EFFECTS OF EXERCISE INTERVENTION ON GAIT KINEMATICS AND LOWER LIMB FUNCTION OF ADOLESCENTS AND YOUNG ADULTS WITH CEREBRAL PALSY

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CP-vamma kulkee ihmisen mukana koko elämän ajan varhaislapsuudesta aikuisuuteen, sillä täysin parantavaa keinoa aivovaurion korjaamiseen ei ole löydetty. Ongelmat näyttäytyvät erityisesti liikkumisessa ja muussa motorisessa toiminnassa. Kasvun myötä cp-vamman oirekuva muuttuu, jonka myötä moniammatillisia kuntoutuskeinoja on kehitetty cp-vammaisten toimintakyvyn ja yleisen terveyden tukemiseksi. Liikunnallisen kuntoutuksen on todettu tutkimuksissa parantavan cp-vammaisten lihasvoimaa, kävelykykyä ja kestävyyskuntoa. Uusien cp-vammaisten liikuntasuositusten mukaisesti tehtävää kävelyharjoittelua ja voimaharjoittelua yhdistämällä ajatellaan saatavan hyötyjä kävelynopeudessa, kävelyn kinematiikassa ja kävelykyvyssä laajemmin kuin pelkästään yhden tyyppisellä harjoittelulla. Lisäksi 3D-kävelyanalyysin avulla voidaan suunnitella voima- ja kävelyharjoittelu yksilöidysti. Tämän tutkimuksen tarkoituksena on tarkastella kahdesta kolmeen viikkoharjoitusta sisältävän yksilöidyn kolmen kuukauden liikuntaintervention vaikutuksia alaraajojen kävelyn kinematiikkaan, voimatasoihin ja kävelysuorituskykyyn kolmella cp-vammaisella.

Kolme spastisen (hemiplegia tai diplegia, GMFCS-tasot I-III) cp-vamman (16-21 vuotta) omaavaa miestä osallistui tutkimukseen osana EXECP-tutkimusta. Kolmen kuukauden liikuntainterventio koostui kahdesta kolmeen valvotusta harjoituskerrasta viikossa. Kukin harjoituskerta sisälsi kävelyharjoittelua moottoroimattomalla kävelymatolla, voimaharjoittelua ja liikkuvuusharjoittelua tärkeimmille alaraajojen lihaksille. Mittausten 3D-liikeanalyysissä tutkittavat kävelivät kuudesti yhden minuutin mittauskävelyn minuutin palautusajalla. Alaraajojen 3D-kinematiikka mitattiin kahdeksan kameran liikkeenkaappausjärjestelmällä 200 Hz mittaustaajuudella, lisäksi samalla mitattiin säären lihasten lihasaktiivisuuksia langattomalla EMG:llä sekä kävelyn reaktiovoimia kahdella upotetulla voimalevyllä 1 kHz mittaustaajuudella. Kinematiikka analysoitiin Visual3D-ohjelmalla. Kuuden minuutin kävelytesti (6MWT) suoritettiin liikeanalyysimittausten jälkeen. Nilkan koukistaja- ja ojentajalihaksien voimantuottoa mitattiin nilkkadynamometrissa erillisenä testikertana. Kaikki mittaukset toistettiin ennen ja jälkeen intervention.

Tutkimuksen perusteella liikuntainterventio voi todennäköisesti kehittää kävelysuorituskykyä ja alaraajojen voimatasoja nuorilla CP-vammaisilla. Kuitenkaan kävelysuorituskyvyn positiiviset muutokset eivät kulje käsi kädessä kinematiikalla arvioidun kävelytekniikan muutoksien kanssa. Suurin osa vahingollisista nivelten kompensaatioista ja epäedullisista kävelymalleista toistuivat myös intervention jälkeen kävelyanalyysimittauksissa. Hemiplegia-tutkittavalla epäsymmetriaa vasemman ja oikean jalan välillä saatiin vähennettyä spatiotemporaalisten kävelymuuttujien ja isometristen voimamuuttujien osalta. Vain yksi tutkittava, diplegikko, pystyi parantamaan heikomman puolen varpaiden nostoa kävelysyklin pääteheilahdusvaiheessa. Säären lihasten aktivoitumisjärjestyksestä havaittiin kolmipäisen pohjelihaksen aktivoituvan kahdella tutkittavalla ennenaikaisesti jo heilahdusvaiheessa, jolloin aktiivisuus meni päällekkäin etummaisen säärilihaksen kanssa. Kävelysuorituskyky kehittyi kahdella tutkittavalla (5,8 ja 8,1 %). Intervention pituus ei mahdollisesti ollut riittävän pitkä aiheuttamaan muutoksia epäedullisissa kävelymalleissa, mutta toi silti muutoksia kävelysuorituskykyyn. Kävely on CP-vammaisilla hyvin yksilöllistä ja tämä tutkimus tarjoaa huomioon otettavia seikkoja CP-vammaisten nuorten harjoittelua ja kuntoutusta yksilöitäessä.

Asiasanat: CP-oireyhtymä, kävely, voimaharjoittelu, liikeanalyysi, kinematiikka

ABSTRACT

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Cerebral palsy (CP) affects individuals throughout their lifetime, usually introducing detrimental changes in ambulatory abilities. Various management strategies to support functional abilities and overall health in order to minimize the effects of the CP have been published. Several studies have shown positive results using different kinds of exercise therapy interventions to increase strength, motor activity or cardiovascular fitness. It is hypothesized that the intervention including both treadmill training and muscle strengthening will enhance walking speed, improve gait kinematics and ankle dorsiflexion. Also, evaluating lower limb functionality with gait analysis could improve the prescription of resistance training exercises for people with CP. The purpose of this study is to show does the three-month tailored exercise therapy intervention, consisting of two to three supervised sessions per week, provide benefits to lower-body gait kinematics, gait performance and lower limb function for three different CP case subjects.

A convenience sample of three male (16-21 years old) with spastic CP (hemiplegic or diplegic, GMFCS I-III) participated in the study. The twelve-week exercise therapy intervention consisted of two to three supervised sessions per week. Each training session started with gait training on a non-motorized incline treadmill with hands supported, was followed by strength and flexibility training for main lower limb muscles. In the gait analysis session, participants performed six times one-minute walking trials with one-minute rest between trials. Lower limb 3D kinematics were acquired with an eight-camera motion capture system at 200 Hz, and in addition to calf muscle wireless EMG, forces were simultaneously measured with two mounted force plates at 1 kHz sampling frequency. Kinematics were analyzed in Visual3D software. Six-minute walk test (6MWT) was performed after the gait analysis. Plantar and dorsiflexor force production was measured in custom-built ankle dynamometer. Measurement results PRE and POST intervention were compared.

Intervention may likely improve the gait performance and strength in adolescents and young adults with CP. However, improvements do not happen hand in hand with gait quality as mostly the same compensations and pathological gait patterns were present also after the intervention. The differences in spatiotemporal gait and isometric torque parameters between affected and non-affected limbs reduced after intervention in the hemiplegic participant. Toe lift of the more affected leg in the terminal swing was slightly improved in one diplegic case. While one diplegic participant had typical EMG onset pattern, triceps surae muscle activity started prematurely in terminal swing in the other two participants overlapping with TA activity. The distance walked in 6MWT improved in two participants (5.8 and 8.1%). This study provides more means and considerations to individualize the training or treatment for children with CP. The intervention period may not be long enough to induce changes on motor patterns and major gait deviations such as crouch gait but may improve gait performance. The underlying neuromechanical and cortical mechanisms should be studied to understand better the changes that are induced because of a training intervention.

