

Equinovarus in Cerebral Palsy

BY

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THESIS

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This thesis is dedicated to my father Joseph “Dusty” Krzak

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LIST OF ABBREVIATIONS

CP	Cerebral Palsy
CS	Change Score
DF	Dorsiflexion
EMG	Electromyography
ER	External Rotation
EV	Eversion
IC	Initial Contact
IR	Internal Rotation
IV	Inversion
GC	Gait Cycle
MFM	Milwaukee Foot Model
PC(A)	Principal Component (Analysis)
PF	Plantar Flexion
PFR	Plantar Fascia Release
PTL	Posterior Tibialis Lengthening
ROM	Range of Motion
SPLATT	Split Transfer of the Anterior Tibialis
SPOTT	Split Transfer of the Posterior Tibialis
TAL	tendo Achilles Lengthening
3-D	Three Dimensional

SUMMARY

The basic objective of locomotion is to move the body from its initial position to a new location. Locomotion consists of a repetitive sequence of multisegmental lower extremity movements functioning to achieve both mobility and stability. The foot and ankle are integral components in human locomotion. Twenty eight bones, 33 joints, and hundreds of muscles and ligaments serve as the primary interface between the individual and the support surface allowing for mobility to absorb forces at the point of initial contact and stability as the body advances during stance phase.

Studying the contributions of the foot and ankle to locomotion in individuals with neuromuscular disorders provides vital insight into how individuals control locomotion and associations between neural impairment and movement dysfunction. For example, studying children with equinovarus secondary to hemiplegic cerebral palsy (CP) provides an association between motor dysfunction of distal musculature and alterations in locomotor kinematics common to this population.

Equinovarus foot and ankle deformity is most common in children diagnosed with hemiplegic CP. A child with equinovarus is limited in gross motor functional skills which can lead to further disability. The cause of this deformity is believed to result from a combination of neuromuscular and biomechanical impairments secondary to an insult to the developing central nervous system. Non-phasic activity of the extrinsic musculature surrounding the foot and ankle create muscular imbalance, atypical positioning and motion of the foot and ankle during locomotion.

Common surgical interventions used to correct an equinovarus foot include musculotendinous lengthenings of the ankle plantar flexors and/or posterior tibialis, transfers of the anterior and/or posterior tibialis, osteotomies to realign the ankle and foot, or arthrodosis to fuse instable joints. These procedures are determined based on a clinician's experience, physical examination,

electromyographic (EMG) data, and/or quantitative gait analysis. Decisions based solely on one of these particular criteria can result in postoperative failure as utilizing an individual's experience or certain physical examination measures are highly subjective and inconsistent. EMG patterns are highly variable, and the methods for attaining the data are prone to criticism. Furthermore, commonly used gait models simplify the foot as one rigid segment providing inadequate data to either identify where specific gait deviations occur within the foot or quantitatively describe changes resulting from surgery. A more complex, segmental foot model is needed to properly track subtle foot motion and positioning during gait.

This dissertation provides fundamental insights into the segmental foot and ankle kinematics during locomotion of children with equinovarus secondary to hemiplegic CP. It will further quantitatively examine the ability of orthopedic surgical intervention to create a more neutral postoperative foot and ankle. This dissertation consists of three experiments. The first experiment looked at the impact of invasive EMG analysis on locomotion. Specifically, we examined temporal-spatial parameters and triaxial hindfoot kinematics with and without the presence of a fine wire electrode inserted into the posterior tibialis. No significant differences in any temporal-spatial parameters or hindfoot kinematics were observed between “with wire” and “without wire” conditions. Our findings suggest that the simultaneous collection of segmental foot and ankle kinematics and fine wire EMG data of the posterior tibialis is acceptable for surgical decision making in this population.

The second experiment used systematic and quantitative methods to identify clinically relevant subgroups that exist among a sample of typically developing children and children with equinovarus due to hemiplegic CP. We used a combination of segmental foot and ankle kinematics, principal component analysis (PCA), and *K*-means cluster analysis to examine the gait deviations associated with

the equinovarus foot. PCA was able to reduce a list of forty clinically relevant variables to six variables that were independent from each other and described the location and plane of involvement in the foot and ankle. Cluster analysis identified five clinically recognizable subgroups with unique segmental involvement, planar motion, and range of motion (ROM).

The third experiment quantitatively described changes in segmental foot and ankle motion during walking that resulted from surgical soft tissue balancing procedures for equinovarus. We used PC scores obtained in Experiment 2 to demonstrate that post-operative foot and ankle kinematics shifted towards those of a control group with a neutral hindfoot and forefoot. PC scores were additionally used to identify whether individuals presented with corrected, partially corrected, or uncorrected post-operative gait deviations. Characteristics such as pre-operative cluster membership and the combination of surgical techniques chosen for correction were then used to explain the presence of uncorrected post-operative gait deviations. Individual cases of uncorrected deviations were attributed to two causes: (1) the specific segmental deformity was not identified pre-operatively, and therefore not surgically addressed, and (2) the deformity was so severe that a soft tissue balancing procedure could not correct it. There was no significant effect of surgery on walking velocity. One significant association was identified between PC5 (Sagittal plane hindfoot ROM) and walking velocity demonstrating that increases in walking velocity were associated with increases in sagittal plane hindfoot range of motion. The results demonstrated that surgical soft tissue balancing procedures result in more neutral hindfoot and forefoot gait kinematics in youth with equinovarus secondary to hemiplegic cerebral palsy and outcomes were not attributed to alterations in walking velocity.

CHAPTER 1

1.0 INTRODUCTION

1.1. Organization of the Dissertation

The overall purpose of this dissertation is to quantitatively evaluate the segmental foot and ankle gait kinematics of equinovarus secondary to hemiplegic cerebral palsy (CP) using the Milwaukee Foot Model (MFM). Chapter 1 reviews previous work that explains requirements of normal locomotion and the role of the foot and ankle in supporting these requirements. Next, I will review the literature on CP and how common impairments associated with CP impact the foot and ankle during locomotion. I will then provide an overview of quantitative assessment techniques used for surgical decision making including 3D gait analysis, segmental foot and ankle kinematic analysis, identifying kinematic subgroups, and electromyography (EMG). Lastly, I will review common surgical techniques used to correct equinovarus and the post-operative outcomes reported in the literature. Chapter 2 investigates the effect of insertion of a fine wire electrode into the posterior tibialis on the gait pattern of children with equinovarus secondary to hemiplegic CP. Chapter 3 investigates the identification of clinically relevant kinematic subgroups among typically developing children and children with equinovarus. Chapter 4 investigates the effect of surgical soft tissue balancing procedures commonly used for equinovarus on hindfoot and forefoot kinematics during gait. All experiments examined segmental foot and ankle kinematics during locomotion using the MFM. Lastly, Chapter 5 summarizes the findings of this dissertation.

1.2. Biomechanical Requirements of Normal Locomotion

According to Perry and Burnfield (2010), during locomotion each weight-bearing limb accomplishes four distinct functions: (1) upright stability which is maintained despite an ever-changing

posture, (2) progression generated by the interaction of selective postures, muscle force, and tendon elasticity, (3) minimize impact with the support surface at the onset of each stride, (4) energy is conservation when these functions are being performed in a manner that reduces the amount of muscular effort required. The simultaneous accomplishment of these four functions depends on distinct motion patterns which represent a complex series of interactions between the upper and lower body. The foot and ankle play an integral role in these functions acting as the principal interface between the individual and the support surface, as well as a mechanical link to more proximal segments.

During locomotion, the foot is required to perform a number of intricate functions allowing for both stability and mobility while minimizing energy expenditure. Stability throughout the foot and ankle requires the functional integration of proprioceptive feedback, joint mobility, and muscle control. While the foot is in contact with the support surface during locomotion, referred to as stance phase, it propagates rotation of more proximal limb segments and minimizes effort of more proximal muscles by providing a stable base during forward progression (Morris 1977). Flexibility of the structures within the foot allows it to absorb forces at initial contact with the support surface and accommodate to alterations in terrain.

The presence of biomechanical and/or neuromuscular foot and ankle dysfunction can impact one or all of the above requirements for normal locomotion. Stebbins et al. (2010) demonstrated that atypical foot and ankle motion and subsequent surgical correction impacted motion of the more proximal segments of the lower extremities during locomotion. Ballaz et al. (2010) showed moderate correlations of ankle kinematic peaks and excursion of motion with energy expenditure during locomotion. The populations used in each of these studies presented with foot and ankle dysfunction

resulting from one of the most prevalent neuromuscular diseases affecting motor control in children and adolescents, cerebral palsy.

1.3. Cerebral Palsy

“Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behavior; by epilepsy, and by secondary musculoskeletal problems” (Rosenbaum et al. 2007). Impairments within the neuromuscular and musculoskeletal systems commonly contribute to mild to severe limitations in functional mobility including locomotion (Boyd et al. 2001). These deficits in functional mobility can lead to further difficulties participating in educational or recreational activities at home or in the community.

1.3.1. Prevalence and Etiology

CP is considered the most prevalent physical disability of childhood with an incidence of 3.6 per 1000 children (Yeargin-Allsopp et al. 2008). The prevalence had risen in the 1990's with a significant increase in two of the primary risk factors for developing CP (survival rate of preterm births and multiple gestations). It is an extremely heterogeneous disorder with various pathological mechanisms of injury resulting in numerous impairments on body systems and limitations in function. Spastic CP is considered to be the most common clinical subtype with incidence of 72%-91%. Among the children diagnosed with spastic CP, 21%-40% were clinically diagnosed as hemiplegic (Odding et al. 2006).

Perinatal vascular incident, i.e. stroke, is considered the leading single cause of hemiplegic CP. Stroke associated with hemiplegia can either be hemorrhagic or ischemic. The causative mechanisms have been extensively investigated but still remain controversial. Fetal distress (adjusted odds ratio >10) and post-maturity (adjusted odds ratio = 7.6) have both been identified as independent predictors of perinatal hemorrhagic stroke (Armstrong-Wells et al. 2009). It was postulated throughout the 1970's and 80's, that arterial blood pressure was associated with cerebral blood flow. In a typically developing infant, as arterial blood pressure increased the body was able to autoregulate the amount of cerebral blood flow. However, in a pre-term infant it was postulated that the ability to autoregulate cerebral blood flow during episodes of increased arterial blood pressure (for example, during fetal distress) was either absent or impaired. Therefore, hypertensive episodes would subsequently increase cerebral blood flow and result in hemorrhage. Another, more current, theory regarding the etiology of hemorrhagic infarction focused on the impact of arterial carbon dioxide levels on blood vessel diameter and cerebral blood flow. Specifically, it was observed that episodes of hypercapnia were associated with vasodilation (Greisen 2009). Neonates who presented with hypercapnia were identified as being at higher risk for hemorrhagic stroke (Zhou and Liu 2008). Furthermore, children and adolescents who presented with hypertension and were subsequently exposed to increased levels of carbon dioxide had diminished cerebrovascular reactivity (Wong et al. 2011).

Ischemic perinatal stroke is defined as, “a group of heterogeneous conditions in which there is focal disruption of cerebral blood flow secondary to arterial or cerebral venous thrombosis or embolism, between 20 weeks of fetal life through the 28th postnatal day, confirmed by neuroimaging or neuropathic studies” (Raju et al. 2007). Ischemic perinatal stroke is considered the leading single cause of CP and accounts for 30% of children diagnosed with hemiplegia (Wu et al. 2006). A multitude of

both maternal and fetal causative mechanisms of thrombophilia exist that predispose a fetus to ischemic perinatal stroke. On the fetal side, plasma/protein based and congenital prothrombotic defects, infections, asphyxia, congenital heart disease, hypoglycemia, and polycythemia all increase risk for thrombotic episodes. These episodes can potentially lead to embolism in the fetal brain secondary to the patency of the foramen ovale and the right-to-left direction of blood flow in the fetal ductus arteriosus (Raju et al. 2007). On the maternal side, pregnancy itself is considered a state of hypercoagulability, and therefore a natural prothrombotic state (Jaigobin and Silver 2000). Expectant mothers can have conditions such as preeclampsia, autoimmune disorders, inherited thrombophilias, or infection that predispose them to a prothrombotic state. Drug abuse and infertility interventions also predispose mothers to thrombus development (Raju et al. 2007).

1.3.2. Pathophysiology

Perinatal stroke disrupts the natural blood flow to cortical and subcortical structures resulting in a cascade of biochemical responses that ultimately kill preoligodendrocytes and develop cystic changes in the cerebral areas that contain the sensory and motor tracts. Following stroke, there is a release in glutamate, oxidative free radicals, and inflammatory cytokines which inhibit the ability of preoligodendrocytes to proliferate, differentiate, and form myelin around the axons of sensory and motor tracts (Khwaja and Volpe 2008). Myelination of axons is incredibly important in periventricular white matter as it allows for fast and effective conduction of action potentials within the sensory and motor tracts. The impact of damage to these tracts has become more apparent with the advent of more sophisticated imaging techniques. Diffusion tensor imaging has become incredibly useful in understanding how microstructural damage to these tracts ultimately results in the clinical manifestation associated with CP especially when conventional magnetic resonance imaging has not shown an identifiable lesion (Glenn et al. 2007). Diffusion metrics (including number of fibers within

a tract, tract fractional anisotropy, transverse diffusivity, and mean diffusivity) have been analyzed in both motor (corticospinal) tracts and sensory (posterior thalamic radiation) tracts in children with CP. Correlations have been identified among diffusion metrics of primarily posterior thalamic radiation, clinical measures of sensation (fine touch and proprioception) and strength (Hoon et al. 2009). Other studies have identified correlations among diffusion metrics and functional severity (Gross Motor Functional Classification Level) in children with CP (Trivedi et al. 2010; Yoshida et al. 2010). Although the initial lesion within the brain is non-progressive, the signs, symptoms, and functional limitations have been known to worsen in individuals as they get older, especially in children who initially present with greater limitations in function (Hanna et al. 2009).

1.3.3. Symptoms of Cerebral Palsy and their Impact on the Foot and Ankle

In hemiplegic CP, the unilateral hemispheric insult results in a disorder of posture and movement on the contralateral side of the body. A task force on pediatric movement disorders established by the American Academy of Cerebral Palsy and Developmental Medicine, developed a systematic framework of movement disorders associated with children with CP (Sanger et al. 2003; Sanger et al. 2006; Deon and Gaebler-Spira 2010; Sanger et al. 2010). The three categories within the framework included: (1) Hypertonic: excessive tone or resistance to passive stretch while attempting to maintain a relaxed state of muscle activity, (2) Hyperkinetic: unwanted excessive movement, and (3) Negative Signs: insufficient muscle activity or insufficient control of muscle activity. Any combination of these clinical symptoms can result in atypical posturing and movement, characteristic locomotor (gait) deviations, and an increase in energy cost of 1.3 times that of aged-matched peers during walking and gross motor tasks (van den Hecke et al. 2007).

In a retrospective review of 492 children with CP, Wren et al. identified the most common gait deviations in this population using quantitative gait analysis (Wren et al. 2005). Overall, equinus was

present in 64% and intoeing was present in 54% of children with CP. Equinus was defined as having an ankle plantar flexion angle > 1 standard deviation of the mean for typically developing children during stance phase, and intoeing was defined as having an internal foot progression angle > 1 standard deviation of the mean. Without previous surgical intervention (N=84), 70% of children with hemiplegic CP presented with equinus and 58% presented with intoeing. There are multiple segments throughout the lower extremity, proximal and distal, that can contribute to intoeing. In a follow-up study of 412 children with cerebral palsy (587 involved sides) by Rethlefsen et al. (2006), two of the leading causes of intoeing in children with hemiplegia (N=82) included pes varus and metatarsus adductus. Pes varus is the inversion of the hindfoot with or without forefoot supination during stance and/or swing and was observed in 43% of children with hemiplegia. Metatarsus adductus is the adduction of the forefoot without associated varus seen in 26%. The segmental contributions of ankle equinus, hindfoot varus, and forefoot adductus collectively result in a characteristic foot and ankle deformity in children with hemiplegic CP, equinovarus. Furthermore, these studies have demonstrated the importance of quantitative gait analysis for objectively identifying the segmental contributions of the foot and ankle to gait deviations in this population.

1.4. Evaluation of Foot and Ankle Deformity

1.4.1. Kinematic Modeling during Locomotion

Quantitative gait analysis typically includes collection of temporal-spatial, kinematic, kinetic and electromyographic (EMG) data during locomotion. Quantitative gait analysis has been extensively utilized across ages and pathologies to guide clinicians' decisions about appropriate interventions, providing objective data about the efficacy of orthopedic surgery, and longitudinally tracking the progression of various disease processes (Wren et al. 2011). A common method for measuring kinematics during quantitative gait analysis includes using three, spherical, non-collinear, retro

reflective markers placed over specific bony prominences to identify a segment in three-dimensional space. The markers are referred to as “passive” because they serve to reflect near-infrared light that is emitted from the periphery of special cinematographic cameras. At a minimum, two cameras are required to track the reflections, i.e. the markers/segments, in three-dimensional (3-D) space as a function of time. The kinematics are reported in degrees of motion and time is normalized to 100% of the gait cycle from initial contact to ipsilateral initial contact. The most commonly tracked segments include the pelvis, thigh, shank, and the foot (Davis et al. 1991).

It is commonly assumed that alterations in kinematic, kinetic and EMG data result in a less efficient gait. Investigators ultimately examined the relationship between gait efficiency and lower limb kinematics during walking (Ballaz et al. 2010). Assessing adolescents with CP, the authors compared a calculated energy expenditure index, based on the linear relationship between heart rate and oxygen consumption, to peaks of sagittal plane joint kinematics during locomotion. Strong correlations existed between energy expenditure index, ankle range of motion ($r = -0.70$, $p < 0.02$) and peak plantar flexion ($r = 0.74$, $p < 0.05$) while walking. These findings suggest that ankle motion during gait is considered a key kinematic factor in gait efficiency in adolescents with CP.

For decades, the accepted approach to quantify foot and ankle kinematics during locomotion has been to represent the entire foot as a single rigid body with a revolute ankle joint (Davis et al. 1991). The proximal and distal segments are identified as the tibia and the foot with surface markers placed on the lateral shaft of the tibia, lateral malleolus, and in-between the second and third metatarsals. At the beginning of data collection, a static standing trial is collected to create additional “virtual” markers located within the knee and ankle joint centers. These markers are created using the location of the known surface markers along with subject-specific anthropometric measurements and function to identify the segment in three-dimensional space. Thus, this process serves to mathematically link the

surface markers with the underlying skeletal anatomy and can be referred to as neutral referencing (Rankine et al. 2008). The motion of the foot during walking (dynamic) trials is based on a vector that passes from the calculated ankle joint center through the marker placed over the second and third metatarsals. The plantar surface of the foot provides a reference for the sagittal plane alignment of this vector, assuming a planatargrade foot. The ankle center and toe marker are intended to provide references for transverse plane alignment.

Ultimately, the foot and ankle kinematics calculated using this method are two-dimensional and consist of plantar/dorsiflexion in the sagittal plane with the position of the foot measured in reference to the tibia, foot rotation in the transverse plane in relation to the tibia, and foot progression angle which describes the transverse plane orientation of the foot in relation to the global coordinate axes (Davis et al. 1991). This process of measuring motion, more specifically rotations, of a distal segment (foot) in relation to the more proximal segment (tibia) is performed through a series of limb rotation algorithms calculated using Euler angles with a y-x-z rotation sequence (Kadaba 1990).

It is well accepted that referring to the “foot” biomechanically as a single segment is an oversimplification of a structure that contains 28 bones, intricate articulations, and hundreds of ligaments and muscles (Nester 2009). Utilization of a single segment model to represent foot motion during gait can potentially neglect to identify deformity within the foot complex. Also, even when abnormalities are identified within the foot and ankle segments, it is not possible to isolate the problem to a particular joint. Thus, to understand the complexities of the of the foot and ankle during gait, a more sophisticated model that describes segmental foot and ankle motion would provide improved quantitative data and more insight into the etiology of various foot deformities.

A potential criticism of using 3-D motion capture technology with passive surface markers to track segmental kinematics of the foot and ankle is that there may be a significant amount of skeletal motion

occurring under the skin's surface that can fail to be accounted for using this technique. Nester et al. (2007) assessed the capability of surface markers to accurately track skeletal motion on cadaver limbs by comparing kinematics collected using three techniques: (1) markers attached directly to the skin, (2) markers attached to rigid plates mounted on the skin, and (3) markers attached to bone pins. The authors determined that there was not a critical difference between locomotive kinematics between the three different techniques. Thus, the utilization of passive marker sets to develop a biomechanical model of the foot and ankle during locomotion should be considered an appropriate method.

Rankine et al. (2008) identified over twenty-three published segmental foot and ankle kinematic models that provide redundant information regarding motion of foot and ankle segments during locomotion. These models divide the foot and ankle into multiple segments that more accurately represent the motion of the foot as opposed to the single segment, rigid body foot model typically used. Factors that distinguish the models include the number of segments used to represent the foot and ankle (ranging from two to ten segments), methods used for mathematically linking the surface marker sets to the underlying skeletal anatomy (i.e. neutral referencing), and methods used to calculate intersegmental angles (Euler angles or the Joint Coordinate System) (Grood and Suntay 1983; Rankine et al. 2008). For example, the Oxford Foot Model is a four segment foot and ankle model (tibia, hindfoot, forefoot, and hallux) that has been adapted for use in children (Carson 2001; Stebbins et al. 2006). An external technical frame is used to standardize the foot and ankle position during the static data collection trial for the purpose of neutral referencing. Intersegmental kinematics are calculated with the segments represented in a distal relative to the next proximal segment relationship using an Euler System.

Another biomechanical model has been developed, referred to as the Milwaukee Foot Model (MFM), and has been validated for use in the pediatric population (Kidder 1996; Myers et al. 2004).

The MFM provides kinematic data of four foot and ankle segments: tibia, hindfoot, forefoot and hallux. Unique to the MFM is the use of weightbearing, roentographic offset measurements in anterior/posterior, lateral, and a unique hindfoot coronal view for the purpose of neutral referencing (Johnson 1999). During neutral referencing, data collection from the static trial is used to mathematically link the orientation of the surface markers to the underlying skeletal anatomy. This becomes problematic when modeling segments such as the hindfoot. Reliable bony landmarks do not exist on the calcaneus to adequately represent the coronal, and sagittal plane orientation of the hindfoot segment. As a result, the MFM uses the measurements obtained from the radiographs to reorient the embedded segmental local coordinate axes to the orientation of the underlying skeletal segments. These radiographic measures are extremely important to the calculation of not only the offsets of the individual segments but also affect the kinematics of other segments and planes of motion (Long et al. 2008). The importance of utilizing this type of technique becomes more apparent when dealing with significant foot deformities found in the pediatric populations where neutral alignment may be impossible to obtain: talipes equinovarus (clubfoot), Charcot-Marie-Tooth, and planovalgus/equinovarus associated with CP. To calculate intersegmental angles, the segmental kinematics are expressed with the tibia referenced to the global coordinate axes. The remaining segments are represented in a distal relative to the next proximal segment relationship using an Euler System. The order of rotations selected for calculation of three-dimensional motion is sagittal, coronal, and then transverse.

