# Evaluating and Improving the Efficacy of Ankle Foot Orthoses for Children with Cerebral Palsy

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**Abstract**

Ankle foot orthoses (AFOs) are commonly recommended for individuals with cerebral palsy (CP) as a means to improve gait. Goals of this dissertation were to evaluate the current efficacy of AFO use for children with CP, investigate the biomechanical mechanism of how AFOs influence gait, and describe new methods for analyzing and improving AFO outcomes as they pertain to gait. Retrospective data analysis, statistical machine learning, and simulation techniques were used to achieve these goals. Data analysis revealed that the general efficacy of AFO use was poor. However, a data driven model developed through machine learning techniques suggests that efficacy can likely be improved by using the model to recommend AFO prescriptions for individuals that are predicted to improve their gait with AFO use and refrain from prescribing AFOs for individuals whose gait will not improve with AFO use. Investigations of gait efficiency and muscle function revealed new factors that could potentially be leveraged to improve the efficacy of AFO use. Finally, an AFO design redundancy between two commonly prescribed AFOs was identified, eliminating misconceptions about the efficacy of a redundant AFO design. The techniques and conclusions presented in this dissertation have the potential to significantly improve the efficacy of AFO use for children with CP.

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# Chapter 1: Evaluating and Improving the Efficacy of Ankle Foot Orthoses for Children with Cerebral Palsy

## 1.1 Purpose and Significance

Cerebral palsy (CP) is a neurological disorder caused by damage to, or abnormal development of, the motor control center of the developing brain [1]. This neurological impairment disrupts an individual's muscle control, and can drastically impair walking ability [2], [3]. As a consequence, both the overall functional ability and quality of life may be significantly impacted for individuals diagnosed with CP [3], [4].

To improve the gait of individual's with CP, the ankle foot orthosis (AFO) if often prescribed [5]. An AFO is a lightweight brace that resists motion at the ankle joint. While the effectiveness of AFOs for improving the gait of individuals with CP has been investigated in case studies and small groups, the general efficacy of AFO use is still not well understood [6], [7].

This thesis evaluates the current efficacy of AFO use for children with CP, investigates the biomechanical mechanism of how AFOs influence gait, and describes new methods for analyzing and improving AFO outcomes as they pertain to gait.

## 1.2 Cerebral Palsy

Worldwide, CP occurs in approximately 1.5 – 3.3 per 1000 live births, and is the most common cause of severe physical disability in childhood [8], [9]. CP is caused by damage to the motor control center of a developing brain, affecting motor function as well as cognitive organization [1]. Additionally, neural pathways that extend outside the brain may also be affected. Spasticity, commonly observed in CP, is a term that describes a hypersensitive stretch reflex caused by damage to the pyramidal tract (which is responsible for transferring motor control signals from the brain to the spinal cord). Spasticity is prohibitive to intentional motion as any agonist muscle motion is often counteracted by a "spastic catch" of the antagonist muscle as it is being stretched. This catch results in a stiff motion for affected limbs, but may also cause the spastic muscles themselves to become stiff and contracted, limiting even slow passive motions. Consequently, having stiff and tight muscles adversely affects the developing skeletal system. Bone growth is influenced by muscle force. When muscle forces are abnormal, as they are in CP, bone growth may be abnormal. Common bony deformities observed in CP are torsional deformities like excessive femoral anteversion or tibial torsion resulting in a "pigeon toed" or "duck footed" gait, or alignment deformities like genu varum (aka "bow-leggedness") or genu valgum ("knock-kneed").

Generally, due to the etiology of CP, distal extremities are affected more severely (Figure 1.1), and, as previously mentioned, bony deformities and muscle contractures are often present as tertiary impairments. While the cause and location of the cerebral damage determine what areas motor deficits are typically manifest, it may not necessarily provide a complete picture of overall impairment, as CP often co-occurs with other developmental disabilities such as seizures or attention-deficit disorder, which increases the level and complexity of impairment [3], [10], [11].

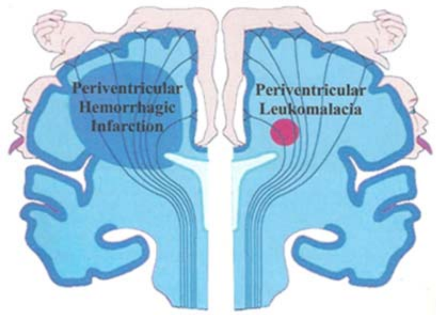


**Figure 1.1 – CP homunculus and typical lesion locations. Typical lesion locations may be widespread as seen for periventricular hemorrhagic infarction or focused as seen for periventricular leukomalacia (Adapted from [1])**

In addition to the physical challenges faced by individuals with CP, significant financial and quality of life costs are also often present. Direct (e.g. physician visits, inpatient hospital stays, assistive devices) and indirect (e.g. workplace limitations, premature death) lifetime medical costs for an individual diagnosed with CP were estimated to be nearly $1,000,000 in 2003 dollars [12]. It has been shown that poorer walking ability in children with CP is correlated with lower quality of life measures (quality of life defined as "*an individual's perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards, and concerns*") [13]. Additionally, other self-reported quality of life metrics, such as physical and psychosocial health, were reported to be significantly lower for children with CP compared to TD children [14].

## 1.3 Gait

Locomotion is an essential part of daily life. For unimpaired and experienced ambulators, walking comes easily and seems intuitive, but in reality it is the coordination of many learned motor skills working together to maintain the stability of a multiple degree of freedom system in a dynamic environment [15]. The coordination of limbs required for balance and stability is already complex, but when pathologies are present, such as those in CP, the task of locomotion can become difficult, inefficient, and energy intensive [16].

Walking, or *gait*, in its most basic form, can be viewed as a dynamic couple of two inverted pendulums [17]. One leg supports the body center of mass (COM) as it falls forward along the arc of an inverted pendulum until the opposite leg swings into a forward position to catch, recover, and again support the COM. In clinical gait analyses, discrete gait events are used to break this cyclic motion into individual gait cycles. Typically, a gait cycle begins the moment a limb strikes the ground - referred to as *initial contact* - and ends the moment the same limb strikes the ground one cycle later. From this, a single gait cycle can be subdivided into various periods and phases related to functional goals that occur throughout a gait cycle (Figure 1.2) [18].

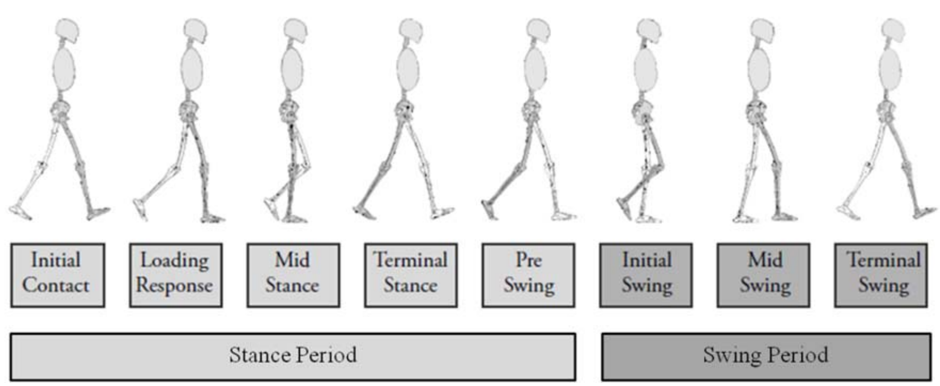


**Figure 1.2 – Phases of Gait (Adapted from [19])**

The gait of typically developing (TD) individuals has been shown to be relatively consistent between individuals [20]. It has also been shown that precise timing of force production and absorption is required to optimize the metabolic cost of walking [21], [22]. For individuals with CP, evidence has emerged showing that these individuals generally use a simplified control strategy during walking when compared to TD individuals [23]. This simplified control strategy, in conjunction with bony and soft tissue deformities, likely limit the precise control necessary to optimally redirect the COM and subsequently minimize metabolic costs or optimize other important gait qualities. As an example, during TD gait, the plantarflexor muscles have been shown to be critically important for generating forward acceleration during walking [24]. However, in CP gait, the plantarflexors are often impaired. Other muscles must compensate to provide adequate forward acceleration during walking which may lead to suboptimal muscle function [25]. If some semblance of "typical plantarflexor function" were able to be imposed, these compensatory muscles may be able to function closer to their optimal function.

### 1.4 Gait Analysis

During gait, motion occurs relatively quickly, and small movements can have large implications on gait. Therefore, precise quantification of movement is required to properly assess an individual's gait. Three dimensional gait analysis provides a means to capture this motion, and reduce this data into an analyzable form. Typical gait analysis methods utilize optical marker tracking systems to track body segment locations through space, and 6 degree of freedom (DOF) force plates to measure ground reaction forces. Optical marker systems use a series of retroreflective markers placed in specific anatomical positions on each limb so that the body may be represented as a simplified biomechanical model. Anatomical points that are difficult or impossible to identify with markers, such as the location of the hip joint center and orientation of the anatomical knee axis, are located using functional range of motion trials where the motion between two segments is used to identify the joint axis [26], [27].

The model used in this thesis is comprised 8 individual segments: a trunk, a pelvis, two thigh segments, two shank segments, and two foot segments. In the model, adjacent anatomical segments are connected by mechanical joints. The trunk/pelvis, hip, and knee joints are all modeled as 3 DOF spherical joints that allow rotation about 3 orthogonal axes while the ankle joint is modeled as a 2 DOF universal joint. Joint axes in the model are strategically oriented to represent standardized physiological definitions of planar limb motion – flexion/extension (sagittal plane), ab/adduction (coronal plane), and internal/external rotation (transverse plane). Using this model, joint angles are calculated for each segment throughout a gait cycle (*kinematics*). When joint locations are combined with ground reaction forces, joint moments and powers may also be calculated (*kinetics*). Both kinematics and kinetics are typically used to analyze gait.

## 1.5 Ankle Function

The anatomical ankle joint is located at the junction of the distal tibia and talus bones which functions much like a saddle joint. Motion at the ankle is primarily sagittal plane flexion and extension but limited inversion and eversion is also possible. However, relative motion of foot bones near the ankle joint can appear as motion through the ankle (e.g. motion between the talus and navicular bones can provide foot flexion/extension and abduction/adduction). As these motions occur relatively close to one another, it is difficult to discern motion through the ankle from motion through the foot through optical marker tracking techniques without using specific foot modeling approaches [28], [29].

For the model used in this thesis, sagittal plane ankle dorsiflexion (upward motion of the foot) and plantarflexion (downward motion of the foot) is defined as the angle between the knee and ankle joint centers and the plantar surface of the foot projected onto a plane normal to the ankle axis (i.e. sagittal plane defined by the ankle axis) (Figure 1.3). Typical ankle motion ranges between 15° of dorsiflexion and 30° of plantarflexion and typical ankle moments range from 0.1 Nm/kg of dorsiflexor moment to a peak plantarflexion moment of 1.5 Nm/kg (Figure 1.3).

The primary muscle that provides ankle dorsiflexion is the anterior tibialis (AT) and the primary muscles that provide ankle plantarflexion are the soleus (SOL) and gastrocnemius (GAS). Both the AT and SOL are uniarticular muscles, meaning they span a single joint. However, the GAS is a biarticular muscle, meaning that it spans two joints, the ankle and knee. Because the GAS is a biarticular joint, its function throughout gait is more difficult to analyze when compared to the AT and GAS. In addition to these primary muscles, there are many secondary muscles around the ankle that provide important functions such as ankle and foot inversion/eversion and toe flexion/extension. However, as these secondary muscles play a smaller role during gait compared to the AT, SOL, and GAS, they will not be discussed in detail in this thesis.

At initial contact, the ankle is generally in a neutral position near 0° of dorsiflexion, and the foot is slightly inclined, allowing the heel to make contact with the floor. When the heel contacts the floor an internal plantarflexion moment is created. To prevent the foot from slapping to the floor, eccentric muscle contraction of the dorsiflexor muscles create a small dorsiflexor moment at the ankle allowing the ankle to undergo controlled plantarflexion until the foot fully contacts the floor. Body weight is shifted to the ipsilateral limb and the contralateral foot lifts from the floor leaving the body in a single support configuration much like an inverted pendulum. As the body progresses forward, the plantarflexor muscles eccentrically contract while the ankle dorsiflexes. An increasing plantarflexion moment is generated until the contralateral limb comes into contact with the floor again putting the body in a double support configuration. Shortly after double support is resumed, the ipsilateral ankle, with a large plantarflexion moment, begins to quickly plantarflex, accelerating the body COM and shifting body weight to the contralateral limb. When body weight has been accepted by the contralateral limb, the ipsilateral foot is picked up off of the floor and begins to swing forward. As the limb swings past the contralateral limb, the ankle is quickly dorsiflexed to prevent the foot from contacting the floor at the bottom of swing. After the foot has cleared the floor, the limb continues to swing forward and the ankle plantarflexes slightly to a neutral position preparing for the next initial contact event. For typical walking, these events typically occur in less than 1 second, so fast and precise motor control is needed to accurately position and control the ankle.

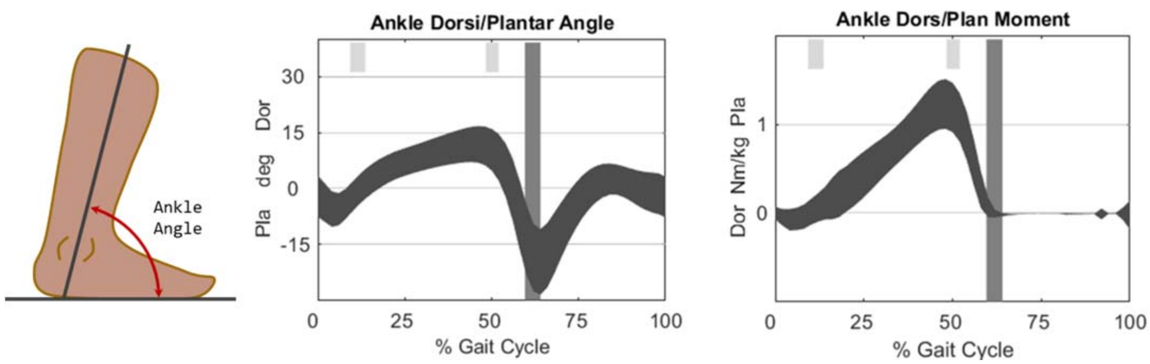


**Figure 1.3 – Typical ankle angle and moment during gait. Dark shaded areas represent typical ankle function +/- 1 SD. Medium shaded areas represent the foot off event. Light shaded blocks at the top of each graph represent the contralateral foot off (1st block) and foot contact (2nd block) events.**

For individuals with CP, control of the AT, GAS, and SOL muscles is compromised. This generally results in ankle positions and moments that do not emulate TD function. Two common gait patterns resulting from ankle dysfunction are foot drop and equinus. For a "foot drop" gait pattern, the ankle dorsiflexors do not properly dorsiflex the ankle during swing (Figure 1.4). This places the foot in a plantarflexed position during a critical moment when the foot needs to clear the floor to prepare for the next step. If the foot drop is severe and the individual is unable to compensate to effectively clear the foot (hip circumduction, vaulting on contralateral limb), the individual may drag their foot across the floor or trip. In addition to the dragging the foot or tripping, the inability of the dorsiflexors to generate even small moments at initial contact may cause the toe to contact the ground before the heel (if the ankle is in a plantarflexed position at initial contact), or cause the ankle to plantarflex uncontrollably, causing the foot to slap the ground (if the ankle is in a dorsiflexed or neutral position at initial contact).

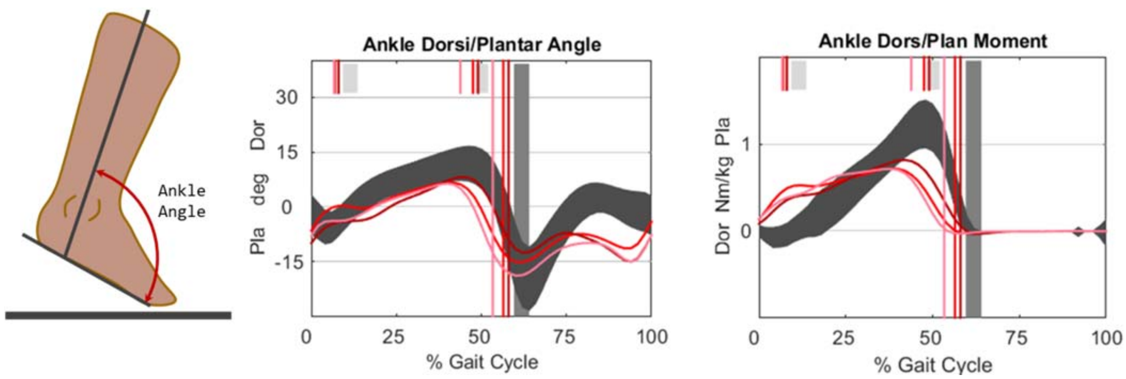


**Figure 1.4 – Foot drop ankle angle and moment during gait. Each shade of red indicates the angles, moments, and gait events for a single gait cycle. Long vertical lines indicate ipsilateral foot off events. Short vertical lines indicate contralateral foot off (1st series of lines) and foot contact (2nd series of lines).**

For an equinus gait pattern, the foot is in a plantarflexed position throughout the gait cycle (Figure 1.5). The plantarflexor muscles are active during all of stance which never allows the heel of the foot to make contact with the floor. Although the plantarflexor muscles are active, they are incapable of generating a significant ankle plantarflexor moment in late stance because the effective lever arm between the ankle and ground reaction force is reduced due to the ankle/foot position.

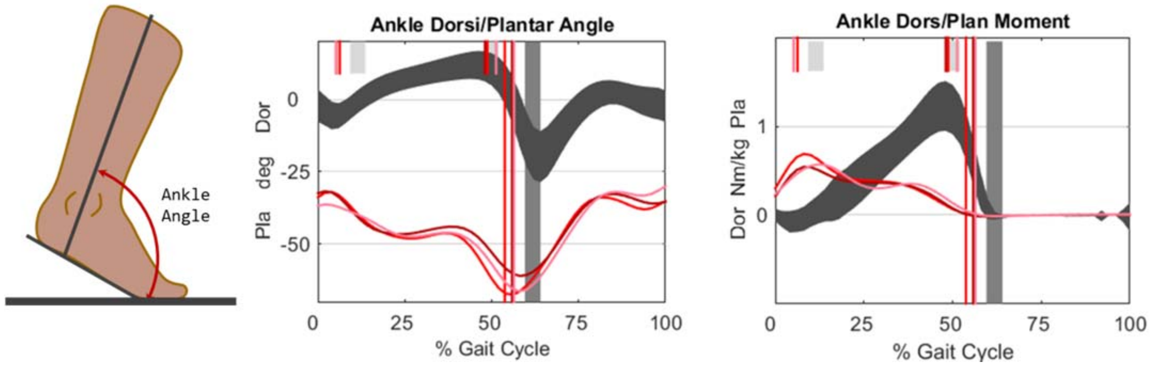


**Figure 1.5 – Equinus ankle angle and moment during gait. Dark shaded areas represent typical ankle function +/- 1 SD. Medium shaded areas represent the foot off event. Light shaded areas at the top of each graph represent the contralateral foot off (1st area) and foot contact (2nd area) events.**

These two examples demonstrate that both proper dorsiflexor and proper plantarflexor control is important for producing a typical gait. While the dorsiflexors typically absorb energy during foot contact and position the foot to clear the floor during swing, the plantarflexors provide the majority of supportive and propulsive energy during stance. From simulation work, the GAS and SOL have been shown to provide the majority of forward COM acceleration during gait as well as a substantial amount of the vertical acceleration required to maintain COM height [24], [30]. Additionally, proper plantarflexion function can also induce knee extension and offload knee extensor muscles (e.g. rectus femoris, vastus lateralis, vastus medius, and vastus intermedius). Active plantarflexion of the foot from the GAS and SOL against a stable foot-floor contact moves the ground reaction force vector to a position in front of the knee which induces knee extension. This, in turn, offloads large knee extensor muscles in favor of the smaller GAS and SOL. When plantarflexor function is impaired, as it is in CP, individuals must compensate for the lack of COM accelerations through a combination of biomechanical configuration and muscle activations/force changes.

## 1.6 Ankle Foot Orthoses

As previously mentioned, AFOs are often prescribed for children with CP as a means to improve gait. An AFO is a brace that encompasses and supports the foot, ankle, and shank. Primarily, an AFO is prescribed to improve both functional ankle-foot movement and foot deformities but is also used to improve balance, stance phase stability, and muscle force imbalances [31]–[34]. Both custom and standard AFO designs exists; common AFO designs are: a solid AFO (SAFO) – which is meant to completely prevent any ankle motion, a posterior leaf spring AFO (PLS) – which is meant to resist but allow some ankle motion, and a hinged AFO (HAFO) – which is meant to allow free motion between fixed stops. However, regardless of their overall design, the primary goal of an AFO is to resist undesirable ankle motion (and thereby promote more desirable ankle motion) [35]. For individuals with CP, the dorsiflexor and/or plantarflexor muscles are often impaired, resulting in undesirable ankle motion during gait. As an example, for individuals with foot drop, an AFO may be able to control excessive plantarflexion during swing when the foot needs to be dorsiflexed to clear the ground (Figure 1.6A). An AFO may also assist weak plantarflexor muscles for individual with a crouch gait by restricting uncontrolled dorsiflexion during stance when large dorsiflexor moments are being generated (Figure 1.6B). The goal of an AFO prescription in these cases is to restrict ankle motion so that the undesirable motion (e.g. uncontrolled dorsiflexion or uncontrolled plantarflexion) will be limited.

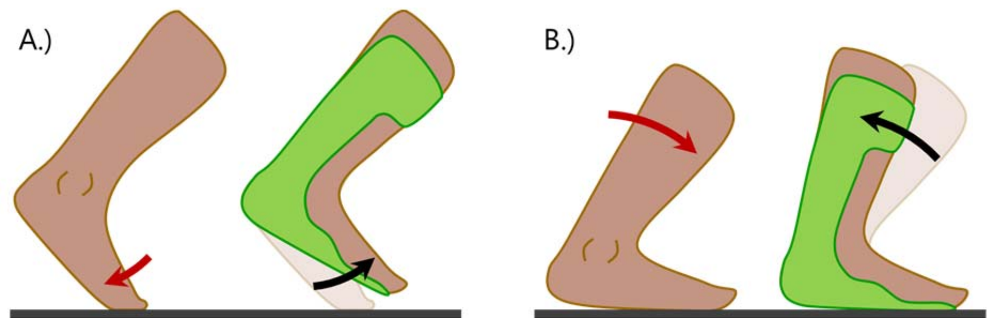


**Figure 1.6 – Influence of AFO on ankle motion. AFOs are primarily designed to control undesirable ankle motion. Two examples of how AFOs control undesirable ankle motion: A) Uncontrolled ankle plantarflexion during swing ("foot drop") is controlled by an AFO that prevents inappropriate amounts of plantarflexion during swing, and B) Uncontrolled ankle dorsiflexion during stance ("crouch") is controlled by an AFO that prevents ankle dorsiflexion during stance.**

AFOs are, by design, passive. As a result, AFOs cannot actively control when they restrict a specific motion and when they do not. If an AFO restricts ankle motions at one point in the gait cycle, it will restrict that same type of motion throughout the entire gait cycle. This is inconvenient. For the foot drop example, if the undesirable ankle plantarflexion during swing is controlled by an AFO, the AFO will also act during stance and prevent active plantarflexion during push off. This may not be a major problem if the individual has full plantarflexor strength, because the plantarflexors are relatively strong, and therefore capable of supplying a much higher ankle moment than the AFO is required to produce to support the foot in swing. But if the plantarflexors are weak, then the AFO may have a detrimental effect during other parts of the gait cycle.

AFOs have other shortcomings. To accommodate the donning of AFOs, individuals generally wear shoes much larger than they typically would. This may be cumbersome and awkward. In addition to the larger and heavier shoes, AFOs can also be relatively heavy, especially for smaller individuals where an AFO's weight can represent a substantial amount of the individual's total weight. Even if formed properly, AFOs may also rub against the skin causing callousing and skin breakdown. Additionally, there is also likely to be some social stigma surrounding the use of AFOs. Although cheaper than many other interventions, AFOs are still relatively expensive so the true physical and social cost of wearing an AFO needs to be carefully weighed against its potential benefits on a case by case basis.

## 1.7 Efficacy of Ankle Foot Orthoses

Many studies have investigated the effects of AFOs on the gait of individuals with CP. To summarize these studies, two literature reviews examined 37 different studies assessing the efficacy of various AFO designs have been performed [6], [7]. The reviews noted that a proper meta-analysis of the literature was difficult to perform due to the variety of study designs and inconsistency of outcome measures. Although a meta-analysis was not performed, the results appeared to be mostly mixed. Energy expenditure was shown to decrease with the use of AFOs in a handful studies [36]–[41] but was also shown to have minimal or no changes in others [36], [40], [42], [43]. Peak ankle power production was reported to decrease during the support phase of gait for many of the studies [44]–[47] but was also reported to increase in another [48]. Peak ankle power absorption during the first half of stance was reported to be reduced in two studies [45], [49] but was reported to increase in another [44]. Gait velocity was also mixed for AFO use where it was shown to increase in some studies [48]–[55] but did not change for others due to a slower cadence [36], [37], [45], [56]. More consistently reported improvements were in ankle position and step/stride length; ankle position at initial contact generally improved with AFO use as the ankle was set in a more appropriate neutral position [45], [46], [48], [49], [54], [56]–[59], and step/stride length also generally increased towards the typical range [36], [37], [48]–[56]. For functional skills, of three studies that included a functional measures (e.g. ability to walk long distances or navigate over curbs), significant improvements were only shown in one [37] and no improvements were noted for the other two [36], [39].