Key words: Cerebral palsy, walking, strength training, gait analysis, kinematics

LIST OF COMMON ABBREVATIONS

6MWT	six-minute walk test
AFO	ankle-foot orthosis
AGLR	approximated generalized likelihood ratio
BoNT-A	botulinum toxin A
BWSTT	bodyweight supported treadmill training
CGA	computerized gait analysis
COG	center of gravity
CP	cerebral palsy
DOF	degrees of freedom
(s)EMG	(surface) electromyography
GCS	global (lab) coordinate system
GMFCS	Gross Motor Function Classification System
GMFM	Gross Motor Function Measure
(v)GRF	(vertical) ground reaction force
LCS	local coordinate system
MG	medial gastrocnemius muscle
NMES	neuromuscular electrical stimulation
RF	rectus femoris
SDR	selective dorsal rhizotomy
SEMLS	single-event multilevel surgery
SOL	soleus muscle
TA	tibialis anterior muscle
TD	typically developed
TKEO	Teager-Kaiser energy operator

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1 INTRODUCTION

Cerebral palsy (CP) affects individuals throughout their lifetime. There is currently no complete cure to prevent or ameliorate the neurodevelopmental impairments caused by the non-progressive brain injury during early development of the child. (Graham et al. 2016.) As the child grows, altered neuromuscular control will introduce detrimental changes in movement, muscle function and musculoskeletal anatomy generating high treatment costs through one's lifespan and making daily life more difficult for the individual and his/her close ones (Rosenbaum et al. 2007; Koop 2009).

A significant amount of research in physical therapy, biomechanics, and exercise science has tried to introduce optimal rehabilitation strategies to support functional abilities and overall health in order to minimize the effects of the disorder. The primary goal of the physical therapy is to improve one's ambulatory abilities (Dodd et al. 2002) because in children with CP weaker muscles have been found especially in lower extremities compared with their age-matched peers (Wiley & Damiano 1998; Mathewson & Lieber 2015). Several studies have shown significant positive results using different kinds of exercise therapy interventions to increase strength, motor activity and cardiovascular fitness (Dodd et al. 2002; Verschuren et al. 2007; Martin et al. 2010; Park & Kim 2014) and first exercise and physical activity recommendations specifically for CP were published by Verschuren et al. in 2016.

Every CP individual has their specific gait deviations, with differing degrees and combinations of impairments (Perry & Burnfield 2010; Graham et al. 2016). Individualized gait assessment can be done using computerized gait analysis (CGA) methods, such as 3D motion capture systems, accelerometry, force plates, and electromyography, to measure human limb muscle functions, kinematics, and forces. Determining the underlying causes of gait deviations, asymmetries and evaluating lower limb functionality could improve the prescription of resistance training exercises and rehabilitation for people with CP. (Armand et al. 2016; Williams et al. 2019.)

Task-specificity, how much one practices the criterion task, is one key element of effective therapeutic interventions for functional improvements. (Krishnan et al. 2019). It is hypothesized that the intervention including both treadmill training and muscle strengthening will enhance walking speed in six-minute walk test (6MWT) (Moreau et al. 2016; Booth et al. 2018), improve gait kinematics towards values of typically developed peers and increase dorsiflexion at initial contact combined wider ankle range of motion (ROM) during gait (Willersley-Olsen et al. 2015; Kirk et al. 2016). However, many studies have not reported the training intervention adequately, or the amount of exercise in the study did not meet current recommendations (Verschuren et al. 2016). Thus, there is an urgent need for more evidence-based research about training interventions in the CP population.

CP has diverse clinical manifestation, and thus analyzing individual responses is also important. No studies have yet researched effects of three-month exercise therapy intervention, featuring multiple different types of training, to lower-body gait kinematics in adolescents and young adults with CP. The purpose of this study is to show does the three-month tailored exercise therapy intervention, based on the Verschuren et al. (2016) recommendations, consisting of two to three supervised sessions per week provide benefits in lower-body gait kinematics and lower limb function for three different CP case subjects.

2 CEREBRAL PALSY

Cerebral palsy (CP) is an umbrella term for permanent disorders of the movement and posture that are caused by a non-progressive brain injury, lesion or other disturbance during the development of fetal, infant or child brain. These disturbances happen to the immature and developing brain before the affected function, such as walking has developed. (Rosenbaum et al. 2007). The clinical indicators of cerebral palsy vary significantly in the type of movement disorder, affected body parts, and the degree of functional ability (Graham et al. 2016). First descriptions of the cerebral palsy syndrome were provided by William J. Little in the mid-19th century. At that time, he identified premature birth and asphyxia neonatorum (respiratory failure in the new-born) as the main underlying factors for cerebral palsy. (Little 1862.) Nowadays, some of the recognized risk factors for CP are low birthweight, premature birth, infections during the fetal period, and multiple births (Odding et al. 2006). Therefore, classically the definition of CP excludes movement and posture disorders that are of short duration, due to progressive disease, or solely because of mental deficiency (Bax 1964). Usually, in addition to weakened movement and posture control, symptoms of CP can include other neurodevelopmental impairments such as problems in sensation, perception, or cognition (Rosenbaum et al. 2007).

As there is currently no cure for CP it will persist through one's lifespan (Rosenbaum et al. 2007) but even without a cure, almost all children with CP survive to adulthood (Graham et al. 2016). Although the brain injury is non-progressive, the functional abilities and weaknesses can progress negatively during maturation for example due to spasticity (involuntarily increased muscle tone) and contractures (i.e., decreased joint range of motion in limbs caused by abnormal muscle/tendon shortening). At birth, children with cerebral palsy have the muscles and bones like those typically developed children without the disorder. During the growth of a child with cerebral palsy altered neuromuscular control leads to altered muscle function and, in the end, to altered musculoskeletal anatomy influencing the development of the skeleton. (Koop 2009.) In addition to spasticity, impaired selective motor control, poor coordination, deficits in sensory function and muscle weakness account greatly for disability and should be taken better into account in management strategies (Graham et al.

2016). Therefore, physical therapy or medical operations are possibly needed multiple times during growth and maturation of children with CP (Bell et al. 2002). The physical and psychological care of children with cerebral palsy is directed towards reducing the effects of the factors making the condition worse and maximizing function and participation in activities (Graham et al. 2016).

Cerebral palsy is one of the most common motor disabilities in children population. In Europe for every 1000 children born the prevalence of CP was ranging around 1,5-2,5 (Surveillance of Cerebral Palsy in Europe 2002) as in the United States of America the prevalence was found somewhat higher, 3,1 per 1000 children (Christensen et al. 2014). Odding et al. (2006) showed that the prevalence of CP has risen above 2,0 per 1000 during the last 40 years. One of the reasons behind the increase in prevalence could be the increased survival of very low birth weight infants due to developed new-born intensive care (Paneth et al. 2006).