1.4.2. Data Reduction and Identification of Kinematic Subgroups

Given the complexity of foot and ankle kinematics during locomotion in children with CP, the MFM produces a large amount of additional data to integrate into the decisions regarding appropriate interventions and measuring the effectiveness of those interventions. The challenge of integrating foot

and ankle kinematics into clinical care then becomes developing effective means of reducing the large amount of heterogeneous data. In whole-body gait applications, previous researchers have developed clinically relevant classification schemes of whole-body gait patterns for individuals with CP. For example, Rodda et al. developed a commonly used classification scheme of gait patterns for children with spastic diplegia using sagittal plane kinematics of the pelvis, hip, knee, and ankle. The subgroups included true equinus, jump gait, apparent equinus, crouch gait, and asymmetrical gait (Rodda et al. 2004). Similar techniques were used by Winters et al. (1987) to identify subgroups of sagittal plane kinematic patterns in children with hemiplegia. Types I-IV hemiplegic gait patterns were based on the amount of distal to proximal involvement of the affected limb. Children with a Type I pattern presented with isolated distal involvement resulting in persistent ankle plantar flexion during the swing phase of gait. Membership to subsequent types was assigned as involvement progressed to the more proximal segments. Thus, children with a Type IV pattern presented with the gait deviations of all the preceding groups (I-III) at the ankle and knee along with deviations extending proximally to the hip and pelvis. The intent was to use these gait classification schemes so that patients can be evaluated and fit into a particular subgroup which subsequently facilitates decision making regarding treatment. However, these previous classifications were often not systematically generated and were derived using qualitative pattern recognition. Additionally, these schemes only used sagittal plane kinematics derived from the whole-body gait model which lacks the complexity necessary to distinguish among the subtle soft tissue imbalances of the ankle plantar flexors and subtalar invertors that result in equinovarus. A quantitative means of classifying gait patterns based on 3-D segmental foot and ankle kinematics is needed to identify subgroups within the hemiplegic CP population with equinovarus.

Recently, principal component analysis (PCA) techniques have been used successfully to develop quantitative gait classification schemes that reduce the large amounts of gait data into a few critical

independent variables (Shemmell et al. 2007; Carriero et al. 2009). This technique takes kinematic/kinetic data points over a time series or an assemblage of relevant gait parameters and transforms them into a smaller number of independent variables. Once these data are reduced, statistical cluster algorithms have been used to identify clinically recognizable subgroups of homogeneous individuals. Multiple clustering techniques are available using hierarchical and non-hierarchical procedures. *K*-means clustering is a non-hierarchical technique that has previously been used as an effective method to identify groups of gait patterns in children with CP (O'Byrne et al. 1998; Kienast et al. 1999; Rozumalski and Schwartz 2009). This mathematical clustering algorithm assigns membership to a group based on the proximity to means maximizing the similarities within the groups and the differences among the groups. The combination of PCA and *K*-means clustering on segmental foot and ankle kinematics in children with equinovarus could be used to systematically identify clinically relevant subgroups. This methodology would result in a classification scheme that would allow segmental foot and ankle kinematics to be seamlessly integrated into the decision making process regarding appropriate interventions and measuring the effectiveness of those interventions.

1.4.3. Electromyography during Locomotion

The use of electromyographic (EMG) activity patterns for clinical decision making in the lower extremities of children with CP has become commonplace, and more recently it has been recommended to assist with understanding the equinovarus foot. EMG data provide information regarding the timing of neuromuscular activity during the gait cycle. A combination of surface electrodes for the more superficial musculature (anterior tibialis and gastrocnemius) and fine wire electrodes for deeper musculature (posterior tibialis) is used to determine the primary neuromuscular contributor(s) of the deformity. The posterior tibialis is a significant source of varus as it has the longest lever arm among the ankle inverters (Piazza et al. 2001). In typical gait, posterior tibialis

activity occurs throughout the midportion of stance phase to control hindfoot medial-lateral instability during a period of unilateral support. In conditions such as hemiplegic CP, atypical electromyographic activity patterns of the tibialis posterior creating equinovarus have been described as continuous firing throughout stance and swing, activity in swing, or reversal of phase where activity occurs when antagonistic anterior tibialis should be active (Scott and Scarborough 2006). Activity of the anterior tibialis, also a powerful ankle inverter, typically occurs immediately following heel strike to allow controlled approximation of the foot with the support surface and during swing phase to facilitate toe clearance. Children with hemiplegic cerebral palsy may demonstrate atypical anterior tibialis patterns characterized by over activity during stance which also contributes to equinovarus (Scott and Scarborough 2006). Previously reported EMG studies have demonstrated that varus deformity in children with hemiplegic cerebral palsy resulted from the anterior tibialis alone in 34% of cases, posterior tibialis alone in 33%, both muscles in 31%, and muscles other than the anterior or posterior tibialis in 2% (Michlitsch et al. 2006). In order to reliably use EMG data for surgical decision making the question must be answered as to whether the introduction of a fine wire electrode alters the existing gait pattern, as young children can experience a combination of pain, anxiety, and discomfort associated with the technique. This becomes problematic when EMG and kinematic data are collected simultaneously for the purpose of surgical decision making.

Fine wire electrodes have been found to result in alterations in temporal-spatial parameters in children with diplegic CP (Young et al. 1989). Specifically, significant reductions were identified in cadence (-9.8 steps/minute), walking velocity (-0.82 m/s), step length of the measured limb (-0.82 m), and step length of the non-measured limb (-0.78 m) when children were instructed to walk at a self-selected velocity. Although these findings implied that caution should be taken when utilizing these data collected simultaneously with 3-D kinematics for surgical decision making, it must be noted that

all of the measures were temporal-spatial parameters. Fatigue might have been another factor for the reported gait alterations since the internal electrode trials were always conducted last. Also, this study only examined children with diplegic CP. Equinovarus deformity is most common in children with hemiplegic CP who have consistently been described as having improved gait and lower extremity function compared to children with diplegia (Damiano et al. 2006). If EMG and kinematic data are to be collected simultaneously to plan surgery, it is important to determine if fine wire insertion into the posterior tibialis affects the gait pattern of children with hemiplegic CP and equinovarus.

1.5. Surgical Interventions for Equinovarus in Children with Hemiplegic Cerebral Palsy

Orthopaedic surgeons provide a significant amount of care for children with cerebral palsy. Routine care often includes surgery with goals to lengthen tight or hypertonic muscles, transfer overactive muscles, osteotomies to correct bony malalignment, and arthrodesis for unstable joints to ultimately improve mobility, function, and independence. Each of these types of procedures plays a significant role in the management of equinovarus. Although non-operative treatments (i.e. physical therapy, serial casting, splinting, taping and Botulinum toxin (Botox®) injections) are initially utilized to address the equinovarus foot and ankle, orthopedic surgery is required if the deformity persists.

Davids (2010) identified that there are three levels of foot and ankle deformity in individuals with cerebral palsy. Each level of deformity was then presented with corresponding treatment options. Individuals with level I deformity present with a dynamic muscular imbalance without fixed contracture and would most likely benefit from pharmacologic/neurologic interventions to address hypertonicity along with split tendon transfers to address the dynamic imbalance at the foot. Individuals with level II deformity present with muscular imbalance including static contracture and would benefit from a combination of musculotendinous lengthening and transfers. Finally, individuals with level III deformity present with muscular imbalance along with skeletal deformity and require a

combination of soft tissue balancing procedures and osteotomies/arthrodeses to reorient the underlying skeletal anatomy. The progression of the deformity through the different levels is associated with development, and as a result, intervention earlier on with surgeries such as musculotendinous lengthenings and tendon transfers may reduce the risk for more invasive procedures as the child ages.

1.5.1. Musculotendinous Lengthenings and Split Transfers

Two surgical techniques used to correct the equinus component of equinovarus include lengthening of the plantar flexors either at the level the myotendinous junction of the gastrocnemius and soleus or the conjoined tendon (tendo Achilles) (Green and McDermott 1942; White 1943; Tachdhian 1972; Olney et al. 1988). The typical objective of isolated equinus correction is to achieve five degrees of passive dorsiflexion with the knee extended while the child is under general anesthesia (Davids 2010). Post-operative reports of lengthening of the plantar flexors in children with CP have demonstrated improvements in physical examination measures, achieving a plantigrade foot, and normalizing ankle kinematics during gait (Tylkowski et al. 2009; Vlachou et al. 2009). However, high rates of post-operative weakness and overcorrection have also been reported, particularly after lengthening of the Achilles tendon (Segal et al. 1989; Borton et al. 2001).

The recommended surgical intervention for children who present with pes varus and/or forefoot adductus leading to intoeing, and are at least 6 years-old, includes a split musculotendinous transfer of the invertors, either the posterior or anterior tibialis (Davids 2010). A split transfer of the posterior tibialis involves dividing the distal portion of the tendon longitudinally. The lateral arm of the divided tendon is then re-routed laterally and sutured into the peroneus brevis (Green et al. 1983). A split transfer of the anterior tibialis, used mainly to correct forefoot adductus, similarly involves dividing the distal portion of the tendon longitudinally. The lateral arm of the divided tendon is then re-routed

laterally through a hole typically drilled into the cuboid (Barnes and Herring 1991). The overall goal of these surgeries is to balance the asymmetrical torque that is generated by the overpowering invertor so that a more neutral foot position is achieved throughout the gait cycle.

A series of cadaveric studies was previously performed to assess the effect of split transfers of these tendons on the invertor/evertor and plantar flexor/dorsiflexor moment arms about the subtalar and talocrural joints, respectively (Piazza et al. 2001; Piazza et al. 2003). Secondly, the authors wanted to assess how variations in tensioning of the medial and lateral arms of the tendon also affect the moment arm distances. Moment arm distances about the subtalar and talocrural joints were calculated via tendon excursion values and were determined incrementally throughout the excursion of passive hindfoot inversion and eversion. In the later study, similar values were obtained while applying various amounts of tension/slack on the arms of the tendons. Their results demonstrated: (1) prior to transfer, moment arm measurements identified that the tibialis posterior was a more powerful ankle invertor than the tibialis anterior; (2) following split transfer of the posterior tibialis to the peroneus brevis, not only did the muscle now act as a “balanced yoke” where in the neutral position it acted as neither an invertor nor an evertor, but as the hindfoot was taken passively into inversion it acted as an evertor and when passively taken into eversion it acted as an invertor; (3) split transfer had no effect on the sagittal plane moment arm in reference to the talocrural joint; and (4) variability in tensioning of the medial and lateral arms of the posterior tibialis only significantly affected the moment arm distance when one arm of the tendon was completely on slack. They concluded that the split transfers of the tibialis anterior and posterior are robust procedures that can effectively create a neutral position of the foot and ankle in the presence of a varus foot deformity.

Clinically reported postoperative outcomes for split musculotendinous transfers have been quite variable and lack consistently good and long-lasting results (Chang et al. 2002). Reports of “excellent”

(where the child presented with a plantigrade foot; no fixed or positional deformity) were observed in 68% of the children who underwent a split posterior tibialis tendon transfer to the peroneus brevis (Scott and Scarborough 2006). The surgical decisions in this case were based primarily on non-phasic firing of the posterior tibialis as observed from fine wire electromyography analysis during quantitative gait analysis. Post-operative outcomes for a split transfer of the anterior tibialis have also been reported in a retrospective study of 69 participants with equinovarus foot (Vogt 1998). Participants' ages ranged from 8-79 years with a post-operative follow-up ranging from 1 to 14 years. The author reported the "foot was perfectly balanced" in 49/73 (67%) feet. Significant functional improvements included increased independent ambulation, decreased need for orthopedic shoes and orthoses, as well as improved ability to wear normal shoes. Rather than attempting to identify the primary neuromuscular contributor(s) to equinovarus among a sample of children with CP, other investigators have reported success rates of 82% when an intramuscular lengthening of the posterior tibialis and a split transfer of the anterior tibialis was consistently used for surgical correction (Barnes and Herring 1991).

1.5.2. Possible Explanations for Variable Post-operative Success Rates

It seems surprising that post-operative outcomes demonstrated variable success rates ranging from 67-82%, and no procedure demonstrated a consistently good and long-lasting result (Barnes and Herring 1991; Vogt 1998; Chang et al. 2002; Scott and Scarborough 2006). A possible explanation for limited postoperative success rates following surgery is the methods used to report outcomes. Successful postoperative outcomes previously have been based on subjective accounts of foot and ankle morphology, as well as, observational analysis of standing and walking. Specifically, post-operative results of "excellent" were determined based on whether the child presented with a plantigrade foot and no fixed or positional deformity, or was "perfectly balanced" (Vogt 1998; Scott

and Scarborough 2006). Other postoperative results have been based on criteria established by Kling et al. (1985) in which surgery was considered successful if the patient was observed to have a plantigrade foot, without fixed or dynamic deformity, used a regular shoe, and had no abnormal callosities. Recent attempts to quantitatively measure the effect of surgery on foot and ankle deformity in children with hemiplegic cerebral palsy have made using the gait analysis. Stebbins et al. (2010) used kinematic analysis to show the effect of soft tissue balancing procedures on the foot using the Oxford Foot Model. They identified postoperative alterations in forefoot adduction and supination; however, any changes at the hindfoot outside of the sagittal plane were inferred. Thus, quantitative gait analysis has the capacity to measure postoperative changes, but significant gaps in the literature still exist that provide a comprehensive, unbiased, and quantitative outcome assessment of surgery for equinovarus due to hemiplegic CP.

CHAPTER 2

2.0 EFFECT OF FINE WIRE INSERTION ON GAIT PATTERNS IN CHILDREN WITH HEMIPLEGIC CEREBRAL PALSY

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2.1. Introduction

The use of electromyographic (EMG) patterns for surgical decision making in the lower extremities of children with cerebral palsy (CP) has become commonplace, and more recently it has been used to assist with understanding the pathomechanics associated with equinovarus foot (McCarthy 2008). A combination of surface electrodes for the more superficial musculature (anterior tibialis and gastrocnemius) and fine wire electrodes for deeper musculature (posterior tibialis) is used to determine the primary neuromuscular contributor(s) of the deformity (Gage 2004). Previously reported EMG studies have demonstrated that varus deformity in children with hemiplegic CP resulted from the anterior tibialis alone in 34% of cases, posterior tibialis alone in 33%, both muscles in 31%, and muscles other than the anterior or posterior tibialis in 2% (Michlitsch et al. 2006). In order to reliably use this assessment technique the question must be answered as to whether the introduction of a fine wire electrode alters the existing gait pattern, as young children can experience a combination of pain, anxiety, and discomfort associated with the technique. This becomes problematic when EMG and kinematic data are collected simultaneously for the purpose of surgical decision making.

Fine wire electrodes have been found to result in alterations in temporal-spatial parameters in children with diplegic CP (Young et al. 1989). Specifically, significant reductions were identified in cadence, walking velocity, step length of the measured limb, and step length of the non-measured limb

when children were instructed to walk at a self-selected velocity. Although these findings implied that caution should be taken when utilizing these data collected simultaneously with 3-D kinematics for surgical decision making, it must be noted that all of the measures were temporal-spatial parameters. Fatigue might have been another factor for the reported gait alterations since the internal electrode trials were always conducted last. Also, this study only examined children with diplegic CP. Equinovarus deformity is most common in children with hemiplegic CP who have consistently been described as having improved gait and lower extremity function compared to children with diplegia (Damiano et al. 2006).

In addition to temporal-spatial parameters, 3-dimensional hindfoot kinematics can provide quantitative data regarding possible alterations in walking due to fine wire insertion into the posterior tibialis. A method for calculating hindfoot kinematics has been previously introduced and validated for use in children (Kidder et al. 1996; Myers et al. 2004). The Milwaukee Foot Model (MFM) is a four-segment foot and ankle kinematic model that uses passive surface markers to quantify motion of the tibia, hindfoot, forefoot and hallux. Unique to the MFM is the use of radiographic offset measurements in anterior/posterior, lateral, and a coronal-plane hindfoot view to relate the underlying orientation of the bony anatomy to the surface markers, i.e. neutral referencing (Johnson et al. 1999). The kinematics are expressed with the tibia referenced to the global coordinate axes, and the remaining segments are represented in a distal relative to the next proximal segment relationship using an Euler System.

The purpose of the present study is to determine if fine wire insertion into the posterior tibialis affects the gait pattern of children with hemiplegic CP and equinovarus. We tested the hypothesis that reductions in cadence, walking velocity and step length will be similar in children with hemiplegia to those previously reported for children with diplegic CP. We also hypothesized that fine wire electrode

insertion will alter hindfoot sagittal, coronal, and transverse plane kinematics during locomotion. Specifically, we expected that the presence of the inserted electrode would result in earlier onsets and reductions in peak motion, as well as, diminished overall hindfoot excursion (ROM) during the gait cycle.

2.2. Methods

2.2.1. Participants

Twelve children with hemiplegic CP (7 males, 5 females, average age: 12.5 yrs, range: 5-17 years). All participants presented with a unilateral equinovarus foot deformity and were recruited for the present study as a part of a diagnostic gait analysis with a plan for possible surgical correction. Four of the participants presented with right-sided hemiplegia, and eight presented with left-sided hemiplegia. Based on the hemiplegic gait classification system established by Winters et al. (1987) 2 participants had a type I pattern, 4 had a type II pattern, 2 had a type III pattern, and 4 had a type IV pattern. All participants had no prior history of orthopedic surgery for equinovarus and had not received botulinum toxin injections within 1 year prior to evaluation. Children were excluded if they presented with cognitive or behavioral impairments that interfered with their ability to understand and follow basic commands necessary to participate in quantitative gait analysis and a standing weight-bearing x-ray series. All participants gave informed consent according to a University approved protocol.

2.2.2. Instrumentation

Subjects underwent 3-D gait analysis using a 14-MX camera motion analysis system (VICON, Oxford, UK) collected at 120 Hz. Cadence, walking velocity, and step length were calculated using Vicon Workstation (version 5.2.4) software and the PlugInGait model.

Simultaneously, the MFM was employed to measure multisegmental foot and ankle motion (Kidder et al. 1996). Twelve passive 9 mm reflective markers were placed on the tibia, calcaneus,

forefoot and hallux. A triad was placed on the proximal phalange to obtain hallux data. Resolution, accuracy, and reliability of the foot and ankle system has been established (Long et al. 2010). The kinematic data were processed and calculated using a custom program in Matlab (Matlab, Mathworks[®], Natick, MA, USA).

Fine wire EMG electrode insertion into the posterior tibialis was performed with participants in a seated, reclined position and the measured lower extremity in external rotation. Needle insertion was performed as reported by Yang et al. (2008) with a posterior approach under the medial tibial shaft and directed deep along the bone where the muscle lies against the interosseous membrane. A 27 Ga., 30 mm hypodermic needle with paired hook wire electrodes was used. Wire electrode placement into the posterior tibialis was confirmed using pulsed electrical stimulation and visual observation of real time raw EMG display during voluntary contraction. Surface electrode placement on the anterior tibialis was 1/3 of the distance from the lower margin of the patella to the lateral malleolus. Medial gastrocnemius surface electrode placement was 1/3 of the distance from the medial femoral condyle to the bisection of the posterior aspect of the calcaneus (Zipp 1982). Surface and fine wire data were captured with Vicon Workstation software at a sampling rate of 2160 Hz.

2.2.3. Experimental Protocol

Participants were instructed to walk “at a comfortable walking speed” over a 30m walkway. A total of 20 to 30 trials were collected until 6 representative trials (3 “with wire” and 3 “without wire”) were obtained for analysis. The presence of fatigue effects was tested by having the first six participants (Wire 1st Group) undergo the “with wire” trials first followed by the “without wire” trials. The second six participants (Wire 2nd Group) underwent the “without wire” trials first followed by the “with wire” trials. All kinematic data was collected with surface EMG electrodes over the anterior tibialis, gastrocnemius, rectus femoris, and medial hamstrings.

Following gait data collection, participants received a series of weight-bearing radiographs of the foot in the anterior-posterior and lateral views along with a modified hindfoot coronal alignment view (Johnson et al. 1999). All fine wire electrode placement and specific radiographic offset measurements were obtained by the same author (JK).

2.2.4. Data Analysis

Group averages were calculated using six representative trials from each participant (3 “with wire” and 3 “without wire”) and were compared across the gait cycle using two-way, repeated measure analyses of variance. This was performed to determine the effect the of presence of a fine wire electrode in the posterior tibialis and trial order on temporal-spatial parameters, as well as, hindfoot sagittal, coronal and transverse plane kinematic peaks, timing of kinematic peaks, and ROM. Individual change scores were also calculated for the amplitude and timing of kinematic peaks, as well as, hindfoot ROM by subtracting the value obtained from the “without wire” trials from the “with wire” trials. A negative score indicates an earlier onset of peak motion, decrease in peak motion, or decrease ROM with the presence of a wire electrode in the posterior tibialis. Conversely, a positive score indicates a delayed onset of peak motion, increase in peak motion, or increase in ROM with the presence of a wire electrode. Due to multiple comparisons a Bonferroni correction was implemented to minimize the risk of a Type I error. This yielded an adjusted alpha value of 0.004.

Once non-significant differences were identified among the variables, Pearson Correlation Coefficients (r) were then calculated to further analyze the association between the gait parameters of the “with wire” trials and the “without wire” trials, as well as, provide an effect size estimate (Volkner 2006). In accordance with Cohen’s Classification, a strong association was defined as an r value of greater than 0.70, a moderate to substantial association was defined as an r value between 0.30 and 0.70, and a weak association was defined as an r value of less than 0.30 (Portney and Watkins 1993).

2.3. Results

2.3.1. Temporal-Spatial Parameters

Table 2.1 shows the temporal-spatial parameters of the measured and non-measured side averaged (with standard error) over all trials for the “with wire” and “without wire electrode” trials. The p -values indicate a non-significant effect of the presence of a fine wire electrode on walking speed, cadence, and step length of the measured and non-measured side. Correlation analysis demonstrated strong associations between “with wire” and “without wire” conditions for walking speed ($r = 0.81, p = 0.001$), step length of the measured leg ($r = 0.96, p \leq 0.0001$), and step length of the non-measured leg ($r = 0.91, p \leq 0.0001$). Correlation of cadence between conditions was not significant.

