Universidade de Lisboa

Faculdade de Motricidade Humana

Plantar Pressure Gait Analysis in Children with Cerebral Palsy

Maria Raquel Branco Raposo

Orientadora: Professora Doutora Filipa Oliveira da Silva João

Tese especialmente elaborada para obtenção do grau de Doutor no ramo Motricidade Humana, na especialidade de Biomecânica

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2023

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Título da Tese: Plantar Pressure Gait Analysis in Children with Cerebral Palsy

Orientadora: Professora Doutora Filipa Oliveira da Silva João

Ano de conclusão: 2023

Ramo de conhecimento do Doutoramento: Motricidade Humana, especialidade de Biomecânica

Faculdade de Motricidade Humana – Universidade de Lisboa

Cruz Quebrada, 2023

Assinatura: _____

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Acknowledgments

"Do the best you can until you know better. Then when you know better, do better!"

Maya Angelou

It has been a long and fruitful journey of learning and doing better every day for the past 6 years. I would like to express my gratitude to all of those with whom I met and supported me, in particular:

- To my supervisor, Professora Filipa João, for all her guidance through the hardest times, and never allowing me to give up! Thank You!
- To my colleague, Diogo Ricardo, for the continuous support and friendly shoulder, thank you!
- To Professor António Prieto Veloso, for the encouragement and support to pursuit roads never before explored, thank you!
- To Sílvia Cabral, for all the availability and technical support, thank you!
- To all the co-authors with whom I had the pleasure to work with, for their support: Júlia Teles, Raul Oliveira, Filomena Carnide, Eduardo Brazete Cruz. Thank you!
- To my co-workers, for the helpful laughs on the dimmest days! Thank you!
- To my wonderful family, for my upbringing that allowed to fight and pursue my dreams and their endless love, thank you!
- To my beautiful daughter, my sweetheart, thank you for bringing an explosion of love into my life!
- To all children and adults with Cerebral Palsy with whom I have had the pleasure to work though the last 12 years, you matter! Thank you!

Abstract

Title: Plantar Pressure Gait Analysis in Children with Cerebral Palsy

Abstract: Cerebral Palsy (CP) is the most common childhood neurologic impairment. Children with CP are often prescribed ankle-foot orthoses (AFO) as an intervention meant to improve gait, but the evidence that supports its generalized use is scarce. Gait analysis is considered a gold standard in CP rehabilitation, and plantar pressure analysis can provide useful information about the way that the foot interacts with the orthoses and the ground. Plantar pressure insoles are a simple and non-invasive technology, but still an underused tool in this context. By combining the use of two rehabilitation tools (AFO and plantar pressure analysis), this PhD thesis aims to contribute to deepen the knowledge and available evidence on the field. Three separate scientific investigations were conducted: a scoping review, aiming to systematize the available evidence about the effects of different types of AFO on the gait of children with spastic bilateral cerebral palsy, showing that AFO have a positive impact in the gait of children with cerebral palsy; a test-retest reliability analysis and minimal detectable change of plantar pressure insoles in a sample of children with CP when walking in regular footwear, that determined high reliability (ICC ≥ 0.60) for 21 of the 24 parameters that were tested; and lastly a descriptive study of the plantar pressure distribution characteristics of children with cerebral palsy, while wearing plantar pressure insoles and walking with AFO, where there were positive changes in plantar pressure measurements, approximating them to the reference percentiles of typically developing children. There is a need to continue to invest in these lines of investigation, namely producing consistent evidence about the effects of AFO, unwavering prescription guidelines and producing a normative database for plantar pressure measurements in children with Cerebral Palsy.

Keywords: “Plantar Pressure “; “Cerebral Palsy”; “Gait”; “Ankle Foot Orthoses”; “Insoles”

Resumo

Título: Análise de Marcha e Pressões Plantares em Crianças com Paralisia Cerebral

Resumo: A Paralisia Cerebral é o comprometimento neurológico mais comum na infância. Ortóteses de pé e tornozelo são comumente prescritas a crianças com Paralisia Cerebral, com o objetivo de melhorar o padrão de marcha, mas a evidência que suporta seu uso generalizado é escassa. A análise da marcha é considerada um *gold standard* na reabilitação da Paralisia Cerebral, e a análise de pressões plantares pode fornecer informações úteis sobre a maneira como o pé interage com as ortóteses e o solo. Neste sentido, as palmilhas de pressão plantar são uma tecnologia simples e não invasiva, mas ainda uma ferramenta subutilizada neste contexto. Ao combinar o uso de duas ferramentas de reabilitação (ortóteses de tornozelo e pé e palmilhas de pressão plantar), esta dissertação visa contribuir para aprofundar o conhecimento e a evidência disponível na área. Foram realizadas três investigações científicas distintas: uma *scoping review*, com o objetivo de sistematizar a literatura sobre os efeitos de diferentes tipos de ortóteses de tornozelo e pé na marcha de crianças com paralisia cerebral bilateral espástica, mostrando que as ortóteses de tornozelo e pé têm um impacto positivo na marcha de crianças com paralisia cerebral; uma análise de confiabilidade teste-reteste e diferença mínima detetável em palmilhas de pressão plantar com uma amostra de crianças com Paralisia Cerebral, utilizando o seu calçado regular, que determinou alta confiabilidade (ICC ≥ 0,60) para 21 dos 24 parâmetros testados; e por último um estudo descritivo das características da distribuição da pressão plantar de crianças com paralisia cerebral, usando palmilhas de pressão plantar e ortóteses tornozelo-pé, onde se registaram mudanças positivas nos valores de pressão plantar, aproximando-os dos percentis de referência de crianças de desenvolvimento típico. É necessário continuar a investir nestas linhas de investigação, nomeadamente produzindo evidência consistente sobre os efeitos das ortóteses de tornozelo e pé, indicações de prescrição concretas e criando uma base de dados normativa para as medições de pressão plantar em crianças com Paralisia Cerebral.

Palavras-Chave: “Pressão Plantar”; “Paralisia Cerebral”; “Marcha” “Ortótese de Tornozelo e Pé”; “Palmilhas”

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List of Abbreviations

AFO - Ankle Foot Orthoses

CCT – Controlled Clinical Trials

COP – Center of Pressure

CP - Cerebral Palsy

DAFO - Dynamic Ankle Foot Orthoses

GMFCS - ER - Gross Motor Function Classification System – Expanded and Revised

GMFM - Gross Motor Function Measure

GRAFO - Ground Reaction Ankle Foot Orthoses

HAFO - Hinged Ankle Foot Orthoses

IC – Initial Contact

IS – Initial Swing

LOA – Limits of Agreement

LR – Loading Response

MDC – Minimal Detectable Change

MS – Mid-Swing

MSt – Mid-Stance

PLS – Posterior Leaf Spring

PS – Pre-Swing

RCT – Randomized Clinical Trials

ROM – Range of Motion

SAFO – Solid Ankle Foot Orthoses

SD – Standard Deviation

SEM - Standard Error of Measurement

TSt – Terminal Stance

TS – Terminal Swing

Chapter I – Introduction

This dissertation reflects the work developed during the doctoral program in Biomechanics, focusing mainly on gait analysis of children with Cerebral Palsy (CP) while wearing ankle-foot orthoses (AFO). While CP is the most common childhood neurologic impairment, there are still many understudied aspects of this condition, one of which, the prescription and use of AFO as an intervention meant to improve gait. After reviewing the available evidence, posing the research questions and hypothesis, we now present the completed original investigation. Certainly, many questions remained unanswered and further studies still need to be conducted.

1.1. Dissertation Objectives

This research is based on the following aspects:

CP is a prevalent condition in the Portuguese population, with very different presentations and needs. This condition has been widely studied and although a huge body of evidence already exists, a lot of questions remain unanswered.

AFO are commonly recommended and prescribed intervention for this population, though evidence regarding its use its still lacking. There are a wide selection of materials and shapes of orthoses in the market, as well as new technologies emerging, that make prescription increasingly difficult.

Gait analysis is considered a gold standard in CP rehabilitation, and plantar pressure analysis can provide useful information about the way that the foot interacts with the orthoses and the ground. Under this aspect, plantar pressure insoles are a simple and non-invasive technology, but still an underused tool in this context.

The need to develop an evidence based clinical practice calls to pose the following research questions:

- Is there available evidence to systemically assess the effects of AFO in the different gait patterns of children with CP?
- Is pedobarography a useful and reliable tool to assess plantar pressure parameters in children with CP?

- What are the effects of AFO on the plantar pressure parameters of different clinical presentations, different gait patterns and different types of orthoses in children with CP?

1.2. Dissertation Overview

The present dissertation aims to display the investigation and research conducted during the PhD process. It includes three studies (either published or submitted for publication in a reference journal), in which we intend to answer the different research questions.

In **Chapter I**, a general introduction is presented, that includes the main research topics, as well as the rationale behind each study.

In **Chapter II**, the candidate presents a literature overview that summarizes the state of evidence for the main topics addressed in the subsequent chapters that include but are not limited to CP physiopathology and epidemiology, pathological gait and laboratory gait analysis (including pedobarography) and the use of AFO.

In **Chapter III,** the methodology of the different investigations is discussed, aiming to justify, based on the best available evidence, the options that were made for each study.

As for **Chapter IV**, entitled "Effects of ankle foot orthoses on the gait patterns in children with spastic bilateral Cerebral Palsy: a scoping review", it comprises a scoping review aiming to systematize the available evidence about the effects of different types of AFO on the gait of children with spastic bilateral CP. The publication can be found in Appendix I.

In **Chapter V** we find the study "Gait Analysis in Children with Cerebral Palsy: Are Plantar Pressure Insoles a Reliable Tool?" which originated our second publication (Appendix II). This study intends to determine to reliability and minimal detectable change of plantar pressure insoles in a sample of children with CP when walking in regular footwear.

Chapter VI shows the final research paper, entitled “Plantar Pressure Analysis in Children with Cerebral Palsy While Wearing Orthoses – a Descriptive Study”, a descriptive study of the plantar pressure distribution characteristics of children with CP, while wearing plantar pressure insoles and walking with AFO. This study is currently submitted in Scientific Reports, pending reviewing and publication.

Finally, in **Chapter VII** we discuss the main findings achieved during the research studies, pointing out the limitations and suggestions for future research. The document ends with a global references (**Chapter VIII**) and the **Appendix**.

Chapter II – Literature Review

2.1. Cerebral Palsy

CP is a complex pathology that describes a group of impairments and motor disorders with different presentations and functional levels. It is the result of a non-progressive insult to the central nervous system on precocious stage of its development that can cause sensory, perceptive, cognitive, communications, feeding impairment, epilepsy and musculoskeletal deformity that consequently lead limited function and difficult on daily activities[1].

CP can be classified by severity, distribution, type of muscle tone, gait pattern, and even by functional abilities and impairment.

Severity refers to how much the motor limitations affect daily function of a person with CP. A commonly used tool to access severity is the Gross Motor Function Classification System – Expanded and Revised (GMFCS – ER). The GMFCS – ER is a tool developed by Palsiano et al. (2008)[2] that classifies self-initiated gross movement of children with CP, in tasks such as sitting, transferring from different positions and surfaces, walking and overall mobility. There are five different levels of severity (level I being the least severe, and level V the most affected) that differentiate the impact of the impairment in the child's daily life. It assess and includes the use of different types of support and mobility and daily life aids. It has been adapted for the Portuguese population from ages under 2 years old up to 18 years of age.

Distribution of CP characterizes the topographic affection of motor impairment and spasticity in the person with CP. It can be labelled Unilateral or Bilateral, referring to affecting mainly one side of the body or both. Unilateral CP usually affects the lower limb and upper limb of one side of the body, with little to no expression on the trunk. Bilateral CP can affect both lower limbs (commonly designated by diplegia), usually with some impact on trunk control, or it can affect 3 or 4 limbs at once (usually called tetraplegia) with trunk and head control being affected too.

All sub-types of CP have some form of movement and posture alterations. Spastic CP presents with increased reflexes and tone that is velocity dependent. Dyskinetic CP presents with involuntary and recurring movements, and a varying muscle tone. The Dyskinetic group encompasses two distinct sub-groups (Dystonic and Choreo-athetotic).

The Dystonic sub-group is characterized by a predominately high, but fluctuating tone and sustained abnormal postures. The Choreo-athetotic CP presents with a combination of rapid, fragmented and irregular movements and slower, reptilian or contorting movements, and a lower muscle tone. Less frequent, Ataxic CP presents significant muscle co-contraction throughout the range of motion, often appearing to shake or tremble and lower tone[3].

Overall, muscle control and tone is severely affected in children with CP (regardless of sub-type), often with spasticity, hyper-reflexia and agonist and antagonist co-contration as positive features, and weakness, difficulties in selective motor control, sensory impairments and poor postural control as negative features. Although spasticity is usually the focus of medical interventions, muscle weakness and muscle control are determinants to the success of acquiring and maintaining walking ability[4].

About half of the population with CP can walk, however most will display visible deviations in the different planes of motion, especially when compared to the gait pattern of their typically developed peers. The specifics of gait pathology and biomechanical deviations will be addressed in the "Typical and Pathological Gait" sub-section of this thesis.

One of the main measures of function in the population with CP is the Gross Motor Function Measure (GMFM). The GMFM is a widely used toll for assessing motor function in children with CP. Its different versions have all showed to be valid and reliable and have good psychometric properties[5,6]. Based on GMFM longitudinal assessments, reference percentiles were created, that may help to predict the motor acquisitions and prognosis of children with CP[7].

Despite CP affecting mainly motor function, it is urgent to use assessment models that can give an overall perspective of the abilities and limitations of the subjects. The International Classification of Functionality (ICF) is the World Health Organization (WHO) framework for measuring health and disability at both individual and population levels, and is used as a common language between different professionals[8]. Specific ICF score sheets have been developed for children and adults with CP[9]. ICF and GMFM have showed a significant correlation, as a better physical function demonstrates a better level of activity and participation and better quality of life[10].

2.2. Epidemiology

The National Cerebral Palsy at 5 years old Surveillance Program ("Programa Nacional de Vigilância em Paralisia Cerebral")[11] collets and treats data of children born in Portugal, between 2001 and 2012, aiming to report the incidence and prevalence of CP and its impact on the Portuguese population. According to the program, it is estimated that 1.61 children in thousand born are diagnosed with CP.

One of the most prevalent identified risk factors for CP is pre-term birth. Especially extremely preterm birth (less than 28 weeks gestation) and very preterm birth (between 28- and 32-weeks' gestation) can increase the risk of CP by 85 and 50 times, when compared to a full-term birth (after 37 weeks gestation). Other risk factors include twin pregnancies, maternal age (women over 39 at the date of birth), congenital malformations and low birth weight.

Most children with CP in Portugal are male (58.1%), with male children being 30% more at risk than female children. Even when associated with other risk factors, like extreme preterm birth, the proportion of male babies affected (49.2%) is higher than that of female babies (44.5%).The most frequent presentation CP is spastic (77%), followed by dyskinetic (10%) and ataxic (5%). Regarding global motor function (GMFCS), about 50% of children present with the more severe levels (III, IV and V), and the other 50% present are able to walk unaided (levels I and II). Over the last few years the percentage of severe cases has been on the rise.

Magnetic resonance imaging is a gold standard in confirming and defining aetiology of CP. About 78% of the population studied had a neuroimaging exam done at least once. From that collected data, it was confirmed that, most often the lesions are located in the white matter of the brain, and consistent with periventricular leukomalacia. Yet a steady percentage of children with CP (10%) do not show any abnormalities in their neuroimaging exams.

Gross motor function and bimanual motor function are more severely affected in children with spastic bilateral CP (with four limb involvement) and dyskinetic CP.

Other associated comorbidities include visual, auditory and cognitive deficits, epilepsy, mal-nutrition and musculoskeletal deformities. A significant portion of the children had a sub-luxation (19%) or luxation (4%) of the hip joint. Other musculoskeletal deformities, like foot deformities, are not directly report, and so, there is not data to assess its prevalence. However, we can infer that most ambulant children with CP will present with one or more musculoskeletal deformities and biomechanical misalignments.

2.3. Typical and Pathologic Gait

Locomotion is a fundamental human need. Humans are bipeds, with a higher gravity center (located in front of the S2 vertebra) and reduced support base, with overall less stability and efficiency than most mammals. This accounts for the prolonged time that takes a baby/child to acquire and consolidate gait. Children begin to develop gait around the first year and this process can mature up to the sixth year of their lives[12].

