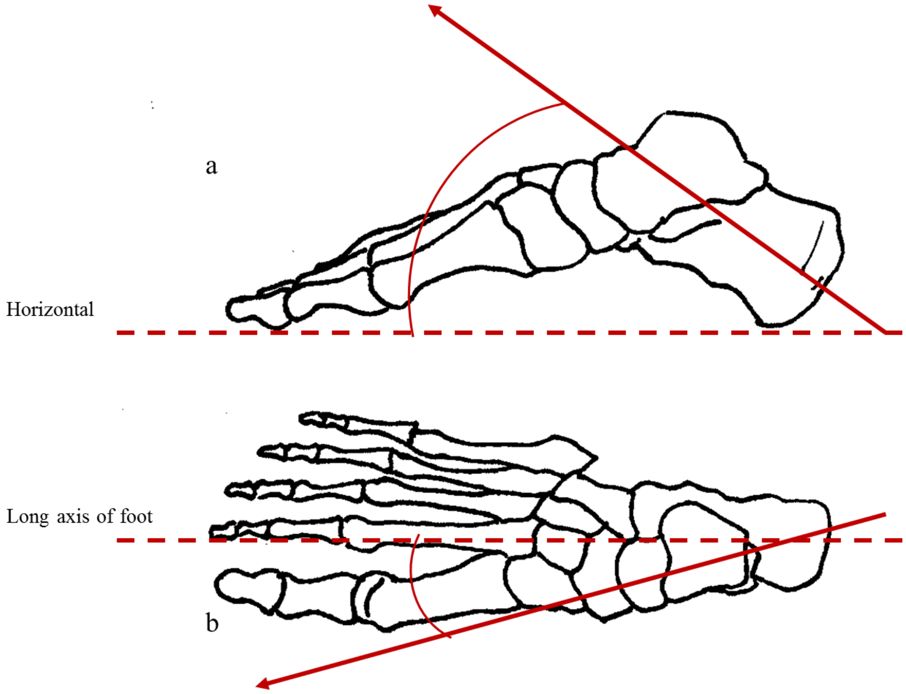


Figure 2.7 Orientation of subtalar axis. Angles a and b vary between individuals and research studies.

Motion of the subtalar joint is complex and is determined by the orientation of the axes, the shapes of articulations and soft tissue restraints, but clinically one usually assesses the subtalar joint by assessing motion of the heel in the coronal plane (inversion/eversion). Due to the oblique orientation of the axes there will also be some motion in the transverse and sagittal planes. Reported ranges of subtalar joint motion vary from 10° up to 50° [123, 138, 190, 199]. The American Medical Association has stated that 20° of eversion and 30° of inversion are normal for an adult [89].

The orientation of the axes of the ankle and subtalar joint allow these joints to act together as a 'torque converter', where transverse plane motion of the tibia is converted to motion of the hindfoot in the coronal plane. This function plays an important role in weight-bearing activities.

Contemporary views of the ankle and subtalar joints suggest that their motions are so closely related it is difficult to apportion specific motions to each joint. Historically the subtalar joint would have been held responsible for all coronal and transverse motions of the hindfoot, however both Lundgren et al. [186] and Nester et al. [230] demonstrated circumstances when the ankle contributed much more to transverse plane motion of the hindfoot than the subtalar joint. In some cases coronal plane motion of the ankle has even been greater than the subtalar coronal plane motion. The variability noted in the contributions of ankle and subtalar joint motions to hindfoot movement has led to the suggestion that instead of regarding each joint separately it is worth considering the hindfoot as a functional unit in its own right.

2.3.3. The Midtarsal Joint

The midtarsal joint, also known as the transverse tarsal joint or Chopart’s joint is actually formed of two articulations: the calcaneocuboid and talonavicular joint. Together they form a functional unit that allows adduction/abduction, a small amount of dorsiflexion/plantarflexion and some supination/pronation.

The talonavicular joint also forms part of the acetabulum pedis (Figure 2.8) [277]. This structure is a deep socket that receives the head of the talus and is formed by the anterior and middle articulating surfaces of the os calcis and navicular with various ligamentous contributions. This modified ball and socket joint intimately links the subtalar and transverse tarsal joints, with the orientation of the subtalar joint essentially dictating the position of the transverse tarsal joints. This relationship was defined by Elftman [87] who described a two-axis model of the midtarsal joint. He suggested that with the heel everted, axes of the talonavicular and calcaneocuboid joints align thereby allowing movement of the transverse tarsal joint. When the heel is inverted, these axes are no longer aligned serving to block motion at the transverse tarsal joint. This is the midtarsal locking mechanism.

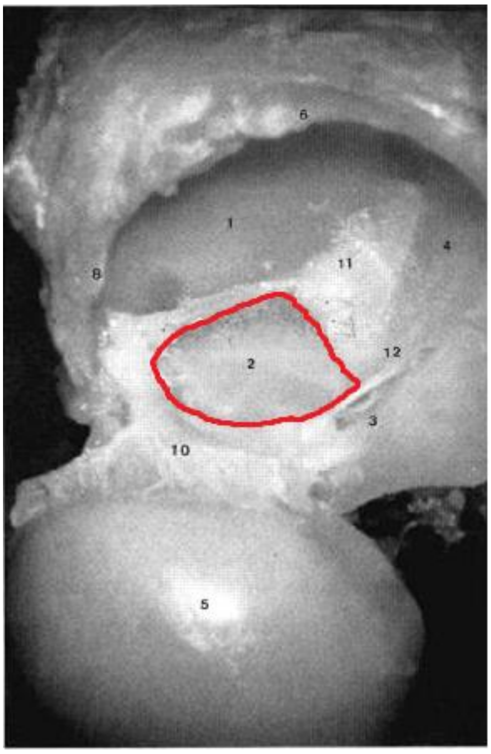


Figure 2.8 The acetabulum pedis, with anterior facet of os calcis highlighted in red (adapted from [277]). 1. Articular facet of navicular. 2. Anterior facet of the os calcis. 3. Middle articular facet of os calcis. 4. Medial wall, calcaneonavicular ligament. 5. Head of Talus enucleated from acetabulum pedis. 6. Roof, talonavicular ligament. 7. Tibialis posterior tendon. 8. Lateral wall. 9. Tendon sheath of flexor halluces longus. 10. Interosseus talocalcaneal ligament. 11. Pulvinar ligament. 12. Plantar ligament (component of ligamentary floor).

Elftman's model of the midtarsal joint is regarded by some as too simplistic. A modern view is that motion at the midtarsal joint is best described as having a single instantaneous axis of rotation, which changes orientation and position over time. Nester et al. [227] demonstrated a wide variation in midtarsal joint motion between subjects, with some subjects demonstrating predominantly coronal plane motion and others predominantly in the transverse plane. The variation in motion was attributed to differing orientations of the joint axis, as that observed in the subtalar joint. This so called planal dominance of the joint was felt to be the result of the wide variety in shapes of the articulating surfaces in the midtarsal joint.

Whilst there is no absolute consensus on the specific mechanisms of the midtarsal joint motion, a number of researchers have demonstrated the existence of midtarsal locking. Blackwood et al. [23] in their study on nine cadaveric specimens showed greater sagittal plane motion of the forefoot when the hindfoot was in a maximally everted position than when maximally inverted. Chi et al. [54] used an inclinometer and found that calcaneal eversion was associated with greater second metatarsal sagittal plane motion. These findings are consistent with the midtarsal locking mechanism.

Recent research studies have challenged the old paradigms of foot and ankle biomechanics, but these studies are not without their own limitations. The most important consideration is that these studies have very small numbers varying from 4 to 13 [186, 230]. These small samples lead to the question of whether the results are truly generalizable. Generalizability is particularly an issue in cadaveric studies as the foot and ankle specimens are usually from older subjects. For example the mean age of specimens in the study by Blackwood et al. [23] was 87.6 years old. One questions whether these findings can be applied to younger age groups. Whilst researchers attempt to load cadaveric specimens similarly to what occurs in vivo this is incredibly difficult and some would argue that even the most elaborate loading jigs fall short of recreating in vivo conditions. In the case of invasive in vivo research using bone pins, aside from any ethical issues, one questions whether a subject would walk normally with pins screwed, or metal balls embedded, into nine bones in their foot and ankle.

2.3.4. Soft tissue contributions to the foot and ankle

The stability and functions of the foot are only possible because of the intricate arrangement of soft tissue structures including joint capsules, ligaments, aponeuroses, muscles and tendons. An in depth description of all these structures is beyond the scope of this thesis, but awareness of certain key structures is required to understand how the foot and ankle complex fulfils its role in load-bearing activities.

Plantar fascia

The plantar fascia is a thick band of connective tissue that arises proximally from the tuberosity of the os calcis, runs across the plantar surface of the foot and distally coalesces with the plantar plates of the metatarsophalangeal joints. This structure plays an important role in stabilising the medial longitudinal arch of the foot and aiding propulsion.

Ligaments

Of the, over 100, ligaments in the foot the three of most importance in the context of flat feet, are the short and long plantar ligaments and the plantar calcaneonavicular or spring ligament (Figure 2.9). The long plantar ligament runs from the under-surface of the os calcis to the tuberosity of the cuboid and bases of the 2nd-4th metatarsals. Deep to this ligament runs the short plantar ligament which runs from the anterior calcaneal tubercle to the cuboid. The spring ligament is a broad band of thick fibres which runs from the anterior margin of the sustentaculum tali of the os calcis to the under-surface of the navicular. As well as connecting these two bones it also forms a sling to support the talar head in the acetabulum pedis. These plantar ligaments provide static support to the medial longitudinal arch (MLA) of the foot.

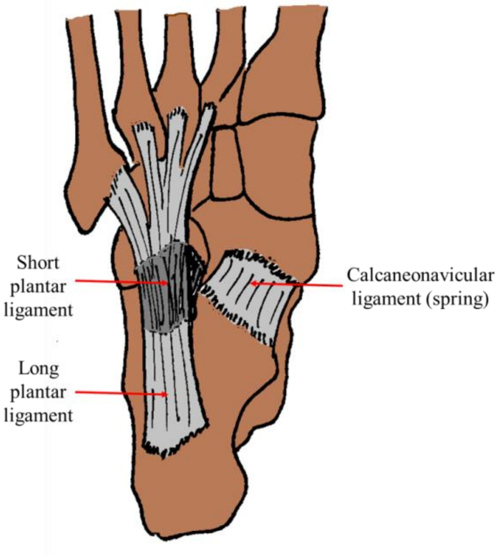


Figure 2.9 Important plantar ligaments for support of the MLA. Short plantar ligament deep to long plantar ligament.

Muscles

There are 19 intrinsic and 10 extrinsic muscles that control movement of the foot. The extrinsic muscles cross more than one joint and have multiple attachments, thus they often affect complex multiplanar movement at multiple joints. The orientation of the extrinsic muscle tendons in relation to the joint axes, in particular the subtalar joint axis will dictate resultant motion (Figure 2.10). In pathological scenarios where either the joint axis or the position of the tendons relative to the joint axis is altered, abnormal motions can result, which can interfere with the dynamic function of the foot and ankle. Table 2.1 summarises the extrinsic muscles and their main actions in the non-pathological setting. The intrinsic muscles have important roles as dynamic stabilisers of the foot. The roles of the extrinsic and intrinsic muscles in normal walking and in the context of a flat foot are discussed in subsequent sections.

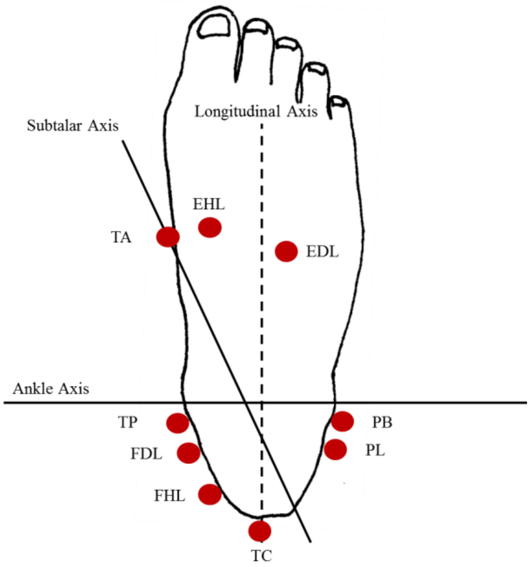


Figure 2.10 Orientation of extrinsic tendons around the ankle with respect to the ankle and subtalar joint axis. Extensor hallucis longus (EHL), Extensor digitorum longus (EDL), Peroneus brevis (PB), Peroneus longus (PL), Achilles tendon (TC), Flexor halluces longus (FHL), Flexor digitorum longus (FDL), Tibialis Posterior (TP) and Tibialis anterior (TA).

Muscle	Origin	Insertion	Action
Extensor Hallucis Longus	Interosseous membrane and middle half of anterior surface of the fibula	Distal phalanx of great toe	Extends hallux, dorsiflexes and inverts foot
Extensor Digitorum Longus	Anterior surface of the fibular and lateral portion of the interosseous membrane	Dorsum of the 2nd-5th toes via extensor expansions	Extends proximal, middle and distal interphalangeal joints, dorsiflexes foot
Peroneus brevis	Lower one third of the lateral surface of the fibula	Base of 5th metatarsal	Plantarflexes and everts foot
Peroneus Longus	Proximal 2/3rds of the lateral fibular surface	Tendon passes medial across the plantar surface of the foot and inserts on the medial cuneiform and base of 1st metatarsal	Plantarflexes first ray, plantarflexes and everts the foot
Peroneus Tertius	Distal part of anterior surface of fibula	Dorsum of the shaft of the 5th metatarsal	Everts the foot
Flexor Digitorum Longus	Middle half of posterior surface of the tibia	Bases of distal phalanges of 2nd-5th toes	Flexes the proximal, middle and distal interphalangeal joints of the 2nd-5th toes and Plantarflexes the foot
Flexor Hallucis Longus	Lower 2/3rds of the posterior surface of the fibula	Distal phalanx of the big toe	Flexes the metatarsophalangeal and proximal interphalangeal joints of the great toe; plantar flexes the foot
Gastrocsoleus	Posterior surface of tibia	posterior aspect of the os calcis via the tendo Achilles	Plantarflexion and some inversion of the foot
Tibialis anterior	Lateral upper surface of tibia	medial surface of the medial cuneiform and 1st metatarsal	Dorsiflexes and inverts the foot
Tibialis posterior	Interosseous membrane posteromedial surface of the fibula and postero-lateral surface of the tibia	Navicular, under-surface of medial cuneiform and 2nd-4th metatarsals	Plantarflexes and inverts the foot. Elevates medial longitudinal arch

Table 2.1 Extrinsic muscles of the foot and ankle; adapted from [193].

2.3.5. The arches of the foot

The foot is imparted further stability by the presence of longitudinal and transverse arches (Figure 2.11). The transverse arch is best visualized at the level of the midfoot and is composed of the three cuneiforms and cuboid. The longitudinal arch of the foot can be divided into the lateral longitudinal arch (LLA) and medial longitudinal arch (MLA). The LLA is formed from the lateral two metatarsals, cuboid and os calcis.

The medial longitudinal arch is formed from the medial three metatarsals, cuneiforms, navicular and os calcis. These arches are supported by inherent stability conferred by conforming shapes of the bones, numerous ligaments, intrinsic muscles and the tendons of extrinsic muscles.

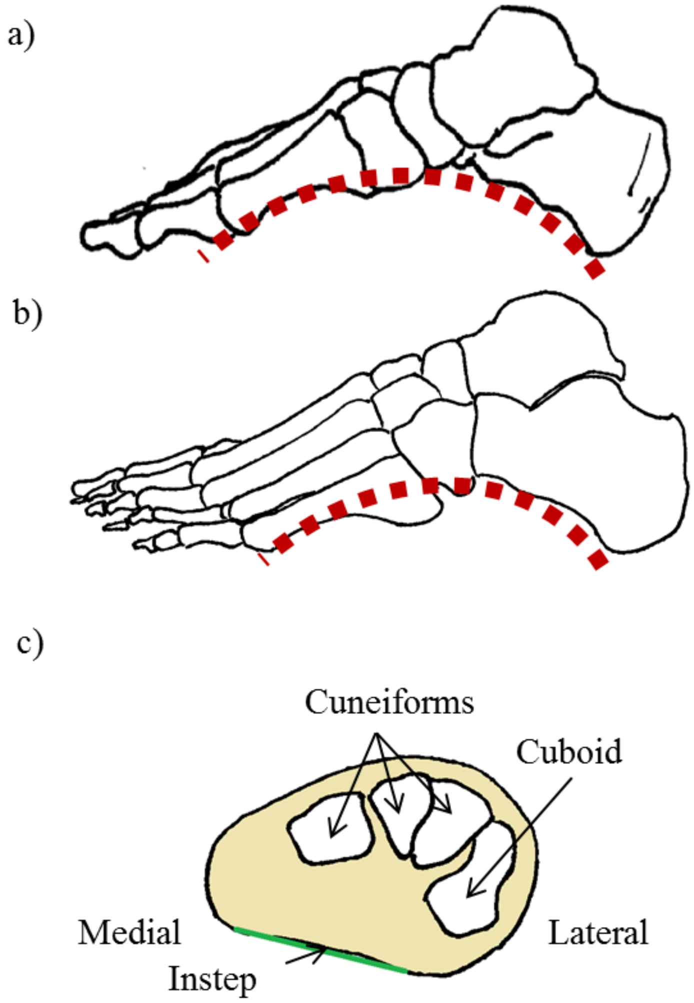


Figure 2.11 Arches of the foot. a) Medial longitudinal, b) Lateral Longitudinal, c) Transverse.

Of the three arches the MLA has the greatest biomechanical role in load-bearing activities. The MLA has an important role in stabilizing the foot and distributing load. The MLA via the plantar fascia also has a role in storing kinetic and potential energy as the foot is loaded in walking and then releasing this energy by elastic recoil to aid propulsion [158].

Hicks [124, 125] described the function of the MLA using beam and truss models. As the foot becomes loaded the MLA behaves as a multi-segment beam and weight is distributed between the forefoot and hindfoot (Figure 2.12 a). The dorsal surface of the arch is under compression and the plantar surface under tension. The tension is resisted by the strong plantar ligaments. With loading the beam ends are forced apart and as this occurs tension increases in the plantar fascia and the MLA acts as a truss (Figure 2.12 b). Tension in the plantar fascia prevents further collapse of the arch. In a normal plantigrade position the arch support is maintained by a combination of beam and truss mechanisms. In walking, as the hallux extends as the foot is pushing off, the truss mechanism takes over as toe extension tightens the plantar fascia via the windlass mechanism (Figure 2.13) [123]. The extent to which soft tissue structures support the arch has been assessed by Huang et al. [132]. In their study they undertook sequential division of soft tissue structures and assessed the change in arch height of cadaveric specimens under static loaded conditions. It was found that the greatest contribution to arch stability was given by the plantar fascia followed by the long and short plantar ligaments and then the spring ligament. Thordarson et al. [315] assessed dynamic arch height in cadaveric specimens and found a similar importance of the plantar fascia. The contribution of muscles was also assessed with the tibialis posterior muscle contributing most to arch support. Tibialis anterior, peroneus longus and peroneus tertius have also been demonstrated to provide dynamic support to the MLA [237, 315].

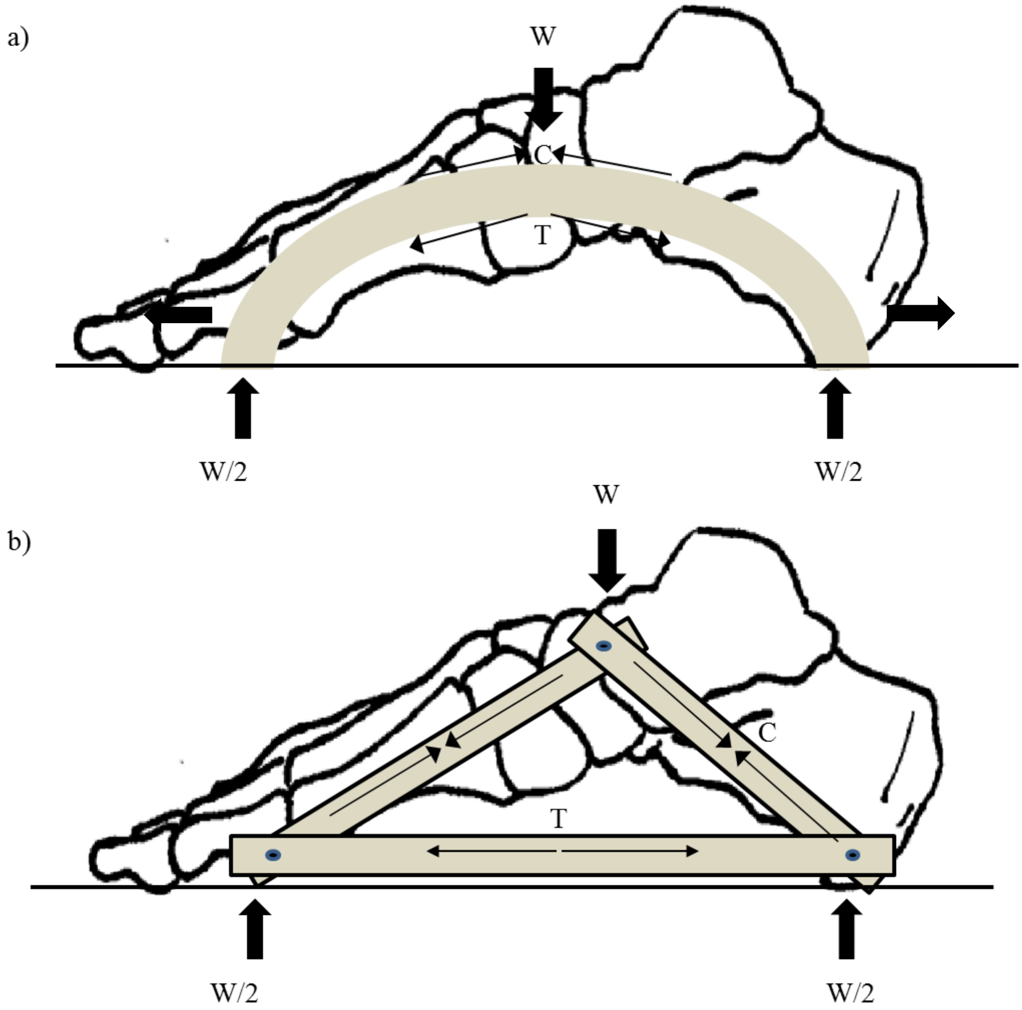


Figure 2.12 Models of the MLA. a) The beam model where loading of the MLA leads to compression (C) superiorly and tension (T) inferiorly, with spreading of the MLA. b) Truss model where the plantar fascia under tension acts as a tie-bar restricting arch deformation. Weight (W).

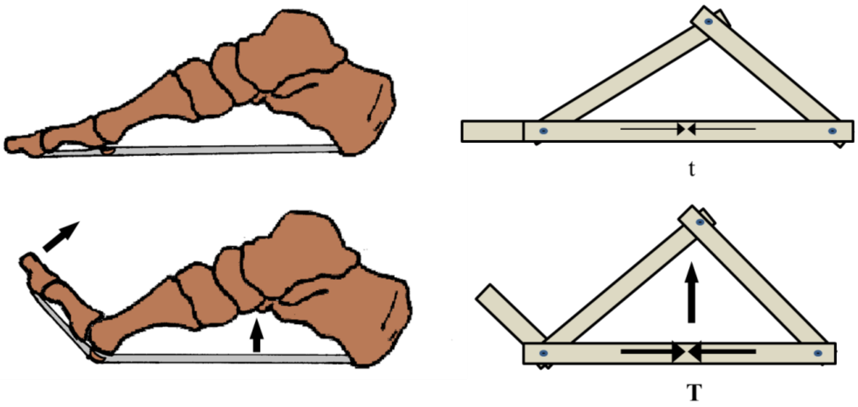


Figure 2.13 Windlass mechanism. Extension of the great toe increases tension (t,T) in the plantar fascia pulling on the 'truss' raising the arch.

2.4. Foot and ankle biomechanics during gait

Thus far the foot and ankle joints and associated motions have been discussed in relative isolation. In practice, activities such as walking rely on all joints in the lower limb, not just the foot and ankle, working in a coordinated fashion to achieve ambulation. Steindler recognized this and coined the term 'kinetic chain', describing it as a "combination of several successively arranged joints constituting a complex motor unit" [302]. Open kinetic chain movement is when the terminal segment moves freely and closed kinetic chain movement is when the terminal segment (foot in this case) meets resistance. When the movement of one segment is directly related to movement at another, usually adjacent, segment the movements are said to be coupled.

Walking is an activity that combines both open and closed kinetic chain movement. A complete gait cycle begins with one foot making contact with the ground and ends when that same foot contacts the ground again. The two phases of gait are stance and swing, with stance occupying just under two thirds of the gait cycle and swing occupying the remainder. The stance phase can be further subdivided into different phases (Figure 2.14), with each phase requiring different functional properties of the foot. The first stage of stance involves loading of the foot and requires the foot to be supple, able to absorb shock and adapt to uneven ground. As stance progresses the foot must transition to a more rigid structure to allow adequate push off and propulsion.

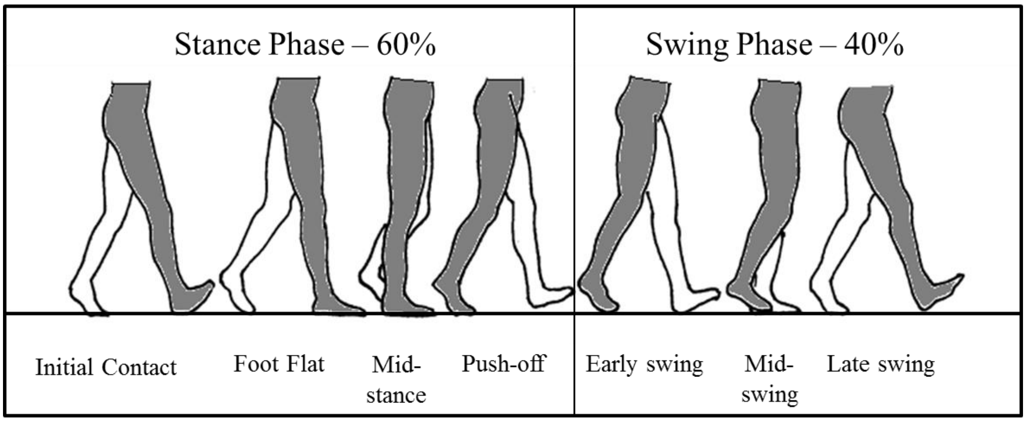


Figure 2.14 The Gait cycle. Demonstrated here for the right leg.

One of the favoured models of how the foot and ankle complex achieves these requirements is the ‘mitred-hinge’ model proposed by Inman in 1978 [137] (Figure 2.15). Inman stated that during the first half of gait internal rotation of the tibia, via ‘torque conversion’ by the acetabulum pedis, leads to hindfoot eversion, lowering of the medial longitudinal arch of the foot, abduction and pronation of the forefoot. As per Elftman’s description of transverse tarsal mechanics, hindfoot eversion leads to alignment of the calcaneonavicular and talonavicular axes resulting in a supple midfoot allowing the foot to absorb shock and adapt to uneven ground. During the second half of stance the tibia undergoes external rotation with coupled inversion of the hindfoot and locking of the midtarsal joint, forefoot adduction and supination to provide a rigid lever for propulsion. This is also referred to as re-supination. This process is facilitated by the insertion of the tendo Achilles being medial to the axis of the subtalar joint, thus the plantarflexion of the foot required in late stance by the gastrocsoleus also helps with hindfoot inversion (Figure 2.10).

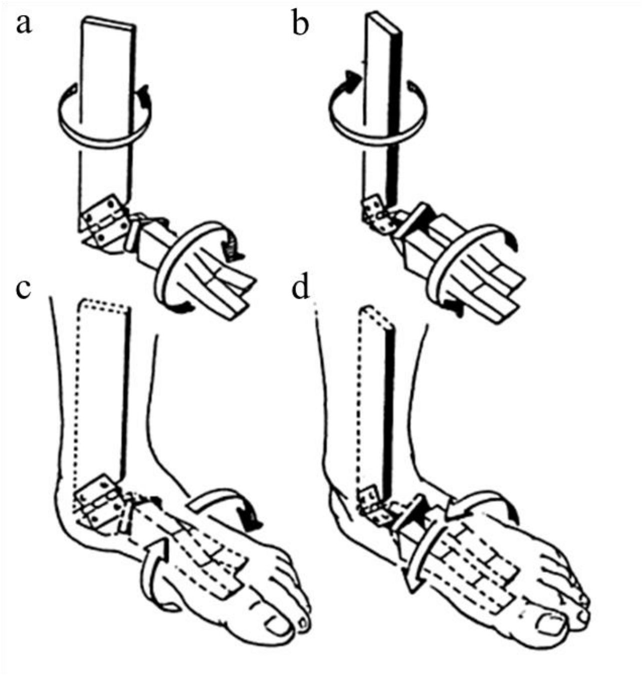


Figure 2.15 The ‘Mitred Hinge’ described by Inman. a) and c) External rotation of the tibia leading to ‘re-supination’. b) and d) Internal rotation of the tibial leading to foot flattening and a supple midfoot. Reproduced from [189].

This one size fits all model of foot function during gait is elegant, but does not highlight the variability of motion between subjects observed in more recent studies [186, 228, 230]. Nonetheless the model seeks to explain clinical observations, and in general terms is accurate about the role of tibial rotation in different phases of gait to allow the transition of the foot from a subtle to rigid structure.

To achieve such a complex movement like walking, considerable coordination is required between muscle groups. The role of intrinsic muscles in gait has been further characterized by Mann et al. [188] who using electromyography demonstrated these muscles to be the principal stabilisers of the foot in propulsion.

Extrinsic muscle activity in gait was well described by Bowker and Hall [31], and is summarized in Figure 2.16. Hunt et al. [135] further investigated extrinsic muscle activity in normal gait using surface electromyography. They demonstrated that greatest demand was placed on extrinsic musculature just before foot flat and after heel rise. The importance of tibialis anterior activity in restraining hindfoot plantarflexion and eversion was also further defined. Activity of peroneus longus in early stance was deemed to be critical in causing eversion after initial contact, and in late stance was felt to be important in stabilising the forefoot.

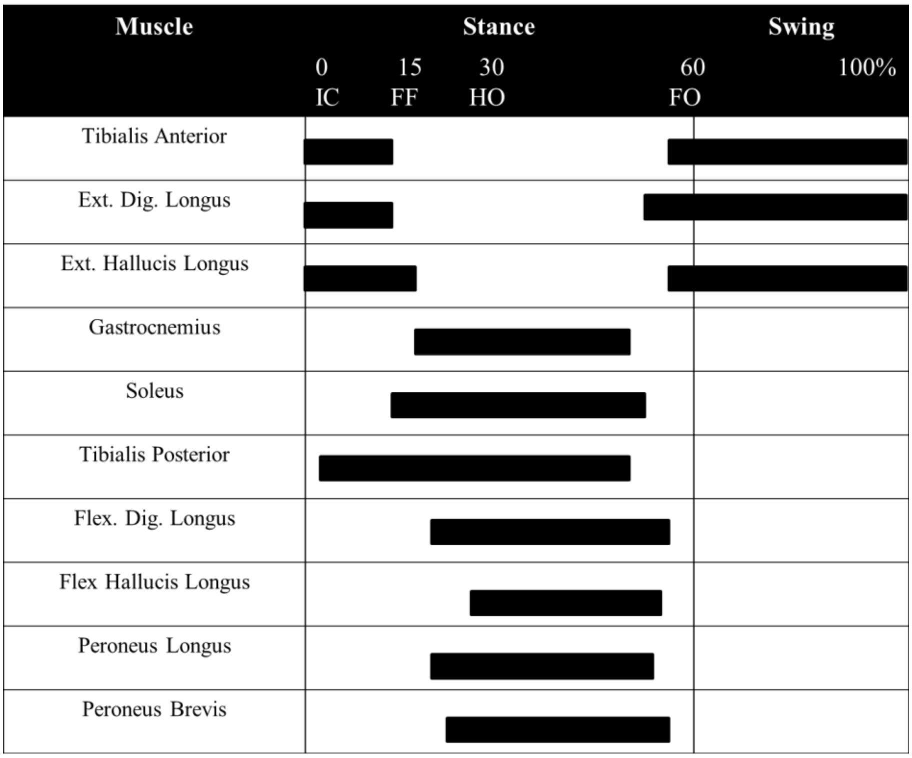


Figure 2.16 Extrinsic muscle activity in normal gait shown with black bands. Initial Contact (IC), foot flat (FF), heel off (HO), foot off (FO).

This description of normal gait relies on the complex interplay of bony articulations, ligaments, muscles and tendons. In the case of foot and ankle deformities, these finely coordinated motions can be disrupted potentially leading to symptoms and disability. Flat foot is one such deformity.

2.5. What constitutes a flat foot?

A flat foot in simple terms is a foot in which the medial longitudinal arch (MLA) is reduced in height or absent. The reduced MLA height is usually in combination with a degree of increased heel valgus. Other features that may be present, but are not pathognomonic include forefoot abduction, prominence of the navicular, and medial curving of the tendo Achilles. Flat foot is quite an all-encompassing term for a variety of conditions which demonstrate some similar morphological features. Whilst on first inspection feet may look similar, the underlying pathology can be dramatically different. As such flat feet can grossly be divided into three categories: rigid, flat and developmental.

2.5.1. Rigid flat foot

A rigid flat foot is one in which the MLA is reduced or absent when the patient is standing and remains so even when the big toe is extended, either passively by the examination or by the subject toe-standing. In otherwise healthy children and young adults this presentation is usually as a result of tarsal coalition. A tarsal coalition is when there is an abnormal fibrous or bony connection between two tarsal bones. This coalition disrupts hindfoot and midfoot mechanics leading to a permanently flat foot. The commonest sites of coalition are between the os calcis and navicular, and a talocalcaneal coalition at the middle facet of the subtalar joint [303]. Not all people with tarsal coalition become symptomatic, and if they do it is usually in the second decade of life as the coalition ossifies further [220]. When tarsal coalitions become symptomatic surgical intervention is commonly undertaken, either in the form of excision of the coalition or selective joint fusion.

In older adults flat foot may be as a result of adult acquired flat foot due to posterior tibial tendon dysfunction (PTTD). In this condition inflammation with subsequent degeneration of the tibialis posterior tendon can lead to development of a flat foot. Over a period of time this may demonstrate features of a rigid flat foot [225]. If symptomatic, this form of rigid flat foot often requires surgical intervention in the form of joint fusions [225].

2.5.2. Flexible flat foot

A flexible flat foot is one in which the arch is flat in the standing position, but extension of the hallux or standing on tiptoes results in the arch being recreated. This presentation contrasts to that of rigid flat foot as it is assumed that in these cases there is no specific underlying pathological process.

2.5.3. Developmental flat foot

All children are born with flat feet. The MLA of the foot continues to develop during the early years of life. There have been a number of studies assessing the development of the MLA in children [103, 241, 250, 258, 300, 332]. Estimates of the age of arch maturation are variable ranging from 2 to 13, but the majority of authors state that after the age of six the arch does not change shape significantly. Thus parents presenting with children below the age of six with suspected flexible flat foot should be reassured that MLA development is ongoing and just because their child is flatfooted currently does not mean they will be flatfooted in the future. In a proportion of children the MLA may not fully develop, and these individuals will subsequently be classified as having PFF.

Of the three forms of flat foot presented developmental does not require intervention and management protocols for rigid flat foot are reasonably well defined. The focus of ongoing controversy, and of this thesis, is flexible flat foot, specifically in children.

2.6. How to decide if a foot is flat or not?

A fundamental question which frequently arises is, ‘How do you decide whether or not a foot is flat?’ The earlier description of the morphological characteristics of PFF highlight the inherently subjective method of deciding foot posture based solely on visual inspection. How low an arch is too low? What amount of heel eversion is too much? Without a frame of reference and description of what is ‘normal’, it is difficult to know what ‘abnormal’ is.

To help the clinician distinguish between normal and abnormal foot postures in the 1970s, Root et al. [267] defined their biophysical criteria for normalcy. According to Root et al. normal alignment was achieved if:

1) The line bisecting the posterior aspect of the os calcis was in line or parallel to a line bisecting the posterior aspect of the lower leg.

2) The plane of the five metatarsal heads was perpendicular to the bisector of the posterior os calcis.

Root et al. [267] specifically stated that this normal alignment would occur when in a subtalar joint neutral position with the midtarsal joint locked. Subsequent work by

Smith-Orrichio & Harris [292] and McPoil et al. [203] demonstrated that these criteria were a bit too stringent and that only a small proportion of presumed normal subjects actually fulfilled Root's criteria. Even Root et al. [267] stated that feet which had minor deviations from the strict criteria they outlined could still be regarded as normal. Caillet [43] also provided criteria for normalcy, stating that a foot should be pain free, have a normal muscle balance, central heel and straight mobile toes. From a dynamic perspective it has previously been stated that even individuals regarded as normal can have variability in joint motion, in part due to the planal dominance of joints. It has also been suggested that functional aspects of the foot cannot necessarily be gleaned from static observation [50, 166]. Thus it seems that finding a gold standard of how normal feet and ankles should look and how they should function is an erroneous concept and much like people vary in anthropometric characteristics so will their joint motion vary [13].

Even though it has proven difficult to define what a normal foot is, many people have devised methods to define foot posture. A 2002 review of the literature came to the conclusion that there is no consensus on the best way of classifying flat foot posture [261]. The number of different foot posture classification methods has dogged research into flat feet. With over 40 different methods used in the literature, comparing and appraising results in the form of a systematic review and meta-analysis becomes problematic. In general terms classification methods can be sub-divided into those based on visual inspection, anthropometric measures, footprint analysis and radiographic angles. The inventors of any foot posture classification method are usually staunch advocates, but deciding which is best is difficult as few, if any, have any direct clinical relevance. Chapter 4 delves into the classification of PFF in further depth.

2.7. Epidemiology of PFF

The focus of this thesis is to define treatment indications for children with PFF thus it is a pre-requisite that the prevalence of the condition is known as well as aetiological factors associated with its development. Appraising this research is hampered by the heterogeneity of classification methods applied in the literature, but nonetheless it is possible to appreciate how common PFF is and gain some understanding of causes it.

2.7.1. The Prevalence of Flat Foot

There are numerous estimates of the prevalence of PFF in children. As previously discussed, developmental flexible flat foot is seen in the majority of toddlers, but once the arch has finished developing the estimates of PFF prevalence vary from 0.6% to 20% [27, 80, 85, 86, 106, 143, 215, 258, 268]. The studies that have investigated this, along with the method used for foot classification, are summarized in Table 2.2. Based on the findings of the studies presented in Table 2.2, 10-20% would seem a reasonable estimate for the prevalence of PFF in children. From the census of 2011 nearly 18% of the population in the UK was age 5-19 (9,393,826 people) [238]. Thus one would expect there to be potentially up to 1.2 million children between 6 and 16 years old in the UK with PFF.

Author, year	Children's age range (years)	Sample size	Classification method	Prevalence of PFF
El, 2006[86]	6-12	579	Footprints (AI)	17.2% moderate or severe 82.8% normal or mild
Echarri, 2003[85]	3-12	1851	Footprints (SI, CA, CSI)	3-4 y, most children have PFF 5-12 y, 10-20%
Rao, 1992[258]	4-13	2300	Footprints (Bespoke)	6.7% Shod (8.6%) vs Unshod (2.8%)
Rose, 1985[268]	5-12	237	Physical examination, Footprints (VI)	9.9%
Morley 1957[215]	5-11	318	Physical examination, Footprints (Bespoke)	10% approx.
Didia, 1987[80]	5-14	990	Footprints (CI2)	0.60% bilateral 2.2% unilateral
Craxford 1984[68]	1-13	100	Plantar pressure	18%
Bordin, 2001[27]	8-10	243	Photography	16.4%
Garcia-Rodriquez, 1999[106]	4-13	1181	Footprints	2.7%
Jerosch, 1998[106]	10-13	345	"Standardized protocol"	19.1%

Table 2.2 Studies citing prevalence of PFF. AI = arch index, SI = Staheli index, CA = Clarke's angle, CSI = Chippaux Smirak index, VI = valgus index, CI2 = contact index 2.

2.7.2. Aetiology of PFF

The reasons why some children have PFF and others do not have still not been completely elucidated, although some associations have been observed.

Numerous studies have demonstrated a correlation between PFF and body weight [82, 85, 250, 313, 330]. In general it is agreed that 'fatter' feet are associated with 'flatter feet'. This would seem reasonable as if the MLA is subjected to more load one would expect greater deformation. In a study of pre-school children Pfeiffer et al. [250] demonstrated significant differences in the prevalence of PFF on the basis of bodyweight. Sixty two percent of obese children had PFF, compared to 51% of overweight children and 42% of children of normal weight. Significant correlation between PFF and body weight in adolescent children was found by Villaroya et al. and Dowling et al. [82, 330]. The majority of studies that demonstrate the association between body weight and PFF use footprint indices as their classification tool. Increased body weight can lead to increased fat deposition around the foot, and thus what appears as a flatter foot may only be due to increased plantar fad pad instead of true lowering of the arch. In an ultrasound study to investigate this, Riddiford-Harland et al. [265] demonstrated that whilst obese children did have an increased medial plantar fat pad which could affect footprint measurement, they also had lowered MLA height.

There is also a strong association between flexibility and PFF. On the basis that the MLA is supported by soft tissues, it seems logical that the more flexible these soft tissues, the flatter the foot when loaded. Flexibility is usually evaluated using the Beighton scale which is a nine point scale assessing specific joint motions [16]. A score of four or more would be classified as having ligamentous laxity or hypermobility. El et al. [86] investigated foot posture and flexibility in a sample of 146 children and demonstrated that hypermobile children had almost double the prevalence of PFF than non-hypermobile children (27.6% vs 13.4%). In a study of preschool children by Lin et al. [180] moderate and severe flat foot was associated with increased joint laxity. In connective tissue disorders associated with joint hypermobility, like Marfan’s syndrome, PFF is a common presentation (59% quoted by Laffargue et al. [170]). When there is a pathological process leading to hypermobility and flat foot these patients cannot be grouped with ‘normal’ children with PFF for investigation or analysis as they essentially come from different populations. This rule has at times been ignored in the literature, making it nearly impossible to interpret the results [29, 276].

Lower limb alignment may also be a factor affecting foot posture with Lin et al. [181] demonstrating an increased incidence of PFF in children with knee valgus than controls. In the transverse plane, increased femoral anteversion and increased external tibial torsion have been associated with the presence PFF, although it is not clear why such an association exists [291, 356].

Heritability has an important role to play in the aetiology of PFF, and often when a child presents with PFF the parents report that they also have flat feet [221]. The exact extent of this hereditary component has not been assessed. Aside from parental foot morphology, there is a genetic component to flexibility which has already been implicated in PFF [3, 93]. Ethnicity also plays a role with there being a higher incidence of PFF in black people compared to Caucasians [221].

An inherited propensity for a flat foot posture may be related to the actual morphology of the foot and ankle joints. In their seminal 1948 paper, Harris and Beath [120] presented a study on tarsal bone morphology by Cates from Toronto University. Cates, using cadaveric specimens, found a wide variety of relationships between the talus and the os calcis, which in turn could affect the way that the head of the talus was supported. Both ‘weak’ and ‘firm’ subtalar joints were described, with the ‘weak’ giving poor support anteriorly to the talar head allowing it to move plantar-medially, and pushing the os calcis into valgus. The ‘firm’ subtalar joint provided better support to the talus, especially anteriorly. It was hypothesized that a weak subtalar joint would lead to a flat foot, whereas a firm subtalar joint would not. This hypothesis was based on the morphology of disarticulated specimens, and was not demonstrated in vivo.

Since the osteological findings of Cates few researchers have investigated the morphological variations of the subtalar joint in the context of foot posture. Bunning et al. [39] were the first to classify variations in the articulating facets of the os calcis in their investigation of adult and foetal specimens. They demonstrated three variants of articulating surface of the os calcis (Figure 2.17). In 1987 Bruckner [36] provided a different classification on the basis of her investigation of 32 dry adult calcanei and the corresponding talus for 26 of them. She demonstrated four general morphological varieties of the subtalar joint (Figure 2.18). Instead of always having the three articulations of anterior, middle and posterior, it was noted that in some cases the anterior and middle articulations were joined and in others the anterior articulation was missing. The clinical implications of this were discussed, but again as it was unclear what foot posture the cadaveric subjects would have had in-vivo, only speculations could be made. Barbaix et al. [10] used a hybrid of Bunning et al. [39] and Bruckner's [36] classifications of the subtalar joint to investigate 256 bone specimens, and again demonstrated significant variability in subtalar joint morphology. They suggested that the orientation of the articulations and the surface contour may predispose some individuals to subtalar instability leading to ankle inversion injury. The role of subtalar morphology in foot and ankle presentations in-vivo is still unknown.

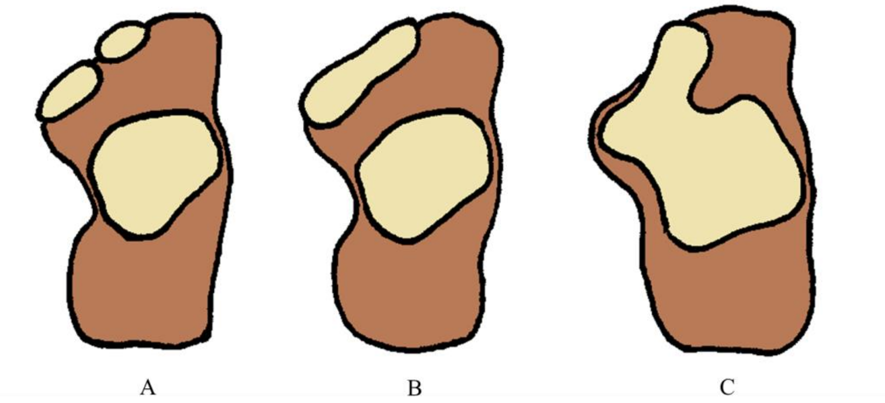


Figure 2.17 Variations in os calcis articulating surface morphology as described by Bunning [39].

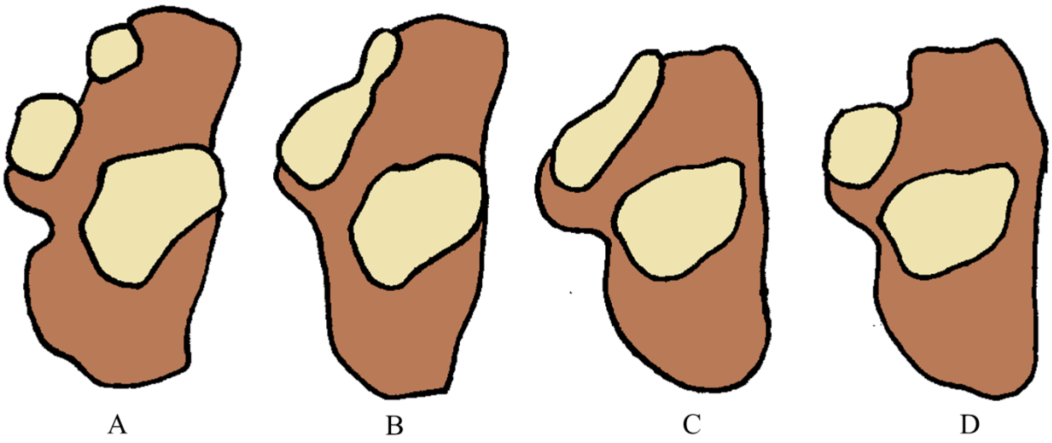


Figure 2.18 Variations in os calcis articulating surface morphology as described by Bruckner [36].

2.8. What's the problem with flat feet?

Even though flexible flat feet are common and there are a number of aetiological factors associated with them, the question of clinical importance still remains. What evidence is there that flat feet cause any problem whatsoever? In this section current research perspectives on the clinical impact of flat feet are discussed as well as the potential mechanisms through which flat feet may cause problems. As there is a relative paucity of evidence in the paediatric population this section will also include relevant literature from the adult flexible flat foot population. As was found with the epidemiology of PFF, the water is again muddied by the numerous classification methods used by researchers.

2.8.1. Clinical associations

There is currently a schism in prevailing views about whether PFF is problematic or not. Some clinicians believe that PFF is just a normal variant and seldom requires intervention, whereas others believe that it could cause significant impairment and disability to some children. There has long been an association of PFF with back and lower limb symptoms, but there is sparse epidemiological evidence to support this. The studies cited in Table 2.2 all provide estimates of the prevalence of PFF, but none actually quote the proportion of children with PFF that get any form of lower limb musculoskeletal symptoms. A number of authors have suggested that PFF is seldom symptomatic, but this has not been backed up by objective evidence, and is merely based on personal experience [106, 221, 297, 318]. Bertani et al. [20] state that of children that have PFF, 10% require treatment to avoid secondary deformity; this statement was again not based on published evidence. Benedetti et al. [18] attempted to quantify the prevalence of symptoms and functional limitations caused by PFF. In their sample of 54 children between the ages of 10 and 14 they demonstrated that 65.3% had symptoms and 68.3% had functional limitation. This study, however, has the inherent flaw of selection bias, as it is more likely that children with problems will present to healthcare services, whereas asymptomatic children are more likely to remain in the community. Tudor et al. [318] investigated motor function of 218 children between 11 and 15 and found no association between foot posture and functional limitation. They did however report a 28% dropout rate, and it is not known whether this represented children who struggled to complete the activities.

This could have significantly biased their results. Currently it is unclear what proportion of children with PFF actually suffer from foot and ankle symptoms and get functional limitation. Aside from not knowing what proportion of children has symptomatic PFF there is no literature describing how severe the symptoms can be and what affect they could have on the child's quality of life. In the context of defining treatment protocols for PFF it is imperative to know what the actual impact on the child could be.

In the adult population the main epidemiological evidence comes from Harris and Beath [120]. In their study of 3619 Canadian army recruits the incidence of flat foot was 23%. Of this group 26% were highlighted as having 'hyper-mobile flat-foot with short tendo achillis'. Harris and Beath [120] stated that this presentation of flat-foot was associated with symptoms and disability, whereas other flat feet were generally asymptomatic.

Aside from non-specific aches and pains, flat feet have been associated with some specific foot and ankle pathologies. Tarsal tunnel syndrome is a nerve entrapment neuropathy of the posterior tibial nerve as it passes through the tarsal tunnel on the medial side of the ankle. Numerous authors believe that increased heel valgus observed in flat feet leads to stretching of the posterior tibial nerve, causing worsening neuropathy [66, 104, 163]. Stretching and increased tension in the posterior tibial nerve has been demonstrated by Daniels et al. [72] in a cadaveric flat foot model.

Flat feet are a proposed etiological factor for plantar fasciitis. Werner et al. [337] demonstrated in a study of 666 assembly plant workers that individuals with a flat foot were 4.2 times more likely to have plantar fasciitis. Prichasuk et al. [255]

demonstrated that individuals with plantar heel pain had flatter feet based on measurements taken from weight-bearing lateral radiographs. Pohl et al. [252] undertook a case control study of 25 female runners with plantar fasciitis and found that flatter feet, based on footprint measures, correlated with symptoms.

The relationship between flat feet and lower limb stress fractures is less clear. Giladi et al. [107] investigated 295 military recruits and found that flat feet were potentially protective for lower limb stress fractures. Subsequently in a follow-up paper from his group it was demonstrated that army recruits, with flat feet were more likely to get metatarsal stress fractures, whereas high arched recruits were more likely to get tibial stress fractures [289]. Other studies have found no relationship between arch height and stress fractures [19, 38].

It is proposed that in closed kinetic chain activities any foot or ankle deformity could lead to problems at more proximal joints. There have been numerous clinical studies in adults investigating the role of foot posture on the proximal joints. In a retrospective study of 97,279 military recruits, it was found that moderate to severe pes planus, as diagnosed by visual inspection, was associated with a doubled incidence of intermittent back pain and almost doubled incidence of anterior knee pain [167].

Investigation of the Framingham foot study cohort demonstrated that adults with planus foot posture as measured using footprint based indices had higher odds of getting knee pain (odds ratio (OR) 1.57) [266]. Gross et al. [114] also showed that flatter feet were associated with increased cartilage loss in the medial compartment of the knee. In this cohort women with planus foot posture also had higher odds of getting low back pain (OR 1.51) [209].

A flat foot posture has also been implicated in anterior cruciate ligament injury, patellofemoral syndrome, iliotibial band syndrome and medial tibial stress syndrome in runners [4, 12, 152, 260].

Thus there is some evidence that foot posture may relate to symptoms at proximal joints, but the majority of the research in this field focuses on adult or late adolescent populations. There is currently little work on how PFF may affect the proximal joints of children.

2.8.2. How could flat feet cause symptoms and functional impairment?

Even with the association between flat feet and back and lower limb symptoms, the pathomechanics of how flat feet cause symptoms and functional impairment have not been fully elucidated. A number of theories have been proposed, and these are reviewed in this section along with the currently limited evidence supporting them.

Foot and Ankle Symptoms

One symptom that children with PFF report is fatigue around the foot and ankle [18, 129]. This may be explained in part by the electromyographic observations of Mann et al. [188]. As well as demonstrating that the intrinsic muscles of the foot were the primary stabilizers of the foot in propulsion, they also found that increased activity of the intrinsic muscles was required to stabilize the midtarsal and subtalar joint in individuals with flat feet. Gray and Basmajian [111] demonstrated increased extrinsic muscle activity in flatfooted individuals, particularly with greater activity of peroneus longus and tibialis anterior. Murley et al. [223] also noted increased activity of tibialis anterior in flatfooted individuals, but actually decreased activity in peroneus longus. Increased activity of the intrinsic and extrinsic muscles in flatfooted people could potentially lead to fatigue during activity. Muscle fatigue may also impair support of the MLA putting extra strain on the plantar ligaments and plantar fascia. Indeed Headlee et al. [122] showed that subjects who had their intrinsic foot muscles fatigued had increased inferior displacement of the navicular when weight-bearing. This was suggestive of impaired dynamic support of the arch by the muscles.

Other pathomechanic theories are based on the effect of flat foot on the joint couplings described previously in the “mitred hinge” model of gait proposed by Inman [137]. This model relies on the smooth transition from an everted hindfoot with a flexible flattened arch to an inverted hindfoot with a rigid arch to allow propulsion. In cases of PFF with excessive hindfoot valgus it is hypothesized that the midtarsal locking system can be disrupted. If the hindfoot does not invert adequately and the midtarsal joint does not lock, the ‘push off’ occurs not on the desired rigid structure, but on a supple and less stable structure. If the active plantarflexors in late stance are working with a flexible lever then some of the energy generated by them will be absorbed in the arch and not used in propulsion. The property of the foot that is so important in early stance is now counterproductive in late stance. The unlocked midfoot is not designed to withstand the forces exerted by push-off and over time it is believed that this could lead to strain, with subsequent attenuation of the capsuloligamentous structures, and deformity. The deformity of midfoot break is often observed in individuals with severe flat foot. Angin et al. [6] used ultrasound to demonstrate thinning and attenuation of the plantar fascia in adults with flexible flat feet.

Myerson [225] hypothesized that with excessive heel valgus the line of action of the tendo Achilles could also change and move laterally to the axis of the subtalar joint (Figure 2.10). If this occurs the gastrocsoleus would become an evertor as well as a plantarflexor. Active plantarflexion is required in late stance, as well as inversion of the hindfoot. If gastrocsoleus acts to evert the hindfoot then locking of the midtarsal joint would be blocked further.

Excessive hindfoot valgus also allows the tendo Achilles to take a ‘short cut’ around the heel and slacken. Muscles exhibit a strong force-length relationship thus slackening of the gastrocsoleus muscle-tendon unit may further impair the ability of the muscle to effect ‘push off’. The tightening of the plantarflexors first observed by Harris and Beath, and subsequently by many others could be as a result of the muscle adapting to restore its force-length relationship [119, 120, 129, 221, 354]. Once this adaptive change occurs it becomes increasingly difficult for the hindfoot to invert in late stance, and the deformity can be perpetuated. Blackman et al. [22] have shown that simulated Achilles contracture in cadaveric specimens gives rise to worsening flat foot deformity, particularly with increasing the severity of hindfoot eversion, arch depression and foot abduction. A restriction of ankle dorsiflexion may lead to increased compensatory forefoot dorsiflexion to attain a plantigrade foot; this may in turn contribute to a midfoot break [129]. This would result in further stretching of the midfoot capsuloligamentous structures leading to discomfort or pain. The excessive hindfoot valgus may also lead to lateral ankle pain if the os calcis impinges on the lateral malleolus.

These pathomechanic theories have been tested using clinical gait analysis, to investigate joint motion (kinematics) and forces causing joint motion (kinetics). Hunt et al. [134] and Levinger et al. [179] demonstrated that flatfooted individuals had reduced forefoot adduction in late stance compared to normal subjects. As adduction is a key component to the re-supination process in late stance both research groups suggested that reduced adduction demonstrated an impairment of this process, which in turn could cause functional problems. Increased internal rotation of the hindfoot was displayed by flatfooted individuals studied by Levinger et al. [178], and this was also deemed to demonstrate an altered coupling relationship with the tibia. Hunt et al. [134] and Hosl et al. [129] also showed reduced hindfoot dorsiflexion during gait in flatfooted individuals, with Hosl et al.'s subjects demonstrating compensatory increased dorsiflexion of the forefoot and mobility of the hallux. Twomey et al. [319] observed increased forefoot relative to midfoot supination during stance, in children with asymptomatic PFF compared to normal arched children, but the significance of this with respect to pathomechanic theories was not commented on. Hosl et al. [129] also found reduced ankle power generation by children with symptomatic PFF compared to normal children, suggestive of impaired 'push-off'. Hunt et al. [134] demonstrated an increased peak ankle plantarflexor moment in late stance in their cohort of flatfooted individuals, but this finding was not felt to have much clinical significance. It is unclear which specific methods were used to calculate the ankle moments.

Increased hindfoot coronal plane motion in flat feet has been observed in a number of studies, with the majority of studies relating a flat foot posture with increased hindfoot eversion [59, 129, 130]. Static hindfoot eversion has also been shown to have a moderate correlation with peak hindfoot eversion in normal walking [133]. Not all studies are in agreement with these findings, with Cobb et al. [63] observing reduced hindfoot eversion in pre-swing in the flat foot group. This is in conflict with the majority of the research findings and the prevailing views of flat foot biomechanics.