2.3.2. Hindfoot Kinematics

Plots of individual sagittal kinematics with and without the presence of a fine wire electrode are presented in Figure 2.1. The first plot shows the Wire 1st group and second plot shows the Wire 2nd group. The “with wire” and “without wire” trials are plotted on top of one another and demonstrate minimal deviation. Figure 2.2 and 2.3 display averages and standard errors of hindfoot kinematic peaks, timing of peaks, and ROM in the sagittal, coronal, and transverse planes. Figures 2.4 and 2.5 display the change score of the Wire 1st and Wire 2nd groups for hindfoot peaks, timing of kinematic peaks, and, excursions in the sagittal, coronal and transverse planes. No significant main effect of fine wire insertion, nor interactions of fine wire insertion and trial order, were found for hindfoot peaks, timing of those peaks, and ROM. Correlation analysis of hindfoot kinematics demonstrated strong associations between the conditions for all gait parameters: Peak Maximum and Timing of Peak

Table 2.1. Group averages and standard errors (SE) of temporal-spatial parameters with and without a fine wire electrode in the posterior tibialis.

Temporal-Spatial Gait Parameters: Group Averages			
Variable	With Wire Trials Average (SE)	Without Wire Trials Average (SE)	p value
Walking Speed (m/s)	0.94 (0.05)	0.95 (0.07)	n.s.
Cadence (steps/min)	116.98 (4.32)	118.70 (4.26)	n.s.
Step Length: Measured Leg (m)	0.47 (0.03)	0.46 (0.04)	n.s.
Step Length: Non-Measured Leg (m)	0.49 (0.02)	0.50 (0.02)	n.s.

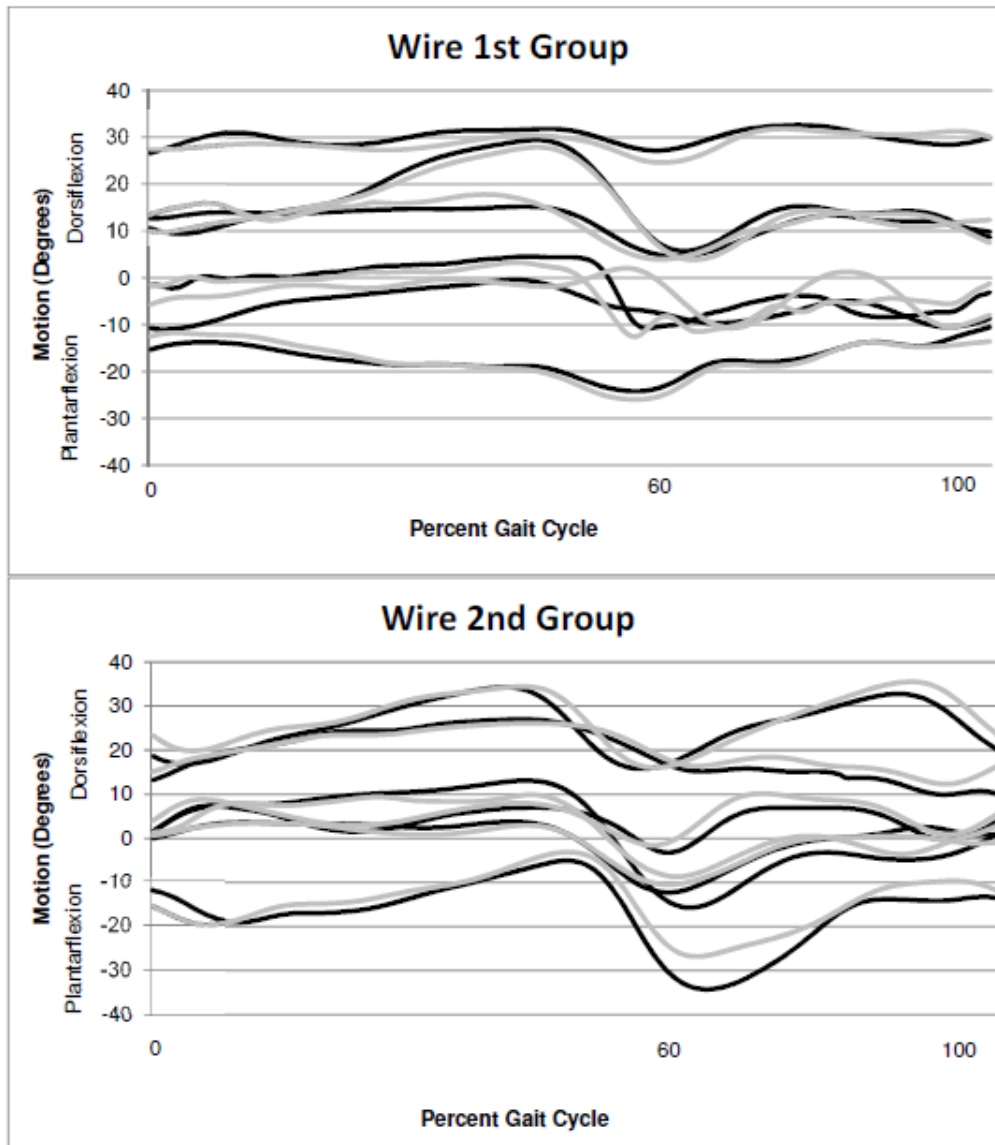


Figure 2.1. Individual sagittal plane hindfoot kinematic plots of the twelve participants with (Black) and without (Grey) a wire electrode in the posterior tibialis. Participants were separated into a “Wire 1st” group or a “Wire 2nd” group depending on when during testing the participants had the wire electrode inserted.

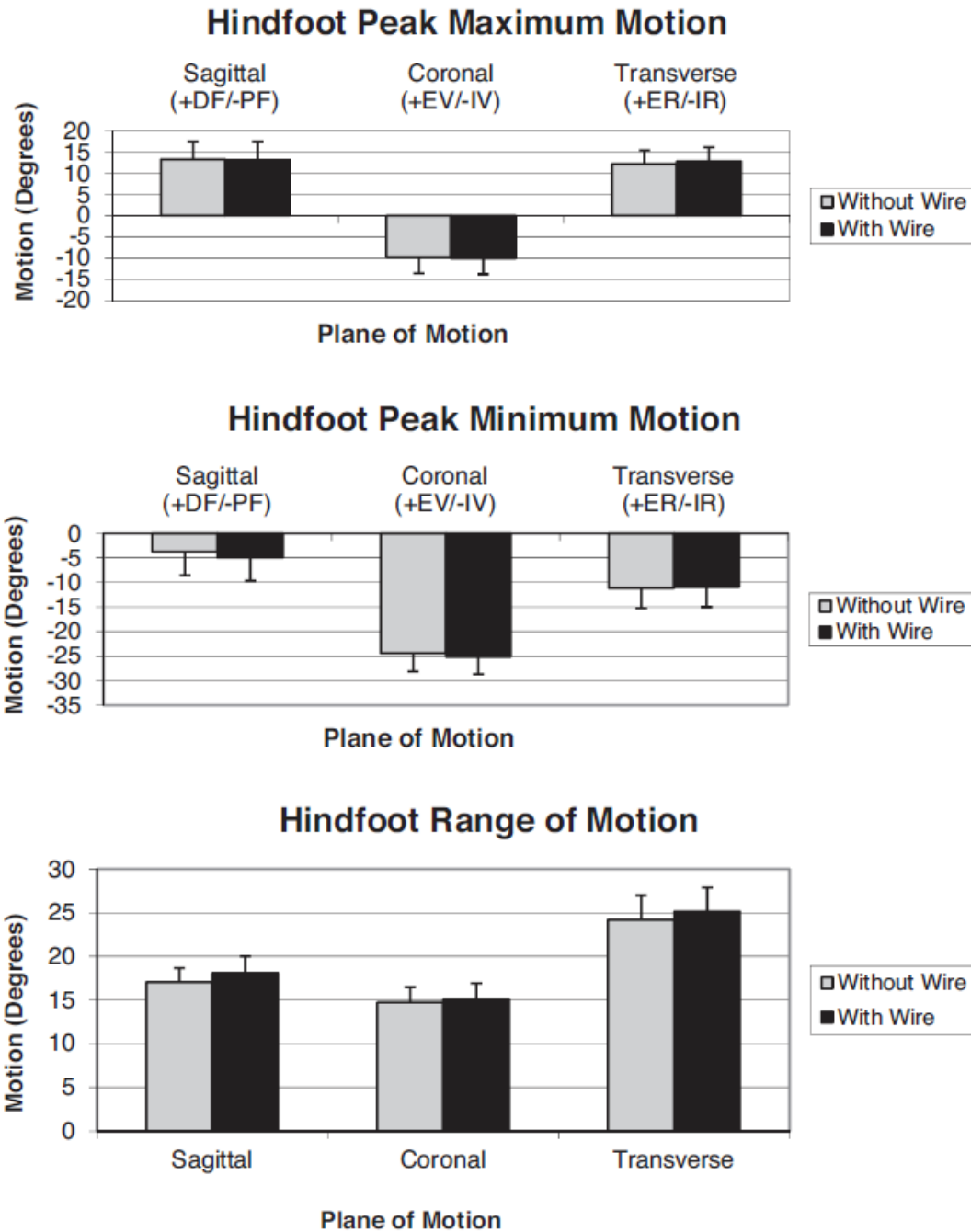


Figure 2.2. Group averages and standard error bars of hindfoot kinematic peaks and range of motion during gait in the sagittal, coronal, and transverse planes with (black) and without (grey) a wire electrode in the posterior tibialis.

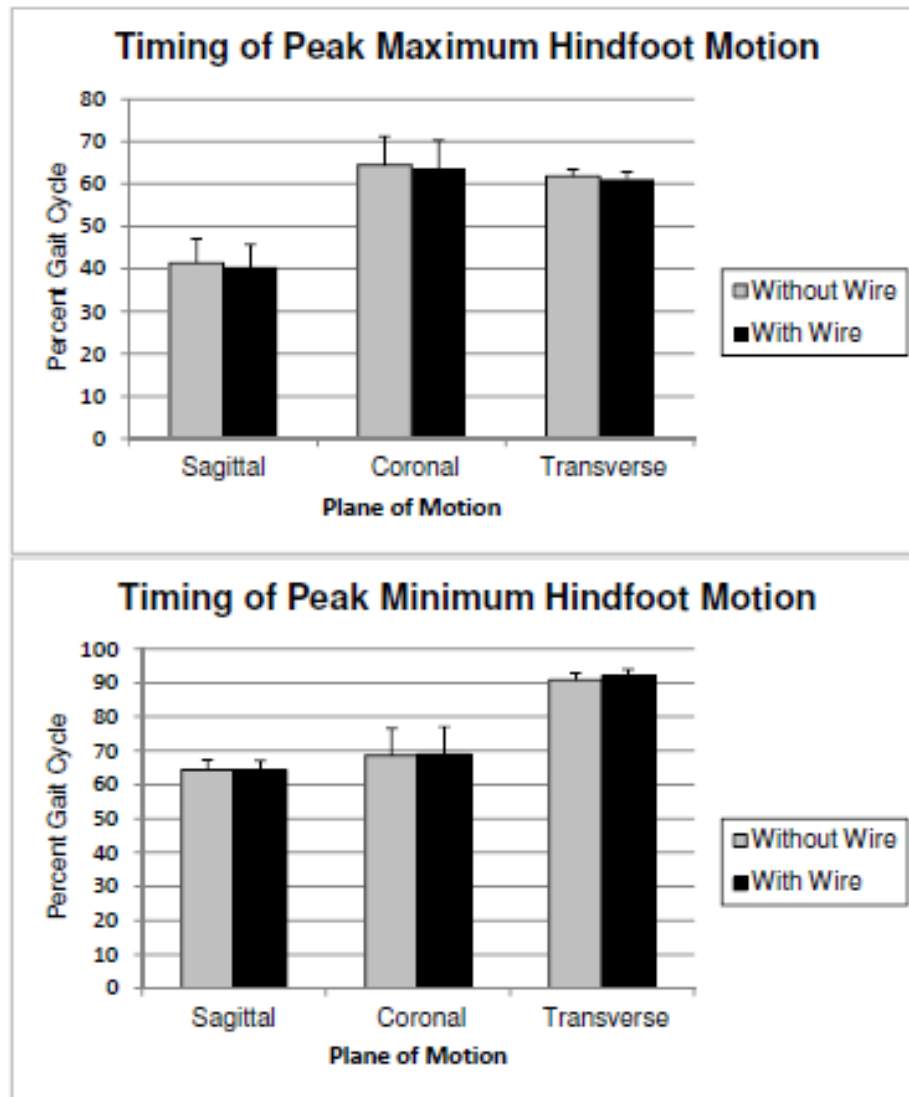


Figure 2.3. Group averages and standard error bars of timing of hindfoot kinematic peaks during gait in the sagittal, coronal, and transverse planes with (black) and without (grey) a wire electrode in the posterior tibialis.

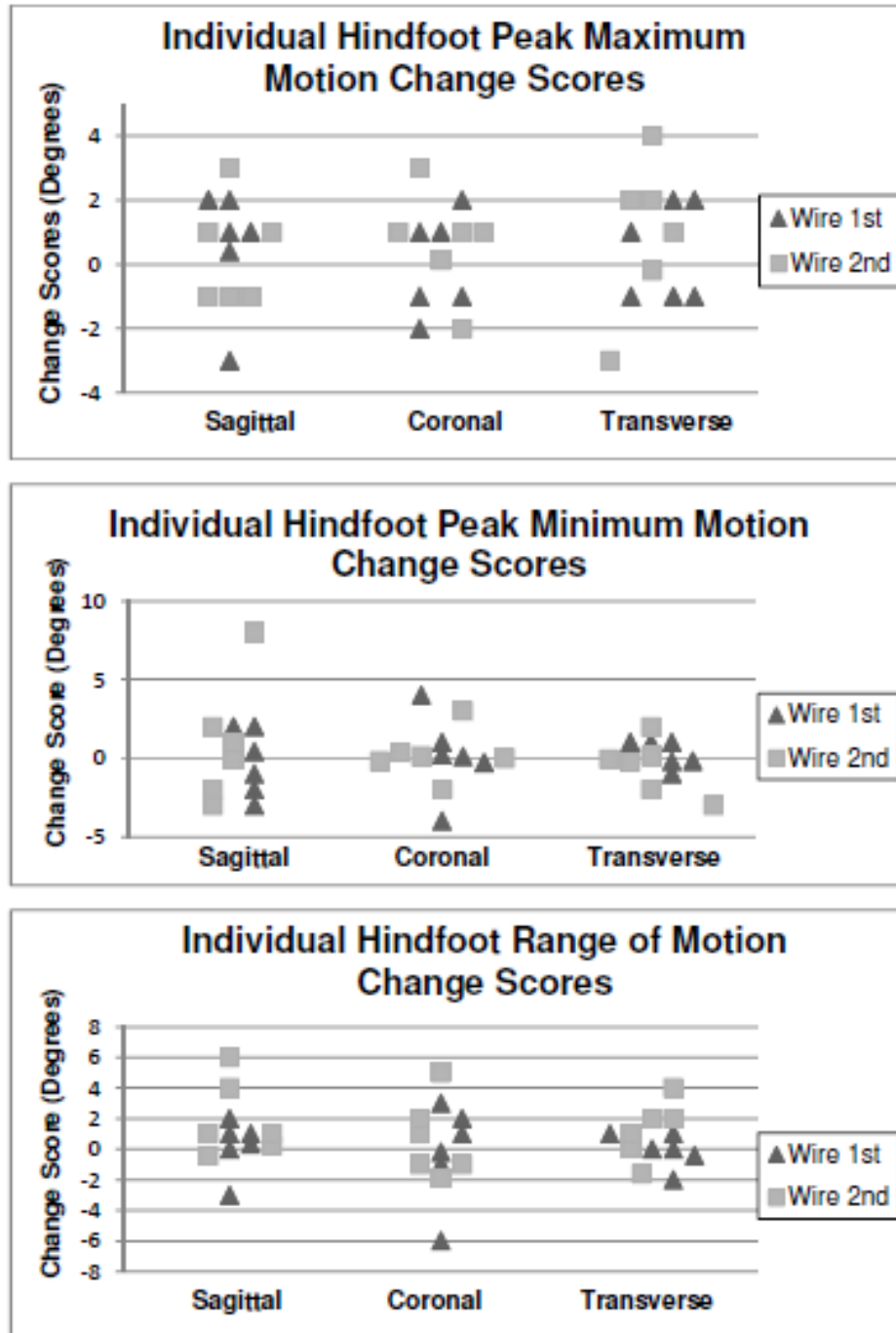


Figure2.4. Individual change scores (CS) in kinematic peaks and range of motion in the sagittal, coronal, and transverse planes stratified by trial order. Black triangles indicate the “Wire 1st” group and the grey squares indicate the “Wire 2nd” group.

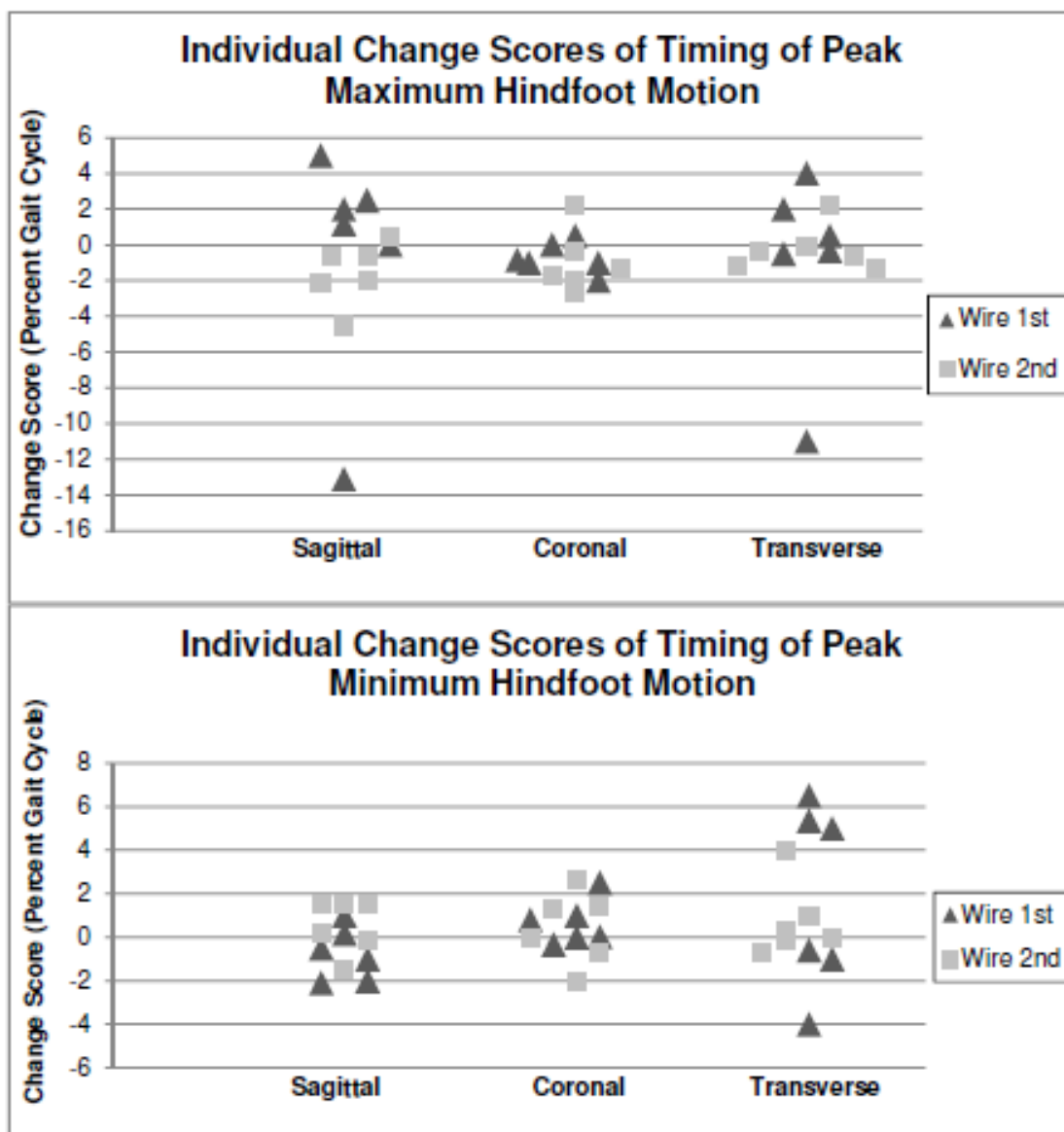


Figure 2.5. Individual change scores (CS) in timing of kinematic peaks in the sagittal, coronal, and transverse planes stratified by trial order. Black triangles indicate the “Wire 1st” group and the grey squares indicate the “Wire 2nd” group.

Maximum (range: $r = 0.78$ to 1.00 , $p \leq 0.0025$), Peak Minimum and Timing of Peak Minimum (range: $r \leq 0.89$ to 1.00 , $p \leq 0.0001$), and ROM (range: 0.89 to 0.99 , $p \leq 0.0001$).

2.4. Discussion

The central finding of this study is that fine wire electrode insertion into the posterior tibialis does not alter the gait patterns of children with equinovarus secondary to hemiplegic CP. No differences were observed in temporal-spatial parameters or hindfoot kinematics with the introduction of a fine wire electrode in the posterior tibialis. The similarities between the “with wire” and “without wire” trials were further supported with strong correlations among both the temporal-spatial and kinematic variables. Thus, these findings suggest that electromyographic (including fine wire analysis of the posterior tibialis) and kinematic data gathered during a gait analysis can be collected simultaneously without the risk of data corruption for children with equinovarus due to hemiplegia. When executed in such a manner, simultaneous data collection efficiently provides a comprehensive evaluation of the multisegmental and multiplanar nature of equinovarus, as well as, identifies the potential neuromuscular contributor(s) that can aid in surgical decision making.

It has been postulated that the presence of a fine wire electrode can result in pain, alterations in muscular activity, and changes in gait mechanics (Gage 2004). Data from the current study identified two potential outliers presenting with either an alteration in peak motion or timing of peak motion isolated to the end of stance phase. After closer observation of their data, the other parameters were consistent between conditions, and any deviations were not considered clinically meaningful. These individuals consistently presented with more severe gait deviations that extended into proximal lower extremity segments (Winters type III and IV gait patterns) (Winters et al. 1987). Without excluding their data from the analysis, strong associations were observed among the temporal-spatial and kinematic parameters.

The current study's findings regarding the effect of fine wire insertion on gait are in contrast to previous reports of children with diplegia who demonstrated reductions in temporal-spatial parameters following fine wire insertion into the posterior tibialis [3]. This discrepancy can be explained when reviewing the fundamental differences in the two patient populations. Damiano et al. (2006) demonstrated that among participants with CP at a similar level of functional mobility, i.e. Gross Motor Functional Classification System (GMFCS) Level, children with hemiplegia had a tendency to perform consistently better at tasks associated with gait (including measures of walking speed and stride length) and lower extremity function than children with diplegia. These functional differences may result from the presence of one higher functioning, if not normal, lower limb in children with hemiplegia (Wiley and Damiano 1998). Thus, the potential response to a treatment, or in this case an evaluation technique, is likely to vary with different distributions of limb involvement. These findings are further supported by the current study where the presence of a wire electrode did not impact temporal-spatial or kinematic parameters during locomotion in children with hemiplegia.

