Universidade de Lisboa

Faculdade de Motricidade Humana

ANKLE-FOOT ORTHOSES: A BIOMECHANICAL APPROACH TO THE EFFECTS OF A NON-INVASIVE THERAPEUTICAL MANAGEMENT OF THE GAIT IN CHILDREN WITH CEREBRAL PALSY

Diogo Filipe dos Reis Ricardo

Orientadora: Professora Doutora Filipa Oliveira da Silva João

Tese especialmente elaborada para obtenção do grau de Doutor em Motricidade Humana, na especialidade de Biomecânica

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Junho de 2022

Para as Mulheres da minha vida!

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A dissertation is a long and lonely journey. We go much of our way into doubts and uncertainties. Yet, the small victories, such as when we manage to make our first field work, or when we see joy as a child collaborates in our data collections and uses an orthosis for the first time, gives us strength and cheer. Today I am certainly a different person, and above all a better professional, than when I started this PhD, and much is due to all the ones who have crossed my path and somehow participated with me in this project.

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Trabalho realizado no âmbito do projeto

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TITLE: Ankle-Foot Orthoses: A biomechanical approach to the effects of a non-invasive therapeutical management of the gait in children with Cerebral Palsy.

ABSTRACT

Three-dimensional gait analysis methodologies are widely used to assess gait and the effects of ankle-foot orthoses (AFO) in the treatment of gait deviations in children with Cerebral Palsy (CP). However, due to the specific requirements for motion capture, AFO characteristics, and the heterogeneity of this population, the wide range of gait parameters present such variability that makes it difficult to interpret its clinical application. This PhD thesis main purpose was to investigate how those assessment methodologies could provide important and clinically relevant data regarding gait analysis with AFO. Four studies were conducted employing exploratory and experimental methods: the first study is a scoping review that presents the immediate and long-term effects of AFO in children with spastic bilateral cerebral palsy; the second study evaluates test-retest reliability of a six-degree-of-freedom marker set in key points of gait kinematics, kinetics, and time-distance parameters in children with CP; the third study demonstrates the use of the gait profile score index to quantify gait quality in children with cerebral palsy wearing several types of AFO; the last study explores two different pose estimation algorithms used to build a 3D model of a child with cerebral palsy wearing a specific AFO. Overall, the findings of our work presented in this dissertation, provided scientific data for the rehabilitation science, demonstrating that the use of gait analysis protocols specific to the characteristics of children with cerebral palsy, and to existing therapeutic interventions, offer less susceptible information to methodological errors. Further research is required to continue exploring the several methodologies to assess and analyse the gait in children with cerebral palsy to support decision making and therefore providing a more effective treatment in the rehabilitation processes.

KEYWORDS: Orthotics device; Cerebral Palsy; Gait; Kinematics; Biomechanics

TÍTULO: Ortóteses do tornozelo: uma abordagem biomecânica aos efeitos de uma intervenção não-invasiva na marcha de crianças com Paralisia Cerebral.

RESUMO

As metodologias tridimensionais de análise da marcha são amplamente utilizadas para avaliar a marcha e os efeitos das ortóteses do tornozelo-pé no tratamento de desvios de marcha em crianças com paralisia cerebral. No entanto, devido aos requisitos específicos para a captura de movimentos, as características das ortóteses e a heterogeneidade desta população, os diversos parâmetros de marcha apresentam tal variabilidade de resultados que dificulta a interpretação da sua aplicação clínica. Esta tese de doutoramento teve como principal objetivo investigar como essas metodologias de avaliação poderiam fornecer dados importantes e clinicamente relevantes no que diz respeito à análise da marcha com ortótese de tornozelo-pé. Foram realizados quatro estudos utilizando métodos exploratórios e experimentais: o primeiro estudo é uma revisão sistemática que apresenta os efeitos imediatos e a longo prazo das ortóteses de tornozelo-pé em crianças com paralisia cerebral bilateral espástica; o segundo estudo avalia a fiabilidade do teste-reteste de um conjunto de marcas refletoras de seis graus de liberdade, definido em pontos-chave da cinética, cinemática e em parâmetros de espácio-temporais em crianças com paralisia cerebral; o terceiro estudo demonstra a utilização do índice de pontuação do perfil de marcha para quantificar a qualidade da marcha em crianças com paralisia cerebral que utilizam vários tipos de ortóteses do tornozelo-pé; o último estudo explora dois diferentes algoritmos de estimativa de posição, usados para construir um modelo tridimensional de uma criança com paralisia cerebral usando um tipo específico de ortótese. Em geral, as conclusões do nosso trabalho apresentado nesta dissertação, forneceram dados científicos para as ciências da saúde, demonstrando que o uso de protocolos de análise de marcha específicos às características das crianças com paralisia cerebral, e às intervenções terapêuticas existentes, oferecem informação menos suscetível a erros metodológicos. No entanto. são necessárias mais pesquisa e investigação para se continuar a explorar as várias metodologias de avaliação e analise da marcha em crianças com paralisia cerebral, por forma a apoiar a tomada de decisão clínica e, portanto, proporcionar um tratamento mais eficaz nos processos de reabilitação.

PALAVRAS-CHAVE: Ortóteses; Paralisia Cerebral; Marcha; Cinemática; Biomecânica.

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LIST OF ABBREVIATIONS

3D	Three Dimensional
6DoF	Six-degree-of-freedom
AFO	Ankle-foot Orthoses
CP	Cerebral Palsy
DAFO	Dynamic Ankle-foot Orthoses
GO	Global Optimization
GPS	Gait Profile Score
GVS	Gait Variable Score
HAFO	Hinged Ankle-foot Orthoses
ICC	Intraclass Correlation Coefficient
MCID	Minimal Clinically Important Difference
PEA	Pose Estimation Algorithms
RoM	Range Of Motion
SAFO	Solid Ankle-foot Orthoses
SO	Segment Optimization
STA	Soft Tissue Artifact

Chapter 1

1. General Overview

1.1 Introduction

This dissertation has a Biomechanical basis and is focused on the instrumented clinical gait analysis of children with cerebral palsy (CP), with a special focus on the use of ankle-foot orthosis (AFO).

Biomechanical gait analysis is an assessment tool that provides detailed and quantitative information, whether from the point of view of kinematics, kinetics or neuromuscular function of the lower limb. The World Health Organization (WHO) International Classification of Functioning (ICF), Disability and Health model identifies the factors that affect disability and should therefore be considered when evaluating an intervention. Instrumented 3D clinical gait analysis is one of the instruments that is recommended in that model [1].

Despite the increase of knowledge about the existing motion capture techniques and methods, and its use in clinical conditions, this work intends to highlight the existing need for a detailed description of gait analysis protocols when assessing subjects with CP that wear AFO. The performed scoping review intended to summarize the published developed studies regarding its quality and transparency in reporting the clinical gait analysis of children with CP using biomechanical parameters. This led us to assess the reliability of measurements in motion capture in a widely heterogenic population with different gait patterns and comorbidities. Our concern about the AFO effects in this population originated a study in which we used a widely applied gait index, not only to assess the gait outcomes, but also to reflect on its limitations in this gait conditions (barefoot vs AFO-footwear combination). The variability of the biomechanical parameters raised some concerns about the pose estimation algorithms. We've tested different biomechanical models regarding the AFO-use, to determine in which way kinematics could be under or overestimated, originating some bias outcomes, and therefore misleading the clinical decision making.

1.1.1 Dissertation objectives

The main purpose of this thesis was to investigate how different three-dimensional gait analysis methodologies (marker set or pose algorithms) can contribute to the assessment of the effects of several types of AFO in gait patterns of children with CP. Accordingly, this research sought also to contribute to further increase evidence-based information regarding the rehabilitation process in CP, and potentially improve the quality of life of these children.

To investigate how those methodologies could provide us important and clinically relevant data, four complementary objectives were defined:

1) To determine if the current literature and scientific data clarifies the effects of AFO in the gait patterns of children with CP.
2) To perform a prospective test-retest reliability study in anthropometric and three-dimensional gait analysis parameters (spatio-temporal, kinematics, and kinetics) in children with CP.
3) To evaluate the acute effect of the use of AFO in the gait parameters (spatio-temporal, kinematics, and kinetics) of children with CP.
4) To compare different pose algorithms in children with CP in AFO-use condition.

1.1.2 Dissertation overview

To achieve our goals, a methodological approach was conducted on a sample of children with cerebral palsy presenting several motor developments disorders. Chapters 1 and 2 are theorical chapters. Chapter 1 presents a research context regarding the most relevant technical background and the major issues behind the proposed research. In Chapter 2 a short summary is developed to reflect upon the methodological options used in this work, which provides a glimpse of the problems and challenges that can be found in this field of research. Four different chapters present the research and field work developed to address the identified problems. An individual and detailed protocol was developed for each study, based on the available scientific data to the best of our knowledge. Two of them (Chapters 3 and 4) were published as scientific papers in international peer-reviewed journals, Chapter 5 is submitted also in an international peer-reviewed journal and Chapter 6 is a work in progress and is expected to be submitted soon.

Hence, Chapter 3 presents an overview of the literature concerning the use of classifications of gait patterns in children with spastic bilateral CP. This study, entitled **"Effects of Ankle Foot Orthoses on the Gait Patterns in Children with Spastic Bilateral Cerebral Palsy: A Scoping Review",** aimed to assess the immediate and long-term effects of AFO in children with spastic bilateral CP. It presents a description of the gait (barefoot and with AFO) regarding the spatio-temporal, kinematic, kinetic, and functional outcomes of randomized controlled trials (RCT) and controlled clinical trials (CCT). This study was preceded of a registration in the PROSPERO platform (CRD42018102670).

The study presented in Chapter 4 **"Test-Retest Reliability of a 6DoF Marker Set for Gait Analysis in Cerebral Palsy Children",** aimed to evaluate test-retest reliability of a six-degree-of-freedom (6DoF) marker set in key points of gait kinematics, kinetics, and time-distance parameters in children with CP. Due to the fact that the same assessor was responsible for the placement of the markers in all the sessions, it was important to assess its natural variability.

The gait trials were performed on two different days within a 10-day period (long enough to minimize the assessor memory bias and short enough to prevent a change in the children's gait pattern). Even though some variations were found between sessions, that seemed to be related mostly to the heterogeneity of their gait patterns and affected sides. For this reason, the marker set that was used was considered feasible for this purpose.

This appreciation led us to Chapter 5, **"The use of Gait Profile Score (GPS) in detecting the effects of Ankle-foot Orthoses in children with Cerebral Palsy"**, a cross-sectional study where we used a gait index (GPS) to quantify gait quality in children with CP wearing several types of AFO. This gait index reflected changes in the gait patterns, regardless of the AFO that was used or affected side, which emphasized it's use as a relevant instrument to support therapeutical interventions in the rehabilitation process.

Chapter 6, "**Sensitivity of kinematic and Kinetic outcomes to different pose estimation algorithms in Children with Cerebral Palsy with the use of AFO: a case study"**, presents a study where two different pose (position and orientation) estimation algorithms are used to build a 3D model of each child wearing a different AFO. We wanted to explore how the different models, with specific constraints, would generate different results in the kinematics and kinetics of gait, and how we could gain more insight of its influence in clinical gait analysis.

The dissertation ends with Chapter 7 summarizing the findings and answering the research questions. We then delineate the work's more general contributions and conclude recommending future research. Chapter 8 presents all the bibliographic references used across the dissertation, and Chapter 9, the Appendix, contains the academic dissemination that derived from this research, in the form of written papers and international congresses communications.

1.2 Background

1.2.1 Gait cycle

Gait is a sequence of movements that is repeated in a cyclic way. Each one of those gait units is called "gait cycle". The gait cycle is defined as the time interval between two successive occurrences of one of the repetitive events of walking, allowing the body to move forward while simultaneously maintaining stability. Although any event could be chosen to define the gait cycle, it is generally convenient to use the instant at which one-foot contacts the ground [2].

Each walking cycle is divided into two periods: the stance phase and the swing phase (Figure 1-1). The stance phase is the term used to nominate the period when the foot is in contact with the ground. The swing phase starts when the foot is in the air, not being in contact with the ground, allowing the limb to advance. In normal gait, the support phase represents about 60% of each walking cycle, while the swing phase represents 40% of this same cycle [2,3].

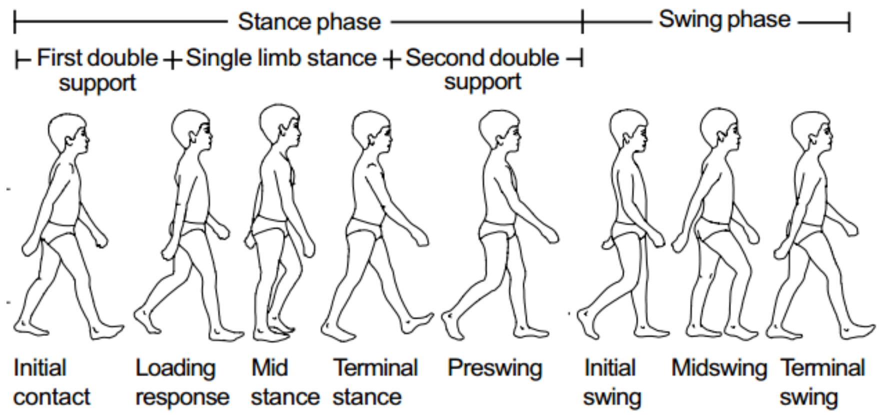

Figure 1-1: Phases and sub-phase of gait [3].

The duration of the gait cycle intervals is called the temporal parameters and may vary between subjects. When gait velocity changes, both phases of gait are shortened or increased, in an inverse relation with the latter. As for the double stance intervals, when gait velocity increases, single support lengthens, and double support diminishes [4].

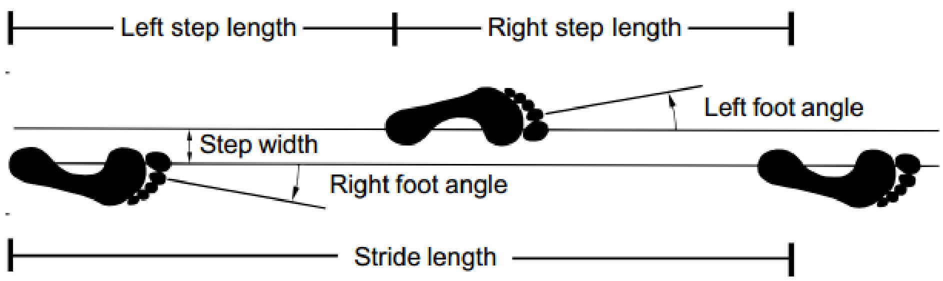

Figure 1-2: Spatial parameters in gait [3].

Spatial parameters related to the placement of the feet can also be useful to describe aspects of the human gait [2]. The stride length is the distance between two successive placements of the same foot and is equivalent of a gait cycle (Figure 1-2) since it is based on the actions of one limb [4]. It consists of two step lengths, left and right, each of which is the distance by which the named foot moves forward in front of the other one. In pathological gait, it is common for the two step lengths to be different [2].

1.2.2 Epidemiology

Cerebral palsy is a group of heterogeneous non-progressive neurodevelopmental conditions that affect the developing fetal or infant brain [5–7] associated with a common and significant disorder of motor development, with an incidence of 2-2.5 per 1000 live births [7]. The prevalence of CP is around 1.5-2.7 per 1000 children on several countries [8]. CP is primarily characterized by central nervous system abnormalities, such as loss of selective motor control and abnormal muscle tone [9]. As a result of growth, these primary characteristics often lead to secondary deficits, including bony deformities, muscle contractures and gait abnormalities [9]. The topographical distribution of motor impairment which these children present (hemiplegia, diplegia or quadriplegia) determines the specifics gait abnormalities [10].

Classifications of the gait patterns in children with Cerebral Palsy are based on how much of the lower limb is affect and the characterization of the abnormal motion that results in abnormal kinematic parameters of the joints [11]. Winters described four pathological gait patterns based on knee motion in the sagittal plane: jump, recurvatum, crouch and stiff knee [11]. Rodda et al. [12] classified the gait patterns in spastic diplegic CP into five groups, based on the kinematic analysis in the sagittal plane of the ankle, knee, hip and pelvis. This type of classifications based on gait patterns or topographical distribution indicates that orthotic recommendation could be more straightforward for children with higher levels of motor function [13].

1.2.3 AFO intervention

The most used lower limb orthoses in CP are AFO which provide direct control of the ankle and foot to improve gait [14–16]. The AFO involves the ankle joint and the entire or part of the foot [17,18], thus preventing the development or progression of structural deformity [19,20] and improving the dynamic efficiency of the child's gait [20]. Therapeutic interventions in motor rehabilitation aim to reduce the effect of increased muscle tone or improve the fluidity of motor control. Their effects might be temporary or permanent, although the quality of this evidence is low [19,21], as clinical practice has often been based on reports from case series and expert guidelines [7]. Although there is a lack of a clinical justification for the type of AFO selected to each case [22], its known that the AFO main properties are characterized according to their design, the type and stiffness of the material that is used for manufacture, and any modification of these factors will change the mechanical behaviour of the AFO, with consequent implications in the child's gait [23]. The clear description of this characteristics, along with a clear rationale for its design may clarify the clinical purposes for its prescription in terms of intervention in the rehabilitation process [24].

AFO are typically fabricated as a solid one-piece (SAFO) or Dynamic (DAFO), or as a two-piece design with a hinged joint (HAFO) [25]. SAFO restricts ankle and foot motion in all three planes, providing in one hand, stability in the stance phase and clearance for the swing limb. In the other hand, these restrictions – inherent in the design – compromise transitions through the three rockers of the stance phase [17]. DAFO can present slightly different trim lines. Although the forefoot is always encased, the proximal trim lines can be just superior to the malleoli or extended to the mid shank. The first case allows a free sagittal movement, and the latter originates a plantarflexion stop and a certain dorsiflexion movement. In either case, the DAFO provides a stable base for more effective motor performance and postural control [26]. HAFO allows a free plantar/dorsiflexion movement within a defined range, by means of a flexible element hinged at the anatomical ankle joint level [27]. As compared with the SAFO, the HAFO also improves mobility in many functional activities, but for some children with CP presenting moderate to severe spastic diplegia, the mobility provided by a HAFO compromises stability in early stance, reinforcing a crouch gait pattern [28].

According to Chui et al. [17] the International Organization for Standardization for consumer and patient protection presented the strength requirements for orthotic and prosthetic components. Although several types of materials, such as leather, metal, have been used in orthotic and prosthetic practice, currently AFO are normally made of lightweight polypropylene or carbon fiber [25]. Intrinsic to the selected type of material, stiffness is probably the most important characteristic of an AFO. It's influence in the desirable stability and shape to the body segments during stance and balance [17] can improve biomechanical outcomes [13]. The stiffness of an AFO may be determined by a number of factors, such as the mechanical properties of the material, the trim lines, the material thickness and the shape of the superstructure [29,30].

An AFO can affect the anatomical structures directly or indirectly. First are the direct effects that result from the anatomical constrains imposed on the joints that are enclosed. The latter can be described as all the other biomechanical gait parameters (proximal joint kinematics, spatio-temporal parameters, GPS, etc) that also affect the gait pattern [18].

1.2.4 Motion Capture in Cerebral Palsy

Patients with cerebral palsy (CP) are the most commonly assessed participants in gait laboratories [9]. Standard clinical gait analysis includes measures of gait in both barefoot and AFO conditions. The data from both gait conditions are commonly used for clinical interpretation and evaluation of AFO prescription and efficacy [31], which supports the often-empirical assessment of gait by introducing an objective and precise information, not only to be used in the decision making process in rehabilitation for the individual patient [16,32], but also to learn about a condition affecting a group of patients or the effect of an intervention [33]. An essential aspect of all these decisions is to know the ongoing interaction between orthopaedic, neurologic, and developmental considerations related to gait. Treatment of gait issues for children with CP with such complex presentations is often greatly enhanced by careful examination of their gait patterns using motion capture in addition to the typical history, clinical examination, and visual observation of gait [34].

Various studies have reported significant improvements in spatio-temporal parameters of velocity, step and stride lengths, and single-limb stance support time when children with CP wear their AFO [35]. In studies that compared either children with CP wearing AFO with their typically developed peers or children with CP wearing AFO and barefoot, it was shown that the use of AFO (regardless of the type) had a significant increase or an approximation to normal reference parameters in walking speed [36,37], step [38] and stride length [36–41], and a significant decrease towards normal cadence [36–38,40]. Nevertheless, there are also studies that reported no significant differences for walking speed [38–41], nor significant differences for cadence [38,39,41] irrespective of AFO type or study design.

In addition to global functional improvements, AFO have been shown to improve abnormal gait parameters specific to the ankle joint function [35]. An increase in the maximum plantar flexion moment in the terminal stance (push-off) was reported, regardless of the type of AFO, with results similar to those of healthy children [37–39,41]. This could indicate an improved ability to support body weight in a more appropriate alignment at the ankle [42].

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Chapter 2

2. Methodological Considerations

2. Methodological Considerations

2.1 Scoping Review

The methodology for conducting full systematic reviews in the area of health care has progressed considerably, leading to an increase in the terminology of the diverse approaches that, despite their different names, share certain essential characteristics, specifically, collecting, evaluating and presenting the available research evidence [1]. While methodologies for the synthesis of evidence in systematic reviews are now relatively sophisticated, much refinement is still possible for the conduct of relatively new techniques such as scoping reviews [2].

According to *Daudt* in States et al. [3], scoping reviews are often used “to examine the extent, range and nature of research activity” and “to identify research gaps in the existing literature”.

Scoping reviews are a useful tool in the evidence synthesis approaches [4] and follow the same methodological steps as systematic reviews, regarding the use of rigorous and transparent methods for data collection, analysis and interpretation of results and the potential for replication [5]. A major difference between scoping and systematic reviews is that scoping reviews focus is on the research findings themselves, as opposed to the means used to obtain them [6]. This allows for wider coverage of body of literature on a given topic and give clear indication of the volume of literature and studies available as well as an overview (broad or detailed) of its focus [7].

Researchers can also opt to conduct a scoping review over a systematic review where the purpose of the review is to identify knowledge gaps, clarify concepts, investigate research conduct [4], principally a body of literature is not sufficiently homogenous to analyze via a systematic review process [3]. However, due to the current overlap of methodologies, there is a need for an internationally agreed set of discrete, coherent and mutually exclusive review types [8].

Some authors [9] have described a framework for scoping reviews, which provided four specific reasons concerning the use this method: 1) To examine the extent, range and nature of research activity; 2) To determine the value of undertaking a full systematic review; 3) To summarize and disseminate research findings; 4) To identify research gaps in the existing literature. To some extent, scoping study methods may represent a shift in methodological focus away from expert knowledge associated with the traditional literature review, towards an approach that highlights competences associated with technical knowledge.

2.2 Intraclass Correlation Coefficients for Reliability Research

Intraclass correlation coefficient (ICC) is a widely used reliability index in conservative care medicine to evaluate test-retest, intrarater, and interrater reliability. These evaluations are fundamental to clinical assessment because, without them, we have no confidence in our measurements, nor can we draw any rational conclusions from our measurements [10].

It is suggested that 2-way mixed-effects model is more applicable for testing intrarater reliability with multiple scores from the same rater [11]. According to Portney et al., similarly, the 2-way mixed-effects model should also be used in test-retest reliability study because repeated measurements cannot be regarded as randomized samples. Still, for the selection of a proper ICC for test-retest Intrarater reliability, it is important to know if its application will be based on a single measurement or the mean of multiple measurements [10] – Eq.2-1., where MS_C = mean square for columns; MS_E = mean square for error; n = number of subjects; k = number of raters/measurements.

$$v = \frac{(cMS_C + dMS_E)^2}{\frac{(cMS_C)^2}{k-1} + \frac{(dMS_E)^2}{(n-1)(k-1)}}$$

Equation 2-1: Formula for calculating ICC considering the two-way mixed model, and absolute agreement (ICC[A,k]).

In addition, absolute agreement definition should always be chosen for both test-retest and intrarater reliability studies because measurements would be meaningless if there is no agreement between repeated measurements [10].

Since ICCs measure a correlating relationship with a value between 0 and 1, it is practically important to have standard criteria used to assess the reliability of measurements. The level of agreement was considered poor, fair, good, and excellent when ICC < 0.40, 0.40 ≤ ICC < 0.60, 0.60 ≤ ICC < 0.75, 0.75 ≤ ICC ≤ 1.00, respectively [12].

2.3 Motion tracking and 3D modeling reconstruction

The marker set that was used followed the calibrated anatomical system protocol (CAST) [13,14] and CODA pelvis [15], as seen in Figure 2-1. It was used to reconstruct the pelvis and both lower limbs [16]. The 22 individual markers and four marker clusters of four embedded markers each (Figure 2-2), allowed the reconstruction of seven body segments: feet, shanks, thighs, and pelvis. Each segment is considered to be independent and to have six degrees of freedom [17]. Lower limb segment masses were determined according to Dempster [18] while the remaining inertial parameters were computed based on Hanavan [19].

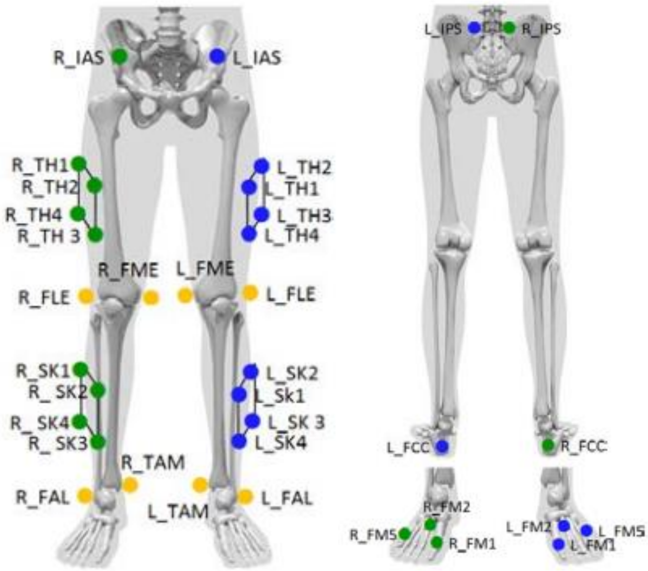

Figure 2-1: Model of the positioning of the retroreflective markers [20].

Figure 2-2: Positioning of the retroreflective markers attached to the subjects in several condition. **A)** Barefoot anterior view; **B)** AFO-shoe lateral view; **C)** Shoe Lateral view; **D)** AFO-shoe posterior view.

Palpation was used to locate the subcutaneous anatomical landmarks on the participants [13] and subsequently to place the marker set. These were 1.25 cm spherical reflective markers with a 1.8 cm semi-flexible width base. Four marker clusters were attached to the lateral part of the thigh and shank to independently track anatomical landmarks of each segment allowing rotational and translational motion at the joints [21]. These types of markers were adequate for the general height of these children given the smaller body parts. Motion capture data were collected with 14 infrared, high-speed cameras (Qualisys Oqus 300, Qualisys AB, Gothenburg, Sweden) at a frequency rate of 100 Hz. This system was synchronized in time and space with two force plates (FP4060-07, FP4060-10, Bertec, Columbus, OH, USA) embedded into the laboratory walkway [22]. Before each dynamic trial, a barefoot static trial in the standing position was recorded in order to determine the participant's joint centres and segmental reference systems, as well as segments' length [21]. Afterwards, the participant was instructed to walk along a 10 m corridor, unassisted at a self-selected pace. The dynamic trials ended when the child successfully achieved a minimum of five complete kinematic and kinetic walking cycles for each side [23–25], considering the natural variation in kinematic and kinetic gait parameters [16].

2.4 Gait Profile Score

GPS is calculated from the GVS, namely pelvic tilt, rotation and obliquity, hip flexion-extension, adduction-abduction and rotation, knee flexion-extension, ankle dorsi- and plantar-flexion, and foot progression of each leg [13]. The GPS is normally distributed for the population without clinically meaningful gait deviations (mean 5.3°) [26]. The root means square difference between a patient's data and the mean value obtained from tests performed on the unaffected population is expressed in degrees. The presentation of each GVS generates a MAP (Table 3a and 3b) which describes the magnitude of deviation of the nine individual variables averaged over the gait cycle, thus providing insight into which variables are contributing to the GPS overall value [27]. Thus, convenience of the MAP and GPS components together with GPS is an advantage in its use in clinical practice since it allows for a simpler overview of some complex kinematic data [27].

The GPS is calculated according to eq.2-2, where GPS is the root mean square average of the GVS variables:

$$\mathrm{GPS} = \frac{1}{N}\sum_{i=1}^{N} \mathrm{GVS}_i^2$$

Equation 2-2. Gait Profile Score calculation formula

Thus, the GPS result is an indicator of the overall quality of gait kinematics (increased GPS corresponds to a larger deviation from a physiological gait pattern). The authors [28] proposed a rationale for defining a minimal clinically important difference (MCID) for the GPS of

1.68. Regarding this MCID, we have calculated the GPS for two test conditions (barefoot and with AFO) as well as the MAP results for each child.

2.5 Pose estimation algorithms (PEA)

According to Capozzo et al. [15] movement analysis in the three dimensions of space requires the determination of the instantaneous position and orientation of systems of axes. The markers attached to the skin are assumed to move rigidly with the body segments to which they are set up [29]. However, some movement can occur between the skin and the underlying skeleton, mostly associated with the interposition of both passive and active soft tissues. This noise is referred as "soft tissue artifact" or STA and can be one of the main causes to poor estimations of pose [30]. Pose algorithms like the Segment Optimization model (SO) and the Global Optimization model (GO) intent to minimize the effect of this noise and improve the estimation of the pose [31].

The pose of the lower limbs and pelvis was estimated using two algorithms: 1) a global optimization (GO) algorithm and 2) a segmental optimization (SO) algorithm.

In the GO algorithm, also known as Inverse Kinematics, the model is built with physically realistic constraints [31,32]. Inverse Kinematics searches for the POSE (position and orientation) that best matches the differences between the measured and the model-determined marker positions. This algorithm is useful if we want to minimize errors due to soft tissue artifact, for instance. However, careful should be taken regarding clinical conditions, where abnormal movements may occur at the joints, so they won't be incorrectly masked. Given a set of measured marker coordinates $\boldsymbol{P}$ on a data frame, the GO at the system level finds a set of generalized coordinates $\boldsymbol{\vartheta}$ such that the error function (eq.2-3) is minimized where $\boldsymbol{W}$ is a positive-definite weighting matrix.

$$f(\vartheta) = \sum_{i=1}^{m}[(P - P'(\vartheta)]^T W[(P - P'(\vartheta)]$$

Equation 2-3: Global optimization algorithm (adapted from [31]).

where $\boldsymbol{P}'(\boldsymbol{\vartheta})$ is the corresponding set of marker coordinates calculated by the following transformation: $P'(\vartheta) = T(\vartheta)P^*$, where $\boldsymbol{T}(\boldsymbol{\vartheta})$ is the combined transformation matrix from segment-embedded frames to laboratory frame and is calculated by the model for a given $\boldsymbol{\vartheta}$.

In the SO algorithm, all the 6DoF for each segment are estimated. Thus, every segment needs at least three non colinear tracking markers. Each segment is independent and there is no linkage between them. SO estimates the segment pose in terms of its transformation matrix by minimizing marker array deformation from its reference shape in a least-squares sense [33]. The transformation is obtained by solving Eq.2-4 and Ep.2-5.

$$f = \sum_{i=1}^{m} (Rx_i + v - y_i)^T (Rx_i v - y_i)$$

Under the orthonormal constraint

$$R'_{seg} R_{seg} = I$$

Equations 2-4 and 2-5: Segmental optimization algorithm (adapted from [33]).

Where $\boldsymbol{x_i}$ and $\boldsymbol{y_i}$ are position vectors of marker $\boldsymbol{i}$ in the marker array at the reference and current positions, respectively, $\boldsymbol{R}$ is the rotation matrix, $\boldsymbol{v}$ is the translation vector and $\boldsymbol{m}$ is the number of markers. The orthonormal constraint indicates that the transformation is orthogonal [33].

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Chapter 3

3. Effects of Ankle Foot Orthoses on the Gait Patterns in Children with Spastic Bilateral Cerebral Palsy: A Scoping Review

Current status: **Published – Children Journal (Appendix 1)**.

Ricardo, D.; Raposo, M.R.; Cruz, E.B.; Oliveira, R.; Carnide, F.; Veloso, A.P.; João, F. Effects of Ankle Foot Orthoses on the Gait Patterns in Children with Spastic Bilateral Cerebral Palsy: A Scoping Review. Children 2021, 8, 903. doi: 10.3390/children8100903

Abstract

Background: Cerebral palsy (CP) is the most common cause of motor disability in children and can cause severe gait deviations. The sagittal gait patterns classification for children with bilateral CP is an important guideline for the planning of the rehabilitation process. Ankle foot orthoses should improve the biomechanical parameters of pathological gait in the sagittal plane. **Methods:** A systematic search of the literature was conducted to identify randomized controlled trials (RCT) and controlled clinical trials (CCT) which measured the effect of ankle foot orthoses (AFO) on the gait of children with spastic bilateral CP, with kinetic, kinematic, and functional outcomes. Five databases (Pubmed, Scopus, ISI Web of SCIENCE, SciELO, and Cochrane Library) were searched before February 2020. The PEDro Score was used to assess the methodological quality of the selected studies and alignment with the Cochrane approach was also reviewed. Prospero registration number: CRD42018102670. **Results:** We included 10 studies considering a total of 285 children with spastic bilateral CP. None of the studies had a PEDro score below 4/10, including five RCTs. We identified five different types of AFO (solid; dynamic; hinged; ground reaction; posterior leaf spring) used across all studies. Only two studies referred to a classification for gait patterns. Across the different outcomes, significant differences were found in walking speed, stride length and cadence, range of motion, ground force reaction and joint moments, as well as functional scores, while wearing AFO. **Conclusions:** Overall, the use of AFO in children with spastic bilateral CP minimizes the impact of pathological gait, consistently improving some kinematic, kinetic, and spatial-temporal parameters, and making their gait closer to that of typically developing children. Creating a standardized protocol for future studies involving AFO would facilitate the reporting of new scientific data and help clinicians use their clinical reasoning skills to recommend the best AFO for their patients.

Keywords: child; cerebral palsy; gait analysis; orthotic devices; biomechanics.

3.1 Introduction

The Cerebral Palsy (CP) is the most common cause of motor disability in children [1–3]. Overall prevalence of CP is around 1 per 500 live births worldwide [2–5]. CP is a complex pathology that describes a group of impairments and motor disorders [5] with different presentations and functional levels [6].

The gait deviations that occur in children with CP are among other factors, due to inadequate muscle action [7]. Instrumented clinical gait analysis has been a great tool for planning intervention and assessing outcomes in the rehabilitation process of children with CP

[2,8]. However, the use of all the outcomes within the three-dimensional kinematics or kinetics data to support the classifying gait patterns in CP is still scarce [8], due to the almost exclusive use of the sagittal plane kinematic outcomes in the majority of the gait classifications systems [9,10].

Among several gait classifications systems in children with CP, and particularly in bilateral spastic CP, Rodda et al. [11] has identified several gait patterns and reported a high intra-rater reliability and moderate inter-rater reliability [9]. More recently Papageorgiou et al. [10] concluded that the characteristics presented by Rodda were considered as the most exhaustive ones, always including information about the co-occurring deviations across all lower limb joints [10].

This classification is based on clinical insight and biomechanical principles and identifies five basic patterns of sagittal plane gait in spastic bilateral CP namely true equinus, jump gait, apparent equinus, crouch gait and asymmetric gait. These definitions are intended to be starting points for the guidelines in the planning of the rehabilitation process of children with CP. This allows not only the assessment of the most suitable orthosis for each case but also other surgical and non-surgical interventions, helping in the clinical decision-making process [11].

The use of ankle foot orthoses (AFO) is commonly prescribed to prevent the development or progression of deformity and to control motion to improve dynamic efficiency of the child's gait [12,13]. There is a wide selection of AFO that can be used in the rehabilitation processes. However, their intended function depends mainly on their configurations, the material used and its stiffness. Any alteration of these three components will alter the control the AFO has on the patient's gait [14]. There are multiple designs, either fabricated as a one-piece of thicker thermoplastic AFO, that restricts ankle and foot motion in all three planes (SAFO), or a flexible and dynamic AFO, that allows some degree of sagittal plane motion (DAFO), or a one piece design with a posterior malleolar trim line (Posterior Leaf Spring-PLS) or as a two-piece design with a hinged joint that typically allows for dorsiflexion (HAFO) or a one piece anterior shelf design that promotes knee extension (GRAFO) [15–17].