ICD-10, the most used classification system in Finland, categorizes the disorder into both the topographical and spasticity characteristics (Pihko et al. 2014a). Most of the children with CP had spastic CP (77,4%), which can be divided to bilateral spastic CP (e.g., diplegic) (63,6%) and unilateral spastic CP (e.g., hemiplegic) (36,4%) (Christensen et al. 2014). In Europe, 85,7% of the children with CP was diagnosed with spastic CP. Also, 58,2% of children with CP could walk independently without support, 11.3% needed to use a hand-held mobility device during walking, and 30.6% had only limited or no walking ability at all. (SCPE 2002). Figure 1 shows the typical areas affected by different types of unilateral and bilateral cerebral palsy. Usually, the lower limbs are more affected than upper limbs. For example, in diplegia, the upper limbs usually show only fine motor impairment. However, in hemiplegia, it is more common to have upper limb as more affected. (Graham et al. 2016.)

As mentioned earlier, CP is many times accompanied by other co-occurring impairments such as epilepsy or musculoskeletal problems. Additional impairments are seen in 25-80% people with CP depending on the subgroup of CP. For example, 41% of children with CP also had co-occurring epilepsy. (Christensen et al. 2014), most commonly among the hemi- and

tetraplegics (Odding et al. 2006). The sensibility of the hands is impaired in about half of the CP population and up to 80% have at least some speech impairment (Odding et al. 2006).

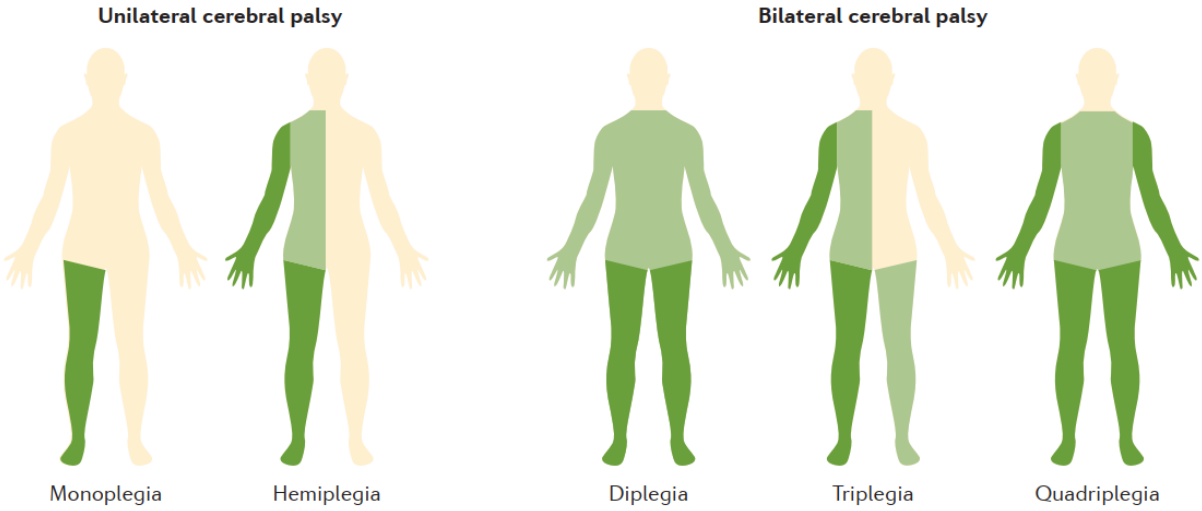

FIGURE 1. Affected areas in different types of cerebral palsy, as per topographical classification. The color shade tells the severity of impairment, darker areas more affected. Prefix word (e.g., mono and di) states the impairment volume, and root word tells the type of impairment (plegia means paralyzed, and paresis means weakened). (Graham et al. 2016.)

2.1 Gross Motor Function Classification System

The functional capabilities of the people with CP are greatly varied because of the heterogeneous characteristics of impairments due to early childhood brain injury. For example, the location of symptoms or number of impaired limbs can vary from one CP individual to another. One of the most used and validated classifications for the ability to function and motor performance of people with CP is Gross Motor Function Classification System (GMFCS) (figure 2). Level I indicates the best level of mobility and Level V, the lowest level of mobility, a situation where you rely on others help for mobility. (Rosenbaum et al. 2007; Damiano et al. 2009.)

The GMFCS divides the individuals with CP into five different levels depending on one's activity limitation and functional mobility. There are different descriptions for children aged

6-12 and 12-18 years. (Burns et al. 2014). As the GMFCS covers mostly the lower limb ambulatory function, there have been developed other classifications for evaluating bimanual hand and arm function such as the Manual Ability Classification System (MACS). (Rosenbaum et al. 2007). GMFCS status usually remains the same in the transition from childhood to adult age, but it may change in response to improvements due to interventions or deterioration consequent of the natural history of the disease (Burns et al. 2014).

The impairments set difficulties for people with CP to deal with everyday life, and decreasing activity levels could lead to lower muscle strength levels in people with CP. Also, rapid force generation is impaired in cerebral palsy, possibly relating to decreased muscle size and activity levels (Moreau et al. 2011). The activity levels of children with CP are directly proportional to their GMFCS Level, but still people with Level I CP have activity levels which are much less than their healthy counterparts (Bjornson et al. 2007). Especially in GMFCS levels IV-V opportunities for performing activities should be emphasized as they are in the weakest position. Also, overall weaker muscles in lower extremities have been identified in children with CP compared with their age-matched peers (Wiley & Damiano 1998). That is why the primary goal of physical therapy is to improve one's ambulatory abilities (Dodd et al. 2002). Aside from physical therapy, currently the care for the CP includes many times multiple medical and surgical interventions to handle spasticity and contractures, especially during childhood (Damiano et al. 2009). Developing and studying effective ways to improve CP patients' walking and other functional abilities would help their everyday life and increase their overall health. In this study, we researched youth with CP in GMFCS level I and III.