The current study also found no effect of trial order on hindfoot kinematic peaks or ROM. Neither the Wire 1st nor Wire 2nd groups demonstrated obvious trends in kinematic change scores. Thus, this patient population should be able to tolerate walking up to 20-30 trials on a 30m walkway without altering their gait pattern.

A limitation in the current study was that localization of wire EMG electrode into the posterior tibialis was not performed with imaging techniques, i.e. ultrasound. Verification was performed with the use of pulsed electrical stimulation and viewing real-time raw EMG output during voluntary contraction of the posterior tibialis and flexor hallucis longus. Also, the results of the current study are based on a relatively small, homogenous group of children with flexible equinovarus deformity due to hemiplegic CP. Therefore, generalization of these results to other patient populations commonly

presenting with equinovarus deformity, such as diplegic CP, talipes equinovarus, and Charcot-Marie-Tooth, should be cautioned.

In summary, the results of the current study demonstrate that fine wire electrode placement into the posterior tibialis did not affect the gait of children with equinovarus secondary to hemiplegic CP. This allows researchers and clinicians to collect multiple forms of data simultaneously during gait analysis to efficiently and effectively determine the etiology of the equinovarus deformity for surgical decision making as well as to measure post-operative outcomes.

CHAPTER 3.

3.0 KINEMATIC FOOT TYPES IN YOUTH WITH EQUINOVARUS SECONDARY TO HEMIPLEGIA

3.1. Introduction

Equinus and varus, often in combination, are the most common foot and ankle deformities in children with hemiplegic cerebral palsy (CP) (Wren et al. 2005). Static or dynamic soft tissue imbalance results in segmental deformities including hindfoot equinus and inversion, midfoot cavus, as well as, forefoot supination and adduction. Beyond atypical foot position, these deformities are associated with gait deviations at more proximal segments, increased mechanical work, and increased energy expenditure in children with cerebral palsy (van den Hecke et al. 2007; Ballaz et al. 2010; Stebbins et al. 2010).

Equinovarus foot deformity in hemiplegic CP is a heterogeneous condition. Because many factors contribute to the complexity of the deformity, previous reports have identified a lack of uniformity in the gait kinematics of children with equinovarus (Theologis and Stebbins 2010). Both the hindfoot and forefoot segments contribute to the deformity in the sagittal, coronal and/or transverse planes. In addition, equinovarus can be present during the stance and/or swing phases of gait. Finally, foot and ankle deformities in children with CP can be characterized by dynamic or static soft tissue imbalance with, or without, skeletal deformity (Davids 2010). Accurate identification of the involved segment(s), plane(s), timing, and the range of motion (ROM) of the deformity is important when defining types and causes of equinovarus deformity to make accurate and effective clinical decisions.

Quantitative gait analysis including multi-segmental foot and ankle kinematics can effectively characterize the equinovarus deformity resulting from hemiplegic cerebral palsy. Several laboratories have implemented multi-segment foot and ankle models to quantify foot kinematics in the sagittal, coronal, and transverse planes (Rankine et al. 2008). However, multi-segment foot and ankle modeling

can be technically difficult in a pediatric population with foot deformity due to the small foot sizes and lack of reliable bony landmarks to attach surface-based markers. The Milwaukee Foot Model (MFM) is a multi-segment foot and ankle model which has previously been used to measure 3-D kinematics during locomotion in this population (Kidder et al. 1996; Krzak et al. 2013). The MFM has demonstrated good inter-laboratory repeatability, was validated for use in children, and is one of the only existing foot and ankle models that has demonstrated external validity using pathologic foot conditions in the literature (Myers et al. 2004; Long et al. 2008; Bishop et al. 2012). It addresses the difficulties in current foot and ankle modeling by using radiographic skeletal indexing to mathematically orient the surface marker-based local coordinate axes to the underlying skeletal anatomy. Accurately linking surface markers to the underlying skeletal anatomy using radiographic indexing will result in accurate foot and ankle segmental kinematics. Such an analysis will identify segmental pathokinematics in children with equinovarus foot deformity when compared to typically developing children.

Given the complexity of foot and ankle kinematics during locomotion, the MFM produces a large amount of additional data to integrate into the decisions regarding appropriate interventions and measuring the effectiveness of those interventions. The challenge of integrating foot and ankle kinematics into clinical care then becomes developing effective means of reducing the large amount of heterogeneous data. In whole-body gait applications, previous researchers have developed clinically relevant classifications of whole-body gait patterns for individuals with CP (Winters et al. 1987; Rodda et al. 2004). Patients can be evaluated and fit into a particular subgroup which subsequently facilitates decision making regarding treatment. However, previous classifications were often not systematically generated, and the whole-body model lacks the complexity necessary to distinguish among the subtle soft tissue imbalances of the ankle plantar flexors and subtalar invertors that result in equinovarus. A quantitative means of classifying gait patterns based on 3-D segmental foot and ankle kinematics is

being proposed to identify subgroups within the hemiplegic cerebral palsy population and facilitate clinical decision making.

Recently, principal component analysis (PCA) techniques have been used successfully to develop quantitative gait classification schemes that reduce the large amounts of gait data into a few critical independent variables (Shemmell et al. 2007; Carriero et al. 2009). This technique takes kinematic/kinetic data points over a time series or an assemblage of relevant gait parameters and transforms them into a smaller number of variables that are independent from one another. Once these data are reduced, cluster analysis can then be used to identify clinically recognizable groups of homogeneous individuals. Multiple clustering techniques are available using hierarchical and non-hierarchical procedures. K-means clustering is a non-hierarchical technique that has previously been used as an effective method to identify groups of gait patterns in children with cerebral palsy (O'Byrne et al. 1998; Kienast et al. 1999; Rozumalski and Schwartz 2009). This mathematical clustering algorithm assigns membership to a group based on the proximity to means maximizing the similarities within the groups and the differences among the groups.

The purpose of the current study was to determine if clinically relevant subgroups exist among a sample of typically developing children and children with equinovarus due to hemiplegic CP using segmental foot kinematics, PCA and *K*-means cluster analysis. We hypothesized that more than one principal component (PC) would be required to account for the variance in segmental gait kinematics of the hindfoot and forefoot among the previously described sample. If more than one PC was identified, *K*-means clustering of the identified principal components would be applied to identify subgroups of participants with varying involvement of the different segments, in three planes, and varying ROM.

3.2. Methods

3.2.1. Participants

Twenty four children/adolescents with hemiplegic CP (13 males, 11 females, average age: 12.0 ± 4.1 years, 13 right-sided, 11 left-sided) and a group of twenty typically developing children/adolescents (11 males, 9 females, average age: 11.8 ± 2.7 years) were included in the present study. All participants with CP presented with a unilateral equinovarus foot deformity as determined by their treating physician and were recruited for the present study as a part of a diagnostic gait analysis with a plan for possible surgical correction. All participants had no prior history of orthopaedic surgery for equinovarus and had not received botulinum toxin (Botox®) injections within one year prior to evaluation. Children were excluded if they presented with cognitive or behavioral impairments that interfered with their ability to understand and follow basic commands necessary to participate in quantitative gait analysis and a standing weight-bearing x-ray series. Informed consent was provided from the participants' legal guardians and, when appropriate, assent/consent was obtained from the participants as approved by an institutional review board.

3.2.2. Instrumentation

Participants underwent quantitative gait analysis using the MFM to measure the motion of four segments (tibia, hindfoot, forefoot and hallux) (Kidder et al. 1996). Twelve passive 9mm reflective markers were placed on the tibia, calcaneus, and forefoot. A special three marker triad was used to obtain hallux data. Resolution and accuracy of the foot and ankle system has been established by our group as well as testing reliability (Long et al. 2008). Marker trajectories were collected at 120 Hz. using a 14-MX camera three-dimensional motion analysis system and Vicon Workstation (version 5.2.4) software (VICON, Oxford, UK). The kinematic data were processed and calculated using a custom program written in Matlab (Mathworks®, Natick, MA, USA).

3.2.3. Experimental Protocol

A static standing trial was collected with the participant standing on a cardboard sheet where a foot tracing was made. Participants were instructed to walk “at a comfortable walking speed” down a 30m walkway. Between twenty and thirty trials were collected, and three representative trials were chosen for analysis. Twenty to thirty trials were collected as twelve of children participated in Experiment 1 where data was collected under two conditions, with and without a fine wire electrode in the posterior tibialis.

Following gait data collection, participants were taken to the radiology department for a series of weight-bearing radiographs of the foot. Anterior-posterior, lateral, and a modified hindfoot coronal alignment view were captured while standing on the foot tracing created during the static standing trial (Johnson et al. 1999). All skeletal alignment measurements were obtained from the radiographs by the same author (JK) to provide static offsets that allow the marker set to better track the underlying skeletal anatomy.

3.2.4. Principal Component Analysis

The input data matrix of the PCA consisted of a subset of thirty-eight segmental foot and ankle kinematic variables, walking speed, and age at the time of the preoperative evaluation. The kinematic variables were chosen via clinical consensus based their ability to identify specific segment(s), plane(s), timing, and the flexibility of the deformity. They included hindfoot and forefoot peaks of motion, ROM, and average position throughout the gait cycle. The resulting raw data matrix consisted of 1760 data points (44 participants * 40 variables). Descriptive statistics including means, standard deviations (SD), and ranges of the 40 variables were computed for initial comparisons between children with cerebral palsy and the typically developing children using Student’s t-tests and Bonferroni-Holm corrections for multiple comparisons (Holm 1979; Aickin and Gensler 1996). Each variable was then normalized by subtracting the mean and dividing by the standard deviation across the entire sample.

The PCs were derived from the correlation matrix of the normalized dataset using a Varimax rotation in IBM SPSS Statistics 20 (Chicago, IL). Calculation of PCs in such a manner is more conducive for data with high variability and results in latent variables that are orthogonal (i.e. uncorrelated). This resulted in a total of 40 initial principal components. Specific criteria to retain variables and PCs have previously been established and were implemented to ensure that the variables were distinct measures of one specific principal component. The criteria used to determine which PCs were retained included: (1) an eigenvalue of ≥ 1.00 (Kaiser 1960), (2) components located to the left of an ‘elbow’ on the scree plot containing the eigenvalues across all PCs (Cattell 1966), (3) retaining enough components so that the cumulative percent of variance accounted for was at least 80% (Kaiser 1960; Jolliffe 2002). Variables were retained in a particular component if: (1) at least 50% of the variance of the normalized variable was accounted for by the retained PCs ($h^2 \geq 0.50$), (2) the variable had a weighting score of ≥ 0.40 on a PC, and (3) the variable demonstrated a simple structure (i.e. the weighting score of the particular variable was not ≥ 0.40 on more than one PC (Stevens 1986). The weighting score is actually the equivalent of the bivariate correlation coefficient between the variable and the PC. Individual PC scores were then derived for each participant across all retained principal components for the subsequent cluster analysis. PC scores were calculated using the following equation:

$$PC\ score_i = \sum_{j=6}^n \bar{x}_{ij} \alpha_j$$

The PC score is the weighted sum of the kinematic variables retained within that particular PC. The PC score ($i = 1$ through 6) is calculated for each participant ($n = 15$) by taking the sum of the products of the averaged kinematic variables (\bar{x}_j) retained within the PC (i) and the PC weighting scores (α) for each variable (j).

3.2.5. Cluster Analysis

An initial hierarchical cluster analysis using squared Euclidian distances and Ward's Method was performed on the standardized principal component scores for all participants (Ward 1963; Kinsella and Moran 2008; Ferrarin et al. 2012). This was done to define the appropriate number of a-priori clusters to be used in the *K*-means cluster analysis. Individual principal component scores were standardized into z-scores by subtracting the means and dividing by the standard deviation within each principal component. Standardization was performed to allow all principal component scores to have equal influence on the initial cluster center locations in the *K*-means analysis. The optimal number of clusters to be used in the *K*-means analysis was determined by calculating the agglomeration distance coefficients across stages as additional cases from 1 to 44 were merged into the clusters. A scree diagram of the distance coefficients across stages was then used to identify the stage where the first significant change occurred in the coefficients as additional cases were added to the clusters. The identified stage was subtracted from the total number of subjects, forty-four, to determine the appropriate number of clusters to be used in the *K*-means analysis.

Once cluster membership was assigned using *K*-means cluster analysis, one-way analyses of variance were performed to determine the effect of cluster membership on principal component scores. Where a main effect of cluster membership was identified, post-hoc Tukey tests were performed to further analyze the pair-wise comparisons. The level of statistical significance was set at 0.05.

3.3. Results

Tables 3.1 and 3.2 show the means, standard deviations (SD), and ranges of the forty chosen variables included in the principal component analysis for children with cerebral palsy and typically

developing children. Comparisons of the means between the two groups using Student's t-tests and the Bonferroni-Holm corrections for multiple comparisons (Holm 1979; Aickin and Gensler 1996) demonstrated expected differences in walking speed and many of the kinematic parameters consistent with an equinovarus foot and ankle deformity. Specifically, participants with cerebral palsy walked slower and presented with a more plantar flexed and inverted hindfoot relative to the tibia, as well as, a forefoot in dorsiflexion and adduction relative to the hindfoot.

3.3.1. Principal Component Analysis

Of the initial forty clinically relevant variables used in the first iteration of the PCA, fourteen were removed from further analyses secondary to either: (1) less than 50% of the variance was accounted for by the principal components ($h^2 < 0.50$), or (2) the variable demonstrated a complex structure (i.e. the variable loaded on more than one PC with weighting scores ≥ 0.40). The remaining twenty-six variables shown in Table 3.3 were ultimately reduced to six PCs (PC1-PC6) with eigenvalues ranging from 8.5 (PC1) to 1.5 (PC6). Weighting scores of the individual variables ranged from 0.70 to 0.97. Additionally, the six retained PCs accounted for 92% of the cumulative variance of the data. Constructs of the PCs were then reviewed to provide a clinically relevant interpretation of the data taking into account the relationship among the variables within each of the six principal components (Table 3.4.).

Table 3.1. Means, standard deviations (SD), and ranges of age, walking velocity, and sagittal plane kinematics used in the initial iteration of the principal component analysis for children with hemiplegic cerebral palsy and typically developing children.

Variables		CP			Typically Developing Children				p value	
		Average	SD	Range		Average	SD	Range		
		Min	Max	Min	Max	Min	Max	Min	Max	
Age		12.0	4.1	5.7	19.7	11.8	2.7	6.1	17.5	n.s.
Walking Velocity		0.9	0.2	0.5	1.2	1.2	0.2	0.8	1.4	< 0.0001
SAGITTAL PLANE KINEMATICS										
	Sagittal hindfoot position at IC	5.3	11.6	-12.5	27.9	13.0	10.0	-5.7	41.0	n.s.
	Peak hindfoot dorsiflexion during stance	12.3	13.3	-11.7	34.5	27.6	9.9	6.1	46.6	n.s.
	Peak hindfoot plantarflexion during stance	-2.8	15.7	-31.6	27.8	9.6	10.2	-12.3	37.2	n.s.
	Peak hindfoot dorsiflexion during swing	7.5	12.7	-13.6	35.5	23.9	9.8	3.3	46.5	< 0.0001
	Peak hindfoot plantarflexion during swing	-4.0	15.5	-34.2	28.3	9.8	10.0	-5.8	38.9	0.0008
	Sagittal hindfoot ROM during stance phase	15.1	5.8	4.7	26.6	18.0	5.2	9.4	29.7	n.s.
	Sagittal hindfoot ROM during swing phase	11.5	5.7	3.6	22.9	14.1	4.9	4.6	22.2	n.s.
	Sagittal hindfoot ROM throughout GC	16.7	6.2	4.7	29.2	18.9	5.3	9.4	29.9	n.s.
	Average sagittal hindfoot position during stance	6.8	12.6	-17.6	28.1	20.6	9.2	0.5	42.4	0.0001
	Average sagittal hindfoot position during swing	2.7	13.4	-21.4	30.3	19.7	9.5	0.9	44.6	0.0001
	Sagittal forefoot position at IC	-30.1	16.5	-50.8	11.7	-35.7	13.4	-54.3	-3.1	n.s.
	Peak forefoot dorsiflexion throughout GC	-13.7	14.7	-39.3	17.6	-29.4	13.7	-50.0	3.5	0.0006
	Peak forefoot plantarflexion throughout GC	-32.8	15.5	-52.8	3.4	-40.3	14.1	-61.2	-6.8	n.s.
	Sagittal forefoot ROM during stance	16.4	7.0	6.2	29.6	9.7	2.2	6.9	15.1	n.s.
	Sagittal forefoot ROM during swing	15.1	7.8	3.4	31.5	6.6	3.1	2.5	16.6	< 0.0001
	Sagittal forefoot ROM throughout GC	19.1	7.0	8.1	31.5	12.2	7.3	6.9	40.4	n.s.
	Average sagittal forefoot position throughout GC	-23.2	14.9	-45.5	11.6	-33.3	14.9	-57.1	-2.4	n.s.

Table 3.2. Means, standard deviations (SD), and ranges of coronal and transverse kinematics used in the initial iteration of the principal component analysis for children with hemiplegic cerebral palsy and typically developing children.

Variables	CP				Typically Developing Children				p value
	Average	SD	Range		Average	SD	Range		
			Min	Max			Min	Max	
CORONAL PLANE KINEMATICS									
Coronal hindfoot position at IC	-16.0	10.0	-39.3	5.1	-4.9	11.4	-23.9	8.4	0.0014
Peak hindfoot eversion throughout GC	-8.9	11.4	-37.8	21.7	6.6	9.3	-10.4	19.4	< 0.0001
Peak hindfoot inversion throughout GC	-21.7	11.8	-47.7	1.4	-7.8	10.8	-24.5	4.8	0.0002
Coronal hindfoot ROM during stance	10.9	6.6	2.1	28.4	11.3	6.9	3.7	27.0	n.s.
Coronal hindfoot ROM during swing	11.3	6.2	3.0	28.0	13.3	7.6	3.1	29.4	n.s.
Coronal hindfoot ROM throughout GC	12.8	6.4	3.7	28.8	14.4	6.8	4.5	29.4	n.s.
Average coronal hindfoot position throughout	-15.5	11.0	-42.1	11.7	-2.1	12.0	-31.1	12.7	0.0003
Coronal forefoot position at IC	6.8	8.8	-12.1	28.4	2.5	5.2	-5.0	15.9	n.s.
Peak forefoot valgus throughout GC	11.5	9.3	-9.7	32.7	6.4	4.4	0.7	18.6	n.s.
Peak forefoot varus throughout GC	-27.0	13.0	-50.5	-0.4	-22.3	9.0	-40.6	-9.8	n.s.
Coronal forefoot ROM during stance	34.6	16.2	2.4	62.9	26.7	11.4	4.6	54.1	n.s.
Coronal forefoot ROM during swing	33.7	16.4	3.9	60.6	25.8	11.6	9.3	50.8	n.s.
Coronal forefoot ROM throughout GC	38.6	16.7	3.9	65.3	28.7	11.7	10.5	56.8	n.s.
Average coronal forefoot position throughout	-2.9	6.7	-19.9	6.6	-3.8	3.8	-12.5	2.5	n.s.
TRANSVERSE PLANE KINEMATICS									
Transverse forefoot position at IC	-26.3	14.9	-50.0	5.6	-14.5	10.9	-35.8	4.1	n.s.
Peak forefoot abduction throughout GC	-17.0	13.7	-46.0	9.0	-3.6	8.4	-25.6	11.2	0.0002
Peak forefoot adduction throughout GC	-34.9	15.9	-60.8	-0.4	-18.4	9.9	-38.1	0.1	0.0001
Transverse forefoot ROM during stance	15.8	9.6	4.9	54.3	12.0	5.0	7.3	26.0	n.s.
Transverse forefoot ROM during swing	15.4	10.3	4.5	51.7	11.2	6.0	4.1	27.8	n.s.
Transverse forefoot ROM throughout GC	17.9	6.4	6.5	29.1	14.1	5.1	8.3	27.8	n.s.
Average transverse forefoot position	-26.2	14.4	-49.5	4.8	-11.4	9.5	-31.4	6.5	0.0002

Table 3.3. Individual weighting scores and the amount of variance accounted for among variables within the retained principal components (h^2). The eigenvalues and cumulative variance are also reported for each principal component.

Variable	Principal Component					
	1 (854, 32.8%)	2 (6.7, 58.5%)	3 (3.0, 70.1%)	4 (2.5, 79.7%)	5 (1.7, 86.3%)	6 (1.5, 91.9%)
Sagittal hindfoot position at IC	0.94					0.96
Peak hindfoot dorsiflexion during stance	0.96					0.98
Peak hindfoot plantarflexion during stance	0.94					0.99
Peak hindfoot dorsiflexion during swing	0.97					0.98
Peak hindfoot plantarflexion during swing	0.93					0.99
Sagittal hindfoot ROM during stance phase					0.94	0.92
Sagittal hindfoot ROM during swing phase					0.87	0.79
Sagittal hindfoot ROM throughout GC					0.94	0.94
Average sagittal hindfoot position during stance	0.97					0.98
Average sagittal hindfoot position during swing	0.97					0.98
Coronal hindfoot position at IC			0.92			0.92
Peak hindfoot eversion throughout GC			0.89			0.98
Peak hindfoot inversion throughout GC			0.90			0.98
Coronal hindfoot ROM during stance		0.93		0.90		0.93
Coronal hindfoot ROM during swing		0.93		0.92		0.96
Coronal hindfoot ROM throughout GC		0.91		0.92		0.98
Average coronal hindfoot position throughout GC			0.94			0.97
Peak forefoot dorsiflexion throughout GC	-0.75					0.79
Coronal forefoot ROM during stance						0.96
Coronal forefoot ROM during swing						0.59
Coronal forefoot ROM throughout GC						0.95
Average coronal forefoot position throughout GC						0.64
Transverse forefoot position at IC		-0.73			-0.70	0.93
Peak forefoot adduction throughout GC		-0.81				0.95
Transverse forefoot ROM during stance						0.83
Transverse forefoot position throughout GC						0.90

Table 3.4. Constructs of the six principal components

Principal Component (PC)	Construct
PC1	Sagittal hindfoot and forefoot equinus
PC2	Transverse forefoot adduction and coronal forefoot ROM
PC3	Coronal hindfoot varus
PC4	Coronal hindfoot ROM
PC5	Sagittal hindfoot ROM
PC6	Coronal/Transverse forefoot supination and transverse forefoot ROM

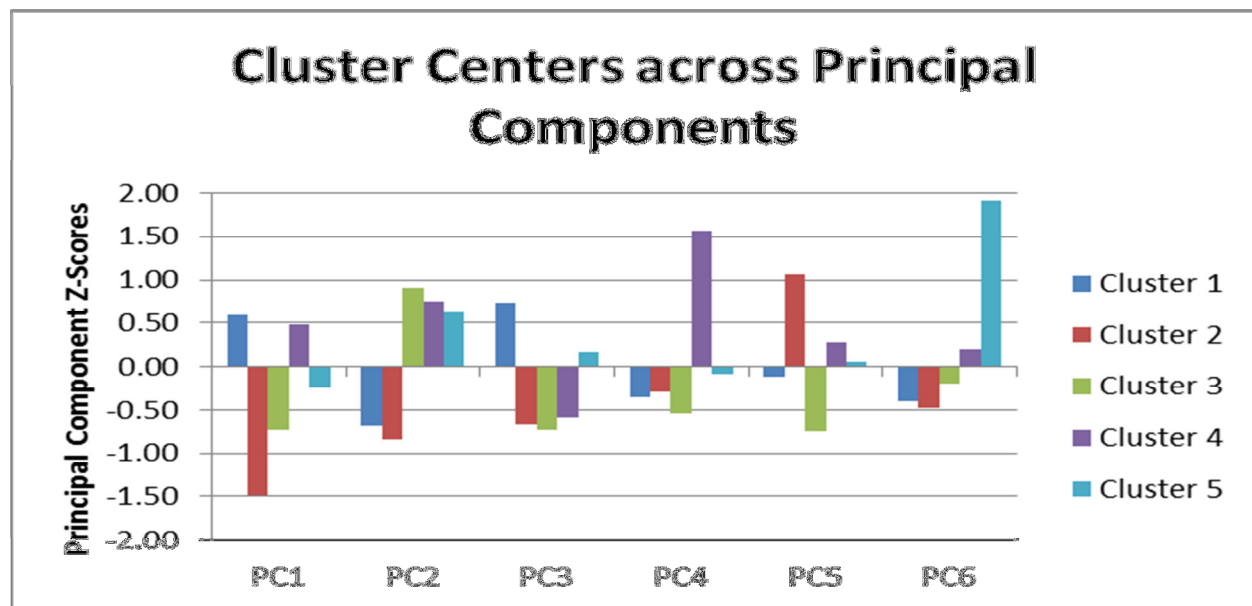
3.3.2. Cluster Analysis

Using the agglomeration schedule from the hierarchical cluster analysis, the first significant change in the distance coefficients was identified at stage thirty-nine. Subtracting thirty-nine from the total number of subjects, forty-four, yielded five clusters for the *K*-means analysis. The *K*-means cluster analysis assigned fifteen of the twenty typically developing children and three children with CP to Cluster #1 and was thus considered as the Control Group. The remaining typically developing children were assigned to Clusters #2 (n=1) and #4 (n=4).