Overall, studies involving gait and kinematic analysis indicated that pathological gait in the sagittal plane can be improved using AFO [2,18,19], however it is not consensual about what factors are improved and how they have been improved. Thus, an assessment of the biomechanical characteristics and functional ability of the participants at baseline is crucial to track existing changes during the use of AFO [20]. Many studies involving orthotic use with CP patients present a wide variety of discrepancies in inclusion criteria or baseline assessments, missing information about orthosis design and construction and how they are used, and different type of outcomes that can bias the indicated results. Previous systematic reviews have not focused on specific CP subgroups or referred to gait pattern classifications, thereby including a wide range of gait abnormalities, or have included the information of lower quality studies [21–24].

Due to the broad specter of physical presentations of children with CP, the aim of this review is to determine the effects of different types of ankle foot orthoses on the gait of children with spastic bilateral CP presenting specific recommendations for this particular subset, and whenever possible refer to its effects on the five different sagittal gait patterns [11].

3.2 Materials and Methods

3.2.1 Search Strategy

A preliminary search was performed to select keywords related to the population, intervention, and outcomes using the PICO framework [25]. The keywords selected from the MeSH database in MEDLINE were: cerebral palsy, child, adolescent, orthotic devices, foot orthoses, splints, gait, kinematics, kinetics, walking, hip, hip joint, knee, knee joint, ankle, ankle joint, articular range of motion, walking speed, and International Classification of Functioning, Disability, and Health (ICF). Subsequent refinement searches were performed to obtain results. The selected keywords were joined by the words "AND" and "OR". The search equation was adapted according to the database where it was applied (Table A1-Appendix 1). The search was performed between January and July 2018, and included all records from the onset of each database. A secondary search was conducted in February 2020 with no other studies meeting the eligibility criteria. A keyword search was performed to match words in (all fields) the title, abstract, or keyword fields. The publication date was not restricted. Whenever possible filters on language were applied (Portuguese and English)(Appendix 1).

The search to identify the relevant articles for this review was carried out in the following databases: Pubmed, Scopus, ISI Web of Science, Cochrane Library, and Scielo. To identify potentially relevant trials that were unpublished or ongoing, a search was also performed in the database of the World Health Organization International Clinical Trials Registry Platform (WHO ICTRP) and in the US National Institutes of Health (ClinicalTrials.gov).

3.2.2 Selection Criteria

3.2.2.1 Eligibility Criteria

The methodology used for this review followed the Cochrane guidelines [26]. The eligibility criteria for the selected articles were randomized clinical trials (RCT) and controlled clinical trials (CCT) (Study Design); written in English, Portuguese or Spanish (Language); with a focus on the paediatric population with bilateral CP (Population) that used an AFO as a therapeutic intervention (Intervention). The exclusion criteria were the use of functional electrical stimulation or robotic assisted therapy and the existence of previous surgical or medical procedures (Intervention). The outcome measures considered were the biomechanical gait parameters and/or functional abilities, including spatial-temporal, kinematic, kinetic, and gross motor function outcomes (Outcomes).

3.2.2.2 Study Selection

The article selection was conducted by two independent reviewers (D.R. and M.R.R.), after duplicate removal and checking the articles' titles and abstracts against the eligibility criteria. The full text of the remaining articles was read. A bibliographic reference software manager (Mendeley V. 1.19.3) was used to assist the selection process. Whenever the two main investigators could not reach a consensus, a third external reviewer (E.B.C.) would intervene.

3.2.3 Methodological Quality (Risk of Bias)

The assessment of the quality of the included studies was the PEDro Risk of Bias Tool [27,28], for a minimum score of ≥5 points, which usually represents an adequate methodological quality study [29]. The rating of the studies and scoring on their methodological consistency were conducted by two reviewers (D.R. and M.R.R.) and, in case of disagreement or any discrepancies in scores, details were discussed with a third reviewer (E.B.C.). Furthermore, alignment between the PEDro scores and the Cochrane approach was verified for a broader assessment of the quality of the included studies [29].

3.2.4 Data Extraction

The characteristics of each selected study were extracted to compare the features across the studies. Author names, date of publication, study type and design, population characteristics and eligibility criteria, sample size, intervention type and duration, variables, measure instruments, and main findings were included.

3.3 Results

3.3.1 Article Selection

The initial search strategy identified 469 articles. After 78 duplicates were excluded, a further screening based on the title and abstract to assess the relevance of the articles excluded 352 articles. These articles did not meet the criteria of population (37), intervention (272), outcomes (4), and study design (39). A full text reading excluded 29 articles based on the criteria of population (3), intervention (2), outcomes (1), study design (21), and language (2). This resulted in a total of 10 articles that met our inclusion criteria and were included in our review flowchart (Figure 3-1).

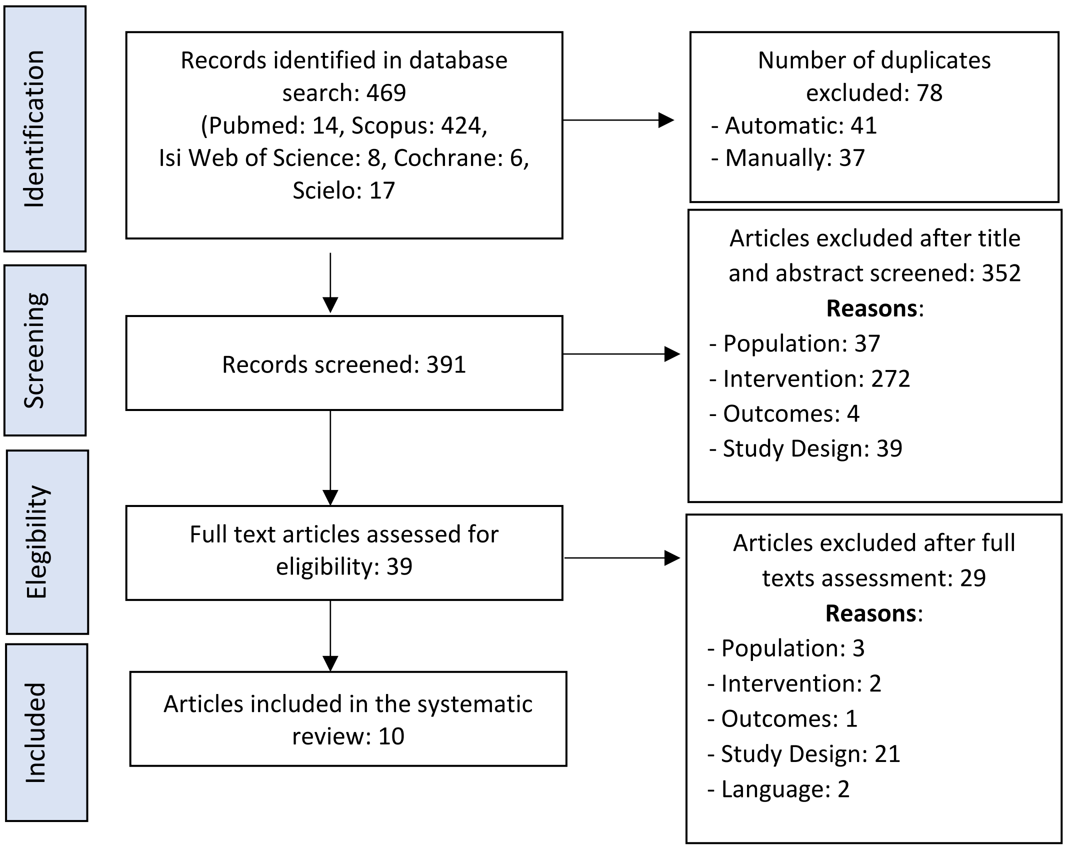

Figure 3-1: Flowchart of the article's selection process.

3.3.2 Article characteristics

The selected articles were published between 1997 and 2016. Of the 10 studies that were included, 5 were RCT [15,30–33] (three with a crossover design) and 5 were CCT [34–38]. The duration of the studies ranged from 1 day to 12 months in total. All studies compared at least one type of AFO intervention with barefoot, shoes or other types of AFO interventions. The range of measurement instruments that were used included: optoeletronic systems, ankle accelerometer, force plates, and the Gross Motor Function Measure (GMFM) tool. The studies reported spatial-temporal parameters (walking speed, stride length and cadence), kinematic outcomes (range of motion), kinetic outcomes (ground reaction force, joint moments and joint power) and functional outcomes (GMFM). This enabled the compilation of data detailed in the Table 3-1.

Table 3-1: Participants, sample details, methods, and main results.

Authors	Year	Study Design	Population Characteristics	Eligibility Criteria	N	Duration	Intervention/ Procedure	Variables	Measurement Instruments	Main Results and Author's Conclusions
Bjornson, 2006 [31]	2006	Randomised crossover controlled trial	23 children with spastic CP (age: 4,3 ± 1,5 years)	Children with spastic diplegia CP, 12 to 96 months, GMFCS I to III, Bilateral use of AFO with free plantarflexion.	23	1 day	DAFO and Shoes. GMFM used once with/without the orthoses during a same day evaluation.	Functional skills (GMFM scores).	GMFM	The GMFM percentage scores for all dimensions were significantly higher with the patients wearing the DAFO (*P* < 0.001). There seems to be a non-significant negative correlation of age to standing skills change, suggesting that DAFO effect may decrease with age, up to the age of approximately 7 years (*P* < 0.001).
Bjornson, 2016 [32]	2016	Randomised crossover controlled trial	11 children with spastic CP (age: 4,3 ± 1,04 years)	Children with spastic diplegia CP; GMFCS I to III; Bilateral use of AFO > 8h/day, >1 month.	11	4 weeks (2 weeks without AFO and 2 weeks with AFO)	SAFO and Shoes. Community based walking with/without AFO with a multiaxis accelerometer.	Functional skills (Average total strides per day; % daytime hours walking; average number strides >30 strides/min; peak activity index).	StepWatch (Ankle accelerometer)	No significant difference was found in the primary outcome of average daily total step count between AFO-ON and AFO-OFF (P = 0.48). AFO did not improve walking activity levels.
Buckon, 2004 [33]	2004	Randomised crossover controlled trial	16 children with spastic CP (age: 8,3 ± 2,3 years)	Children with spastic diplegia CP; GMFCS I to II; Bilateral use of AFO, 6 to 12h daily >3 month.	16	1 year (a baseline assessment after three months of no AFO wear, and an assessment at the end of each AFO three-month wearing period	Barefoot or HAFO or PLS or SAFO	Functional skills (GMFM scores); Gait analysis data (Kinematic variables at the pelvis, hip, knee, and ankle; Kinetic variables at the hip, knee, and ankle; Velocity, stride length, step length, and cadence)	Optoelectronic system; Force plates; GMFM.	AFO use, regardless of configuration, did not significantly alter pelvic and hip kinematics and/or kinetics from the barefoot condition. At the knee there was no significant kinematic change. All AFO configurations significantly altered ankle kinematics during the stance and swing phases of gait: dorsiflexion at initial contact (p=0.0001), peak dorsiflexion in stance (p<0.009), timing of peak dorsiflexion in stance (p<0.003), peak dorsiflexion in swing (p<0.0002), and dynamic ankle range (p<0.0001) compared with barefoot. Between the configurations, peak dorsiflexion in stance was significantly greater in the HAFO than the SAFO

										(p=0.01), and the timing of peak dorsiflexion in stance was significantly later in the stance phase in the HAFO compared with the SAFO (p=0.005). In conjunction with the changes in ankle kinematics, ankle kinetics (peak dorsiflexion moment in early stance [p=0.0001], peak plantarflexion moment in early stance [p=0.0001], peak power generation in stance [p<0.008], and the timing of peak power generation [p<0.005]) changed significantly in all the AFO configurations compared with barefoot. All of the AFO configurations significantly increased step (p<0.005) and stride length (p<0.006) compared with barefoot, while significantly decreasing cadence (p<0.0005). Therefore, velocity did not increase significantly with AFO use compared with barefoot. Velocity was significantly slower in the HAFO compared with the PLS (p=0.009), owing to a 17% decrease in cadence in the HAFO, an 11% decrease in the PLS, and a 13% decrease in the SAFO, compared with barefoot. AFO use did not significantly improve skills within the Standing dimension of the GMFM. However, all AFO configurations significantly improved skills within the W/R/J dimension compared with the barefoot condition (p<0.002).
Degelean, 2012 [34]	2012	Non-randomised controlled clinical trial plus healthy controls (repeated	20 children with spastic diplegic CP (mean age: 7,6 ± 1,7 years) + 20 typically developing children	Children with CP of the spastic diplegia type within the age of 4 and 12 years; No history of orthopaedic surgery; No botulinum toxin	20 + 20	1 day	Spring AFO or SAFO vs Barefoot. Participants walked at a comfortable speed an 8-meter walkway with AFO and barefoot.	Gait analysis data (Trunk movements; Angular velocities; Peak-to-peak excursions in trunk angular displacements;	Optoeletronic system.	Children with CP showed greater trunk sway excursion and angular velocity in both the sagittal and frontal directions compared to the control group (*P* < 0.05). Children with CP have greater sagittal and frontal trunk movements compared to typically developing children, but the

		measures design)	(mean age: 7,8; ± 1,4 years)	injections within the last year; GMFCS level I or II; Use of posterior leaf spring-type or solid AFO either in habitual walking or during physical therapy sessions.			The task was recorded using an optoelectronic system detecting passive retro-reflective markers.	Elevation angles of the thigh, shank, and foot).		difference in frontal motion was higher than in sagittal motion (*P* < 0.05). The use of any of AFO improved lower limb intersegmental coordination during gait in children with spastic diplegia by making it closer to a typical, mature gait pattern (*P* < 0.05). This was indicated in a significant greater ROM of the shank and a decreased ROM the foot. However, wearing AFO results in increased trunk motion, which may be problematic in the context of difficult postural control.
El-Kafy, 2014 [15]	2014	Randomised parallel group controlled trial	57 children with spastic diplegic (mean age: 7,3 ± 1,3 years)	Children with CP of the spastic diplegia type within the age of 6-8 years old; Under 40 kg; Cognitively able to understand simple instructions; No recurrent medical issues; No allergic reactions to the adhesive tape or any other materials; No visual, auditory, or perceptual deficits or seizures; No previously use of TheraTogs orthotic undergarment, or strapping system and ground reaction ankle foot orthosis; No botulinum toxin in the lower extremity musculature during the past 6 months or other spasticity medication within 3	19 + 19 + 19	2h/day, 5 days/week for a total of 12 weeks	Control group (A) - traditional neuro-developmental physical therapy. Study group (B) – A + TheraTogsTM orthotic undergarment and strapping system for both lower extremities. Study group (C) – B + received GRAFO in both lower limbs. Participants walked at a comfortable speed an 8-meter walkway with AFO and barefoot. The task was recorded using an optoelectronic system detecting passive retro-reflective markers.	Gait analysis data (Gait speed; Cadence; Stride length; Hip and knee flexion angles).	Optoeletronic system.	There were significant differences among the 3 groups pretreatment in all measured variables (gait speed, cadence, stride length, and bilateral hip and knee flexion angles), and that they were present post-treatment (*P* < 0.05). This is due to the improvement of the plantar flexion, knee extension coupling and knee and hip extension angle in mid stance provided by the GRAFO. The statistically significant differences post-treatment, in all parameters, were greater in group C than that in both groups A and B (*P* < 0.05). The results concerning the mean values of bilateral hip and knee rotational angles between both groups B and C revealed that there were no statistically significant differences in either pre- or post-treatment evaluation times (*P* < 0.05).

				months of pre-treatment testing.						
Lam, 2005 [35]	2005	Non-randomised controlled clinical trial plus healthy controls (repeated measures design)	7 boys and 6 girls with spastic diplegic CP (mean age: 5,9 ± 1,81 years) + 18 typically developing children (age matched)	Spastic diplegia CP with mainly moderate dynamic equinus (modified Ashworth scale 1–3); No significant coronal or rotational deformities; No botulinum toxin injections within the preceding 5 months; Good vision; The ability to comprehend instructions; Be able to walk independently.	13 + 18	1 day	AFO and DAFO. Barefoot (healthy subjects control group).	Gait analysis data (Stride length; Stride time; Speed; Stance time; Swing time; Stance/Swing ratio; Cadence; Range of motion parameters; Moment parameters; Power parameters).	Optoeletronic system; Force platform.	CP patients had significantly shorter stride length than normal. Both AFO and DAFO conditions significantly increased stride length (*P* < 0.05). The mean stride length in CP patients walking barefoot (0.69 ± 0.14) was 65% of the healthy age matched children. The stride length was significantly increased when the subjects were wearing AFO (0.74 ± 0.15) or DAFO (0.81 ± 0.15). Concerning the total ROM there was a reduction of range of motion at the ankle joint between the barefoot (22.39 ± 6.78), AFO (12.44 ± 5.55) and DAFO (19.72 ± 4.46). At initial contact children with DAFO presented a significantly increased knee and hip flexion by 4.8˚ (*P* < 0.016) and 5.3˚ (*P* = 0.012), respectably, when compared to barefoot walking. No significant difference was found at the ROM in the knee and hip between the AFO and DAFO . There was a significantly higher ground reaction force at the second peak wearing an AFO (0.97 ± 0.06) than when walking barefoot (0.89 ± 0.11). Both the AFO (0.96 ± 0.27) and the DAFO (1.11 ± 0.43) showed a significant improvement in the maximum plantarflexion moment compared to barefoot (0.69 ± 0.25). It was 0.28 Nm/kg higher in the AFO and 0.42 Nm/kg higher in the DAFO. There was no significant difference determined among barefoot, SAFO and DAFO in all knee and hip power parameters.

Radtka, 1997 [37]	1997	Non-randomised controlled clinical trial (repeated measures design)	10 children with spastic CP (6 diplegic; 4 hemiplegic) (mean age: 6,5 ±1,86 years)	Spastic diplegia and unilateral CP; Community ambulatory with plantigrade foot in standing, excessive plantar flexion during the stance, passive dorsiflexion of 5 degrees or more with knee extended, passive hip extension of 10 degrees or more, passive hamstring muscle length of 60 degrees or more in straight leg raise, mild to moderate spasticity in lower limb; No use of assistive device in ambulation; No orthopaedic surgery in the previous year.	10	3 months (2weeks barefoot +1 month with AFO + 2 weeks barefoot +1 month with DAFO)	AFO and DAFO.	Gait analysis data (Walking speed; stride length; cadence; range of motion of the trunk, pelvis, hip, knee, and ankle at initial contact and mid-stance).	Contact closing foot- switches; Optoelectronic system.	There was as increased stride length wearing the AFO (0.97 ± 0.16) and DAFO (0.93 ± 0.13) compared with the barefoot condition (0.82 ± 0.13). The cadence was higher barefoot (148.33 ± 15.73) than with the AFO (140.10 ± 8.79) and DAFO (136.55 ± 10.96). The excessive ankle plantar flexion with no orthoses (8.54 ± 5.61) was over reduced with AFO (-2.62 ± 3.93) and DAFO (-1.66 ± 6.23). There were no differences ($P < 0.002$) at the level in joint motions of the knee, hip, and pelvis at initial contact and mid-stance with AFO or DAFO. The amount of ankle plantar flexion that occurred at initial contact and mid-stance in the interventions with no orthoses was reduced with both AFO and DAFO. No differences were found for the gait variables when comparing the two orthoses ($P < 0.02$).
Radtka, 2005 [36]	2005	Non-randomised controlled clinical trial (repeated measures design)	12 children with spastic diplegic CP (mean age: 7,5 ± 3,83 years)	Spastic diplegia CP; Community ambulatory with ankle dorsiflexion to 0 degrees during static standing, excessive ankle plantar flexion of 5 degrees or more during stance in gait, passive ankle dorsiflexion to 5 degrees with knee extended passive hip extension to −10 degrees or less in the Thomas test,	12	3 months (2weeks barefoot +1 month with AFO + 2 weeks barefoot +1 month with HAFO)	SAFO and HAFO.	Gait analysis data (Range of motion of the knee and ankle during the stance phase; walking velocity; stride length; cadence; knee and ankle sagittal joint moments and powers during the stance phase).	Optoelectronic system; Force plates.	The mean stride length was increased with both SAFO (0.87 ± 0.19) and HAFO (0.90 ± 0.19) when compared to no AFO (0.79 ± 0.19). No significant differences in walking velocity, cadence and stride length when comparing no AFO, SAFO and HAFO ($P < 0.05$). At the knee joint there were no findings of significant differences between barefoot, SAFO or HAFO. When compared to the barefoot condition, at the ankle joint there were significant differences with the AFO and HAFO. The HAFO produced more normal dorsiflexion at the terminal stance phase than the SAFO and more

				passive hamstring length of 50 degrees or more as measured by a straight leg raise; mild spasticity of the triceps surae, hamstrings and quadriceps; No surgical procedures in the past or any other orthopaedic surgery during the year prior to the study.					excessive dorsiflexion during loading phase than barefoot. There were significant differences when comparing no AFO (0.69 ± 0.14), SAFO (0.96 ± 0.22) and HAFO (0.94 ± 0.25) in the peak ankle moments. There was a significant difference in peak ankle moments during the terminal stance phase between barefoot (-1.30 ± 6.59) and SAFO (11.50 ± 4.28) and barefoot and HAFO (16.13 ± 6.17). The mean values were similar between both AFO.
Smith, 2009 [38]	2009	Non-randomised controlled clinical trial plus healthy controls (repeated measures design)	15 children with spastic diplegic CP (mean age: 7,5 ± 2,9 years) + 20 typically developing children (mean age: 10,6 ±2,8 years)	Spastic diplegia CP; Able to walk independently without an assistive device; Jump gait pattern; GMFCS level I; No orthopaedic surgery in the past 12 months; No botulinum toxin injections in the past 6 months; Range of ankle dorsiflexion to at least neutral on static physical examination with the knee extended.	15 + 20	2,5 months (barefoot baseline + 4 weeks with DAFO or HAFO + 2 weeks barefoot + 4 weeks with DAFO or HAFO)	DAFO and HAFO. Barefoot (healthy subjects control group).	Gait analysis data (Walking speed; Cadence; Stride length; range of motion; joint moments; Joints powers); Functional skills (GMFM scores).	Optoelectronic system; Force plates; GMFM.	Significant improvements in gait metrics were seen during brace wear (*P* ≤ 0.05). When compared with barefoot condition, CP children wearing HAFO and DAFO showed a significant increase in stride length (0.98 ± 0.05) and (1.01 ± 0.05) and walking speed (1.09 ± 0.6) and (1.11 ± 0.6). When using HAFO or DAFO there was a significant decrease in normal cadence (*P* ≤ 0.006) compared with the children with CP in barefoot condition. When comparing gait cycles of children with CP and healthy children there was no significant difference in terms of stride length, walking speed or cadence. At the ankle significant differences between the HAFO or DAFO and the barefoot condition were found during the stance and swing phase (P ≤ 0.05). The knee peak flexion during swing was significantly different between de DAFO and barefoot condition (P ≤ 0.05). Children with CP using HAFO or DAFO had no significant effect on hip ROM. No significant differences were seen between the two different braces used

										($P \leq 0.05$). The barefoot and braced conditions differed most significantly in terms of ankle kinematics and kinetics ($P \leq 0.05$). During the terminal stance of pre-swing, the ankle moment was significantly increased for both DAFO (0.98 ± 0.1) and HAFO (1.05 ± 0.1) when compared to the barefoot condition (0.80 ± 0.1). When compared to healthy children, in the barefoot and AFO condition, CP children presented a significant increase in plantar flexor moment during the initial contact ($P \leq 0.05$). No significant differences in ankle powers were found between DAFO and HAFO.
Zhao, 2013 [30]	2013	Randomised parallel group controlled trial	70 boys and 42 girls with spastic diplegic CP (mean age: 2,69 ± 0.81 years)	Spastic diplegic CP; Between 1 and 4 years of age; Ability to walk independently, with or without an assistive Device; GMFCS levels I-II; Able to accept and follow AFO treatment strategy; No unstable seizures; No orthopaedic surgery for spasticity within the preceding 6 months; No botulinum toxin injections within the preceding 3 months; Without any other diseases that interfered with physical activity, and existence of	56 + 56	5 to 8 weeks	Day AFO. Night and Day AFO.	Gait analysis data (Passive ankle dorsiflexion angle).	Sections D and E of the 66-item GMFM.	No evidence was found that the prolonged wearing time with AFOs leads to increased benefits ($P < 0.05$). The GMFM-66 improvement in the day-night AFO-wearing group was lower than in the day AFO-wearing group rather than higher. AFO day-night use was not more effective than daytime use alone in children with spastic diplegia at GMFCS levels I to II.

	serious cognitive disabilities.

Abbreviations: AFO - Ankle Foot Orthoses; CP - Cerebral Palsy; DAFO - Dynamic Ankle Foot Orthoses; GRAFO - Ground Reaction Ankle Foot Orthoses; GMFCS - Gross Motor Function Classification System; GMFM - Gross Motor Function Measure; HAFO - Hinged Ankle Foot Orthoses; ROM - Range of Motion; SAFO - Solid Ankle Foot Orthoses;

The studies with fair to strong methodological quality were as follows: six studies with 4-5/10, one study with 6/10 and three studies with 8/10 in the PEDro scale (Table 3-2). All articles specified their "eligibility criteria", "follow-up", "intention to treat" and "statistical comparison". The "blind distribution", "blind subject", "blind therapist" and "blind assessor" were the items most often not verified. Three studies [15,30,31] managed to create blind assessment conditions, only two studies [15,30] had "blind distribution" and only one study [31] had unknowing therapist. No studies had "blind subjects" as it is not possible to use AFO without knowing it. Three studies [34,35,38] did not have equal circumstances at baseline ("similar prognosis") for their groups as they used typically developed children for control group.

Table 3-2: Methodological quality for studies in the review.

Article ID	PEDro Score											Total Score
	Eligibility Criteria*	Random Allocation	Blind Distribution	Similar Prognosis	Blind Subject	Blind Therapist	Blind Assessors	85% Follow-up	Intention to treat	Statistical Comparisons	Point of measure/ Measures of Variability	
Bjornson, 2006 [31]	Yes	Yes	No	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	8/10
Bjornson, 2016 [32]	Yes	Yes	No	Yes	No	No	No	Yes	Yes	Yes	No	5/10
Buckon, 2004 [33]	Yes	Yes	No	Yes	No	No	No	Yes	Yes	Yes	Yes	6/10
Degelean, 2012 [34]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
El-Kafy, 2014 [15]	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	Yes	8/10
Lam, 2005 [35]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
Radtka, 1997 [37]	Yes	No	No	Yes	No	No	No	Yes	Yes	Yes	Yes	5/10
Radtka, 2005 [36]	Yes	No	No	Yes	No	No	No	Yes	Yes	Yes	Yes	5/10
Smith, 2009 [38]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
Zhao, 2013 [30]	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	Yes	8/10

*This criterion is cited but not used to compute the total PEDro score.

3.3.2.1 Characteristics of the Participants (Sagittal gait patterns)

Across all studies, there was a total of 347 participants (289 children with CP and 58 typically developing children [34,35,38]). Most studies included only children with spastic bilateral CP (285). Despite this, one study [37] presented a heterogeneous population, with 4 children with spastic unilateral CP. However, as the results were presented separately, we did not include them in this review. Only a small percentage of the total of participants had their gait patterns identified. Two studies referred to the sagittal gait patterns classification [32,38], identifying in total 18 participants with jump gait pattern, 5 true equinus and 3 crouch gait patterns.

3.3.2.2 Types of AFO

The majority of interventions were centered in the comparison of gait when using ankle-foot orthosis and when walking barefoot [15,33–37] or using conventional shoes [31,32,38]. The type of AFO is central on most studies [15,30,33–38], but information about AFO construction, design and materials, as well as overall lower limb alignment and footwear are partially missing in every study.

We identified five different types of orthoses: 178 participants used Solid Ankle Foot Orthoses (SAFO) [30,32–37], 57 participants used Dynamic Ankle Foot Orthoses (DAFO) [31,35,37,38] 24 participants used Posterior Leaf Spring (PLS) [33,34], 46 participants used Hinged Ankle Foot Orthoses (HAFO) [33,36,38] and 19 participants used Ground Reaction Ankle Foot Orthoses (GRAFO) [15]. We found that overall, studies had no clear and consensual definition of the different types of AFO, and there was more than one description and configuration for the same terminology. In some of the studies, participants wore more than one type of orthoses [33,35–38], and in other studies some participants did not use any type of AFO [15].

3.3.2.3 Type of Outcomes

The main outcomes that were found were the following: spatial-temporal parameters [15,33,35–38], range of motion (RoM) [33,35–38], ground reaction forces [35], joint moments [33,35,36,38] and joint power [33,35,36,38]. Secondarily some studies presented functional parameters, isolated or correlated with the biomechanical analysis [38]. The most frequently used tool was the Gross Motor Function Measure scale (GMFM) [30–33].

Most articles do not directly relate the reported outcomes with changes of the gait pattern in children with CP. Still, whenever possible, outcomes observed in the sagittal plane were associated with changes in the gait pattern.

Spatial-temporal parameters

One study compared gait in children with CP barefoot at baseline and after 4 weeks of DAFO or HAFO wear and found significant differences ($P \leq 0.006$) across all measured spatial-temporal parameters (walking speed, stride length and cadence) [38]. In studies that compared either children with CP wearing AFO with their typically developed peers or children with CP wearing AFO and barefoot, it was shown that use of AFO (regardless of the type) had a significant increase or an approximation to normal reference parameters in walking speed [15,38], step [33] and stride length [15,33,35–38] and a significant decrease towards normal cadence [15,33,37,38].

Nevertheless, there were studies that reported no significant differences for walking speed [33,35–37] nor significant differences for cadence [33,35,36] irrespective of AFO type or study design.

Kinematic outcomes

The most often used kinematic parameter was RoM of the lower limb joints. For instance, significant improvement towards dorsiflexion of the ankle at the initial contact, and swing phase was observed [33,35–38] but, because the orthoses limit the plantarflexion, there was a significant decrease in RoM of the push-off stage of the pre-swing phase [35]. Maximal dorsiflexion in stance phase improved significantly with the use of SAFO [33,35,36]. It was also reported that the HAFO can produce excessive dorsiflexion during the stance phase [36].

While the most significant changes when wearing AFO are in the ankle RoM, in the knee RoM some differences were found, particularly in knee flexion on initial contact when compared to barefoot condition [35,38]. Also, children with CP wearing AFO showed a significantly greater range of motion of the shank [34]. No significant difference at knee RoM was found between the different types of AFO [33,35].

One study showed that children wearing DAFO were found to have a significantly greater hip flexion at initial contact [35], but overall, most studies found no significant changes at the hip joint, regardless the type of AFO [33,36–38].

Kinetic outcomes

Only four studies reported kinetic parameters. One study reported that when using a SAFO or DAFO there was a significant increase in the ground reaction force at the push-off when compared with the barefoot condition in children with CP [35]. An increase in the maximum plantarflexion moment in the terminal stance (push-off) was also reported, regardless of the type of AFO, with results similar to those of healthy children [33,35,36,38]. Peak knee extensor moment in early stance was significantly increased in the HAFO configuration compared with barefoot condition [33].

Regarding joint power, no significant difference was found in any of the analyzed joints between barefoot condition and AFO condition [33,35,38]. However, it was also reported that the peak of ankle power (that occurs at the push-off phase) when wearing a HAFO was similar to the barefoot condition [36] and between the configurations, the SAFO decreased peak power generation in stance significantly more than the PLS [33].

Functional Outcomes

To complement the biomechanical data, we were also interested in functional outcomes that the CP children may have reported with the use of AFO. The GMFM was the most often used tool, and studies showed it is responsive to change and can be used to evaluate the progress of a child while wearing AFO [39]. Although some of the included studies presented poor biomechanical data, they used this measure to evaluate the progress of AFO use in the rehabilitation [30,31,33]. Most of the studies showed that the percentage scores for this scale were significantly higher when the patients wore the AFO [30–32], with the exception of one study whereas the AFO use did not significantly improve skills within the standing dimension of the GMFM [33]. The changes in some dimensions and total score of GMFM were also significantly higher for independent walkers compared to children with CP using assistive devices while wearing DAFO [31].

3.4 Discussion

The main focus of this review was to assess the effects of AFO on gait in children with spastic bilateral CP, with particular attention to effects on different sagittal gait patterns. Identifying the gait type is useful in guiding orthotic options [40] and its use, coupled with the three-dimensional gait analysis, has been helpful in the clinical decision-making process. As a result, we have selected sagittal gait pattern classification [11] to help gather and systematize information. However, very few studies referred to such classification, making it difficult to summarize the data in the way planned in the protocol.

Fundamentally, clinical gait analysis for children with bilateral CP is very complex since bilateral impairment of the lower limbs is often met with different sagittal gait patterns in each limb, sometimes even overlapping, due to multiple gait abnormalities.

The lack of gait pattern classification makes it more difficult to determine the mechanical and functional AFO characteristics needed to improve the different gait phases and overall performance. Two studies [32,38] did use the sagittal gait patterns [11] to identify and categorize clinical subsets, although only one [38] provided the participants with the type of AFO indicated in the classification.

The appropriate AFO prescription is a practice that requires the clinician to perform a thorough physical examination and observational gait analysis, regardless of the age or Gross Motor Function Classification System (GMFCS) level of the child with CP [40]. Although consistent guidelines are lacking in this field [41], when applying an AFO, the aim is to correct and stabilize the biomechanical alignment of the foot and ankle, prevent the appearance or worsening of a musculoskeletal deformity, maintain the outcome of a surgical procedure, and ultimately improve gait [13].

The rationale behind the selection of each AFO and its prescription is missing in most studies. One study used the GMFCS to select the AFO to be used [34]; one study used the AFO already owned by the children with CP but without describing criteria [32]; two used the results of similar studies made previously [31,36]: one study made their own recommendations after a clinical and biomechanical assessment [37]; and three studies did not declare the criteria followed [30,35,37].

Nevertheless, results suggest that overall, AFO use may impact positively the gait of children with spastic bilateral CP. Spatial-temporal parameters, such as walking speed and stride length, revel an approximation to normal reference [34–37], suggesting a better gait efficiency and probably less energy expenditure [33].

Overall, children with CP wearing any type of AFO presented significant differences in the range of motion of the ankle, when compared to the barefoot condition. Regardless of the AFO type, its use appears to reduce pathological plantarflexion, common in several of the bilateral CP gait patterns [35]. However, some types of orthoses (DAFO, SAFO and GRAFO) are particularly more effective in controlling tibial progression and consequently promote knee extension during stance [32]. This can impact and modify the crouch gait pattern of CP children, approximating it to that of healthy subjects.

In children with spastic bilateral CP, there were significant increases in ground-reaction force and joint moments at push-off, while wearing different AFO [35]. This demonstrates that up to 5 degrees of dorsiflexion of the ankle inside the AFO, is more advantageous and induces an optimal muscle length on the calf muscles, approximating the plantar flexion moment to that of normal values [35,37].

Of the ten studies included in this review, only three focused on functional gains, and only one of the studies presented both biomechanical and functional data. Functional assessments are widely use in the rehabilitation of children with CP and should be more often correlated with biomechanical variables.

3.5 Methodological considerations of this review

We identified methodological limitations that are common in this type of study. Due to our eligibility criteria, the number of articles included was lower than other similar reviews. Of the 10 studies included, there was no common primary outcome between them. Although biomechanical and/or functional outcomes were found in all studies, the study designs are vastly heterogeneous (different samples sizes, wide range of age of participants, typically develop children control group versus children with CP barefoot control group; one-day studies versus 12 months follow up). This limits our ability to compare results due to the wider confidence intervals and a lower precision of the outcome measurements [42]. The point of statistical significance may be misleading, and this analysis may be leaving out some rehabilitation issues.

In CP research, CCT compares changes between groups to evaluate the efficacy of any treatment, but usually they lack reliable measures to detect changes that occur, and which may be important from a clinical point of view [43]. In evidence-based medicine the RCT is the highest level of evidence to be provided [44] and is the design of choice when comparing two or more healthcare interventions [29,44]. However, randomization may sometimes be affected by the number of participants, number of comparison groups, duration of the protocol and the overall study design, when studying AFO intervention. This may be a challenge because of differing clinical gait presentations and AFO requirements, thus we found that CCT are the more common for this population. The concealment of the allocation from parents and health care teams is a problem that practically limits this type of research [45,46].