Gait requires a complex central control system that starts in the motor cortex and ends in the motor neuron, an efficient energy source that must provide oxygen and metabolic fuel for the muscular system to process, and levers and forces that when acted upon produce a moment. Walking is a compromise between the internal moments generated by the muscles and the external moments generated by the ground reaction and inertial forces[12].

Initially small children walk with a wide base of support, little balance and flat feet on the floor, so called stepping. As the child grows, central nervous systems and musculoskeletal systems matures, and the gait pattern will progressively resemble that of an adult[12].

Gait can be divided in two main phases, stance and swing with each one taking 60% and 40% of the gait cycle, respectively. The typical gait cycle initiates when the foot strikes the ground (Initial Contact – IC), progresses and bares weigh through the stance phase (Loading Response – LR; Mid-Stance – MSt; Terminal Stance – TSt; Pre-Swing - PS), swings in the air while advancing in space (Initial Swing – IS; Mid-Swing – MS; Terminal Swing – TS) and finishes when that same foot hits the ground again[12,13].

The normal gait cycle joint kinetics and kinematics has been thoroughly described, and involves sequence of muscle and joint responses that originate proximally and evolve distally. Temporal parameters, such as walking speed, cadence, step-length and stride length are also useful in characterizing the gait cycle[13].

Pathological gait frequently lacks prerequisites that make typical gait fluid and energy efficient, such as a stable stance phase, foot clearance and foot pre-positioning in swing phase, an adequate step length and the means to conserve energy[14].

About half of the population with CP can walk, however most will display visible deviations in the different planes of motion, especially when compared to the gait pattern of their typically developed peers. The brain insult that occurs at an early age affects tone, balance and muscular control, imposing abnormal internal forces on a growing skeleton that overtime result in musculoskeletal deformities that lead to increasing gait deviations[4]. This cycle can be moderated by early intervention, and therefore it is of the utmost importance to fully understand the different gait patterns of children with CP.

A few different authors have proposed abnormal gait pattern classifications for children with CP, but the two most used are Winters et al. (1987), revised by Rodda and Graham (2001)[15] for spastic hemiplegia and Rodda et al. (2004)[16] for spastic diplegia. These classifications are frequently used and quoted in the available literature and a common language for research in gait analysis of children and adults with CP. They summarize the complex characteristics of pathological gait in a manageable format.

Rodda et al., (2004)[16], sagittal gait patterns in spastic diplegia classification proposes five different groups, describing the abnormal motion on the hip, knee and ankle joints, as shown in Table 1.

Group I, True Equinus	The ankle is in equinus. The knee extends fully or goes into mild recurvatum. The hip extends fully and the pelvis is within the normal range or tilted anteriorly.
Group II, Jump Gait	The ankle is in equinus, particularly in late stance. The knee and hip are excessively flexed in early stance and then extend to a variable degree in late stance, but never reach full extension. The pelvis is either within the normal range or tilted anteriorly.

Group III, Apparent Equinus	The ankle has a normal range but the knee and hip are excessively flexed throughout stance. The pelvis is normal or tilted anteriorly.
Group IV, Crouch Gait	The ankle is excessively dorsiflexed throughout stance and the knee and hip are excessively flexed. The pelvis is in the normal range or tilted posteriorly.
Group V, Asymmetric Gait	The gait pattern is asymmetrical to the degree that the subject's two lower limbs are classified as belonging to different groups; e.g. right lower limb group III, apparent equinus and left lower limb group II, jump gait.

Table 1: Classification of Sagittal Gait Patterns in Spastic Diplegia

Winters et al. (1987) classification, later revised by Rodda and Graham, (2001)[15] describes four different sagittal gait patterns for spastic hemiplegia:

Type I Drop Foot	There is an accentuated drop foot in the swing phase of gait. No contractures are present and there is free dorsiflexion range of motion.
Type IIa True Equinus	There is a plantar-flexed foot in the swing phase of gait as well as restricted dorsiflexion in stance. No knee or hip involvement.
Type IIb True Equinus with Recurvatum Knee	Similar to Type 2a, but presents with a hyperextended knee during stance.
Type III True Equinus w/ Jump Knee	This type of gait is characterized by gastrocnemius-soleus spasticity or contracture, resulting in impaired ankle dorsiflexion in the swing phase and a flexed, stiff knee gait as the result of hamstring/quadriceps co-contraction.
Type IV True Equinus w/ Jump Knee and Internal Rotation	It encompasses the characteristics described in the previous types, as well as hip involvement with limited hip extension in terminal stance and increasing anterior pelvic tilt and internal hip rotations in stance.

Table 2: Classification of Sagittal Gait Patterns in Spastic Hemiplegia

Still the currently available classification systems do not encompass all patients, as CP is highly heterogeneous and asymmetric. Further analysis, investigating separate joint motion patterns and functional impairment in patients is needed[17].

2.4. Pedobarography

Gait analysis has been historically associated with children with CP. In its advent, it was associated with the need to assess pre and post chirurgical results of complex orthopaedic surgery, and slowly expanded to other types of surgery and medical and rehabilitation interventions, including AFO[13].

Laboratory gait analysis tools, such as optoelectronic cameras, reflective markers, motion sensors, pressure mats and electromyography sensors have provided very useful information about biomechanical alignment and function of the lower limb of ambulant children with CP, and has allowed the body of available evidence to be ever growing in the last few years[12,13].

Although instrumented clinical gait analysis has been a great tool for planning intervention and assessing outcomes in the rehabilitation process of CP children[1,18], very few studies include foot pressures assessment.

Foot deformity is a prevalent problem in people with CP, but still very few studies illustrate the foot-ground interaction, namely with parameters like contact area and time, plantar pressure distribution or progression during the stance phase of gait. Even fewer studies reported the effects on plantar pressure distribution when introducing an AFO.

Under this aspect, dynamic pedobarography is a relatively simple and non-invasive technology that measures the change in plantar pressure distribution throughout the stance phase of gait[19,20]. It is a reliable tool[20,21] and has been widely used to obtain data from both healthy adult and children. In clinical settings it is mostly used to monitor and assess patients with foot pathology over time and less often monitor the effects of prescribed orthoses[22].

In the past years, several studies have tried to produce normative age-dependent databases[19,21–23], fundamental in order to assess and compare pathologic populations.

In fact, more evidence is now surfacing about the foot characteristics of typically developed children. Firstly, foot pressures change dramatically throughout the life cycle, especially in the early years (up to 6 years old). The evidence shows that while younger

children present with a flatfoot pattern, older children tend to develop a more curvilinear pattern[22]. Also older children show higher values in the main plantar pressure variables, compared to younger children[23].

One of the challenges of standardizing this tool, is that there are multiple footprint segmentation models[19]. There is still no consensus about which foot model may provide the most detailed information, without losing the functional aspects of the foot[24]. Most authors propose an anatomical division, corresponding to the foot joint positions, that ranges from 5 to 8 subdivisions of the footprint (among the most often used are heel, midfoot (medial and lateral), forefoot (medial and lateral), toes (2^{nd}–5^{th} and 1^{st} toe)[19,22,24–26].

Another challenge of systematically using this tool and provide comparisons between groups is the selection of the different pedobarography variables. From the available studies[19,22,23,27] we can identify variables like, contact time, contact area, average and maximum force, average and peak pressure, force-time integral, pressure-time integral, Center of Pressure (CoP) progression and the Arch Index.

Contact area is defined as the area covered by foot during one step and contact time is defined as the time interval between initial ground contact and toe off. Normal foot tends to have larger contact area covered and shorter contact time, which in addition leads to less rigid and more stable foot to absorb impact.

Maximal and average force, peak and average plantar pressures represent the maximal and average load in an area under the foot during one step. These forces may represent up to 120% of body weight, and are definite contributors for the formation of the foot[23].

Additionally, the influence of high pressures for a short duration of time versus lower pressures for longer duration is also important to consider. On that note, force-time integral and pressure-time integral are variables which describe the cumulative effect of force and pressure over time in a certain area of the foot, additionally providing a value for the total load exposure of a foot sole area during one step. Previous studies have shown that cumulative effect of force and pressure can lead to tissue damage and increase the risk of skin trauma[23].

The CoP progression provides a picture of foot pressure distribution throughout the stance phase and has been used to predict regions of the foot that are at risk for overload. The Arch Index (AI), is defined as the ratio between the midfoot area and the overall contact area and it provides useful information on the position of the foot, classifying it as flatfeet/ planus feet (lower or missing arch), normal feet (with normal arch) and cavus feet (with high arch)[27].

Still from the few available studies concerning the CP population, data shows that overall there is an increased pressure towards the forefoot and toes[27,28].

Femery et al. (2002)[29] reports that children with CP have significant differences in plantar pressure loads in both feet, whether on the most affected limb or on the unaffected one. Also, as opposed to the typically developing foot, that tends to have larger contact area and shorter contact time[22,23], children with CP tend to have a smaller foot area distribution and increased contact time, due to the asymmetry of gait and the difficulties in postural control[22,23].

2.5. The Use of Ankle-foot Orthoses in Children with Cerebral Palsy

CP is a clinical condition responsible for significant functional deficits and has a huge impact in daily living of those who live with it. It is very representative of the practice of a Paediatric Physiotherapist.

Optimizing the gait pattern of children with CP is a primordial goal in rehabilitation, as a higher functional mobility has been associated with a higher social participation[10]. There are countless interventions that aim to improve selective motor control and muscle coordination, strength and endurance, biomechanical alignment and overall gait efficiency. Medical and surgical interventions include application of botulinum toxin, phenol, muscle and tendon stretching and dorsal rizothomy. Orthoses are also commonly used and one of the most prescribed are the AFO.

AFO main objective is to improve the gait pattern by controlling and positioning the ankle and foot, during the different phases of gait. AFO increase stability of the lower limb, amending for muscle weakness and biomechanical misalignment, moderating the

deforming forces common to CP. They can work either by restricting excessive ankle plantarflexion, improving valgus/varus of the foot, and sometimes aiming to influence the positioning of the knee, by allowing a better knee extension during stance[14,30–32].

AFO are likely more effective at preventing, rather than alleviating, contractures and deformity in the foot and ankle, although the existing literature is not yet conclusive. One of the challenges of providing strong evidence on AFO is its prescription. One of the existing difficulties relates to the "dosing" of orthoses (duration of wear and design) in ambulatory CP[33]. They may be used for smaller or longer periods of time, and often enough users report maladaptation. Still there is a valid concern about the restriction of movement and its relation to muscle waste and limiting the development of typical movement patterns[33].

These orthoses may come in a multitude of materials and configurations, and may be pre-made or customised according to the identified issues. 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[34]. 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)[35–37].

AFO have demonstrated positive effects in multiple parameters, like gait speed, step length, knee and ankle joint range or energy expenditure[14,30].

Even though AFO is a frequently-prescribed intervention for children with CP, rigorous evidence of their efficacy is limited[38], mainly because of the heterogeneity of outcome measures among researchers, which limits comparison between studies[39]. 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[40,41].

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Chapter III – Methodological Considerations

3.1. Scoping Review

Evidence-based healthcare is the gold standard and in the CP research, an expanding field. With the continual increase of primary research, the conduct of reviews has also increased and evolved. Different objectives and questions have led to the development of new approaches that are designed to more effectively and rigorously synthesize the evidence. Aiming to review the existing body of literature about the use of AFO in children with CP, a scoping review protocol was constructed based on PRISMA guidelines[1] and registered in PROSPERO (International prospective register of systematic reviews).

A registered scoping review protocol is important, as it pre-defines the objectives, methods, and reporting of the review and allows for transparency of the process, and preventing anyone else to do the exact same review.

According to Aromataris and Pearson (2014)[2], the main characteristics of a well-conducted review are:

- Clearly articulated objectives and questions to be addressed;

- Inclusion and exclusion criteria, stipulated a priori (in the protocol), that determine the eligibility of studies;

- A comprehensive search to identify all relevant studies, both published and unpublished;

- Appraisal of the quality of included studies, assessment of the validity of their results, and reporting of any exclusions based on quality;

- Analysis of data extracted from the included research;

- Presentation and synthesis of the findings extracted;

- Transparent reporting of the methodology and methods used to conduct the review;

Bearing these recommendations in mind, a preliminary search was performed to select keywords related to the population, intervention and outcomes using the PICO framework[3]. The keywords selected from the MeSH database in MEDLINE. 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. 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).

The article selection was conducted by two independent reviewers and a third external reviewer would resolve any disagreements. The resulting articles were assessed by the PEDro Risk of Bias Tool[4,5], for a minimum score of ≥5 points, which usually represents an adequate methodological quality study[6].

The main findings of the scoping review, alongside with any clinical practise guidelines and implications for future studies were clearly discussed and stated as a conclusion at the end of the paper.

3.2. Test-retest Reliability Analysis

Reliability analysis means to assert that an assessment tool produces stable and consistent results overtime. Test-retest reliability is a method of measuring of reliability obtained by administering the same test twice over a period of time to the same group of individuals.

The correlation between the different assessments may be expressed by the Intraclass Correlation Coefficient (ICC). The ICC is an index that reflects both correlation and agreement between measurements[7].

For the second research paper ICC considering the two-way mixed model with absolute agreement and accounting for the mean of multiple measurements[7] were calculated for all variables and masks, and a critical level of $p < 0.05$ was considered significant. The ICC statistical analysis was performed using the following formula:

$$ICC = \frac{MS_R - MS_E}{MS_R + \frac{MS_C - MS_E}{n}}$$

where MS_R represents the mean square between lower limbs; MS_E represents the mean square for error; MS_C represents the mean square within lower limbs, concerning the selected pedobarograph variables; and n is the total number of lower limbs assessed (two lower limbs for each of the eight participants).The level of agreement was considered poor, fair, good, and excellent when $ICC < 0.40$, $0.40 \leq ICC < 0.60$, $0.60 \leq ICC < 0.75$, and $0.75 \leq ICC \leq 1.00$, respectively [29]. Calculations also included the mean difference between measurements ($Mean_{diff}$), the 95% CI for the $Mean_{diff}$, the standard deviation of the differences (SD_{diff}), and the 95% Bland and Altman limits of agreement (95% LOA).

The absolute measure of reliability standard error of measurement (SEM) was

$$SEM = \frac{SD_{diff}}{\sqrt{2}}$$

calculated using the following equation:

where SD_{diff} represents the standard deviation of the difference.

For clinical purposes, it is very important to have a limit to determine when significant changes occur and the outcomes are meaningful[9]. So, to determine the smallest amount of change that must be achieved to reflect a true change, outside the error of the tests, the minimal detectable change (MDC) was calculated using the following equation:

$$MDC = 1.96 \cdot \sqrt{2} \cdot SEM$$

3.3. Plantar Pressure Insoles for Pedobarography Analysis

Plantar pressure collection devices are a fairly recent instrument that has gained popularity in scientific studies with typical developed population and with individuals with pathology alike. Both in-shoe (insoles) systems and pressure-platform/mat systems have been used, and typically measure pressure, vertical force, and foot contact area during the stance phase of the gait cycle[10].

This technology consists of capacitive sensors with electrical properties, which are sensible to pressure changes, creating a signal proportional to the pressure exerted. Similar to other systems, it also uses a method of normalization and calibration[10].

Data collection and processing was similar in both studies in included in this dissertation. The participants wore the foot insoles Pedar-X system® (Novel, Munich, Germany), inside their usual footwear and/or orthotic device (adequate to their feet size) and no socks, wearing the same combination of footwear and/or orthoses for all required trials. The batteries and the wireless transmitter were strapped or placed inside a backpack on the participant's back. A schematic picture and a photograph illustrate the experimental setup used (Figure 1). The insoles were calibrated using the Pedar X Standard (v 25.3.6, Novel, Munich, Germany) protocol (before the beginning of each trial, the participant was asked to lift one foot at a time off the ground for approximately 15 s). Data were sampled at 100 Hz. The participants were instructed to walk back and forth, along a 5 m line drawn on a smooth and regular floor, unassisted and at a self-selected speed, without running. A chair was placed at either end of the walkway, in case the participants needed to stop. Data collection stopped after 2 min if the children achieved a minimum of 15 steps with each lower limb.