Table 2.3 summarises the relevant studies comparing walking foot and ankle biomechanics between normal and flat feet. It should be noted that the majority of these studies were undertaken in adult populations. The sample sizes are also small which impacts on the statistical power to detect small and moderate effect sizes. Another criticism is that just because flat feet demonstrate different biomechanics to normal feet, it does not necessarily mean that these cause or will cause problems. Of all the papers summarized in Table 2.3 only two assessed function in the context of symptoms [129, 134]. Of those, Hunt et al. [134] compared symptomatic flat feet with normal feet, but without having an asymptomatic flat foot group it is impossible to know whether groups' differences were because of foot posture, symptoms or both. Hosl et al. [129] pooled feet from subjects in their analysis, but did not compensate for this in their statistical analysis. This runs the risk of artificially inflating sample size leading to erroneous results [208].

Aside from the Hosl et al. [129] study there are a limited number of studies investigating either the structure or function of flexible flat foot in the context of symptoms. Moraleda et al. [213] undertook a retrospective review of 145 children with PFF to assess the relationship between bony alignment as assessed in weight-bearing radiographs and the presence of symptoms. The main finding of their work was that children with symptomatic PFF had greater medial uncovering of the talar head in the transverse plane as assessed using the talonavicular coverage angle. Yan et al. [354] compared the weight-bearing radiographs of 50 children with symptomatic PFF to 50 children with asymptomatic PFF and demonstrated similar findings to Moraleda et al. [213]. In the clinical assessment of the study participants by Yan et al. [354] it was noted that shortening of the plantarflexors was only observed in children with symptomatic PFF, but not in all. Whilst the significant findings of both these studies only relate to static alignment of the foot it is suggested by the authors that these findings would affect the dynamic function of the foot. It is believed that lateral deviation of the navicular with respect to the talus affects the shape of the acetabulum pedis thereby impairing load transmission from the hindfoot to the forefoot through the talonavicular joint.

Pehlivan et al. [249] also undertook radiographic comparison of symptomatic and asymptomatic flexible flat feet in young adults, but only measured angles in the sagittal plane. They demonstrated that the talus was more plantarflexed in relation to the forefoot in symptomatic flat feet compared to asymptomatic flat feet. Their study however only included 28 subjects and feet were again pooled without statistical compensation. The significance of the radiological findings with respect to generation of symptoms was not discussed.

Proximal Joint Symptoms

With regards to proximal joint symptoms, it is hypothesized that excessive hindfoot eversion throughout the gait cycle would lead to excessive internal rotation of the tibia. This in turn would lead to increased rotational shear on the articular contact areas in the knee as well as increasing articular contact in the patellofemoral joint. A coupling between hindfoot position and tibia rotation has frequently been demonstrated [137, 226, 229, 233]. A further coupling between the hindfoot/tibia complex and the hip joint has been demonstrated by Souza et al. [296], with a more everted hindfoot leading to more internal rotation of the hip during stance. Increased internal rotation of the hip may in turn detrimentally alter the lever arm of the hip abductors, and increase the Q-angle at the knee further overloading the lateral patellar facet. In the case of the lumbar spine it has been demonstrated that arch height can affect the magnitude of accelerations during running, pelvic alignment and gluteal and erector spinae muscle activity [21, 160, 239].

Author	Participants number (age (yrs.) ± standard deviation)	Foot type classification method	Main findings
Cobb et al.[63]	NF = 11 (25.2 ± 3.2) FF = 11 (24.5 ± 6.1)	Arch height index and relative arch deformity	FF vs NF Reduced midfoot abduction during midstance Reduced hindfoot eversion excursion during pre-swing Greater rearfoot inversion excursion during pre-swing
Houck et al.[130]	NF = 7 (22 ±1) FF = 14 (22.2 ± 1.3)	Classified as FF is > 10° forefoot varus and navicular drop ≥ 10mm	FF vs NF Greater rearfoot eversion during early stance Greater first metatarsal dorsiflexion
Hunt et al.[134]	NF = 15 (25 ± 5) FF = 18 (26 ± 7) All subjects symptomatic	Visual inspection	FF vs NF Less midfoot adduction during toe off Less midfoot range of motion Increased plantarflexor moment at push-off Increased invertor moment at foot flat
Levinger et al.[179]	NF = 10 (24.3 ± 8.7) FF = 9 (20.1 ± 1.3)	Groups assigned using weight-bearing radiographic angles	FF vs NF Greater forefoot plantarflexion in late stance Greater forefoot abduction in midstance Greater rearfoot internal rotation in late stance Less forefoot adduction peak angle in terminal stance
Hosl et al.[129]	NF = 11 age-matched ASFF = 21 (11.0 ± 2.6) SFF = 14 (11.6 ± 2.0)	Visual inspection	ASFF + SFF vs NF Increased hindfoot eversion Reduced hindfoot dorsiflexion Increased forefoot mobility Hypermobile hallux SFF vs NF + ASFF Reduction in positive ankle energy in propulsion
Twomey et al.[319]	NF = 25 (11.1 ± 1.2) FF = 27 (11.2 ± 1.2)	Footprint index, arch index and navicular height	FF vs NF Increased forefoot supination at initial contact, maximum and minimum values

Table 2.3 Findings summary of relevant studies comparing flat foot (FF) and neutral foot (NF) biomechanics.

The association of flat feet with increased internal rotation of the tibia has been shown by Levinger et al. [178] in their study of people with medial compartment osteoarthritis. It however was unclear whether the foot posture was a potential cause of knee problems or actually a compensation for varus malalignment of the knee to achieve a plantigrade foot. Nigg et al. [233] investigated the effect of arch height on the running biomechanics of 30 individuals. Arch height was not related to maximal internal rotation of the tibia, however, arch height was noted to affect the coupling and movement transfer between hindfoot eversion and tibial rotation. Flatfooted individuals were noted to have reduced knee adduction, but greater flexion in running, by McClay et al. [196]. Significantly increased mean peak angular velocities around the knee were found. This led to the authors' suggestion that flat feet could put runners at increased risk or injury. Shih et al. [286] investigated lower extremity kinematics in children with PFF and found no significant differences compared to children with normal arches. Twomey et al. [320] demonstrated increased external hip rotation in children with PFF contradicting prevailing pathomechanic theories.

In conclusion a flat foot posture appears to having significant clinical associations with lower extremity problems. There are also sound biomechanical theories as to why people with a flat foot posture may develop symptoms. What is lacking is the link between these two statements, as essentially there is very little evidence relating foot posture and biomechanical functioning in the context of the presence or absence of symptoms. Whilst a number of research studies have been discussed in this section when it comes to flexible flat feet in children there are only three studies which describe differences between asymptomatic and symptomatic PFF. Two studies make the not too insignificant leap of inferring dynamic function from static findings, and the final study is hampered by a small sample of symptomatic flat feet as well as other methodological issues.

2.9. Treatment for children with PFF

A number of guidelines have been developed to help clinicians with the management of PFF. The published guidance is very general, partly reflecting the lack of evidence about the pathogenicity of PFF. Of note, guidelines have been developed by Clinical Practice Guideline Pediatric Flatfoot Panel of the American College of Foot and Ankle Surgeons [119]. They suggest that treatment for PFF should be considered if the child is symptomatic, and should start with non-operative measures like stretching of the plantarflexors and orthotics. If these treatments are not adequate, then surgical intervention can be considered. They also suggest that in certain cases children with asymptomatic PFF should be periodically monitored for progression to symptomatic PFF. Indications for monitoring asymptomatic PFF include tightness of the Achilles, excessive heel eversion and talonavicular joint instability. Exactly what constitutes excessive heel eversion and talonavicular joint instability is not discussed. They also state that some children with asymptomatic PFF may benefit from treatment with either stretching or orthoses. In summary children with both asymptomatic and symptomatic PFF may benefit from treatment! It is unclear whether this guideline provides the clinician with much help when planning management. Evans [92] has also provided clinical guidelines using a traffic light system. A red light represents children with symptomatic PFF who require treatment, amber for asymptomatic non-developmental PFF who require monitoring for progression and green for developmental flat foot who can be left alone. These guidelines are again vague, and one would hope that with more evidence about how PFF relates to symptoms, it may be possible to refine these guidelines to help the clinician further.

At a local level, a recent audit of physiotherapist and orthotist practices for children with PFF demonstrated a wide variety of approaches with little consistency in treatment methods [159]. This potentially highlights the non-specific nature of published guidelines, or that the evidence base does not always sway personal clinical opinions and experience. This was particularly highlighted in an informal survey of the local orthotic department staff members who all stated that they had prescribed orthoses for children with knee, hip and back pain; even though there is little evidence to support this practice.

The aim of this thesis is to evaluate children with PFF and help further define treatment indications for children with flexible flat feet. To successfully achieve this, it is important to consider three key questions:

1) Is PFF linked to symptoms and functional impairment, and if so to what extent?

This fundamental question was considered earlier in this chapter, and whilst there are a number of differing views, the general consensus is that PFF could cause foot and ankles problems in some individuals. Whether it causes problems higher up the kinetic chain is unclear. There is no literature quantifying the severity and extent of problems that individuals may get. Historically, children have been labelled as either symptomatic or asymptomatic, but this gives no indication on symptom severity. Without knowing the potential clinical impact of PFF on a child it is very difficult to define treatment guidelines. If symptoms associated with PFF were very mild, then watchful waiting or a cautious approach to management may be appropriate. However if PFF was leading to significant impairment and disability, then a much more aggressive approach may be suitable. It is also unclear whether children with asymptomatic PFF are truly asymptomatic. As Benedetti et al. [18] have discovered, if one carefully questions children with PFF it turns out that more have problems than previously thought.

2) Does PFF cause long-term impairment?

If it was known that PFF would eventually lead to impairment and disability in later life, then the decision to treat would be simple. Unfortunately there are no published longitudinal studies investigating the consequences of untreated PFF, thus this rationale cannot be used to mandate treatment. Nonetheless there are some anecdotal suggestions that some individuals with PFF will have long term problems, joint ankyloses and arthropathy [212]. This has guided the approach to treatment for some clinicians, but raises some ethical issues, particularly when surgical intervention is considered.

3) What determines whether a child with PFF gets symptoms?

This is the ultimate unanswered question of PFF. Children with symptomatic PFF do not start this way; they start with asymptomatic feet, and over a period of time start getting problems. No-one knows why, of those children with apparently similar foot types, some develop symptoms and others remain asymptomatic. From the limited evidence that exists, plantarflexor contracture is implicated in symptomatic PFF, but is only part of the story. It is a general assumption that there are specific structural and functional factors that cause a child with PFF to develop symptoms, but these are yet to be fully elucidated. If it were possible to identify these factors, then it might be possible to construct more objective clinical practice guidelines. This would particularly be useful in identifying the asymptomatic children who were potentially at risk of developing problems. If specific mechanisms of symptom generation were identified, then it could also help guide treatment strategies. Currently the majority of treatments focus on making the foot appear more normal, but if it is still unclear how the structure of PFF relates to symptoms, then this may not be the correct strategy.

2.10. Aims of thesis

Thus, the overall aim of this thesis is to discover if there are structural or functional factors associated with PFF that relate to the presence of symptoms. By doing this, it is hoped that the treatment indications for children with flexible flat feet can be better defined.

This broad aim can be met by consideration of the following objectives:

i) To classify foot posture using objective methods.

ii) To assess the clinical impact of PFF on a child's quality of life.

iii) To investigate the aetiology of PFF including the relationship of subtalar joint morphology to foot posture.

iv) To test the biomechanical theories of how PFF function could cause symptoms.

Chapter 3. Study design and methods

3.1. Introduction

The aim of this project was to gain a better understanding of the structure and function of PFF and to investigate how this relates to symptoms and health related quality of life. There are a number of ways that this could be achieved, and this chapter begins with a discussion of potential experimental techniques and the rationale behind the ultimate choice of methods. This is followed by a discussion of the principles behind the study design and subject recruitment. Subsequently the general experimental protocols used to collect data are described in detail including problems and technical difficulties encountered during data collection. Validation of the semi weight-bearing MRI technique, as well as the data processing methods are described towards the end of this chapter. Specific methods involving the analysis of the collected data are included within subsequent chapters.

3.2. Experimental methods

In the previous chapter relevant literature about flat feet was discussed. The work presented demonstrated that numerous experimental methods can be utilized to gain insight into the structure and function of flat feet. The aim of this study was to place the structure and function of PFF into the clinical context. Achieving this precludes the use of ex-vivo techniques such as analysis of osteological specimens, cadaveric studies or mathematical or computational methods [22, 23, 36, 39, 230, 308, 333]. As this investigation was conducted on a paediatric population there were also other ethical issues which to a certain extent dictated the methods that could be used.

3.2.1. Assessment of structure

Assessing the structure of PFF in-vivo can be achieved in two main ways. Foot morphology can be assessed clinically by visual inspection of the foot and anthropometric measurement in a non-weightbearing or weightbearing setting. This can help describe the size and shape of the foot, as well as distinctive features and deformity associated with foot and ankle pathology. However to understand what dictates the observed topographical anatomy, it is important to gain insight into the orientation, alignment and morphometry of the foot and ankle bones. Thus the second method for assessing foot structure is by using radiological imaging. Weight-bearing antero-posterior and lateral foot and ankle radiographs have been used routinely in the assessment of PFF in clinical and research settings [17, 213, 325]. These are useful in helping to identify bony structural abnormalities, and to assess relative alignment of segments, and are regarded as the gold standard technique for classifying foot posture [224]. This method is also relatively quick and cheap. It does however come with some drawbacks. Firstly radiographs use ionizing radiation, which dependent on dose, can lead to neoplasia. Soft tissues often have similar appearances on a radiograph, which can make it difficult to discriminate normal from pathological soft tissue. Radiographs also are a two-dimensional representation of a three-dimensional structure. This requires careful positioning of the x-ray beam and patient, to minimise superimposition of bones. Radiographs are also susceptible to changes in the perspective and projection of anatomic structures depending on the orientation of the x-ray beam (parallax error) [156]. For example a true antero-posterior radiograph of the foot can distort the appearance of the tarsometatarsal joint [156]. Parallax error and bony superimposition is further compounded in cases of foot and ankle deformity.

The preferred technique for visualising bony anatomy of the foot and ankle is computed tomography (CT) [162]. Like radiographs, CT utilizes ionizing radiation, but instead of a two-dimensional picture displaying the whole three-dimensional volume, CT generates regularly spaced stacked cross-sectional images. These can be combined to generate highly detailed three-dimensional reconstructions of bony anatomy, and have been used occasionally in flat foot research [5, 98, 162, 248]. The main drawback is that a high resolution CT of the foot and ankle results in significant amounts of ionizing radiation, which preclude its use in research in children [34].

More attractive imaging modalities from the ethical point of view are those that do not use ionizing radiation, and as such pose no potential health risk to the patient. The two most commonly used modalities are ultrasound scanning and magnetic resonance imaging (MRI).

Ultrasound is a safe non-invasive technique that uses high frequency sound waves to visualize internal body structures. This has been used in the context of flat feet to describe the plantar fascia and muscles, but this modality is unable to describe bony anatomy in any detail [6].

MRI is regarded as the best non-ionizing technique to assess the anatomy of the foot and ankle [139]. The principles of MRI are based around the detection of hydrogen nuclei (protons) in water, the most abundant compound in the body. Protons, spin around an axis (nuclear spin), and can be regarded as a miniature bar magnets with a positive and negative pole. Under normal conditions the axes around which protons spin are randomly orientated. When these protons are subject to the strong magnetic field of an MRI scanner their axes partly align with the long axis of the field and the nuclei rotate about their axis like a gyroscope at a characteristic frequency (known as precession). At this stage the protons are not precessing in phase, so there is no net magnetic field. When a radiofrequency pulse is applied, proton precessions step into phase, and the axes all align, inducing signal currents in the receiving coils of the scanner. When the radiofrequency pulse stops, the protons return to their previous alignment and the manner and time in which this is done affects the signal induced in the coils giving an indication of the tissue type. The scanning space can be regarded as a three dimensional grid of boxes, called voxels, and by ascertaining the tissue type for each voxel it becomes possible to construct an accurate anatomical picture. The size of the voxel will also determine spatial resolution.

A number of different MRI image sequences exist, with T1 or T2 weight images being the most commonly used. T1-weighted images are optimal for assessing anatomy, whereas T2 images are more suitable for assessing pathology.

MRI scans, like CT, are usually performed with the patient in the recumbent position with the joints unloaded. It is well accepted that joint alignment will change on loading, and this is particularly the case in PFF where the arch flattens as the foot is loaded [70, 205, 246]. Therefore a number of researchers have attempted to simulate weight-bearing conditions to gain further insight into the loaded joint alignment of the foot and ankle [5, 98, 162]. This has not previously been attempted in a paediatric population.

3.2.2. Assessment of function

Lower limb function in children can be assessed in a qualitative or quantitative manner. Qualitative methods include assessing performance in certain motor tasks.

For example Lin et al. [180] investigated the ability of pre-schoolers with and without flat feet to perform a number of different motor tasks including squatting, one-leg standing and one-leg hopping. These tests, whilst useful, do not elucidate the mechanisms by which symptoms and functional impairment may be caused. Quantitative methods of assessing function are more likely to provide insight on the pathomechanics. The technique of instrumented gait analysis is regarded as the method of choice to obtain quantitative data on function of the lower limbs. It is this technique that has furthered the field of foot and ankle biomechanics as presented in Chapter 2. Instrumented gait analysis consists of a number of biomechanical methods and incorporates clinical examination as standard. Used individually or often in combination these methods can provide information of the effect of functional activities in five areas:

1. Temporal and spatial parameters of gait (spatio-temporal parameters)

2. The motion of limb segments and joints (kinematics)

3. The forces, moments and powers that produce motion (kinetics)

4. The way pressure is distributed through the sole of the foot (pedobarography)

5. Muscle function (electromyography)

1. Temporal and spatial parameters of gait (spatio-temporal parameters)

These parameters are the basic descriptors of gait and include step length and width, cadence, and walking speed. These parameters are described and illustrated in Figure 3.1. They often provide the simplest method to evaluate and characterize gait and can be obtained using only a tape measure and stopwatch, or can be calculated using more complex methods using force plates and three-dimensional gait analysis (3DGA) described later in this chapter. These parameters are routinely used to evaluate the development of walking in children, and have been used in a number of studies investigating PFF [129, 179, 309, 319, 340].

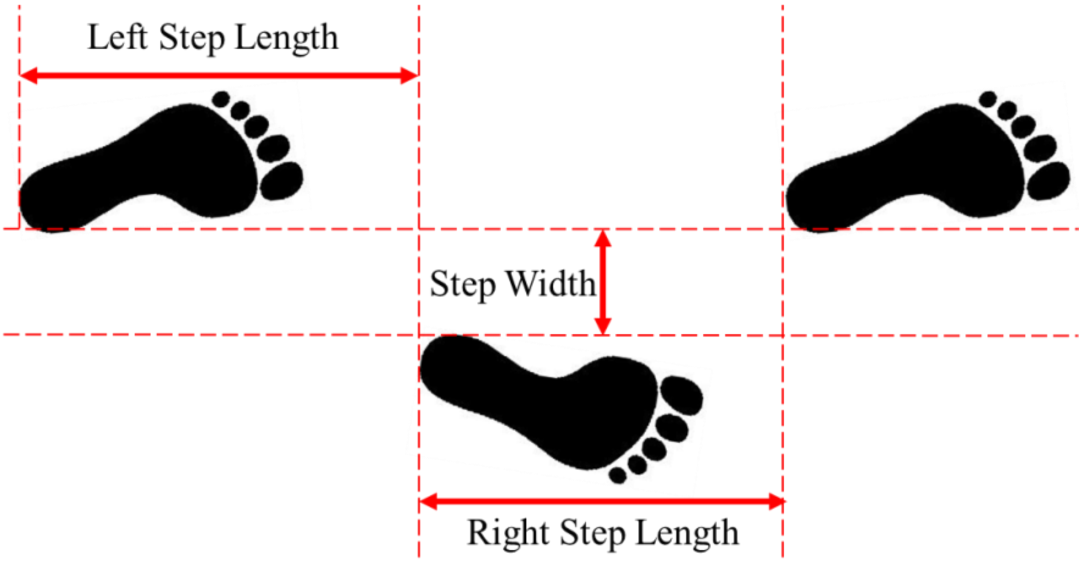


Figure 3.1 Definition of spatio-temporal parameters. Step length is the distance from the point of initial contact of one foot and the point of initial contact of the opposite foot. Step width is a measure of the medio-lateral separation of the feet. Walking speed is calculated as the step length divided by step duration. Cadence is the number of steps per minute.

2. Kinematics

The position and orientation of limb segments and joint angles can be measured using a number of different techniques. Some methods necessitate the direct attachment of joint angle measuring devices to the subject. Such devices include electrogoniometers. These methods are simple in terms of the hardware required, but can be bulky encumbering the subject. They are most suited to providing information about the sagittal plane motion of large joints. For a more comprehensive three dimensional (3D) description of joint motion, camera-based systems are now the preferred option.

These systems have increased in precision and complexity over the last 100 years, with optoelectronic systems now being the industry standard. The cameras used in optoelectronic systems are designed to track markers attached to the test subject. The images taken by each camera are two-dimensional; as such, one camera cannot track the 3D position of a marker in space. If however two or more cameras can track the marker, then its position in 3D space can be triangulated. During functional activities there will be times when markers are obscured from the cameras. Having arrays of cameras distributed around the gait laboratory ensures that, in the most part, at any one time at least two cameras can track the marker. The more cameras, the better the marker tracking.

Optoelectronic systems use either passive or active markers. With a passive marker system, cameras emit visual or infra-red light and register light reflected from markers coated in reflective material. In active systems markers themselves emit light usually by a light emitting diode which is tracked by the camera system.

For the purpose of clinical gait analysis, groups or clusters of markers are used to define limb segments, and limb segments are treated as rigid bodies. Having at least three non-collinear markers allows the segment to be tracked for all six degrees of freedom of motion. Marker data are then combined with anthropometric data to form a virtual skeleton. With the 3D position of each limb segment defined in the global laboratory coordinate frame and with some knowledge and assumptions of how the marker positions relate to the underlying bony and joint anatomy, it is then possible to calculate the angles between the segments (joint kinematics). A full description of the mathematical methods by which this is done can be found in Cappozzo et al.[47].

A number of marker configurations have been described to track lower limb movement with the most frequently used being the Plug in Gait marker set (PiG) [149]. This marker set however only models the foot as a single line vector, and thus it is only possible to assess dorsiflexion/plantarflexion and internal/external rotation of the whole foot relative to the shank/tibia. Modelling the whole foot as a single line is a gross over-simplification of a complex structure. This, up until recently, has been necessary as markers could not be placed too close together, due to limitations in the spatial resolution of gait analysis hardware. This in turn made the investigation of foot and ankle biomechanics difficult. However in the last decade, with significant technological advances, it has become possible to track smaller markers and much higher densities of them. This has facilitated the development of multi-segment foot models (MSFM) designed to gain further understanding of the motion of individual foot and ankle segments. A number of multi-segment foot models have been devised including the Oxford Foot Model (OFM), Heidelberg Foot Measurement Method (HFMM), Milwaukee Foot Model (MFM), the Instituto Orthopedico Rizzoli (IOR) model and the modified Shriner's Hospital of Children Greenville model (mSHCG) [49, 161, 172, 275, 290]. Of these models the OFM has undergone the most rigorous testing in normal and pathological populations, and demonstrates strong reliability in adult and paediatric populations, making it the preferred model of many gait laboratories [69, 77, 351]. It is also endorsed by the world leader in gait analysis system manufacture, Vicon (Vicon, Oxford, UK). In the literature, the OFM is also the most commonly used MSFM in the gait analysis of flexible flat feet [129, 179].

The OFM models the foot as three segments, the hindfoot, forefoot and hallux. Ankle joint motion is defined as the relative movement of the hindfoot with respect to the tibia, representing the combined motion of both the ankle and subtalar joints. Movement of the forefoot relative to the hindfoot occurs across numerous joints, and can be thought of as rotation of these segments about a theoretical midfoot joint. Finally, the first metatarso-phalangeal motion is modelled as the movement of the hallux relative to the forefoot [301].

Even though these methods are well established, there are a number of potential sources of error which could affect the results. Skin mounted markers are subject to skin and soft tissue motion artefact, which is worse in areas with larger amounts of soft tissue between the skin and the bone and in areas near joints. A number of studies have quantified the potential magnitude of soft tissue artefact and these are summarized by Leardini et al. [173]. The absolute displacement depends on the marker location and the amount of overlying soft tissue, and can vary from one to two millimetres on the metatarsals to over five centimetres on the thigh. In practice this is minimized by placing markers in areas where there is less soft tissue, like bony prominences. Attaching markers directly into bone will also remove soft tissue artefact but is not a practical option in children, or in other populations, for obvious reasons.

To adequately model limb segments markers are placed at specific anatomical locations or aligned with specific bone axes. Error in marker placement may lead to significant changes in joint angles measured. Before undertaking this project, measurement of potential marker placement error of the OFM around the foot and ankle was measured, and was found to generally be acceptable in most places except for placement of markers around the heel [168]. Knowledge of this highlighted the importance of training and a systematic reproducible approach to marker placement.

There will also be systematic errors in marker tracking related to the motion capture system. Careful calibration of the motion capture system before use can minimise these errors. A well calibrated motion capture system has a mean camera tracking error of less than 0.1 mm [55].

3. Kinetics

Aside from measuring joint kinematics it can be useful to measure the interaction between limb segments caused by external forces applied by the surroundings, and internal forces applied, either actively by muscles, or passively by stretch of periarticular ligaments and the joint capsule. The quantities relating to these interactions include joint forces, moments and powers and are collectively known in the field of gait analysis as kinetics. Knowledge of joint kinetics may be instrumental to understanding the primary cause of, or compensation as a result of, deviations in gait patterns.

The external force applied by the surroundings in this context is the reaction of the ground to the force exerted on it, by the body (ground reaction force (GRF)). According to Newton’s third law of motion, the GRF is equal in magnitude and opposite in direction to the force exerted. In the gait laboratory this is measured using force plates set into the ground. The force plate transduces the force applied to it into an electrical signal. The transducers may be based on strain gauge, capacitance gauge, piezo-electric or piezo-resistive technologies and can be classified as either single or multiple pedestal. The simplest force plates only measure the vertical component of the GRF, which alone is not adequate to calculate joint kinetics using conventional methods. More advanced force plates are able to measure the 3D components of the single equivalent GRF (resultant GRF) and the point of application (centre of pressure (COP)), as well as the vertical moment of force, also known as the free vertical torque (T_{GRF}). The resultant GRF, COP and T_{GRF} can then be used with segment linear/angular acceleration (obtained from marker data), segment masses and moments of inertia to calculate the forces and moments at the joint. This process is inverse dynamics analysis (IDA), an approach first pioneered by Braune and Fischer [33]. A full description of the computation of lower limb joint kinetics has been made by Winter [345].

Inverse dynamics is a powerful technique for understanding movement, but does have some inherent limitations. Firstly the technique relies on the assumption that all joints are pin joints, with no friction at the joint, and all the forces produced by joint elements such as ligaments and joint capsule pass through the joint centres. Another significant limitation is that the body segment parameters (relative mass, mass moment of inertia or radius of gyration of each segment as well as position of their COM) used to calculate joint kinetics are usually determined from published cadaveric studies [345]. As such body segment parameters are approximations and generalizations. Inverse dynamic analysis is also sensitive to errors in joint centre of rotation estimation, and soft tissue motion artefact of the skin mounted markers [127, 128]. As the IDA process begins at the foot and works proximally, it is evident that any errors affecting distal joint calculations will affect the proximal joints, leading to a propagation of error up the kinetic chain. Finally the method will only provide net joint moments, thus it is not possible to assess the contribution of individual muscles, or evidence of co-contraction which may be more pronounced in neuromuscular pathologies with spasticity like cerebral palsy. For that, full musculoskeletal modelling of the lower-limb is required.

The calculation of joint kinetics based on the PiG model is common place, and often incorporated and output by the gait analysis software. As previously stated, PiG models the foot as a single line vector, and this oversimplification results in significant inaccuracies in the computation of ankle kinetics, as demonstrated by Dixon et al. [81]. With the advent of MSFMs it is now possible to calculate kinetic quantities for individual foot segments. This approach has been used by some researchers, but is yet to reach mainstream clinical practice [81, 187]. Nonetheless, the use of kinetics, derived from MSFMs, to gain further insight into interactions between different foot segments is attractive when researching foot and ankle pathology.

4. Pedobarography

As well as measuring the force going through the foot, it is also desirable to measure the plantar pressure (pedobarography), i.e. the way the force is distributed across the sole of the foot. In some pathologies, for example, diabetes and rheumatoid arthritis, it is pressure that is the critical quantity that determines the harm done by the force [322, 329].

The earliest investigations of foot pressure patterns involved the imprint in Plaster of Paris, but these methods were crude and only allowed qualitative assessment of foot shape [15]. Subsequent methods involved air filled chambers, succeeded by arrays of deformable materials that either left an ink print or could be recorded by optical methods [48, 191]. Whilst some simple mechanical methods like the Harris mat [120] are still in use, the general trend has been towards the use of more sophisticated systems (pedobarograph) using electromechanical transducers. The pedobarograph can either be floor mounted or in-shoe, and consists of arrays of transducer sensors which communicate with a central processing unit. Much like the force plate, the transducers can be based on a number of technologies, but commonly are either capacitive or piezoelectric. Unlike the force plate these sensors only measure the normal component of the GRF, not shear. The size and number of sensors in the array will determine the resolution of the pedobarograph, with Davis et al. [74] advising that sensors size should always be less than 6.4 by 6.2 mm to allow adequate characterization of the pressure. The measured force is divided by the sensor surface area to obtain the pressure in Pascals (Pa). Large sensors can under-estimate real pressure values of defined pressure peaks under small anatomic structures (e.g. metatarsal heads), as a result of lower pressures around these peaks also contained within the sensor area [216].

Data collection is determined by the specific type of pedobarograph in use, but can be either static (standing) or dynamic (walking/running). The in-shoe pedobarographs have the benefit of being able to measure multiple sequential steps, but only measure the interaction between the sole of the foot and the shoe. A floor mounted pressure plate can measure the, at times, more desirable interaction between foot and floor. The relatively small area of the pressure plate requires a degree of ‘warming-up’ and practice, to ensure the whole foot lands perfectly on the plate. This is not necessary for the in-shoe device.

A number of different parameters can be extracted from the pedobarograph measurements, and these can be applied to the whole foot or specific regions of interest. For example the Novel pressure plate software (Novel GmbH, Munich, Germany) can output over 50 different parameters, thus deciding which is appropriate to use is dependent on the specific clinical or research aims. The simplest measure is the peak pressure during gait for each sensor. This has proven particularly useful in the diabetic neuropathy and rheumatoid arthritis populations to identify areas of excessive pressure that can lead to tissue breakdown and ulceration [322, 329]. For other applications the temporal characteristics of foot loading may be of interest, and the combination of time and amplitude factors is reflected in the pressure-time-integral, or the force-time integral. The COP can also be calculated from the pressure pattern, and the progression of the COP may be useful in providing further insight into foot function [56, 140]. The utility of pedobarographic measures in clinical populations aside from patients with diabetes and rheumatoid arthritis continues to be debated. In foot and ankle research plantar pressure has been used in a number of ways, including investigating the sequelae of trauma, effect of surgical intervention and investigation of foot and ankle deformities [14, 28, 32, 270, 339].

5. Electromyography

It is often important to understand how different muscles contribute to functional activities, especially in the context of PFF where certain muscle groups may fatigue early. Electromyography (EMG) is used to measure the electrical activity of muscles during activity. The electrical signal gives an indication of the timing and relative intensity of muscle activation, but one cannot infer the force of muscle contraction. EMG can be measured using surface electrodes or fine wire electrodes directly into the muscle. The former has the benefit of being non-invasive, but it is only suitable for superficial muscles and can be subject to movement artefact and interference from adjacent muscle groups. The later can give a better signal, and be used for deeper muscle groups, but is invasive in nature and gives a very local measurement. Both are susceptible to electrode placement error [341].

Electromyography data was not collected in this study as monitoring of the majority of the muscles of interest in PFF, including tibialis posterior, the intrinsics and peroneus longus would have required fine wire electrodes. This is not possible in a paediatric population, and it was felt that surface EMG alone would not have given an adequate picture of the relevant muscle activation in PFF.

3.2.3. Assessment of symptoms

For some children PFF may just be an incidental finding; the child may experience no symptoms or problems at all. Other children however may get a variety of symptoms that could significantly impair quality of life [18, 20]. The potential symptoms with which a child with PFF may present are numerous and nebulous. Hosl et al. [129] recorded the symptoms experienced by 14 children with PFF and found that the predominant problem was pain followed by fatigue and discomfort. When the site of pain was investigated further, a total of six different anatomical locations, around the foot and ankle, were identified. Benedetti et al. [18] investigated 54 children with PFF and found that most patients had foot symptoms (65.3% of feet) and 68.3% of feet were associated with functional limitation. A sixth of the symptomatic children had discomfort and/or tiredness associated with activity and the remainder complained of pain. In this group the pain was found predominantly on the plantar aspect of the foot (28.7%), but also medial hindfoot (18.8%), medial forefoot (12.9%), lateral hindfoot and forefoot (5% and 2% respectively). These two studies highlight the variability in symptomatology of children with PFF.

The nature of symptoms in PFF can be established through medical history taking. However from both a clinical and research perspective it would be useful to have a metric that could quantify symptoms. Not only could this help guide management, but it would also aid in the investigation of potential factors that may cause symptoms. When symptom profiles are so heterogeneous, quantifying symptoms becomes fraught with difficulty. As such, in all the literature where symptoms in PFF have been investigated, when it comes down to statistical analysis, children have been labelled as either symptomatic or asymptomatic [17, 129, 213, 339]. This fails to be an adequate representation of the clinical picture. Such a binary definition would not be able to distinguish a child with slight discomfort after walking several miles from a child with foot and ankle pain experienced whenever they walk even short distances.

In terms of quantifying pain in children a number of validated methods exist including the Wong-Baker faces rating scale [348], 0 - 10 numeric pain rating scale [245] and the visual analogue and verbal pain intensity scale [254]. A problem with using any of these is that firstly they only quantify one dimension of the multidimensional symptom profile of PFF. Secondly the measures only take a ‘snapshot’ of symptoms at the current time point. Children with symptomatic PFF would usually develop symptoms during certain activities, with symptoms easing with rest. These pain measures may not accurately represent this. Measures also exist for measuring fatigue in certain patient populations, but again are limited by assessing only one dimension of the symptom profile.

An alternative approach is to measure the health-related quality of life (HRQOL) in children with PFF. Numerous definitions of HRQOL exist, but the majority consensus is that HRQOL represents the functional effect of a medical problem upon a patient [117, 247, 279, 349]. By definition this may be subjective and spans physical, occupational, psychological and social dimensions.

As Osoba and King [242] eloquently argue, “The ultimate goal of health care is to restore or preserve functioning and well-being related to health, that is health-related quality of life”. If it is the HRQOL that ultimately guides management then it seems appropriate to measure it. Indeed measuring HRQOL in clinical and research practice is becoming more widely spread, and regarded by many as the gold standard method of measuring the impact of a condition on the patient [75]. In this study validated HRQOL measures have been used to quantify symptoms severity in participants. In the following section a description of HRQOL measures is given, including important factors related to their construction and use.

3.2.3.1. Overview of HRQOL measures

Health related quality of life measures usually take the form of a structured questionnaire with carefully composed questions designed to predominantly capture information on one or more domains of health (physical, social and psychological/emotional). The structure and content of the measure is carefully determined and should have the following properties [306]:

1) Validity. Does the instrument measure what it purports to measure?

2) Appropriateness and acceptability. Is the measure suitable for measuring what it purports to measure?

3) Reliability. Will the results of the measure be consistent when repeated in the same population?

4) Responsiveness to change. Is the measure sensitive to clinically important changes?

5) Interpretability. Do the results from the measure have clinical significance?

To ensure that a HRQOL measure has these properties it undergoes rigorous and strict validation. Only once this is completed can such a measure be used safely.

HRQOL measures can be split into two broad categories: generic or site/disease-specific. Generic measures are generally quite broad in capturing various aspects of health and the consequence of disease on the individual. This design makes it possible to be used in a wide variety of patient groups with a wide variety of diseases giving quite generic HRQOL information. An example of the most widely used generic tool of this type in adults is the Short Form 36 [334]. As tools like this can be used in a number of populations it makes it easier to compare HRQOL of one patient group with another. A disadvantage of such a broad approach, in capturing HRQOL, is that this may be at the expense of detail in terms of relevance to a specific illness. It is expected that a generic tool is likely to have fewer relevant items to the disease in question thereby making it less sensitive to change. It is for this reason that site and disease specific measures have been developed.

Site/disease specific measures adopt a much more focused approach in measuring HRQOL, asking only questions that would be directly relevant to individuals with that disease or problems with that site. An example of a site-specific tool used in adults is the Oxford Hip Score [154]. Tools of this type tend to be more sensitive than generic tools in tracking change in the region experienced by patients as a result of intervention.

Generic and site/disease specific tools both have their respective advantages and disadvantages, and as such often a combination of these tools is used to capture HRQOL. This approach is advocated by a number of trialists [25, 102, 116]. The main disadvantage of using multiple tools is that patient compliance may go down if he or she is asked to complete too many questionnaires.

To add meaning to HRQOL measures, comparison can be made with any existing normative database as well as comparison with previous published results. It is also important to be aware of the minimally important difference (MID) of the HRQOL. The MID is defined as the minimum change in questionnaire score that represents a clinically important or relevant change in health status of the patient. Part of the extensive HRQOL measure validation process usually involves establishing the MID values.

The MID values can be calculated using anchor-based methods or a distributional approach [198]. The anchor-based approach as pioneered by researchers at McMaster University assesses changes in scores using an “anchor” as a reference [148]. The anchor in a longitudinal study is usually a simple question to assess whether the patient feels better or worse than they did before. Assessing which questionnaire scores correspond to the patient feeling better or worse will guide the MID. In cross-sectional studies patients can be divided according to diagnosis, disease severity or functional status among other potential anchors and mean difference in HRQOL scores between groups can help define the MID [274]. A distributional approach involves using statistical methods to assess the spread of questionnaire scores to calculate the MID. The majority of the statistical approaches calculate MID on the basis of the effect size, defined as the difference in group mean scores divided by its standard deviation. In general anchor-based methods are favoured to distributional in calculating MID [274].

3.2.3.2. Assessing health status in children

A number of health related quality of life (HRQOL) measures have been developed to assess general health status in children. One of the most commonly used generic HRQOL measures used in the paediatric population is the PedsQL™ 4.0 generic core scales [327]. This tool developed by Varni et al. [326] has been designed to measure the core dimensions of health as outlined by the World Health Organization as well as school functioning. The PedsQL™ 4.0 has been extensively validated and is proposed to be reliable, valid, responsive and developmentally appropriate. The tool can also be used in healthy populations as well as those with pathology.

In the context of assessing foot and ankle problems in children there is currently only one site-specific HRQOL measure, the Oxford Ankle Foot Questionnaire for children (OxAFQ-C) [219]. The OxAFQ-C has been proposed to measure HRQOL in children between the ages of 5-16 with foot and ankle pathology. Morris et al. [217, 218] have validated the questionnaire by assessing scaling and reliability as well as responsiveness and longitudinal validity. To assess construct validity, Morris et al. [218] undertook a process similar to the method of convergent and discriminant validity described by Campbell and Fiske [45]. In this method domains of a new questionnaire are cross-correlated with those of a well-established one. If the new questionnaire has reasonable construct validity then domains that measure similar constructs are expected to show a higher correlation than those measuring dissimilar constructs. Morris et al. used the well-established Kidscreen HRQOL in this comparison demonstrating good convergent and discriminant validity of the OxAFQ-C.

Thus, the rationale behind using HRQOL measures in this study has been discussed. Further details of the OxAFQ-C and PedsQL™ 4.0 are found in section 3.5.1 (p.78).

3.3. Study design

This study was prospective, cross-sectional and observational in design. The study was approved by the local NHS Research Ethics Committee (Oxford B, reference: - 12/SC/0334). Approval was also obtained from the Research and Development Department of Oxford University Hospitals NHS Trust. Letters of approval can be found in Appendix 1. Participants were not coerced into participating and all provided informed assent. Parent/Guardians of the participants all provided informed consent for their child/ward to take part in the study. Research practices as outlined by the good clinical practice (GCP) guidelines were adhered to. Data collection occurred over the period of August 2013 to April 2014.

To ensure consistency I personally undertook the data collection as follows:-

Subject recruitment

Physical examination, including assessment of flexibility

Instrumented gait analysis

Administration of questionnaires

3.4. Subject recruitment

Children between age eight and fifteen years old inclusive were recruited into this study. This age range was used for a number or reasons. As outlined in Chapter 2 (p.31-32), arch development is usually complete by the age of six years old. It was hoped that by adopting a conservative lower age limit of eight years old, all developmental flat feet would be excluded from the study. An upper age limit of fifteen years old was adopted because at the age of sixteen, the patient would transition from paediatric orthopaedic/physiotherapy services to adult services, and thus the term PFF would no longer be correct. The resulting age range is also representative of the population attending clinic with symptomatic PFF [159].

Inclusion and exclusion criteria for this study as well as potential sources of entry into the study are shown in Figure 3.2.

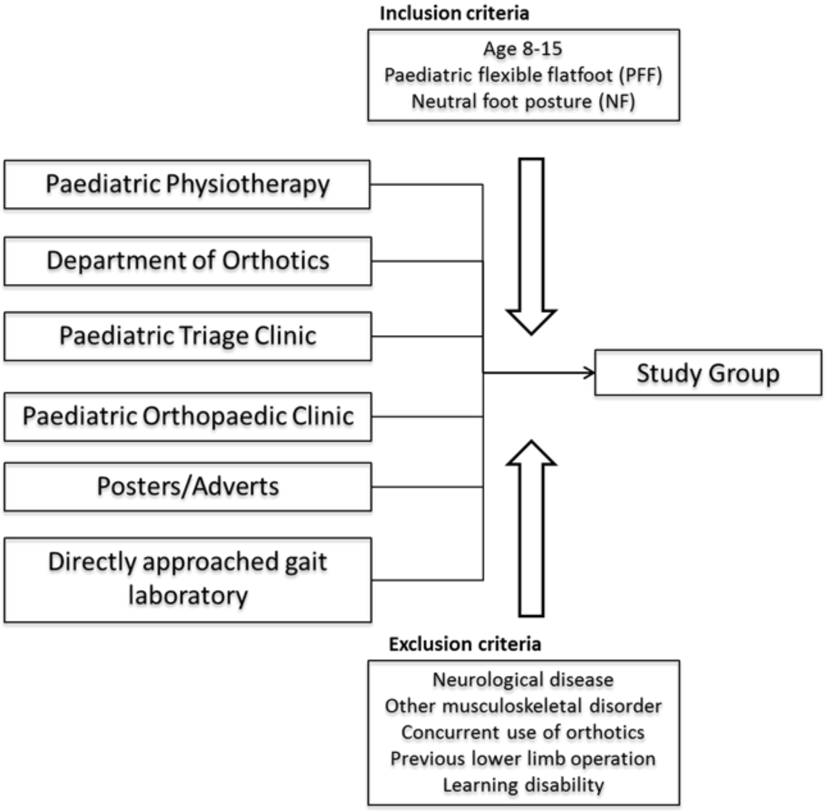


Figure 3.2 Selection criteria and route of entry of children into study.

The first forty subjects were recruited sequentially via the routes outlined in Figure 3.2, and data from these subjects were then used to devise an objective foot classification technique as outlined in Chapter 4 (p.93-120). Subsequently as each further subject entered the study the objective classification technique was used to define foot posture. As recruitment proceeded, attempts were made to have similar final NF and PFF group sizes. Thus during recruitment, when it appeared that there were a greater number of children defined as having PFF than NF by the objective classification tool, priority was made to recruit children from the community with neutral foot posture (on the basis of parental opinion). Similarly when it appeared that there were a greater number of children defined as having NF than PFF, focus was placed on recruiting children from clinics who had been referred because of a flat foot posture. Foot posture was subsequently always formally defined using the objective classification technique and this did not always agree with parental or clinician opinion. However by formally defining foot posture as recruitment progressed it was possible to react to any in balance in PFF and NF group sizes by adapting recruitment methods as outlined.

In total 95 children were recruited into this study. Care was taken not to bias recruitment on the basis of symptom severity, and impaired quality of life, thus HRQOL measures were only completed after recruitment. Subjects were recruited sequentially, with no exclusions for reasons outside the exclusion criteria shown in Figure 3.2.

3.5. Data Collection

3.5.1. Questionnaire evaluation

Participants were each administered two questionnaires to assess health-related quality of life; the Oxford Ankle Foot Questionnaire for children (OxAFQ-C) and the Pediatric Quality of Life Inventory™ (PedsQL™ 4.0) (Appendix 2) [219, 327]. Parents of participants completed the proxy component of these HRQOL measures. Questionnaires were administered according to published guidelines. A brief summary of the HRQOL measures is given below.

The OxAFQ-C has a total of 15 items, the first 14 of which are used to calculate domain scores, the three domains being:

- Physical (6 items , 1-6)
- School & Play (4 items, 7-10)

- Emotional (4 items, 11-14)

The three domain scores are reported separately.

The scoring system is designed to assess how frequently each issue (represented by an item in the questionnaire) is a problem. The response options to each item are on a 5-point scale rated from never (4), rarely (3), sometimes (2), very often (1) to always (0), where the number in brackets represents the value that is applied by the scorer to each response. Domain scores are calculated as the total of the scale item scores divided by the maximum for each domain (i.e. Physical 24, and School & Play and Emotional 16). Domain scores can be transformed to a percentage scale (0-100) to aid interpretation. A higher score for a domain represents better functioning.

The final item (item 15 – Has your foot or ankle stopped you wearing any shoes you wanted to wear?) has been added to reflect the concern by many children that they cannot wear the footwear they prefer. This issue is important to children and therefore adds face validity, but psychometrically it does not fit into any of the domains. Therefore, this final item 15 is reported as a single item.

The PedsQL™ 4.0 Generic Core Scales tool has been developed through focus groups and cognitive interviews to capture general HRQOL in children aged 2-18 (ages 2-5 solely proxy reported) [327]. There are 23 items which encompass, physical functioning (8 items), emotional functioning (5 items), social functioning (5 items) and school functioning (5 items). Each item consists of a 5 point Likert scale which is then reverse scored and linearly transformed to a 0-100 scale, so that higher scores indicate better functioning. Domain scores can be reported separately as the mean score of all items in the domain. A total score can also be reported as the mean score of all item scores in the questionnaire.

3.5.2. History

A thorough history was taken from the child with help from the parent/guardian. Any previous medical problems, previous operations and family history were documented. If the participant had symptomatic PFF then if applicable the worse side was recorded. Using a basic Likert scale of, never (1), rarely (2), sometimes (3), often (4) and on a daily basis (5), participants were questioned about pain in the knee, hip and back in the last month.

3.5.3. Clinical examination and anthropometric measurements

The participant underwent a standard clinical examination. Lower limb joint range of motion was measured using a standard goniometer. As per standard methods outlined by Reider [263], passive hip, knee and ankle joint range motions were measured. In the case of the ankle, the Silfverskiöld test was performed to assess ankle dorsiflexion with the knee extended and flexed to 90° [288].

Craig’s test was used to measure femoral anteversion [263]. Tibial torsion was measured as the angle between the long axis of the foot and the intermalleolar line. This was undertaken with the child sitting on the edge of the examination couch with their knees at 90° flexion. Knee coronal alignment was measured to the nearest five degrees with the participant standing.

Flexibility was assessed according to the Beighton score and the Lower Limb Assessment Score (LLAS) [16, 97]. The Beighton score assesses flexibility on the basis of a nine point scale with measures from upper and lower limbs. Figure 3.3 summarises the components of the Beighton score. The LLAS focuses on lower limb hypermobility and is based on 12 separate points with a separate total score per limb. Table 3.1 summarises the components of the LLAS.

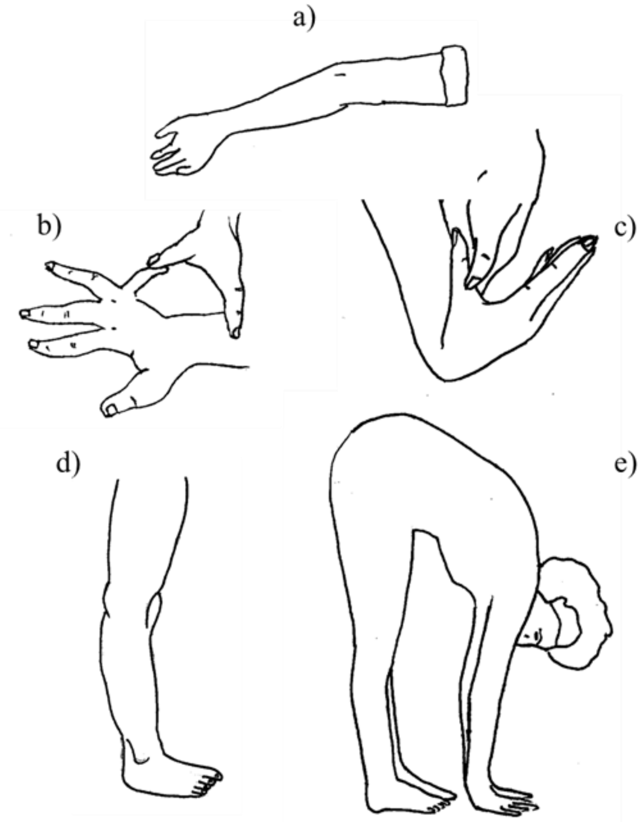


Figure 3.3 Beighton score components. a) Hyperextension of elbows >10° (one point per elbow, b) Hyperextension of little finger to >90° (one point per hand), c) Thumb to forearm without flexion of the interphalangeal joint (one point per hand), d) Hyperextension of the knees >10° (one point per knee), e) Palms to floor without bending knees (one point). Nine points in total.

Foot posture was assessed non weight-bearing with the hindfoot controlled in neutral and weight bearing. Hindfoot alignment (varus/valgus), arch height (planus/neutral/cavus), transverse foot alignment (abduction, adduction) and foot pronation/supination were recorded.

HIP FLEXION. Patient supine, examiner flexes one hip fully, other leg extended. Does the anterior thigh drop easily onto chest with minimum/moderate force application?
HIP ABDUCTION. Patient supine, knees and hips flexed. Knees are allowed to fall outwards with soles of feet staying together. Examiners hands on lateral aspect of knee With minimal force application can the lateral femoral condyle of the knee come down sufficiently to let the back of the examiner’s hand touch the couch?
KNEE HYPEREXTENSION. With the patient supine can the knee by hyperextended so that the heel is at least 3cm off the couch (2 finger breadths)?
KNEE ANTERIOR DRAWER TEST. When the anterior draw test is performed Is there obvious forward movement of the tibia against the femur with palpable clunking of the joint?
KNEE ROTATION. Patient supine, knee and hip at 90°. Lower leg held by malleoli and shank rotated laterally and medially. Does the tibial tubercle move easily beyond 1cm in any direction or >2cm in total?
ANKLE JOINT DORSIFLEXION. With the knee flexed to 45° can the ankle be dorsiflexed to 15° or greater with moderate to strong force?
ANKLE ANTERIOR DRAWER TEST. Knee flexed to 45°, one hand gripping the foot around the back of the heel, and the other stabilising the tibia. Can the calcaneum and talus be brought forward on the tibia using a strong anterior force?
SUBTALAR JOINT INVERSION. Is the greater than 45° of subtalar joint inversion?
MIDTARSAL JOINT INVERSION. With the hindfoot controlled by one foot does the midtarsal joint invert beyond 45° so that the plantar surface of the metatarsal heads can be brought inwards by 45°?
MIDTARSAL JOINT AB/ADDUCTION AND DORSI/PLANTARFLEXION. Movement of the forefoot with one hand, with the hindfoot controlled with the other. Does the forefoot move in either ab/adduction or dorsi/plantarflexion planes >1cm or “wobble” when minimal force is applied?
METATARSOPHALANGEAL MOVEMENT. Can the big toe be extended to 90° or greater?
EXCESSIVE SUBTALAR JOINT PRONATION. Children are asked to march on the spot and then on command invert their foot and hold the position close to subtalar joint neutral. Then they are asked to relax. Does the arch lower and flatten fully, excessively and easily with the talus bulging medially, and with no further pronation possible?

Table 3.1 LLAS components. Subject scores one point if the answer for the question is yes. Scores out of twelve for each limb. Reproduced from [97].

Participants were also assessed on the basis of whether they could undertake a single heel rise (SHR). This functional test is commonly used in the assessment of adult acquired flat foot [225]. Participants were asked to stand facing the wall with their hands against the wall for balance. They were asked to stand on tiptoes (double heel rise) to confirm the presence of a medial longitudinal arch of the foot (pre-requisite to the diagnosis of PFF). Subsequently, from a standing position they were asked to lift one foot off the ground by flexing the ipsilateral knee to 90°, and they were asked to perform an SHR. The participant was deemed to have failed the SHR if one or more of the following criteria had been met.

1) Inability to attain the same heel rise height as observed in the double heel rise.

2) Use of the other leg to help attain the heel rise.

3) A technique where two discrete movements were used to complete the task. With the first stage only partly elevating the heel off the ground, while the heel is still in valgus. The next stage, involving a forcible movement to swing the heel into varus, allowing further plantarflexion and completion of the task.

The ability to perform an SHR was recorded as a binary outcome.

Anthropometric measurements included inter-ASIS distance, leg length, knee and ankle width and height. Weight was measured on the force plate in the gait laboratory and the BMI was calculated as per standard methods and this was cross-referenced on child specific WHO BMI charts [350]. Specific foot measures were taken using calipers including foot length, truncated foot length and arch height whilst weight-bearing. Saltzman et al. have demonstrated excellent repeatability for these measurements [272]. Figure 3.4 demonstrates the foot measurements taken.

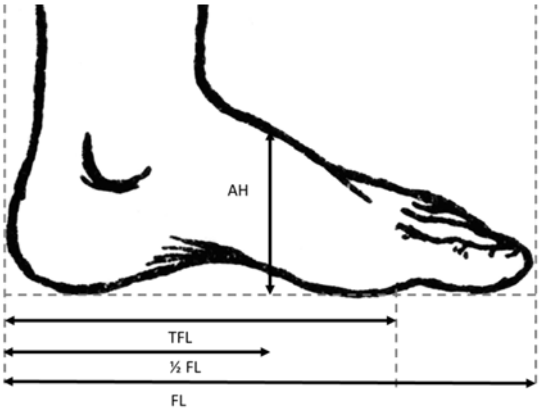
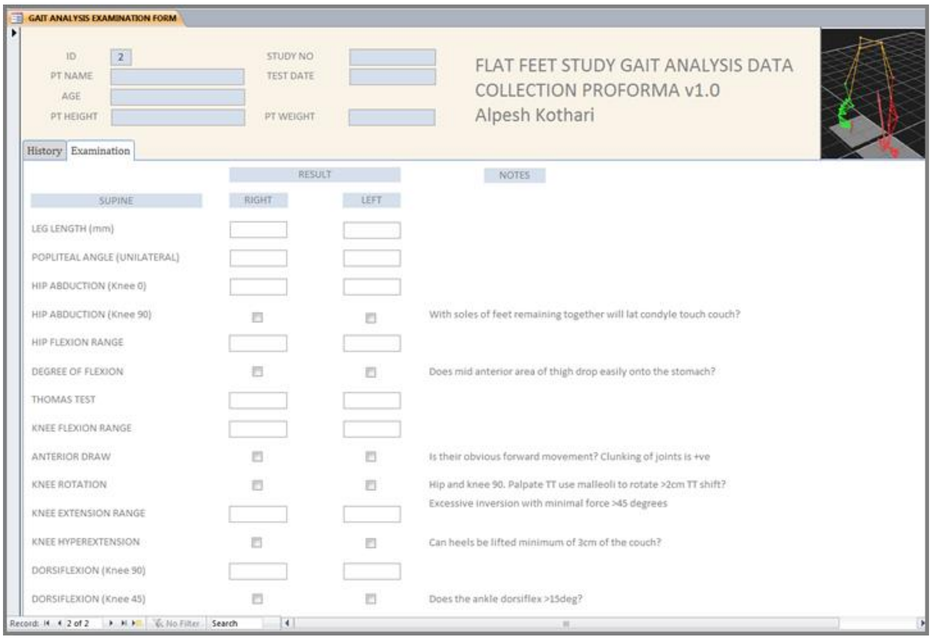


Figure 3.4 Figure demonstrating foot measures. Arch Height (AH) or Dorsal Arch Height (DAH) measured as the distance from the plantar aspect of the foot to the dorsal surface at ½ foot length (½ FL). Truncated foot length is the foot length minus toes (TFL).

History and clinical examination data were entered into a bespoke database (Figure 3.5).



FLAT FEET STUDY GAIT ANALYSIS DATA COLLECTION PROFORMA v1.0
Alpesh Kothari

ID 2
PT NAME
AGE
PT HEIGHT
STUDY NO
TEST DATE
PT WEIGHT

SUPINE	RESULT RIGHT	RESULT LEFT	NOTES
LEG LENGTH (mm)			
POPLITEAL ANGLE (UNILATERAL)			
HIP ABDUCTION (Knee 0)			
HIP ABDUCTION (Knee 90)	☐	☐	With soles of feet remaining together will lat condyle touch couch?
HIP FLEXION RANGE			
DEGREE OF FLEXION	☐	☐	Does mid anterior area of thigh drop easily onto the stomach?
THOMAS TEST			
KNEE FLEXION RANGE			
ANTERIOR DRAW	☐	☐	Is their obvious forward movement? Clunking of joints is +ve
KNEE ROTATION	☐	☐	Hip and knee 90. Palpate TT use malleoli to rotate >2cm TT shift?
KNEE EXTENSION RANGE			Excessive inversion with minimal force >45 degrees
KNEE HYPEREXTENSION	☐	☐	Can heels be lifted minimum of 3cm of the couch?
DORSIFLEXION (Knee 90)			
DORSIFLEXION (Knee 45)	☐	☐	Does the ankle dorsiflex >15deg?