Because the efficacy of AFO is highly mixed and quite convoluted, each of the subsequent chapters in this thesis contain a detailed literature review that is focused on the points of the study presented in that chapter.

## 1.8 Primary Aims

This thesis aims to provide new methods for understanding and improving AFO outcomes as they pertain to gait.

## 1.9 Outline

The chapters of this thesis describe five individual studies related to assessing and improving the current efficacy of AFO use on gait.

In **Chapter 2**, data from a large cross sectional sample of children diagnosed with diplegic CP from a single center was used to **investigate the current efficacy of AFO use**. Changes in an individual's gait between walking barefoot and with their clinically prescribed AFO (SAFO, PLS, HAFO designs) were used to investigate the impact of an AFO on multiple gait related outcome measures.

In **Chapter 3**, an orthosis prescription algorithm was developed to **recommend the optimal orthosis design** that would maximally improve the gait for a child diagnosed with diplegic CP. The algorithm selected an orthosis design from pool of 5 common designs (3 AFO designs, 2 non-AFO orthoses designs), and included the option of that the patient not wear an orthoses if none of the designs were predicted to provide an advantage over barefoot walking. The potential level of benefit to the diplegic CP population was estimated under the assumption that the algorithm's recommendations are followed.

In **Chapter 4**, mechanical work and walking efficiency were analyzed for a large group of children diagnosed with diplegic CP in order to better **understand the source of the excessive metabolic demands** during walking, typically observed for these individuals. The study also investigated different strategies in work production and absorption that may help to explain why AFOs often reduce metabolic demand and lead to improved AFO prescription.

In **Chapter 5**, a musculoskeletal simulation is used to **investigate changes in muscle function** for individuals who have an observed qualitative improvement in gait when wearing their AFOs. The study uses induced acceleration analysis to determine changes in muscle function on center of mass accelerations caused by AFO use.

In **Chapter 6**, statistical analysis is used to **investigate differences in performance between two common AFO designs** that are thought of "clinically" as being distinct, but "mechanically" should have identical functions. The study uses stepwise linear regression to identify significant factors that contribute to the performance gap between solid and ground reaction AFO designs for the correction of crouch gait for individuals diagnosed with CP.

**Chapter 7** contains **general discussion** about the work contained in this thesis. Main findings, methodological considerations, and clinical implications are addressed.

# Chapter 2: The Efficacy of Ankle-Foot Orthoses on Improving the Gait of Children with Diplegic Cerebral Palsy: a Multiple Outcome Analysis

*PM&R. 7(9), 2015*

## 2.1 Introduction

The prescription of an ankle-foot orthosis (AFO) is common practice in the treatment of patients diagnosed with cerebral palsy (CP). AFOs are normally made of lightweight polypropylene or carbon fiber. They are typically fabricated as a one piece solid (SAFO) or posterior leaf spring (PLS) design, or as a two piece design with a hinged joint (HAFO). Typical objectives of AFO use are to improve overall function [60], enhance general gait quality and energy economy by improving ankle-foot function [61], correcting positional pathologies of the foot by maintaining correct ankle-foot alignment [61], and preventing contractures by slowly stretching spastic muscles [62].

While AFOs have been recommended for the aforementioned purposes, their use is not necessarily benign. With side effects including skin abrasion, social stigma, and a theoretical risk of disuse atrophy – especially for SAFOs, any benefits provided may be outweighed by deleterious effects. Additionally, annual costs for replacement AFOs during childhood and adolescent growth can be substantial. By knowing the overall effectiveness of AFO use for various treatment objectives, centers can establish realistic treatment goals for an AFO prescription. Estimates for the likelihood of achieving a specific treatment goal from an AFO can then be extrapolated from this information. This knowledge would help to maximize the efficacy of AFOs for the CP population (*e.g.* minimize over-prescription when detrimental side effects have the potential to exceed functional benefits).

Two literature reviews [7], [6] have summarized what is known about the effects of AFO use on gait for children with CP. The reviews identified 39 studies examining a total of 737 subjects. Study sizes ranged from 1 to 115 subjects, with a median size of 15. The studies were comprised of various sample populations, AFO designs, control conditions, and outcome measures. This heterogeneity among study designs makes meta-analysis impractical. As a consequence, main effects and interactions are unclear. As an example of the discordance among study designs, consider the three largest studies which contained a cross section of spastic CP subjects using a combination of SAFO, PLS or HAFO AFO designs. White [55] reported velocity, stride length, cadence, step length, and single limb stance time, Buckon [36] reported sagittal plane ankle and knee kinematics, velocity, stride length, step length, and cadence, and Desloovere [63] reported hip, knee, ankle kinematics and stride length. The only mutually reported outcome was stride length.

Another factor that makes a comparative analysis challenging is that many of the typical outcome measures currently used to define “general gait quality” are not consistently reported. Primarily, this is because most studies focus on evaluating a single specific outcome measure (*i.e.* knee flexion, step length, ankle ROM) rather than the overall effect from AFO use. Additionally, many of these studies predate the development of accepted general gait quality metrics such as the Gait Deviation Index (GDI) [64] or Gait Variable Score (GVS) [65].

This study focuses on assessing the effectiveness of AFOs for a large representative sample of children with diplegic CP seen at one center. The goal was to compile gait outcomes related to AFO use in a representative clinical setting by: (1) reporting the statistical parameters of general gait quality metrics and other commonly used outcome measures, and (2) studying the effect of AFO design and ambulatory status (*i.e.* independent or assistive device dependent) on these outcomes.

## 2.2 Methods

### *Study Group*

Retrospective data was compiled from a comprehensive search of the clinical database at one center. Inclusion criteria were:

- primary diagnosis of diplegic CP
- prescription of an SAFO, PLS, or HAFO
- same prescription bilaterally to minimize any compensatory actions due to inconsistent bracing conditions
- walking motion trials collected both barefoot (BF) and wearing an AFO during a single visit
- ambulated either with or without assistive device

Walking motion data was collected using an instrumented three dimensional motion capture system (Vicon, Oxford, UK). Multiple visits from an individual were allowed if the visits were more than 6 months apart. The time after a surgical or tone altering intervention was not explicitly controlled for. However, individuals at our center are typically undergoing a one-year postoperative assessment or routine follow-up which generally requires that between 9 and 18 months has passed between an intervention and data collection. Additionally, because the BF and AFO trials were collected during the same visit, each individual acted as their own control.

### *Outcome Measures*

Outcome data for each limb was compiled by averaging all trials for each walking condition (BF or AFO) during each visit; normally 3 trials per condition. The change score for each limb was then calculated by subtracting the BF from the AFO outcome values (ΔOutcome = AFO Outcome Value – BF Outcome Value). Outcome measures, described below, were the GDI, ankle GVS, knee GVS, nondimensional (ND) walking speed, and ND step length.

The GDI [64] is a single number that represents the deviation from a normal kinematic profile and is an accepted way of representing overall gait pathology. GDI scores over 100 represent normal kinematics and each decrement of 10 points represents one standard deviation from normal. A +4.3 GDI point change corresponds to an improvement in one level of the Gillette Functional Assessment Questionnaire (FAQ) score – a validated functional measurement tool [60], [64]. Conservatively, a +5 GDI point improvement is generally considered the minimal clinically important difference (MCID) for improvement after a surgery performed to improve function.

The GVS [65] is similar to the GDI. It is a single number that represents gait pattern deviation, but it is a *joint specific* measure. Ankle and knee joint adaptations to AFO use were identified by calculating sagittal plane ankle GVS, which is a measure of the direct influences of an AFO on the ankle, and sagittal plane knee GVS, which is a measure of the indirect influences of an AFO on the knee through the plantarflexion/knee extension couple. The two GVS measures along with GDI allowed for a more thorough evaluation of kinematic changes when wearing an AFO. Lower GVS values represent a more normal kinematic profile, which is opposite to the GDI where larger numbers are better. Therefore, improvements in GVS result in negative values for this analysis. Baker et al. [66] reported MCID in GVS for ambulatory children with CP corresponding to changes of a single FAQ level. This is analogous to the method used to determine the MCID for the GDI. The MCID for sagittal plane ankle GVS is 1.5° and the MCID for sagittal plane knee GVS is 3.4°.

Walking speed is often used as a surrogate measure of overall gait quality. By nondimensionalizing walking speed, subjects of varying height can be compared on a single scale. The ND speed was computed by dividing walking speed measured during the motion trial by $\sqrt{gL}$, where g is acceleration due to gravity and L is leg length [67]. Oeffinger et al. [68] reported the MCID for speed was 9.1% of normal. The ND speed for unimpaired controls, measured as 0.432 from Schwartz et al. [69], was used to calculate the MCID for ND speed of 0.039.

Step length is also commonly used to characterize gait quality. The ND step length was calculated by dividing step length by average leg length [67]. Oeffinger et al. [68]

identified stride length MCID as 5.8% of normal. Assuming that step length changes for the right and left limbs occur in equal proportions (*i.e.* symmetrical changes), the MCID for stride length can be applied to step length. Using step length for unimpaired controls, interpolated as 0.75 from Schwartz et al. [69], the MCID for ND step length was calculated to be 0.044.

Paired t-tests were used to determine statistical significance among changes in all six outcome measures between walking barefoot and with an AFO. The MCID values were used to establish clinical significance for changes in all outcomes. Additionally, standard ANOVA techniques were used to determine main effects and interactions affecting the amount of change in each outcome. Grouping variables for the ANOVA included two categorical groups – AFO design (Levels: SAFO, PLS, or HAFO) and ambulation type (Levels: independent or assistive device dependent), and a single continuous covariate – the BF value of the outcome measure being analyzed. Significance level was set at $\alpha = .05$. Calculations were performed using the statistics toolbox for Matlab 2012a (Mathworks, Natick, MA).

## 2.3 Results

The database search resulted in a data pool consisting of 601 visits derived from 378 individuals (215 male: 163 female). Mean age at time of visit was 9.8 years (SD 3.8) and the median time between multiple visits was 1.4 years. The 601 visits were grouped according to AFO and ambulation type (Table 2.1).

**Table 2.1 – Visits Grouped by AFO Design and Ambulation Type**

| | Ambulation Type | | |
|---|---|---|---|
| **AFO Design** | Independent | Assistive Device Dependent | Total |
| SAFO | 132 | 98 | 230 |
| PLS | 162 | 49 | 211 |
| HAFO | 103 | 57 | 160 |
| Total | 397 | 204 | 601 |

The FAQ levels for the visits are also shown and demonstrate the range of functional abilities within the sample (Table 2.2).

**Table 2.2 - Visits Grouped by FAQ Level**

| FAQ Level | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| 10 | 9 | 8 | 7 | 6 | 5 | 4 | 3 | 2 | NR | Total |
| 44 | 209 | 165 | 62 | 71 | 12 | 2 | 4 | 2 | 30 | 601 |

FAQ: 10 - keeps up with peers, 2 - does not routinely walk, NR - Not Recorded

Gross Motor Functional Classification Score (GMFCS) information was not available. A random limb for each visit was selected for outcome analysis.

### *General Outcome Changes*

Wearing an AFO provided statistically significant improvements compared to walking barefoot for GDI, ND speed, and ND step length (Table 2.3). However, only the improvements for ND speed and ND step length exceeded the established MCID levels.

**Table 2.3 – Overall Outcome Changes for AFO Use**

| | Mean BF Value | Mean Change in AFOs | p-value | MCID | % > MCID |
|---|---|---|---|---|---|
| **GDI** | 73.8 | 1.4 (6.6) | <.001* | 5 | 27% |
| **Ankle GVS** | 10.78° | -0.05° (-0.05) | .78 | -1.5° | 31% |
| **Knee GVS** | 5.94° | 0.14° (2.89) | .20 | -3.4° | 10% |
| **ND Speed** | 0.316 | 0.042 (0.059)† | <.001* | 0.039 | 48% |
| **ND Step Length** | 0.636 | 0.115 (0.103)† | <.001* | 0.044 | 76% |

Change is calculated as AFO value - BF Value; †Mean change surpassed MCID Value;*Statistical Significance at p <.05; MCID direction represents improvement

No systematic benefits from an AFO were identified for GDI, ankle GVS, or knee GVS (Figures 2.1A-C). With AFO intervention, average ND speed improved, surpassing the MCID of 0.039 (Figure 2.1D).

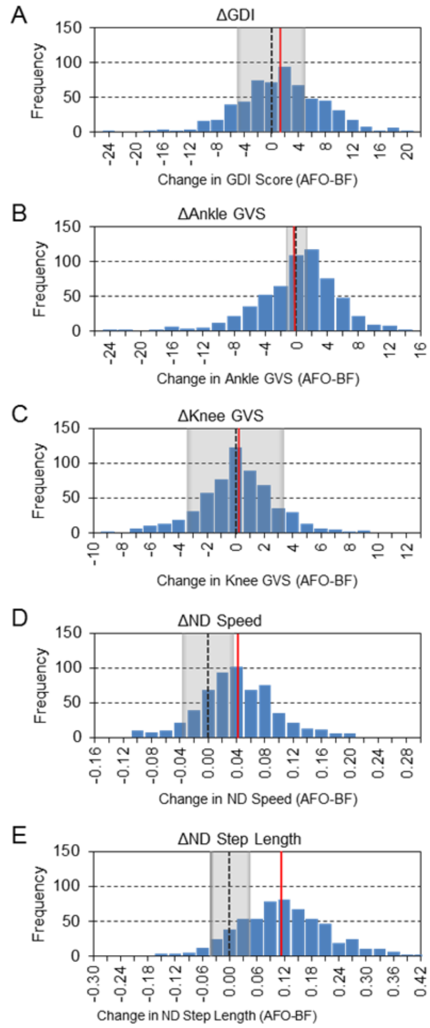


**Figure 2.1 – Changes in outcome between walking barefoot and with an AFO. The shaded areas represent change less than the MCID. The solid vertical lines represent mean change and the dashed lines represent zero change. A) Average GDI improved slightly (statistically but not clinically) – For the MCID of +5, 27% of limbs had clinically meaningful improvements, while 15% clinically worsened. B) Mean ankle GVS did not change with an AFO – For the MCID of 1.5°, 31% of limbs experienced meaningful improvements while 42% of limbs clinically worsened. C) Mean knee GVS change reflected minimal influence on knee function from an AFO – With an MCID of 3.4°, only 10% of limbs received a clinical benefit, while 11% of limbs experienced a meaningful worsening of knee function. D) Mean ND speed was clinically improved – For the MCID of 0.039, 48% of limbs clinically improved while only 6% worsened. E) Mean ND step length was clinically improved – For the MCID of 0.044, 76% of limbs clinically improved while only 5% worsened.**

The mean BF ND walking speed for the group was slow compared to typical ND walking speed for unimpaired controls, 0.315 compared to 0.363-0.500 [69]. Using an AFO increased mean ND speed by 14% to just under the normal range, 0.358. When analyzing only the slower subjects (BF speed < 0.363), almost all (88%) increased speed with an average speed increase of 34%. Similar to ND speed, average ND step length increased, surpassing the MCID of 0.044 (Figure 2.1E). Relative step length increased by 18%, and 88% of limbs experienced a step length increase. The correlation between change in ND speed and ND step length resulted in an $r^2$ value of 0.40.

***Main Effects***

The ANOVA identified the *barefoot value* for each outcome measure (*i.e.* BF GDI, BF ankle GVS, etc.) as the most significant main effect on the change in outcome (Table 2.4). For all outcomes, the worse the barefoot value, the larger the improvement tended to be.

**Table 2.4 – Main Effects and Interactions Between AFO Design, Ambulation Type, and BF Value**

| | ANOVA Main Effects (p-value) | | | ANOVA Interactions (p-value) | | |
|---|---|---|---|---|---|---|
| | BF Value | AFO Design | Ambulation Type | AFO Design & BF Value | Ambulation Type & BF Value | AFO Design & Ambulation Type |
| **GDI** | <.001* | .16 | <.001* | .19 | <.001* | .74 |
| **Ankle GVS** | <.001* | <.05* | .25 | .11 | .92 | .08 |
| **Knee GVS** | <.001* | .41 | .40 | .06 | .82 | .30 |
| **ND Speed** | <.001* | <.05* | .30 | <.05* | .34 | <.01* |
| **ND Step Length** | <.001* | .47 | .42 | .32 | .21 | <.01* |

Change is calculated as AFO value - BF Value; †Mean change surpassed MCID Value;*Statistical Significance at p <.05; MCID direction represents improvement

The analysis identified a significant main effect of *AFO design* on changes in Ankle GVS and ND speed (Table 2.4, Figure 2.2A-B). For ankle GVS, Tukey's HSD test (95% CI) identified significant differences between the mean changes for the SAFO group and PLS group (Figure 2.2A). However, neither group surpassed the MCID; ankle function for the SAFO group generally improved, -0.93, while the PLS group ankle function was hindered, +0.93. For ND speed, Tukey's HSD test identified significant differences in the mean change between the SAFO group and PLS/HAFO groups (Figure 2.2B). The SAFO group improved speed the most, +0.052, compared to the other two groups, +0.037 and +0.037.

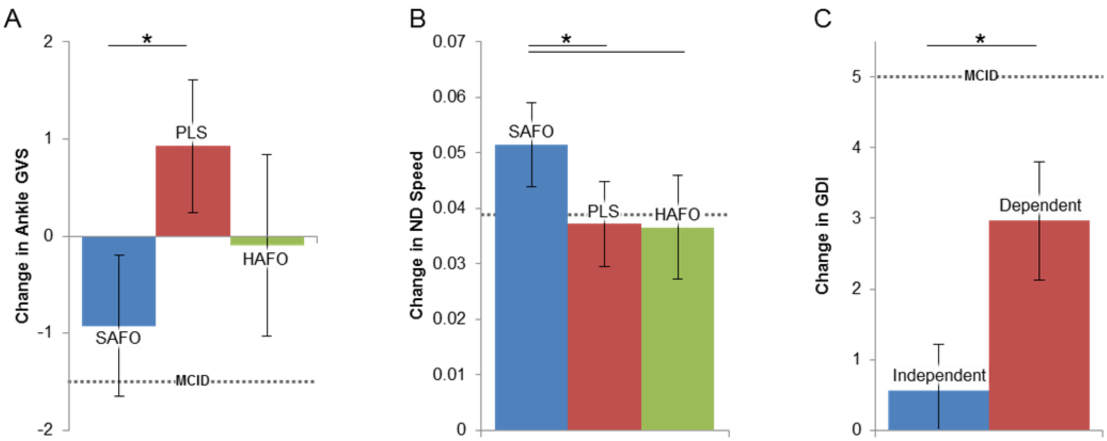


**Figure 2.2 – Significant main effects for AFO Design and ambulation type on outcomes. A) Effect of AFO design on Ankle GVS. B) Effect of AFO design on ND Speed. C) Effect of Ambulation type on GDI. The asterisk and line above the plots identify significant differences between groups as determined by Tukey's HSD test (α = .05). Error bars represent the 95% CI of the actual mean change in outcome for each group and dashed lines represent the MCID levels for improvement (the MCID for GDI is off of the plot at +5).**

The analysis identified significant effects of *ambulation type* on GDI (Table 2.4, Figure 2.2C). Assistive device dependent ambulators had the largest response with an increase of +3.0 GDI points where 37% of limbs experienced clinically meaningful improvements. For the independent ambulators, the average GDI increase was only +0.6 points where 22% experienced a meaningful level of improvement. Neither group average surpassed the MCID despite having statistically different outcomes.

### *Interactions*

In addition to the main effects, the ANOVA identified significant interactions for 3 of the 5 outcomes (Table 2.4). For the general gait quality metrics (*i.e.* GDI, ankle GVS, knee GVS), there was only significant interaction between ambulation type and BF GDI

(Figure 2.3A); no interactions were identified for Ankle GVS or knee GVS (Figure 2.3B). A comparison of the coefficients of determination between change in GDI, change in ankle GVS, and change in knee GVS were all low ($r^2 < 0.13$).

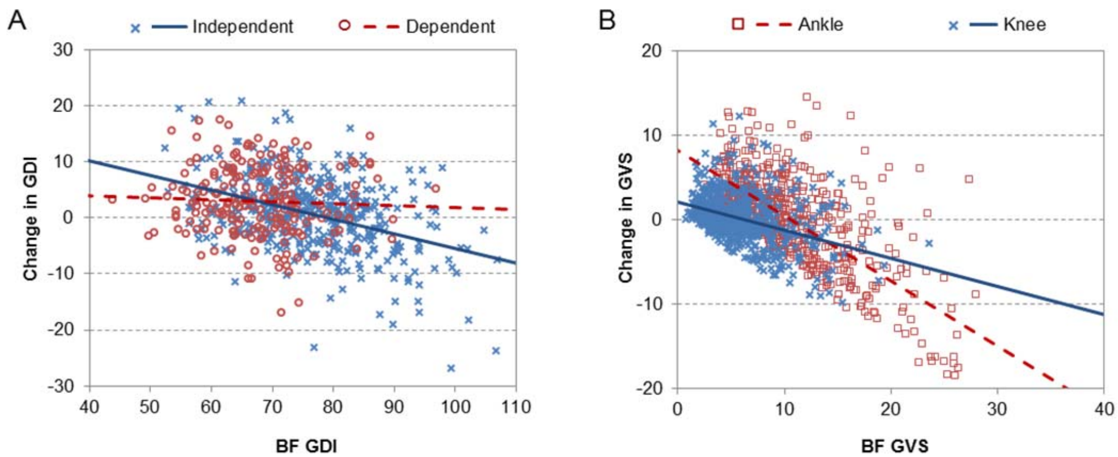


**Figure 2.3 – Significant interactions on general gait quality metrics. A) There was a significant interaction between BF GDI and ambulation type. Independent ambulators had the most sensitivity to BF GDI whereas there was little effect for dependent ambulators. B) The main effects of ankle GVS and knee GVS are shown (there were no significant interactions for either). There was a strong main effect of BF ankle GVS and change in ankle GVS ($r^2$ = .45). The main effect of BF knee GVS and change in knee GVS was low ($r^2$ = .15).**

For the spatio-temporal metrics (*i.e.* ND speed, ND step length), a significant interaction between AFO design and BF ND speed was identified (Figure 2.4A). An interaction between AFO design and ambulation type was also indicated for ND speed. For ND step length, an interaction between AFO design and ambulation type was identified; there were no interactions related to the main effect of BF ND step length (Figure 2.4B).

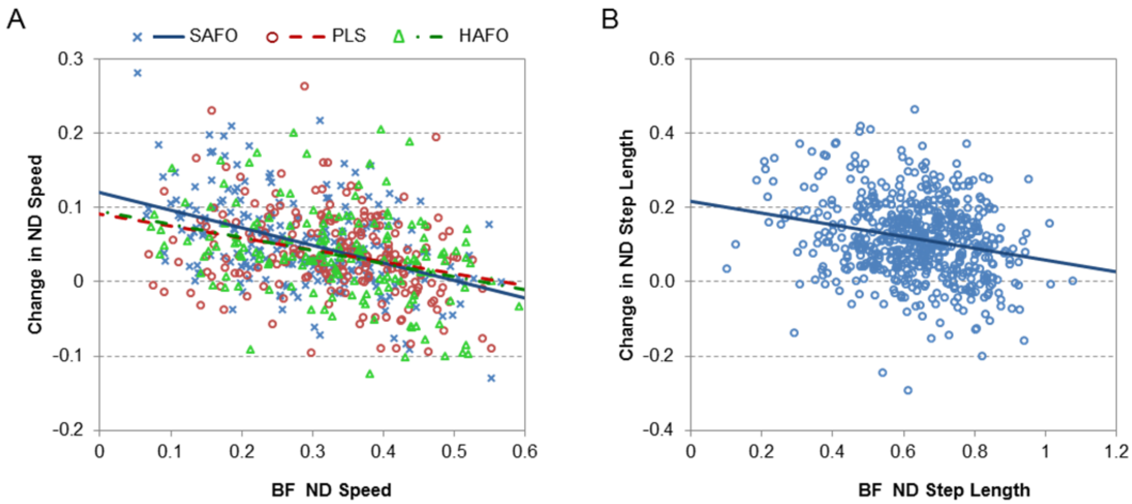


**Figure 2.4 – Significant interactions on spatio-temporal metrics. A) The main effect for BF ND speed on change in ND speed is apparent. There was a significant interaction between AFO design and BF ND speed. The SAFO was slightly more effective at improving walking speed when speed was slower than unimpaired controls (< 0.363). However, the SAFO group had greater negative speed changes for normal or fast walkers due to the larger interaction. B) The main effect of BF ND step length on change in ND step length is shown (there were no significant interactions for ND step length).**

Overall, the analysis showed that speed and step length were the only consistently improved outcomes from AFO use. The most significant influence to the amount of benefit provided by an AFO was determined to be the barefoot outcome value (*i.e.* ceiling effect). Using only the barefoot value main effects, thresholds were identified as when to begin expecting positive results from wearing an AFO: BF GDI below 70, BF ankle GVS greater than 10°, BF knee GVS greater than 6°, BF ND speed lower than 0.50, and BF ND step length less than 1.2. Additionally, *ambulatory status* had a significant effect on GDI while *AFO design* had a significant effect on ankle GVS and ND speed. In addition to the main effects, the analysis also identified the interactions of ambulation type and BF GDI on change in GDI and AFO design and BF ND speed on change in ND speed. These interactions provide additional information for the expected influence of AFOs on gait. By using the statistical parameters for each outcome (Table 2.3), the probability of achieving a desired outcome level can be estimated.

## 2.4 Discussion

This study was conducted to assess the performance of AFOs for children with diplegic CP. The primary finding from this analysis is that, other than step length, AFOs do not appear to provide clear and consistent improvements in the gait of children with diplegic CP. Speed and step length are clearly important, however, there is an implicit assumption that other outcomes, such as ankle motion, change in response to an AFO. Data from this study shows that clinically meaningful benefits for other outcomes are achievable, but only for a small percentage of AFO users.