Figure 1: Experimental Set Up

Data were extracted and processed using the Novel Multiprojects-e (v 24.3.34, Novel, Munich, Germany), which enabled the creation of a database and processing of each participant's individual footprint. Each data set was reviewed and amiss footprints and directional changes were wiped out of the original records. The average of the selected variables (force–time integral, pressure–time integral, maximum force, peak pressure, contact area, and contact time) was automatically calculated by the software for the whole foot. A mask then divided the foot into three regions (hindfoot, middle foot, and forefoot), according to the length of the foot (0 to 30%, 30 to 60%, and 60 to 100% of total length, respectively), as shown in Figure 2. These masks were applied automatically by the software, and average scores were calculated for each variable and zone. The software also produced 3D plantar pressure maps for each participant, allowing a visual comparison of the first and second trial (Figure 3).

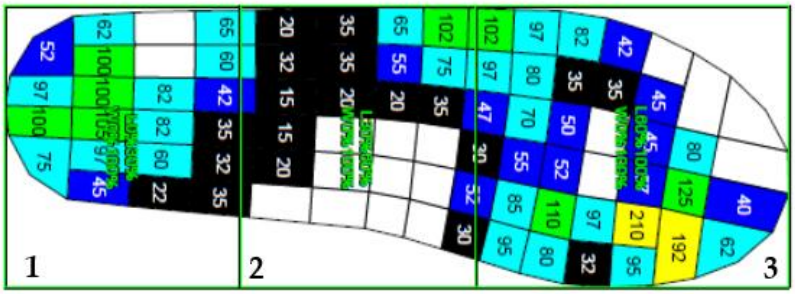

Figure 2: Three zones of segmentation of the foot (1 – 0% to 30% of total length; 2 – 30% to 60 % of total length; 3 – 60% to 100% of total length). Obtained from Novel Multiprojects-e (v 24.3.34, Novel, Munich, Germany).

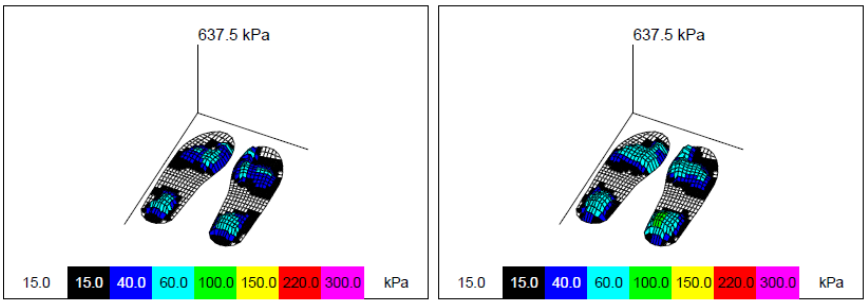

Figure 3: Three-dimensional plantar pressure mapping for test–retest results of participant 008. Obtained from Novel Multiprojects-e (v 24.3.34, Novel, Munich, Germany).

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Chapter IV – Effects of Ankle Foot Orthoses on the Gait Patterns of Children with Spastic Bilateral Cerebral Palsy: a Scoping Review.

Current Status: published in *Children* (Appendix I)

Ricardo D, **Raposo MR**, Cruz EB, Oliveira R, Carnide F, Veloso AP, João F. Effects of Ankle Foot Orthoses on the Gait Patterns in Children with Spastic Bilateral Cerebral Palsy: A Scoping Review. Children (Basel). 2021 Oct 10; 8(10):903. doi: 10.3390/children8100903. PMID: 34682168; PMCID: PMC8534539.

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 (AFO) 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 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 to 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 5 RCT. 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 report 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

4.1. 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 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 ankle foot orthoses (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].

4.2. Materials and Methods

4.2.1. 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. 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 keywords 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).

4.2.2. Selection criteria

4.2.2.1. 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).

4.2.2.2. Study Selection

The article selection was conducted by two independent reviewers (D.R. and M.R.R.), after duplicates 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.

4.2.3. 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].

4.2.4. 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.

4.3. Results

4.3.1. 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 4).

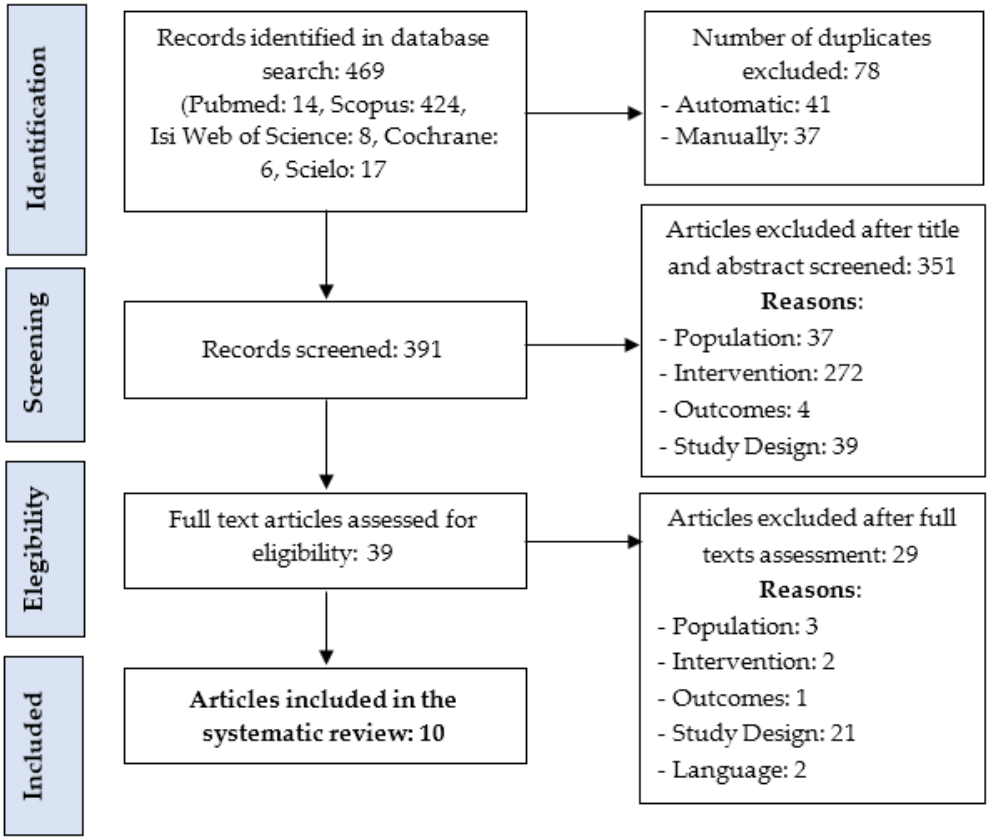

Figure 4: Flowchart of the article's selection process

4.3.2. 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: optoelectronic 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 1.

Table 3: 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[72]	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[73]	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[74]	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[75]	2012	Non-randomised 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 orthopaedic 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 an 8-meter 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 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[35]	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	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.	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

				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.			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.			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).
Lam, 2005[76]	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[78]	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	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.

				assistive device in ambulation; No orthopaedic surgery in the previous year.						No differences were found for the gait variables when comparing the two orthoses ($P < 0.02$).
Radtka, 2005[77]	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, 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.	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 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[79]	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 ($P \leq 0.05$). 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 de 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≤0.05). No significant differences in ankle powers were found between DAFO and HAFO.
Zhao, 2013[71]	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 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 in 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 2 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 4: 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[72]	Yes	Yes	No	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	8/10
Bjornson, 2016[73]	Yes	Yes	No	Yes	No	No	No	Yes	Yes	Yes	No	5/10
Buckon, 2004[74]	Yes	Yes	No	Yes	No	No	No	Yes	Yes	Yes	Yes	6/10
Degelean, 2012[75]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
El-Kafy, 2014[35]	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	Yes	8/10
Lam, 2005[76]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
Radtka, 1997[78]	Yes	No	No	Yes	No	No	No	Yes	Yes	Yes	Yes	5/10
Radtka, 2005[77]	Yes	No	No	Yes	No	No	No	Yes	Yes	Yes	Yes	5/10
Smith, 2009[79]	Yes	No	No	No	No	No	No	Yes	Yes	Yes	Yes	4/10
Zhao, 2013[71]	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.

4.3.2.1. 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 pattern.

4.3.2.2. Types of AFO

The majority of interventions were centred 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].

4.3.2.3. 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.

4.3.2.3.1. 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≤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.

4.3.2.3.2. 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].

4.3.2.3.3. 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 analysed 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].

4.3.2.3.4. 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].

4.4. 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. There is a wide variety of variables and outcomes within this area of rehabilitation studies which

makes difficult the comparison between studies and consequently to access the effectiveness between AFO.

4.4.1. 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 characterised 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].

4.5. Conclusions

In this review, we found that AFO use seem 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 a creation 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, report 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.

4.6. References

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Chapter V – Gait Analysis in Children with Cerebral Palsy: Are Plantar Pressure Insoles a Reliable Tool?

Current Status: published in *Sensors* (Appendix II)

Raposo MR, Ricardo D, Teles J, Veloso AP, João F. Gait Analysis in Children with Cerebral Palsy: Are Plantar Pressure Insoles a Reliable Tool? Sensors (Basel). 2022 Jul 13; 22(14):5234. doi: 10.3390/s22145234. PMID: 35890913; PMCID: PMC9319716.

Abstract**:** Cerebral palsy (CP) is a common cause of motor disability, and pedobarography is a useful, non-invasive, portable, and accessible tool; is easy to use in a clinical setting; and can provide plenty of information about foot–soil interaction and gait deviations. The reliability of this method in children with CP is lacking. The aim of this study is to investigate test–retest reliability and minimal detectable change (MDC) of plantar pressure insole variables in children with CP. Eight children performed two trials 8 ± 2.5 days apart, using foot insoles to collect plantar pressure data. Whole and segmented foot measurements were analyzed using intraclass correlation coefficients (ICC). The variability of the data was measured by calculating the standard error of measurement (SEM) and the MDC/ICC values demonstrated high test–retest reliability for most variables, ranging from good to excellent (ICC ≥ 0.60). The SEM and the MDC values were considered low for the different variables. The variability observed between sessions may be attributed to the heterogeneous sub-diagnosis of CP.

Keywords: plantar pressure; cerebral palsy; gait analysis; reliability; insoles

5.1. Introduction

Cerebral palsy (CP) is the most common cause of motor disability in children[1–3]. CP is a complex pathology that describes a group of impairments and motor disorders, which are permanent but not immutable, resulting from a nonprogressive cerebral disorder[4] with different presentations and functional levels[5].

CP presents both positive features such as spasticity, hyper-reflexia, and co-contraction, and negative features including weakness, difficulties in motor control, and sensory and balance impairments[6]. The lack of control is obvious at the lower limb joints, especially the ankle joint. These alterations are the main cause of limb contractures, musculoskeletal deformity, and gait deviations[7].

Foot deformities, along with hip displacement, are the most common musculoskeletal occurrences in CP. Among the most common foot deformities in this population are equinus, planovalgus, and equinovarus, which can vary from very mild and flexible to severe and rigid[8]. These deformities, which cause the foot to abnormally lay on the ground, can significantly impair function and quality of life; however, very few

studies have systematically investigated the foot morphology and the ground–foot interaction during the stance phase in this population[7].

Instrumented clinical gait analysis has been an excellent tool for planning intervention and assessing outcomes in the rehabilitation process of children with CP[1,2]. Though the gold standard for gait analysis in children with CP would be a quantitative three-dimensional analysis of movement and respective articular moments and power (kinematics and kinetics), possibly alongside muscle activation (electromyography) and oxygen consumption[9], it is not always possible to conduct such an assessment in a clinical setting. More accessible and portable methods have been recently used such as inertial sensors[10,11] and plantar pressure recording devices[7,12–15].

Under this aspect, dynamic pedobarography is a relatively simple, portable, and non-invasive technology that measures the change in plantar pressure distribution throughout the stance phase of gait[16]. It is an easy method to use in a clinical setting; can provide plenty of information about foot–soil interaction; and, alongside other gait analysis methods, can help assess the impact of a medical intervention, a rehabilitation program, or the effects of an orthotic device. Several studies tested its reliability[16,17] for both healthy adult and children, but none have assessed subjects with CP. The few existing clinical studies in participants with CP use mainly plantar pressure mats/platforms instead of insoles[7,12–15].

In the past years, several studies have tried to produce normative age-dependent gait databases[18–20], which are fundamental to assess and compare with pathologic situations. In fact, more evidence is now surfacing about the foot characteristics of typically developed children. Foot pressure changes dramatically throughout the life cycle, especially in the early years (up to 6 years old). The evidence shows that, while younger typically developing children present with a flatfoot pattern, older children tend to develop a more curvilinear pattern[18]. Moreover, older children show greater values in the main plantar pressure variables when compared with younger children[20].

Even fewer studies have included plantar pressure measurements in children with CP. There has been no attempt to create any kind of database, which is fundamental to assess and compare the natural progression of the condition and the results of medical and therapeutic interventions. Nevertheless, data collected across the

existing studies show that there is a variability in foot pressure distribution depending on spasticity overall, there is an increase in pressure towards the toes and forefoot as well as a significant reduction towards the heel[7,14,21].

Reports of plantar pressure data in the literature are highly heterogeneous. One of the challenges of standardizing this tool is that there are multiple footprint segmentation models[19]. There is still no consensus about which foot model may provide the most detailed information, without losing the functional aspects of the foot[15]. Most authors propose an anatomical/functional segmentation, corresponding to the foot joint positions, which ranges from as few as 3 to as many as 12 subdivisions of the footprint (the most often used are the hind-foot, mid-foot (medial and lateral), forefoot (medial and lateral), and toes (toes 2–5 and the first toe)[7,13–15,17–19,21–24].

The absence of systematized evidence regarding the reliability of foot pressure insoles on this specific population and the need to assess the dimension of error measurement with this tool calls for further investigation. In so, the aim of this study is to investigate test–retest reliability and minimal detectable change of plantar pressure insoles in a sample of children with CP when walking in regular footwear.

5.2. Materials and Methods

5.2.1. Design

Prospective intra-rater test–retest reliability and minimal detectable change study.

5.2.2. Participants Selection

A convenience sample of 10 children with cerebral palsy was selected from a Portuguese rehabilitation center to participate in this study. The selected participants followed the eligibility criteria: male or female children between 4 and 12 years of age, foot length ranging from 15 to 20 cm (because of equipment constraints), with a clinical diagnosis of bilateral (lower limb predominance) or unilateral cerebral palsy, grades I

and II on the Gross Motor Function Classification System (GMFCS)[25], able to walk independently for 5 m without walking aids, and able to comprehend and comply with simple instructions. Children should also have not been subjected to orthopedic surgery or botulinum toxin treatment in the previous 6 months. The protocol was approved and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019). All procedures were previously explained to both the child and the legal guardian, an informed consent form was filled and signed by the legal guardian, and verbal consent was given by the child.

5.2.3. Data Collection Protocol

Data collection was performed on two different days within a period of 7 to 14 days (8 ± 2.5 days) to minimize the assessor memory bias and to prevent a change in the children's gait pattern or clinical condition. Clinical history and a brief physical exam (mass, height, lower limb posture, selective motor control tests, gastrocnemius length, and spasticity)[9] were conducted in the first session.

Children wore the foot insoles Pedar-X system® (Novel, Munich, Germany), inside their usual footwear (adequate to their feet size) and no socks. The children wore the same pair of shoes for both trials. The batteries and the wireless transmitter were strapped or placed inside a backpack on the child's back. A schematic picture and a photograph illustrate the experimental setup used (Figure 1). The insoles were calibrated using the Pedar X Standard (v 25.3.6, Novel, Munich, Germany) protocol (before the beginning of each trial, the participant was asked to lift one foot at a time off the ground for approximately 15 s). Data were sampled at 100 Hz. Children were instructed to walk back and forth, along a 5 m line drawn on a smooth and regular floor, unassisted and at a self-selected speed, without running. A chair was placed at either end of the walkway, in case the participants needed to stop. Data collection stopped after 2 min if the children achieved a minimum of 15 steps with each lower limb.

5.2.4. Data Processing

Data were extracted and processed using the Novel Multiprojects-e (v 24.3.34, Novel, Munich, Germany), which enabled the creation of a database and processing of each participant's individual footprint. Each data set was reviewed and amiss footprints and directional changes were wiped out of the original records. The average of the selected variables (force–time integral, pressure–time integral, maximum force, peak pressure, contact area, and contact time) was automatically calculated by the software for the whole foot. A mask then divided the foot into three regions (hindfoot, middle foot, and forefoot), according to the length of the foot (0 to 30%, 30 to 60%, and 60 to 100% of total length, respectively), as shown in Figure 2. These masks were applied automatically by the software, and average scores were calculated for each variable and zone. The software also produced 3D plantar pressure maps for each participant, allowing a visual comparison of the first and second trial (Figure 3).