Figure 3.5 Screenshot of gait analysis data collection proforma v1.0.

3.5.4. Gait analysis

Gait analysis was undertaken at the Oxford Gait Laboratory (OGL) at the Nuffield Orthopaedic Centre in Oxford. This is one of only nine gait labs in England accredited by the Clinical Movement Analysis Society UK and Northern Ireland (CMAS). Accreditation requires the institution to adhere to the quality, standards and training outlined by CMAS. This is particularly important in the field of gait analysis as the integrity of data is highly dependent on consistency and accuracy of methods [109].

Before I was able to personally undertake instrumented gait analysis I underwent a period of training under the supervision of Dr Julie Stebbins, operational lead of the OGL. Over a two month period this involved instruction on laboratory calibration, marker placement, data collection and processing. Only after I fulfilled strict criteria outlined by Dr Stebbins was I allowed to independently undertake testing.

The OGL system as of August 2013 consisted of a twelve camera Vicon MX motion capture system (Vicon, Oxford, UK). In December 2013 the hardware was upgraded to a sixteen camera Vicon T40 system (Vicon, Oxford, UK). Despite the changes in the lab, there was no adverse effect on the data. The OGL also had three AMTI force plates positioned in the central walkway (Advanced Medical Technology, Inc. Watertown, MA, USA). In keeping with normal clinical data collection settings, the cameras sampled at 100 Hz, and the force plate at 1000 Hz. Plantar pressure was measured using the Novel EMED-M pressure plate (Novel GmbH, Munich, Germany), sampling at 50 Hz.

Testing procedure

Before the participant attended for his/her assessment session I undertook full calibration of the gait laboratory to minimise motion capture camera image error and ensure accurate functioning of the force plates.

After the history, clinical examination and anthropometric measures were recorded, 9 mm diameter reflective markers were attached using double sided wig tape to the standard locations as dictated by the PiG and OFM gait models (Figure 3.6). The OFM does not give direct information of midfoot function. As such in the first 55 subjects, a marker was also attached to the navicular tuberosity of each foot.

Anthropometric measures were entered into the computer and with the subject standing a static calibration trial was captured. Care was taken to ensure that the knees were not hyperextended, as this can lead to the erroneous definition of joint axes which would in turn alter the subsequent kinematic and kinetic output.

In the cases where subjects had navicular markers as well, a static trial was taken with the child in a sitting position with knees flexed at 90°.

After the static trials D1M, MMA and PCA markers (Figure 3.6) were removed from both feet and then dynamic trials were captured.

For the dynamic trials the participant was instructed to walk normally at a self-selected pace along the 10 m long central walkway in the gait laboratory. Walking trials were repeated until at least five trials were collected which had clean force plate strikes, with both left and right feet. If during data collection any markers required reapplication a second static trial was captured at the end of the walking trials.

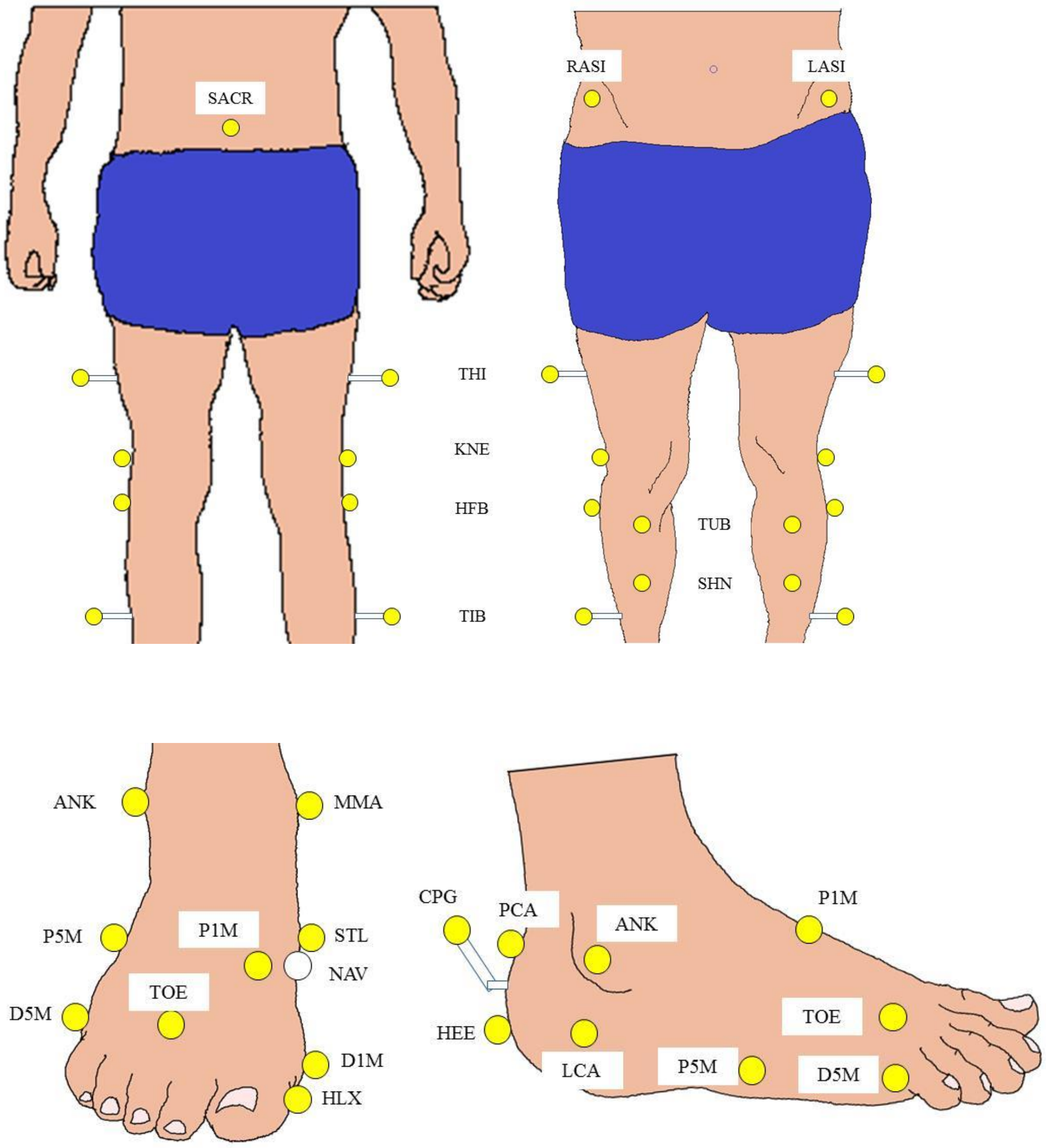


Figure 3.6 Marker locations. SACR - sacrum, ASI -anterior superior iliac spine (R - right, L- left), THI - lateral surface of thigh wand marker, KNE - lateral femoral epicondyle, HFB - head of fibula, SHN - anterior surface of tibia, TIB - lateral surface of shank wand marker, TUB - tibial tubercle, ANK - lateral malleolus, MMA - medial malleolus, HEE - postero-inferior surface of os calcis, PCA - postero-superior surface of os calcis, CPG posterior surface of os calcis wand marker, LCA - lateral surface of os calcis, STL - medial surface of os calcis, TOE - between 2nd and 3rd metatarsal head, D5M - head of 5th metatarsal, P5M - base of 5th metatarsal, P1M - base of 1st metatarsal medial to extensor hallucis longus tendon, D1M - 1st metatarsal head, NAV - optional navicular tuberosity marker.

Dynamic plantar pressure data were collected from each foot. Where possible a three-step technique was used to capture dynamic pressure data [40]. If the participant did not understand the three step technique then a one-step technique was utilized. Whilst the three-step technique is historically regarded as the gold standard technique for plantar pressure measurement, Bus et al. [40] have demonstrated that one-step, two-step or three-step techniques generally yielded similar results. A total of five trials were recorded for each foot. If subjectively it was thought that the participant had altered their stride pattern for a specific trial, "stamped" or just placed their foot on the pressure plate, the trial was discarded. Figure 3.7 gives a pictorial representation of the laboratory setup.

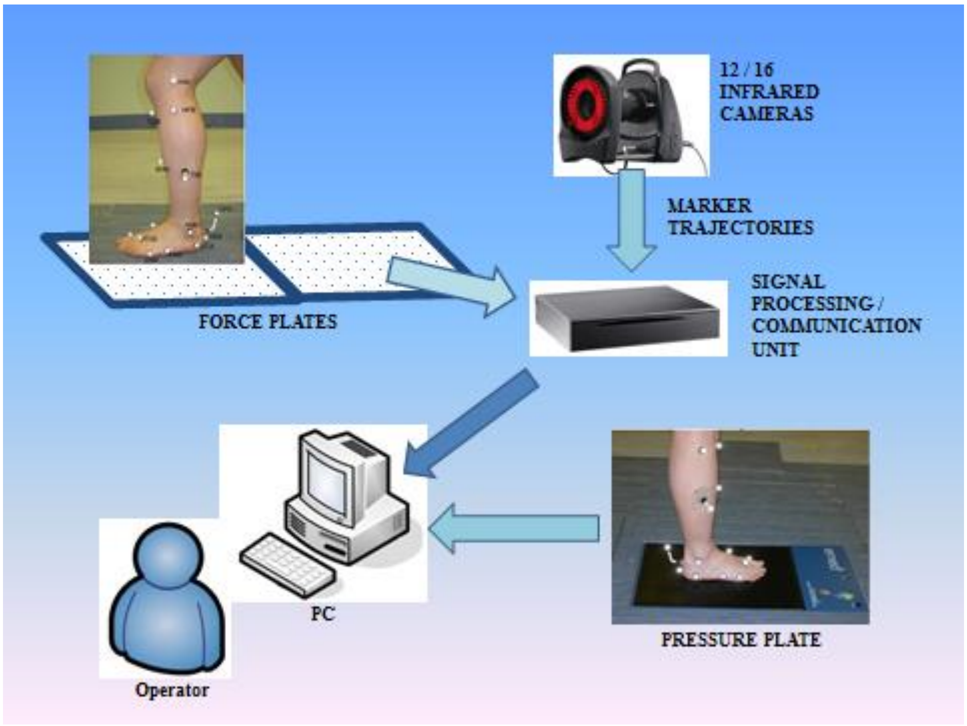


Figure 3.7 Pictorial representation of OGL setup.

3.5.5. Magnetic Resonance Imaging

Magnetic resonance imaging of participants' feet and ankles was undertaken in the radiology department of the Nuffield Orthopaedic Centre, Oxford University Hospitals Trust, Oxford. Imaging was performed using the Philips Achieva 3.0 Tesla MRI scanner (Philips Medical Systems, DA Best, Netherlands).

The imaging protocol included three scout images and a sagittal T1-weighted spin-echo (T1-W) sequence of the foot and ankle to just proximal to the malleoli. The sagittal plane was aligned with long axis of the foot. A SENSE Flex L coil was used to maximise spatial resolution. The T1-W sequence had a pixel size of 0.27 mm, slice thickness 1.5 mm - 2.5 mm, slice increment 2.2 mm - 3.1 mm. Sequence duration was never longer than 10 minutes to minimise distress in the study participants.

To simulate weight-bearing the subject was positioned in a custom-built rig (Figure 3.8). An extra raised footplate was constructed to house one element of the Flex L coil. Ideally the subject was to be loaded with 40% body-weight to each foot based on [346]. Because of the imaging acquisition time this could not be tolerated by the participants, thus a load of 10 kg was applied across both feet. This was validated by comparing the standing arch height with the arch height measured on the MRI scans for 65 feet. Agreement between the two arch height measures (one observer, two observations) was assessed using a Bland-Altman plot as well as calculation of the intra-class correlation coefficient (ICC) using a two-way mixed model with measures of absolute agreement [24]. The method for measuring standing arch height was described in section 3.5.3 (p.83). Measurement of the arch height from the MRI scan firstly involved the identification of the sagittal image with the longest profile of the foot as measured from the posterior-most aspect of the heel to the anterior-most aspect of the toe. Referencing from the posterior-most aspect of the foot, a point at half the foot length was identified at the level of the foot plate (corresponding to plantar aspect of foot) and a line perpendicular to this, bisecting the foot, was drawn. The height of this line from the foot plate to the dorsum of the foot was the provisional arch height. Sagittal images of the foot were then scrolled through medially and laterally to ensure that this was the highest point of the arch. If it was not, the height of the superior-most projection of the arch the same distance along the foot-plate was used as the final arch height measurement.

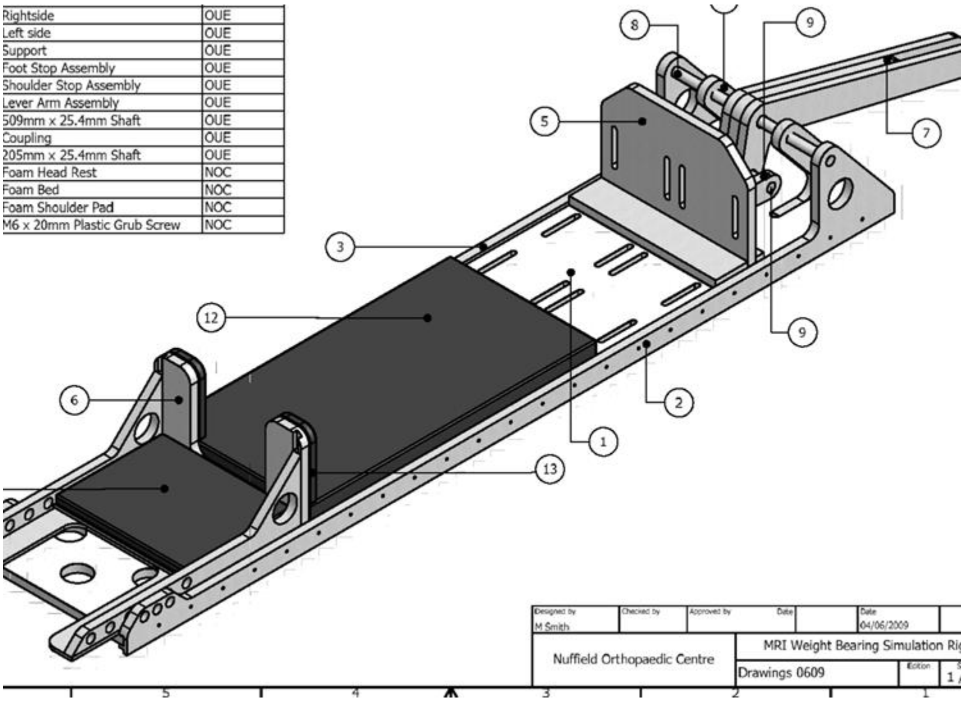


Figure 3.8 MRI Weight-bearing simulation rig [176].

The mean between-subject difference between measurement techniques (standing, MRI) was not significant (0.6 mm, $p > 0.05$) (Figure 3.9). There was no systematic bias in the calculation of arch height between the techniques with the majority of measurement differences falling between the 95% confidence intervals (Figure 3.9).

The ICC was 0.952, which is better than the intra-observer reliability of directly measuring arch height described by Saltzman et al. of 0.91 [272].

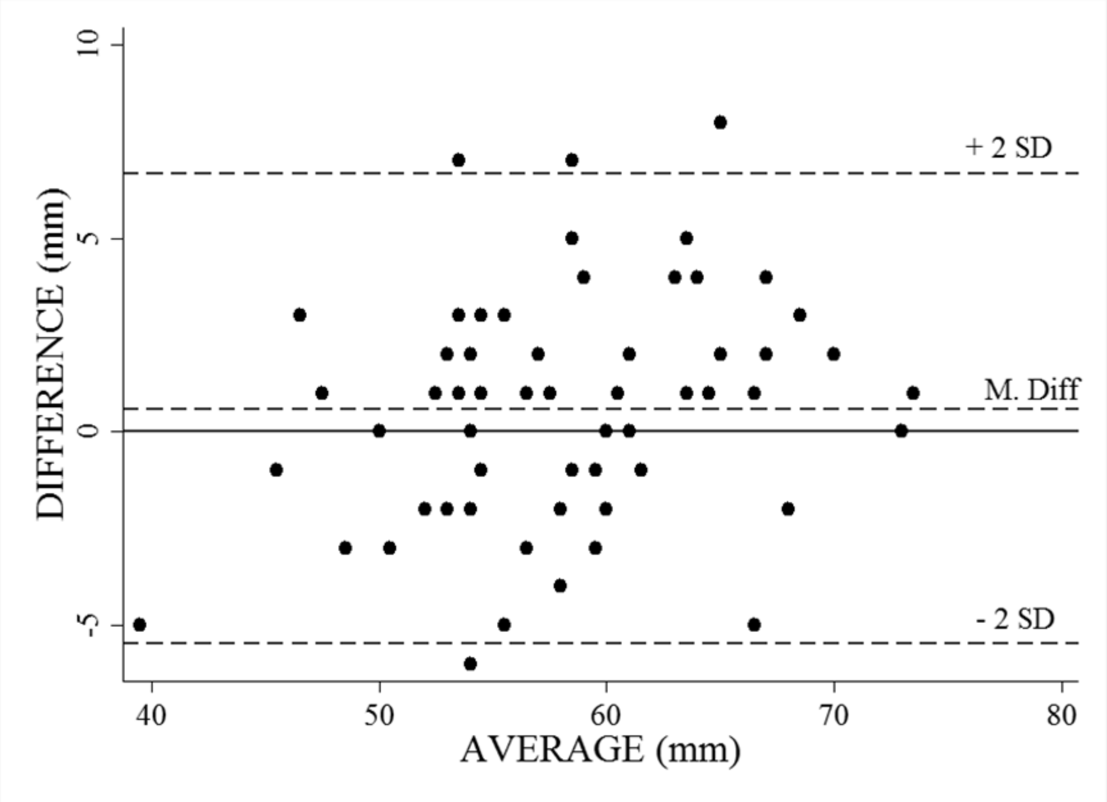


Figure 3.9 Bland-Altman plot demonstrating agreement between two measurement techniques. Confidence intervals are plotted as ± 2 standard deviations from the mean.

3.6. Gait analysis data processing

Vicon Nexus version 1.8.4 (Vicon Motion Systems, Oxford UK) was used to collect and process marker coordinate data. Trials were reconstructed and small gaps were filled using cubic spline interpolation [222]. To fill larger gaps it was assumed that there was a constant distance between markers on the same segment and their trajectories were used to infer missing marker trajectories [78]. The frequency of gaps in marker trajectories was greatly reduced with the upgrade of the OGL motion capture equipment in December 2013. If a gap in marker trajectory was greater than

10 frames the trial was discarded. A Woltring filtering algorithm was used to reduce marker trajectory noise (predicted mean square error setting of 20 mm^2) [347]. Gait events (initial contact and foot off), were determined using force plate data. Events occurring away from the force plates were defined manually. An iterative process was used to alter rotational alignment of the thigh segment by minimising knee abduction and adduction [8].

3.7. Summary

In this chapter the choice of experimental methods has been rationalized, along with a description of study design and the recruitment process. This has been followed by an in depth account of the method by which data was collected and processed. A large amount of data on each study participant was collected, but before it can be utilized to achieve the main objectives of this thesis, the fundamental issue of the classification of a flat foot posture needs to be addressed. This is the focus of the following chapter.

Chapter 4. The classification of paediatric flexible flat feet

4.1. Introduction

Defining foot posture continues to be a source of much debate. With no agreed consensus on the best classification method and over 30 different ways of defining foot posture, deciding which method to use for this thesis was difficult. The modes of classification employed in research and clinical practice can generally be split into four methods.

i) Visual inspection methods

ii) Footprint measures

iii) Radiographic measures

iv) Anthropometric measures

What follows is a brief description of some commonly used measurement indices which were under consideration for use in this study. Published results on the reliability of these measures are summarized in Table 4.1.

i) Visual inspection methods

Classification of foot posture by visual inspection is probably the most common method used in clinical practice [58]. This method usually forms part of the standard clinical assessment of the patient and involves inspecting the foot and ankle in coronal, transverse and sagittal planes. Most methods do not have explicit instructions about how to classify foot posture, therefore the process relies on the clinician's prior experience and training and is inherently subjective. Even when explicit criteria are used to classify foot posture, consistency between observers is variable, with Dahle et al. [71] reporting good inter-rater reliability (Cohen's Kappa 0.72), but Cowan et al. [67] reporting poor reliability (Kendall's Tau 0.35).

To further refine clinical foot posture classification methods and to serve as a benchmark, Redmond et al. [262] devised the foot posture index (FPI). The score was originally based on the sum of individual scores of eight criteria (scored from −2 to +2), but was subsequently refined to six criteria (FPI-6), with a maximum score of +12 and a minimum score of −12 [155]. The inter and intra-rater reliability of the FPI-6 has been reported as being good to excellent [44, 96]. The main criticism with the FPI-6 is that even though the scores give good overall reliability, it is ultimately determined by inherently subjective methods, evidenced by poor individual component reliability [95, 207].

ii) Footprint measures

The footprint method popularized by Harris and Beath [120], has been used for many years as a quick and simple way of assessing foot posture. The simplest method involved an inkpad, but this has now been superseded by the use of pressure plates constructed from pressure transducers (Chapter 3 p.64). The ease of capturing a footprint has resulted in this method being used in many of the larger epidemiological studies of PFF [80, 85, 86]. The footprint can be obtained in the static setting or dynamically with the subject walking across the measuring apparatus. The inherent assumption is that the footprint is representative of the structure and function of the foot and changes in either of these will be reflected in the resulting footprint. A number of footprint indices have been devised to classify foot posture, and the following section provides a brief explanation of five commonly used ones. Figure 4.1 illustrates how these indices are calculated.

The Arch Index

The Arch Index (AI) defined by Cavanagh et al. [52] is a simple ratio of the midfoot contact area divided by the overall contact area of the foot; based on the toeless footprint (Figure 4.1A). The midfoot region is defined as the middle third of the toeless footprint measured along the long axis of the foot. The AI generally has excellent inter- and intra-rater reliability (Table 4.1)[146, 243, 256].

Footprint angle

Also known as the Alfa, or arch angle, the footprint angle as defined by Clarke et al. [61] is measured as the angle between the medial border of the foot and a line drawn from the most medial point of the metatarsal region to the apex of the MLA print concavity (Figure 4.1B). There is some reported variability in the repeatability of this angular measure ranging from fair to excellent (Table 4.1) [243, 256]. This inconsistency is potentially due to the difficulty in identifying the points of interest required to calculate the angle.

Staheli Index (SI) and Chippaux-Smirak Index (CSI)

These two indices are both based on the ratio between the minimum width of the midfoot and the maximum width of the forefoot (CSI) or the heel (SI) (Figure 4.1C) [103, 298]. Both measures are reported to have excellent inter- and intra-rater reliability [243, 256]. These measures rely on continuous foot contact between heel and forefoot; however this is not always present when using the pedobarograph. Any discontinuity in the midfoot will lead to an index value of zero, casting little light on actual foot morphology.

Valgus index

The valgus index as described by Rose et al. [268] relates the transverse plane position of the intermalleolar axis of the ankle with the footprint (Figure 4.1D). A positive index indicates a medial shift of the ankle and a negative one a lateral shift (relative to the foot). Whilst this technique is reported as being repeatable with a standard deviation of 1.2, it is not thought to be a particularly representative classifier of foot posture, as it does not assess the actual midfoot component of the footprint [336].

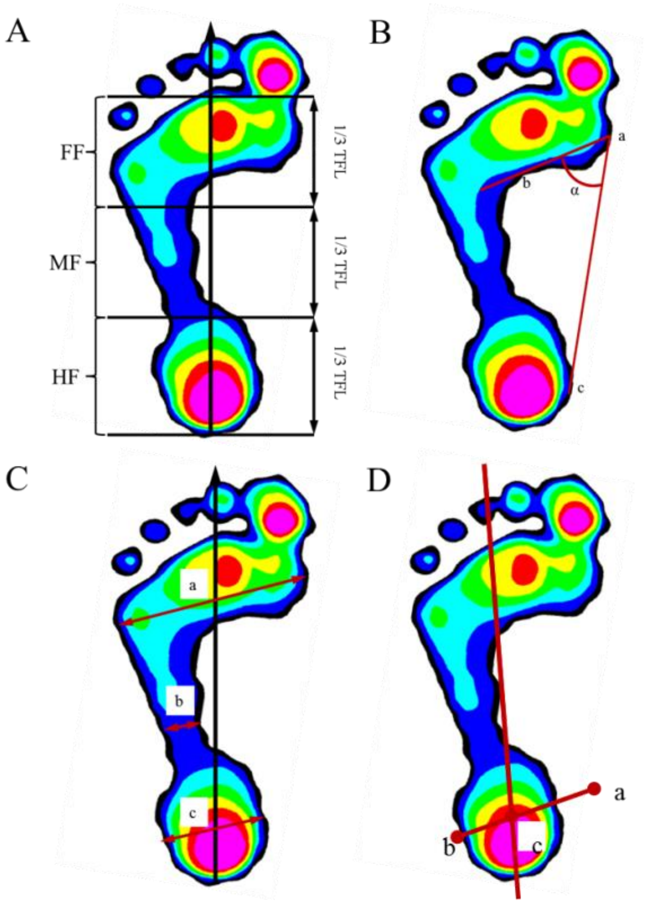


Figure 4.1 Footprint indices. A. Arch index = MF/(FF+MF+HF). TFL is truncated foot length. B. Footprint angle. C. Staheli index = b/c, Chippaux-Smirak index = b/a. D. Valgus index = 0.5ab - ac x (100/ab).

iii) Radiographic measures

A number of different radiographic parameters have been described to classify PFF, based on weight-bearing lateral and AP radiographs of the foot and ankle [325]. Radiographic measures are an attractive option as they make it possible to describe the underlying bony anatomy without interference by soft tissues. Two of the most commonly used parameters are the talo-1st metatarsal angle (T1MA) and the calcaneal inclination angle (CIA) (Figure 4.2). The CIA is measured as the angle between the tangent of the inferior surface of the calcaneus and the platform on which the foot rests. The T1MA is the angle between the long axis of the first metatarsal and the talus. Both measures have good to excellent repeatability (Table 4.1), but as stated in Chapter 3, radiographic techniques use ionising radiation which may preclude their use in research practice [66, 73, 355]. Even in clinical practice radiographs of children with PFF may not always be routinely undertaken.

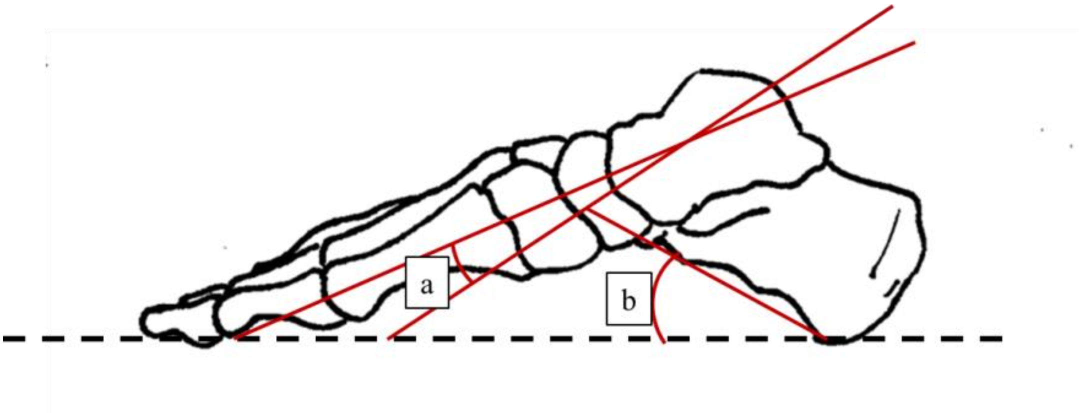


Figure 4.2 Diagram demonstrating the measurement of the T1MA (a) and CIA (b).

iv) Anthropometric measures

Measurement using calipers or goniometers represents a simple method to classify foot posture which can be performed readily in the clinical setting. The dorsal arch height (DAH) is measured as the vertical height of the dorsal aspect of the foot halfway along the length of the foot. This value is divided by the truncated foot length (without the toes) to get the arch height index (AHI) as described by Williams and McClay [343].

As the navicular bone is regarded as the keystone of the MLA the vertical height of the navicular tuberosity (NH) has also been described as a good measure of foot posture, especially when normalized to foot length (NNH) [224].

With hindfoot valgus being another important component of the PFF deformity, the coronal alignment of the hindfoot in standing, also known as the resting calcaneal stance position (RCSP), or rearfoot angle is another way of classifying foot posture.

The majority of anthropometric measures have good to excellent inter- and intra-rater reliability (Table 4.1).

4.1.1. Relationship between measuring methods

A number of researchers have investigated the relationship between different foot classifying methods and have demonstrated a certain amount of redundancy, with some measures correlating quite strongly with others. Table 4.2 summarises these correlations. Chuckpaiwong et al. [58] assessed the correlation between twelve different foot measurement parameters and visual inspection. They demonstrated significant correlations between visual inspection and both anthropometric and footprint measures with Pearson’s R generally greater than 0.65. There also seems to be generally good correlation between different footprint measures [285]. The majority of comparisons between measurement methods have been made with either NH or NNH, and it seems that both correlate well with certain footprint measures and radiological measures [53, 150, 224, 256, 330].

Class of method	Specific method	Inter-rater reliability	Intra-rater reliability
Visual inspection	Clinical assessment	Kendall's Tau: 0.35[67] Cohen's Kappa: 0.72 [71]	
	Foot posture index	0.69 - 0.86 [44, 96]	0.88 - 0.94 [44, 96]
Footprint measures	Arch index	0.89 - 0.96 [243, 256]	0.38 - 1.00 [52, 146, 243]
	Footprint angle	0.61 - 0.81 [243, 256]	0.82 - 0.99 [243]
	Chippaux-Smirnak index	0.96 - 0.97 [243, 256]	0.99 - 1.00 [243]
	Staheli index	0.96 - 0.98 [243, 256]	0.99 – 1.00 [243]
	Valgus index	0.97 [295]	0.98 [295]
Radiographic measures	Calcaneal inclination	0.54 - 0.99 [272, 355]	0.99 - 1.00 R = 0.60 [272, 355]
	Talo-1st metatarsal angle	0.76 [355]	0.86 R = 0.98 [73, 355]
Anthropometric measures	Dorsal arch height	0.56 - 0.99 [62, 201, 202, 204, 272, 314, 342]	0.82 - 0.99 [62, 201, 202, 204, 272, 342]
	Arch height index	0.77 - 0.99 [62, 272, 314, 342]	0.69 - 0.98 [62, 272, 342]
	Resting calcaneal stance position	0.61 - 0.91 [292, 323]	> 0.8 [323]
	Navicular height	0.61 - 0.99 [62, 272, 287, 342]	0.69 - 0.98 [62, 272, 287, 342]
	Normalized navicular height	0.75 - 0.94 [62, 272, 342]	0.62 -0.92 [62, 272, 342]

Table 4.1 Table demonstrating inter- and intra-rater reliability for foot posture measures. Values shown represent the intra-class correlation coefficient unless specified otherwise. R = Pearson's correlation coefficient.

4.1.2. Static measures to assess dynamic function

A criticism of the measurement methods already discussed is that static measures are potentially poor at predicting dynamic function. Cavanagh et al. [51] demonstrated that only about a third of the variation in peak plantar pressures under the heel and first metatarsal head during walking, could be explained by static foot posture measures from standing radiographs. Cashmere et al. [50] reported a poor correlation between static and dynamic DAH measurement. In contrast Williams et al. [343] found that the AHI was the most reliable and valid measure across different weight-bearing conditions.

Due to these concerns, other measures have been developed to gain further insight into the foot. These include navicular drop, dynamic navicular drop and navicular drift [35, 65, 331]. All these measures quantify the motion of navicular in different loading conditions. A full description of these methods is found in Chapter 7 in which they have been utilized to investigate the relationship between midfoot function and foot posture.

	Clinical	FPI	AI	FA	CSI	SI	VI	DAH	AHI	RCSP	NH	NNH	CIA	T1MA
Clinical														
FPI														
AI	0.66 [58]													
FA	0.74 [58]		−0.497 [285]											
CSI	0.66 [58]		0.560 [285]	−0.538 [285]										
SI	0.66 [58]		0.430 [285]	−0.241* [285]	0.468 [285]									
VI			0.039* [150]											
DAH	−0.74 [58]					−0.302 [285]								
AHI	0.699 [58]													
RCSP			0.04* [150]				0.140 [150]							
NH	0.51 [58]	0.44 to 0.45 [231, 285]	−0.46 to −0.73 [108, 197, 285]	0.55 [285]	−0.48 [285]									
NNH	0.77 [58]	0.60 [256]	−0.58 to −0.61 [224, 256]	0.63 [256]	−0.65 [256]	0.07* to −0.62 [256] [271]				0.32 [271]				
CIA			-0.24* to - 0.60 [150, 224]	-0.503 to - 0.51 [330]	−0.21* to −0.51 [53, 330]	−0.27 [53]			0.58 [272]			−0.31 to −0.57 [271, 272]		
T1MA				−0.51 to −0.54 [330]	0.17* to −0.54 [53, 330]	0.37 [53]						−0.57 [271]		

Table 4.2 Published correlations between foot posture classification indices. * Where correlation not significant.

4.1.3. Selecting an ideal measurement method

For the purposes of this study choosing a method to classify foot posture was difficult. There are numerous ways one could measure foot posture and there is no consensus on which is the best. A perfect method for posture classification would be quick and easy, safe, reliable, and clinically relevant. Most methods are quick, easy and reliable, and most are safe apart from the potential risks of using ionizing radiation posed by radiographs. However of all these ideal characteristics, clinical relevance is one which is seldom demonstrated by the methods described.

An option would be to arbitrarily select an individual published method and proceed with that. Another option would be to use an alternative approach like the Delphi method to get a consensus of expert opinion [183]. Neither of these options necessarily take into account whether the method is good at defining foot posture groups. Chuckwaipong et al. [58] and Murley et al. [224] suggest that the best way to classify foot posture is to employ a number of different indices. Again it is not clear which indices are the best to use. An alternative approach is to define foot posture using statistical inference. If one wants to classify the foot posture of a population of children, there are methods, like cluster analysis which assess the spread of the data from multiple variables and split the population into groups [235]. The end result is groups or clusters in which subjects are arranged such that those in the same group/cluster are more similar to each other (with respect to foot posture), than to those in other groups/clusters. Using a data driven approach can also provide further insight into how different measures relate to each other. This is the approach used in this chapter.

4.2. Aim

The primary aim of this chapter is to construct a tool using a data-driven approach to provide an objective method to classify foot posture.

Secondary aims are to assess how different foot posture classification methods relate to each other and how the classification tool compares to clinical assessment.

4.3. Methods

As there is still no consensus on the best way to classify foot posture, a method was devised to incorporate radiological, footprint and anthropometric measurements. This tool was constructed using data from each foot of the first forty participants of the study (80 feet). The mean age with standard deviation of children in this sample was 12.10 (± 2.06) years old and the male : female ratio was 21:19. In total six separate indices were calculated (Table 4.3).

Foot posture classification index	Index acronym
Arch height index	AHI
Arch index	AI
Calcaneal inclination angle	CIA
Normalized navicular height	NNH
Resting calcaneal stance position	RCSP
Talo-1st metatarsal angle	T1MA

Table 4.3 Summary of foot posture classification indices used in this chapter.

Visual classification of foot posture (neutral or flat) was also performed based on whether increased hindfoot valgus and planus were observed when the participant was standing. Based on visual classification of the 80 feet, 52 were classified as flat and 28 as neutral.

4.3.1. Calculation of classification indices

The AHI was calculated as described earlier in this chapter, using the measurement methods described in Chapter 3 (p.83). The AI was calculated and outputted directly from the EMED-M pressure plate software (Gaitline, Geometry, Novel GmbH, Munich, Germany), and based on the maximum pressure footprint (p.97). The AI for a representative dynamic pedobarography trial was used for each foot. Navicular height was recorded as the vertical height of the reflective marker placed on the navicular during quiet standing. This was obtained from the motion capture software (Vicon Nexus v1.7, Vicon, Oxford UK). This was then normalized to foot length to calculate NNH. The average OFM hindfoot/tibia angle in the coronal plane during quiet standing (static hindfoot valgus), was used as the RCSP.

The CIA and T1MA were calculated from the simulated weight-bearing MRI using Mimics v14.0 (Materialise, Leuven, Belgium). To measure the CIA the sagittal images of the foot were scrolled through until the image with the most inferior projection of the anterior tubercle of the calcaneus was identified. A line from this point to the most inferior point of the medial calcaneal process was then drawn and the angle between this and the plantar surface of the foot was measured (Figure 4.3 i)).

The T1MA was calculated by subtracting the angle between the long axis of the talus and the horizontal, from the angle formed by the first metatarsal and the horizontal. The long axis of the talus was defined by finding the sagittal image of the foot with the largest cross-section of the talus. The superior and inferior margins of the articular surface were marked and then tangents of the superior and inferior surface of the talus were marked. The long axis of the talus was the line lying halfway in-between these lines (Figure 4.3 ii)). The long axis of the first metatarsal was defined as the line running through the middle, and parallel to the shaft of the bone when it was in largest profile on the sagittal images (Figure 4.3 ii)).

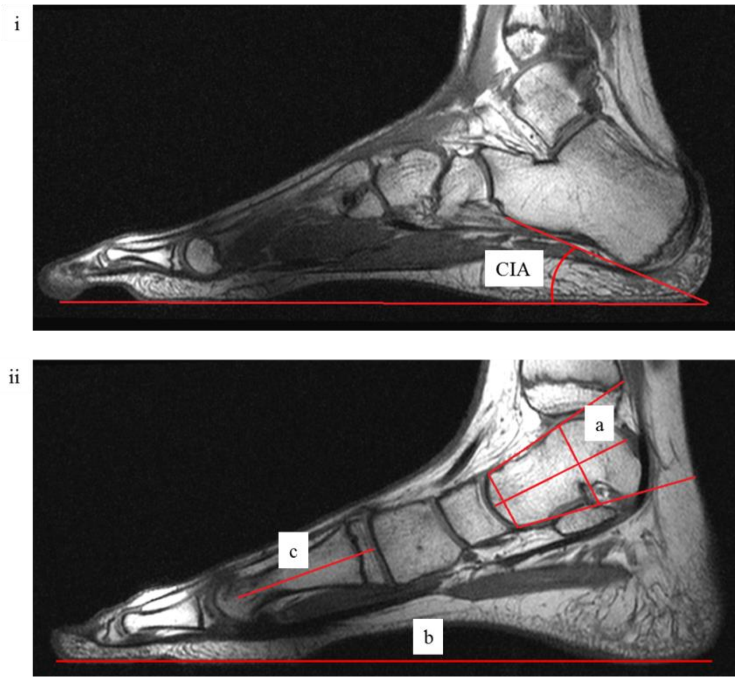


Figure 4.3 i) Measurement of CIA. ii) T1MA calculated from subtracting the angle between long axis of talus (a) and horizontal (b), from the angle between the long axis of the 1st metatarsal (c) and horizontal (b).

4.3.2. Statistical analysis

As the method for describing the CIA and T1MA from MRI scans was novel, assessment of inter-observer reliability of angle measurement was performed by two raters; the author (AK) and SB, a consultant radiologist. Intra-rater reliability was also assessed by AK repeating angle measurements on a separate occasion separated by one month. Reliability was quantified using the intra-class correlation coefficient, using a two-way mixed model with measures of absolute agreement, for twenty feet.

In this study a cluster analysis procedure was used to reveal natural groupings in foot posture inherent in the data. A two-step cluster analysis method was used due to its unique advantage of automatically selecting the number of clusters that best fit the data, and a mechanism to handle outliers if present. Feet were clustered using the log-likelihood distance measure. The number of clusters was determined by the software using Bayesian Information Criterion as the clustering criterion. The outlier handling option was used to identify feet that did not fit well into any of the clusters. One of the assumptions of cluster analysis is that input variables should not be collinear, as this can adversely affect the analysis and distort the results. For this analysis, collinearity was regarded as problematic if Pearson's correlations between two variables was greater than $R = 0.6$ [147]. In cases where such correlations were observed a practical approach was used to reduce the number of variables used in the cluster analysis. Subjects were recruited into the study on the basis of having either a flat or normal foot posture, thus it was hypothesized that the cluster analysis method would output only two clusters; with foot measurement indices consistent with those expected for flat and normal foot postures.

Once the clusters had been established, logistic regression was used to construct a foot posture classification tool. The ability of the individual classifiers used in the regression model to predict foot posture was assessed by measuring the area under the receiver operator characteristic (ROC) curves.

Cohen’s Kappa was used to assess agreement between the foot posture classification tool and classification using a visual inspection method [171].

4.4. Results

4.4.1. Reliability of radiological measurements

The radiological measurements both demonstrated excellent inter- and intra-rater reliability as documented in Table 4.4, ICCs of greater than 0.9.

	Type of ICC	Inter-rater	Intra-rater
CIA	Single measures	0.919	0.970
	Averaged measures	0.958	0.985
T1MA	Single measures	0.945	0.979
	Averaged measures	0.972	0.989

Table 4.4 Reliability of radiological measurement within and between raters. The ICCs for both single and average measures are given.

4.4.2. Cross-correlation between indices

Statistically significant correlations were noted between most of the pairs of foot posture indices used in this study (Table 4.5). The only non-significant correlations were between AI and RCSP (R = 0.051 p = 0.663), and CIA and RCSP (R = 0.058, p = 0.637). Pearson’s correlation coefficients of above 0.6 were observed between AHI and NNH, AHI and T1MA and NNH and TIMA. To avoid violation of aforementioned statistical assumptions, two of these indices had to be excluded from the cluster analysis. As the AHI is the simplest and easiest index to measure out of the three this was retained for further analysis.

	AHI	**AI**	**NNH**	**CIA**	**T1MA**	**RCSP**
AHI	1					
AI	-0.477 <0.001	1				
NNH	0.673* <0.001	-0.477 <0.001	1			
CIA	0.529 <0.001	-0.580 <0.001	0.478 <0.001	1		
T1MA	-0.703* <0.001	0.498 <0.001	-0.734* <0.001	-0.573 <0.001	1	
RCSP	0.343 <0.001	0.051 0.663	0.273 0.025	0.058 0.637	-0.379 0.002	1

Table 4.5 Cross correlation matrix for footprint classification indices. Correlation coefficient with significance level below. * signifies absolute value of R >0.6.

4.4.3. Results of cluster of analysis

The optimal number of clusters to describe the data was two, and there were no outliers identified in the data. Table 4.6 summarises the descriptive statistics for the foot posture indices for all the feet used in the cluster analysis as well as the two groups. Group 1 was defined as NF and group 2 as PFF. The table includes NNH and T1MA even though they were not used in the clustering to allow further appraisal of groups.

	All Feet (n=80)		Group 1 (n=35)		Group 2 (n=45)	
Variable	Mean	SD	Mean	SD	Mean	SD
AHI	0.30	0.03	0.32	0.03	0.28	0.02
AI	0.19	0.09	0.15	0.07	0.26	0.08
CIA(^{o})	18.03	4.54	20.12	3.20	14.33	4.19
RCSP(^{o})	-5.78	5.26	-3.84	3.73	-9.19	5.84
T1MA	7.21	7.60	3.2	4.48	13.83	7.08
NNH	0.14	0.04	0.15	0.03	0.11	0.03

Table 4.6 Summary of foot classification indices for all feet and cluster groups. Negative values for RCSP indicate an everted heel. Group 1 is NF and group 2 is PFF. T1MA and NNH not used in cluster analysis, but data present for group appraisal.

4.4.4. Foot classification tool

As two groups were defined using the cluster analysis method logistic regression was used to assess the relative contribution of each variable to groups. The resulting group membership prediction tool was:

$$Group\ = (AHI * -72.06) + (AI * 14.77) + (CIA * -0.51) + (RCSP * -0.28) + 25.45 \qquad 4.1$$

The equation to quantify the specific group membership probability:

$$Probability = \frac{1}{(1+\exp(group))} \qquad 4.2$$

If the probability < 0.5, then the foot belongs to group 1 (NF). Otherwise, the foot belongs to group 2 (PFF).

The ROC curves for each individual index to predict group membership as defined by the cluster analysis are illustrated in Figure 4.4. The area under the curve represents how good the index is at classifying foot posture with unity being perfect classification (only possible with all four classifiers). As suggested by the coefficients in equation 4.1, AHI was the best at classifying foot posture with it correctly classifying over 90% of cases. The worse individual classifier was RCSP which could only correctly classify 75% of cases.

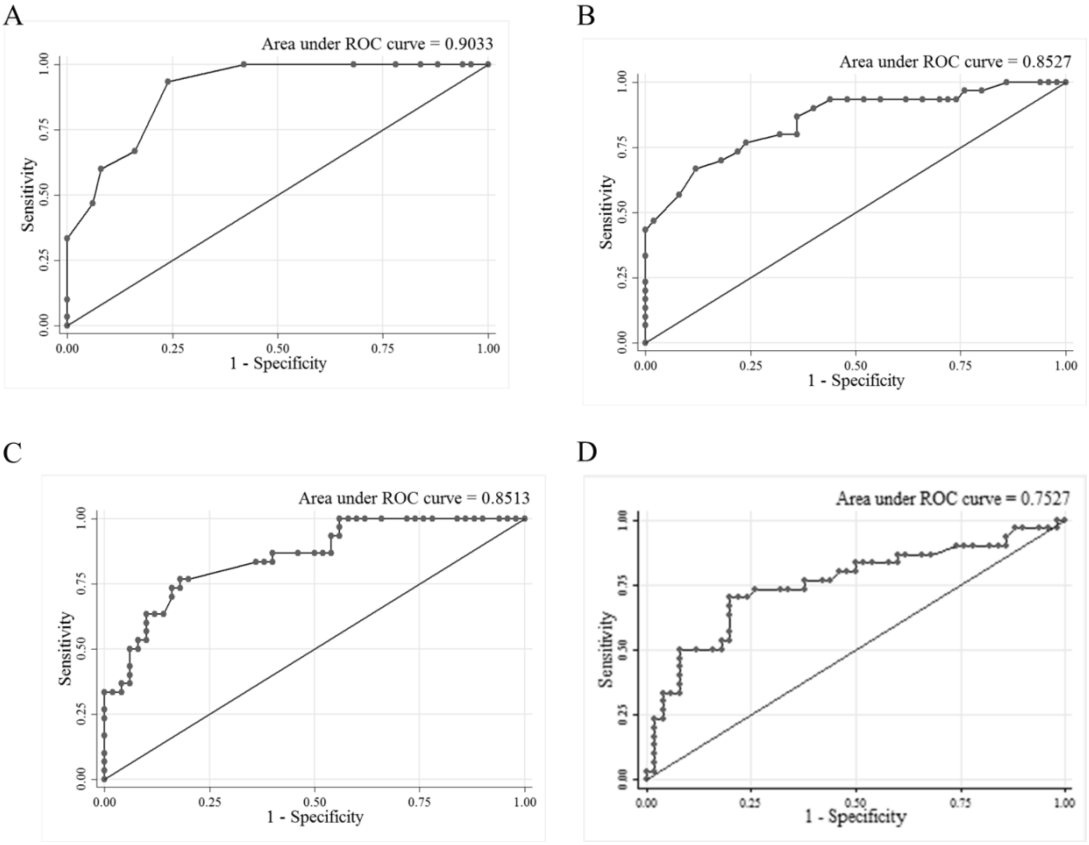


Figure 4.4 Receiver operating curves. A. AHI alone, B. AI alone, C. CIA alone, D. RCSP alone.

4.4.5. Comparison between cluster analysis and expert opinion

	Expert Flat	Expert Neutral	Total
Cluster Flat	40	5	45
Cluster Neutral	12	23	35
Total	52	28	80

Table 4.7 2 x 2 table demonstrating agreement between expert clinical assessment and foot posture classification tool.

There was only agreement between clinical examination and the cluster analysis results for 63 of 80 feet (78.8%) (Table 4.7). Cohen’s Kappa to measure agreement was 0.588, regarded as moderate agreement according the criteria outlined by Landis and Koch [171].

4.5. Discussion

The aim of this chapter was to utilise an objective method to classify foot posture. The rationale behind this was that, firstly visual inspective methods have been proven to be subjective and of poor reliability, and secondly there is no absolute consensus on which method is the best. Children recruited into this study had an a priori classification of either a flat or normal foot posture, and a data driven approach using the 80 feet of the first 40 subjects was used to construct a classification tool that could best distinguish between groups with different foot posture. Six commonly used foot posture measurement methods were used in this study, AHI, AI, NNH, T1MA, CP and RCSP.

The first stage of the classification process involved assessing for correlation between the measurement indices. Whilst this was a necessary procedure for the cluster analysis process, it also provided further insight into the inter-relation between foot measurement indices. There were significant correlations between most pairs of indices, and this was in keeping with much of the previously published literature. The strongest correlations were between AHI and T1MA, AHI and NNH, and NNH and T1MA. The navicular forms the keystone of the MLA, thus the fact that NNH and AHI correlated strongly was unsurprising. Radiological measures like the T1MA are felt to be superior to other measures as they actually assess the underlying bony alignment, without being affected by the soft tissue. The fact that both NNH and AHI correlated so strongly with T1MA would suggest that the variability of soft tissue contribution is much less than the variability in arch height. There was no significant correlation between RCSP and AI, or between RCSP and CIA, highlighting the fact that the RCSP quantifies a different aspect of the flat foot deformity compared to the other two measures. This emphasises the benefit of using a classification tool that utilises multiple indices.

The next stage involved using a hierarchical cluster analysis algorithm to group subjects into separate clusters. The number of clusters was not pre-determined, and a method that would identify outliers was selected. Reassuringly only two clusters were defined with no specific outliers, confirming the belief that children with two types of foot posture were recruited into the study. The resulting foot measurement index values for the 80 feet in total and two separate foot posture groups are summarized in Table 4.6. On the basis of the groupings, group 1 was classified as NF and group 2 as PFF.

The mean AHI for the flat group was 0.28 compared to the neutral group of 0.32, both with standard deviations of 0.03. There is no published set of normative AHI values for children, but normative values have been published in adults. Williams and McClay [343], who developed the measure, suggested that values of AHI over 1.5 times the standard deviation less than the mean population value should be regarded as having a flat foot posture. In Williams and McClay’s initial normative database of 102 feet the mean AHI was 0.316, but in numerous, larger studies the population mean appears to be closer to 0.343 with a standard deviation of 0.030 [42, 202, 335].

Thus the mean AHI of the PFF group of 0.28, and mean AHI of the NF group of 0.32 are consistent with the published literature.

With respect to the AI, Cavanagh and Rodgers [52] stated that a flatfoot posture in adults would have an AI of greater than or equal to 0.26, and a neutral foot posture had an AI of between 0.21 and 0.26. These published values were based on footprints obtained using an inkpad and paper. The AI in this study was calculated using the pedobarograph. The first difference is that compared to the near infinite spatial resolution of inkpad and paper, the pedobarograph has a sensor size of 0.25 cm^2. This ultimately limits how well the foot can be mapped for calculation of the AI. Secondly each sensor has a trigger threshold of 10 kPa, thus part of the foot may conceivably be in contact with the plate without triggering the sensor. Urry et al. [321] have demonstrated that these factors result in consistent under-estimation between the AI calculated using dynamic pedobarography and classical techniques like the inkpad. Bearing this in mind, a mean AI of the PFF group of 0.26 seems reasonable.

It is generally agreed that the normal value of the CIA is between 18° and 20°, with values less than that consistent with a flat foot [151]. In this sample, PFF subjects had a mean CIA of 14°, which was similar to findings by a number of studies [213, 249, 354]. The mean CIA of the NF group was also similar to that found in the literature [224].

Zero degrees is regarded as the normal value of the T1MA; with greater than four degrees being classified as a flat foot posture in adults [110, 206]. Vanderwilde et al. [325] studied normal ranges of numerous foot and ankle radiographs in children and demonstrated that the mean T1MA of nine year olds was between five and six degrees. The PFF group mean T1MA of 14° is similar to that of flatfooted subjects in the existing literature [53, 110, 213, 354].

Root et al. [267] suggested that a RCSP of greater than four degrees of valgus in adults was consistent with a flat foot posture, whereas Sobel et al. [294] in their study of 150 normal children demonstrated a mean population RCSP of four degrees of valgus with a range of zero to nine degrees. The mean RCSP of the NF group in this study was four degrees of valgus which is consistent with Sobel et al. [294]. The mean RCSP of the PFF was just over nine degrees of valgus, falling outside of the range of observed values by Sobel et al., and similar to values observed in a group of flatfooted children by Jay et al. [142].

The NNH values of the NF and PFF group were almost exactly the same as those observed by Xiong et al. [353] in their investigation of the foot posture of 48 adults.

A comparison was made between the foot classification tool and visual inspection of foot posture. Cohen’s Kappa was 0.558, which, based on cut-offs by Landis and Koch [171] would be classified as moderate agreement. Visual inspection only had 78.8% agreement with the classification tool. As there is no well accepted gold standard classification method one cannot say that the cluster analysis method is better than the clinical assessment, just that there is a difference between the two methods. The method used in this study is however quantitative and thus makes it easier to compare with existing literature, and it is based on the actual data as opposed to a subjective internal representation of what constitutes PFF. For cases of particularly severe PFF, visual inspection and the foot classification tool would be equivalent. The classification tool is likely to be more relevant when the foot posture is equivocal.

The resultant PFF and NF groups from the cluster analysis had foot measurement indices consistent with previously published literature. The logistic regression classification tool was subsequently used to classify feet as PFF or NF as new participants entered the study. In four cases, the subject was close to the dividing line between groups, such that one foot was classified as PFF and the other as NF. This highlights a problem with dichotomisation as even though both feet were very similar in all respects, a small difference between feet led to the straddling of the dividing line between groups. From previous literature, it is well documented that both feet tend to have the same foot posture, and for the purposes of statistical analysis it was important to define whether the subject, not individual was flatfooted or not [213, 221, 354]. Thus in the few cases where each subject's foot was classified differently the subject was given the overall classification of having PFF. This decision was made because the foot posture classification tool was very strict in its classification of PFF and the measurement indices of the NF group were erring towards the flatter side. This decision was also consistent with the clinical appearance of the feet.

At the termination of this study 95 children had been recruited. Figure 4.5 demonstrates the distribution of the groups as well as source of recruitment into the study, and subject demographics.

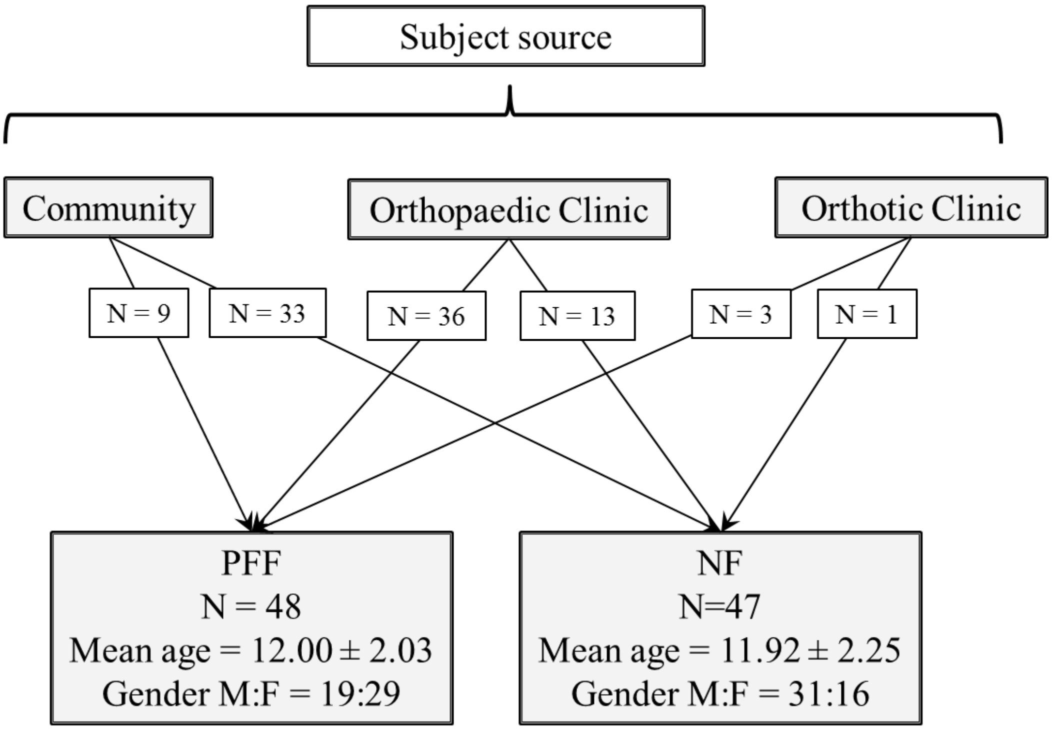


Figure 4.5 Flow chart demonstrating origin of study participants and group characteristics.

A dichotomous classification is also not representative of the fact that foot posture is continuously distributed. From a clinician's perspective, a simple binary outcome or classifier can be helpful in guiding management. However a problem is that measuring a continuous variable with a binary measure results in significant loss of data resolution. This can result in reduced statistical power, requiring larger sample sizes with an increased risk of a type II error. It has been demonstrated that using binary instead of continuous outcomes is at best only 67% as efficient in statistical analyses [307].

For the purposes of this thesis a continuous measure of foot posture for data analysis was desirable. To maintain consistency with the methods used to group feet, the continuous measure of foot posture could have been the probability of group membership calculated using equation 4.2. However this is quite an esoteric quantity, which is removed from the practicalities of clinical practice. The AHI was selected as the best single variable for quantify foot posture in a continuous manner as it was the strongest predictor of foot posture group membership, being able to correctly classify over 90% of feet (Figure 4.4).

4.6. Limitations

The methods used in this study to calculate some of the foot measurement indices did vary somewhat from the classical description of the methods. The most striking difference was with the methods used to measure CIA and T1MA, which are conventionally measured from standing radiographs, but in this case were measured from semi-weight bearing MRI. In Chapter 3 (p.90-91) assessment was made as to whether the MRI jig simulated weight-bearing conditions adequately, and it was found that there was no systematic bias between arch height measured standing and that measured on the MRI. The protocol for measuring CIA and T1MA from the MRI scans was adapted to closely resemble that used when measuring from radiographs, although the absolute validity of this has not been tested. The values of CIA and T1MA for the groups were consistent with previous literature, thus the conclusion was that these measures were comparable. The measurement of CIA and T1MA also had similar intra- and inter-rater reliability to the standard measurement methods [73, 272, 355].