Multiple studies in the literature reported step length increases with AFO use [36], [37], [55], [63], [48], [52], [53], [70], while about half as many reported speed increases [36], [55], [52], [53]. These reports are consistent with the results from our analysis. The average levels of change for the remaining outcome measures – GDI, ankle GVS, and knee GVS – were not reported in previous studies as they relate to AFO use.

By definition, AFOs are meant to directly impact ankle motion, but they are also thought to indirectly influence other joints [71]. First, when analyzing ankle function alone, the AFO design most restrictive to movement, the SAFO, tended to normalize ankle function the most. This indicates that for many individuals with diplegic CP, ankle kinematics may be better normalized by simply restricting the ankle to a single neutral angle than by allowing it to move when using a more compliant AFO design. Secondly, the indirect effect caused by an AFO on the knee was low. The coefficients of determination between changes in ankle GVS, knee GVS, and GDI were all less than 0.13. In other words, changes in one outcome do not appear to systematically influence the other outcomes. This indicates that although the knee, hip, and pelvis may be indirectly affected by AFOs, such as through the plantarflexion/knee extension couple, the joint adaptations to AFO use appear to be independent and not predictable. We conclude from this low correlation that a standard adaptation strategy does not exist for AFO users in general, and that functional adaptations to AFO use are unique to subgroups or individuals.

This study found that ambulators who used assistive devices essentially had a ‘fixed’ level of response and did not experience the same ceiling effect for changes in GDI as independent ambulators. The study also found that speed generally increased when using an AFO while GDI did not. If the speed with which one moves from place is a primary goal, then walking speed makes sense as a primary outcome measure. It may be tempting to use speed and step length improvements as proxies for reductions in the energy cost of walking or other outcome measures not assessed by this study. For energy costs, Brehm et al. [40] investigated the interaction between speed and energy costs for children with CP who use AFOs, and showed that for all of the participants that sped up while using an AFO, approximately 1/4 experienced disproportionate increases in energy costs. In other words, the energy cost of walking for some individuals increased more than was explained by their increase in speed. For other measures, Sullivan et al. [72] identified correlations between speed and step length with other common functional outcome measures for children with CP. Speed had a strong correlation with Gross Motor Functional (GMFM) level ($0.6 \le r < 0.8$), but all other outcomes had moderate ($0.4 \le r < 0.6$) to weak correlations ($r < 0.4$) with speed and step length. The results presented in these studies should caution those that would consider speed and step length as strong proxies for energy cost or other functional outcome measures.

There are several limitations to this study which are primarily attributable to its retrospective nature. It is important to restate that the children were typically undergoing a one-year postoperative assessment or routine follow-up. The majority of studies identified in the literature reviews [6], [7] use inclusion criteria for the individual to be at least 1 year postoperative from major surgery. Therefore, using data collected during the one year postoperative or follow-up assessments at our center is consistent with previous study designs. Additionally, we felt that the inclusion of multiple visits for an individual better represented the typical outcomes from AFO prescriptions rather than analyzing only a “snapshot” from a single point in their care path. The children analyzed in this study were also wearing AFOs that were “standard-of-care”; meaning that there were heterogeneous prescription guidelines and goals as recommended by the patient’s physician. Unfortunately, AFO prescription goals (*e.g.* prevent drop foot, promote normal ankle rocker mechanics, etc.) were not available. It is noteworthy that AFO prescription goals were listed for only 15% of the individuals identified in the literature reviews [6], [7]. The lack of clinical context for why an AFO was prescribed in our analysis is consistent with the majority other retrospective AFO studies. Finally, the use of the barefoot condition instead of a “shoes only” control condition may lead to concerns. As this was a retrospective analysis, it was limited to the standard data collection methodology at our center. In addition, the use of barefoot as the control condition is widespread throughout the literature; 24 of the 39 studies (which included nearly 75% of the participants) in the literature reviews [6], [7] used barefoot walking as the control condition. Therefore, we conclude that the weaknesses in this study related to the retrospective nature are wholly consistent with previous retrospective analyses of this type.

Ultimately, the goal from any intervention must be determined by weighing the desires of the child, family, and practitioner. Depending on the goal, the likelihood of receiving a benefit from an AFO may be inferred from past results. This study showed that step length is highly likely to be improved with an AFO (76%) and that some individuals are also likely to experience clinical benefit for speed (48%). Clinical benefit is less likely for the other general gait quality measures (27% for GDI, 31% for ankle GVS, and 10% for knee GVS). Therefore, one must be sensible when forming expectations of how likely an AFO will be at achieving the desired goal.

This study provides substantial insight into changes in gait with AFO use. It is not meant to be a guide for prescription, nor does it attempt to explain the biomechanical basis for good or poor performance. The nature of the responses to AFO use emphasizes the need to utilize knowledge gained from past performance to critically evaluate how effective AFOs are at achieving their intended goal on a case-by-case basis. Furthermore, if general gait quality improvements are desired, these results should highlight the need for an improved approach to selecting or fabricating an AFO. For example, there has been much interest in AFO ‘tuning’, where the characteristics of the AFO are customized for an individual. This approach looks promising; however, current tuning methods appear to lack the evidence necessary to understand the effects of each adjustment made during the tuning process. Instead, current methods appear to be based primarily on heuristic or anecdotal evidence. Further work is needed to fully understand the potential of AFO tuning to improve gait outcomes.

# Chapter 3: A Data Driven Model for Optimal Orthosis Selection in Children with Cerebral Palsy

*Gait & Posture, 40(4), 2014*

## 3.1 Introduction

Orthoses are commonly prescribed for individuals with cerebral palsy (CP). Typical goals of the prescription include enhancing gait quality and energy economy [37], correcting positional pathologies of the foot [61], and preventing contractures by stretching of spastic muscles [62]. Orthoses influence the ankle and foot by providing a control moment opposing ankle motion, and also stabilize the motions of the mid- and forefoot joints. Common orthosis designs for children with CP are the ankle-foot orthosis (AFO) and foot orthosis (FO). Common AFO designs are the solid (SAFO), posterior leaf spring (PLS), hinged (HAFO), and supra-malleolar orthosis (SMO), while a typical FO is the University of California, Biomechanics Laboratory design (UCBL). There is limited guidance to aid in the selection of an optimal orthosis for an individual patient when the goal is improving overall gait quality.

While various studies have analyzed the effect of orthoses on gait quality [6], none have been shown to reliably predict improvements. Buckon compared the SAFO, PLS, and HAFO designs in spastic CP [37]. The study concluded that AFOs improved many of the outcomes analyzed, but that no single AFO design was optimal for every individual. Buckon's results emphasize the need for customized orthosis prescriptions. However, without an estimate of how an orthosis will affect gait, a patient would need to be tested in all AFO designs. This is not practical for the clinical setting.

Rodda and Graham [73] presented a biomechanically based orthotic management algorithm which used gait and postural patterns to determine the appropriate orthosis design. However, the proposed system's clinical benefit has never been presented. In addition, the algorithm is ambiguous, recommending multiple AFO designs for a single gait pattern.

Utilizing statistical machine learning techniques to construct a model from a large pool of retrospective data may be a more effective way to develop a clinically useful orthosis prescription tool.

The Random Forest Algorithm (RFA) is a statistical classification method that has been applied to a variety of fields, from gene expression [74] to terrain classification [75]. More recently, it has been harnessed to predict likelihood of good outcomes from orthopaedic surgeries [76], [77].

The RFA works by utilizing a large group of independent classification and regression trees (CARTs) built from measured features (e.g. walking speed) to predict responses (e.g. change in gait) for a set of observations (e.g. limbs) [78]. Each CART is built using a random sample of observations and features. This randomness renders the CARTs independent of one another. The CARTs are collected to form an ensemble or 'forest'. The response predicted by the ensemble is based on vote aggregation. Each individual CART has only a small influence on the overall prediction. If more CARTS vote for outcome A over outcome B, then the ensemble predicts outcome A over B. Using RFA methodology versus a single CART is beneficial because it generally gives more accurate and robust predictions [78].

A common measure used to quantify overall gait quality is the Gait Deviation Index (GDI) [64]. The GDI is a single number that represents the difference between the gait of an individual and that of a typically developing control group. A GDI over 100 reflects normal kinematics, and each decrement of 10 GDI points represents one standard deviation from normal. An improvement in GDI of +5 points is considered clinically meaningful for surgical interventions that are performed to improve function [64]. In this study, we chose the change in GDI between walking with an orthosis and walking barefoot as the outcome measure ($\Delta GDI = GDI_{Orthosis} - GDI_{Barefoot}$).

The goals of the study were to: (1) use the RFA to build a model that can predict changes in GDI for individuals in various orthosis designs, without having to fabricate and test each design on the individual, and (2) estimate the potential clinical benefit of the model.

## 3.2 Methods

The study consisted of two steps; Step 1: Build a model based on retrospective data from limbs with an existing orthosis prescription, and Step 2: Evaluate the potential benefit of the model by applying it to a representative sample of patients.

### *Step 1: Model Development*

Data from a large sample of individuals walking barefoot and with one of the five orthosis designs was needed to build the predictive model using the RFA. A search of our clinical database was conducted to identify this '*modeling sample'*. Each individual's data was then then separated into one of five groups, depending on which orthosis design was prescribed. We then used the RFA to build and test five independent predictive ensembles corresponding to the five orthosis designs. The ensembles were then combined to form a single predictive model (Figure 3.1A). The model simultaneously predicts the change in GDI for all five orthosis designs based on an individual's barefoot walking data. The model makes a recommendation for a specific orthosis design (or predicts no orthosis will provide benefit) based on the highest predicted response for the limb (Figure 3.1B).

A good outcome was defined as $\Delta$GDI ≥ +3 points. We decided that the low cost, relatively low invasiveness and ease of intervention of an orthosis prescription warranted a slightly lower threshold for benefit than the $\Delta$GDI ≥ +5 generally required of a surgical intervention. It should be noted that the methodology in this study would be unchanged if a higher (or lower) threshold was chosen.

### *Step 2: Benefit Estimation*

A representative sample of the diplegic CP population was then needed to estimate the clinical benefit of the model. A second search of the clinical database was conducted to identify this '*benefit sample'*. Individuals were included in the benefit sample regardless of whether or not they had an existing orthosis prescription, since only barefoot walking data is required by the model to make predictions. All limbs from the benefit sample were then processed by the model (Figure 3.1C). The benefit was calculated by comparing the average ∆GDI from existing orthosis prescriptions to the average predicted ∆GDI from the orthosis designs recommended by the model.

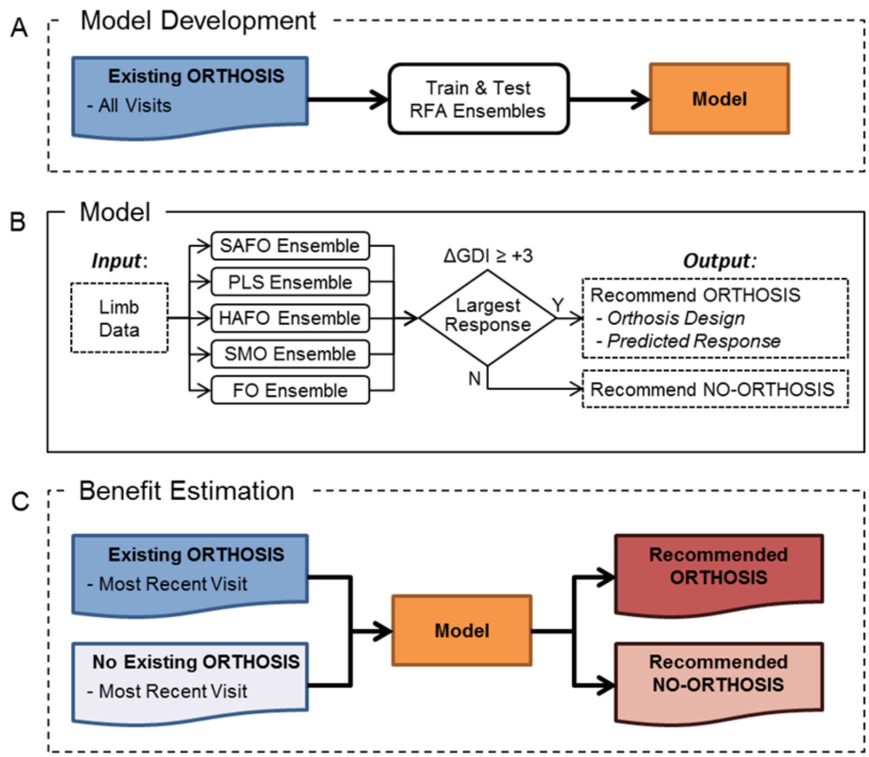


**Figure 3.1 – Optimal orthosis selection model development and benefit estimation. A) All visits from individuals with an existing orthosis prescription of an SAFO, PLS, HAFO, SMO, or FO were used to train five RFA ensembles and develop the orthosis prescription model ('Modeling Sample').B) The prescription model utilized the five RFA ensembles to recommend the optimal orthosis design. First, data for an individual limb was provided to the model. The ensembles simultaneously process the data and predict the ΔGDI response for each orthosis design. The responses were compared and the design with the largest predicted response greater than or equal to + 3 ΔGDI points was recommended by the model. If no design was predicted to surpass the +3 ΔGDI threshold, a "No Orthosis" recommendation was made instead. C) To estimate the benefit of the model to the diplegic CP population, a representative sample was used ('Benefit Sample'). The most recent visit for individuals with diplegic CP seen at the center was used as the benefit sample. All limbs in the benefit sample were processed by the model and orthosis recommendations were made. Benefit was then assessed by comparing the average ΔGDI for limbs with their existing orthosis prescription to the average predicted ΔGDI for limbs with the orthosis design recommended by the model. *Due to the overlapping inclusion criteria, some of the data used to develop the model (Existing ORTHOSIS group in Model Development) was also used to estimate benefit (Existing ORTHOSIS group in Benefit Estimation).**

### *Model Development*

Modeling data for constructing RFA ensembles was compiled from the clinical database at our center. Inclusion criteria were:

- diagnosis of diplegic CP
- walking motion trials collected barefoot
- walking motion trials collected wearing an orthosis during same visit as the barefoot trials
- prescription of an SAFO, PLS, HAFO, SMO, or FO
- same orthosis design worn bilaterally

Multiple visits from individuals were allowed since highly correlated observations do not adversely affect the performance of the RFA [78].

Five RFA ensembles were constructed; one for each of the five orthosis designs (SAFO, PLS, HAFO, SMO, or FO). Each ensemble predicts the ΔGDI for a limb wearing the specified orthosis compared to walking barefoot. For example, the HAFO model predicts the change from barefoot gait to gait wearing an HAFO, while the SAFO model predicts the change from barefoot gait to gait wearing an SAFO, etc. The ensembles were provided with features drawn from medical history, physical exam measures, and kinematic data derived from a three-dimensional gait analysis. Features were objectively ranked in order of their importance by the RFA [78], [79]. Model reduction was carried out by systematically reducing the number of features available to each ensemble until removing additional features significantly decreased performance accuracy. Performance metrics were calculated using estimates derived from samples not randomly selected for the construction of a CART (unbiased out-of-bag estimates). As only about 63% of the modeling sample was directly used for the construction of a CART, the remaining out-of-bag samples were used to calculate performance. Using these out-of-bag estimates eliminates the need for a separate test set [78], [80]. Each ensemble's performance was assessed using standard diagnostic metrics based on (1) outcome classification (good/poor based on the +3 ΔGDI threshold), and (2) predicted GDI change. Classification metrics consisted of accuracy, sensitivity, specificity, positive predictive value (PPV), negative predictive value (NPV), and Matthews correlation coefficient (MCC). Metrics for regression analysis were coefficient of determination (R2) and root mean squared error (RMSE). Modeling was performed using Matlab with the Statistics Toolbox (2012a).

A single orthosis prescription model was then constructed by combining the five RFA ensembles in parallel (Figure 3.1B). Data from a limb was provided to the model, evaluated by all five ensembles, and the ΔGDI response for each design was made. The orthosis design corresponding to the largest predicted response ≥ +3 GDI points was recommended by the model. If no orthosis design met the +3 ΔGDI threshold, a '*No Orthosis*' recommendation was made.

***Benefit Estimation***

A representative sample of the diplegic CP population was needed to estimate the benefit of the model. A second search of the clinical database was conducted. Inclusion criteria for the benefit sample were:

- diagnosis of diplegic CP
- walking motion trials collected in the barefoot condition
- Note: the benefit sample also includes individuals with no existing orthosis prescription

For individuals with multiple visits, only the most recent visit was used. Due to the overlapping inclusion criteria for the modelling sample and the benefit sample, the most recent visit from the individuals used to develop the model (Existing ORTHOSIS in Model Development – Figure 3.1A) was also used to estimate benefit (Existing ORTHOSIS in Benefit Estimation – Figure 3.1C).

Each limb in the benefit sample was then evaluated by the orthosis prediction model. Benefit was assessed by comparing the average ΔGDI for limbs in their existing orthosis prescription with the average ΔGDI for limbs in the orthosis design recommended by the model.

## 3.3 Results

### *Model Sample*

The database search returned 476 individuals meeting the inclusion criteria. Of these 476 individuals, 227 had multiple visits. This resulted in 857 individual/visit combinations, for a total of 1714 limbs.

### *Model Development*

For each orthosis design, an RFA ensemble of 100 CARTs was created using 301 potential predictor features. Following model reduction, the finalized ensembles utilized between 4 and 9 predictor features (Table 3.1). All but one of the predictor features were derived from kinematic walking data (ND speed depends on leg length).

**Table 3.1 – Model Performance Characteristics and Features Used for Each RFA Ensemble. Features are listed in order of importance according to the RFA.**

| | SAFO | PLS | HAFO | SMO | FO |
|---|---|---|---|---|---|
| **Accuracy** | 0.67 | 0.73 | 0.71 | 0.81 | 0.82 |
| **Sensitivity** | 0.60 | 0.50 | 0.52 | 0.22 | 0.40 |
| **Specificity** | 0.72 | 0.85 | 0.83 | 0.94 | 0.94 |
| **PPV** | 0.61 | 0.62 | 0.66 | 0.45 | 0.66 |
| **NPV** | 0.71 | 0.77 | 0.73 | 0.85 | 0.85 |
| **MCC** | 0.32 | 0.37 | 0.38 | 0.22 | 0.41 |
| **$R^2$** | 0.20 | 0.19 | 0.23 | 0.24 | 0.28 |
| **RMSE** | 6.2 | 5.7 | 6.0 | 5.1 | 5.2 |
| **# of Features** | 9 | 9 | 4 | 6 | 4 |
| **Features** | BF GDI | BF GDI | Mean Sta. KF | BF GDI | Mean FP |
| | Ankle GVS | Mean Sta. FP | Mean Ankle Dor. | Mean FP | BF GDI |
| | Min Sta. FP | Max Sta. FP | I.C. Foot Prg. | Min FP. | Max Sta. FP |
| | Speed | ROM Foot Prg. | Max Sta. FP | ROM Swi. FP | Mean Swi. FP |
| | ND Speed | F.O. KF | | Mean Swi. FP | |
| | Mean FP | Mean Sta. Ankle Dor. | | F.O. FP | |
| | Mean Sta. FP | t_Max Sta. Hip Rot. | | | |
| | ROM Hip Add. | Min Sta. FP | | | |
| | Foot Off | Mean Ankle Dor. | | | |

**Variable abbreviations and codes are: BF - barefoot, GDI - gait deviation index, GVS - gait variable score [65], ND - Nondimensionalized, Sta. - stance, Swi. - swing, I.C. - initial contact, F.O. - foot off, ROM - range of motion, t_ - time of, FP - foot progression, KF - knee flex, Add. - adduction, Rot. - rotation, Dor. - dorsiflexion**

In terms of outcome classification, the ensembles performed well, with accuracy ranging from 67 - 82%. The SMO and FO ensembles had the highest accuracy, and were particularly good at ruling out limbs unlikely to benefit from their use (specificity = 0.94). However, these ensembles were less useful at identifying limbs likely to benefit from their use (sensitivity SMO = 0.22 and FO = 0.40). Where large imbalances between numbers of good and poor outcomes exist, MCC values, which represent the quality of prediction independent of class balance, are preferred. Values for MCC were relatively high in all but the SMO model.

In terms of predicted GDI changes, RMSE values ranged from 5.1 GDI points for the SMO to 6.2 for the SAFO ensemble. The $r^2$ values indicated between 19% and 28% of the variance was explained by the ensembles.

Of the features used to predict outcomes, barefoot GDI is a factor in all but the HAFO ensemble, and foot progression features appeared consistently in all RFA ensembles. Greater improvements in GDI tended to be predicted for limbs with lower barefoot GDIs. The SAFO, PLS, and HAFO designs generally provided a level of benefit proportionate to the amount of deviation from normal progression for both internal and external foot progression angles. The SMO and FO designs tended to provide benefit for external foot progression only.

***Benefit Estimation***

The search of the database for the benefit sample identified 1016 individuals, age: 12.6 (6.7) years. At the time of their visit, 476 had an existing orthosis prescription (ORTHOSIS group) and 540 did not have an existing orthosis prescription (NO-ORTHOSIS group). This resulted in 2032 limbs being analyzed by the model. Limbs wearing a currently prescribed orthosis had an average gait improvement of +0.4 GDI points, while the average GDI improvement for all limbs, including those without an orthosis, was +0.2 points. Application of the model was estimated to render an overall improvement of +2.1 GDI points (+0.2 → +2.3) for the average individual with diplegic CP (Table 3.2).

**Table 3.2 – Comparison Between the Existing and Model Recommended Condition for All Limbs**

| | | Recommended Condition | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|
| | | SAFO | PLS | HAFO | SMO | FO | NO-ORTHOSIS | Total | |
| Existing Condition | **SAFO** | 73 [+5.4] | 34 [+5.8] | 26 [+6.9] | 1 [+5.0] | 4 [+5.5] | 116 [0.0] | **254** {+1.9} | |
| | **PLS** | 26 [+5.1] | 23 [+5.5] | 18 [+6.3] | 0 [0.0] | 14 [+4.1] | 157 [5.3] | **238** {+0.7} | |
| | **HAFO** | 22 [+4.8] | 16 [+5.3] | 27 [+7.1] | 0 [0.0] | 4 [+3.9] | 91 [0.0] | **160** {+0.3} | |
| | **SMO** | 12 [+5.9] | 8 [+6.2] | 9 [+5.9] | 1 [+4.5] | 0 [0.0] | 79 [0.0] | **116** {-1.2} | |
| | **FO** | 14 [+4.3] | 12 [+5.0] | 8 [+5.7] | 0 [0.0] | 14 [+5.2] | 136 [0.0] | **184** {-1.1} | |
| | **NO-ORTHOSIS** | 159 [+4.9] | 186 [+6.0] | 88 [+6.3] | 2 [+3.8] | 34 [+5.0] | 611 [0.0] | **1080** {0.0} | |
| | **Total** | 306 [+5.0] | 279 [+5.9] | 176 [+6.5] | 4 [+4.3] | 77 [+4.8] | 1190 [0.0] | **2032** {+0.2} | [+2.3] |

[ ] = predicted ΔGDI in recommended condition; { } = actual ΔGDI in existing condition

Five groups were identified based on the existing orthosis condition and the model-recommended orthosis (*Existing Condition* → *Recommended Condition*) (Table 3.2, Figure 2.2). By analyzing each of these groups individually, the contributions to the +2.1 overall improvement in ΔGDI was determined (Figure 2.3A). Of the 2032 limbs in the dataset, 37% were members of the NO-ORTHOSIS → NO-ORTHOSIS or ORTHOSIS → SAME-ORTHOSIS groups, and as such, were recommended to remain in their existing prescription. These two groups represent the portion of limbs where the existing clinical prescription was determined to be optimal. The model provides no added benefit for these limbs since there is no change in condition. However, for the remaining 63% of limbs, significant improvements were predicted. There were 469 limbs in the NO-

ORTHOSIS → ORTHOSIS group. These limbs were predicted to have an average ΔGDI of +5.6 points, improving from a ΔGDI of 0 to +5.6. There were 235 limbs that had an existing orthosis but were recommended a different design, improving their ΔGDI by +2.7 points from +2.8 → +5.5. Finally, 579 limbs were members of the ORTHOSIS → NO-ORTHOSIS group and improved their ΔGDI by +1.5 points, from -1.5 → 0, by not wearing an orthosis. The gait of these limbs worsened in their existing orthosis prescription, but the model was unable to identify any design that would result in ΔGDI greater than +3 points.