5.2.5. Statistical Analysis

Statistical analysis to assess the test–retest reliability of plantar pressure data was carried out using the methodology described by Koo and Li (2015)[26], similar to the methods used by Fernandes et al. (2015)[27] and Ricardo et al. (2021)[28] in their works.

Intraclass correlation coefficients (ICCs) considering the two-way mixed model with absolute agreement and accounting for the mean of multiple measurements were calculated for all variables and masks, and a critical level of $p < 0.05$ was considered significant. The *ICC* statistical analysis was performed using SPSS (version 28.0.0; IBM, Chicago, IL, USA), using the following formula:

$$ICC = \frac{MS_R - MS_E}{MS_R + \frac{MS_C - MS_E}{n}}$$

where MS_R represents the mean square between lower limbs; MS_E represents the mean square for error; MS_C represents the mean square within lower limbs, concerning the selected pedobariografic variables; and n is the total number of lower limbs assessed (two lower limbs for each of the eight participants).The level of agreement was considered poor, fair, good, and excellent when $ICC < 0.40$, $0.40 \leq ICC < 0.60$, $0.60 \leq ICC < 0.75$, and $0.75 \leq ICC \leq 1.00$, respectively[29]. Calculations also included

the mean difference between measurements ($Mean_{diff}$), the 95% CI for the $Mean_{diff}$, the standard deviation of the differences (SD_{diff}), and the 95% Bland and Altman limits of agreement (95% LOA).

The absolute measure of reliability standard error of measurement (*SEM*) was calculated using the following equation:

$$SEM = \frac{SD_{diff}}{\sqrt{2}}$$

where SD_{diff} represents the standard deviation of the difference.

To determine the smallest amount of change that must be achieved to reflect a true change, outside the error of the tests, the minimal detectable change (*MDC*) was calculated using the following equation:

$$MDC = 1.96 \cdot \sqrt{2} \cdot SEM$$

The *SEM* and *MDC* were calculated using Microsoft Excel 2013 (Microsoft Corporation, Redmond, WA, USA).

5.3. Results

The participants of the study were a convenience sampling of ten children with CP (nine spastic unilateral, one spastic bilateral; four females, six males; age 57.9 ± 13.4 months; height 110.4 ± 7.6 cm; mass 18.1 ± 2.4 kg) (Table 1), two of which dropped out of the study as they could not complete the trials in the same time frame as the other participants (one because of Covid-19 prophylactic quarantine and the other because of loss of contact). Data from each limb were processed separately (N = 16), because of the heterogeneous physical presentation of unilateral CP that composed most of the selected sample. On average, we assessed 75.8 ± 27.9 steps on each trial.

Table 5: Participants' characteristics.

Participant	Gender	Age (Months)	Diagnosis	Affected Side	GMFCS Level[107]	Interval between Trials (Days)	Mass (kg)	Height (cm)	Sagittal Gait Pattern[16,110]		Gastrocnemius Spasticity (Modified Ashworth Scale)[111]		Foot Length (cm)		Number of Steps (Average from Both Trials)		Status
									Right	Left	Right	Left	Right	Left	Right	Left	
001	Male	54	Unilateral CP	Right	I	14	16.5	105	Drop Foot	-	1	0	15	16	70	70	Completed trials
002	Male	65	Unilateral CP	Left	II	9	20	118	-	True Equinus	0	4	19	17	52	59	Completed trials
003	Female	41	Unilateral CP	Right	II	7	19	105	True Equinus	-	1+	0	16	17	55	52	Completed trials
004	Female	56	Bilateral CP	Both	II	7	18	110	Apparent Equinus	Apparent Equinus	1	1	17	17	55	55	Completed trials
005	Female	65	Unilateral CP	Right	I	7	20.4	120	True Equinus	-	1	0	20	19	64	65	Completed trials
006	Male	45	Unilateral CP	Left	I	-	13	97	-	True Equinus	0	1	15	15	-		Dropped out
007	Male	41	Unilateral CP	Right	I	-	16	103	True Equinus	-	1	0	15	16	-		Dropped out
008	Male	74	Unilateral CP	Right	I	7	20.5	115	True Equinus	-	1	0	20	20	75	74	Completed trials
009	Male	80	Unilateral CP	Right	I	6	20.1	115	True Equinus	-	1+	0	19	19	122	120	Completed trials
010	Female	58	Unilateral CP	Right	I	7	17	116	Equinus/Jump Knee	-	2	0	16	18	112	119	Completed trials

5.3.1. Reliability of Whole Foot Measurements

As shown in Table 6, all selected variables calculated for the whole footprint showed an excellent ICC (ICC ≥ 0.75), except for the contact time variable (ICC = 0.36, 95% CI 0 to 0.784). The SEM and MDC values were within an acceptable range for each of the variables.

Table 6: Reliability values for pedobarography measurements (whole foot).

Pedobarography Measurements	ICC	ICC 95% CI	Mean	Mean Diff	SD Diff	95% LOA	SEM	MDC
Force–time integral (N·)	0.76	(0.30; 0.92)	73.72	−2.08	18.57	(−38.47; 34.31)	13.13	36.39
Pressure–time integral (kPa·s)	0.89	(0.70; 0.96)	55.40	0.63	10.04	(−19.05; 20.31)	7.10	19.68
Maximum force (N)	0.79	(0.42; 0.93)	161.30	−7.61	25.00	(−56.61; 41.40)	17.68	49.00
Peak pressure (kPa)	0.81	(0.47; 0.93)	136.45	6.84	27.48	(−47.01; 60.70)	19.43	53.85
Contact area (cm^2)	0.83	(0.53; 0.94)	56.80	−3.69	8.15	(−19.66; 12.27)	5.76	15.97
Contact time (ms)	0.37	(0; 0.78)	669.93	4.29	137.30	(−264.81; 273.40)	97.08	269.11

5.3.2. Reliability of Segmented Foot Measurements

Overall ICC values for the segmented foot measurements fit in the good to excellent range (ICC values ≥ 0.60), except for peak pressure (ICC = 0.439, 95% CI 0 to 0.807) and contact time (ICC = 0.552, 95% CI 0 to 0.845) at the forefoot (Table 7). The SEM and MDC values were within an acceptable range for each of the variables.

Table 7: Reliability values for pedobarography measurements (three zones of the segmented foot).

	Pedobarograpy Measurements	ICC	ICC 95% CI	Mean	Mean Diff	SD Diff	95% LOA	SEM	MDC
Hindfoot	Force–time integral (N·s)	0.83	(0.51; 0.94)	17.44	−1.43	11.35	(−23.67; 20.82)	8.02	22.24
	Pressure–time integral (kPa·s)	0.97	(0.92; 0.99)	21.41	0.62	12.01	(−22.93; 24.16)	8.49	23.54
	Maximum force (N)	0.92	(0.77; 0.97)	70.50	−6.38	28.65	(−62.53; 49.77)	20.26	56.15
	Peak pressure (kPa)	0.88	(0.65; 0.96)	78.56	−3.84	18.48	(−40.06; 32.37)	13.07	36.22
	Contact area (cm^2)	0.91	(0.75; 0.97)	13.68	−1.76	6.19	(−13.89; 10.36)	4.38	12.13
	Contact time (ms)	0.86	(0.62; 0.95)	365.79	38.16	272.29	(−495.53; 571.85)	192.54	533.69
Middle Foot	Force–time integral (N·s)	0.91	(0.75; 0.97)	15.32	0.52	3.14	(−5.63; 6.67)	2.22	6.15
	Pressure–time integral (kPa·s)	0.97	(0.92; 0.99)	30.19	0.91	5.95	(−10.75; 12.57)	4.21	11.66
	Maximum force (N)	0.91	(0.74; 0.97)	47.92	−2.32	7.84	(−17.69; 13.05)	5.54	15.37
	Peak pressure (kPa)	0.97	(0.92; 0.99)	74.89	1.19	8.31	(−15.09; 17.47)	5.87	16.28
	Contact area (cm^2)	0.98	(0.94; 0.99)	16.54	−0.34	2.07	(−4.39; 3.72)	1.46	4.06
	Contact time (ms)	0.73	(0.25; 0.90)	621.32	9.79	118.82	(−223.09; 242.67)	84.02	232.88
Forefoot	Force–time integral (N·s)	0.73	(0.25; 0.90)	40.95	−1.18	11.14	(−23.02; 20.66)	7.88	21.84
	Pressure–time integral (kPa·s)	0.97	(0.92; 0.99)	42.35	1.53	7.30	(−12.77; 15.83)	5.16	14.30
	Maximum force (N)	0.73	(0.26; 0.90)	123.44	−5.93	23.40	(−51.80; 39.95)	16.55	45.87
	Peak pressure (kPa)	0.44	(0; 0.81)	124.59	8.68	28.00	(−46.19; 63.55)	19.80	54.87
	Contact area (cm^2)	0.68	(0.07; 0.89)	25.59	−3.57	7.21	(−17.70; 10.55)	5.10	14.12
	Contact time (ms)	0.55	(0; 0.85)	578.39	22.66	194.57	(−358.70; 404.02)	137.58	381.36

5.4. Discussion

The main objective of the current study was to assess the intersession and intra-rater reliability of plantar pressure variables when using pressure foot insoles and, to the best of the authors' knowledge, it is the first study to do so. Plantar-pressure-related data for children with CP are still scarce in published evidence. Alongside other gait analysis tools, pedobarographic measurements are useful in assessing pre- and post-surgical outcomes, treatment with botulinum toxin, and orthotic management, as they provide important information about foot pressure

distribution, postural control, center of pressure (COP) displacement, and the foot–soil interaction. Nonetheless, if this type of data is to be used for assessing clinical or therapeutic interventions, it is of high importance to establish reliability levels for this specific method and population[22].

The reliability of foot pressure platforms or mats for typically developing children and healthy adults has been previously established by Cousins et al. (2012)[33], Hafer et al. (2013)[16], and Niller et al. (2016)[17]. Other similar studies assessed likewise reliability for both typically developing children and children with CP, also using a plantar pressure mat[14,34]. However, the use of plantar pressure foot insoles presents with different benefits, such as the possibility of their use inside shoes or orthotic devices recording a higher number of gait cycles, as well as overall being easier to use with smaller children.

Our results show high reliability (ICC ≥ 0.60) for 21 of the 24 parameters that were tested. Still, three of the outcome measures for whole foot and forefoot showed lower values (whole foot contact time variable and peak pressure and contact time variables at the forefoot).

The number of participants included in this study was small, but similar to other researches[14,23]. However, because of the heterogeneity of children with CP, we opted to conduct a separate analysis of right and left feet. This increases the total sample to sixteen (feet). Post-hoc power analysis with $\alpha = 0.05$ revealed good power (≥0.90) for most variables, except for the three variables mentioned above. Post-hoc statistical analysis was carried out using R software (version 4.1.3., R Core Team 2022)[35] and the "ICC.Sample.Size" package (version 1.0.)[36].

The poor reliability results for the contact time variable (whole foot and forefoot region) may be explained by the heterogeneous gait pattern with which the participants presented. Most of our sample were children with unilateral CP, who present with a slower pace and abnormal weight shift between the affect side and less affected size. As a separate limb analysis was conducted, the diminished weight shift to the more affected side may have led to an increased contact time on the opposite side, and thus the contact time variable registered a wider range of values. Moreover, although we asked the children to walk at a self-selected comfortable pace, their pace varied.

The lower ICC values obtained from the forefoot peak pressure can be attributed to the slight discrepancy between the total foot length and the length of the available insole. Foot length across our sample ranges from 15 cm to 20 cm, but the same pair of 20 cm insoles was used throughout the investigation. This means that the fit was not always perfect, leaving vacant pressure cells at the top of the insoles, which can reflect in the forefoot values. Moreover, the total weight of the equipment was 0.5 kg, which may impact the trials of some of the smaller children and those with greater locomotion difficulties and gait deviations.

The *SEM* and *MDC* values were determined to quantify the amount of error associated with each variable in this population. Even though the *SEM* and *MDC* values for each variable showed a clinically acceptable level of error[20], they were transformed into a percentage for comparison purposes:

$$SEM\% = \frac{SEM}{Mean} \cdot 100$$

And

$$MDC\% = \frac{MDC}{Mean} \cdot 100$$

Please refer to Ayán-pérez, C. and Bouzas-rico, S. (2019)[37] for more information. For reference purposes, *MDC*% scores >30% were considered poor, from 10 to 30% were considered acceptable, and <10% were considered excellent[38]. The obtained values for *MDC*% were all considered to be poor, except for the contact area variable for the whole foot and peak pressure and contact area for the midfoot, which were within the acceptable range. These results are equivalent to other similar studies[37–39].

Various foot segmentation models have been reported in recent literature[7,13,24,14,15,17–19,21–23]. Complex masking usually involves anatomical and functional segmentation, including external references (for example, retroreflective markers and an optoelectronic system) that were not available for this specific study. Smaller areas of division may provide with less detailed information, and they are also more error-prone[17]. A three identical part division masking was selected for this study, similar to that of Galli et al. [7], allowing to differentiate force, pressure, and spatio-temporal values between the hind-foot, midfoot, and forefoot. Knowing that most

participants presented an equinus gait pattern, we expected altered values in these three areas, and that division allowed the retrieval of more specific data.

The absence of previous reliability studies with this population and method precludes comparisons with similar *SEM* and *MDC* data. These preliminary results could prove useful to determine clinical changes in foot pressure and understand how those changes differentiate from the error of measurement. This is particularly important in studies where we have a pre- and post-assessment of the participant to see the effect of an intervention process. If the post results are superior to the reported error of the measurement, we can be confident in stating that there was a significant effect caused by the intervention.

5.5. Conclusions

This study is the first that establishes plantar pressure insoles as a reliable tool for measuring different gait-related variables in children with CP. The results indicate a good reliability for most variables, except for whole foot contact time and peak pressure and contact time at the forefoot. These lower values observed may be attributed to the heterogeneous gait pattern of children with CP and the above-mentioned equipment limitations of the study.

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Chapter VI – Plantar Pressure Analysis in Children with Cerebral Palsy While Wearing Orthoses – A Descriptive Study

Current Status: submitted in Scientific Reports

Raposo MR, Ricardo D, Veloso AP, João F. (2023). Plantar Pressure Analysis in Children with Cerebral Palsy While Wearing Orthoses – A Descriptive Study. (In revision process)

Abstract**:** *Background:* Cerebral Palsy (CP) is the most common cause of motor disability in children, often leading to different musculoskeletal abnormalities, including foot deformities. Ankle-foot orthoses (AFO) are commonly prescribed to minimize abnormal foot posture and to minimize the impact of spasticity on daily function. Dynamic pedobarography may provide new data to better assess the changes in plantar pressure distribution throughout the stance phase of gait in children with CP. *Methods*: Nine children with CP walked wearing plantar pressure insoles inside their orthoses and regular footwear. Mean values and standard deviation were calculated for each variable in a total and a segmented foot analysis. Clusters based on clinical distribution of spasticity, gait pattern and type of orthoses were created to allow for further analysis. *Results*: Overall data was consistent across all participants and clusters. The use of AFO did not significantly impact any of the mean values for the variables in study, when referencing to the means of the same variables in children with CP walking in regular footwear. The cluster analysis revealed increased pedobarography values in Unilateral CP, Apparent Equinus gait pattern and Dynamic AFO sub-groups. In the segmented foot analysis, all variables increased from heel to the fore foot. *Conclusions*: The use of AFO in children with CP produce positive changes in plantar pressure measurements, approximating them to the reference percentiles of typically developing children.

Keywords: Plantar pressure; Cerebral Palsy; orthoses; gait analysis; insoles;

6.1. Introduction

Cerebral Palsy (CP) is the most common cause of motor disability in children[1–3]. CP is a complex pathology that describes a group of impairments and motor disorders, that are permanent but not immutable resulting from a nonprogressive cerebral disorder[4] with different presentations and functional levels[5]. CP presents both positive features such as spasticity, hyper-reflexia and co-contraction, and negative features including weakness, difficulties in motor control, sensory and balance impairments[6]. The lack of control is obvious at the lower limb joints, especially the ankle joint. These

alterations are the main cause of limb contractures, musculoskeletal deformity and gait deviations[7].

