Clinical Meaningfulness of the Oxford Foot Model to Assess Foot Deformity during Gait

Jennifer L. McCahill

The research presented in this thesis was a collaboration between:

1. Amsterdam University Medical Centers, Amsterdam Movement Science, Dept Rehabilitation Medicine, Netherlands
2. Oxford Gait Laboratory, Oxford University Hospitals NHS Foundation Trust, UK
3. Nuffield Department of Orthopaedics, Rheumatology and Musculoskeletal Sciences, University of Oxford, UK

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CLINICAL MEANINGFULNESS OF THE OXFORD FOOT MODEL TO ASSESS FOOT DEFORMITY DURING GAIT

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Jennifer Leigh McCahill

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promotoren:	prof.dr.ir. J. Harlaar
	prof.dr.ir. C. Lavy
copromotoren:	prof.dr. T. Theologis
	dr. J. Stebbins
promotiecommissie:	prof.dr. B.J. van Royen
	dr. M.M. van der Krogt
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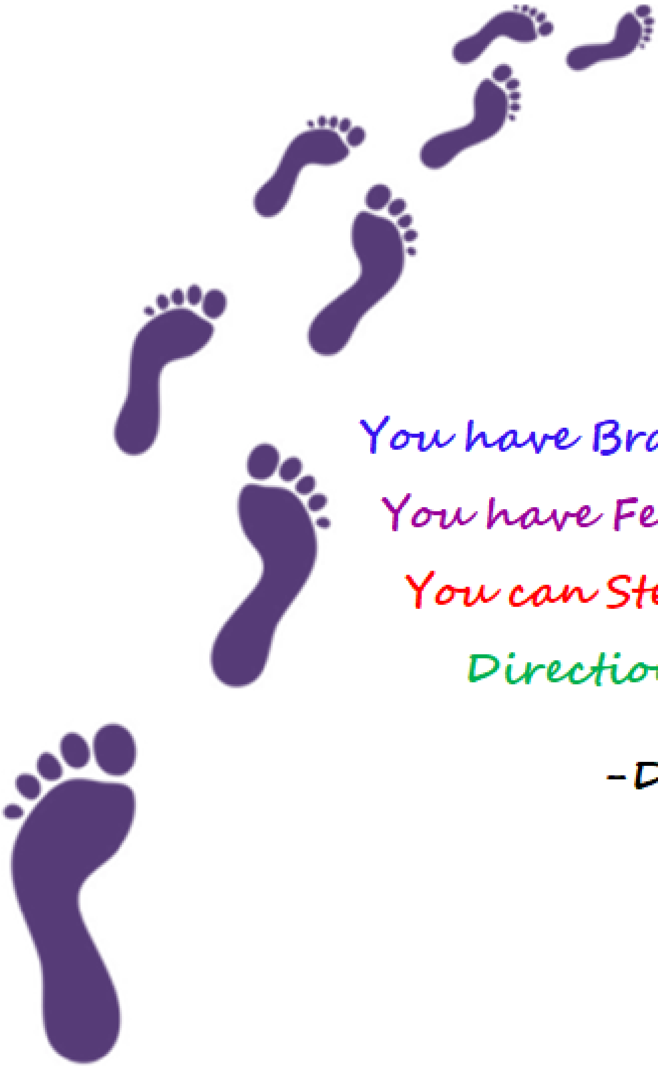

You have Brains in your Head
You have Feet in your Shoes.
You can Steer yourself any
Direction you choose.

-Dr Seuss

The prevalence of foot deformity is globally high affecting populations across the lifespan. Foot deformity can be present from birth (such as clubfoot), emerge during growth/ present over-time (flat foot, hallux valgus), or occur following an injury or neurological event (cerebral palsy, stroke). A person with a foot deformity has altered foot structure and potentially foot function. This in turn may limit their activities in daily life due to long-lasting pain and diminished walking capacity. Despite the common clinical presentation of foot deformity altering an individual's function, the correlation between the amount of structural deformity and its effects on quality of life remains unclear. The challenge for health professionals is to identify a clinically meaningful level of deformity that warrants intervention to maximise an individual's participation in society.

The available assessments of foot deformity in the literature are largely static measures despite previous studies showing significant differences between static structure and dynamic foot function. Optimal assessment of abnormal foot structure could be achieved through assessment in three dimensions during gait and function. Three-dimensional gait analysis is an assessment tool which measures dynamic deformity in the lower limbs. More recently, three-dimensional multi-segment foot models have been developed to improve our understanding of foot motion during gait, such as the Oxford Foot Model (OFM). The OFM was developed to measure tibia, hindfoot, forefoot and hallux motion in a clinical setting. As a relatively recent development in the assessment of dynamic foot motion, rigorous clinimetric testing of the OFM is still lacking, limiting its full potential for clinical applications and research utility. Therefore the general aim of this thesis was to establish the clinical role of the Oxford Foot Model to assess foot function during gait in the presence of deformity.

Chapter I is the introduction of the thesis detailing the prevalence of foot deformity and the lack of dynamic foot assessments available in the literature. Gait, gait analysis and foot kinematics are discussed, leading to the clear gap in the literature providing the basis of the research aim. Chapter II is a repeatability study to justify the use of the OFM in populations with known foot deformity. Previously the repeatability of the OFM had been assessed in adults and children healthy populations. The OFM was designed to be adaptable in its application to measure different types of foot deformity therefore, it is important to know its repeatability in pathological conditions. This study assessed the intra and inter-rater repeatability of marker placement in children with clubfoot and in children with hemiplegic cerebral palsy compared to a typically developing population. The results of this study show that the OFM provides repeatable results in healthy children, as well as in children with either congenital or acquired foot deformity.

Chapter III builds on previous research completed in Oxford evaluating the repeatability of the hindfoot marker in the OFM suggesting that the axes of the hindfoot are most sensitive to marker placement on the posterior aspect of the heel. Since other multi-segment foot models also use a similar marker, it is important to find methods to place this as accurately as possible. The aim of this pilot study was to test two different ‘jigs’ (anatomical alignment devices) against the eyeball marker placement method to improve reliability of heel marker placement and calculation of hindfoot angles. Two gait analysts (one beginner and one experienced with the foot model) completed this repeatability study on 10 healthy adult subjects using a ratio caliper and heel mould, both designed by three-dimensional printing, against eyeball marker placement. The intra-tester and inter-tester repeatability of hindfoot marker placement were assessed for 5 clinically relevant variables of the OFM. Overall the results showed there was low intra-tester and inter-tester variability suggesting good sensitivity of the OFM to detect meaningful clinical differences. The use of the ratio caliper may improve intra-tester variability, but did not seem superior to the eyeball method of marker placement for inter-tester variability. The use of a heel mould was discouraged.

Chapter IV addresses the lack of available dynamic assessment tools of foot function in the literature. To summarise the quality of foot motion over the gait cycle, the Foot Profile Score (FPS) was defined as a single score based on the OFM kinematics expressing the overall deviation of foot function relative to the norm. The aim of this study was to define and validate the FPS by studying its properties and design, and analyse it against a clinical assessment of foot deformity. Concurrent validity was established for the FPS analysing the relationship with Clinical Foot Deformity Score (CFDS) in 60 subjects with a condition affecting the lower limbs. Content validity was established for the six Foot Variable Scores (FVS) that make up the FPS using a multiple regression of the CFDS on the 6 FVS in the 60 subjects. Predictive validity was established analysing the relationship of the FPS and GPS comparing 60 lower limb involvement subjects with 60 subjects with isolated foot deformity. The FPS has become the first validated score of dynamic foot motion.

Chapter V analyses the responsiveness of the FPS in a clinical population. The FPS enables clinicians and researchers to quantify deviations of foot motion during gait, to monitor change in foot/ankle motion over time, and to measure the outcome of intervention. With the creation of a new outcome measure, it is important to test its responsiveness to intervention in a clinical population. Firstly, we defined the minimal clinically important difference (MCID) for the FPS based on the regression of the FPS on the Clinical Foot Deformity Scale (CFDS) presented in Chapter IV. Using the MCID, we applied it to a clinical population of 37 children with cerebral palsy, spastic hemiplegia, comparing their FPS before and after isolated foot and ankle surgery. A regression analysis looked at potential relationships between the change in FPS and their pre-operative FPS, age at surgery, and time since surgery. An MCID of 2.4 degrees for the FPS indicated a clinically meaningful improvement in foot function, which was evident in 76% of children with hemiplegia post isolated foot/ankle surgery. Moreover, the FPS responded with larger improvements for more deformed feet. These findings suggest the FPS is sufficiently responsive in a clinical population and should be considered when indicating and evaluating foot surgery.

Chapter VI investigates if older symptomatic children with clubfoot deformity differ in perceived disability and foot function during gait, depending on initial treatment with Ponseti or surgery, compared to a control group. The second aim was to investigate correlations between foot function during gait and perceived disability in this population. Foot function was assessed by the OFM kinematics and plantar pressure and correlated with parent-reported outcome measures including the Oxford Ankle Foot Questionnaire, the Disease Specific Index for clubfoot and the Pediatric Quality of Life Inventory 4.0. Our findings suggest that symptomatic children with clubfoot deformity present with similar degrees of gait deviations and perceived disability regardless of whether they had previously been treated with the Ponseti Method or surgery. The presence of sagittal and coronal plane hindfoot deformity and coronal plane forefoot deformity were associated with higher levels of perceived disability, regardless of their initial treatment. This was the first study to compare outcomes between Ponseti and surgery in a symptomatic older clubfoot population seeking further treatment. In addition, it was the first paper to correlate foot function during gait and perceived disability to establish a link between deformity and subjective outcomes.

In Chapter VII, the main findings of the presented studies were critically discussed, leading to clinical implications and ideas for future research. To summarise, this thesis was able to establish the Oxford Foot Model (OFM) and its summary score, the Foot Profile Score (FPS), provide clinically meaningful information for treatment indication and evaluation of dynamic foot deformity during gait in the presence of foot deformity.

General Introduction

The human foot is a complex structure comprising of 26 bones, 33 joints, and more than a hundred muscles, tendons and ligaments (1). It can be subdivided into the hindfoot (talus and calcaneus), the midfoot (five tarsal bones) and the forefoot (five metatarsals and 14 phalanges or toes) (1). The adaptability of the foot is essential to normal human walking to provide shock absorption as the heel contacts the ground through to a rigid lever for propulsion as the foot leaves the floor. Any injury or deformity of the foot can alter its natural biomechanics and ability to generate intrinsic forces, which can lead to long-term pain, walking difficulties and disability.

Picture of newborn feet: Alfred James McDonnell born 12.07.2016

INCIDENCE OF FOOT DEFORMITY

Foot deformity can be present from birth (such as clubfoot), emerge during growth/ present over-time (flat foot, hallux valgus), or occur following an injury or neurological event (cerebral palsy, stroke). The incidence of foot deformities present at birth has been reported as high as 4.2% (2) and can include clubfoot, metatarsus adductus, calcaneovalgus, and vertical talus. Flexible flat feet are common in young children with an incidence reported between 2.7% and 18.1% (3,4), and this often spontaneously corrects as their foot posture continues to mature until the age 8 years (5). Some children however maintain their flat foot postures, especially if they are genetically pre-disposed (1). Adults can acquire a flat foot posture with reported rates >3% in women over the age of 40 and >10% in adults over the age of 65 years (6,7). The incidence of acquired foot deformity following a neurological injury is high in children and adults (>80%) due to progressive abnormal forces (spasticity and weakness) across their foot and ankle joints (8).

THE IMPACT OF FOOT DEFORMITY ON AN INDIVIDUAL AND SOCIETY

The International Classification of Functioning, Disability and Health (WHO, 2002) highlights the interactive relationship between human functioning and disability (Figure 1).

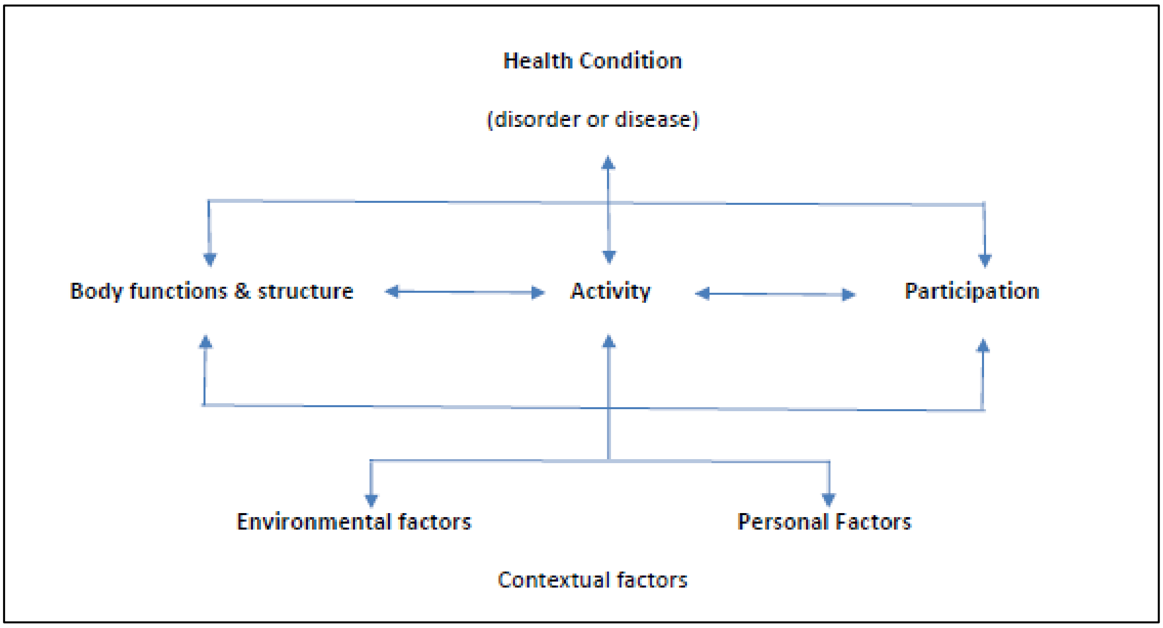

Figure 1: The International Classification of Functioning, Disability and Health (ICF). Source: World Health Organisation Geneva 2002, 'Towards a Common Language for Functioning, Disability and Health: ICF'.

A person with a foot deformity has altered foot structure and potentially foot function. This in turn may limit their activities in daily life due to long-standing pain and diminished walking capacity, thus reducing their participation within society. There is strong evidence in the literature that foot deformities can significantly negatively impact a person's quality of life (9-12). In individuals with flat foot, there is evidence suggesting their altered foot postures lead to abnormal movement patterns and forces in more proximal lower limb joints contributing to knee pain (13-16), hip pain (14) and back pain (15-19).

Despite the common clinical presentation of foot deformity altering an individual's function, the correlation between the severity of structural deformity and its effects on quality of life is not clear (11). The challenge for health professionals is to identify a clinically meaningful level of deformity that may warrant intervention to maximise an individual's participation in society (20). The difficulty in defining this is that often the severity of foot deformity does not correlate directly with foot function or subjective reports of quality of life (11). Seemingly the same amount of deformity in two people can lead to very different outcomes: for example, one is painful, and one is not. So, the questions remain: How can we improve our measurement of foot function to better inform treatment decisions that target foot deformities which lead to functional deficits and disability? What is the relationship between foot structure, dynamic foot function and perceived disability?

STANDARD MEASUREMENTS OF FOOT DEFORMITY

A recent systematic review by Banwell and colleagues (2018) investigated how paediatric foot posture is defined and measured in the literature (20). In the 27 studies reviewed, the authors found 40 definitions of paediatric flat foot indicating little consensus for the amount of deformity that is considered atypical. They defined four groupings of available assessments for foot related deformity: plain film radiographs, foot print indices, static foot measures, and plantar pressure analysis (20). All these available assessments are based on static measurements despite previous studies showing significant differences between static structure and dynamic foot function (20,21). This highlights a need for dynamic assessment of foot function (20), which may be more clinically relevant providing a better relationship between atypical foot function and perceived disability.

Hijji and colleagues (2020) recently completed a systematic review of the adult foot and ankle literature with the aim to consolidate the outcome measures used in foot and ankle medicine (22). The authors summarised:

"It is well established that evidence-based practice and patient-centred outcomes are essential in health care.
The ideal outcome measurement tool should be relevant, reliable, valid, and responsive to a given pathology. Additionally, it should be able to detect a clinically meaningful difference in varying disease states, thus enabling comparisons between studies and permitting accurate assessments of different treatment modalities."

[Hijji et al 2020]

The outcome measures from 541 studies were grouped into three categories: generic (eg. Visual Analogue Scale), foot and ankle specific (eg. American Orthopaedic Foot and Ankle Society Scale), and disease-specific (eg. Ankle Osteoarthritis Scale) (22). Their review of the literature suggested a higher level of evidence was associated with studies who used a disease-specific outcome measure in combination with a generic outcome measure (such as Harlaar and colleagues (23)), whereas the use of foot and ankle specific measure alone was associated with lower level of evidence (22). In addition, the authors concluded that patient reported outcome measures have become increasingly prevalent in the published foot and ankle literature, however substantial variability exists among reported outcome measurement tools (22).

MEASURING HUMAN GAIT

Research suggests that static measurement of the foot shape does not correlate to how a foot will function during walking (20,21). Human walking is a complex synergy of the lower limbs with the foot and ankle an integral component. Saunders, Inman and Eberhart in 1953 (24) described gait as the following:

> *"Human locomotion is a phenomenon of the most extraordinary complexity in which so great are the multitude of individual motions occurring simultaneously in the three planes of space … locomotion is the translation of the center of gravity through space along a pathway requiring the least expenditure of energy"*

Figure 2 depicts a complete gait cycle for the right leg. The gait cycle consists of a stance and a swing phase, with single and double support times. The gait cycle begins with initial contact on the floor with one foot and ends with the same foot contacting the floor again.

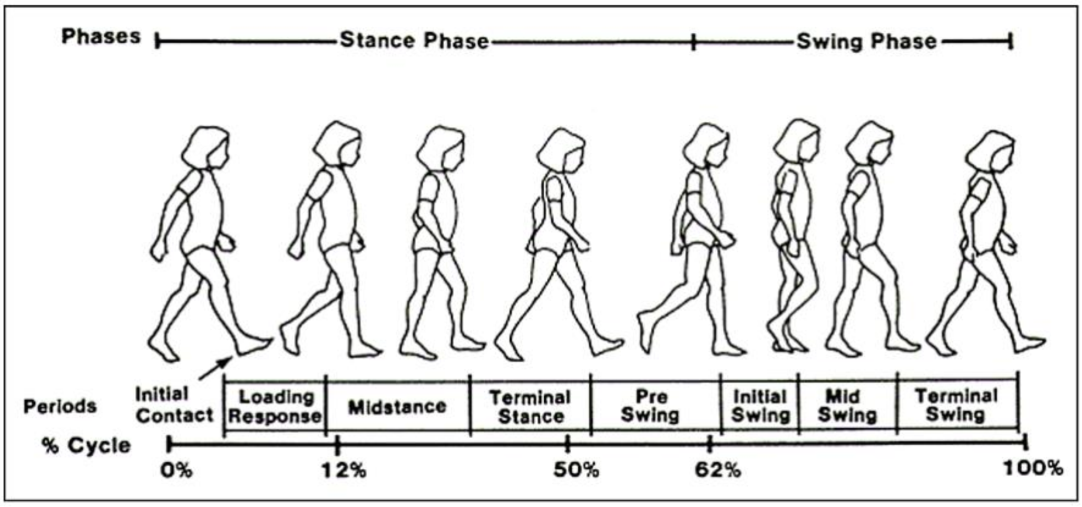

Figure 2: A complete gait cycle for the right leg
Reference: Perry, J. and Burnfield, J. (1992) Gait Analysis: Normal and Pathological Function. SLACK Incorporated, New Jersey.

The term *gait analysis* can be used for:

1. Observational gait analysis- watching someone walk in clinic with the naked eye
2. Video gait analysis- recording someone walking so it can be replayed in front view (coronal plane) and side view (sagittal plane)
3. Three-dimensional gait analysis (3DGA)- using specialised 3D cameras, force plates and anatomical markers. This offers the most accurate assessment giving objective kinematic, kinetic and temporal spatial data in the three anatomical planes at the same time.

Observational and video analyses are unable to capture detailed movement patterns within the foot due to its complex anatomical structure. Three-dimensional gait analysis offers potential to quantify foot and ankle movement patterns more accurately.

3-DIMENSIONAL GAIT ANALYSIS AND FOOT MODELLING

Three-dimensional gait analysis (3DGA) has been used widely to identify deformity in a variety of conditions including osteoarthritis (25), clubfoot (26), and cerebral palsy (27) through defining atypical movement patterns and assisting in management planning. However, the traditional lower limb kinematic models used in 3DGA represent the foot as a single rigid segment with just two degrees of freedom (Figure 3). This allows for measurement of whole foot dorsiflexion relative to the tibia and whole foot adduction relative to the tibia. Foot progression measures the position of the foot (internal/ external) relative to the gait laboratory.

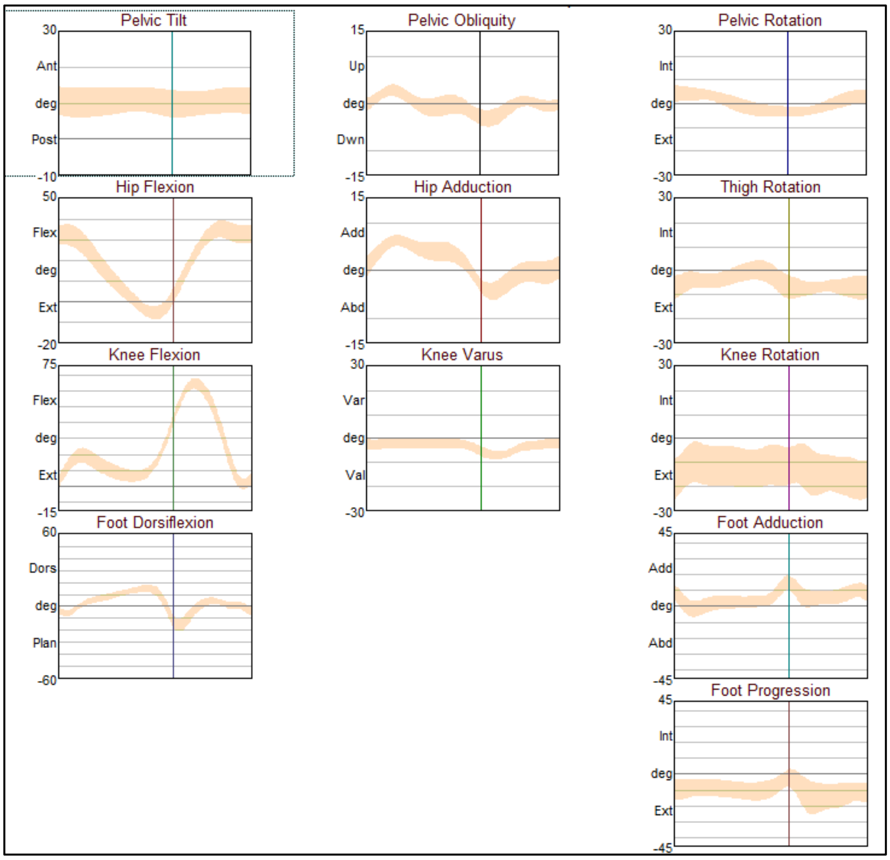

Figure 3: An example of normative lower limb kinematic data using a modification of the Helen Hayes (28) marker set obtained from the Oxford Gait Laboratory.