Figure 3.1 shows the standardized principal component z-scores among the five clusters. PC1 was able to identify the presence of significant equinus in Clusters #2 and #3. PC2 identified increased forefoot adduction and coronal forefoot ROM in Clusters #3, #4, and #5. PC3 identified significant hindfoot varus in Clusters #2, #3, and #4. PC4 identified significant increased coronal hindfoot ROM in Cluster #4. PC5 identified increased sagittal hindfoot ROM between Clusters #2 and #3. PC6 identified an increased forefoot coronal varus and transverse adduction (supination) throughout the gait cycle along with increased transverse forefoot ROM in Cluster #5. The standardized principal component z-scores demonstrated that as the distance from the Control Group (Cluster #1) increased, the deviations in the actual segmental kinematics throughout the gait cycle became more severe (Figures 3.2 and 3.3). A summary of the number of participants among the clusters, cluster descriptions, and principal component scores are provided in Table 3.4.

3.4. Discussion

The current study identified five distinct, clinically recognizable subgroups among a sample of typically developing children and children with equinovarus due to hemiplegic CP using 3-D



PC1						PC2					
	Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5		Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5
Cluster 1						Cluster 1					
Cluster 2	< 0.001					Cluster 2	n.s.				
Cluster 3	0.001	n.s.				Cluster 3	< 0.001	0.001			
Cluster 4	n.s.	< 0.001	0.011			Cluster 4	< 0.001	0.002	n.s.		
Cluster 5	n.s.	n.s.	n.s.	n.s.		Cluster 5	0.005	0.014	n.s.	n.s.	
PC3						PC4					
	Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5		Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5
Cluster 1						Cluster 1					
Cluster 2	0.009					Cluster 2	n.s.				
Cluster 3	0.001	n.s.				Cluster 3	n.s.	n.s.			
Cluster 4	0.003	n.s.	n.s.			Cluster 4	< 0.001	< 0.001	< 0.001		
Cluster 5	n.s.	n.s.	n.s.	n.s.		Cluster 5	n.s.	n.s.	n.s.	0.001	
PC5						PC6					
	Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5		Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5
Cluster 1						Cluster 1					
Cluster 2	n.s.					Cluster 2	n.s.				
Cluster 3	n.s.	0.01				Cluster 3	n.s.	n.s.			
Cluster 4	n.s.	n.s.	n.s.			Cluster 4	n.s.	n.s.	n.s.		
Cluster 5	n.s.	n.s.	n.s.	n.s.		Cluster 5	<0.001	<0.001	<0.001	0.001	

Figure 3.1. Principal component z-scores among the five clusters. PC1: Sagittal hindfoot and forefoot equinus, PC2: Transverseforefoot adduction and coronal forefoot ROM, PC3: Coronal hindfoot varus, PC4: Coronal hindfoot ROM, PC5: Sagittal hindfoot ROM, and PC6: Coronal/Trasverse forefoot supination and transverse forefoot flexibility. P-values resulting from the one-way analyses of varaince demonstrate the differences among the clusters.

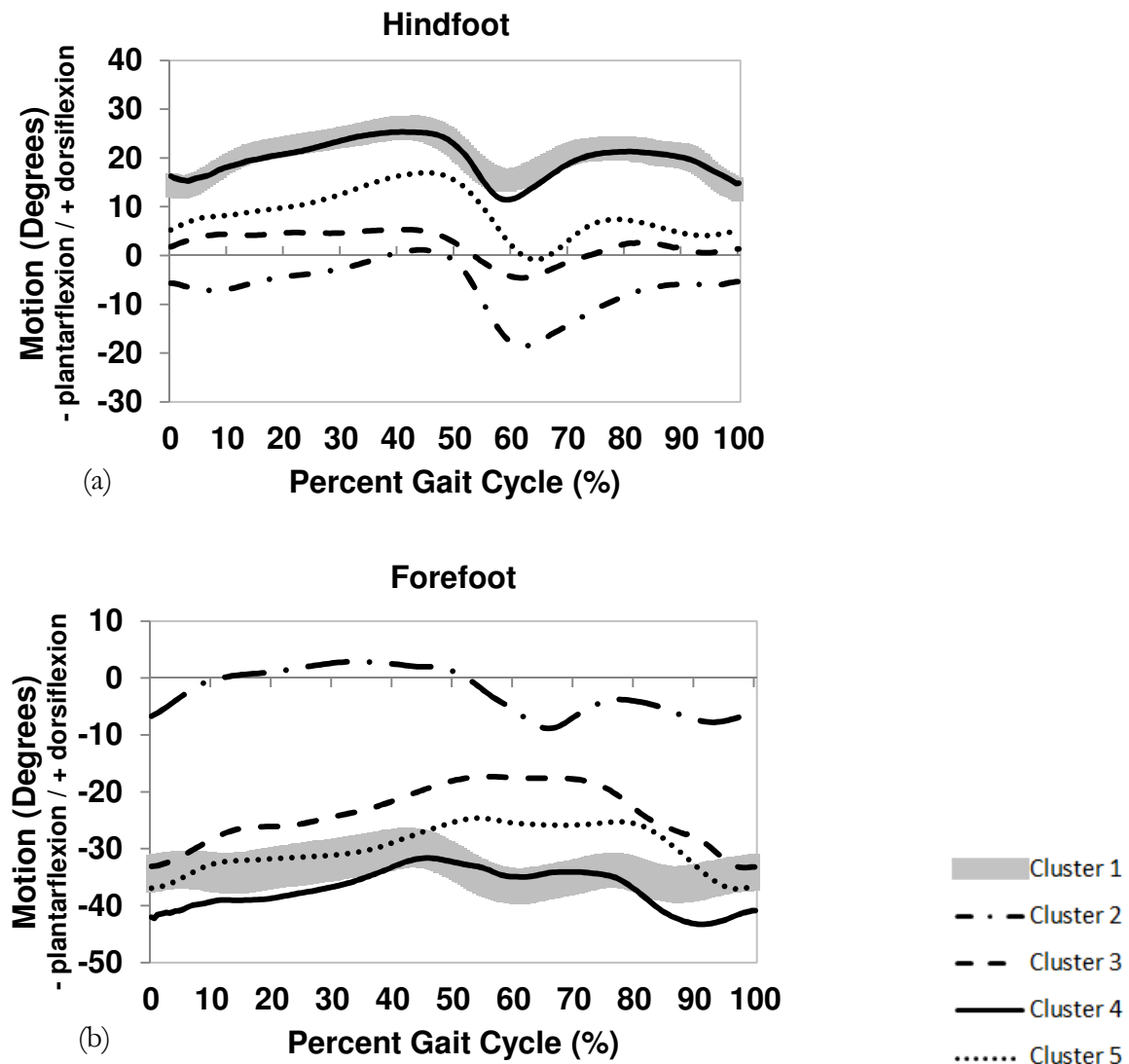


Figure 3.2. Summary of mean sagittal hindfoot (a) and forefoot (b) kinematics among clusters #2 through #4 and the plus/minus one standard error (grey band) for Cluster #1 (Control Group).

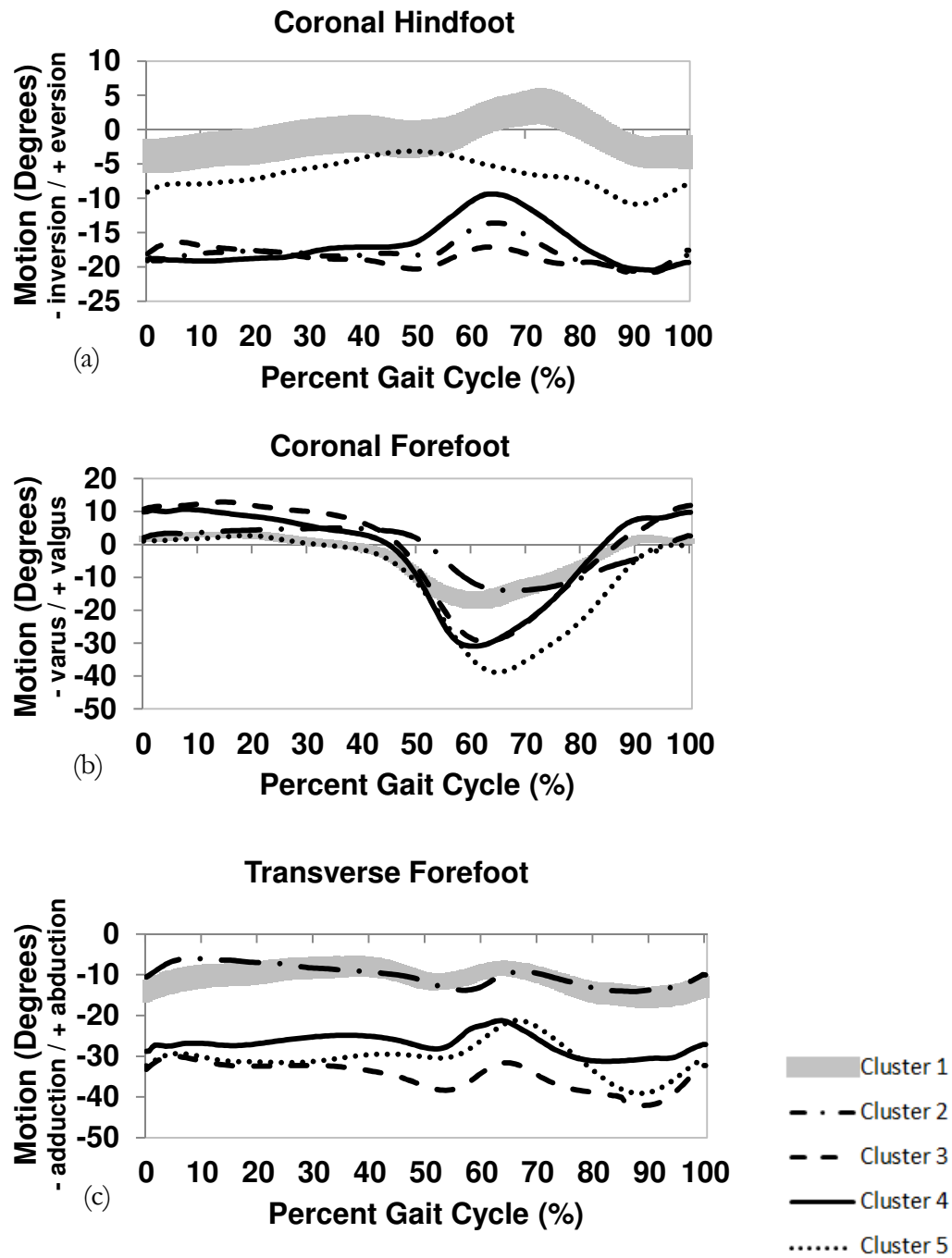


Figure 3.3. Summary of mean coronal hindfoot (a), coronal forefoot (b) and transverse forefoot (c) kinematics among clusters #2 through #4 and the plus/minus one standard error (grey band) for Cluster #1 (Control Group).

Table 3.5. Number of participants assigned to each cluster, interpretation of the clusters, and the means (SE) of the individual principal component scores

Cluster (n=44)	Description	PC1	PC2	PC3	PC4	PC5	PC6
#1 (n=18)	Control Group (Rectus)	114.3 (12.5)	86.2 (9.5)	-12 (7.9)	27.9 (2.1)	41.7 (3.7)	236 (18)
#2 (n=5)	Flexible equinovarus deformity with hindfoot involvement	-75.1 (24.5)	77.0 (22.5)	-65.0 (24.8)	28.9 (8.4)	58.9 (4.1)	225 (3.3)
#3 (n=8)	Equinovarus deformity with both hindfoot and forefoot involvement	-6.4 (23.2)	182.3 (16.5)	-67.8 (10.8)	24.5 (3.7)	32.6 (2.8)	26.4 (3.1)
#4 (n=8)	Flexible varus deformity with both hindfoot and forefoot involvement (Cavus)	104.1 (33.7)	172.5 (12.5)	-61.3 (7.4)	62.0 (5.4)	47.4 (4.5)	314 (4.3)
#5 (n=5)	Varus deformity with forefoot involvement	39.0 (15.7)	164.7 (14.7)	-26.7 (20.8)	32.6 (5.2)	44.1 (5.9)	54.0 (5.8)

segmental foot and ankle gait kinematics, PCA, and *K*-means cluster analysis. PCA was able to reduce a list of forty clinically relevant variables to six independent variables that described the location and plane of involvement in the foot and ankle. The cluster analysis identified distinct groups that had similar segmental involvement, planar motion, and range of motion. These data should be helpful in better characterizing the deformity by identifying the primary segments involved, and grading severity. We anticipate that these groupings can further be utilized to improve treatment planning and outcomes, and to track progression over time.

Interestingly, although most of the typically developing children were assigned to Cluster #1, not all were statistically isolated into the same cluster. Fifteen were assigned to Cluster #1, four to Cluster #4, and one to Cluster #2. This is explained by the inherent variability of healthy feet that are asymptomatic. Three biomechanical foot types have been commonly found in healthy adults: planus (low arched with valgus hindfoot and/or varus forefoot), rectus (well aligned hindfoot and forefoot), and cavus (high arched with a varus hindfoot and/or valgus forefoot) (Root 1977; Hillstrom et al. 2012; Mootanah et al. 2012). In the current study, Cluster #1 can be identified as having a rectus foot type with a well aligned hindfoot and forefoot (Figures 2 and 3). Cluster #4 is consistent with a cavus foot type which includes coronal hindfoot varus throughout the gait cycle, coronal forefoot valgus during stance, increased peak coronal forefoot varus at the end of stance phase, and transverse forefoot adduction throughout the gait cycle (Figure 3). In the current study 4/20 (20%) of typically developing feet were identified as cavus which is consistent with previous research on larger samples of healthy adults (Ledoux et al. 2005; Hillstrom et al. 2012).

It has been reported that there are multiple types of equinovarus in children with CP (Theologis and Stebbins 2010). This variability results from the combination of possible neuromuscular contributors that affect the biomechanics of the foot. EMG studies have demonstrated that varus deformity in children with hemiplegic CP resulted from the anterior tibialis activation alone in 34% of

cases, posterior tibialis alone in 33%, both muscles in 31%, and muscles other than the anterior or posterior tibialis in 2% (Michlitsch et al. 2006). Additionally, the ankle plantar flexors, particularly the soleus, are potential contributors to equinovarus as they act as subtalar invertors due to a medial insertion of the Achilles tendon on the calcaneus (Perry and Burnfield 2010). Non-phasic firing patterns or contracture of any or all of these muscles can impact the segmental motion of the foot in multiple planes. Thus, it is fitting that distinct subgroups within a sample of children with equinovarus were identified in the present study when segmental foot and ankle kinematic analysis was performed.

Cluster #1 was mainly comprised of typically developing children, whereas, participants with CP were primarily assigned to Clusters #2, #3, #4 and #5. Participants in both Clusters #2 and #3 presented with equinus (PC1) and hindfoot varus (PC3) (Figures 1, 2, and 3a). However, participants in Cluster #3 additionally exhibited forefoot involvement (PC2). The combination of equinus and hindfoot varus is consistent with involvement of the plantar flexors and/or the posterior tibialis (Piazza et al. 2001; Perry and Burnfield 2010). Cadaveric studies have identified that the posterior tibialis has the largest inversion moment arm of all of the subtalar joint invertors and additionally acts as a plantar flexor of the talocrural joint (Piazza et al. 2001). Thus, treatment of the feet in Clusters #2 and #3 should target the plantar flexors and the posterior tibialis to address the combination of equinus, hindfoot varus, and forefoot adduction. Participants in Cluster #4 also presented with hindfoot varus (PC3), but they did not have equinus (PC1) (Figures 1 and 3a). The lack of equinus can eliminate the involvement of the plantar flexors, and along with forefoot involvement (PC2), directs attention to the anterior tibialis. The anterior tibialis' insertion on the first metatarsal also creates an inversion moment about the subtalar joint. Additionally, the anterior tibialis' insertion on the forefoot creates a dorsiflexion moment about the talocrural joint. This dorsiflexion moment arm is larger than the plantar flexion moment arm of the posterior tibialis (Piazza et al. 2001). Participants in Clusters #3, #4, and #5 each demonstrated significant coronal and transverse forefoot deviations, as well as,

increased forefoot ROM (PC2 and PC6). Thus, the anterior tibialis most likely contributes to the deformity in these individuals, and surgical interventions including a split transfer of the anterior tibialis to the cuboid may be indicated.

A limitation in the current study was that the results were based on a specific patient population with a flexible equinovarus deformity secondary to hemiplegic CP. Therefore, generalization of these results to other patient populations commonly presenting with equinovarus deformity, such as diplegic cerebral palsy, talipes equinovarus, and Charcot-Marie-Tooth, should be cautioned. Another limitation was that even in an effort to create an objective method for identifying homogeneous groups of children with equinovarus, some level of subjective interpretation was still required. For example, to determine the a-priori number of K clusters, identification of the first significant change in distance coefficients following the hierarchical cluster analysis was required. This was performed by looking at a scree diagram of the agglomeration schedule produced by the hierarchical cluster analysis and choosing the point where the first significant change occurred. Regardless, highly significant differences were observed in the final comparisons of the principal component scores among the clusters, findings were consistent with previous reports, and a clear clinical interpretation of the results was made.

In summary, the current study presented an objective means to classify the segmental foot and ankle kinematics in children with equinovarus deformity secondary to hemiplegic CP and typically developing children. Five distinct kinematic clusters were identified with involvement of the different foot segments, in different planes, and varying degrees of ROM when compared to a control group. These quantitative methods can ultimately be used to analyze severity, track progression of deformity, and plan treatments in this common foot and ankle deformity in children with hemiplegia.

CHAPTER 4.

4.0 EFFECT OF SOFT TISSUE BALANCING SURGERIES ON FOOT MOTION IN YOUTH WITH EQUINOVARUS

4.1. Introduction

Children with hemiplegic cerebral palsy (CP) often present with a plantar flexed ankle, a varus hindfoot, and/or adducted forefoot on their involved side resulting in an equinovarus deformity. Orthopaedic surgery, including soft tissue balancing procedures, is typically indicated when conservative interventions are no longer effective. Soft tissue balancing procedures include combinations of musculotendinous lengthenings and split tendon transfers. They are used to reduce tightness and balance overpowering torques generated at the foot and ankle. The goal is to correct foot and ankle position during standing and motion during walking.

Two surgical techniques used to correct the equinus component of equinovarus include lengthening of the plantar flexors either at the level the intramuscular aponeurosis of the gastrocnemius and soleus or the conjoined tendon (tendo Achilles) (Green and McDermott 1942; White 1943; Tachdhian 1972; Olney et al. 1988). The objective of isolated equinus correction is to achieve five degrees of passive dorsiflexion with the knee extended while the child is under general anesthesia (Davids 2010). The decision as to which technique is indicated is made by determining whether the deformity is dynamic or fixed. The Silfverskiöld test is a common physical examination measure used to assess whether the deformity is dynamic or fixed by comparing the amount of passive ankle dorsiflexion with the knee flexed and extended (Silfverskiöld 1924). If a limitation in passive dorsiflexion is observed only when the knee is extended, the deformity is considered to be dynamic secondary to tightness or hypertonicity of the biarticular gastrocnemius, and lengthening at the level of the intramuscular aponeurosis of the gastrocnemius and soleus is indicated. Conversely, if limitations

in passive dorsiflexion are observed when the knee is flexed and extended, the deformity is considered static and a lengthening at the conjoined tendo-Achilles is indicated.

Surgical options for addressing varus and adductus include lengthening the posterior tibialis or a split transfer of the anterior or posterior tibialis. An intramuscular lengthening of the posterior tibialis can be performed at the distal third of the lower leg to correct mild varus (Majestro et al. 1971; Ruda and Frost 1971). Lengthening of the posterior tibialis weakens the muscle, reduces varus-producing torque about the subtalar joint, and results in a neutral, plantigrade foot. A split transfer of the posterior tibialis is indicated for a moderate deformity. The transfer involves dividing the distal portion of the posterior tibialis tendon longitudinally. The lateral arm of the divided tendon is then re-routed laterally and sutured into the peroneus brevis (Green et al. 1983). A split transfer of the anterior tibialis, used to address forefoot adductus, similarly includes dividing the distal portion of the tendon longitudinally. The lateral arm of the divided tendon is then re-routed laterally through a hole typically drilled into the cuboid (Barnes and Herring 1991). The objective of split transfers is to balance the asymmetrical torque that is generated by the overpowering invertor muscle so that a more neutral foot position is achieved throughout the gait cycle. To determine which muscle(s) are contributing to the deformity, current recommendations include the use of electromyographic (EMG) patterns for surgical decision making (McCarthy 2008). A combination of surface electrodes for the more superficial musculature (anterior tibialis and gastrocnemius) and fine wire electrodes for deeper musculature (posterior tibialis) is used to determine the primary neuromuscular contributor(s) of the deformity (Gage 2004). Previously reported EMG studies have demonstrated that varus deformity in children with hemiplegic CP resulted from the anterior tibialis alone in 34% of cases, posterior tibialis alone in 33%, both muscles in 31%, and muscles other than the anterior or posterior tibialis in 2% (Michlitsch et al. 2006). Furthermore, the use of visual observation alone was insufficient in determining which muscle(s) were responsible for the deformity.