Foot deformities, along with hip displacement, are the most common musculoskeletal occurrences. Amongst the most common foot deformities in this population there are equinus, planovalgus and equinovarus, that can vary from very mild and flexible to severe and rigid[8]. These deformities, that cause the foot to abnormally lay on the ground, can significantly impair function and quality of life. However, very few studies have systematically investigated the ground-foot interaction during the stance phase in this population[7].

Gait patterns, for both unilateral and bilateral CP have been thoroughly described by Rodda and Graham (2001)[9] and Rodda, Graham, Carson, Galea and Wolfe, (2004)[10], respectively. Each gait pattern has its own unique characteristics. For example, in the true equinus gait pattern, we can assert that initial contact of the foot is usually performed with the forefoot, and therefore it is expected for this region to display increased pressure and longer contact time. Heel contact occurs in late stance or does not occur at all. In apparent equinus, and although the ankle usually presents with a normal range of motion, the knee and hip are excessively flexed throughout stance. This means that initial contact may also be anterior, but the foot will lay flat on the ground in an earlier phase of stance, and probably displaying larger contact areas than the equinus gait pattern.

Optimizing the gait pattern of children with cerebral palsy is a primordial goal in rehabilitation. As we have seen before, there are countless interventions that aim to improve selective motor control and muscle coordination, strength and endurance, biomechanical alignment and overall gait efficiency[11].One of the most prescribed interventions is the use of ankle foot orthoses (AFO). Ankle foot orthoses' main goal is to improve the gait pattern by controlling and positioning the ankle and the foot, during the different phases of the gait cycle. AFO increase stability of the lower limb, amending for muscle weakness and biomechanical misalignment. They can work either by restricting excessive ankle plantarflexion, improving valgus/varus of the foot, and sometimes aiming to influence the positioning of the knee, by allowing a better knee extension during stance[12]. They have demonstrated positive effects in multiple

parameters, like gait speed, step length, knee and ankle joint range or energy expenditure[11,13].

These orthoses may come in a multitude of materials, configurations and may be pre-made or customized according to the identified issues[11,14–18].Among the most used there are the dynamic ankle-foot orthoses (DAFO) that encompasses the posterior region of the leg, ankle and foot, manufactured from a malleable plastic, which restricts both ankle plantarflexion and dorsiflexion, and allows mediolateral stabilization. Another example is the bimaleolar ankle-foot orthoses (BAFO), a shorter version that only reaches the malleoli and allows plantarflexion and dorsiflexion, but provides additional support and heel stabilization.

Instrumented clinical gait analysis has been an excellent tool for planning intervention and assessing outcomes in the rehabilitation process of children with CP[1,2]. Thought the gold standard for gait analysis in children with CP would be a quantitative three-dimensional analysis of movement and respective articular moments and powers (kinematics and kinetics), it is not always possible to conduct such assessment in a clinical setting[19,20]. More accessible and portable methods such as plantar pressure recording devices have been recently used[7,21–24]. Dynamic pedobarography is a relatively simple, portable and non-invasive technology that measures plantar pressure distribution and force throughout the stance phase of gait[25,26]. It also can provide information about contact time and contact area, as well as data that can give us insight about the influence of high and lower pressures for a short or longer duration of time (with the use of parameters like force-time integral and pressure-time integral)[26,27].

A few studies have contributed to produce a normative database[27–30], and variables such as peak pressure, peak force, pressure-time integral, force-time integral, contact area and contact time have consistently showed good reliability for typically developed children[27,29]. Studies on plantar pressure parameters for gait assessment of persons with CP are scarce, and to the best of the authors' knowledge, no investigation on plantar pressures behavior while wearing ankle-foot orthoses has been undertaken. The absence of systematized evidence regarding the different results of plantar pressure measurements on this specific population calls for further investigation. In so, the aim

of this study is to describe the plantar pressure distribution characteristics using insoles, in a sample of children with CP, when walking with AFO's.

6.2. Materials and Methods

6.2.1. Design

Cross-sectional descriptive study.

6.2.2. Participants Selection

A convenience sample of 9 children with cerebral palsy, were recruited from the main hospitals and rehabilitation centers in Lisbon, Portugal, to participate in this study. The selected participants followed the eligibility criteria: male or female children between 4 and 12 years of age, foot length ranging from 15 to 20 cm (due to equipment constraints), with a clinical diagnosis of bilateral (lower limb predominance) or unilateral cerebral palsy, grades I and II on the Gross Motor Function Classification System (GMFCS)[31], habitual users of AFO, able to walk independently for 5 m without walking aids, and able to comprehend and comply with simple instructions. Children should also have not been subjected to orthopedic surgery or botulinum toxin treatment in the previous 6 months. The protocol was approved and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019). All procedures were previously explained to both the child and the legal guardian, an informed consent form was filled and signed by the legal guardian and verbal consent was given by the child.

6.2.3. Data Collection Protocol

Data collection was performed in a clinical setting where the participants usually had their physical therapy sessions. Clinical history and a brief physical exam (mass, height, lower limb posture, selective motor control tests and gastrocnemius length and spasticity)[26] were conducted before pedobarographic data collection.

Children wore the foot insoles Pedar-X system® (Novel, Munich, Germany), inside their usual orthoses, no socks, and their usual footwear (large enough to accommodate the orthoses). The batteries and the wireless transmitter were strapped or placed inside a backpack on the child's back. A schematic picture and a photograph illustrate the experimental setup used – Figure 1. The insoles were calibrated using the Pedar X Standard (v 25.3.6, Novel, Munich, Germany) protocol: before the beginning of each trial, the participant was asked to lift one foot at the time off the ground for approximately 15 s. Data was sampled at 100 Hz. Children were instructed to walk back and forth, along a 5 m line drawn on a smooth and regular floor, unassisted and at a self-selected speed, without running. A chair was placed at either end of the walkway, in case the participants needed to stop. Data collection stopped after 2 min or if the children achieved a minimum of 15 steps with each lower limb.

6.2.4. Data Processing

Data was extracted and processed using the Novel Multiprojects-e (v 24.3.34, Novel, Munich, Germany), that enabled the creation of a database with each participant's individual footprint. Each data set was reviewed and amiss footprints and directional changes were wiped out of the original records. The average of the selected variables (force-time integral, pressure-time integral, maximum force, peak pressure, contact area and contact time) was automatically calculated by the software for the complete foot. A mask was applied to segment the foot into three regions (heel, middle foot and forefoot), according to the length of the foot (0% to 30%, 30% to 60% and 60% to 100% of total length, respectively) – Figure 2. These masks were applied automatically by the software, and average scores were calculated for each variable and region. The software also produced 3D plantar pressure maps for each participant that allowed a visual comparison of the trials, whenever possible – Figure 3.

6.2.5. Statistical Methods

A descriptive exploratory analysis was carried out to identify the behavior of the pedobarography variables under study, for the total of the participants and within the

different sub-set of conditions: type of Cerebral Palsy, gait pattern and type of orthoses used during the trials. Mean values and standard deviation for each variable and participant sub-group was calculated. To identify relevant changes, and since no reference values were found in current literature, the previously calculated values for Minimal Detectable Change (MDC) for pedobarography measurements in children with CP while walking in regular footwear were used[32].

6.3. Results

The nine children with CP, that took part in this study, were characterized as follows: three spastic unilateral, six spastic bilateral with lower limb predominance; two females, seven males; age 85.33± 23.09 months; height 119.44 ± 10.58 cm; mass 24.79 ± 6.68 kg - Table 8. Participant's data from each limb was processed separately (n=18), because of the heterogeneous physical presentation of unilateral CP or the type of orthoses used, that comprised some of the selected cluster samples.

Table 8: Participants' Characteristics

Subject Code	Age (months)	Sex	Clinical Diagnosis	Affected Side	Gait Pattern		GMFCS Level	Mass (kg)	Height (cm)	Insole	AFO	
					Left	Right					Left	Right
PC_AFO_001	72	Male	Spastic Bilateral CP	Both	True Equinus	True Equinus	I	25	109	R-209l-208r	Bimaleolar dynamic AFO	Bimaleolar dynamic AFO
PC_AFO_002	84	Male	Spastic Unilateral CP	Right	-	True Equinus	I	26	114	R-209l-208r		Non-articulated dynamic AFO
PC_AFO_003	108	Female	Spastic Bilateral CP	Both	Apparent Equinus	Apparent Equinus	II	26	132	S-245l-246r	Non-articulated dynamic AFO	Non-articulated dynamic AFO
PC_AFO_005	122	Male	Spastic Bilateral CP	Both	Apparent Equinus	Apparent Equinus	II	33	135	S-245l-246r	Non-articulated dynamic AFO	Non-articulated dynamic AFO
PC_AFO_006	120	Male	Spastic Unilateral CP	Right	-	True Equinus w/ recurvatum knee	I	37	132	S-245l-246r		Articulated dynamic AFO
PC_AFO_008	77	Male	Spastic Bilateral CP	Both	Jump Gait	Jump Gait	II	18	110	R-209l-208r	Non-articulated dynamic AFO	Non-articulated dynamic AFO
PC_AFO_010	65	Male	Spastic Bilateral CP	Both	True Equinus	True Equinus	II	20	118	R-209l-208r	Non-articulated dynamic AFO	Bimaleolar dynamic AFO
PC_AFO_012	56	Female	Spastic Bilateral CP	Both	Apparent Equinus	Apparent Equinus	II	18	110	R-209l-208r	Articulated rigid AFO	Articulated rigid AFO
PC_AFO_017	64	Male	Spastic Unilateral CP	Right	-	True Equinus	I	20,1	115	R-209l-208r		Bimaleolar dynamic AFO

The participants were divided into Clusters, according to the type of Cerebral Palsy, the gait pattern or the type of orthoses used during the trials (Table 9). Cluster 1 focused on the clinical diagnosis and sub-type of Cerebral Palsy (Spastic Bilateral versus Spastic Unilateral). Cluster 2 differentiated between different gait patterns (True Equinus and Apparent Equinus). Cluster 3 compared different types of orthoses used during the trials (Non-articulated dynamic AFO versus Bimaleolar dynamic AFO). Some of the participants presented different types of gait patterns and used different types of orthoses, and in such, their data was excluded from cluster comparisons.

Overall data was consistent across all participants and clusters. As the use of pedobarography is fairly recent in gait analysis of CP children, and there are no published reference values, the authors applied the previously calculated total mean and Minimal Detectable Change (MDC) values for the pedobarography variables in children with CP while walking in regular footwear, for reference purposes – Table 10.

The use of AFO did not significantly impact any of the mean values for the variables in study, when referencing to the means of the same variables in children with CP walking in regular footwear. Similar to the previous study[32], the contact time has a wide range of mean and standard deviation values - Figure 11.

In cluster 1 (distribution of spasticity) the data from the Unilateral group shows overall higher values than the Bilateral group, with pressure time-integral (50,7 kPa*s), maximum force (241,7 N) and contact area (70,6 cm2) showing minimal detectable changes.

Regarding cluster 2 (gait pattern), the Apparent Equinus participants also show higher values in the contact time variable than the True Equinus group. This seems consistent as the Equinus sub-group has two participants with unilateral CP, and therefore naturally shorter contact time.

In cluster 3 and when compared to the Bimaleolar AFO participants, the Dynamic AFO participants have higher values across all variables, with particular significance in the force-time integral (119,1 N*s), maximum force (215,9 N), contact area (63,3 cm^2) and contact time (958,7 s) variables.

Moreover, the Bimaleolar AFO sub-group (cluster 3) displays the lowest values across all variables and all sub-groups, but only the the maximum force value (123,5 N) and the contact area value (40,1 cm^2) register a difference bellow the minimal detectable change value.

Table 9: Total mean and Minimal Detectable Change (MDC) for Pedobarography variables in Children with CP while walking in regular footwear.

Pedobarograpy Measurements	Total Mean	MDC
Force–time integral (N*s)	72,7	36,4
Pressure–time integral (kPa*s)	55,7	19,7
Maximum force (N)	157,5	49,0
Peak pressure (kPa)	139,9	53,9
Contact area (cm^2)	55,0	16,0
Contact time (ms)	672,1	269,1

Table 10: Total and Cluster Pedobarography Measurements.

Pedobarography Measurements	*Total*		*Cluster 1: Distribution of Spasticity*				*Cluster 2: Gait Pattern*				*Cluster 3:Type of Orthoses*			
			Bilateral		*Unilateral*		*True Equinus*		*Apparent Equinus*		*Bimaleolar AFO*		*Dynamic AFO*	
	Mean	*SD*	*Mean*	*SD*	*Mean*	*SD*	*Mean*	*SD*	*Mean*	*SD*	*Mean*	*SD*	*Mean*	*SD*
Force-time integral (N*s)	103,6	47,6	99,8	51,4	100,9	37	88,7	41	115,7	47	69,6	41,2	119,1	56,4
Pressure-time integral (kPa*s)	66,2	43,2	70,5	52,3	50,7	22	60,1	44,5	73,7	43,4	58	58,7	73,6	46,1
Maximum force (N)	193,6	41,1	172,4	41,3	241,7	41,3	174,5	33,7	217,5	41,5	123,5	29,5	215,9	53,5
Peak pressure (kPa)	121,6	32,3	117,7	32,2	132,2	35,6	113,2	30,6	136,8	32,5	97	29,6	132,2	37,2
Contact area (cm^2)	60,5	10,5	52,6	10,9	70,6	9,1	50	8	68,2	10,7	40,1	8,5	63,3	12,9
Contact time (ms)	831	742,4	883,4	899,6	651,2	383,3	670,5	659,1	904	753,1	612,3	838,2	958,7	874,7

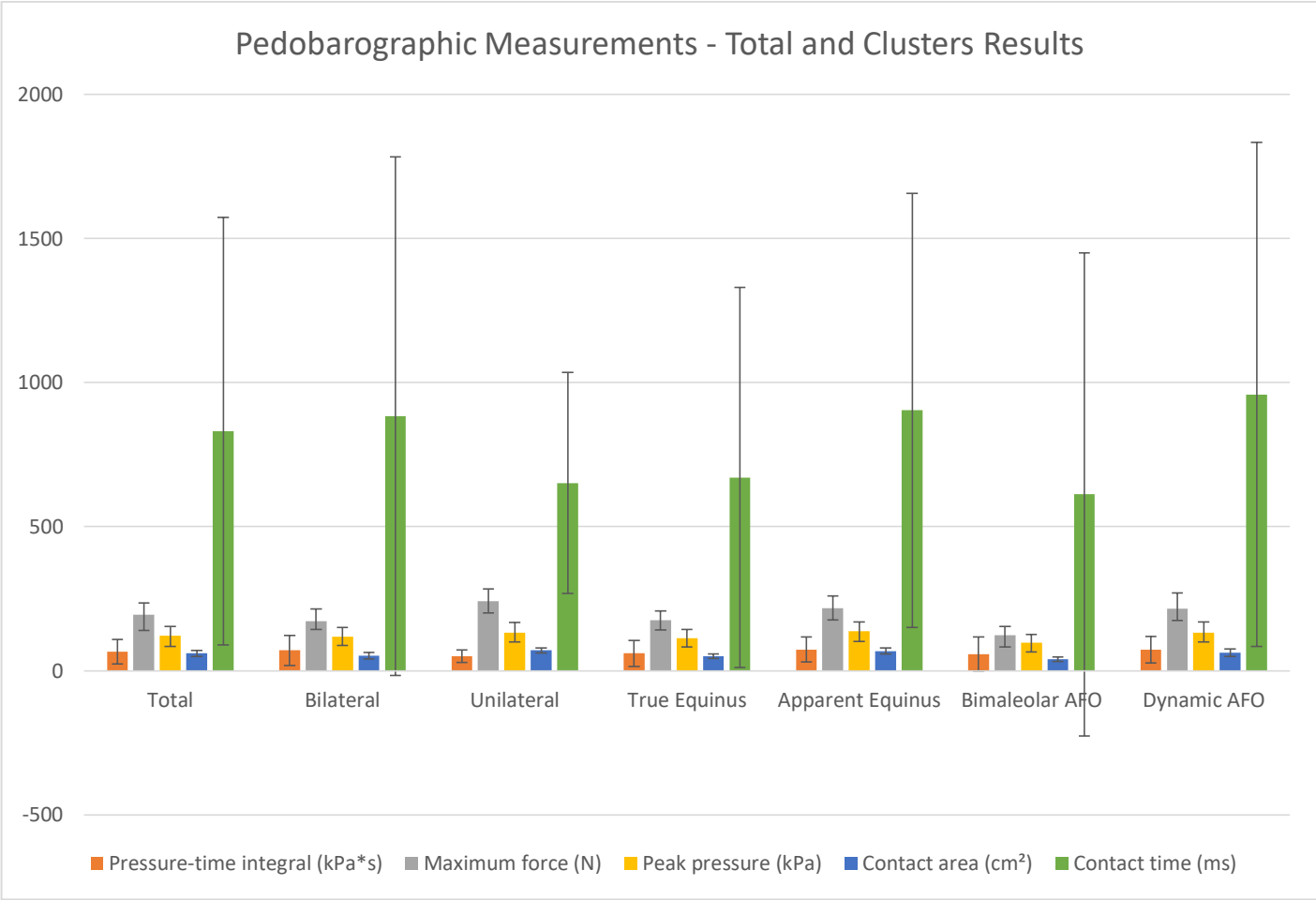

Figure 5: Pedobarography Measurements of Total and Cluster sub-groups.