This two-dimensional analysis of the foot cannot measure the complex movement patterns of the foot and ankle that assist with shock absorption at initial contact, propulsion in terminal stance, and ground clearance during swing.

More recently, 3D multi-segment foot models have been developed to better reflect the complexity of foot motion during gait. The Oxford Foot Model (OFM) is a multi-segment, three-dimensional kinematic model that assesses dynamic motion of the foot (29). It was developed to measure tibia, hindfoot, forefoot and hallux motion in a clinical setting (Figure 4).

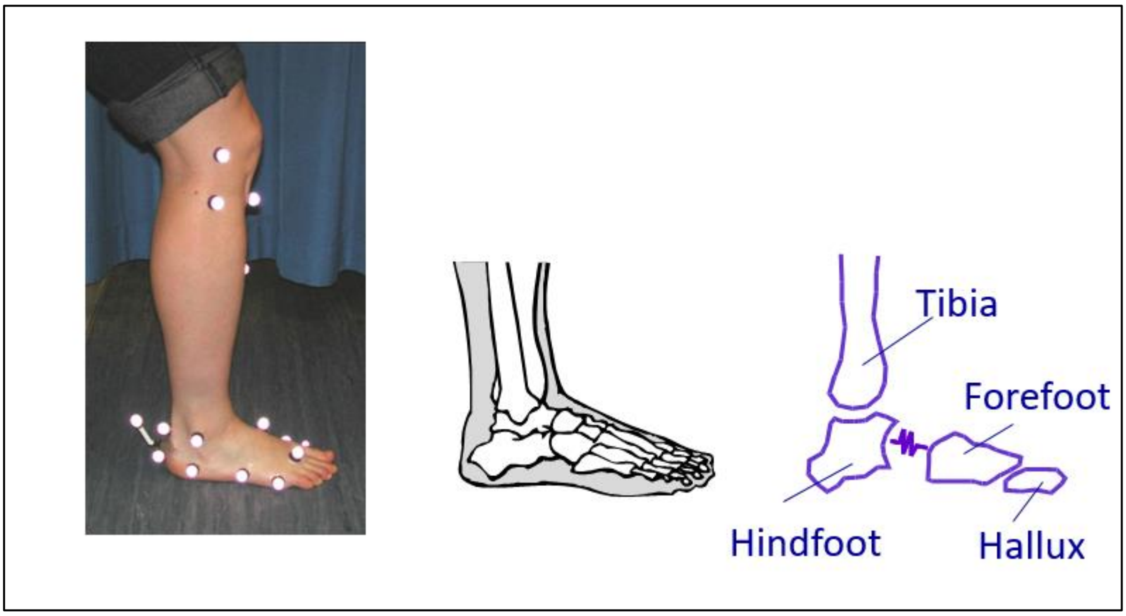

Figure 4: Oxford Foot Model (29) with markers and segment identification. Image used with permission of the author.

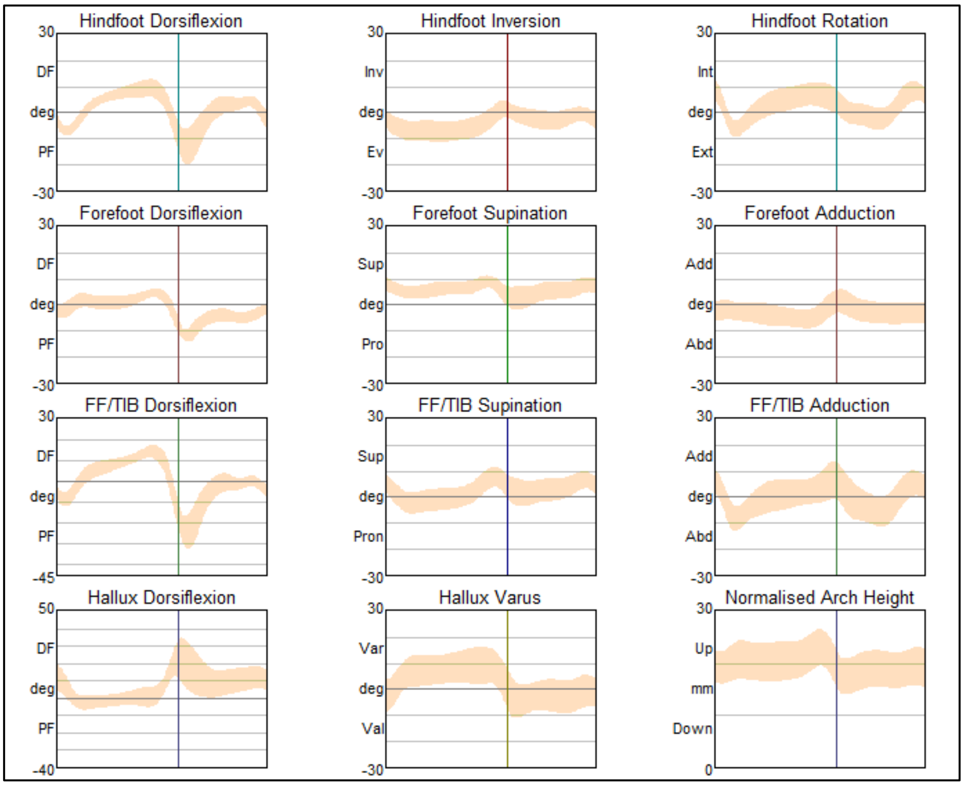

Figure 5: An example of normative kinematic data of the Oxford Foot Model obtained from the Oxford Gait Laboratory.

Figure 5 shows the kinematic output of the OFM including 12 kinematic analyses of movement patterns of the foot: the hindfoot relative to the forefoot in 3 planes, the forefoot relative to the tibia in 3 planes, and the hallux relative to the forefoot in 2 planes, and an arch height calculation.

Since the initial publication of the OFM by Carson and colleagues in 2001 (30), it has been utilised in clinical settings and research studies world-wide (cited 597 times in PubMed- online search 8.10.2021). Since its original publication in 2001, the OFM has undergone a second version (29) and had repeatability testing in healthy adult populations by groups outside of Oxford (31,32). Despite its popularity within the gait analysis industry, as a relatively recent development in the assessment of dynamic foot motion, rigorous clinimetric testing of the OFM is still lacking. This would further improve its clinical applications and research utility.

AIM of THESIS

The primary aim of this thesis was to establish the clinical role of the Oxford Foot Model to assess foot deformity during gait. To achieve this, we evaluated the OFM's clinimetric properties and created and tested a new outcome measure based on the OFM kinematics: the Foot Profile Score. A secondary aim was to explore a potential relationship between altered foot structure/ function during gait and perceived disability in a clinical population.

CHAPTER OUTLINES

The following chapters culminate to address the overall aims of this thesis.

Chapter II is a repeatability study to justify the use of the OFM in populations with known foot deformity. Intra and inter-rater repeatability of marker placement was assessed in children with clubfoot and in children with hemiplegia cerebral palsy and compared to a typically developing population. This study was completed at the Oxford Gait Laboratory in Oxford, UK.

Chapter III builds on previous research completed in Oxford evaluating the repeatability of the hindfoot marker in the OFM. Two jigs were created to improve the repeatability of the heel marker placement. Two gait analysts (one beginner and one experienced with the foot model) completed this repeatability study comparing the two jigs to the traditional method of eyeballing marker placement on 10 healthy adult subjects. This study was completed at VU Medical Centre in Amsterdam, Netherlands.

Chapter IV is a validation study of a new summary score of dynamic foot motion during gait based on the OFM kinematics- the Foot Profile Score (FPS). THE FPS was defined, then studied for its properties and design, and analysed against a clinical assessment of foot deformity. This study was completed at the Oxford Gait Laboratory in Oxford, UK.

Chapter V defines a minimal clinically important difference (MCID) for the Foot Profile Score based on the regression of the FPS on the Clinical Foot Deformity Scale (CFDS) presented in Chapter IV. The FPS was then assessed for its responsiveness in children with hemiplegia, cerebral palsy who underwent gait analysis pre-and post-surgery for correction of their foot deformities.

Chapter VI utilises the FPS to analyse recurrent foot deformities in children previously treated for clubfoot deformity, presenting at the Oxford Gait Laboratory for consideration of further treatment due to ongoing symptoms. Foot function was assessed by the OFM kinematics and plantar pressure and correlated with parent-reported outcome measures including the Oxford Ankle Foot Questionnaire, the Disease Specific Index for clubfoot and the Pediatric Quality of Life Inventory 4.0.

Chapter VII discusses the overall aim of this thesis and considerations for future research are given.

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Repeatability of the Oxford Foot Model in children with foot deformity

Jennifer McCahill

Julie Stebbins

Bart Koning

Jaap Harlaar

Tim Theologis

Gait & Posture. 2018 Mar;61:86-89.

ABSTRACT

INTRODUCTION: The Oxford Foot Model (OFM) is a multi-segment, kinematic model developed to assess foot motion. It has previously been assessed for repeatability in healthy populations. To determine the OFM's reliability for detecting foot deformity, it is important to know repeatability in pathological conditions. The aim of the study was to assess the repeatability of the OFM in children with foot deformity.

METHODS: Intra-tester repeatability was assessed for 45 children (15 typically developing, 15 hemiplegic, 15 clubfoot). Inter-tester repeatability was assessed in the clubfoot population. The mean absolute differences between testers (clubfoot) and sessions (clubfoot and hemiplegic) were calculated for each of 15 clinically relevant, kinematic variables and compared to typically developing children.

RESULTS: Children with clubfoot showed a mean difference between visits of 2.9° and a mean difference between raters of 3.6° Mean absolute differences were within one degree for the intra and inter-rater reliability in 12/15 variables. Hindfoot rotation, forefoot /tibia abduction and forefoot supination were the most variable between testers. Overall the clubfoot data were less variable than the typically developing population.

Children with hemiplegia demonstrated slightly higher differences between sessions (mean 4.1°), with the most reliable data in the sagittal plane, and largest differences in the transverse plane.

CONCLUSIONS: The OFM was designed to measure different types of foot deformity. The results of this study show that it provides repeatable results in children with foot deformity. To be distinguished from measurement artifact, changes in foot kinematics as a result of intervention or natural progression over time must be greater than the repeatability reported here.

INTRODUCTION

Foot deformities are prevalent in children and can be either congenital or acquired. Clubfoot is the most common congenital musculoskeletal deformity in children occurring in 1-2 out of 1000 live births [1]. It can result in foot and ankle stiffness, pain and arthritis which tend to increase over the lifespan [2]. Other examples of congenital foot deformities include vertical talus, cavus and metatarsus adductus. Flat foot deformity can be acquired, first becoming obvious as a child begins to walk. In general it is noted that the majority of toddlers have flat feet [3,4] which improves as they mature such that the adult prevalence is nearer 20% [5]. Acquired foot deformity is also very common in children with neurological problems such as cerebral palsy. Cerebral palsy (CP) is the most common motor disability in childhood with international prevalence estimates ranging from 1.5 to more than 4 per 1,000 live births [6]. At birth CP children's feet have normal postures, but over time the effects of their abnormal neurology leads to increasing lower limb deformity [7].

Three-dimensional gait analysis is an assessment tool to measure dynamic deformity in the lower limbs. It is widely used to identify lower limb deformity in children with clubfoot [8-14] and cerebral palsy [15,16] to assist in treatment planning. Traditionally the foot has been measured as a single rigid segment in a two-dimensional kinematic model. More recently, three-dimensional multi-segment foot models have been developed to improve our understanding of foot motion during gait. Fifteen foot models have been reported in the literature [17] with up to 9 segments being proposed [18]. Baker [18] reports 3 or 4 segment foot models are gaining preference for use in clinical gait analysis. Despite numerous foot models being available in the literature, very few are being used in centres outside of where they were developed [18].

The Oxford Foot Model (OFM) is a multi-segment, three-dimensional kinematic model that assesses dynamic motion of the foot [19]. It was developed to measure tibia, hindfoot, forefoot and hallux motion in a clinical setting. It can identify the presence of dynamic deformity compared to a healthy population, monitor change of an individual's foot posture over time, and measure change in foot motion before and after intervention. Published literature confirms the OFM is being used world-wide to evaluate various populations with foot deformity such as flat foot [20,21,22] clubfoot [23] and calcaneal fractures [24]. The OFM has already been shown to be repeatable in healthy populations (adults and children) for both intra-tester and inter- tester repeatability [19,25,26,27]; however, to date there is no published literature of its repeatability in pathological conditions.

The aim of this study was to assess the repeatability of the OFM in children with hemiplegic cerebral palsy and in children previously treated for clubfoot deformity, and compare it to a healthy population. Our hypothesis is that the repeatability of the OFM in children with foot deformity will be similar to previously reported values of the OFM's repeatability in healthy populations in the literature. For the purpose of this study, repeatability is defined as the difference between two repetitions of testing.

METHODS

Subjects- Typically Developing Fifteen typically developing children (mean age 9.5 years, range 6-14 years; 10 female and 5 male) were assessed with the OFM during level walking at self- selected velocity using a 12 camera Vicon 612 system (sampling at 100Hz) and 14mm passive markers. Each child was measured on two occasions by the same tester with the visits spaced between two and four weeks apart. The typically developing children were recruited from friends and colleagues of the Oxford Gait Laboratory.

Hemiplegia

Fifteen children with hemiplegic CP (mean age 10.2 years, range 6-15 years; 9 male, 6 female; 8 left side and 7 right side affected) were assessed with the OFM during level walking at self-selected velocity using a 12 camera Vicon 612 system (sampling at 100Hz) and 14mm passive markers. This was a convenience sample and we did not exclude any subjects on the basis of severity of foot deformity. The data was collected from routine clinical referrals- children referred to the gait laboratory for consideration of further management. The referrals were asking for clarification on orthotic management as well as potential surgical management for both populations- indicating a range of severity. Inclusion criteria were a confirmed diagnosis of hemiplegic cerebral palsy, presence of foot deformity on the affected side, appropriate level of co-operation and behaviour with no subjective reported deterioration or botulinum toxin/ surgery between visits. Each child was measured on two occasions by the same tester with visits spaced no more than six months apart as a part of their clinical pathway. Written, informed consent was obtained from subjects agreeing to participate in the project on the day of their first appointment in the gait laboratory.

Clubfoot

Fifteen children with clubfoot were assessed (mean age 8.8 years, range 4-14 years; 8 male, 7 female; 9 bilateral, 2 left, 4 right side affected). For the bilateral subjects- 1 side was randomly chosen resulting in 8 left and 7 right feet for analysis. OFM data were collected during level walking at self-selected velocity using a 16 camera Vicon MX/T-series system and 9.5mm passive markers. The subjects were chosen from consecutive routine clinical referrals- children referred to the gait laboratory for consideration of further management. The referrals were asking for clarification on orthotic management as well as potential surgical management indicating a range of foot deformity. We did not exclude any subjects on the basis of severity of foot deformity. Inclusion criteria were a confirmed structural idiopathic clubfoot deformity diagnosed at birth, no other musculoskeletal or neurological diagnoses, and the children and parents reported no change in symptoms between gait analysis visits.

Each child was measured on two occasions by the same tester, and once by a second tester. Written, informed consent was obtained prior to placing markers during their clinical visit to the gait laboratory. After clinical data collection was complete and the markers were removed by the primary marker placer, they had the markers replaced by the secondary placer for inter-rater repeatability with 6 new walking trials recorded. On a separate occasion, the child revisited the gait lab to complete 6 walks again with the primary marker placer (intra-rater data). On average the visits were 2.5 months a part (SD 1.9). Written, informed consent was obtained from subjects agreeing to participate in the project on the day of their first appointment in the gait laboratory.

Data Collection

The typically developing and hemiplegic groups were collected at the time when the Oxford Foot Model was being initially validated in 2002-2003 by a single tester with approximately 1 year of experience in placing OFM markers (JS). The clubfoot group was collected more recently (2013 - 2015) by someone with 7+ years experience with the OFM (JM) as the primary marker placer who put the markers on twice for each subject (intra-rater), and a third tester (JL) (3+ years experience with the OFM), who placed the markers once on each subject (inter-rater).

Data Processing

All data were processed for all populations by one of the authors (JS) who was also the tester during the initial phase of data collection (CP and TD groups). Three representative trials were chosen for analysis for each subject as the trials closest to the mean for that subject (ie with the lowest root mean square difference to the mean trace). The intra-tester repeatability was analysed for the hemiplegia, clubfoot and healthy populations, and the inter-tester repeatability was analysed for the clubfoot population. The data from both the hemiplegia and clubfoot populations were compared to the data of the typically developing children.

Fifteen clinically relevant kinematic variables (Table 1) were calculated and then averaged across the three trials. The mean absolute differences between sessions were calculated for each variable for all three populations, and as well as the mean absolute differences between raters for each variable for the clubfoot population.

We chose to report 15 kinematic variables which we deem to be clinically relevant when interpreting the Oxford Foot Model, and are consistent with the variables reported in Stebbins et al (2006). These incorporate all three anatomical planes and report on five variables each for hindfoot motion relative to the tibia, forefoot motion relative to the hindfoot, and forefoot motion relative to the tibia. In the sagittal plane we reported on range of dorsiflexion as well as maximum dorsiflexion achieved in stance and in swing. In the coronal and transverse planes we reported on average positioning of the segments due to less overall foot motion expected in these planes- with the position (ie. supinated/ abducted) being more clinically relevant.

RESULTS

Children with hemiplegic cerebral palsy were assessed for intra-rater repeatability across two sessions. The mean difference across all variables was 4.1° (Table 1). The largest difference was in hindfoot rotation (6.0°) and the smallest differences were seen in the sagittal plane.

Children previously treated for clubfoot deformity were tested for intra-rater repeatability with a mean difference between visits of 2.9°, with a range between 1.8 to 3.5° for the fifteen kinematic variables (Figure 1). There was no difference in variability between the sagittal, coronal and transverse planes for intra-rater repeatability.

Inter-rater repeatability in children previously treated for clubfoot deformity had a mean difference between raters of 3.6 degrees, with a range of 2.1 to 7.6 degrees for the 15 kinematic variables (Figure 1). Three outliers were above 4 degrees including average hindfoot rotation (transverse plane), forefoot /tibia abduction (transverse plane) and forefoot supination (coronal plane).

The mean absolute differences were within one degree for the intra and inter-rater reliability in 12/15 variables. Overall the clubfoot data was less variable than the healthy data.

TD – (intra)	**4.8 (2.2)**
Hemiplegia – (Intra)	**4.1 (2.2)**
Clubfoot- (Intra)	**2.9 (1.2)**
Clubfoot – (Inter)	**3.6 (2.0)**

Table 1: Mean absolute differences in degrees averaged across all the fifteen kinematic variables and their standard deviations.

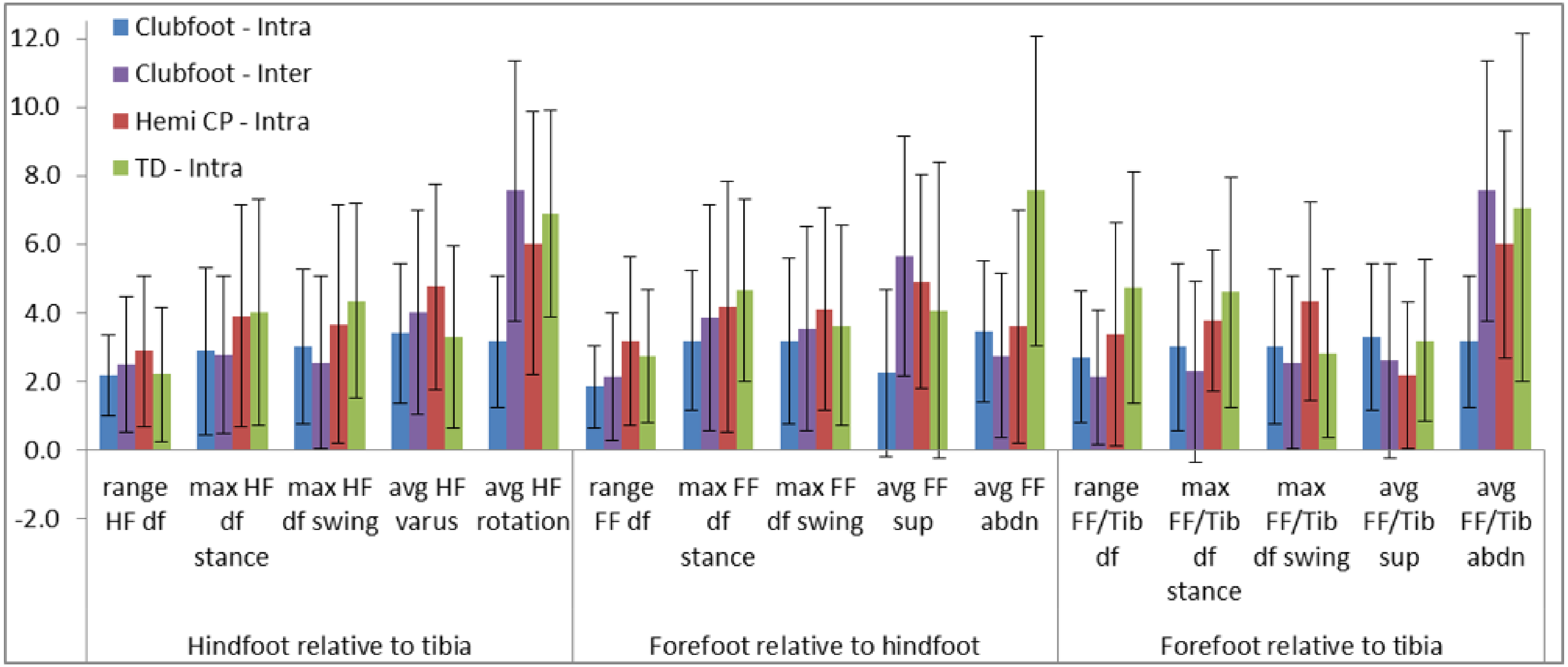

Figure 1: Inter and intra-rater repeatability for foot model kinematic variables. HF = hindfoot relative to tibia angles, FF = forefoot relative to hindfoot angles, FF/Tib = forefoot relative to tibia angles, df = dorsiflexion, avg=average.

DISCUSSION

This study shows the OFM has good intra-tester repeatability in typically developing children, as well as in children with cerebral palsy (hemiplegia) and in children with clubfoot deformity. The mean absolute difference in typically developing children was 4.8°, which improved to 4.1° in children with hemiplegia, and further improved to 2.9° in children with clubfoot. Overall the intra-tester variability of the clubfoot population was less than the hemiplegic and typically developing populations. Numerous factors could influence this including the clubfoot data being collected more recently, with a newer camera system including more cameras (16 instead of 12), and with smaller markers (9.5mm as opposed to 14mm). In addition, the experience of the marker placer may have contributed as the clubfoot population markers placed by the clinician with the most experience (ten years, compared to less than one year in the typically developing and CP populations).

In the clubfoot population, the inter-tester mean absolute differences across all fifteen kinematic variables was all less than 4°, with some variables being better than the intra-tester results; however there were three outliers from this trend. The first outlier was average hindfoot rotation. This measurement is dependent on the position of the heel marker which has been shown to have the most variability of the hindfoot markers [28]. Average forefoot relative to tibia abduction is directly affected by this measurement as it is the forefoot relative to the tibia in the transverse plane. The third outlier, average forefoot supination, is interesting for clubfoot as this is dependent on the placement of the distal first metatarsal marker which can be difficult in the presence of foot deformity when the forefoot cannot achieve a neutral position. Often residual clubfoot deformity does not allow the forefoot to rest flat on the floor during marker placement, either because of excessive supination or over-pronation. However it is worth noting that the intra-tester results were still very good for these three outliers which indicates that training, or the use of an aid to standardise marker placement, may improve these measurements further.