Since various combinations of surgical techniques are used to correct equinovarus, it is important to objectively measure results to assess post-operative outcomes. However, the current evidence for the effectiveness of orthopaedic surgery to correct equinovarus deformity in children with CP is inconsistent. Post-operative reports of lengthening of the plantar flexors in children with CP have demonstrated improvements in physical examination measures, achieving a plantigrade foot, and normalizing ankle kinematics during gait (Tylkowski et al. 2009; Vlachou et al. 2009). However, high rates of post-operative weakness and overcorrection have also been reported, particularly after lengthening of the Achilles tendon (Segal et al. 1989; Borton et al. 2001). Split musculotendinous transfers of the tibialis anterior and posterior have been identified as effective procedures in creating a neutral foot position in cadaveric models (Piazza et al. 2001; Piazza et al. 2003). However, post-operative outcomes of surgery to address varus and adductus have demonstrated variable success rates from 67%-82%, and no procedure demonstrated a consistently good and long-lasting result (Barnes and Herring 1991; Vogt 1998; Chang et al. 2002; Scott and Scarborough 2006).

A possible explanation for these discordant results is the method of reporting outcomes. Although 3-D gait analysis was commonly used to quantitatively evaluate the effectiveness of plantar flexor lengthenings on ankle kinematics in individuals with CP, previous methods for measuring varus and adductus of the foot were unsatisfactory. Post-operative outcomes were often subjective accounts of foot and ankle morphology, as well as, observational analysis of standing and walking. Subjective accounts of post-operative outcomes are of significant concern due to inter-observer variability, and in fact, success may actually be overstated if not measured objectively. One study reported post-operative results of “excellent” if the child presented with a plantigrade foot and no fixed or positional deformity (Scott and Scarborough 2006). Another study reported that surgery was successful if the foot appeared “perfectly balanced” (Vogt 1998). Still, other postoperative results have been based on criteria established by Kling et al. (1985) in which surgery was considered successful if

the patient was observed to have a plantigrade foot, without fixed or dynamic deformity, used a regular shoe, and had no abnormal callosities. An objective criterion to measure post-operative success would yield a non-biased and comparable result.

In the previous chapter, the Milwaukee Foot Model (MFM) (Kidder 1996; Myers et al. 2004) and principal component analysis (PCA) were used to characterize equinovarus gait patterns in children with hemiplegic CP. Characterization was performed by initially reducing a subset of thirty-eight segmental foot and ankle kinematic variables, walking speed, and age at the time of the preoperative evaluation to six independent principal components (PCs) using a sample of typically developing children and children with hemiplegic cerebral palsy. The six PCs included:

- PC1: Sagittal hindfoot and forefoot equinus
- PC2: Transverse forefoot adduction and coronal forefoot ROM
- PC3: Coronal hindfoot varus
- PC4: Coronal hindfoot range of motion
- PC5: Sagittal hindfoot range of motion
- PC6: Coronal/Transverse forefoot supination and transverse forefoot ROM

PC scores (PC1-PC6) were then calculated for each participant and five subgroups of kinematic patterns were identified using *K*-means cluster analysis. Each subgroup presented with distinct combinations of segmental involvement in multiple planes and various ranges of motion. Membership to a particular subgroup offers individual pre-operative characteristics that can assist with surgical decision making or explain why certain post-operative results were observed. Among the subgroups, Cluster #1 was primarily comprised of typically developing children with a hindfoot and forefoot aligned in neutral. Participants in Cluster #1 presented with segmental foot characteristics consistent with one of the three biomechanical foot types identified in a normal population as

described by Hillstrom, a ‘rectus’ foot type (Root 1977; Hillstrom et al. 2012; Mootanah et al. 2012). Given the neutral alignment, the rectus group is an appropriate control group for comparing pre and post-operative kinematics in youth with equinovarus. If surgical soft tissue balancing procedures are able to effectively decrease or redistribute the excessive plantar flexion, adduction, and varus producing torques associated with equinovarus, it is plausible to expect that post-operative kinematics should approach neutral and resemble those of the rectus group.

It is widely accepted that temporal-spatial parameters, specifically walking velocity, influence gait patterns (Stansfield et al. 2006; Schwartz et al. 2008). Gait kinematics at the hip, knee, and ankle are known to be different in individuals who walk with faster or slower velocities. Schwartz et al. (2008) described differences in gait kinematics as amplifications of peak values with increasing velocity. However, this relationship has not been established with segmental foot and ankle kinematics. It is important to know if changes in segmental foot and ankle kinematics occur due to surgery, and whether those changes are secondary to surgery or merely a result of alterations in walking velocity.

The purpose of the present study is to quantitatively describe the effect of surgical soft tissue balancing procedures for equinovarus deformity on gait patterns in youth with hemiplegic CP. Specifically, gait deviations relative to a control group (the rectus group described in Chapter 3) were compared pre- and post-operatively in children with hemiplegic CP using the six PC scores that describe equinovarus foot and ankle kinematics. We tested the hypothesis that pre-operative PC scores would have greater deviations from a rectus foot type than post-operative PC scores. We further evaluated the effect of surgical soft tissue balancing procedures on walking velocity along with the relationship between walking velocity and the six PC scores. We hypothesized that increases in walking velocity would be correlated with increases in PC scores describing segmental foot and ankle kinematic peaks and range of motion. Finally, we evaluated individual changes in gait pattern resulting from surgery. Characteristics such as pre-operative cluster membership and the

combinations of surgical techniques chosen for correction were used to explain why certain individuals presented with uncorrected gait deviations, post-operatively.

4.2. Methods

4.2.1. Participants

Fifteen children and adolescents with equinovarus foot secondary to hemiplegic CP were included in the present study. They represented a sample of convenience comprised of participants with unilateral equinovarus secondary to hemiplegic CP, scheduled for a diagnostic gait analysis with a plan for surgical correction including soft tissue balancing procedures, and were willing to commit to pre and post-operative visits. Evaluations included quantitative gait analysis and segmental foot and ankle kinematic analysis using the MFM. All subjects had no prior history of orthopaedic surgery for equinovarus and had not received botulinum toxin (Botox®) injections within 1 year prior to evaluation. Participants attended a single visit to the Motion Analysis Laboratory for pre-operative assessment. Participants returned for post-operative assessment at an average of one year following surgery. The timing of the post-operative visit was chosen to allow sufficient time for patients to recover from surgery and to experience any potential benefit while minimizing the effects of growth and maturation. Children were excluded if they had bony surgeries including osteotomies or fusions. Additionally, children were excluded if they presented with cognitive or behavioral impairments that interfered with their ability to understand and follow basic commands necessary to participate in quantitative gait analysis and a standing weight-bearing x-ray series. Informed consent was provided from the participants' legal guardians and, when age-appropriate, assent/consent was obtained from the participants as approved by an institutional review board.

4.2.2. Surgical Intervention

All participants underwent surgery at Shriners Hospitals for Children® - Chicago by one group of four surgeons. Each surgeon had greater than 15 years of experience working with children with

cerebral palsy. Surgeries included the following procedures performed either in isolation or combination to address equinovarus: lengthening of the plantar flexors (at the level of the intramuscular aponeurosis of the gastrocnemius and soleus or at the Achilles tendon), intramuscular lengthening of the posterior tibialis, split transfer of the posterior tibialis to the peroneus brevis, split transfer of the anterior tibialis to the cuboid, and/or plantar fasciotomy. The choice of surgical procedure(s) was not standardized. Choices were based on a combination of available observational gait, kinematic, kinetic, EMG, and physical examination data. Following surgery, participants were allowed full weight-bearing in a short-leg, fiberglass cast for six weeks. Following cast removal, they each received a custom articulated ankle-foot-orthosis and a home exercise program consisting of active range of motion exercises for the ankle and plantar flexor stretching.

4.2.3. Instrumentation

Participants underwent pre- and post-operative gait analysis using a 14-MX camera three-dimensional motion analysis system (VICON, Motion Analysis Corp., Oxford, UK) collected at 120 Hz. Walking velocity was calculated using Vicon Workstation (version 5.2.4) software and the PlugInGait model. Simultaneously, the MFM was employed to measure the motion of multiple segments of the foot and ankle (Kidder et al. 1996). Twelve passive 9mm reflective markers were placed on the tibia, calcaneus, forefoot and hallux. A special three-marker triad was used to obtain hallux data. Resolution and accuracy of the foot and ankle system has been established by our group as well as testing reliability (Long et al. 2010). The kinematic data were processed and calculated using a custom program in Matlab (Matlab, Mathworks[®], Natick, MA, USA).

4.2.4. Experimental Protocol

A static standing trial was collected with the participant standing on a cardboard sheet where a foot tracing was made. Participants were instructed to walk “at a comfortable walking velocity” down a 30m walkway. A maximum of thirty trials were collected, and three representative trials were chosen

for analysis. Up to thirty trials were collected, pre-operatively, secondary to participation in Experiment 1 where data was collected under two conditions, with and without a fine wire electrode in the posterior tibialis. Post-operatively, between ten and twelve trials were collected.

Following gait data collection, participants were taken to the radiology department for a series of weight-bearing radiographs of the foot. Anterior-posterior, lateral, and a modified hindfoot coronal alignment view were captured while standing on the foot tracing created during the static standing trial (Johnson et al. 1999). All skeletal alignment measurements were obtained from the radiographs by the same author (JK) to provide static offsets that allow the marker set to be directly referenced to the underlying skeletal anatomy.

4.2.5. Data Analysis

Group Data

Pre and post-operative PC scores (PC1-PC6) were calculated for all participants to provide quantitative measures of the segmental and planar components of equinovarus during gait. First, averages of each of the kinematic variables associated with PC1-PC6 were computed for the involved limb using three representative gait trials. Second, PC scores were calculated using the following equation:

$$PC\ score_i = \sum_{j=1}^n \bar{x}_{ij} \alpha_j$$

The PC score is the weighted sum of the kinematic variables retained within that particular PC. The PC score ($i = 1$ through 6) is calculated for each participant ($n = 15$) by taking the sum of the products of the averaged kinematic variables (\bar{x}_j) retained within the PC (i) and the PC weighting scores (α) for each variable (j). The PC weighting scores were obtained from Experiment 2 (Table

3.3.). Finally, individual kinematic Deviance Scores were calculated to quantitatively describe gait deviations from the rectus group.

$$\textit{Kinematic Deviance Score}_{in} = \overline{PC_{Ri}} - PC_{CPin}$$

Kinematic Deviance Scores for each of the six PC Scores (*i*) were calculated for a participant (*n*) by subtracting the individual PC score of the participant with CP (PC_{CP}) from the corresponding mean PC score of the previously reported rectus group ($\overline{PC_R}$). Kinematic Deviance Scores further from zero represent greater gait deviations from the rectus group, and scores closer to zero represent gait patterns similar to the rectus group. Six, one-way repeated-measure analyses of variance were used to compare pre and post-operative Kinematic Deviance Scores for PCs 1 through 6 to determine if soft tissue balancing procedures resulted in improved post-operative gait patterns.

Effect size estimates of changes in Kinematic Deviance Scores using Cohen's *d* were calculated to measure the magnitude of the effect of surgery. Cohen's *d* values represent how many standard deviations' difference there is between the means of the pre-operative and post-operative kinematic deviance scores. In accordance with the classification scheme for interpreting effect sizes for Cohen's *d*, a large effect size was defined as having a coefficient (*d*) greater than 0.80, a medium effect size was defined as having a coefficient (*d*) between 0.50 and 0.79, and a small effect size was defined as having a coefficient (*d*) less than 0.20 (Cohen 1988).

To determine the effect of surgery on walking velocity, group averages of walking velocity were obtained using the same trials from the analysis of the kinematic data. Pre and post-operative group averages were compared using paired Student's *t*-tests. Pearson correlation coefficients (*r*) were then calculated to further analyze the association between walking velocity and the six PC scores. In accordance with Cohen's Classification for interpreting Pearson's *r*, a strong association was defined as an *r* value of greater than 0.70, a moderate association was defined as an *r* value between 0.30 and 0.70,

and a weak association was defined as an r value of less than 0.30 (Portney and Watkins 1993; Portney 1993). A probability value less than 0.05 was chosen to indicate significant differences and correlations.

Individual Data

Individual, normalized, Kinematic Deviance Scores were used to identify participants who presented with uncorrected gait deviations, post-operatively.

$$Kinematic\ Deviance\ Score_{Normalized(in)} = \frac{Kinematic\ Deviance\ Score_{in}}{sd_{CPi}}$$

Kinematic Deviance Scores for each of the six PC Scores (i) were normalized for a participant (n) by dividing the individual's post-operative Kinematic Deviance Score by the standard deviation of the Kinematic Deviance Score for the post-operative group (sd_{CP}). Normalizing allows all PCs to be on the same scale. Normalizing also facilitates establishing criteria to identify individuals who had uncorrected gait deviations by explaining the distribution of post-operative results relative to the standard deviation. The distribution of post-operative results were divided into three categories: (1) Corrected, (2) Partial Correction, and (3) Uncorrected. Figure 4.1 shows the criteria chosen to define post-operative correction using the normalized Kinematic Deviance Scores. The normalized Kinematic Deviance Scores across the six PCs were then individually evaluated for the degree of correction. When an uncorrected score was identified, individual characteristics including pre-operative cluster membership and the combination of surgical techniques chosen to correct equinovarus were evaluated to explain the uncorrected gait deviation.

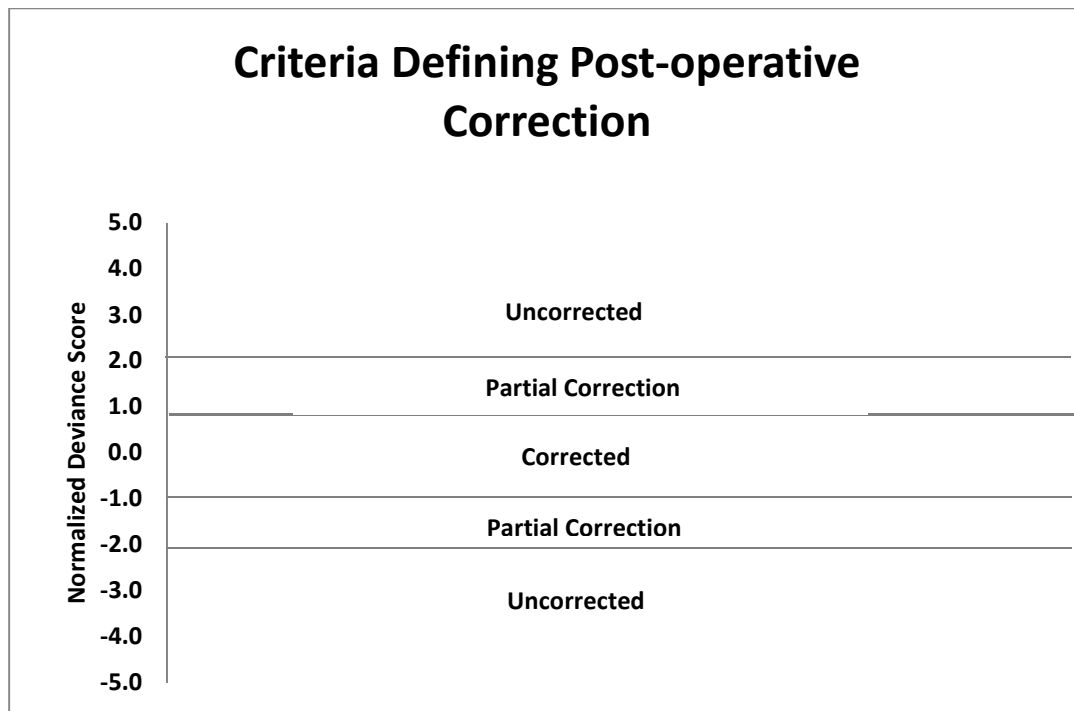


Figure 4.1. Criteria used to define post-operative correction using the normalized Kinematic Deviance Score.

4.3. Results

Among the participants, eight presented with right-sided hemiplegia and seven with left-sided hemiplegia. Based on the hemiplegic gait classification system established by Winters et al., one participant had a type I pattern, seven had a type IIa pattern, two had a type IIb pattern, three had a type III pattern, and two had a type IV pattern (Winters et al. 1987). Based on cluster membership established in Experiment 2, three participants were in Cluster #2 (Equinovarus with hindfoot involvement), six were in Cluster #3 (Equinovarus with hindfoot and forefoot involvement), one was in Cluster #4 (Flexible varus with hindfoot and forefoot involvement), and five were in Cluster #5 (Varus with forefoot involvement). The average age at the time of the pre-operative evaluation was 12.3 ± 4.5 years, and the post-operative evaluation occurred at an average of 1.0 ± 0.3 years following surgery (Table 4.1.). The surgical procedures for each participant are additionally documented in Table 1. There were six procedures either performed in isolation or combination to address equinovarus. One patient with a type IV hemiplegic gait pattern additionally underwent a transfer of the rectus femoris to the gracilis for a stiff knee gait pattern. There were no reported complications associated with surgery throughout the post-operative period.

4.3.1. Kinematics

Post-operative improvements in sagittal hindfoot/forefoot equinus (PC1, $p < 0.001$) and coronal hindfoot varus (PC3, $p = 0.02$) were observed (Figure 4.2.a and 4.2.c). Larger pre-operative Kinematic Deviance Scores were consistent with increased equinus and varus. Surgery had a large effect on reducing the degree of equinus ($d = 1.89$) and a medium effect on reducing the degree of hindfoot varus ($d = 0.70$). However, along with reductions in the degree of equinus and varus, surgery had a large effect on reducing range of motion at the hindfoot in both the coronal (PC4, $p = 0.008$; $d = 0.94$) and sagittal (PC5, $p = 0.03$; $d = 0.88$) planes (Figure 4.2.d and 4.2.e). The negative pre-operative Kinematic Deviance Scores represent increased range of motion throughout the gait cycle relative to the rectus

Table 4.1. Demographic characteristics including hemiplegic side, pre-operative cluster membership, Winter classification, age at pre-operative evaluation, surgery(ies) used to correct equinovarus, and length of time until post-operative follow-up. Surgeries used included: Split transfer of the anterior tibialis (SPLATT), lengthening of the posterior tibialis (PTL), split transfer of the posterior tibialis (SPOTT), lengthening of the myotendinous junction of the gastrocnemius and soleus (Vulpus), tendo-Achilles lengthening (TAL), and plantar fascia release (PFR).

Participant	Hemiplegic Side	Pre-operative Cluster	Winter Classification	Age at Preop
1	Right	5	I	16.7
2	Right	3	IIa	12.1
3	Right	2	IIa	10.9
4	Left	4	IIa	7.3
5	Left	5	IV	17.0
6	Left	3	IIa	10.6
7	Right	5	IIb	6.2
8	Left	3	IIa	6.8
9	Right	3	IIa	19.7
10	Left	2	IIb	11.6
11	Left	5	IIa	5.7
12	Right	3	III	17.3
13	Left	2	III	12.8
14	Right	5	III	14.3
15	Right	3	IV	16.0
Averages/Totals		8 Right/7 Left		12.3 (4.5)

Table 4.1. (continued)

Participant	SPLATT	PTL	SPOTT	Vulpus	TAL	PFR	Postoperative Follow-up (years)
1			x				1.7
2	X	x			x		1.0
3	X	x				x	0.6
4			x				1.0
5			x				0.5
6					x		1.1
7	X	x					0.9
8			x	x			0.9
9		x			x		1.0
10			x	x			0.8
11			x	x		x	0.8
12			x	x		x	0.8
13		x	x		x		1.0
14	X	x				x	0.9
15			x		x		0.7
Averages/Totals	4	6	9	4	5	4	1.0 (0.3)

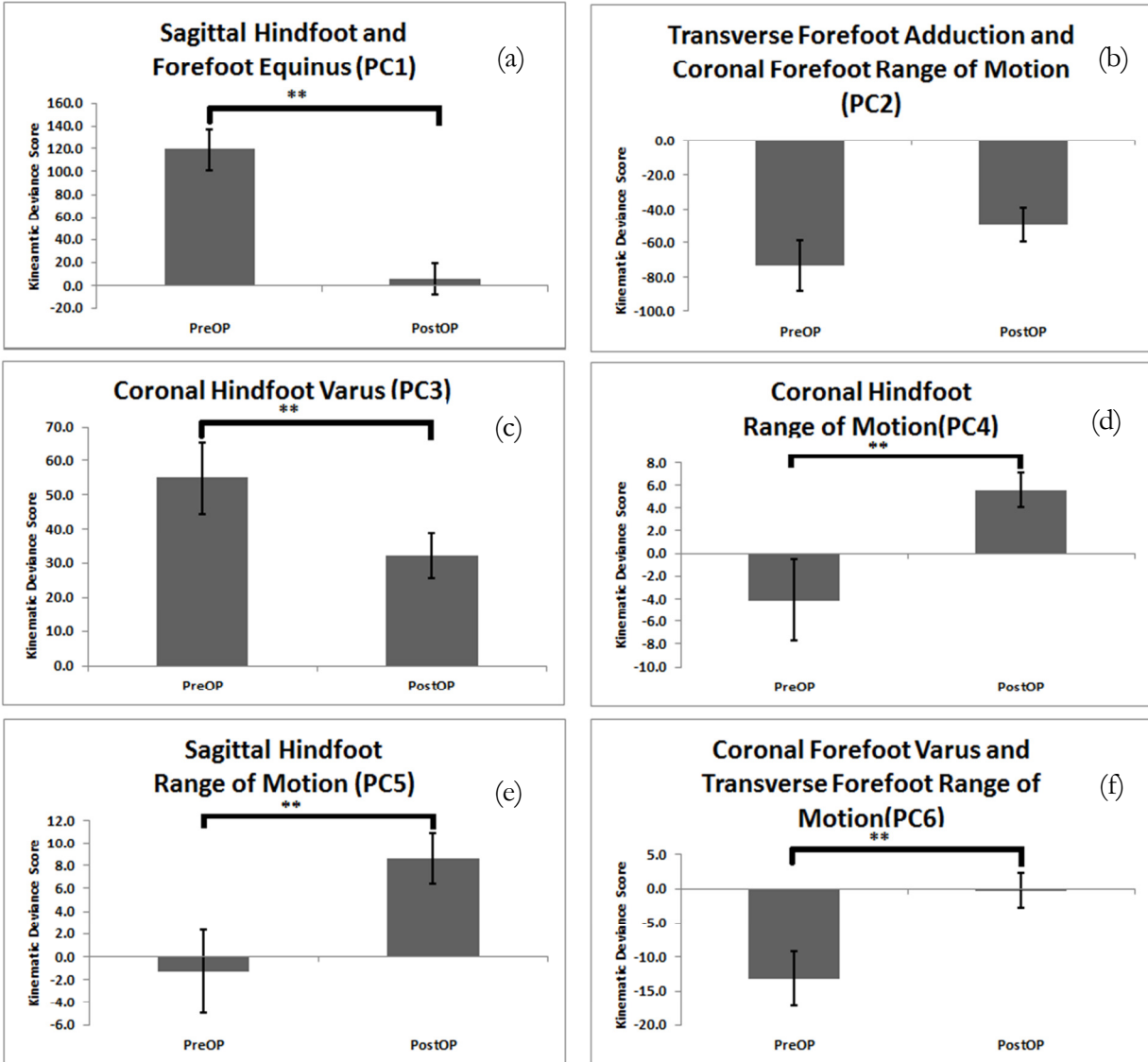


Figure 4.2. Group averages and standard error bars of pre and post-operative hindfoot and forefoot kinematic deviance scores among the six PCs. ** indicates a significant difference between pre and post-operative values $p \leq 0.05$.

group. Post-operatively, the large positive Kinematic Deviance Scores represent a reduction in range of motion relative to the rectus group. At the forefoot, improvements were observed in the coronal plane as surgery had a large effect on the degree of postoperative forefoot varus (PC6, $p=0.006$; $d=1.03$). Surgery did not have a significant effect on the amount of transverse plane forefoot adduction (PC2, $p=0.23$).