Normalized navicular height and RCSP were also measured using an alternative method, 3DGA. Using such a method may potentially be more precise than standard caliper methods, because a well-calibrated motion analysis laboratory has an image error of less than 0.1 mm, which is significantly better than human precision. A recent study also demonstrated no significant difference in the navicular height measured using a marker based method compared to an ultrasound method to locate the navicular tuberosity [312]. Using ultrasound to locate the most prominent point of the navicular tuberosity would be regarded as more accurate than the standard method of measuring navicular height based on palpation alone. Thus one would expect the method used in this study to measure navicular height to be at least comparable to standard methods. The protocol and landmarks for applying the hindfoot markers from which the RCSP was computed were the same as the standard measurement method, with markers defining the heel segment being placed on a line bisecting the heel as viewed posteriorly. The OFM actually outputs the position of the heel relative to the tibia, as opposed to standard RCSP measurements which reference the position of the heel to the vertical. It is accepted that this is a potential source of discrepancy, especially in cases of significant lower limb malalignment.

4.7. Conclusion

The main aim of this chapter was to provide a robust method to classify foot posture. PFF and NF groups were defined using a composite measure of the arch height index, arch index, calcaneal inclination angle and resting calcaneal stance position. As a singular measure, the arch height index proved to be the best method to classify foot posture, and was deemed to be the most appropriate method of measuring foot posture in a continuous fashion. The results of this chapter are applied throughout the rest of this thesis.

Chapter 5. What is the impact of PFF on health-related quality of life?

5.1. Introduction

To justify further investigation into, and definition of management protocols for, PFF one must be able to demonstrate that this presentation can adversely affect children. There is sparse literature confirming the pathological nature of PFF, and indeed there is no published literature evaluating the impact of PFF on the HRQOL of children. Up until recently it would have been only possible to measure HRQOL in these children using generic tools. Now with the advent of the OxAFQ-C it is also possible to assess the impact of PFF on HRQOL using a site-specific tool. These tools also have proxy questionnaires to be completed by the parent or guardian as well as patient self-reported questionnaires. The main utility of the proxy questionnaire is in circumstances where the child may not be able to complete the form themselves, for example if the child is too young, cognitively impaired, too unwell or too fatigued [121]. In the context of this study these circumstances did not occur, however from the research perspective it is still desirable to get the perspective of the parents/guardians. It is a widely held belief that PFF causes more anxiety to the parents than it causes problems to the child [100, 297]. As it is usually the parent/guardian who accesses the healthcare system on behalf of the child, it is of interest to compare self-report and proxy questionnaires for consistency and bias [46, 141, 328].

5.2. Aims and hypotheses

The aim of this chapter is to use the PedsQL™ 4.0 and OxAFQ-C to investigate HRQOL in study participants. There are two main hypotheses:

1. There will be good reliability and no systematic bias between patient and proxy questionnaire scores in PFF and NF subject groups.

2. A significantly decreased HRQOL will be demonstrated in children with PFF compared with children with NF. It is expected that the biggest differences will be in the physical domain scores.

5.3. Methods

5.3.1. Data collection and participants

In Chapter 3 (p.78-80) there is an in depth description of the PedsQL™ 4.0 and OxAFQ-C as well as a description on how questionnaire scores are calculated. Patient self-reported were completed for all 95 children recruited to this study, with 48 having PFF and 47 NF as defined by the foot posture classification technique described in Chapter 4. Participant characteristics are summarised in Figure 4.5 (p.118). Proxy questionnaires were collected from the parents of 80 children (PFF = 44, NF =36).

For comparative purposes self-reported OxAFQ-C questionnaires were collected in two other patient groups: 1) Children with symptomatic treated clubfoot (n=13, mean age 9.7 years old, Male : Female, 13:0) and 2) Children with foot and ankle problems associated with cerebral palsy (n=8, mean age 11.4 years old, Male : Female, 6:2).

5.3.2. Statistical Analysis

Reliability between patient and proxy questionnaires was assessed using the absolute agreement intraclass correlation coefficient (ICC) and comparing mean absolute differences with a paired two-tailed t-test as per recommendations by Marshall et al. [192]. According to the recommendations by Fleiss et al. [101] and Cicchetti et al. [60] an ICC of < 0.40 was classified as poor agreement, 0.4 - 0.59 as fair, 0.60 - 0.74 as good and > 0.74 as excellent. Questionnaire domain scores were compared between subject groups using an unpaired two-tailed t-test. As AHI is used as a continuous measure of foot posture in subsequent chapters, Pearson's R was used to assess the correlation between it and OxAFQ-C scores. Pearson's R was also used to assess the correlation between OxAFQ-C and PedsQL™ 4.0 domain scores. Alpha was set at 0.05 to define significance.

The sample size was calculated using previously published data by Morris et al. [219]. With the published upper limit of minimally important difference of 17% in the OxAFQ-C physical domain, and a maximum standard deviation of 25.2% a sample size of 47 was required in each group for 90% study power. Statistical analysis was undertaken using Stata v.13.0 (Statacorp LP, Texas, USA) and SPSS v 22.0 (IBM, Chicago, IL, USA).

5.4. Results

Patient self-reported questionnaires for both the OxAFQ-C and the PedsQL™ 4.0 were obtained from all study participants. Proxy questionnaires were obtained from 80 of 95 participants. A full set of proxy questionnaires was not obtained as some parents only attended to sign the consent form for the study and then had to depart for work-related reasons. None of the questionnaires that were completed had any missing data.

5.4.1. Comparison between patient and proxy questionnaire scores

Indices quantifying agreement between the patient and proxy questionnaires for all the study participants are summarized in Table 5.1. There was fair to excellent agreement between subjects and parents/guardians for the majority of questionnaire domains except for PedsQL™ 4.0 social and emotional domains which only had poor agreement (ICC = 0.38). The physical domains for the OxAFQ-C and PedsQL™ 4.0 demonstrated the highest ICC with excellent agreement between proxy and self-reported questionnaires (0.87 and 0.86 respectively). As a combined population there were no significant absolute differences between self-reported and proxy domain scores for any of the OxAFQ-C questionnaire domains. There was however a significant difference between self-reported and proxy PedsQL™ 4.0 physical and emotional domain scores with parents/guardians consistently reporting worse functioning than the child.

All subjects	n	Mean diff.	Sig.	SD of difference	95% Confidence interval		ICC
OxAFQ-C Physical	80	−0.50	0.979	17.60	−3.97	3.87	0.87
OxAFQ-C School & Play	80	−1.60	0.242	12.40	−4.40	1.13	0.84
OxAFQ-C Emotional	80	−2.10	0.313	18.60	−6.24	2.02	0.64
OxAFQ-C Extra Q.	80	−0.94	0.750	26.20	−6.77	4.89	0.81
PedsQL™ 4.0 Physical	80	−3.28	0.010*	11.20	−5.76	−0.81	0.86
PedsQL™ 4.0 Emotional	80	−5.93	0.005*	18.47	−10.01	−1.84	0.38
PedsQL™ 4.0 Social	80	−0.09	0.962	16.40	−3.72	3.54	0.38
PedsQL™ 4.0 School	80	0.68	0.711	16.40	−2.96	4.31	0.60

Table 5.1 Reliability between patient self-reported and proxy questionnaire scores for all subjects for which self-report and proxy questionnaire data was collected. n denotes sample size. Mean diff. is the average of the difference of patient self-reported questionnaire subtracted from the paired proxy questionnaire. Significance of mean diff. is given (sig.), as well as the standard deviation of the difference (SD of difference) and the 95% confidence intervals of the mean diff. * denotes statistical significance. ICC denotes the intraclass correlation coefficient.

Table 5.2a and 5.2b summarise the indices quantifying agreement between the self-reported and proxy questionnaires when the population was split into PFF and NF groups. In general there was a tendency for parents of children in the PFF group to over-estimate the impairment in HRQOL experienced by the children. This reached statistical significance in the PFF group for the PedsQL™ 4.0 Physical (p = 0.026) and the OxAFQ-C School & Play (p = 0.041) domains scores. In the NF group negative bias was only observed in absolute mean score difference in the PedsQL™ 4.0 emotional domain (p = 0.002). Better ICCs were observed for OxAFQ-C domain scores than PedsQL™ 4.0 domain scores. Self-reported and proxy questionnaire scores for physical domains again demonstrated greater agreement in the groups than alternative domains. Due to any potential bias in proxy questionnaire scores only self-reported questionnaire scores were used in subsequent analyses.

PFF	N	Mean diff	Sig	SD of difference	95% Confidence interval		ICC
OxAFQ-C Physical	44	−3.50	0.230	19.10	−9.31	2.30	0.83
OxAFQ-C School & Play	44	−4.68	0.041*	14.77	−9.17	−0.20	0.81
OxAFQ-C Emotional	44	−5.82	0.091	22.32	−12.60	0.96	0.57
OxAFQ-C Extra Q.	44	−5.11	0.262	29.82	−14.18	3.95	0.75
PedsQL™ 4.0 Physical	44	−4.09	0.026*	11.90	−7.67	−0.52	0.85
PedsQL™ 4.0 Emotional	44	−3.67	0.236	20.46	−9.81	2.48	0.52
PedsQL™ 4.0 Social	44	0.29	0.920	19.19	−5.47	6.05	0.27
PedsQL™ 4.0 School	44	2.77	0.277	16.94	−2.31	7.87	0.65

a)

NF	N	Mean diff	Sig	SD of difference	95% Confidence interval		ICC
OxAFQ-C Physical	36	4.17	0.100	14.18	−0.84	9.18	0.78
OxAFQ-C School & Play	36	2.08	0.103	7.47	−0.44	4.61	0.84
OxAFQ-C Emotional	36	2.43	0.206	11.30	−1.39	6.26	0.60
OxAFQ-C Extra Q.	36	4.17	0.226	20.27	−2.69	11.02	0.81
PedsQL™ 4.0 Physical	36	-2.26	0.200	10.37	−5.76	1.25	0.76
PedsQL™ 4.0 Emotional	36	-8.75	0.002*	15.40	−14.00	−3.51	0.46
PedsQL™ 4.0 Social	36	−0.55	0.790	12.40	−4.75	3.64	0.52
PedsQL™ 4.0 School	36	−1.94	0.461	15.64	−7.23	3.35	0.38

b)

Table 5.2 Reliability between patient self-reported and proxy questionnaire scores for PFF (2a) and NF (2b) groups. Abbreviations as per Table 5.1. * denotes statistical significance.

5.4.2. Correlation between OxAFQ-C and PedsQL™ 4.0 domain scores

Correlations between the OxAFQ-C domain scores and PedsQL™ 4.0 domain scores were all positive and statistically significant at $p \leq 0.01$ (Table 5.3). Thus higher domain scores in one HRQOL were associated with higher domain scores in the other and vice versa. The correlation between the physical domains of the OxAFQ-C and the PedsQL™ 4.0 was high ($R = 0.803$). The corresponding questionnaire's emotional domains demonstrated moderate correlation ($R = 0.447$). Both OxAFQ-C emotional and school & play domains also had moderate correlations with the PedsQL™ 4.0 physical domain scores (R = 0.520 and R = 0.677 respectively). The remainder of the domains with dissimilar constructs had weak correlations of R < 0.4.

		PedsQL™ 4.0 Domain			
		Physical	Social	Emotional	School
OxAFQ-C Domain	Physical	0.803	0.295	0.314	0.371
	School & Play	0.677	0.326	0.309	0.296
	Emotional	0.520	0.254	0.447	0.444

Table 5.3 Pearson's correlation coefficients between HRQOL questionnaire domain scores.

5.4.3. Comparison of HRQOL measures between PFF and NF children

PFF children had significantly lower mean scores in all OxAFQ-C questionnaire domains compared to the NF children. The differences were most marked for the physical domain and extra question item, with PFF children scoring around 20% less than NF children. In the emotional and school & play domains differences between PFF and NF children were just under 10%. Results are summarized in Table 5.4.

Analysis of the PEDSQL™ 4.0 domain scores showed significantly lower mean scores for the PFF children compared to the NF children in all but the social domain, which itself was tending towards significance (p = 0.097) (Table 5.4). The differences in mean scores were most marked for the physical domain (12.96%, p < 0.001).

	PFF		**NF**			
	Mean Score	SD	Mean Score	SD	Mean Difference	Sig
OxAFQ-C Physical	63.45	23.78	83.24	20.31	19.79	<0.001
OxAFQ-C School & Play	86.06	18.56	94.54	10.39	8.48	0.007
OxAFQ-C Emotional	85.28	17.59	94.81	11.46	9.52	0.002
OxAFQ-C Extra Q.	66.15	32.82	87.23	23.81	21.09	<0.001
PedsQL™ 4.0 Physical	78.06	15.78	91.02	10.02	12.96	<0.001
PedsQL™ 4.0 Emotional	82.08	19.53	88.4	15.78	6.32	0.043
PedsQL™ 4.0 Social	88.02	16.30	91.7	10.44	3.68	0.097
PedsQL™ 4.0 School	78.64	17.97	87.98	12.88	9.33	0.005

Table 5.4 Mean domain scores with standard deviation (SD) for PFF and NF groups. Mean difference as well as significance level (sig) tabulated. * denotes statistical significance.

Correlations between AHI and OxAFQ-C domains scores were all statistically significant such that a reduced AHI (flatter foot) was associated with a worse HRQOL (Table 5.5).

OxAFQ-C Questionnaire Domain	Correlation coefficient (R)	Significance (p)
Physical	0.37	< 0.001*
Emotional	0.29	0.005*
School & Play	0.25	0.015*
Extra question	0.25	0.012*

Table 5.5 Correlation between OxAFQ-C domain scores and subject average AHI. * Denotes statistical significance.

5.4.4. Comparison of OxAFQ-C domain scores between children with PFF and children with symptomatic clubfoot and children with foot and ankle problems associated with cerebral palsy

A broad range of OxAFQ-C domain scores were observed in the PFF, clubfoot and cerebral palsy groups (Figure 5.1). The children with cerebral palsy generally had worse scores in all domains as compared to the other two groups. The PFF and clubfoot groups had similar domain scores.

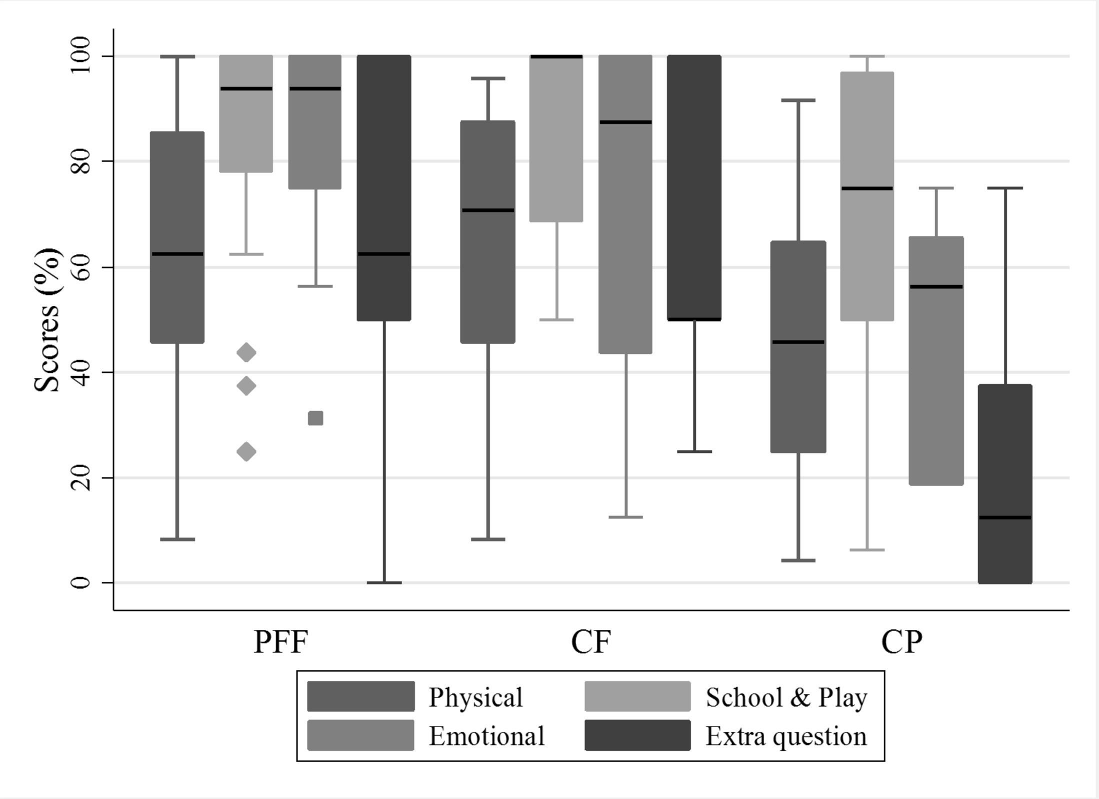


Figure 5.1 Box-whisker plot demonstrating OxAFQ-C domain scores of PFF group compared to that of children with clubfoot (CF) and foot problems dues to cerebral palsy (CP).

5.5. Discussion

The classification and management of PFF continues to be the subject of considerable debate in the paediatric orthopaedic community. Whilst it is evident that there are structural and functional differences between NF and PFF, whether these are of any clinical relevance remains unclear. In the limited literature assessing the clinical impact of PFF children are grouped on the basis of presence or absence of symptoms [17, 129, 213, 339]. Clinicians would agree that children with symptomatic PFF may present in a variety of ways and that the distinction is not black and white. Using binary groups in such a fashion results in loss of data resolution, and does not accurately represent the more continuously distributed nature of symptoms [305]. The impact of symptoms on HRQOL is important. In this chapter, better insight into HRQOL of PFF children has been obtained through the use of two validated HRQOL measures, the OxAFQ-C and the PedsQL™ 4.0 [218, 327].

In the first part of the chapter, the consistency between patient self-reported and proxy questionnaire domain scores was evaluated. As a combined group there was, generally, fair to excellent consistency as measured by the ICC. Consistency was better for the OxAFQ-C than the PedsQL™ 4.0. Significant negative bias was found between patient self-reported and proxy scores for the physical and emotional domains of the PedsQL™ 4.0 with parents/guardians suggesting worse functioning than the child reported. Achenbach et al. [1] found that parents were much better at judging more observable external problems in their child, like aggressiveness, than internalized problems like anxiety or sadness. This might explain why overall we see better consistency with more observable problems like physical impairment than other less observable issues like emotional or social functioning.

When the subject groups were split into PFF and NF, an interesting phenomenon was observed. Parents of children with PFF consistently gave scores lower than the children themselves. This was statistically significant for the OxAFQ-C school and play, and PedsQL™ 4.0 physical domains. These findings suggest that parents perceive PFF to have more negative consequences than do the children themselves. Similar findings were also found by Ennett et al. [90] when they demonstrated that mothers of children with juvenile idiopathic arthritis felt that their child was more affected by the disease than did the child.

The main purpose of this chapter was to compare the HRQOL between PFF and NF children. It was hypothesized that children with PFF would have worse HRQOL than NF children. This was demonstrated both with the OxAFQ-C and PedsQL™ 4.0 questionnaires. When using HRQOL measures such as these, statistical differences between subject/group domains scores need to be put in the context of what the MID for the domain is. Anything below this difference may not have any clinical importance and as such not be an important finding. Published MID for the PedsQL™ 4.0 for each domain is 4 points [327]. For the OxAFQ-C, the MID varies between domains, with it being 10-17% for the physical domain and 7-9% for the school & play and emotional domains [217]. In this study, mean questionnaire domain scores differences between NF and PFF groups were all at or above the MID values, except for the PedsQL™ 4.0 social domain. The greatest differences between the groups as hypothesized were observed in the physical domain scores for both HRQOL measures.

Statistical analysis also demonstrated significant positive correlations between averaged AHI and OxAFQ-C domain scores. Thus the flatter the foot the worse the HRQOL.

The OxAFQ-C has been used in two recent studies to quantify burden of disease in other patient groups. Duffy et al. [84] used it to assess HRQOL in children treated for clubfoot. In their sample, children who were surgically treated had mean child reported OxAFQ-C domain scores between 74.0% and 88.4%. The Ponseti group had mean OxAFQ-C domain scores between 81.9% and 95.7%. The PFF group in this study had worse child-reported OxAFQ-C scores in all domains compared to the Ponsetti group and in all but the emotional domain compared to the surgically managed group. Kennedy et al. [157] assessed HRQOL related to foot and ankle abnormalities in Hurler’s Syndrome. They reported an average OxAFQ-C score of 44.7 out of 60; however, as an overall combined score has not been validated, direct comparison with the children in this study cannot be made.

To put the PFF OxAFQ-C domain scores in further context a descriptive comparison was made with questionnaire scores from a small sample of children with symptomatic treated clubfoot and children with foot and ankle problems due to cerebral palsy (Figure 5.1). For all groups, broad ranges of domain scores in the OxAFQ-C were observed. This highlights the heterogeneity of the populations and the need to assess HRQOL. The median scores of the PFF were similar to the clubfoot group. As a group, cerebral palsy children generally had a worse HRQOL. The broad range of PFF domain scores demonstrated that some individuals with PFF may have as bad if not worse domains scores than clubfoot and cerebral palsy children.

In this study, the OxAFQ-C has been used in a normal population (NF) as well as in a pathological group (PFF). The tool itself was developed in a pathological population and as such there are no published normative data. As this is the case, the absolute domain scores are harder to interpret. To legitimise use of the tool in this context, we also used the well-established and validated PedsQL™ 4.0 which does have normative data. In comparison with this normative data, it firstly seems that the NF population scores better in all domains than the healthy children in the study by Varni et al. [327]. It is unclear why this is the case, although it may be related to the relatively small sample size and specific inclusion criteria in our study. When looking at individual domain scores, as a group, PFF children have worse mean physical domain scores than the published values for acutely unwell children and similar scores to chronically unwell children. For the other domains, the PFF children have similar domain scores to the healthy children. Correlation between PedsQL™ 4.0 and OxAFQ-C domain scores was particularly strong for the physical domain, and thus the impairment seen in the PFF can be placed in a broader context. Correlations in other domains were less strong, and thus the scores in the other OxAFQ-C domains are less widely applicable. Discrepancies between the two HRQOL measures may be in part due to the fact that the PedsQL™ 4.0 is a generic tool whereas the OxAFQ-C pertains solely to foot and ankle pathology. If the correlations were perfect then the tools would be measuring the same thing, and thus there would be no point having both.

In this chapter, children with PFF, as a group, demonstrated a broad range of questionnaire scores in all domains. The impairment observed varied from essentially normal function in some children with PFF, all the way to significant deterioration in quality of life seen in others. These findings highlight the heterogeneity in clinical presentation of children with PFF, which in turn makes it difficult to define treatment protocols. In an attempt to simplify things, clinicians have further divided PFF children into those who are asymptomatic and those who are symptomatic. The evidence presented in this chapter suggests that this is inappropriate, as a binary definition is not sensitive to the variety of impairments that PFF can cause in children. A binary classifier may also be insensitive to clinical change or improvement – just because a symptomatic child does not become asymptomatic does not mean that his or her symptoms have not improved. As a result it is suggested that when assessing a child with PFF the clinician should spend time elucidating the nature and extent of symptoms and the effect on quality of life. Use of the OxAFQ-C to achieve this in such a context could be very helpful. If treatment is instituted, repeat administering of the questionnaire will also make it possible to chart improvement. From a research and audit point of view, this is particularly important in the context of PFF as the benefit of treatment remains uncertain.

5.6. Limitations

An important limitation of this study is that there may have been some selection bias in the recruiting of subjects. As the majority of children with PFF were recruited from an orthotic or orthopaedic clinic, this may only represent the tip of the ‘clinical iceberg’, with the asymptomatic or mildly symptomatic majority in the community not represented. An objective method to classify foot posture was used to minimise any additional clinician bias in subject selection. Even if there remains some selection bias, the aim of this chapter was not to describe the epidemiology of PFF, but to demonstrate that a proportion of affected children have significantly impaired

HRQOL compared to healthy controls. This impairment may be equivalent to, or worse than, acutely or chronically unwell children.

A final limitation is that as this study is cross-sectional in nature the natural history of foot and ankle symptoms and impaired quality of life due to PFF is unknown. Only a longitudinal study would be able to assess whether these symptoms remain static, get worse, or improve over time.

5.7. Conclusion

In conclusion, even though parents may overestimate the severity of their child’s impairment, children with PFF do have significantly impaired HRQOL when compared to NF children. This is particularly evident with respect to physical functioning and confirms the belief that PFF cannot always be regarded as just a benign normal variant. The diagnosis of PFF alone, however, is not enough to guide clinical management and careful consideration should be given to the child’s symptom profile and HRQOL.

It has now been demonstrated that children with PFF have significantly impaired HRQOL compared to children with NF. The next step is to understand the relationship between the structure and function of PFF and HRQOL. This is investigated in subsequent chapters of this thesis.

Chapter 6. Clinical and anatomical associations with PFF

6.1. Introduction

In the previous chapter the PedsQL™ 4.0 and the OxAFQ-C were used to illustrate that children with PFF had significantly worse HRQOL than children with a normal foot posture. Having shown this, it is important to establish the clinical and anatomical factors that relate to PFF. Identifying these factors may help, firstly to further our understanding of the pathomechanics of PFF and secondly may help guide management strategies.

From the literature presented in Chapter 2 (p.34-37), commonly accepted determinants of PFF include gender, ethnicity, BMI and flexibility [86, 180, 221, 250]. Paediatric flexible flatfoot has also been associated with genu valgum, increased external tibial torsion and increased femoral anteversion, although the evidence supporting these findings is limited and conflicting [181, 291, 356]. The main theories of the pathogenesis of PFF focus on the effect of ligamentous laxity, and muscle control influencing the static and dynamic alignment of the bones in the foot. However, some authors support the notion that bony structure may also be important in determining foot posture. Particular interest is focused on the anterior subtalar articulation, with some authors speculating that the absence of this articulation may afford less support to the talar head resulting in a flat foot deformity [10, 36]. This has yet to be demonstrated in vivo.

Aside from clarifying the aetiology of PFF, the ongoing challenge for the clinician is to differentiate between children who would benefit from intervention and those who would not. Any clinical sign which relates to impaired HRQOL or function would be helpful in the management decision making process. Currently, contracture of the plantarflexors is the only purported indicator of a potentially symptomatic flatfoot [120, 129]. The sensitivity and specificity of this finding to identify problematic PFF has not been tested.

A second clinical factor that could be useful in the assessment of children with PFF is the single heel rise test (SHR). This test is used in adults with acquired flatfoot to identify those with different disease stages, but its routine use in the paediatric population is not well established [164, 225].

6.2. Aims and hypotheses

The first aim of this chapter is to gain further insight into the aetiology of PFF by assessing the association between clinical and anatomical factors and PFF. It is hypothesized that increased flexibility, increased BMI, lower limb malalignment and altered subtalar joint morphology will be associated with a flatter foot posture.

The second aim is to assess the utility of two clinical signs, tight plantarflexors and failure of the SHR test, to identify children with more problematic PFF. It is hypothesized that both tests will demonstrate good sensitivity and specificity for identifying children with PFF and significantly reduced HRQOL.

6.3. Methods

6.3.1. Data collection

Clinical examination of study participants was undertaken as per the methods described in Chapter 3 (p.55-92). Age, BMI, gender, ankle range of motion and SHR were collected for all 95 study participants (48 = PFF, 47 = NF). Skeletal alignment data (including coronal knee alignment, femoral anteversion and tibial torsional profile), and LLAS (p.82) were collected for all but one participant (n = 94, 47 = PFF, 47 = NF).

Assessing ankle range of motion using standard goniometry has been proven to be of poor reliability [88, 352]. As such contracture of the plantarflexors was quantified in all study participants using a binary method, namely whether on clinical examination, whilst controlling the hindfoot in an inverted position, both ankles could passively achieve plantigrade with the knees extended in the supine position.

Eighty four of the 95 children underwent MRI scans of their feet with their feet in a simulated weight bearing posture (46 = PFF, 38 = NF). Some participants only had one foot scanned as they could not tolerate any longer in the MRI scanner due to discomfort or claustrophobia. Images of 127 feet in total were obtained.

Using the physical domain scores of the self-reported OxAFQ-C questionnaires the study participants in the PFF group were dichotomized into two groups, more symptomatic (MS) and less symptomatic (LS) study participants. More symptomatic participants were defined as those with OxAFQ-C physical domain scores at, or worse than, the minimally important difference (MID) for the questionnaire compared to the NF group. The mean OxAFQ-C physical domain score of the NF group was 83.24%, thus children with questionnaire scores less than (83.24% − MID) were grouped together as MS and the remainder grouped together as LS. As the published MID for the OxAFQ-C physical domain is 10% - 17% both values were used in subsequent analysis. Thus when 10% was used as the MID of the 48 children with PFF 31 were classified as MS, and 17 in the LS. When 17% was used as the MID, 25 were classified as MS and 23 as LS.

6.3.2. Assessment of subtalar joint morphology

The sagittal T1-weighted MRI images were sequentially inspected using Mimics v14.0 (Materialise, Leuven, Belgium). A protocol similar to that described by Shahabpour et al. [283] was used to classify the subtalar joint of each foot. The classification used in this thesis was that described by Bruckner et al. [36] (Figure 6.1).

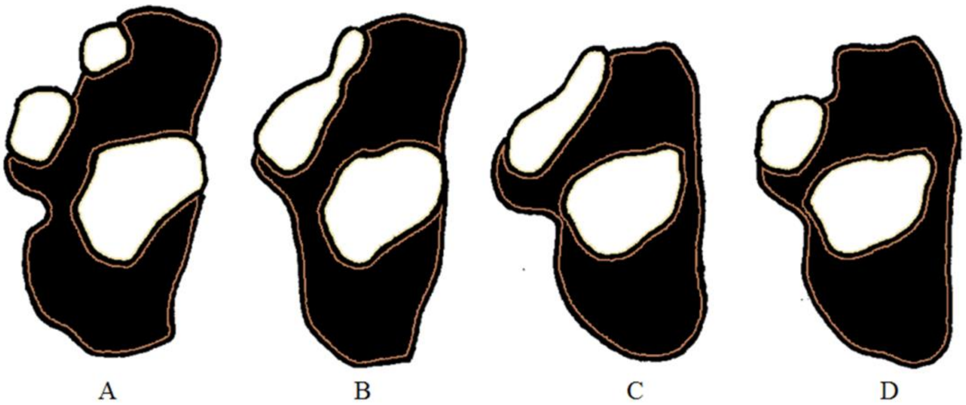


Figure 6.1 Variation in os calcis articulating surface morphology described by Bruckner. A has three separate articular surfaces (anterior/middle/posterior). B and C have the anterior and middle articular surfaces joined. D is missing the anterior articular surface (adapted from [36]).

An example of the visualisation of the three articular facets of the subtalar joint in one subject is shown in Figure 6.2 As these articular facets were not in continuity this corresponded to a Bruckner A classification.

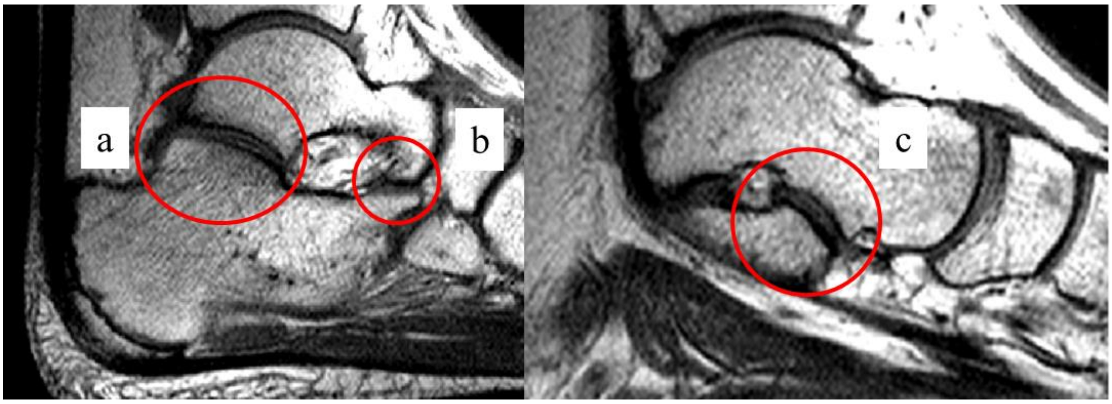


Figure 6.2 Subtalar joint morphology. Example of one subject. Discrete posterior (a), middle (c), and anterior (b) articular facets of the os calcis can be seen clearly.

To assess the validity of this method to define subtalar morphology intra- and inter-rater reliability of subtalar joint classification in 20 feet was assessed, using two raters; the author (AK) and a consultant radiologist (SB). Intra-rater reliability was assessed by AK repeating the classification on two occasions separated by a two month gap. Cohen’s kappa was used to quantify agreement, and this was reported as per recommendations by Landis and Koch [171].

The inter-rater reliability for the two raters was found to be, Kappa = 0.718, 95% CI [0.600 - 0.836]. Intra-rater reliability was found to be, Kappa = 0.863, 95% CI [0.773 - 0.953]. Whilst both were regarded as, at least, substantial agreement, difficulty was experienced in differentiating between Bruckner type B and C articular morphologies. On this basis, and because the main feature of interest was presence or absence of the anterior articulation, the articular morphology classification was further simplified to a binary outcome; supportive (Bruckner A, B, C) and unsupportive (Bruckner D). A supportive articular surface had an anterior articulation and the unsupportive did not. Using this classification inter-rater reliability and intra-rater reliability both improved to 0.886, 95% CI [0.776 - 0.996]. This would be regarded as almost perfect agreement. The binary classification was used in subsequent regression analysis.

6.3.3. Data analysis

6.3.3.1. Predictors of AHI

The first stage of data analysis involved assessing the relationship between individual clinical factors and foot posture as measured with the AHI. For continuous variables this involved construction of scatter plots with linear regression lines (including 95% confidence intervals). Pearson's correlation coefficients were calculated for each pair of variables. Subject specific measures of age and BMI were plotted against mean AHI of both left and right feet. Absolute BMI was used instead of age and gender specific centiles as it was normally distributed. Limb specific measures including LLAS and measures of skeletal alignment were plotted with right and left lower limbs pooled. For the categorical measures, box-whisker plots were constructed. An unpaired, two-tailed Student's t-test was used to compare mean AHI values between categorical groups.

Clinical factors that demonstrated an association with AHI, with a significance level of < 0.1 (subtalar joint morphology, knee coronal alignment and lower limb flexibility measured using the LLAS) were then assessed in combination as potential predictors for AHI using stepwise multiple linear regression. Regression standard errors were adjusted to account for the existence of paired limb data, where required. This was essential to avoid the statistical violations of paired data discussed in previous chapters. Regression diagnostics were used to ensure that model residuals were normally distributed and that the residual versus fitted values did not demonstrate any heteroscedasticity. Interaction between individual predictors was also assessed. The multiple linear regression analysis of 84 subjects had 80% power to detect an overall model effect size (f^2) of 0.12, which represents a moderate effect size [64]. The regression effect size (f^2) is calculated as follows:

$$f^2 = \frac{R^2}{1- R^2} \tag{6.1}$$

Sample size and power calculations were made using GPower (v3.1.9.2 Universität Kiel, Germany).

6.3.3.2. Clinical tests and PFF

Two by two contingency tables were constructed for both the ability to perform SHR, and whether it was possible for the ankle to passively achieve plantigrade, by foot posture. Two by two contingency tables were also constructed for clinical sign versus level of symptoms (MS or LS) in the PFF group, and Fisher's exact test was used to assess the significance of the distribution. Sensitivity and specificity of the clinical signs to identify MS children were calculated including 95% confidence intervals. As the published MID for the OxAFQ-C physical domain is 10% - 17%, both values were used to assess how adjusting the threshold affected the ability of the clinical signs to identify MS children [218].

As there was no previous literature to indicate what the expected proportion of children able to perform the SHR test and achieve plantigrade is, no formal power analysis could be made.

Data analysis was undertaken using SPSS v22 (IBM) and Stata v13.0 (Statacorp Texas). Alpha was set as 0.05.

6.4. Results

6.4.1. Relationship between clinical and anatomical factors and AHI

Figure 6.3 and 6.4 demonstrate the relationship between individual clinical and anatomical factors and AHI. Of the continuous variables assessed, only LLAS ($R = -0.37$, $p < 0.001$) and knee varus/valgus ($R = 0.23$, $p = 0.002$) demonstrated correlation with AHI with significance levels < 0.1 (Figures 6.3 c) and e) respectively). Of the categorical variables a significant level of < 0.1 was only found for the relationship between AHI and subtalar joint morphology (Figure 6.4 b)).

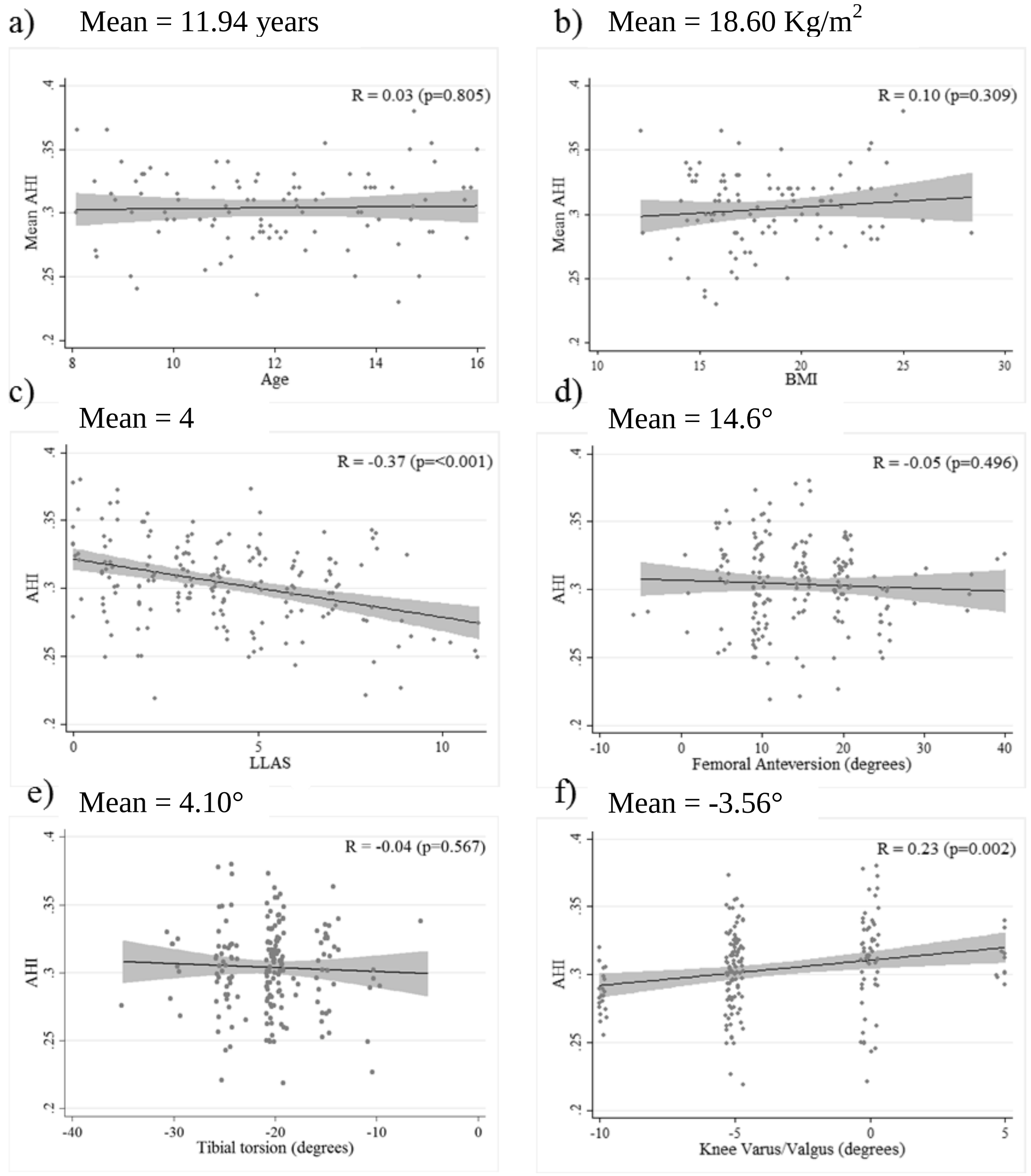


Figure 6.3 Scatter plots demonstrating, a) Mean AHI vs Age, b) Mean AHI vs BMI, c) AHI vs LLAS, d) AHI vs Femoral Anteversion, e) AHI vs Tibial Torsion and f) AHI vs Knee Varus/Valgus (varus +ve, valgus -ve). Line of best fit plotted on each graph as well as 95% confidence intervals. Plots annotated with Pearson's R and significance level (p). Jitter applied to graph d, e and f to avoid superimposition of data points. Mean values of variables above individual graphs.

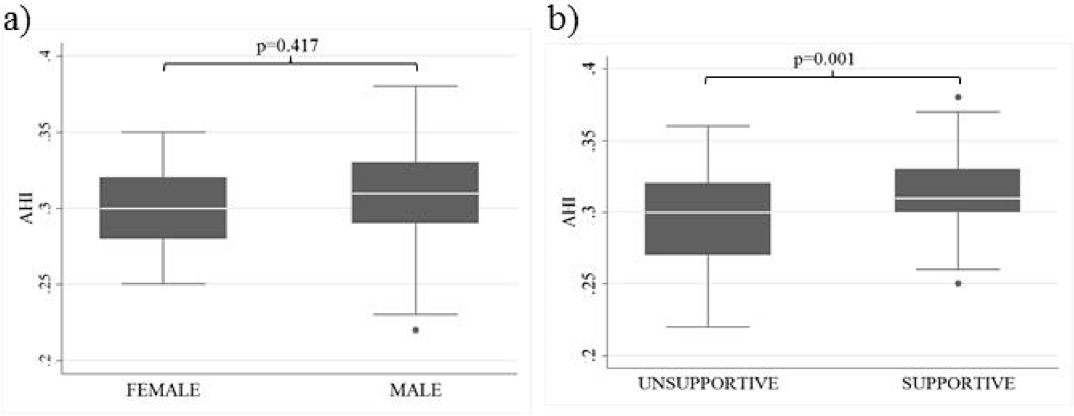


Figure 6.4 Box-whisker plots demonstrating a) AHI vs gender and b) AHI vs subtalar morphology. Significance to Student's t-test is shown (p).

6.4.2. Regression analysis of potential predictors of AHI

The multiple linear regression model demonstrated that LLAS, subtalar morphology and knee varus/valgus were all significant predictors of AHI. The regression output is summarized in Table 6.1. The overall $R^2 = 0.28$ is consistent with a large effect size ($f^2 = 0.39$). Thus increased knee valgus and lower limb flexibility as well as an unsupportive subtalar joint morphology were significantly associated with a flatter foot posture as measured using the AHI. There was no evidence of interaction between individual predictors. Of the overall variance accounted for by the model LLAS contributed the most ($R^2 = 0.15$, $f^2 = 0.18$) and both subtalar joint morphology and knee alignment contributed similar amount of variance ($R^2 = 0.06$, $f^2 = 0.06$)[†].

[†] Individual predictor variances sum to more than overall model variance due to rounding up and down phenomenon.

Predictor	Coefficient	Robust std. error	Sig. (p)	95% Confidence interval	
LLAS	−0.0044	0.0012	<0.001*	−0.0067	-0.0020
ST Morphology	0.0137	0.0053	0.012*	0.0031	0.0243
Knee varus/valgus	0.0016	0.0007	0.016*	0.0003	0.0029

Table 6.1 Summary of results of multiple linear regression, including regression coefficient, standard err (std. error), significance (Sig) and 95% confidence intervals for coefficient. * Denotes significant predictors. Subtalar morphology (ST Morphology), is a binary variable, one is supportive (Bruckner A,B,C) and zero is unsupportive (Bruckner D).

6.4.3. Plantarflexor tightness

Out of all 95 children, only 11 were unable to achieve plantigrade with one or both of their feet with their knee in extension when supine. Of this group, nine were classified as flatfooted (Table 6.2).

	FOOT POSTURE		
PLANTIGRADE	PFF	NF	TOTAL
NO	9	2	11
YES	39	45	84
TOTAL	48	47	95

Table 6.2 2 x 2 table demonstrating foot posture against ability to attain plantigrade.

Fisher’s exact test demonstrated a statistically significant greater proportion of children with PFF were unable to achieve plantigrade (p=0.05).

When the MID of the OxAFQ-C was taken to be 17%, seven of the nine children who could not achieve plantigrade were in the MS group (Table 6.3 a)). This was not statistically significant (p = 0.140). Sensitivity to identify a participant in the MS group was 78%, 95% CI [40% - 97%]. Specificity was 54%, 95% CI [37% - 70%].

When the MID was taken to be 10% all nine children who could not achieve plantigrade were in the MS group (Table 6.3 b)). This was statistically significant (p = 0.018). This resulted in 100% sensitivity, 95% CI [66% - 100%] and specificity of 44%, 95% CI [28% - 60%].

a)

	PLANTIGRADE		
SYMPTOMS	NO	YES	TOTAL
MS	7	18	25
LS	2	21	23
TOTAL	9	39	48

b)

	PLANTIGRADE		
SYMPTOMS	NO	YES	TOTAL
MS	9	22	31
LS	0	17	17
TOTAL	9	39	48

Table 6.3 2 x 2 tables demonstrating ability to attain plantigrade against more or less symptoms; in PFF group. a) When MID is 17%, and b) when MID is 10%.

6.4.4. SHR Difficulty

Of the 95 study participants 23 failed the SHR test. Of these, 21 were classified as having PFF (Table 6.4). Fisher's exact test demonstrated a statistically significant greater proportion of children with PFF failed the SHR test (p < 0.001).

	FOOT POSTURE		
FAILED SHR	PFF	NF	TOTAL
YES	21	2	23
NO	27	45	72
TOTAL	48	47	95

Table 6.4 2x2 table demonstrating foot posture against whether SHR failed or not.

When the MID of the OxAFQ-C was taken to be 17%, 17 of the 23 children that failed the SHR test were in the MS group (Table 6.4 a)). This was statistically significant (p = 0.001). Sensitivity of the SHR test to identify a member of the MS group was 81%, 95% CI [58% - 94%]. Specificity was 70%, 95% CI [50% - 86%]. When the MID was 10% there was no change in the number of the children that failed the SHR test in the MS group (Table 6.5 b)). However there was a change in the proportion of children that passed the SHR test in the MS group. As such the differences were no longer statistically significant (p = 0.067). Sensitivity of the SHR test to identify a member of the MS remained the same, but specificity was less at 48%, 95% CI [29% - 68%].

a)

	FAILED SHR		
SYMPTOMS	YES	NO	TOTAL
MS	17	8	25
LS	4	19	23
TOTAL	21	27	48

b)

	FAILED SHR		
SYMPTOMS	YES	NO	TOTAL
MS	17	14	31
LS	4	13	17
TOTAL	21	27	48

Table 6.5 2 x 2 contingency tables demonstrating failed SHR test against symptoms; in the PFF group a) When MID of 17%, and b) when MID of 10%.

6.5. Discussion

The inter-relationship between both anatomical, and clinical factors, and PFF have been explored in this chapter. Investigation into the determinants of a flat foot posture demonstrated that knee alignment, lower limb flexibility and subtalar morphology were all significant predictors of AHI. This is the first clinical study to demonstrate that an absent anterior subtalar articulation is associated with a PFF.

Failing the SHR test and inability to achieve plantigrade were both significantly more likely in the PFF group compared to the NF group. However the sensitivity and specificity of these tests, to identify children with more symptomatic PFF was dependent on the threshold of MID applied to the OxAFQ-C physical questionnaire domain.

6.5.1. The determinants of PFF

Investigation of the determinants of PFF was undertaken in two stages. The first stage involved plotting the relationship between individual clinical or anatomical factors and foot posture as measured by the AHI, and undertaking correlational or Student t-test analysis. Eight different factors were investigated in total, and the inclusion of all except subtalar morphology was on the basis of the findings of previously published studies [86, 181, 221, 250, 291, 356]. The inclusion of subtalar morphology was due to the hypothesized, but unproven relationship between aberrant subtalar morphology and a flat foot posture put forward by Bruckner et al. [36] and Barbaix et al. [10].

Of the eight factors initially investigated, age, gender, femoral anteversion, tibial torsion and BMI were not significantly related to AHI. As age was not a significant predictor, it can safely be assumed that the study population was out of the age range of developmental flat foot. This would be consistent with the majority of the literature, but some reports suggest that arch development can continue until the age of fifteen [300]. The fact that BMI was not related to foot posture went against our initial hypothesis and previous literature. Much of the literature describing the link between foot posture and BMI use footprint measures [82, 250, 330]. It is accepted that increased BMI is associated with increased adiposity which will affect the footprint parameters accordingly [265]. Morphologically the foot may also appear flatter, when in reality underlying bony architecture is relatively normal. An index like the AHI may be less sensitive to adiposity than footprint parameters, and as a result may be a better index for clinical practice. Alternatively these findings may be related to the BMI distribution of the study population, with a relatively low mean

BMI of 18.6 kg/m^2. If more children with a greater BMI had been recruited into the study then perhaps the results would have been different.

In this chapter femoral anteversion and tibial torsion did not have significant correlations with AHI. This conflicts with the findings of Zafiropoulos et al. [356] and Singh et al. [291], although there is no other clinical evidence confirming a link between rotational profile and foot posture. It may just be the case that there is no relationship between these two factors, or the effect size was too small for this study. Measuring the rotational profile of the lower limb is also notoriously difficult and prone to significant error. Using a standard goniometer to measure femoral anteversion and tibial torsion potentially does not have adequate precision and accuracy for this purpose [311]. Using a radiological method like CT or MRI to measure these parameters would be preferable. This could be an avenue for future investigation.

The three factors that appeared to have a significant relationship with AHI were subsequently placed together as predictors in a multiple linear regression model. The benefit of doing this was firstly that it would be possible to assess for any interaction between variables, and secondly to quantify the effect size demonstrated by each individual variable. Increased lower limb flexibility was associated with a reduced AHI, with the LLAS demonstrating a better than moderate effect size. Increased knee valgus and a less supportive subtalar joint were also associated with a reduced AHI although the effect size was only small to moderate, and less than half that of the LLAS.

The finding that increased flexibility is associated with a flatter foot posture is consistent with previously published literature [86, 180]. Whilst the majority of studies assess flexibility using the Beighton score [16], a different measure, the LLAS was utilized in this analysis [97]. The LLAS was favoured as all its components assess lower limb flexibility, unlike the Beighton score which only has two of nine points relating to the lower limb. The Beighton score was also measured in this study and it correlated strongly with the LLAS (R = 0.6 $p < 0.001$). As discussed in Chapter 2 (p.18-25), soft tissue structures play an important role in the maintenance of the MLA. It seems logical, therefore that the more compliant these structures the flatter the foot.

The association between PFF and valgus knee alignment has been raised in textbooks [7, 210]. The evidence supporting this view in a clinical setting is however conflicting, with Morley et al. [215] demonstrating no association in their cohort of 1000 children, and Lin et al. [181] demonstrating a highly significant relationship between the two ($p = 0.001$) in their sample of 305 children. In this chapter a significant relationship between foot posture and knee varus/valgus alignment is in keeping with the prevailing view that knee valgus is associated with a flat foot posture. The reason why this relationship is observed may be in part explained by Guichet et al. [115] who demonstrated that hindfoot valgus leads to greater lateral deviation of the lower limb mechanical axis. At the level of the knee this could drive valgus knee deformity.

This contrasts somewhat with the findings of other authors who have observed an increased prevalence of flat foot in adults with medial knee arthritis and varus knee deformities [114, 177]. In these cases it seems that the hindfoot adopting a valgus foot posture is an adaptation to a primary problem at the knee, potentially to offload the medial compartment, or merely to get the foot flat on the ground. This is further evidenced by the fact that after knee alignment is restored by arthroplasty, hindfoot valgus improves [118].

The above findings demonstrate the intimate relationship between the foot and ankle and the knee, and how deformities at one level may lead to adaptations at another. The relationship between PFF and the proximal joints will be investigated further in Chapter 10.

The role of subtalar morphology in the determination of foot posture has previously been hypothesized on the basis of observations in osteological specimens, but never confirmed in-vivo. [36, 120]. The anterior articulation of the os calcis is an integral component of the talocalcaneonavicular joint also known as the acetabulum pedis [277]. Dissection of foetal cadaveric specimens by Epelegui and Delgado [91] demonstrated that the anterior facet of the os calcis normally forms part of the osseous floor of the AP, supporting the talar head. They demonstrated that variations in subtalar morphology would affect subsequent foot shape. Barbaix et al. [10] 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 of os calcis provided reduced bony support to the talar head. Thus an absent anterior articulation of the subtalar joint places greater reliance on the plantar ligaments to support the talar head. It is believed that over time these will stretch, allowing plantar-medial deviation of the talar head with reduction of the medial longitudinal arch as seen in PFF. Whilst no statistical interaction was noted between flexibility and subtalar morphology, one might have expected there to be an additive effect of both to further reduce arch height.

As a flatter foot posture is associated with worse HRQOL, and flexibility and subtalar morphology are significant determinants, some important clinical considerations need to be highlighted. Increased flexibility may result in impaired static stabilisation of the foot and ankle joint by capsuloligamentous structures, thereby permitting increased joint motion, potentially to pathological levels. This would be especially important in subtalar and midtarsal articulations. Treatments focusing on strengthening the muscles, acting as dynamic stabilisers of the joints would reduce dependence on capsuloligamentous structures. Indeed muscle strengthening regimes have been utilized by some clinicians, with a suggestion that these may improve foot posture [264].

With respect to subtalar joint morphology, there is clearly no therapeutic intervention that can be undertaken to add an anterior articulation. However, there has been concern that lateral column lengthening may encroach upon the subtalar articular surface [41, 257]. This may be less of a concern if there is no anterior articulation. If, however, the os calcis articular surface is a Bruckner type B or C, then no modification of the lateral column lengthening technique can save the surgeon from cutting the articular surface. Whilst it remains unclear whether cutting the subtalar articular surface is of any clinical importance, an alternative surgical approach, like the ‘triple C’ (calcaneal cuboid cuneiform osteotomy) would avoid this potential issue [259]. Pre-operative imaging is recommended to assess articular morphology in PFF if surgical intervention is planned.

6.5.2. SHR Difficulty and plantarflexor tightness to identify those with symptomatic PFF

Any simple clinical test that can identify children with more symptomatic PFF would be a helpful adjunct to the assessment of this condition. The two tests assessed in this chapter were the ability to attain plantigrade with the knee extended, and the single heel rise test. It was hypothesized that these tests would be both sensitive and specific for identifying children with more symptomatic PFF. The specificity for neither test was good, although both demonstrated moderate to good sensitivity, in part dependent on the MID used to differentiate MS from LS participants.

Tightness of the plantarflexors, particularly the gastrocsoleus complex, is a well-accepted potential cause for symptomatic PFF [120, 129]. The exact mechanism by which this leads to symptoms has not been fully elucidated. However it is believed that over pull of the plantarflexors will increase the flat foot deformity, and disrupt the normal re-supination process, leading to increased loading of the plantar-medial foot structures [22]. In the study population only a small proportion of study participants were unable to attain plantigrade (12%). Nonetheless a greater proportion of children with PFF were unable to attain plantigrade, than children with NF. Ability to attain plantigrade had poor specificity to identity children with more symptomatic PFF, and sensitivity ranged from moderate to excellent (78% - 100%) dependent on which MID was used to define MS and LS. Thus if a child was unable to attain plantigrade then he or she was more likely to have more symptomatic PFF, but the ability to attain plantigrade did not rule out the child having more symptomatic PFF. These findings highlight that other factors, aside from plantarflexor tightness are also important in the development of symptomatic PFF.

The successful achievement of a single heel rise is dependent on the complex interaction between both active and passive structures. In the first stage of the SHR, tibialis posterior must invert the hindfoot. This subsequently combines with increased tension in the plantar fascia and intrinsic muscle activity to lock the midtarsal joint. This provides a rigid lever for the second stage of the SHR in which the gastrocsoleus contracts further elevating the heel to its final position. The SHR test is used routinely in the assessment of adult acquired flat foot to evaluate dysfunction of the tibialis posterior tendon [164, 225]. It has also been used to assess outcomes after tendo Achilles rupture [240]. The SHR test is preferred to manual muscle testing by some authors, as it represents an important function required for numerous activities including climbing stairs [145].

As PFF may be associated with stretching of the tibialis posterior tendon and weakness/fatigue of certain intrinsic and extrinsic muscle groups, the SHR was used as another potential screening test to identify children with more symptomatic PFF. It was also hypothesized that the SHR test would demonstrate good sensitivity and specificity for identifying children with more symptomatic PFF. As also observed with the ability to attain plantigrade, the SHR test demonstrated poor specificity for identifying MS children. The SHR test did however demonstrate good sensitivity for identifying MS children (81%), and this was irrespective of the MID used to differentiate MS and LS groups. Thus inability to achieve a SHR makes it more likely that the child has more symptomatic PFF, but the ability to achieve SHR does not rule out the child being more symptomatic.

The SHR test findings echo those observed with the ability to attain plantigrade, with both tests having high true positive, and high false negative rates. Thus they are poor as pure screening tests for children with more symptomatic PFF, because they will miss a large proportion of MS children. However as a simple adjunct to the clinical assessment they may be helpful because if a child with PFF is unable to attain plantigrade or achieve a SHR it is likely that they will be more symptomatic, and thus warrant treatment.

6.6. Limitations

The main limitations of these findings are firstly that the numbers, especially with the inability to attain plantigrade, are small. As such this part of the study is significantly underpowered to demonstrate anything but very large effect sizes.

Secondly, Maurer et al. [195] investigating plantarflexor stamina in children aged seven to nine using the SHR test, showed that on average a child could complete thirteen single heel rises before fatiguing. This contrasts significantly with this chapter where many children we unable to attain one satisfactory heel rise. The discrepancy with these findings is based on the definition of successfully achieving a SHR, and represents more of a conflict than true limitation. Maurer et al. accepted ≥ 50% of their maximum heel rise height. In this chapter the SHR had to reach the same height as the double heel rise. If Maurer et al. had adopted these more strict criteria it is questionable whether the children would have been able to achieve as many repetitions.