McGinley et al [29] offered guidelines for acceptable error of measurement following a systematic review of lower limb reliability studies in three-dimensional gait analysis. They state that when interpreting clinical gait analysis, error under two degrees is acceptable, error between two and five degrees is reasonable, and error over five degrees is concerning [29]. Based on these guidelines, this study has shown that the OFM has acceptable to reasonable reliability in typically developing children, as well as children with foot deformity. McGinley and colleagues [29] also identified that the highest reliability of lower limb kinematics was in the sagittal plane, and the lowest was in the transverse plane. This trend was not totally supported in our multi- segment foot kinematic data. In the clubfoot population, the heel marker placement contributed to lowest repeatability, but forefoot adduction/ abduction had reasonable repeatability.

The results of this study are comparable to other OFM repeatability studies in children in the published literature. Curtis et al [30] studied the repeatability of typically developing children with the OFM at discrete points in the gait cycle and found the typical errors of measurement ranged from 0.93-8.56 degrees for maximal, minimal and mean joint angles. Mahaffey et al [26] evaluated three different foot models concurrently, including the OFM, in typically developing children. The authors found the OFM demonstrated acceptable mean error over repeated sessions with a standard error of measurement of 4.61 +/-2.86 degrees [26]. This compared to 3DFoot [31] with a standard error of measurement of 3.88 +/-2.18 degrees [24]. However Kinfoot [32] had a higher standard error of measurement of 5.08 +/-1.53 degrees [26]. Deschamps et al [33] studied the repeatability of the model by Leardini et al [31] in adults with foot deformity. Their study showed the intra-rater variability was higher in the deformity population than the healthy population [33]. The authors attributed this to the subjective complaint of pain or fatigue during the testing sessions of the deformity population.

Our hypothesis was correct that our repeatability values for the typically developing population were similar to previously reported values [19, 25, 26, 27] however our study found that both the deformity populations (hemiplegia and clubfoot) had less variability than the typically developing population. Of particular note is the inter-tester repeatability in the clubfoot population was better than both the intra-tester repeatability in the TD and hemiplegic populations. This is reassuring as in a clinical gait analysis setting children often attend for comparison analysis with a different member of the team assessing them at each visit.

STUDY LIMITATIONS

As stated in the discussion, the main limitations of the study include a more experienced marker placer for the clubfoot data and the clubfoot data was collected more recently with an upgraded VICON system with more cameras and smaller markers. Both improved technology and clinician experience play an important role in improving repeatability. This is why the clubfoot data has the best reported repeatability, even better than the typically developing population. A third limitation is the potential for bias with the TD and hemiplegic populations being collected and processed by a single person (JS), during the course of development of the OFM. A final limitation to note is the difference between testing sessions for all three populations. This was due to assessing children in accordance with their clinical pathway or routine hospital visits- the authors ensured the inclusion criteria of no subjective or obvious clinical deterioration in symptoms was adhered to.

CONCLUSIONS

Three-dimensional gait analysis is widely used to guide the management of older children with congenital foot deformities such as clubfoot, and in children with acquired foot deformities such as flat foot and cerebral palsy. In order to measure the dynamic foot motion in detail, a multi-segment foot model must be used. The Oxford Foot Model was designed to be adaptable in its application to measure different types of foot deformity. In order to determine the reliability of the model for detecting foot deformity, it is important to know repeatability in pathological conditions. The results of this study show that the OFM provides repeatable results in healthy children, as well as in children with either congenital or acquired foot deformity. To be distinguished from measurement artifact, changes in foot kinematics as a result of intervention or as natural progression over time must be greater than the repeatability reported here.

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Reliability testing of the heel marker in three-dimensional gait analysis

Jennifer McCahill

Wouter Schallig

Julie Stebbins

Robin Prescott

Tim Theologis

Jaap Harlaar

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ABSTRACT

INTRODUCTION: In three-dimensional gait analysis, anatomical axes are defined by and therefore sensitive to marker placement. Previous analysis of the Oxford Foot Model (OFM) has suggested that the axes of the hindfoot are most sensitive to marker placement on the posterior aspect of the heel. Since other multi-segment foot models also use a similar marker, it is important to find methods to place this as accurately as possible.

The aim of this pilot study was to test two different 'jigs' (anatomical alignment devices) against eyeball marker placement to improve reliability of heel marker placement and calculation of hindfoot angles using the OFM.

METHODS: Two jigs were designed using three-dimensional printing: a ratio caliper and heel mould. OFM kinematics were collected for ten healthy adults; intra-tester and inter-tester repeatability of hindfoot marker placement were assessed using both an experienced and inexperienced gait analyst for 5 clinically relevant variables.

RESULTS: For 3 out of 5 variables the intra-tester and inter-tester variability was below 2 degrees for all methods of marker placement. The ratio caliper had the lowest intra-tester variability for the experienced gait analyst in all 5 variables and for the inexperienced gait analyst in 4 out of 5 variables. However for inter-tester variability, the ratio caliper was only lower than the eyeball method in 2 out of the 5 variables. The mould produced the worst results for 3 of the 5 variables, and was

particularly prone to variability when assessing average hindfoot rotation, making it the least reliable method overall.

CONCLUSIONS : Overall there was low intra-tester and inter-tester variability suggesting good sensitivity of the OFM to detect meaningful clinical differences. The use of the ratio caliper may improve intra-tester variability, but does not seem superior to the eyeball method of marker placement for inter-tester variability. The use of a heel mould is discouraged.

INTRODUCTION

In three-dimensional gait analysis, anatomical axes are defined by and therefore sensitive to marker placement (1). In most kinematic multi-segment foot models, the posterior heel marker is used to help define the hindfoot segment by placing the marker centrally on the posterior aspect of the calcaneus. For the Oxford Foot Model (OFM) hindfoot segment, the heel (HEE), proximal heel (PCA), lateral calcaneus (LCA) and sustentaculum tali (STL) markers are used to define the axes of the calcaneus (2).

Previous research has shown that misplacement of the calcaneal markers has a profound effect on the kinematic output (3-5). Paik and colleagues used radiopaque monitoring electrodes placed on the feet at the locations specified by the OFM and CT images to investigate how changes in marker placement affect the orientation of the OFM hindfoot segment axes (5). Their results suggest changing the anterio-posterior position of either the LCA or the STL marker by 1mm induced 0.2° of change in the anterior-posterior (A-P) axis. Whereas, when the HEE marker position was moved in mediolateral direction by 1mm, it induced 4° of change in the orientation of the A-P axis (5). Since the orientation of the A-P axis is more sensitive to the location of the HEE marker than to the locations of the LCA and STL markers, it is essential to ensure that the HEE marker is placed accurately.

Intra-tester and inter-tester repeatability of hindfoot marker placement has been shown to be improved when using an alignment device or jig to assist in marker placement, compared to using a manual palpation/ eyeball method; however both jigs were designed to align the medial and lateral calcaneal markers, and not the central heel marker (4,6).

After reviewing the available alignment devices in the literature, and using the authors' expert experience with foot anatomy and the OFM, we designed two jigs that could potentially improve the repeatability of HEE marker placement: a ratio caliper and mould. The aim of this pilot study was to test these two jigs against the conventional method of eyeball marker placement to improve marker placement repeatability of the HEE marker when using the OFM. We hypothesized that the ratio caliper and mould would not improve an experienced gait analyst's repeatability, but that they would improve an inexperienced gait analyst's repeatability as well as the inter-tester error.

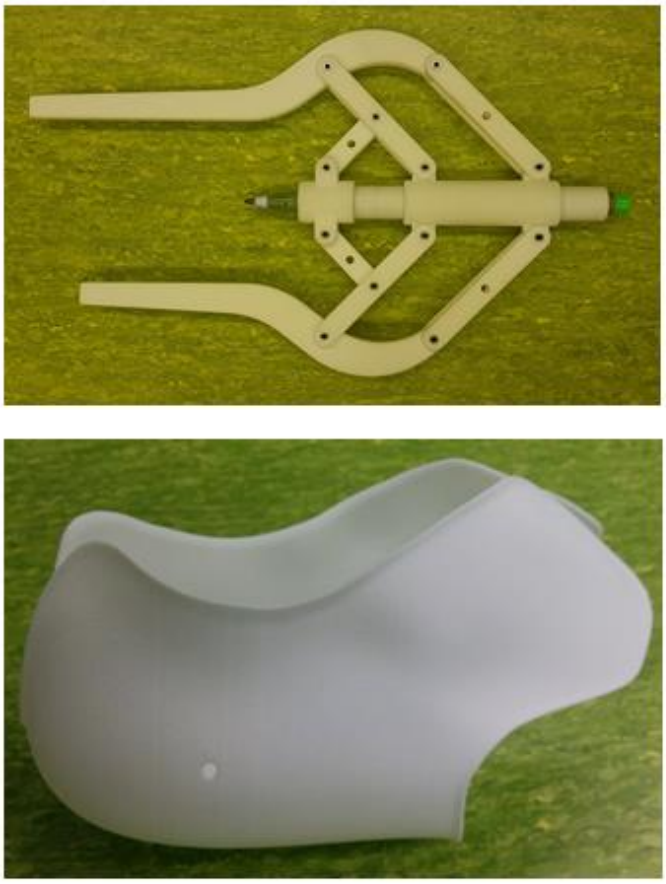
METHODS

2.1 Specifications of two jigs

Two jigs were constructed using three-dimensional printing to specifications designed by the authors.

a) Ratio caliper: The longer fixed arm was placed on the lateral border of the foot to the base of the 5th metatarsal- while the shorter moving arm was brought in to the medial hindfoot. A mid-point at 50% between the 2 arms determined the midline of the calcaneus where the HEE marker was placed. The caliper was used with the subject in weight bearing. (Picture 1a).

b) Heel mould: The foot shape for the mould was determined by a 3D light scan of the skin surface of a female with asymptomatic feet and an EU shoe size of 36. The mould was scaled to three different sizes to accommodate different shoe size ranges (small, medium, large) and 3D printed. The mould had a central hole in the middle of the calcaneus to mark with pen so the heel marker could be placed over the mark upon removal of the mould. It also had holes for the LCA and STL markers to be placed over as well. The mould was placed on the foot with the subject in a seated position (Picture 1b).



Picture 1: a) ratio caliper b) heel mould

2.2 Definition of eyeball method

Heel marker placement for the OFM uses an eyeball technique with manual palpation to place the heel marker in the middle of the calcaneus at the same height above the plantar surface of the foot as the TOE marker (between the heads of the 2nd and 3rd metatarsals).

2.3 Repeatability testing

Ten healthy adult subjects (6 female, age: 26.8 (SD 2.6) years, height: 176.4 (8.1), weight: 67.2 (8.5) with a normal foot posture index (2.4 (1.4)) were recruited for this study (7). The subjects did not have any foot or ankle complaints, did not wear insoles, and did not have any concerns that would affect their gait pattern. Informed consent was obtained for all subjects and ethical approval was provided by the local ethics committee. All subjects were assessed during level walking at self-selected velocity using a 12 camera motion capture system (Vicon Motion Systems Ltd., Oxford, UK) (sampling at 100Hz) and 9.5mm passive markers with 9.5 mm diameter bases were placed by two different gait analysts for OFM kinematics: an experienced analyst (over 10 years) and an inexperienced analyst (less than 6 months) experience with the OFM.

Each subject attended the gait laboratory for one visit. The experienced gait analyst put all the lower limb and OFM markers on initially using the eyeball method. All markers except for the calcaneal (hindfoot) markers stayed in place for the rest of the session. In order to not bias the placement of the HEE marker, all of the calcaneal markers were replaced each session. The HEE, CPG, PCA, STL, LCA markers (2) were replaced for the additional walking trials so both gait analysts used the eyeball, the ratio caliper, and the mould methods of marker placement for two walking sessions each (12 sessions for each subject in total). Within each session, five walking trials were recorded, and three walks (three strides) were averaged for data analysis.

All data were processed by the same person with the OFM pipeline implemented in Vicon Nexus (v2.9.3), in which the hindfoot flat option was not checked. The data were analysed using five clinically relevant variables of hindfoot motion during the gait cycle: maximum hindfoot dorsiflexion in stance, maximum hindfoot dorsiflexion in swing, range of hindfoot motion in the sagittal plane, average hindfoot varus, and average hindfoot rotation. Inter-tester repeatability was taken from the first marker application for both the gait analysts.

Initially, statistical parametric mapping was used to demonstrate an absence of significant order effects within raters and an absence of a systematic difference between testers, evaluated over the full gait cycle. Subsequent analysis was applied to each of the five derived variables, in combination with each of the three methods of marker placement. A series of Bland-Altman plots were produced, one for each tester and for between the first of the tester assessments, none of which showed that differences varied with the magnitude of the observations. The standard deviations (SDs) for within tester differences and between tester differences were calculated from the root mean square of the differences. We also report variance components for each intra-tester component and inter-tester component for all combinations of variable and marker method, as suggested by Chia and Sangeux (8) using restricted maximum likelihood. A pooled estimate of the intra- and inter-tester components for each of the three marker methods was obtained using the mean of the five separate estimates.

RESULTS

The SDs from the intra- and inter-tester differences are shown in Figure 1. These show similar, low intra-tester differences for both gait analysts and inter-tester differences in the sagittal plane variables for all three methods of marker placement with the majority of differences under 2.0 degrees, and all differences under 2.5 degrees.

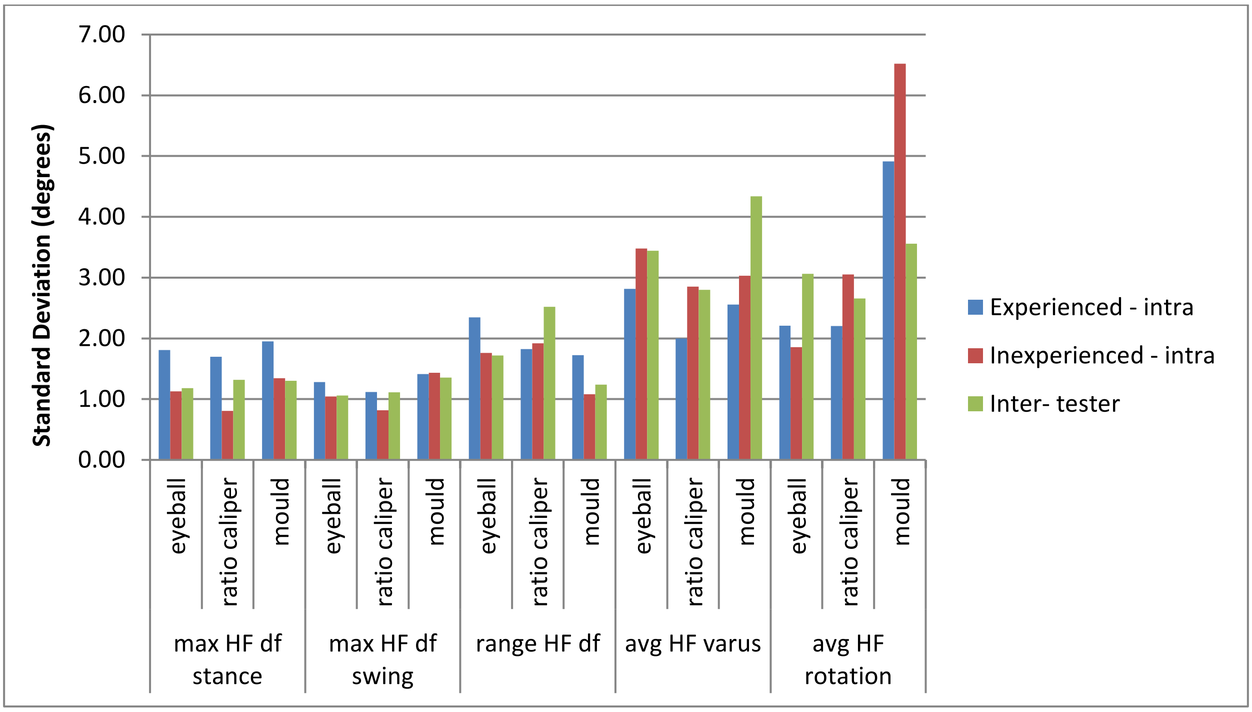


Figure 1: Standard deviations (N=10) in degrees of both intra-tester (experienced and inexperienced gait analysts) and inter-tester differences max= maximum, HF=hindfoot, df=dorsiflexion, avg=average

Compared to the eyeball method, the ratio caliper and the mould reduced intra-tester variability for both gait analysts for average hindfoot varus. However the mould had the highest inter-tester variability in the coronal plane compared to the other clinical variables. Average hindfoot rotation showed the highest intra-tester variability for both gait analysts, and the second highest inter-tester variability when using the mould compared to the other two methods. These findings were reinforced by the variance components, shown in Supplementary Material: Table 1. For five of the 15 combinations of variable and method, the estimate of the inter-rater component was zero.

DISCUSSION

Overall our results show that in a healthy population, the ratio caliper method of marker placement produced the lowest intra-tester variability for the experienced gait analyst in all five variables and for the inexperienced gait analyst in four out of five variables (all but the transverse plane). However for inter-tester variability, the ratio caliper was only lower than the eyeball method in two out of the five variables. The mould produced the worst results for 3 of the 5 variables, and was particularly prone to variability when assessing average hindfoot rotation, making it the least reliable method overall. Therefore we can only partly accept our hypotheses.

The concept behind the heel mould was that it serves as a morphological template of the hindfoot. It's relatively poor results might be due to its maneuverability on the subject's foot. Despite having it available in three different sizes, there was medial/lateral play of the mould on the hindfoot when placing it on the subject in non-weight bearing which would have affected its repeatability. This was evident from outliers in the raw data using this method of marker placement causing occasional very wide deviations which affects the estimates of reliability. These outliers were examined and found to be true values for both gait analysts. However, even without these outlying observations, variability was still greater with this method.

Our analysis showed zero inter-rater variance components for 5 of the 15 combinations of variable and method. When these components are zero or very small, chance variation can result in inter-tester reliability being paradoxically better than intra-tester variability, as we saw in Figure 1. Although surprising, we have also found this trend when analyzing the inter-tester repeatability of OFM marker placement in children with clubfoot, using an experienced and inexperienced gait analyst as well (9).

It is common practice to place the OFM using the eyeball method with palpation for marker placement for all segments. The Heidelberg foot measurement model uses a heel alignment device to place the medial and lateral calcaneal markers (6). The authors describe its use in a non-weight bearing position with the main axis extending from the heel to the toe marker and the secondary axis aligned with the Achilles tendon. This may be appropriate in healthy populations, but in foot deformity the Achilles tendon is often mal-aligned in relation to the calcaneus; a common clinical picture of ‘escape valgus’. Like Deschamps and colleagues (4), we believe the hindfoot markers should be placed in weight bearing, therefore using devices such as the mould or the

Heidelberg heel alignment device in non-weight bearing, may negatively affect the marker placement and therefore axis definition of a three-dimensional foot model. Since the ratio caliper is used in weight-bearing and seems useful in improving repeatability of the central HEE marker placement in this study, the use of this jig warrants further testing.

The inexperienced analyst was generally less repeatable compared to the experienced analyst for hindfoot varus and rotation. This could be due to reduced knowledge of anatomy during marker placement of the HEE and PCA markers. It was surprising to the authors that the inexperienced gait analyst was the most repeatable with the eyeball method. This does reinforce its original design for marker placement and suggest the eyeball method can be used reliably with only six months of experience. Our data also suggests that a jig may not improve the repeatability for an inexperienced analyst. Maybe the task of placing a jig on a foot further complicates the task of marker placement for inexperienced analysts.

We recognise this study only included a healthy adult population, a small sample size and only two raters. We would recommend this study be repeated comparing the eyeball method to the ratio caliper, with more gait analysts placing markers, a larger sample size including adults and children, with a range of foot postures. A population with foot deformity may yield different results due to difficulties with marker placement in abnormal standing anatomical alignment (9).

Despite it not being an aim of our study, the authors feel it's important to note our intra-tester and inter-tester variability was quite low (mostly under 3 degrees for all methods tested) suggesting good sensitivity to detect clinically meaningful differences, and lower than other published studies (10).

CONCLUSIONS

In a healthy adult population, the ratio caliper improved the intra-tester repeatability of hindfoot marker placement for an experienced and an inexperienced gait analyst. However, both ratio caliper and eyeballing yielded good inter-tester repeatability.

SUPPLEMENTARY MATERIAL

Variable	Method	Intra-experienced (degrees)	Intra-inexperienced (degrees)	Inter-tester (degrees)
Max HF df stance	Eyeball	1.17	0.54	0^{+}
	Ratio Caliper	1.44	0.33	0.30
	Mould	1.73	0.86	0^{+}
Max HF df swing	Eyeball	0.82	0.54	0.01
	Ratio Caliper	0.63	0.33	0.24
	Mould	0.95	0.98	0^{+}
Range HF df	Eyeball	2.16	1.32	0^{+}
	Ratio Caliper	1.67	1.84	1.13
	Mould	1.48	0.59	0.36
Avg HF varus	Eyeball	3.97	6.06	0.68
	Ratio Caliper	1.99	4.07	0.98
	Mould	3.27	4.59	0.99
Avg HF rotation	Eyeball	2.43	1.72	4.79
	Ratio Caliper	2.43	4.65	0.03
	Mould	9.8	15.53	0^{+}
Pooled*	Eyeball	2.32	2.10	1.48
	Ratio Caliper	1.63	2.24	0.54
	Mould	3.95	5.68	0^{+}

Table 1: Variance component estimates by variable and marker placement method
max= maximum, HF=hindfoot, df=dorsiflexion, avg=average

∗ Pooled estimates were obtained allowing individual negative variances and so do not agree exactly with the means of the individual estimates in the table.
$^{+}$ Negative estimate constrained to be zero.

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Validation of the Foot Profile Score

Jennifer McCahill

Julie Stebbins

Andrew Lewis

Robin Prescott

Jaap Harlaar

Tim Theologis

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ABSTRACT

BACKGROUND: There are numerous static measures of foot posture but there is no published score of dynamic foot motion. Three-dimensional gait analysis can include a multi-segment foot model like the Oxford Foot Model (OFM) to comprehensively quantify foot kinematic deviations across the gait cycle but it lacks an overall score, like the Gait Profile score (GPS), used to summarize the quality of lower extremity motion.

RESEARCH QUESTION: This paper introduces the Foot Profile Score (FPS), a single number, analogous to the GPS based on kinematic data of the OFM. The aim of this study is to validate the FPS by studying its properties and design, and analyse it against a clinical assessment of foot deformity.

METHODS: Concurrent validity was established for the FPS analysing the relationship with Clinical Foot Deformity Score (CFDS) in 60 subjects with a condition affecting the lower limbs globally. Content validity was established for the six Foot Variable Scores (FVS) that make up the FPS using a multiple regression of the CFDS on the 6 FVS in the 60 subjects. Predictive validity was established analysing the relationship of the FPS and GPS comparing 60 global involvement subjects with 60 subjects with isolated foot deformity.

RESULTS: Pearson correlation between the FPS and CFDS was significant at 0.62 ($p < 0.001$). Each element of FVS contributes positively to predicting the CFDS with $R2=0.456$ ($p < 0.001$). FPS contributed independently to the prediction of CFDS ($t=3.9$, $p < 0.001$). The correlation between the GPS and FPS in the global involvement group was significant at $r=0.64$ ($p < 0.001$), while there was no correlation found with $r=0.08$ ($p=0.54$) in the foot deformity group.