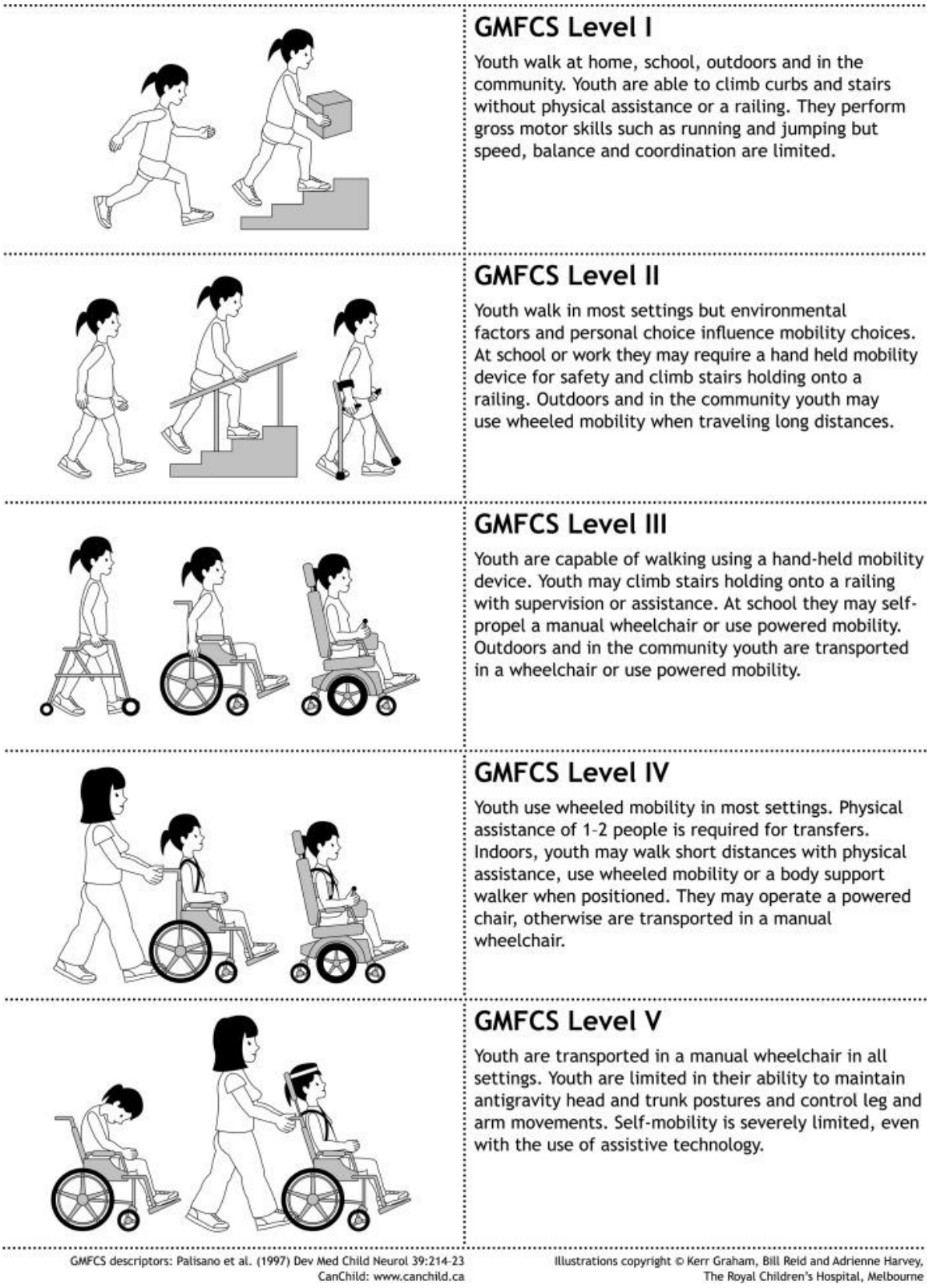

FIGURE 2. Gross Motor Function Classification System (GMFCS) for children aged 12-18 years (Burns et al. 2014).

2.2 Physiologic classification

Apart from using GMFCS to evaluate the ability to function and motor performance of people with CP or topographical distribution of impairment to understand affected body parts (figure 1), there are also other CP classification systems which approach the condition from different perspectives. Cerebral palsy can be divided based on the physiological motor function to two main groups, spastic CP (pyramidal) and non-spastic CP (extrapyramidal). Spastic CP and its different variations consist of 78-85% all CP population (SCPE 2002; Christensen et al. 2014) and is clinically characterized by increased muscle tone (i.e. spastic hypertonus) many times resulting in very stiff limbs (Pakula et al. 2009) and extensor plantar response (Gulati & Sondhi 2017). In spasticity, the muscles overreact and increase muscle activity due to the disorder of velocity-dependent stretch reflex (Graham et al. 2016). More broadly, spasticity is suggested by SPASM consortium to be defined as "disordered sensorimotor control, resulting from an upper motor neuron lesion, presenting as intermittent or sustained involuntary activation of muscles" (Pandyan et al. 2005). The upper motor neuron lesion results predominantly loss of corticospinal tract connections to lower motor neurons and therefore to skeletal muscles but may also induce loss of inhibitory descending input to the lower motor neurons (Graham et al. 2016). The ambiguity of the spasticity definition in much of the research has caused some discussion on how to correctly define and measure spasticity with reliable and valid methods because lack of correspondence between definition and measurement methods can compromise the internal validity of the research. Combining biomechanical measures with simultaneous muscle activity monitoring and controlling environmental conditions and time of testing is recommended. (Malhotra et al. 2009). Distinction from rigidity (hypertonia at low movement speeds) is important as in spasticity hypertonia increases with the increasing speed of muscle stretch or joint motion beyond a specific critical angle (Bonow et al. 2018, 753). Spastic CP is sometimes called as pyramidal CP as it is commonly connected to defects or damage occurring in the brain's corticospinal pathways and white matter. (figure 3; Jones et al. 2007; Eunson et al. 2016). Pyramidal tract (corticospinal tract) starts in the motor cortex, and upper motor neurons of the corticospinal tract are the most important pathways for voluntary motor function (Gilroy et al. 2013, 641).

Non-spastic CP includes two different CP groups (dyskinetic and ataxic), which are the second most prevalent CP types after spastic CP (dyskinetic 6,5% and ataxic 4,3%) (SCPE 2002). In addition to that, there appears to be a small percentage of people with mixed forms CP (e.g., combination of spasticity with dyskinesia), where no single tone abnormality or movement disorder predominate. Different forms of non-spastic CP are characterized by particular impairments, but common features are involuntary movement and reduced muscle tone (Pakula et al. 2009; Wimalasundera & Stevenson 2016). Dyskinetic CP description includes recurring, uncontrolled, and involuntary movements, but tone abnormality varies in dystonic and choreoathetotic variants of dyskinetic CP (Wimalasundera & Stevenson 2016). Non-spastic CP is commonly seen after damage to neurons in the extrapyramidal system, i.e., basal ganglia, thalamus or the cerebellum. (Bax et al. 2006; Jones et al. 2007). Extrapyramidal system is a part of the motor system network causing involuntary actions. Consists of motor-modulation systems outside the corticospinal tract, the basal ganglia, and cerebellum. This tract controls muscle tone and balance reflexes. (Gilroy et al. 2013, 641). Ataxic CP has been associated with damage to neurons in the cerebellum, seen as oral motor difficulties, balance, and depth perception problems (Jones et al. 2007).

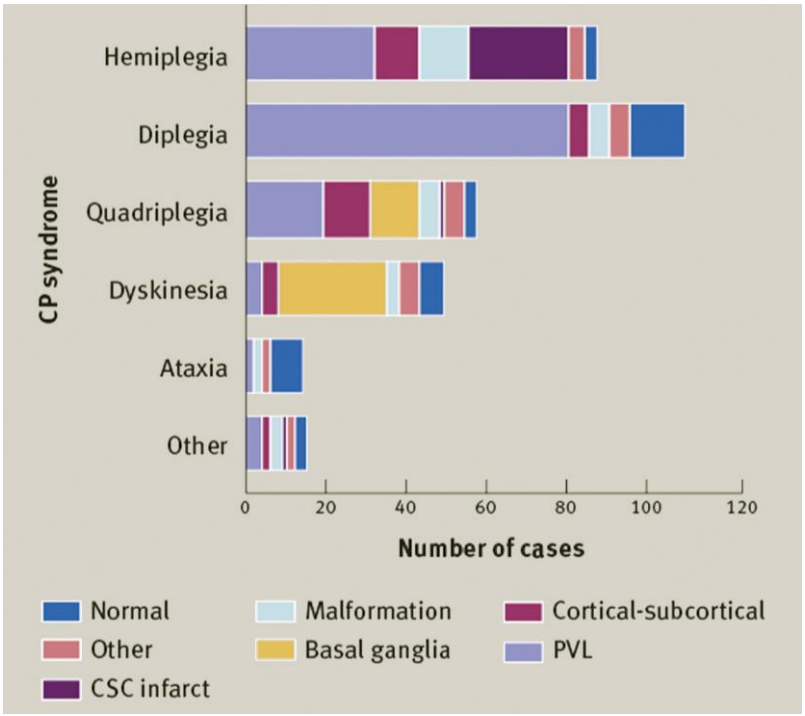

FIGURE 3. Periventricular leukomalacia (PVL) is one type of white matter damage of immaturity and was the most common MRI finding in spastic CP (Eunson et al. 2016).