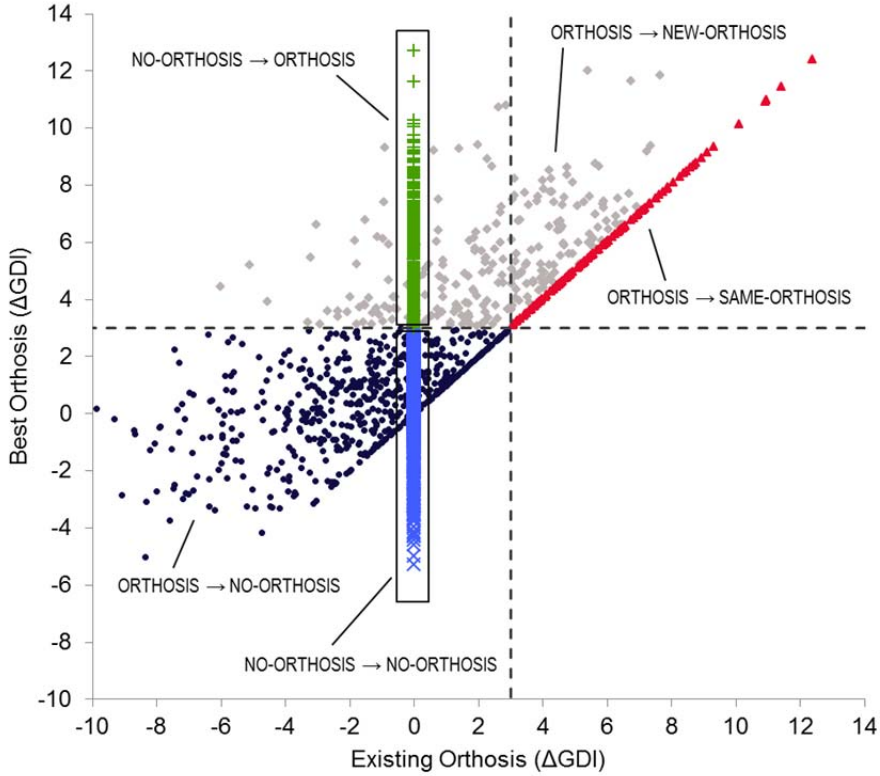


**Figure 3.2 – Model identified prescription groups based on the existing and recommended conditions (Existing Condition → Recommended Condition). The horizontal axis represents the predicted outcome level for the existing orthosis prescription. The vertical axis represents the predicted outcome level for the orthosis design with the highest predicted response. The dashed vertical and horizontal lines identify the respective +3 GDI point "good response" threshold. The triangles along the identity line (x=y) in the upper right quadrant are limbs for which the existing prescription is also the best orthosis prescription (ORTHOSIS → SAME-ORTHOSIS). The diamonds scattered on the upper two quadrants are limbs with an existing orthosis prescription for which a different orthosis design is predicted to result in a better outcome beyond the +3 GDI point threshold (ORTHOSIS → NEW-ORTHOSIS). The circles scattered in the lower left quadrant are the limbs with an existing orthosis for which no orthosis is predicted to have a good response. These limbs are recommended to have "No Orthosis" (ORTHOSIS → NO-ORTHOSIS). The + symbols in the upper left quadrant are limbs without an existing orthosis that are predicted to have a good response with an orthosis (NO-ORTHOSIS → ORTHOSIS). The × symbols in the lower left quadrant are limbs without an existing orthosis that are not predicted to have a good response in any orthosis (NO-ORTHOSIS → NO-ORTHOSIS).**

In total, 41% of limbs were recommended to wear an orthosis by the model compared to 47% who had an existing orthosis prescription (Figure 3.3A). The model indicates that almost 25% of the limbs in the benefit sample would be expected to benefit from an orthosis but didn't have one prescribed. Another 25% of individuals were expected to benefit by changing their existing prescription. These two large groups suggest that current prescription methodologies are not optimal for improving GDI in this population.

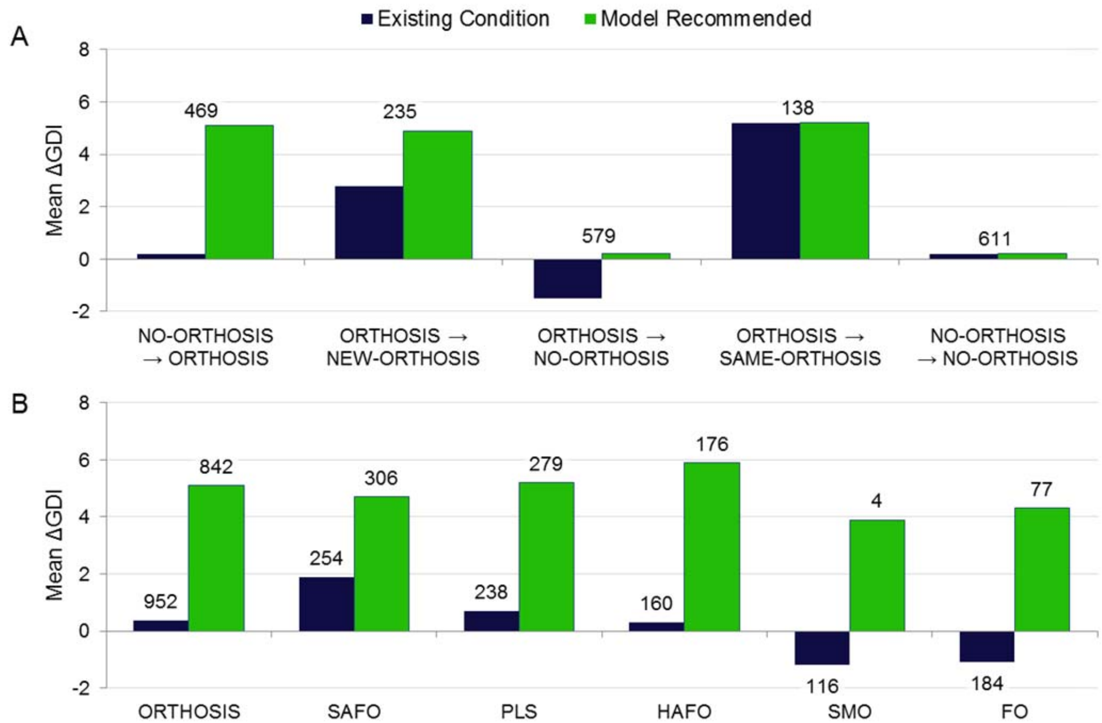


**Figure 3.3 – Predicted responses and limb count for various orthosis conditions and designs. The number above the bar indicates the number of limbs in the group while the bar height represents the average response (ΔGDI) in that group.A) Change in GDI for the five groups identified by the model. The ΔGDI for the first three groups improve significantly. The NO-ORTHOSIS → NO-ORTHOSIS and ORTHOSIS→SAME-ORTHOSIS groups have no change in outcome since there is no recommended change in the prescription (existing prescription = best prescription). The predicted average ΔGDI for all 2032 limbs improves to +2.3 by using the model recommendations compared to +0.2 for the existing condition. B) Breakdown of the designs of orthosis used between the existing orthosis prescriptions and model recommended orthosis prescription groups. The predicted ΔGDI for limbs wearing an orthosis improves to +5.6 from +0.4. Total orthosis prescriptions numbers decrease slightly with the majority of prescriptions recommended for the PLS – 36%, HAFO – 33%, and SAFO – 21%.**

## 3.4 Discussion

The study set out to answer the question, "*Can we improve gait outcomes by recommending the orthosis design with the highest predicted response?*". To address this question we developed a statistical model capable of predicting changes in GDI for a limb when wearing five different orthosis designs. The model uses a patient's barefoot gait data to predict the optimal orthosis for improving overall gait quality. The predictive model was comprised of five ensembles developed using RFA techniques and data from 476 individuals. All ensembles utilized between 4 and 9 features and demonstrated excellent predictive performance with accuracies ranging from 67 - 82%. The RMSE values from 5.1 – 6.2 GDI points indicate modest prediction errors for an individual limb. However, these prediction errors should be viewed in light of the poor efficacy of the standard of care orthosis prescription scheme (ΔGDI = +0.4). Overall, the model provides significant benefit to the population, even if it does not perfectly predict the response for each individual limb.

The complexities involved in combining multiple CARTs makes it difficult to generalize the roles of each feature used by the RFA ensembles [78]. The model does not necessarily provide a biomechanical basis for change. Nevertheless, it is evident that certain conditions appear to make the chance of a positive outcome more likely. Barefoot GDI was a feature used in all but the HAFO ensemble, and foot progression features appeared consistently in all RFA ensembles. This suggests that barefoot GDI and foot progression are critical predictors for orthosis outcomes in children with diplegic CP.

To estimate the effects of the model on the diplegic CP population, we applied the model to a representative sample of patients seen by our clinical gait analysis service. The model recommended 41% of the limbs use an orthosis with a mean predicted ΔGDI of +5.6 points, improved from the ΔGDI of +0.4 for existing orthosis users. Overall, the total number of orthosis prescriptions remained nearly constant. Interestingly, 90% of limbs recommended an orthosis were recommended either the SAFO, PLS, or HAFO design. While the SMO and FO designs may provide other significant benefits (*e.g.* sit-to-stand motions), they appear to almost always be outperformed by the other designs when it comes to improving GDI.

Though performance of the model was excellent, it does have limitations. Although the model is derived from individuals with a symmetric prescription, it recommends orthoses on a limb-by-limb basis. Clinicians and patients will need to weigh the benefits of symmetric versus asymmetric prescription in a treatment plan. The role of shoes is a confounder in understanding the effects of orthoses on gait. The majority of orthosis studies use barefoot gait as the control condition [6], [7]. Data was drawn from a historical database, so study design was limited to data collected using standard practices at our center. As a result, the effects of shoes could not be isolated. The influence of shoe design is likely to have been randomized across all five orthosis designs, and therefore unlikely to have a significant impact on the overall study findings. Another problem, also common in orthosis research, is the lack of standardization in orthosis design [81]. The majority of orthoses at our center are custom fabricated and differences can exist even within same design (*e.g.* shank-to-vertical angle, heel height, trim lines, ankle stiffness). The model presented here does not take into account any of these differences. Further work is needed to understand the effects of these properties on gait before improvements to the model can be made.

In summary, this study suggests that the current standard of care results in orthosis prescriptions that maximize GDI only 37% of the time. The model we have proposed here not only maximizes the effect of orthosis on gait, but also identifies patients that are not likely to benefit from an orthosis. Information gained by using this model can assist clinicians by providing clinically meaningful, patient specific, optimal orthosis prescriptions. Further validation of the model is warranted using data from other centers and a prospective study.

# Chapter 4: Low Gait Efficiency, Not High External Mechanical Work Rate, Explains Increased Metabolic Demand During Gait in Children with Cerebral Palsy

## 4.1 Introduction

As the most common childhood disability, cerebral palsy (CP) affects approximately 3.3 individuals per 1000 live births in the United States [4]. Children diagnosed with CP use two to three times more metabolic energy during walking than typically developing (TD) children [16], [82], [83]. Individuals with CP use the same amount of energy to walk as TD individuals use to climb stairs or run at a moderate pace [84]. These high metabolic demands have a significant impact on participation, activity, and quality of life.

Traditionally, interventions, such as the selective dorsal rhizotomy or orthopaedic surgery, have been used to reduce these high metabolic demands. The proposed mechanism underlying these treatments is that the rhizotomy will reduce spasticity and co-contraction, orthopaedic surgery will correct skeletal malalignment, and as a result, metabolic demand will drop [85]. However, while metabolic demand drops following rhizotomy it is not normalized. Even after subsequent orthopaedic surgery, metabolic demand is still excessive [86]. This indicates that the origins of the high metabolic demand are still largely unknown.

The gait of individuals with CP is often referred to as being “inefficient”, due to the high metabolic work rates measured during walking [83], [87], [88]. In fact, poor gait economy, rather than gait efficiency, more appropriately describes this phenomenon. Economy is the *distance* that can be traveled from a given *energy supply*. Gait economy, then, is the overall distance that a person can walk from a given metabolic energy supply. Efficiency, on the other hand, is the amount of *useful energy* that can be derived from a given *energy supply*. Gait efficiency, then, is the amount of energy that can be used to move the body's center of mass (COM) from a give metabolic energy supply. An individual with an efficient gait would be able to produce more COM work than an individual with an inefficient gait, given the same metabolic energy supply. While a higher efficiency is likely to lead to a greater economy, the subtle difference between the two terms is important.

Gait economy provides a “big picture” assessment relating the total energy it takes to move the entire body from point A to point B, only accounting for the distance the body moves between the points. Economy may be useful for assessing the functional impact of high energy demand, but it is not useful for identifying the underlying cause, and possible treatments. Gait efficiency, on the other hand, accounts for the relative motion of the COM and provides a precise picture of energy utilization.

Gait economy is often assessed during clinical gait evaluations, and its value is typically presented as metabolic cost (metabolic energy expended per unit distance) [82]. However, unlike gait economy, gait efficiency is not routinely calculated during clinical gait evaluations; although the data required for the calculation is regularly collected. An assessment of gait efficiency in CP may be beneficial in explaining some individual's metabolic energy expenditure is so high.

Gait efficiency is determined by how much metabolic energy is used for moving the COM and how much metabolic energy is converted to other forms of energy. For example, co-contraction, poor muscle timing, and excessive arm swing can all lower gait efficiency as they increase metabolic energy demands but do not increase the total mechanical energy of the body [89].

Because the point of walking is to get "*from there to here, from here to there*"[90], we define 'useful energy' for gait as the energy that is used to move the body's COM relative to the environment. During gait, metabolic energy (*i.e.* energy supply), primarily derived from aerobic activity, is continuously converted to various forms of energy which include internal work (work that does not change the total mechanical energy of the body), external work (work that does change the total mechanical energy of the body), thermal work (body heat), and even sound (muscle by-product) [91], [92]. Specifically, the portion of external work used to move the COM relative to the environment is called external mechanical work (EMECH). Therefore, we define useful energy as the time-rate at which EMECH is being performed during walking (EMECH rate). Additionally, we define energy supply as the time-rate at which metabolic energy is being consumed during walking (MET rate). Using these definitions, gait efficiency can be defined as the EMECH rate divided by the MET rate.

$$\text{Gait Efficiency} = \frac{Useful\ Energy\ Rate}{Energy\ Supply\ Rate} = \frac{EMECH\ Rate}{MET\ Rate} \qquad \text{Eq. 4.1}$$

In the clinical setting, the EMECH rate can be computed using force plate data and step-to-step transition analysis techniques, while the MET rate can be measured through breath-by-breath gas analysis and indirect calorimetry [82], [93], [94]. Previous studies have used similar methods for estimating gait efficiency for both typically developed and hemiparetic adults [21], [95].

In short, step-to-step transition analysis uses ground reaction force vectors and the COM trajectory measured during walking to calculate the EMECH performed on the COM [94]. In the analysis of a single step, the work performed on the COM is often separated into five components: pre-propulsion, propulsion, collision, rebound, and pre-load [21]. These components are often helpful for understanding work production and absorption phases during a step. In previous studies of TD adults, EMECH was shown to be a major determinant of metabolic consumption; explaining 89% of the metabolic variance in TD adults [21]. Additionally, EMECH was shown to be sensitive to changes in step length, step width, step frequency, and walking speed [17], [21], [94], [96], [97]. Using EMECH and MET rates, gait efficiency for TD adults was estimated to be 50% [21]. Children with diplegic CP have been shown to produce more EMECH and consume more metabolic energy than TD children during walking [16], [82], [87]. However, the increase in work was not compared to changes in metabolic expenditure, and thus, the gait efficiency of children with diplegic CP has not previously been evaluated.

The purpose of this study was to investigate gait efficiency for children with diplegic CP in order to better understand the source of excessive MET rates typically observed for these individuals. We hypothesized that gait efficiency would be lower in children with CP. Furthermore, we hypothesized that both reduced efficiency and excess EMECH would contribute to the high MET rates.

## 4.2 Methods

### *Study Group*

A representative sample of the pediatric ambulatory diplegic CP population seen at a regional treatment center was assembled by conducting a comprehensive search of our clinical database. Inclusion criteria were:

- diagnosis of diplegic CP
- three-dimensional instrumented gait analysis performed for level ground walking at self-selected speed
- consecutive force plate strikes achieved during gait analysis trial
- metabolic assessment performed for level ground walking at self-selected speed during the gait analysis visit

Individuals walked barefoot and did not use assistive devices during data collection. To minimize the confounding effects of speeding up or slowing down, the speed at the start of a step was required to be within 15% of the speed at the end of the step. Additionally, individuals having a clinical diagnosis noting asymmetric involvement (e.g. asymmetric dipelgia) were excluded from the analysis. During data collection, the gait analysis and metabolic assessment were performed at different times on the same day, and an individual's data was excluded if the speed difference between the gait analysis and metabolic assessment was greater than 15%. For individuals with data from multiple visits, only data from the first visit was used.

### *Control Group*

In addition to the pediatric CP sample, a pediatric control sample was assembled from retrospective gait data collected at the same regional center during a previous study of 83 TD children [69]. Data was available for these children walking at 5 different speeds – very slow, slow, free, fast, and very fast. To match the speed ranges from the CP group, only the slow and free speed trials for the control group were used in this analysis. Metabolic assessment data was not collected for the TD individuals. Instead, metabolic data was calculated using a historical regression equation derived from a similar sample of pediatric TD controls (see ‘Metabolic Work Rate’ section).

### *External Mechanical Work Rate*

The external mechanical work rate contributing to COM motion was calculated for both the CP and TD groups using the independent limbs step-to-step transition method [94]. A step was defined as the foot strike of one limb to the foot strike of the contralateral limb. In short, to calculate the EMECH rate,

1. The step power produced by each limb over a single step was calculated using ground reaction forces and the COM trajectory [94]. Ground reaction forces were collected using force plates (AMTI, Watertown, MA) sampling at 1080 Hz.

2. The positive and negative components of EMECH for each limb were calculated by integrating step power over the periods corresponding to pre-propulsion, propulsion, collision, rebound, and pre-load [21].

3. The EMECH *rate* was calculated by multiplying the EMECH components measured for a single step by the step frequency.

We assumed that the work provided for each step was the same for both the left limb leading and right limb leading configurations, due to the exclusion of individuals with clinically noted asymmetric involvement, and the requirement of matched start and end of step speed. Therefore, work values were averaged where multiple trials for an individual were available. Work rates were non-dimensionalized according to Hof [67].

### *Metabolic Work Rate*

Metabolic data for the CP group was collected using a breath-by-breath gas analyzer (Medgraphics, St. Paul, MN). Oxygen consumption rates were converted to MET rates assuming standard temperature and pressure, and aerobic muscle activity (20.1 J/ml $O_2$) [98]. The protocol for measuring the walking net MET rate for individuals with CP consisted of a quiet period sitting in a reclined chair, followed by a 6 minute free speed walk [99]. The walking net MET rate was calculated as the difference in metabolic consumption between walking and sitting rates, and was non-dimensionalized according to Hof [67].

As mentioned, metabolic data was not collected for the control group. In lieu of measured data, the dimensionless MET rate was calculated using a regression derived from metabolic data collected from a sample of 168 TD children during a previous study,

$$Metabolic\ Work\ Rate = 1.766v^3 - 1.166v^2 + 0.394v + 0.007 \qquad \text{Eq. 4.2}$$

where $v$ is the average dimensionless walking speed ($v = speed/\sqrt{gL_{leg}}$) measured during the walking trial [98].

### *Efficiency Analysis*

Mean gait efficiency for the CP and TD groups was determined using linear regression. Each regression was required to intersect the MET rate axis at the respective group's average standing net MET rate, as this represents the MET rate when no EMECH is being performed. Standing net MET rates were determined for each group by calculating the difference between sitting and standing MET rates for 1489 children with diplegic CP and 139 TD children previously seen at the same regional center.

### *Analysis*

Statistical analyses were performed using Matlab with the Statistics Toolbox (2013b). Between-group comparisons were made using independent samples t-tests unless otherwise noted, and statistical significance was set at $\alpha = .05$.

## 4.3 Results

From a pool of approximately 1,700 children diagnosed with diplegic CP seen at our center, a total of 306 children met the inclusion criteria and were included in the study group. Primary reasons for being excluded from the study were speed differences between the gait analysis and energy assessment and non-consecutive force plate strikes. For the control group, a total of 73 TD children walking at their free walking speed and 59 TD children walking at a slow speed met the inclusion criteria and were included in the control group.

Statistically, the CP group had a 27% higher gross EMECH rate and consumed metabolic energy at a rate 97% higher than the TD group average (Table 4.1). There were no significant differences in age or walking speed between the pediatric CP and TD groups.

**Table 4.1 – Group Characteristics of Data Used for Efficiency Regression**

| | Free Speed CP | All TD | p-value (CP vs. All TD) |
|---|---|---|---|
| **n** | 306 | 132 | - |
| **Age [years]** | 11.4 (3.3) | 11.2 (3.1) | 0.59 |
| **Male:Female** | 180:126 | 75:57 | 0.78† |
| **Mass [kg]** | 39.0 (16.1) | 42.7 (15.3) | **0.03*** |
| **Leg Length [m]** | 0.74 (0.11) | 0.78 (0.11) | **<0.01*** |
| **GDI** | 78.4 (9.8) | 99.4 (8.0) | **<0.001*** |
| **Walking Speed** | 0.368 (0.058) | 0.373 (0.069) | 0.45 |
| **Negative Work Rate** | 0.021 (0.009) | 0.016 (0.005) | **<0.001*** |
| **Positive Work Rate** | 0.021 (0.009) | 0.016 (0.005) | **<0.001*** |
| **Gross Work Rate** | 0.042 (0.016) | 0.033 (0.010) | **<0.001*** |
| **Metabolic Work Rate** | 0.175 (0.063) | 0.088 (0.019) | **<0.001*** |

All values are dimensionless unless otherwise noted
() indicates ±SD,
* indicates statistical significance,
† p-value calculated using a Chi-squared test

### *Efficiency*

Children with CP had a significantly lower mean gait efficiency compared to TD children (CP 24% vs. TD 40%, $p < .001$) (Figure 4.1). The mean gait efficiency measured for TD adults, 48% [adapted from [21]], was higher than either the CP ($p < .001$) or pediatric TD ($p < .001$) groups.

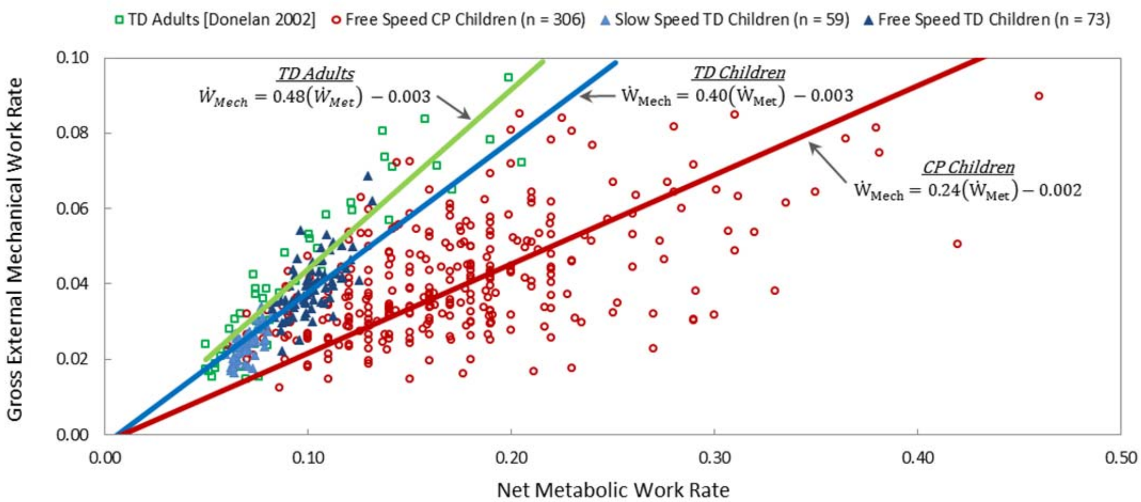


**Figure 4.1 – Gait efficiency analysis for 306 CP and 74 TD children. Children with CP had a mean gait efficiency of 24% while TD children had a mean efficiency of 40%. Coefficient of determination (r2) was 0.15 for the CP group and was 0.89 for the TD adult group, showing heterogeneity of efficiency between CP individuals. An r2 value was not computed for the TD control group since their metabolic work rates were based on historic data (Equation 5). The regressions were forced to pass through their respective average standing net metabolic work rate measured using data previously collected at our center (0.007 for the CP group and 0.009 for the TD group). Experimentally, no statistical differences were found between the standing net metabolic work rates of the two groups (p = .10).**

### *Step Analysis*

The typical power produced and absorbed during a single step for the CP and TD groups was similar in overall pattern (Figure 4.2A). The CP pattern can be generally described as lacking pre-propulsion and propulsion, while having excessive collision, rebound, and preload. The CP group tended to not produce positive power until after contralateral heel strike (0% and 100% of step), while the TD group started positive power production much earlier, before the end of single support, at approximately 90% of the step. Overall, the CP group tended to produce the largest amount of positive work during the rebound portion of the step, whereas the TD group produced the largest amount of positive work during propulsion/pre-propulsion (Figure 4.2B).

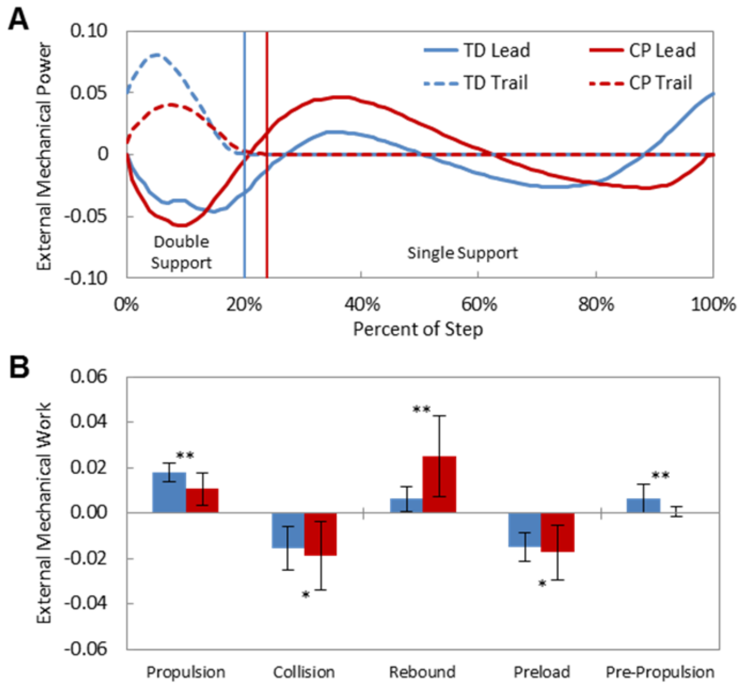


**Figure 4.2 – Step power and work measurements. A) Median power curves for the TD and CP groups from foot strike of one limb (0% of step) to the foot strike of the contralateral limb (100% of step). The curves were calculated by taking the median value every 2% of the gait cycle (GC) for the leading and trailing limbs of each group. Contralateral limb foot off times separating double and single support periods are indicated by the vertical lines. Conceptually, step work (the area between zero and the step power curve) can be broken up into 5 components: propulsion, collision, rebound, preload, and pre-propulsion [21]. Propulsion is positive work performed by the trailing limb during double support. Collision work is performed by the leading limb primarily during double support and is characterized by negative step power. Rebound work occurs just after collision, when step power becomes positive again. Preload work begins at approximately 60% of single support, when step power dips back to negative values. Finally, pre-propulsion work is performed by the supporting limb just prior to double support. In this analysis, the TD individuals began to produce propulsive power at approximately 90% of a step, which was approximately 10% of the cycle sooner than their CP counterparts. The CP individuals, in fact, did not generally produce positive power until the onset of double support. B) The delay in the production of propulsive work results in CP individuals producing less total propulsive work (propulsion + pre-propulsion) during a single step. For CP individuals, the deficit in propulsive work was compensated for by producing more rebound work than the TD group. The '*' and '**' symbols indicate statistical significance at $p < .05$ and $p < .001$, respectively. All values have been non-dimensionalized.**

## 4.4 Discussion

The findings of this study confirmed our hypotheses that gait efficiency is substantially lower for children diagnosed with diplegic CP, and that these children perform more external mechanical work compared to TD children. Our second hypothesis was also confirmed, that both decreased efficiency and excess external mechanical work were responsible for the increased metabolic demand.