Data was also explored in plantar segments by dividing the foot in three sections: heel (0% to 30% of the total length of the footprint), middle foot (30% to 60% of the total length of the footprint) and fore foot (60% to 100% of the total length of the footprint) –Table 11.

Maximum force shows a relevant change from heel to the fore foot, when accounting for the MDC difference of 49 N. Again, contact time reveals large and disperse values that are reflected in the obtained means and standard deviations – Figure 12.

Table 11: Mean values for mask measurements (heel, middle foot and fore foot).

Pedobarography Measurements	**Heel (0%-30%)**		**Middle Foot (30%-60%)**		**Fore foot (60%-100%)**	
	Mean	**SD**	**Mean**	**SD**	**Mean**	**SD**
Force-time integral (N*s)	20,4	22,4	38	20,5	45,1	29,7
Pressure-time integral (kPa*s)	35,1	36,2	43,7	26,3	49,2	24,9
Maximum force (N)	52,9	22,1	78,4	20,7	121,5	42,2
Peak pressure (kPa)	66,2	21,3	82,5	26,1	112,3	37,5
Contact area (cm^2)	12,9	4,4	24,1	4,9	23,0	5,3
Contact time (ms)	739,2	749,7	775,8	607,7	780,0	603,2

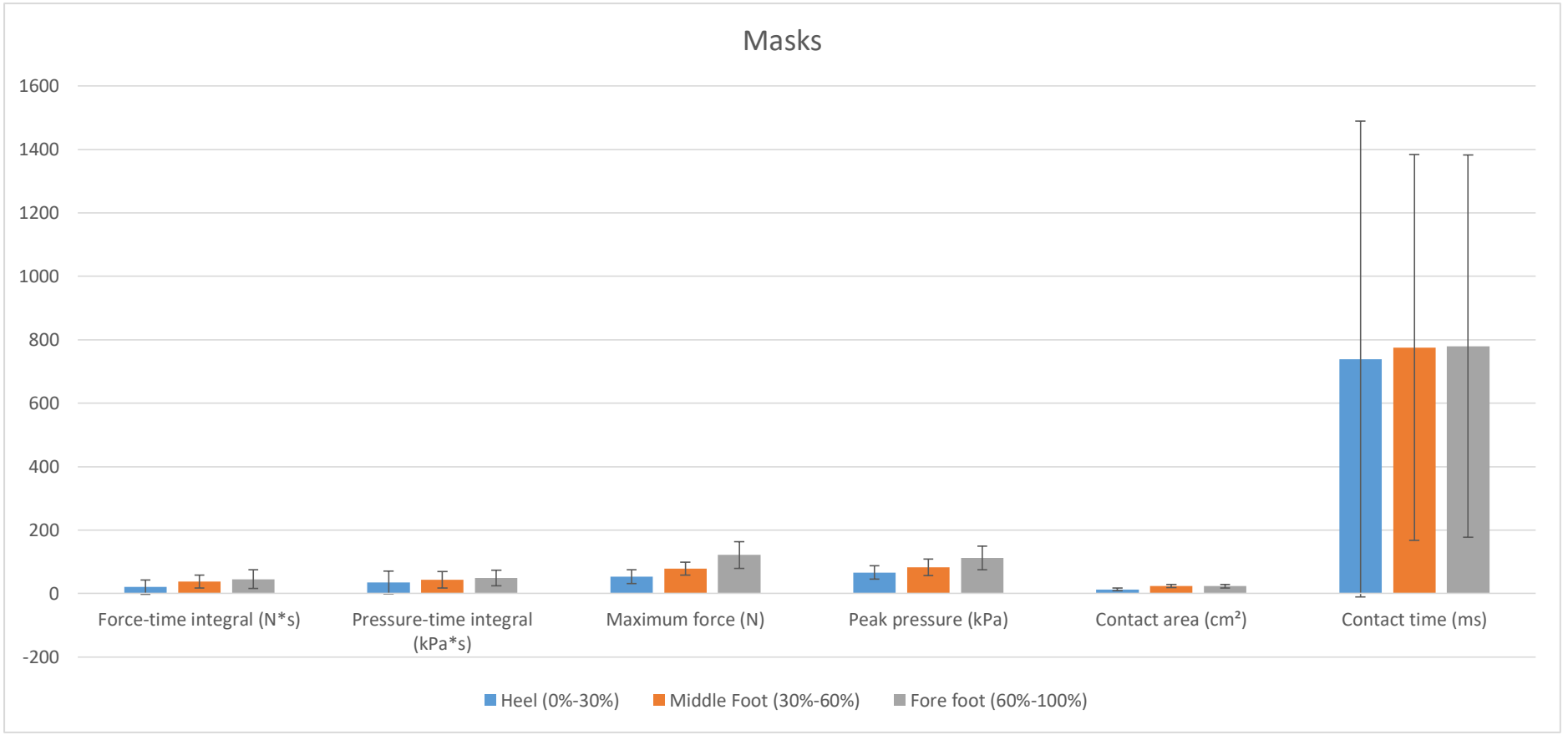

Figure 65: Pedobarographic mean results for mask measurements (Heel, middle foot and fore foot).

6.4. Discussion

Our results seem to be consistent with previous studies that described the changes in plantar pressure in children with CP. To the authors' best knowledge, this is the first study to assess plantar pressure parameters in children with CP while wearing AFO. Knowing how the foot interacts with the AFO may prove substantial information for tuning and optimizing the orthoses to each individual. With that purpose, studies that assess plantar pressure within the AFO provide valuable information.

The existing literature regarding plantar pressure is sparse, even in typically developing children. Kasović, Štefan and Zvonár (2020)[27] published a normative data base with percentiles for different parameters of plantar pressure analysis for children ages between 6 and 14 years old. When compared to their healthy peers, and as expected, the participants of this sample (mean 85.33 months or 7.1 years old), when wearing AFO's, seem to present lower pedobarography results than typically developed children for all variables, with the exception of contact time, which was above of the 95th percentile. Force-time integral was on the 10th percentile, and pressure-time integral, peak pressure and contact area were below the 5th percentile.

These results accurately represent the natural deviation of plantar pressures in the gait pattern of children with CP while wearing AFO's. While the AFO strives to minimize the cumulative force and pressure dislocated towards the forefoot region, our results seem to show that it still does not equals to those of their healthy peers. Overtime these deviations may lead to the musculoskeletal deformity often found in children with CP.

Also, as opposed to the typically developed foot, that tends to have larger contact area and shorter contact time[27], children with CP seem to have a smaller foot area distribution and increased contact time, due to the asymmetry of gait and the difficulties in postural control.

AFO´s purpose is to restrict excessive ankle plantarflexion, promoting a more plantigrade footprint which will naturally increase the contact area and maximum force. AFO use seems to be beneficial, particularly in groups with asymmetric gait or in groups

were the equinus position of the foot can be managed (i.e. Unilateral CP distribution, Apparent Equinus gait pattern or the Dynamic AFO group).

Also, the influence of an affected lower limb on its contralateral cannot be disregarded. In Unilateral CP, contact time, stride length and step duration, can be deviated as the less affected limb will support a higher percentage of the load during an extended period. This asymmetry in stance and therefore contact time, is not so evident in Bilateral CP, since both lower limbs are similarly affected[33,34].

The segmental analysis using the footprint masks revealed the increase of all variables towards the forefoot, as it was expected, since most of the sample is composed by participants with equinus gait patterns, even when wearing the AFO.

The use of AFO, does not seem to change the pedobarographic parameters in order to claim that a relevant change has occurred. Nevertheless, there was an increase in the values of force-time integral, pressure-time integral, peak pressure and contact area while wearing AFO, which brings them closer to the reference percentiles for the typically developing children.

6.4.1. Limitations

The sample size of this study was small, and thus, making it hard to draw additional conclusions. Nonetheless it provides a starting point for much needed further investigation in the area. Due to the lower sample size and the heterogeneous presentation of CP, a separate lower limb analysis was conducted. This means that in the cluster analysis, the “healthy” lower limbs of Spastic Unilateral CP and data from participants that did not fit the cluster criteria (Jump Gait and Articulated AFO) were excluded, and therefore may have influenced the final outcome.

6.5. Conclusion

The use of AFO in children with CP can produce positive changes in plantar pressure measurements, approximating them to the reference percentiles of typically

developing children. The use of AFO seems to be beneficial, but further investigations with larger sample size in these 3 cluster groups and in controlled conditions are due.

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Chapter VII – General Discussion

Ankle-foot orthoses are widely used as a rehabilitation device for children and adults with Cerebral Palsy, but they lack the evidence to support its generalized use. On the other hand, gait analysis has proven to be a universally used and strong tool, considered a gold standard in CP rehabilitation. By combining the use of two rehabilitation tools (AFO's and plantar pressure analysis), this PhD thesis aims to contribute to deepen the knowledge and available evidence on the field.

The body of evidence on the clinical and rehabilitation aspects of Cerebral Palsy is ever growing, but practical difficulties still represent a huge set back when it comes to systemize new evidence. Cerebral Palsy is a heterogeneous condition, with multiple clinical presentations, and each individual is unique. This makes studies with large number of participants hard to accomplish, as eligibility criteria often excludes part of the available sample.

This chapter is therefore divided in two parts. On the first part we can find an overview of the main findings of the investigation and the limitations of the several studies are exposed, and the overall difficulties of doing research in this field. The second part elaborates on the practical implications of the main findings in the clinical rehabilitation setting and daily life of people with Cerebral Palsy, and also suggests new lines of investigation that can be undertaken in the near future.

7.1. Main Findings and Limitations

In **Chapter IV**, we found that 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[1–4], suggesting a better gait efficiency and probably less energy expenditure[5].

As expected, wearing any type of AFO meant significant differences in the range of motion of the ankle, when compared to the barefoot condition. AFO's are designed to reduce pathological plantarflexion, common in several of the bilateral CP gait patterns[2]. However, some types of orthoses (DAFO, SAFO and GRAFO) are particularly more effective in controlling tibial progression and consequently promote knee

extension during stance[6]. This can impact and modify the crouch gait pattern of CP children, approximating it to that of healthy subjects.

There were also significant increases in ground-reaction force and joint moments at push-off, while wearing AFO[2]. 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[2,3].

Only three studies focused on functional gains, and often these variables are under reported and are not correlated with biomechanical variables of gait analysis. Research in Cerebral Palsy is often complex, and this aspect is frequently reflected in the identified limitations of the research conducted with this specific population.

The methodology used for the scoping review followed the Cochrane guidelines and was previously registered in PROSPERO (International prospective register of systematic reviews). Nonetheless, due to our eligibility criteria, the number of articles included was lower than other similar reviews. Of the 10 studies that were included, there was no common primary outcome between them.

Studies often comprised heterogeneous groups, with different gait patterns and different approaches to the use of AFO. There is a wide variety of variables and outcomes which makes difficult the comparison between studies and consequently to access the effectiveness between AFO.

Studies also did not report the gait pattern classification and the type of AFO used nor the clinical reasoning behind the AFO prescription, which makes it more difficult to systematically assess the effects of the AFO in gait performance of children with CP. A wide variety of AFO are used in clinical practice, which are characterized by their design and construction, materials used (including stiffness, thickness, and other mechanical properties) and AFO/footwear combinations and may produce different outcomes. 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 likewise may have impacted the results of their study.

Overall AFO's seem to have an immediate and a long-term effect in improving the sagittal gait patterns in children with spastic bilateral CP. Further investigation about

what are the effects of the AFO and how the foot lays and moves inside the orthoses was needed.

This led to the work developed in **Chapter V**. The initial research showed that plantar-pressure-related data for children with CP are still scarce in published evidence. There was a fundamental need to establish the plantar pressure insoles as a reliable tool to be used in this specific population, as they provide important information about foot pressure distribution, postural control, center of pressure (COP) displacement, and the foot–soil interaction.

The use of this particular tool may prove useful in assessing the effects of orthotic options and medical and surgical interventions, as they allow the possibility to use inside shoes or orthotic devices recording a higher number of gait cycles, as well as overall being easier to use with smaller children.

Trials to assess intersession and intra-rater reliability testing of plantar pressure variables when using pressure foot insoles were conducted. The results showed high reliability (ICC ≥ 0.60) for 21 of the 24 parameters that were tested. Still, three of the outcome measures for whole foot and forefoot showed lower values (whole foot contact time variable and peak pressure and contact time variables at the forefoot).

Due to the lower sample size and the heterogeneous presentation of CP, a separate lower limb analysis was conducted. The separate analysis of right and left feet increases the total sample to sixteen (feet) in the reliability study. Post-hoc power analysis with $\alpha = 0.05$ revealed good power (≥0.90) for most variables, except for the three variables mentioned above (whole foot contact time variable and peak pressure and contact time variables at the forefoot).

The poor results obtained for the contact time and peak pressure may be explained by different factors. The heterogeneous gait pattern and the difficulties in weight bearing on the most affected limb, especially in children with unilateral CP. Moreover, although we asked the children to walk at a self-selected comfortable speed, their pace varied. Other constraints may have arisen form the slight discrepancy between the total foot length and the length of the available insole. Also, limitations concerning the total weight of the equipment (about 0.5 kg), which may impact the trials of some of the smaller children and those with greater locomotion difficulties and gait deviations.

The obtained values for MDC% were all considered to be poor, but these results are equivalent to other similar studies[7,8]. The absence of previous reliability studies with this population and method precludes comparisons with similar SEM and MDC data. Still these preliminary results could prove useful to determine clinical changes in foot pressure and understand how those changes differentiate from the error of measurement. This is particularly important in studies where we have a pre- and post-assessment of the participant to see the effect of an intervention process. If the post results are superior to the reported error of the measurement, we can be confident in stating that there was a significant effect caused by the intervention.

Another limitation to research, particularly in pedobarography, is foot segmentation. Various foot segmentation models have been reported in recent literature[9–19]. Complex masking usually involves anatomical and functional segmentation, including external references. Smaller areas of division may provide with less detailed information, and they are also more error-prone[14]. A three identical part division masking was selected for this study, similar to that of Galli et al. [9], allowing to differentiate force, pressure, and spatio-temporal values between the hind-foot, midfoot, and forefoot.

Establishing plantar pressure insoles as a reliable tool to be used with children with CP was a significant step forward into better understanding how the pathological foot interacts with the ground and the AFO during gait.

Lastly, in **Chapter VI** we aimed to assess plantar pressure parameters in children with CP while wearing AFO. Knowing how the foot interacts with the AFO may prove substantial information for tuning and optimizing the orthoses to each individual. Globally, children with CP wearing AFO's presented lower pedobarographic results than their typically developed peers for all variables, except for contact time, which was above of the 95th percentile. Force-time integral was on the 10th percentile, and pressure-time integral, peak pressure and contact area were below the 5th percentile.

While the AFO strives to minimize the cumulative force and pressure dislocated towards the forefoot region, our results show that it still does not equals to those of their healthy peers. Overtime these deviations may lead to the musculoskeletal deformity often found in children with CP[20]. Also, children with CP seem to have a

smaller foot area distribution and increased contact time, due to the asymmetry of gait and the difficulties in postural control.

Also, the influence of an affected lower limb on its contralateral cannot be disregarded. In Unilateral CP, contact time, stride length and step duration, can be deviated as the less affected limb will support a higher percentage of the load during an extended period. This asymmetry in stance and therefore contact time, is not so evident in Bilateral CP, since both lower limbs are similarly affected[18,21].