2.3 Treatment for spastic CP

Although there is no definitive cure for CP, rehabilitation and medical care can minimize the effects during child- and adulthood. Also, in recent years there has been progress in the means of prevention and amelioration of the brain injury. For example, fetal exposure to magnesium sulfate in mothers at risk for preterm delivery reduces the risk of developing cerebral palsy (Constantine & Weiner 2009). Another important recent finding in reducing mortality and prevalence of cerebral palsy was the application of head or body cooling for 72 hours in newborns who were diagnosed with hypoxic-ischaemic encephalopathy (Jacobs et al. 2013). After infancy, treatment for CP focuses on two themes: surgical treatment and non-surgical management, including physical therapy, tone-reducing drugs, or botulinum injections.

It has been stated that central nervous system injury is resulting in cerebral palsy, but clinical symptoms are observed in the peripheral neuromuscular system, especially in skeletal muscles (Graham et al. 2016). The cumulative effect of spasticity causes problems in muscles and tendons with time. As children with CP grow older, despite best clinical practices, they often develop contractures, which are defined as limited joint range of motion caused by high passive muscle force and morphological changes, e.g. shortening of the tissue. Contractures are one of the main functional complications of cerebral palsy. (Bonow et al. 2018.) Muscle spasticity is still thought to be the main reason leading to muscle contractures (Smith et al. 2011; Graham et al. 2016), but the development of contractures is likely related to more factors (Smith et al. 2011; Gough & Shortland 2012). Tedroff et al. (2009) have shown in their study of long-term effects of botulinum toxin A (BoNT-A) treatment in children with cerebral palsy that contractures can develop without spasticity.

Secondly, review by Gough & Shortland (2012) showed the effects of muscle growth impairment and physical inactivity to muscle deformities in children with CP. Muscle tissue is stiffer in contracture compared to age-matched children, but titin and individual fiber stiffnesses are unaltered. Adaptations of the extracellular matrix are more probable reasons behind contractures than changes in the muscle cell itself. The increased collagen content of the muscle and in vivo sarcomere length was found to increase the stiffness of the contracture

tissue. (Smith et al. 2011). Muscle fascicle lengths can be the same as in typically developed (TD) peers, but sarcomere lengths have been found up to almost twice the length of the normal in different lower limb muscles. This puts TD and CP in very different parts of the theoretical sarcomere length-tension curve (figure 4), which could be one of the reasons force production capacity is impaired in CP. (Mathewson et al. 2015.) Also, the muscles' reduced satellite cell quantities in CP has been proposed to lessen the longitudinal growth of muscles and lead to decreased muscle strength and developing fixed contractures. (Smith et al. 2013). The clinical management on tackling these functional challenges in children with CP always includes physiotherapy, and often surgeries and BoNT-A therapy (Graham et al. 2016).

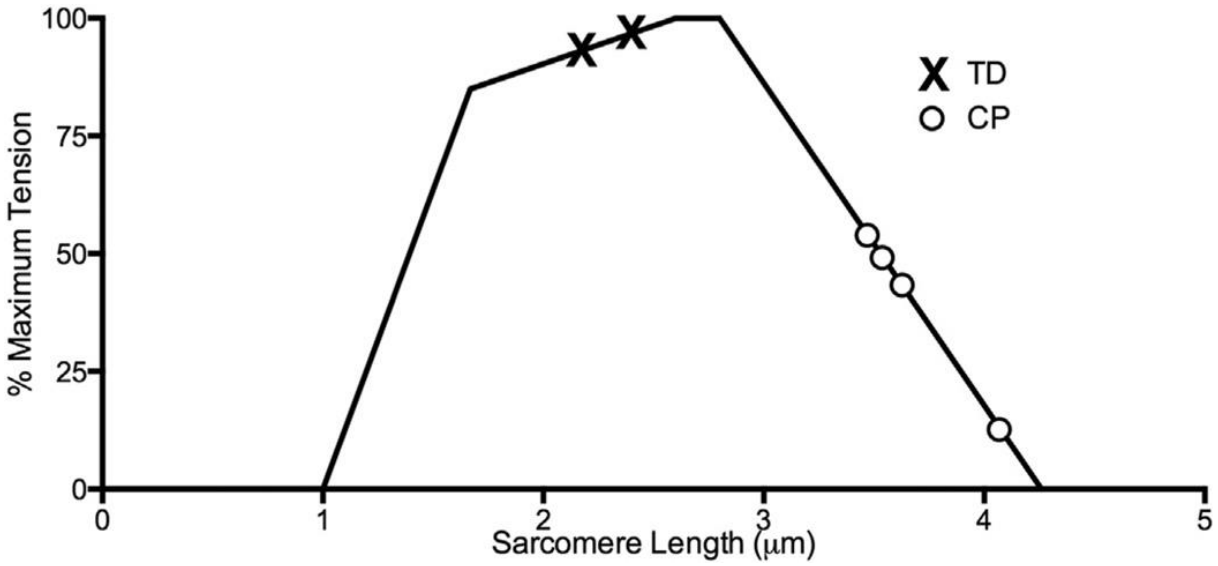

FIGURE 4. The schematic length-tension curve of skeletal muscle comparing TD and CP sarcomere length data (Mathewson & Lieber 2015).

2.3.1 Physiotherapy

Physiotherapy is the central element of multi-professional clinical management of children with cerebral palsy, combining different modalities to enhance the functional motor activity, motor coordination, and fitness for improved daily functioning (Graham et al. 2016). Physiotherapy for children with CP works many times in conjunction with occupational therapy, especially when there is a need for improving hand function. Occupational therapy

concentrates on fostering independence by developing an individual's abilities to manage daily living, education, and work. (Gulati & Sondhi 2017). A lot of different interventions have been investigated over the decades, and it is concluded that effective physical therapy interventions should reflect current neuroscience knowledge (Novak et al. 2013) as electrophysiological studies have shown that both somatosensory and motor systems are impaired in CP (Pihko et al. 2014b; Willerslev-Olsen 2015). Nowadays, evidence recommends two types of activity-based physical therapy interventions: task-specific skill training and physical training.

Task-specific skill training aims to develop children's motor coordination and performance, and physical training addresses secondary impairments, for example, muscle weaknesses and decreased cardiorespiratory fitness (Graham et al. 2016). Improving fitness in children with CP is important as even the most functional children with this impairment are weaker and have lower cardiorespiratory fitness compared with their TD peers. (Wiley & Damiano 1998; Balemans et al. 2013). This forces people with cerebral palsy to work close to their maximal capacity, already in normal daily activities like walking. Task-specific skill training is done using constraint-induced movement therapy, which combines restraint of the unaffected limb to encourage the intensive use of affected limb when going through therapeutic tasks (Graham et al. 2016.) For example, restraints can be created using casting or physically restraining the normal hand or leg by holding it in place. (Gulati & Sondhi 2017). Other effective possibilities for activity-based interventions depending on the goals are, for example, bimanual training, context-focused therapy using changing task and environment characteristics, and home programs. Virtual reality methods, for example, built virtual environments, have been proven effective in improving motor function and engagement in therapy. Thus, virtual reality probably will be incorporated into more training interventions in the future. (Novak et al. 2013; Chen et al. 2018.)