Most studies included in this review were long-term follow-up studies [15,30,32,33,36–38] investigating the effects of the AFO for more than four weeks [47]. Studies with longer follow-up periods have also accounted for two weeks of rest between different orthosis [36,37]. This is relevant as there were trials with a crossover design, where more than one type of orthosis was tested on the same day, raising concerns about the issue of carry-over effect between the different orthosis [31,32]. We suggest that future studies account for a proper wash-out period between trials [48].

Few authors advocate an acclimatization period to ensure that the gait pattern is completely adapted to the altered ankle function as induced by the prescribed AFO which may have impacted the results of their study [49]. Three studies allowed the children to wear the AFO one to three months prior to the first gait assessment so that the participants could gradually adapt to wearing them for the entire test day [33,36–38]. In two studies, children were already wearing their currently prescribed AFO [31,34]. Only one study reported the number of hours per/day/week that the subjects wore their AFO, but in all others that information was missing [15].

There are a wide variety of AFOs used in clinical practice, which are characterized by their design, the material used and the stiffness of that material [14]. We´ve encountered at least five different types of AFO, but their definition was not always clear. The lack of nomenclature standardization also makes communication between researchers difficult [50].

Only one study used a prefabricated standard AFO [32] and in the remaining custom-made AFO were assigned for each participant [15,30,33,35–38]. Recent studies suggest that the initial outcomes are the immediate biomechanical response to the effect to the physical constraint imposed by the standard AFO, particularly the AFO stiffness [19,49]. On the other hand, custom-made AFO can be optimized, with fine adjustments to its design and/or to the footwear prescription, in order to focus on optimal stiffness and increase its effects on gait pattern [14,51].

Even though an AFO is a frequently-prescribed intervention for children with CP, rigorous evidence of their efficacy is limited [52], mainly because of the heterogeneity of outcome measures among researchers, which limits comparison between studies [53]. Although previous reviews have reported similar results and identified some of the limitations described above, still none has not reported consistent guidelines for future studies [10,21–24]. Particularly the absence of information about the clinical reasoning behind the AFO prescription, the selection of AFO design and construction, materials (including stiffness and thickness), AFO/footwear combinations, tuning and acclimatization periods, makes it difficult to compare results within studies [50,54]. For instance, Kerkum et al. [47] reported that ankle ROM was significantly less reduced by both stiff and flexible spring-hinged AFO, and there was also a reduction of the ankle power when using a more rigid AFO. In this study, the authors used an instrument to measure the mechanical properties of the AFO and reported all the parametrization that was used for the AFO design. The differences found in gait kinematics and kinetics due to the stiffness of the AFO are only possible to compare with studies that also report the mechanical characteristics of the AFO and that seems to be one of the greatest flaws in research regarding this topic [50].

Generically, the gait analysis protocols are not standard and have systematics errors related to extrinsic and intrinsic factors [55]. Regarding the use of 3D gait analysis in children with CP, several reliability studies identified that in the barefoot condition, kinematic and kinetic variables present with deviation between sessions due to number of gait trials [56], biomechanical models and marker setup [57] or gait patterns and affected sides [58,59]. In turn, many studies report difficulties in 3D motion analyses when children with CP are wearing an AFO (especially when modeling ankle kinematics). While assessing the gait of children with CP wearing AFO, the marker setup usually sits on the surface of the AFO and shoe, making the assumption that they are the same rigid segment [60]. This may cause the interaction shank/ankle/AFO to present with some deviations. Ries et al. [16] attempted to minimize the influence of the AFO on shank and ankle kinematics, by placing technical markers in a way that they were not to be covered or moved when the AFO was worn. By measuring the angle between the plantar surface of the shoe and the tibia, this study presented an alternative of measuring the true ankle position or the true neutral angle of the AFO.

Even thought, some methodological limitations are well reported, studies involving 3D gait analysis with the use of AFO should implement processes to minimize the error associated with their protocols, and state what measures they have to assure that the outcomes of their research singles out the AFO effect.

It is also important to use tools like International Classification of Functioning, Disability and Health (ICF) to standardize the report of results within the health-related domains [61]. Currently, there are specific ICF Core sets for CP patients, therefore future studies should summarize the outcomes in this framework and create a common language across healthcare professionals [62].

Overall, we considered there is need to standardize the AFO research, which can optimize the biomechanical properties and simplify future studies, making it possible to replicate results and provide better options for children with CP and their families [50].

3.6 Conclusions

In this review, we found that AFO use seems to have an immediate and a long-term effect in improving the sagittal gait patterns in children with spastic bilateral CP. However, most studies included heterogeneous groups with different gait patterns, and there were different approaches to the use of AFO. There is a need for future studies to invest in higher methodological quality protocols.

We propose the creation of a standardized protocol for future studies involving AFO and children with CP. There is a need to develop consistent AFO prescription algorithms that are designed specifically for each gait pattern. It should also include information about periods for AFO acclimatization and the need for fine tuning, appropriate follow-up periods to ensure full effect of AFO, appropriate wash-out periods, reports on hours per day of AFO usage, and AFO design, materials, and construction. This would facilitate the report and replication of new scientific data and help clinicians use their clinical reasoning skills to recommend the best AFO for their patients.

The rationale for these options needs to be more objective and evidence-based, which in the future may represent both improved assessment tools as well as a more effective therapeutic intervention.

3.7 References

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Chapter 4

4. Test-Retest Reliability of a 6DoF Marker Set for Gait Analysis in Cerebral Palsy Children

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Abstract

Background: Cerebral palsy (CP) is a complex pathology that describes a group of motor disorders with different presentations and functional levels. Three-dimensional gait analysis is widely used in the assessment of CP children to assist in clinical decision making. Thus, it is crucial to assess the repeatability of gait measurements to evaluate the progress of the rehabilitation process. The purpose of the study is to evaluate test-retest reliability of a six-degree-of-freedom (6DoF) marker set in key points of gait kinematics, kinetics, and time-distance parameters in children with CP.

Methods: trials were performed on two different days within a period of 7.5 ± 1.4 day. Motion capture data was collected with 14 infrared, high-speed cameras at a frequency rate of 100 Hz, synchronized in time and space with two force plates. Intraclass correlation coefficients considering the two-way mixed model, and absolute agreement (ICC[A,k]) were calculated for anthropometric, time–distance, kinematic and kinetic parameters of both lower limbs.

Results: the majority of gait parameters demonstrated a good ICC, and the lowest values were in the kinematic variables.

Conclusions: this study indicates wide-ranging reliability values for lower limb joint angles and joint moments of force during gait, especially for frontal and transverse planes. Although the use of a 6DoF-CAST in CP children was shown to be a feasible method, the gait variation that can be observed between sessions in CP children seems to be related not only to the extrinsic factors but also to their different gait patterns and affected sides.

Keywords: cerebral palsy; gait; reliability; kinematic model; biomechanics; kinematics; kinetics.

4.1 Introduction

Cerebral palsy (CP) is the most common cause of motor disability in children [1–3]. The average incidence of cerebral palsy is estimated to range between 1.5 to 3.3 per 1000 live births in European countries [4], whereas this number is around 1 per 500 live births worldwide [2,3,5]. CP is a complex pathology that describes a group of impairments and motor disorders [6] with different presentations and functional levels [7]. The gait deviations that occur in CP children are mainly originated by an inadequate muscle action [8]. Three-dimensional gait analysis is the widely accepted technique used in the assessment of ambulant patients with CP to assist in clinical decision making and assessing outcomes in the rehabilitation process [9], supporting a complete biomechanical analysis of the time-distance, kinematic and kinetic parameters of gait [10].

The purpose of each clinical gait measurement technology is to provide data free from measurement errors that may create uncertainty about the possible clinical interpretations. Thus, reliability addresses to which extent gait measurements are consistent or free from variation across time [11]. However, most of these clinical variables are not reliable [12], either due to their own intrinsic variations, namely in the intra-individual oscillations that occur in trial-to-trial sessions, or due to extrinsic variations, such as, marker placement [13]. CP children are intensively studied in gait analysis, but unlike other populations with gait abnormalities [14] there are no specific biomechanical models to their gait characteristics. It is known that there are significant differences among the techniques, but the gait laboratories still opt to use their typical protocols, regardless of the population.

It is essential to understand the possible errors associated with the different techniques of marker sets and underlying anatomical models [15] to reproduce the clinical gait measurements with confidence [16]. Significant differences exist in biomechanical models used in different laboratories. These include measured variables, degrees of freedom assigned to the joints, anatomical reference frames, and joint rotation conventions [17]. The conventional gait model (CGM) is a very widely used biomechanical model to calculate kinematic and kinetic variables in gait analysis [16]. It has been extensively validated but there are still some issues regarding its reliability, mainly due to its unconstrained segment dimensions which makes it more exposed to sources of errors [18]. The six-degree-of-freedom (6DoF) models are the most common alternative to the CGM that, despite needing more extensive validation [18], assumes that the segments are rigid and do not constrain the joints motions [19]. Several 6DoF modeling techniques were used in the assessment of repeatability in participants with motor and physical characteristics limiting the normal gait [14,20,21].

These 6DoF models address the known limitations of the CGM, but unlike the latter it still needs to be better researched. However, some results have indicated some of those claims (e.g., the segments have a fixed length and soft tissue artifact is reduced). Soft tissue artifact between markers is certainly eliminated by using rigid clusters, but a different form of soft tissue artifact will affect the orientation and position of the whole cluster in relation to the bones [22]. In children in particular, the amount of soft tissue surrounding the limb segments is not the major reason for some oscillations, but the smaller distance between clusters and anatomical markers which do not minimize the magnitude of this type of error. According to a systematic review of McGinley et al. [11] about the repeatability studies of kinematic models, the majority of the included studies used the CGM or some variant of it. In previous test-retest reliability studies performed in CP children, the biomechanical models were based in CGM [23] and similar models such as the Helen-Hayes [24] and the Vicon Clinical Manager [25]. One study that used a 6DoF variant (the Cleveland clinic marker set) [26] did not compare kinetic data and the authors assessed repeatability using a coefficient of multiple correlation (CMC) which has recently been determined not to be suitable as a tool for assessing reliability in gait measurements [27].

The lack of evidence regarding the reliability of 6DoF models in subjects with abnormal gait patterns, particularly in kinetic variables, was the motivation to develop this research. Moreover, knowing that errors associated with kinematic variables have tremendous consequences in the estimation of the kinetic parameters, it is essential to assess the magnitude of these errors. Considering these issues, the aim of this study is to evaluate the test-retest reliability of a 6DoF model in key kinematic and kinetic gait cycle parameters in CP children.

4.2 Materials and Methods

4.2.1 Design

Prospective controlled study.

4.2.2 Participants Selection

A convenience sampling of eight children (two females and six males) with cerebral palsy was recruited from two Portuguese cerebral palsy centres to participate in the study. Firstly, the participants' clinical history was reviewed, and a clinical exam was performed with the subject laid on the table, seated on a chair, or standing. The eligibility criteria were as follows: male and female children, between 4 and 16 years of age; with a clinical diagnosis of Unilateral Spastic Cerebral Palsy or Bilateral Spastic Cerebral Palsy of crural predominance, grades I and II in the Gross Motor Function Classification System (GMFCS) [28]; able to walk independently with or without walking aids; cooperative and able to comply with simple orders; feet size between 20 and 33; who had a clinical recommendation to use an ankle foot orthosis, but have never used it before, or during the trials; who have not undergone orthopaedic surgery of the lower limb in the last 12 months, and who are not expecting to have a surgical intervention in the next 6 months; and who were not given botulinum toxin in the last 6 months [29]. The protocol was approved by and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019). An informed consent was previously signed by the parent or the legal guardian of the participant.

4.2.3 Gait Protocol

The trials were performed on two different days within a period of 7.5 ± 1.4 days to minimize the assessor memory bias and short enough to prevent a change in the children's gait pattern or clinical condition [21]. Upon the participants' arrival, instruction was given about the protocol, the risks and benefits, as well as the informed consent.

The initial clinical exam consisted of a sequence of measures to assess bone and joint deformities, muscle length, muscle force, selective motor control and spasticity [2]. Two experienced researchers performed the clinical assessment while the same assessor was responsible for the placement of the markers in all the sessions. Palpation was used to locate the subcutaneous anatomical landmarks on the participants [30] and subsequently to place the marker set. These were 1.25 cm spherical reflective markers with a 1.8 cm semi-flexible width base. Four marker clusters were attached to the lateral part of the thigh and shank to independently track anatomical landmarks of each segment allowing rotational and translational motion at the joints [19]. These types of markers were adequate for the general height of these children given the smaller body parts. Motion capture data were collected with 14 infrared, high-speed cameras (Qualisys Oqus 300, Qualisys AB, Gothenburg, Sweden) at a frequency rate of 100 Hz. This system was synchronized in time and space with two force plates (FP4060-07, FP4060-10, Bertec, Columbus, OH, USA) embedded into the laboratory walkway [31]. Before each dynamic trial, a barefoot static trial in the standing position was recorded in order to determine the participant's joint centres and segmental reference systems, as well as segments' length [19]. Afterwards, the participant was instructed to walk along a 10 m corridor, unassisted at a self-selected pace. The dynamic trials ended when the child successfully achieved a minimum of five complete kinematic and kinetic walking cycles for each side [14,32,33], considering the natural variation in kinematic and kinetic gait parameters [34].

4.2.4 Data Processing

Gait cycles were extracted using Qualysis Track Manager (QTM) (v2020.3 build 6020, Qualisys AB, Gothenburg, Sweden). The subsequent analysis and processing were done using Visual 3D software (Professional Version v4.80.00, C-Motion, Inc., Rockville, MD, USA). The marker set (Figure 4-1) that was used followed the calibrated anatomical system protocol (CAST) [30,35] and CODA pelvis [36]. It was used to reconstruct the pelvis and both lower limbs [34]. The 22 individual markers and four marker clusters of four embedded markers each, allowed the reconstruction of seven body segments: feet, shanks, thighs, and pelvis. Each segment is considered to be independent and to have six degrees of freedom [37]. Lower limb segment masses were determined according to Dempster [38] while the remaining inertial parameters were computed based on Hanavan [39].

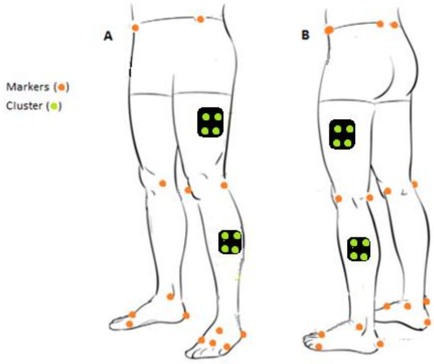

Figure 4-1: Positioning of the retroreflective markers attached to the subjects. Adapted from [40]: (A) anterior view; (B) posterior view.

The pelvic anatomical coordinate system was defined by surface markers placed on the right and left anterior superior iliac spines (ASIS) and on the right and left posterior superior iliac spines (PSIS) and can be described as the origin at the midpoint between the right ASIS and the left ASIS; the Z-axis points from the origin to the right ASIS; the X-axis lies in the plane defined by the right ASIS, left ASIS, and the midpoint of the right PSIS and left PSIS markers and points ventrally orthogonal to the Z-axis; and the Y-axis is orthogonal to the previous two [41]. The hip joint centers were computed using the pelvis markers, according to Bell's regression equations [36]. Anatomical reference frames of the lower limb segments were defined in accordance with the International Society of Biomechanics (ISB) recommendations to the standard description of joint kinematics [41].

The thigh anatomical coordinate system was defined by the hip joint centers previously computed using the pelvis markers and the lateral and medial femur condyles; the origin was the hip joint center; the Z-axis points from the midpoint between the lateral and medial femur condyles and the origin; the Y-axis is perpendicular to the Z-axis and the frontal plane of the thigh (defined by an axis between the lateral and medial femur condyles and the hip joint center); the X-axis is orthogonal to the previous two.

The shank anatomical coordinate system was defined by the femur condyles and malleolli markers; the origin was the knee joint center defined as the midpoint of the medial and lateral femur condyles; the Z-axis points from the midpoint between the lateral and medial malleoli and the origin; the Y-axis is perpendicular to the frontal plane of the shank and Z-axis; X-axis is orthogonal to the previous two.

The foot anatomical coordinate system was defined by the malleolli markers and the metatarsal markers; the origin was the ankle joint center defined by the midpoint between the lateral and medial malleoli markers; the Z-axis points from the midpoint between the 1st and 5th metatarsal heads and the origin; the Y-axis is perpendicular to the frontal plane of the foot and the Z-axis; X-axis is orthogonal to the previous two [42].

Lower limb and pelvis joint angles (calculated using a XYZ Cardan sequence) and moments (determined through inverse dynamics and normalized to subjects' body mass) were computed and expressed relative to the proximal segment. The XYZ Cardan sequence was used due to the ISB recommendations regarding its clinical and anatomical meaning [43], since the description of X, Y and Z are equal to flexion-extension, abduction-adduction and longitudinal internal-external rotation, respectively.

A cubic spline smoothing routine was used to filter both kinematic and kinetic data. The segment length was defined as the distance between the proximal and distal ends of the segment. Kinematic and kinetic data were normalized to 100% of the gait cycle. Peak values for lower limb angles and moments, as well as time–distance parameters, were computed for each cycle and averaged for each subject [21]. All data were considered assuming the lower limbs as independent to evaluate the variation of each one, even if they participated jointly during the gait cycle.

4.2.5 Statistical Methods

Statistical analysis to assess test-retest reliability of the gait kinematic and kinetic data was carried out using the method described by Quigley et al. [44] and Fernandes et al. [21]. Intraclass correlation coefficients considering the two-way mixed model, and absolute agreement (ICC[A,k]) [45,46] were calculated for anthropometric, time-distance, kinematic and kinetic parameters of both lower limbs. The level of agreement was considered poor, fair, good, and excellent when ICC < 0.40, 0.40 ≤ ICC < 0.60, 0.60 ≤ ICC < 0.75, 0.75 ≤ ICC ≤ 1.00, respectively [47]. The absolute measure of reliability standard error of measurement (SEM) was calculated using the following equation: SEM = $\mathrm{SD}_{diff}/\sqrt{2}$. The indicated levels of error for kinematic data were considered acceptable if SEM ≤2°, reasonable between 2° and 5°, and concerning if SEM ≥5° [20]. From each trial, 97 individual values of clinical interest were extracted. The calculated key points included the mean difference between measurements and the 95% confidence interval (CI) for mean difference, the standard deviation of the differences (SD_{diff}) and the 95% Bland and Altman limits of agreement (95% LOA). All the statistical tests were conducted using SPSS (version 26.0; IBM, Chicago, IL, USA) and $p < 0.05$ was considered statistically significant.

4.3 Results

The participants of the study were a convenience sampling of eight CP children (Table 4-1) able to walk independently (three hemiplegic, five diplegic; two females, six males; age 87.88 ± 25.56 months; height 1.17 ± 0.14 m; mass 24.25 ± 8.26 kg). Two trials were performed on two different days within period of 7.5 ± 1.4 days.

4.3.1 Reliability of Anthropometric Parameters

The ICCs were ≥0.96 for anthropometric measurements (Table 4-2). The lowest were the right (0.97, 95% CI 0.86 to 0.99) and left foot segment length (0.96, 95% CI 0.83 to 0.99) and SEM values were ≤0.64 cm.

4.3.2 Reliability of Time-Distance Parameters

For time-distance parameters, ICCs were ≥0.75 (Table 4-3) except for right step length (0.64, 95% CI 0.00 to 0.92) and right stride length (0.64, 95% CI 0.00 to 0.92). The SEM values were 0.06 m and 0.11 m, respectively.

4.3.3 Reliability of Kinematic Parameters

Most joint angle peaks demonstrated excellent ICCs ≥0.75 (Table 4-4). Good ICCs were also shown in both sides of the lower limbs. On the right lower limb, the pelvic obliquity up was (0.67, 95% CI 0.00 to 0.94) and the hip internal and external rotation (0.73, 95% CI 0.00 to 0.95) and (0.67, 95% CI 0.00 to 0.93), respectively. Similarly on the left side, hip abduction was (0.60, 95% CI 0.00 to 0.92) and internal rotation (0.67, 95% CI 0.00 to 0.93). At the knee joint, its internal rotation was (0.64, 95% CI 0.00 to 0.92) and ankle eversion (0.60, 95% CI 0.00 to 0.91). However, a few of the ICCs variables were poor, the majority on the right side, with hip flexion (0.14, 95% CI 0.00 to 0.84), knee abduction (0.37, 95% CI 0.00 to 0.88), adduction (0.33, 95% CI 0.00 to 0.87), internal rotation (0.00, 95% CI 0.00 to 0.69) and ankle plantar flexion (0.00, 95% CI 0.00 to 0.81) and inversion (0.00, 95% CI 0.00 to 0.80). In the left side, only the ankle plantar flexion (0.27, 95% CI 0.00 to 0.92) presented similar values in this range. The SEM values ranged between 1.8° to 14.7° and average between 3.2° e 7.9°.

Table 4-1: Subject characteristics.

				Left Lower Limb			Right Lower Limb		
Subject	Affected Side	Height (m)	Mass (Kg)	True Leg Length (cm)	Sagittal Gait Pattern	Gastrocnemius Spasticity (Modified Ashworth Scale)	True Leg Length (cm)	Sagittal Gait Pattern	Gastrocnemius Spasticity (Modified Ashworth Scale)
001	Bilateral	1.09	19.5	52.5	True equinus [48]	1+	54.5	True equinus [48]	2
002	Unilateral	1.14	26	54.6	Normal	0	54.3	True equinus [49]	2
003	Bilateral	1.32	26	66	Apparent equinus [48]	1+	66	Apparent equinus [48]	1+
004	Unilateral	0.98	13.5	46	True equinus [48]	1+	45	Normal	0
005	Bilateral	1.37	34	71	Apparent equinus [48]	2	70.5	Apparent equinus [48]	2
006	Unilateral	1.32	37	70.2	Normal	0	70.1	True equinus with recurvatum knee [49]	1+
007	Bilateral	1.06	15.5	52	True equinus [48]	3	52.7	True equinus [48]	3
008	Bilateral	1.10	18	54	Jump gait [48]	2	54.5	Jump gait [48]	2

Table 4-2: Reliability values for anthropometric measurements.

Anthropometric Parameters	ICC	ICC 95% CI	Mean	Mean Diff	Mean Diff 95% CI	SD_{diff}	95% LOA	SEM
Pelvis Segment Depth (cm)	0.98	(0.93, 0.99)	13.2	0.2	(−0.2, 0.7)	0.6	(−0.97, 1.40)	0.4
Inter ASIS Distance (cm)	0.98	(0.94, 0.99)	17.3	−0.1	(−0.7, 0.3)	0.6	(−1.50, 1.13)	0.4
Right Tight Segment Length (cm)	0.99	(0.97, 0.99)	26.6	−0.1	(−0.7, 0.4)	0.7	(−1.55, 1.20)	0.5
Left Tight Segment Length (cm)	0.99	(0.89, 0.99)	26.7	−0.5	(−0.9, 0.1)	0.4	(−1.50, 0.42)	0.3
Right Leg Segment Length (cm)	0.99	(0.95, 0.99)	25.8	0.1	(−0.7, 0.8)	0.9	(−1.68, 1.85)	0.6
Left Leg Segment Length (cm)	0.99	(0.97, 0.99)	25.9	0.3	(−0.0, 0.7)	0.4	(−0.53, 1.23)	0.3

Right Foot Segment Length (cm)	0.97	(0.86, 0.99)	8.8	0.1	(−0.2, 0.4)	0.4	(−0.76, 0.96)	0.3
Left Foot Segment Length (cm)	0.96	(0.83, 0.99)	9.0	0.1	(−0.3, 0.5)	0.5	(−1.01, 1.21)	0.4
Average	0.98							0.4

Intraclass correlation coefficient, ICC; 95% CI, 95% confidence interval for the ICC; mean, mean of measurements at baseline trial and retest trial; mean diff, mean of the differences between measurements at times 1 and 2 and the 95%. CI for mean diff, the standard deviation of the differences (SD_{diff}); 95% LOA, Bland and Altman 95% limits of agreement; SEM, standard error of measurement.

Table 4-3: Reliability values for time-distance parameters.

Time-Distance Parameters	**ICC**	**ICC 95% CI**	**Mean**	**Mean Diff**	**Mean Diff 95% CI**	SD_{diff}	**95% LOA**	**SEM**
Speed (m/s)	0.78	(0.08, 0.99)	0.82	−0.08	(−0.21, 0.06)	0.16	(−0.40, 0.24)	0.12
Cycle Time (s)	0.86	(0.34, 0.97)	0.92	0.04	(−0.06, 0.13)	0.11	(−0.19, 0.26)	0.08
Double Limb Support Time (s)	0.84	(0.01, 0.97)	0.2	0.05	(0.01, 0.09)	0.05	(−0.05, 0.15)	0.03
Stride Length (m)	0.94	(0.65, 0.99)	0.74	−0.04	(−0.08, 0.01)	0.05	(−0.14, 0.07)	0.04
Stride Width (m)	0.94	(0.73, 0.99)	0.12	0.01	(0.00, 0.02)	0.02	(−0.02, 0.04)	0.01
Average	0.87							0.06
				Left lower Limb				
Cycle Time (s)	0.84	(0.31, 0.97)	0.92	0.06	(−0.05, 0.16)	0.12	(−0.19, 0.30)	0.09
Stance Time (s)	0.85	(0.33, 0.97)	0.58	0.05	(−0.03, 0.13)	0.10	(−0.15, 0.25)	0.07
Swing Time(s)	0.76	(0.00, 0.95)	0.35	0.01	(−0.03, 0.04)	0.04	(−0.07, 0.08)	0.03
Step Time (s)	0.79	(0.00, 0.96)	0.45	0.01	(−0.04, 0.06)	0.06	(−0.11, 0.13)	0.04
Step Length (m)	0.93	(0.63, 0.99)	0.38	0.00	(−0.03, 0.03)	0.04	(−0.08, 0.08)	0.03
Stride Length (m)	0.93	(0.63, 0.99)	0.75	0.00	(−0.07, 0.07)	0.08	(−0.16, 0.16)	0.06
Average	0.85							0.05

				Right lower Limb				
Cycle Time (s)	0.86	(0.30, 0.97)	0.93	0.02	(−0.08, 0.12)	0.12	(−0.21, 0.25)	0.08
Stance Time (s)	0.87	(0.44, 0.97)	0.57	0.04	(−0.03, 0.10)	0.08	(−0.12, 0.19)	0.05
Swing Time(s)	0.84	(0.24, 0.97)	0.36	−0.02	(−0.06, 0.02)	0.05	(−0.11, 0.08)	0.03
Step Time (s)	0.79	(0.00, 0.96)	0.46	0.00	(−0.07, 0.07)	0.09	(−0.16, 0.17)	0.06
Step Length (m)	0.64	(0.00, 0.93)	0.36	−0.05	(−0.12, 0.02)	0.08	(−0.21, 0.11)	0.06
Stride Length (m)	0.64	(0.00, 0.93)	0.72	−0.11	(−0.24, 0.03)	0.16	(−0.42, 0.21)	0.11
Average	0.73							0.07

Intraclass correlation coefficient, ICC; 95% CI, 95% confidence interval for the ICC; mean, mean of measurements at baseline trial and retest trial; mean diff, mean of the differences between measurements at time 1 and 2 and the 95% CI for mean diff, the standard deviation of the differences (SD_{diff}); 95% LOA, Bland and Altman 95% limits of agreement; SEM, standard error of measurement.

Table 4-4: Reliability values for kinematic parameters.

Kinematic Parameters	**ICC**	**ICC 95% CI**	**Mean**	**Mean Diff**	**Mean Diff 95% CI**	SD_{diff}	**95% LOA**	**SEM**
				Pelvic joint angle (°)				
				Left lower Limb				
Anterior Tilt +	0.40	(0.00, 0.88)	16.0	−0.1	(−5.2, 5.0)	6.1	(−12.24, 12.02)	4.3
Posterior Tilt -	0.83	(0.20, 0.97)	10.4	−1.2	(−5.2, 2.8)	4.7	(−10.58, 8.19)	3.3
Obliquity Up +	0.84	(0.20, 0.97)	2.7	0.5	(−1.7, 2.7)	2.6	(−4.69, 5.69)	1.8
Obliquity Down -	0.75	(0.00, 0.95)	−4.5	0.2	(−1.9, 2.3)	2.5	(−4.87, 5.28)	1.8
External Rotation -	0.44	(0.00, 0.89)	−6.6	0.2	(−6.4, 7.0)	8.0	(−15.55, 16.10)	5.3
Internal Rotation +	0.76	(0.00, 0.95)	13.7	1.1	(−5.0, 7.2)	7.3	(−13.32, 15.55)	5.2
Average	0.67							3.6

				Right lower Limb				
Anterior Tilt +	0.51	(0.00, 0.91)	16.1	−0.8	(−6.1, 4.3)	6.2	(−13.15, 11.37)	4.4
Posterior Tilt -	0.84	(0.31, 0.97)	10.3	−2.2	(−6.1, 1.6)	4.6	(−11.30, 6.82)	3.2
Obliquity Up +	0.67	(0.00, 0.94)	3.8	0.1	(−2.2, 2.5)	2.8	(−5.44, 5.81)	2.0
Obliquity Down -	0.85	(0.31, 0.97)	−2.7	−0.7	(−2.9, 1.3)	2.5	(−5.78, 4.20)	1.8
External Rotation -	0.88	(0.44, 0.98)	−12.0	−1.8	(−6.2, 2.4)	5.2	(−12.06, 8.32)	3.6
Internal Rotation +	0.85	(0.21, 0.97)	7.5	−4.2	(−8.7, 0.2)	5.4	(−14.86, 6.30)	3.8
Average	0.77							3.1
				Hip Joint angle (°)				
				Left lower Limb				
Flexion +	0.79	(0.00, 0.96)	45.0	−1.4	(−6.2, 3.5)	5.8	(−12.78, 9.98)	4.1
Extension -	0.78	(0.00, 0.96)	1.3	−0.7	(5.8, 4.3)	6.1	(−12.72, 11.24)	4.3
Abduction -	0.60	(0.00, 0.92)	−10.4	0.3	(−4.2, 4.9)	5.5	(−10.41, 11.15)	3.9
Adduction +	0.76	(0.00, 0.95)	4.8	0.8	(−2.7, 4.4)	4.3	(−7.62, 9.27)	3.0
External Rotation -	0.58	(0.00, 0.90)	−8.9	4.3	(−7.3, 18.0)	15.1	(−24.37, 35.08)	9.7
Internal Rotation +	0.67	(0.00, 0.92)	3.9	4.9	(−4.1, 16.0)	12.0	(−17.69, 29.66)	8.5
Average	0.70							5.6
				Right lower Limb				
Flexion +	0.14	(0.00, 0.85)	45.5	−0.9	(−9.1, 7.1)	9.7	(−20.10, 18.11)	6.9
Extension -	0.82	(0.12, 0.96)	1.5	−1.8	(−7.6, 3.9)	6.9	(−15.46, 11.80)	4.9
Abduction -	0.75	(0.00, 0.95)	−9.9	0.2	(−3.5, 4.1)	4.6	(−8.78, 9.37)	3.2
Adduction +	0.79	(0.00, 0.96)	6.9	−0.4	(−3.9, 3.0)	4.1	(−8.62, 7.71)	2.9
External Rotation -	0.67	(0.00, 0.93)	−10.7	−6.1	(−16.8, 4.4)	12.7	(−31.10, 18.77)	9.0

Internal Rotation +	0.73	(0.00, 0.95)	1.0	−4.0	(−14.5, 6.3)	12.4	(−28.54, 20.40)	8.8
Average	0.65							5.9
Knee Joint angle (°)								
Left lower Limb								
Flexion +	0.75	(0.00, 0.95)	70.6	0.2	(−6.5, 7.0)	8.1	(−15.71, 16.17)	5.7
Extension -	0.85	(0.17, 0.97)	8.6	0.4	(−3.6, 4.5)	4.9	(−9.15, 10.04)	3.4
Abduction -	0.48	(0.00, 0.90)	−7.4	0.5	(−5.1, 6.1)	6.7	(−12.68, 13.74)	4.7
Adduction +	0.46	(0.00, 0.90)	5.8	1.5	(−7.7, 10.9)	11.2	(−20.27, 23.42)	6.8
External Rotation -	0.75	(0.00, 0.95)	−8.4	−0.6	(−7.9, 6.6)	8.7	(−17.73, 16.45)	6.1
Internal Rotation +	0.62	(0.00, 0.92)	4.7	3.0	(−5.0, 11.0)	9.7	(−15.91, 21.94)	6.8
Average	0.65							5.6
Right lower Limb								
Flexion +	0.86	(0.25, 0.97)	68.5	−0.1	(−8.3, 8.0)	9.8	(−19.38, 19.13)	5.9
Extension -	0.98	(0.88, 0.99)	6.4	1.5	(−0.6, 3.6)	2.5	(−3.50, 6.50)	1.8
Abduction -	0.37	(0.00, 0.88)	−6.9	−2.0	(−10.2, 6.1)	9.8	(−21.31, 17.13)	6.9
Adduction +	0.33	(0.00, 0.87)	4.7	−3.8	(−14.2, 6.6)	12.4	(−28.21, 20.54)	8.7
External Rotation -	0.76	(0.00, 0.95)	−7.5	3.5	(−4.9, 12.1)	10.2	(−16.43, 23.61)	7.2
Internal Rotation +	0.00	(0.00, 0.69)	5.4	0.8	(−11.4, 13.0)	14.6	(−27.87, 29.49)	9.3
Average	0.55							6.6
Ankle Joint angle (°)								
Left lower Limb								
Dorsiflexion +	0.46	(0.00, 0.90)	9.8	3.3	(−9.0, 15.7)	14.8	(−25.69, 32.37)	10.4
Plantar Flexion -	0.27	(0.00, 0.86)	−11.1	2.6	(−10.5, 15.7)	15.7	(−28.22, 33.48)	11.1

Eversion -	0.60	(0.00, 0.91)	1.2	2.4	(−2.0, 7.0)	5.4	(−8.13, 13.11)	3.8
Inversion +	0.75	(0.00, 0.94)	13.0	1.6	(−3.1, 6.3)	5.6	(−9.44, 12.68)	3.9
Foot Internal Progression +	0.95	(0.75, 0.99)	3.8	−0.4	(−4.1, 3.1)	4.4	(−9.13, 8.14)	3.1
Foot External Progression -	0.87	(0.34, 0.97)	−14.3	1.4	(−7.5, 10.3)	10.6	(−19.40, 22.29)	6.5
Average	0.65							6.5
				Right lower Limb				
Dorsiflexion +	0.40	(0.00, 0.82)	7.7	2.3	(−12.5, 17.1)	17.7	(−32.48, 37.14)	12.6
Plantar Flexion -	0.00	(0.00, 0.81)	−13.5	4.5	(12.8, 21.9)	20.7	(−36.15, 45.23)	14.6
Eversion -	0.43	(0.00, 0.76)	1.1	0.0	(−5.7, 5.7)	6.9	(−13.48, 13.52)	4.8
Inversion +	0.00	(0.00, 0.80)	14.1	0.0	(−3.9, 3.8)	4.6	(−9.11, 8.99)	3.2
Foot Internal Progression +	0.95	(0.78, 0.99)	−11.7	−3.1	(−9.0, 2.7)	7.0	(−16.97, 10.67)	4.9
Foot External Progression -	0.94	(0.72, 0.99)	29.3	−4.6	(−13.9, 4.6)	11.0	(−26.37, 17.05)	6.8
Average	0.45							7.8

Intraclass correlation coefficient, ICC; 95% CI, 95% confidence interval for the ICC; mean, mean of measurements at baseline trial and retest trial; Mean Diff, mean of the differences between measurements at time 1 and 2 and the 95% CI for mean diff, the standard deviation of the differences (SD_{diff}); 95% LOA, Bland and Altman 95% limits of agreement; SEM, standard error of measurement.

Table 4-5: Reliability values for kinetic parameters.