In total, the metabolic work rate was nearly two times as high for the CP group compared to TD controls and the CP group produced about one third more external mechanical work than the TD group. This means that the extra external mechanical work produced by the CP group is responsible for, at most, one third of the metabolic demand increase. The reduction in efficiency, then, accounts for the remaining two thirds of metabolic demand increase. Examining the relative sizes of these two effects (Figure 4.3), it is clear that low gait efficiency is the primary factor that explains why metabolic demand is nearly 100% higher for individuals with CP. In other words, although individuals with CP perform extra external mechanical work during gait, the conspicuous inability to efficiently convert metabolic energy into useful external mechanical work is the major factor which leads to the high metabolic demand.

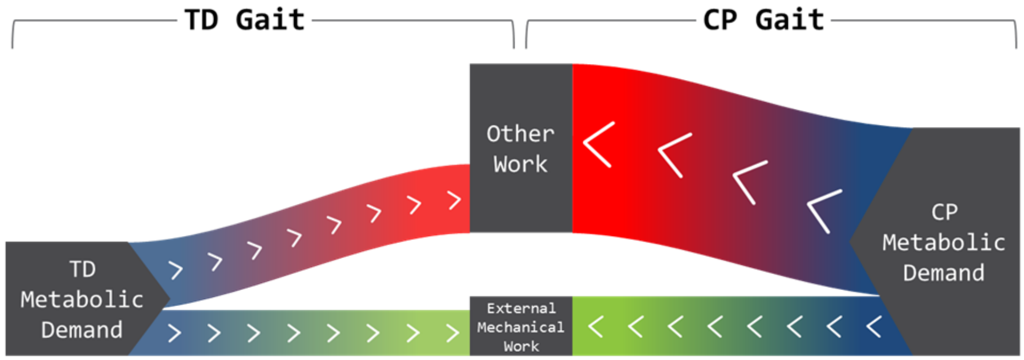


**Figure 4.3 – Energy conversion differences between TD and CP children. Most of additional metabolic demand of individuals with CP is primarily converted to forms of energy other than external mechanical work (e.g. internal work). This imbalance between external mechanical work and other work decreases gait efficiency.**

Previously, a single study has shown that individuals with spastic hemiplegic CP had similar gait efficiencies to TD individuals, concluding that all increases in metabolic cost were associated with proportional increases in mechanical work [100]. However, instead of using external mechanical work in the calculation, the study utilized total mechanical work (i.e. external mechanical work + internal mechanical work) as their "useful energy" term. We believe that in addition to studying a different population (hemiplegia vs diplegia), the difference in approach may have led to the differences in conclusion between their study and ours. If individuals with hemiplegic CP perform excessive amounts of internal work (which is likely), the inclusion of internal work in the efficiency calculation would inflate the group's useful energy term (numerator) which would boost the calculated efficiency. To properly compute gait efficiency, the work used for directly moving the COM (i.e. external mechanical work) must be isolated from other sources. For example, in internal combustion engines, engine efficiency calculations do not include the internal work required to redirect the pistons (although the work is necessary for the function of the engine). Engine efficiency is calculated by measuring only the work at the engine drive shaft divided by the energetic potential of the fuel being supplied to the engine. For our analysis, excessive internal work may indeed be the cause of the low gait efficiency, but to properly quantify gait efficiency, internal work must be excluded.

It is intriguing that CP children perform significantly more work during single support and significantly less work during double support compared to TD controls. We hypothesize that CP children lack the precise double support limb control, mediated by muscle activation, timing, and force generation/absorption, to optimally transition from one limb to another. Previous work has shown that children with CP are 33% less efficient at recovering mechanical energy when transitioning between potential and kinetic energy states [101]. As Steele et al. has shown, during stance, the plantarflexors contribute to nearly all of forward COM acceleration and nearly two-thirds of the vertical acceleration required to support the COM [24]. If the plantarflexors are impaired, as they commonly are in diplegic CP, the vasti take over to provide the required vertical acceleration to support the COM. However, the vasti also produce substantial backwards COM acceleration which the authors likened to "*driving with your parking brake on*". Moreover, to compensate for the backwards COM acceleration of the vasti, other muscles must act when they would normally be at rest. This cascade of muscle compensations is unlikely to be efficient. From these previous findings, we suspect that the inability of individuals with CP to provide significant plantarflexor COM acceleration is a primary factor that contributes to the decreased gait efficiency.

As previously mentioned, metabolic energy is converted to forms of energy other than external mechanical work. During walking, these primarily include internal mechanical work (e.g. arm motion relative to the COM), thermal energy (e.g. heat generated as a metabolic by-product), and workless energy (e.g. muscle co-contraction). These energy sinks may indirectly affect COM motion. For example, co-contractions can 'lock' a joint and allow energy transfer through limbs [102]. However, by definition, these energy sinks do not directly contribute to COM motion, and are, therefore, not included in the efficiency calculation. Nevertheless, their cumulative effect on efficiency may still be examined. This is because the metabolic energy not accounted for by external mechanical work is due to the combined effect of all remaining factors (i.e. metabolic inefficiency, excessive internal work, and excessive co-contraction can increase metabolic demands). By understanding how these remaining factors influence metabolic demand, it may be possible to better understand the mechanisms that cause gait efficiency to decrease.

There are some important limitations of our study. First, in our analysis of 306 individuals with CP, only 14% had 3 or more steps included in the analysis, 32% had 2 steps, and 54% had only 1 step. Comparatively, within the TD group, 66% had 3 or more steps, 14% had 2 steps, and 20% had 1 step. By averaging the work measurement over multiple steps, the effect of an individual's natural walking variability on the resultant work measurement is reduced. Assuming differences between steps for an individual are random, this experimental shortcoming would not affect the computed efficiency, and therefore would not change the overall conclusions of this study. Second, our analysis was restricted to individuals with a generally symmetric gait. This limits the applicability to other gait patterns. By collecting force plate data over an entire stride instead of a single step, the efficiency analysis can be adapted for those with an asymmetric gait. Third, the mechanical and metabolic measurements for CP children were made during separate tests on the same day. We limited speed differences between the motion analysis trials and metabolic assessment to 15% (similar results were observed using 10% and 20% speed differences). However, simultaneously collecting these two quantities on an instrumented treadmill would be preferred. As this was a retrospective analysis, we were limited by the historical data collection methods at our center. Finally, individuals were not forced to walk at a constant speed. Speed changes within a step were limited to 15% by the inclusion criteria, but most individuals did exhibit some slight speed changes. While there were no speed changes for any group as a whole, using an instrumented treadmill to fix walking speed would allow for efficiency and step work calculations to be isolated from this confounding influence.

This study investigated gait efficiency, defined as the relationship between external mechanical work and metabolic work rates, for children with diplegic CP. Compared to TD children, the CP group had a substantially lower gait efficiency despite performing slightly more work during walking. This means that the metabolic rate is exacerbated by both of these factors. We suspect that the low efficiency in the CP group is partly due to factors that are treatable, such as spasticity, weakness, and biomechanical malalignment. However, low efficiency may also be due to factors for which no current treatments exist, such as poor underlying motor control. Clinically, a proper gait efficiency evaluation, as described in this study, could identify the relative influence of gait efficiency and external mechanical work on an individual's metabolic work rate. This type of analysis could help select targeted interventions which address the primary cause of the increased metabolic work rate. For example, if excess external mechanical work is being performed during a step, selection of an intervention that targets a reduction in work (e.g. ankle-foot orthoses) might be effective at reducing the metabolic work rate. On the other hand, if efficiency is low, then perhaps spasticity reduction may be more effective at reducing the metabolic work rate. Such an approach to reducing the metabolic work rate is clearly speculative right now, but the analytical method outlined here provides a means for testing such hypotheses. By investigating the effectiveness of common interventions designed to improve gait efficiency, researchers may be able to better understand the underlying mechanisms related to the excessive metabolic energy rates seen in children with diplegic CP.

# Chapter 5: Ankle Foot Orthoses Allow Proximal Muscles to Compensate for Poor Plantarflexor Function in Children with Diplegic Cerebral Palsy

## 5.1 Introduction

Ankle foot orthoses (AFO) are commonly prescribed for individuals diagnosed with cerebral palsy (CP). Typical objectives of AFO use are to improve overall ankle function and thereby enhance general gait quality and energy economy [103], [61], [40]. While AFOs have been shown to improve gait, the biomechanical bases for many of the improvements are not well understood [6], [7]. A critical evaluation of how AFO use influences a muscle's ability to contribute to center of mass (COM) accelerations may help to explain the mechanisms behind these improvements.

Fundamentally, AFOs are designed to restrict undesirable ankle motion. During gait, AFOs passively store energy when the AFO is flexed and then release that energy as the AFO returns to its molded position (i.e. the AFO's *neutral angle*). It is commonly thought that this energy storage and return mechanism is responsible for lowering metabolic energy costs. However, the amount of energy that an AFO is capable of storing is only a small fraction of the total energy produced at the ankle during gait [47], [104], [105]. Additionally, the timing of the energy return has been shown to be critical to reducing metabolic costs; and passive AFOs have no way of controlling the timing of energy release [17], [106]. These two factors make the AFO energy storage and return mechanism unlikely to provide significant improvements in gait.

Through simulation, the plantarflexor muscles have been shown to be critical for generating much of the vertical and forward accelerations used to support and propel the body during gait [24]. For optimal plantarflexor function, good motor control, sufficient strength, and good range of motion are required. These plantarflexor qualities are diminished for children with CP. We hypothesize that AFOs, by their semi-rigid design, improve gait by providing and individual with a stabilized ankle which allows for more functional proximal muscles to compensate for diminished plantarflexor function.

One way to investigate if AFOs allow for proximal muscles to have a greater influence on COM motion during gait is to use simulation and induced acceleration analysis (IAA). An IAA is, in essence, a perturbation analysis where the influence of muscles on COM acceleration can be elicited. Previous IAA work using motion capture, EMG, and ground reaction data along with biomechanical simulation investigated how muscle function changed for children with CP walking with various levels of crouch gait [24]. The analysis concluded that for crouch gait, if ankle plantarflexors are weak, other muscles must compensate to provide the necessary supportive force for the COM. Interestingly, it was also shown that the muscle compensations were not entirely beneficial. For example, the compensatory muscles used to provide COM support, also functioned to accelerate the body *backwards*. This meant that the muscles used to support the COM were also working against the goal of moving the COM forward. The inability to provide high levels of plantarflexor power during gait and the compensatory reaction of other muscles were brilliantly likened to "*driving with your parking brake on*". Using a similar methodology to investigate changes in muscle function due to AFO use may provide a clearer rationale for AFO prescription, and may eventually lead to improved AFO designs.

Therefore, the goal of this study was to investigate if lower limb muscle function could be influenced by the use of AFOs. Our primary research question was: Do the supportive and propulsive functions of muscles change when wearing an AFO? Our central hypothesis was that AFOs can change the basic function of the proximal muscle groups so that they can compensate for plantarflexor deficits.

### 5.2 Methods

This study used three dimensional motion analysis, EMG, and ground reaction data, coupled with biomechanical simulation and IAA to evaluate the impact of wearing an AFO on muscle function. The study was a repeated measures design where each subject was compared to themselves (barefoot vs. wearing AFOs), and therefore acted as their own control.

Five participants were recruited for this study. Participants were screened during their standard of care clinical appointment at Gillette Children's Specialty Healthcare's Center for Gait and Motion Analysis (CGMA). Individuals were considered for recruitment if they had a "qualitatively better gait" while walking with their AFOs as assessed by an experienced physical therapist in the CGMA. As this study was designed as a small proof of concept experiment, no explicit criteria were defined to distinguish an individual as having a qualitatively better gait with their AFOs and only required the physical therapist to perceive a general sense of improvement in gait with AFO use.

IRB approval was obtained prior to study recruitment. In addition to being subjectively critiqued as having a better gait with their AFO, each subject met the following inclusion criteria:

- Diagnosis of Cerebral Palsy
- Current prescription of a Solid/Rigid style AFO
- Age 7-17
- Gross Motor Function Classification System (GMFCS) level of I or II
- > 9 months post-surgery
- > 6 weeks post-botulinum toxin injection or hardware removal

For each study participant, three dimensional motion capture, ground reaction force, and EMG data was collected for two conditions: walking barefoot, and walking with their clinically prescribed AFOs (and athletic shoes). Motion capture data was collected at 120 Hz using a 12-camera system (Vicon Motion Systems, Oxford, UK) and processed using a standard motion capture model (Vicon Plug-in Gait). Functional range of motion trials were used to defined hip joint centers and knee joint axes [27], [107]. Ground reaction forces were collected using 6 force plates embedded in an 8 meter walkway (AMTI, Watertown, MA) sampling at 1080 Hz. Surface EMG data was collected for the vastus lateralis, rectus femoris, hamstrings, gastrocnemius, and tibialis anterior at 1080 Hz and bandpass filtered between 20 and 400 Hz (Motion Laboratory Systems, Baton Rouge, LA). In addition to the motion capture data, the ankle stiffness of each AFO used in the study was quantified using the Bi-articular Reciprocating Universal Compliance Estimator (BRUCE), which is an instrument developed to assess the mechanical properties of AFOs [108]. For each AFO, a singular value of ankle stiffness was quantified using the slope of a linear model fit to the AFO ankle torque versus ankle angle data as measured by BRUCE (Figure 5.1A). As the BRUCE mechanical ankle does not precisely align with an individual's anatomical ankle, the neutral angle of each AFO (i.e. the angle at which the AFO holds the ankle when no external loads are applied) was estimated using the mean ankle angle during swing as measured by the motion capture Plug-in Gait model instead of using the BRUCE data when AFO torque was zero (Figure 5.1B).

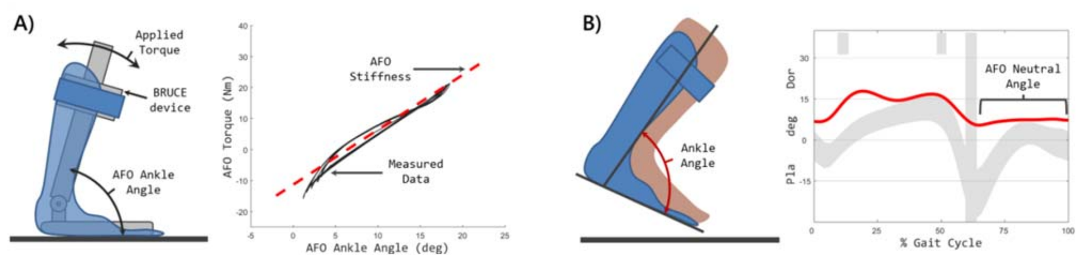


**Figure 5.1 – Identification of AFO mechanical properties. A) Using the BRUCE device, the foot plate of the AFO is held fixed (fixation mechanism not shown) and torque is applied to the leg of the device to move the upper portion of the AFO through a modest range of motion. Both AFO torque and AFO ankle angle data was sampled at 100 hz. The AFO ankle stiffness was determined as the slope of a linear model fit to the AFO torque and angle data. B) The AFO neutral angle was determined by the mean ankle angle during swing, when it is expected that minimal loads are applied to the AFO.**

Using a biomechanical modeling and simulation software package (OpenSim, opensim.stanford.edu), body segment motion, ground reaction forces, EMG, AFO neutral angle, and AFO ankle stiffness data collected during motion capture trials were used to develop two dynamic musculoskeletal models for each individual: one model to simulate walking barefoot, and another model to simulate walking with AFOs. Each musculoskeletal model in the simulation was developed from a generic 23 degree of freedom (DOF) model consisting of 92 musculotendon actuators [109]. Model DOFs included a 6-DOF pelvis, a 3-DOF ball and socket joint at the pseudo torso/pelvis joint, a 3-DOF ball and socket joint at each hip, a 1-DOF coupled translation/rotation joint at each knee, and a 1-DOF revolute joint at each ankle. Joints at the subtalar and metatarsophalangeal joints (4-DOF in total) were constrained for the simulation, reducing the total model DOF to 19. To incorporate the contribution of an AFO into the model, a custom torque actuator was added to each ankle joint. The torque actuator allowed for a prescribed time variant torque equal to the product of the experimentally determined AFO stiffness value and the angular difference between the AFO's neutral angle and the model's ankle angle – which varies over the gait cycle – to be applied at the ankle. The torque applied to the ankle by the actuator (simulated AFO) was equal to,

$$\tau(t)_{AFO} = k_{AFO}(\theta(t)_{Ankle\ Angle} - \theta_{AFO\ Neutral\ Angle}) \qquad \text{Eq. 5.1}$$

where $\tau(t)_{AFO}$ is the torque applied by the actuator on the ankle throughout gait, $k_{AFO}$ is the stiffness of the AFO, $\theta(t)_{Ankle\ Angle}$ is the time ankle angle measured throughout gait, and $\theta_{AFO\ Neutral\ Angle}$ is the neutral angle of the AFO identified by the mean ankle angle during swing.

### *Simulation Details*

For the simulation, each model was scaled according to the anthropometric body size of the individual, and joint angles were calculated for the entire gait cycle using an inverse kinematics process [110]. This process places the model in the optimal pose at each motion capture frame which minimizes errors between the simulated and experimental markers and coordinates. Next, inverse dynamics was used to calculate joint moments (i.e. kinetics) based on the modeled kinematics and ground reaction forces [111]. As consecutive force plate strikes were needed to create a simulation for an entire gait cycle for both limbs, only trials that contained consecutive force plate strikes were used in the analysis. After kinetics were calculated for the model, the residual reduction algorithm (RRA) was used to minimize the effects from modeling and marker data errors which may lead to high residual forces being applied to the model [112]; residual forces may be applied to the pelvis during the inverse dynamics process to force the kinetics to accurately match the ground reaction data. The RRA systematically alters the torso mass center and body kinematics by small amounts to reduce the residual forces by providing a better match between the ground reaction forces and kinetics.

The computed muscle control (CMC) algorithm was then used with the RRA adjusted model to calculate the muscle excitations and forces necessary to track the simulated kinematics [110]. The experimental EMG data collected during the motion capture session was used to constrain the muscle excitations for the CMC to encourage the simulation to mimic the experimentally measured activations. Muscle on and off periods were determined by visual inspection of processed motion capture EMG data for each trial consisting of the normalized motion capture EMG data superimposed with the linear enveloped EMG signal.

Finally, after muscle activations and corresponding muscle forces were calculated using CMC, IAA was used to identify the contribution of each muscle to the total COM acceleration during stance. By modeling the foot-floor contact as a rolling without slipping foot contact constraint type, the IAA algorithm permutes the force of a muscle by one newton independent of the other muscles and calculates the corresponding change in COM accelerations of the model. This permutation process is repeated for all muscles and results in an assessment of the acceleration potential of each muscle per newton of force throughout the gait cycle. Each muscle's potential over the gait cycle is then multiplied by the muscle force estimated during CMC to calculate the muscle's total supportive and propulsive contribution to COM acceleration during gait. The functional contributions of individual muscles to supportive and propulsive COM accelerations were elicited by comparing a muscle's function between the two simulations (i.e. barefoot walking versus walking with an AFO).

***Additional Data***

Muscle strength values assessed during a comprehensive physical examination were used to investigate the potential influence of muscle strength on COM acceleration.

### *Data Analysis*

One randomly selected limb from each individual was used for analysis. Analysis of the simulations and IAA results consisted of: 1) a comparison of the experimentally measured kinematics and EMG to the simulated model kinematics and EMG to identify how well the simulated musculoskeletal model represented measured gait, 2) a comparison of joint angles between walking with an AFO and walking without an AFO, and 3) a comparison of muscle contributions to vertical and horizontal COM accelerations between walking barefoot and walking with an AFO. Quantitative comparisons between barefoot walking and walking with an AFO were made using paired sample t-tests ($\alpha = .05$). All statistical analyses were performed using the Matlab Statistics Toolbox (Mathworks, Natick, MA).

## 5.3 Results

Of the 5 individuals recruited for the study, one participant was not able to complete the protocol, and was excluded from the analysis. The 4 remaining participants varied in age from 11 – 17 years old and consisted of 2 males and 2 females (Table 5.1). All individuals in the group were functioning at GMFCS level 2 and all had a prescription for bilateral solid AFOs (B-SAFO). The neutral angle of the AFOs ranged from 6–12° of dorsiflexion and had stiffness ranging from 1.3 Nm/deg to 8.3 Nm/deg.

**Table 5.1 – Participant Details and their AFO Characteristics**

| | | | | | | | | Left AFO | | Right AFO | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| Sub # | Age (yrs) | Sex | Height (cm) | Weight (kg) | GMFCS | FAQ | AFO Type | Neutral Angle (deg) | Stiffness (Nm/deg) | Neutral Angle (deg) | Stiffness (Nm/deg) |
| 1 | 14.3 | F | 153 | 37.3 | II | 10 | B-SAFO | 8 | 7.8 | 10 | 8.1 |
| 2 | 11.3 | F | 143 | 33.3 | II | 8 | B-SAFO | 10 | 1.3 | 7 | 1.5 |
| 3 | 17.4 | M | 170 | 55.7 | II | 8 | B-SAFO | 6 | 3.0 | 11 | 2.7 |
| 4 | 14.6 | M | 156 | 78.6 | II | 9 | B-SAFO | 11 | 7.8 | 12 | 8.3 |

Joint angles from the simulated musculoskeletal model matched the experimentally captured data well; RMS errors were below 1° for pelvic obliquity, pelvic tilt, pelvic rotation, hip flexion, hip rotation, knee flexion, and ankle dorsiflexion for all individuals. On/off periods of the simulated EMG matched the experimentally collected EMG moderately well.

### *Kinematic Changes*

The influence of the AFO on kinematics varied for all 4 subjects. Substantial kinematic changes were observed for Subject 3 with the use of an AFO, such as greater hip and knee extension, more normal hip rotation, and a slightly more plantarflexed ankle (Figure 5.2).

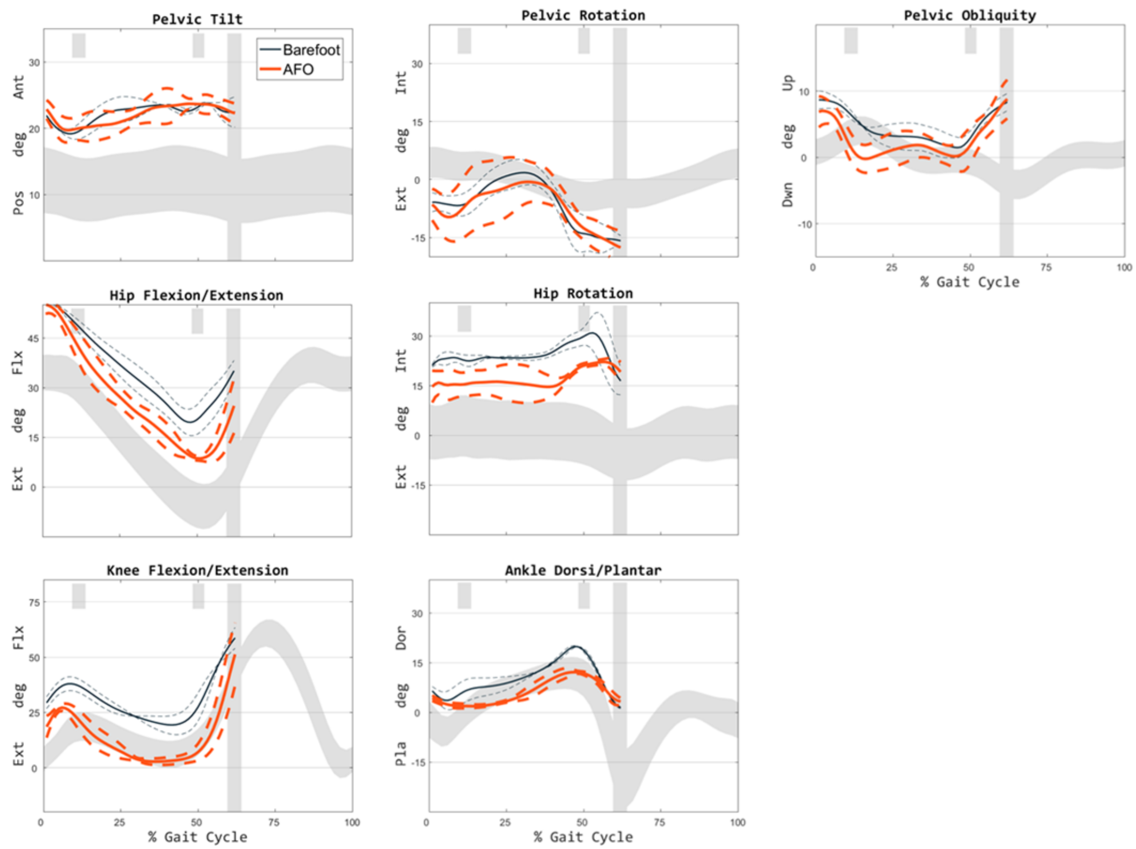


**Figure 5.2 – Kinematic plots for Barefoot and AFO walking simulations for Subject 3. The solid line is the mean of all trials collected and the dashed line is 1 standard deviation from the mean. Grey bands represent normal gait ± 1 SD, full scale vertical bands represent typical toe off, and short vertical bands at the top of each plot represent opposite toe off and opposite heel strike, respectively.**

For Subject 2, although categorized as having a qualitatively improved gait in their AFOs by the physical therapist conducting the gait analysis, the use of an AFO did not substantially change the kinematic pattern other than promoting slightly more hip flexion during late stance (Figure 5.3). Kinematic changes for Subject 1 and Subject 4 are shown in the Appendix (Figures A1 and A2).