2.3.2 Surgery

Although physiotherapy and medical management are a vital part of the care instantly after the diagnosis at a young age, growing children with spastic CP will frequently require surgical

therapy to correct deformities such as contractures or reduce spasticity through selective dorsal rhizotomy neurosurgical operation (Gulati & Sondhi 2017). Orthopedic surgery operations for contractures include, for example, tendon-lengthening or tendon transfers to improve range of motion (Bonow et al. 2018). Functional improvements are seen after surgery for ankle equinus (i.e., impaired ankle dorsiflexion), hip flexion contracture, knee flexion contracture and rectus femoris (RF) (stiff knee gait due to RF spasticity). Even though surgical operations usually have good outcomes, surgical manipulation of the muscle-tendon unit is shown to alter muscle sarcomere length and change force production capacity of the muscle. Muscle's sarcomere length was found dramatically longer in children with CP (after tendon lengthening surgery for ankle equinus) compared with TD children when the muscle fascicle length was similar. Reduced serial sarcomere number and increased sarcomere length are probable contributors to muscle weakness in people with CP. (Mathewson et al. 2015.)

Aside from tendon-lengthening surgeries, osteotomies may be required to correct bone alignment (i.e., hip disposition). Applying computerized gait analysis (CGA) in surgical decision making has improved functional outcome evaluation and postoperative outcomes greatly. (Filho et al. 2008; Aversano et al. 2017.) Although having promising studies, predicting the effects of surgery on gait is still challenging, and there is a need for better validated predictive models (Khouri & Desailly et al. 2017). Normally orthopedic surgery for contractures are done only after six years of age as the overall picture of the movement disorder (dystonic and spastic contributions), and surgical outcomes are then more predictable, but also CGA can then be utilized (Graham et al. 2016; Bonow et al. 2018). However, hip displacements are an exception which should be treated already before that age. Bony surgeries usually do not need to be repeated as the case is with tendon lengthening during the child's growth. To avoid yearly surgeries and constant recovery and rehabilitation from them, an approach named single-event multilevel surgery (SEMLS) is recommended. In SEMLS, multiple procedures (i.e., bony surgery, soft tissue lengthening) are conducted in one surgery. (Lynn et al. 2009.) When conducting multiple procedures in one surgery, CGA is an important tool to guide the development of multilevel surgery plan as studies have shown that up to 80% of plans are changed after CGA data acquisition (Khouri & Desailly 2017). Additionally, the positive surgical and functional outcomes are tied to the extent to which

CGA recommendations were followed in the treatment plan (Filho et al. 2008; Wren et al. 2013).

Selective dorsal rhizotomy (SDR) is a neurosurgical procedure that is used to ease the spasticity. Reducing spasticity without causing paralysis is acquired by selective transection of lumbar and sacral sensory nerve roots of the spinal cord. Nowadays, the extent of required nerve root sectioning is defined by using both physiologic information and electrophysiologic responses to rootlet stimulation. People with spastic diplegia have improved their gait, balance and motor function after SDR operation but the patient should have adequate underlying strength levels to ambulate once the tone is removed. (Lynn et al. 2009; Rumberg et al. 2016; Bonow et al. 2018.) It should be noted that any effect by different treatment methods on tone is temporary. SDR can produce a durable reduction in spasticity for over ten years, but improvement permanence in ROM or motor function depends on the GMFCS level. (Ailon et al. 2015).

2.3.3 Botulinum toxin A

Aside from SDR, intramuscular injections of botulinum toxin A (BoNT-A) is routinely used as a local way of managing spasticity. BoNT-A has been a standard treatment method for hypertonia in CP longer than 20 years, and for most children, it is an effective and safe intervention (Novak et al. 2013; Paget et al. 2018). When BoNT-A is injected into affected muscles, it blocks the presynaptic release of acetylcholine from motor endplates of the lower motor neuron, producing chemodenervation. By limiting muscle contraction, the tone decrease is immediate (Gulati & Sondhi 2017), and it is also an effective way to induce a reduction of long-term spasticity. This would possibly offer a rehabilitation period for developing better movement patterns and economy of gait. Although repeated BoNT-A injections are reducing muscle tone over a longer period, it is not preventing the development of contractures or sustaining improvements ROM in spastic muscles the same way. (Tedroff et al. 2009). Also, the emphasis on the treatment of children with CP has changed from focusing the impairments towards promoting activity and participation in everyday life. BoNT-A reduces long-term spasticity, but the spasticity reduction has not meant

improvements in gait or function, and improvements in passive ankle dorsiflexion are short-lived. (Löwing et al. 2017). Thus, more sustainable and preferable way of treating these problems in children with CP would be strengthening weaker muscles through targeted resistance training rather than correcting imbalances by blocking or weakening the stronger muscle with, for example, BoNT-A injections (Graham et al. 2016). Concurrent use of BoNT-A and strength training or goal-directed physiotherapy has shown better results in both reducing spasticity and improving strength and functional measures than BoNT-A treatment alone (Löwing et al. 2017; Fonseca et al. 2018). Strength training should also be targeted for muscles which are injected with BoNT-A (Williams et al. 2012).

Spasticity is also managed with tone-reducing, systemic oral medication such as baclofen or diazepam which target generalized or regional spasticity. These tone-reducing drugs inhibit reflexes that lead to increased tone by binding to receptors in spinal cord. Diazepam has been shown to be effective medication for spasticity management, however it is not widely used for long-term management. Oral baclofen has not as rigid evidence-based justification while it is still generally considered as first line drug. (Novak et al. 2013.) Baclofen can be delivered also intrathecally directly to its site of effect on spinal cord inhibitory pathways. Intrathecal baclofen is favored in non-ambulatory CP in GMFCS level IV and V. (Hurwitz et al. 2014.)

2.4 Training interventions

Several studies and systematic reviews have shown significant positive results using different kinds of exercise therapy interventions to increase strength, motor activity and cardiovascular fitness in children with CP (Dodd et al. 2002; Martin et al. 2010; Park & Kim 2014; Verschuren et al. 2016). Children with CP require exercise therapy because of functional deficits in force production and range of motion in addition to alterations of muscle at a structural level, such as having less muscle mass, smaller-diameter fibers and stretched sarcomeres compared with TD children (Mathewson & Lieber 2015). For example, tibialis anterior muscle size has been linked to fast gait velocity and required ankle dorsiflexion for foot clearance during gait (Bland et al. 2011). Muscle weaknesses are one of the characteristics of people with CP that can hinder the possibility to move freely during the day

and increase daily physical activity levels. For some time now exercise programs including more intense training such as strength training with free weights and weight machines or aerobic training using treadmills have been utilized to maximize the functional capabilities of people with CP. (Dodd et al. 2002; Damiano et al. 2007.) Although exercise programs might not lessen the muscle tone abnormalities (i.e, spasticity) or developing contractures which need to be corrected with neurosurgical operations, exercise can make positive changes in muscle strength or aerobic capacity which could lead to better ambulatory abilities. (Damiano et al. 2007). In this study, the effects of versatile exercise intervention in the gait of adolescents with CP are investigated. Thus, exploring the effects of different types of training interventions on functional abilities are needed.