Finally both the assessment of the SHR test and the ability to attain plantigrade were only assessed by one rater. If these tests are investigated in the future, then larger numbers of affected subjects would be required, and inter and intra-tester reliability would also need to be performed.

6.7. Conclusion

A thorough investigation of the aetiological factors of a flat foot posture, and an exploratory investigation into the utility of two clinical tests to identify children with more symptomatic PFF have been undertaken. The results have illustrated potentially helpful adjuncts to the clinical assessment of patients with PFF, and have also identified factors which guide therapeutic intervention. Thus far in the thesis, PFF has been analysed in a static manner. The next stages of investigation involve assessing the function of PFF in a more dynamic fashion to further define the mechanisms by which symptoms may be caused.

Chapter 7. Investigating midfoot dysfunction in PFF

7.1. Introduction

The navicular has an important biomechanical function in the tarsus. It acts as the keystone of the medial longitudinal arch of the foot (MLA), and is said to be ‘a global indicator of rearfoot and midfoot components of foot pronation and supination’ [65]. Due to the navicular’s intimate relationship with the MLA there has been much focus on the biomechanical function of this bone.

As previously discussed in Chapter 4, concerns have been raised by some researchers that standard static measures may not be the best way to classify foot posture. Therefore to help further characterize the function of the foot and to investigate alternative methods to classify foot posture, the relative position of the navicular in the foot has been investigated in several ways. To quantify how the navicular moves vertically in the MLA, Brody et al. [35] introduced the quasi-static, navicular drop (NDro) test. This test quantifies the vertical movement of the navicular when standing from a subtalar joint neutral position to the resting calcaneal stance position (RCSP). Brody et al. found that runners with increased NDro were more prone to injury. NDro has also been assessed, using an electromagnetic motion analysis system during walking by Cornwall and McPoil [65], and this measure is referred to as dynamic navicular drop (DyNDro). They found that NDro was a poor predictor of DyNDro, and therefore, DyNDro may be a better measure of dynamic foot function.

Much of the research on navicular motion characterises its position in the sagittal plane. Navicular displacements are not purely in this plane and only a few researchers have also investigated movement in the transverse plane. The index, navicular drift (NDri), aims to quantify this and is defined as the distance the navicular moves medio-laterally as the foot goes into RCSP from a subtalar neutral position [331]. Increased values of NDri have been associated with ‘shin splints’, but there is no consensus on normal values [76]. Measuring NDri is technically difficult. To date it relies on accurately marking a projection of the most medial position of the navicular onto a piece of paper on the floor and then measuring how much this projection moves medially as the foot progresses from subtalar joint neutral to RCSP. Vinicombe et al. [331] investigated the repeatability of NDri and found it to be only moderately reliable with intra-tester intraclass correlation coefficients of 0.3-0.62. Due to the technical limitations of measuring NDri using the method above, it has not been assessed in normal walking as DyNDro has. If dynamic measures do not relate well to the quasi-static measures, as observed with DyNDro, then quantifying NDri during walking may also cast more light on the dynamic function of the midfoot.

The majority of the research on navicular motion has been in adult populations and often adult runners. Evans et al. [95] are the only group to have investigated navicular indices in the paediatric population, although much of their work is focused on reliability of navicular height and NDro measures.

As the position of the navicular can give information about the relative orientation of the foot segments it is useful to see how this varies in patients with foot and ankle pathology. Increased motion of the navicular may indicate midfoot instability which may relate to the development of symptoms in pes planovalgus. In the previous chapter, flexibility was shown to be an important determinant of a flat foot posture, and thus may also affect navicular motion. Whilst BMI was not proven to significantly relate to overall foot posture, it may still be of importance when addressing the midfoot, indeed Adhikari et al. [2] demonstrated a statistically significant correlation between NDro and BMI.

A limitation of the OFM multi-segment foot model is that the motion of the midfoot segment including the navicular bone is not measured. Thus to utilise 3DMA to gain further insight into navicular motion the model needs to be adapted. In this chapter a method for dynamically quantifying navicular motion presented. These results are used to gain further understanding of midfoot function in PFF.

7.2. Aims and Hypotheses

The aim of this chapter is to investigate motion of the navicular in different foot postures in a proportion of the study population. It is hypothesized that altered patterns of motion of the navicular in children with PFF, compared to NF will be observed. It is also hypothesized that dynamic navicular motion will correlate poorly with the standard static measures of navicular displacement. The final hypothesis is that navicular motion will relate to flexibility and BMI, with increased magnitudes of navicular motion observed with greater BMI and greater flexibility.

7.3. Materials and methods

7.3.1. Participants and protocol

Data collection was undertaken as per the protocol outlined in Chapter 3 (p.55-92). To assist in quantifying navicular motion, an extra marker was attached to the navicular tuberosity (Figure 7.1).

Based on the power calculations in 7.3.3, the first 55 study participants were selected for this study. However as the navicular was not palpable in four subjects, analysis of only 51 subject's data was possible. Foot classification was based on the methods outlined in Chapter 4 (p.93-120). This yielded 25 subjects being classified as having PFF and 26 subjects as having NF. Table 7.1 summarises demographic, BMI and flexibility characteristics of the subjects. The PFF group was significantly more flexible than the NF group ($p < 0.01$).

Parameters	PFF (n=25)	NF (n=26)	p
Age (years) median [IQR]	11.1 [10.0 - 12.2]	12.4 [9.4 - 13.9]	0.500
BMI mean (SD)	17.7 (3.5)	17.4 (2.7)	0.25
LLAS median [IQR]	5 [2 - 7]	3 [1 - 4]	< 0.01*
Gender (M:F)	11:14	17:9	0.125

Table 7.1 Characteristics of study participants, separated into PFF and NF groups. IQR, interquartile range, BMI, body mass index, SD standard deviation. Significance values (p) for differences between groups also illustrated. *demonstrates statistically significant results.

7.3.2. Navicular Index Calculation

The navicular marker coordinates were used to calculate all navicular indices. NDro was calculated as the difference in the vertical height (h) of the navicular marker as the participant went from sitting to standing:

$$\mathrm{NDro} = h_{SIT} - h_{STAND} \tag{7.1}$$

DyNDro was calculated as the difference between sitting navicular height and the minimum navicular height during the stance phase of gait:

$$\mathrm{DyNDro} = h_{SIT} - \mathrm{Min}(h_{WALK}) \tag{7.2}$$

Both NDro and DyNDro are positive when *h* decreases with weightbearing.

NDri and DyNDri were calculated as the change in the perpendicular distance (d) of the navicular marker from the heel-toe axis in the transverse plane of the laboratory coordinate system (Figure 7.1):

$$\mathrm{NDri} = d_{STAND} - d_{SIT} \tag{7.3}$$

$$\mathrm{DyNDri} = \mathrm{Max}(d_{WALK}) - d_{SIT} \tag{7.4}$$

Positive values of NDri and DyNDri indicate movement of navicular marker in the medial direction, consistent with the sign convention used in previous studies [76, 331].

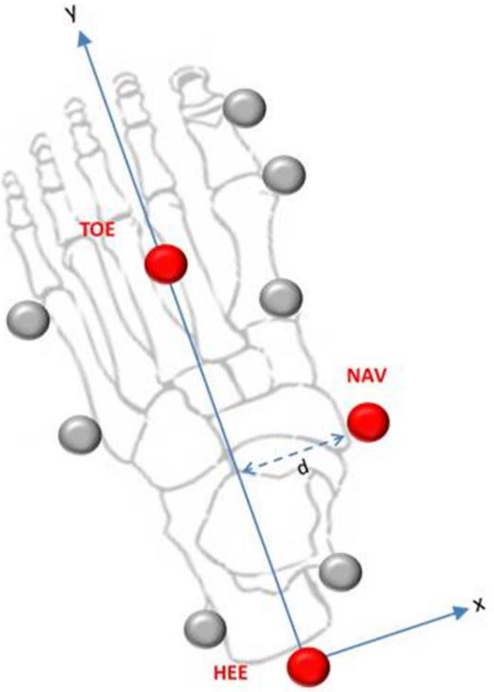


Figure 7.1 The perpendicular distance (d) in the transverse plane of the navicular marker (NAV) from the heel-toe axis (HEE-TOE, as used in the calculations of NDri and DyNDri. Position of Oxford foot model markers also shown. NDri, navicular drift; DyNDri, dynamic navicular drift.

Calculations were all performed in Matlab (R2012b, The Mathworks Inc., Natick, USA 2012). Each navicular index was calculated for both feet, but an averaged value for both feet was used for data analysis. Values in millimetres for navicular motion are reported to aid comparison with previously published work. Nielsen et al. [232] recommend normalization of navicular drop by TFL to account for varying foot size. Navicular indices were normalized for comparison between groups and with flexibility and BMI such that:

$$\text{Normalized Index} = \frac{Index}{TFL} \text{ x } 100 \qquad 7.5$$

7.3.3. Statistical analysis

Data were assessed for normality and kurtosis. The data were parametric, thus Pearson product moment correlation and two-tailed, unpaired Student t-tests were used for data analysis. Statistical analysis was undertaken using Stata v.13.0 (Statacorp LP, Texas, USA).

Based on the standard deviation of normalized navicular drop (NNDro) of 2.5 (based on pilot data presented at the European Society of Movement Analysis for Adults and Children annual meeting (see Appendix 3)), with a minimum of 25 subjects in each group and an α of 0.05 this analysis had an 80% power to detect a difference in means of 2.0. For normalized navicular drift (NNDri) with a standard deviation of 1, a minimum of 25 subjects in each group and an α of 0.05, this study had 80% power to detect a minimum difference in mean values of 0.80.

7.4. Results

A larger magnitude and range of motion of the navicular in the sagittal plane was noted compared to movement of the navicular in the transverse plane (Table 7.2). There was no significant difference between the mean values of DyNDro compared to NDro. Over all subjects, the mean value of DyNDri was 0.5 mm greater than NDri, but this was also not statistically significant. When normalized for foot length the PFF group had significantly reduced NNDri compared to the NF group (0.7 ± 1.1 vs 1.5 ± 1.0, $p = 0.007$). This equates to a difference of between 1.2 mm and 1.8 mm depending on TFL.

Parameters	**All (n=51)**		**PFF (n=25)**	**NF(n=26)**
	Mean ± SD	Range	Mean ± SD	Mean ± SD
NDro (mm)	10.1 (± 4.7)	2.0 to 23.5	10.9 (± 4.6)	9.4 (± 4.7)
DyNDro (mm)	9.8 (±5.0)	1.0 to 22.8	10.7 (± 5.6)	8.9 (± 4.3)
NDri (mm)	2.1 (±2.0)	−3.2 to 8.3	1.4 (± 2.1)*	2.7 (± 1.8)*
DyNDri (mm)	2.6 (±1.8)	−3.5 to 7.9	2.5 (± 2.1)	2.9 (± 1.4)

Table 7.2 Summarising navicular measures for all subjects and subjects by group membership. The values marked with the asterisk denote where there was a significant difference in normalized NDri between groups (p = 0.007).

7.4.1. Correlation between static and dynamic measures

There was a strong linear relationship between DyNDro and NDro for all subjects (Figure 7.2 a)) with a Pearson’s R of 0.71 ($p < 0.001$). There was a similarly strong relationship between NDri and DyNDri (Figure 7.2 b)) with a Pearson’s R of 0.69 ($p < 0.001$). Due to the strong relationship and collinearity between quasi-static and dynamic measures, solely quasi-static measures were used in subsequent analyses.

7.4.2. Relationship between navicular motion in sagittal and transverse plane

In the case of neutral feet there was a strong positive correlation between normalized navicular drop (NNDr) and normalized navicular drift (NNDri) (Figure 7.3) with a Pearson’s R of 0.56 ($p = 0.003$). No such relationship was noted between NNDr and NNDri in the flat footed group with a non-significant Pearson’s R of 0.18.

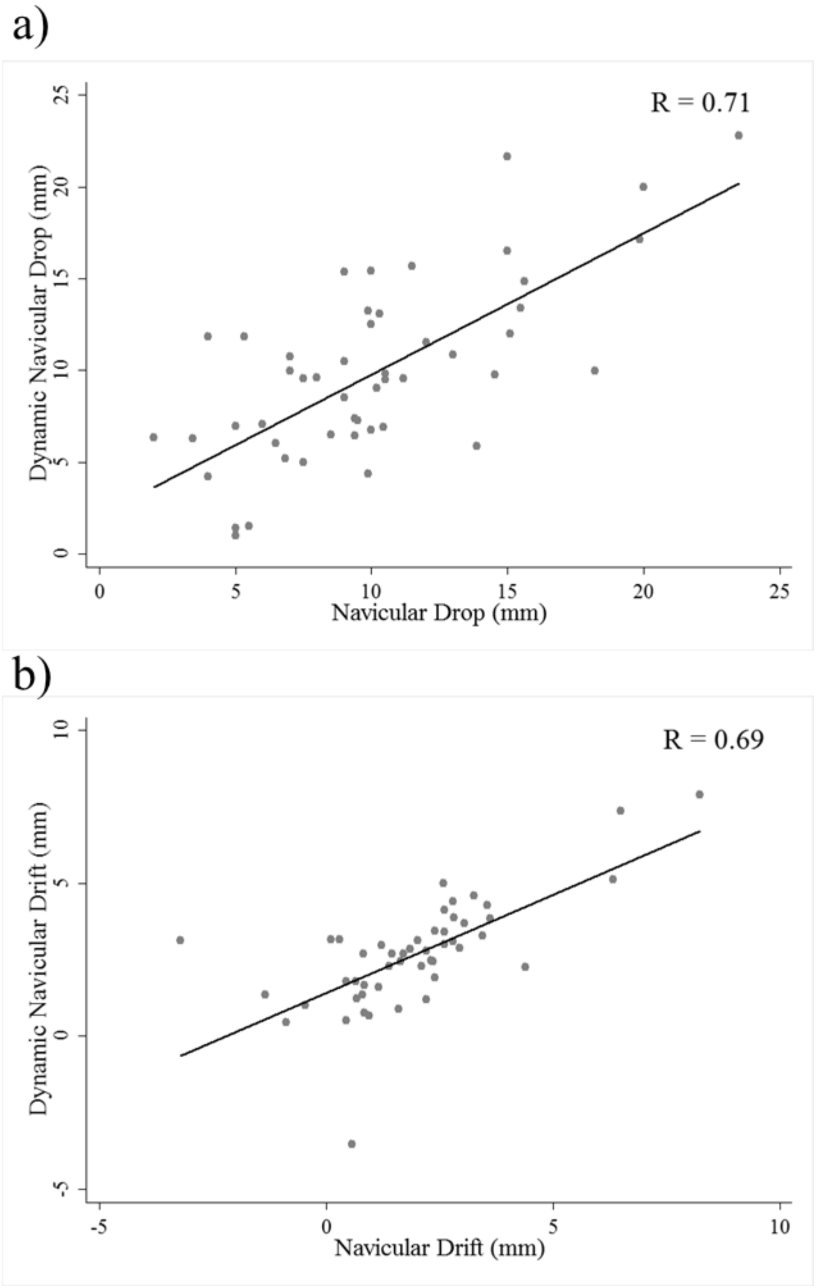


Figure 7.2 a) Scatter plot of dynamic navicular drop vs navicular drop b) Scatter plot of dynamic navicular drift vs navicular drift. Lines of best fit shown as well as Pearson’s correlation coefficient (R).

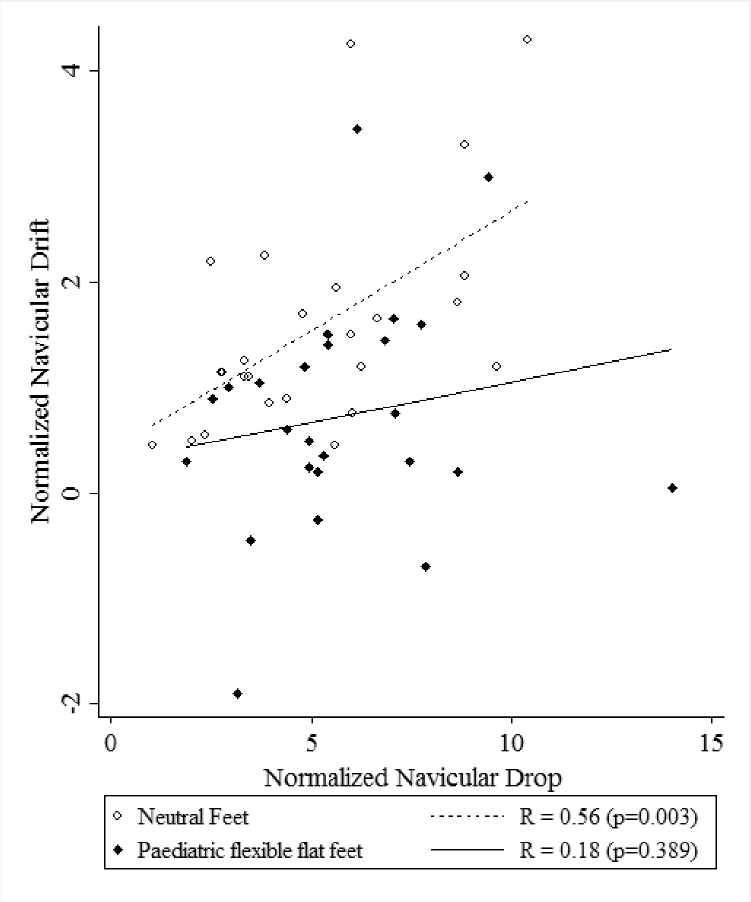


Figure 7.3 Scatter plot of NNDri (positive direction indicates medial displacement) vs NNDro. Indices are normalized to foot length. NF and PFF plotted separately, with linear best-fit line. Pearson's correlation coefficient R noted in graph legend.

7.4.3. Relationship of BMI and flexibility with navicular indices for all subjects

Body mass index did not correlate significantly with either navicular index (Figure 7.4). The lower limb assessment score did however have a significant correlation with NNDri (R = 0.29, p = 0.04), with increased flexibility being associated with reduced medial drift of the navicular.

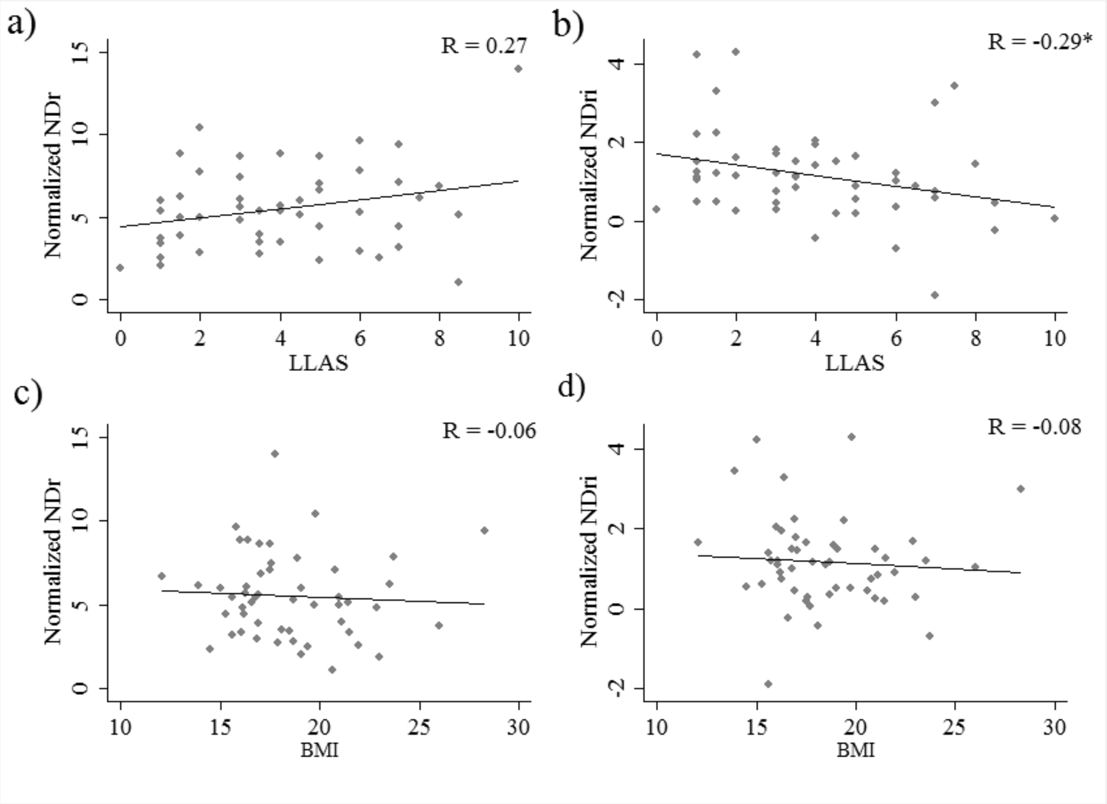


Figure 7.4 Scatter plots demonstrating the relationship between a) Normalized NDr vs LLAS, b) Normalized NDri vs LLAS, c) Normalized NDr vs BMI (kg/m2) and d) Normalized NDri vs BMI (kg/m2). The scatter plots are illustrated with the line of best fit and correlation coefficient (R). * denotes statistically significant correlation (p = 0.04).

7.5. Discussion

In this chapter, navicular motion in children with different foot postures was investigated and this motion was related to BMI and flexibility. Two indices describing the sagittal motion of the navicular were calculated, NDro and DyNDro. These have previously been investigated by several researchers in the adult population, with a variety of normal values being quoted. Brody et al. [35] quoted 10 mm as being normal and 15 mm as potentially abnormal. There have also been some smaller estimates of what is regarded as normal (8.1 mm, 6.8 mm and 6-9 mm [4, 184, 293]). Increased NDro has been associated with an increased risk of injury when running [35]. In the case of DyNDro similar ranges of values of navicular drop have been noted by Dicharry et al. [79], and Cornwall et al. [65], however, they found a poor correlation between the quasi-static and the dynamic measures. This led them to the conclusion that DyNDro gave a better indication of functional movement of the navicular. The mean values of NDro and DyNDro obtained for children in this study appear to be similar to what has been described in the literature with mean values of 10.1 mm and 9.8 mm respectively. The NDro and DyNDro values measured were independent of foot posture and this was similar to recent findings in adult subjects by McPoil et al. [200].

This chapter adopts a slight variation in the classical technique of measuring NDro as described by Brody et al. [35]. In the original description the subject is standing and the subtalar joint is manipulated into a neutral position [35]. The navicular height is measured in this position and then measured again when the subject is allowed to drop into the RCSP. The NDro is the difference between these two measures. This technique has been subject to various criticisms because it has been shown to be very difficult to know whether the subtalar joint has been placed in a neutral position [251], which may contribute to poor reproducibility of NDro measurement. In response some researchers have adopted altered techniques to measure NDro including the sit-to-stand protocol that we used in this study [317]. Sit-to-stand has been found to lead to better reproducibility when measuring arch height, and consequently is believed to be better for measuring NDro [200].

In this chapter the movement of the navicular in the transverse plane was assessed by measuring NDri and the novel DyNDri. NDri is a measure of the medial displacement of the navicular as the foot goes from subtalar joint neutral to RCSP. DeLarceda et al. [76] used NDri measurements in a comparative study of runners with and without shin splits. They found that runners with shin splints had a mean NDri of 8.9 mm compared to 5.5 mm in healthy subjects. Vinicombe et al. [331] described a normal range of NDri of 0-9 mm. These values are quite different from those obtained in this chapter (−3.2 mm to 8.3 mm). In this chapter some negative values of NDri were demonstrated implying lateral translation of the navicular as the foot is loaded. The discrepancy between measures may be due to the variation in measurement technique. The classical NDri does not solely measure the medial translation of the navicular but is actually a composite measure of hindfoot valgus and forefoot abduction which may occur as the foot adopts the RCSP. The technique used in this study attempts to measure the absolute position of the navicular relative to the heel-toe axis and it is believed to be more representative of medio-lateral translation of the navicular. The sit-to-stand protocol is also a variation of the originally described technique, similar to that for NDro. In this chapter less medial translation of the navicular, and sometimes even lateral translation in the PFF group was observed compared to the neutrals. This is logical as the more flatfooted a person becomes, the less well-aligned the talonavicular joint, and it is the talar head which moves medially with the navicular moving relatively laterally with the rest of the forefoot. This greater lateral movement of the navicular in symptomatic flat footed subjects has been observed on plain radiographs by Moraleda et al. [213].

A number of authors have found that in adults there is a poor relationship between NDro and DyNDro, with DyNDro being a better measure of foot function [65, 79]. These findings prompted the investigation of this relationship in children and also an assessment of DyNDri as NDri’s dynamic counterpart. In children it appears that the standard static measures correlate very well with dynamic measures and, as such, dynamic measurement may not confer any added benefit. Deviation from this relationship, however, may be observed during other motor tasks including fast walking or running and is worth investigating.

There is an established link between foot posture, BMI and flexibility in children. The heavier a person is, and the more flexible, the flatter his or her feet [86, 250]. The navicular is the keystone of the MLA, thus the relationship of BMI and flexibility and the amount of navicular excursion was assessed. Increased flexibility was associated with a reduction in NDri, with values at times going into the negative. In Chapter 6 (p.136-157) it was demonstrated that increased flexibility was a determinant of foot posture, and this deformity is known to be associated with lateral subluxation of the navicular on the talus [213]. Therefore the findings are consistent with the observation of a reduction in NDri in the PFF group.

No significant correlations were found between BMI and the navicular indices, which contrasts with the findings of Adhikari et al. [2] but is consistent with what was observed in adult subjects by Neilsen et al. [232]. The finding that BMI did not correlate with navicular excursion is consistent with the finding in Chapter 6 that BMI was not a significant determinant of PFF. These findings are also subject to the same limitations outlined in Chapter 6 (p.156).

In the introduction it was hypothesized that altered navicular motion would be observed in flat compared to neutral feet. As described earlier there were differences in the magnitude of NDri, and also a disruption of the normal linear relationship between NNDri and NNDro. Inman and Mann [137] were the first to describe the important coupling relationships between the joints of the foot. For adequate push off the hindfoot must invert, which in turn alters the orientation of the calcaneocuboid and talonavicular joint making the midfoot and forefoot a rigid lever to allow propulsion. Mosca [221] has subsequently hypothesized that increased hindfoot eversion may disrupt this coupling leading to impaired push off. The uncoupling of NNDri and NNDro adds further evidence to this hypothesis demonstrating dysfunction of the midfoot in flat-footed individuals. Those with impaired push off may subsequently bear more stresses in their midfoot eventually leading to the midfoot break observed in severely flatfooted individuals. It is believed that symptoms associated with flatfeet are worse in children where this derangement of navicular motion is more extreme. Ongoing biomechanical derangement may lead to secondary degeneration, and therefore these children may benefit from intervention. Some clinicians believe that, in some cases, adult acquired flat foot deformity may lead on from flexible flatfoot [175]. Thus while the population assessed in this study was from 8-15 years of age, the findings may have relevance to the continuation of flexible flatfoot into adulthood, with follow-up and treatment implications.

7.6. Limitations

The motion of the navicular was assessed using 3DMA. This technique allows the measurement of dynamic navicular motion in a non-invasive fashion and has been utilized in adults by several groups [9, 65, 232, 260]. This study represents the first time that it has been used in children. The measurement error of a well-calibrated 3DMA system is less than 0.1 mm, and from a precision perspective is superior to the classical methods for measuring these indices. This measurement method does, however, rely on the accurate placement of the navicular marker on the navicular tuberosity and, as with 3DMA in general, markers would also be subject to soft tissue motion artefact. In this study care was taken to minimize skin motion artefact by using low-mass markers attached with the subject standing [105, 194], however the reliability of navicular placement needs further investigation.

Another potential source of error is the effect of variability in foot size in children aged 8 to 15 on navicular motion. To minimize the effect of this the navicular indices were normalized to foot length, as per the recommendations of Neilsen et al. [232].

The absolute ranges of motion of the navicular observed are also small and potential sources of error are likely to interfere with the results obtained, however they were the same for both study groups and as such it is believed that the results demonstrated are a true representation of the relationship of navicular indices between groups. Nonetheless further validation of this method to quantify navicular motion is required.

7.7. Conclusion

In children, dynamic navicular motion in straight walking is similar to sit-to-stand navicular motion; thus measurement of DyNDro and DyNDri does not necessarily give any added information.

This chapter demonstrates that motion of the navicular in the transverse as well as the sagittal plane is important when investigating foot function. The uncoupling of these movements observed in the PFF population may indicate an impairment of midfoot biomechanical function, with the reduced medial translation of the navicular observed in flat feet probably related to altered alignment of the talonavicular joint.

The next stage of this investigation is to formally assess foot and ankle joint motion using 3DGA and put the findings in a more clinical context.

Chapter 8. Is PFF associated with clinically relevant deviations in foot and ankle joint motion?

8.1. Introduction

As discussed in Chapter 2 (p.38-50), there are a number of differences between neutral and flat feet when it comes to form and function. On the basis that foot posture is defined using various morphological characteristics one might expect there to be differences in the kinematics between PFF and NF. Even if a flat foot posture is associated with altered function this may not necessarily be detrimental, and deviation from the norm does not always equate to dysfunction. As such it is important to put any significant findings in context. This can be done, firstly, by relating observations to the hypothesized pathomechanic theories of excessive hindfoot valgus and impaired midtarsal locking leading to increased forefoot motion. Secondly, and more importantly, significant findings should be related to the clinical picture - in this case HRQOL. If there are certain key deformities relating to an impaired HRQOL, then not only is it possible to get further insight into the mechanism by which PFF causes symptoms, it will also provide a relevant target for therapeutic intervention.

Intimately linked with foot and ankle joint kinematics are the spatio-temporal parameters: walking speed, cadence, step length and stride width. If a flat foot posture is truly associated with biomechanical dysfunction then one would expect this to also be reflected in spatio-temporal parameters.

8.2. Aims and hypotheses

The aims of this chapter are two-fold: firstly to investigate how spatio-temporal parameters and joint kinematics relate to foot posture, and secondly, to investigate the relationship between spatio-temporal parameters and relevant joint kinematics with HRQOL.

The hypotheses are as follows:

1. PFF will be associated with slower self-selected walking speed and this will be more marked the more symptomatic the child. The reduced walking speed will be due to reduced step length and cadence. PFF will also be associated with increased step width.

2. It is hypothesized that a flat foot posture will be associated with the following dynamic joint angle differences:

    Increased hindfoot eversion

    Decreased hindfoot dorsiflexion

    Increased forefoot dorsiflexion, abduction, supination and overall forefoot mobility

    Increased hallux Dorsiflexion

3. Of the kinematic differences, it is hypothesized that increased hindfoot eversion and increased forefoot motion will be associated with worse HRQOL in the PFF group.

8.3. Methods

The general data collection methods used for this chapter are outlined in Chapter 3 (p.55-92). Spatio-temporal parameters were obtained for all 95 subjects. Oxford Foot Model kinematics were obtained for only 94 subjects. This was due to the fact that the retroreflective markers required for the OFM failed to remain adhered to one subject's feet during testing procedures. This resulted in equal group sizes for kinematic analysis of 47 subjects in each of the PFF and NF groups.

8.3.1. Spatio-temporal parameters

A number of spatio-temporal parameters were measured during 3DGA, these included walking speed, cadence, step length and step width. The values for the spatio-temporal parameters were calculated and outputted directly from Vicon Polygon (v. 3.5.1,Vicon, Oxford UK). The distance parameters were calculated from the positions of the TOE marker (see Figure 3.6). To account for any potential intra-subject variability of these parameters, during testing, the spatio-temporal parameters for three normal walking trials were averaged. As there were no statistically significant differences between left and right sides the parameters were also averaged across sides.

As the age of the study participants ranged from eight to fifteen years old, considerable variability in subject height and consequently leg length was observed. As this could potentially interact with the spatio-temporal parameters, values were normalized according to the methods described by Hof et al. [126] such that:

$$nSL = \frac{SL}{LL} \qquad 8.1$$

$$nSW = \frac{SW}{LL} \qquad 8.2$$

$$nCad = \frac{Cad}{\sqrt{(g/LL)}} \qquad 8.3$$

$$nWS = \frac{WS}{\sqrt{(g*LL)}} \qquad 8.4$$

SL= step length, SW = step width, Cad = cadence, WS = walking speed, g = gravity (9.81m/s^2) and n = normalized parameter.

Stepwise multiple linear regression was used to assess how foot posture related to the spatio-temporal parameters, in all subjects. Age and BMI were also included in the regression models as it is reported in the literature that they can affect spatio-temporal parameters [83, 214]. The effect of OxAFQ-C physical domain scores, age and BMI on spatio-temporal parameters, solely in the PFF group, was also assessed using a stepwise multiple linear regression approach. Regression diagnostics were used to ensure that model residuals were normally distributed and that the residual versus fitted values did not demonstrate heteroscedasticity.

8.3.2. Comparison between PFF and NF OFM angles

Oxford Foot Model angles were computed using dedicated software pipelines in the Vicon Nexus (v1.7, Vicon, Oxford UK) software environment. Table 8.1 summarises the angles output from the OFM. From the minimum of five normal walking trials, one trial was selected as the representative trial for the subject. This was achieved by computing the root mean squared difference (RMSD) between each curve and the mean curve for each OFM angle. The trial with the overall minimum average RMSD was selected as the representative trial. The gait cycle from initial contact to initial contact was normalized to 100%.

OFM Angle	Suffix	Motion	Axis
Hindfoot relative to tibia (HFTBA)	X	Hindfoot plantarflexion/dorsiflexion	Transverse axis of the tibia
	Z	Hindfoot inversion/eversion	Longitudinal axis of the hindfoot
	Y	Hindfoot internal/external rotation	Axis perpendicular to the previous two axes
Forefoot relative to hindfoot (FFHFA)	X	Forefoot plantar/dorsiflexion	Transverse axis of the hindfoot
	Z	Forefoot supination/pronation	Longitudinal axis of the forefoot
	Y	Forefoot abduction/adduction	Axis perpendicular to previous two axes
Hallux relative to forefoot (HXFFA)	X	Hallux plantar/dorsiflexion	Transverse axis of the forefoot
	Y	Hallux abduction/adduction	Superior/inferior axis of the forefoot

Table 8.1 Summary of OFM model output including specific joint motion and axis about which motion occurs.

The first stage of kinematic data analysis involved visual inspection of gait curves pooled within groups for each OFM joint angle. The mean curves and 95% confidence interval bands for the PFF and NF groups were plotted. Regions of interest were identified on the graphs where potentially significant differences between PFF and NF groups were observed. These were defined as areas where a section of mean curve of one group was not within the 95% confidence interval of the other. This also had to correspond to a readily defined value or point in the gait cycle. The points included local maxima (MAX), and minima (MIN), initial contact (IC), midstance (MIDS), foot off (FO) and range of motion (ROM). Where MAX or MIN values were around the 60% point (end of stance phase) in the gait cycle angle, values at FO were used in preference. Midstance was simply defined as the point at 50% of the stance phase of gait.

Discrete joint angle values were then correlated, using Pearson’s correlation coefficient, with the AHI as a continuous measure of foot posture. Where a statistically significant correlation was noted between AHI and a joint angle measure, the confounding effect of age, gender and BMI was assessed. Where necessary, stepwise multiple linear regression was undertaken to control for the confounding effects of these variables.

8.3.3. Correlation of gait kinematics with HRQOL

From the processes outlined in section 8.3.2 a list of discrete joint angular measures were obtained that had a statistically significant relationship with arch height as measured using the AHI. These values were then correlated with OxAFQ-C physical domain scores solely in the PFF group using Pearson’s correlation coefficient to identify whether deviations in foot and ankle joint motion were related to HRQOL.

8.3.4. Sample size and power calculations

Sample size and power calculations were made using GPower (v3.1.9.2 Universität Kiel, Germany). Correlations with the entire sample (95 subjects) had an 80% power to detect an effect size (R) of 0.28. Correlations just in the PFF group had 80% power to detect an effect size of (R) 0.38 (Figure 8.1). According to Cohen, an R of 0.10 represents a small effect size, 0.30 a moderate effect size and 0.5 a large effect size [64].

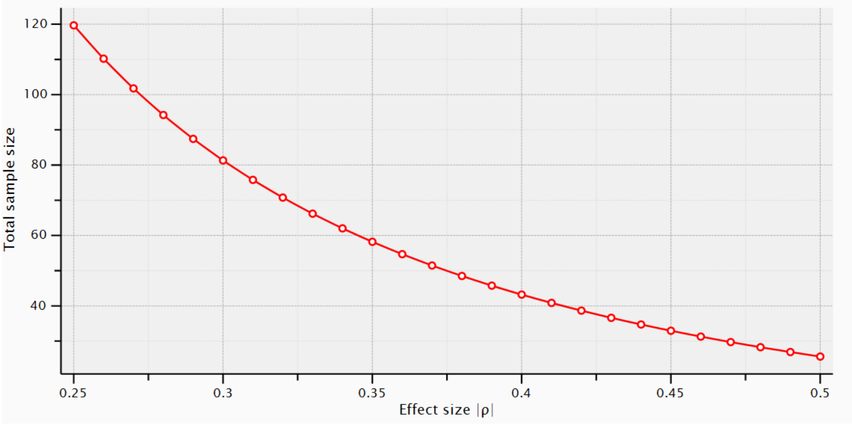


Figure 8.1 Sensitivity plot demonstrating the relationship between sample size and effect size in correlation where power is set as 80% and alpha as 0.05.

In the case of multiple linear regression, analysis of the entire sample using three test predictors had an 80% power to detect an effect size (f^2) of 0.09. Analysis of just the PFF group using three predictors had an 80% power to detect an effect size (f^2) of 0.17 (Figure 8.2). According to Cohen an f^2 of 0.02 represents a small effect size, 0.15, a moderate effect size and 0.30 a large effect size [64].

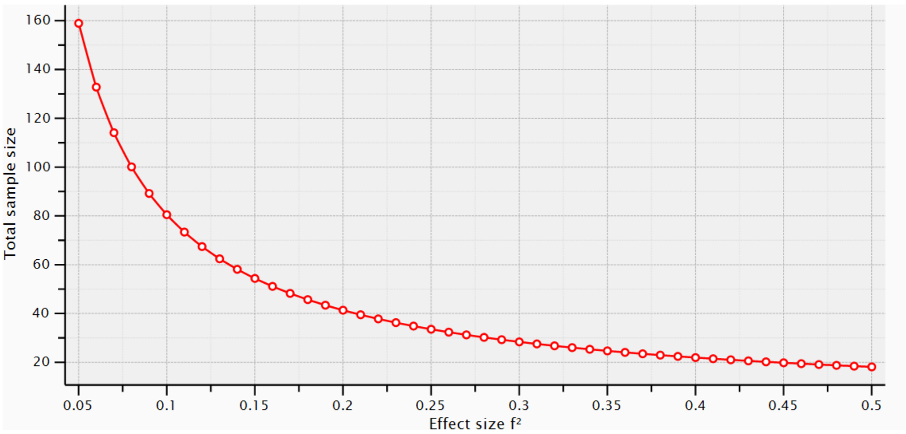


Figure 8.2 Sensitivity plot demonstrating the relationship between sample size and effect size for multiple linear regression with three predictors, where power is set as 80% and alpha as 0.05.

8.4. Results

8.4.1. Spatio-temporal parameters

Children with PFF had a statistically significant reduction in normalized step length compared to NF children (0.75 vs 0.78, p = 0.03), with an effect size of 0.05. There was also a tendency for PFF children to have a slower normalized walking speed (0.44 vs 0.47; p = 0.09), but this did not reach statistical significance. Normalized cadence and step width were similar between groups. Table 8.2 summarizes spatio-temporal parameters by group.

Normalized parameter	NF	PFF
Walking speed (mean ± SD)	0.47 (± 0.07)	0.44 (± 0.06)
Cadence (mean ± SD)	0.59 (± 0.04)	0.59 (± 0.05)
Step length (mean ± SD)	0.78 (± 0.08)*	0.75 (± 0.07)*
Step width (mean ± SD)	0.19 (± 0.04)	0.19 (± 0.04)

Table 8.2 Summary of mean spatio-temporal parameters for NF and PFF groups with association standard deviations (SD). * denotes statistically significant difference between groups ($p < 0.05$).

Body mass index was significantly associated with reduced step length (p = 0.03). Increased age was a significant predictor of reduced step width (p = 0.04).

Stepwise multiple linear regression demonstrated that the OxAFQ-C physical domain score was a significant predictor for normalized walking speed ($R^2 = 0.10$ p = 0.03), and normalized cadence ($R^2 = 0.08$ p = 0.05) in the PFF group. This relationship was not observed for normalized stride length or stride width. Thus the lower the OxAFQ-C physical domain scores, the slower the normalized walking speed, secondary to a reduced cadence.

8.4.2. Oxford Foot Model Kinematics

A number of regions of interest were identified from the OFM curves, and have been highlighted in Figures 8.3 to 8.5. The most striking difference between groups was observed in Figure 8.4 b), where children in the PFF group were noted to have greater forefoot supination relative to the hindfoot compared to the NF group, with no overlap of confidence interval bands throughout the gait cycle. In total 20 potential regions of interest were identified in the graphs. These are summarized in Table 8.3 along with the Pearson’s correlation coefficient and significance value for correlation with AHI.

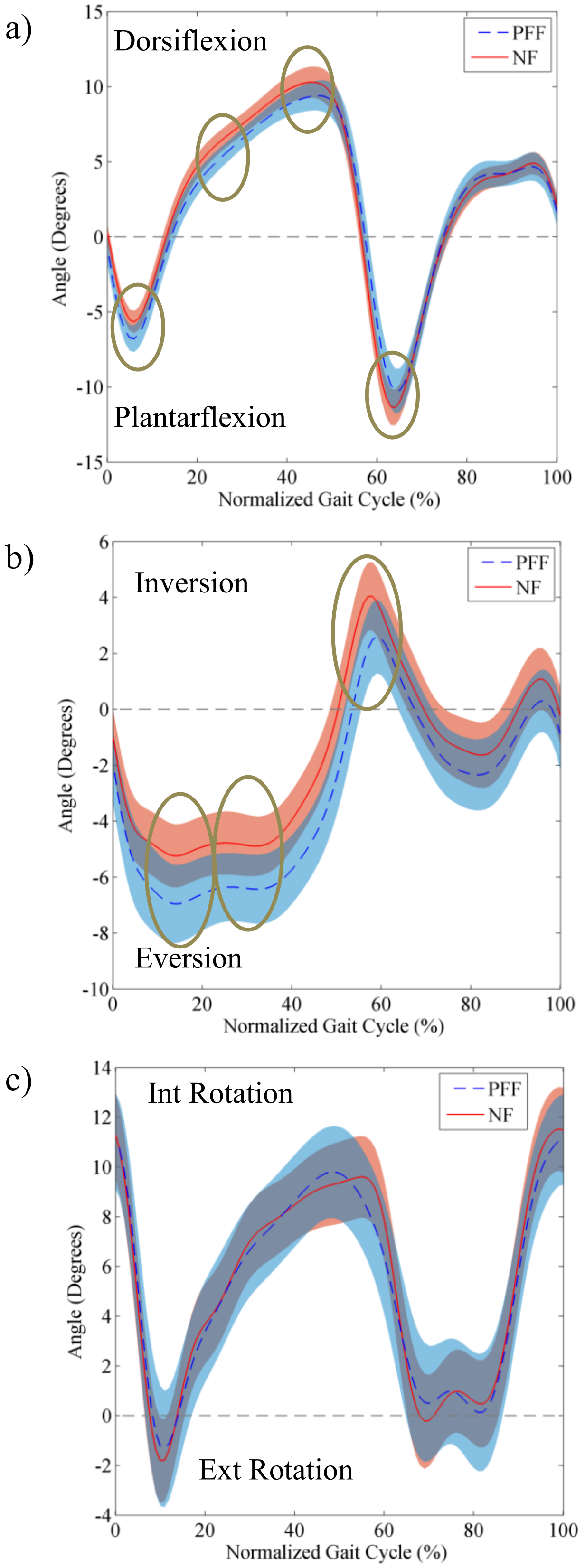


Figure 8.3 Normalized gait cycle curves for motion of the hindfoot relative to the tibia. Mean curve for each group along with 95% confidence bands plotted. Regions of interest (ROI) marked.

a) HFTBAx, with ROIs of local minima during Loading response (LRMIN), MIDS, MAX and FO.

b) HFTBAz, with ROIs MIN, MIDS and FO.

c) HFTBAy, with no ROIs.

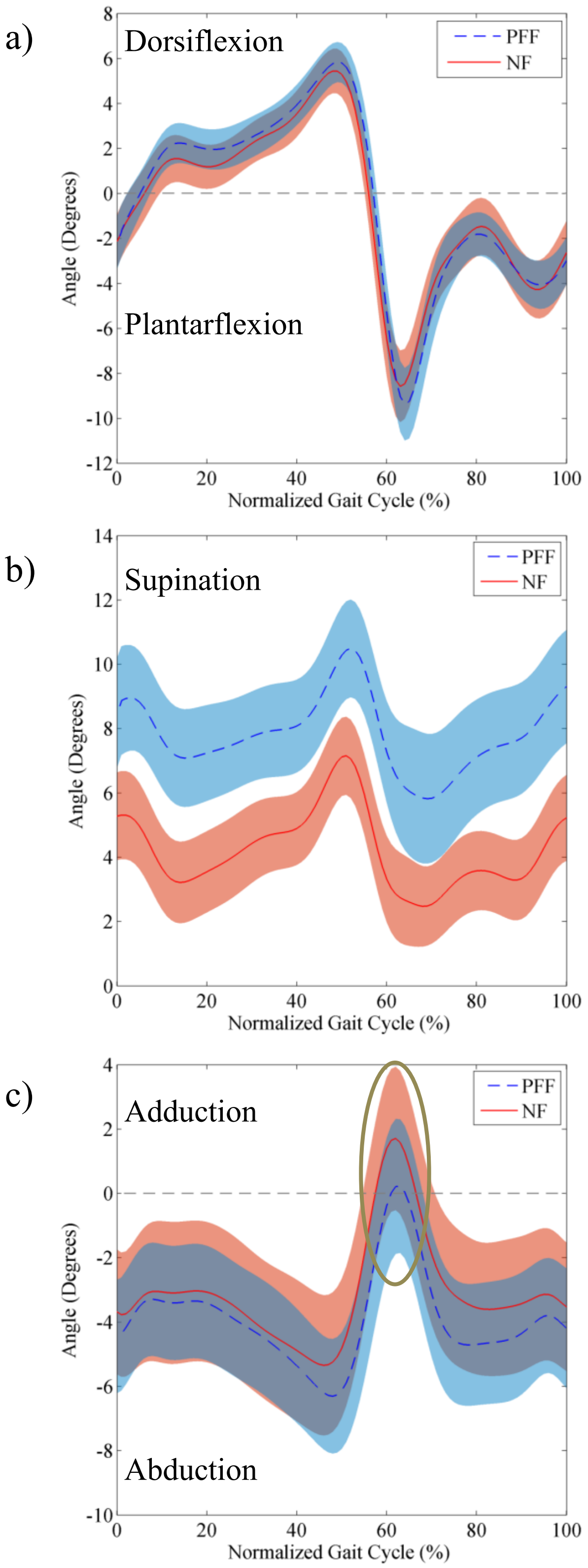


Figure 8.4 Normalized gait cycle curves for motion of the forefoot relative to the hindfoot. Mean curve for each group along with 95% confidence bands plotted.

a) FFHFAx, with ROI of ROM.

b) FFHFAz. As no overlap between 95% confidence bands the whole gait curve is of interest. As such MIN, MAX, IC, MIDS, FO and ROM are specific ROIs.

c) FFHFAy, with ROI of FO.

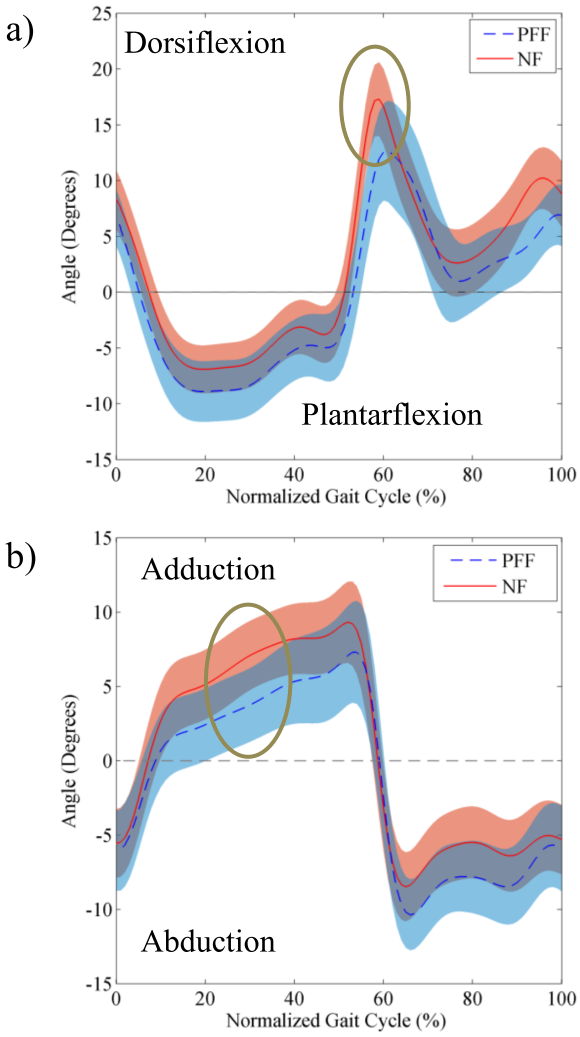


Figure 8.5 Normalized gait cycle curves for motion of the hallux relative to the forefoot. Mean curve for each group along with 95% confidence bands plotted. Regions of interest (ROI) marked.

a) HXFFAx, with ROI of FO and ROM.

b) HXFFAy with ROI of MIDS.

Of the twenty discrete parameters listed in Table 8.3, ten had significant correlations with AHI. Thus a flatter foot posture was related to the following findings:

i) Increased hindfoot plantarflexion in the loading response.

ii) Decreased minimum hindfoot inversion (equivalent to increased maximum eversion) and decreased midstance inversion.

iii) Increased forefoot dorsiflexion/plantarflexion range of motion.

iv) Increased maximum and minimum forefoot supination as well as increased forefoot supination at initial contact, midstance and foot off.

v) Reduced forefoot adduction at foot off.

Angle	**Parameter**	**Correlation with AHI**	
		R	Sig (p)
HFTBAx	MAX	0.01	0.90
	LRMIN	0.21	0.04*
	FO	−0.18	0.09
	ROM	0.13	0.21
HFTBAz	MIN	0.26	0.01*
	MIDS	0.23	0.03*
	FO	0.14	0.19
	ROM	−0.16	0.13
FFHFAx	ROM	−0.28	<0.01*
FFHFAz	MAX	−0.32	<0.01*
	MIN	−0.24	0.02*
	IC	−0.22	0.04*
	MIDS	−0.28	0.01*
	FO	−0.28	0.01*
	ROM	−0.13	0.21
FFHFAy	FO	0.34	<0.01*
	ROM	0.00	1.00
HXFFAx	FO	0.06	0.55
	ROM	−0.15	0.14
HXFFAy	MIDS	0.00	0.98

Table 8.3 Regions on interest identified by visual inspection of OFM joint kinematic graphs. The relationship between ROIs and AHI quantified with Pearson’s R and significance (Sig). * Denotes a statistically significant result.

There were significant differences by gender for forefoot dorsiflexion/plantarflexion (FFHFAx) range of motion as well as for all forefoot pronation/supination (FFHFAz) variables (Table 8.4). Body mass index had a significant negative correlation with forefoot dorsiflexion/plantarflexion ROM (FFHFAx)(Table 8.4). Significant positive correlations between forefoot adduction at FO and both BMI and age were observed (FFHFAy).

Angle	Parameter	Gender		BMI		Age	
		Diff (°)	Sig (p)	R	Sig (p)	R	Sig (p)
HFTBAx	LRMIN	0.86	0.17	0.05	0.63	−0.10	0.32
HFTBAz	MIN	−0.74	0.40	−0.08	0.42	−0.01	0.93
	MIDS	−0.52	0.55	−0.11	0.28	−0.04	0.74
FFHFAx	ROM	2.52	0.01*	−0.23	0.03*	−0.11	0.31
FFHFAz	MAX	2.71	0.01*	0.06	0.58	−0.03	0.74
	MIN	2.82	0.01*	0.06	0.57	0.02	0.84
	IC	2.72	0.01*	0.02	0.89	−0.01	0.91
	MIDS	2.27	0.03*	0.04	0.68	−0.04	0.67
	FO	2.62	0.01*	0.00	0.99	0.03	0.81
FFHFAy	FO	−0.85	0.59	0.24	0.02*	0.31	<0.01*

Table 8.4 Table demonstrating the relationship between gender, BMI and age with OFM joint angles.

When gender, age and BMI were controlled for, using multiple linear regression, AHI continued to be a statistically significant predictor for all angles except for FFHFAz at IC which was now non-significant (p = 0.088). Thus of the initial twenty values of interest, only nine were significantly associated with AHI.

8.4.3. Correlation between kinematic parameters and HRQOL scores

Significant negative correlations were observed between the OxAFQ-C physical domain score and MAX and MIN supination, as well as supination at MIDS and FO (Table 8.5). These correlations demonstrated moderate to large effect sizes with the strongest being observed with MIN supination (R = −0.39, p = 0.01). Thus the greater amount of forefoot supination during the stance phase of gait, the lower the questionnaire scores. None of the other kinematic parameters that correlated significantly with AHI had subsequent correlation with OxAFQ-C domain scores.

Angle	Parameter	Correlation (R) [95% C.I.]	Sig (p−value)
HFTBAx	LRMIN	0.06 [−0.23 to 0.34]	0.69
HFTBAz	MIN	0.19 [−0.10 to 0.45]	0.20
	MIDS	0.16 [−0.13 to 0.43]	0.28
FFHFAx	ROM	−0.17 [−0.43 to 0.13]	0.26
FFHFAz	MAX	−0.34 [−0.57 to −0.06]	0.02*
	MIN	−0.39 [−0.61 to −0.12]	0.01*
	MIDS	−0.35 [−0.58 to −0.07]	0.02*
	FO	−0.34 [−0.57 to −0.05]	0.02*
FFHFAy	FO	−0.12 [−0.39 to 0.18]	0.44

Table 8.5 Correlation between OFM joint angle measures and OxAFQ-C domain scores. Pearson's R tabulated as well as significance (Sig). * Denotes statistically significant result.

8.5. Discussion

In this chapter the relationship between foot posture and both joint kinematics and spatio-temporal parameters were assessed. There were a number of significant findings which in part are consistent with prevailing pathomechanic theories and previous published literature. Of the findings, however, only walking speed, cadence and forefoot supination in the PFF group were related to HRQOL. The work presented in this chapter is the first of its kind to relate HRQOL in children with PFF with dynamic function.

8.5.1. Spatio-temporal parameters

Study participants with PFF demonstrated a statistically significant reduction in normalized stride length compared to those with NF. On this basis one would also have expected to see a concomitant reduction in normalized walking speed, cadence or both. However, whilst there was a tendency for reduced normalized walking speed in the PFF group, this did not reach statistical significance, and no difference in cadence was observed. The study was underpowered to identify small effect sizes, and thus it is believed that this contributed to the non-significant difference in walking speed.

When the spatio-temporal parameters of the PFF group were analysed separately, worsening OxAFQ-C physical domain scores were significantly associated with slower normalized walking speed and cadence.

Of the studies using 3DGA to investigate differences between a flat and neutral foot posture only three have reported spatio-temporal parameters. Twomey et al. [319] did not demonstrate any significant difference in spatio- temporal parameters between flat and neutral foot posture groups in their population of nine to twelve year olds. They did not, however, normalize their parameters for leg length, which may have affected the results of their analysis. Levinger et al. [179] also found no significant differences in walking speed, cadence or stride length between groups. Their study population only consisted of ten subjects with neutral arches and nine with flat arches which suggests their study was underpowered for identifying group differences. The findings in this chapter are in part consistent with those published by Hosl et al. [129] who demonstrated that subjects with symptomatic flat feet (SFF) had slower normalized walking speed than typically developing feet (TDF) and asymptomatic flatfeet (ASFF). This was attributed to reduced normalized step length, although cadence was not reported. Increased normalized step width was also noted in the ASFF and SFF groups compared to the TDF, but no such differences were observed in the current study. The effect sizes for differences between groups, quoted by Hosl et al. [129] were moderate to large, but spatio-temporal parameters for each foot of each subject were treated as independent samples and pooled together. For reasons discussed previously, this is not necessarily appropriate, and will artificially inflate observed effect sizes.

The spatio-temporal findings observed in this study are possible evidence of biomechanical impairment caused by PFF in some individuals. Walking speed is known to correlate with the magnitude of joint moments, thus the adaptive behaviour of reducing one's self-selected walking speed may reduce the abnormal joint moments caused by altered joint kinematics, thereby serving to limit foot and ankle symptoms [280]. The effect of foot posture on foot and ankle joint moments will be investigated in Chapter 9.

8.5.2. The relationship between joint kinematics and foot posture

The overall shapes of the kinematic curves and the range of joint motions observed in this study were similar to previous studies using the OFM and other multi-segment foot models. It was hypothesized that a flat foot posture would be associated with a number of dynamic kinematic findings (section 8.2), including excessive hindfoot eversion, and a disrupted midtarsal locking mechanism, with concomitant effects on forefoot motion. However not all of the hypothesized relationships were observed.

The hindfoot in the PFF group was generally more everted that the NF group, which was consistent with the findings of Houck et al. [130] and Hosl et al. [129] (Figure 8.3 b)). Correlational analysis demonstrated significant positive correlations between AHI and MIN hindfoot inversion (maximum hindfoot eversion) and MIDS inversion. There was however no significant correlation at FO.