Kinetic Parameters	**ICC**	**ICC 95% CI**	**Mean**	**Mean Diff**	**Mean Diff 95% CI**	SD_{diff}	**95% LOA**	**SEM**
				Hip Joint Moment (N m/Kg)				
				Left lower Limb				
Flexion -	0.95	(0.76, 0.99)	−0.46	−0.02	(−0.11, 0.06)	0.10	(−0.22, 0.17)	0.07
Extension +	0.67	(0.00, 0.94)	0.50	0.02	(−0.12, 0.16)	0.17	(−0.31, 0.34)	0.12
Abduction +	0.79	(0.00, 0.96)	0.43	0.01	(−0.08, 0.10)	0.11	(−0.20, 0.22)	0.08

Adduction -	0.00	(0.00, 0.75)	−0.21	−0.05	(−0.28, 0.18)	0.28	(−0.60, 0.50)	0.20
Average	0.61							0.12
Right lower Limb								
Flexion -	0.84	(0.11, 0.97)	−0.37	0.01	(−0.12, 0.13)	0.15	(−0.29, 0.30)	0.11
Extension +	0.40	(0.00, 0.86)	0.47	0.08	(−0.13, 0.30)	0.26	(−0.43, 0.59)	0.18
Abduction +	0.73	(0.00, 0.95)	0.48	0.00	(−0.13, 0.12)	0.15	(−0.29, 0.29)	0.11
Adduction -	0.79	(0.16, 0.96)	−0.12	−0.04	(−0.11, 0.02)	0.08	(−0.20, 0.11)	0.06
Average	0.69							0.12
Knee Joint Moment (N m/Kg)								
Left lower Limb								
Flexion -	0.69	(0.00, 0.94)	−0.27	0.03	(−0.04, 0.11)	0.09	(−0.15, 0.21)	0.07
Extension +	0.79	(0.00, 0.96)	0.41	−0.02	(−0.19, 0.15)	0.21	(−0.42, 0.38)	0.15
Valgus +	0.72	(0.00, 0.95)	0.17	0.01	(−0.09, 0.11)	0.12	(−0.23, 0.25)	0.09
Varus -	0.76	(0.00, 0.95)	−0.16	0.06	(0.00, 0.12)	0.07	(−0.07, 0.20)	0.05
Average	0.74							0.09
Right lower Limb								
Flexion -	0.49	(0.00, 0.90)	−0.26	0.13	(−0.06, 0.32)	0.23	(−0.32, 0.58)	0.16
Extension +	0.92	(0.63, 0.98)	0.31	−0.06	(−0.19, 0.07)	0.16	(−0.36, 0.24)	0.11
Valgus +	0.00	(0.00, 0.78)	0.27	−0.13	(−0.39, 0.13)	0.31	(−0.74, 0.48)	0.22
Varus -	0.61	(0.00, 0.92)	−0.14	−0.04	(−0.12, 0.03)	0.09	(−0.23, 0.14)	0.07
Average	0.51							0.14
Ankle Joint Moment (N m/Kg)								
Left lower Limb								

Dorsiflexion -	0.72	(0.00, 0.95)	−0.02	0.01	(−0.01, 0.04)	0.03	(−0.05, 0.08)	0.02
Plantar Flexion +	0.93	(0.61, 0.99)	0.85	0.00	(−0.12, 0.11)	0.14	(−0.27, 0.26)	0.10
Eversion +	0.57	(0.00, 0.92)	0.07	0.02	(−0.05, 0.08)	0.08	(−0.14, 0.17)	0.06
Inversion -	0.75	(0.00, 0.95)	−0.13	0.02	(−0.06, 0.09)	0.09	(−0.16, 0.19)	0.06
Average	0.74							0.06
				Right lower Limb				
Dorsiflexion -	0.00	(0.00, 0.77)	−0.02	−0.02	(−0.05, 0.02)	0.04	(−0.10, 0.06)	0.03
Plantar Flexion +	0.78	(0.00, 0.96)	0.75	−0.01	(−0.15, 0.13)	0.17	(−0.34, 0.32)	0.12
Eversion +	0.85	(0.21, 0.97)	0.04	0.00	(−0.03, 0.03)	0.04	(−0.07, 0.07)	0.03
Inversion -	0.55	(0.00, 0.91)	−0.16	−0.03	(−0.18, 0.13)	0.18	(−0.39, 0.33)	0.13
Average	0.55							0.08

Intraclass correlation coefficient, ICC; 95% CI, 95% confidence interval for the ICC; mean, mean of measurements at baseline trial and retest trial; mean diff, mean of the differences between measurements at time 1 and 2 and the 95% CI for mean diff, the standard deviation of the differences (SD_{diff}); 95% LOA, Bland and Altman 95% limits of agreement; SEM, standard error of measurement.

4.3.4 Reliability of Kinetic Parameters

For the ICCs of kinetic parameters, the results were higher than those for the kinematic data, where the majority were ≥0.75 (Table 4-5). The lowest ICCs between sessions were found in right knee joint valgus moment (0.00, 95% CI 0.00 to 0.78), right ankle dorsiflexion (0.00, 95% CI 0.00 to 0.77) and left hip joint adduction moment (0.00, 95% CI 0.00 to 0.75). The SEM values ranged between 0.1 Nm/Kg to 14.7 Nm/Kg and averaged between 0.1 Nm/Kg and 0.1 Nm/Kg.

4.4 Discussion

The purpose of this study was to evaluate the inter-session reliability and measurement error of a 3D gait analysis protocol in a group of CP children, in order to better understand the causes of intrinsic and extrinsic variation. Knowing this variability is crucial to improve clinical analysis that supports decision-making in the rehabilitation process.

Ferrari et al. [17] have found that when comparing five protocols on the same gait cycles, the main cause for the variability of outcomes between variables was the biomechanical model used and its definitions, regardless of the number of raters or even different laboratories. These different biomechanical models make it more difficult to compare results between reliability studies, as they present different sources of variability [17]. Repeated testing of a single subject allows for a clinical usefulness of the data, since it provides some understanding into the extent of variation of the measured outcomes that can be expected due to the pathology and those that are truly a consequence of a therapeutic intervention [15].

Despite extreme caution and compliance with the protocol instructions regarding the marker placement procedure, some inconsistency is still unavoidable [16], while possible sources of error can occur due to subjects' natural oscillations or skin motion [13] or movement between the skin markers and the underlying bones [50,51]. This source of error is totally disruptive for the joints with a limited range of motion, such as knee abduction–adduction, internal–external rotation, and linear displacements [52,53].

CP children can demonstrate different gait patterns in each leg. This occurs not only in unilateral spastic CP, where each legs presents different kinematic values [23], but also in some bilateral spastic CP children with an asymmetrical gait pattern, combining at least two different types of gait pattern [48]. A previous study by Mackey et al. [26] used the 6DoF Cleveland marker set with unilateral CP children and presented similar results at both normal and hemiplegic limbs, where the highest repeatability was at the sagittal plane (CMC values of 0.96–0.99) and lower in the transverse and frontal planes (CMC ≥0.7), In this study, the CP children presented different gait patterns (Table 4-1): five had bilateral spasticity, two had unilateral spasticity with their right limb affected and one was affected in the left limb, which contributed to some degree of variation of the data. The overall ICC results of kinematic and kinetic variables were lower on the right side, which can indicate that—to some degree—the instability of the affected lower limbs could influence the propagation of the STA. Reinschmidt and co-investigators reported that the soft tissue motion can originate additional movement, resulting in an overestimation in kinematic peak values of the segments by as much as 100% [54]. This is in accordance with our research, where a larger variation was noted in the transverse and frontal planes of the knee (Table 4-4). In the 6DoF models it is assumed that the limbs' segments are independent and do not share a fixed joint centre, which often originates non-physiological translations between the proximal and distal bones at some joints [22]. However, in pathological gait, care should be taken because non physiological movements may occur.

Typically, true equinus gait patterns constrain CP children to stand with the ankle in a neutral position [48]. However, according to Schlough et al. [55] when passive dorsiflexion is detected in the clinical examination, it is possible for some subjects to walk with their feet flat on the ground upon request. This variability in walking pattern during development is considered typical. Nevertheless, when unable to perform heel contact, some biomechanical compensation is detected, mainly in the coordination of movement at the hip, knee and ankle joints. In this study, one subject presented mild spastic diplegia and a considerable gastrocnemius tone (as seen in Table 4-1), which often shows similar characteristics to idiopathic toe walking. In the first session, the subject was able to perform a normal heel strike at initial contact and during the stance phase of walking. However, during the dynamic trials in the second session, the gastrocnemius stiffness was significantly higher which caused some motion restriction at the ankle. As, in the static calibration trial, the subject was able to stand with both feet flat on the floor, the range of motion differences were wider from the start. The magnitude of this variation is visible in the scatter plots of the dorsi/plantar flexion (Figure 4-2a; 4-2b; 4-2c; 4-2d). When we compare the kinematic data between sessions, there was an increase of 8° in hip flexion, a decrease of 13° in knee flexion and a total absence of ankle dorsiflexion in both lower limbs. These results are in accordance with the study of Hicks et al. [56] where CP children with toe walking often exhibited increased hip flexion and a decrease in knee flexion throughout the walking cycle. Furthermore, excessive plantar flexion may be responsible for changes in flexion, internal rotation and adduction of the hip as well as in the pelvic anterior tilt [33] which explains the reduced ICC on left and right anterior tilt (0.40 and 0.51, respectively) compared with the other kinematic variables of this segment, as seen in Table 4-4.

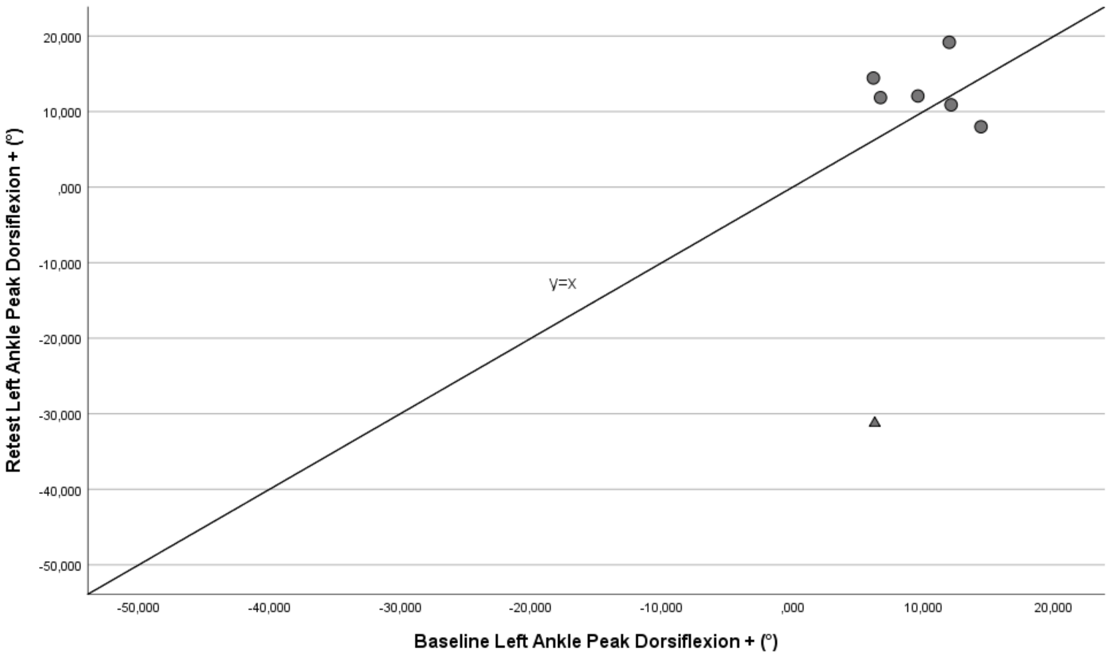

Figure 4-2a: Scatter plot for left ankle peak joint angles for dorsiflexion. Subject with increased gastrocnemius stiffness values is represented with a different symbol from the rest.

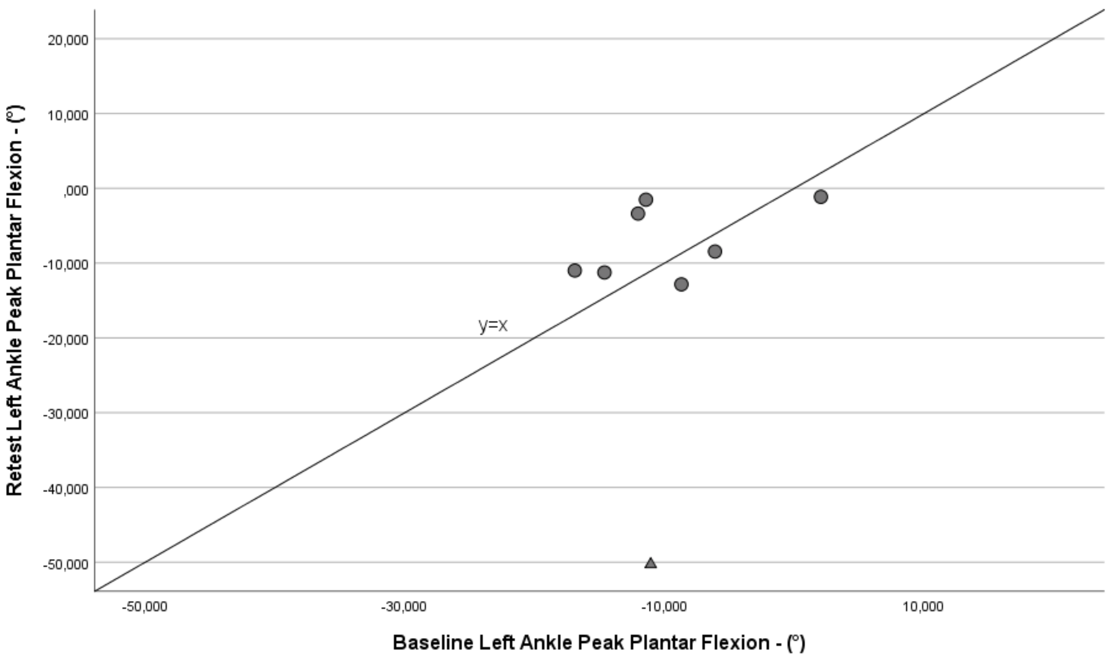

Figure 4-2b: Scatter plot for left ankle peak joint angles for plantar flexion. Subject with increased gastrocnemius stiffness values is represented with a different symbol from the rest.

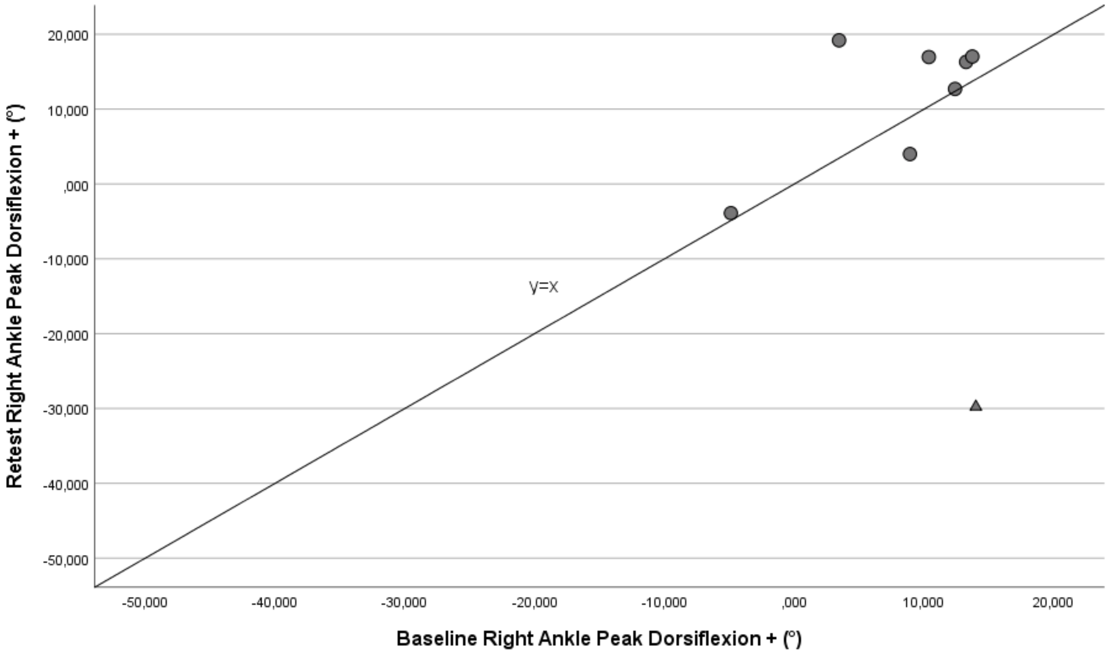

Figure 4-2c: Scatter plots for right ankle peak joint angles for dorsiflexion. Subject with increased gastrocnemius stiffness values is represented with a different symbol from the rest.

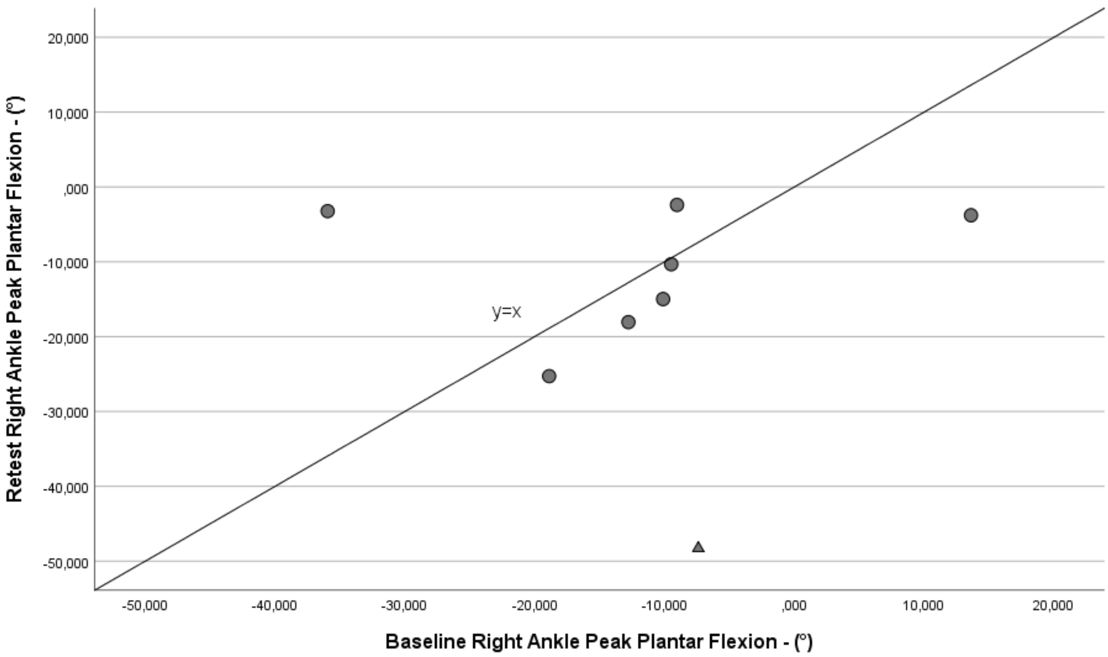

Figure 4-2d: Scatter plots for right ankle peak joint angles for plantar flexion. Subject with increased gastrocnemius stiffness values is represented with a different symbol from the rest.

Yet, due to co-spasticity of the muscles causing reciprocal movements across the joints and originating a wider variation in kinematic data, CP children are not able to change joint moments which results in a more reliable measure between the two assessment days [57]. This is evident in our results where the kinetic variables presented less variation (Table 4-5), in accordance with similar studies [16,23]. Although there is no reliability analysis published with a 6DoF model and kinetics variables, these results may be partially attributed to the small variations of the anthropometric measurements. Even though the two recorded sessions occurred several days apart, there was a small variation in marker placement between sessions (Table 4-2). Anthropometric measurements were considered excellent regarding ICC (ICC average ≈0.98) and an absolute error of approximately 4 mm.

4.5 Limitations

The number of CP children included in similar studies varies from 5 to 20 [23–26,44] and even though this gait protocol was performed with 8 CP children, the analysis of the right and left legs imply distinguished experiments, involving independent landmark identification, marker attachment, anthropometric measurements, and data processing [17]. Consequently, the current research should be considered as an independent analysis of sixteen legs.

Given that every gait research laboratory uses its own marker set and gait model, in order to compare gait analysis data, all the specific methodology used in each process must be considered. Regardless of the set of techniques chosen, there will always be different measurement errors that can influence the outcomes and consequently, a clinical interpretation. These differences have a greater impact in the kinematic and kinetic outcome measures (e.g., joint angles and moments). Thus, gait protocols should be described in detail to allow a contextualized interpretation of the results and comparison between similar investigations. This should be done in a critical manner on all the variables during the gait cycle, rather than only interpret the absolute values presented, regardless of the measures of repeatability or correlation used [15]. It is of great relevance when it comes to gait assessment of CP children who have an intrinsic gait variability due to their neuromuscular impairments. In these cases, it is crucial to differentiate the methodological errors (raters' error) from the participants' natural variability and from the effect of a rehabilitation process.

Due to the different gait analysis protocols used, the influence of the number of gait cycles in test–retest reliability measurements [11] remains to be determined. Although in general, repeatability increases with a higher number of gait cycles, this is true mainly for the kinematic data. All the time-distance and kinetic parameters do not reveal significant differences from the fifth gait cycle onwards. In addition, the assessment of more than five gait cycles in a clinical setting may be difficult to accomplish due to the preparation of the subject [34]. Regarding CP children, this can be a very complex and difficult task, therefore the five gait cycles used in this protocol were shown to be quite good in achieving reliable results.

4.5 Conclusions

This study indicates wide-ranging reliability values for lower limb joint angles and joint moments of force during gait, especially for frontal and transverse planes. Although the use of a 6DoF-CAST in CP children was shown to be a feasible method, the gait variation that can be observed between sessions in CP children seems to be related not only to the extrinsic factors but also to their different gait patterns and affected sides. In future research, it could be interesting to assess the reliability of these models using different groups of subjects, according to their gait pattern, for instance. These models and their technical characteristics still require some improvements in order to support clinical decision-making.

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Chapter 5

5. The use of Gait Profile Score (GPS) in detecting the effects of Ankle-foot Orthoses in children with Cerebral Palsy

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Abstract

Background: The ankle-foot orthosis is the most common non-invasive therapeutic intervention used to correct gait deviations, especially in children with Cerebral Palsy. Several studies demonstrated that its use improved several spatio-temporal and kinematic outcomes. However, the biomechanical reports can be complex and may need some experience to correctly interpret the results. Gait deviations indices, such as the Gait Profile Score intend to summarize some clinically meaning parameters and clarify in which way an ankle-foot orthosis impacts the pathological gait in children with Cerebral Palsy. The aim of this study was to assess if the gait profile score can reflect the effect of the AFO-use in spatio-temporal and kinematic outcomes on a sample of children with cerebral Palsy while walking.

Methods: This was a prospective study of children with spastic Cerebral Palsy (unilateral and bilateral). All children had an AFO prescription (Solid ankle-foot orthosis; Dynamic ankle-foot orthosis or Hinged ankle-foot orthosis) but never used any type of AFO. The primary outcomes were some spatio-temporal parameters, the Gait Profile Score and de Gait Variable Scores, collect in a one-day session and with different conditions (Barefoot and AFO-use). Data collection was performed with the use of 14 infrared, high-speed cameras with a frequency rate of 100 Hz.

Results: Eight children with spastic Cerebral Palsy (three unilateral and five bilateral) with an age range: 4-10 years (height 1.17 ± 0.14 m; mass 24.25 ± 8.26 kg) and grades I and II in the Gross Motor Function Classification System participated in this study. The trials were performed on a one-day session in each condition. Overall, gait speed increased in three subjects (0.089 ± 0.034) and decreased in five subjects (0.177 ± 0.129). GPS score decreased in five subjects (3.38 ± 2.3) and increased in three subjects (2.1 ± 1.1). According to the indicated MCID value (1.68°), four subjects showed an improvement in the GPS overall score of equal or superior value, ranging from 1.7° to 6.6°, and two subjects worsen their GPS overall values.

Conclusions: Our findings suggest that GPS detected clinical meaningful effects of the AFO-use in gait variables in unilateral and bilateral Cerebral Palsy children. However, due to the heterogeneity of this population, a broader classification system should be used across all gait analysis studies to better understand what type of changes should be expected according to the type of AFO. Regarding GPS, there should be caution for its use in the assessment of AFO effects in children with CP, especially due to methodological considerations.

Keywords: Cerebral Palsy; Children; AFO; Orthotics; Gait index.

5.1 Introduction

Cerebral Palsy is the most common physical disability of childhood with and incidence of 2-2.5 per 1000 live births [1]. It is a heterogeneous disorder with many associated comorbidities, particularly in the lower limbs (unilateral or bilateral) which originates different gait patterns [2,3]. The ankle–foot orthoses (AFO) are the most common type of non-invasive interventions in the gait of children with CP to prevent the development or progression of deformities and to improve dynamic efficiency of the child's gait [4]. However, the wide variety of AFO types, and the different structural characteristics, such as material stiffness and design [5,6] makes it more difficult to support the prescription process, which is commonly influenced by a strong empirical factor [7].

Although gait research demonstrates the impact of AFO in gait variables [8–10], the lack of effectiveness evidence in a large scale of the interventions in use within standard care is still a problem for children with CP [11]. This occur mainly due to the heterogeneous outcome measures applied across all gait studies [12]. Regarding this, there is an existing need to improve the reporting detail and protocol transparency regarding AFO interventions [13], allowing a more precise assessment of which ones are most effective in improving the functional outcomes of children with CP [7,14].

3D instrumental gait analysis is a method to collect several kinematic and kinetic variables to understand the level of functional limitation due to pathology. However, the analysis and interpretation of this data can be a quite complex process. Despite its objectivity with the usual summary of measures and graphic displays, the large number of specific parameters may be complicated to interpret for inexperient clinicians. Thus, there is a growing need for the use of summary gait indexes, that can provide a single score of gait function for clinical evaluation, screening, and outcome assessment [15]. There are several gait indices developed based on 3D motion capture kinematics like the Gait Deviation Index (GDI) [16] originated from clinical gait analysis data, which resumes the gait quality of patients with CP into a single index. Among such indexes, the gait profile score (GPS) is a validated index, widely used in clinical gait analysis and research, consisting in a ponderation on the individual contribution of gait variables scores (GVS) collected during gait trials. This is different from other index, like the GDI, given that instead of assessing the RMS difference between the entire gait vector and the average gait vector for people with no gait pathology, it calculates the RMS difference for each single gait variable [17]. Finally, the movement profile scores (MAP) summarizes the overall deviation from normal gait, based upon the GVS of nine key relevant kinematic variables for the right and left legs [17,18].

Gait indices represent the overall gait pathology, and therefore, can potentially reflect the effect of AFO on alignment and gait [19]. McMulkin et al. demonstrated that the GPS is one of the most sensitive measures in assessing differences pre/post-treatment on several multiple clinical paediatric populations. However, this study focused in invasive treatments [20]. Recently, Joanna et al. found that the GDI was sensible to changes in spatio-temporal parameters and gait kinematics in spastic hemiplegic CP children using AFO [21].

Based on these considerations, our study intended to use of GPS to assess the quality of gait function with and without AFO, as well as to quantify gait deviations through MAP parameters in children with CP.

5.2 Methods

5.2.1 Subjects

A convenience sample of 8 children with spastic CP (5 bilateral, 3 unilateral, 6 male and 2 female, age range: 4-10 years) with grades I and II in the Gross Motor Function Classification System (GMFCS) [22], was recruited to participate in the study (Table 1). Inclusion criteria were: male and female children, between 4 and 16 years of age; able to walk independently with or without walking aids; cooperative and able to comply with simple orders; feet size between 20 and 33; who had a clinical recommendation to use an ankle foot orthosis, but have never used it before; who have not undergone orthopaedic surgery of the lower limb in the last 12 months, and who are not expecting to have a surgical intervention in the next 6 months; who were not given botulinum toxin in the last 6 months [23].

The protocol was approved by and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019). An informed consent was previously signed by the parent or the legal guardian of the participant.

5.2.2 Orthoses characteristics

Each child used their owned shoe with an AFO, which was selected on the basis of a previous study of Rodda and Graham [24]. The AFO were pre-fabricated (Figure 5-1) according to the orthopaedic company production guidelines and tunned to AFO-footwear combination parameters (i.e. angle of the ankle in the AFO, tibia inclination, calcaneal and forefoot positioning, shank-to-vertical-angle, among others [25]), by an experienced Certified Prosthetist and Orthotist.

Figure 5-1: A) Dynamic ankle-foot orthosis B) Solid ankle-foot orthosis C) Hinged ankle-foot orthosis. Accessed 25 November 2021, {https://www.orliman.com/en/dafo-2/}. **A)** DAFO can control varus or valgus deformities of the hindfoot and compensate for forefoot deformities. It provides arch support and hindfoot varus and valgus control. DAFO occasionally lessens footdrop in swing to some degree, even though it does not passively control sagittal plane ankle joint alignment. **B)** SAFO is used primary for completely incompetent plantarflexor function. It allows no ankle joint motion and is appropriate for use in this child whose strength and motor control distally are poor and whose balance mechanisms are impaired. **C)** HAFO uses a plantarflexion stop that can prevent footdrop in swing [25].

5.2.3 Study Design

The gait trials were performed in the Biomechanics and Functional Morphology Laboratory of the Faculty of Human Kinetics. Upon the participants' arrival, instruction was given about the protocol, the risks and benefits, as well as the informed consent. Initially, the participants' clinical history was reviewed, and a clinical exam was performed with the subject laid on the table, seated on a chair, or standing [26]. Motion capture was performed with 14 infrared, high-speed cameras (Qualisys Oqus 300, Qualisys AB, Gothenburg, Sweden) with a frequency rate of 100 Hz. Palpation was used to locate the subcutaneous anatomical landmarks on the participants [27] and subsequently to place the marker set. These were 1.25 cm spherical reflective markers with a 1.8 cm semi-flexible width base. Four marker clusters were attached to the lateral part of the thigh and shank to independently track anatomical landmarks of each segment allowing rotational and translational motion at the joints [28]. These types of markers were adequate for the general height of these children given the smaller body parts. Before each dynamic trial, a barefoot static trial in the standing position was recorded in order to determine the participant's joint centres and segmental reference systems, as well as segments' length [28]. Afterwards, the participant was instructed to walk barefoot (first part of the session) and wearing the AFO (second part of the session) along a 10 m corridor, unassisted at a self-selected pace. The dynamic trials ended when the child successfully achieved a minimum of five complete kinematic and kinetic walking cycles for each side [29–31], considering the natural variation in kinematic gait parameters [32].

5.2.4 Spatio-temporal and kinematic data collection and processing

The marker set that was used followed the calibrated anatomical system protocol (CAST) [27,33] and CODA pelvis [34]. It was used to reconstruct the pelvis and both lower limbs [32]. The 22 individual markers and four marker clusters of four embedded markers each, allowed the reconstruction of seven body segments: feet, shanks, thighs, and pelvis. Each segment is considered to be independent and to have six degrees of freedom [35]. Lower limb segment masses were determined according to Dempster [36] while the remaining inertial parameters were computed based on Hanavan [37]. Gait cycles were extracted using Qualysis Track Manager (QTM) (v2020.3 build 6020, Qualisys AB, Gothenburg, Sweden). The subsequent analysis and processing were performed using Visual 3D software (Professional Version v4.80.00, C-Motion, Inc., Rockville, MD, USA).

5.2.5 Gait Profile Score (GPS)

GPS is calculated from the GVS, namely pelvic tilt, rotation and obliquity, hip flexion-extension, adduction-abduction and rotation, knee flexion-extension, ankle dorsi- and plantar-flexion, and foot progression of each leg [13]. The GPS is normally distributed for the population without clinically meaningful gait deviations (mean 5.3°) [15]. The root mean square difference between a patient’s data and the mean value obtained from tests performed on the unaffected population is expressed in degrees. The presentation of each GVS generates a MAP (Table 3a and 3b) which describes the magnitude of deviation of the nine individual variables averaged over the gait cycle, thus providing insight into which variables are contributing to the GPS overall value [18]. Thus, convenience of the MAP and GPS components together with GPS is an advantage in its use in clinical practice, since it allows for a simpler overview of some complex kinematic data [18].

The GPS is calculated according to eq.5-1, where GPS is the root mean square average of the GVS variables:

$$\mathrm{GPS} = \frac{1}{N}\sum_{i=1}^{N} \mathrm{GVS}_i^2$$

Equation 5-1. Gait Profile Score calculation formula

Thus, the GPS result is an indicator of the overall quality of gait kinematics (increased GPS corresponds to a larger deviation from a physiological gait pattern). The authors[38] proposed a rationale for defining a minimal clinically important difference (MCID) for the GPS of 1.68. Regarding this MCID, we have calculated the GPS for two test conditions (barefoot and with AFO) as well as the MAP results for each child.

5.3 Results

The participants of the study were a convenience sample of eight children with CP (Table 5-1) able to walk independently (three hemiplegic, five diplegic; two females, six males; height 1.17 ± 0.14 m; mass 24.25 ± 8.26 kg). All the children completed the gait trials successfully and the GPS and MAP was calculated for each test condition.

Table 5-1: Subjects' characteristics and gait patterns.

				Left Lower Limb		Right Lower Limb	
Subject	**Affected Side**	**Height (m)**	**Mass (Kg)**	**Sagittal Gait Pattern**	**AFO**	**Sagittal Gait Pattern**	**AFO**
001	Bilateral	1.09	19.5	True equinus [2]	DAFO	True equinus [2]	DAFO
002	Unilateral	1.14	26	Normal	None	True equinus [3]	SAFO
003	Bilateral	1.32	26	Apparent equinus [2]	DAFO	Apparent equinus [2]	DAFO
004	Unilateral	0.98	13.5	True equinus [3]	HAFO	Normal	None
005	Bilateral	1.37	34	Apparent equinus [2]	SAFO	Apparent equinus [2]	SAFO
006	Unilateral	1.32	37	Normal	None	True equinus with recurvatum knee [3]	HAFO
007	Bilateral	1.06	15.5	True equinus [2]	SAFO	True equinus [2]	SAFO
008	Bilateral	1.10	18	Jump gait [2]	HAFO	Jump gait [2]	HAFO

5.3.1 Spatio-temporal outcomes

Table 5-2 summarizes the comparative temporal parameters in the barefoot and AFO-use condition. Unlike a gait index such as the Gillette Gait Index (GGI), where spatio-temporal parameters are used to calculate the final score [39], this does not occur in the GPS calculation [17]. Due to this fact, it is important to report self-selected speed during the gait trials [15,17] which reflects different domains of gait quality [17]. Overall, gait speed increased in three subjects (002, 005 and 006) and decreased in five subjects (001, 003, 004, 007 and 008). The step length increased in both lower limbs in subject 001, 003, 005 and 006. For subject 002, 004 and 007, step length increased on the right lower limb and decreased on the left side. In the case of subject 008, step length decreased on both lower limbs. Step time increase in both lower limbs in five subjects (001, 002, 003, 004 and 008) and decreased on the right lower limb on subject 005 and on the left lower limbs of the remaining subjects (005, 006 and 007).

Table 5-2: Temporal and spatial data.

				Left Lower Limb		Right Lower Limb	
Subject	**Condition**	**Gait Speed (m/s)**	**Cycle Time (s)**	**Step Length (m)**	**Step Time (s)**	**Step Length (m)**	**Step Time (s)**
001	Barefoot	1.033	0.790±0.051	0.411±0.021	0.400±0.060	0.380±0.038	0.379±0.072
	AFO	0.813	1.044±0.101	0.425±0.068	0.492±0.035	0.435±0.021	0.540±0.070
002	Barefoot	0.613	1.018±0.179	0.414±0.215	0.461±0.231	0.113±0.250	0.407±0.160
	AFO	0.657	1.050±0.118	0.328±0.090	0.462±0.109	0.355±0.043	0.602±0.050
003	Barefoot	0.908	0.907±0.097	0.444±0.060	0.463±0.061	0.379±0.020	0.443±0.045
	AFO	0.870	1.002±0.089	0.472±0.044	0.499±0.043	0.383±0.061	0.499±0.058
004	Barefoot	1.013	0.756±0.063	0.406±0.022	0.403±0.015	0.354±0.010	0.370±0.032
	AFO	0.893	0.894±0.051	0.387±0.012	0.467±0.022	0.412±0.025	0.423±0.035
005	Barefoot	0.826	1.056±0.053	0.422±0.024	0.506±0.048	0.461±0.062	0.551±0.025
	AFO	0.952	1.027±0.104	0.452±0.051	0.483±0.037	0.531±0.032	0.540±0.075
006	Barefoot	0.846	0.977±0.069	0.414±0.030	0.441±0.024	0.413±0.019	0.536±0.054
	AFO	0.942	1.005±0.053	0.478±0.025	0.430±0.034	0.467±0.027	0.574±0.027
007	Barefoot	0.824	0.709±0.048	0.252±0.026	0.352±0.029	0.325±0.041	0.360±0.043
	AFO	0.724	0.888±0.088	0.154±0.383	0.341±0.223	0.495±0.394	0.520±0.209
008	Barefoot	0.579	1.032±0.134	0.295±0.037	0.549±0.100	0.303±0.074	0.482±0.043
	AFO	0.485	1.171±0.146	0.282±0.097	0.556±0.009	0.269±0.032	0.548±0.070

5.3.2 Gait Profile Score

Table 5-3 summarizes the overall GPS score. According to these results, GPS score decreased in five subjects (3.38 ± 2.3) and increased in three subjects (2.1 ± 1.1). According to the indicated MCID value (1.68°)[38], four subjects (001, 002, 006 and 007) showed an improvement in the GPS overall score of equal or superior value, ranging from 1.7° to 6.6°. Subject 008 showed a minor improvement of 0.1°. On the other hand, subjects (004 and 005) worsen their GPS overall values from 3.4° to 1.9°, respectively. Subject 003 also had a slight inferior value of 0.9°.