Changes in Kinematic Deviance Scores were consistent with changes in the hindfoot and forefoot kinematics during walking. Reductions in the amount of equinus (PC1) and varus (PC3) were also apparent in the sagittal hindfoot/forefoot and coronal forefoot kinematic plots as the postoperative curves approached those of the rectus group (Figure 4.3.a, b, and c). This was also observed at the forefoot in the coronal plane with a reduced mean postoperative PC6 Kinematic Deviance Score and improved coronal forefoot kinematics over the gait cycle (Figure 4.3.d). The lack of significant change in the amount of transverse forefoot adduction was reinforced by the gait kinematics with minimal change in the degree of forefoot adduction throughout the gait cycle (Figure 4.3.e).

4.3.2. Walking Velocity

Comparisons between pre and post-operative walking velocities demonstrated non-significant differences (pre-operative: $0.91 \pm 0.18 \text{ m/s}$, post-operative: $0.96 \pm 0.18 \text{ m/s}$; $t_{14} = -1.71$, $p = \text{n.s.}$, $d = 0.29$). Correlation analysis demonstrated only one significant association between walking velocity and the six PC scores. A positive, moderate correlation between walking velocity and PC5 was observed ($r = 0.33$, $p = 0.02$) demonstrating that increases in walking velocity were associated with increases in sagittal plane hindfoot range of motion.

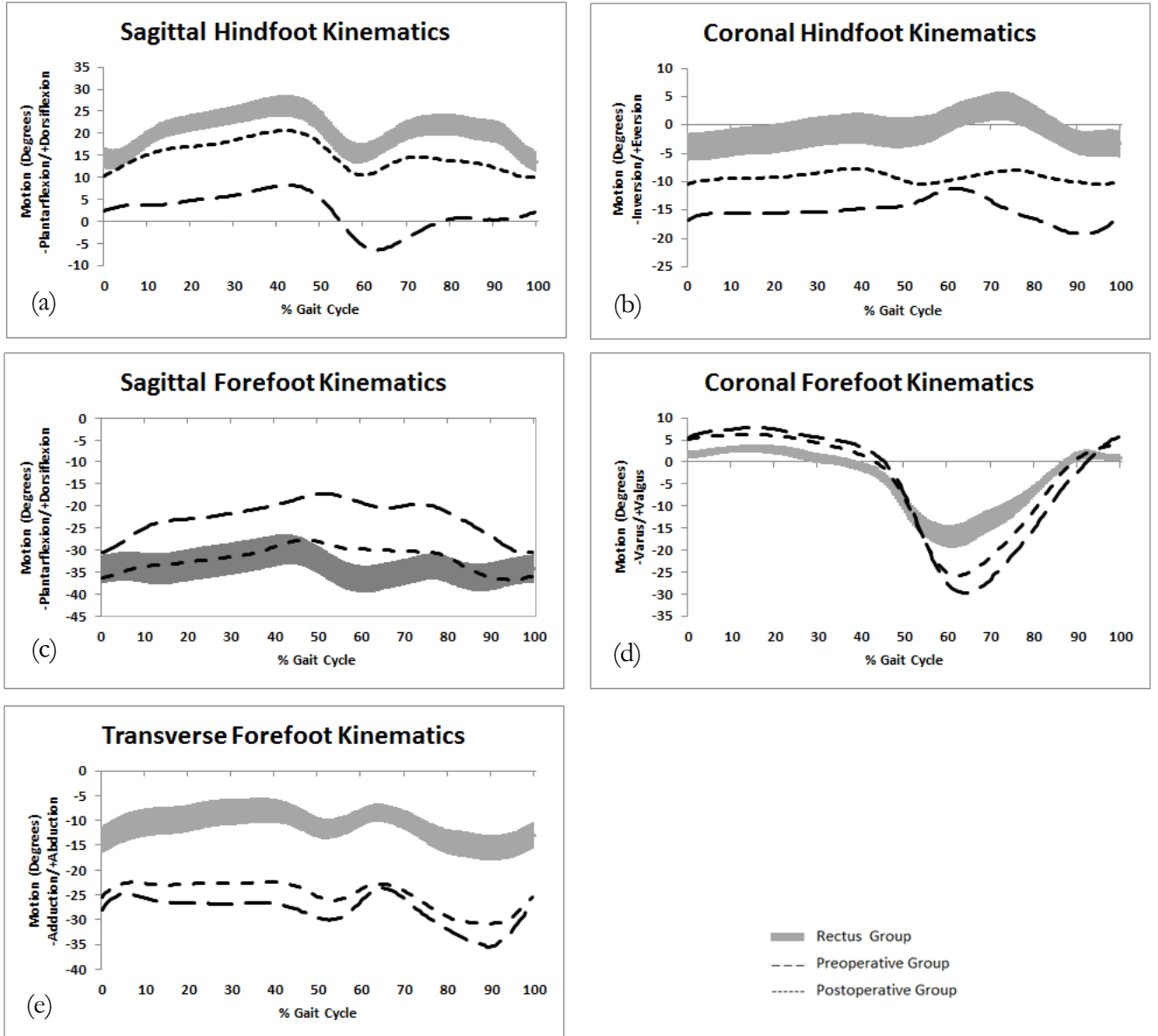


Figure 4.3. Group averages of hindfoot and forefoot gait kinematics for the Rectus (grey band \pm 1 standard error), pre-operative (thin dash) and post-operative (thick dash) groups.

4.3.3. Individual Kinematic Results

Figure 4.4. shows the individual normalized Kinematic Deviance Scores across the six PCs. There were seven participants (47%) with post-operative gait deviations, and one of the participants presented with deviations in two of the PCs (PC4 and PC5).

PC2

There were four participants who had uncorrected forefoot adductus (PC2). Each participant was identified as being a member of a pre-operative subgroup that indicated forefoot adductus. Two participants were in Cluster #3 (Equinovarus with hindfoot and forefoot involvement), one participant was in Cluster #4 (Flexible varus with hindfoot and forefoot involvement), and one participant was in Cluster #5 (Varus with isolated forefoot involvement). However, when reviewing the combinations of surgeries used to correct the foot deformity, none of these individuals had a split transfer of the anterior tibialis to address forefoot adductus.

PC3

There was one participant who had uncorrected hindfoot varus (PC3). This individual was identified as being a member of a pre-operative subgroup that indicated hindfoot varus, Cluster #2 (Equinovarus with isolated hindfoot involvement). The participant underwent both an intramuscular lengthening of the posterior tibialis and a split transfer of the posterior tibialis to the peroneus brevis. Post-operative change in PC3 Score (Coronal Hindfoot Varus) was consistent with the group average (Individual Change Score: 42.5; Group Average: 35.5 ± 19.0). However, this individual had the greatest amount of pre-operative varus when compared to the rest of the sample (Individual PC3 Score: 150.7; Group Average: 48.2 ± 32.1). Even though the combination of procedures used to address hindfoot varus resulted in an average change in PC3 Score, a more robust procedure to address such a severe deformity may have been indicated, such as a lateral closing wedge osteotomy of the calcaneus (Dwyer osteotomy) (Barenfeld et al. 1967).

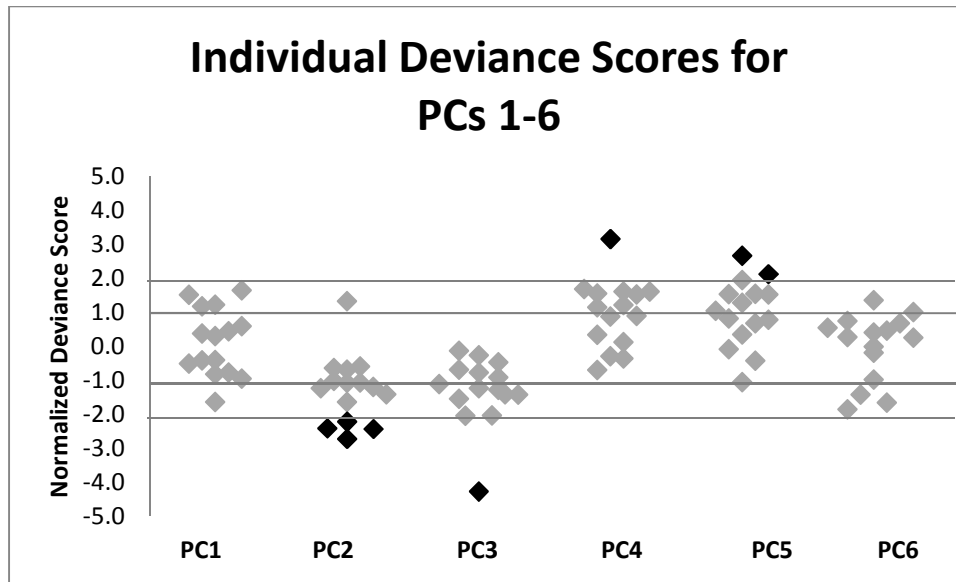


Figure 4.4. Individual Normalized Kinematic Deviance Scores for the six Principal Component Scores. Grey diamonds indicate the participants who had a “Corrected” or “Partially Corrected” foot following surgery. Black diamonds indicate individual deformities that were “Uncorrected” following surgery.

PC4

The group mean demonstrated an overall decrease in post-operative coronal hindfoot ROM (PC4) with one participant possessing a significantly greater limitation. This individual was placed in a pre-operative subgroup that identified forefoot adductus, Cluster #5 (Varus with isolated forefoot involvement). Pre-operative EMG data demonstrated continuous activity of the anterior and posterior tibialis throughout the stance phase of the gait cycle. This participant underwent a split transfer of the posterior tibialis to the peroneus brevis. Continuous anterior tibialis EMG activity throughout stance phase persisted, post-operatively. One possible explanation for limited coronal hindfoot motion, post-operatively, is that the untreated contribution of the anterior tibialis continued to pull against the transferred lateral arm of the posterior tibialis, preventing any excursion of the hindfoot in the coronal plane. Pre-operative hindfoot varus was not identified based on cluster membership; therefore, an isolated split transfer of the anterior tibialis may have resulted in a better post-operative correction.

PC5

The group average demonstrated an overall decrease in sagittal hindfoot ROM (PC5), and two participants had significantly greater reductions in ROM. One of the individuals also had limitations in coronal hindfoot ROM (PC4). Pre-operatively, this individual presented with a Winter type IV hemiplegic gait pattern and decreased pre-operative sagittal plane ROM at the hip, knee and ankle, as well as a drop foot during swing phase. In addition to a split transfer of the posterior tibialis, a transfer of the rectus femoris to the gracilis was also included to address limitations in knee ROM during swing phase. The decreased ROM persisted, post-operatively, which explains why this participant demonstrated such significant deficits in ROM. The other participant was placed into Cluster #2 (Equinovarus with isolated hindfoot involvement). This individual underwent a tendo-Achilles lengthening, intramuscular lengthening of the posterior tibialis, and a split transfer of the anterior tibialis to the cuboid. High rates of post-operative plantar flexor weakness and overcorrection have

been reported following surgical lengthening, particularly after lengthening of the Achilles tendon (Segal et al. 1989; Borton et al. 2001). Lengthening of the posterior tibialis also results in weakness of a muscle that is not only an invertor of the subtalar joint but also an ankle plantar flexor. The diminished post-operative ROM of the hindfoot in the sagittal plane can be explained by the residual weakness of the plantar flexors following the tendo-Achilles and posterior tibialis lengthenings.

4.4. Discussion

The current study presented quantitative methodology to evaluate the effect of surgical soft tissue balancing procedures for equinovarus on gait patterns in youth with hemiplegic CP. Group means of pre and post-operative Kinematic Deviance Scores that quantitatively described segmental foot and ankle deviations demonstrated that post-operative hindfoot and forefoot kinematics were more neutral than pre-operative values. However, along with improvement in the degree of deformity, surgery resulted in reduced post-operative ROM at the hindfoot in both the sagittal and coronal planes. The fact that walking velocity did not change appreciably after surgery and only one association was found between walking velocity and PC scores suggests that speed was not a factor in post-operative changes. Individual cases of uncorrected deviations can be attributed to two causes: (1) the specific segmental deformity was not identified pre-operatively, and therefore not surgically addressed, and (2) the deformity was so severe that a soft tissue balancing procedure could not correct it. Together, these data provide objective measures of the effectiveness of soft tissue balancing procedures in this patient population and methodology to identify post-operative success.

Piazza et al. conducted a series of cadaveric studies to assess the effect of split transfers of the anterior and posterior tibialis on the invertor/evertor and plantar flexor/dorsiflexor moment arms about the subtalar and talaocrural joints (Piazza et al. 2001; Piazza et al. 2003). Their results demonstrated that prior to transfer, moment arm measurements using tendon excursion values identified that the posterior tibialis was a more powerful ankle invertor than the anterior tibialis.

Attention to the strong contribution of the posterior tibialis to hindfoot varus was demonstrated in the current study as 14/15 (93%) of participants had surgical lengthening and/or split transfer of the posterior tibialis to address varus. One of the participants in the current study was observed to have significant residual hindfoot varus (PC3) despite having both a split transfer and an intramuscular lengthening of the posterior tibialis. Even though the amount of surgical correction was consistent with the group mean, the combination of surgical procedures was not robust enough to address such a severe pre-operative deformity. Thus, in the presence of a severe varus deformity, soft tissue balancing may not be sufficient and a more invasive technique such as a calcaneal osteotomy may be more appropriate.

The cadaveric series also demonstrated that following split transfer, the posterior tibialis acted as a “balanced yoke” where in the neutral position it acted as neither an invertor nor an evertor. Furthermore, when the hindfoot was taken passively into inversion, the posterior tibialis acted as an evertor and when passively taken into eversion, it acted as an invertor. This finding assists in the explanation of the salient post-operative reduction in range of motion at the hindfoot (PCs 3 and 4) observed in the current study, particularly in the coronal plane. The lateral arm of the transferred tendon restricts the excessive varus that was observed pre-operatively and ultimately reduces the hindfoot range of motion in the coronal plane. When the restriction of the lateral arm is coupled with continuous activation of the other subtalar invertor, the loss of motion can become greater. This was the case with an individual who underwent a split transfer of the posterior tibialis and presented with continuous EMG activity of the anterior tibialis throughout stance phase. It is plausible that the untreated contribution of the anterior tibialis continued to pull against the transferred lateral arm of the posterior tibialis, further preventing excursion of the hindfoot in the coronal plane. The decrease in the sagittal plane hindfoot range of motion represents the combination of reduced post-operative plantar flexion motion at preswing and reduced hindfoot dorsiflexion during swing phase. It is widely

accepted that one of the most frequent undesired results of intramuscular lengthening surgeries is high rates of post-operative weakness and overcorrection, particularly after lengthening of the Achilles tendon (Segal et al. 1989; Borton et al. 2001). One participant in the current study presented with greater loss of sagittal plane hindfoot motion following lengthening of both the Achilles tendon and the posterior tibialis. Post-operative weakness resulted in limitations in peak ankle plantarflexion contributing to the overall loss of motion. In another participant, significant limitations in motion were identified pre-operatively with a reduction in swing phase dorsiflexion resulting in a drop foot. Split transfer of the invertors in cadaver models, had no effect on the sagittal plane moment arm in reference to the talocrural joint. Thus, an identified a pre-operative drop foot during swing phase remained post-operatively.

Finally, the cadaveric series demonstrated that variability in tensioning of the medial and lateral arms of subtalar invertors only significantly affected the moment arm distance when one arm of the tendon was completely on slack. The authors concluded that the split transfers of the anterior and posterior tibialis are robust procedures that can effectively create a neutral position of the foot and ankle in the presence of a varus foot deformity. These findings are further supported in the current study, where even in the presence of four different surgeons; soft tissue balancing procedures were able to consistently result in improvements in segmental kinematics of the hindfoot and forefoot in the sagittal and coronal planes.

Using the Oxford Foot Model, Stebbins et al. identified improvements in forefoot kinematics in all three planes following foot surgery in children with spastic hemiplegic CP (Stebbins et al. 2010). The current study also identified improvements in sagittal and coronal forefoot kinematics (PCs 1 and 6); however, significant changes in transverse forefoot adduction were not observed (PC2). This discrepancy can be explained by the types of surgical procedures chosen in the two studies to correct the equinovarus foot deformity. In the Stebbins study, nine of the twelve participants (75%) had a

split transfer of the anterior tibialis to the cuboid; whereas, in the current study only four of fifteen (27%) participants underwent split transfer of the anterior tibialis. During a split transfer of the anterior tibialis, the lateral arm of the transferred tendon is inserted into a hole typically drilled in the cuboid. The function of this lateral insertion is to restrict forefoot adduction during the gait cycle. However, previous reports have identified long-term overcorrection with severe valgus deformity as one of the most common findings (50% incidence) in individuals with CP who were considered to have failed surgery (Chang et al. 2002). Effect size calculations using Cohen's *d* did identify a moderate effect of surgery on transverse forefoot kinematics. Given this effect size calculation, surgery can potentially have a moderate effect on transverse forefoot kinematics. However, given the decreased utilization of the split transfer of the anterior tibialis in the current study, statistical significance was not met in the group comparison and four individuals demonstrated greater forefoot adduction post-operatively.

There was not a statistically significant change in walking velocity following surgery. This finding is consistent with other reports on the effects of orthopaedic surgery on walking velocity in children with CP (Gorton et al. 2009). The correlation analysis demonstrated that walking velocity had a moderate, positive association with PC5, sagittal plane hindfoot range of motion. Sagittal hindfoot kinematics using the MFM are calculated by measuring the rotation of the hindfoot segment relative to the tibia. These calculations can be considered similar to the sagittal plane kinematics of the ankle when using the standard single-segment foot model. Given the similarities between the two models, it is reasonable to expect comparable associations between the kinematics and walking velocity. Shwartz et al. (2008), using a single-segment foot model, demonstrated that increasing walking velocities resulted in amplifications in kinematic peaks (Schwartz et al. 2008). The range of motion measurements in the current study are calculated as a peak-to-peak measurements throughout the gait cycle, thus increases in kinematic peaks result in increased range of motion. However, since non-

significant changes in walking velocity and post-operative reductions in sagittal hindfoot range of motion were observed in the current study, it can be determined that all post-operative changes were not associated with alterations in walking velocity and were secondary to surgery.

Several limitations in the current study warrant discussion. The surgical procedures used were not standardized across the participants. Procedures were chosen based on available data including physical examination, 3-D gait analysis and EMG data as interpreted by the treating surgeon. This methodology is consistent with current recommendations for surgical decision making for equinovarus (McCarthy et al. 2008). The surgeons did not have access to the pre-operative cluster membership. Additionally, individual variations in surgical techniques are possible among the four surgeons who performed the operations in the current study. Tensioning variations of the medial and lateral arms of the tendons of the anterior and posterior tibialis have been well documented (Piazza et al. 2001; Piazza et al. 2003). However, even in the presence of multiple surgeons, consistent improvements in post-operative kinematics were identified when using these technically robust procedures. Another limitation in the current study was that a single, post-operative assessment was performed at an average of one-year post-operatively. The timing of the post-operative evaluation was chosen to allow sufficient time for patients to recover from surgery and to experience any potential benefit while minimizing the effects of growth and maturation. Future longitudinal studies including a larger sample size that assess the effect of the timing of surgery, taking pre-operative foot type and growth into consideration, would be clinically beneficial.

In summary, previous reports describing the effects of surgical soft tissue balancing procedures on equinovarus foot and ankle deformity are commonly subjective with variable success rates. Quantitative assessments of the effectiveness of these procedures are isolated to cadaveric modeling which fail to take into account dynamic foot deformities. The results of the current study provide researchers and clinicians with quantitative data that describes changes in segmental foot and ankle

motion during walking that result from surgical soft tissue balancing procedures for equinovarus deformity. The results demonstrated that surgical soft tissue balancing procedures result in more neutral hindfoot and forefoot gait kinematics in youth with equinovarus secondary to hemiplegic cerebral palsy and were not associated with alterations in walking velocity.

CHAPTER 5.

5.0 CONCLUSIONS

Three different experiments were conducted focusing on the locomotion of children with equinovarus secondary to hemiplegic cerebral palsy (CP). The first used temporal-spatial parameters and 3-D hindfoot kinematics to investigate the effects of fine wire insertion into the posterior tibialis on gait patterns in children with equinovarus. The second used 3-D hindfoot and forefoot kinematics, PCA and *K*-means cluster analysis to identify kinematic subgroups in a sample of typically developing children and children with equinovarus secondary to hemiplegic CP. The third experiment used PC scores derived in the second experiment to quantitatively investigate the effects of surgical soft tissue balancing procedures on the gait patterns of children with equinovarus secondary to hemiplegic CP. The following sections discuss conclusions for each of these experiments and possible directions for future work.

5.1. Chapter 2 Conclusions

Fine wire electromyography (EMG) is commonly used for surgical decision making in equinovarus foot deformity. Due to the invasive nature of this technique, it may have unwanted effects that alter the gait pattern. The experiment in Chapter 2 examined if fine wire insertion into the posterior tibialis muscle affects temporal-spatial parameters and hindfoot kinematics during gait in children with equinovarus secondary to hemiplegic CP.