A reduction in hindfoot dorsiflexion due to a flat foot posture as previously demonstrated by Hosl et al. [129] was also hypothesized. Figure 8.3 a) was suggestive of a potential reduction in dorsiflexion in mid and late stance, but this did not prove to be statistically correlated with AHI. The only region of interest where a statistically significant relationship between AHI and hindfoot dorsiflexion angle was noted was plantarflexion during loading response (LRMIN), with flatter feet being more plantarflexed. Such a finding was also noted by Hosl et al. [129] but the significance of this is unclear. The effect size of this relationship was only small to moderate. In Chapter 6 (p.136-157) it was shown that only a small proportion of study participants had tightness of their plantarflexors such that their ability to attain plantigrade with the knee extended was impaired. This small proportion may be the reason why significant differences in dynamic dorsiflexion in stance were not been observed. Further subgroup analysis may demonstrate restricted dynamic hindfoot dorsiflexion in this group, but the sample size would be too small for meaningful results.

Significant correlations were observed between AHI and forefoot motion relative to the hindfoot in all three planes. The main finding was that foot posture correlated strongly with forefoot supination, such that the flatter the foot, the more the foot was supinated relative to the hindfoot. This proved to be the case for MIDS, FO, MAX and MIN, with a, verging on, moderate effect size observed. These findings were similar to those of Hosl et al.[129]. Twomey et al. [319] also found a significant difference between forefoot coronal plane motion between flat and neutral foot postures, although they found that both groups had forefoot pronation throughout the gait cycle. The stark contrast between study findings undoubtedly will be due to the method used to calculate these angles, as Twomey et al. [319] used the Heidelberg foot measurement method instead of the OFM. As such absolute angle values are not necessarily equivalent to those presented in this chapter. Nonetheless the shape of the kinematic curves presented by Twomey et al. [319] and the differences between flat and neutral feet are in part comparable to what was observed in this study.

With respect to forefoot motion in the transverse plane, there did not appear to be any difference in the amount of forefoot abduction throughout the majority of stance. A flatfoot posture was however associated with reduced adduction of the forefoot at FO, which was also consistent with some published findings [134].

One of the pathomechanic theories of why PFF can cause problems relates to the impaired midtarsal locking. It is hypothesized that increased hindfoot eversion, especially in late stance inhibits adequate locking of the midtarsal joint leading to a supple, instead of rigid midfoot thereby allowing increased forefoot abduction, dorsiflexion and supination. Excessive hindfoot eversion was noted in the PFF group, and this as well as the abnormal forefoot motions could all be potentially a result of impaired midtarsal locking.

8.5.3. Correlation between kinematics HRQOL

Out of the numerous kinematic parameters which correlated with AHI only forefoot supination had significant correlations with the OxAFQ-C physical domain scores. Negative correlations at MIDS and FO as well as MAX and MIN values were observed with the largest effect size noted with MIN ($R = -0.39$; moderate to large effect size). Thus increased forefoot supination was associated with worse HRQOL. In this observational study is it hard to assess the causality between hindfoot eversion and forefoot supination. To compensate for the hindfoot eversion and ensure the foot is flat to the ground in stance, a degree of forefoot supination would be expected. However, even in the swing phase of gait, the forefoot still remained more supinated in the PFF group. This would imply that in PFF children, the orientation of the forefoot relative to the hindfoot has changed, and it is not simply reacting to the hindfoot eversion in the stance phase of gait. In adult acquired flat foot, Parsons et al. [244] have drawn attention to the degree of resting forefoot supination observed. In their classification, the greater the supinatory deformity, the worse the disease stage. The pathology of adult acquired flat foot is distinct from PFF, but development of a supinatory deformity may be a common theme and relate to impaired biomechanical function of the foot and development of symptoms.

An important muscle stabilising the MLA by plantarflexion of the first ray is peroneus longus. Hunt et al. [134] demonstrated weakness of peroneus longus in adults males with flexible flat feet, and Angin et al. [6] demonstrated a reduction in cross-sectional area of this muscle in individuals with a flat foot posture. Excessive loading of the medial forefoot driving the increased forefoot supination could stretch peroneus longus leading to both these findings. Part of the symptom profile may be caused by stretching of the peroneus longus and medial foot capsuloligamentous structures.

8.5.4. Clinical significance

The evidence presented in this chapter not only demonstrates important functional differences between PFF and NF children, but also relates them to HRQOL as measured using the OxAFQ-C. As, ultimately, the goal of health care is to restore, or preserve HRQOL, these findings support the close monitoring and treatment of PFF in some children. The findings of this study also highlight which dynamic deformities are associated with symptoms, thereby guiding potential treatment strategies. Indeed, in a clinical setting, Mosca [221] has observed the existence of a forefoot supination deformity in some children with PFF. If surgical intervention is undertaken, it is his recommendation that additional procedures are required to address this deformity alongside the lateral column lengthening calcaneal osteotomy. A technique used successfully to achieve this is the plantar closing wedge osteotomy of the medial cuneiform [182]. However to reiterate the findings of Chapter 6 (p.136-157), a lateral column lengthening may encroach on the subtalar articular surface and thus an alternative procedure may be more appropriate.

8.6. Limitations and other considerations

The OFM is a much simplified representation of the foot and ankle [49]. As discussed in Chapter 2 (p.42-43) the midfoot including the midtarsal joint is likely to play an important role in PFF. Ideally direct information about the midfoot position and orientation would be useful, but the OFM does not model this segment. This was in part the motivation behind Chapter 7 (p.158-173). There are some MSFMs that include a midfoot segment and further subdivisions of the foot, and these intuitively may seem like a better option to use in such a study. However having more segments leads to more potential sources of error and no MSFM has been as extensively validated as the OFM. Thus in this chapter the relative motion of the forefoot to the hindfoot was used as a surrogate measure of midfoot function.

Symptoms associated with PFF may not manifest themselves during short duration straight line walking, and may only develop during more strenuous activities over longer periods. This is partially measured by the OxAFQ-C with questions specifically interrogating the effect of play and sports on HRQOL. Early muscle fatigue may be a contributory factor to symptom development, and it has been demonstrated that fatigue can alter gait parameters [253]. Differences in kinematics and spatio-temporal parameters between NF and PFF children may be even more marked during activities like running or hopping, and the effect of other functional activities on the 3DGA differences between NF and PFF children would be an interesting topic for future investigation.

Defining causation in a cross-sectional study is difficult, and at times it is only possible to state the relationship between two variables. Nonetheless one would expect any adaptation in spatio-temporal parameters to be caused by either biomechanical dysfunction, symptoms or both. To confirm that increased forefoot supination does cause symptoms it would be necessary to treat this deformity and reassess the patient using 3DGA and the OxAFQ-C. This is beyond the scope on this study.

8.7. Conclusion

Increased dynamic forefoot supination during gait is associated with worse HRQOL in children with PFF. The impairment in HRQOL may simply be caused by stretching of capsuloligamentous structures due to abnormal joint motion, however joint malalignment may also affect the loading of the foot and ankle joints. In Chapter 9, the effect of foot posture on foot and ankle loading will be assessed, as this may provide further biomechanical insight into why some children with PFF develop symptoms and impaired HRQOL.

Chapter 9. Do children with PFF demonstrate abnormal foot and ankle loading, instability or impaired propulsion?

9.1. Introduction

The prevailing theories behind why PFF can result in foot and ankle functional impairment and symptoms relate to an impaired midtarsal locking mechanism, with failure of adequate re-supination in late stance, and muscle fatigue [6, 111, 122, 137, 188, 225, 316]. The kinematic differences observed in the previous chapter have been presented as preliminary evidence of impaired midtarsal locking, and the purpose of this chapter is to investigate this further. It has been theorized that impaired midtarsal locking would lead to instability of the foot and ankle complex from mid to late stance affecting propulsion and altering joint loading. Analysis of the magnitude and point of application of the ground reaction force (GRF) and foot and ankle joint moments make it possible to investigate whether these characteristics are observed in the PFF population.

As previously stated in Chapter 3 (p.64), the GRF can be resolved into three orthogonal components. Of these the anteroposterior (APGRF) can provide insight into propulsion, and the vertical (VGRF), and mediolateral (MLGRF) can help provide insight into stability. The APGRF consists of two phases (Figure 9.1). From initial contact to midstance is a braking phase in which the centre of mass (COM) is decelerated, and from midstance to foot off, a propulsive phase is observed as the centre of mass is accelerated. The force time integral of each phase represents the braking and propulsive impulses, which give an indication of the rate of change of linear momentum of the COM [269]. These measures have been used to quantify the limbs' contribution to forward propulsion in gait after stroke, but have not been used in the context of PFF [30]. The peak value of APGRF is usually around 20% BW [269].

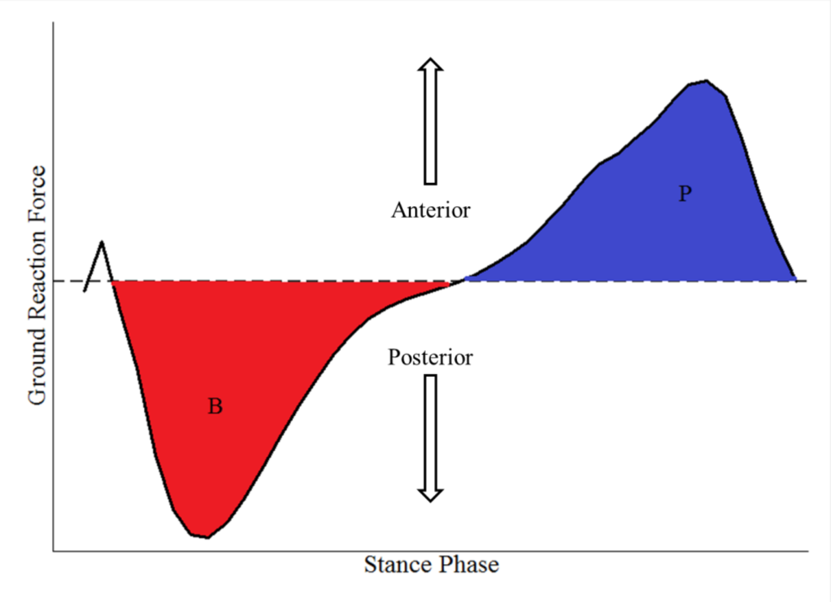


Figure 9.1 Anteroposterior GRF graph over stance phase of gait with direction of GRF marked. Red area B is the braking impulse, and blue area P is the propulsive impulse.

The VGRF curve has a characteristic double bump pattern with the maxima above BW and minimum below BW in non-pathological gait (Figure 9.2). These peaks and troughs are due to the response of the ground to upward and downward acceleration of the body COM during phases of single and double support [269]. Reduction in the height of the second peak of the VGRF has been observed in children with cerebral palsy and been described as 'deceleration deficiency' [344]. This refers to the inability to control the downward velocity of the COM in late stance, which may be as a result of weakness or instability. If the downward velocity of the COM is not counteracted the child could collapse to the ground. Thus in certain circumstances the contralateral leg intervenes to prevent this unsatisfactory outcome. In the context of PFF, an unlocked unstable midfoot, pain, or a combination of both could result in failure to adequately slow the path of the COM leading to reductions in the second peak of the VGRF.

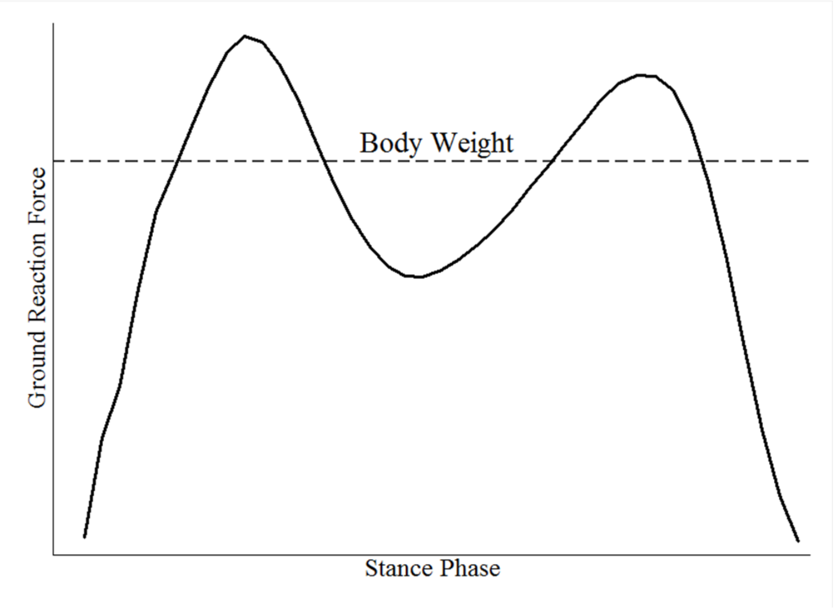


Figure 9.2 Vertical GRF graph over stance phase of gait, demonstrating characteristic double bump pattern in relation to body weight.

The mediolateral GRF (MLGRF) is the final component of the GRF, and the smallest in magnitude. It acts to dynamically balance the effect of the VGRF and body weight force couple on body rotation during gait [269]. Increased mediolateral motion of the body COM has been demonstrated in a number of pathological groups including cerebral palsy, Down’s syndrome and adolescent idiopathic scoliosis [57, 131, 169]. Failure to control the MLGRF may be a marker of instability, with an increased range of MLGRF acting as a surrogate marker of increased mediolateral motion of the body COM [144].

The GRF can be analysed in isolation or can be utilized to calculate joint kinetics (moments and powers) using the inverse dynamics method described in Chapter 3 (p.65). Knowledge of these kinetic quantities can be helpful in appreciating compensatory mechanisms and secondary abnormalities as a result of primary gait deviations. Standard gait models treat the foot as single segment, and thus tend to incorrectly estimate ankle joint kinetics [81]. As such the prospect of using a multi-segment foot model to gain insight into the joint kinetics within the foot and ankle segments is attractive. Few have tried this approach, and the reporting of MSFM kinetics is not yet mainstream in clinical practice [37, 187].

In the context of PFF it is theorized that dysfunction of gastrocsoleus due to excessive hindfoot valgus, and lever arm dysfunction due to a supple midfoot may lead to a reduction in peak internal ankle and midfoot moments in the sagittal plane. This would then manifest as reduced peak ankle and midfoot power generation (‘push off’). Alterations in foot segment position as demonstrated in the previous chapter may also lead to internal moments that counteract or attenuate the desired re-supination process.

The analysis of joint moments often involves assessment of peak moments during gait. However peak moments only reflect a single time point, and the total exposure to loading that occurs through stance may be more important. Integrating the moment over time to provide the moment impulse may provide more clinically relevant data. This approach has been utilized in knee osteoarthritis where the knee adduction moment impulse has been found to be more sensitive at distinguishing different severities of disease compared to the peak adduction moment [153, 165]. This method has not yet been used in the foot and ankle.

The influence of the GRF on joint moments is directly related to the point of application of the GRF, also known as the centre of pressure (COP). In normal heel-toe gait, foot loading begins under the heel, progressing through stance to the metatarsal heads and finally the hallux [234]. In pathological situations deviations may be observed in the location, path of progression of the COP, as well as its duration in each foot region (hindfoot, midfoot and forefoot) [282]. In the context of PFF the COP progression (COPP) has been utilized as a measure to assess the outcome of flat foot surgery [339]. Jameson et al. constructed a reference map of the position of the path of COPP from 60 children with a normal foot posture [140]. In cases of PFF where some medial deviation of the path of COPP was noted during dynamic pedobarography, part of a successful surgical outcome was rectification of the path of COPP closer to the mean path of COPP in normal individuals. This practice is based on the assumption that PFF children have a significantly medially deviated path of COPP compared to NF children. This is something that has yet to be formally demonstrated. If there is evidence of midfoot instability then this may also affect how the COP progresses through the foot.

Thus the GRF, COPP and joint kinetics are tools to aid investigation of propulsion, stability and foot and ankle joint loading. As well as describing the relationship between a flat foot posture and these measures it is imperative to put the findings into a clinical context. The HRQOL data collected on each subject makes this comparison possible.

9.2. Aims and hypotheses

The aim of this chapter is to use the GRF, joint kinetics and COPP to investigate whether children with PFF demonstrate foot and ankle complex instability leading to postural instability, reduced propulsion and abnormal foot and ankle loading. The positive findings will also be related to HRQOL.

The hypotheses are as follows:

1. A flat foot posture will be associated with the following GRF findings:

Decreased second peak VGRF

Decreased peak propulsive APGRF and ↓ APGRF propulsive impulse

Increased range of MLGRF

2. The COPP of children with PFF will be medially deviated and show delayed progression from the midfoot to forefoot.

3. A flat foot posture will be associated with the following main kinetic findings:

Decreased peak ankle plantarflexor moment

Decreased peak midfoot plantarflexor moment

Increased Ankle eversion moment impulse

Decreased peak ankle inversion moment

Decreased peak midfoot pronation moment

Decreased peak midfoot and ankle power generation

4. Based on the kinematic results it is hypothesized that the peak midfoot pronation moment will have the strongest correlation with HRQOL in the PFF group.

9.3. Methods

The general methods for data collection have previously been outlined in Chapter 3 (p.55-92). Specific methods are noted below.

9.3.1. Ground Reaction Force

Ground reaction force data were collected for all 95 study participants. The GRF was resolved into its three components (APGRF, MLGRF and VGRF), and reported as a percentage of body weight. A representative GRF trial was selected using the minimum RMSD between trial and mean trial for each subject as per the method outlined in Chapter 8 (p.176-179). The stance phase of gait was normalized to 101 data points.

Preliminary data analysis began with the inspection of GRF graphs with subjects pooled by foot type. Regions of interest (ROI) were identified, as in Chapter 8, with particular focus on the regions where differences were hypothesized. Discrete GRF ROI parameters were then correlated, using Pearson’s correlation coefficient, with the AHI as a continuous measure of foot posture. Where a statistically significant correlation was noted between AHI and GRF ROI, the confounding effect of age and gender was assessed. Where necessary stepwise multiple linear regression was undertaken to control for the confounding effect of these variables.

9.3.2. Centre of Pressure Progression Calculation

The position of the COP can be calculated in a number of ways, but for ease of comparison with previously published work in the paediatric population the COPP was calculated using dynamic pedobarography [140, 339]. Data were collected on 47 children with PFF and 47 children with NF. It was not possible to collect dynamic pressure plate data for one child with PFF due to compliance issues, and inability to adopt a normal walking pattern over the pedobarograph.

A representative plantar pressure trial was selected, arbitrarily, for the left foot of each participant. This trial was selected by visual inspection of consistency with other trials, and invariably was the final trial that the child had undertaken, as at this stage they were proficient with the technique of contacting the pressure plate properly. The first stage of quantifying the position of the centre of pressure progression line was to identify the long axis of the foot. The foot axis was determined as the line that bisected the long plantar angle on the maximum pressure print (MPP), with the long plantar angle being defined as the angle formed by the point of intersection of the tangents of the medial and lateral borders of the foot (Figure 9.3). Two points on this line were identified automatically in the heel and forefoot (O and O’) by the Novel software [236]. A rotation matrix was constructed to align the long axis of the MPP with the longitudinal axis of the pressure plate and the print was translated such that the pre-defined point O became the origin. Foot length and width were calculated as the absolute range of y and x values respectively of the maximum pressure footprint.

The coordinates of the centre of pressure for each frame of the dynamic pedobarograph with respect to the pressure plate itself were output directly from the

Novel software as an ASCII file. These coordinates underwent the same rotation and translation to align them with the foot-axis-based coordinate system. The position of the COPP with respect to the long axis of the foot as a proportion of foot length (vertical component) and width (horizontal component) were then calculated for each frame, and then interpolated to 100 points to correspond to percentage foot length. The location of the COP with respect to time was also normalized to 100 points and the percentage of roll over process (ROP) in each foot region was calculated (ROP equivalent to stance phase of gait). The foot regions were calculated according to the Novel pressure plate software default settings. The division between heel and midfoot was at 27% along the length of the foot from the heel and the division between mid and forefoot at 55% [236].

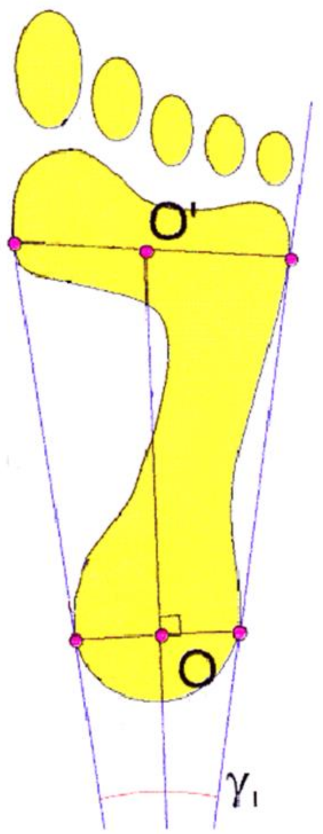


Figure 9.3 Figure demonstrating method to determine long axis of the foot γ_1 represents long plantar angle. Point O represents the midpoint of heel on the long plantar angle bisector, and O' represents the midpoint of the forefoot on the long plantar angle bisector. (adapted from [236]).

This process was performed for each subject representative trial and trials were pooled by foot posture (PFF or NF). Mean COPP with 95% percent confidence bands were constructed for both groups and the same procedure was undertaken for the difference between group means for each percent along the foot. Where the confidence band of the difference between groups did not cross zero there was a statistically significant difference in the position of the COP between groups. An unpaired, two-tailed Student's t-tests was used to compare duration of the COP in each foot region between groups.

9.3.3. Oxford Foot Model Kinetics

The OFM kinetics were calculated according to the methods outlined in Chapter 3 (p.64-66), using the application of the inverse dynamics method to the OFM described by Dixon et al. [81]. Moments and powers were normalized to body weight. Midfoot kinetics were cropped and normalized to the final 40% of stance as per the recommendations of Dixon et al. [81]. Kinetic analysis had a similar flow to that of the kinematic and GRF analysis, with the identification of ROIs by visual inspection of kinetic graphs, followed by formal correlational analysis. Where the significance of correlation was $p < 0.1$ the effect of confounding by age and gender was assessed, and where required, controlled for using stepwise multiple linear regression. The potential regions of interest were limited to peak joint moments, and the evertor moment impulse. The peak moments were all normally distributed; however the distribution of the evertor moment impulse demonstrated marked positive skewness. As such a cube root transformation of the evertor moment impulse was undertaken for data analysis.

Oxford Foot Model kinetics could only be calculated for 94 of 95 subjects for the reason stated in Chapter 8 (p.176).

9.3.4. Correlation of gait kinetics with HRQOL

From the processes outlined in section 9.3.3 a list of kinetic parameters were obtained that had a statistically significant relationship with arch height as measured using the AHI. These values were then correlated with OxAFQ-C physical domain scores solely in the PFF group using Pearson’s correlation coefficient to identify whether deviations in foot and ankle joint kinetics were related to HRQOL.

Differences in GRF and COPP between groups were not correlated independently with HRQOL, as these both are used to calculate joint moments. The belief was that any statistically significant associations between either GRF or COPP and foot posture would be borne out through the joint moments.

9.3.5. Power Calculation for Centre of Pressure Progression

The power analysis outlined in the previous chapter applied similarly for the GRF and kinetic analysis. In the case of COPP, t-test analysis had 80% power to detect an effect size (Cohen's d) of 0.58 (medium effect size) [64]. This was based on a combined sample size of 94 with equal allocation to NF and PFF groups.

9.4. Results

9.4.1. Ground Reaction Force

A number of regions of interest were identified from the GRF curves and highlighted in Figure 9.4. The most striking differences were, increased peak medial GRF, and reduced second peak of the VGRF in the PFF group. The APGRF propulsive impulse and range of MLGRF were impossible to assess visually, thus were included in subsequent correlational analysis. In total seven ROIs were identified and are summarized in Table 9.1 with the Pearson's correlation coefficient and significance value when correlated with AHI.

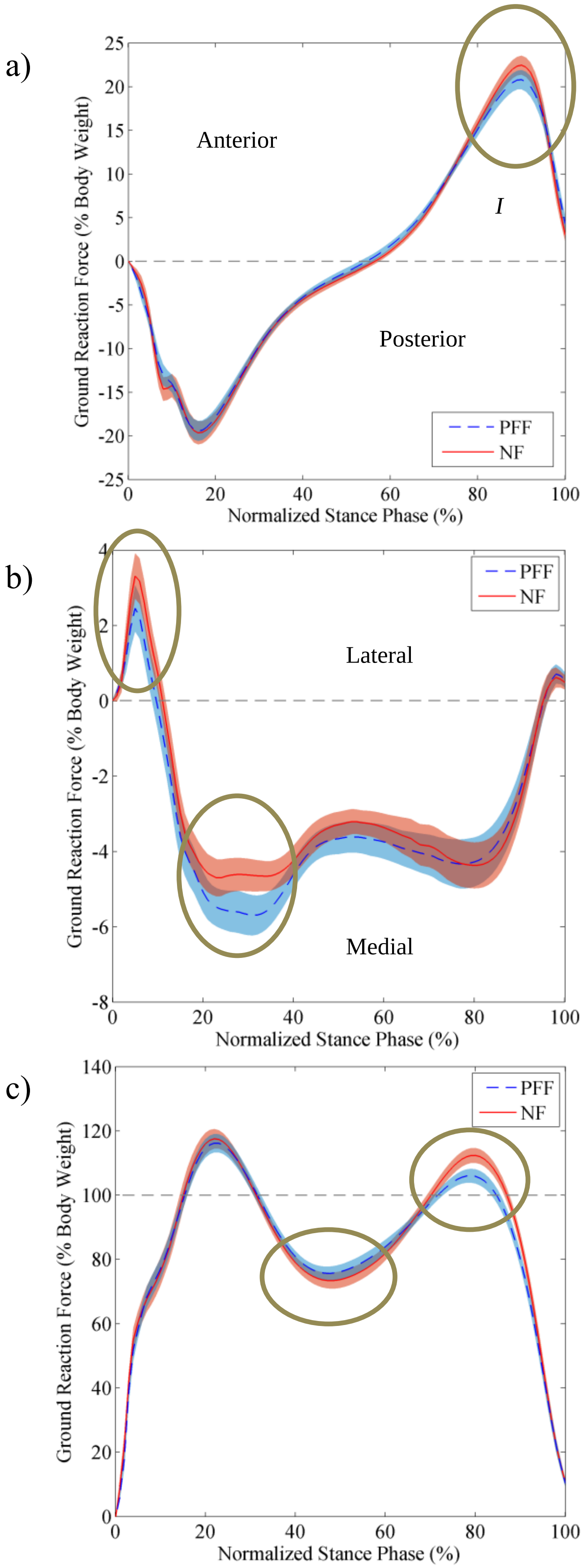


Figure 9.4 Normalized stance phase curves for the GRF. Mean curve for each group along with 95% confidence bands plotted. Regions of interest marked.

a) APGRF, with ROIs of peak propulsive force (AMAX), and propulsive impulse (*I*).

b) MLGRF, with ROIs of peak medial GRF (MMAX) and peak lateral GRF (LMAX). Range of MLGRF (MLRANGE).

c) VGRF, with ROIs, midstance trough (VMIN), and second peak (VMAX2).

GRF component	Parameter	Correlation with AHI	
		R	Sig (p)
X	AMAX	-0.02	0.86
	I	0.04	0.74
Y	LMAX	0.21	0.04*
	MMAX	-0.20†	0.05*
	MLRANGE	0.03	0.76
Z	VMAX2	0.24	0.02*
	VMIN	-0.05	0.66

Table 9.1 Regions of interest identified from GRF graphs correlated with AHI. Pearson's R and significance (Sig) given. * Denotes a statistically significant result. † Correlation made with absolutes value of MMAX.

Of the seven ROIs only three (LMAX, MMAX and VMAX2) had significant correlations with AHI. Investigation of the potential confounding effect of gender and age only showed that age may have confounded VMAX2 (Table 9.2).

GRF Component	Parameter	Gender		Age	
		Diff (% BW)	Sig (p)	R	Sig (p)
Y	LMAX	-0.12	0.08	0.10	0.34
	MMAX	-0.68	0.61	-0.19†	0.06
Z	VMAX2	0.07	0.97	0.29	0.01*

Table 9.2 Table demonstrating the relationship between gender and age with GRF parameters. * Denotes a statistically significant results. † Correlation made with absolute values of MMAX.

When age was controlled for, using multiple linear regression, AHI continued to be a statistically significant predictor of VMAX2 (p = 0.018).

Thus a flatter foot posture was associated with a significant reduction in the second peak VGRF, a significant reduction in peak lateral GRF, and a significant increase in peak medial GRF.

9.4.2. Centre of Pressure Progression

The centre of pressure progression analysis could only be performed on 45 of the 47 children with PFF, as two of the children had a COPP path that did not follow the standard heel toe trajectory (Figure 9.5). For the remainder, the overall shape of the mean COPP curve between groups was similar, with the COPP in close proximity to the long axis of the foot as it progressed through the hindfoot and midfoot (Figure 9.6). As the roll over process progressed the COP moved to the forefoot and demonstrated marked medial deviation to locate it under the 1st ray.

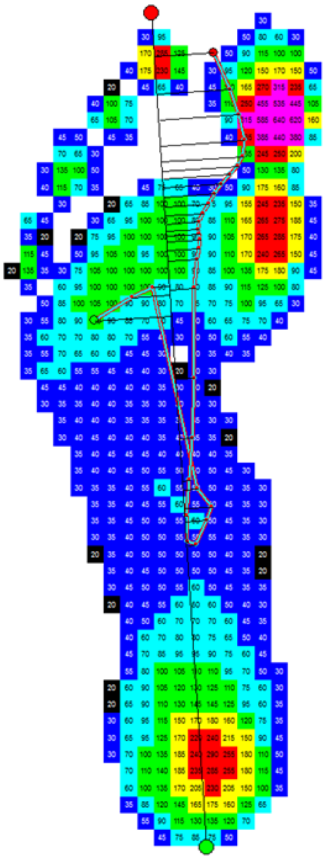


Figure 9.5 Example of COPP of one of the subjects excluded for data analysis. COPP does not progress from heel to toe in one direction making the analysis used in this chapter impossible. In this case initial contact occurs at the midfoot, with the COPP moving heel-ward before it progresses to the Hallux region.

Figure 9.7 demonstrates the difference in mean COPP between groups with associated 95% confidence intervals. The mean COPP line demonstrated that overall, there was a tendency for the PFF group to have a more laterally placed COPP compared to the NF group. This persisted until the middle third of the forefoot when the overall trend was for the COPP in the PFF group to be more medially placed than the NF group. Aside from a small region in the hindfoot the 95% confidence intervals always crossed zero implying no statistically significant difference between groups.

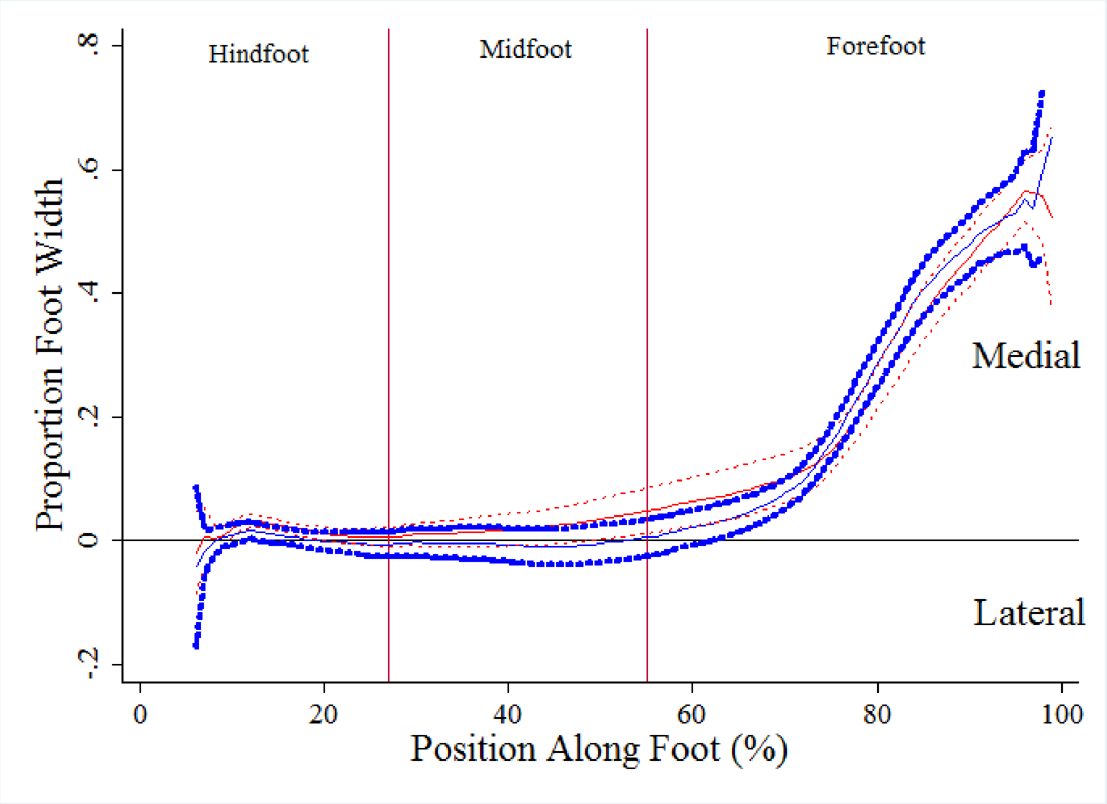


Figure 9.6 Graph demonstrating position of the COPP versus position along foot (%) by foot type. Solid lines represent mean COPP for group (red = NF and blue = PFF), and dotted lines mark 95% confidence bands. Medial and lateral directions marked on the graph.

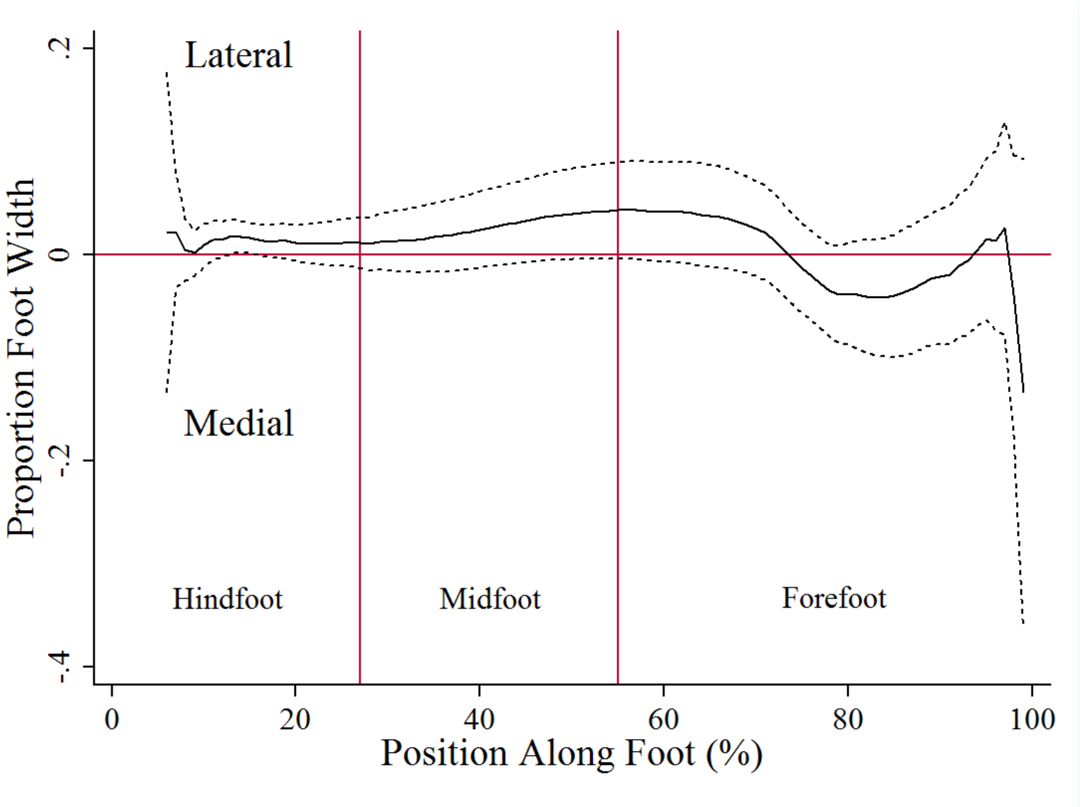


Figure 9.7 Graph demonstrating mean difference (solid line) in COPP versus position along foot (PFF – NF) with 95% confidence bands (dotted lines). Foot regions marked. Lateral deviation is positive on y-axis.

There were no significant differences in the proportion of the ROP spent in each foot region between foot posture groups (Table 9.3).

Foot Region	Foot Type PFF % ROP Mean (± SD)	NF % ROP Mean (± SD)	Significance (p)
Hindfoot	28.4 (± 8.9)	26.3 (± 9.3)	0.3
Midfoot	21.1 (± 6.9)	21.6 (± 8.0)	0.8
Forefoot	50.0 (± 9.6)	52.6 (± 9.6)	0.2

Table 9.3 Percentage of ROP spent in each foot region by foot type. Mean values and standard deviation (SD) given as well as significance value (p) based on comparison of groups with unpaired, two-tailed Student’s t-test.

9.4.3. Oxford Foot Model Kinetics

The main findings from visual inspection of the OFM ankle kinetic graphs (Figure 9.8) were in the coronal and transverse plane moments, with the PFF generally having greater eversion and internal rotation moments, with reduction in peak inversion and external rotation moments. In the sagittal plane there was a tendency for the PFF group to have reduced peak hindfoot dorsiflexion moment. From these graphs four ROIs were identified (DMAX, IMAX, EvI and ExtMAX).

At the level of the midfoot the magnitude of midfoot sagittal moments was markedly smaller than the ankle sagittal moments, but the coronal and transverse moments were equivalent in magnitude (Figure 9.9). There was a general trend for a reduction in dorsiflexion, pronation and adduction moment, but only one ROI fulfilling the a priori criteria was identified (PROMAX).

Visual inspection of ankle and midfoot power graphs (Figure 9.10) was essentially unremarkable aside from the tendency for the PFF group to have reduced peak ankle power generation (APowMax).

Correlation between kinetic ROI parameters and AHI demonstrated a significant relationship for peak ankle inversion moment (IMAX, $p = <0.01$) and eversion moment impulse (EvI, $p <0.01$), such that a flatter foot posture was associated with a smaller IMAX, and a larger EvI. A significant correlation was also noted between AHI and the peak ankle external rotation moment (ExtMAX, $p = 0.05$). Peak ankle power generation (APowMAX) demonstrated a near significant correlation with AHI ($p = 0.07$). These findings are summarized in Table 9.4. Controlling for the effect of gender and age on ankle kinetics did not alter the significance of any of the above relationships.

Thus in summary a flatter foot posture was associated with a reduction in peak ankle inversion moment, an increase in eversion moment impulse and a reduction in peak external rotation moment.

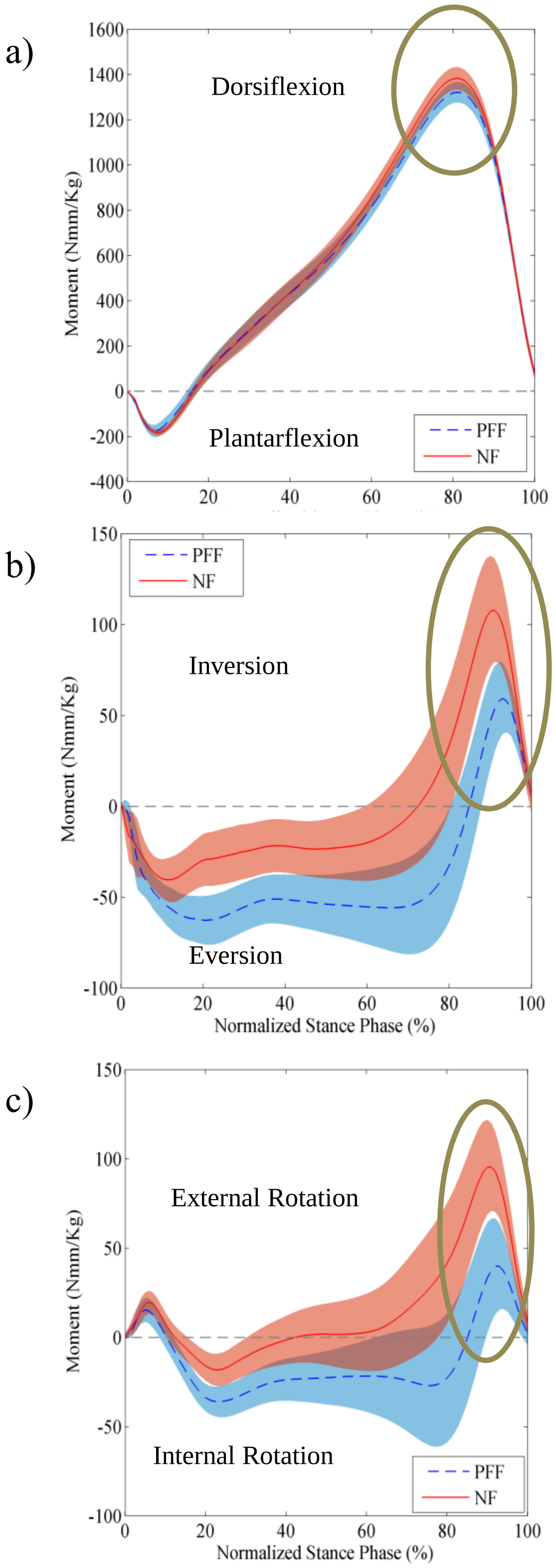


Figure 9.8 Normalized stance phase curves for ankle moments. Mean curve for each group along with 95% confidence bands plotted. Regions of interest marked.

a) Ankle moment x, with ROI of peak dorsiflexor moment (DMAX).

b) Ankle moment y, with ROIs of peak invertor moment (IMAX) and evertor moment impulse (EvI).

c) Ankle moment z, with ROI of peak external rotation moment (ExtMAX).

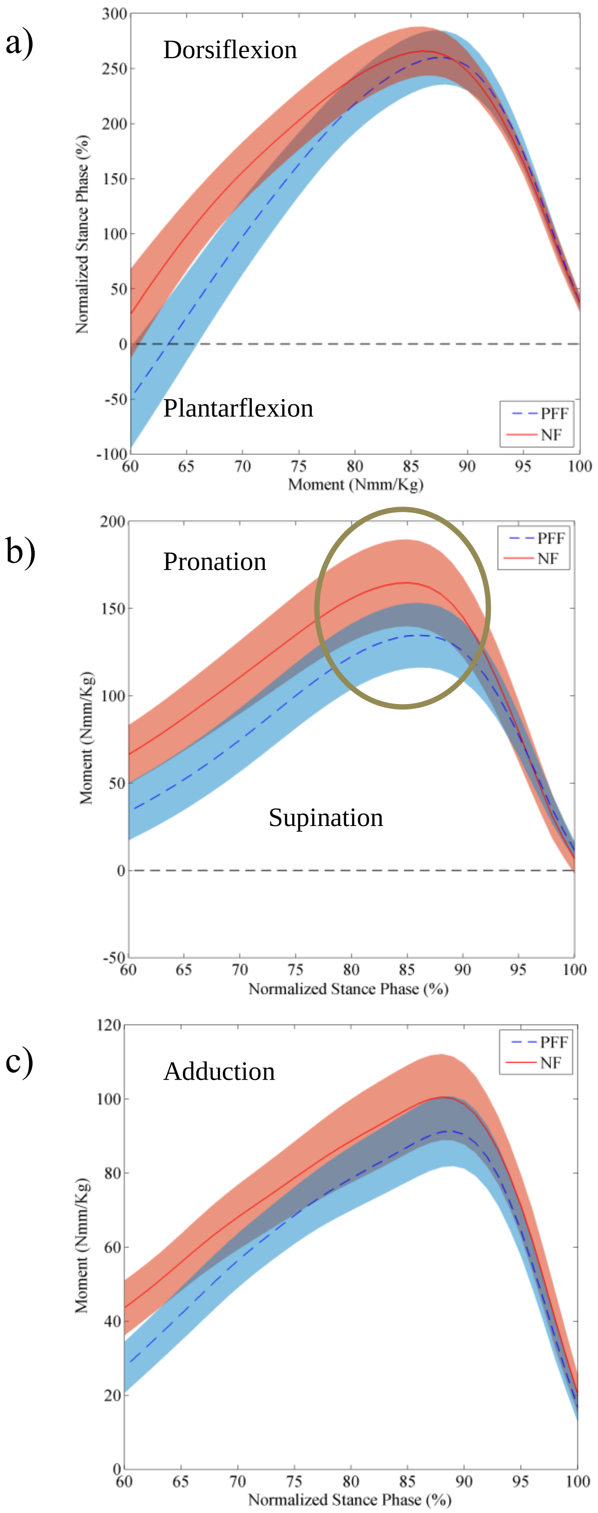


Figure 9.9 Midfoot moments for 60% - 100% of normalized stance phase. Mean curve for each group along with 95% confidence bands plotted. Regions of interest marked.

a) Midfoot moment x, with no ROIs.

b) Midfoot moment y, with ROI of peak pronation moment (PROMAX).

c) Midfoot moment z, with no ROIs.

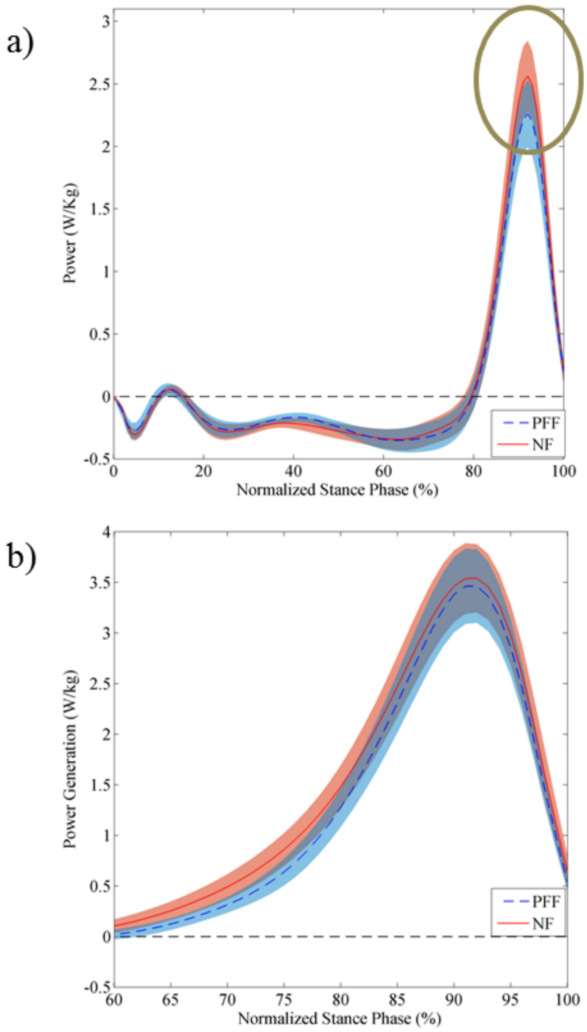


Figure 9.10 Ankle power graph (a) for normalized stance phase of gait and midfoot power graph (b) from 60% to 100% of normalized stance phase. Mean curve for each group along with 95% confidence bands plotted. Region of interest is peak ankle power generation (APowMax).

Moment/Power	Parameter	Correlation (R)	Sig (p-value)
Ankle Moment x	DMAX	0.09	0.41
Ankle Moment y	IMAX	0.42	<0.01*
	EvI	-0.50	<0.01*
Ankle Moment z	ExtMAX	0.20	0.05*
Midfoot Moment y	ProMAX	0.13	0.20
Ankle Power	APowMAX	0.19	0.07†

Table 9.4 Regions of interest identified by visual inspection of OFM kinetic graphs. The relationship between ROIs and AHI quantified with Pearson’s R and significance (Sig). * Denotes a statistically significant result. † denotes significance 0.05 < p < 0.1.

Moment/Power	**Parameter**	**Gender**		**Age**	
		Diff	Sig (p)	R	Sig (p)
Ankle Moment y	IMAX	-42.29 (Nmm/Kg)	0.02*	0.08	0.45
	EvI	1.9 (Nmm/Kg*%)	0.07	-0.07	0.50
Ankle Moment z	ExtMAX	-32.90 (Nmm/Kg)	0.06	0.10	0.31
Ankle Power	APowMAX	-0.10 (Watts/Kg)	0.60	0.21	0.04*

Table 9.5 Table demonstrating the relationship between gender and age with kinetic parameters. * Denotes a statistically significant result.

9.4.4. Correlation between kinetic parameters and HRQOL scores

None of the shortlisted kinetic parameters from section 9.4.3 had significant correlations with the OxAFQ-C physical domain scores (Table 9.6).

Moment	**Parameter**	**Correlation (R) [95% C.I.]**	**Sig (p-value)**
Ankle Moment y	IMAX	0.07 [-0.22 to 0.35]	0.65
	EvI	-0.11 [-0.40 to 0.18]	0.44
Ankle Moment z	ExtMAX	-0.02 [-0.31 to 0.27]	0.88

Table 9.6 Correlation between OFM joint kinetic parameters and OxAFQ-C physical domain scores. Pearson's R tabulated with 95% C.I. and significance (Sig).

9.5. Discussion

The aim of this chapter was two-fold. Firstly the GRF measurements, position and time-course of the COP and foot and ankle joint kinetics were used to investigate the presence of foot and ankle instability, impaired propulsion and abnormal joint loading in the PFF population. Secondly any positive findings in joint kinetics were to be put in a clinical context by correlating them with HRQOL.

Vertical GRF, MLGRF and ankle coronal and transverse moments all had a significant correlation with arch height. However subsequent subgroup analysis of the PFF participants failed to demonstrate a significant relationship between HRQOL and ankle moments.

9.5.1. Foot and ankle instability

The presence of foot and ankle instability was assessed by investigating vertical and mediolateral ground reaction forces. As hypothesized a flat foot posture was associated with a reduction in VMAX2, although the ES was only medium to small. In normal gait the VMAX2 is generally of a similar magnitude to the first peak [310]. It has been suggested that reduction in the VMAX2 as observed in many children with cerebral palsy, represents a degree of failure to adequately decelerate the body COM in late stance [344]. If the second peak drops below BW this represents significant instability, or weakness and may relate to the severity of the pathology. In this chapter, whilst the VMAX2 was significantly reduced in children with a flatter foot posture, the mean value of the PFF group was still above BW. In fact only a small proportion of the PFF group had their VMAX2 less than BW. This suggests that whilst there may have been some ‘decelerator deficiency’ in the PFF group it was not at the level which would truly threaten collapse. It is conceivable that this reduction in VMAX2 may be related to impaired midtarsal locking with a more supple midfoot failing to adequately resist the downward acceleration of the COM. Alternatively this behaviour may be adaptive to reduce symptoms or discomfort caused by the PFF. That said, if it were purely related to symptoms then one might expect a reduction in the first GRF peak as well. Deceleration deficiency may require compensation by the contralateral foot during loading response, but there were no significant differences between groups at loading response to suggest this. Presumably this was because the reduction in VMAX2 did not go below BW.

Stansfield et al. [299] refute the suggestion that a reduced VMAX2 is pathological as they intermittently observed a reduced VMAX2 in a sample of 25 normal children they observed for seven years in a longitudinal study. Interestingly they had not assessed foot posture in their sample so it is unclear whether PFF could have resulted in these observations.

Increased medio-lateral excursion of the COM has been associated with a number of pathologies, and failure to control mediolateral motion can cause sideways falls in the elderly [113]. The MLGRF counteracts deviations of the COM in the mediolateral direction, and thus its magnitude may give an indication of the relative motion of the COM [144]. Increasing walking velocity leads to an increasing range of MLGRF, which itself may be a marker of increased instability. It was hypothesized that instability as a result of an excessively supple midfoot in the PFF group would result in an increased MLGRF range. Reduced AHI did correlate with a significant reduction in LMAX and increase in MMAX (medium to small ES); however AHI did not correlate significantly with the MLGRF range. The medial shift of the MLGRF curve in early stance in the PFF group was likely to be due to the increased hindfoot eversion inherent to this population.

9.5.2. Impaired Propulsion

To evaluate whether a flat foot posture resulted in impaired propulsion in gait a combination of methods was used including quantifying the APGRF, sagittal plane OFM moments, OFM power generation and assessment of the progression of the COP along the foot.

It was also hypothesized that a flat foot posture would be associated with a reduced AP impulse and AMAX, however this was not observed. This component of GRF was investigated as it has been used to measure the contribution of the paretic leg to walking after stroke, and has been shown to reflect the severity of paresis [30]. Hemiplegia caused by stroke is clearly a much more severe pathology than PFF, and for some subjects Bowden et al. [30] only found small differences between healthy and paretic legs. As such this mode of analysis may be relatively insensitive to impairments in propulsion caused by PFF. Indeed propulsion is a result of contributions from the whole limb, and this may outweigh the influence of foot posture alone.

A reduction in peak ankle and midfoot dorsiflexion moments and power generation would have been consistent with impaired propulsion in the PFF population. Whilst there was an initial suggestion of reduced DMAX and APowMax on the basis of visual inspection (Figures 9.9 a) and 9.10 a)) formal statistical analysis did not prove this to be the case.

There is limited literature on the kinetics of PFF to compare with our findings, with different methods and conflicting results. Hunt et al. [134] demonstrated an increased peak ankle plantarflexor moment at push-off in their flat foot group compared to normals, but Hosl et al. [129] demonstrated reduced ankle power in their symptomatic PFF group. Both Hunt et al. [134] and Hosl et al. [129] calculated ankle joint moments using the standard lower limb gait model (i.e. non-segmented foot). Saraswat et al. [276] calculated segmented foot and ankle joint kinetics in a group of ten subjects with pes planovalgus. They demonstrated a reduced ankle plantarflexion moment and ankle power in their pes planovalgus group compared to normals. Foot posture was not related to a reduced plantarflexion moment in this chapter, but not only did Saraswat et al. [276] pool feet for statistical analysis, they had a heterogeneous group of flat feet, including those with over-corrected clubfoot and connective tissue disorders. As such it is difficult to make a direct comparison.

The majority of studies reporting the COP focus on differences in mediolateral position of the COP along the foot in different patient groups [56, 140, 339]. The COP actually has another dimension, that of time. In this chapter the temporal progression of the COP along the foot was also investigated with the belief that in PFF an excessively supple midfoot resulting in lever arm dysfunction would manifest as delayed progression from the second to third rocker, with the COP remaining in the midfoot for an increased duration. Formal comparison of PFF and NF groups however did not demonstrate such findings, and thus as a group children with PFF did not demonstrate rocker dysfunction.

9.5.3. Abnormal foot and ankle loading

The loading of the foot and ankle was also evaluated by investigating the OFM coronal and transverse plane moments, and COPP to quantify any deviation in the point of application of the GRF.

The COP was investigated in this context for two reasons. Firstly knowledge of the path of COPP may provide further insight into observed joint kinetics. Secondly as Westberry et al. [339] use the COPP as a measure of successful outcome for surgically managed pes planovalgus, it was expected to provide further insight into the functional differences between PFF and NF groups.

In this chapter no major differences were found between PFF and NF groups for either position of the COPP or transition through the foot. Whilst it was hypothesized that the PFF group would demonstrate greater medial deviation of the COP than the

NF group, particularly in the midfoot, the opposite tendency was observed, although this did not reach statistical significance. The only area where a statistically significant difference between groups existed was a very small region in the hindfoot, but this was not felt to be clinically significant and did not correspond to any hypothesis.

One of the factors that may have affected the results was the heterogeneity in the dynamic footprint of the PFF group. Figure 9.11 demonstrates the maximum pressure print with COPP for two subjects from the PFF population. In Figure 9.11 a) increased pressure is observed under the locality of the navicular bone. This will have the tendency to pull the COPP medial to the long axis of the foot. In contrast 9.11 b) has increased pressure under the locality of the cuboid bone and this has the tendency to shift the COPP lateral to the long axis of the foot. Thus both subjects are classified as having PFF, but both have markedly different COPP, particularly in the midfoot. As such when results are pooled as a group these deviations are cancelled out. As Jameson et al. [140] observed medial deviation of the COPP in their flat foot population it may have been as a result of having more patients like Figure 9.11 a).

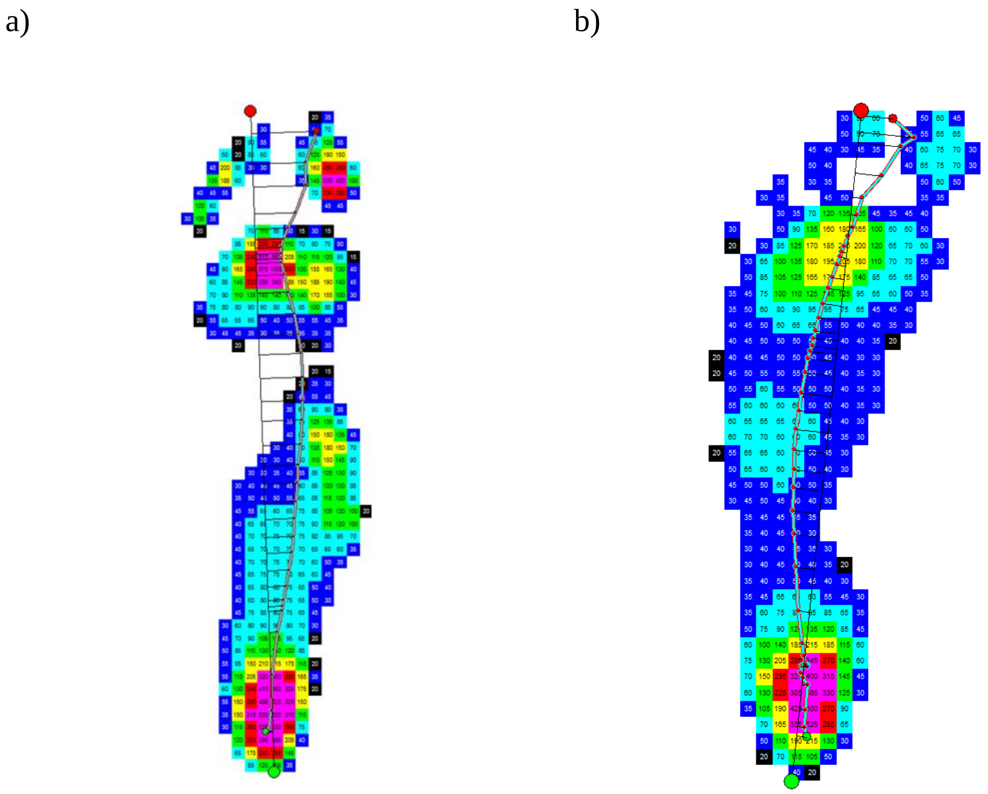


Figure 9.11 Figure demonstrating variability in COPP depending on whether increased pressure is observed under the navicular (a) or cuboid (b).