Table 5-3: Gait Profile Score.

Subject	Condition	GPS Left	GPS Right	GPS Overall	GPS Overall Diff
1	Barefoot	7.4	10.2	9.2	-1.7
	AFO	6.4	8.4	7.9	
2	Barefoot	23.6	12.2	19.3	**-6.6**
	AFO	11.9	12.6	12.7	
3	Barefoot	13.5	11.2	13.2	+0.9
	AFO	12.9	13.2	14.1	
4	Barefoot	8.1	9.8	9.1	**+3.4**
	AFO	13.9	9.6	12.5	
5	Barefoot	7.5	7.4	8.0	**+1.9**
	AFO	10.0	7.6	9.9	
6	Barefoot	9.4	17.2	14.7	**-4**
	AFO	10.5	10.2	10.7	
7	Barefoot	16.2	15.5	17.2	**-4.6**
	AFO	12.2	9.9	12.6	
8	Barefoot	10.6	16.1	16.0	-0.1
	AFO	12.3	17.5	15.9	

Tables 5-4a and 5-4b presents the results of GVS of the left and right lower limb, respectively, and the GVS*diff* between conditions.

According to Table 5-4a, MAP shows that the left lower limbs have worsen their GVS, mainly hip flexion and knee flexion. As the first parameter, only subject 001 and 003 had a slight improvement, and all other subjects aggravated GVS values, ranging GVS*diff* from 0.3° to 9.1°. As for the latter, only subject 001 improved the GVS in 4.3°. The ankle dorsiflexion also had a wider range of inferior GVS values between conditions, from 1.7° to 7.9°. All the GVS*diff* were higher than the stabilised MCID (1.6°). On the other hand, GVS value of foot progression shows an improvement on most of the subjects, with the exception of subject 004 (11.4°) and subject 005 (6.1°). From the 72 GVS*diff* values, 40 GVS*diff* were ≥1.6°, where 12 GVS improved and 28 GVS aggravated between conditions.

Regarding Table 5-4b, none of the GVS shows a tendency across all subjects, whereas to the same GVS, there are improvements and decreases for the respective values. This is evident in foot progression, where subject 003 had a decrease in the GVS between conditions (9.1°) and subject 006 improved the same GVS in 24.3°. The same scenario occurs in ankle dorsiflexion, with improvements ranging from 1.3° to 29.1° and decreases from 0.3° to 11.9°. Concerning this GVS, subject 007 had the improvement of 29.1° and subject 008 aggravated it in 11.9°. From the 72 GVS*diff* values, 37 GVS*diff* were ≥1.6°, where 18 reduced and 19 aggravated their values between conditions.

Table 5-4a: Left Lower Limb Movement Analysis Profile.

Left Lower Limb																			
Subject	**Condition**	**GVS Pelvis Tilt**	**GVS Diff**	**GVS Hip Flexion**	**GVS Diff**	**GVS Knee Flexion**	**GVS Diff**	**GVS Ankle Dorsiflexion**	**GVS Diff**	**GVS Pelvis Obliquity**	**GVS Diff**	**GVS Hip Abduction**	**GVS Diff**	**GVS Pelvis Rotation**	**GVS Diff**	**GVS Hip Rotation**	**GVS Diff**	**GVS Foot Progression**	**GVS Diff**
1	Barefoot	4.1	+1.9	6.8	-1.5	10.2	-4.3	4.5	-0.4	2.7	-0.1	3.5	-0.1	5.1	-0.2	7.5	+1.2	12.4	-2.5
	AFO	6.0		5.3		5.9		4.1		2.6		3.4		4.9		8.7		9.9	
2	Barefoot	2.3	+3.1	6.9	+1.2	7.7	+4.3	19.0	-9.4	5.8	+0.1	9.0	0.0	20.1	-6.6	26.3	-9.2	40.8	-24.3
	AFO	5.4		8.1		12.0		9.6		5.9		9.0		13.5		17.1		16.5	
3	Barefoot	4.1	-1.2	4.1	-0.8	15.6	+3.7	6.8	+4.2	6.1	-1.6	9.0	-1.0	7.0	+2.3	5.0	0.0	32.2	-10.2
	AFO	2.9		4.9		19.3		11.0		4.5		8.0		9.3		5.0		22.0	
4	Barefoot	4.3	+5.5	8.5	+9.1	14.1	+11.7	6.9	+7.9	1.5	+0.1	4.0	-1.0	3.6	+1.0	13.7	-3.1	5.1	+11.4
	AFO	9.8		17.6		25.8		14.8		1.6		3.0		4.6		10.6		16.5	
5	Barefoot	3.2	-1.1	5.3	+0.3	15.2	+4.1	5.4	+3.4	4.1	-0.1	5.2	-0.9	2.9	+1.8	8.3	-2.5	7.9	+6.1
	AFO	2.1		5.6		19.3		8.8		4.0		4.3		4.7		5.8		14.0	
6	Barefoot	10.4	-0.1	15.5	+0.8	12.3	+1.9	9.9	+1.7	4.1	+2.4	6.9	+4	5.9	-0.2	5.4	+1.1	4.7	0.0
	AFO	10.3		16.3		14.2		11.6		6.5		10.9		5.7		6.5		4.7	
7	Barefoot	4.6	-2.8	8.6	+3.3	8.9	+10.2	41.9	-32.8	3.5	-1.0	3.5	+2.6	12.7	+0.9	6.2	-1.0	11.2	-2.3
	AFO	1.8		11.9		19.1		9.1		2.5		6.1		13.6		5.2		8.9	
8	Barefoot	11.2	-1.8	9.5	+1.6	15.6	+4.7	9.6	+4.2	3.1	+1.1	5.8	+1.4	9.5	+2.9	7.7	+5.3	10.5	-0.8
	AFO	9.4		11.1		20.3		13.8		4.2		7.2		12.4		13.0		9.7	

GVS *Diff* (°) – difference of the GVS score from the first session (barefoot) to the second session (AFO-use); (-) minus – improved the GVS; (+) aggravate the GVS

Table 5-4b. Right Lower Limb Movement Analysis Profile

Right Lower Limb																			
Subject	**Condition**	**GVS Pelvis Tilt**	**GVS *Diff***	**GVS Hip Flexion**	**GVS *Diff***	**GVS Knee Flexion**	**GVS *Diff***	**GVS Ankle Dorsiflexion**	**GVS *Diff***	**GVS Pelvis Obliquity**	**GVS *Diff***	**GVS Hip Abduction**	**GVS *Diff***	**GVS Pelvis Rotation**	**GVS *Diff***	**GVS Hip Rotation**	**GVS *Diff***	**GVS Foot Progression**	**GVS *Diff***
1	Barefoot	4.0	+1.8	8.1	-1.4	9.9	-1.8	5.7	-2.1	3.3	+0.1	6.2	-1.3	7.3	-1.2	21.5	-4.8	10.5	-1.3
	AFO	5.8		6.7		8.1		3.6		3.4		4.9		6.1		16.7		9.2	
2	Barefoot	2.3	+4.5	10.0	+0.5	9.7	-1.0	14.6	-8.7	5.6	+2.0	4.3	+3.7	21.0	-8.3	10.5	+11.7	12.7	-0.4
	AFO	6.8		10.5		8.7		5.9		7.6		8.0		12.7		22.2		12.3	
3	Barefoot	4.1	-1.3	7.2	+0.2	21.8	+3.7	7.4	+3.7	5.3	-0.4	9.3	-1.6	6.2	+4.7	5.4	-1.0	8.7	+9.1
	AFO	2.8		7.4		25.5		11.1		4.9		7.7		10.9		4.4		17.8	
4	Barefoot	5.7	+3.9	8.9	+6.3	13.1	+1.1	7.6	-1.3	2.5	-0.6	3.5	+0.2	4.4	+2.4	19.4	-8.0	5.8	+1.2
	AFO	9.6		15.2		14.2		6.3		1.9		3.7		6.8		11.4		7.0	
5	Barefoot	2.9	-0.8	7.1	+0.5	15.8	-0.7	5.8	+0.3	4.3	+0.1	7.1	+1.0	3.2	+0.7	4.4	+1.7	4.9	+1.2
	AFO	2.1		7.6		15.1		6.1		4.4		8.1		3.9		6.1		6.1	
6	Barefoot	10.5	-0.4	10.9	-2.2	9.6	-2.4	7.5	+7.0	4.2	+1.7	5.8	-1.5	8.8	-1.2	12.4	-4.0	41.1	-24.3
	AFO	10.1		8.7		7.2		14.5		5.9		4.3		7.6		8.4		16.8	
7	Barefoot	4.5	-2.7	9.9	-2.7	9.4	+3.4	39.0	-29.1	3.2	-0.5	4.3	-0.9	8.7	+2.5	12.1	-5.2	10.5	-2.0
	AFO	1.8		7.2		12.8		9.9		2.7		3.4		11.2		6.9		8.5	
8	Barefoot	10.9	-1.3	7.0	+0.2	22.1	+9.6	11.3	+11.9	2.5	+0.6	5.2	-1.2	16.1	-5.8	26.6	-1.3	21.7	-5.5
	AFO	9.6		7.2		31.7		23.2		3.1		4.0		10.3		25.3		16.2	

GVS *Diff* (°) – difference of the GVS score from the first session (barefoot) to the second session (AFO-use); (-) minus – improved the GVS; (+) aggravate the GVS

5.4 Discussion

The aim of this study was to use a summarize measure to evaluate the effects of AFO on the kinematic variables of children with CP walking in a straight line at self-selected speed.

Of the analysed subjects where an improvement in the GPS was clinically significantly (Table 5-3), gait speed outcomes were not concordant to data presented in similar studies. There is a general consensus that the increase in gait speed [40,41] is considered to be an important factor in the children's motor development given its approximation to the data of typically developed children. However, Subject 001 and 007 – both bilateral with a True equinus gait pattern - decreased their gait speed between trials setup (0.22 and 0.10, respectively). Morris et al. [42] developed a bibliography review in search of an consensus regarding the AFO use in children with CP, and reported that the majority of studies included in his analysis, found increases in gait speed of unilateral spastic children with CP, but did not found alterations in gait speed in bilateral children with CP. This highlights the importance in reporting the characteristics of the children that participate in this type of research, given that not all the results in rehabilitation should be compared crosswise.

Previous studies have demonstrated evidence supporting the use of the GPS to describe the pathological gait in children with CP and the effects of surgery [17,43] or AFO-use [44]. The latter showed that bilateral children with spastic CP did not evidence different values regarding the overall GPS between barefoot and AFO-use condition. In our study, of the five subjects with bilateral CP, subject 001 and subject 007 presented a meaningful clinical value improvement ≥1.6° (1.6° and 4.6°, respectively). On the other hand, subject 005 had a decreased in the overall value of 1.9°. Several factors can explain such differences between studies. The referenced study [44] did not clarify which gait patterns may influence the therapeutic purposes and AFO effects [45]. We have identified three types of gait patterns in our sample group, whereas two presented a true equinus gait, one jump gait and two subjects with apparent equinus [24], and consequently the AFO type was also different. This fact highlights the importance of the uniformization and clarification of the sample study characteristics and AFO prescription to enable a better comparison between studies with less outcomes variability [7].

As presented by Danino et al. [19] GPS measures the deviations from normal kinematics, and, as expected, either with or without the use of an AFO, the GPS overall score was in most cases, greater than the control group. An AFO prescription tends to accept some abnormal gait parameters with the expectation to improve other spatio-temporal and kinematic parameters. An example of this fact is the physiological behavior of the ankle joint during gait, in which the

AFO-use can impose mechanical constraints on the ankle joint, bringing the sagittal gait parameters closer to normal gait. However, neither the foot accomplishes any of the three rockers during stance, nor substantial changes in kinematics or kinetics at the pelvis, hip, or knee have been identified [25].

It is not clear what were the walking conditions (barefoot or with shoe) of the assessed subjects (typically developed children and children with gait deviations) in the GPS validations studies [17,38]. Such feature is fundamental to understand the scope of this tool in assessing the effects of AFO in children with CP. Either in shoe condition or with an AFO-footwear combination, these will significantly change the sagittal gait parameters [46], furthering them from the start of the normal values. The role of the footwear becomes at least equal to that of the orthosis because footwear design is so highly influential on the kinematics of the segments throughout the gait cycle. Consequently, the AFO-footwear combination is fundamental to the effectiveness of such intervention. Features, like “heel-sole differential” or “shank vertical angle” significantly influence the shank inclination at midstance, and if not properly intervened can forcibly impose a greater or a smaller amount of inclination [46]. Thus, some caution should be considered when using only gait indices, as the GPS, to analyze the quality of gait.

5.5 Conclusions

Our study findings suggest that the gait index GPS detected clinical meaningful effects of the AFO-use in gait variables in unilateral and bilateral Cerebral Palsy children. However, due to the heterogeneity of this population, a broader classification system should be used across all gait analysis studies to better understand what type of changes should be expected according to the type of AFO. Regarding GPS, there should be some precautions for its use in the assessment of AFO effects in children with CP due to methodological considerations.

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Chapter 6

6. Effect of different pose estimation algorithms in gait kinematics of cerebral palsy children using AFO: A case study

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Current status: **In preparation to be submitted.**

Abstract

Objective: To compare two pose estimation algorithms used in clinical gait analysis and discuss sources of gait deviations in a case of a children with cerebral palsy wearing a DAFO.

Methods: A case study of a 6-year-old male diagnosed with spastic bilateral cerebral palsy undergoing a gait analysis while wearing a supramalleolar DAFO. A 3D motion capture with the CAST marker set and CODA pelvis was used during an AFO-use condition walking along a 10 m walkway ate self-selected speed pace. Two different pose algorithms (Segment optimization and Global optimization) were used to quantify lower limb joint kinematics.

Results: There is some indication that different pose algorithms can differ in the outcomes according to which plane of movement is being assessed. Apparently, GO have a propensity to overestimate sagittal plane movements and SO tends to overestimate movements in the frontal and transverse planes.

Conclusions: The orthoses configuration to prevent plantarflexion thus improving clearance in swing and first ankle rocker seems to be more in accordance when using a model with constraints that closely match that configuration.

6.1 Introduction

Errors of measurement can occur in any type of gait analysis, however its importance is largely more relevant in clinical testing, where it is important to have data in which the clinicians can rely upon to take informed decisions in the therapeutic process of patients [1]. One of the major causes for errors in measurement is the soft tissue artifact (STA). Human gait analysis assesses the movement of skin point and no that of the underlying bone, thus there is a need to minimize the movement of skin relative to the bone [2,3]. Different methods are used to minimize STA. Its efficacy depends on which joint constraints are applied in accordance with a specific model [4]. Pose estimation algorithms (PEA) are fundamental when using rigid bodies' models to assess the kinematics of human movement [5–7]. According to Robertson et al. [8] segment optimization (SO) and global optimization (GO) are the most effective class of algorithms for estimating pose. Even though these issues have been addressed in pathological populations [9], among which children with Cerebral Palsy (CP) [10,11]. The most common non-invasive treatment in gait deviations is the AFO [12] and its effectiveness has been analysed in several studies [13]. However, the used PEA of the lower limb segments usually follows the same modelling approach despite the constraints that the orthosis may cause. To overcome this limitation, the purpose of this work is to compare the kinematic data of two different PEA models (Global Optimization and Segment Optimization) [14] during gait of children with CP wearing AFO.

6.2 Methods

6.2.1 Study design

The study was developed in a one-day trial. The first part of the protocol included a clinical history review. After a clinical exam, which was performed by an experienced physiotherapist, with the subject laid on the table, seated on a chair, or standing [15] to evaluate bone and joint deformities, muscle length and force, selective motor control and spasticity. A pre-fabricated DAFO was then tunned to AFO-footwear combination parameters, by an experienced Certified Prosthetist and Orthotist. Before the gait trials, an acclimatization period was given [16]. The subject was instructed to walk along a 10-meter corridor, at self-selected speed wearing the DAFO with the usual footwear, referred to as the AFO-footwear combination. After 5 successful trials of both lower limbs, the trial session was over.

6.2.2 Subject

A 6-year-old male diagnosed with spastic bilateral CP was referred for gait analysis to assess the first time use of a type of AFO. The subject presented a True equinus gait pattern [17], with grade I in the Gross Motor Function Classification System (GMFCS) [18]. The clinical exam found no abnormal function of the lower limbs, with the exception of the ankle foot joints. According to the score in Modified Ashworth Scale [19], the left ankle and right ankle joint, revealed spasticity in the gastrocnemius in a determine plantarflexion degree (1+ at 10° and 1+ at 30°, respectively). The protocol was approved by and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019). An informed consent was previously signed by the parent or the legal guardian of the participant.

6.2.3 Orthoses characteristics

The DAFO is a thin flexible supramalleolar orthosis with a posterior cut-out allowing several degrees of movement in the sagittal plane [20]. This type of AFO is recommended in cases of mild to moderate spastic bilateral CP [21]. The DAFO is effective at correcting dynamic equinus in stance and swing and less restrictive of ankle movement compared with other types of AFO [22]. DAFO application is more significant in cases of hindfoot and midfoot deformities.

It provides an arch support, and hindfoot varus and valgus control. The DAFO can reduce plantarflexion in the swing phase, even though it does not passively control sagittal plane ankle joint alignment [23].

6.2.4 Motion capture protocol

Gait analysis was performed with the use of a 14 camera-based optoelectronic system Qualysis Track Manager (QTM) (v2020.3 build 6020, Qualisys AB, Gothenburg, Sweden) at 100Hz and 3 force plates (FP4060-07, FP4060-10, Bertec, Columbus, OH, USA) embedded into the laboratory walkway [6]. Palpation was used to locate the subcutaneous anatomical landmarks on the participants [7] and subsequently to place the marker set. These were 1.25 cm spherical reflective markers with a 1.8 cm semi-flexible width base. Four marker clusters were attached to the lateral part of the thigh and shank to independently track anatomical landmarks of each segment allowing rotational and translational motion at the joints [24], according to CAST protocol [7,25] and CODA pelvis [26], allowing the reconstruction of seven body segments [27]. Each segment is considered to be independent and to have six degrees of freedom [28].

6.2.5 Pose estimation algorithms

The pose of the lower limbs and pelvis was estimated using two algorithms: 1) a global optimization (GO) algorithm and 2) a segmental optimization (SO) algorithm.

In the GO algorithm, also known as Inverse Kinematics, the model is built with physically realistic constraints [29,30]. Inverse Kinematics searches for the POSE (position and orientation) that best matches the differences between the measured and the model-determined marker positions. This algorithm is useful if we want to minimize errors due to soft tissue artifact, for instance. However, careful should be taken regarding clinical conditions, where abnormal movements may occur at the joints, so they won't be incorrectly masked. Given a set of measured marker coordinates $\boldsymbol{P}$ on a data frame, the GO at the system level finds a set of generalized coordinates $\boldsymbol{\vartheta}$ such that the error function (eq.6-1) is minimized where $\boldsymbol{W}$ is a positive-definite weighting matrix.

$$f(\vartheta) = \sum_{i=1}^{m}[(P - P'(\vartheta)]^T W[(P - P'(\vartheta)]$$

Equation 6-1: Global optimization algorithm (adapted from [30]).

where $\boldsymbol{P}'(\boldsymbol{\vartheta})$ is the corresponding set of marker coordinates calculated by the following transformation: $P'(\vartheta) = T(\vartheta)P^*$, where $\boldsymbol{T}(\boldsymbol{\vartheta})$ is the combined transformation matrix from segment-embedded frames to laboratory frame and is calculated by the model for a given $\boldsymbol{\vartheta}$.

In the SO algorithm, all the 6DoF for each segment are estimated. Thus, every segment needs at least three non colinear tracking markers. Each segment is independent and there is no linkage between them. SO estimates the segment pose in terms of its transformation matrix by minimizing marker array deformation from its reference shape in a least-squares sense [31]. The transformation is obtained by solving eq.6-2 and eq.6-3.

$$f = \sum_{i=1}^{m} (Rx_i + v - y_i)^T (Rx_i v - y_i)$$

Under the orthonormal constraint

$$R'_{seg} R_{seg} = I$$

Equations 6-2 and 6-3: Segmental optimization algorithm (adapted from [31]).

Where $\boldsymbol{x_i}$ and $\boldsymbol{y_i}$ are position vectors of marker $\boldsymbol{i}$ in the marker array at the reference and current positions, respectively, $\boldsymbol{R}$ is the rotation matrix, $\boldsymbol{v}$ is the translation vector and $\boldsymbol{m}$ is the number of markers. The orthonormal constraint indicates that the transformation is orthogonal [31].

6.2.6 Data Processing and Models

Gait cycles were extracted using Qualysis Track Manager (QTM) (v2020.3 build 6020, Qualisys AB, Gothenburg, Sweden). The subsequent analysis and processing were done using Visual 3D software (Professional Version v4.80.00, C-Motion, Inc., Rockville, MD, USA). Different pose algorithms – 6DoF and IK model – were used to reconstruct each segment. The 6DoF model [25] was created to allow all rotations and translations in the segments. The IK model [30] the pelvis had 3 rotational DoF and 3 translations; no translations were allowed to the thigh, shank and foot segments, the hip and the knee joints were allowed to rotate in the 3 axes, and the ankle joint was only allowed to rotate in the medio-lateral axis (dorsiflexion and plantarflexion movements). The latter was intended to recreate the constraints imposed by the specific AFO that the participant was wearing (DAFO). Lower limb segment masses were determined according to Dempster [32] while the remaining inertial parameters were computed based on Hanavan [33].

6.3 Results

In this section it will be presented the preliminary results of this study.

Table 6-1: Participant characteristics.

Morpho functional parameters				
Affected Side	**Height (m)**	**Mass (Kg)**	**Sagittal Gait Pattern**	**GMFCS**
Bilateral	1.09	19.5	True equinus [17]	I

Due to the fact of being a pre-fabricated AFO, the tunning and optimization measures were the followed: angle of the ankle in the AFO, tibia inclination, calcaneal and forefoot positioning, shank-to-vertical-angle [23].

The lower limb joint angles in the sagittal, frontal, and transverse planes are shown in Figures 6-1, 6-2 and 6-3 respectively. Both SO (6DoF) and the GO (IK) algorithm were used to calculate the position of the segments of the subject while wearing a DAFO on both lower limbs. Only the results of the left side will be presented, as in this case, the right side was similar. To be noted that given the restrictions described above (6.2.6 Data Processing and Models) there are no values for the ankle joint in the frontal and transverse planes.

Regarding the sagittal plane (Figure 6-1), the ankle joint presented values with greater differences between algorithms. Flexion (dorsiflexion) and extension (plantarflexion) peak values were superior for the SO (13.9° and 17.2°, respectively). As for the GO, peak values were between 7.7° (dorsiflexion) and 11.5° (plantarflexion). At the knee joint, flexion peak value for the SO (68.6°) was similar to the GO (70°). The extension peak values were equal (1.4°), but in different time during the gait, given that in the GO this value occur at mid-stance and in the SO at the terminal swing. As for the hip joint angles, flexion was higher in the SO value (39.6°) and smaller in the GO (34,3°). Extension peak value were higher for the GO (14.2°) and smaller for the SO (9.5°).

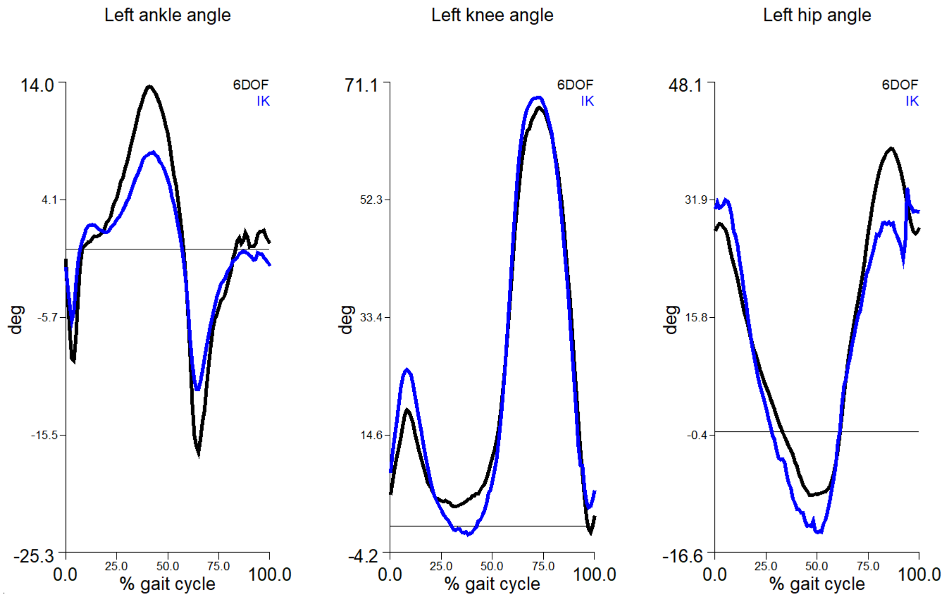

Figure 6-1: Ankle, knee and hip joint angles in the sagittal plane. Flexion (+) and Extension (–). Black line corresponds to SO and the blue line to the GO.

At frontal plane (Figure 6-2), the knee joint presented the most different angles, most notable the fact that the GO had no adduction, and the closest value was -1.4°. Still, the abduction values were similar between SO and GO (18.1° and 22°, respectively). Even so, SO presented an adduction of 3.7°.

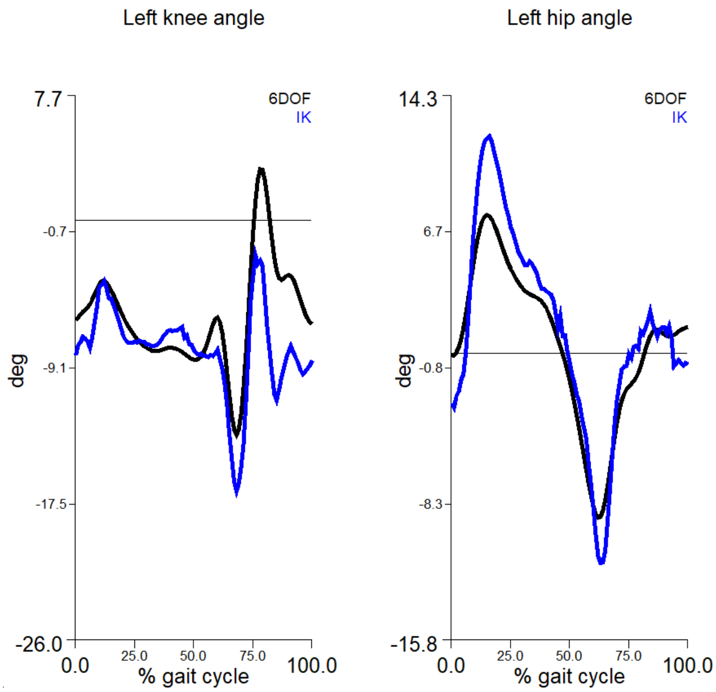

Figure 6-2: Knee and hip joint angles in the frontal plane. Adduction (+) and Abduction (–). Black line corresponds to SO and the blue line to the GO.

In the transverse plane (Figure 6-3) the values were more disperse throughout the gait at the knee and hip joints. Even so, the knee internal rotation peak values in SO (2.3°) and GO (3.7°) and external rotation peak values in SO (18.1°) and GO (-22°) were similar. At the hip joint, SO did not register any internal rotation and presented a peak external rotation (10.6°). as for the GO, it showed a small internal rotation (1.6°) and a wider external rotation (17.2°) when compared to SO.

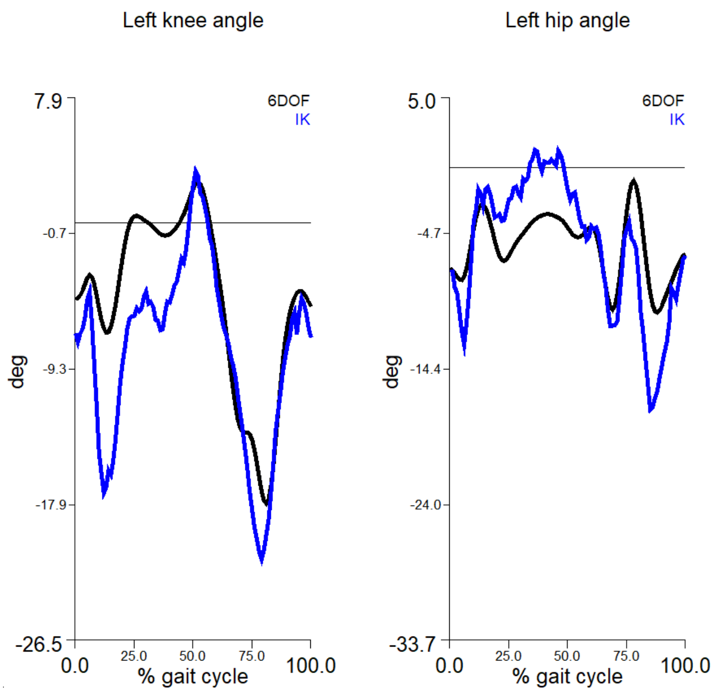

Figure 6-3: Knee and hip joint angles in the transverse plane. Internal rotation (+) and External rotation (–). Black line corresponds to SO and the blue line to the GO.

6.4 Discussion

The main differences between the two models were found in the ankle joint kinematics. The dorsiflexion peak in the stance phase and the plantarflexion peak in the push off period seems to be overestimated using the 6DOF model.

The proximal lower limb joints also presented differences between the two models, more pronounced in the stance phase for the knee joint and at the midstance and initial swing for the hip joint. Kainz et al. [10] suggested that the differences at knee joint can occur due to the different joint constrains.

Regarding the DAFO, both algorithms showed that the therapeutic objective was successful. Despite the subject presented a true equinus [17] gait with a dynamic ankle joint and predominance of the gastrocnemius muscle, the supramalleolar DAFO improved clearance in swing and first ankle rocker, which seems to be more in accordance when using a model with constraints that closely match that configuration. The results show a closer to normal RoM in the sagittal plane of the ankle.

As a result of co-spasticity of the muscles in children with CP, some high intensity movements across the joints can originate a wider variation in kinematic data [34], particularly in the SO algorithm. Even so, the presented behaviour of the kinematic parameters was similar to other studies [11], in which the variation of the transverse plane values, was wider and more prolonged during the two gait phases (stance and swing). This can be noted in Figure 6-3, where possibly there is some overestimation in the IK which is not related with the movement made by the participant.

In the near future, we intend to extend this research to the assessment of the effects of other AFO models, increased the number of participants for each group of AFO types and analyse the results using more robust methods (root-mean-square-differences between mean kinematic waveforms and absolute differences in discrete gait parameters).

6.5 Conclusions

The orthoses configuration to prevent plantarflexion thus improving clearance in swing and first ankle rocker seems to be more in accordance when using a model with constraints that closely match that configuration, yet a deeper treatment and analysis is needed to validate such assumptions.

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Chapter 7

7. General Discussion and Conclusion

7. General Discussion

This dissertation intended to investigate the changes in the gait of children with CP using an AFO and to provide a significant contribution to the optimization of the motion capture methodologies generally used in this population. The current chapter is divided in two sections. The first section provides an overall overview of the main findings of the four studies (from chapters 3 to 6) that were developed during the PhD process. The second section presents the main recommendations for future research.

7.1 Main findings

In **Chapter 3**, we intended to determine the effects of different types of ankle foot orthoses on the gait of children with spastic bilateral CP. Previous systematic reviews have not focused on specific CP subgroups or referred to gait pattern classifications, thereby including a wide range of gait abnormalities, or have included the information of lower quality studies [1–4]. The lack of gait pattern classification makes it more difficult to determine the mechanical and functional AFO characteristics needed to improve the different gait phases and overall performance. We found no more than two studies [5,6] that referred the sagittal gait patterns classification [7] to identify and categorize clinical subsets. Of these, only one [6] provided the participants with the type of AFO indicated in the classification. More worrisome is that the rationale behind the selection of each AFO and its prescription is missing in most studies, ranging from the use of a classification without evidence supporting of its use in AFO prescription [8], to suggestions based on empirical know-how [9], or not declaring any criteria at all [9–11]. Yet, some results suggest that the AFO use may produce a positive impact in spatial-temporal parameters [8–10,12], kinematic parameters [6,9,10,12,13] but on kinetic parameters there should be some caution when comparing and extrapolating the results to similar populations [6,10,12,13]. Even still, given the diversity of methodological options and the error associated with experimental protocols, some measures to assure that the outcome of the research singles out the AFO effect, should be considered. This led us to **Chapter 4**, where we performed the test-retest reliability of a 6DoF marker set in key points of gait biomechanical parameters in children with CP. It is known that most of the clinical variables can be influenced by intrinsic variations, namely in the intra-individual oscillations that occur in trial-to-trial sessions, or due to extrinsic variations, such as, marker placement [14]. There are significant differences between the existing biomechanical models used in different laboratories, for which is essential to understand the possible errors associated with the different techniques of marker sets and underlying anatomical models [15]. We addressed a lack of evidence regarding the reliability of 6DoF marker set in children with CP, to improve the reliability of a 3D gait analysis protocol and therefore improving the clinical analysis that supports decision-making in rehabilitation process. In this study, the CP children presented different gait patterns which contributed to some degree of variation of the data. The overall ICC results of kinematic and kinetic variables were lower on the right side, which can indicate that - to some degree – the instability of the affected lower limbs could influence the propagation of the STA. Given that in the 6DoF there is no linkage between segments, some non-physiological translations can occur [16] and result in an overestimation in kinematic peak values of the segments by as much as 100% [17]. Some specific gait patterns i.e., true equinus, can present a wide variation in its gait parameters [18], due to fluctuations in the gastrocnemius stiffness. Depending on which gait trials are selected, the data can be misrepresented of the true gait patterns and that may influence the type of intervention to be taken. These results represent an even bigger challenge in clinical gait analysis, where some pathologies can originate non-physiological movements that are not visible through visual inspection. The natural variability of gait in children with CP should require a repeated testing of a single subject to understand its own gait variation, because only in this way it is possible to identify when a change in gait is due to an intervention and not caused by the natural variability of the subject [19]. **Chapter 5** presents a study where a gait index commonly used in children with CP, is implemented to assess the clinical influence of the AFO-use. There is a general consensus that the increase in gait speed is considered to be an important factor in the children's motor development given its approximation to the data of typically developed children [12,13]. However, these results can be different according to the affected sides, where many studies reported no variations in the gait speed in bilateral children with CP [20]. Even though GPS measures the deviations from normal kinematics, an AFO prescription tends to accept some abnormal gait parameters with the expectation to improve other spatio-temporal and kinematic parameters [21]. The physiological behaviour of the ankle joint during gait, in which the AFO-use can impose mechanical constraints on the ankle joint, brings the sagittal gait parameters closer to normal gait. However, neither the foot accomplishes any of the three rockers during stance, nor substantial changes in kinematics or kinetics at the pelvis, hip, or knee have been identified [22]. A gait index from which validation and use was done in specific conditions, should be used in the same terms. Either barefoot, in shoe condition or with an AFO-footwear combination, these will significantly change the sagittal gait parameters [23]. The role of the footwear becomes at least equal to that of the orthosis because footwear design is so highly influential on the kinematics of the segments throughout the gait cycle. Consequently, the AFO-footwear combination is fundamental to the effectiveness of such intervention.