Twelve children with equinovarus secondary to hemiplegic CP (mean age 12.5 yrs, 4 right-sided, 8 left-sided) were recruited. Temporal-spatial parameters and 3-D segmental foot and ankle kinematic gait data were collected utilizing standard gait analysis and the Milwaukee Foot Model (MFM). Three representative trials with and without fine wire electrode insertion were compared to determine the effect of electrode placement in the posterior tibialis on temporal spatial-parameters and hindfoot

sagittal, coronal and transverse plane kinematic peaks, timing of kinematic peaks, and range of motion (ROM).

No significant differences in any temporal-spatial or kinematic parameters were observed between "with wire" and "without wire" conditions. Strong correlations were observed among the gait parameters, with the exception of cadence, for the two conditions.

Fine wire insertion into the posterior tibialis had no measurable effect on the gait of individuals with equinovarus secondary to hemiplegic CP. This suggests that the simultaneous collection of segmental foot and ankle kinematics and fine wire EMG data of the posterior tibialis is acceptable for surgical decision making in this patient population.

5.2. Chapter 3 Conclusions

Equinus, varus, and adductus, often in combination, are the most common foot and ankle deformities in children with hemiplegic CP. Accurate measurement of the involved segment(s), plane(s), timing, and the range of motion (ROM) of the deformity is important when choosing appropriate interventions. Identification of segmental contributions can be performed using segmental foot kinematics; however, the challenge of integrating additional kinematic data into clinical care is developing systematic means of reducing the large amount of heterogeneous data. Previous studies have simplified kinematic data by identifying subgroups within a sample of patients. Membership to a particular subgroup was then used to assist with surgical planning. The experiment in Chapter 3 examined if clinically relevant subgroups exist among a sample of typically developing children and children with equinovarus due to hemiplegic cerebral palsy using segmental foot kinematics, principal component analysis (PCA) and *K*-means cluster analysis.

The input data matrix of the PCA consisted of a subset of thirty-eight segmental foot and ankle kinematic variables, walking speed, and age at the time of the preoperative evaluation. The kinematic variables were chosen via clinical consensus based their ability to identify specific segment(s), plane(s),

timing, and the ROM associated with the deformity. They included hindfoot and forefoot peaks of motion, ROM, and average position throughout the gait cycle. PC scores were calculated for each of the six components and defined as the weighted sum of each PC variable's weighting score and the associated raw data. A combination of hierarchical cluster analysis and *K*-means cluster analysis was used to identify the kinematic subgroups.

The data were reduced to six principal components (PC1-PC6) with eigenvalues ranging from 8.5 (PC1) to 1.5 (PC6). Weighting scores of the individual variables ranged from 0.70 to 0.97. The six PCs included:

- PC1: Sagittal hindfoot and forefoot equinus
- PC2: Transverse forefoot adduction and coronal forefoot ROM
- PC3: Coronal hindfoot varus
- PC4: Coronal hindfoot ROM
- PC5: Sagittal hindfoot ROM
- PC6: Coronal/Transverse forefoot supination and transverse forefoot ROM

Based on the *K*-means cluster analysis each subgroup presented with distinct combinations of segmental involvement in multiple planes and various ROM. The five kinematic subgroups were:

Cluster#1: Control Group (Rectus)

Cluster #2: Flexible equinovarus deformity with hindfoot involvement

Cluster #3: Equinovarus deformity with both hindfoot and forefoot involvement

Cluster #4: Flexible varus deformity with both hindfoot and forefoot involvement (Cavus)

Cluster #5: Varus deformity with forefoot involvement

These data should be helpful in better characterizing the deformity by identifying the primary segments involved, and grading severity. We anticipate that these groupings can further be utilized to

improve treatment planning and outcomes, and to track progression over time. Since segmental foot pathology is present in a wide variety of childhood neuromuscular and musculoskeletal disorders, future research would be to conduct experiments with similar systematic methods using segmental foot kinematics, PCA and *K*-means cluster analysis to identify subgroups of kinematic patterns. These patterns will identify specific segmental and planar contributions to foot deformity and allow segmental kinematics to be integrated into clinical care.

5.3. Chapter 4 Conclusions

Orthopaedic surgery, including soft tissue balancing procedures, is typically indicated to correct equinovarus foot deformity when conservative interventions are no longer effective. Soft tissue balancing procedures include combinations of musculotendinous lengthenings and split tendon transfers. The current evidence for the effectiveness of orthopaedic surgery to correct equinovarus deformity in children with hemiplegia is inconsistent. Post-operative outcomes have demonstrated variable success rates from 67%-82%, and no procedure demonstrated a consistently good and long-lasting result. Such variability could be explained by the common use of subjective descriptions of foot morphology in the literature to measure the effectiveness of surgery for equinovarus. The experiment in Chapter 4 examined the six PC scores derived in Chapter 3 describing equinovarus foot and ankle kinematics during walking, pre- and post-operatively. Reference data from the rectus group (Cluster #1) described in Chapter 3 was also used to determine if post-operative kinematics became more neutral as a result of surgery. The experiment in Chapter 4 then examined how foot and ankle kinematics change with alterations in walking velocity by evaluating the associations between walking velocity and the six PC scores. Finally, individual post-operative results were evaluated to identify whether participants presented with corrected, partially corrected, or uncorrected post-operative gait deviations. Characteristics such as pre-operative cluster membership and the combination of surgical

techniques chosen for correction were then used to explain the presence of uncorrected post-operative gait deviations.

The findings demonstrated surgical soft tissue balancing procedures for equinovarus secondary to hemiplegic CP resulted in a shift towards neutral hindfoot and forefoot kinematics during walking at a post-operative follow up of one year. However, along with improvement in the degree of deformity, surgery resulted in reduced range of motion at the hindfoot in both the sagittal and coronal planes. Individual cases of uncorrected deviations can be attributed to two causes: (1) the specific segmental deformity was not identified pre-operatively, and therefore not surgically addressed, and (2) the deformity was so severe that a soft tissue balancing procedure could not correct it. The fact that walking velocity did not change after surgery and only one association was found between walking velocity and PC scores suggests that speed was not a factor in post-operative changes. These data provide objective measures of the effectiveness of soft tissue balancing procedures in correcting foot and ankle motion during walking in children with equinovarus secondary to hemiplegic CP. Future work would include the development of software to automate the generation of segmental kinematics, calculating PC Scores, and assigning cluster membership. We are also interested in examining the effectiveness of using this methodology as a forward model. It is anticipated that identifying specific segmental deviations, using pre-operative cluster membership, in conjunction with EMG and physical examination data will guide surgeons to the appropriate surgical combinations and improve post-operative outcomes.

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7.0 APPENDIX

IRB Approval

UNIVERSITY OF ILLINOIS AT CHICAGO

Office for the Protection of Research Subjects (OPRS)
Office of the Vice Chancellor for Research (MC 672)
203 Administrative Office Building
1737 West Polk Street
Chicago, Illinois 60612-7227

Approval Notice Continuing Review

January 11, 2013

Joseph Krzak, PT PCS
Department of Kinesiology and Nutrition
Shriners Hospital for Children
2211 N. Oak Park Ave.
Chicago, IL 60707
Phone: (773) 385-5892

RE: Protocol # 2010-0266
"Equinovarus in Cerebral Palsy"

Dear Mr. Krzak:

Your Continuing Review was reviewed and approved by the Expedited review process on January 9, 2013. You may now continue your research.

Please note the following information about your approved research protocol:

Protocol Approval Period: January 9, 2013 - January 9, 2014
Approved Subject Enrollment #: 0 (0 @ UIC; 24 @ Shriner's Hospital)
Additional Determinations for Research Involving Minors: The Board determined that this research satisfies 45CFR46.404, research not involving greater than minimal risk. Therefore, in accordance with 45CFR46.408, the IRB determined that only one parent's/legal guardian's permission/signature is needed. Wards of the State may not be enrolled unless the IRB grants specific approval and assures inclusion of additional protections in the research required under 45CFR46.409. If you wish to enroll Wards of the State contact OPRS and refer to the tip sheet.
Performance Sites: UIC, Shriners Hospital for Children, Chicago
Sponsor: None
Research Protocol(s):
a) Equinovarus in Cerebral Palsy, Version #4, 6/19/12
Assent(s):
a) Subject enrollment will occur at Shriners Hospital for Children - Chicago, and assent will be obtained via Shriners Hospital for Children - Chicago IRB-approved documents. Research activities at UIC are limited to data analysis.
Parental Permission(s):
a) Subject enrollment will occur at Shriners Hospital for Children - Chicago, and parental permission will be obtained via Shriners Hospital for Children - Chicago IRB-approved

Phone: 312-996-1711

<http://www.uic.edu/depts/ovcr/oprs/>

FAX: 312-413-2929

APPENDIX (continued)

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documents. Research activities at UIC are limited to data analysis.

HIPAA Authorization(s):

- a) Subject enrollment will occur at Shriners Hospital for Children - Chicago, and HIPAA Authorization will be obtained via Shriners Hospital for Children - Chicago IRB-approved documents. Research activities at UIC are limited to data analysis.

Your research meets the criteria for expedited review as defined in 45 CFR 46.110(b)(1) under the following specific categories:

(4) Collection of data through noninvasive procedures (not involving general anesthesia or sedation) routinely employed in clinical practice, excluding procedures involving X-rays or microwaves. Where medical devices are employed, they must be cleared/approved for marketing. (Studies intended to evaluate the safety and effectiveness of the medical device are not generally eligible for expedited review, including studies of cleared medical devices for new indications.),

(5) Research involving materials (data, documents, records, or specimens) that have been collected, or will be collected solely for nonresearch purposes (such as medical treatment or diagnosis).

Please note the Review History of this submission:

Receipt Date	Submission Type	Review Process	Review Date	Review Action
01/07/2013	Continuing Review	Expedited	01/09/2013	Approved

Please remember to:

→ Use your **research protocol number** (2010-0266) on any documents or correspondence with the IRB concerning your research protocol.

→ Review and comply with all requirements on the enclosure,
"UIC Investigator Responsibilities, Protection of Human Research Subjects"

Please note that the UIC IRB has the prerogative and authority to ask further questions, seek additional information, require further modifications, or monitor the conduct of your research and the consent process.

Please be aware that if the scope of work in the grant/project changes, the protocol must be amended and approved by the UIC IRB before the initiation of the change.

We wish you the best as you conduct your research. If you have any questions or need further help, please contact OPRS at (312) 996-1711 or me at (312) 996-0865. Please send any correspondence about this protocol to OPRS at 203 AOB, M/C 672.

Sincerely,

Tricia Hermanek, BS
IRB Coordinator, IRB # 1
Office for the Protection of Research Subjects

APPENDIX (continued)

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Enclosure(s):

1. UIC Investigator Responsibilities, Protection of Human Research Subjects

cc: Charles B. Walter, Department of Kinesiology and Nutrition, M/C 517
Daniel Corcos, Faculty Sponsor, M/C 994
Allan Jackimek, Director, Environmental Health and Safety Office, M/C 932

8.0 VITA

NAME

Joseph J. Krzak, PT PCS

EDUCATION

Ph.D., Musculoskeletal Biomechanics and Motor Control
University of Illinois at Chicago
Chicago, IL, 2013

B.S., Physical Therapy
University of Illinois at Chicago
Chicago, IL, 2000

LICENCES AND CERTIFICATIONS

Licensed Physical Therapist in the State of Illinois
License #: 070.012181

Board Certified Specialist in Pediatric Physical Therapy (PCS) by the American Physical Therapy Association (APTA), 2005

SOCIETY MEMBERSHIPS

American Physical Therapy Association (APTA),
Pediatric Section

Illinois Physical Therapy Association (IPTA)

Gait and Clinical Motion Analysis Society
(GCMAS)

WORK EXPERIENCE

2007-Present, Senior Motion Analysis Laboratory
Physical Therapist
Motion Analysis Laboratory (MAL)
Shriners Hospitals for Children®-Chicago
Chicago, IL

2000-2007, Staff Physical Therapist
Department of Rehabilitation
Shriners Hospitals for Children®-Chicago
Chicago, IL

VITA (continued)

PUBLICATIONS

Flanagan A, Krzak J, Peer M, Urban M, Johnson P. Evaluation of short-term intensive orthotic garment use in children who have cerebral palsy. *Pediatr Phys Ther.* 2009; 21(2):201-4.

Graf A, Hassani S, Krzak J, Caudill A, Flanagan A, Bajorunaite R, Harris G, Smith P. Gait characteristics and functional assessment of children with type I osteogenesis imperfecta. *J Orthop Res.* Sep 2009;27(9):1182-1190.

Graf A, Hassani S, Krzak J, Long J, Caudill A, Flanagan A, Eastwood D, Kuo KN, Harris G, Smith P. Long-term outcome evaluation in young adults following clubfoot surgical release. *J Pediatr Orthop.* Jun 2010;30(4):379-385.

Krzak J, Graf A, Caudill A, Flanagan A, Smith P, Harris GF. Analysis of push-off power during Locomotion in Children with Type 1 Osteogenesis Imperfecta. *Journal of Experimental and Clinical Medicine.* 2011;3(5):195-199.

Konop K, Strifling K, Krzak J, Graf A, Harris GF. Upper extremity joint dynamics during walker assisted gait: a quantitative approach towards rehabilitative intervention. *Journal of Experimental and Clinical Medicine.* 2011;3(5):213-217.

Hassani S, Krzak J, Flanagan A, Bagley A, Gorton G, Romness M, Tykowski C, Abel M, Johnson B, Oeffinger D. Assessment of strength and function in ambulatory children with cerebral palsy by GMFCS level and age: a cross-sectional study. *Critical Reviews in Physical and Rehabilitation Medicine.* 2011;23(1):1-14.

VITA (continued)

Flanagan A, Krzak J, Hassani S, Bagley A, Gorton G, Romness M, Tykowski C, Abel M, Johnson B, Oeffinger D. Relationships among strength, body composition and measures of activity in ambulatory children with cerebral palsy: a cross-sectional study. *Critical Reviews in Physical and Rehabilitation Medicine*. 2011;23(1):15-29.

Graf A, Wu KW, Smith PA, Kuo KN, Krzak J, Harris G. Comprehensive review of the functional outcome evaluation of clubfoot treatment: a preferred methodology. *J Pediatr Orthop B*. Jan 2012;21(1):20-27.

Krzak JJ, Corcos DM, Graf A, Smith P, Harris GF. Effect of fine wire electrode insertion on gait patterns in children with hemiplegic cerebral palsy. *Gait and Posture*, Feb 2013;37(2):251-257.

Ackman J, Altiok H, Flanagan A, Peer M, Graf A, Krzak J, Hassani S, Eastwood D, Harris GF. Long-term follow up of Van Nes rotationplasty for congenital proximal focal femoral deficiency. *J Bone Joint Surg Br* 2013;95-B:192-198.

Cohen T, Altiok H, Wang M, McGrady L, Tarima S, Krzak J, Graf A, Smith P, Harris G. Evaluation of cast creep occurring during simulated clubfoot correction. *Journal of Engineering in Medicine* [in press].

Anissipour AK, Hammerberg KW, Caudill A, Kostiuik T, Tarima S, Zhao HS, Krzak JJ, Smith PA. Behavior of scoliosis during growth in children with Osteogenesis Imperfecta. *J Bone Joint Surg* [in review].

VITA (continued)

Hassani S, Krzak JJ, Johnson B, Flanagan A, Gorton G, Bagley A, Öunpuu S, Romness M, Tytkowski C, Oeffinger D. One minute walk test and timed up & go in ambulatory children with diplegic cerebral palsy: performance and minimum clinically important differences. *Dev med Child Neurol*. [in review].

BOOK CHAPTERS

Graf A, Krzak J, Harris G. Response to balance perturbation: A strategy for pediatric assessment. *Foot and Ankle Motion Analysis: Clinical Treatment and Technology*. Boca Raton, FL: CRC Press, 2007.

CONFERENCE PRESENTATIONS

Krzak J, Flanagan A, Johnson P, Jacoby N. Study of The preliminary effects of short-term, intensive treadmill training for children with cerebral palsy. American Academy for Cerebral Palsy and Developmental Medicine. New Orleans, LA, 2002.

Flanagan A, Jacoby N, Johnson P, Krzak J. Short-term intensive physical therapy to address balance and gait efficiency in an adolescent who has cerebral palsy. American Academy for Cerebral Palsy and Developmental Medicine, Los Angeles, CA, 2004.

Flanagan A, Krzak J, Peer M, Urban M, Johnson P. The effect of Theratogs on pediatric balance, gross motor skills and gait quality. American Physical Therapy Association (APTA) Combined Sections Meeting (CSM), Boston, MA, 2007.

Bustamante Valles K, Long J, Riedel S, Hassani S, Graf A, Krzak J, and Harris G. Postural control under altered sensory conditions in children with cerebral palsy. RESNA 2008 Annual Conference, Arlington, VA, 2008.

VITA (continued)

Sturm P, Hammerberg K, Hassani S, Graf A, Krzak J, Bustamante Valles K, Harris G. Gait and postural stability assessment in children with idiopathic scoliosis undergoing posterior spine instrumentation. American Academy of Pediatrics, Boston, MA, 2008.

Smith P, Graf A, Krzak J, Hassani S, Harris G. Long term follow-up after comprehensive clubfoot surgery. Gait and Clinical Movement Society. Denver, CO, 2009.

Sturm P, Hammerberg K, Hassani S, Krzak J, Graf A, Riordan M, Zaharski K, and Harris, GF. Adolescents with idiopathic scoliosis spinal range of motion compared to age-matched controls, International Research Society of Spinal Deformities. Montreal, BC, 2010.

Krzak J, Sturm P, Graf A, Hammerberg K, Gupta P, Hassani S, Riordan M, Zaharski K, Harris G. A comparison of computerized and clinical assessment of weight symmetry in adolescent idiopathic scoliosis. International Meeting on Advanced Spine Techniques. Toronto, Canada, 2010.

Krzak J, Sturm P, Graf A, Hammerberg K, Gupta P, Hassani S, Riordan M., Zaharski K, Harris G. Post-operative postural symmetry improvements in adolescent idiopathic scoliosis. International Meeting on Advanced Spine Techniques. Toronto, Canada, 2010.

Krzak J, Corcos DM, Graf A, Smith P, Harris G. The effect of wire electrode placement on hindfoot kinematics during gait in children with hemiplegic cerebral palsy. Gait and Clinical Movement Analysis Society. Bethesda, MD, 2011.

VITA (continued)

Flanagan A, Altiok H, Krzak J, Ackman J, Peer M, Hassani S. Long-term follow up of Van Nes rotationplasty for congenital PFFD. Gait and Clinical Movement Analysis Society. Bethesda, MD, 2011.

Graf A, Hassani S, Flanagan A, Caudill A, Krzak J, Long J, Kuo KN, Harris GF, Smith P. Long term outcomes of comprehensive surgical intervention vs. Ponseti casting in the treatment of idiopathic clubfoot. Gait and Clinical Movement Analysis Society. Bethesda, MD, 2011.

Slavens B, Graf A, Krzak J, Vogel L, Harris G. Upper extremity wheelchair kinematics in children with spinal cord injury. International Conference of the IEEE Engineering in Medicine and Biology Society (EMBC'11). Boston, MA, 2011.

Kuo K, Graf A, Hassani S, Krzak J, Caudill A, Long J, Dietz F, Morcuende J, Harris GF, Smith P. Long term outcomes of comprehensive surgical intervention vs Ponseti casting in the treatment of idiopathic clubfoot. International Clubfoot Congress. Prague, Czech Republic, 2011.

Krzak J, Graf A, Hassani A, Caudill A, Smith P, Harris GF. Analysis of push off power reduction in children with type 1 Osteogenesis Imperfecta. International Conference Osteogenesis Imperfecta, Dubrovnik, Croatia, 2011.

Altiok H, Riordan A, Bland M, DiPatri A, Flanagan A, Graf A, Krzak J, Hassani S. Gait analysis and its utilization in the decision making process for Tethered Cord Syndrome: a case study. Gait and Clinical Movement Analysis Society, Grand Rapids, MI, 2012.

VITA (continued)

Slavens B, Paul A, Graf A, Krzak J, Vogel L, Harris G. Upper extremity kinematics in children with spinal cord injury during wheelchair mobility. Gait and Clinical Movement Analysis Society. Grand Rapids, MI, 2012.

Hassani S, Graf A, Krzak J, Hammerberg K, Sturm P, Gupta P, Riordan M, Harris G. Assessing spinal motion at different fusion levels in adolescents with idiopathic scoliosis. Gait and Clinical Movement Analysis Society, Grand Rapids, MI, 2012.

Konop K, Long J, Krzak J, Graf A, Harris G. Segmental kinematic assessment of pediatric pes planovalgus with radiographic skeletal indexing. Gait and Clinical Movement Analysis Society. Grand Rapids, MI, 2012.

INVITED LECTURES & TEACHING EXPERIENCE

Static and Dynamic Balance Assessment Tools. The First Symposium on Pediatric Vestibular Function and Balance. Nemours/Alfred I. DuPont Hospital for Children. Wilmington, DE, 2006.

The Use of Computerized Dynamic Posturography (CDP) in the Pediatric Population: NeuroCom Midwest Users Symposium. Shriners Hospitals for Children®-Chicago. Chicago, IL 2007.

Advances in the Assessment and Rehabilitation of Balance Disorders with an Emphasis on the Patient with Neurologic Disease. Aurora Healthcare System. Milwaukee, WI, 2007.

Clinical Integration Seminar (CIS). NeuroCom International, Inc. Clackamas, OR, 2/07, 3/07, 2/08, 3/08, 6/09, and 7/10.

Postural Stability in Children – Beyond Single Leg Stance. American Physical Therapy Association, MN Chapter. Minneapolis, MN, 2008.

VITA (continued)

Applications of Motion Analysis for Children with Spinal Cord Injury. The Lawrence C. Vogel Pediatric Spinal Cord Injury Symposium. Shriners Hospitals for Children®-Chicago. Chicago, IL, 2011.

Clinical Applications of Segmental Kinematic Foot and Ankle Analysis. RUSH University Medical Center, Department of Orthopedics. Chicago, IL, 2012.

Wheelchair Seating and Mobility. Occ Thpy 593: Introduction to Biomedical and Rehabilitation Instrumentation, University of Wisconsin -Milwaukee. Milwaukee, WI, 2012.

Quantitative Assessment of the Upper Extremity. The Lawrence C. Vogel Pediatric Spinal Cord Injury Symposium. Shriners Hospitals for Children®-Chicago. Chicago, IL, 2012.

The Basics of Postural Stability Assessment and Rehabilitation – A Pediatric Focus. eSeminar, Neurocom®, a division of Natus®. 2012.

Equinovarus in Cerebral Palsy. Grand Rounds Lecture. Department of Orthopaedics, Shriners Hospitals for Children®-Chicago. Chicago, IL; Department of Biomedical Engineering, Marquette University. Milwaukee, WI, 2013.