For the reasons outlined above application of COPP using this method may not be entirely appropriate to assess group differences in PFF, although may have some utility on a case by case basis. In this chapter the COPP has been referenced to the MPP. It is known from the previous chapter that foot and ankle kinematics vary significantly with a flat foot posture, thus assessing the COP in relation to the foot and ankle joint centres may demonstrate quite different findings, and would be an interesting avenue for future investigation.

Of the hypotheses made in section 9.2, significant results were only found in the hindfoot coronal plane kinetics. Foot posture as measured using the AHI demonstrated highly significant correlations with peak ankle inversion moment and evertor moment impulse, with moderate to large, and large ES respectively. Whilst not formally tested, from visual inspection of the ankle coronal plane moment graph (Figure 9.9 b)) it appeared that a net inversion moment began much later in stance in the PFF group. These findings add further weight to the suggestion that the re-supination process is affected in individuals with PFF.

Significant differences were not initially hypothesized in the transverse plane ankle moments, however a reduction in peak ankle external rotation moment was significantly associated with a flatter foot posture. The ES for this observation was however only moderate to small. A delay in transitioning to a net external rotation moment in the PFF group was observed in Figure 9.9, which echoes what was observed in the coronal plane. As ankle external rotation coupled with hindfoot inversion and locking of the midtarsal joint is required for re-supination in late stance, these findings are also of significance.

The transverse plane findings are consistent with those of Saraswat et al. [276]; however, they did not observe such differences in the ankle coronal plane. The magnitude of both coronal and transverse plane ankle moments are small relative to the sagittal moment, and thus contribute little to ankle power generation and propulsion.

Visual inspection of the midfoot kinetic graphs demonstrated trends to potentially support the hypotheses in section 9.2, but these were not subsequently shown to be statistically significant. This again contrasts with the findings of Saraswat et al. [276] who demonstrated a reduction in midfoot power generation in their flat foot group.

9.5.4. Correlation between kinetics and HRQOL

None of the three kinetic parameters identified as having a significant relationship with AHI correlated with HRQOL in the PFF group. This does not support the hypothesis made in section 9.2. Thus whilst abnormal foot and ankle joint loading is observed in the PFF group this is not directly contributing to the presence of foot and ankle symptoms.

9.6. Limitations and other considerations

Implementing the methods utilized by Jameson et al. [140] and Westberry et al. [339] to calculate the COPP led to a key difficulty; the method relies on the COPP only travelling along the foot in one direction. As was observed in the PFF population this is not always the case (Figure 9.5). The two subjects that demonstrated similar COPP patterns both had evidence of plantarflexor contracture on clinical examination, and as such did not attain a normal heel strike. As plantarflexor contracture is associated with more symptomatic PFF it is likely that this method of analysis would not be appropriate when this is feature is present [120, 129].

The limitations of the kinetics methods have been described in part in Chapter 3 (p.65). The calculation of midfoot kinetics is in relative infancy, and their inclusion in this chapter was mainly for exploratory purposes. The significant limitation of the method utilized in this chapter is that midfoot kinetics can only be described after heel lift in gait, which is approximately after 60% of stance phase. As such midfoot kinetics for the majority of stance are not calculated. To quantify midfoot kinetics for the whole of stance plantar pressure and force plate measurements can be combined using methods similar to those described by MacWilliams et al. [187], but this is computationally difficult.

Using inverse dynamics methods also relies on numerous assumptions, and processes which can lead to the propagation of error (discussed in Chapter 3 p.65). What results, is an apparently noisy signal, and particularly in the context of joint moments with small magnitudes, the signal to noise ratio becomes very small. This is evidenced by the broad 95% confidence bands observed in many of the figures presented in this chapter. Larger standard errors mean that to even register a moderate difference in joint kinetics between groups, sample sizes need to be very large. This increases the likelihood of type II errors especially for the more subtle kinetic differences between groups. It is likely that several hundred more study participants would be required to mitigate the adverse signal to noise ratio. This goes beyond the scope of this thesis.

Ground reaction forces and joint kinetics are sensitive to walking speed, with increasing walking speed leading to increased ranges and magnitudes of these measures [144, 280, 299]. In the previous chapter differences were noted in spatio-temporal parameters between PFF and NF groups, and the more symptomatic a flatfoot the slower the normalized walking speed. For the purposes of this study walking speed was not used as a covariate in statistical analysis as it was felt to be intimately related to the PFF deformity, and controlling for it would be artificial. It is likely that alterations in walking speed would have had concomitant effects on joint moments, but with the data available it is impossible to speculate on the magnitude of the effect. In the context of GRF, the reduction in VMAX2 was not associated with a similar reduction in VMAX1 thus it is unlikely that this was due to any variation in walking speed (which would cause reduction or elevation in both peaks).

9.7. Conclusion

Thus in conclusion PFF was associated with a reduction in the second peak of the VGRF, a reduction in peak ankle inversion and external rotation moments, and an increase in peak ankle eversion moment impulse. These findings add support to the belief that PFF leads to instability and abnormal foot and ankle loading. There was however no evidence that a flat foot posture impairs propulsion. Kinetic parameters associated with PFF do not appear to relate to the generation of foot and ankle symptoms.

Chapter 10. PFF and the proximal joints

10.1. Introduction

The focus of the thesis thus far has been on the foot and ankle complex. As the majority of weight-bearing activities involve the closed chain coupling of the foot/ankle, knees and hips, it is conceivable that a change in foot posture can lead to a change in posture and motion of the proximal joints [137, 226, 229, 233, 296]. It is hypothesized that in the case of PFF, excessive hindfoot eversion throughout the gait cycle would lead to excessive internal rotation of the tibia, with knock on effects higher up the kinetic chain [316]. As a result it is common practice to attempt amelioration of proximal joint musculoskeletal symptoms by altering the alignment of the foot and ankle with the use of orthotics. The evidence linking foot posture with altered lower limb biomechanics and symptoms is however sparse, especially in the paediatric population. In this chapter this relationship is explored further.

10.2. Aims and hypotheses

Thus the aim of this chapter is to investigate the relationship between arch height and the proximal joints in children. As well as looking at the association of symptoms with foot posture, 3DGA is used to identify biomechanical factors that may help explain how symptoms are caused.

The three main hypotheses tested are:

1) Children with reduced arch height are more likely to have proximal joint symptoms.

2) Children with reduced arch height will demonstrate altered gait kinematics and kinetics particularly in the transverse plane at the knee and hip (based on abnormal coupling relationships postulated in Chapter 2, p.6-54).

3) Kinematic and kinetic deviations in the proximal joints demonstrated by the PFF group will relate to the presence of proximal joint symptoms.

10.3. Methods

The general data collection methods used for this chapter are outlined in Chapter 3 (p.55-92). Data for this chapter were collected on all 95 study participants.

10.3.1. Proximal joint symptoms

The distributions of proximal joint symptom scores were markedly skewed and thus to adhere to the assumptions of parametric analysis these variables were converted to binary variables; either the presence or absence of symptoms. The presence of hip or back symptoms were also pooled as participants found it was difficult to differentiate between anatomical regions. Indeed, in the literature, spinal pathology can lead to hip pain and vice versa, thus the specific location of the pain may not correctly identify the cause of pain [281].

Logistic regression was used to assess the effect of the predictors, AHI, age, BMI, gender and flexibility, measured using the LLAS, on the presence of proximal joint symptoms. The presence of statistical interaction between variables was also assessed using the likelihood ratio test. Unpaired, two-tailed Student’s t-tests were used to assess whether the presence of proximal joint symptoms was associated with impaired foot and ankle HRQOL.

10.3.2. Comparison between PFF and NF proximal joint kinematics and kinetics

Pelvis, hip and knee kinematics, and hip and knee moments and powers were computed using dedicated software pipelines in the Vicon Nexus (v1.7, Vicon, Oxford UK) software environment. The method of selection of a representative trial per subject was the same as that utilized in Chapter 8 (p.177-180). In the case of kinematics, the entire gait cycle was normalized to 101 data points. For the kinetics the stance phase of gait was normalized to 101 data points.

The process of analysing kinematic and kinetic data also took the form outlined in Chapters 8 and 9 (p.177-179). The process started with identifying regions of interest on the pooled by group gait graphs. This was followed by formal correlational analysis with AHI using Pearson's R. Where the significance of correlation was $p < 0.1$ the effect of confounding by age, gender and BMI (not with kinetics) was assessed, and where required controlled for using multiple linear regression.

10.3.3. Relationship between gait kinematics/kinetics and symptoms

From the processes outlined in section 10.3.2 a short-list of kinematic and kinetic parameters which had statistically significant correlation with AHI were identified. Then within the PFF group the effect of deviations in these parameters on the presence of lower limb symptoms were assessed using an unpaired, two-tailed Student's t-test. Kinematic and kinetic parameters related to the pelvis or hip were tested with respect to the presence of hip or back symptoms and those related to the knee with the presence of knee symptoms accordingly.

10.3.4. Power Calculations

Power analysis for logistic regression requires a priori knowledge of the relationship between the response variable (knee/hip or back symptoms) and the main predictor variable (AHI) [64]. There were no pilot data to help with the power calculation in this case. The power analysis outlined in Chapter 8 (p.179-180) applied similarly for analysis of the proximal joint kinematics and kinetics.

As the proximal joint symptoms were converted to a binary variable, the analysis outlined in section 10.3.3 was only adequately powered to identify large effect sizes (Cohen's d of > 0.8).

10.4. Results

10.4.1. Proximal joint symptoms

Thirty-seven of 95 children had knee symptoms (39%) and 33 had hip or back symptoms (35%). The distribution of symptoms by foot type is demonstrated in Tables 10.1 a) and 10.1 b). There was a significant association between AHI and knee symptoms with odds ratio (OR) of 0.76 (95% CI: 0.64-0.90, p = 0.002). A significant association was also observed between AHI and hip or back symptoms with OR of 0.78 (95% CI: 0.65-0.93, p = 0.007). Of the other predictors BMI was also found to have a significant association with hip or back symptoms with OR of 1.23 (95% CI: 1.03-1.47, p = 0.020).

a)

	FOOT POSTURE		
KNEE SYMPTOMS	PFF	NF	TOTAL
YES	25	12	37
NO	23	35	58
TOTAL	48	47	95

b)

	FOOT POSTURE		
HIP/BACK SYMPTOMS	PFF	NF	TOTAL
YES	22	11	33
NO	26	36	62
TOTAL	48	47	95

Table 10.1 Tables demonstrating distribution of proximal joint symptoms by foot posture. a) Knee symptoms and b) Hip or back symptoms.

Children with knee symptoms had significantly lower HRQOL scores in all OxAFQ-C questionnaire domains (Figure 10.1). Children with hip or back symptoms had significantly lower HRQOL scores in all OxAFQ-C questionnaire domains aside from the emotional domain (Figure 10.2).

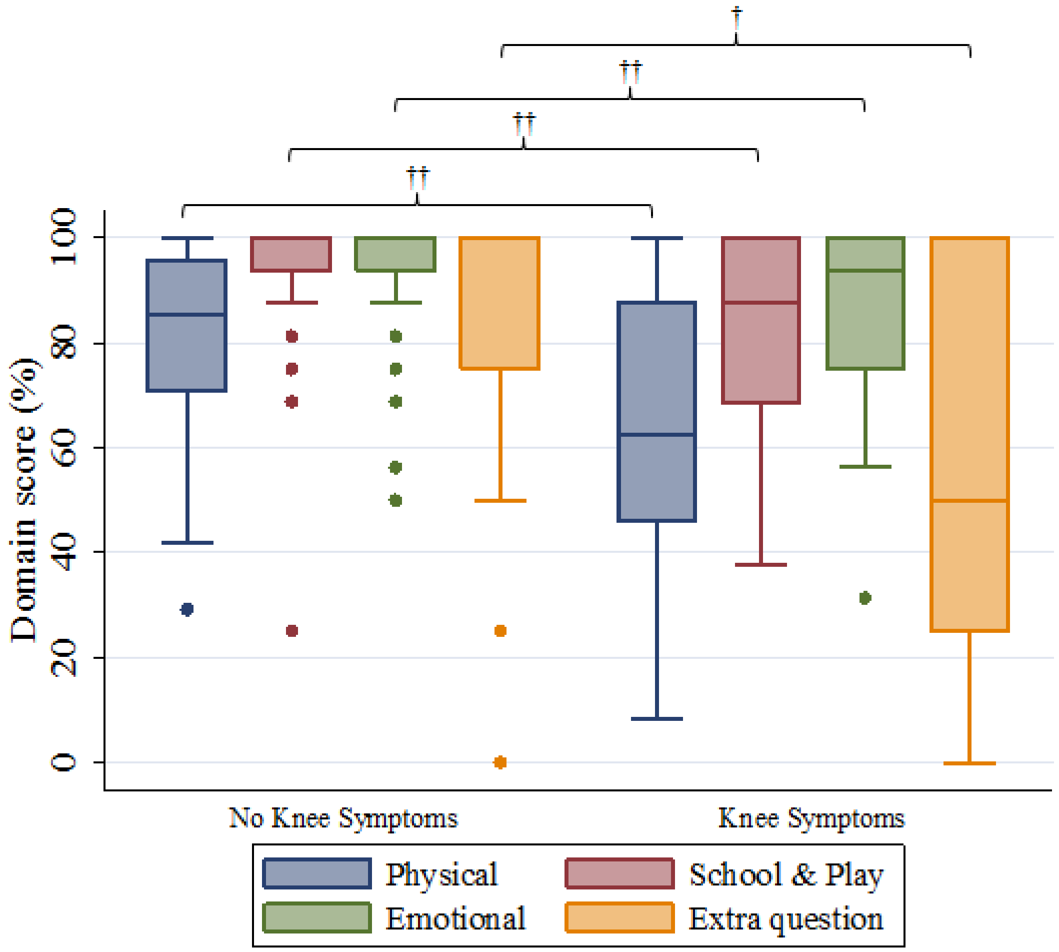


Figure 10.1 Box-whisker plot comparing OxAFQ-C questionnaire domain scores for all study participants with no knee symptoms and those with knee symptoms. † denotes $p < 0.05$ and †† denotes $p < 0.001$.

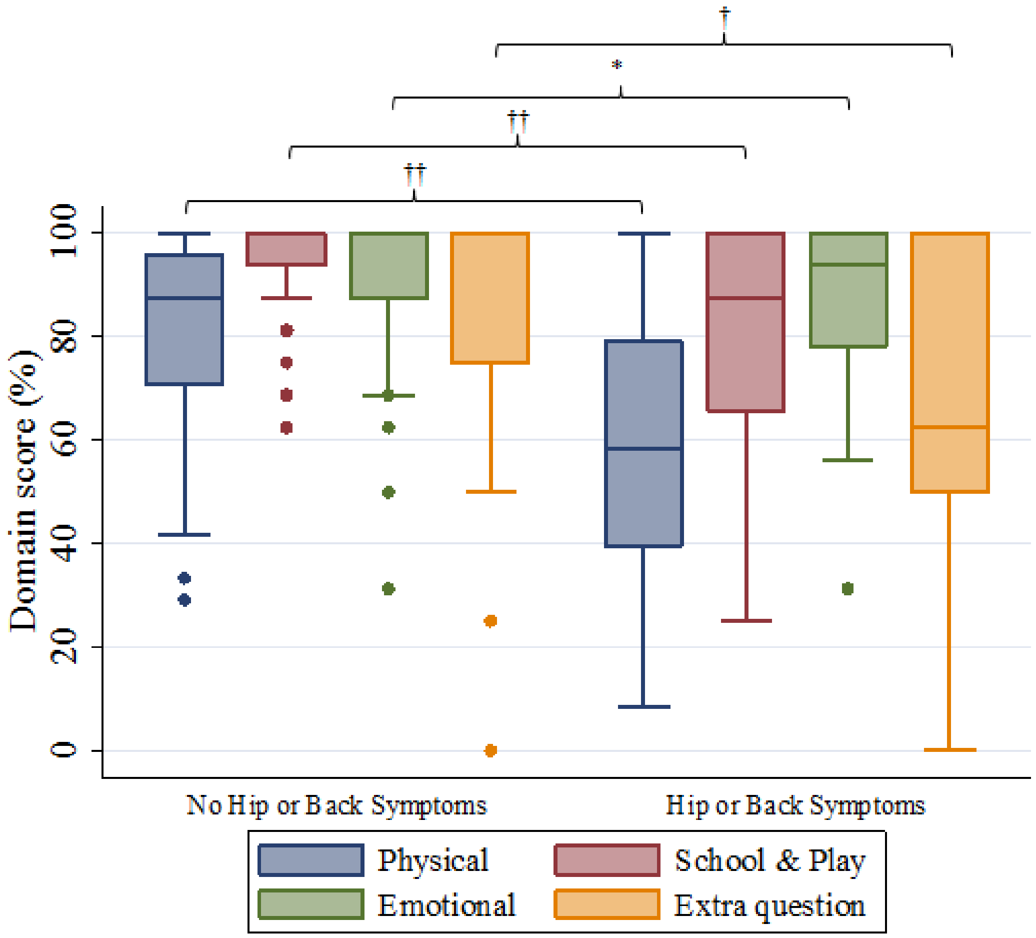


Figure 10.2 Box-whisker plot comparing OxAFQ-C questionnaire domain scores for all study participants with no hip or back symptoms and those with hip or back symptoms. * denotes P > 0.05, † denotes p < 0.01 and †† denotes p < 0.001.

10.4.2. Proximal Joint Kinematics

Visual inspection of the proximal joint kinematic graphs (Figures 10.3 to 10.5) only showed modest differences between PFF and NF groups at the pelvis and hip. At the knee there was a tendency for the PFF group to be in more valgus throughout stance. It appeared that there was a reduction in peak knee internal rotation in the PFF group when compared with the NF group. Five specific ROIs were identified and the correlation between kinematic parameters and AHI is summarized in Table 10.2.

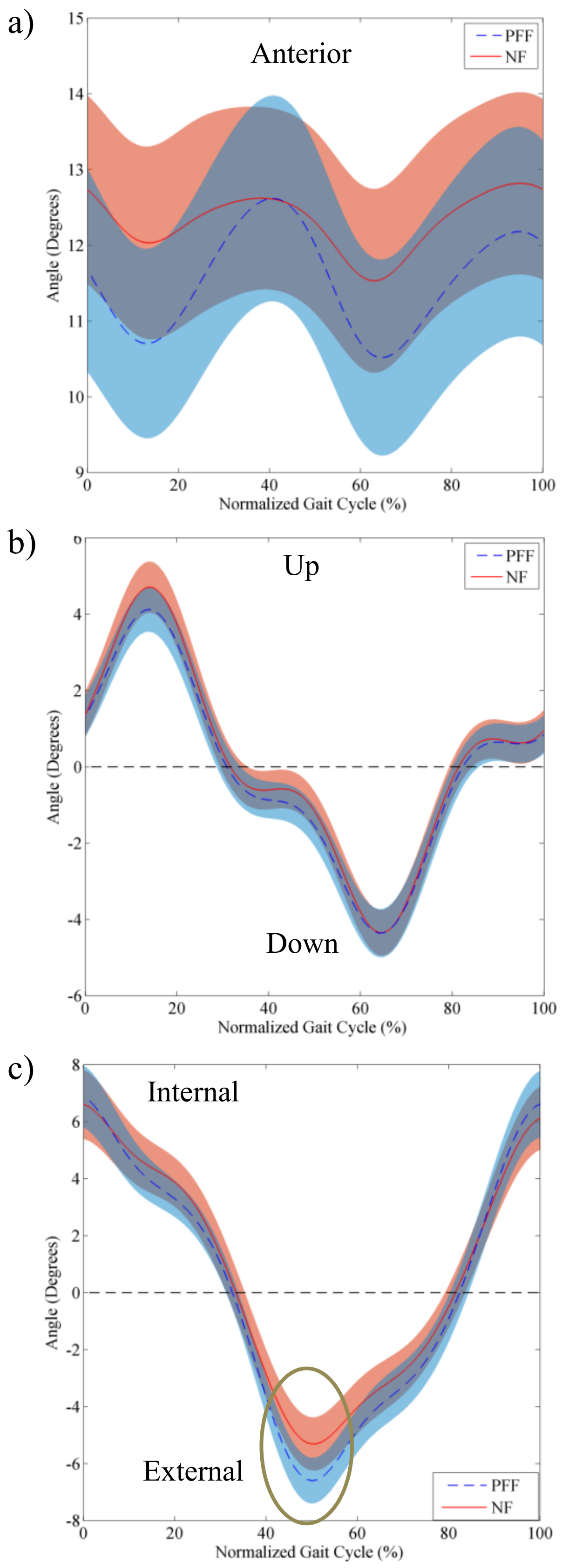


Figure 10.3 Normalized gait cycle curves for pelvis motion. Mean curve for each group along with 95% confidence bands plotted. Regions of interest marked.

a) Pelvic tilt, no ROIs.

b) Pelvic obliquity, no ROIs.

c) Pelvic rotation, with ROI or peak external rotation (PERMAX).

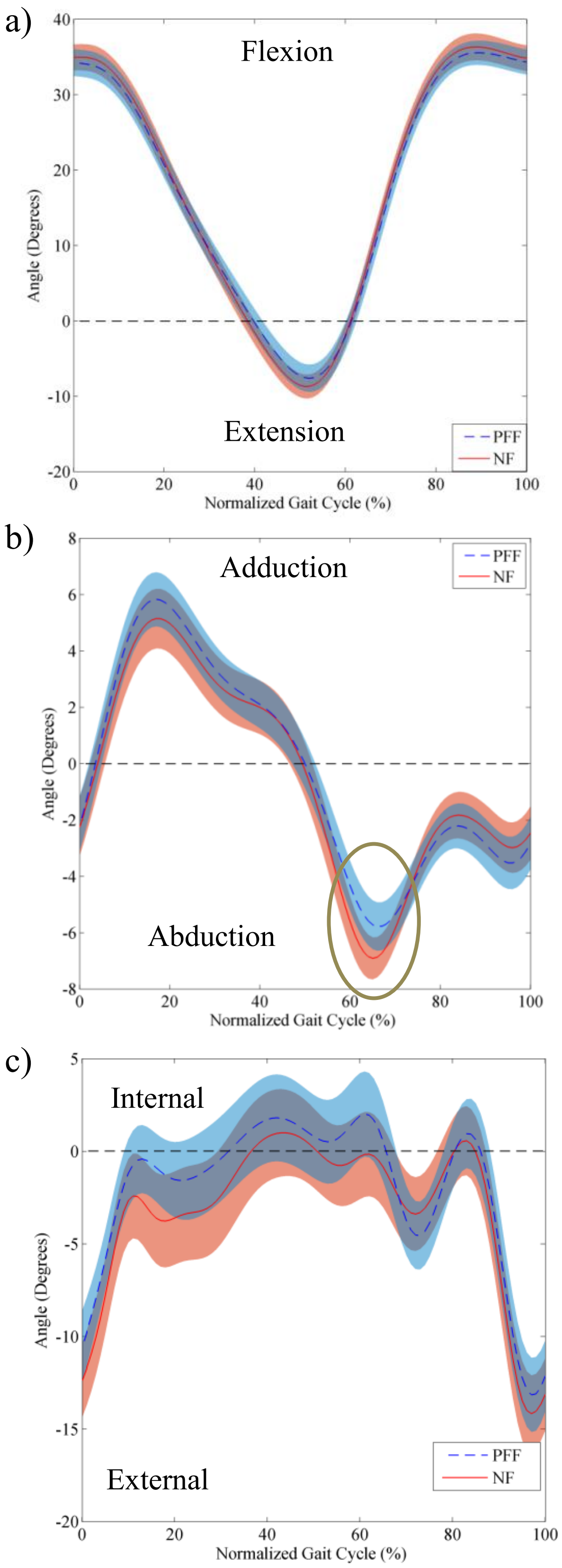


Figure 10.4 Normalized gait cycle curves for hip motion. Mean curve for each group along with 95% confidence bands plotted. Regions of interest marked.

a) Hip flexion/extension, no ROIs.

b) Hip adduction/abduction, with ROI of peak abduction (HABMAX).

c) Hip internal/external rotation, no ROIs.

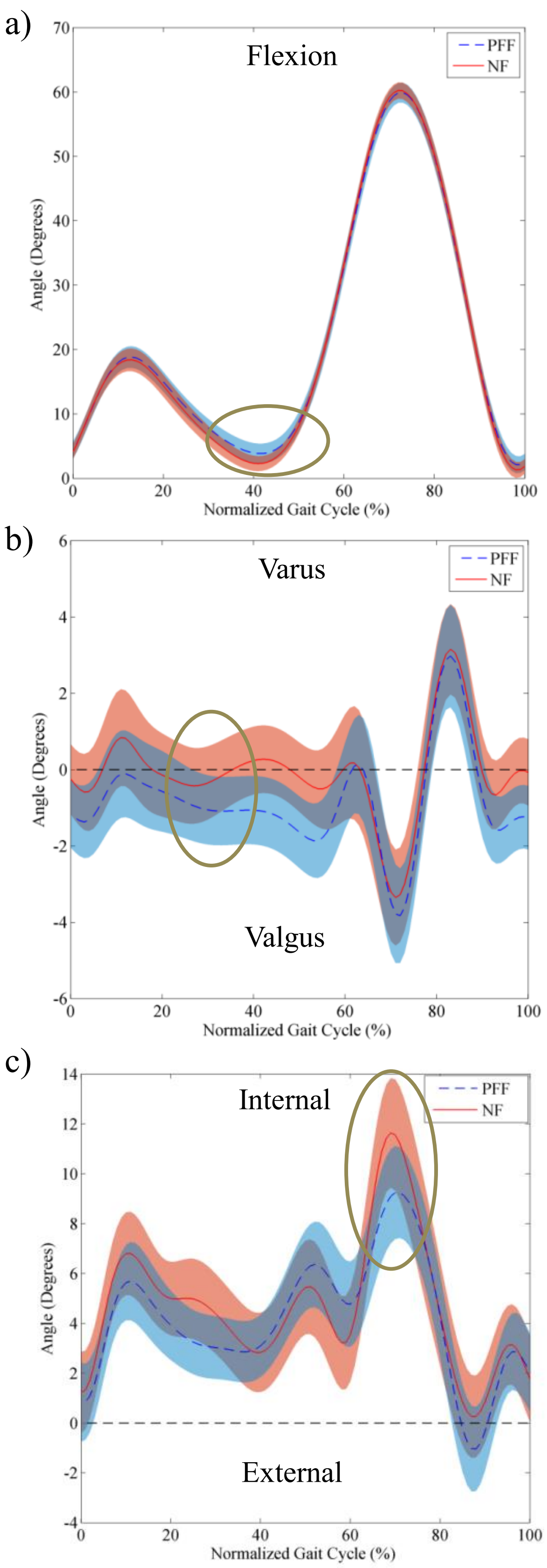


Figure 10.5 Normalized gait cycle curves for knee motion. Mean curve for each group along with 95% confidence bands plotted. Regions of interest marked.

a) Knee flexion/extension, with ROI of flexion trough (KFTROUGH).

b) Knee varus/valgus, with ROI of midstance varus (KVMID).

c) Knee internal/external rotation, with ROI of peak internal rotation (KINTMAX).

Angle	Parameter	Correlation with AHI	
		R	Sig (p)
Pelvis_z	PERMAX†	−0.20	0.05*
Hip_y	HABMAX†	0.14	0.19
Knee_x	KFTROUGH	−0.08	0.44
Knee_y	KVMID†	−0.23	0.02*
Knee_z	KINTMAX	0.15	0.14

Table 10.2 Regions of interest identified by visual inspection of proximal joint kinematic graphs. The relationship between ROIs and AHI quantified with Pearson's R and significance (Sig). * Denotes a statistically significant result. †correlations made with absolute values of parameter.

Of the five ROIs significant correlations were noted for PERMAX and KVMID. The confounding effect of gender, BMI and Age on the angles were assessed (Table 10.3), and after controlling for potential confounders AHI was a statistically significant predictor for only PERMAX (p = 0.05).

Angle	Parameter	Gender		BMI		Age	
		Diff (°)	Sig (p)	R	Sig (p)	R	Sig (p)
Pelvis_x	PERMAX	0.18	0.76	−0.12	0.23	0.82	0.02*
Knee_y	KVMID	−1.43	0.03*	−0.12	0.25	0.25	0.01*

Table 10.3 Table demonstrating the relationship between gender, BMI and age with kinematic parameters. * Denotes as statistically significant result.

Thus the only kinematic parameter that has a statistically significant relationship with AHI was peak pelvic external rotation (pelvic retraction). Such that a flatter foot posture was associated with increased peak pelvic external rotation.

10.4.3. Proximal Joint Kinetics

Visual inspection of the proximal joint kinetic graphs (Figures 10.6 to 10.8) demonstrated larger differences between PFF and NF groups than that observed with the kinematics. At the hip the PFF appeared to have a reduction in peak flexion/extension moments (HFMAX and HEMAX), second peak adduction moment

(HAMAX2) and internal rotation moments (HIMAX). At the knee the PFF group have an increased peak knee flexion moment (KFMAX) and demonstrate similar coronal and transverse moment patterns to those observed at the hip. The PFF group also demonstrate reduced peak power generation at the hip and knee (HPGEN and KPGEN), as well as reduced peak power absorption and the hip (HPABS). From the kinetic graphs ten ROIs were identified. The correlations between the kinetic parameters and AHI are summarized in Table 10.4.

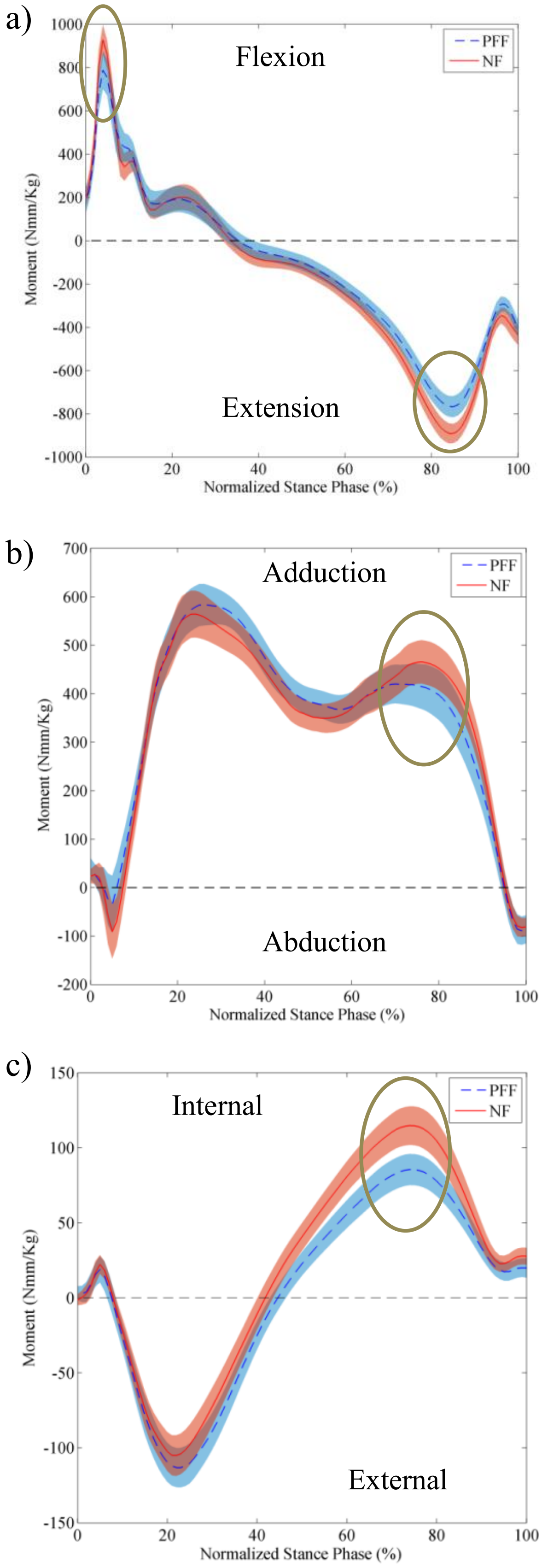


Figure 10.6 Normalized stance phase curves for hip moments. Mean curve for each group along with 95% confidence bands plotted. Regions of interest marked.

a) Hip moment x, with ROI of peak flexion moment (HFMAX) and peak extension moment (HEMAX).

b) Hip moment y, with ROI of second peak adduction moment (HAMAX2).

c) Hip moment z, with ROI of peak internal rotation moment (HIMAX).

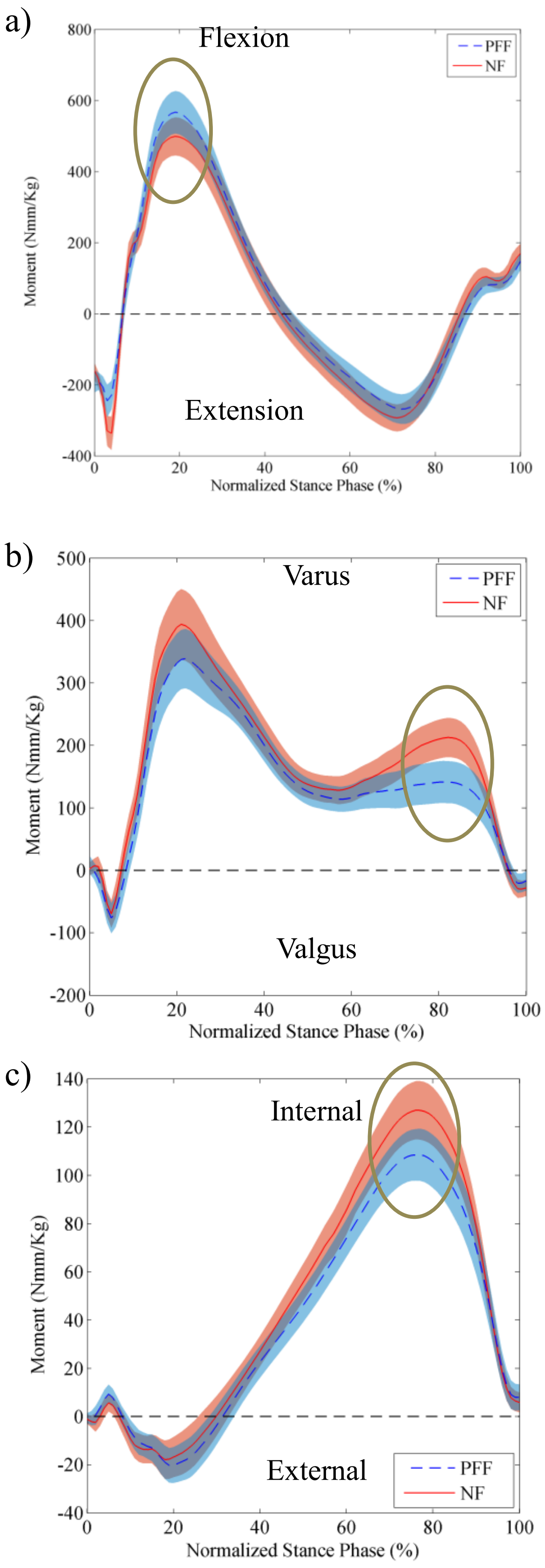


Figure 10.7 Normalized stance phase curves for knee moments. Mean curve for each group along with 95% confidence bands plotted. Regions of interest marked.

a) Knee moment x, with ROI of peak flexion moment (KFMAX).

b) Knee moment y, with ROI of second peak varus moment (KVMAX2).

c) Knee moment z, with ROI of peak internal rotation moment (KIMAX).

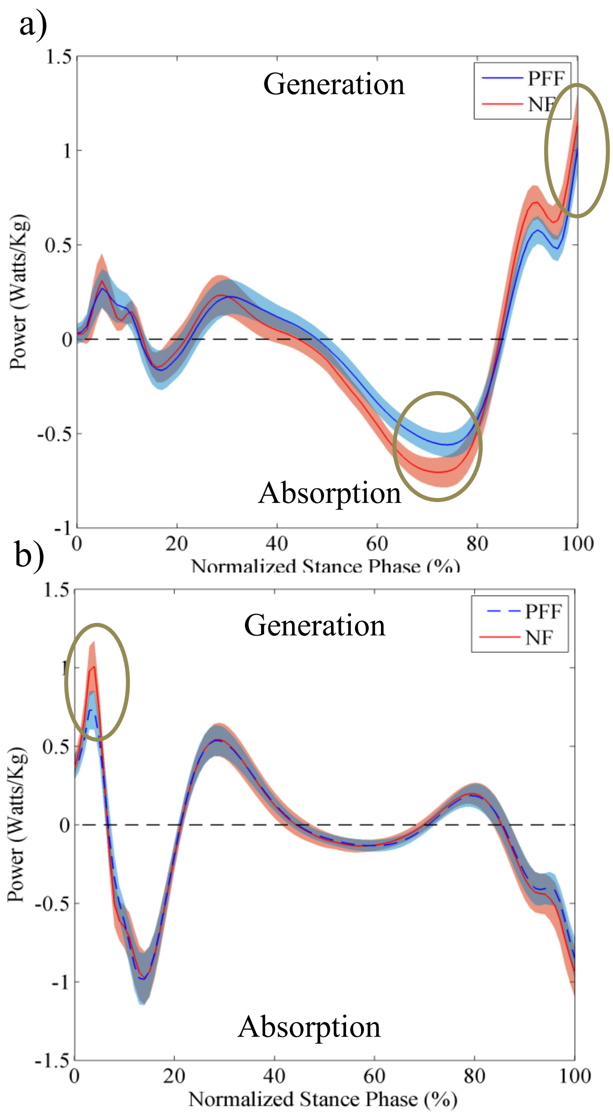


Figure 10.8 Hip power graph (a) and knee power graph (b) for normalized stance phase of gait. Mean curve for each group along with 95% confidence bands plotted. a) Regions of interest peak hip power generation (HPGEN) and peak hip power absorption (HPABS).

b) Region of interest peak knee power generation (KPGEN).

Moment/Power	Parameter	Correlation (R)	Sig (p-value)
Hip Moment x	HFMAX	0.10	0.35
	HEMAX†	0.35	<0.01*
Hip Moment y	HAMAX2	0.08	0.47
Hip Moment z	HIMAX	0.18	0.08‡
Knee Moment x	KFMAX	−0.17	0.1
Knee Moment y	KVMAX2	0.29	<0.01*
Knee Moment z	KIMAX	0.26	0.01*
Hip Power	HPGEN	0.06	0.58
	HPABS†	0.26	0.01*
Knee Power	KPGEN	0.14	0.16

Table 10.4 Regions of interest identified by visual inspection of proximal joint kinematic graphs. The relationship between ROIs and AHI quantified with Pearson's R and significance (Sig). * Denotes a statistically significant result. †correlations made with absolute values of parameter. ‡ denotes significance $0.05 < p < 0.1$.

Of the ten ROIs, four had statistically significant correlations with AHI, with HIMAX tending to significance ($p = 0.08$) (Table 10.4). The confounding effect of gender, and age on these kinetic parameters are summarized in Table 10.5. Age correlated significantly with HIMAX, KVMAX2 and KIMAX. When age was controlled for using regression AHI still was not a significant predictor of HIMAX ($p = 0.06$).

Moment/Power	Parameter	Gender		Age	
		Diff	Sig (p)	R	Sig (p)
Hip Moment x	HEMAX†	−12.14	0.75	0.16	0.12
Hip Moment z	HIMAX	0.06	0.99	0.45	<0.01*
Knee Moment y	KVMAX2	−25.97	0.19	0.42	<0.01*
Knee Moment z	KIMAX	−7.44	0.38	0.54	<0.01*
Hip Power	HPABS†	0.02	0.77	0.16	0.12

Table 10.5 Table demonstrating the relationship between gender, and age with kinetic parameters. * Denotes as statistically significant result. † absolute values of parameter used for analysis.

Thus after statistical analysis a flatter foot posture was associated with the following kinetic findings:

1) Reduced peak hip extension moment.

2) Reduced second peak knee adduction moment.

3) Reduced peak knee internal rotation moment.

4) Reduced peak hip power absorption.

10.4.4. Relationship between kinematic/kinetic parameters and proximal joint symptoms.

From the statistical analysis in sections 10.4.2 and 10.4.3, one kinematic and four kinetic parameters were found to have a statistically significant relationship with AHI. In subsequent analysis of just the PFF group no statistically significant difference was noted for these parameters in relation to the presence of absence of proximal joint symptoms. The results are summarized in Table 10.6.

Parameter	Difference in means	Significance (p)
PERMAX*	0.72 (°)	0.34
HEMAX*	18.66 (Nmm/Kg)	0.70
KVMAX2†	−33.54 (Nmm/Kg)	0.20
KIMAX†	−15.45 (Nmm/Kg)	0.15
HPABS*	−0.02 (Watts/Kg)	0.78

Table 10.6 Summary of differences between gait parameters in PFF group with respect to proximal joint symptoms. * Denotes where comparison was made with presence or absence of hip or back symptoms. † denotes where comparison was made with presence of absence of knee symptoms.

10.5. Discussion

The purpose of this chapter was to investigate the relationship between foot posture and the proximal joints. The first stage was to explore the association between a flat foot posture and proximal joint symptoms. Subsequently the aim was to establish a potential causative pathway from a flattened arch, to abnormal biomechanics, with these being directly associated with proximal joint symptoms. Whilst it was possible to demonstrate the association between a reduced AHI and abnormal lower limb biomechanics (particularly hip and knee kinetics), the parameters identified did not seem to relate to the presence or absence of proximal joint symptoms.

10.5.1. Foot posture versus proximal joint symptoms

Logistic regression demonstrated that AHI was a significant predictor of both knee symptoms and hip/back symptoms in the study participants. Based on the odds ratios presented in 10.4.1, for every 0.01 reduction in AHI, the odds of having knee symptoms were increased by just under a third. The same phenomenon was observed with hip or back symptoms. Odds ratios of this magnitude can be approximated to a Cohen’s d of 0.15, which is consistent with a small effect size. Unsurprisingly proximal joint symptoms were not mutually exclusive of foot and ankle symptoms, with there being a strong association between the presence of proximal joint symptoms and lower OxAFQ-C scores.

The link between foot posture and proximal joint symptoms presented in this chapter is consistent with a common perspective in clinical practice, and something observed in a number of adult populations [12, 114, 167, 178, 209].

In this chapter it was also noted that increased BMI was associated with an increased risk of hip or back pain. The association between BMI and musculoskeletal symptoms is well established in the literature; indeed Stovitz et al. [304] demonstrated that the back was the most common site of pain in their cohort of overweight children. This gives another potential avenue for therapeutic intervention in an overweight child with PFF.

After demonstrating the clinical association between a flat foot posture and proximal joint symptoms, the next stage was to confirm the pathomechanic theories about how a flat foot posture may lead to proximal joint symptoms.

10.5.2. Relationship between foot posture and proximal joint kinematics

The coupling relationships between hindfoot motion and tibial rotation are well established [137, 229]. As a flat foot posture is associated with increased hindfoot eversion in the static and dynamic settings, it was hypothesized that increased dynamic internal tibial rotation would also be observed. This however was not the case, with little difference in knee rotation between the NF and PFF groups for the majority of stance. In fact, it initially appeared that the PFF group had reduced peak internal knee rotation, although this relationship was not statistically significant. This contrasts with the findings of Levinger et al. [178] who did demonstrate increased tibial internal rotation with an everted hindfoot in a population of adults with medial knee osteoarthritis. However, this is a significantly different population to that investigated in this thesis. It is also unclear whether the adults in Levinger et al.’s [178] study had flexible flat feet or adult acquired rigid flat foot secondary to posterior tibial tendon dysfunction. In comparison with the literature from similar populations neither, Twomey et al. [320] nor Shih et al. [286] demonstrated increased internal knee rotation in their PFF populations. Twomey et al. [320] however did observe increased external hip rotation in their PFF group, which contradicts the observations of Souza et al. [296] who found increased hindfoot eversion was associated with increased internal hip rotation. In this chapter no relationship was noted between hip rotation and foot posture.

At the level of the pelvis a flatter foot posture was associated with increased external pelvis rotation. This peak was observed in late stance. The significance of this finding is unclear, but it may be related to proximal adaptation to assist propulsion as discussed in the previous chapter.

In the coronal plane it was hypothesized that a flat foot posture would be associated with increase knee valgus in stance. Whilst the overall trend was observed on visual inspection of the graphs, at midstance there was no correlation between AHI and knee varus/valgus. Statistical analysis of the whole curve in a continuous fashion may have identified differences between groups, but either way group differences would have been small and of questionable clinical significance.

Foot posture did not correlate with any of the other proximal joint kinematic ROIs. This echoes the general findings in the paediatric population that foot posture does not hugely affect kinematics higher up the kinetic chain than the ankle [286, 320].

10.5.3. Relationship between foot posture and proximal joint kinetics

It was again hypothesized that the majority of differences in joint moments would be in the transverse and coronal planes. However unlike the kinematics, the proximal joint kinetics demonstrated much larger group deviations. The reasons underlying this (cf. Chapter 3, p.64-66) are due to either the GRF or differences in angular momentum of the limb segment. Reassessing the kinetic graphs it is observed that all three of the ROIs that demonstrated a significant relationship with AHI are all in late stance at around 80%. This coincides with the reduced second peak of the VGRF observed in the previous chapter. This would explain why a reduction in AHI is associated with a reduction in peak joint moment for these ROIs. This also explains why there is a reduction in hip power absorption at this point. Without assessing the correlation between the kinetic deviations and the presence of symptoms (to be discussed in 10.5.4) one would think that it is unlikely that these gait adaptations are the cause of the proximal joint symptoms, and are merely the response to the reduced VGRF second peak. On the other hand the suggestion of an increased knee peak flexion moment in Figure 10.7 a) may have been of greater significance, but the study was not powered adequately to identify smaller effect sizes.

10.5.4. Relationship between kinematic/kinetic parameters and proximal joint symptoms

None of the kinematic or kinetic parameters identified as having a significant relationship with AHI, correlated significantly with the presence of proximal joint symptoms in the PFF group. Due to the nature of dichotomous symptom data the minimum effect size that this portion of the chapter was adequately powered to identify was very large. This meant it was difficult to detect differences between groups, although prior to this stage it was clear that the expected/hypothesized kinetic and kinematic adaptations to a flat foot posture were not present. The adaptations that were present were not expected to relate to the presence of symptoms and any significant results would have been hard to explain from a mechanistic perspective.

10.6. Limitations and other considerations

These findings raise the obvious question, ‘why do children with PFF develop proximal joint symptoms?’. Firstly the method by which we can measure joint kinematics and kinetics may not be sensitive enough to pick up small but clinically significant differences. The errors associated with a skin mounted marker based system were discussed in the Chapter 3 (p.63), and issues regarding signal to noise ratio were discussed in the previous chapter. As observed at the foot and ankle, coronal and transverse plane kinematics and kinetics are much smaller in range and magnitude compared to the sagittal plane, leading to relatively broad confidence intervals, and marked within group variability. Indeed the variability of joint moments particularly in the transverse plane has been the subject of debate, with the resulting moments being highly sensitive to change in reference planes [278]. These factors act against this large sample size (in the context of gait analysis studies) reducing statistical power. Standard gait models also do not model the patellofemoral joint. Patellar kinematics and kinetics may play an important role in the development of knee symptoms, but have not been assessed. Finally formal assessment of trunk kinematics and kinetics may cast further light on the presence of back symptoms.

It is also conceivable that the pathomechanic theories about why PFF would lead to proximal joint symptoms are incorrect, and that there may be other mechanisms that can account for the symptoms. There may also be other confounding factors that relate to both a flat foot posture and proximal joint symptoms. The obvious candidates in this case would be increased joint mobility and increased BMI, although these variables could not always significantly predict the presence of proximal joint symptoms. Or simply it may the case that PFF are relatively common in the general paediatric population as are proximal joint symptoms, and the association between the two observed in this study is merely a chance occurrence and represents a type 1 error.

It is accepted that the method of measuring proximal joint symptoms had potential flaws, in that a simple question was used as opposed to a formally validated questionnaire. The main reservation about giving further validated questionnaires as well as the OxAFQ-C and PedsQL 4.0 was that the child would reach questionnaire saturation and the data obtained from the questionnaires would not be representative of the true clinical picture. The risk of using too many questionnaires in parallel has been highlighted by Fitzpatrick et al. [99]. Also, currently there are no appropriate validated site-specific knee and hip HRQOL measures for the paediatric population.

10.7. Conclusion

Children with a flatter foot posture are more likely to have pain or discomfort at the hip, knee and back. This in turn is associated with increased foot and ankle symptom severity. Using 3DGA a number of kinematic and kinetic variables were noted to correlate with arch height, but the findings were not consistent with prevailing pathomechanic theories. The majority of positive kinetic findings could be explained by the reduction in the second peak of vertical ground reaction force observed in Chapter 9 (p.208). Without explicit understanding of how PFF relates to proximal joint symptoms, planning and delivering treatment is difficult, and further work is required in this area.

Chapter 11. Summary and conclusions. Who to treat?

11.1. What this thesis adds to knowledge on paediatric flexible flat foot

The key new findings of this thesis are as follows:-

1) Parents of children with PFF may over-estimate the degree of impairment their child has. However Children with PFF have significantly impaired HRQOL compared to healthy controls.

2) A flat foot posture in associated with an absent anterior subtalar articulation.

3) Tight plantarflexors and difficulty performing a SHR are sensitive tests for identifying children with more symptomatic PFF.

4) The more symptomatic a child with PFF the slower their walking speed.

5) Symptoms in PFF are strongly associated with the amount of dynamic forefoot supination.

6) A flat foot posture is associated with a reduced second peak in the vertical ground reaction force, which has concomitant effects on lower limb and foot and ankle moments. This may be due to pain or instability.

7) There is no evidence that PFF cause impaired propulsion.

8) Whilst children with PFF are more likely to have symptoms at their knees, hips and back, conventional gait analysis does not demonstrate any mechanism by which this may occur.

11.2. Summary

Flexible flat feet are a common presentation in children, but considerable debate continues about the aetiology, diagnosis and management of this condition. Ultimately treatment for this condition can be only be rationalized if one or both of the following assumptions are correct: Firstly that PFF will lead to progressive joint arthropathy and problems into adulthood, and secondly that during childhood PFF can cause significant symptoms and impaired quality of life. Whilst Bertani et al. [20] among other authors have suggested that the first assumption is true, currently there is no longitudinal evidence to support their proposition [211]. The purpose of this thesis was to investigate the second assumption, but to also identify whether there are structural or functional characteristics of the lower limbs of children with PFF which could cause, or pre-dispose to, symptoms. A prospective cross-sectional observational study was used to investigate the entity of PFF using a multimodal approach including clinical examination, questionnaire assessment, MRI imaging and 3DGA. It was hoped that by doing this a better understanding of PFF could be achieved and that this could help better define treatment protocols.

After reviewing all relevant literature on the subject of PFF, and describing the general methods utilized for this thesis, a technique to classify foot posture was sought using objective, robust methods. This was deemed necessary, firstly because there was concern about the subjectivity of using visual assessment to define foot posture, and secondly because there is no consensus on the ideal classification method [67, 261]. Using a data driven approach a classification method was devised using multiple indices as recommended by Chuckwaipong et al. [58] and Murley et al. [224]. This proved helpful in dichotomising the study participants' foot postures as either neutral of flat. Interestingly it demonstrated strong, significant correlations between the majority of classification methods. As well as having a binary classification it was also deemed desirable to have a continuous measure of foot posture. The AHI was used as the continuous measure [343].

Having defined a flat foot, the next stage of investigation involved assessing the impact of foot posture on HRQOL in the study population (consisting of 48 PFF and 47 NF). Whilst the use of HRQOL measures is increasing in popularity in clinical and research practice, this approach had not previously been used in the context of PFF. Using the OxAFQ-C [219] and the PedsQL™ 4.0 [327] it was demonstrated that children with PFF had worse HRQOL compared to children with NF. This was particularly marked in the physical domains of the questionnaires, and was potentially as severe as a number of other pathologies [84, 327]. Moreover the correlational analysis demonstrated that the flatter the foot the worse the HRQOL. This supports the currently limited evidence that the severity of the deformity relates to the presence of symptoms [213, 354]. The findings of this chapter serve as an objective, evidence based foundation to refute the personal views of a number of authors that PFF is a benign normal variant [100, 297]. The findings also confirm the overall rationale to treat PFF in certain cases, as well as adding further justification for identifying structural or functional characteristics that may contribute to impaired HRQOL.

In Chapter 6 the aetiology of PFF was investigated to gain further insight into how this condition may cause functional impairment, and to guide potential treatment strategies. Flexibility and knee coronal plane alignment were shown to be significant predictors of AHI, consistent with previous published literature [86, 180]. The results also demonstrated the hitherto only speculated relationship between an absent anterior subtalar articulation and a flat foot posture [10, 36]. This finding has greater implications if surgical intervention is to be undertaken, and it is conceivable that the outcome from joint preserving surgery, in terms of operative correction, would be dependent of the shape of the subtalar joint. This is relevant, not only in the PFF population, but also in neuromuscular and adult acquired flat foot populations.

In Chapter 6 the utility of two simple clinical tests were also assessed as tools to identify children with more symptomatic PFF. Both the inability to attain plantigrade with the knee extended and failure of the single heel rise test demonstrated high sensitivity for identifying more symptomatic children, thus representing useful adjuncts to standard clinical assessment.

The remainder of the chapters investigated the function of PFF from a more dynamic perspective, with Chapters 7 to 9 focusing on the foot and ankle, and Chapter 10 addressing the proximal joints. The overarching hypothesis of these chapters was that PFF would demonstrate altered biomechanics compared to NF, and that this would be representative of functional impairment which may be responsible for impaired HRQOL. The hypothesized mechanistic pathway for symptom generation at the foot and ankle involves abnormal loading of capsuloligamentous structures around the plantar-medial aspect of the foot caused by impaired re-supination, disruption of the midtarsal locking mechanism, and push-off on a supple foot. It was hypothesized that abnormal foot and ankle joint motions would be coupled with abnormal joint motion of the pelvis, hips and knees which in turn could account for the development of proximal joint symptoms.

Tracking the motion of the navicular in Chapter 7 gave further insight into midfoot function in children with PFF. Children with PFF had reduced medial translation of the navicular, and this may be related to the finding in Chapter 8 that children with PFF had reduced forefoot adduction at foot off. This finding is also consistent with the findings of Moraleda et al. [213] who demonstrated that children with PFF had greater uncovering of the talar head due to lateral subluxation of the navicular. The uncoupling of sagittal and transverse plane motion of the navicular was observed in the PFF children which was also suggestive of impaired midfoot function. The dynamic function of the foot and ankle complex as a whole in the context of HRQOL was subsequently assessed in Chapters 8 and 9.

A number of differences were demonstrated in foot and ankle kinematics between children with PFF and those with NF. A flat foot posture was associated with dynamic deformities at the hindfoot and forefoot. Of these deformities, only increased forefoot supination in the PFF had a significant correlation with impaired HRQOL. Spatio-temporal parameters demonstrated slower normalized walking speed in children with symptomatic PFF which may be a further reflection of biomechanical dysfunction.

The loading of the foot and ankle was assessed primarily focusing on the presence of instability and impaired propulsion as well any abnormal joint moments that related to impaired health related quality of life. One of the main findings was a reduced second VGRF peak, suggestive of ‘decelerator deficiency’ of the loaded limb [344]. This may have been a protective adaptation to reduce foot and ankle joint moments. Increased ankle evertor moment impulse, peak ankle evertor moment and peak external rotation moment was further evidence of impaired re-supination in the PFF population.

In Chapter 10 the important question of whether PFF is related to proximal joint symptoms was addressed. This chapter provided conflicting results, in that children with PFF had increased likelihood of having proximal joint symptoms. However whilst 3DGA demonstrated some differences in kinematics and kinetics between children with PFF and those with NF these were not in keeping with the hypothesized mechanisms by which it was thought the symptoms occurred. The potential reasons behind the slightly conflicting finding were addressed in the chapter discussion.

Thus is it evident that there are a number of structural and functional differences between a flat and neutral foot posture. It is possible to directly relate some of these findings to the presence of lower limb symptoms, but not in all cases. The findings which did not correlate with symptoms are not necessarily of no clinical significance as over time they may cause problems. Unfortunately without following up the study cohort longitudinally it is impossible to know to what extent these differences could impinge on the subject. Armed with these data it is now possible to refine treatment protocols for children with PFF.

11.3. Who to treat?

One of the aims of this thesis is to better define treatment indications for children with flexible flatfeet. To achieve this, a cohort of children has been thoroughly investigated to identify key structural and functional characteristics that may relate to impaired function or symptoms. In a broader sense the work presented in this thesis seeks to understand PFF in as much detail as possible and to cast further light on this common, but poorly understood presentation.

Before deciding who should be treated, the question of whether treatment is at all indicated should be addressed, i.e. is PFF a pathological presentation or normal variant? The work presented in Chapter 5 confirms that PFF is associated with impairment in HRQOL at such a level to warrant intervention. Using the OxAFQ-C as a tool for deciding who should receive treatment then it seems reasonable that any child with PFF and an OxAFQ-C physical domain score of less than around 75% would be a candidate for treatment (based on a domain MID of 10% [218]). If the OxAFQ-C is not being used, then inability to attain plantigrade and/or failure of the SHR test would highlight children who could potentially benefit from treatment. It is also evident that the worse the flat foot deformity as measured by the AHI, the worse the HRQOL. Sensitivity analysis demonstrated that an AHI of less than or equal to 0.27 had approximately 80% sensitivity to identify more symptomatic children. This was similar to the sensitivity demonstrated by the SHR test and ability to attain plantigrade.

A key dynamic deformity (forefoot supination) was associated with more symptomatic PFF, and the correlation of this to impaired HRQOL adds further rationale to treat. It is accepted that instrumented gait analysis may not be routinely undertaken to identify this abnormality, but nonetheless significant deformity may be identified by systematic visual gait analysis. The question then arises, ‘What should one do if the dynamic deformity is present, but the child remains asymptomatic?’. This highlights the main short-fall of this thesis which is that the study was cross-sectional in nature. As such it is not possible to assess the natural history of PFF, in general terms, or also in the context of asymptomatic individuals who present with factors with known association with impaired HRQOL. Nonetheless it seems reasonable to monitor these children for the development of symptoms in an outpatient setting and treat accordingly if they become symptomatic.