It is not clear what were the walking conditions (barefoot or with shoe) of the assessed subjects (typically developed children and children with gait deviations) in the GPS validations studies [24,25]. Such feature is fundamental to understand the scope of this tool in assessing the effects of AFO in children with CP. Consequently, the AFO-footwear combination is fundamental to the effectiveness of such intervention. Features, like "heel-sole differential" or "shank vertical angle" significantly influence the shank inclination at midstance, and if not properly intervened can forcibly impose a greater or a smaller amount of inclination [23]. Regarding this evidence, in **Chapter VI** we started to explore the effects of different pose estimation algorithms to reconstruct the pelvis and lower limb segments for gait analysis. As an example, it was modeled a supramaleolar AFO using two different pose algorithms. With the global optimization, we found that ankle sagittal plane movements can be overestimated. With segment optimization the results show that it tends to overestimate movements in the frontal and transverse plane. These preliminary results seem to point out that it is extremely important to acknowledge the type of optimization that is used when modeling the lower limb with an AFO, since the results may be misleading according to which plane of movement is being studied.

7.2 Future research

Scientifically, the contribution of this thesis is: 1) to increase the knowledge and insight about the existing methodologies in gait analysis in children with CP, particularly regarding the variability of measurements; 2) to investigate the quality of gait with AFO using a gait index and 3) to explore the pose estimation algorithms in 3D modelling of subjects walking with AFO. The next steps would certainly be to continue the work in the research and development of methods in gait assessments in cerebral palsy children to improve the experimental protocols used in the CP population. This would be of major importance, to be able to translate the real effects of the therapeutical proceedings applied or to be applied in such an impactful pathology in our society.

In addition, for my next goals, I would like to continue the work regarding the pose estimation algorithms, widening it to other types of AFO and other gait patterns. Exploring the better ways to manipulate these parameters can give us a clearer insight about which factors can affect the variability of outcomes with this population.

Moreover, I believe that subject-specific modelling is the best way to direct research on this topic, so I intend to study and explore those pathways in the near future.

7.3 Other publications

7.3.1 Poster presentations in international congress

Ricardo, D.; Teles, J.; Raposo, M.R.; Veloso, A.P.; João, F.

ISPO 18th World Congress. Poster “Reliability of lower extremity kinematics in three-dimensional gait measurements in children with Cerebral Palsy”.

7.3.2 Congress Proceedings

Ricardo, D.; Teles, J.; Raposo, M.R.; Veloso, A.P.; João, F. **(Appendix 3)**

ISPO 18th World Congress Abstract Book. *Prosthetics and Orthotics International*, 45 (2021) 1-293. https://doi.org/10.1097/01.PXR.0000799072.18452.79.

João, F.; **Ricardo, D.**; Raposo, M.R.; Veloso, A.P. **(Appendix 4)**

ESMAC Abstract Book. *Gait & Posture*, 90 Suppl. 1 (2021) 112-113. https://doi.org/10.1016/j.gaitpost.2021.09.057

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Chapter 8

8. General References

8. General References

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Chapter 9

9. Appendix

Appendix 1

Ricardo, D.; Raposo, M.R.; Cruz, E.B.; Oliveira, R.; Carnide, F.; Veloso, A.P.; João, F. Effects of Ankle Foot Orthoses on the Gait Patterns in Children with Spastic Bilateral Cerebral Palsy: A Scoping Review. Children 2021, 8, 903. doi: 10.3390/children8100903

children

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Review

Effects of Ankle Foot Orthoses on the Gait Patterns in Children with Spastic Bilateral Cerebral Palsy: A Scoping Review

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Abstract: Background: Cerebral palsy (CP) is the most common cause of motor disability in children and can cause severe gait deviations. The sagittal gait patterns classification for children with bilateral CP is an important guideline for the planning of the rehabilitation process. Ankle foot orthoses should improve the biomechanical parameters of pathological gait in the sagittal plane. Methods: A systematic search of the literature was conducted to identify randomized controlled trials (RCT) and controlled clinical trials (CCT) which measured the effect of ankle foot orthoses (AFO) on the gait of children with spastic bilateral CP, with kinetic, kinematic, and functional outcomes. Five databases (Pubmed, Scopus, ISI Web of SCIENCE, SciELO, and Cochrane Library) were searched before February 2020. The PEDro Score was used to assess the methodological quality of the selected studies and alignment with the Cochrane approach was also reviewed. Prospero registration number: CRD42018102670. Results: We included 10 studies considering a total of 285 children with spastic bilateral CP. None of the studies had a PEDro score below 4/10, including five RCTs. We identified five different types of AFO (solid; dynamic; hinged; ground reaction; posterior leaf spring) used across all studies. Only two studies referred to a classification for gait patterns. Across the different outcomes, significant differences were found in walking speed, stride length and cadence, range of motion, ground force reaction and joint moments, as well as functional scores, while wearing AFO. Conclusions: Overall, the use of AFO in children with spastic bilateral CP minimizes the impact of pathological gait, consistently improving some kinematic, kinetic, and spatial-temporal parameters, and making their gait closer to that of typically developing children. Creating a standardized protocol for future studies involving AFO would facilitate the reporting of new scientific data and help clinicians use their clinical reasoning skills to recommend the best AFO for their patients.

Keywords: child; cerebral palsy; gait analysis; orthotic devices; biomechanics

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1. Introduction

Cerebral palsy (CP) is the most common cause of motor disability in children [1–3]. Overall prevalence of CP is around 1 per 500 live births worldwide [2–5]. CP is a complex pathology that describes a group of impairments and motor disorders [5] with different presentations and functional levels [6].

The gait deviations that occur in children with CP are among other factors, due to inadequate muscle action [7]. Instrumented clinical gait analysis has been a great tool for planning intervention and assessing outcomes in the rehabilitation process of children with CP [2,8]. However, the use of all the outcomes within the three-dimensional kinematics or kinetics data to support classifying gait patterns in CP is still scarce [8], due to the almost exclusive use of the sagittal plane kinematic outcome in the majority of the gait classification systems [9,10].

Among several gait classification systems in children with CP, and particularly in bilateral spastic CP, Rodda et al. [11] identified several gait patterns and reported a high intra-rater reliability and moderate inter-rater reliability [9]. More recently Papageorgiou et al. [10] concluded that the characteristics presented by Rodda were considered as the most exhaustive ones, always including information about the co-occurring deviations across all lower limb joints [10].

This classification is based on clinical insight and biomechanical principles, and identifies five basic patterns of sagittal plane gait in spastic bilateral CP, namely true equinus, jump gait, apparent equinus, crouch gait, and asymmetric gait. These definitions are intended to be starting points for the guidelines for the planning of the rehabilitation process of children with CP. This allows not only the assessment of the most suitable orthosis for each case but also other surgical and non-surgical interventions, helping in the clinical decision-making process [11].

The use of ankle foot orthoses (AFO) is commonly prescribed to prevent the development or progression of deformity, and to control motion to improve dynamic efficiency of the child's gait [12,13]. There is a wide selection of AFO that can be used in the rehabilitation processes. However, their intended function depends mainly on their configurations, the material used, and its stiffness. Any alteration of these three components will alter the control that the AFO has on the patient's gait [14]. There are multiple designs, either fabricated as a one-piece of thicker thermoplastic AFO that restricts ankle and foot motion in all three planes (SAFO), or a flexible and dynamic AFO that allows some degree of sagittal plane motion (DAFO); a one piece design with a posterior malleolar trim line (posterior leaf spring-PLS), a two-piece design with a hinged joint that typically allows for dorsiflexion (HAFO), or a one piece anterior shelf design that promotes knee extension (GRAFO) [15–17].

Overall, studies involving gait and kinematic analysis have indicated that pathological gait in the sagittal plane can be improved using AFO [2,18,19], however it is not consensual about what factors are improved and how they have been improved. Thus, an assessment of the biomechanical characteristics and functional ability of the participants at baseline is crucial to track existing changes during the use of AFO [20]. Many studies involving orthotic use with CP patients present a wide variety of discrepancies in inclusion criteria or baseline assessments; missing information about orthosis design and construction, and how they are used; and different types of outcomes that can bias the indicated results. Previous systematic reviews have not focused on specific CP subgroups or referred to gait pattern classifications, thereby including a wide range of gait abnormalities, or have included the information of lower quality studies [21–24].

Due to the broad specter of physical presentations of children with CP, the aim of this review is to determine the effects of different types of ankle foot orthoses on the gait of children with spastic bilateral CP, presenting specific recommendations for this particular subset, and whenever possible refer to its effects on the five different sagittal gait patterns [11].

2. Materials and Methods

2.1. Search Strategy

A preliminary search was performed to select keywords related to the population, intervention, and outcomes using the PICO framework [25]. The keywords selected from the MeSH database in MEDLINE were: cerebral palsy, child, adolescent, orthotic devices, foot orthoses, splints, gait, kinematics, kinetics, walking, hip, hip joint, knee, knee joint, ankle, ankle joint, articular range of motion, walking speed, and International Classification of Functioning, Disability, and Health (ICF). Subsequent refinement searches were performed to obtain results. The selected keywords were joined by the words "AND" and "OR". The search equation was adapted according to the database where it was applied

(Table A1). The search was performed between January and July 2018, and included all records from the onset of each database. A secondary search was conducted in February 2020 with no other studies meeting the eligibility criteria. A keyword search was performed to match words in (all fields) the title, abstract, or keyword fields. The publication date was not restricted. Whenever possible filters on language were applied (Portuguese and English) (Appendix A).

The search to identify the relevant articles for this review was carried out in the following databases: Pubmed, Scopus, ISI Web of Science, Cochrane Library, and Scielo. To identify potentially relevant trials that were unpublished or ongoing, a search was also performed in the database of the World Health Organization International Clinical Trials Registry Platform (WHO ICTRP) and in the US National Institutes of Health (ClinicalTrials.gov).

2.2. Selection Criteria

2.2.1. Eligibility Criteria

The methodology used for this review followed the Cochrane guidelines [26]. The eligibility criteria for the selected articles were randomized clinical trials (RCT) and controlled clinical trials (CCT) (study design); written in English, Portuguese, or Spanish (language); with a focus on the pediatric population with bilateral CP (population) that used an AFO as a therapeutic intervention (intervention). The exclusion criteria were the use of functional electrical stimulation or robotic assisted therapy, and the existence of previous surgical or medical procedures (intervention). The outcome measures considered were the biomechanical gait parameters and/or functional abilities, including spatial-temporal, kinematic, kinetic, and gross motor function outcomes (outcomes).

2.2.2. Study Selection

The article selection was conducted by two independent reviewers (D.R. and M.R.R.), after duplicate removal and checking the articles' titles and abstracts against the eligibility criteria. The full text of the remaining articles was read. A bibliographic reference software manager (Mendeley V. 1.19.3) was used to assist the selection process. Whenever the two main investigators could not reach a consensus, a third external reviewer (E.B.C.) would intervene.

2.3. Methodological Quality (Risk of Bias)

The assessment of the quality of the included studies was the PEDro Risk of Bias Tool [27,28], for a minimum score of $\geq$5 points, which usually represents an adequate methodological quality study [29]. The rating of the studies and scoring of their methodological consistency were conducted by two reviewers (D.R. and M.R.R.), and, in case of disagreement or any discrepancies in scores, details were discussed with a third reviewer (E.B.C.). Furthermore, alignment between the PEDro scores and the Cochrane approach was verified for a broader assessment of the quality of the included studies [29].

2.4. Data Extraction

The characteristics of each selected study were extracted to compare the features across the studies. Author names, date of publication, study type and design, population characteristics and eligibility criteria, sample size, intervention type and duration, variables, measure instruments, and main findings were included.

3. Results

3.1. Article Selection

The initial search strategy identified 469 articles. After 78 duplicates were excluded, a further screening based on the title and abstract to assess the relevance of the articles excluded 352 articles. These articles did not meet the criteria of population (37), intervention (272), outcomes (4), and study design (39). A full text reading excluded 29 articles based on the criteria of population (3), intervention (2), outcomes (1), study design (21), and language (2). This resulted in a total of 10 articles that met our inclusion criteria and were included in our review flowchart (Figure 1).

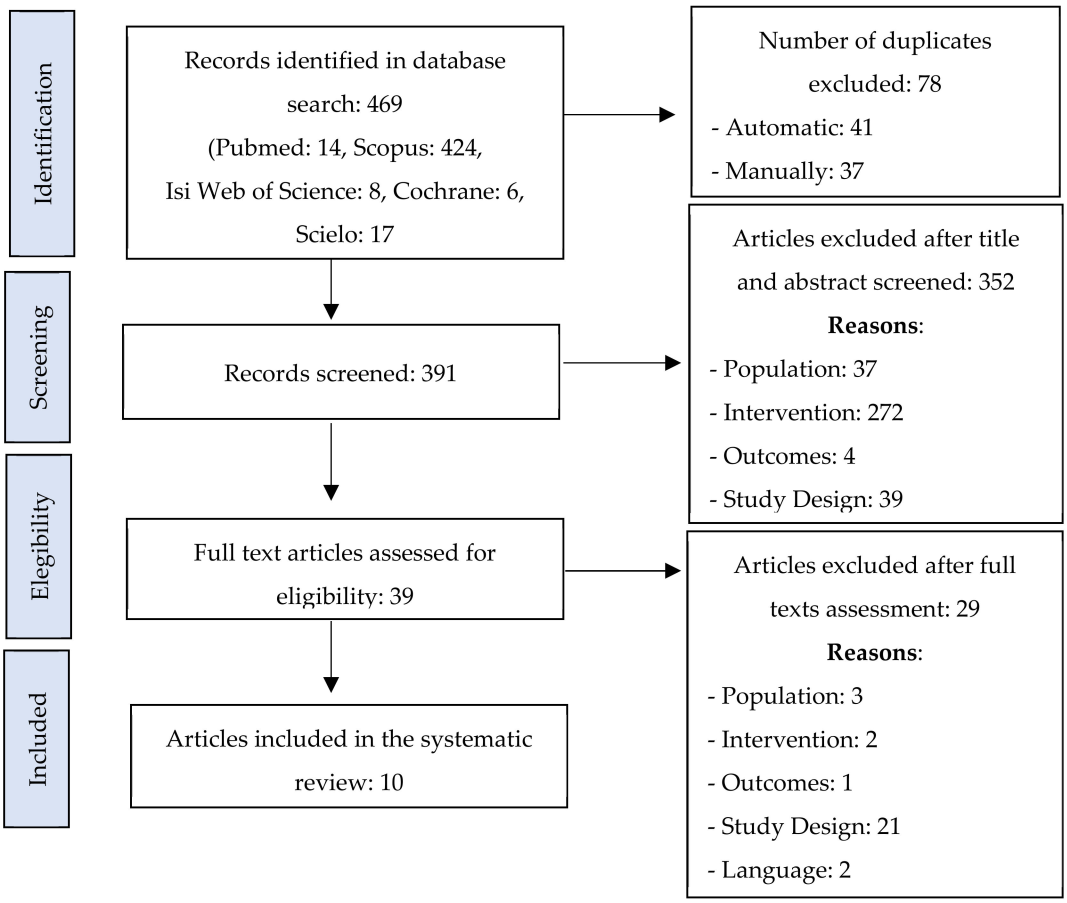

Figure 1. Flowchart of the article selection process.

3.2. *Article Characteristics*

The selected articles were published between 1997 and 2016. Of the 10 studies that were included, five were RCT [15,30–33] (three with a crossover design) and five were CCT [34–38]. The duration of the studies ranged from 1 day to 12 months in total. All studies compared at least one type of AFO intervention with barefoot, shoes, or other types of AFO interventions. The range of measurement instruments that were used included: optoeletronic systems, ankle accelerometer, force plates, and the Gross Motor Function Measure (GMFM) tool. The studies reported spatial-temporal parameters (walking speed, stride length, and cadence), kinematic outcomes (range of motion), kinetic outcomes (ground reaction force, joint moments, and joint power), and functional outcomes (GMFM). This enabled the compilation of data detailed in Table 1.

Table 1. Participants, sample details, methods, and main results.

Authors	Year	Study Design	Population Characteristics	Eligibility Criteria	N	Duration	Intervention/ Procedure	Variables	Measurement Instruments	Main Results and Author's Conclusions
Bjornson, 2006 [31]	2006	Randomized crossover controlled trial	23 children with spastic CP (age: 4.3 ± 1.5 years)	Children with spastic diplegia CP, 12 to 96 months, GMFCS I to III, bilateral use of AFO with free plantarflexion.	23	1 day	DAFO and shoes. GMFM used once with/without the orthoses during a same day evaluation.	Functional skills (GMFM scores).	GMFM.	The GMFM percentage scores for all dimensions were significantly higher with the patients wearing the DAFO ($p < 0.001$). There seems to be a non-significant negative correlation of age to standing skills change, suggesting that DAFO effect may decrease with age, up to the age of approximately 7 years ($p < 0.001$).
Bjornson, 2016 [32]	2016	Randomized crossover controlled trial	11 children with spastic CP (age: 4.3 ± 1.04 years)	Children with spastic diplegia CP; GMFCS I to III; bilateral use of AFO > 8 h/day, >1 month.	11	4 weeks (2 weeks without AFO and 2 weeks with AFO)	SAFO and shoes. Community based walking with/without AFO with a multiaxis accelerometer.	Functional skills (average total strides per day; % daytime hours walking; average number strides >30 strides/min; peak activity index).	StepWatch (Ankle accelerometer).	No significant difference was found in the primary outcome of average daily total step count between AFO-ON and AFO-OFF ($p = 0.48$). AFO did not improve walking activity levels.
Buckon, 2004 [33]	2004	Randomized crossover controlled trial	16 children with spastic CP (age: 8.3 ± 2.3 years)	Children with spastic diplegia CP; GMFCS I to II; bilateral use of AFO, 6 to 12 h daily >3 month.	16	1 year (a baseline assessment after three months of no AFO wear, and an assessment at the end of each AFO three-month wearing period	Barefoot or HAFO or PLS or SAFO.	Functional skills (GMFM scores); gait analysis data (kinematic variables at the pelvis, hip, knee, and ankle; Kinetic variables at the hip, knee, and ankle; Velocity, stride length, step length, and cadence).	Optoelectronic system; force plates; GMFM.	AFO use, regardless of configuration, did not significantly alter pelvic and hip kinematics and/or kinetics from the barefoot condition. At the knee there was no significant kinematic change. All AFO configurations significantly altered ankle kinematics during the stance and swing phases of gait: dorsiflexion at initial contact ($p = 0.0001$), peak dorsiflexion in stance ($p < 0.009$), timing of peak dorsiflexion in stance ($p < 0.003$), peak dorsiflexion in swing ($p < 0.0002$), and dynamic ankle range ($p < 0.0001$) compared with barefoot. Between the configurations, peak dorsiflexion in stance was significantly greater in the HAFO than the SAFO ($p = 0.01$), and the timing of peak dorsiflexion in stance was significantly later in the stance phase in the HAFO compared with the SAFO ($p = 0.005$). In conjunction with the changes in ankle kinematics, ankle kinetics (peak dorsiflexion moment in early stance [$p = 0.0001$], peak plantarflexion moment in early stance [$p = 0.0001$], peak power generation in stance [$p < 0.008$], and the timing of peak power generation [$p < 0.005$]) changed significantly in all the AFO configurations compared with barefoot. All of the AFO configurations significantly increased step ($p < 0.005$) and stride length ($p < 0.006$) compared with barefoot, while significantly decreasing cadence ($p < 0.0005$). Therefore, velocity did not increase significantly with AFO use compared with barefoot. Velocity was significantly slower in the HAFO compared with the PLS ($p = 0.009$), owing to a 17% decrease in cadence in the HAFO, an 11% decrease in the PLS, and a 13% decrease in the SAFO, compared with barefoot. AFO use did not significantly improve skills within the standing dimension of the GMFM. However, all AFO configurations significantly improved skills within the W/R/J dimension compared with the barefoot condition ($p < 0.002$).

Table 1. *Cont.*

Authors	Year	Study Design	Population Characteristics	Eligibility Criteria	N	Duration	Intervention/Procedure	Variables	Measurement Instruments	Main Results and Author's Conclusions
Degelean, 2012 [34]	2012	Non-randomized controlled clinical trial plus healthy controls (repeated measures design)	20 children with spastic diplegic CP (mean age: 7.6 ± 1.7 years) + 20 typically developing children (mean age: 7.8; ± 1.4 years)	Children with CP of the spastic diplegia type within the age of 4 and 12 years; no history of orthopedic surgery; no botulinum toxin injections within the last year; GMFCS level I or II; use of posterior leaf spring-type or solid AFO either in habitual walking or during physical therapy sessions.	20 + 20	1 day	Spring AFO or SAFO vs. barefoot. Participants walked at a comfortable speed on an 8 m walkway with AFO and barefoot. The task was recorded using an optoelectronic system detecting passive retro-reflective markers.	Gait analysis data (trunk movements; angular velocities; peak-to-peak excursions in trunk angular displacements; elevation angles of the thigh, shank, and foot).	Optoeletronic system.	Children with CP showed greater trunk sway excursion and angular velocity in both the sagittal and frontal directions compared to the control group ($p < 0.05$). Children with CP have greater sagittal and frontal trunk movements compared to typically developing children, but the difference in frontal motion was higher than in sagittal motion ($p < 0.05$). The use of any of AFO improved lower limb intersegmental coordination during gait in children with spastic diplegia by making it closer to a typical, mature gait pattern ($p < 0.05$). This was indicated by a significant greater ROM of the shank and a decreased ROM foot. However, wearing AFO results in increased trunk motion, which may be problematic in the context of difficult postural control.
El-Kafy, 2014 [15]	2014	Randomized parallel group controlled trial	57 children with spastic diplegic (mean age: 7.3 ± 1.3 years)	Children with CP of the spastic diplegia type within the age of 6–8 years old; under 40 kg; cognitively able to understand simple instructions; no recurrent medical issues; no allergic reactions to the adhesive tape or any other materials; no visual, auditory, or perceptual deficits or seizures; no previously use of TheraTogs orthotic undergarment, or strapping system and ground reaction ankle foot orthosis; no botulinum toxin in the lower extremity musculature during the past 6 months or other spasticity medication within 3 months of pre-treatment testing.	19 + 19 + 19	2 h/day, 5 days/week for a total of 12 weeks	Control group (A)—traditional neuro-developmental physical therapy. Study group (B)—A + TheraTogsTM orthotic undergarment and strapping system for both lower extremities. Study group (C)—B + received GRAFO in both lower limbs. Participants walked at a comfortable speed on an 8 m walkway with AFO and barefoot. The task was recorded using an optoelectronic system detecting passive retro-reflective markers.	Gait analysis data (gait speed; cadence; stride length; hip and knee flexion angles).	Optoeletronic system.	There were significant differences among the 3 groups pre-treatment in all measured variables (gait speed, cadence, stride length, and bilateral hip and knee flexion angles), and that they were present post-treatment ($p < 0.05$). This is due to the improvement of the plantar flexion, knee extension coupling, and knee and hip extension angle in mid stance provided by the GRAFO. The statistically significant differences post-treatment, in all parameters, were greater in group C than that in both groups A and B ($p < 0.05$). The results concerning the mean values of bilateral hip and knee rotational angles between both groups B and C revealed that there were no statistically significant differences in either pre- or post-treatment evaluation times ($p < 0.05$).

Table 1. *Cont.*

Authors	Year	Study Design	Population Characteristics	Eligibility Criteria	N	Duration	Intervention/Procedure	Variables	Measurement Instruments	Main Results and Author's Conclusions
Lam, 2005 [35]	2005	Non-randomized controlled clinical trial plus healthy controls (repeated measures design)	7 boys and 6 girls with spastic diplegic CP (mean age: 5.9 ± 1.81 years) + 18 typically developing children (age matched)	Spastic diplegia CP with mainly moderate dynamic equinus (modified Ashworth scale 1–3); no significant coronal or rotational deformities; no botulinum toxin injections within the preceding 5 months; good vision; the ability to comprehend instructions; be able to walk independently.	13 + 18	1 day	AFO and DAFO. Barefoot (healthy subjects control group).	Gait analysis data (stride length; stride time; speed; stance time; swing time; stance/swing ratio; cadence; range of motion parameters; moment parameters; power parameters).	Optoeletronic system; force platform.	CP patients had significantly shorter stride length than normal. Both AFO and DAFO conditions significantly increased stride length ($p < 0.05$). The mean stride length in CP patients walking barefoot (0.69 ± 0.14) was 65% of the healthy age matched children. The stride length was significantly increased when the subjects were wearing AFO (0.74 ± 0.15) or DAFO (0.81 ± 0.15). Concerning the total ROM, there was a reduction in range of motion at the ankle joint between the barefoot (22.39 ± 6.78), AFO (12.44 ± 5.55), and DAFO (19.72 ± 4.46). At initial contact children with DAFO presented a significantly increased knee and hip flexion by 4.8° ($p < 0.016$) and 5.3° ($p = 0.012$), respectably, when compared to barefoot walking. No significant difference was found at the ROM in the knee and hip between the AFO and DAFO. There was a significantly higher ground reaction force at the second peak wearing an AFO (0.97 ± 0.06) than when walking barefoot (0.89 ± 0.11). Both the AFO (0.96 ± 0.27) and the DAFO (1.11 ± 0.43) showed a significant improvement in the maximum plantar flexion moment compared to barefoot (0.69 ± 0.25). It was 0.28 Nm/kg higher in the AFO and 0.42 Nm/kg higher in the DAFO. There was no significant difference determined among barefoot, SAFO, and DAFO in all knee and hip power parameters.

Table 1. *Cont.*

Authors	Year	Study Design	Population Characteristics	Eligibility Criteria	N	Duration	Intervention/Procedure	Variables	Measurement Instruments	Main Results and Author's Conclusions
Radtka, 1997 [37]	1997	Non-randomized controlled clinical trial (repeated measures design)	10 children with spastic CP (6 diplegic; 4 hemiplegic) (mean age: 6.5 ± 1.86 years)	Spastic diplegia and unilateral CP; community ambulatory with plantigrade foot in standing, excessive plantar flexion during the stance, passive dorsiflexion of 5 degrees or more with knee extended, passive hip extension of 10 degrees or more, passive hamstring muscle length of 60 degrees or more in straight leg raise, mild to moderate spasticity in lower limb; no use of assistive device in ambulation; no orthopedic surgery in the previous year.	10	3 months (2 weeks barefoot + 1 month with AFO + 2 weeks barefoot + 1 month with DAFO)	AFO and DAFO.	Gait analysis data (walking speed; stride length; cadence; range of motion of the trunk, pelvis, hip, knee, and ankle at initial contact and mid-stance).	Contact closing foot switches; optoelectronic system.	There was an increased stride length wearing the AFO (0.97 ± 0.16) and DAFO (0.93 ± 0.13) compared with the barefoot condition (0.82 ± 0.13). The cadence was higher barefoot (148.33 ± 15.73) than with the AFO (140.10 ± 8.79) and DAFO (136.55 ± 10.96). The excessive ankle plantar flexion with no orthoses (8.54 ± 5.61) was over reduced with AFO (−2.62 ± 3.93) and DAFO (−1.66 ± 6.23). There were no differences ($p < 0.002$) at the level in joint motions of the knee, hip, and pelvis at initial contact and mid-stance with AFO or DAFO. The amount of ankle plantar flexion that occurred at initial contact and mid-stance in the interventions with no orthoses was reduced with both AFO and DAFO. No differences were found for the gait variables when comparing the two orthoses ($p < 0.02$).
Radtka, 2005 [36]	2005	Non-randomized controlled clinical trial (repeated measures design)	12 children with spastic diplegic CP (mean age: 7.5 ± 3.83 years)	Spastic diplegia CP; community ambulatory with ankle dorsiflexion to 0 degrees during static standing, excessive ankle plantar flexion of 5 degrees or more during stance in gait, passive ankle dorsiflexion to 5 degrees with knee extended passive hip extension to −10 degrees or less in the Thomas test, passive hamstring length of 50 degrees or more as measured by a straight leg raise; mild spasticity of the triceps surae, hamstrings and quadriceps; no surgical procedures in the past or any other orthopedic surgery during the year prior to the study.	12	3 months (2 weeks barefoot + 1 month with AFO + 2 weeks barefoot + 1 month with HAFO)	SAFO and HAFO.	Gait analysis data (range of motion of the knee and ankle during the stance phase; walking velocity; stride length; cadence; knee and ankle sagittal joint moments and powers during the stance phase).	Optoelectronic system; force plates.	The mean stride length was increased with both SAFO (0.87 ± 0.19) and HAFO (0.90 ± 0.19) when compared to no AFO (0.79 ± 0.19). No significant differences in walking velocity, cadence, and stride length when comparing no AFO, SAFO, and HAFO ($p < 0.05$). At the knee joint there were no findings of significant differences between barefoot, SAFO, or HAFO. When compared to the barefoot condition, at the ankle joint there were significant differences with the AFO and HAFO. The HAFO produced more normal dorsiflexion at the terminal stance phase than the SAFO, and more excessive dorsiflexion during loading phase than barefoot. There were significant differences when comparing no AFO (0.69 ± 0.14), SAFO (0.96 ± 0.22), and HAFO (0.94 ± 0.25) in the peak ankle moments. There was a significant difference in peak ankle moments during the terminal stance phase between barefoot (−1.30 ± 6.59) and SAFO (11.50 ± 4.28) and barefoot and HAFO (16.13 ± 6.17). The mean values were similar between both AFO.

Table 1. *Cont.*

Authors	Year	Study Design	Population Characteristics	Eligibility Criteria	N	Duration	Intervention/Procedure	Variables	Measurement Instruments	Main Results and Author's Conclusions
Smith, 2009 [38]	2009	Non-randomized controlled clinical trial plus healthy controls (repeated measures design)	15 children with spastic diplegic CP (mean age: 7.5 ± 2.9 years) + 20 typically developing children (mean age: 10.6 ± 2.8 years)	Spastic diplegia CP; able to walk independently without an assistive device; jump gait pattern; GMFCS level I; no orthopedic surgery in the past 12 months; no botulinum toxin injections in the past 6 months; range of ankle dorsiflexion to at least neutral on static physical examination with the knee extended.	15 + 20	2.5 months (barefoot baseline + 4 weeks with DAFO or HAFO + 2 weeks barefoot + 4 weeks with DAFO or HAFO)	DAFO and HAFO. Barefoot (healthy subjects control group).	Gait analysis data (walking speed; cadence; stride length; range of motion; joint moments; joints powers); functional skills (GMFM scores).	Optoelectronic system; force plates; GMFM.	Significant improvements in gait metrics were seen during brace wear ($p \leq 0.05$). When compared with barefoot condition, CP children wearing HAFO and DAFO showed a significant increase in stride length (0.98 ± 0.05) and (1.01 ± 0.05) and walking speed (1.09 ± 0.6) and (1.11 ± 0.6). When using HAFO or DAFO there was a significant decrease in normal cadence ($p \leq 0.006$) compared with the children with CP in barefoot condition. When comparing gait cycles of children with CP and healthy children there was no significant difference in terms of stride length, walking speed, or cadence. At the ankle significant differences between the HAFO or DAFO and the barefoot condition were found during the stance and swing phase ($p \leq 0.05$). The knee peak flexion during swing was significantly different between the DAFO and barefoot condition ($p \leq 0.05$). Children with CP using HAFO or DAFO had no significant effect on hip ROM. No significant differences were seen between the two different braces used ($p \leq 0.05$). The barefoot and braced conditions differed most significantly in terms of ankle kinematics and kinetics ($p \leq 0.05$). During the terminal stance of pre-swing, the ankle moment was significantly increased for both DAFO (0.98 ± 0.1) and HAFO (1.05 ± 0.1) when compared to the barefoot condition (0.80 ± 0.1). When compared to healthy children, in the barefoot and AFO condition, CP children presented a significant increase in plantar flexor moment during the initial contact ($p \leq 0.05$). No significant differences in ankle powers were found between DAFO and HAFO.

Table 1. *Cont.*

Authors	Year	Study Design	Population Characteristics	Eligibility Criteria	N	Duration	Intervention/Procedure	Variables	Measurement Instruments	Main Results and Author's Conclusions
Zhao, 2013 [30]	2013	Randomized parallel group controlled trial	70 boys and 42 girls with spastic diplegic CP (mean age: 2.69 ± 0.81 years)	Spastic diplegic CP; between 1 and 4 years of age; ability to walk independently, with or without an assistive device; GMFCS levels I-II; able to accept and follow AFO treatment strategy; no unstable seizures; no orthopedic surgery for spasticity within the preceding 6 months; no botulinum toxin injections within the preceding 3 months; without any other diseases that interfered with physical activity, and existence of serious cognitive disabilities.	56 + 56	5 to 8 weeks	Day AFO. Night and day AFO.	Gait analysis data (passive ankle dorsiflexion angle).	Sections D and E of the 66-item GMFM.	No evidence was found that the prolonged wearing time with AFOs leads to increased benefits ($p < 0.05$). The GMFM-66 improvement in the day-night AFO-wearing group was lower than in the day AFO-wearing group rather than higher. AFO day-night use was not more effective than daytime use alone in children with spastic diplegia at GMFCS levels I to II.

Abbreviations: AFO—ankle foot orthoses; CP—cerebral palsy; DAFO—dynamic ankle foot orthoses; GRAFO—ground reaction ankle foot orthoses; GMFCS—Gross Motor Function Classification System; GMFM—Gross Motor Function Measure; HAFO—hinged ankle foot orthoses; ROM—range of motion; SAFO—solid ankle foot orthoses.

The studies with fair to strong methodological quality were as follows: six studies with 4–5/10, one study with 6/10, and three studies with 8/10 on the PEDro scale (Table 2). All articles specified their "eligibility criteria", "follow-up", "intention to treat", and "statistical comparison". The "blind distribution", "blind subject", "blind therapist", and "blind assessor" were the items most often not verified. Three studies [15,30,31] managed to create blind assessment conditions, only two studies [15,30] had "blind distribution", and only one study [31] had unknowing therapist. No studies had "blind subjects", as it is not possible to use AFO without knowing it. Three studies [34,35,38] did not have equal circumstances at baseline ("similar prognosis") for their groups, as they used typically developed children for control group.

3.2.1. Characteristics of the Participants (Sagittal Gait Patterns)

Across all studies, there was a total of 347 participants (289 children with CP and 58 typically developing children [34,35,38]). Most studies included only children with spastic bilateral CP (285). Despite this, one study [37] presented a heterogeneous population, with four children with spastic unilateral CP. However, as the results were presented separately, we did not include them in this review.

Only a small percentage of the total participants had their gait patterns identified. Two studies referred to the sagittal gait patterns classification [32,38], identifying in total 18 participants with jump gait pattern, 5 true equinus, and 3 crouch gait pattern.

3.2.2. Types of AFO

The majority of interventions were centered in the comparison of gait when using ankle-foot orthosis and when walking barefoot [15,33–37], or using conventional shoes [31,32,38]. The type of AFO is central in most studies [15,30,33–38], but information about AFO construction, design and materials, as well as overall lower limb alignment and footwear are partially missing in every study.

We identified five different types of orthoses: 178 participants used solid ankle foot orthoses (SAFO) [30,32–37], 57 participants used dynamic ankle foot orthoses (DAFO) [31,35,37,38], 24 participants used posterior leaf spring (PLS) [33,34], 46 participants used hinged ankle foot orthoses (HAFO) [33,36,38], and 19 participants used ground reaction ankle foot orthoses (GRAFO) [15]. We found that overall, studies had no clear and consensual definition of the different types of AFO, and there was more than one description and configuration for the same terminology. In some of the studies, participants wore more than one type of orthoses [33,35–38], and in other studies some participants did not use any type of AFO [15].

3.2.3. Type of Outcomes

The main outcomes that were found were the following: spatial-temporal parameters [15,33,35–38], range of motion (RoM) [33,35–38], ground reaction forces [35], joint moments [33,35,36,38], and joint power [33,35,36,38]. Secondarily, some studies presented functional parameters, isolated or correlated with the biomechanical analysis [38]. The most frequently used tool was the Gross Motor Function Measure scale (GMFM) [30–33].

Most articles did not directly relate the reported outcomes with changes in the gait pattern in children with CP. Still, whenever possible, outcomes observed in the sagittal plane were associated with changes in the gait pattern.

Table 2. Methodological quality for studies in the review.