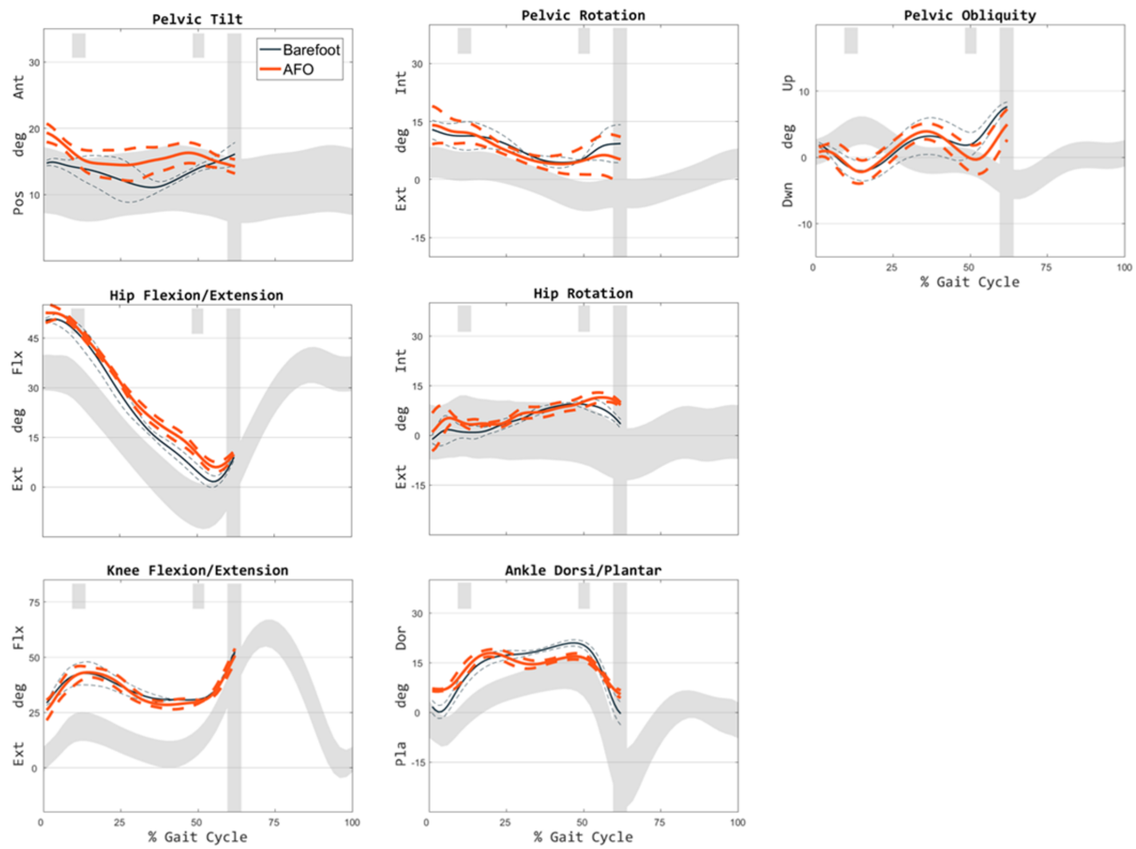


**Figure 5.3 – Kinematic plots for Barefoot and AFO walking simulations for Subject 2. The solid line is the mean of all trials collected and the dashed line is 1 standard deviation from the mean. Grey bands represent normal gait ± 1 SD, full scale vertical bands represent typical toe off, and short vertical bands at the top of each plot represent opposite toe off and opposite heel strike, respectively.**

### *COM Acceleration Changes*

Similar to the kinematics, the influence of an AFO on a muscle's influence on COM acceleration during gait (i.e. muscle acceleration potential multiplied by muscle force) varied for all 4 individuals. For Subject 3, substantial differences in vertical acceleration for nearly all of the major muscle groups were measured (Figure 5.4). For the proximal muscle groups, the gluteus maximus and gluteus medius contributed considerably more to vertical COM acceleration during the first double support period (HS to OTO) when wearing AFOs.

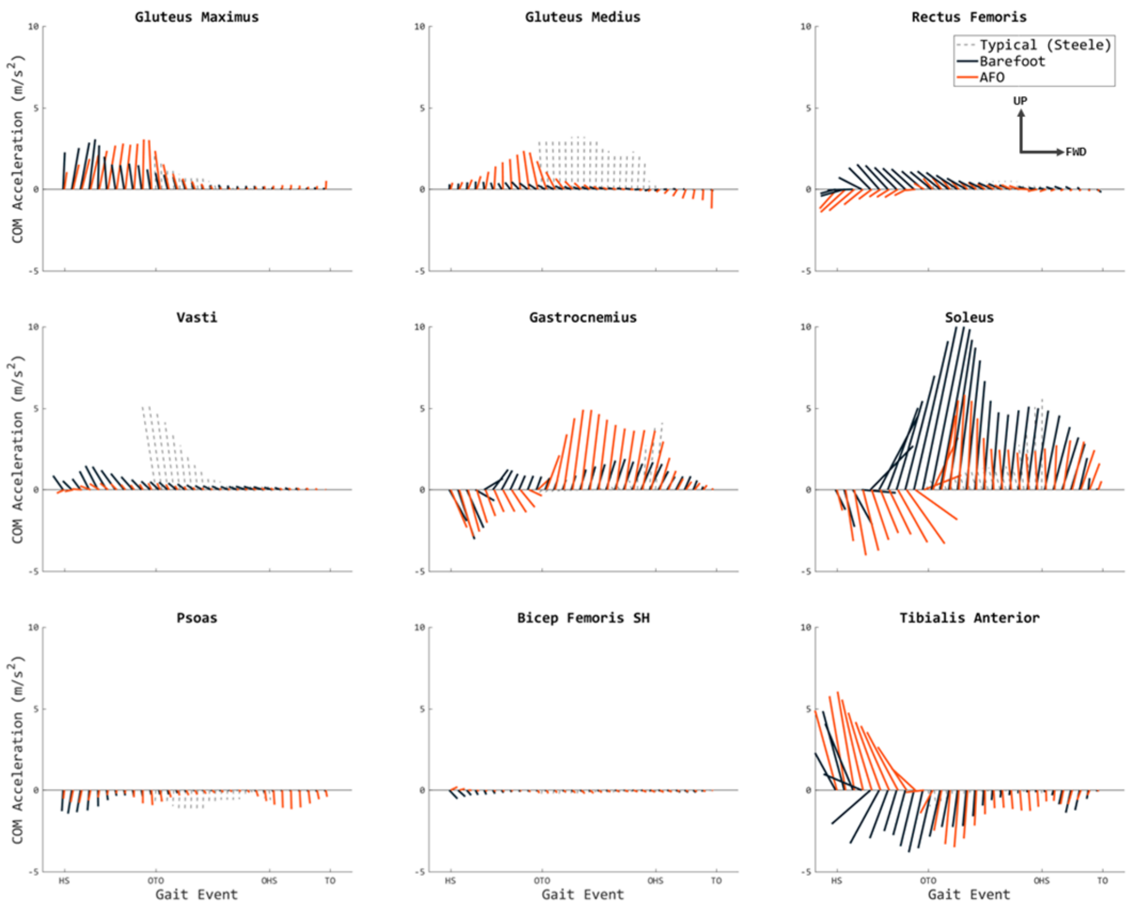


**Figure 5.4 – Total COM accelerations for Barefoot and AFO walking for Subject 3. Muscles with the largest changes between conditions were the soleus, gastrocnemius, and tibialis anterior. Smaller changes are present for nearly all of the remaining muscles. The dashed grey lines represent data from typically developing individuals during single support adapted from Steele et al. [25]. The vertical displacement of each vector from zero represents the vertical COM acceleration induced during stance while the horizontal displacement of the vector represents the fore-aft induced COM acceleration. Vectors are plotted at approximately each 3% of stance. Abbreviations: HS – heel strike, OTO – opposite toe off, OHS – opposite heel strike, TO – toe off.**

For Subject 2, there were fewer changes in COM accelerations when wearing an AFO (Figure 5.5). The soleus muscle contribution to vertical COM acceleration was noticeably reduced and the gluteus maximus, gluteus medius, and gastrocnemius contributions to total vertical COM acceleration were reduced as well but to a lesser degree. The COM acceleration changes for Subject 1 and Subject 4 are shown in the Appendix (Figures A3 and A4).

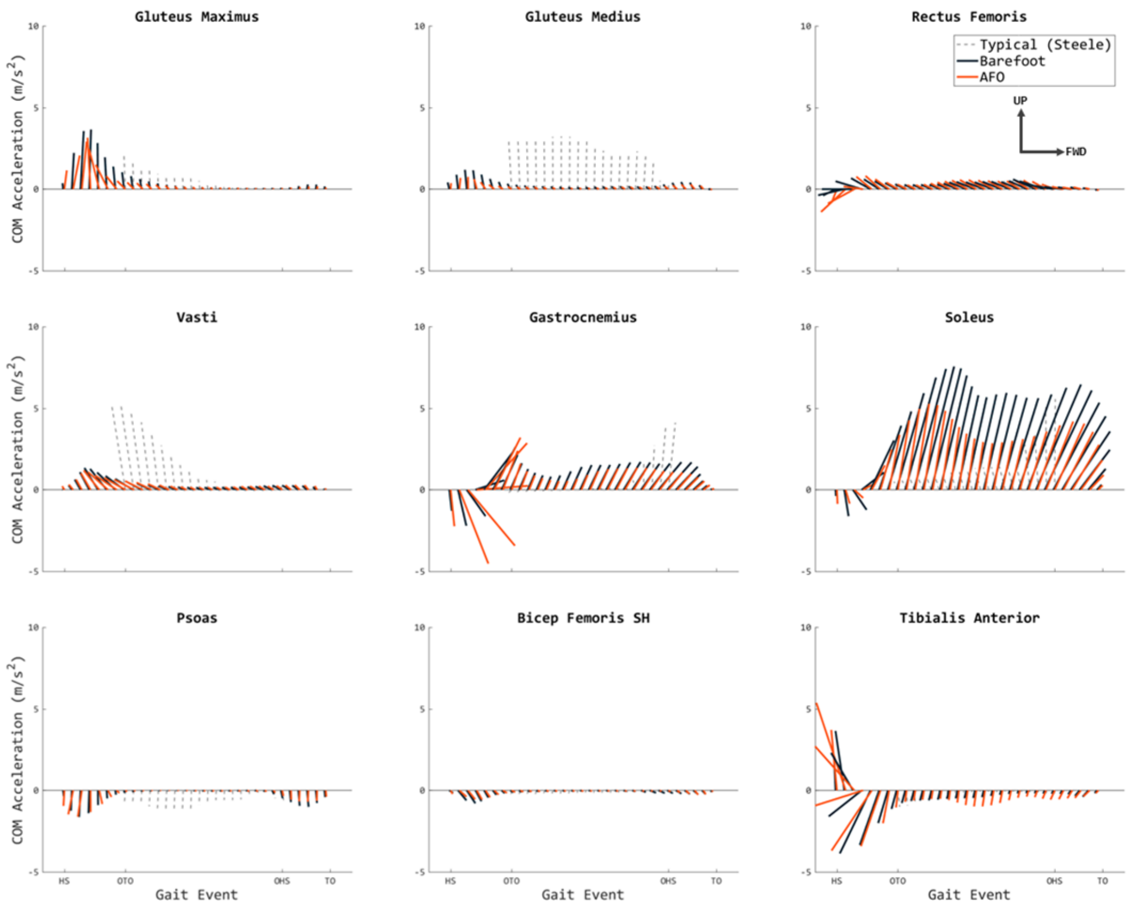


**Figure 5.5 – Total COM accelerations for Barefoot and AFO walking for Subject 2. The only muscle group with a noticeable change was the soleus. All other muscle groups experienced minimal changes. The vertical displacement of each vector from zero represents the vertical COM acceleration induced during stance while the horizontal displacement of the vector represents the fore-aft induced COM acceleration. Vectors are plotted at approximately each 3% of stance. Abbreviations: HS – heel strike, OTO – opposite toe off, OHS – opposite heel strike, TO – toe off.**

When analyzing the *potential* of a muscle to accelerate the COM, significant changes were observed for all individuals. For Subject 3, changes in the COM acceleration potential for most muscle groups were statistically significant in the vertical direction and AFO use appears to have some effects in the horizontal direction as well, although most of the horizontal changes were not statistically significant (Figure 5.6). For vertical COM acceleration potential, AFO use increased the potential of the proximal gluteus maximus and medius while reducing the potential of the distal gastrocnemius, soleus, and tibialis anterior muscles. For horizontal COM acceleration, the same trend appears to hold true where the distal muscles appear to have a lower potential during AFO use while the proximal muscles appear to have a higher potential.

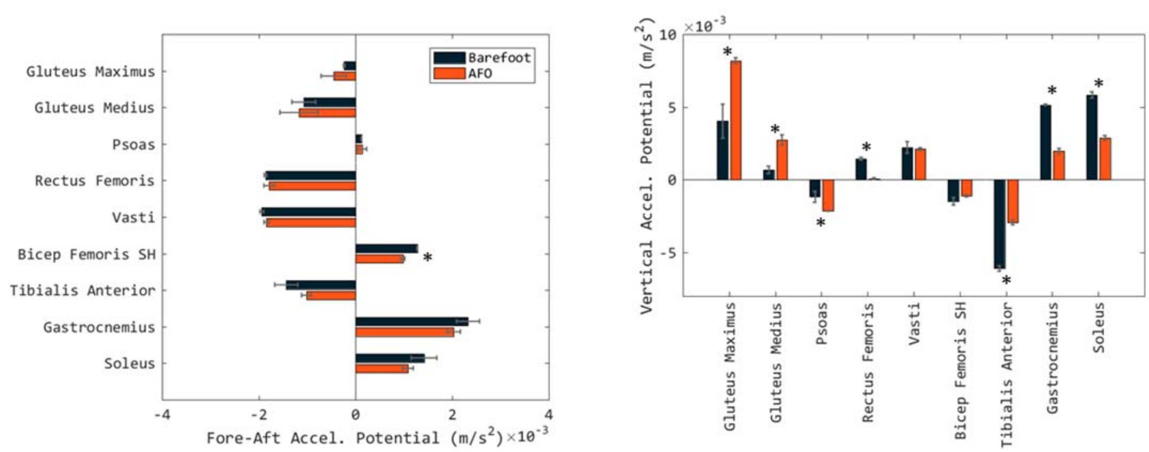


**Figure 5.6 – Muscle COM potentials for Barefoot and AFO walking for Subject 3. The muscle potentials for each trial were averaged for the entire stance phase and then individual trials were averaged.**

For Subject 2, statistically significant changes in muscle COM acceleration potentials were limited primarily to the horizontal (fore-aft) direction but some significant differences appear to exist for vertical acceleration potentials as well (Figure 5.7). The use of AFOs reduced the fore-aft potential for the majority of muscles with the exception of most proximal muscles: the gluteus maximus, gluteus medius, and psoas. For vertical COM accelerations, muscle potentials were significantly reduced for the distal gastrocnemius and tibialis anterior, similar to the trend observed for Subject 3.

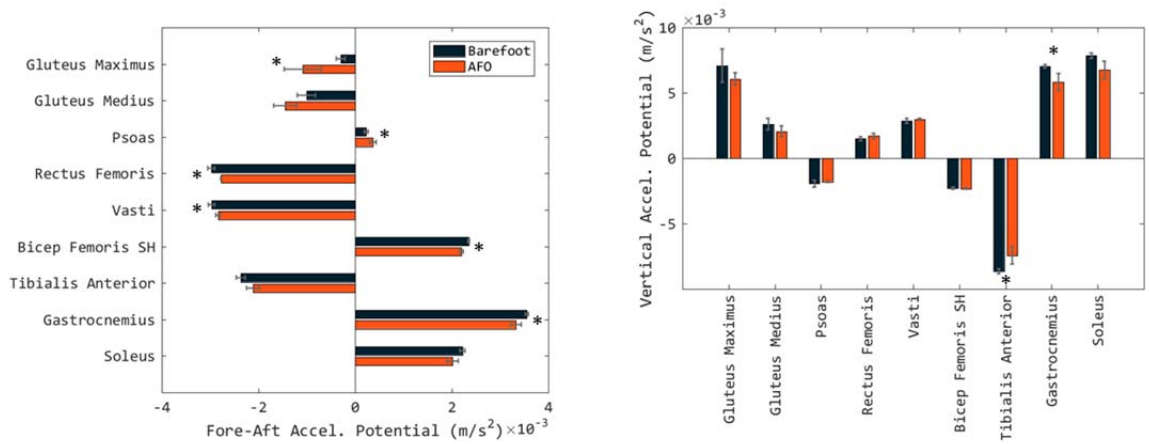


**Figure 5.7 – Muscle COM potentials for Barefoot and AFO walking for Subject 2. The muscle potentials for each trial were averaged for the entire stance phase and then individual trials were averaged.**

The muscle potential changes for Subject 1 were that AFO use increased the fore-aft potential of the distal muscles and increased the vertical potential of the proximal muscles (Appendix Figure A5). For Subject 4, AFO use reduced nearly all muscle vertical acceleration potentials and didn't meaningfully change fore-aft potentials (Appendix Figure A6).

Consistent with the literature, manual muscle testing (MMT) showed that the distal ankle plantarflexors were consistently the weakest muscle and that proximal muscle groups (hip flexor and knee extensor) were the strongest for the individuals in the study [113].

Subject 3 showed only MMT deficits for knee flexors and ankle plantarflexors while Subject 2 had additional deficits at the hip extensors and ankle dorsiflexors (Table 5.2).

**Table 5.2 – Manual Muscle Testing Results**

| Sub # | Hip Flexion | Hip Extension | Knee Flexion | Knee Extension | Ankle Plantar | Ankle Dorsiflexion |
|---|---|---|---|---|---|---|
| 1 | 4 | 4- | 3- | 4+ | 2+ | 4- |
| 2 | 5 | 4- | 4 | 5 | 2+ | 4 |
| 3 | 5 | 5 | 4- | 5 | 2+ | 5 |
| 4 | 5 | 2+ | 4- | 5 | 1 | 3+ |

## 5.4 Discussion

This simulation study investigated the influence of AFOs on lower limb muscle function. We created musculoskeletal simulations for a group of 4 study participants with diplegic CP and used IAA to investigate changes in muscle supportive and propulsive functions between barefoot walking and walking with AFOs. An analysis of the data revealed that AFOs can alter both the total COM acceleration and the COM acceleration potential of a muscle.

Principally, we found that for some individuals, the COM acceleration provided by the distal muscles was reduced with AFO use and that the COM acceleration provided by the higher functioning proximal muscles, such as the gluteus maximus and medius, was increased. MMT identified the plantarflexors as weak and showed that the proximal muscle groups were less affected by weakness. This may be a selective “strategic shift” by each individual to move the burden of COM acceleration from the more impaired distal muscles to the less affected proximal muscle groups.

It appears that the total COM acceleration provided by a muscle changes the most when AFO use is accompanied by a shift in lower limb kinematics. Previous work has shown that a kinematic shift towards a crouch gait (e.g. excessively flexed knees and ankles) maximizes the ground reaction forces that can be generated by muscles [114]. Biomechanically, kinematic changes influence muscle function by modifying the orientation of the force vector that is generated by a limb. A similar compensation mechanism appears to be plausible when wearing an AFO. By stabilizing the ankle, AFO use appears to allow an individual to position themselves in a more advantageous kinematic position which allows them to better compensate for their less functional distal muscles with more functional, proximal muscles. However, as previously mentioned, this kinematic shift was not present for all individuals and it remains unclear why there is a heterogeneous response to AFO use.

Substantial kinematic or total COM acceleration changes were not observed for Subject 2 when wearing an AFO. Interestingly, the muscle potentials did change, albeit primarily in the fore-aft direction. It is difficult to understand if the small changes in potential reflect any meaningful benefit, but they may help explain why AFOs appear to improve gait for some individuals and not others. As noted, most of the potential changes were in the fore-aft direction and only limited statistically significant changes were observed in the vertical direction. Perhaps Subject 2 had sufficient muscle capacity from the plantarflexors to support the COM and did not need other muscles to compensate. Additionally, Subject 2 had the most flexible AFOs of all the individuals in the study. Perhaps, the AFOs were not the built to a stiffness or neutral angle that would allow the individual to modify their gait enough to better utilize the more functional proximal muscles, similar to Subject 3.

There were a few limitations to the study. The first limitation is that the sample size was small. With the wide heterogeneity of responses to an AFO observed in the literature, we did not expect these four individuals in the study to be representative of the entire CP population [6], [7]. Our primary goal was to investigate if lower limb muscle function *could* be influenced by the use of AFOs. To develop generalizations about how AFO use typically influences muscle COM accelerations, a much larger sample would need to be evaluated. The second limitation is that we do not know how accurately the muscle activations in the simulation correspond to real muscle activations. We assume that the simulated muscle activations are reasonable representations of measured muscle activations (experimental EMG was used to restrict the on/off periods for the simulated muscles), but non-monitored muscle groups could be generating irregular forces in the simulation (e.g. gluteus maximus was not constrained by experimental EMG). However, the set of muscles used to constrain simulated EMG is consistent with previous IAA work, and as our goal was only to provide a basic level of biomechanical understanding of AFO use we do not expect that irregularities in the non-monitored muscle groups would impact the conclusions drawn from the IAA. A final limitation of the study is that it is impossible to separate the influence of shoes from the AFO without using a "shoes only" control condition. The experimental data for this study was collected during a standard clinical visit to the CGMA, and therefore barefoot walking and walking with an

AFO and shoes were the only two conditions collected. Future work should focus on isolating the confounding influence of shoes on muscle COM accelerations.

This study showed that AFOs can change muscle function between barefoot walking and walking with an AFO. Through simulation work, it appears that AFOs have the potential to change muscle function by shifting COM accelerations used to move the body from weaker, distal plantarflexors, to stronger, proximal muscle groups.

Future work should focus on analyzing muscle function for a larger sample of individuals to be able to generalize about how an AFO affects COM accelerations for the CP population as a whole. Additionally, future work investigating how different AFO designs affect muscle function is needed.

# Chapter 6: Ground Reaction Ankle Foot Orthoses and Solid Ankle Foot Orthoses are Equivalent for the Correction of Crouch Gait in Children with Cerebral Palsy

## 6.1 Introduction

Crouch gait is characterized by excessive knee and ankle flexion during stance, and can lead to significant orthopaedic pathologies such as patella alta and fixed knee contractures [115]. Solid ankle foot orthoses (SAFO) and ground reaction ankle foot orthoses (GRAFO) (also called floor reaction orthoses) are often recommended to reduce excessive knee flexion (KF) for individuals with cerebral palsy (CP) who exhibit a crouch gait [5], [73]. Understanding the differences in effectiveness of SAFO and GRAFO will lead to improved orthosis prescription for individuals with a crouch gait.

Both SAFO and GRAFO are specific types of ankle foot orthoses (AFOs). Functionally, SAFO and GRAFO are similar in that they both apply a corrective internal plantarflexion moment to the ankle in response to dorsiflexion of the ankle. However, this function is achieved via slightly different mechanical designs. The SAFO uses a posterior shell with an anterior strap that engages the tibia to prevent dorsiflexion, while the GRAFO utilizes an anterior tibial shell as the means to prevent dorsiflexion.

The mechanism which AFOs utilize to correct crouch gait has been termed the *plantarflexion/knee extension couple* (PF/KE couple). The PF/KE couple works through simple physics. Essentially, for any rigid object, if the ground reaction force (GRF) vector and weight vector are offset from one another, a net moment is induced on the object. This net moment either causes the object to rotate or generate a reaction force to balance the net moment. For AFOs, when body weight is applied to the ground, the GRF vector is often offset anterior to the body weight vector. This induces a net moment which causes the AFO and tibia to drive the knee in a posterior direction, and thereby, extend the knee.

A direct comparison of the effectiveness of the SAFO and GRAFO to reduce knee flexion in crouch gait has only recently been performed [116]. The comparison concluded that GRAFOs promoted a larger reduction of excessive knee flexion during stance, with a mean reduction of 8.9°, and that the SAFO was an inferior design for reducing knee flexion, with a mean reduction of 4.2°. These results struck us as somewhat surprising, given the mechanical equivalence of the two designs. It was expected that both designs would improve knee flexion, but we were intrigued as to why individuals would respond better to the GRAFO compared to the SAFO. Essentially, if the stiffness of the ankle and the AFOs neutral angle (the angle at which the AFO holds the ankle when no external forces are applied) is the same between two AFO designs, the whether the tibia is restricted from a strap or an anterior shell should have no effect on AFO function (Figure 6.1). In other words, the ankle stiffness and neutral angle should determine the performance of the AFO, not differences in design for how the tibia is restricted.