SIGNIFICANCE: The FPS is the first validated score of dynamic foot motion.

1. INTRODUCTION

Measuring foot deformity in a clinical or research setting has always been challenging due to numerous factors. Some available measures are condition specific, such as the Pirani Score for clubfoot [1] or measure only one element of a deformity, such as the Arch Height Index [2] or hindfoot valgus. A recent systematic review of the measurement of paediatric flat foot by Banwell and colleagues found four groupings of available assessments- plain film radiographs, foot-print indices, static foot measures, and plantar pressure analysis [3]. The authors found that all groups were based on static analysis of foot postures. The authors concluded that dynamic measurement of foot motion is needed to improve our understanding of foot structure [3].

Three-dimensional (3D) gait analysis is a tool to measure dynamic motion in the lower limbs. It is widely used to identify deformity in a variety of conditions including osteoarthritis [4], clubfoot [5] and cerebral palsy [6] to assist in treatment planning. Traditionally the foot has been represented as a single rigid segment with just two degrees of freedom. More recently, 3D multi-segment foot models have been developed to improve our understanding of foot motion during gait.

The Oxford Foot Model (OFM) was developed as a multi-segment, 3D kinematic model that assesses dynamic motion of the foot [7]. It was developed to measure tibia, hindfoot, forefoot, and hallux motion in a clinical setting. It can identify the presence of deformity compared to a healthy population, monitor change of foot posture over time, and measure change in ankle and foot motion before and after intervention [7]. Published literature confirms the OFM is being used clinically and in research settings world-wide to evaluate populations with foot deformity [8-11]. The OFM has been shown to be repeatable in healthy populations (adults and children) for both intra-tester and inter-tester repeatability [7,12,13]; and children with foot deformity including clubfoot and cerebral palsy [14]. The OFM is a comprehensive measure of foot/ ankle motion with each segment being measured in 3D. With the large amount of data available, it can be difficult to quantify as an outcome measure. Therefore, an overall score of foot motion using the kinematic data from the OFM would be beneficial.

The Gait Profile Score (GPS) was developed to provide a single measurement of the quality of an individual's gait pattern based on lower limb kinematics [15]. The GPS is calculated as the root mean square average of 9 key kinematic variable scores (Gait Variable Scores- GVS), each calculated as the root mean square difference between a patient's data and normative data for both legs [15]. Since its creation, the GPS has been widely used in clinical and research settings. However, the GPS includes the traditional measurement of the foot as a single segment. An additional score representing detailed foot and ankle motion may therefore be beneficial, particularly in patients with foot deformity as the predominant pathology.

This paper introduces the Foot Profile Score (FPS), a single measurement of dynamic foot motion, constructed similarly to the GPS, but based on OFM kinematics. The aim of this study is to validate the FPS by evaluating its inherent properties and design, and analyse it against a global clinical assessment of foot deformity.

2. METHODS

2.1. Construction of the foot variable scores, the foot profile score, and the foot movement analysis profile

The Foot Variable Scores (FVS), the Foot Profile Score (FPS), and the Foot Movement Analysis Profile (F-MAP) were calculated using the same formula as the construction of the GPS [15] but using 6 key kinematic variables from the Oxford Foot Model for both the right and left legs.

Thus if $x_{i,t}$ is the value of foot variable i calculated at a specific point in the gait cycle t, and $\bar{x}_{i,t}^{ref}$ is the mean value of that variable at the same point in the gait cycle for the reference population then the i^{th} foot variable score (FVS) is given by:

$$FVS_i = \sqrt{\frac{1}{T}\sum_{t=1}^{T} (x_{i,t} - \bar{x}_{i,t}^{ref})^2}$$

where T is the number of instants into which the gait cycle has been divided. The FPS is then the RMS average of the FVS variables:

$$FPS = \sqrt{\frac{1}{N}\sum_{i=1}^{N} FVS_i^2}$$

where N is the number of FVS variables used, in this case 6 (hind foot dorsiflexion, forefoot dorsiflexion, hind foot inversion, forefoot supination, hind foot rotation, forefoot adduction)

The 6 FVS represent the motion of the hindfoot relative to the tibia in the sagittal, coronal and transverse planes, as well as the motion of the forefoot relative to the hindfoot in the sagittal, coronal and transverse planes. The more the foot deviates from the reference data, the higher the FPS. The FVS and the FPS do not reflect the direction of the deviation (e.g. plantarflexion or dorsiflexion). The F-MAP is a bar chart of the 6 FVS for each foot and the FPS to provide a visual representation of where a subject deviates from the normative data (Figure 1).

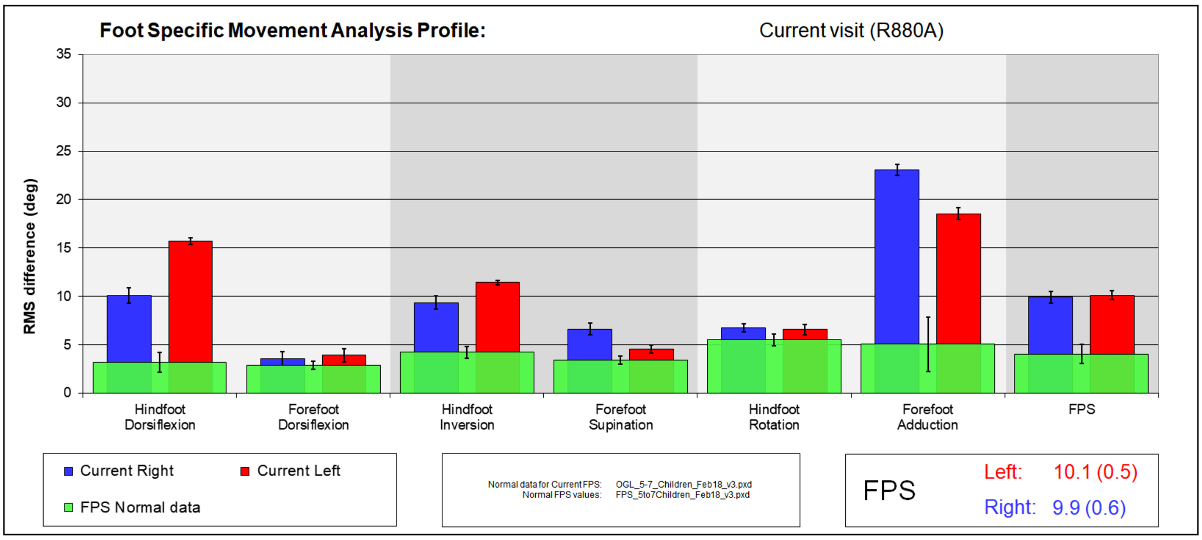

Figure 1: An example of a Foot Movement Analysis Profile (F-MAP)

2.2 Validation of the Foot Profile Score

As with the GPS, the FPS already has high face validity as it is based on the kinematic data of the OFM. The repeatability of the FPS is also inherent as the marker placement of the OFM has been extensively studied with good results [7,12, 13,14]. Therefore, the formal validation process included analysing concurrent validity, content validity, and predictive validity.

<u>Concurrent validity</u>

There is no published dynamic foot deformity scale to which it is appropriate to correlate the FPS [3]. In the absence of this- we created a clinical rating scale of foot deformity to use in the validation process. We sent sagittal and coronal close-up foot videos of 60 subjects to 10 gait analysts affiliated with a 3D gait laboratory from 4 countries (5 physiotherapists, 2 orthopaedic surgeons, 2 clinical scientists/ engineers and 1 paediatric physiatrist). Each subject was scored by 5 gait analysts. The subjects included a range of demographics and severity of foot deformity. We made sure to represent the full range of deformities, varying from planovalgus to cavo-varus foot deformities. There were 30 children and 30 adults; 36 males and 24 females. 23 Subjects had orthopaedic diagnoses, 21 had cerebral palsy and 16 had neurological diagnoses. For each of the 60 subjects, the gait analysts scored both feet separately. We used right leg data in 31 subjects/ and left leg data in 29 subjects. There were no markers on the feet in the videos. We asked the gait analysts to rate the overall appearance of the foot using a scale from 0-3, which we termed the Clinical Foot Deformity Scale (CFDS: 0=normal, 1=mild, 2=moderate, 3=severe foot deformity) with no further instructions. All 60 subjects had OFM kinematics [7] collected using a Vicon T-series motion capture system (Vicon Motion Systems Ltd.) including 16 cameras collecting at 100Hz. Subjects walked at self-selected speed over level ground for both the video and motion capture trials.

The CFDS was taken as the mean of all 5 gait analysts' ratings for each subject. The FPS was calculated by the average root mean square difference between a patient's data and normative data taken over 6 key kinematic graphs for the same leg as used for the CFDS scoring for each subject. Pearson correlation coefficient was used to analyse the relationship between FPS and CFDS as a measure of concurrent validity.

We hypothesised that FPS would correlate moderately with CFDS as FPS also contains transverse plane information not easily visible in a clinical assessment.

Content validity

To analyse the FVS (sagittal plane- hindfoot dorsiflexion and forefoot dorsiflexion; coronal plane- hindfoot inversion and forefoot supination; transverse plane- hindfoot internal rotation and forefoot adduction) - we looked at a multiple regression of the CFDS on the 6 FVS for the above mentioned 60 subjects.

We hypothesised that the 6 FVS chosen would contribute positively to CFDS.

Predictive validity

We analysed the relationship between FPS and GPS to evaluate if the measurement of foot deviation during gait provides additional information to the measurement of the overall gait pattern. We collected 2 groups for the analysis: the above mentioned 60 subjects who had predominantly global involvement (deviations at more than one joint including proximal involvement) and a group with isolated foot deformity. The foot deformity group consisted of children with clubfoot aged 5-16 (mean 10 years), with 45 males/ 15 females and 39 right legs/ 21 left legs analysed. For children with bilateral clubfoot we analysed their more involved foot (the foot with the higher FPS). All subjects had a conventional lower limb model [28] and OFM kinematics collected using a 16 camera Vicon T-series motion capture system (Vicon Motion Systems Ltd.). GPS and FPS were calculated for both groups. Pearson correlation coefficient was used to analyse the relationship between bilateral GPS and unilateral FPS.

Additionally, to consider the correlation coefficients in the groups, we also report the ratio of the variances for FPS and GPS. As GPS and FPS are likely to be correlated, 95% confidence intervals for the ratio in each population can be calculated using a method based on the Pitman-Morgan test [16,17] illustrated by Snedecor and Cochran [18].

We hypothesised that FPS will give new information not offered by GPS and therefore the correlation of GPS and FPS will be higher in the global involvement group than the foot deformity group. Additionally, the ratio of variances (FPS:GPS) should be higher in the foot deformity group than the global involvement group.

RESULTS

Concurrent validity

The mean CFDS scores for each pathological group were cerebral palsy 1.5 (SD 1.0), orthopaedic 1.1 (SD 1.1) and neurological 1.9 (SD 1.0) indicating a range of deformity within each group, and the amount of deformity was consistent across groups. The mean FPS score for all 60 subjects was 8.6 degrees (SD 3.0) indicating a range of deformity within the group (normal FPS is 4.1 degrees (SD 0.8). The Pearson correlation between FPS and CFDS was significant at 0.66 with p<0.001 (Figure 2).

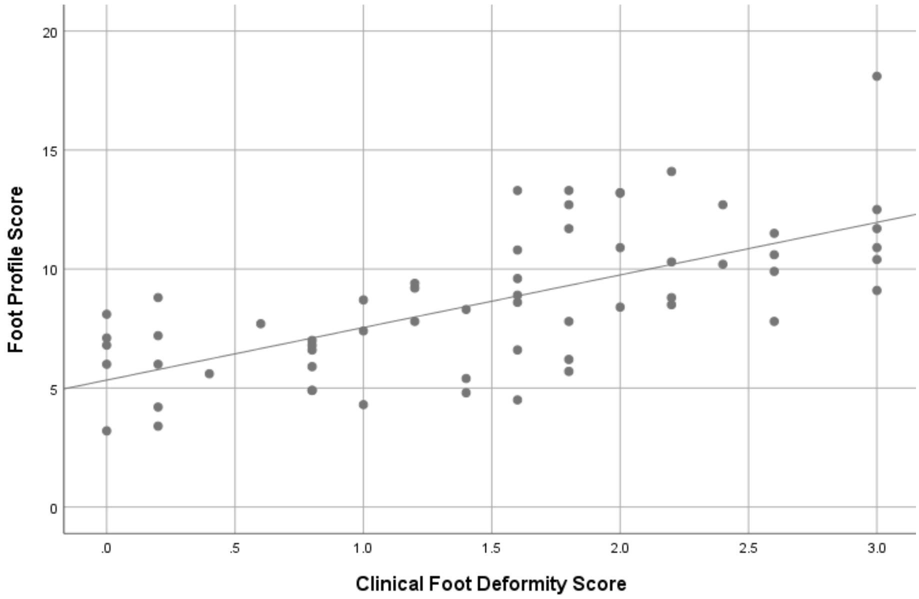

Figure 2- Scatterplot of the Foot Profile Score and the Clinical Foot Deformity Score

<u>Content validity</u>

Table 1 shows the multiple regression of CFDS on the 6 component scores of the FPS. For comparison, the corresponding regression with FPS as predictor yields identical coefficients for each component of 0.033 with standard errors of 0.005. Thus, although they differ in individual statistical significance, we see that the regression coefficients in Table 1 are all within one standard error of the combined regression coefficient. Furthermore, the residual standard deviation is marginally smaller when fitting with FPS as predictor (0.69) than using the 6 independent terms (0.71). This is confirmed using analysis of variance where the reduction in the sum of squares from fitting 5 additional coefficients is non-significant ($F(5,53)=0.34$, $p=0.89$).

This confirms that the FPS, which assigns equal weight to each constituent component, performs better than a model with separate weights for each component and that each component is contributing positively to predicting the clinical scores.

	Regression Coefficient	Standard Error	p
(Constant)	*-0.099*	*0.301*	
Hindfoot sagittal	0.040	0.016	0.014
Forefoot sagittal	0.033	0.029	0.255
Hindfoot coronal	0.052	0.021	0.019
Forefoot coronal	0.019	0.026	0.462
Hindfoot transverse	0.019	0.021	0.377
Forefoot Transverse	0.024	0.014	0.095

Table 1- Regression of the CFDS on the 6 component scores of the FPS ($R2 = 0.456$ ($p<0.001$))

We have already shown the significant correlation between CFDS and FPS, but due to the way the GPS is constructed, we note there is also a significant correlation of CFDS with GPS ($r=0.63$, $p<0.001$). It is therefore necessary to show that the association between FPS and CFDS is not a simple consequence of the mutual association with GPS. Table 2 shows that when analyzing the regression of the CFDS on the GPS and FPS, the

FPS is contributing independently to the prediction of the CFDS, ($t=4.3$, $p<0.001$).

	Regression Coefficient	Standard Error	t	p
(Constant)	*-0.686*	*0.277*		
GPS	0.120	0.033	3.6	0.001
FPS	0.138	0.032	4.3	<0.001

Table 2- Regression of the CFDS on the FPS and GPS

Predictive validity

The global involvement group had a mean GPS of 8.3 degrees (SD 2.9) and a mean FPS of 8.6 degrees (SD 3.0) indicating that at group level both distal and proximal joints contributed to gait abnormalities. The foot deformity group had a mean GPS of 5.7 (SD 1.3) and a mean FPS of 8.0 (SD 2.9) indicating foot specific problems in this group. The Pearson correlation between GPS and FPS in the global involvement group was significant at $r = 0.68$ with $p<0.001$ (Figure 3). The correlation between GPS and FPS in the foot deformity group was much lower at $r = 0.27$ with $p=0.04$ (Figure 4).

The ratio of the variances for FPS and GPS in the global involvement group was 1.07 (95% CI 0.69, 1.67), while in the foot deformity group it was 4.91 (95% CI 2.97, 8.10). With a wide difference between their confidence intervals, predictive validity of the FPS was established.

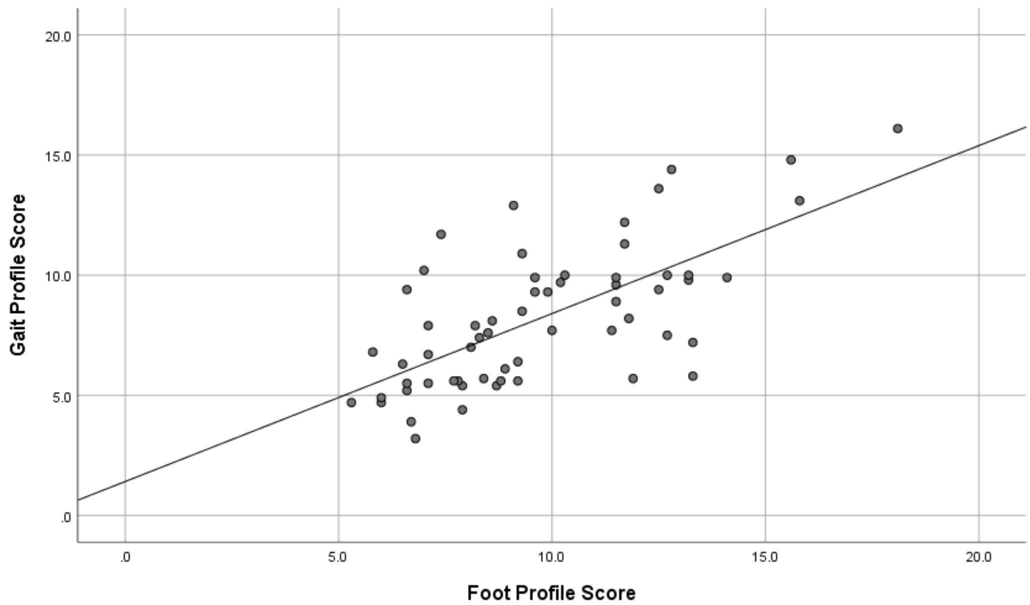

Figure 3: Global involvement group: scatterplot of GPS (bilateral) and FPS (unilateral)

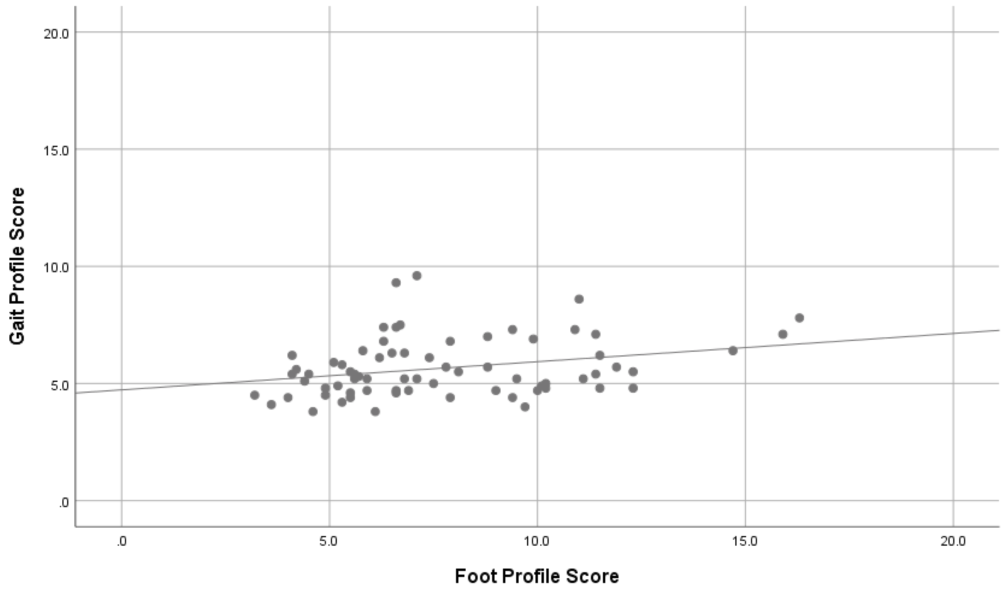

Figure 4: Foot deformity group: scatterplot of GPS (bilateral) and FPS (unilateral)

DISCUSSION

This paper introduces the Foot Profile Score- a measurement tool aiming to represent dynamic foot motion as a single meaningful numerical value. Through our validation process we have proven our stated hypotheses were true. The FPS showed good correlation with CFDS and all of the 6 FVS contributed positively to the prediction of CFDS. The FPS does offer different information than GPS, especially in populations where foot deformity is dominant.

Due to the lack of an available dynamic measure to validate the FPS against, we created the Clinical Foot Deformity Score. The CFDS expresses expert opinion and is based on a visual impression of foot deformity that clinicians use in their daily practice. It is encouraging the FPS correlates well with the mean CFDS, scored by 5 experts. We wouldn't expect a perfect correlation as the FPS gives quantitative information on all three planes of movement; in particular the transverse plane which is difficult to evaluate clinically or with 2D video.

The regression of CDFS on the 6 FVS shows that each of the score components contributes positively to predicting the clinical scores. It also reaffirms that all 6 kinematic variables are appropriate to include in the FPS, with equal weights. Due to the limited sample size and high levels of correlation of the individual components of the FPS, it is unsurprising that several of the FVS did not reach statistical significance. However, the FPS produced a better fit to the CFDS than the model with separate components. The positive and substantial contribution of FVS in predicting the CFDS is indicative of the content validity of FPS.

The fact that GPS also correlates to CFDS isn't surprising as GPS does model the foot crudely as a single rigid segment calculating ankle dorsiflexion, foot adduction and foot progression. In addition, if severe foot deformity is present, this can induce compensations at more proximal joints (hip and knee) which will influence the GPS. This is why we felt it important to include the regression of the CFDS on GPS and FPS (Table 2) proving FPS contributed independently of the GPS to the prediction of the CFDS.

A strong case for FPS validity is that the correlation between FPS and GPS was different when comparing the foot deformity and global involvement groups. The difference in correlations between these groups indicates that whilst FPS represents gait deviations not reflected by GPS in both groups, this is particularly evident in individuals where foot deformity is dominant. We also reported a more powerful approach to analyzing predictive validity of FPS that we believe to be new. When considering the ratio of the variances for FPS and GPS, we found the ratio to be increased in the foot deformity group compared to the global involvement group. This is because FPS should be sensitive in identifying differences between patients whose underlying problem is related directly to their feet, compared to differences in their GPS.

In patients with an isolated foot condition with little effect on proximal joints the GPS would be relatively unaffected. This reinforces the need for a separate foot specific outcome measure. A combination of GPS and FPS may be appropriate to report in these situations to more meaningfully describe an individual's gait pattern. A future consideration could be to remove the conventional lower limb model ankle kinematics from the GPS calculation when GPS and FPS are reported together.

Since its introduction in 2009, the Gait Profile Score has been used in clinical practice world-wide, particularly in children with cerebral palsy to improve understanding of their complex gait patterns [19], evaluate the use of Botulinum toxin [20] and evaluate surgical outcomes [21]. In addition to cerebral palsy, there is published research using GPS in other populations such as Charcot-Marie Tooth [22], Hereditary Spastic Paraplegia [23], Multiple Sclerosis [24], Parkinson's Disease [25] , and amputees [26]. The GPS has also been used to create or validate other outcome measures [27,28].

We envision the Foot Profile Score will be used similarly to GPS, to quantify foot specific deformity during gait in a wide range of populations, and demonstrate the outcome of intervention strategies. These may include conditions with global involvement and progressive foot deformity, such as cerebral palsy and stroke. Moreover, the FPS would be a useful clinical and research outcome measure in foot-specific conditions that affect dynamic foot motion and gait, such as pes cavus or flat foot.