2.4.1 Resistance training

Dodd et al. (2002) suggested in their review article that strength training can improve strength and motor activity of people with CP without having negative effects. Current position statement paper on school-aged youth resistance training recommends performing 1-3 sets of 6-15 repetitions on a variety of upper- and lower-body strengthening exercises, 2-3 times per week in the beginning. Exercises should be performed at an intensity of 50-85% of one-repetition maximum (1 RM), first starting with lower intensity, and resistance should be increased gradually as strength improves. At least eight weeks but preferably 12 weeks of training is required to observe an increase in muscle strength. (Faigenbaum et al. 2009.)

The first cerebral palsy specific physical activity and exercise recommendations were presented by Verschuren et al. in 2016. These recommendations are largely consistent with the guidelines for typically developed children (Faigenbaum et al. 2009), i.e. training frequency 2-3 times per week. Because one-repetition maximum testing in CP population can often be challenging or unsafe, assigning exercise intensity by increasing loading progressively within a prescribed range of repetitions (i.e., 8-12) is recommended. Many times, people with CP need more time to adapt to strenuous level resistance training and hence training familiarization period of 2-4 weeks helps them to reach the recommended training intensities and volumes. Thus, interventions or training programs for people with CP

should be designed to last longer (e.g., 12-16 weeks) to observe meaningful increases in muscle strength. For weaker individuals and at the beginning of the training phase (e.g., familiarization) simple, single-joint exercises are suggested and later add more complex, multi-joint exercises through gradual progressions. Still, it should be noted that single-joint exercises can be difficult for people with CP lacking the sensitive motor control needed to isolate and perform given single-joint motion, for example, in the ankle joint. (Verschuren et al. 2016.)

Walking ability has been shown to be related to muscle strength in children with CP. Muscle weakness was the most noticeable in muscle groups around the ankle, which contribute to plantar- and dorsiflexion during gait, and in the hip area. Problems in plantar- and dorsiflexion force production can negatively affect many fundamentals of normal gait, such as stance stability, push-off propulsion, foot clearance in swing, adequate step length, and conservation of energy. (Eek & Beckung 2008.) Gillett et al. (2018) suggested that maximum isometric plantarflexion strength in CP could be one of the most important independent variables explaining the variance in the distance walked on the 6MWT. One major component contributing to gait inefficiency in CP in addition to muscle weakness is erroneous co-contraction of antagonistic muscles (Unnithan et al. 1996).

Co-contraction has been found to decrease after 6-week eccentric strength training, but this covered only upper-limbs (Reid et al. 2010). Developing greater awareness of voluntary control by EMG biofeedback training consisting of lower extremity exercises with trials of contracting tibialis anterior and relaxing the spastic triceps surae muscles increased gait velocity and stride length significantly in three months. (Dursun et al. 2004). In another study, Eek et al. (2008) had 16 children with spastic CP (GMFCS I-II) doing strength training exercises for lower limbs using free weights, rubber bands and body weight three times a week for eight weeks. Participants started with strength levels below normal, especially at the ankle and hip muscles. Muscle strength and gross motor function measured with Gross Motor Function Measure (GMFM) were better after training. Eek et al. (2008) also used 3D gait analysis successfully to individualize strength training to concentrate on the weakest muscles and gait abnormalities found in the analysis. Notably, gait analysis proved an increased power produced at a push off, although gait velocity did not increase. Eek et al. (2008), Damiano et

al. (2010), Taylor et al. (2013), and Kirk et al. (2016) are the few studies looking on gait kinematics changes after strength training programs. Even though muscle strength has increased, there have not been significant changes in gait kinematics, except for Kirk et al. (2016), in which explosive resistance training increased the toe lift during swing phase. However, sample sizes have been only moderate at its best, and more studies are needed in the topic.

Walking velocity did not increase in Eek et al. (2008) study, regardless of muscle strength increases in hip muscles and power growth in the push off. However, Morton et al. (2005) noticed increased walking velocities after 6-week progressive resistance training of quadriceps and hamstring muscles. Testing showed improved isometric muscle strength and increased walking velocity and step rate in the 10-meter walking test and retaining changes at four-week follow-up. Increases in both walking velocity and step rate in Morton et al. (2005) were almost opposite results compared with Eek et al. (2008).

Gross motor function and spasticity are thought to be inversely related, as higher the spasticity the lower the functional abilities. Ross & Engsberg (2007) noticed that maximum strength levels in the ankle, knee, and hip were significantly more accounted for gait variance than the spasticity of the children with spastic diplegia CP. Regardless of the gait analysis done with or without assistive devices, strength was better related to function than spasticity was to function, measured with GMFM. Strength was also highly related to stride length, moderately related to gait speed, pelvic tilt ROM, and knee flexion at initial contact. Results could probably vary depending on the severity of subjects' spasticity, and one needs to take caution when drawing clinical implications based on the results of a regression analysis. (Ross & Engsberg 2007.) Results like this could emphasize the role of muscle strength development in interventions for people with CP.

The effects of resistance training to improve functional abilities such as walking function in children and adolescents with CP is still disputable by current evidence. In some studies, there may have been positive changes in muscle strength, but no significant changes in gait ability were found. Scholtes et al. (2012) using functional progressive resistance exercise training for

12 weeks, including leg press and functional exercises (sit-to-stand, lateral step-up, half knee-rise) with weight vest loading found significant effects in total isometric strength and leg-press strength compared with usual care. However, they did not find a carry-over effect on walking ability (i.e., walking speed, cadence, or step length). (Scholtes et al. 2012). For example, Boyd (2012) and Damiano et al. (2010) point out that strength training should be task-specific and intense enough to offer an effective way to improve gait ability. In Scholtes et al. (2012) study, the intensity was probably enough, but it can be argued if the training was specific enough when thinking about the outcome measures.

Gym training in weight machines will not transfer to better walking ability, especially when talking quite well capable Level 1 CP patients. Instead, children with spastic CP, who had a GMFCS level of II or III may benefit from getting stronger lower extremities to help their ambulatory capabilities (Hoffman et al. 2018). Independent walkers on GMFCS I level might have sufficient muscle strength reserve for walking, at which level further increases will not provide additional increases in walking ability, especially when measured with rather short walking tests. Also, children with CP on GMFCS I have more difficulties with coordination and motor planning than muscle strength sufficiency for walking. Thus, training in a context-specific manner would possibly enhance motor learning and functional performance more than training with resistance in non-functional tasks. (Damiano et al. 2010; Boyd 2012.) As an example, Peungsuwan et al. (2017) used combined strength and endurance exercise training containing everyday like functional movements to result in significant improvements in walking ability, balance, and functional lower limb strength within eight weeks of training. Williams et al. (2019) and van Vulpen et al. (2017) have suggested strength training with high movement velocity (e.g., power training) would be more effective for improving walking than traditional resistance training with relatively high loads and slow movements because sufficient power generation is vital for walking. Also, the energetics of step-to-step transitions in gait suggest that rehabilitative training should not focus only on strength needs but also abilities to produce mechanical power with the proper timing (Kuo & Donelan 2010). This training type would fill all factors in the concept of specificity in resistance training: target muscle groups, range of motion, speed, and muscle actions are specific enough to the movement. Correctly prescribed and modified ballistic resistance exercises may match the angular velocities occurring in the lower limbs during walking, but the exercises might suit

only for mildly impaired people. (Williams et al. 2019.) This concept of specificity of resistance training considers factors such as (1) the muscle groups that are targeted, (2) the range of motion through which the movement is performed, (3) the speed of movement, and (4) the muscle actions involved.