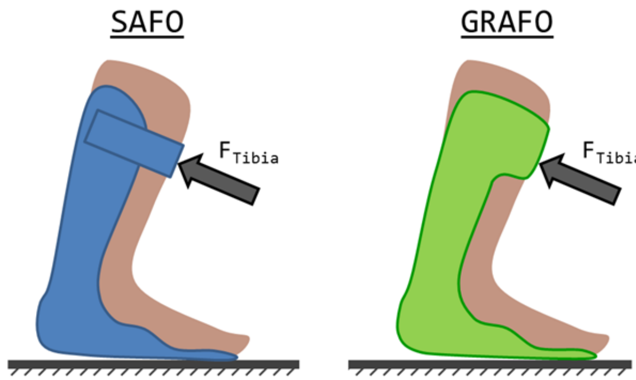


**Figure 6.1 - Function of SAFO and GRAFO designs. Mechanically, the total restrictive force on the tibia by the AFO should be the same between the two designs.**

One possible explanation for the differences in performance is that the AFO neutral angle is systematically different between the two designs. Hypothetically, an AFO with a more plantarflexed neutral angle will induce more knee extension. For a dorsiflexed ankle, a more plantarflexed AFO will provide a larger corrective plantarflexion moment and will work to drive the knee into more extension compared to the same AFO set in a more dorsiflexion position. We suspect that the GRAFO is generally set in a more plantarflexed position compared to the SAFO, which leads to the appearance of different effectiveness for the two designs.

Our study was performed to independently assess the effectiveness of two common orthoses designs for reducing excessive knee flexion in crouch gait using three dimensional motion capture data. We hypothesized that there will be measurable differences in performance between the two designs at our center, but that any differences in performance will be explained by factors other than the AFO design, such as neutral angle or stiffness.

## 6.2 Methods

A search of our center's clinical database was conducted to identify a cross sectional sample of children diagnosed with diplegic CP, who had a bilateral prescription of either a GRAFO or SAFO, and had a crouch gait pattern for at least one limb. Crouch gait was defined as excessive KF coupled with excessive dorsiflexion in stance for a limb (*i.e.* true crouch). As such, the objective criteria used to identify crouch gait was minimum KF in stance angle greater than 14.5° (more than 2 SD from normal) and maximum ankle dorsiflexion in stance angle greater than 7.3° (more than 1 SD from normal) [73].

Study inclusion criteria included:

- Diagnosis of Diplegic CP
- Prescription of bilateral SAFO or bilateral GRAFO
- Crouch gait (analyzed on a per limb basis)
    - KF in stance > 14.5°
    - Dorsiflexion in stance > 7.3°
- Gait data collected walking barefoot and walking in AFOs

For each study participant, three dimensional motion capture data was collected for two conditions: walking barefoot and walking with their clinically prescribed AFOs (and athletic shoes). If only one limb of an individual met all the inclusion criteria (e.g. one limb met the objective criteria for crouch gait but the other did not), only that limb was used in the analysis; if both limbs met the inclusion criteria, both limbs were included.

Kinematic angles included in the analysis (KF and ankle dorsiflexion) were quantified using a standard motion capture model (Vicon Plug-in Gait).

The average change in knee flexion ($\Delta$KF) between walking barefoot and with an AFO for each orthosis design was examined using an independent samples t-test. Negative $\Delta$KF values indicate a reduction in knee flexion with AFO use, which is the desired direction of change. Possible explanatory variables included in the analysis were: age, GMFCS [117], FAQ (level of function) [103], mass, minimum barefoot KF in stance (i.e. level of crouch), AFO neutral angle (estimated by using the mean ankle angle in swing when wearing the orthosis (Figure 6.2)), AFO range of motion (ROM) during stance (surrogate for AFO stiffness), max hip extension by physical exam (i.e. evidence of hip flexion contracture), and maximum knee extension by physical exam (i.e. evidence of KF contracture). Comparison of the possible explanatory variables between SAFO and GRAFO groups was performed using independent samples t-test. The influence of possible explanatory variables on $\Delta$KF was examined using stepwise linear regression, and included the additional possible predictive variable of AFO design (SAFO vs. GRAFO) to determine if the AFO design was, in fact, a significant influence on AFO performance. Statistical significance was set at $\alpha = .05$. All statistics were performed using the Matlab Statistics Toolbox (Mathworks, Natick, MA).

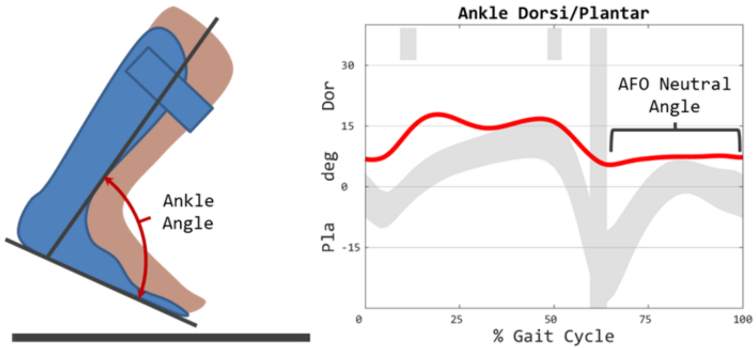


**Figure 6.2 – AFO neutral angle estimation. AFO neutral angles were estimated by taking the average ankle angle measured during swing when walking with their AFO. Significant forces typically aren't present during swing so the AFO will be in its neutral position.**

## 6.3 Results

The database search returned data from 153 limbs (from 96 individuals) wearing a SAFO and 58 limbs (from 32 individuals) wearing a GRAFO who exhibited a crouch gait pattern according to our objective crouch criterion (Table 6.1).

**Table 6.1 – Group Characteristics**

| | SAFO | GRAFO | p-value |
|---|---|---|---|
| **N** | 153 | 57 | - |
| **ΔKF (deg)** | -4.7 (7.2) | -7.5 (7.0) | **.02** |
| **Age (yrs)** | 10.9 (3.8) | 12.9 (4.9) | **.002** |
| **GMFCS†** | 2.5 (0.7) | 2.5 (0.7) | .92 |
| **FAQ‡** | 7.8 (1.4) | 7.6 (1.8) | .35 |
| **Mass (kg)** | 37.8 (16.5) | 45.1 (16.3) | **.005** |
| **Min KF in Stance (deg)** | 26.4 (10.6) | 32.0 (11.0) | **<.001** |
| **AFO Neutral Angle (deg)** | 7.6 (5.9) | 4.1 (7.3) | **<.001** |
| **AFO ROM in Stance (deg)** | 11.1 (4.5) | 8.9 (3.4) | **<.001** |
| **Hip Flex Contracture by PE (deg)** | 2.4 (12.8) | 4.1 (10.2) | .45 |
| **Knee Flex Contracture by PE (deg)** | 3.3 (8.2) | 5.4 (9.5) | .12 |

Values are Mean (±SD); Negative ΔKF reflect an improvement in KF
p-values were determined using 2 sample t-tests
† = GMFCS was not assigned for 44% of SAFO and 66% of GRAFO limbs
‡ = FAQ was not assigned for 9% of SAFO and 9% of GRAFO limbs

T-tests revealed significant differences in AFO performance for improving ∆KF between the SAFO (-4.7°) and GRAFO (-7.8°) groups (Table 6.1). Significant group differences for other measures were also identified. In summary, the SAFO group was younger, lighter, had less knee flexion during stance, had more dorsiflexed AFOs, and had greater AFO ROM during stance compared to the GRAFO group. No significant differences between groups were detected for function, as quantified by the GMFCS and FAQ, or contractures at the hip and knee, as measured by physical exam.

The stepwise regression identified AFO neutral angle, minimum barefoot KF in stance (i.e. severity of crouch), and level of knee flexion contracture as significant predictors for ΔKF (Table 6.2). AFO design was not a significant predictor for improvement in ΔKF after accounting for the other predictors (p = .49).

**Table 6.2 – Stepwise Linear Regression Results**

| | Coefficient | Standard Error | p-value | Effect Size (adjusted) |
|---|---|---|---|---|
| **Intercept** | -4.52 | 1.52 | .003 | - |
| **Neutral Angle (deg)** | 0.32 | 0.07 | <.001 | 13.4 |
| **Min KF in Stance (deg)** | -0.15 | 0.05 | .004 | -9.6 |
| **Knee Flexion Contracture (deg)** | 0.26 | 0.07 | <.001 | 16.7 |

$r^2 = .16$

## 6.4 Discussion

The purpose of this study was to evaluate differences in performance between SAFO and GRAFO designs for improving excessive knee flexion in crouch gait. Previous studies have shown a performance gap between the two AFO designs, with the GRAFO outperforming the SAFO [116]. We found similar differences in AFO performance at our center. However, after further examination of the data, we found that the AFO design itself does not explain the performance difference, but rather it is the AFO neutral angle, and its systematic difference between designs, that primarily determines change in knee flexion with an AFO. This confirms our study hypothesis. The GRAFO is, on average, set in 3.5° more plantarflexion compared to the SAFO which is why the GRAFO induces 2.8° more knee extension compared to the SAFO. The more plantarflexed position allows the GRAFO to utilize the plantarflexion/knee extension mechanism to extend the knee to a greater degree.

Other factors were also shown to contribute to the difference in performance between AFO designs. The level of crouch was positively associated with an improvement in ΔKF outcomes (i.e. min KF in stance has a negative coefficient and negative changes in ΔKF represent improvement). This means that the deeper in crouch an individual is, the larger the expected improvement will be. A similar effect response has been seen for a wide array of gait variables in children with CP [23], [118]. Presence of a significant knee flexion contracture, as would be expected, had a negative impact on ΔKF (i.e. the larger the knee flexion contracture, the smaller the expected improvement from an AFO).

It is not clear why the GRAFO is typically set in more plantarflexion than the SAFO when the goal for both designs is improving excessive knee flexion. The primary purpose of the GRAFO is to correct excessive knee flexion, and GRAFOs are not typically recommended to correct other gait deviations [73]. We suspect that when an individual is prescribed a GRAFO, the orthotist making the GRAFO, whether aware or not, is more aggressively positioning the ankle angle towards plantarflexion to promote a greater improvement in knee extension. Our data shows that, on average, GRAFOs are prescribed for individuals with 5.6° deeper crouch compared to SAFOs. Conversely, when an individual is prescribed a SAFO, the goal of the SAFO may not be as clear as it is for the GRAFO, and the orthotist may be less aggressive at positioning the SAFO ankle. Just as misperceptions about the GRAFO being better for correcting crouch gait could have led to the GRAFO being more aggressively positioned, misperceptions about the SAFO being less effective for correcting crouch gait could have led to the ankle being less aggressively positioned. We suspect that a combination of these two subtle biases led to the apparent performance gap, and that this is a case of “self-fulfilling prophecy” where the false perception of the efficacy of the GRAFO vs. SAFO actually caused the differences in performance between the two designs.

Interestingly, according to a 2007 study, United States Medicare reimbursement for a GRAFO was nearly twice that of a SAFO ($739 vs. $417). We did not find a significant difference in performance specifically attributable to the GRAFO design. As a consequence, from the findings of this study, future studies need to provide evidence that there is a clear benefit to GRAFO use over the SAFO to preserve the GRAFO as a viable AFO design.

The primary limitation of this study was that AFO ankle stiffness was not directly quantified, and the ankle ROM during gait was used as a surrogate. Benchtop equipment, such as the bi-articular reciprocating universal compliance estimator (BRUCE), do exist for directly measuring an AFO's ankle stiffness [108]. However, for this study, only a small fraction of the individuals included in this study had the stiffness their AFOs benchtop tested as the BRUCE device was only recently added to the clinical examination protocol. We suspect that a directly measured ankle stiffness value would substantially affect the predictive value of "ankle stiffness" in the stepwise regression model. But, improving the ankle stiffness value would not likely cause AFO design to suddenly become a significant influence on AFO performance and change the conclusions of this study.

This study shows evidence that a simple mechanical analysis can be applied to debunk the misconception that the GRAFO outperforms the SAFO for the correction of crouch gait for individuals with CP. We have shown that the GRAFO design is not superior to the SAFO design. Therefore, a valuable avenue for future research would be to determine optimal AFO design parameters (e.g. stiffness and neutral angle) for augmenting knee extension.

# Chapter 7: General Discussion

Ankle foot orthoses (AFOs) are often recommended for individuals with cerebral palsy (CP) as a means to improve gait. Typically, the goals of an AFO prescription are to enhance gait quality, improve overall function, improve energy economy, correct positional pathologies, and prevent contractures. This thesis approached AFO optimization in three ways. First, I evaluated the current efficacy of AFO use in children with CP (Chapter 2), next, I described new methods for analyzing and improving AFO outcomes as they pertain to gait (Chapters 3 and 4), and finally, I investigated biomechanical mechanisms related to the mechanisms by which AFOs influence gait (Chapters 5 and 6).

The result of these investigations can be summarized as follows: AFOs don't work as well as we think, and the current clinical prescription paradigm is not improving AFO efficacy. By utilizing advanced machine learning techniques to prescribe AFOs and simulations to understand how AFOs work, the efficacy of AFOs can likely be improved.

## 7.1 Main Findings

### *1. Current AFO outcomes are mixed, at best*

In Chapter 2, data from a large cross sectional sample of children diagnosed with diplegic CP from a single center was used to investigate the current efficacy of AFO use. Changes in an individual's gait between walking barefoot and with their clinically prescribed AFO (SAFO, PLS, HAFO designs) were used to investigate the impact of an AFO on multiple gait related outcome measures.

The primary outcome measures evaluated for 378 individuals were: gait deviation index (GDI) – which is a single number that represents the overall deviation from a normal kinematic profile, ankle gait variable score (GVS) – which is a single number that represents the joint-specific deviation from a normal ankle joint motion profile, knee GVS – similar to the ankle GVS but for the Knee, walking speed, and step length.

The majority of outcome measures were found to show mixed results from AFO use. Mean improvements in GDI, ankle GVS, and knee GVS were all below clinically meaningful levels. However, speed and step length did improve by a clinically meaningful amount with AFO use. While speed and step length are good indicators of overall performance, they do not necessarily reflect the traditional goals of AFO prescription such as to correct positional pathologies of the foot and ankle (i.e. ankle GVS) and enhance overall gait quality (i.e. GDI).

While the average individual in the study population did not improve for GDI, ankle GVS, and knee GVS, the results did show that a small number of individuals at the extremes of the response distribution experienced improvements beyond the clinical threshold (conversely, a substantial number of individuals experienced a decline in performance beyond the clinical threshold as well). These results suggest that an immediate way to improve the efficacy of AFO use overall, would be to prospectively identify the individuals that would have a “good” response to AFO use, and not prescribe an AFO for individuals that would have a “poor” response to AFO use. We believe that the model proposed in Chapter 3 has the potential to fulfill this strategy.

***2. Advanced data driven prescription models are likely to improve AFO outcomes***

In Chapter 3, an orthosis prescription algorithm was developed to recommend the optimal orthosis design that would maximally improve the gait for a child diagnosed with diplegic CP. The algorithm selected an orthosis design from pool of 5 common designs (3 AFO designs, 2 non-AFO orthoses designs), and included the option of that the patient not wear an orthoses if none of the designs were predicted to provide an advantage over barefoot walking. The potential level of benefit to the diplegic CP population was estimated under the assumption that the algorithm’s recommendations were followed.

The singular goal of the orthosis prescription algorithm (unlike the multiple goals of clinical care) was to improve GDI. Development of the orthosis prescription algorithm was achieved using a relatively new yet already widely used statistical machine learning technique called the random forest algorithm (RFA). Essentially, the RFA utilizes a set of classification or regression trees built from randomly selected features (e.g. measured values) to predict outcomes (e.g. change in GDI) for a set of observations (e.g. limbs).

The final orthosis prescription model was accurate at predicting whether or not a limb would receive at least a +3 GDI point improvement from a specific orthosis design. Prediction accuracy for each orthosis design (i.e. from each prediction ensemble) ranged from 67% for the SAFO to 82% for the foot orthosis (an orthosis that supports only the foot). Due to the underlying nature of the RFA it is difficult to discern “how” each ensemble used the predictive variables to estimate the orthosis response. Nevertheless, the ensembles can shed light onto “what” variables are important, or at least what variables are surrogates of important qualities that determine how an orthosis will affect GDI. In this case, the RFA demonstrated that changes in performance can be reasonably estimated using only a few predictive variables (between 4 and 9) without the need to fabricate and test an actual orthosis designs on an individual, which would be expensive and impractical.

The majority of limbs analyzed by the model (63%) were not in their optimal orthosis prescription as recommended by the model. By far, the vast majority of limbs that were not currently in the recommended optimal orthosis design either had an orthosis but did not need one (45%), or needed an orthosis but did not have one (37%). The remaining 18% of limbs currently had an orthosis and were simply recommended a different orthosis design. As a consolation, orthotists and those concerned about the business of orthosis prescription and fabrication, should be relieved that the number of orthoses recommended for individuals remained in-line with current levels: the model recommended that 842 limbs have an orthosis, which was only slightly lower than the current prescription methodology which resulted in 952 limbs having an orthosis prescription (-12% relative change).

Interestingly, 90% of the limbs that were recommended an orthosis were recommended one of the 3 AFO designs (i.e. SAFO, PLS, HAFO). It appears that while the SMO and FO designs may provide other benefits to an individual such as rising from the floor or climbing stairs, it appears that if the goal is to improve GDI, an AFO design will almost always outperform the non-AFO (foot support only) designs.

The estimated potential clinical benefit to the CP population from the model was high. For over 2000 limbs analyzed, the average GDI change for a limb currently prescribed an orthosis rose from +0.4 GDI points to +5.6 GDI points if the model recommendations were followed. This is impactful as the overall change of +5.2 GDI points is above the accepted clinically relevant +5 GDI point threshold. This indicates that there can be a clinically significant impact on AFO efficacy if we simply *put the right orthoses on the right limbs and don't put the wrong orthoses on the wrong limbs*.

### ***3. An analysis of gait efficiency may identify individuals who can benefit from an AFO***

In Chapter 4, mechanical work and walking efficiency were analyzed for a large group of children diagnosed with diplegic CP in order to better understand the source of excessive metabolic demands during walking, typically observed for these individuals. The study also investigated different strategies in work production and absorption that may help to explain why AFOs often reduce metabolic demand.

The primary goal of this study was to investigate gait efficiency for children with diplegic CP. Gait efficiency, in its most basic sense, was defined as the external mechanical work rate divided by the net walking metabolic work rate.

Gait efficiency was found to be significantly lower for children with diplegic CP when compared to typically developing (TD) age matched peers. The gross external mechanical work rate for the CP group was measured to be 27% higher compared to the TD group. However, the metabolic work rate was 97% higher in the CP group compared to the TD group. In general, it can be said that although individuals with CP perform extra work during gait, the conspicuous inability to efficiently convert metabolic energy into useful external mechanical work is the major factor which leads to high metabolic work rates.

One of the mechanisms that may be responsible for low walking efficiency is a lack of propulsion work. Overall, an analysis of the work production during a step identified that children with diplegic CP generally have a significant propulsion work deficit, and do most of their positive step work during rebound in single stance. This is opposite to what occurs for TD individuals, where most of the positive work occurs during propulsion. This lack of propulsion work may limit potential energy saving mechanisms, such as energy storage and return by the plantarflexors and Achilles tendon that occurs during propulsion.

As is common in CP, there was a substantial amount of heterogeneity in efficiency values for the CP group. This heterogeneity may influence the efficacy of specific treatments for reducing metabolic demands, a major goal of intervention. Many individuals had a high metabolic work rate and low walking efficiency. It may be possible that, for individuals such as these with a low gait efficiency, interventions that target a reduction in external mechanical work rate, such as AFOs, may not be as effective at reducing metabolic demands as a spasticity reduction intervention. This is because the metabolic work rate for individuals with a low gait efficiency is primarily exacerbated by factors other than external mechanical work (e.g. “workless work” from co-contractions/spasticity). However, many individuals were observed to have a high metabolic work rate and high efficiency, comparable to the TD group. For individuals such as these with gait efficiency near TD levels, interventions that target a reduction in work, such as an AFO, may be a more effective intervention, as the higher metabolic work rate is primarily exacerbated by increases in external mechanical work rate.

***4. The supportive and propulsive functions of muscles are influenced by AFOs***

In Chapter 5, a musculoskeletal simulation was used to investigate changes in muscle function for individuals who have an observed qualitative improvement in gait when wearing their AFOs. The study used induced acceleration analysis (IAA) to determine changes in muscle function on center of mass (COM) accelerations caused by AFO use.

The primary goal of this study was to use musculoskeletal simulation to investigate the supportive and propulsive functions of muscles, and to see if those functions can change when wearing an AFO. Essentially, the study aimed to investigate a possible biomechanical mechanism that an individual use to improve their gait when wearing an AFO.

Two musculoskeletal models were created for each individual in the analysis: one model to simulate walking barefoot, and another to simulate walking with AFOs. A simulation was created for each model using motion capture data (collected for both walking barefoot and walking with AFOs conditions) and the influence of muscles on COM accelerations were calculated using IAA. An IAA is, in essence, a perturbation analysis in which the influence of muscles on COM acceleration can be elicited by modulating the force produced by a muscle and then evaluating the corresponding changes in body accelerations. Muscle function differences between the two conditions were used to understand the influence of an AFO on muscle function.

From the IAA, some individuals tended to shift supportive and propulsive functions from distal muscles (e.g. plantarflexors) to more proximal muscle groups (e.g. gluteals). Additionally, this shift tended to be associated with a change in kinematic position. The shift of function to more proximal muscles make sense, since distal muscle groups tend to be more severely impaired in CP. By shifting to more functional proximal muscles, individuals may be better able to control COM accelerations. While this kinematic and muscle function change was not observed for all individuals, it nevertheless provides a biomechanical basis for why some individuals benefit from AFO use while others do not.

***5. Some AFO designs are redundant***

In Chapter 6, we investigated performance differences between two common AFO designs that are thought of clinically as being distinct, but mechanically have identical functions. The study used stepwise linear regression to identify significant factors that contributed to the performance gap between solid and ground reaction AFO designs for the correction of crouch gait for individuals diagnosed with CP.

The primary goal of this study was to investigate differences in performance between the solid ankle foot orthosis (SAFO) and ground reaction ankle foot orthosis (GRAFO) for reducing excessive knee flexion (KF) for individuals with a crouch gait, and to investigate possible factors that could explain any performance gap. Possible predictive factors were: age, functional ability as measured by the FAQ [103], mass, minimum barefoot KF in stance (i.e. level of crouch), AFO design (i.e. SAFO or GRAFO), AFO neutral angle (i.e. the angle at which the AFO holds the ankle when no external forces are applied), AFO range of motion (i.e. surrogate for AFO ankle stiffness), maximum hip extension by physical exam (i.e. evidence of hip flexion contracture), and maximum knee extension by physical exam (i.e. evidence of knee flexion contracture).

Using a large cross-sectional sample of individuals, a significant performance gap between the two AFO designs was found. The SAFO corrected excessive knee flexion by 4.7°, on average, while the GRAFO outperformed the SAFO, correcting excessive knee flexion by 7.5°, on average. These results were similar to those reported in a previously published abstract [116].

Using a stepwise linear regression, AFO design (i.e. SAFO vs. GRAFO) was found to *not* be a significant factor on changes in KF from an AFO. Instead, AFO neutral angle, minimum KF in stance, and maximum knee extension by physical exam were identified as significant factors for predicting changes in KF from an AFO. The GRAFOs were found to be fabricated in a more plantarflexed position compared to the SAFOs. This led the GRAFOs to *appear* to be more effective at reducing KF for crouch gait, although the design itself had no influence on performance. The precise reasons were not clear as to why the GRAFO was typically set in more plantarflexion compared to the SAFO. However, a possible reason may be that when a GRAFO is prescribed, it is clear that the objective of the prescription is to correct excessive KF. The orthotist fabricating the GRAFO understands that a more plantarflexed ankle will be better at correcting excessive KF, and aggressively positions the GRAFO ankle in more plantarflexion. The objective of the SAFO prescription may be less clear, or multi-faceted, which may lead the SAFO to be less aggressively plantarflexed.

This study identified the GRAFO design as being redundant for improving KF in crouch gait compared with the SAFO, as it was the AFO neutral angle, and not AFO design, which primarily influenced performance. Interestingly, as reported in a 2007 study, the United States Medicare reimbursement rate for a GRAFO was nearly twice that of a SAFO ($739 vs. $417). Therefore, future studies need to provide evidence that there is a clear benefit to GRAFO use over the SAFO design to be able to preserve the GRAFO as a viable AFO design alternative to the less expensive SAFO.

### *Conclusions*

Each study described in this thesis contributes to improving the efficacy of AFO use. I believe that the development of the data driven orthosis prescription model, described in Chapter 3, has the largest potential to impact the current state of AFO efficacy. In light of the current worldwide push to harness the power of Big Data, it would not be surprising to see advance data driven prescription algorithms transform medicine over the next decade. Advanced models, similar to the one described in Chapter 3, have the potential to significantly increase the efficiency of medical practice, saving both time and resources. Instead of using a clinical expert to analyze and review ever increasing amounts of complex data, advanced data driven models would exploit trends in data to provide precise, patient specific, guidance without the need for expert interpretation. I envision these advanced models to be used as companion guides to clinical experience, informing and guiding the clinician during the decision making process.

## 7.2 Methodological Considerations

This thesis used standard statistical techniques, advanced machine learning methods, and musculoskeletal simulation to better understand how AFOs influence gait and how to improve AFO efficacy. Both retrospective and prospective study designs were used.