STUDY LIMITATIONS

A limitation of this study is that we have only considered the OFM to create the FPS. It is unclear how it would work with other foot models, but theoretically it could be applied in a similar way. As with any summary gait index, there is a trade-off between simplicity and information content, therefore we still recommend the FPS is used in conjunction with the full kinematic data.

Hallux motion is not included in the FPS due to numerous factors. The hallux is a short segment measured with 2 markers in 2D. During a clinical session of 3D gait analysis it also has a high tendency to get knocked and replaced during pathological gait, making it less reliable than the other segments. Further study into how the addition of the hallux influences the overall FPS would be beneficial.

A second consideration for future work is to represent the FVS as a positive or negative value depending on the direction of deviation from the normative data. This may be particularly interesting when analysing foot motion over time or pre/post a surgical intervention as it is possible for an overall FPS to remain abnormal- but the foot posture to have changed (for example from equino-varus to excessive dorsiflexion with hindfoot valgus).

The FPS was specifically designed to offer a dynamic score of foot motion. Our previous work has shown that standing foot posture does not necessarily correlate to dynamic foot movement [29], therefore further work could be done to understand the differences of static foot mal-alignment versus abnormal dynamic foot motion. A method that could be applied to evaluate this has recently been suggested [30].

CONCLUSIONS

This study successfully validated the Foot Profile Score by studying its inherent properties and design, and by analysing it against a global clinical assessment of foot deformity.

The FPS is the first validated outcome measure of dynamic foot motion. It is a single measurement based on OFM kinematics. The FPS gives additional information to GPS and should be presented alongside other gait data to offer a better understanding of an individual's gait deviations.

The FPS has the potential to assist clinicians and researchers in quantifying foot abnormalities during gait, to monitor change over time, and to measure the outcome of intervention.

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Responsiveness of the Foot Profile Score in children with hemiplegia

Jennifer McCahill

Julie Stebbins

Robin Prescott

Jaap Harlaar

Tim Theologis

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ABSTRACT

BACKGROUND: The Foot Profile Score (FPS) is a single score that summarises foot posture and dynamic foot motion during the gait cycle based on the kinematic data of the Oxford Foot Model. The FPS enables clinicians and researchers to quantify foot abnormalities during gait, to monitor change in foot/ankle motion over time, and to measure the outcome of intervention. With the creation of a new outcome measure, it is important to test its responsiveness in a clinical population for whom it may be sensitive to change.

AIM: To evaluate the responsiveness of the FPS in a clinical population following isolated foot and ankle surgery.

METHODS: Using previous work completed to validate the FPS, we defined the minimal clinically important difference (MCID) for the FPS. Using this MCID, we applied it to a clinical population of 37 children with cerebral palsy, spastic hemiplegia, comparing their FPS before and after foot and ankle surgery. A regression analysis looked at potential relationships between the change in FPS and their pre-operative FPS, age at surgery, and time since surgery.

RESULTS: An MCID of 2.4 degrees was calculated through regression analysis. The mean change from the pre-operative FPS to the post-operative FPS was 4.6 (SD 3.7 with a range from -0.1 to 13.4). Twenty-eight children (76%) had a change in their FPS greater than the MCID. A regression analyses only showed a clear regression between pre-operative FPS and change in FPS ($R2 = 0.58$ $p<0.01$).

INTRODUCTION

The Foot Profile Score was created and validated in 2019 as a single score of foot posture and dynamic foot motion during gait (1) based on the kinematics of the Oxford Foot Model (2). The FPS is calculated as the root mean square average of 6 key kinematic variable scores (Foot Variable Scores- FVS), each calculated as the root mean square difference over the gait cycle between a patient's data and normative data individually for right and left legs (1). The 6 variables included in the FPS represent the motion of the hindfoot relative to the tibia in the sagittal, coronal, and transverse planes, as well as the motion of the forefoot relative to the hindfoot in the sagittal, coronal and transverse planes (1).

Hijji and colleagues state that an ideal outcome measurement tool should be 'relevant, reliable, valid, and responsive to a given pathology' (3). In addition, the FPS should be able to detect a clinically meaningful difference when analysing a progression in dynamic foot deformity over time, or a change in foot motion following an intervention (3). The Oxford Foot Model has been shown to be repeatable in both adult and child healthy populations (2,4-6), as well as in children with foot deformity (7). The FPS has been shown to be relevant and valid, particularly in populations where foot deformity is the predominant contributor to an altered overall gait pattern (1). What hasn't yet been demonstrated is the responsiveness of the FPS to detect changes within individuals following an intervention.

Children with cerebral palsy who experience walking problems are commonly referred for three-dimensional gait analysis (8,9). It is well documented that children with cerebral palsy develop musculoskeletal problems over time (9,10) often including progressive foot deformities requiring surgical intervention (11). For example, children with spastic hemiplegia can present with a variety of foot deformities including equinus, cavo-varus and planovalgus and often benefit from isolated foot correction (11,12). For this reason, the FPS is a relevant outcome measure for this population.

The aim of this study is to analyse the responsiveness of the FPS following isolated foot and ankle surgery in children with cerebral palsy, spastic hemiplegia.

METHODS

Defining the MCID for the FPS

The dataset previously reported by McCahill et al (1) in the original validation of the FPS was used to define the minimal clinically important difference (MCID). The Clinical Foot Deformity Scale (CFDS) was created by the authors to validate the FPS in the absence of another published dynamic foot deformity scale as described in McCahill and colleagues (1). Foot videos of 60 subjects were sent to 10 gait analysts affiliated with a 3D gait laboratory from 4 countries (5 physiotherapists, 2 orthopaedic surgeons, 2 clinical scientists/ engineers and 1 paediatric physiatrist).

Each subject was scored by 5 gait analysts. The subjects (30 adults and 30 children) included a range of demographics and severity of foot deformity ranging from planovalgus to cavovarus. 23 Subjects had orthopaedic diagnoses, 21 had cerebral palsy and 16 had neurological diagnoses. The gait analysts rated the overall appearance of the foot using a scale from 0 to 3, which was termed the Clinical Foot Deformity Scale (CFDS: 0=normal, 1=mild, 2=moderate, 3=severe foot deformity) with no further instructions. The CFDS was taken as the mean of all 5 gait analysts' ratings for each subject. The FPS was calculated for the same leg as used for the CFDS scoring for each subject (1). The MCID for the FPS was defined through linear regression of the FPS on the CFDS, corresponding to the change in FPS associated with a one unit change in the CFDS.

Responsiveness of the FPS pre-post intervention

A separate group of thirty-seven children with cerebral palsy, spastic hemiplegia was included for this study (mean age 11.9 years, SD 3.03, age range 7-17 years; 21 females/ 16 males; 18 left, 19 right side affected). All children had a pre-op and post-op gait analysis with OFM kinematics (2) collected using a Vicon MX/T-series motion capture system (Vicon Motion Systems Ltd.) including 16 cameras collecting at 100 Hz. Subjects walked at self-selected speed over level ground. The predominant foot deformities (as defined by the gait kinematics) requiring surgical correction included pure equinus (12 children), planovalgus (8 children) and cavovarus (17 children). Surgeries included only procedures below the knee: muscle and tendon lengthenings, tendon transfers, bony osteotomies, and supra-malleolar tibial de-rotations (Supplementary Information). The post-op analyses were completed on average 7.7 months following surgical intervention (range 6-15 months) for all 37 subjects.

For the purposes of this study, the definition of *responsiveness* is – the percentage of subjects where the change in FPS exceeds the MCID following surgery. This would indicate what percentage of subjects had a clinically meaningful change in their dynamic foot function post-surgery. All 37 subjects were analysed for their pre- post-surgical differences in their FPS. The change in FPS was also regressed on the subjects' age at surgery, time since surgery, and on their pre-operative FPS.

All analyses were completed using SPSS version 25, IBM, Chicago. Significance level was set at $p<0.05$.

RESULTS

Defining the MCID for the FPS

The MCID for the FPS was defined at 2.4 degrees with a significance of $p<0.001$ (Figure 1) as the regression coefficient, corresponding to a one unit change in the CFDS.

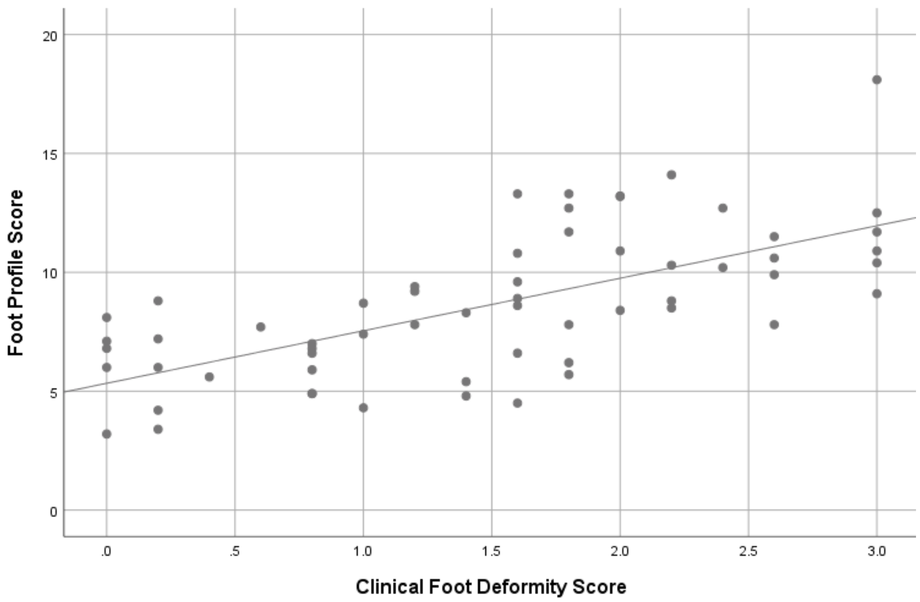

Figure 1: Regression of the Foot Profile Score on the Clinical Foot Deformity Score (Reprinted with permission from McCahill et al 2019).

Responsiveness of the FPS pre-post intervention

The mean change from the pre-operative FPS to the post-operative FPS was 4.6 degrees (SD 3.7 with a range from -0.1 to 13.4 degrees). Nine children (24%) did not reach the MCID of 2.4 degrees, one of whom worsened in their FPS by 0.1 degree. For the 9 children who did not reach the MCID, their pre-operative FPS ranged from 5.2 to 13.5 degrees and their pre-operative foot postures were: 3 cavovarus (18% of cavovarus feet), 3 planovalgus (38% of planovalgus feet), 3 equinus (25% of equinus feet) (Figure 2). The mean change for all children treated for cavovarus foot deformities was 5.2 degrees (SD 3.9 with a range from -0.1 to 13.4), equinus was 4.9 degrees (SD 4.3 with a range from 0.2 to 12.6) and planovalgus was 3 degrees (SD 1.5 with a range from 1.3 to 5.1).

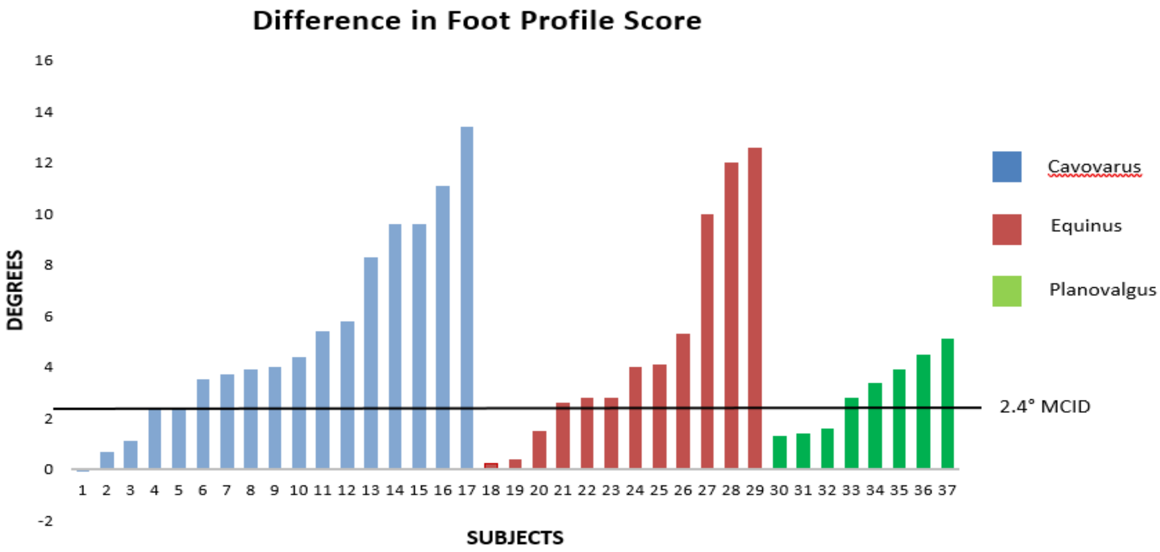

Figure 2: The difference in the FPS for all subjects, grouped into cavovarus, equinus and planovalgus pre-operative foot deformities.

Regressing the change in FPS on the pre-operative value of the FPS yielded a significant result with B=0.67 (SE 0.10) at $p<0.01$, and $R^2=0.58$, indicating 58% of the variance in the FPS difference can be explained by the pre-operative value of the FPS (Figure 3). Regressing the change in FPS on the subjects' age at surgery suggested a trend towards significance with B=–0.362 (SE 0.197) at p=0.074. Regression of the change in FPS on the time since surgery was not significant, B= –0.048 (SE 0.271) p=0.86.

Figure 3: Regression of the difference in the Foot Profile Score on the pre-operative Foot Profile Score in degrees.

DISCUSSION

Children with cerebral palsy, spastic hemiplegia commonly have isolated surgery to the foot/ ankle and are therefore an appropriate population to determine the responsiveness of the FPS without the confusing factor of additional surgeries. The results showed in our cohort of 37 children with spastic hemiplegia that 28 children (76%) met or exceeded the MCID of the FPS indicating a clinically meaningful improvement in the dynamic function of their feet following isolated foot and ankle surgery.

Our data shows, when regressing the change in FPS on the pre-operative FPS, the FPS fits with an established trend found by Rutz et al when analysing the change in Gait Profile Score in children with cerebral palsy post multi-level surgery (13). As well as an expected regression to the mean effect, a greater degree of abnormality in the FPS prior to surgery means a greater scope for improvement following surgery. This strengthens the confidence that the FPS is a responsive outcome measure. It also suggests a potential floor effect, as once the kinematics near the normal range, further improvements become less detectable. This raises an interesting dilemma about an MCID in general as the clinically important change in an outcome measure may be proportional to the original degree of deviation from norm, therefore those with minor deviations prior to surgery may not be expected to exceed a fixed MCID.

It is important to highlight that that although the FPS offers an objective assessment of foot shape and dynamic motion during gait, it does not capture other aspects such as pain. There are other factors that influence the subjective success of a procedure; therefore, the FPS should always be considered in combination with other outcome measures as a part of pre/post-surgical assessment.

Interestingly our results suggest that two factors may have a role in the outcomes following foot corrective surgery in children with hemiplegia, which require further investigation. Firstly, we will consider the type of pre-operative deformity: cavovarus, equinus, planovalgus. Sees and Miller (11) state that foot deformity is the most common orthopaedic problem in children with cerebral palsy. Many authors suggest that equinus and cavovarus deformities are the most common in hemiplegia (11,14), and our convenience sampling supports this. However, a natural tendency to planovalgus does exist in this population, and it can also occur due to over-correction of an equinus or cavovarus foot posture (15). Interestingly, no published study seems to compare the results of foot deformity correction based on the initial deformity. Further long-term follow up research is therefore justified to consider if one type of foot deformity in cerebral palsy is easier than the other to correct and maintain its correction.

The second factor which may influence the results of surgery is the age of the child at the time of surgery. Our results suggest that the younger children in our cohort (age range 7-17 years), had a greater difference in their FPS post-surgery then our older children, without this achieving statistical significance at the conventional 5% level. The FPS does not directly measure how well the foot was corrected but how well it is moving dynamically after treatment. Surgery in older children can be more extensive due to fixed deformity and stiffness. Therefore, surgery may improve the overall alignment of the foot, but not improve joint range of motion, or even come at the cost of that. This is particularly true if the surgery is more extensive (leading to more scarring) and/or if it includes bony surgery including joint fusions.

Contrary to this, minor soft tissue surgery in younger children will often correct the foot shape but also improve range of joint motion. Two recent review papers have looked at longer term results of foot surgery in cerebral palsy. Review papers by both Koman et al (16) and Shore et al (15) concluded that age at first surgery is the greatest predictor of recurrent equinus deformity in children with CP, and therefore conservative methods of management should precede any surgical intervention. Both of their review papers included children with spastic diplegia and spastic hemiplegia and both sets of authors acknowledge this makes it very difficult to make recommendations on individual cerebral palsy subtypes (15,16). In addition, the age at surgery for our included cohort is older than the majority of the reviewed papers indicating conservative management was likely employed prior to embarking on surgical intervention.

A limitation of this paper could be the MCID based on the association with the CFDS created to validate the FPS in a previous paper (1). We chose to base the MCID on a full unit in the CFDS, corresponding to a difference in grade that was agreed by all five assessors. It might be argued that if four of five assessors were assessing at a higher grade, this is indicating a difference that is of clinical importance, and an MCID might be set at 2 degrees or lower. Therefore, the value of the MCID warrants further investigation to rigorously evaluate the change in FPS required to make a clinically meaningful difference in a large cohort of subjects. In addition, the repeatability of the FPS is assumed to be good as the repeatability of the OFM has been shown to be good; however, a follow up study of the test-retest repeatability of the FPS would be beneficial.

Lastly, we have only assessed the responsiveness of the FPS in one clinical population, therefore we would recommend repeating this study in other populations.

CONCLUSIONS

An MCID of 2.4 degrees for the FPS indicated a clinically meaningful improvement in 76% of children with hemiplegia post isolated foot/ankle surgery. Moreover, the FPS responded with larger improvements for more deformed feet. These findings suggest the FPS is sufficiently responsive in a clinical population and should be considered when indicating and evaluating foot surgery. Further testing of the MCID is suggested, as a lower value may still be indicative of clinically meaningful improvement.

Supplementary Information:

Patient	pre FPS	post FPS	change FPS	Foot posture	Surgery
1	11.6	11.7	-0.1	cavovarus	Gastrocnemius, tibialis posterior, FHL, FDL and plantarfascia lengthenings, SPLATT, reconstruction of peroneal tendon sheath
2	13.5	12.8	0.7	cavovarus	Posterior-medial release, SPLATT
3	8.6	7.5	1.1	cavovarus	Tendo Achilles and tibialis posterior lengthenings, TATT
4	14.3	11.9	2.4	cavovarus	Gastrocnemius and tibialis posterior lengthenings, SPLATT
5	16	13.6	2.4	cavovarus	Posterior release, SPLATT
6	10.2	6.7	3.5	cavovarus	Tendo Achilles, FHL, FDL, and tibialis posterior lengthening, and a SPLATT
7	10.8	7.1	3.7	cavovarus	Tendo Achilles FHL, FDL and tibialis posterior lengthenings, SPLATT
8	14.5	10.6	3.9	cavovarus	TATT
9	10.8	6.8	4	cavovarus	Gastrocnemius and tibialis posterior lengthenings
10	16.9	12.5	4.4	cavovarus	Tendo Achilles and tibialis posterior lengthening, SPLATT, plantarfascia release
11	12.8	7.4	5.4	cavovarus	Tendo Achilles and tibialis posterior lengthenings, SPLATT
12	16.8	11	5.8	cavovarus	Tendo Achilles, FHL, FDL, tibialis posterior lengthenings, SPLATT
13	15.5	7.2	8.3	cavovarus	Gastrocnemius and tibialis posterior lengthenings, SPLATT
14	18.5	8.9	9.6	cavovarus	Tendo Achilles and tibialis posterior lengthenings, TATT
15	15	5.4	9.6	cavovarus	Tibialis anterior and posterior lengthenings, SPLATT
16	15.5	4.4	11.1	cavovarus	Tendo Achilles and tibialis posterior lengthenings
17	21.6	8.2	13.4	cavovarus	Posterior release, SPLATT
18	6.4	6.2	0.2	equinus	Tendo Achilles lengthening
19	10.6	10.2	0.4	equinus	Gastrocnemius lengthening
20	5.2	3.7	1.5	equinus	Tendo Achilles lengthening
21	11	8.4	2.6	equinus	Tendo Achilles lengthening, FDL transfer
22	5.8	3	2.8	equinus	Tendo Achilles lengthening and transfer of FDL to the dorsum of the foot
23	7.9	5.1	2.8	equinus	Tendo Achilles lengthening
24	10	6	4	equinus	Tendo Achilles lengthening
25	10.8	6.7	4.1	equinus	Tendo Achilles lengthening
26	12.5	7.2	5.3	equinus	Tendo Achilles lengthening
27	18.1	8.1	10	equinus	Tendo Achilles lengthening
28	17.2	5.2	12	equinus	Tendo Achilles lengthening
29	18.5	5.9	12.6	equinus	Tendo Achilles lengthening
30	6.9	5.6	1.3	planovalgus	Calcaneal lengthening, Tendo Achilles lengthening, flexor digitorum longus transfer
31	6	4.6	1.4	planovalgus	Calcaneal lengthening osteotomy, Tendo Achilles lengthening, tibial derotation osteotomy
32	9.8	8.2	1.6	planovalgus	Calcaneal lengthening osteotomies, 1st metatarsal osteotomy
33	9.1	6.3	2.8	planovalgus	Tendo Achilles lengthening, calcaneal lengthening and medial cuneiform shortening
34	7.4	4	3.4	planovalgus	Calcaneal and tibial osteotomies
35	7.6	3.7	3.9	planovalgus	Lateral column lengthening, flexor digitorum longus transfer, Tendo Achilles lengthening
36	9.1	4.6	4.5	planovalgus	Calcaneal lengthening osteotomy, tibialis posterior reefing
37	11.6	6.5	5.1	planovalgus	Calcaneal lengthening osteotomy, Tendo-Achilles lengthening

Table of surgical procedures for each subject including pre-post and change in Foot Profile Score

Key: SPATT- split anterior tibialis tendon transfer TATT- tibialis anterior tendon transfer FDL- flexor digitorum longus FHL- flexor halluces longus

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Foot function during gait and parental perceived outcome in older children with symptomatic clubfoot deformity

Jennifer McCahill

Julie Stebbins

Jaap Harlaar

Robin Prescott

Tim Theologis

Chris Lavy

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ABSTRACT

AIMS: To assess if older symptomatic children with clubfoot deformity differ in perceived disability and foot function during gait, depending on initial treatment with Ponseti or surgery, compared to a control group. Second aim was to investigate correlations between foot function during gait and perceived disability in this population.

PATIENTS AND METHODS: Seventy-three children with idiopathic clubfoot were included: 31 children treated with the Ponseti method (mean age 8.3 years; 24 male; 20 bilaterally affected, 13 left and 18 right sides analysed) and 42 treated with primary surgical correction (mean age 11.6 years; 28 male; 23 bilaterally affected, 18 left and 24 right sides analysed).

Foot function data was collected during walking gait and included Oxford Foot Model kinematics (Foot Profile Score and the range of motion and average position of each part of the foot) and plantar pressure (peak pressure in five areas of the foot). Oxford Ankle Foot Questionnaire, Disease Specific Index for clubfoot, Pediatric Quality of Life Inventory 4.0 were also collected. The gait data were compared between the two clubfoot groups and compared to control data. The gait data were also correlated with the data extracted from the questionnaires.