There is a clear association between foot posture and proximal joint symptoms; however the precise mechanisms by which these symptoms occur is unclear. As such planning and delivering effective treatment is difficult. That being said, in principle it seems sensible to trial simple non-operative measures for PFF to ameliorate proximal joint symptoms. However it would be best to monitor the outcomes from treatment carefully and it would be best to do this in a research setting.

Figure 11.1 summarises a proposed algorithm for the management of PFF in children aged 8 to 15 years old based on the research findings of this thesis. A traffic light scheme has been adopted highlighting in red, those who should undergo treatment, in orange those who one should monitor or consider treatment for proximal joint symptoms and in green, those individuals who can be discharged from healthcare services. An important tenet of this algorithm is the use of the OxAFQ-C to monitor select asymptomatic individuals and the response to treatment in symptomatic individuals. In cases where treatment has been instigated for proximal joint symptoms it is recommended that an alternative HRQOL measure to the OxAFQ-C is used to chart progress appropriate to the joint at which symptoms are being experienced.

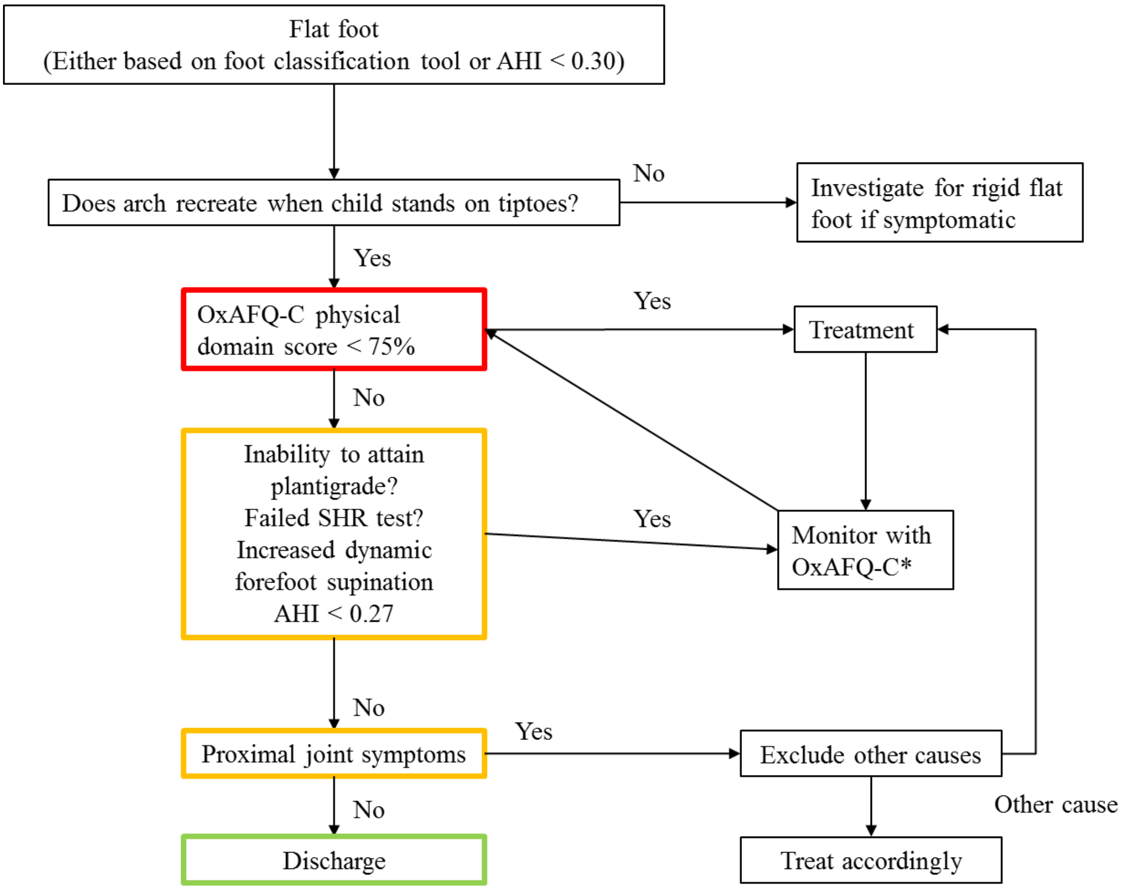


Figure 11.1 Figure demonstrating proposed management pathway for PFF in children 8-15 years old.* Alternative HRQOL measure to be used to monitor proximal joint symptoms.

11.4. How to treat?

In Figure 11.1 the treatment box does not define which treatment should be utilized. There is sparse clinical evidence supporting current non-operative and operative treatment for PFF. Thus even though the indications for treatment have been better defined, whether the treatment itself can improve symptoms remains to be seen. In a recent Cochrane systematic review [94] it was suggested that the evidence was too limited to draw definitive conclusions about the benefit of non-surgical interventions for the treatment of PFF. No such review, to date, has been undertaken for the benefit of surgical intervention for PFF.

Whilst it may not be clear which interventions could best improve symptoms related to PFF, the findings of this thesis do provide a number of potential therapeutic targets. It has once again been demonstrated that increased flexibility is a key determinant of a flat foot posture. From a treatment perspective, muscle strengthening and proprioceptive exercises to improve the dynamic stability of the foot and ankle joint complex seem worthwhile, but have seldom been undertaken [94]. Plantarflexor contracture, whilst not particularly prevalent in the study population, was also associated with more symptomatic PFF. This finding is consistent with a number of other studies, and indeed stretching of the plantarflexors is also advocated by a number of authors, but its efficacy is yet to be properly examined [120, 129, 221]. Finally the kinematic findings suggest that the key deformities of PFF which should be addressed are hindfoot valgus and forefoot supination, the exact methods by which this could be done in terms of non-operative or operative intervention is beyond the scope of this thesis.

11.5. Recommendations for future work

An in depth study has been undertaken into PFF, but as is often the case with research more questions have been raised than answered. Some directions for future work have been identified in individual chapters; however in general terms there are two main avenues for further research. Firstly a prospective long-term cohort study of children with PFF would be very useful to understand the natural history of PFF. Does PFF actually lead to degenerative arthropathy? If one has PFF is one more likely to develop tibialis posterior tendon dysfunction leading to a painful rigid flat foot? The answers to these questions may add further rationale for the timely treatment of PFF to avoid morbidity. Secondly, and probably of more pressing importance is to evaluate current treatment protocols. Undoubtedly millions of pounds in the UK are spent on the treatment of PFF per year. However we do not know whether this represents value for money, as we do not truly know the efficacy of treatments. The methods utilized in this study act as a framework around which future studies can be based. It is hoped that the clarification of foot posture classification, and the use of clear outcome measures in terms of HRQOL measures and gait parameters, will help provide the evidence which is so highly sought after in this field. The high prevalence of PFF also reduces the complexity of undertaking a prospective randomized controlled study (RCT) evaluating the effect of different treatments. This would be an endeavour which could be achieved in a timely fashion. Currently a prospective audit of the outcomes of orthotic use in PFF is being undertaken in our unit to provide pilot data from which a formal RCT can be designed.

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Appendix 1. Ethical and trust approval

Oxford University Hospitals NHS
NHS Trust

From the R & D Lead
OUH Research & Development
Joint Research Office
Block 60, Churchill Hospital
Old Road, Headington
Oxford OX3 7LE

Tel: (01865) 572970
Fax: (01865) 572242
Email: oliver.dove@ouh.nhs.uk

HH/VR/OD/10166

Alpesh Kothari
Nuffield Orthopaedic Centre
Windmill Road
Headington
Oxford
OX3 7LD

Monday 9th July 2012

Dear Dr Kothari,

Re: Defining surgical indications for children with flexible flatfeet
Research and Development Reference: 10166
Research Ethics Committee Reference: 12/SC/0334

Confirmation of Trust Management Approval

On behalf of the Oxford University Hospitals NHS Trust, I am pleased to confirm Trust Management Approval and Indemnity for the above research on the basis described in the application, protocol and other supporting documents.

Conditions of Approval

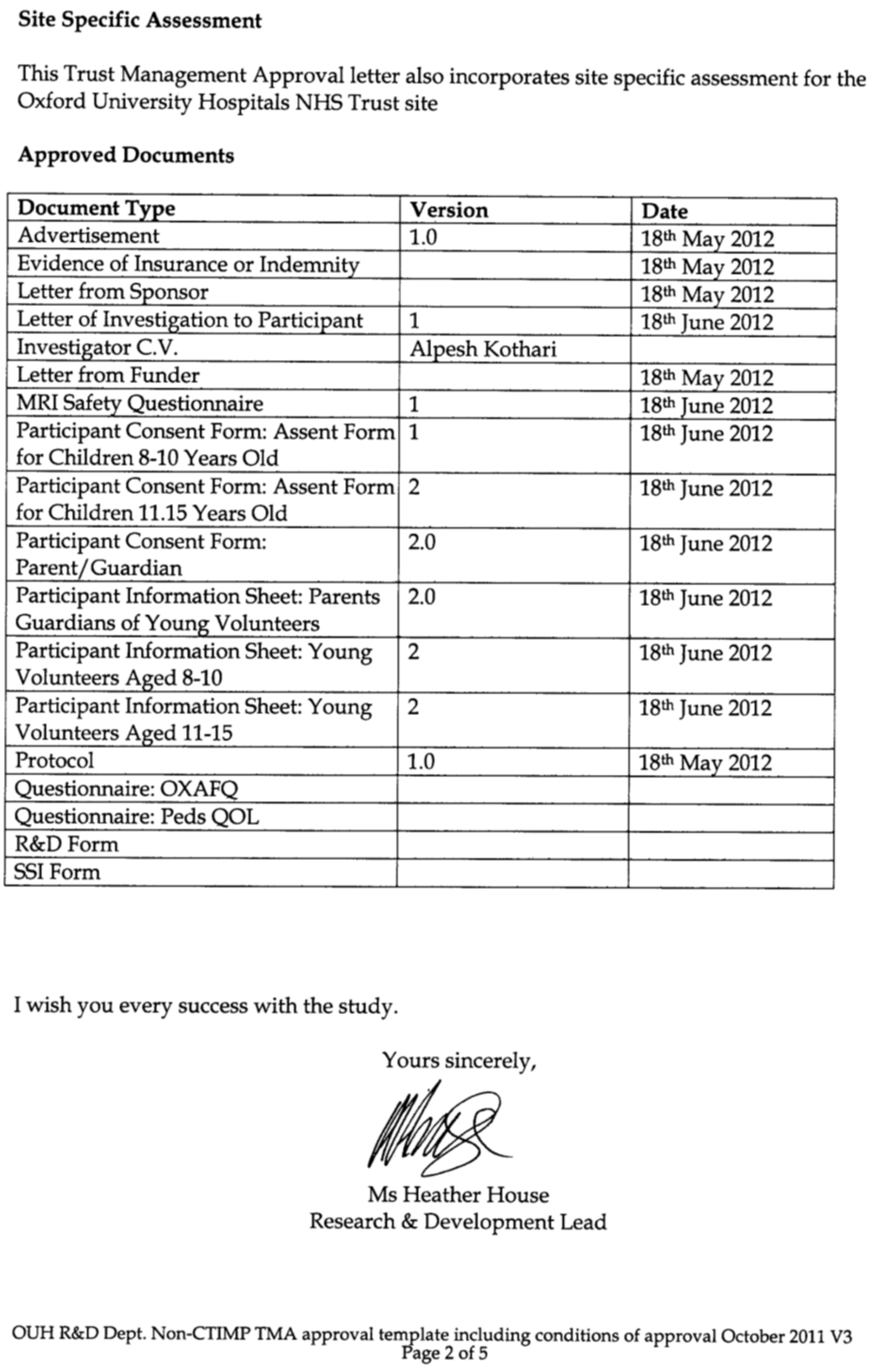
Your attention is drawn to the attached conditions of approval. Breach of these conditions may result in Trust Management Approval being revoked.

Ethics Correspondence

In order to facilitate good communications and avoid unnecessary delays please copy all correspondence with the Research Ethics Committee (REC) to R&D, providing copies of all relevant documents.

Research Sponsorship

The Oxford University Hospitals NHS accepts the role of Research Sponsor for this study in accordance with the requirements of Department of Health Research Governance Guidelines.



Site Specific Assessment

This Trust Management Approval letter also incorporates site specific assessment for the Oxford University Hospitals NHS Trust site

Approved Documents

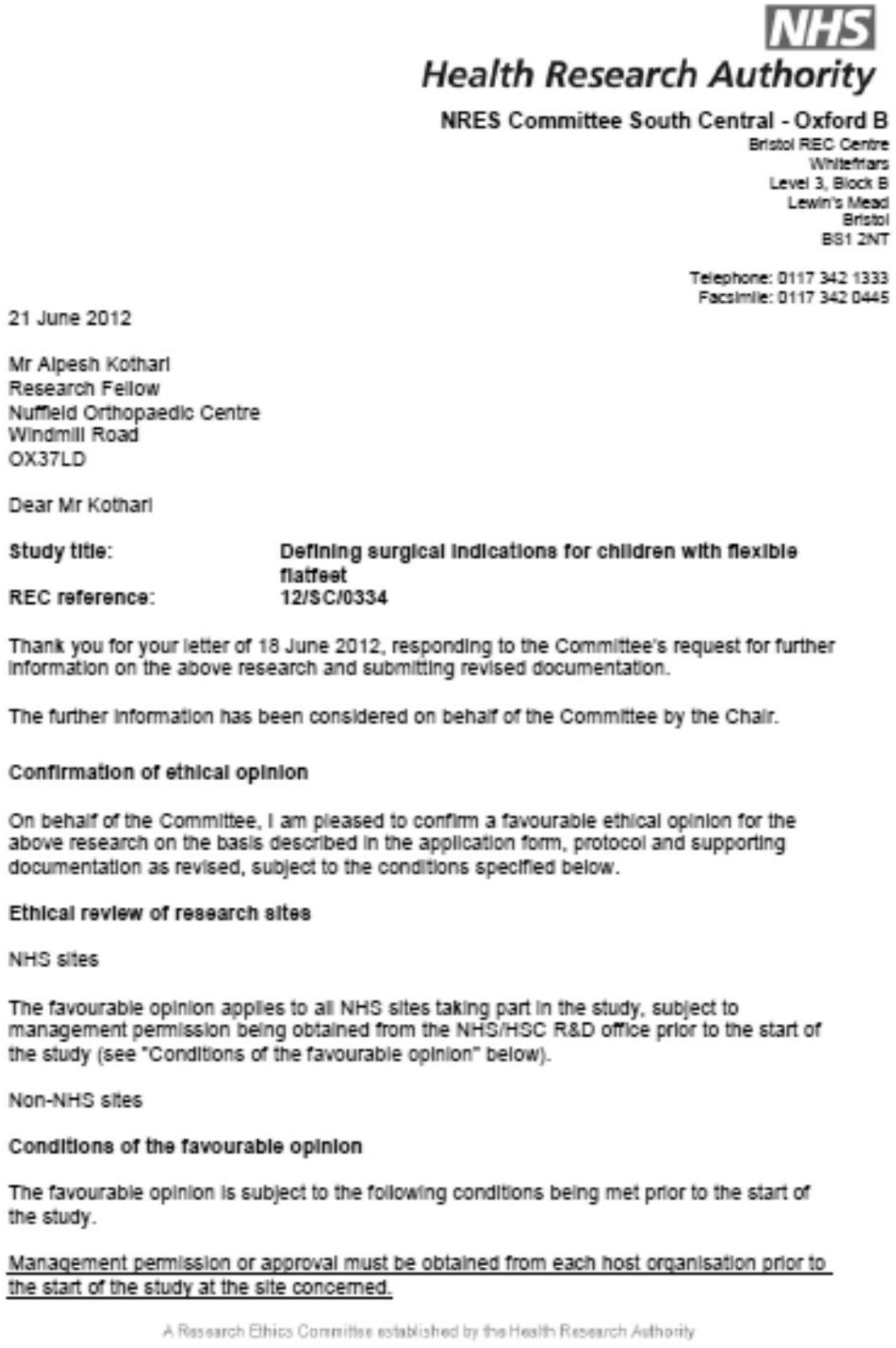
Document Type	**Version**	**Date**
Advertisement	1.0	18th May 2012
Evidence of Insurance or Indemnity		18th May 2012
Letter from Sponsor		18th May 2012
Letter of Investigation to Participant	1	18th June 2012
Investigator C.V.	Alpesh Kothari	
Letter from Funder		18th May 2012
MRI Safety Questionnaire	1	18th June 2012
Participant Consent Form: Assent Form for Children 8-10 Years Old	1	18th June 2012
Participant Consent Form: Assent Form for Children 11.15 Years Old	2	18th June 2012
Participant Consent Form: Parent/Guardian	2.0	18th June 2012
Participant Information Sheet: Parents Guardians of Young Volunteers	2.0	18th June 2012
Participant Information Sheet: Young Volunteers Aged 8-10	2	18th June 2012
Participant Information Sheet: Young Volunteers Aged 11-15	2	18th June 2012
Protocol	1.0	18th May 2012
Questionnaire: OXAFQ		
Questionnaire: Peds QOL		
R&D Form		
SSI Form		

I wish you every success with the study.

Yours sincerely,

Ms Heather House
Research & Development Lead

OUH R&D Dept. Non-CTIMP TMA approval template including conditions of approval October 2011 V3
Page 2 of 5



NHS
Health Research Authority

NRES Committee South Central - Oxford B
Bristol REC Centre
Whitefriars
Level 3, Block B
Lewin's Mead
Bristol
BS1 2NT

Telephone: 0117 342 1333
Facsimile: 0117 342 0445

21 June 2012

Mr Alpesh Kothari
Research Fellow
Nuffield Orthopaedic Centre
Windmill Road
OX37LD

Dear Mr Kothari

Study title:	**Defining surgical indications for children with flexible flatfeet**
REC reference:	**12/SC/0334**

Thank you for your letter of 18 June 2012, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Ethical review of research sites

NHS sites

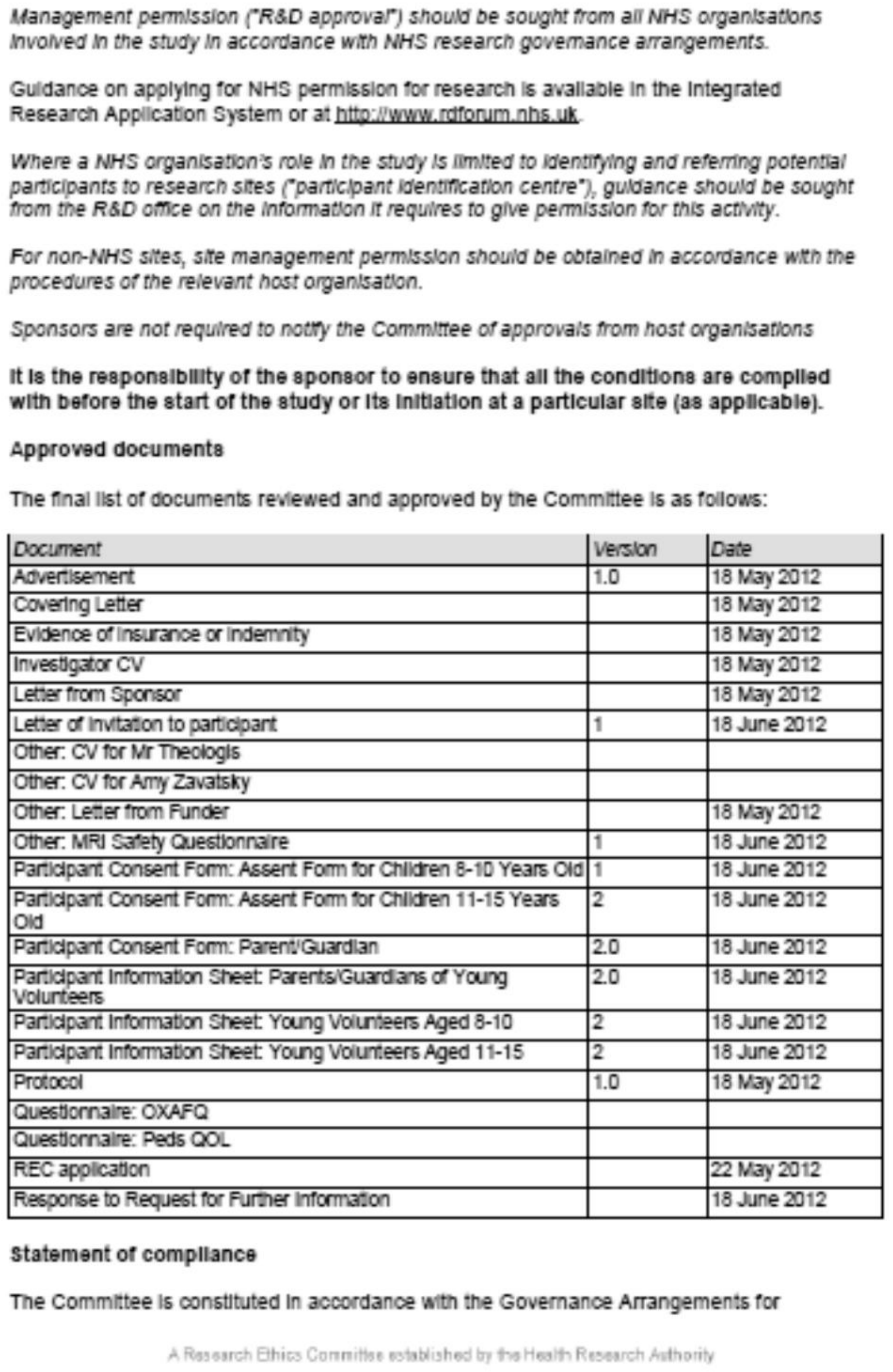
The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.



Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

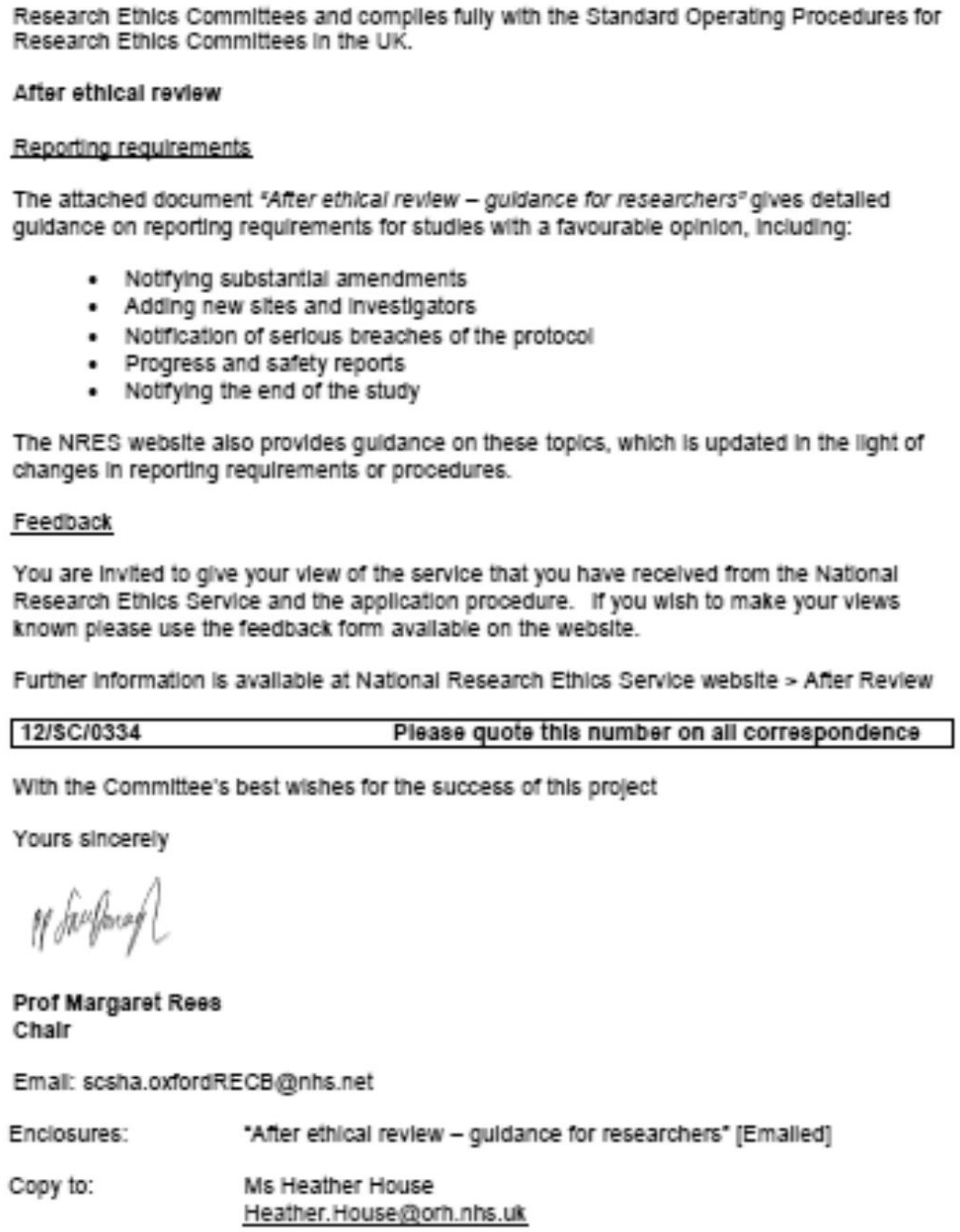
The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Advertisement	1.0	18 May 2012
Covering Letter		18 May 2012
Evidence of Insurance or Indemnity		18 May 2012
Investigator CV		18 May 2012
Letter from Sponsor		18 May 2012
Letter of invitation to participant	1	18 June 2012
Other: CV for Mr Theologis		
Other: CV for Amy Zavatsky		
Other: Letter from Funder		18 May 2012
Other: MRI Safety Questionnaire	1	18 June 2012
Participant Consent Form: Assent Form for Children 8-10 Years Old	1	18 June 2012
Participant Consent Form: Assent Form for Children 11-15 Years Old	2	18 June 2012
Participant Consent Form: Parent/Guardian	2.0	18 June 2012
Participant Information Sheet: Parents/Guardians of Young Volunteers	2.0	18 June 2012
Participant Information Sheet: Young Volunteers Aged 8-10	2	18 June 2012
Participant Information Sheet: Young Volunteers Aged 11-15	2	18 June 2012
Protocol	1.0	18 May 2012
Questionnaire: OXAFQ		
Questionnaire: Peds QOL		
REC application		22 May 2012
Response to Request for Further Information		18 June 2012

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for

A Research Ethics Committee established by the Health Research Authority



Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

12/SC/0334	**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project

Yours sincerely

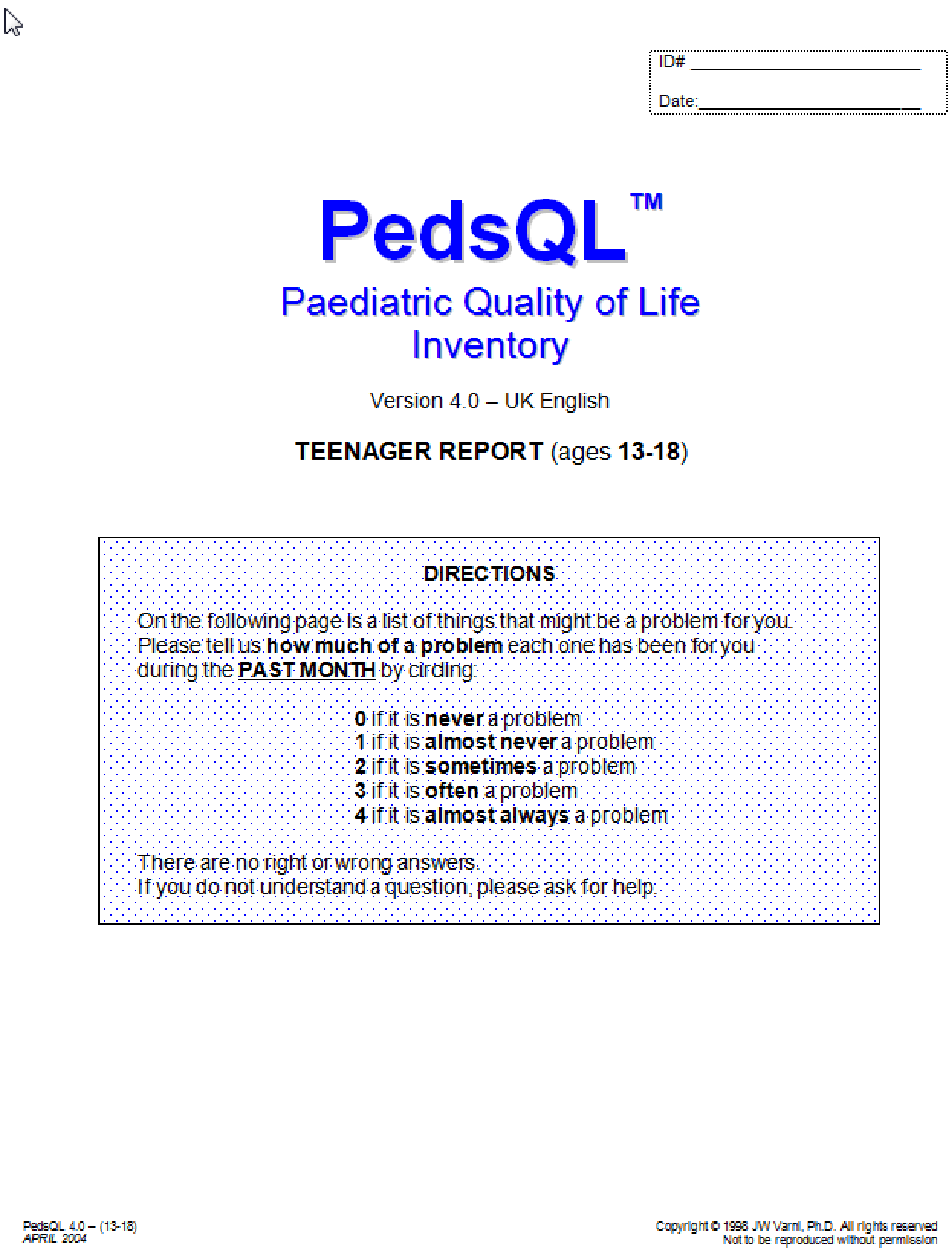
Prof Margaret Rees
Chair

Email: scsha.oxfordRECB@nhs.net

Enclosures: "After ethical review – guidance for researchers" [Emailed]

Copy to: Ms Heather House
Heather.House@orh.nhs.uk

Appendix 2. Health-related quality of life measures



ID# ______________________

Date: ______________________

PedsQL™

Paediatric Quality of Life Inventory

Version 4.0 – UK English

TEENAGER REPORT (ages **13-18**)

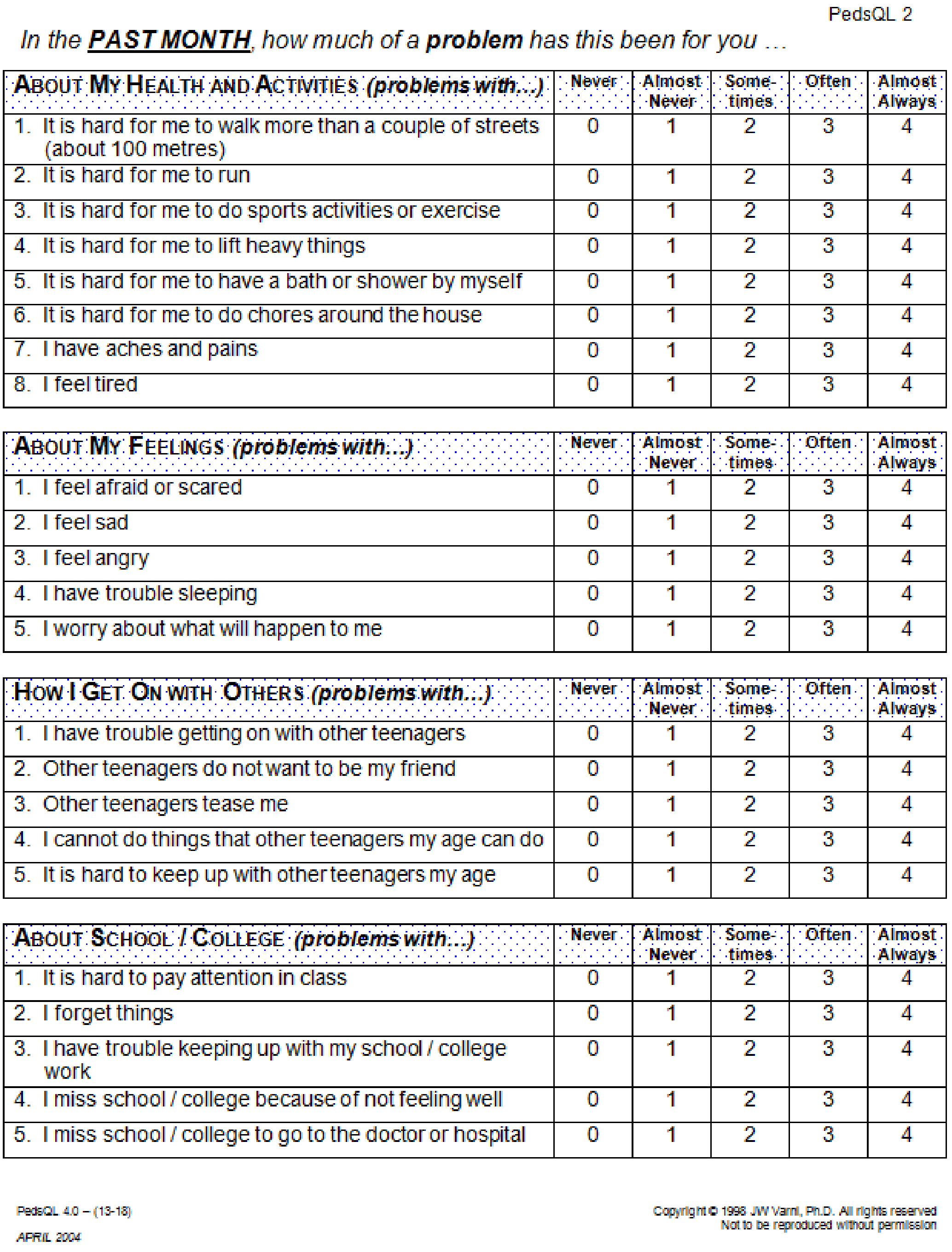
DIRECTIONS

On the following page is a list of things that might be a problem for you.
Please tell us **how much of a problem** each one has been for you
during the **PAST MONTH** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

PedsQL 4.0 – (13-18)
APRIL 2004



PedsQL 2

In the ***PAST MONTH****, how much of a* ***problem*** *has this been for you …*

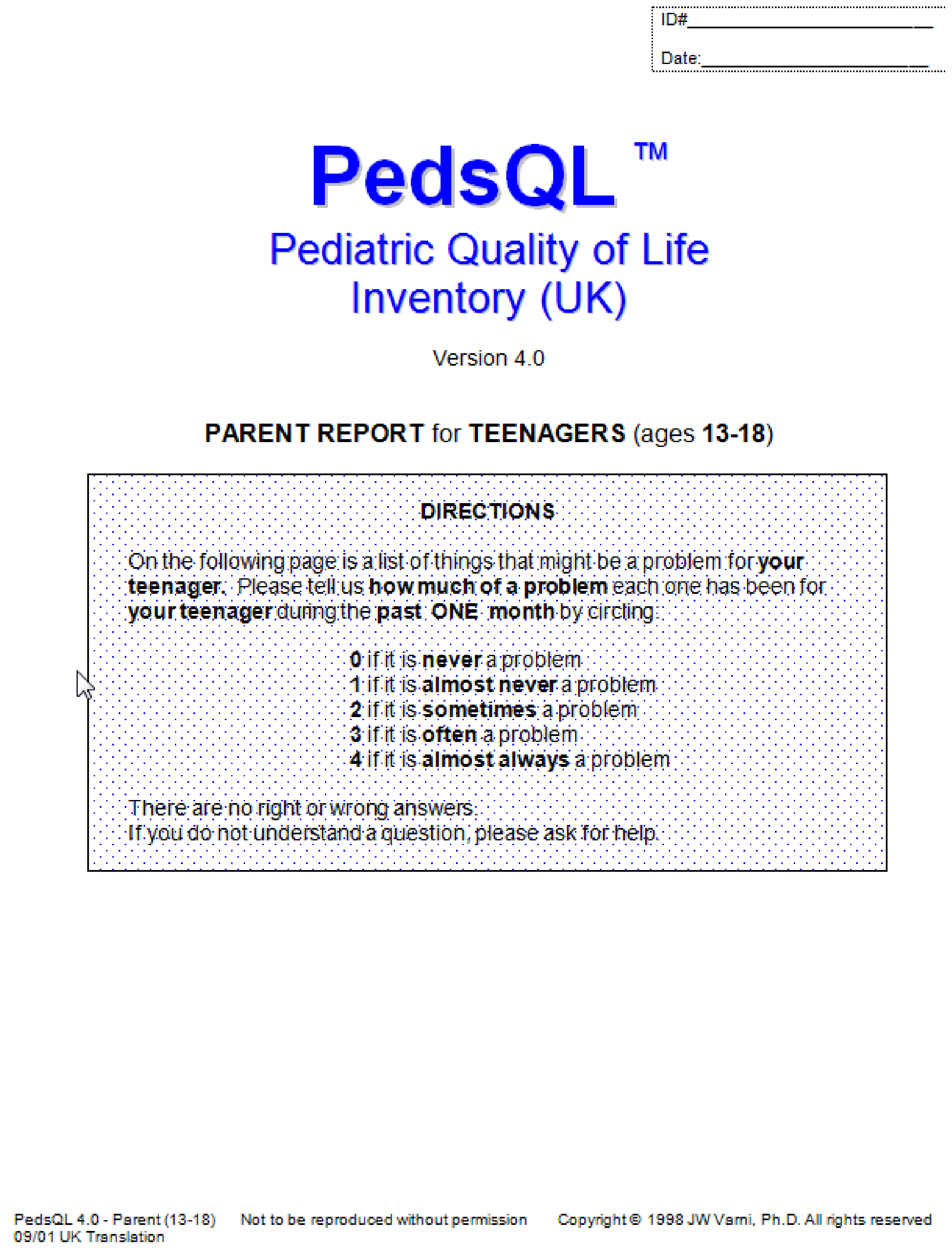
ABOUT MY HEALTH AND ACTIVITIES ***(problems with…)***	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than a couple of streets (about 100 metres)	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activities or exercise	0	1	2	3	4
4. It is hard for me to lift heavy things	0	1	2	3	4
5. It is hard for me to have a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I have aches and pains	0	1	2	3	4
8. I feel tired	0	1	2	3	4

ABOUT MY FEELINGS ***(problems with…)***	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

HOW I GET ON WITH OTHERS ***(problems with…)***	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting on with other teenagers	0	1	2	3	4
2. Other teenagers do not want to be my friend	0	1	2	3	4
3. Other teenagers tease me	0	1	2	3	4
4. I cannot do things that other teenagers my age can do	0	1	2	3	4
5. It is hard to keep up with other teenagers my age	0	1	2	3	4

ABOUT SCHOOL / COLLEGE ***(problems with…)***	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my school / college work	0	1	2	3	4
4. I miss school / college because of not feeling well	0	1	2	3	4
5. I miss school / college to go to the doctor or hospital	0	1	2	3	4

PedsQL 4.0 – (13-18)
APRIL 2004



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Date:_______________________

PedsQL ™

Pediatric Quality of Life Inventory (UK)

Version 4.0

PARENT REPORT for **TEENAGERS** (ages **13-18**)

DIRECTIONS

On the following page is a list of things that might be a problem for **your teenager.** Please tell us **how much of a problem** each one has been for **your teenager** during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

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*In the past **ONE month,** how much of a **problem** has your teenager had with …*

PHYSICAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking down the road a little bit	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports or running games	0	1	2	3	4
4. Lifting heavy things	0	1	2	3	4
5. Having a bath or shower by him or herself	0	1	2	3	4
6. Tidying up around the house	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Feeling very tired	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or unhappy	0	1	2	3	4
3. Feeling angry or cross	0	1	2	3	4
4. Trouble sleeping at night	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Getting on with other teenagers	0	1	2	3	4
2. Other teenagers not wanting to be his or her friend	0	1	2	3	4
3. Getting bullied by other teenagers	0	1	2	3	4
4. Not able to do things that other teenagers his or her age can do	0	1	2	3	4
5. Keeping up with other teenagers during activities	0	1	2	3	4

SCHOOL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with schoolwork	0	1	2	3	4
4. Having days off school because of not feeling well	0	1	2	3	4
5. Having days off school to go to the doctor or hospital	0	1	2	3	4

ID# ______________________

Date:______________________

PedsQL™

Paediatric Quality of Life Inventory

Version 4.0 – UK English

CHILD REPORT (ages **8-12**)

DIRECTIONS

On the following page is a list of things that might be a problem for you. Please tell us **how much of a problem** each one has been for you during the **PAST MONTH** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

PedsQL 2

In the ***PAST MONTH****, how much of a* ***problem*** *has this been for you …*

ABOUT MY HEALTH AND ACTIVITIES ***(problems with…)***	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than a couple of streets (about 100 metres)	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activities or exercise	0	1	2	3	4
4. It is hard for me to lift heavy things	0	1	2	3	4
5. It is hard for me to have a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I have aches and pains	0	1	2	3	4
8. I feel tired	0	1	2	3	4

ABOUT MY FEELINGS ***(problems with…)***	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

HOW I GET ON WITH OTHERS ***(problems with…)***	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting on with other children	0	1	2	3	4
2. Other children do not want to be my friend	0	1	2	3	4
3. Other children tease me	0	1	2	3	4
4. I cannot do things that other children my age can do	0	1	2	3	4
5. It is hard to keep up when I play with other children	0	1	2	3	4

ABOUT SCHOOL ***(problems with…)***	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my schoolwork	0	1	2	3	4
4. I miss school because of not feeling well	0	1	2	3	4
5. I miss school to go to the doctor or hospital	0	1	2	3	4

PedsQL 4.0 – (8-12)
APRIL 2004

ID# ______________________

Date: ______________________

PedsQL ™

Pediatric Quality of Life Inventory (UK)

Version 4.0

PARENT REPORT for **CHILDREN** (ages **8-12**)

DIRECTIONS

On the following page is a list of things that might be a problem for **your child**. Please tell us **how much of a problem** each one has been for **your child** during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month,** how much of a **problem** has your child had with …*

PHYSICAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking down the road a little bit	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports or running games	0	1	2	3	4
4. Lifting heavy things	0	1	2	3	4
5. Having a bath or shower by him or herself	0	1	2	3	4
6. Tidying up around the house	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Feeling very tired	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or unhappy	0	1	2	3	4
3. Feeling angry or cross	0	1	2	3	4
4. Trouble sleeping at night	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Getting on with other children	0	1	2	3	4
2. Other kids not wanting to be his or her friend	0	1	2	3	4
3. Getting bullied by other children	0	1	2	3	4
4. Not able to do things that other children his or her age can do	0	1	2	3	4
5. Keeping up when playing with other children	0	1	2	3	4

SCHOOL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with schoolwork	0	1	2	3	4
4. Having days off school because of not feeling well	0	1	2	3	4
5. Having days off school to go to the doctor or hospital	0	1	2	3	4

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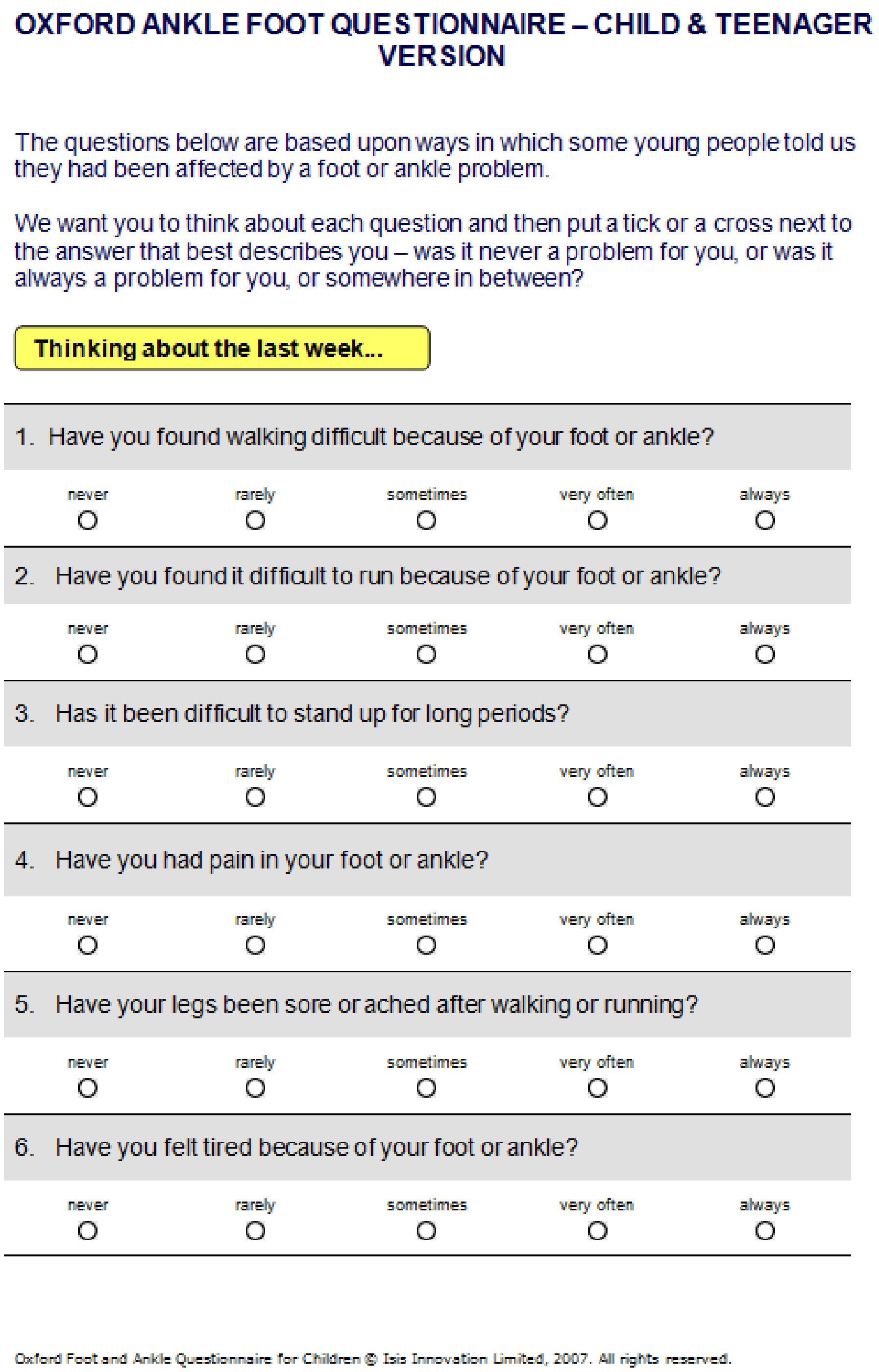

OXFORD ANKLE FOOT QUESTIONNAIRE – CHILD & TEENAGER VERSION

The questions below are based upon ways in which some young people told us they had been affected by a foot or ankle problem.

We want you to think about each question and then put a tick or a cross next to the answer that best describes you – was it never a problem for you, or was it always a problem for you, or somewhere in between?

Thinking about the last week...

1. Have you found walking difficult because of your foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

2. Have you found it difficult to run because of your foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

3. Has it been difficult to stand up for long periods?

never	rarely	sometimes	very often	always
○	○	○	○	○

4. Have you had pain in your foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

5. Have your legs been sore or ached after walking or running?

never	rarely	sometimes	very often	always
○	○	○	○	○

6. Have you felt tired because of your foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

7. Has your foot or ankle stopped you joining in with others in the playground?

never	rarely	sometimes	very often	always
○	○	○	○	○

8. Has your foot or ankle stopped you playing in the park or outside?

never	rarely	sometimes	very often	always
○	○	○	○	○

9. Has your foot or ankle stopped you taking part in PE lessons?

never	rarely	sometimes	very often	always
○	○	○	○	○

10. Has your foot or ankle stopped you taking part in any other lessons at school?

never	rarely	sometimes	very often	always
○	○	○	○	○

11. Have you been bothered by how your foot or ankle looks?

never	rarely	sometimes	very often	always
○	○	○	○	○

12. Has the way you walk bothered you?

never	rarely	sometimes	very often	always
○	○	○	○	○

13. Have you been embarrassed because of your foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

14. Has anyone been unkind to you because of your foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

15. Has your foot or ankle stopped you wearing any shoes you wanted to wear?

never	rarely	sometimes	very often	always
○	○	○	○	○

OXFORD ANKLE FOOT QUESTIONNAIRE – PARENT VERSION

The questions below are based upon ways in which some young people told us they had been affected by a foot or ankle problem.

We want you to think about each question and then put a tick or a cross next to the answer that best describes your child – was it never a problem for them, or was it always a problem, or was it somewhere in between?

Thinking about the last week...

1. Has your child found walking difficult because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

2. Has your child found it difficult to run because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

3. Has it been difficult for your child to stand up for long periods?

never	rarely	sometimes	very often	always
○	○	○	○	○

4. Has your child had pain in their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

5. Have your child's legs been sore or ached after walking or running?

never	rarely	sometimes	very often	always
○	○	○	○	○

6. Has your child felt tired because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

Parent Questionnaire Version 1.1 April 2008

7. Has your child's foot or ankle stopped them joining in with others in the playground?

never	rarely	sometimes	very often	always
○	○	○	○	○

8. Has your child's foot or ankle stopped them playing in the park or outside?

never	rarely	sometimes	very often	always
○	○	○	○	○

9. Has your child's foot or ankle stopped them taking part in PE lessons?

never	rarely	sometimes	very often	always
○	○	○	○	○

10. Has your child's foot or ankle stopped them taking part in any other lessons at school?

never	rarely	sometimes	very often	always
○	○	○	○	○

11. Has your child been bothered by how their foot or ankle looks?

never	rarely	sometimes	very often	always
○	○	○	○	○

12. Has the way your child walks bothered them?

never	rarely	sometimes	very often	always
○	○	○	○	○

13. Has your child been embarrassed because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

14. Has anyone been unkind to your child because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

15. Has your child's foot or ankle stopped them wearing any shoes they wanted to wear?

never	rarely	sometimes	very often	always
○	○	○	○	○

Appendix 3. Publications and presentations

Published

Kothari A, Dixon P Stebbins J, Zavatsky A, Theologis T. The relationship between quality of life and foot function in children with flexible flatfeet. Gait and Posture. 2015;41(3):786:790.

Kothari A, Stebbins J, Zavatsky A, Theologis T. Health related quality of life in children with flexible flatfeet: a cross-sectional study. Journal of Children's Orthopaedics. 2014;8(6):489-96.

Kothari A, Dixon P, Stebbins J, Zavatsky A, Theologis T. Motion analysis to track navicular displacements in the paediatric foot: Relationship with foot posture, body mass index and flexibility. Foot and Ankle International 2014 Sept;35(9):929-37.

Kothari A, Kerr C, Stebbins J, Zavatsky A, Theologis T. Centre of pressure progression and gait parameter deviations may be related to second rocker dysfunction in children with flat feet. Journal of Foot and Ankle Research. 2014; 7(Suppl1):A47

Invited for resubmission

Kothari A, Bhuva B, Stebbins J, Zavatsky AB, Theologis T. Flexible flatfeet and impaired quality of life: the role of subtalar joint morphology. Bone and Joint Journal. Log No. 36059

Kothari A, Dixon P, Stebbins J, Zavatsky AB, Theologis T. Are flexible flat feet associated with proximal joint problems in children? Gait and Posture Log No. GAIPOS-D-15-00477

Presentations

Kothari A, Stebbins J, Zavatsky A, Theologis T. Health related quality of life in children with flexible flatfeet: a cross-sectional study. European Paediatric Orthopaedic Society. Marseille, April 2015 (podium)

Kothari A, Stebbins J, Zavatsky A, Theologis T. Aberrant subtalar morphology is associated with flexible flatfeet in children. European Paediatric Orthopaedic Society. Marseille, April 2015 (e-poster)

Kothari A, Stebbins J, Zavatsky A, Theologis T. Subtalar morphology and the determinants of flexible flatfeet in children. American Academy of Orthopaedic Surgeons. Las Vegas, March 2015 (podium)

Kothari A, Dixon P, Stebbins J, Zavatsky A, Theologis T. The relationship between quality of life and foot kinematics in flatfooted children. European Society of Movement Analysis for Adults and Children. Rome, October 2014 (podium)

Kothari A, Kerr C, Stebbins J, Zavatsky A, Theologis T. Centre of pressure progression and gait parameter deviations may be related to second rocker dysfunction in children with flatfeet. European Society of Movement Analysis for Adults and Children. Rome, October 2014 (podium)

Kothari A, Stebbins J, Zavatsky A, Theologis T. Using the Oxford Ankle Foot Questionnaire to evaluate children with flexible flatfeet:- A comparison with typically developing feet, treated clubfoot and cerebral palsy. Paediatric Orthopaedic Society of North America. Los Angeles, April 2014 (podium)

Paik A, Stebbins J, Kothari A, Zavatsky A. Effect of marker placement on Oxford Foot Model hindfoot segment axes. International Foot and Ankle Biomechanics Congress. Busan, April 2014 (podium)

Kothari A, Stebbins J, Zavatsky A, Theologis T. Kinematic differences between neutral and flatfooted children and their relationship with quality of life. British Society of Children's Orthopaedic Surgery. Aberdeen, March 2014 (podium)

Kothari A, Dixon P, Stebbins J, Zavatsky AB, Theologis T. Motion analysis to track navicular displacements in the paediatric foot: relationship with foot posture and flexibility. European Society of Movement Analysis for Adults and Children. Glasgow, September 2013 (podium)

Appendix 4. List of abbreviations

3D	Three-dimensional
3DGA	Three-dimensional gait analysis
3DMA	Three-dimensional motion analysis
AH	Arch height
AHI	Arch Height Index
AI	Arch Index
AMAX	Peak anterior ground reaction force
APGRF	Antero-posterior ground reaction force
APowMax	Peak ankle power
BMI	Body mass index
BW	Body weight
CF	Clubfoot
CI	Confidence interval
CIA	Calcaneal inclination angle
CMAS	Clinical Movement Analysis Society UK and Northern Ireland
COM	Centre of mass
COP	Centre of pressure
COPP	Centre of pressure progression
CP	Cerebral palsy
CSI	Chippaux-Smirak Index
CT	Computed tomography
DAH	Dorsal arch height
DMAX	Peak ankle dorsiflexor moment
DyNDri	Dynamic navicular drift
DyNDro	Dynamic navicular drop
EMG	Electromyography
ES	Effect size
EvI	Hindfoot evertor moment impulse
ExtMAX	Peak hindfoot external rotation moment
FF	Foot flat
FF	Flexible flat feet
FFHFA	Forefoot hindfoot angle
FL	Foot length
FPI	Foot Posture Index
GCP	Good clinical practice
GRF	Ground reaction force
HABMAX	Peak hip abduction
HAMAX2	Second peak hip adduction moment
HEMAX	Peak hip extension moment
HFMAX	Peak hip flexion moment
HFMM	Heidelberg foot measurement method
HFTBA	Hindfoot tibia angle
HIMAX	Peak hip internal rotation moment
HO	Heel-off

HPABS	Peak hip power absorption
HPGEN	Peak hip power generation
HRQOL	Health-related quality of life
HXFFA	Hallux forefoot angle
I	Anterior propulsive impulse
ICC	Intraclass correlation
IDA	Inverse dynamic analysis
IMAX	Peak ankle invertor moment
IOR	Instituto Orthopaedico Rizzoli
IQR	Inter-quartile range
KFMAX	Peak knee flexion moment
KFTROUGH	Knee flexion extension trough
KIMAX	Peak knee internal rotation moment
KINTMAX	Peak knee internal rotation
KPGEN	Peak knee power generation
KVMAX2	Second peak knee varus moment
KVMID	Knee midstance varus
LLA	Lateral longitudinal arch
LLAS	Lower limb assessment score
LMAX	Peak lateral ground reaction force
LRMIN	Loading response minimum
LS	Less symptomatic
MAX	Maxima
MFM	Milwaukee foot model
MID	Minimally important difference
MIDS	Midstance
MIN	Minima
MLA	Medial longitudinal arch
MLGRF	Medio-lateral ground reaction force
MLRANGE	Range of medio-lateral ground reaction force
MMAX	Peak medial ground reaction force
MPP	Maximum pressure print
MRI	Magnetic resonance imaging
MS	More symptomatic
MSFM	Multi-segment foot model
mSHCG	Modified Shriner’s Hospital of Children Greenville
NDri	Navicular drift
NDro	Navicular drop
NF	Neutral feet
NH	Navicular height
NHS	National health service
NNDri	Normalized navicular drift
NNDro	Normalized navicular drop
NNH	Normalized navicular height
OFM	Oxford Foot Model
OGL	Oxford Gait Laboratory
OR	Odds ratio
OxAFQ-C	Oxford Ankle Foot Questionnaire for Children

Pa	Pascals
PedsQL™ 4.0	Pediatric quality of life inventory
PERMAX	Peak pelvis external rotation
PFF	Paediatric flexible flat feet
PiG	Plug in Gait
PROMAX	Peak midfoot pronation moment
PTTD	Posterior tibialis tendon dysfunction
RCSP	Resting calcaneal stance position
RMSD	Root mean squared difference
ROC	Receiver operative characteristic
ROI	Region of interest
ROM	Range of motion
ROP	Roll over process
SD	Standard deviation
SHR	Single heel rise
SI	Staheli Index
T1MA	Talo-1st metatarsal angle
TFL	Truncated foot length
FO	Foot off
VGRF	Vertical ground reaction force
VI	Valgus Index
VMAX2	Second peak of vertical ground reaction force
VMIN	Midstance trough of vertical ground reaction force
WHO	World Health Organisation