### *Selection Bias*

Most of the data used in this thesis was drawn from a historical database composed of gait data collected from patients seen at the Center for Gait and Motion Analysis (CGMA) at Gillette Children's Specialty Healthcare in Saint Paul, MN. The database was beneficial to performing the research work in this thesis, since it allowed for access to large study samples without the need to fund costly prospective studies. Additionally, the availability of the large dataset allowed for the studies in this thesis to have significantly higher sample sizes compared to similar works. However, as the database was constructed using patient data, a potential for selection bias in the data exists. First, the individuals seen at the CGMA vary widely in scope and level of ability. However, some groups within the CP population may not be seen as frequently as others in the CGMA.

For example, individuals that are not being considered for an intervention (e.g. mild levels of impairment) or individuals that are not being seen for longitudinal tracking (e.g. stable, manageable conditions) are underrepresented in the historical data. While these groups may be undersampled with respect to the CP population as a whole, differences referral patterns between clinicians to the CGMA likely provides a reasonable sampling of these groups. Second, if there were any bias in the data, the broad inclusion criteria used for each study design would work to limit the influence of any inherent selection bias. This means that the individuals seen in the CGMA and included in each retrospective analysis very likely comprise a high quality representative “as treated” cross-sectional sample of the individuals that are prescribed an AFO in the ambulatory CP population as a whole.

### *Confounding Influences*

Precautions were taken to minimize the effect of confounding influences in the studies presented in this thesis. Primarily, when changes due to an AFO were used as the outcome measure, individuals were used as their own control, similar to a matched study design. In these types of studies, the principal confounding influence unaccounted for was the effect of shoes on gait. As a standard clinical practice, individuals at the CGMA are tested walking barefoot. Optionally, if an individual is to have additional tests in their AFOs, they must wear shoes with their AFOs. Testing an individual walking barefoot is advantageous from a clinical perspective as data for subsequent visits can be compared to the same baseline barefoot condition without the need to account for shoes. However, from a research perspective, wearing an AFO with shoes adds a confounding influence on gait as the effect of shoes cannot be separated from the AFO retrospectively. This confounder can only be controlled for by using a *shoes only* condition, which would only be possible by using prospectively collected data. While the effect of the shoe is not directly quantifiable, the majority of the individuals seen at the CGMA wear an athletic type shoe which should have a reasonably fixed effect on gait. Furthermore, the shoes plus AFO condition is the standard condition reported in the literature for AFO outcome studies. While not optimal, the inclusion of the shoe confounder, at a minimum, maintains the status quo for outcome reporting.

When potential confounders could not be controlled for through study design, data analysis techniques were used to account for confounders. In Chapter 6, the performance of a group of individuals wearing a SAFO was compared to the performance of a group of individuals wearing a GRAFO. In this case, individuals could not be matched and a significant performance gap was measured between the groups. A multivariate regression was therefore used to account for group differences. In this case, the regression subsequently identified the AFO neutral angle (a potential confounder) as the primary influence on AFO performance instead of AFO design.

### ***Information Bias***

Much of the effort put forth in this thesis was spent challenging typical AFO paradigms by showing the true level of data sophistication required to understand and improve AFO efficacy. Information bias is as a type of cognitive bias where it is believed that acquiring more information for a decision, whether irrelevant or not, improves the decision. From its inception, clinical gait analysis has strived to provide the clearest picture of impairment by using objective data aided by subjective assessments. New procedures, assessments, and surveys are constantly being added to the data collection scheme with the hope that this *new* data will improve outcomes. This study and many previous studies have come to the conclusion that outcomes from AFO use is mixed. However, despite all the new information added to the data collection scheme over the years to try and understand AFO performance, AFO prescription paradigms have remained largely unchanged, and remain focused on simple, unproven ad hoc approaches.

The objective of this thesis was to improve prescription paradigms through a more directed and efficient use of data (Chapters 3 and 4), and to provide a better understanding of how AFOs work (Chapters 5 and 6). This work has shown that data currently collected during routine gait analysis has the potential to improve outcomes without needing to collect any “new” information.

***Generalizability***

There are good reasons to expect that the results, conclusions, and methodologies presented in this thesis will be applicable to the general CP population. First, as previously mentioned, the data used in this thesis consists of individuals seen at the CGMA and vary widely in scope and level of ability. Second, broad inclusion criteria were used to select the study samples. These two factors provide confidence that the thesis projects are applicable to a broad section of the CP population.

## 7.3 Clinical Implications

The first step to improving AFO outcomes is to recognize that current prescription paradigms are not optimal (*Step 1 of every 12 Step program is admitting that there is a problem). This thesis has shown that AFOs do not generally improve the types of outcomes that are often sought from an AFO prescription, such as improvements in GDI or ankle function. Yet, prescription methodologies will likely be slow to change even after this evidence is presented. While intuition is an important factor in the current clinical decision making process, there are inherent bias that are difficult, if not impossible, to control, such as confirmation bias, recall bias, outcome bias, and selective perception. One bias that should be highlighted in light of the evidence presented in this thesis is optimism bias. This thesis has shown that AFO outcomes are mixed; for example, only a small percentage of individuals are expected to improve their GDI with an AFO using the current prescription methodology. However, optimism bias will affect many clinicians by causing them to ignore the low probability of a good outcome from an AFO and to think “this time is different – the AFO will improve gait”. Simply acknowledging the sub-optimality with the way AFOs are currently prescribed is the first step to improving AFO efficacy.

The second step to improving AFO outcomes is to embrace machine learning and statistical data driven processes. Understandably, it may be uncomfortable for many clinicians to trust a machine when making a medical decision. However, unlike current prescription paradigms, a data driven process, such as the optimal orthosis prescription algorithm developed in Chapter 3, is not in itself affected by cognitive biases. Data driven processes tell us what is actually happening instead of what is thought to be happening. While there are often drawbacks to using a data driven approach as opposed to a traditional approach (e.g. an intuitive explanation for a data driven approach may not be perceptible), the benefits of these data driven approaches often clearly outweigh their shortcomings. For example, a data driven model was recently used to optimize the triage procedure at a Level I trauma center [119]. The model was shown to be more accurate at triaging compared to existing procedures, but at a cost. The model's decision making criterion was not able to be precisely summarized so that it was easily understandable to clinicians. While the model's methodology was essentially masked to clinicians, improvement in triage accuracy was expected to lead to a more efficient use of the trauma system and result in both life and cost savings, clearly outweighing the masked decision making criteria drawback. Although this thesis indicates that the implementation of the optimal orthosis algorithm could improve the efficacy of AFOs by a clinically significant amount, these improvements will only be realized if clinicians have the patience and vision to rigorously tests algorithms like this in a proper prospective manner, and if validated, trust and follow the algorithm's recommendations.

The final step to improve AFO outcomes must be to understand that AFOs may not work the way they are often thought to work. As a simple example, this thesis shows that two common AFO designs that were traditionally thought of as having distinct functions, in fact, have exactly the same function. Traditionally, improvements in gait from an AFO are thought to be provided by the energy storage and return mechanism of the AFO as it flexes and extends. However, the amount of energy capable of being stored by an AFO is small in comparison to the total energy typically provided by the plantarflexors. This thesis showed that instead, AFOs can move center of mass acceleration control to the more functional proximal muscles and away from the more affected distal muscles for some individuals. This shift may explain why some individuals benefit from an AFO while others do not. Finally, this thesis showed that some individuals with CP have low gait efficiency compared to TD individuals, while others have gait efficiencies comparable to TD. This suggests that an AFO may affect individuals differently based on their gait efficiency. Only by exploring these new avenues of research will improvements in AFO efficacy be realized.

In summary, the work contained in this thesis demonstrates that current AFO prescription paradigms do not lead to consistent improvements in gait, but that both significant and meaningful improvements in AFO efficacy are possible for children with CP. By utilizing data driven approaches over existing ad hoc approaches significant improvements in AFO efficacy for children with CP can be realized.

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# Appendix

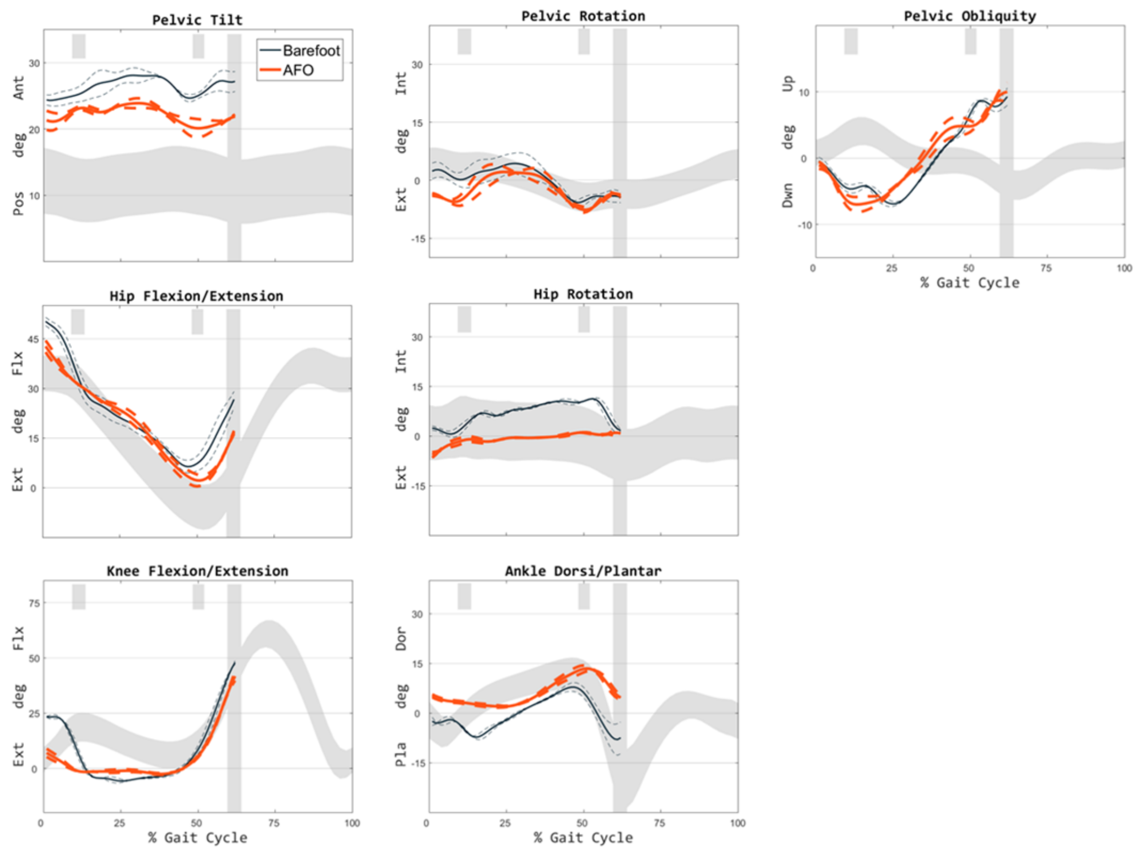


**Figure A1 – Kinematic plots for Barefoot and AFO walking simulations for Subject 1. The solid line is the mean of all trials collected and the dashed line is 1 standard deviation from the mean. Grey bands represent normal gait ± 1 SD, full scale vertical bands represent typical toe off, and short vertical bands at the top of each plot represent opposite toe off and opposite heel strike, respectively.**

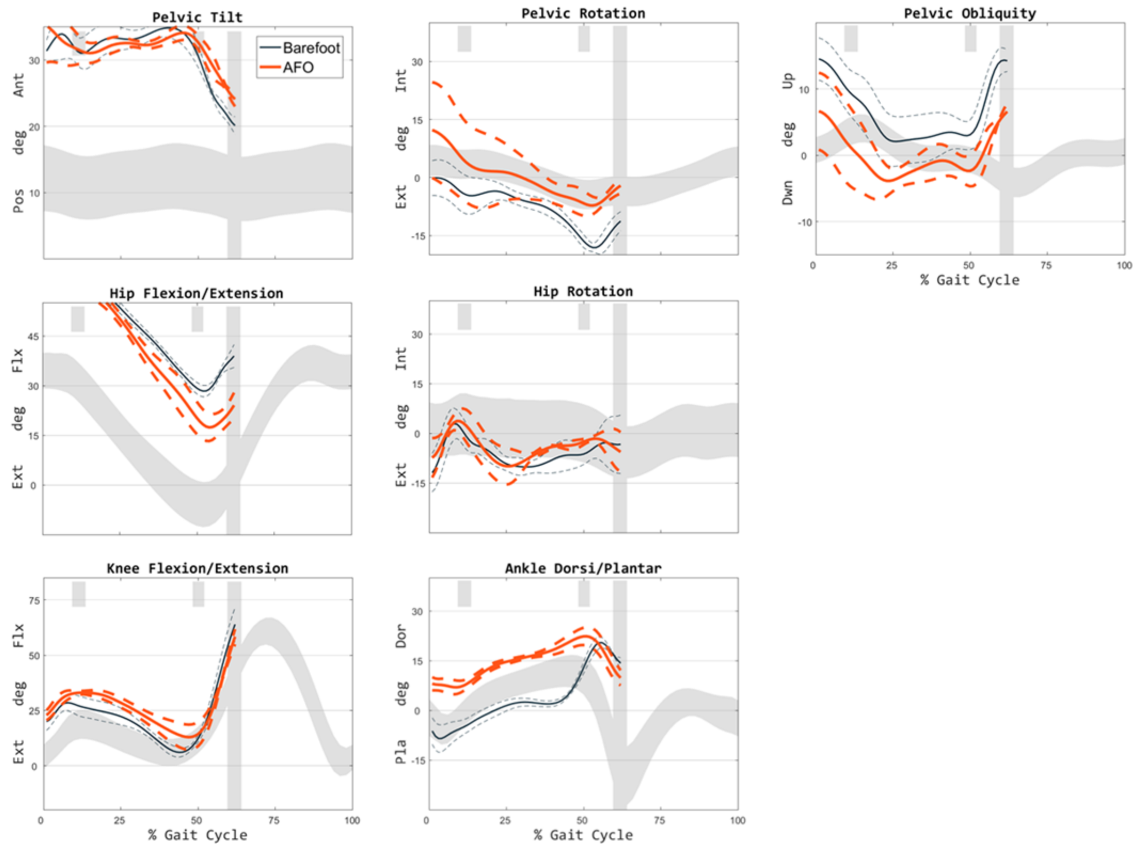


**Figure A2 – Kinematic plots for Barefoot and AFO walking simulations for Subject 4. The solid line is the mean of all trials collected and the dashed line is 1 standard deviation from the mean. Grey bands represent normal gait ± 1 SD, full scale vertical bands represent typical toe off, and short vertical bands at the top of each plot represent opposite toe off and opposite heel strike, respectively.**

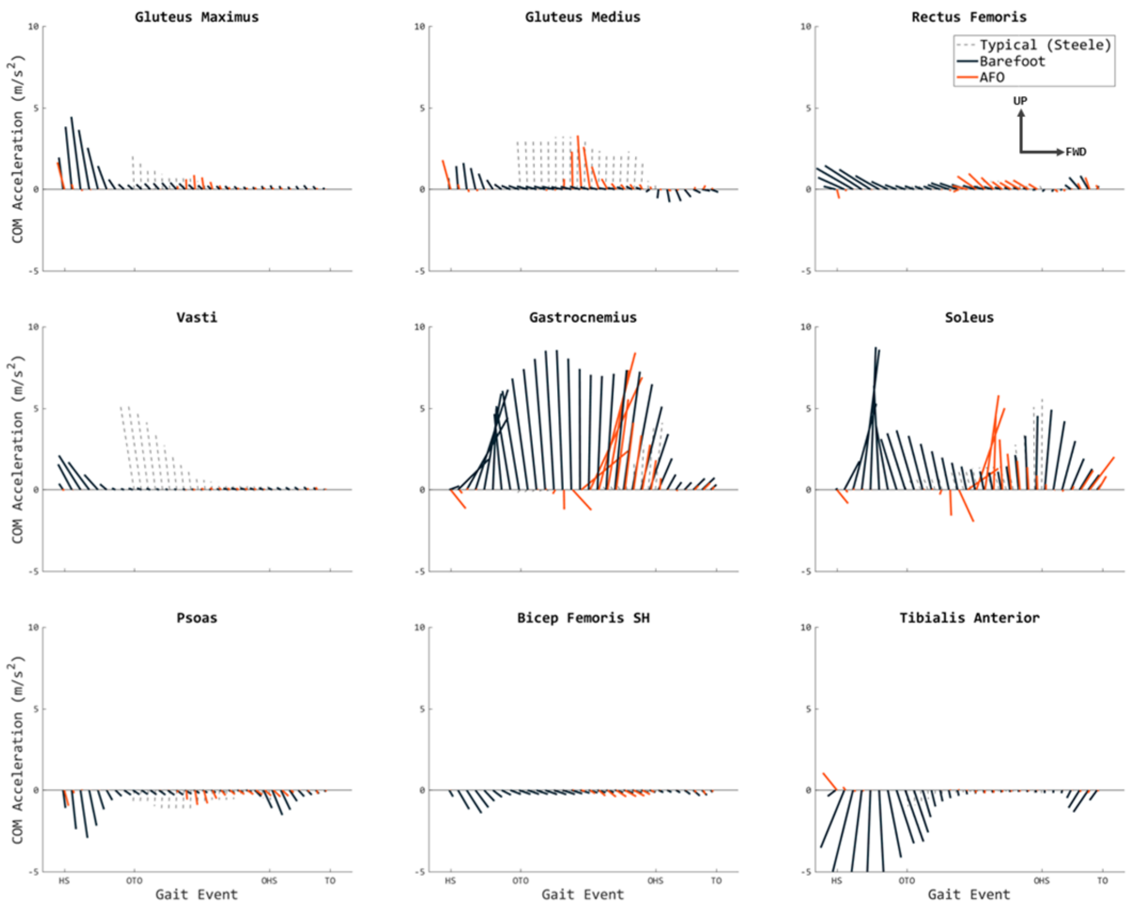


**Figure A3 – Total COM accelerations for Barefoot and AFO walking for Subject 1. The only muscle group with a noticeable change was the soleus. All other muscle groups experienced minimal changes. The vertical displacement of each vector from zero represents the vertical COM acceleration induced during stance while the horizontal displacement of the vector represents the fore-aft induced COM acceleration. Vectors are plotted at approximately each 3% of stance. Abbreviations: HS – heel strike, OTO – opposite toe off, OHS – opposite heel strike, TO – toe off.**

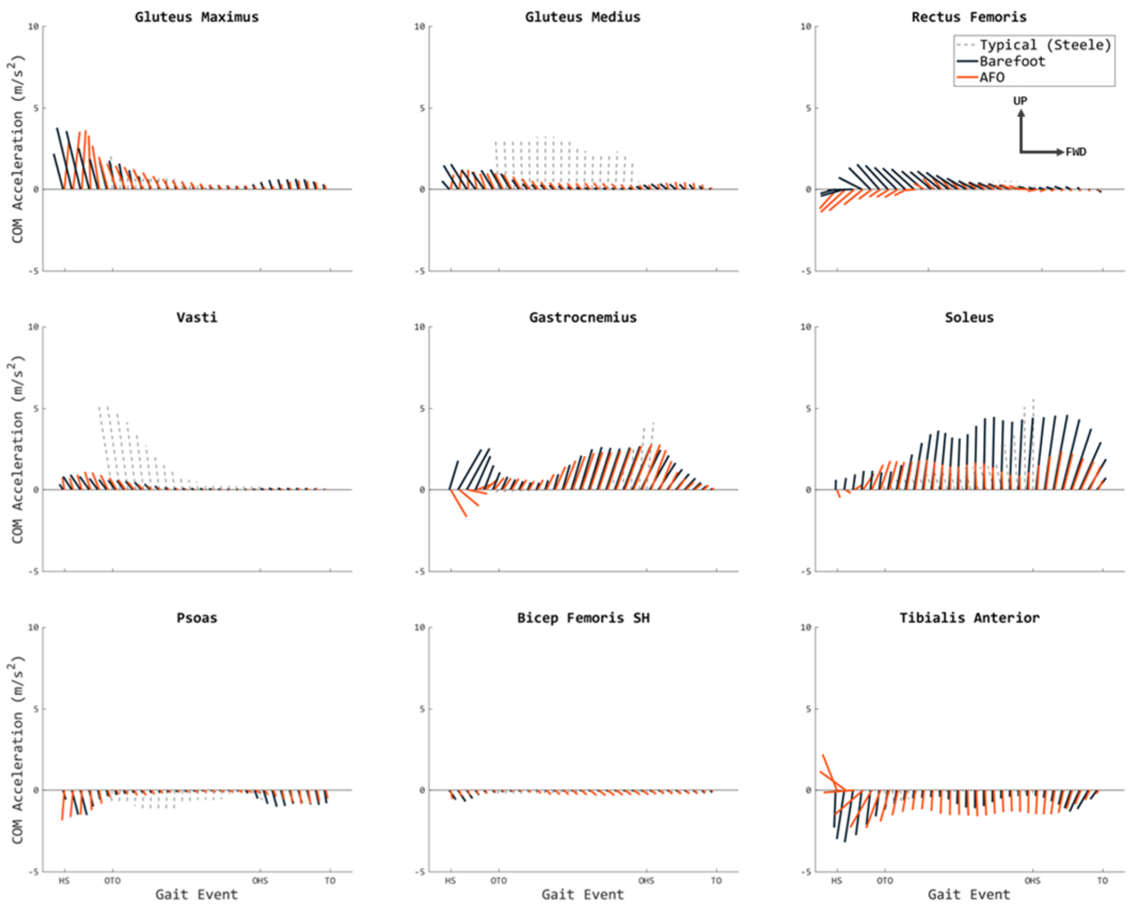


**Figure A4 – Total COM accelerations for Barefoot and AFO walking for Subject 4. The only muscle group with a noticeable change was the soleus. All other muscle groups experienced minimal changes. The vertical displacement of each vector from zero represents the vertical COM acceleration induced during stance while the horizontal displacement of the vector represents the fore-aft induced COM acceleration. Vectors are plotted at approximately each 3% of stance. Abbreviations: HS – heel strike, OTO – opposite toe off, OHS – opposite heel strike, TO – toe off.**

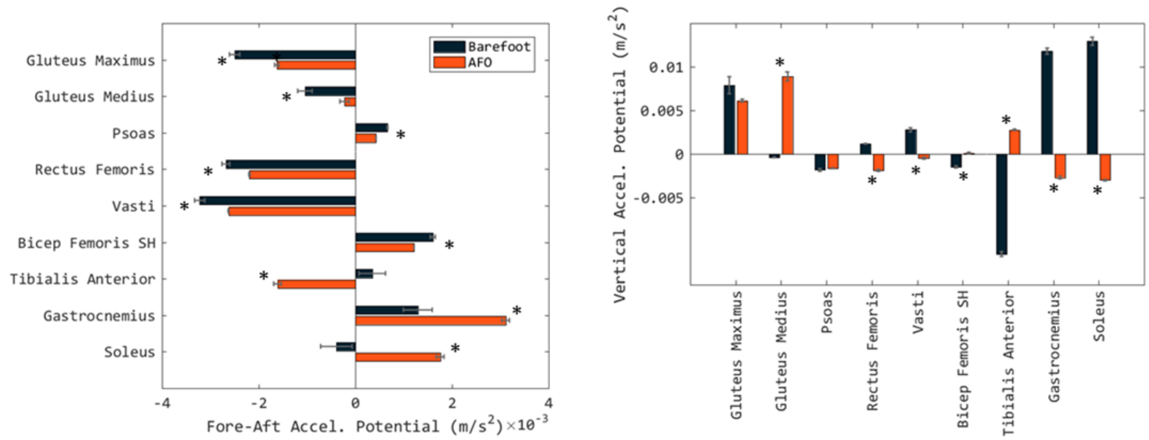


**Figure A5 – Muscle COM potentials for Barefoot and AFO walking for Subject 1. The muscle potentials for each trial were averaged for the entire stance phase and then individual trials were averaged.**

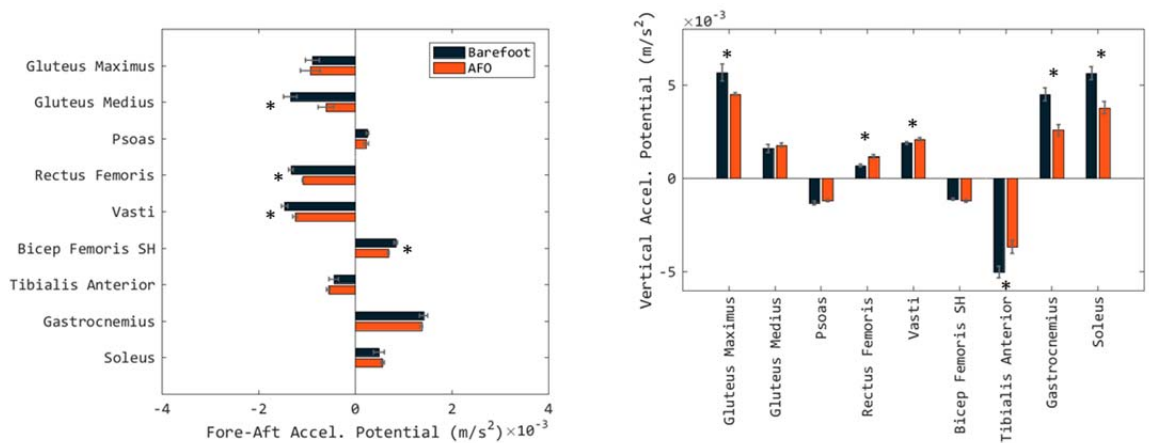


**Figure A6 – Muscle COM potentials for Barefoot and AFO walking for Subject 4. The muscle potentials for each trial were averaged for the entire stance phase and then individual trials were averaged.**