RESULTS AND CONCLUSIONS: Our findings suggest that symptomatic children with clubfoot deformity present with similar degrees of gait deviations and perceived disability regardless of whether they had previously been treated with the Ponseti Method or surgery. The presence of sagittal and coronal plane hindfoot deformity and coronal plane forefoot deformity

were associated with higher levels of perceived disability, regardless of their initial treatment.

- First paper to compare outcomes between Ponseti and surgery in a symptomatic older clubfoot population seeking further treatment
- First paper to correlate foot function during gait and perceived disability to establish a link between deformity and subjective outcomes

INTRODUCTION

Children treated in infancy for idiopathic clubfoot can present with residual, relapsed, or over-corrected foot deformity. Follow-up at 11 years post initial surgery has shown 56% required at least one additional procedure at a mean of four years following the initial surgery (1). In a prospective study comparing surgical versus Ponseti results, 38% of Ponseti and 30% of surgical subjects required additional procedures after three years of follow-up (2). The results also showed the severity of recurrent deformity in the surgical group was higher than the Ponseti group; resulting in the surgery group requiring more corrective procedures to treat the persistent deformities (2). More recently Hayes et al, reported a risk of over-correction following the Ponseti method of 12% after at least 8 years of follow-up (3).

Due to a lack of evidence to guide clinical decision making, current practices managing older children vary. How does a clinician decide who should receive additional surgical or conservative management, and who can be left untreated? There is a known association between the number of surgical interventions and level of perceived disability, however, the deformity may continue to progress if left untreated, causing disability into adulthood (4,5).

To date, outcome studies in older children with clubfoot have focused on comparing different types of treatment using pedobarography (6-9), lower limb kinematics and kinetics (1,10-16), multi-segment foot kinematics (16,17), and subjective questionnaires (15-18). However, these have focused on children who are doing well. No published literature exists analysing a symptomatic population of children previously treated for clubfoot deformity. We don't know if 'failed' Ponseti presents similarly to 'failed' surgery. In addition, no previous study has investigated the relationship between foot function (assessed by foot kinematics and plantar pressure) and patient reported outcome measures. Therefore our study hypotheses were:

1. There will be a difference in foot function during gait in older symptomatic children with clubfoot between those who have been previously treated by Ponseti compared to surgery; and that both will be different to a control group.
2. There will be a difference in perceived disability in older symptomatic children with clubfoot between those who have been previously treated by Ponseti compared to surgery.

If a correlation between foot function during gait and perceived disability could be established, it would give insight into the specific elements of foot deformity that are associated with poor subjective outcomes. This would enable treatment to target specific elements of the foot deformity, or else give evidence to reassure a family that no further treatment is indicated. Such correlations have not been established, and would need large numbers. Therefore, our third research question was more exploratory, with an aim to generate hypotheses for future studies:

3. What are the associations between foot function during gait and perceived disability in older symptomatic children with clubfoot who have been previously treated by Ponseti or surgery?

METHODS

Subjects

Seventy-three children with idiopathic clubfoot were included (mean age 10.2 years, range 5-16 years; 51 male; 43 bilateral, 12 left, 18 right side affected). For bilateral subjects, the worst-affected foot as assessed by the Foot Profile Score (19) was included, resulting in 42 right and 31 left feet being analysed. The sample included routine referrals- children referred to the clinical service for consideration of further management due to residual deformity, pain or reduced function. The gait laboratory is part of a tertiary hospital receiving referrals from multiple centres requiring this specialist service. The reasons for referral were to clarify residual foot abnormalities, advice on orthotic management, as well as potential surgical management. This indicates that a range of foot deformity were included in the sample. Inclusion criteria were subjects between the ages of 5 and 16 years old with a confirmed structural idiopathic clubfoot deformity diagnosed at birth, and no other musculoskeletal or neurological diagnoses.

Of the 73 children, 31 were previously treated with the Ponseti method with the treatment starting within the first 4 months following birth. 83% of the Ponseti group had an Achilles tenotomy (26 children) and 32% subsequently had an anterior tibialis tendon transfer (ATTT) (10 children). One of the children, who did not undergo a tenotomy following the initial casting, had a limited Achilles tendon lengthening at 2 years old. The Ponseti group had a mean age 8.3 years (range 5-16 years); 24 male; 20 bilaterally affected, with a total of 13 left and 18 right sides analysed.

The surgery group had 42 children treated with primary surgery before the age of 1 year old, following either strapping or below-knee casting. 24 children underwent postero-medial releases, 17 children had posterior releases, and 1 child had an Achilles tenotomy combined with a medial release. 19 of these children underwent subsequent surgery; 2 ATTT in isolation, 10 with an ATTT in combination with more extensive soft tissue release, capsular release, and tibial de-rotation osteotomies. The surgery group had a mean age 11.6 years (range 5-16 years); 28 male; 23 bilaterally affected, with a total of 18 left and 24 right sides analysed.

Two control groups were used in the assessment of foot function selected from the gait laboratory's normal databases. The kinematic data control group consisted of 30 children, mean age 10.7 years (range 5-16 years). The plantar pressure control group consisted of 30 children, mean age 10.6 years (range 5-16 years). For both control groups, the participants included healthy children with no known diagnoses or orthopaedic conditions. In order to match the gender and age distribution of the clubfoot group, 9 female and 21 male controls were selected for each group, using a stratified random sample (15 right and 15 left legs randomly selected).

<u>**Data Collection**</u>

1. **Foot function during gait**

<u>Foot kinematic data</u>

All 73 children had multi-segment foot kinematic data collected using the Oxford Foot Model (OFM) (20) during level walking at self-selected speed

using a 16 camera Vicon T-series system (Vicon Motion Systems Ltd, Oxford, United Kingdom) sampling at 100Hz with 9.5mm passive markers.
The *Foot Profile Score* (FPS) and 6 Foot Variable Scores were then calculated from the kinematic data of the OFM (19).

Since the FVS and FPS are absolute deviations from normal, we also calculated the *average position of each segment* during the gait cycle in each plane, which additionally gave the direction of deviation.
We also computed the overall *flexibility of each inter-segment* joint by calculating the range of motion in each plane.

<u>Plantar pressure data</u>

Plantar pressure data were collected using an EMED-M pressure plate (Novel, Munich, Germany) sampling at 50Hz. Total peak pressure and force-time integral were collected in 70 subjects. Due to technical difficulties, plantar pressure data from three subjects were not collected. Peak pressure in five areas of the plantar surface of the foot, defined by the kinematic markers: were measured in 59 subjects: medial and lateral hindfoot, midfoot, medial and lateral forefoot (21). Due to technical difficulties we could not calculate pressure variables for sub-areas of the foot in 11 children, resulting in data from 28 Ponseti and 31 surgical subjects.

2. Perceived disability

Oxford Ankle Foot Questionnaire (OxAFQ) (22) was collected in all 73 subjects. The OxAFQ comprises three domain scores (physical, school &

play, emotional). Roye's Disease Specific Index for clubfoot (DSI) (23) was collected in 38 subjects. This score measures the outcome of treatment of clubfoot and is comprised of a satisfaction subscale and function subscale. In addition, the Pediatric Quality of Life Inventory 4.0 SF15 Generic Core Scales (PedsQL) (24) was collected in 34 subjects, comprising a psychosocial health summary score, physical health summary score and a total score.

<u>Data Analysis</u>

1. **Foot function during gait**

The FPS, FVS, average position of each segment, flexibility of each segment, peak plantar pressure and force time integral data were compared between all three groups (the two clubfoot groups and the control group) using Welch's Analysis of Variance. Where significant differences were found, post hoc independent t-tests were used with unequal variances assumed. Log transformation was performed prior to the analysis for the FPS, FVS, flexibility score of each segment and plantar pressure data, because of marked positive skewness in these variables.

2. **Perceived disability**

An independent t-test was used to compare the means of the two clubfoot groups for each of the three subjective outcome measures with equal variances not assumed.

3. **Association of foot function and perceived disability**

For convenience in examining a large number of associations, Pearson Correlation Coefficients were used to explore the association between the independent variables (FPS, FVS, RoM of each foot joint in each plane, and

plantar pressure) and the dependent variables extracted from the parent-reported questionnaires. They yield the same p-values as a corresponding linear regression and provide a convenient measure of effect size. Due to the exploratory nature of this research question, we identified a priori the following components of foot deformity which we hypothesised would be associated with the dependent variables: hindfoot equinus, hindfoot varus, forefoot supination, forefoot adduction and increased midfoot pressure. When interpreting the data we took into account any outliers that affected the associations and checked scatter diagrams for non-linearity.
All analyses were completed using SPSS version 25, IBM, Chicago. Significance levels were set at $p<0.05$.

RESULTS

1. **Foot function during gait**

ANOVA results revealed a significant difference between the FPS and all six FVS (Table 1). Post hoc t-tests showed a significant difference for all variables between the surgical and control groups, as well as between the Ponseti and control groups, with the only exception being the forefoot in the coronal plane. When comparing the Ponseti and surgical groups, there were no statistically significant differences.

	Mean and range			Welch ANOVA (p values)	Independent t-tests (p values)		
	Control n=30	Ponseti n=31	Surgery n=42	3 groups	Ponseti vs Control	Surgery vs Control	Ponseti vs Surgery
Foot Profile Score (°)	4.8 (2.3-7.3)	8.3 (3.3-18.1)	9.3 (4.0-18.3)	<0.001*	<0.001*	<0.001*	0.11
Hindfoot sagittal (°)	3.7 (2.1-7.9)	4.9 (2.0-10.9)	5.5 (2.5-21.5)	0.006*	0.03*	0.003*	0.50
Forefoot sagittal (°)	3.4 (2.0-8.8)	5.3 (1.7-12.0)	4.8 (2.1-17.1)	0.001*	0.001*	0.002*	0.47
Hindfoot coronal (°)	3.7 (1.2-8.7)	8.0 (2.2-19.4)	7.3 (1.7-18.3)	<0.001*	<0.001*	<0.001*	0.49
Forefoot coronal (°)	4.9 (1.3-9.9)	6.9 (2.1-19.2)	9.1 (2.1-33.4)	0.001*	0.07	<0.001*	0.07
Hindfoot transverse (°)	5.8 (2.5-15.5)	9.7 (2.7-20.8)	10.8 (3.4-23.7)	<0.001*	0.001*	<0.001*	0.39
Forefoot transverse (°)	5.0 (1.1-11.0)	9.1 (1.5-20.5)	10.6 (1.4-26.2)	0.001*	0.02*	<0.001*	0.26

Table 1: The mean and range of the Foot Profile Score and the six Foot Variable Scores for all three groups (prior to log transformation). Welch ANOVA for all 3 groups and independent t-test (unequal variances assumed) between groups following log transformation (* $=p<0.05$). A higher number indicates greater deformity.

The comparison of the average position of each segment throughout the gait cycle between the clubfoot groups and control group (Appendix 1-Figure 1; Table 2) showed the surgery group had significantly increased forefoot supination relative to the tibia compared to the control group ($p=0.008$). Both the Ponseti and the surgery groups had increased hindfoot internal rotation compared to the control group ($p<0.001$). The Ponseti group had significantly increased forefoot adduction relative to the tibia compared to the control group (p=0.001) and compared to the surgery group (p=0.04).

	Mean and range			Welch ANOVA (p values)	Independent t-tests (p values)		
	Control n=30	Ponseti n=31	Surgery n=42	3 groups	Ponseti vs Control	Surgery vs Control	Ponseti vs Surgery
Hindfoot dorsiflexion (°)	2.1 (-4.5-8.1)	0.6 (-10.3-13.6)	1.4 (-19.9-12.4)	0.254			
Forefoot dorsiflexion (°)	-1.2 (-6.9-7.5)	-1.0 (-13.1-5.9)	-2.8 (-11.9-14.1)	0.159			
Forefoot/ Tibia dorsiflexion (°)	1.0 (-5.7-10.7)	1.4 (-15.3-8.2)	0.0 (-31.0-7.6)	0.540			
Hindfoot varus (°)	-3.3 (-10.6-5.4)	0.7 (-18.0-17.0)	-1.6 (-16.1-15.8)	0.055			
Forefoot supination (°)	6.6 (-2.4-14.7)	4.5 (-12.4-25.0)	8.6 (-5.4-36.4)	0.142			
Forefoot/Tibia supination (°)	3.3 (-1.8-9.8)	5.5 (-10.4-14.9)	6.8 (-7.0-32.5)	0.012*	0.071	0.008*	0.427
Hindfoot internal rotation (°)	2.4 (-6.6-15.8)	9.0 (-3.9-24.0)	8.6 (-6.0-26.9)	<0.001*	<0.001*	<0.001*	0.799
Forefoot adduction (°)	1.3 (-7.7-13.5)	4.7 (-18.1-35.5)	-1.6 (-26.5-28.2)	0.095			
Forefoot/Tibia adduction (°)	3.7 (-5.1-13.1)	13.5 (-17.8-45.1)	6.7 (-29.3-34.4)	0.002*	0.001*	0.206	0.040*

Table 2: The mean and range of the average position of each segment in the gait cycle for all three groups. Welch ANOVA for all 3 groups and independent t-test (unequal variances assumed) between groups (* =p<0.05). Positive numbers= dorsiflexion, varus, supination, internal rotation, adduction. Negative numbers= plantarflexion, valgus, pronation, external rotation, abduction.

There were no significant differences in range of forefoot motion between the groups in all three planes (Table 3). The hindfoot in the surgery group had significantly reduced RoM compared to the control group in the sagittal and coronal planes (p=0.004 and p=0.012 respectively). Interestingly, the hindfoot in the transverse plane showed increased range of motion in both the Ponseti and surgery groups compared to controls (p=0.003 and p<0.001 respectively). In no instance was there a statistically significant difference between the Ponseti and surgery groups.

	Mean and range			Welch ANOVA (p values)	Independent t-tests (p values)		
	Control n=30	Ponseti n=31	Surgery n=42	3 groups	Ponseti vs Control	Surgery vs Control	Ponseti vs Surgery
Hindfoot sagittal (°)	22.7 (14.8-34.4)	21.0 (13.3-30.8)	19.7 (13.7-33.1)	0.014*	0.135	0.004*	0.170
Forefoot sagittal (°)	16.1 (10.9-23.0)	15.7 (6.9-26.7)	15.1 (7.6-26.3)	0.299			
Hindfoot coronal (°)	10.5 (7.2-17.3)	10.8 (4.3-24.2)	9.1 (3.8-16.8)	0.036*	0.864	0.012*	0.069
Forefoot coronal (°)	8.1 (4.2-13.2)	8.6 (3.7-15.3)	9.9 (3.8-21.9)	0.218			
Hindfoot transverse (°)	16.0 (6.6-25.8)	20.4 (10.3-34.7)	23.3 (11.5-60.9)	<0.001*	0.003*	<0.001*	0.084
Forefoot transverse (°)	9.1 (4.5-16.6)	8.6 (4.2-25.6)	8.7 (3.3-24.7)	0.329			

Table 3: The mean and range of the flexibility (range of motion) of each inter-segment angle during the gait cycle for all three groups (prior to log transformation). Welch ANOVA for all 3 groups and independent t-test (unequal variances assumed) between groups following log transformation (* =p<0.05).

Significant differences were found across the three groups for all pressure measures except lateral forefoot pressure (Table 4). Both the medial and lateral hindfoot pressures were reduced for the Ponseti compared to the control group ($p<0.001$ for both) and the surgery compared to the control group ($p=0.017$ and $p<0.001$ respectively). Midfoot pressures were significantly increased in both Ponseti and surgery groups compared to the control group ($p<0.001$). Medial forefoot pressure was reduced in the Ponseti group compared to the control group ($p=0.008$) and compared to the surgery group ($p=0.008$). Total peak pressure was reduced in the Ponseti group compared to the control group ($p<0.001$) and compared to the surgery group ($p=0.005$). Force time integral was increased in the surgery group compared to control group ($p=0.002$) and compared to the Ponseti group ($p=0.013$).

	Mean and range			Welch ANOVA (p values)	Independent t-tests (p values)		
	Control n=30	Ponseti n=28	Surgery n=31	3 groups	Ponseti vs Control	Surgery vs Control	Ponseti vs Surgery
Medial Hindfoot (kPa)	394.5 (175-605)	231.0 (88-402)	336.4 (53-998)	<0.001*	<0.001*	0.017*	0.053
Lateral Hindfoot (kPa)	348.7 (200-585)	198.9 (88-333)	231.1 (111-471)	<0.001*	<0.001*	<0.001*	0.109
Midfoot (kPa)	38.7 (0-130)	118.7 (10-398)	132.0 (67-313)	<0.001*	<0.001*	<0.001*	0.348
Medial Forefoot (kPa)	387.7 (155-940)	290.0 (115-555)	428.6 (100-1151)	0.010*	75% 0.008*	68% 0.761	0.008*
Lateral Forefoot (kPa)	260.0 (140-760)	246.5 (143-527)	319.7 (133-980)	0.136			
Total Peak Pressure (kPa)	481.7 (290-940)	357.4 (195-1067)	493.3 (230-1151)	0.001*	<0.001*	0.637	0.005*
Force time integral (kPa.s)	184.0 (73-405)	195.6 (103-408)	231.7 (66-433)	0.004*	0.244	0.002*	0.013*

Table 4: The mean and range of the plantar pressure measurements of all 3 groups (prior to log transformation). Welch ANOVA for all 3 groups and independent t-test (unequal variances assumed) between groups following log transformation (* =p<0.05).

2. Perceived disability

Overall, the surgery group scored lower than the Ponseti group in the DSI and the OxAFQ, but the only statistically significant differences between the groups were in the Satisfaction subscale of the DSI (p=0.031) and the Emotional domain of the OxAFQ (p=0.016) (Table 5).

		Mean (range) Ponseti	Surgery	P-value
PedsQL	PhysHealth	77.0 (47-100)	75.7 (31-100)	0.852
Ponseti n=22	PsychSoc	75.1 (16-100)	82.9 (61-100)	0.228
Surgery n=13	Total Score	75.5 (31-97)	80.4 (54-100)	0.418
DSI	Satisfaction	65.6 (40-100)	54.4 (26-80)	0.031*
Ponseti n=25	Function	62.1 (20-100)	56.9 (6-100)	0.394
Surgery n=13	Total Score	65.1 (37-90)	55.6 (33-90)	0.107
OxAFQ	Physical	64.1 (12.5-100)	58.2 (12.5-100)	0.323
Ponseti n=31	School & Play	80.0 (19-100)	78.4 (25-100)	0.771
Surgery n=42	Emotional	83.1 (19-100)	69.7 (12.5-100)	0.016*

Table 5: The mean and range of the Pediatric Quality of Life Questionnaire (PedsQL), Disease Specific Index (DSI) and Oxford Ankle Foot Questionnaire (OxAFQ) for the Ponseti and surgical groups. Independent t-test (unequal variances assumed) between groups (* =p<0.05).

3. Association of foot function and perceived disability

The correlations of the gait data with subjective outcome measures are presented in Tables 6, 7 and 8 in Appendix 2. We were particularly interested in the associations with foot function variables that we identified a priori in our hypotheses. The variables representing hindfoot equinus (RoM in the sagittal plane, hindfoot sagittal FVS, and reduced pressure in the heel regions) all demonstrated significant associations with each of the subjective questionnaire scores, although these differed according to clubfoot group and gait variable being considered. This was similarly the case for variables representing hindfoot varus (coronal hindfoot RoM and peak pressure under the medial aspect of the foot compared to the lateral), forefoot supination (coronal forefoot FVS and RoM), forefoot adduction (transverse forefoot FVS and RoM), and midfoot pressure. Results overall indicated that the foot function variables we identified were associated with poorer subjective outcomes.

DISCUSSION

Our findings suggest that children with symptomatic clubfoot deformity, whether treated by Ponseti or surgery, present with similar degree of deficits in foot function during gait as well as a similar level of perceived disability. Therefore we accept the hypothesis that both clubfoot treatment groups are different to controls. However, we cannot conclude that the two clubfoot groups are different to each other with respect to foot function or subjective outcomes.

This is the first study to investigate children who are symptomatic following their initial clubfoot correction- regardless of whether they were treated with the Ponseti method or surgery. The uniqueness of our cohort is confirmed by our lower DSI scores compared to the literature (25, 21).

Both clubfoot groups had increased FPS and FVS compared to normal, which indicates impaired foot function during walking. However, they were not statistically significantly different to each other. The position of the forefoot and hindfoot showed that under-correction or over-correction occurred in both clubfoot groups. The only statistically significant difference between the groups was increased forefoot adduction relative to the tibia in the Ponseti group compared to both the surgical and the control groups. Both clubfoot groups showed significantly reduced peak hindfoot pressure and increased midfoot pressure compared to controls. The Ponseti group had reduced medial forefoot pressure compared to both surgery and controls groups.

Other clubfoot studies have reported stiffness in the sagittal hindfoot using the OFM in a surgical population compared to a Ponseti population (17), and Mindler et al (16) found this in a Ponseti population compared to controls. Jeans et al (9) investigated a Ponseti population and found compared to controls, similar to our results, they had reduced plantar pressure in the hindfoot and increased pressure in the midfoot. Converse to our results, Salazar et al (8) compared Ponseti and surgery groups using plantar pressure and found the Ponseti group had reduced peak hindfoot pressure and increased midfoot pressure compared to their surgical population. Differences are likely due to the populations studied.

This is the first study to correlate gait data with perceived disability in children treated for clubfoot. Multiple exploratory correlations were assessed to identify relationships between the gait data and subjective questionnaires. It is important to note that the OxAFQ had the most responses and therefore the most emphasis should be put on associations found using this outcome measure. Despite the similarities in gait and subjective outcomes between

the clubfoot groups, the Ponseti and surgery groups behaved differently in how their gait deviations related to subjective outcomes.
In the Ponseti group perceived disability was associated with hindfoot equinus, increased peak midfoot pressures, reduced peak medial forefoot pressures, and reduced RoM of the hindfoot in the coronal plane. This suggests that children who have these residual deformities are more likely to have poor subjective outcomes. Therefore good initial correction of hindfoot equinus with a tenotomy, as well as full subtalar correction in the casting phase may be important in this population.

In the surgical group perceived disability was associated with coronal forefoot deformity, reduced RoM of the forefoot in the sagittal plane and of the hindfoot in the sagittal and coronal planes. This suggests that post-surgical correction, children who have residual forefoot supination or residual stiffness of the forefoot and hindfoot in the sagittal plane, or stiffness of the hindfoot in the coronal plane are likely to have poor subjective outcomes.

It is important to acknowledge the large inter-individual variation within the clubfoot subjects (Figure 1). It is therefore difficult to make generalisations and recommendations based on a child's previous treatment (Ponseti or surgery) as both contain the entire spectrum of deformity with no specific pattern. This supports the view that each child should receive an individualized approach when seeking further management.

An interesting outcome of our study was that the three subjective outcome measures showed very little agreement in correlations with the gait data. This might be expected with a generic health measure like the PedsQL, but the DSI was designed for use in clubfoot (23), and the OxAFQ was validated using clubfoot as one of its populations (22). One possibility is that these measures are not sensitive enough to correlate with foot function defined by 3D gait analysis. The link between body function, participation and quality of life has not yet been well defined for this population, which justifies future research in this area.

STUDY LIMITATIONS

Specific details of severity of the original deformity, such as the Pirani Score, and initial success of the Ponseti method or surgery were unknown due to the nature of tertiary referral. We recognise the many correlations examined may bring up false positive associations. Therefore, we only put emphasis on those we had hypothesised a priori. A larger study would be needed to further explore our preliminary findings. Lastly, due to subdividing the clubfoot subjects into two groups and only having a subset of data for the PedsQL and DSI, some of the associations were more prone to outliers. We did our best to acknowledge when outliers were affecting statistically significant associations.