Article ID	PEDro Score										Total Score
	Eligibility Criteria *	Random Allocation	Blind Distribution	Similar Prognosis	Blind Subject	Blind Therapist	Blind Assessors	85% Follow-Up	Intention to Treat	Statistical Comparisons	Point of Measure/Measures of Variability	
Bjornson, 2006 [31]	Yes	Yes	No	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	8/10
Bjornson, 2016 [32]	Yes	Yes	No	Yes	No	No	No	Yes	Yes	Yes	No	5/10
Buckon, 2004 [33]	Yes	Yes	No	Yes	No	No	No	Yes	Yes	Yes	Yes	6/10
Degelean, 2012 [34]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
El-Kafy, 2014 [15]	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	Yes	8/10
Lam, 2005 [35]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
Radtka, 1997 [37]	Yes	No	No	Yes	No	No	No	Yes	Yes	Yes	Yes	5/10
Radtka, 2005 [36]	Yes	No	No	Yes	No	No	No	Yes	Yes	Yes	Yes	5/10
Smith, 2009 [38]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
Zhao, 2013 [30]	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	Yes	8/10

* This criterion is cited but not used to compute the total PEDro score.

Spatial-Temporal Parameters

One study compared gait in children with CP barefoot at baseline and after 4 weeks of DAFO or HAFO wear, and found significant differences ($p \leq 0.006$) across all measured spatial-temporal parameters (walking speed, stride length, and cadence) [38]. In studies that compared either children with CP wearing AFO with their typically developed peers or children with CP wearing AFO and barefoot, it was shown that use of AFO (regardless of the type) had a significant increase or an approximation to normal reference parameters in walking speed [15,38], step [33] and stride length [15,33,35–38], and a significant decrease towards normal cadence [15,33,37,38].

Nevertheless, there were studies that reported no significant differences for walking speed [33,35–37], nor significant differences for cadence [33,35,36] irrespective of AFO type or study design.

Kinematic Outcomes

The most often used kinematic parameter was RoM of the lower limb joints. For instance, significant improvement towards dorsiflexion of the ankle at the initial contact, and swing phase was observed [33,35–38], but, because the orthoses limit the plantar flexion, there was a significant decrease in RoM in the push-off stage of the pre-swing phase [35]. Maximal dorsiflexion in stance phase improved significantly with the use of SAFO [33,35,36]. It was also reported that the HAFO can produce excessive dorsiflexion during the stance phase [36].

While the most significant changes when wearing AFO are in the ankle RoM, in the knee RoM some differences were found, particularly in knee flexion on initial contact when compared to the barefoot condition [35,38]. Furthermore, children with CP wearing AFO showed a significantly greater range of motion of the shank [34]. No significant difference in knee RoM was found between the different types of AFO [33,35].

One study showed that children wearing DAFO were found to have a significantly greater hip flexion at initial contact [35], but overall, most studies found no significant changes at the hip joint, regardless the type of AFO [33,36–38].

Kinetic Outcomes

Only four studies reported kinetic parameters. One study reported that when using a SAFO or DAFO, there was a significant increase in the ground reaction force at the push-off when compared with the barefoot condition in children with CP [35]. An increase in the maximum plantar flexion moment in the terminal stance (push-off) was also reported, regardless of the type of AFO, with results similar to those of healthy children [33,35,36,38]. Peak knee extensor moment in early stance was significantly increased in the HAFO configuration compared with barefoot condition [33].

Regarding joint power, no significant difference was found in any of the analyzed joints between barefoot condition and AFO condition [33,35,38]. However, it was also reported that the peak of ankle power (that occurs at the push-off phase) when wearing a HAFO was similar to the barefoot condition [36], and between the configurations, the SAFO decreased peak power generation in stance significantly more than the PLS [33].

Functional Outcomes

To complement the biomechanical data, we were also interested in functional outcomes that the CP children may have reported with the use of AFO. The GMFM was the most often used tool, and studies showed it is responsive to change and can be used to evaluate the progress of a child while wearing AFO [39]. Although some of the included studies presented poor biomechanical data, they used this measure to evaluate the progress of AFO use in rehabilitation [30,31,33]. Most of the studies showed that the percentage scores for this scale were significantly higher when the patients wore the AFO [30–32], with the exception of one study where the AFO use did not significantly improve skills within the standing dimension of the GMFM [33]. The changes in some dimensions and total score of

GMFM were also significantly higher for independent walkers compared to children with CP using assistive devices while wearing DAFO [31].

4. Discussion

The main focus of this review was to assess the effects of AFO on gait in children with spastic bilateral CP, with particular attention to effects on different sagittal gait patterns. Identifying the gait type is useful in guiding orthotic options [40], and its use, coupled with the three-dimensional gait analysis, has been helpful in the clinical decision-making process. As a result, we have selected sagittal gait pattern classification [11] to help gather and systematize information. However, very few studies referred to such classification, making it difficult to summarize the data in the way planned in the protocol.

Fundamentally, clinical gait analysis for children with bilateral CP is very complex, since bilateral impairment of the lower limbs is often met with different sagittal gait patterns in each limb, sometimes even overlapping due to multiple gait abnormalities.

The lack of gait pattern classification makes it more difficult to determine the mechanical and functional AFO characteristics needed to improve the different gait phases and overall performance. Two studies [32,38] did use the sagittal gait patterns [11] to identify and categorize clinical subsets, although only one [38] provided the participants with the type of AFO indicated in the classification.

The appropriate AFO prescription is a practice that requires the clinician to perform a thorough physical examination and observational gait analysis, regardless of the age or Gross Motor Function Classification System (GMFCS) level of the child with CP [40]. Although consistent guidelines are lacking in this field [41], when applying an AFO, the aim is to correct and stabilize the biomechanical alignment of the foot and ankle, prevent the appearance or worsening of a musculoskeletal deformity, maintain the outcome of a surgical procedure, and ultimately improve gait [13].

The rationale behind the selection of each AFO and its prescription is missing in most studies. One study used the GMFCS to select the AFO to be used [34]; one study used the AFO already owned by the children with CP but without describing criteria [32]; two used the results of similar studies made previously [31,36]; one study made their own recommendations after a clinical and biomechanical assessment [37]; and three studies did not declare the criteria followed [30,35,37].

Nevertheless, results suggest that overall, AFO use may positively impact the gait of children with spastic bilateral CP. Spatial-temporal parameters, such as walking speed and stride length, reveal an approximation to normal reference [34–37], suggesting a better gait efficiency and probably less energy expenditure [33].

Overall, children with CP wearing any type of AFO presented significant differences in the range of motion of the ankle, when compared to the barefoot condition. Regardless of the AFO type, its use appears to reduce pathological plantarflexion, common in several of the bilateral CP gait patterns [35]. However, some types of orthoses (DAFO, SAFO, and GRAFO) are particularly more effective in controlling tibial progression and consequently promote knee extension during stance [32]. This can impact and modify the crouch gait pattern of CP children, approximating it to that of healthy subjects.

In children with spastic bilateral CP, there were significant increases in ground-reaction force and joint moments at push-off while wearing different AFO [35]. This demonstrates that up to 5 degrees of dorsiflexion of the ankle inside the AFO is more advantageous and induces an optimal muscle length in the calf muscles, approximating the plantar flexion moment to that of normal values [35,37].

Of the ten studies included in this review, only three focused on functional gains, and only one of the studies presented both biomechanical and functional data. Functional assessments are widely use in the rehabilitation of children with CP and should be more often correlated with biomechanical variables.

Methodological Considerations of This Review

We identified methodological limitations that are common in this type of study. Due to our eligibility criteria, the number of articles included was lower than other similar reviews. Of the 10 studies included, there was no common primary outcome between them. Although biomechanical and/or functional outcomes were found in all studies, the study designs are vastly heterogeneous (different samples sizes; wide range of age of participants; typically developed children control group versus children with CP barefoot control group; one day studies versus 12 months follow-up). This limits our ability to compare results due to the wider confidence intervals and a lower precision of the outcome measurements [42]. The point of statistical significance may be misleading, and this analysis may be leaving out some rehabilitation issues.

In CP research, CCT compares changes between groups to evaluate the efficacy of any treatment, but usually they lack reliable measures to detect changes that occur, which may be important from a clinical point of view [43]. In evidence-based medicine, the RCT is the highest level of evidence to be provided [44], and is the design of choice when comparing two or more healthcare interventions [29,44]. However, randomization may sometimes be affected by the number of participants, number of comparison groups, duration of the protocol, and the overall study design when studying AFO intervention. This may be a challenge because of differing clinical gait presentations and AFO requirements, thus we found that CCT are the more common for this population. The concealment of the allocation from parents and healthcare teams is a problem that practically limits this type of research [45,46].

Most studies included in this review were long-term follow-up studies [15,30,32,33,36–38] investigating the effects of the AFO for more than four weeks [47]. Studies with longer follow-up periods have also accounted for two weeks of rest between different orthosis [36,37]. This is relevant, as there were trials with a crossover design, where more than one type of orthosis was tested on the same day, raising concerns about the issue of carry-over effect between the different orthosis [31,32]. We suggest that future studies account for a proper wash-out period between trials [48].

Few authors advocate for an acclimatization period to ensure that the gait pattern is completely adapted to the altered ankle function as induced by the prescribed AFO, which may have impacted the results of their study [49]. Three studies allowed the children to wear the AFO one to three months prior to the first gait assessment so that the participants could gradually adapt to wearing them for the entire test day [33,36–38]. In two studies, children were already wearing their currently prescribed AFO [31,34]. Only one study reported the number of hours per/day/week that the subjects wore their AFO, but in all others that information was missing [15].

There are a wide variety of AFOs used in clinical practice, which are characterized by their design, the material used, and the stiffness of that material [14]. We have encountered at least five different types of AFO, but their definition was not always clear. The lack of nomenclature standardization also makes communication between researchers difficult [50].

Only one study used a prefabricated standard AFO [32], and in the remaining custom-made AFO were assigned for each participant [15,30,33,35–38]. Recent studies suggest that the initial outcomes are the immediate biomechanical response to the effect to the physical constraint imposed by the standard AFO, particularly the AFO stiffness [19,49]. On the other hand, custom-made AFO can be optimized with fine adjustments to its design and/or to the footwear prescription, in order to focus on optimal stiffness and increase its effects on gait pattern [14,51].

Even though an AFO is a frequently prescribed intervention for children with CP, rigorous evidence of their efficacy is limited [52], mainly because of the heterogeneity of outcome measures among researchers, which limits comparison between studies [53]. Although previous reviews have reported similar results and identified some of the limitations described above, still none have reported consistent guidelines for future studies [10,21–24].

Particularly, the absence of information about the clinical reasoning behind the AFO prescription, the selection of AFO design and construction, materials (including stiffness and thickness), AFO/footwear combinations, tuning, and acclimatization periods, makes it difficult to compare results within studies [50,54]. For instance, Kerkum et al. [47] reported that ankle ROM was significantly less reduced by both stiff and flexible spring-hinged AFO, and there was also a reduction in the ankle power when using a more rigid AFO. In this study, the authors used an instrument to measure the mechanical properties of the AFO and reported all the parametrization that was used for the AFO design. The differences found in gait kinematics and kinetics due to the stiffness of the AFO are only possible to compare with studies that also report the mechanical characteristics of the AFO, and that seems to be one of the greatest flaws in research regarding this topic [50].

Generically, the gait analysis protocols are not standard and have systematic errors related to extrinsic and intrinsic factors [55]. Regarding the use of 3D gait analysis in children with CP, several reliability studies identified that in the barefoot condition, kinematic and kinetic variables present with deviation between sessions, due to number of gait trials [56], biomechanical models and marker setup [57], or gait patterns and affected sides [58,59]. In turn, many studies report difficulties in 3D motion analyses when children with CP are wearing an AFO (especially when modeling ankle kinematics). When assessing the gait of children with CP wearing AFO, the marker setup usually sits on the surface of the AFO and shoe, making the assumption that they are the same rigid segment [60]. This may cause the interaction shank/ankle/AFO to present with some deviations. Ries et al. [16] attempted to minimize the influence of the AFO on shank and ankle kinematics by placing technical markers in a way that they were not to be covered or moved when the AFO was worn. By measuring the angle between the plantar surface of the shoe and the tibia, this study presented an alternative of measuring the true ankle position or the true neutral angle of the AFO.

Even though some methodological limitations are well reported, studies involving 3D gait analysis with the use of AFO should implement processes to minimize the error associated with their protocols, and state what measures they have included to assure that the outcomes of their research singles out the AFO effect.

It is also important to use tools such as the International Classification of Functioning, Disability, and Health (ICF) to standardize the report of results within the health-related domains [61]. Currently, there are specific ICF core sets for CP patients, therefore future studies should summarize the outcomes in this framework and create a common language across healthcare professionals [62].

Overall, we considered that there is need to standardize the AFO research, which can optimize the biomechanical properties and simplify future studies, making it possible to replicate results and provide better options for children with CP and their families [50].

5. Conclusions

In this review, we found that AFO use seems to have an immediate and a long-term effect in improving the sagittal gait patterns in children with spastic bilateral CP. However, most studies included heterogeneous groups with different gait patterns, and there were different approaches to the use of AFO. There is a need for future studies to invest in higher methodological quality protocols.

We propose the creation of a standardized protocol for future studies involving AFO and children with CP. There is a need to develop consistent AFO prescription algorithms that are designed specifically for each gait pattern. It should also include information about periods for AFO acclimatization and the need for fine tuning, appropriate follow-up periods to ensure full effect of AFO, appropriate wash-out periods, reports on hours per day of AFO usage, and AFO design, materials, and construction. This would facilitate the report and replication of new scientific data and help clinicians use their clinical reasoning skills to recommend the best AFO for their patients.

The rationale for these options needs to be more objective and evidence-based, which in the future may represent both improved assessment tools as well as a more effective therapeutic intervention.

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Appendix A

PICO Question Key Words

Population: cerebral palsy; cp; children; children with cerebral palsy; adolescent; diplegia; spastic diplegia.

Intervention: ankle foot orthoses; AFO; orthoses; orthotics; orthosis; ground force reaction orthoses; GRAFO; hinged ankle foot orthoses; HAFO; dynamic ankle foot orthoses; DAFO; solid ankle foot orthoses; SAFO.

Comparison: (none).

Outcome: gait; kinematics; kinetics; walking; functionality; functional activities; gait pattern; gait velocity; trunk sway; maximum knee extension; maximum hip extension; ankle; knee; hip; range of motion; ROM; gross motor function; GMFM; walking speed; stride length; energy expenditure.

Search Strategies (MeSh terms; word truncation; relevance of key words).

1. "cerebral palsy" [mh]
2. child *[mh]
3. adolescent
4. #1–#3
5. "sagittal gait patterns"
6. "spastic diplegia"
7. "true equinus"
8. "jump gait"
9. "apparent equinus"
10. "crouch gait"
11. "asymmetric gait"
12. #5–#11
13. "ankle foot orthoses"
14. AFO
15. "orthotic devices" [mh]
16. "foot orthoses" [mh]
17. splints [mh]
18. #12–#17
19. gait [mh]
20. walking [mh]

21. kinematics [mh]
22. kinetics [mh]
23. "spatiotemporal analysis"
24. functionality
25. "functional activities"
26. ICF
27. "gross motor function measure"
28. #19–#27
29. "randomised controlled trial" [pt]
30. "controlled clinical trial" [pt]
31. "clinical trial" [pt]
32. "comparative study" [pt]
33. #29–#32
34. #1–#3 AND #5–#11 AND #12–#17 AND #19–#27 AND #29–#32

Search Questions used in different data sources

#1:

(("cerebral palsy" [mesh] OR child* [mesh] OR adolescent [mesh]) AND ("sagittal gait patterns" OR "spastic diplegia" OR "true equinus" OR "jump gait" OR "apparent equinus" OR "crouch gait" OR "asymmetric gait") AND ("ankle foot orthoses" OR AFO OR "orthotic devices" [mesh] OR "foot orthoses" [mesh] OR splints [mesh]) AND (gait [mesh] OR walking [mesh] OR kinematics [mesh] OR kinetics [mesh] OR "spatiotemporal analysis" OR functionality OR "functional activities" OR ICF OR "gross motor function measure") AND ("randomized controlled trial" [pt] OR "controlled clinical trial" [pt] OR "clinical trial" [pt] OR "comparative study" [pt]))

#2

("cerebral palsy" OR child* OR adolescent OR youth) AND ("sagittal gait patterns" OR "spastic diplegia" OR "true equinus" OR "jump gait" OR "apparent equinus" OR "crouch gait" OR "asymmetric gait") AND ("ankle foot orthoses" OR AFO OR "orthotic device*" OR orthos* OR "foot orthos*" OR splint*) AND (gait OR "walking speed" OR walking OR ambulation OR kinematics OR kinetics OR biomechanical OR "spatiotemporal analysis" OR functionality OR "functional activities" OR ICF OR "gross motor function measure") AND ("randomized controlled trial" OR "controlled clinical trial" OR "clinical trial" OR "comparative study")

#3:

("cerebral palsy") AND ("sagittal gait patterns") OR ("spastic diplegia") AND ("ankle foot orthoses") OR (gait) OR (kinematics) OR (kinetics)

Table A1. Search Results.

Date	Source	Search Question	N° of Results	Notes
13 January 2020	Pubmed	#1	14	
27 January 2020	Scopus	#2	363	
27 January 2020	Isi Web of Science	#1	8	No filter
27 January 2020	Scielo	#3	17	No filter
27 January 2020	Cochrane	#1	6	

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Appendix 2

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Test-Retest Reliability of a 6DoF Marker Set for Gait Analysis in Cerebral Palsy Children

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Abstract: Background: Cerebral palsy (CP) is a complex pathology that describes a group of motor disorders with different presentations and functional levels. Three-dimensional gait analysis is widely used in the assessment of CP children to assist in clinical decision making. Thus, it is crucial to assess the repeatability of gait measurements to evaluate the progress of the rehabilitation process. The purpose of the study is to evaluate test-retest reliability of a six-degree-of-freedom (6DoF) marker set in key points of gait kinematics, kinetics, and time-distance parameters in children with CP. Methods: trials were performed on two different days within a period of 7.5 ± 1.4 day. Motion capture data was collected with 14 infrared, high-speed cameras at a frequency rate of 100 Hz, synchronized in time and space with two force plates. Intraclass correlation coefficients considering the two-way mixed model, and absolute agreement (ICC[A,k]) were calculated for anthropometric, time–distance, kinematic and kinetic parameters of both lower limbs. Results: the majority of gait parameters demonstrated a good ICC, and the lowest values were in the kinematic variables. Conclusions: this study indicates wide-ranging reliability values for lower limb joint angles and joint moments of force during gait, especially for frontal and transverse planes. Although the use of a 6DoF-CAST in CP children was shown to be a feasible method, the gait variation that can be observed between sessions in CP children seems to be related not only to the extrinsic factors but also to their different gait patterns and affected sides.

Keywords: cerebral palsy; gait; reliability; kinematic model; biomechanics; kinematics; kinetics

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1. Introduction

Cerebral palsy (CP) is the most common cause of motor disability in children [1–3]. The average incidence of cerebral palsy is estimated to range between 1.5 to 3.3 per 1000 live births in European countries [4], whereas this number is around 1 per 500 live births worldwide [2,3,5]. CP is a complex pathology that describes a group of impairments and motor disorders [6] with different presentations and functional levels [7]. The gait deviations that occur in CP children are mainly originated by an inadequate muscle action [8]. Three-dimensional gait analysis is the widely accepted technique used in the assessment of ambulant patients with CP to assist in clinical decision making and assessing outcomes in the rehabilitation process [9], supporting a complete biomechanical analysis of the time-distance, kinematic and kinetic parameters of gait [10].

The purpose of each clinical gait measurement technology is to provide data free from measurement errors that may create uncertainty about the possible clinical interpretations. Thus, reliability addresses to which extent gait measurements are consistent or free from variation across time [11]. However, most of these clinical variables are not reliable [12], either due to their own intrinsic variations, namely in the intra-individual oscillations that occur in trial-to-trial sessions, or due to extrinsic variations, such as, marker placement [13].

CP children are intensively studied in gait analysis, but unlike other populations with gait abnormalities [14] there are no specific biomechanical models to their gait characteristics. It is known that there are significant differences among the techniques, but the gait laboratories still opt to use their typical protocols, regardless of the population.

It is essential to understand the possible errors associated with the different techniques of marker sets and underlying anatomical models [15] to reproduce the clinical gait measurements with confidence [16]. Significant differences exist in biomechanical models used in different laboratories. These include measured variables, degrees of freedom assigned to the joints, anatomical reference frames, and joint rotation conventions [17]. The conventional gait model (CGM) is a very widely used biomechanical model to calculate kinematic and kinetic variables in gait analysis [16]. It has been extensively validated but there are still some issues regarding its reliability, mainly due to its unconstrained segment dimensions which makes it more exposed to sources of errors [18]. The six-degree-of-freedom (6DoF) models are the most common alternative to the CGM that, despite needing more extensive validation [18], assumes that the segments are rigid and do not constrain the joints motions [19]. Several 6DoF modeling techniques were used in the assessment of repeatability in participants with motor and physical characteristics limiting the normal gait [14,20,21].

These 6DoF models address the known limitations of the CGM, but unlike the latter it still needs to be better researched. However, some results have indicated some of those claims (e.g., the segments have a fixed length and soft tissue artifact is reduced). Soft tissue artifact between markers is certainly eliminated by using rigid clusters, but a different form of soft tissue artifact will affect the orientation and position of the whole cluster in relation to the bones [22]. In children in particular, the amount of soft tissue surrounding the limb segments is not the major reason for some oscillations, but the smaller distance between clusters and anatomical markers which do not minimize the magnitude of this type of error. According to a systematic review of McGinley et al. [11] about the repeatability studies of kinematic models, the majority of the included studies used the CGM or some variant of it. In previous test-retest reliability studies performed in CP children, the biomechanical models were based in CGM [23] and similar models such as the Helen-Hayes [24] and the Vicon Clinical Manager [25]. One study that used a 6DoF variant (the Cleveland clinic marker set) [26] did not compare kinetic data and the authors assessed repeatability using a coefficient of multiple correlation (CMC) which has recently been determined not to be suitable as a tool for assessing reliability in gait measurements [27].

The lack of evidence regarding the reliability of 6DoF models in subjects with abnormal gait patterns, particularly in kinetic variables, was the motivation to develop this research. Moreover, knowing that errors associated with kinematic variables have tremendous consequences in the estimation of the kinetic parameters, it is essential to assess the magnitude of these errors. Considering these issues, the aim of this study is to evaluate the test-retest reliability of a 6DoF model in key kinematic and kinetic gait cycle parameters in CP children.

2. Materials and Methods

2.1. Design

Prospective controlled study.

2.2. Participants Selection

A convenience sampling of eight children (two females and six males) with cerebral palsy was recruited from two Portuguese cerebral palsy centres to participate in the study. Firstly, the participants' clinical history was reviewed, and a clinical exam was performed with the subject laid on the table, seated on a chair, or standing. The eligibility criteria were as follows: male and female children, between 4 and 16 years of age; with a clinical diagnosis of Unilateral Spastic Cerebral Palsy or Bilateral Spastic Cerebral Palsy of crural predominance, grades I and II in the Gross Motor Function Classification System

(GMFCS) [28]; able to walk independently with or without walking aids; cooperative and able to comply with simple orders; feet size between 20 and 33; who had a clinical recommendation to use an ankle foot orthosis, but have never used it before, or during the trials; who have not undergone orthopaedic surgery of the lower limb in the last 12 months, and who are not expecting to have a surgical intervention in the next 6 months; and who were not given botulinum toxin in the last 6 months [29]. The protocol was approved by and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019). An informed consent was previously signed by the parent or the legal guardian of the participant.

2.3. Gait Protocol

The trials were performed on two different days within a period of 7.5 ± 1.4 days to minimize the assessor memory bias and short enough to prevent a change in the children's gait pattern or clinical condition [21]. Upon the participants' arrival, instruction was given about the protocol, the risks and benefits, as well as the informed consent.

The initial clinical exam consisted of a sequence of measures to assess bone and joint deformities, muscle length, muscle force, selective motor control and spasticity [2]. Two experienced researchers performed the clinical assessment while the same assessor was responsible for the placement of the markers in all the sessions. Palpation was used to locate the subcutaneous anatomical landmarks on the participants [30] and subsequently to place the marker set. These were 1.25 cm spherical reflective markers with a 1.8 cm semi-flexible width base. Four marker clusters were attached to the lateral part of the thigh and shank to independently track anatomical landmarks of each segment allowing rotational and translational motion at the joints [19]. These types of markers were adequate for the general height of these children given the smaller body parts. Motion capture data were collected with 14 infrared, high-speed cameras (Qualisys Oqus 300, Qualisys AB, Gothenburg, Sweden) at a frequency rate of 100 Hz. This system was synchronized in time and space with two force plates (FP4060-07, FP4060-10, Bertec, Columbus, OH, USA) embedded into the laboratory walkway [31]. Before each dynamic trial, a barefoot static trial in the standing position was recorded in order to determine the participant's joint centres and segmental reference systems, as well as segments' length [19]. Afterwards, the participant was instructed to walk along a 10 m corridor, unassisted at a self-selected pace. The dynamic trials ended when the child successfully achieved a minimum of five complete kinematic and kinetic walking cycles for each side [14,32,33], considering the natural variation in kinematic and kinetic gait parameters [34].

2.4. Data Processing

Gait cycles were extracted using Qualysis Track Manager (QTM) (v2020.3 build 6020, Qualisys AB, Gothenburg, Sweden). The subsequent analysis and processing were done using Visual 3D software (Professional Version v4.80.00, C-Motion, Inc., Rockville, MD, USA). The marker set (Figure 1) that was used followed the calibrated anatomical system protocol (CAST) [30,35] and CODA pelvis [36]. It was used to reconstruct the pelvis and both lower limbs [34]. The 22 individual markers and four marker clusters of four embedded markers each, allowed the reconstruction of seven body segments: feet, shanks, thighs, and pelvis. Each segment is considered to be independent and to have six degrees of freedom [37]. Lower limb segment masses were determined according to Dempster [38] while the remaining inertial parameters were computed based on Hanavan [39].

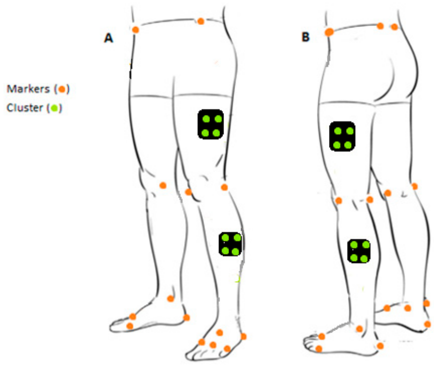

Figure 1. Positioning of the retroreflective markers attached to the subjects. Adapted from [40]: (**A**) anterior view; (**B**) posterior view.

The pelvic anatomical coordinate system was defined by surface markers placed on the right and left anterior superior iliac spines (ASIS) and on the right and left posterior superior iliac spines (PSIS) and can be described as the origin at the midpoint between the right ASIS and the left ASIS; the Z-axis points from the origin to the right ASIS; the X-axis lies in the plane defined by the right ASIS, left ASIS, and the midpoint of the right PSIS and left PSIS markers and points ventrally orthogonal to the Z-axis; and the Y-axis is orthogonal to the previous two [41]. The hip joint centers were computed using the pelvis markers, according to Bell's regression equations [36]. Anatomical reference frames of the lower limb segments were defined in accordance with the International Society of Biomechanics (ISB) recommendations to the standard description of joint kinematics [41].

The thigh anatomical coordinate system was defined by the hip joint centers previously computed using the pelvis markers and the lateral and medial femur condyles; the origin was the hip joint center; the Z-axis points from the midpoint between the lateral and medial femur condyles and the origin; the Y-axis is perpendicular to the Z-axis and the frontal plane of the thigh (defined by an axis between the lateral and medial femur condyles and the hip joint center); the X-axis is orthogonal to the previous two.

The shank anatomical coordinate system was defined by the femur condyles and malleolli markers; the origin was the knee joint center defined as the midpoint of the medial and lateral femur condyles; the Z-axis points from the midpoint between the lateral and medial malleoli and the origin; the Y-axis is perpendicular to the frontal plane of the shank and Z-axis; X-axis is orthogonal to the previous two.

The foot anatomical coordinate system was defined by the malleolli markers and the metatarsal markers; the origin was the ankle joint center defined by the midpoint between the lateral and medial malleoli markers; the Z-axis points from the midpoint between the 1st and 5th metatarsal heads and the origin; the Y-axis is perpendicular to the frontal plane of the foot and the Z-axis; X-axis is orthogonal to the previous two [42].

Lower limb and pelvis joint angles (calculated using a XYZ Cardan sequence) and moments (determined through inverse dynamics and normalized to subjects' body mass) were computed and expressed relative to the proximal segment. The XYZ Cardan sequence was used due to the ISB recommendations regarding its clinical and anatomical meaning [43], since the description of X, Y and Z are equal to flexion-extension, abduction-adduction and longitudinal internal-external rotation, respectively.

A cubic spline smoothing routine was used to filter both kinematic and kinetic data. The segment length was defined as the distance between the proximal and distal ends of the segment. Kinematic and kinetic data were normalized to 100% of the gait cycle. Peak values for lower limb angles and moments, as well as time–distance parameters, were computed for each cycle and averaged for each subject [21]. All data were considered assuming the lower limbs as independent to evaluate the variation of each one, even if they participated jointly during the gait cycle.

2.5. Statistical Methods

Statistical analysis to assess test-retest reliability of the gait kinematic and kinetic data was carried out using the method described by Quigley et al. [44] and Fernandes et al. [21]. Intraclass correlation coefficients considering the two-way mixed model, and absolute agreement (ICC[A,k]) [45,46] were calculated for anthropometric, time-distance, kinematic and kinetic parameters of both lower limbs. The level of agreement was considered poor, fair, good, and excellent when ICC < 0.40, 0.40 ≤ ICC < 0.60, 0.60 ≤ ICC < 0.75, 0.75 ≤ ICC ≤ 1.00, respectively [47]. The absolute measure of reliability standard error of measurement (SEM) was calculated using the following equation: SEM = $\boldsymbol{SD_{diff}}/\sqrt{2}$. The indicated levels of error for kinematic data were considered acceptable if SEM ≤ 2°, reasonable between 2° and 5°, and concerning if SEM ≥ 5° [20]. From each trial, 97 individual values of clinical interest were extracted. The calculated key points included the mean difference between measurements and the 95% confidence interval (CI) for mean difference, the standard deviation of the differences ($\boldsymbol{SD_{diff}}$) and the 95% Bland and Altman limits of agreement (95% LOA). All the statistical tests were conducted using SPSS (version 26.0; IBM, Chicago, IL, USA) and $p < 0.05$ was considered statistically significant.

3. Results

The participants of the study were a convenience sampling of eight CP children (Table 1) able to walk independently (three hemiplegic, five diplegic; two females, six males; age 87.88 ± 25.56 months; height 1.17 ± 0.14 m; mass 24.25 ± 8.26 kg). Two trials were performed on two different days within period of 7.5 ± 1.4 days.

3.1. Reliability of Anthropometric Parameters

The ICCs were ≥0.96 for anthropometric measurements (Table 2). The lowest were the right (0.97, 95% CI 0.86 to 0.99) and left foot segment length (0.96, 95% CI 0.83 to 0.99) and SEM values were ≤0.64 cm.

3.2. Reliability of Time-Distance Parameters

For time-distance parameters, ICCs were ≥0.75 (Table 3) except for right step length (0.64, 95% CI 0.00 to 0.92) and right stride length (0.64, 95% CI 0.00 to 0.92). The SEM values were 0.06 m and 0.11 m, respectively.

3.3. Reliability of Kinematic Parameters

Most joint angle peaks demonstrated excellent ICCs ≥0.75 (Table 4). Good ICCs were also shown in both sides of the lower limbs. On the right lower limb, the pelvic obliquity up was (0.67, 95% CI 0.00 to 0.94) and the hip internal and external rotation (0.73, 95% CI 0.00 to 0.95) and (0.67, 95% CI 0.00 to 0.93), respectively. Similarly on the left side, hip abduction was (0.60, 95% CI 0.00 to 0.92) and internal rotation (0.67, 95% CI 0.00 to 0.93). At the knee joint, its internal rotation was (0.64, 95% CI 0.00 to 0.92) and ankle eversion (0.60, 95% CI 0.00 to 0.91). However, a few of the ICCs variables were poor, the majority on the right side, with hip flexion (0.14, 95% CI 0.00 to 0.84), knee abduction (0.37, 95% CI 0.00 to 0.88), adduction (0.33, 95% CI 0.00 to 0.87), internal rotation (0.00, 95% CI 0.00 to 0.69) and ankle plantar flexion (0.00, 95% CI 0.00 to 0.81) and inversion (0.00, 95% CI 0.00 to 0.80). In the left side, only the ankle plantar flexion (0.27, 95% CI 0.00 to 0.92) presented similar values in this range. The SEM values ranged between 1.8° to 14.7° and average between 3.2° e 7.9°.

Table 1. Subject characteristics.

				Left Lower Limb			Right Lower Limb		
Subject	**Affected Side**	**Height (m)**	**Mass (Kg)**	**True Leg Length (cm)**	**Sagittal Gait Pattern**	**Gastrocnemius Spasticity (Modified Ashworth Scale)**	**True Leg Length (cm)**	**Sagittal Gait Pattern**	**Gastrocnemius Spasticity (Modified Ashworth Scale)**
001	Bilateral	1.09	19.5	52.5	True equinus [48]	1+	54.5	True equinus [48]	2
002	Unilateral	1.14	26	54.6	Normal	0	54.3	True equinus [49]	2
003	Bilateral	1.32	26	66	Apparent equinus [48]	1+	66	Apparent equinus [48]	1+
004	Unilateral	0.98	13.5	46	True equinus [48]	1+	45	Normal	0
005	Bilateral	1.37	34	71	Apparent equinus [48]	2	70.5	Apparent equinus [48]	2
006	Unilateral	1.32	37	70.2	Normal	0	70.1	True equinus with recurvatum knee [49]	1+
007	Bilateral	1.06	15.5	52	True equinus [48]	3	52.7	True equinus [48]	3
008	Bilateral	1.10	18	54	Jump gait [48]	2	54.5	Jump gait [48]	2

Table 2. Reliability values for anthropometric measurements.

Anthropometric Parameters	ICC	ICC 95% CI	Mean	Mean Diff	Mean Diff 95% CI	SD_{diff}	95% LOA	SEM
Pelvis Segment Depth (cm)	0.98	(0.93, 0.99)	13.2	0.2	(−0.2, 0.7)	0.6	(−0.97, 1.40)	0.4
Inter ASIS Distance (cm)	0.98	(0.94, 0.99)	17.3	−0.1	(−0.7, 0.3)	0.6	(−1.50, 1.13)	0.4
Right Tight Segment Length (cm)	0.99	(0.97, 0.99)	26.6	−0.1	(−0.7, 0.4)	0.7	(−1.55, 1.20)	0.5
Left Tight Segment Length (cm)	0.99	(0.89, 0.99)	26.7	−0.5	(−0.9, 0.1)	0.4	(−1.50, 0.42)	0.3
Right Leg Segment Length (cm)	0.99	(0.95, 0.99)	25.8	0.1	(−0.7, 0.8)	0.9	(−1.68, 1.85)	0.6
Left Leg Segment Length (cm)	0.99	(0.97, 0.99)	25.9	0.3	(−0.0, 0.7)	0.4	(−0.53, 1.23)	0.3
Right Foot Segment Length (cm)	0.97	(0.86, 0.99)	8.8	0.1	(−0.2, 0.4)	0.4	(−0.76, 0.96)	0.3
Left Foot Segment Length (cm)	0.96	(0.83, 0.99)	9.0	0.1	(−0.3, 0.5)	0.5	(−1.01, 1.21)	0.4
Average	0.98							0.4

Intraclass correlation coefficient, ICC; 95% CI, 95% confidence interval for the ICC; mean, mean of measurements at baseline trial and retest trial; mean diff, mean of the differences between measurements at times 1 and 2 and the 95%. CI for mean diff, the standard deviation of the differences (SD_{diff}); 95% LOA, Bland and Altman 95% limits of agreement; SEM, standard error of measurement.

Table 3. Reliability values for time-distance parameters.