In this chapter, the separate limb analysis increased the sample size to 18 feet. However, in the subsequent cluster analysis, the "unaffected" lower limbs of Spastic Unilateral CP and data from participants that did not fit the cluster criteria (Jump Gait and Articulated AFO) were excluded, and therefore may have influenced the final outcome.

For both experimental studies (chapter V and chapter VI), the sample size were small, and thus, making it hard to draw additional conclusions. Nevertheless, there was an increase in the values of force-time integral, pressure-time integral, peak pressure and contact area while wearing AFO, which brings them closer to the reference percentiles for the typically developing children.

7.2. Practical Implications and Future Research

Overall, the growing body of evidence in CP research supports the use of AFO, as a way to prevent deformities, improve energy efficiency and approximate the gait pattern of children with CP to that of their typically developing peers.

However, small sample sizes and heterogeneous population increase the difficulty in producing high quality and strong methodological evidence. 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. Future studies should invest in higher methodological quality protocols that account for the limitations stated throughout this dissertation.

Also, the use of a common language may prove very useful. The International Classification of Functionality (ICF) is the World Health Organization (WHO) framework for measuring health and disability at both individual and population levels and is used as a common language between different professionals. 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.

Pedobarography, particularly plantar pressure insoles, proved to be a reliable tool for assessing plantar pressure variables in children with CP, and a normative database for this population would prove valuable in future research.

Alongside with the construction of the normative database, further research about the effects of AFO use in larger sample sizes and in the specific sub-groups (divided by distribution of spasticity, gait pattern, AFO type and other distinctive characteristics) is called upon.

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Appendix I

Ricardo D, **Raposo MR**, Cruz EB, Oliveira R, Carnide F, Veloso AP, João F. Effects of Ankle Foot Orthoses on the Gait Patterns in Children with Spastic Bilateral Cerebral Palsy: A Scoping Review. Children (Basel). 2021 Oct 10; 8(10):903. doi: 10.3390/children8100903. PMID: 34682168; PMCID: PMC8534539.

children

MDPI

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

Citation: 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. https://doi.org/10.3390/children8100903

Academic Editor: Hilde Van Waelvelde

Received: 1 August 2021
Accepted: 7 October 2021
Published: 10 October 2021

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).

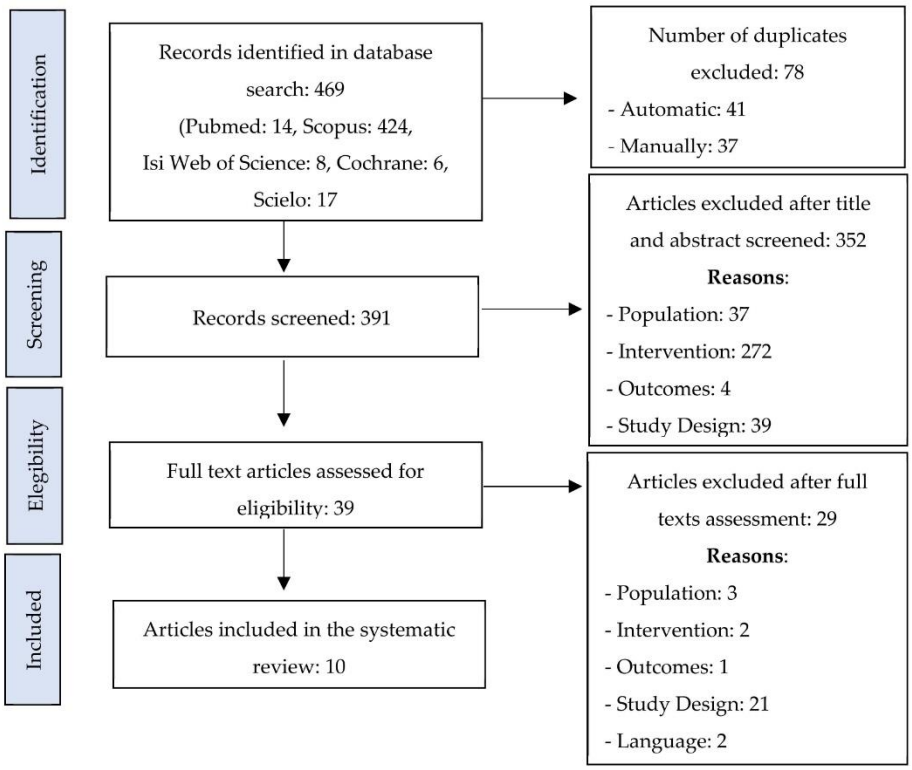

Figure 1. Flowchart of the article selection process.

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 ($p < 0.05$). 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 ($p \leq 0.05$). 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 ($p < 0.05$). 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 scizures; 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.

Author Contributions: Conceptualization, D.R., M.R.R., R.O., F.C., A.P.V. and F.J.; methodology, D.R., M.R.R. and E.B.C.; software D.R., M.R.R. and E.B.C.; validation, A.P.V. and F.J.; formal analysis, E.B.C., R.O., F.C., A.P.V. and F.J.; investigation, D.R., M.R.R. and E.B.C.; resources, A.P.V. and F.J.; data curation, D.R., M.R.R., E.B.C. and F.J.; writing—original draft preparation D.R., M.R.R., E.B.C. and F.J.; writing—review and editing, D.R., M.R.R., R.O., F.C., E.B.C. and F.J.; visualization, R.O., F.C., A.P.V. and F.J.; supervision, E.B.C. and F.J.; project administration, F.J.; funding acquisition, A.P.V. All authors have read and agreed to the published version of the manuscript.

Funding: This work was supported by the Fundação para a Ciência e Tecnologia (FCT), through CIPER (unit 447) with the reference project UIDB/00447/2020.

Institutional Review Board Statement: Not applicable.

Informed Consent Statement: Not applicable.

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

Conflicts of Interest: The authors declare no conflict of interest.

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 II

Raposo MR, Ricardo D, Teles J, Veloso AP, João F. Gait Analysis in Children with Cerebral Palsy: Are Plantar Pressure Insoles a Reliable Tool? Sensors (Basel). 2022 Jul 13; 22(14):5234. doi: 10.3390/s22145234. PMID: 35890913; PMCID: PMC9319716.

sensors

MDPI

Article

Gait Analysis in Children with Cerebral Palsy: Are Plantar Pressure Insoles a Reliable Tool?

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Abstract: Cerebral palsy (CP) is a common cause of motor disability, and pedobarography is a useful, non-invasive, portable, and accessible tool; is easy to use in a clinical setting; and can provide plenty of information about foot–soil interaction and gait deviations. The reliability of this method in children with CP is lacking. The aim of this study is to investigate test–retest reliability and minimal detectable change (MDC) of plantar pressure insole variables in children with CP. Eight children performed two trials 8 ± 2.5 days apart, using foot insoles to collect plantar pressure data. Whole and segmented foot measurements were analyzed using intraclass correlation coefficients (ICC). The variability of the data was measured by calculating the standard error of measurement (SEM) and the MDC/ICC values demonstrated high test–retest reliability for most variables, ranging from good to excellent (ICC ≥ 0.60). The SEM and the MDC values were considered low for the different variables. The variability observed between sessions may be attributed to the heterogeneous sub-diagnosis of CP.

Keywords: plantar pressure; cerebral palsy; gait analysis; reliability; insoles

Citation: Raposo, M.R.; Ricardo, D.; Teles, J.; Veloso, A.P.; João, F. Gait Analysis in Children with Cerebral Palsy: Are Plantar Pressure Insoles a Reliable Tool? *Sensors* **2022**, *22*, 5234. https://doi.org/10.3390/s22145234

Academic Editor: Carlo Ricciardi

Received: 14 June 2022
Accepted: 11 July 2022
Published: 13 July 2022

1. Introduction

Cerebral palsy (CP) is the most common cause of motor disability in children [1–3]. CP is a complex pathology that describes a group of impairments and motor disorders, which are permanent but not immutable, resulting from a nonprogressive cerebral disorder [4] with different presentations and functional levels [5].

CP presents both positive features such as spasticity, hyper-reflexia, and co-contraction, and negative features including weakness, difficulties in motor control, and sensory and balance impairments [6]. The lack of control is obvious at the lower limb joints, especially the ankle joint. These alterations are the main cause of limb contractures, musculoskeletal deformity, and gait deviations [7].

Foot deformities, along with hip displacement, are the most common musculoskeletal occurrences in CP. Among the most common foot deformities in this population are equinus, planovalgus, and equinovarus, which can vary from very mild and flexible to severe and rigid [8]. These deformities, which cause the foot to abnormally lay on the ground, can significantly impair function and quality of life; however, very few studies have systematically investigated the foot morphology and the ground–foot interaction during the stance phase in this population [7].

Instrumented clinical gait analysis has been an excellent tool for planning intervention and assessing outcomes in the rehabilitation process of children with CP [1,2]. Though the gold standard for gait analysis in children with CP would be a quantitative three-dimensional analysis of movement and respective articular moments and power (kinematics and kinetics), possibly alongside muscle activation (electromyography) and

oxygen consumption [9], it is not always possible to conduct such an assessment in a clinical setting. More accessible and portable methods have been recently used such as inertial sensors [10,11] and plantar pressure recording devices [7,12–15].

Under this aspect, dynamic pedobarography is a relatively simple, portable, and non-invasive technology that measures the change in plantar pressure distribution throughout the stance phase of gait [16]. It is an easy method to use in a clinical setting; can provide plenty of information about foot–soil interaction; and, alongside other gait analysis methods, can help assess the impact of a medical intervention, a rehabilitation program, or the effects of an orthotic device. Several studies tested its reliability [16,17] for both healthy adult and children, but none have assessed subjects with CP. The few existing clinical studies in participants with CP use mainly plantar pressure mats/platforms instead of insoles [7,12–15].

In the past years, several studies have tried to produce normative age-dependent gait databases [18–20], which are fundamental to assess and compare with pathologic situations. In fact, more evidence is now surfacing about the foot characteristics of typically developed children. Foot pressure changes dramatically throughout the life cycle, especially in the early years (up to 6 years old). The evidence shows that, while younger typically developing children present with a flatfoot pattern, older children tend to develop a more curvilinear pattern [18]. Moreover, older children show greater values in the main plantar pressure variables when compared with younger children [20].

Even fewer studies have included plantar pressure measurements in children with CP. There has been no attempt to create any kind of database, which is fundamental to assess and compare the natural progression of the condition and the results of medical and therapeutic interventions. Nevertheless, data collected across the existing studies show that there is a variability in foot pressure distribution depending on spasticity overall, there is an increase in pressure towards the toes and forefoot as well as a significant reduction towards the heel [7,14,21].

Reports of plantar pressure data in the literature are highly heterogeneous. One of the challenges of standardizing this tool is that there are multiple footprint segmentation models [19]. There is still no consensus about which foot model may provide the most detailed information, without losing the functional aspects of the foot [15]. Most authors propose an anatomical/functional segmentation, corresponding to the foot joint positions, which ranges from as few as 3 to as many as 12 subdivisions of the footprint (the most often used are the hind-foot, mid-foot (medial and lateral), forefoot (medial and lateral), and toes (toes 2–5 and the first toe) [7,13–15,17–19,21–24].

The absence of systematized evidence regarding the reliability of foot pressure insoles on this specific population and the need to assess the dimension of error measurement with this tool calls for further investigation. In so, the aim of this study is to investigate test–retest reliability and minimal detectable change of plantar pressure insoles in a sample of children with CP when walking in regular footwear.

2. Materials and Methods

2.1. Design

Prospective intra-rater test–retest reliability and minimal detectable change study.

2.2. Participants Selection

A convenience sample of 10 children with cerebral palsy was selected from a Portuguese rehabilitation center to participate in this study. The selected participants followed the eligibility criteria: male or female children between 4 and 12 years of age, foot length ranging from 15 to 20 cm (because of equipment constraints), with a clinical diagnosis of bilateral (lower limb predominance) or unilateral cerebral palsy, grades I and II on the Gross Motor Function Classification System (GMFCS) [25], able to walk independently for 5 m without walking aids, and able to comprehend and comply with simple instructions. Children should also have not been subjected to orthopedic surgery or botulinum toxin

treatment in the previous 6 months. The protocol was approved and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019). All procedures were previously explained to both the child and the legal guardian, an informed consent form was filled and signed by the legal guardian, and verbal consent was given by the child.

2.3. Data Collection Protocol

Data collection was performed on two different days within a period of 7 to 14 days (8 ± 2.5 days) to minimize the assessor memory bias and to prevent a change in the children's gait pattern or clinical condition. Clinical history and a brief physical exam (mass, height, lower limb posture, selective motor control tests, gastrocnemius length, and spasticity) [9] were conducted in the first session.

Children wore the foot insoles Pedar-X system® (Novel, Munich, Germany), inside their usual footwear (adequate to their feet size) and no socks. The children wore the same pair of shoes for both trials. The batteries and the wireless transmitter were strapped or placed inside a backpack on the child's back. A schematic picture and a photograph illustrate the experimental setup used (Figure 1). The insoles were calibrated using the Pedar X Standard (v 25.3.6, Novel, Munich, Germany) protocol (before the beginning of each trial, the participant was asked to lift one foot at a time off the ground for approximately 15 s). Data were sampled at 100 Hz. Children were instructed to walk back and forth, along a 5 m line drawn on a smooth and regular floor, unassisted and at a self-selected speed, without running. A chair was placed at either end of the walkway, in case the participants needed to stop. Data collection stopped after 2 min if the children achieved a minimum of 15 steps with each lower limb.

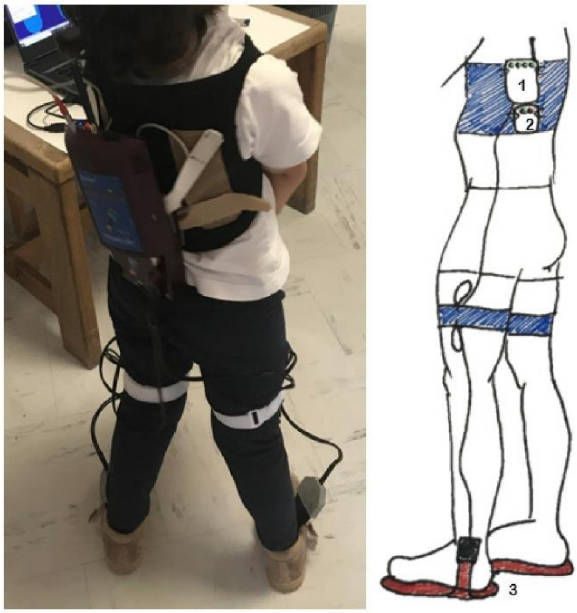

Figure 1. Experimental set up (1—wireless transmitter; 2—batteries; 3—plantar pressure insoles).

2.4. Data Processing

Data were extracted and processed using the Novel Multiprojects-e (v 24.3.34, Novel, Munich, Germany), which enabled the creation of a database and processing of each participant's individual footprint. Each data set was reviewed and amiss footprints and directional changes were wiped out of the original records. The average of the selected variables (force–time integral, pressure–time integral, maximum force, peak pressure, contact area, and contact time) was automatically calculated by the software for the whole

foot. A mask then divided the foot into three regions (hindfoot, middle foot, and forefoot), according to the length of the foot (0 to 30%, 30 to 60%, and 60 to 100% of total length, respectively), as shown in Figure 2. These masks were applied automatically by the software, and average scores were calculated for each variable and zone. The software also produced 3D plantar pressure maps for each participant, allowing a visual comparison of the first and second trial (Figure 3).

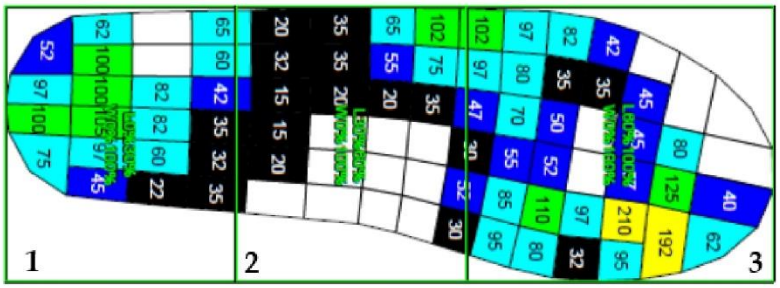

Figure 2. Three zones of segmentation of the foot (1—0% to 30% of total length; 2—30% to 60% of total length; 3—60% to 100% of total length). Obtained from Novel Multiprojects-e (v 24.3.34, Novel, Munich, Germany).