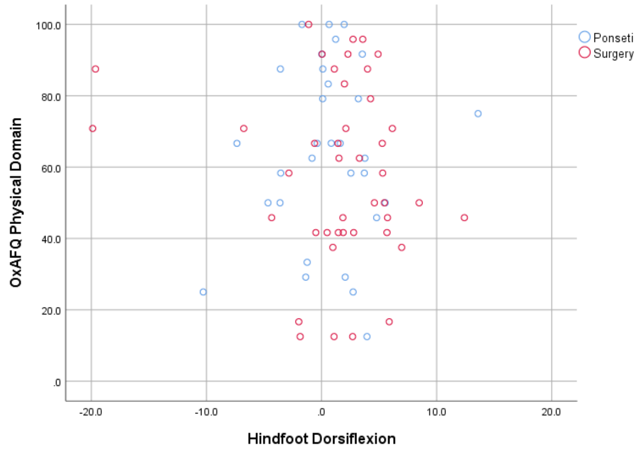
CONCLUSIONS

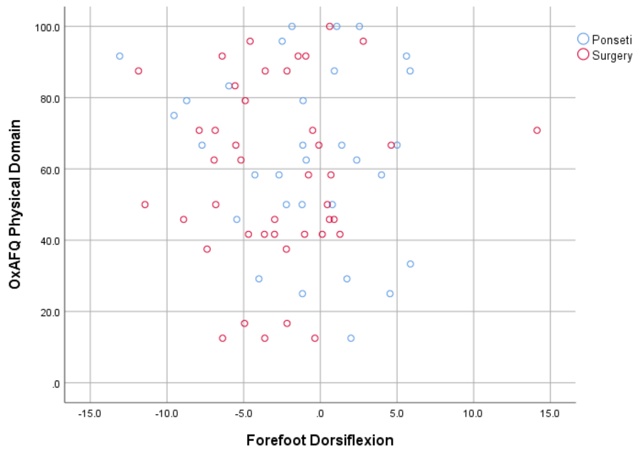
Our findings suggest that children with symptomatic clubfoot deformity present with a similar amount of gait deviations and perceived disability whether treated by the Ponseti method or surgery. Hindfoot deformity in the sagittal plane and forefoot and hindfoot deformity in the coronal plane were associated with perceived disability, regardless of whether they had received the Ponseti method or surgery.

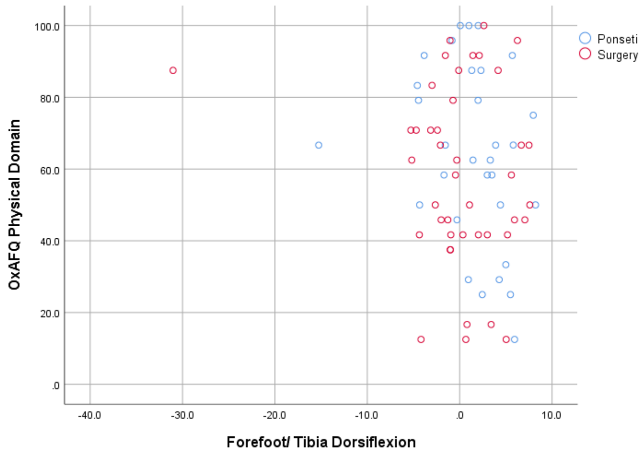
We would like to acknowledge funding of this research from Newlife the Charity for Disabled Children.

Appendix 1- Figure 1- Scatterplots of the average position of the forefoot and hindfoot throughout the gait cycle in the sagittal, coronal and transverse planes

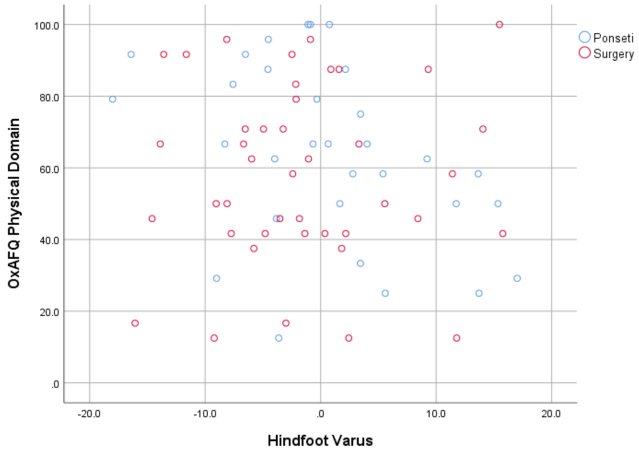
Sagittal plane:

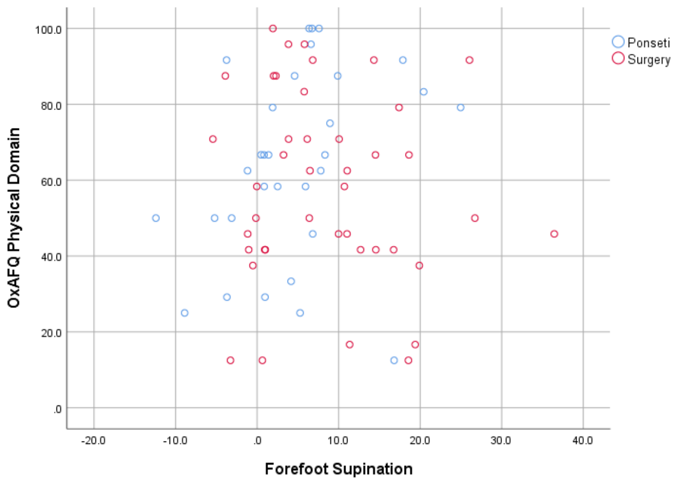


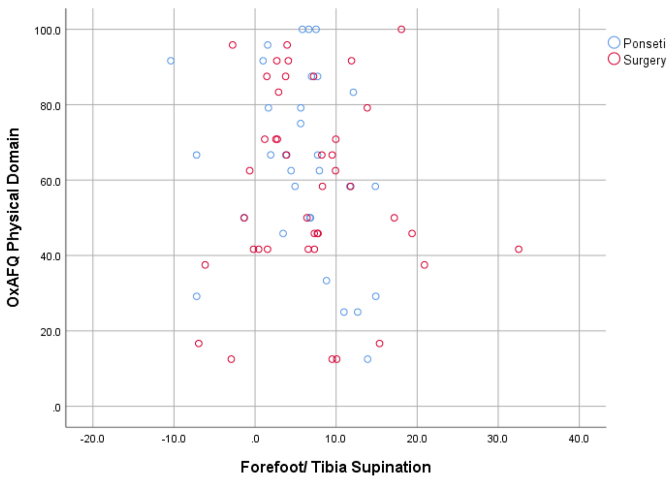




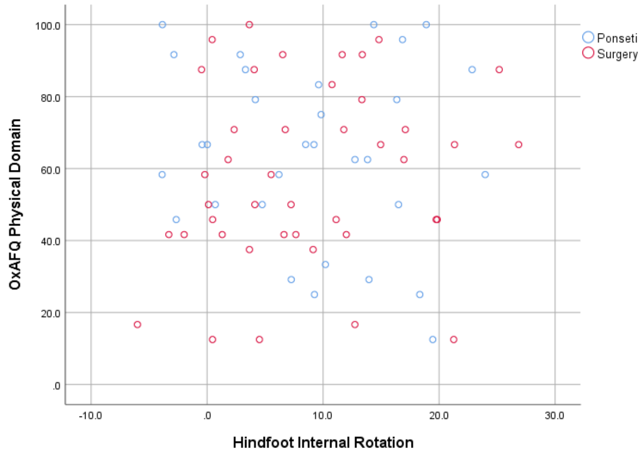
Coronal plane:

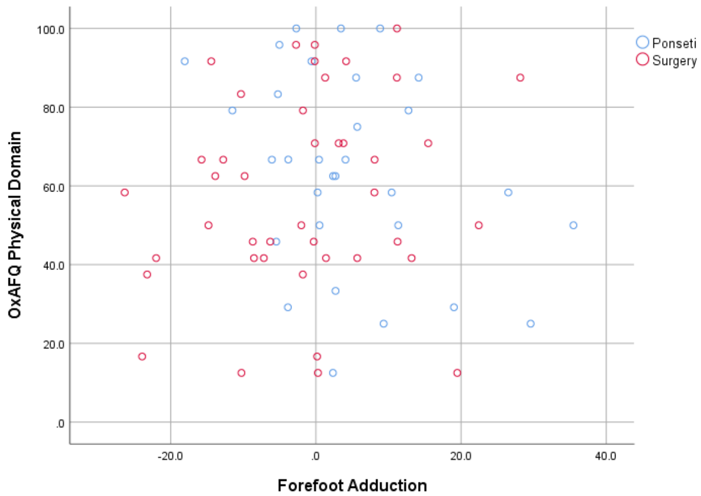


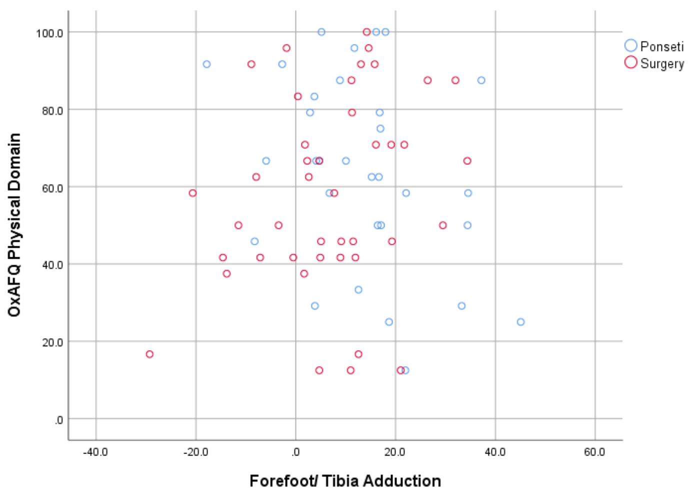




Transverse plane:







Appendix 2–

			Foot Variable Scores						Range						Plantar pressure						
		FPS	HF sag	FF sag	HF cor	FF cor	HF trans	FF trans	HF sag	FF sag	HF cor	FF cor	HF trans	FF trans	med HF	lat HF	MF	med FF	lat FF	total PP	FTI
DSI-satisfaction	r=	-0.036	-0.218	0.053	-0.142	0.118	-0.145	0.018	-0.098	0.075	-0.107	0.261	0.024	**.449***	0.217	-0.157	-0.337	**.488***	-0.206	0.263	0.230
	p=	0.866	0.295	0.800	0.500	0.575	0.490	0.932	0.642	0.722	0.611	0.207	0.908	0.024	0.309	0.463	0.108	0.016	0.335	0.215	0.291
	N	25	25	25	25	25	25	25	25	25	25	25	25	25	24	24	24	24	24	24	23
DSI-function	r=	0.009	-0.212	0.168	-0.075	0.016	-0.205	0.178	-0.339	-0.010	-0.111	0.083	-0.081	0.170	0.042	-0.061	-0.082	0.114	-0.169	0.063	0.048
	p=	0.965	0.310	0.422	0.722	0.939	0.326	0.395	0.098	0.963	0.596	0.693	0.702	0.417	0.847	0.777	0.702	0.595	0.431	0.771	0.828
	N	25	25	25	25	25	25	25	25	25	25	25	25	25	24	24	24	24	24	24	23
DSI-total score	r=	-0.012	-0.255	0.139	-0.124	0.072	-0.212	0.127	-0.275	0.033	-0.130	0.192	-0.041	0.348	0.137	-0.120	-0.225	0.323	-0.216	0.175	0.148
	p=	0.953	0.219	0.507	0.555	0.731	0.310	0.545	0.183	0.876	0.536	0.358	0.847	0.088	0.522	0.576	0.290	0.124	0.312	0.414	0.501
	N	25	25	25	25	25	25	25	25	25	25	25	25	25	24	24	24	24	24	24	23
PedsQL psychsocial	r=	-0.055	-0.052	-0.018	0.088	-0.186	-0.358	0.283	-0.252	0.198	**-.515***	0.295	0.064	0.380	0.283	0.270	0.058	0.342	-0.039	0.281	0.344
	p=	0.813	0.824	0.937	0.704	0.420	0.111	0.215	0.270	0.391	0.017	0.194	0.783	0.090	0.227	0.249	0.809	0.140	0.870	0.229	0.149
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	20	20	20	20	20	20	19
PedsQL physhealth	r=	-0.227	**-.446***	0.073	-0.280	-0.169	-0.336	0.121	-0.233	0.088	**-.430***	0.123	-0.203	0.299	0.349	0.261	-0.279	0.245	-0.412	-0.004	0.103
	p=	0.309	0.037	0.745	0.206	0.451	0.127	0.592	0.296	0.697	0.046	0.584	0.366	0.176	0.120	0.253	0.220	0.285	0.064	0.987	0.665
	N	22	22	22	22	22	22	22	22	22	22	22	22	22	21	21	21	21	21	21	20
PedsQL total score	r=	-0.145	-0.174	-0.001	-0.045	-0.239	-0.414	0.252	-0.288	0.204	**-.543***	0.272	-0.018	0.392	0.330	0.297	-0.028	0.357	-0.153	0.231	0.317
	p=	0.530	0.451	0.998	0.848	0.297	0.062	0.270	0.205	0.374	0.011	0.233	0.937	0.079	0.156	0.204	0.906	0.122	0.519	0.327	0.187
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	20	20	20	20	20	20	19
OxAFQ-emotional	r=	-0.162	-0.272	-0.101	-0.209	-0.204	-0.156	0.062	0.042	0.067	-0.203	0.287	0.046	0.250	0.339	0.368	-0.173	0.211	-0.115	0.026	0.052
	p=	0.384	0.139	0.589	0.260	0.272	0.401	0.738	0.822	0.719	0.274	0.118	0.806	0.175	0.078	0.054	0.377	0.280	0.561	0.892	0.788
	N	31	31	31	31	31	31	31	31	31	31	31	31	31	28	28	28	28	28	30	29
OxAFQ-sch+play	r=	-0.173	-0.331	-0.008	-0.271	-0.117	-0.314	0.152	-0.216	0.006	-0.286	0.107	-0.191	0.202	0.345	**.379***	-0.237	0.156	-0.367	-0.050	-0.032
	p=	0.353	0.069	0.968	0.141	0.531	0.085	0.414	0.243	0.974	0.118	0.566	0.304	0.276	0.072	0.046	0.226	0.427	0.055	0.792	0.869
	N	31	31	31	31	31	31	31	31	31	31	31	31	31	28	28	28	28	28	30	29
OxAFQ-physical	r=	-0.255	-0.321	0.045	-0.345	-0.254	-0.173	-0.096	-0.086	0.201	-0.017	0.083	-0.020	**.403***	**.492****	**.402***	**-.446***	0.253	**-.487****	0.008	-0.052
	p=	0.166	0.078	0.809	0.058	0.168	0.353	0.607	0.646	0.278	0.928	0.656	0.915	0.024	0.008	0.034	0.017	0.194	0.009	0.966	0.790
	N	31	31	31	31	31	31	31	31	31	31	31	31	31	28	28	28	28	28	30	29

Table 6: Correlations of the gait data and subjective outcome measures are presented for the Ponseti group

			Foot Variable Scores						Range						Plantar pressure						
		FPS	HF sag	FF sag	HF cor	FF cor	HF trans	FF trans	HF sag	FF sag	HF cor	FF cor	HF trans	FF trans	med HF	lat HF	MF	med FF	lat FF	total PP	FTI
DSI-satisfaction	r=	0.080	-0.049	-0.031	0.370	0.058	-0.210	-0.082	0.459	-0.330	**-.685****	0.132	0.449	0.366	-0.553	-0.526	0.159	**-.685***	-0.308	-0.571	-0.162
	p=	0.795	0.873	0.921	0.214	0.850	0.491	0.791	0.115	0.271	0.010	0.667	0.123	0.218	0.097	0.119	0.661	0.029	0.387	0.067	0.654
	N	13	13	13	13	13	13	13	13	13	13	13	13	13	10	10	10	10	10	11	10
DSI-function	r=	0.394	-0.018	0.115	**.754****	-0.152	0.025	0.369	-0.351	-0.175	-0.398	0.460	0.242	0.221	-0.489	-0.137	0.564	0.280	0.378	0.284	0.009
	p=	0.183	0.955	0.707	0.003	0.620	0.935	0.215	0.240	0.567	0.178	0.113	0.426	0.469	0.152	0.707	0.090	0.434	0.282	0.398	0.981
	N	13	13	13	13	13	13	13	13	13	13	13	13	13	10	10	10	10	10	11	10
DSI-total score	r=	0.356	-0.034	0.083	**.768****	-0.102	-0.063	0.271	-0.107	-0.275	**-.600***	0.432	0.378	0.327	-0.614	-0.293	0.552	0.025	0.235	0.007	-0.057
	p=	0.232	0.912	0.788	0.002	0.740	0.839	0.370	0.729	0.362	0.030	0.141	0.203	0.275	0.059	0.412	0.098	0.945	0.514	0.985	0.876
	N	13	13	13	13	13	13	13	13	13	13	13	13	13	10	10	10	10	10	11	10
PedsQL psychsocial	r=	0.478	-0.016	-0.014	0.389	-0.073	0.265	0.513	-0.172	-0.158	-0.144	0.351	0.032	0.331	-0.133	0.173	0.411	-0.023	0.225	0.105	-0.174
	p=	0.098	0.960	0.964	0.189	0.813	0.381	0.073	0.575	0.605	0.639	0.240	0.918	0.270	0.714	0.633	0.237	0.950	0.532	0.758	0.631
	N	13	13	13	13	13	13	13	13	13	13	13	13	13	10	10	10	10	10	11	10
PedsQL physhealth	r=	0.329	0.036	0.066	0.530	-0.268	-0.023	0.411	-0.302	-0.250	-0.221	0.391	-0.044	0.334	-0.362	0.021	0.556	0.241	0.444	0.231	-0.120
	p=	0.272	0.907	0.830	0.062	0.376	0.942	0.163	0.317	0.411	0.468	0.186	0.886	0.265	0.304	0.953	0.095	0.502	0.199	0.494	0.741
	N	13	13	13	13	13	13	13	13	13	13	13	13	13	10	10	10	10	10	11	10
PedsQL total score	r=	0.432	0.007	0.021	0.469	-0.163	0.148	0.490	-0.238	-0.206	-0.185	0.384	-0.001	0.346	-0.246	0.108	0.495	0.101	0.337	0.169	-0.155
	p=	0.140	0.983	0.945	0.106	0.594	0.630	0.089	0.434	0.499	0.546	0.195	0.998	0.247	0.493	0.767	0.145	0.782	0.341	0.619	0.669
	N	13	13	13	13	13	13	13	13	13	13	13	13	13	10	10	10	10	10	11	10
OxAFQ-emotional	r=	-0.058	-0.180	-0.201	-0.175	-0.271	**.344***	-0.212	-0.216	-0.150	-0.011	-0.036	-0.069	-0.070	0.036	0.077	-0.026	0.035	-0.004	0.146	-0.099
	p=	0.720	0.260	0.207	0.274	0.086	0.027	0.183	0.176	0.350	0.948	0.822	0.669	0.664	0.852	0.686	0.893	0.855	0.984	0.375	0.560
	N	41	41	41	41	41	41	41	41	41	41	41	41	41	30	30	30	30	30	39	37
OxAFQ-sch+play	r=	-0.046	-0.061	-0.049	-0.087	-0.142	0.168	-0.080	**-.316***	-0.236	0.155	**.382***	-0.008	0.154	0.129	0.262	-0.052	0.114	0.195	0.193	-0.136
	p=	0.777	0.703	0.762	0.590	0.374	0.295	0.617	0.044	0.138	0.334	0.014	0.961	0.336	0.499	0.162	0.786	0.547	0.302	0.240	0.423
	N	41	41	41	41	41	41	41	41	41	41	41	41	41	30	30	30	30	30	39	37
OxAFQ-physical	r=	-0.074	-0.037	-0.104	-0.069	**-.335***	0.264	-0.264	-0.178	**-.348***	0.112	0.154	0.079	0.188	-0.158	-0.116	0.038	-0.085	0.232	0.042	-0.312
	p=	0.640	0.814	0.514	0.663	0.030	0.092	0.091	0.259	0.024	0.479	0.331	0.621	0.234	0.397	0.535	0.840	0.648	0.210	0.796	0.057
	N	42	42	42	42	42	42	42	42	42	42	42	42	42	31	31	31	31	31	40	38

Table 7: Correlations of the gait data and subjective outcome measures for the surgery group

			Foot Variable Scores						Range						Plantar pressure							
		FPS	HF sag	FF sag	HF cor	FF cor	HF trans	FF trans	HF sag	FF sag	HF cor	FF cor	HF trans	FF trans	med HF	lat HF	MF	med FF	lat FF	total PP	FTI
DSI-satisfaction	r=	-0.049	-0.222	0.045	0.041	0.047	-0.201	-0.037	0.115	0.064	-0.147	0.203	-0.005	**.421****	-0.037	-0.247	-0.282	0.069	-0.283	-0.113	0.051
	p=	0.771	0.181	0.789	0.807	0.778	0.227	0.828	0.492	0.704	0.380	0.221	0.976	0.008	0.835	0.160	0.107	0.698	0.104	0.520	0.777
	N	38	38	38	38	38	38	38	38	38	38	38	38	38	34	34	34	34	34	35	33
DSI-function	r=	0.137	-0.150	0.152	0.265	-0.069	-0.137	0.231	**-.325***	-0.022	-0.164	0.219	-0.026	0.199	-0.227	-0.096	0.089	0.159	0.025	0.072	-0.002
	p=	0.413	0.367	0.362	0.108	0.683	0.411	0.162	0.046	0.894	0.324	0.187	0.876	0.232	0.196	0.588	0.615	0.370	0.886	0.680	0.992
	N	38	38	38	38	38	38	38	38	38	38	38	38	38	34	34	34	34	34	35	33
DSI-total score	r=	0.072	-0.215	0.129	0.207	-0.025	-0.196	0.145	-0.173	0.016	-0.188	0.254	-0.021	**.347***	-0.183	-0.184	-0.065	0.148	-0.112	-0.003	0.023
	p=	0.666	0.194	0.439	0.213	0.882	0.239	0.385	0.300	0.926	0.258	0.123	0.901	0.033	0.299	0.297	0.716	0.404	0.528	0.987	0.898
	N	38	38	38	38	38	38	38	38	38	38	38	38	38	34	34	34	34	34	35	33
PedsQL psychsocial	r=	0.142	0.014	-0.025	0.163	-0.090	-0.137	**.356***	-0.207	0.045	**-.428***	0.314	0.122	0.336	0.131	0.235	0.148	0.250	0.100	0.289	0.267
	p=	0.424	0.936	0.886	0.356	0.611	0.439	0.039	0.239	0.802	0.012	0.070	0.490	0.052	0.492	0.211	0.436	0.182	0.599	0.114	0.162
	N	34	34	34	34	34	34	34	34	34	34	34	34	34	30	30	30	30	30	31	29
Peds QL physhealth	r=	0.001	-0.199	0.072	0.064	-0.209	-0.213	0.227	-0.265	-0.023	-0.313	0.227	-0.146	0.315	-0.008	0.153	-0.035	0.221	-0.057	0.050	0.012
	p=	0.997	0.251	0.681	0.714	0.227	0.219	0.190	0.124	0.896	0.068	0.189	0.403	0.065	0.967	0.412	0.850	0.232	0.763	0.784	0.950
	N	35	35	35	35	35	35	35	35	35	35	35	35	35	31	31	31	31	31	32	30
PedsQL total score	r=	0.093	-0.053	0.000	0.133	-0.165	-0.191	**.342***	-0.258	0.035	**-.431***	0.316	0.039	**.360***	0.090	0.231	0.114	0.274	0.065	0.245	0.214
	p=	0.599	0.764	1.000	0.454	0.351	0.280	0.048	0.140	0.845	0.011	0.069	0.829	0.036	0.635	0.219	0.548	0.143	0.735	0.185	0.265
	N	34	34	34	34	34	34	34	34	34	34	34	34	34	30	30	30	30	30	31	29
OxAFQ-emotional	r=	-0.151	-0.231	-0.126	-0.151	**-.289***	0.103	-0.132	-0.062	-0.042	-0.029	0.044	-0.073	0.046	0.107	0.174	-0.130	0.045	-0.091	0.005	-0.122
	p=	0.206	0.051	0.291	0.206	0.014	0.391	0.267	0.607	0.723	0.809	0.712	0.541	0.700	0.425	0.191	0.332	0.736	0.496	0.969	0.328
	N	72	72	72	72	72	72	72	72	72	72	72	72	72	58	58	58	58	58	69	66
OxAFQ-sch+play	r=	-0.116	-0.184	-0.025	-0.167	-0.135	-0.066	0.025	**-.259***	-0.112	-0.054	**.253***	-0.101	0.173	0.219	**.315***	-0.157	0.130	-0.057	0.071	-0.095
	p=	0.332	0.121	0.837	0.160	0.260	0.580	0.838	0.028	0.350	0.655	0.032	0.399	0.146	0.099	0.016	0.238	0.330	0.673	0.560	0.448
	N	72	72	72	72	72	72	72	72	72	72	72	72	72	58	58	58	58	58	69	66
OxAFQ-physical	r=	-0.177	-0.160	-0.024	-0.172	**-.317****	0.063	-0.205	-0.118	-0.090	0.079	0.108	0.011	**.270***	0.073	0.104	-0.235	0.030	-0.089	-0.016	-0.238
	p=	0.133	0.176	0.842	0.146	0.006	0.599	0.082	0.321	0.447	0.505	0.361	0.925	0.021	0.581	0.432	0.074	0.819	0.501	0.896	0.052
	N	73	73	73	73	73	73	73	73	73	73	73	73	73	59	59	59	59	59	70	67

Table 8: Correlations of the gait data and subjective outcome measures for the combined clubfoot group

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General Discussion

SUMMARY OF AIM

Foot deformity affects a high percentage of the population at some point in their lifetime. It is the most prevalent musculoskeletal problem at birth (1), it has a high incidence following a neurological insult (2), and it is known to be prevalent in the natural aging process from childhood into adulthood (3-5). Recent systematic reviews of the foot and ankle literature agree we are limited in functional ways to assess foot deformity (6) and we lack consensus on the best way to measure foot deformity for meaningful outcome analysis (7-9). In addition to this, we also lack understanding of how the structural deformity correlates to aberrant foot motion and an individual's subjective perspective on foot function and perceived disability.