Time-Distance Parameters	ICC	ICC 95% CI	Mean	Mean Diff	Mean Diff 95% CI	SD_{diff}	95% LOA	SEM
Speed (m/s)	0.78	(0.08, 0.99)	0.82	−0.08	(−0.21, 0.06)	0.16	(−0.40, 0.24)	0.12
Cycle Time (s)	0.86	(0.34, 0.97)	0.92	0.04	(−0.06, 0.13)	0.11	(−0.19, 0.26)	0.08
Double Limb Support Time (s)	0.84	(0.01, 0.97)	0.2	0.05	(0.01, 0.09)	0.05	(−0.05, 0.15)	0.03
Stride Length (m)	0.94	(0.65, 0.99)	0.74	−0.04	(−0.08, 0.01)	0.05	(−0.14, 0.07)	0.04
Stride Width (m)	0.94	(0.73, 0.99)	0.12	0.01	(0.00, 0.02)	0.02	(−0.02, 0.04)	0.01
Average	0.87							0.06
				Left lower Limb				
Cycle Time (s)	0.84	(0.31, 0.97)	0.92	0.06	(−0.05, 0.16)	0.12	(−0.19, 0.30)	0.09
Stance Time (s)	0.85	(0.33, 0.97)	0.58	0.05	(−0.03, 0.13)	0.10	(−0.15, 0.25)	0.07
Swing Time(s)	0.76	(0.00, 0.95)	0.35	0.01	(−0.03, 0.04)	0.04	(−0.07, 0.08)	0.03
Step Time (s)	0.79	(0.00, 0.96)	0.45	0.01	(−0.04, 0.06)	0.06	(−0.11, 0.13)	0.04
Step Length (m)	0.93	(0.63, 0.99)	0.38	0.00	(−0.03, 0.03)	0.04	(−0.08, 0.08)	0.03
Stride Length (m)	0.93	(0.63, 0.99)	0.75	0.00	(−0.07, 0.07)	0.08	(−0.16, 0.16)	0.06
Average	0.85							0.05
				Right lower Limb				
Cycle Time (s)	0.86	(0.30, 0.97)	0.93	0.02	(−0.08, 0.12)	0.12	(−0.21, 0.25)	0.08
Stance Time (s)	0.87	(0.44, 0.97)	0.57	0.04	(−0.03, 0.10)	0.08	(−0.12, 0.19)	0.05
Swing Time(s)	0.84	(0.24, 0.97)	0.36	−0.02	(−0.06, 0.02)	0.05	(−0.11, 0.08)	0.03
Step Time (s)	0.79	(0.00, 0.96)	0.46	0.00	(−0.07, 0.07)	0.09	(−0.16, 0.17)	0.06
Step Length (m)	0.64	(0.00, 0.93)	0.36	−0.05	(−0.12, 0.02)	0.08	(−0.21, 0.11)	0.06
Stride Length (m)	0.64	(0.00, 0.93)	0.72	−0.11	(−0.24, 0.03)	0.16	(−0.42, 0.21)	0.11
Average	0.73							0.07

Intraclass correlation coefficient, ICC; 95% CI, 95% confidence interval for the ICC; mean, mean of measurements at baseline trial and retest trial; mean diff, mean of the differences between measurements at time 1 and 2 and the 95% CI for mean diff, the standard deviation of the differences (SD_{diff}); 95% LOA, Bland and Altman 95% limits of agreement; SEM, standard error of measurement.

Table 4. Reliability values for kinematic parameters.

Kinematic Parameters	ICC	ICC 95% CI	Mean	Mean Diff	Mean Diff 95% CI	SD_{diff}	95% LOA	SEM
				Pelvic joint angle (°)				
				Left lower Limb				
Anterior Tilt +	0.40	(0.00, 0.88)	16.0	−0.1	(−5.2, 5.0)	6.1	(−12.24, 12.02)	4.3
Posterior Tilt −	0.83	(0.20, 0.97)	10.4	−1.2	(−5.2, 2.8)	4.7	(−10.58, 8.19)	3.3
Obliquity Up +	0.84	(0.20, 0.97)	2.7	0.5	(−1.7, 2.7)	2.6	(−4.69, 5.69)	1.8
Obliquity Down −	0.75	(0.00, 0.95)	−4.5	0.2	(−1.9, 2.3)	2.5	(−4.87, 5.28)	1.8
External Rotation −	0.44	(0.00, 0.89)	−6.6	0.2	(−6.4, 7.0)	8.0	(−15.55, 16.10)	5.3
Internal Rotation +	0.76	(0.00, 0.95)	13.7	1.1	(−5.0, 7.2)	7.3	(−13.32, 15.55)	5.2
Average	0.67							3.6
				Right lower Limb				
Anterior Tilt +	0.51	(0.00, 0.91)	16.1	−0.8	(−6.1, 4.3)	6.2	(−13.15, 11.37)	4.4
Posterior Tilt −	0.84	(0.31, 0.97)	10.3	−2.2	(−6.1, 1.6)	4.6	(−11.30, 6.82)	3.2
Obliquity Up +	0.67	(0.00, 0.94)	3.8	0.1	(−2.2, 2.5)	2.8	(−5.44, 5.81)	2.0
Obliquity Down −	0.85	(0.31, 0.97)	−2.7	−0.7	(−2.9, 1.3)	2.5	(−5.78, 4.20)	1.8
External Rotation −	0.88	(0.44, 0.98)	−12.0	−1.8	(−6.2, 2.4)	5.2	(−12.06, 8.32)	3.6
Internal Rotation +	0.85	(0.21, 0.97)	7.5	−4.2	(−8.7, 0.2)	5.4	(−14.86, 6.30)	3.8
Average	0.77							3.1
				Hip Joint angle (°)				
				Left lower Limb				
Flexion +	0.79	(0.00, 0.96)	45.0	−1.4	(−6.2, 3.5)	5.8	(−12.78, 9.98)	4.1
Extension −	0.78	(0.00, 0.96)	1.3	−0.7	(5.8, 4.3)	6.1	(−12.72, 11.24)	4.3
Abduction −	0.60	(0.00, 0.92)	−10.4	0.3	(−4.2, 4.9)	5.5	(−10.41, 11.15)	3.9
Adduction +	0.76	(0.00, 0.95)	4.8	0.8	(−2.7, 4.4)	4.3	(−7.62, 9.27)	3.0
External Rotation −	0.58	(0.00, 0.90)	−8.9	4.3	(−7.3, 18.0)	15.1	(−24.37, 35.08)	9.7
Internal Rotation +	0.67	(0.00, 0.92)	3.9	4.9	(−4.1, 16.0)	12.0	(−17.69, 29.66)	8.5
Average	0.70							5.6
				Right lower Limb				
Flexion +	0.14	(0.00, 0.85)	45.5	−0.9	(−9.1, 7.1)	9.7	(−20.10, 18.11)	6.9
Extension −	0.82	(0.12, 0.96)	1.5	−1.8	(−7.6, 3.9)	6.9	(−15.46, 11.80)	4.9
Abduction −	0.75	(0.00, 0.95)	−9.9	0.2	(−3.5, 4.1)	4.6	(−8.78, 9.37)	3.2
Adduction +	0.79	(0.00, 0.96)	6.9	−0.4	(−3.9, 3.0)	4.1	(−8.62, 7.71)	2.9
External Rotation −	0.67	(0.00, 0.93)	−10.7	−6.1	(−16.8, 4.4)	12.7	(−31.10, 18.77)	9.0
Internal Rotation +	0.73	(0.00, 0.95)	1.0	−4.0	(−14.5, 6.3)	12.4	(−28.54, 20.40)	8.8
Average	0.65							5.9

Table 4. *Cont.*

Kinematic Parameters	ICC	ICC 95% CI	Mean	Mean Diff	Mean Diff 95% CI	SD_{diff}	95% LOA	SEM
Knee Joint angle (°)								
Left lower Limb								
Flexion +	0.75	(0.00, 0.95)	70.6	0.2	(−6.5, 7.0)	8.1	(−15.71, 16.17)	5.7
Extension −	0.85	(0.17, 0.97)	8.6	0.4	(−3.6, 4.5)	4.9	(−9.15, 10.04)	3.4
Abduction −	0.48	(0.00, 0.90)	−7.4	0.5	(−5.1, 6.1)	6.7	(−12.68, 13.74)	4.7
Adduction +	0.46	(0.00, 0.90)	5.8	1.5	(−7.7, 10.9)	11.2	(−20.27, 23.42)	6.8
External Rotation −	0.75	(0.00, 0.95)	−8.4	−0.6	(−7.9, 6.6)	8.7	(−17.73, 16.45)	6.1
Internal Rotation +	0.62	(0.00, 0.92)	4.7	3.0	(−5.0, 11.0)	9.7	(−15.91, 21.94)	6.8
Average	0.65							5.6
Right lower Limb								
Flexion +	0.86	(0.25, 0.97)	68.5	−0.1	(−8.3, 8.0)	9.8	(−19.38, 19.13)	5.9
Extension −	0.98	(0.88, 0.99)	6.4	1.5	(−0.6, 3.6)	2.5	(−3.50, 6.50)	1.8
Abduction −	0.37	(0.00, 0.88)	−6.9	−2.0	(−10.2, 6.1)	9.8	(−21.31, 17.13)	6.9
Adduction +	0.33	(0.00, 0.87)	4.7	−3.8	(−14.2, 6.6)	12.4	(−28.21, 20.54)	8.7
External Rotation −	0.76	(0.00, 0.95)	−7.5	3.5	(−4.9, 12.1)	10.2	(−16.43, 23.61)	7.2
Internal Rotation +	0.00	(0.00, 0.69)	5.4	0.8	(−11.4, 13.0)	14.6	(−27.87, 29.49)	9.3
Average	0.55							6.6
Ankle Joint angle (°)								
Left lower Limb								
Dorsiflexion +	0.46	(0.00, 0.90)	9.8	3.3	(−9.0, 15.7)	14.8	(−25.69, 32.37)	10.4
Plantar Flexion −	0.27	(0.00, 0.86)	−11.1	2.6	(−10.5, 15.7)	15.7	(−28.22, 33.48)	11.1
Eversion −	0.60	(0.00, 0.91)	1.2	2.4	(−2.0, 7.0)	5.4	(−8.13, 13.11)	3.8
Inversion +	0.75	(0.00, 0.94)	13.0	1.6	(−3.1, 6.3)	5.6	(−9.44, 12.68)	3.9
Foot Internal Progression +	0.95	(0.75, 0.99)	3.8	−0.4	(−4.1, 3.1)	4.4	(−9.13, 8.14)	3.1
Foot External Progression −	0.87	(0.34, 0.97)	−14.3	1.4	(−7.5, 10.3)	10.6	(−19.40, 22.29)	6.5
Average	0.65							6.5
Right lower Limb								
Dorsiflexion +	0.40	(0.00, 0.82)	7.7	2.3	(−12.5, 17.1)	17.7	(−32.48, 37.14)	12.6
Plantar Flexion −	0.00	(0.00, 0.81)	−13.5	4.5	(12.8, 21.9)	20.7	(−36.15, 45.23)	14.6
Eversion −	0.43	(0.00, 0.76)	1.1	0.0	(−5.7, 5.7)	6.9	(−13.48, 13.52)	4.8
Inversion +	0.00	(0.00, 0.80)	14.1	0.0	(−3.9, 3.8)	4.6	(−9.11, 8.99)	3.2
Foot Internal Progression +	0.95	(0.78, 0.99)	−11.7	−3.1	(−9.0, 2.7)	7.0	(−16.97, 10.67)	4.9
Foot External Progression −	0.94	(0.72, 0.99)	29.3	−4.6	(−13.9, 4.6)	11.0	(−26.37, 17.05)	6.8
Average	0.45							7.8

Intraclass correlation coefficient, ICC; 95% CI, 95% confidence interval for the ICC; mean, mean of measurements at baseline trial and retest trial; Mean Diff, mean of the differences between measurements at time 1 and 2 and the 95% CI for mean diff, the standard deviation of the differences (SD_{diff}); 95% LOA, Bland and Altman 95% limits of agreement; SEM, standard error of measurement.

Table 5. Reliability values for kinetic parameters.

Kinetic Parameters	ICC	ICC 95% CI	Mean	Mean Diff	Mean Diff 95% CI	SD_{diff}	95% LOA	SEM
Hip Joint Moment (N m/Kg)								
Left lower Limb								
Flexion −	0.95	(0.76, 0.99)	−0.46	−0.02	(−0.11, 0.06)	0.10	(−0.22, 0.17)	0.07
Extension +	0.67	(0.00, 0.94)	0.50	0.02	(−0.12, 0.16)	0.17	(−0.31, 0.34)	0.12
Abduction +	0.79	(0.00, 0.96)	0.43	0.01	(−0.08, 0.10)	0.11	(−0.20, 0.22)	0.08
Adduction −	0.00	(0.00, 0.75)	−0.21	−0.05	(−0.28, 0.18)	0.28	(−0.60, 0.50)	0.20
Average	0.61							0.12
Right lower Limb								
Flexion −	0.84	(0.11, 0.97)	−0.37	0.01	(−0.12, 0.13)	0.15	(−0.29, 0.30)	0.11
Extension +	0.40	(0.00, 0.86)	0.47	0.08	(−0.13, 0.30)	0.26	(−0.43, 0.59)	0.18
Abduction +	0.73	(0.00, 0.95)	0.48	0.00	(−0.13, 0.12)	0.15	(−0.29, 0.29)	0.11
Adduction −	0.79	(0.16, 0.96)	−0.12	−0.04	(−0.11, 0.02)	0.08	(−0.20, 0.11)	0.06
Average	0.69							0.12
Knee Joint Moment (N m/Kg)								
Left lower Limb								
Flexion −	0.69	(0.00, 0.94)	−0.27	0.03	(−0.04, 0.11)	0.09	(−0.15, 0.21)	0.07
Extension +	0.79	(0.00, 0.96)	0.41	−0.02	(−0.19, 0.15)	0.21	(−0.42, 0.38)	0.15
Valgus +	0.72	(0.00, 0.95)	0.17	0.01	(−0.09, 0.11)	0.12	(−0.23, 0.25)	0.09
Varus −	0.76	(0.00, 0.95)	−0.16	0.06	(0.00, 0.12)	0.07	(−0.07, 0.20)	0.05
Average	0.74							0.09
Right lower Limb								
Flexion −	0.49	(0.00, 0.90)	−0.26	0.13	(−0.06, 0.32)	0.23	(−0.32, 0.58)	0.16
Extension +	0.92	(0.63, 0.98)	0.31	−0.06	(−0.19, 0.07)	0.16	(−0.36, 0.24)	0.11
Valgus +	0.00	(0.00, 0.78)	0.27	−0.13	(−0.39, 0.13)	0.31	(−0.74, 0.48)	0.22
Varus −	0.61	(0.00, 0.92)	−0.14	−0.04	(−0.12, 0.03)	0.09	(−0.23, 0.14)	0.07
Average	0.51							0.14
Ankle Joint Moment (N m/Kg)								
Left lower Limb								
Dorsiflexion −	0.72	(0.00, 0.95)	−0.02	0.01	(−0.01, 0.04)	0.03	(−0.05, 0.08)	0.02
Plantar Flexion +	0.93	(0.61, 0.99)	0.85	0.00	(−0.12, 0.11)	0.14	(−0.27, 0.26)	0.10
Eversion +	0.57	(0.00, 0.92)	0.07	0.02	(−0.05, 0.08)	0.08	(−0.14, 0.17)	0.06
Inversion −	0.75	(0.00, 0.95)	−0.13	0.02	(−0.06, 0.09)	0.09	(−0.16, 0.19)	0.06
Average	0.74							0.06
Right lower Limb								
Dorsiflexion −	0.00	(0.00, 0.77)	−0.02	−0.02	(−0.05, 0.02)	0.04	(−0.10, 0.06)	0.03
Plantar Flexion +	0.78	(0.00, 0.96)	0.75	−0.01	(−0.15, 0.13)	0.17	(−0.34, 0.32)	0.12
Eversion +	0.85	(0.21, 0.97)	0.04	0.00	(−0.03, 0.03)	0.04	(−0.07, 0.07)	0.03
Inversion −	0.55	(0.00, 0.91)	−0.16	−0.03	(−0.18, 0.13)	0.18	(−0.39, 0.33)	0.13
Average	0.55							0.08

Intraclass correlation coefficient, ICC; 95% CI, 95% confidence interval for the ICC; mean, mean of measurements at baseline trial and retest trial; mean diff, mean of the differences between measurements at time 1 and 2 and the 95% CI for mean diff, the standard deviation of the differences (SD_{diff}); 95% LOA, Bland and Altman 95% limits of agreement; SEM, standard error of measurement.

3.4. Reliability of Kinetic Parameters

For the ICCs of kinetic parameters, the results were higher than those for the kinematic data, where the majority were $\geq$0.75 (Table 5). The lowest ICCs between sessions were found in right knee joint valgus moment (0.00, 95% CI 0.00 to 0.78), right ankle dorsiflexion (0.00, 95% CI 0.00 to 0.77) and left hip joint adduction moment (0.00, 95% CI 0.00 to 0.75). The SEM values ranged between 0.1 Nm/Kg to 14.7 Nm/Kg and averaged between 0.1 Nm/Kg and 0.1 Nm/Kg.

4. Discussion

The purpose of this study was to evaluate the inter-session reliability and measurement error of a 3D gait analysis protocol in a group of CP children, in order to better understand the causes of intrinsic and extrinsic variation. Knowing this variability is crucial to improve clinical analysis that supports decision-making in the rehabilitation process.

Ferrari et al. [17] have found that when comparing five protocols on the same gait cycles, the main cause for the variability of outcomes between variables was the biomechanical model used and its definitions, regardless of the number of raters or even different laboratories. These different biomechanical models make it more difficult to compare results between reliability studies, as they present different sources of variability [17]. Repeated testing of a single subject allows for a clinical usefulness of the data, since it provides some understanding into the extent of variation of the measured outcomes that can be expected due to the pathology and those that are truly a consequence of a therapeutic intervention [15].

Despite extreme caution and compliance with the protocol instructions regarding the marker placement procedure, some inconsistency is still unavoidable [16], while possible sources of error can occur due to subjects' natural oscillations or skin motion [13] or movement between the skin markers and the underlying bones [50,51]. This source of error is totally disruptive for the joints with a limited range of motion, such as knee abduction–adduction, internal–external rotation, and linear displacements [52,53].

CP children can demonstrate different gait patterns in each leg. This occurs not only in unilateral spastic CP, where each legs presents different kinematic values [23], but also in some bilateral spastic CP children with an asymmetrical gait pattern, combining at least two different types of gait pattern [48]. A previous study by Mackey et al. [26] used the 6DoF Cleveland marker set with unilateral CP children and presented similar results at both normal and hemiplegic limbs, where the highest repeatability was at the sagittal plane (CMC values of 0.96–0.99) and lower in the transverse and frontal planes (CMC $\geq$ 0.7), In this study, the CP children presented different gait patterns (Table 1): five had bilateral spasticity, two had unilateral spasticity with their right limb affected and one was affected in the left limb, which contributed to some degree of variation of the data. The overall ICC results of kinematic and kinetic variables were lower on the right side, which can indicate that—to some degree—the instability of the affected lower limbs could influence the propagation of the STA. Reinschmidt and co-investigators reported that the soft tissue motion can originate additional movement, resulting in an overestimation in kinematic peak values of the segments by as much as 100% [54]. This is in accordance with our research, where a larger variation was noted in the transverse and frontal planes of the knee (Table 4). In the 6DoF models it is assumed that the limbs' segments are independent and do not share a fixed joint centre, which often originates non-physiological translations between the proximal and distal bones at some joints [22]. However, in pathological gait, care should be taken because non physiological movements may occur.

Typically true equinus gait patterns constrain CP children to stand with the ankle in a neutral position [48]. However, according to Schlough et al. [55] when passive dorsiflexion is detected in the clinical examination, it is possible for some subjects to walk with their feet flat on the ground upon request. This variability in walking pattern during development is considered typical. Nevertheless, when unable to perform heel contact, some biomechanical compensation is detected, mainly in the coordination of movement at the hip, knee and ankle joints. In this study, one subject presented mild spastic diplegia and a considerable gastrocnemius tone (as seen in Table 1), which often shows similar characteristics to idiopathic toe walking. In the first session, the subject was able to perform a normal heel strike at initial contact and during the stance phase of walking. However, during the dynamic trials in the second session, the gastrocnemius stiffness was significantly higher which caused some motion restriction at the ankle. As, in the static calibration trial, the subject was able to stand with both feet flat on the floor, the range of motion differences were wider from the start. The magnitude of this variation is visible in the scatter plots of the dorsi/plantar flexion (Figure 2). When we compare the kinematic data between sessions, there was an increase of 8° in hip flexion, a decrease of 13° in knee flexion and a total absence of ankle dorsiflexion in both lower limbs. These results are in accordance with the study of Hicks et al. [56] where CP children with toe walking often exhibited increased hip flexion and a decrease in knee flexion throughout the walking cycle. Furthermore, excessive plantar flexion may be responsible for changes in flexion, internal rotation and adduction of the hip as well as in the pelvic anterior tilt [33] which explains the reduced ICC on left and right anterior tilt (0.40 and 0.51, respectively) compared with the other kinematic variables of this segment, as seen in Table 4.

Yet, due to co-spasticity of the muscles causing reciprocal movements across the joints and originating a wider variation in kinematic data, CP children are not able to change joint moments which results in a more reliable measure between the two assessment days [57]. This is evident in our results where the kinetic variables presented less variation (Table 5), in accordance with similar studies [16,23]. Although there is no reliability analysis published with a 6DoF model and kinetics variables, these results may be partially attributed to the small variations of the anthropometric measurements. Even though the two recorded sessions occurred several days apart, there was a small variation in marker placement between sessions (Table 2). Anthropometric measurements were considered excellent regarding ICC (ICC average $\approx$ 0.98) and an absolute error of approximately 4 mm.

Limitations

The number of CP children included in similar studies varies from 5 to 20 [23–26,44] and even though this gait protocol was performed with 8 CP children, the analysis of the right and left legs imply distinguished experiments, involving independent landmark identification, marker attachment, anthropometric measurements, and data processing [17]. Consequently, the current research should be considered as an independent analysis of sixteen legs.

Given that every gait research laboratory uses its own marker set and gait model, in order to compare gait analysis data, all the specific methodology used in each process must be considered. Regardless of the set of techniques chosen, there will always be different measurement errors that can influence the outcomes and consequently, a clinical interpretation. These differences have a greater impact in the kinematic and kinetic outcome measures (e.g., joint angles and moments). Thus, gait protocols should be described in detail to allow a contextualized interpretation of the results and comparison between similar investigations. This should be done in a critical manner on all the variables during the gait cycle, rather than only interpret the absolute values presented, regardless of the measures of repeatability or correlation used [15]. It is of great relevance when it comes to gait assessment of CP children who have an intrinsic gait variability due to their neuromuscular impairments. In these cases, it is crucial to differentiate the methodological errors (raters error) from the participants' natural variability and from the effect of a rehabilitation process.

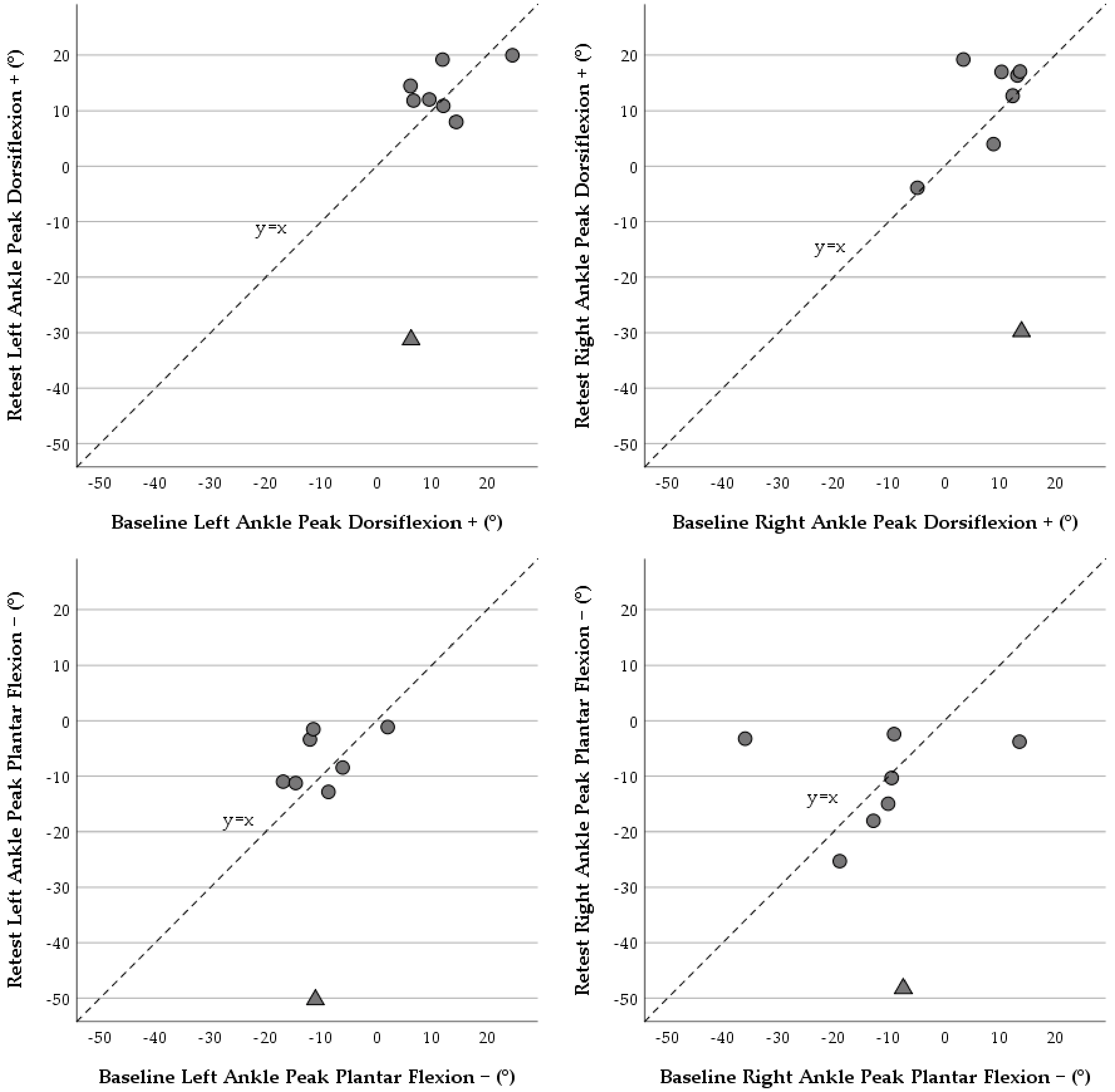

Figure 2. Scatter plots for ankle peak joint angles for dorsiflexion and plantar flexion. Subject with increased gastrocnemius stiffness values is represented with a different symbol from the rest.

Due to the different gait analysis protocols used, the influence of the number of gait cycles in test–retest reliability measurements [11] remains to be determined. Although in general, repeatability increases with a higher number of gait cycles, this is true mainly for the kinematic data. All the time-distance and kinetic parameters do not reveal significant differences from the fifth gait cycle onwards. In addition, the assessment of more than five gait cycles in a clinical setting may be difficult to accomplish due to the preparation of the subject [34]. Regarding CP children, this can be a very complex and difficult task, therefore the five gait cycles used in this protocol were shown to be quite good in achieving reliable results.

5. Conclusions

This study indicates wide-ranging reliability values for lower limb joint angles and joint moments of force during gait, especially for frontal and transverse planes. Although the use of a 6DoF-CAST in CP children was shown to be a feasible method, the gait variation that can be observed between sessions in CP children seems to be related not only to the extrinsic factors but also to their different gait patterns and affected sides. In future research, it could be interesting to assess the reliability of these models using different groups of subjects, according to their gait pattern, for instance. These models and their technical characteristics still require some improvements in order to support clinical decision-making.

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Institutional Review Board Statement: The study was conducted according to the guidelines of the Declaration of Helsinki and approved by and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019).

Informed Consent Statement: An informed consent was signed by the parent or the legal guardian of the participant.

Data Availability Statement: All data generated or analysed during this study are included in this published article.

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Appendix 3

Ricardo, D.; Teles, J.; Raposo, M.R.; Veloso, A.P.; João, F.

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5.85
Reliability of Lower Extremity Kinematics in Three-Dimensional Gait Measurements in Children with Cerebral Palsy

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BACKGROUND
Cerebral Palsy is a complex pathology that describes a group of motor disorders[1] with different presentations and functional levels[2]. Three-dimensional gait analysis is used in the assessment of CP children to assist in clinical decision making and assessing outcomes in the rehabilitation process[3]. Due to the CP intra-subject gait variability[4], it's crucial to access the repeated gait measurements to evaluate the response to therapeutic interventions in the rehabilitation process.

AIM
The objective of this study was to determine the intra-subject measurement reliabilities of the kinematic parameters of children with hemiplegic and diplegic type of cerebral palsy.

METHOD
The study group was composed of a convenience sample of 8 CP children able to walk independently (2 hemiplegic, 6 diplegic; 2 female, 6 males; age 87.88±25.56months; height 1.17±0.14m; mass 24.25±8.26kg). Two trials were performed on two different days within period of 7.5±1.4days. Two assessors positioned the reflective marker set based on the CAST protocol[5]. Data was collected with 8 infrared, high-speed cameras working at 200 Hz. Intraclass correlation coefficients considering the two-way mixed model, absolute agreement (ICC[2,k])[6,7] for these trials were calculated for kinematic parameters. The ICC level of clinical significance was considered poor, fair, good, and excellent when ICC<0.40, 0.40<ICC<0.59, 0.60<ICC<0.74, 0.75<ICC<1.00[8].

RESULTS
Intraclass correlation coefficients were calculated for the joint angles peak values in the gait cycle (from heel strike to terminal swing) on both sides. The considered joint angles were in the range of motion of the flexion/extension, abduction/adduction, and rotations, in the pelvis, hip, knee, and ankle. Reliability of kinematics was excellent in the most variables (22 of 48), good (10 of 48), fair (8 of 48) and poor (8 of 48). Overall, joint angles of the right lower limb showed the greatest differences and variability, mainly in the Hip flexion (0.141, 95%CI – 0.000 to 0.842); Knee abduction (0.370, 95%CI–0.000 to 0.879) and adduction (0.332, 95%CI – 0.000 to 0.807); Ankle plantar flexion (0.000, 95%CI – 0.000 to 0.807) and inversion (0.000, 95%CI – 0.000 to 0.755).

DISCUSSION AND CONCLUSION
In this study, there were greater differences in frontal and transverse planes of the right lower limb. The inclusion of two hemiplegic CP children affected on the right lower limb, may have contributed to a larger variability of the assessments due to the irregularity of muscle spasticity that can affect the participants gait patterns and consequently the joint angles. Presented data shows that most of the assessed kinematics measurements can be replicated to a moderate extent.

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Appendix 4

João, F.; **Ricardo, D.**; Raposo, M.R.; Veloso, A.P.

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Effect of different pose estimation algorithms in gait kinematics of cerebral palsy children using ankle foot orthosis (work in progress)

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1. Introduction

Pose estimation algorithms are fundamental when using rigid bodies models to assess the kinematics of human movement [1–3]. Different methods are used to minimize issues as the soft tissue artefact. Its efficacy depends of which joint constraints are applied in accordance with a specific model [3]. The ankle-foot orthoses (AFO) are widely used in cerebral palsy (CP) children with an abnormal gait pattern. However, the pose estimation of the lower limb segments usually follows the same modelling approach despite the constraints that the orthosis may cause. To overcome this limitation and having in mind the constraints that the AFO is supposed to inflict in the lower limb, the purpose of this work in progress is to compare the kinematic data of two different pose estimation algorithms models (Global Optimization and Segment

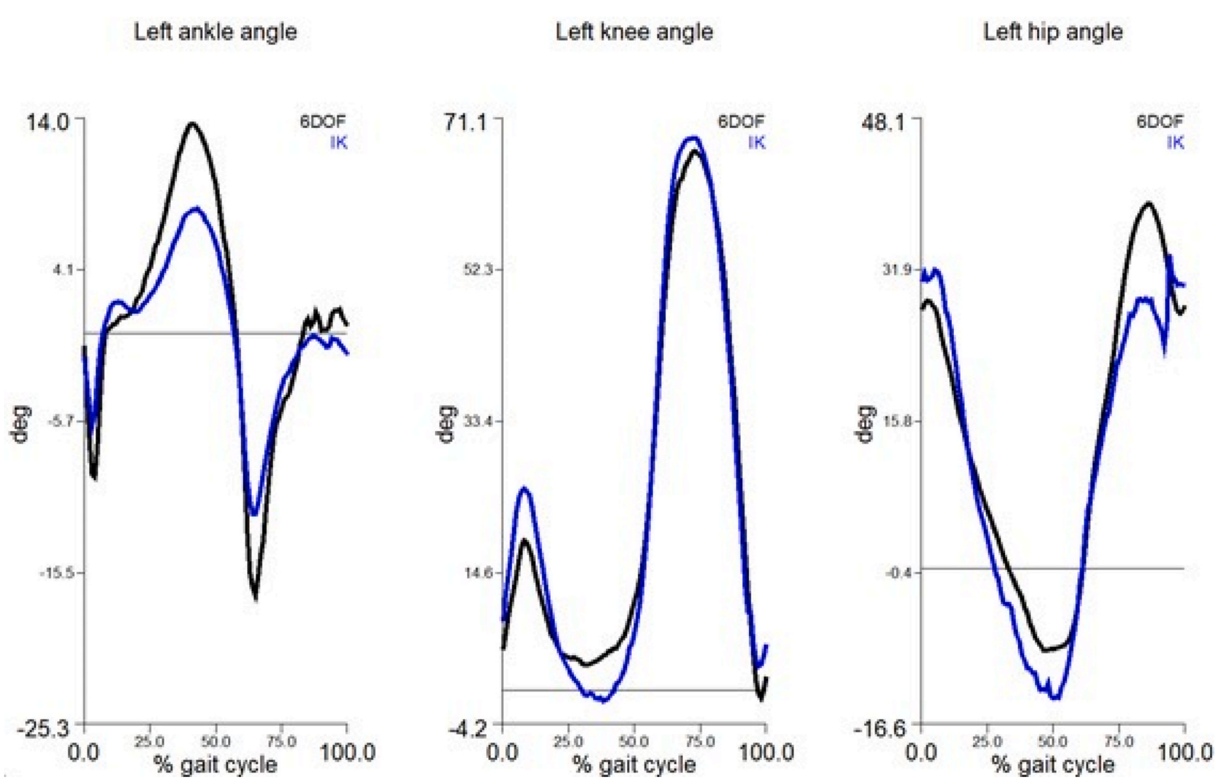

Fig. 1. Ankle, knee and hip kinematics with the 2 optimization models.

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Optimization) [4] during gait of CP children wearing AFO.

2. Research question

What are the differences in gait kinematics of cerebral palsy children wearing AFO, when using two different pose estimation algorithms models: Global Optimization and Segment Optimization?

3. Methods

Gait analysis was performed on 8 CP children using AFO, aged between 5-10 years old. It was used a 14 camera-based system and 3 force plates. 53 passive reflective markers were placed on specific anatomic places, using CAST protocol [5] and CODA pelvis, allowing the reconstruction of seven body segments. Children were instructed to walk along a 10m corridor, at self-selected speed wearing the AFO with the usual footwear. The pose of the lower limbs and pelvis was estimated using two algorithms: 1) a global optimization where each segment had 6 degrees of freedom (6dof), 3 rotations and 3 translations and 2) a segment optimization where no translations were allowed to the thigh, shank and foot segments, the hip and the knee joints were allowed to rotate in the 3 axis, and the ankle joint could only rotate in the flex/ext axis. All the kinematic variables were calculated using Visual 3D software (v4.80.00, C-Motion, Inc, Rockville, USA).

4. Results

The lower limb joint angles in the sagittal plane are shown in Fig. 1 for one subject wearing a supramaleolar AFO.

5. Discussion

The main differences were found in the ankle joint kinematics. The dorsiflexion peak in the stance phase and the plantarflexion peak in the push off seems to be overestimated using the 6DOF model. The proximal lower limb joints also presented differences between the two models, more pronounced in the stance phase for the knee joint and at the midstance and initial swing for the hip joint. The orthoses configuration to prevent plantarflexion thus improving clearance in swing and first ankle rocker seems to be more in accordance when using a model with constraints that closely match that configuration.

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