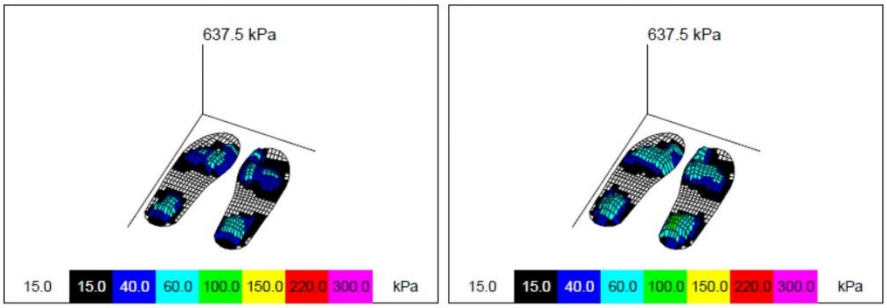

Figure 3. Three-dimensional plantar pressure mapping for test–retest results of participant 008. Obtained from Novel Multiprojects-e (v 24.3.34, Novel, Munich, Germany).

2.5. Statistical Analysis

Statistical analysis to assess the test–retest reliability of plantar pressure data was carried out using the methodology described by Koo and Li (2015) [26], similar to the methods used by Fernandes et al. (2015) [27] and Ricardo et al. (2021) [28] in their works.

Intraclass correlation coefficients (ICCs) considering the two-way mixed model with absolute agreement and accounting for the mean of multiple measurements [26] were calculated for all variables and masks, and a critical level of $p < 0.05$ was considered significant. The *ICC* statistical analysis was performed using SPSS (version 28.0.0; IBM, Chicago, IL, USA), using the following formula:

$$ICC = \frac{MS_R - MS_E}{MS_R + \frac{MS_C - MS_E}{n}}$$

where MS_R represents the mean square between lower limbs; MS_E represents the mean square for error; MS_C represents the mean square within lower limbs, concerning the selected pedobariografic variables; and n is the total number of lower limbs assessed (two lower limbs for each of the eight participants).The level of agreement was considered poor, fair, good, and excellent when $ICC < 0.40$, $0.40 \leq ICC < 0.60$, $0.60 \leq ICC < 0.75$, and $0.75 \leq ICC \leq 1.00$, respectively [29]. Calculations also included the mean difference

between measurements ($Mean_{diff}$), the 95% CI for the $Mean_{diff}$, the standard deviation of the differences (SD_{diff}), and the 95% Bland and Altman limits of agreement (95% LOA).

The absolute measure of reliability standard error of measurement (*SEM*) was calculated using the following equation:

$$SEM = \frac{SD_{diff}}{\sqrt{2}}$$

where SD_{diff} represents the standard deviation of the difference.

To determine the smallest amount of change that must be achieved to reflect a true change, outside the error of the tests, the minimal detectable change (*MDC*) was calculated using the following equation:

$$MDC = 1.96 \cdot \sqrt{2} \cdot SEM$$

The *SEM* and *MDC* were calculated using Microsoft Excel 2013 (Microsoft Corporation, Redmond, WA, USA).

3. Results

The participants of the study were a convenience sampling of ten children with CP (nine spastic unilateral, one spastic bilateral; four females, six males; age 57.9 ± 13.4 months; height 110.4 ± 7.6 cm; mass 18.1 ± 2.4 kg) (Table 1), two of which dropped out of the study as they could not complete the trials in the same time frame as the other participants (one because of COVID-19 prophylactic quarantine and the other because of loss of contact). Data from each limb were processed separately (N = 16), because of the heterogeneous physical presentation of unilateral CP that composed most of the selected sample. On average, we assessed 75.8 ± 27.9 steps on each trial.

Table 1. Participants' characteristics.

Participant	Gender	Age (Months)	Diagnosis	Affected Side	GMFCS Level [25]	Interval between Trials (Days)	Mass (kg)	Height (cm)	Sagittal Gait Pattern [30,31]		Gastrocnemius Spasticity (Modified Ashworth Scale) [32]		Foot Length (cm)		Number of Steps (Average from Both Trials)		Status
									Right	Left	Right	Left	Right	Left	Right	Left	
001	Male	54	Unilateral CP	Right	I	14	16.5	105	Drop Foot	-	1	0	15	16	70	70	Completed trials
002	Male	65	Unilateral CP	Left	II	9	20	118	-	True Equinus	0	4	19	17	52	59	Completed trials
003	Female	41	Unilateral CP	Right	II	7	19	105	True Equinus	-	1+	0	16	17	55	52	Completed trials
004	Female	56	Bilateral CP	Both	II	7	18	110	Apparent Equinus	Apparent Equinus	1	1	17	17	55	55	Completed trials
005	Female	65	Unilateral CP	Right	I	7	20.4	120	True Equinus	-	1	0	20	19	64	65	Completed trials
006	Male	45	Unilateral CP	Left	I	-	13	97	-	True Equinus	0	1	15	15		-	Dropped out
007	Male	41	Unilateral CP	Right	I	-	16	103	True Equinus	-	1	0	15	16		-	Dropped out
008	Male	74	Unilateral CP	Right	I	7	20.5	115	True Equinus	-	1	0	20	20	75	74	Completed trials
009	Male	80	Unilateral CP	Right	I	6	20.1	115	True Equinus	-	1+	0	19	19	122	120	Completed trials
010	Female	58	Unilateral CP	Right	I	7	17	116	Equinus/ Jump Knee	-	2	0	16	18	112	119	Completed trials

3.1. Reliability of Whole Foot Measurements

As shown in Table 2, all selected variables calculated for the whole footprint showed an excellent ICC (ICC ≥ 0.75), except for the contact time variable (ICC = 0.36, 95% CI 0 to 0.784). The SEM and MDC values were within an acceptable range for each of the variables.

Table 2. Reliability values for pedobarography measurements (whole foot).

Pedobarograpy Measurements	ICC	ICC 95% CI	Mean	Mean Diff	SD Diff	95% LOA	SEM	MDC
Force–time integral (N·s)	0.76	(0.30; 0.92)	73.72	−2.08	18.57	(−38.47; 34.31)	13.13	36.39
Pressure–time integral (kPa·s)	0.89	(0.70; 0.96)	55.40	0.63	10.04	(−19.05; 20.31)	7.10	19.68
Maximum force (N)	0.79	(0.42; 0.93)	161.30	−7.61	25.00	(−56.61; 41.40)	17.68	49.00
Peak pressure (kPa)	0.81	(0.47; 0.93)	136.45	6.84	27.48	(−47.01; 60.70)	19.43	53.85
Contact area (cm^2)	0.83	(0.53; 0.94)	56.80	−3.69	8.15	(−19.66; 12.27)	5.76	15.97
Contact time (ms)	0.37	(0; 0.78)	669.93	4.29	137.30	(−264.81; 273.40)	97.08	269.11

3.2. Reliability of Segmented Foot Measurements

Overall ICC values for the segmented foot measurements fit in the good to excellent range (ICC values ≥ 0.60), except for peak pressure (ICC = 0.439, 95% CI 0 to 0.807) and maximum force (ICC = 0.552, 95% CI 0 to 0.845) at the forefoot (Table 3). The SEM and MDC values were within an acceptable range for each of the variables.

Table 3. Reliability values for pedobarography measurements (three zones of the segmented foot).

	Pedobarograpy Measurements	ICC	ICC 95% CI	Mean	Mean Diff	SD Diff	95% LOA	SEM	MDC
Hindfoot	Force–time integral (N·s)	0.83	(0.51; 0.94)	17.44	−1.43	11.35	(−23.67; 20.82)	8.02	22.24
	Pressure–time integral (kPa·s)	0.97	(0.92; 0.99)	21.41	0.62	12.01	(−22.93; 24.16)	8.49	23.54
	Maximum force (N)	0.92	(0.77; 0.97)	70.50	−6.38	28.65	(−62.53; 49.77)	20.26	56.15
	Peak pressure (kPa)	0.88	(0.65; 0.96)	78.56	−3.84	18.48	(−40.06; 32.37)	13.07	36.22
	Contact area (cm^2)	0.91	(0.75; 0.97)	13.68	−1.76	6.19	(−13.89; 10.36)	4.38	12.13
	Contact time (ms)	0.86	(0.62; 0.95)	365.79	38.16	272.29	(−495.53; 571.85)	192.54	533.69
Middle Foot	Force–time integral (N·s)	0.91	(0.75; 0.97)	15.32	0.52	3.14	(−5.63; 6.67)	2.22	6.15
	Pressure–time integral (kPa·s)	0.97	(0.92; 0.99)	30.19	0.91	5.95	(−10.75; 12.57)	4.21	11.66
	Maximum force (N)	0.91	(0.74; 0.97)	47.92	−2.32	7.84	(−17.69; 13.05)	5.54	15.37
	Peak pressure (kPa)	0.97	(0.92; 0.99)	74.89	1.19	8.31	(−15.09; 17.47)	5.87	16.28
	Contact area (cm^2)	0.98	(0.94; 0.99)	16.54	−0.34	2.07	(−4.39; 3.72)	1.46	4.06
	Contact time (ms)	0.73	(0.25; 0.90)	621.32	9.79	118.82	(−223.09; 242.67)	84.02	232.88
Forefoot	Force–time integral (N·s)	0.73	(0.25; 0.90)	40.95	−1.18	11.14	(−23.02; 20.66)	7.88	21.84
	Pressure–time integral (kPa·s)	0.97	(0.92; 0.99)	42.35	1.53	7.30	(−12.77; 15.83)	5.16	14.30
	Maximum force (N)	0.73	(0.26; 0.90)	123.44	−5.93	23.40	(−51.80; 39.95)	16.55	45.87
	Peak pressure (kPa)	0.44	(0; 0.81)	124.59	8.68	28.00	(−46.19; 63.55)	19.80	54.87
	Contact area (cm^2)	0.68	(0.07; 0.89)	25.59	−3.57	7.21	(−17.70; 10.55)	5.10	14.12
	Contact time (ms)	0.55	(0; 0.85)	578.39	22.66	194.57	(−358.70; 404.02)	137.58	381.36

4. Discussion

The main objective of the current study was to assess the intersession and intra-rater reliability of plantar pressure variables when using pressure foot insoles and, to the best of the authors' knowledge, it is the first study to do so. Plantar-pressure-related data for children with CP are still scarce in published evidence. Alongside other gait analysis tools, pedobarographic measurements are useful in assessing pre- and post-surgical outcomes, treatment with botulinum toxin, and orthotic management, as they provide important information about foot pressure distribution, postural control, center of pressure (COP) displacement, and the foot–soil interaction. Nonetheless, if this type of data is to be used for assessing clinical or therapeutic interventions, it is of high importance to establish reliability levels for this specific method and population [24].

The reliability of foot pressure platforms or mats for typically developing children and healthy adults has been previously established by Cousins et al. (2012) [33], Hafer et al. (2013) [16], and Niller et al. (2016) [17]. Other similar studies assessed likewise reliability for both typically developing children and children with CP, also using a plantar pressure mat [14,34].

However, the use of plantar pressure foot insoles presents with different benefits, such as the possibility of their use inside shoes or orthotic devices recording a higher number of gait cycles, as well as overall being easier to use with smaller children.

Our results show high reliability (ICC $\geq$ 0.60) for 21 of the 24 parameters that were tested. Still, three of the outcome measures for whole foot and forefoot showed lower values (whole foot contact time variable and peak pressure and contact time variables at the forefoot).

The number of participants included in this study was small, but similar to other researches [14,22]. However, because of the heterogeneity of children with CP, we opted to conduct a separate analysis of right and left feet. This increases the total sample to sixteen (feet). Post-hoc power analysis with α = 0.05 revealed good power ($\geq$0.90) for most variables, except for the three variables mentioned above. Post-hoc statistical analysis was carried out using R software (version 4.1.3., R Core Team 2022) [35] and the "ICC.Sample.Size" package (version 1.0.) [36].

The poor reliability results for the contact time variable (whole foot and forefoot region) may be explained by the heterogeneous gait pattern with which the participants presented. Most of our sample were children with unilateral CP, who present with a slower pace and abnormal weight shift between the affect side and less affected size. As a separate limb analysis was conducted, the diminished weight shift to the more affected side may have led to an increased contact time on the opposite side, and thus the contact time variable registered a wider range of values. Moreover, although we asked the children to walk at a self-selected comfortable pace, their pace varied.

The lower ICC values obtained from the forefoot peak pressure can be attributed to the slight discrepancy between the total foot length and the length of the available insole. Foot length across our sample ranges from 15 cm to 20 cm, but the same pair of 20 cm insoles was used throughout the investigation. This means that the fit was not always perfect, leaving vacant pressure cells at the top of the insoles, which can reflect in the forefoot values. Moreover, the total weight of the equipment was 0.5 kg, which may impact the trials of some of the smaller children and those with greater locomotion difficulties and gait deviations.

The *SEM* and *MDC* values were determined to quantify the amount of error associated with each variable in this population. Even though the *SEM* and *MDC* values for each variable showed a clinically acceptable level of error [20], they were transformed into a percentage for comparison purposes:

$$SEM\% = \frac{SEM}{Mean} \cdot 100$$

And

$$MDC\% = \frac{MDC}{Mean} \cdot 100$$

Please refer to Ayán-pérez, C. and Bouzas-rico, S. (2019) [37] for more information. For reference purposes, *MDC*% scores >30% were considered poor, from 10 to 30% were considered acceptable, and <10% were considered excellent [38]. The obtained values for *MDC*% were all considered to be poor, except for the contact area variable for the whole foot and peak pressure and contact area for the midfoot, which were within the acceptable range. These results are equivalent to other similar studies [37,39].

Various foot segmentation models have been reported in recent literature [7,13–15,17–19,21–24]. Complex masking usually involves anatomical and functional segmentation, including external references (for example, retroreflective markers and an optoelectronic system) that were not available for this specific study. Smaller areas of division may provide with less detailed information, and they are also more error-prone [17]. A three identical part division masking was selected for this study, similar to that of Galli et al. [7], allowing to differentiate force, pressure, and spatio-temporal values between the hind-foot, midfoot, and forefoot. Knowing that most participants presented an equinus gait pattern, we expected altered values in these three areas, and that division allowed the retrieval of more specific data.

The absence of previous reliability studies with this population and method precludes comparisons with similar *SEM* and *MDC* data. These preliminary results could prove useful to determine clinical changes in foot pressure and understand how those changes differentiate from the error of measurement. This is particularly important in studies where we have a pre- and post-assessment of the participant to see the effect of an intervention process. If the post results are superior to the reported error of the measurement, we can be confident in stating that there was a significant effect caused by the intervention.

5. Conclusions

This study is the first that establishes plantar pressure insoles as a reliable tool for measuring different gait-related variables in children with CP. The results indicate a good reliability for most variables, except for whole foot contact time and peak pressure and contact time at the forefoot. These lower values observed may be attributed to the heterogeneous gait pattern of children with CP and the above-mentioned equipment limitations of the study.

Author Contributions: Conceptualization, M.R.R., A.P.V. and F.J.; methodology, M.R.R., J.T. and F.J.; software, M.R.R.; validation, M.R.R., D.R. and F.J.; formal analysis M.R.R.; investigation, M.R.R.; resources, M.R.R. and F.J.; data curation, M.R.R.; writing—original draft preparation, M.R.R.; writing—review and editing, D.R., J.T. and F.J.; supervision, A.P.V.; project administration, A.P.V. and F.J.; funding acquisition, A.P.V. and F.J. All authors have read and agreed to the published version of the manuscript.

Funding: This work was supported by the Fundação para a Ciência e Tecnologia (FCT), through CIPER (unit 447) with the reference projects UIDB/00447/2020 and PTDC/EMD-EMD/5804/2020.

Institutional Review Board Statement: The protocol of this study was approved and executed in accordance with the Faculty of Human Kinetics Ethics Committee (CEFMH-2/2019).

Informed Consent Statement: Written informed consent was obtained from all participants involved in the study.

Data Availability Statement: The data presented in this study are available on request from the corresponding author. The data are not publicly available due to privacy issues.

Conflicts of Interest: The authors declare no conflict of interest.

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