More recently, instrumented gait analysis including a three-dimensional foot model has allowed dynamic analysis of foot posture and motion. The Oxford Foot Model (OFM) (10) is a multi-segment kinematic foot model that has been used widely in clinical and research applications, including outside of the institution it was developed (11). The primary aim of this thesis was to establish the clinical role of the Oxford Foot Model to assess foot deformity during gait. A secondary aim was to explore a potential relationship between altered foot structure/ function during gait and perceived quality of life in a clinical population.

To address the primary aim, first we investigated if the OFM was repeatable in children with foot deformity. Secondly, we built upon previous published work and investigated options to improve the marker placement repeatability of the heel marker on the calcaneus. Thirdly we created and validated a single score of dynamic foot function based on the OFM kinematics, the Foot Profile Score (FPS), offering a clinically meaningful interpretation of foot function relative to healthy controls, and to measure the decline or improvement in foot function over time or following intervention. For the primary aim we finally investigated the responsiveness of the FPS in children with cerebral palsy, hemiplegia, before and after foot corrective surgery. Our final study addressed both the primary and secondary aim. Based on the foundation of our previous four studies, we were able to compare the dynamic foot motion of children previously treated for clubfoot who were symptomatic and considering further management, to a control population. These children had a range of initial treatment including both conservative (Ponseti method) and surgery. In addition, we began to explore a potential relationship between dynamic foot function (FPS and plantar pressure) and perceived quality of life in children with residual clubfoot deformity.

In this final chapter the main findings of the presented studies are critically discussed and the clinical implications and ideas for future research are considered.

MAIN FINDINGS

Clinimetric testing of the Oxford Foot Model

In chapter II and chapter III, we conducted two repeatability studies for the OFM that were lacking in the literature and important to validate our methodologies for our other chapters.

Firstly, the repeatability of the OFM in clinical populations with foot deformity was compared to a typically developing population. The OFM has published repeatability in healthy adults and children but had limited testing in a population with deformity (10,12-14). Our results showed good intra-tester repeatability with the mean absolute difference in typically developing children at 4.8°, which improved to 4.1° in children with hemiplegia, and further improved to 2.9° in children with clubfoot. Inter-rater repeatability in children previously treated for clubfoot deformity had a mean difference between raters of 3.6 degrees. The mean absolute differences were within one degree for the intra and inter-rater reliability in 12/15 variables for the clubfoot data. Our results fall within the acceptable error of measurement suggested by McGinley et al (15) in three-dimensional gait analysis and are comparable to other published studies (13,16). Overall, the clubfoot data was the least variable which we attributed to a more experienced marker placer for the clubfoot data as well as improved technology.

Secondly, chapter III investigates the repeatability of placing the heel (HEE) marker when using the OFM. This study followed on from previous work investigating the hindfoot segment of the OFM. Paik and colleagues (17) showed the misplacement of the HEE marker induced the most change in the orientation of the anterior-posterior axis when compared to the other calcaneal markers, indicating the importance of ensuring the HEE marker is placed accurately. Two alignment devices (jigs) were created to optimise marker placement: a mould and a ratio caliper. The aim of this pilot study was to test these two jigs against the conventional method of eyeball marker placement, to improve marker placement repeatability of the HEE marker when using the OFM in 10 healthy adult subjects. We hypothesized that the ratio caliper and mould would not improve an experienced gait analyst's repeatability (as experience generally improves repeatability), but that they would improve an inexperienced gait analyst's repeatability as well as the inter-tester error.

Overall, the results were surprising and did not fully align with our hypotheses and discussion from chapter II. The ratio caliper produced the lowest intra-tester variability for both the experienced and inexperienced gait analyst. However, both ratio caliper and eyeballing yielded good inter-tester repeatability. The mould was the most variable for both analysts. We were surprised our results suggested the common practice of the eyeball method with palpation of bony landmarks for marker placement of the OFM was not improved significantly by using a jig, especially for the inexperienced gait analyst as hypothesised. The fact that we investigated a healthy control population instead of a population with foot deformity may have influenced the repeatability results of the gait analysts.

A recent study by Reay and colleagues (18) investigated marker placement repeatability of the OFM in 10 healthy adults, applied by three assessors not native to Oxford (physiotherapist, mechanical engineer training to be a clinical scientist, and Master of biomedical kinesiology), with no practical experience of the OFM (18). The authors agreed with our results- that despite varying experience with anatomy and marker placement, the OFM is repeatable in an adult healthy population (18). This strengthens the clinical utility of the OFM and supports its original design to be used with the eyeball method of marker placement. The authors do comment on a common trend where the transverse plane remains the least repeatable for all groups testing repeatability (10,12,18,19), likely due to the placement of the heel marker (16,18). This suggests that since the caliper did not significantly improve marker placement, and the mould actually worsened it, it may be worth considering further ways to improve the marker placement of the heel marker that haven't been considered as yet (17,18,20,21).

Development and responsiveness of the Foot Profile Score

In chapter IV, we introduced the FPS, a new summary score of dynamic foot motion during gait based on the Oxford Foot Model kinematics (10,22). The FPS was constructed similarly to the Gait Profile Score (GPS), a single measurement of the quality of an individual's overall gait pattern based on lower limb kinematics (23). The FPS was defined, then studied for its properties and design, and analysed against a clinical assessment of foot deformity during gait (clinical foot deformity score- CFDS). Our results showed a significant correlation between the FPS and CFDS with all 6 Foot Variable Scores contributing positively and independently to the prediction of the CFDS. Correlation between the FPS and the GPS was then investigated in both a total lower limb involvement population (neurological diagnoses such as cerebral palsy), and an isolated foot deformity population (clubfoot). This revealed a moderate correlation between the FPS and GPS in the lower limb involvement population, but no correlation was found in the group with isolated structural and dynamic foot deformity. This indicates the FPS represents new information that the GPS does not capture, especially in populations with isolated foot deformity.

As a single summary score, the FPS is more intuitive to clinicians who are not trained in interpreting three-dimensional kinematic graphs. As a single number, the FPS is a clinically meaningful outcome measure to identify the presence of deformity during gait, monitor change in foot function over time, and measure change following an intervention.

Following the validation process of the FPS, chapter V investigates the responsiveness of the FPS in a clinical population: children with cerebral palsy, spastic hemiplegia, before and after surgical correction of their foot deformity. The minimal clinically important difference (MCID) of the FPS was calculated from the regression of the FPS on the CFDS from chapter IV and defined as no more than 2.4 degrees. The difference in the FPS was then analysed using this MCID to indicate the success of surgical outcome. We also looked at potential relationships between the change in FPS and their pre-operative FPS, age at surgery, and time since surgery. Seventy-six percent of children had a change in their FPS greater than the MCID. A regression analyses only showed a clear relationship between pre-operative FPS and change in FPS indicating a greater degree of abnormality in the FPS prior to surgery means a greater scope of improvement following surgery. This confirms the FPS is a responsive outcome measure.

Foot function and perceived outcomes

The final study of this thesis is presented in chapter VI. This study aims to define the residual dynamic deformity in symptomatic children following initial correction of their clubfoot deformity compared to a healthy control population. The study group is further subdivided into children previously treated with the Ponseti method (conservatively) or with surgery. Foot function was assessed by the OFM kinematics, FPS, and plantar pressure and then correlated with parent-reported outcome measures to identify if there was a relationship between foot function and perceived disability. The subjective outcome measures included the Oxford Ankle Foot Questionnaire, the Disease Specific Index for clubfoot and the Pediatric Quality of Life Inventory 4.0. A secondary aim of this study was to investigate correlations between foot function during gait and perceived disability in this population. Interestingly, our findings suggest that older, symptomatic children following clubfoot treatment present with similar degrees of gait deviations and perceived disability regardless of whether they were treated with the Ponseti Method or surgery. The presence of sagittal and coronal plane hindfoot deformity (equinus and varus respectively) and coronal plane forefoot deformity (supination) were associated with higher levels of perceived disability, regardless of their initial treatment.

Reflecting on the findings of this final study, the gait deformities most linked with poor perceived outcomes are not unsurprising as they are a component of the original structural deformity. It is well accepted that recurrence rates are high even following a good initial correction, and can be as high as 40%

(24). In addition, when treating this population, an atypical or resistant clubfoot deformity can be present from birth, or it can be created if abduction of the foot is initiated before the cavus element of the deformity is fully corrected (25). These complex clubfeet are known to have high recurrence rates regardless of treatment type, leading to poor outcomes (25). Since publishing this paper, Grin and colleagues (26) have agreed with our findings. The authors investigated kinematic data during gait in children with relapsed clubfoot using the OFM and showed that forefoot adduction and forefoot supination were the kinematic biomarkers for relapsed feet (26) however they did not link their gait findings with perceived outcomes.

I am left with 2 questions. The first is: 'Why didn't the Ponseti cohort do better than surgery?' I do believe the answer to this question could be achieved by a larger international cohort study investigating older children with the full spectrum of outcomes, including fully corrected through to significant residual deformity, and asymptomatic through to symptomatic cases, using the same research methodology as used in our study. This would analyse the full breadth of deformity correction and what is achievable from a deformity correction and satisfaction stand-point.

The second question is trickier: 'Why were the subjective outcome measures used in our study *NOT* better correlated with each other, and with foot function during gait?'. This is despite two out of the three subjective measures used being created and validated in the clubfoot population. Does this bring in doubt the validity of the measures? Or (more likely) does this reflect the difficulty of relating the three domains of the ICF- body and structure to activity and participation? Foot structure *may* affect foot function and therefore walking capacity (amongst many other factors) … and walking capacity *may* affect quality of life (amongst many other factors) …. Therefore, much deeper insight into this dilemma is needed to be able to link structure, function and perception if we are hoping to improve subjective outcomes in our clinical populations.

METHODOLOGICAL CONSIDERATIONS

Study populations

A strong element to our methodology of four of the five studies in this thesis was the use of a wide age range (adults and children) as well as a variety of clinical populations including a congenital, idiopathic foot deformity (clubfoot) as well as foot deformities that develop over-time due to a neurological insult (cerebral palsy and acquired head injury). Chapter III would benefit from being repeated in a clinical population. Investigating the repeatability of marker placement using experienced and less-experienced gait analysts may be better illuminated when foot deformity is present. A neutral hindfoot in a healthy population could inherently improve intra-rater and inter-rater repeatability and likely masks the advantage of the superior knowledge of foot deformity that comes with experience in both clinical assessment of feet and marker-placement experience.

Modelling

Despite repeatability testing and validating kinematic outcome measures, our study results are only as good as the kinematic models in their current form. Constant improvement in technology and development of the models themselves are improving our ability to replicate and interpret human motion. One example of this may be markerless kinematic models- where we are no longer limited by placing markers on the skin over bony landmarks (27,28). The ISB (International Society of Biomechanics) recommendations have recently cited: 'Bi-planar video fluoroscopy methods either with invasively inserted intracortical markers or using marker-less tracking have shown great potential … but are at present limited by the radiation exposure, equipment and personnel costs, and tracking volumes.' (29).

Study design

As stated earlier, our study populations are widely representative of those who commonly access a three-dimensional gait laboratory for clinical management decisions. However, our studies all included convenience sampling, as opposed to being representative of those who are living in society with varying levels of foot deformity. Chapter IV was the first study in the literature to investigate children with ongoing, symptomatic clubfoot deformity. Following the results of this paper, the findings may be strengthened by looking at the full range of children post clubfoot treatment, including those who have good functional results and are subjectively pleased with their outcomes. This may provide thresholds of deformity that are problematic/ or not problematic, especially in the longer term. The knowledge of when not to treat can be just as important as knowing when to treat; especially in populations we know can have disastrous long-term results due to stiff and painful feet.

CLINICAL IMPLICATIONS

The Oxford Foot Model (OFM) was originally designed to measure dynamic motion of the tibia, hindfoot and forefoot in three-dimensions in a clinical setting where anatomical abnormalities of the foot structure is common (10,12). The primary aim of this thesis was to establish the clinical meaningfulness of the OFM to assess foot deformity during gait. Through the course of this thesis, we have proven the OFM is repeatable in populations with known foot deformity; whether the foot deformity was congenital, acquired, or secondary to abnormal neurology. We have established the eyeball method of marker application is the most repeatable over available anatomical alignment devices, for both experienced and inexperienced marker placers. The Foot Profile Score (FPS) - a single summary score of the OFM kinematic data- was defined and validated as an outcome measure of foot structure and dynamic function. The FPS was then tested for its responsiveness in a clinical population with a convincing result. Finally, the OFM was key in determining differences in foot function between children with clubfoot treated initially with the Ponseti Method or surgery, compared to a healthy population. Our secondary aim was to then correlate these gait findings with perceived outcomes, highlighting which residual deformities are more likely to be associated with poor outcomes in this population.

Over the span of the thesis, the included published work has been cited numerous times. The clinimetric testing has been cited in review papers of foot modelling (30), the development of new outcome measures for clubfoot deformity (31), further repeatability studies (18,32) and ISB recommendations for skin-marker-based multi-segment foot kinematics (29), which also cited the FPS validation study. The FPS validation paper has been further cited in outcome-based studies (26,33) and in a systematic review of assessing foot-types (34). The study analysing foot function and perceived outcomes in symptomatic clubfoot has been cited in review papers of clubfoot management (35,36) and outcome-based studies (26,37).

FUTURE RESEARCH

The aims of the overall thesis span many disciplines from engineering to orthopaedics to social-based medicine. Therefore, the potential future research recommendations are wide and exciting:

- Improving the modelling of the foot during three-dimensional gait analysis. We have proven the OFM *is* repeatable and clinically meaningful in analysing foot function during gait, including pre-post surgical intervention. However, as technology improves, so must our modelling techniques. Therefore, it is likely improvements can and will be made to the OFM over time.

- A rigorous investigation of the minimal clinically important difference (MCID) for the Foot Profile Score (FPS). In Chapter V, an MCID of no more than 2.4 degrees was calculated based on the validation results. However, a prospective trial to rigorously test the MCID is warranted as a lower value may be indicative of a clinically meaningful improvement.

- A larger, international, multi-centre clubfoot study based on the same methodology as Chapter VI; to further investigate what element of the clubfoot deformity correlates to poor perceived outcomes. To understand thresholds of *how much* deformity leads to poor outcomes, a wider clubfoot population is needed – those with good results through to very poor results. This may be the only way to understand which residual deformities are best to target with further treatment, and which ones are better left alone.

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About the Author

CURRICULUM VITAE

Jennifer Leigh McCahill was born on April 4th, 1975, in London, Ontario, Canada. She graduated with a Bachelor of Science (Physical Therapy) from the University of Toronto, Canada in 1998. Following graduation, she travelled to Australia and then Europe on working holidays. Jennifer returned to Australia and completed a Masters of Physiotherapy (Paediatrics) at the University of Melbourne, in 2004 while working at the Royal Children's Hospital. Following her Masters, she left Australia and worked at Sinai Hospital in Baltimore, USA, treating children with clubfoot following training in Iowa with Dr Ponseti.

In 2006, Jennifer migrated to the United Kingdom on a work-sponsorship visa for the Oxford Gait Laboratory. Alongside her work in motion analysis, she was seconded to develop a Ponseti clinic at the Royal Berkshire Hospital in Reading. From 2006 to 2013, Jennifer taught on clubfoot courses internationally (Egypt, USA, Bangladesh, Zimbabwe) in addition to her work in Oxford. She also undertook a job-share position, Managing the Children's Physiotherapy Service in Oxfordshire for five years from 2009-2014. In 2015, Jennifer formally enrolled at VU Medical Centre to complete her PhD via publication.

Alongside her contracted roles, Jennifer has been the treasurer, and is currently the chairperson of the Clinical Movement Analysis Society of the UK & Ireland. She has also held the position of Evidence-Base Officer for the

Association of Paediatric Chartered Physiotherapists, a professional group under the Chartered Society of Physiotherapists in the UK.

In July 2016, Jennifer undertook the most challenging, and rewarding, role of her life, becoming a mother to Alfred James McDonnell. Georgia Leigh McDonnell was quick to follow in December 2017.

In January 2020, Jennifer became a part-time lecturer at University College London, in Advanced Paediatric Physiotherapy Studies. In April 2021, Jennifer gave up her permanent position in the Oxford Gait Laboratory and began an honorary contract to continue her research. She continues to run a successful, private physiotherapy business, and is about to launch a new venture: the Watlington Wellness Centre in her hometown.

LIST OF PUBLICATIONS

Articles

1. * **McCahill J**, Stebbins J, Prescott R, Harlaar J, Theologis T. Responsiveness of the Foot Profile Score in children with hemiplegia. Gait Posture. 2022: 95;160-163.
2. * **McCahill J**, Schallig W, Stebbins J, Prescott R, Theologis T, Harlaar J. Reliability testing of the heel marker in three-dimensional gait analysis. Gait Posture. 2021 Mar;85:84-7.
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1. **McCahill J**, Stebbins J, Theologis T, Harlarr J, Lavy C. Pressure distributions in symptomatic clubfoot. Volume 73, Supplement 1, September 2019, Pages 175-176.
2. **McCahill J**, Stebbins J, Lewis A, Prescott R, Harlaar J, Theologis T. Validation of the Foot Profile Score. Gait Posture. 2018;65.
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Contributions to (inter)national scientific conferences:

Oral Presentations

1. **McCahill J**, Stebbins J, Prescott R, Harlaar J, Theologis T. Responsiveness of the Foot Profile Score in children with hemiplegia. CMAS conference May 2022 Edinburgh, UK
2. **McCahill J**, Stebbins J, Theologis T, Harlaar J, Lavy C. Correlation of structural abnormalities and quality of life in children with clubfoot. EPOS conference April 2019 Tel Aviv, Israel
3. **McCahill J**, Stebbins J, Theologis T, Harlaar J, Lavy C. Pressure distributions in symptomatic clubfoot.
   ESMAC conference September 2019, Amsterdam, Netherlands
4. **McCahill J**, Stebbins J, Lewis A, Harlaar J, Theologis T. Correlation of the foot profile score and gait profile score.
   ESMAC conference September 2018, Prague, Czech Republic
5. **McCahill J**, Stebbins J, Koning B, Harlaar J, Theologis T. Repeatability of the Oxford Foot Model in children with foot deformity.
   CMAS conference April 2016, Belfast, Ireland

Poster Presentations

1. **McCahill J**, Stebbins J, Lewis A, Prescott R, Harlaar J, Theologis T.
   Validation of the Foot Profile Score.
   ESMAC conference September 2018, Prague, Czech Republic
2. **McCahill J**, Stebbins J, Theologis T, Harlaar J.
   Repeatability of the Oxford Foot Model in children with clubfoot.
   MOVE conference, February 2016, Amsterdam, Netherlands

Grants and Prizes

CMAS conference April 2016, Belfast, Ireland- best paper prize.

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Since beginning this journey in January 2015, my life has completely changed. In January 2015 I had been married to Andrew David McDonnell for just one year, and we had recently purchased our ‘forever home’ which has required extensive landscaping and building work to become our beautiful family home. Since this time, we have also endured two IVF cycles (one failed/ one successful), two healthy pregnancies, and the birth of our two beautiful children: Alfred James McDonnell on 12.07.2016 and Georgia Leigh McDonnell on 5.12.2017. Unfortunately, also during this time, I have lost both of my parents. My mother unexpectedly died in a tragic accident at home in Canada in July 2015, and more recently in August 2022, my father passed away in my arms after a battle with vascular dementia. It was actually at his hospital bedside in London, Ontario, Canada, where I ‘finished’ my thesis ….

Thank you doesn’t seem to be enough for the people who have inspired me and kept me going through the past 8 years, both personally and professionally. Naturally I will start with my family: my wonderful **husband** and **children**, for sacrificing time with me and putting up with ‘grumpy mummy’. **Susan** (my mother-in-law), **Salome** and **Jayne** for keeping our children blissfully entertained while mummy was working. My **sisters** (Tracy and Lisa) – thank you for supporting me from afar. **Karen & Fred and Tom**, for supporting me and my family like we were your own. **My closest friends** (you know who you are) for supporting me and never judging. **Jane**, for being my NHS job-share, co-director, confidant, and a true friend. **Helen**, **Roz, Charlotte and Emma**, for helping me navigate motherhood and becoming a part of my UK family. **Brenda**, for being the other 'half' of me- our friendship is one of the foundations of my life.

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What would my professional life look like if I had stayed in Melbourne…?

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*"It's worth remembering that it is
often the small steps, not the giant leaps,
that bring about the most
lasting change."*
Queen Elizabeth II