Effectiveness of strengthening interventions are especially seen in individual muscle level (Park & Kim 2014), and some improvement is also seen in GMFM (Eek et al. 2008) but the similar effect has not found consistently in gait parameters such as gait speed (Moreau et al. 2016). The probable reason behind the absent of carryover to functional activities could be the short intervention periods in many studies. Many studies also may have failed to report the volume, intensity, and progression of exercise prescribed, reported participant's normal physical activity vaguely, or the amount of exercise in the study did not meet current recommendations for resistance training for people with CP (Verschuren 2016). Diversity in the type of training (e.g., home, school, or gym-based, and functional or machine training programs) might also explain lack of effect seen after training interventions in children with CP (Scholtes et al. 2012). Also, poor quality of available evidence and small sample sizes might explain the negative results in a recent systematic review of randomized controlled trials assessing the effects of exercise interventions. Resistance training appeared to improve muscle strength and walking endurance but did not improve motor function, gait speed, or physical activity levels. Apart from just strengthening muscles resistance training is usually meant to improve activity capacity by reducing activity limitations. These results do not support the view that impairment-based interventions would improve activity. (Ryan et al. 2017.) Especially long-term effectiveness of resistance training is yet poorly investigated. The longest found trial by Verschuren et al. (2007) evaluated the effects of an 8-month training including anaerobic and aerobic as well as muscle-strengthening exercises in a circuit. Functional muscle strength measured with lateral step-up and sit-stand 30-second repetition maximums showed significant positive results after the intervention.

2.4.2 Stretching

Stretching has been an integral part of therapy programs in CP populations as muscle contractures cause loss of joint range of motion and stretching is thought to help in preserving the range of motion and perhaps even preventing surgical operations. Preserving the mobility of the joints would help daily function and limb positioning. (Wiart et al. 2008; Damiano et al. 2009.) In 2008 Wiart et al. showed that there is not enough evidence about the effectiveness of passive or active stretching in children with CP. Later Katalinic et al. (2011) and Novak et al. (2013) have concluded in their reviews that regular stretching does not produce clinically significant long-term changes in joint mobility, pain, spasticity, or physical activity levels. Theis et al. (2015) identified an increase in maximal passive dorsiflexion and decrease in ankle joint stiffness after six-week passive stretching in children with CP. Alterations in the mechanical properties of muscle were found as muscle and fascicle strain increased, but not in the tendon. Still, Harvey et al. (2017) concluded that stretching is probably not effective intervention to prevent contractures or provide short-term positive effects in pain or quality of life when looking at randomized controlled trials and controlled clinical trials. Orthopedic surgery is many times needed to lengthen tendons after contractures have developed and impaired function (Damiano et al. 2009). Stretching cannot probably be effective alone, and thus, Wiart et al. (2008) recommend interventions accompanying stretching to strength training or other more functional activities for flexibility purposes. Combining stretching with electrical stimulation might have marginal positive effects on spasticity and contractures (Khalili & Hajihassanie 2008). Also, innovative ways to combine traditional passive stretching and active movement training by robotic rehabilitation have shown promising results in ankle mobility (Wu et al. 2011).

2.4.3 Aerobic and gait training

Walking speed is a great indicator for functional abilities and quality of life in different populations (Fritz & Lusardi 2009). Therefore, improving walking speed and diminishing the effect of other gait abnormalities should be one of the main focuses of training interventions in CP. Usage of aerobic exercise as a training method for children with CP has slowly

increased after concerns for the possible detrimental effects of intense training in people with disabilities have been proven as partly groundless fear. Strength training interventions in CP are many times impairment-based, but it should be noted that task-specificity is one key element of effective therapeutic interventions for functional improvements. Specificity, how much one practices the criterion task, is more important for learning new gait patterns than difficulty progressions and variations in the practiced tasks (Krishnan et al. 2019). For example, Booth et al. (2018) found gait training improving walking speed more than standard physical therapy. Moreau et al. (2016) showed that gait training is a more effective way to improve gait speed than strength training alone. Even though the intensity in strength training was enough, the improvements in muscular strength were not transferring as positive changes in gait speed.

Furthermore, significant increases in aerobic fitness parameters have been found in the CP population after aerobic training interventions. Heart rate during submaximal and maximal tests was lowered (Bar-Or et al. 1976), peak and maximal oxygen uptake were increased (Bar-Or et al. 1976), and gait efficiency was improved (Kim et al. 2015). In van der Berg-Emons et al. (1998) study the participants had a 9-month training period done twice, first four times per week and after 2-month rest period for two times per week. These sessions included a variety of different sports such as running, swimming, and wheelchair skills. The peak aerobic power output measured with either cycle or arm crank ergometer was increased significantly at the end of both training periods and after a 2-month rest period, the values of four times trained individuals were significantly higher than in the start. Also, Verschuren et al. (2007) showed that children with CP could benefit from cardiovascular fitness exercise programs concentrating on lower-extremity work by increasing their both aerobic and anaerobic capacity via circuit-training of functional exercises. Younger children might have an advantage for muscle plasticity after gait training and therefore demonstrating better walking speed improvements (Hoffman et al. 2018). Especially people with GMFCS Level I CP can benefit from aerobic exercise as they are almost fully capable of doing a variety of different sports, but people with lower functional capabilities can find it less practical. Due to muscle weaknesses, people with CP need to generate relatively higher submaximal force outputs to maintain their walking pace, and thus especially the lower leg muscles are more prone to fatigue than in TD. With more severe CP, muscle fatigue of the shank muscles during walking

possibly could account for limited walking capacity and limited training response. (Eken et al. 2019.)

Overall it seems that aerobic exercise can enhance physiological outcomes such as cardiovascular capacity and endurance both short- and long-term, but it may not have a carryover effect on physical activity levels in children with cerebral palsy. (Rogers et al. 2008; Butler et al. 2010.) Closing a gap between motor capacity (i.e., potential) and daily activity (i.e., actual performance) may need promotion of both participation in physical activity and aerobic exercise altogether rather than just one-time prescribed exercise program. Thus, the regular daily physical activity probably impacts more the motor capacity than motor capacity influences the daily physical activity levels. (Ryan et al. 2017.)

Gait training studies vary for example by type of training used, usually including either over ground or treadmill gait training. Additionally, treadmill gait training can be done with partly body weight supported (BWSTT, bodyweight supported treadmill training). BWSTT has produced smaller effect sizes in improving gait speed than treadmill training without body weight support (e.g., over ground, instrumented treadmill training). However, populations using BWSTT and non-support training usually differ in their ambulatory capabilities and frequency using assistive devices (e.g., crutches) (Moreau et al. 2016). Still, more high-level evidence, large-scale controlled trials, on BWSTT studies are required to be claimed as an effective method of treatment in children with CP. (Martin et al. 2010.) For both treadmill-based gait training types, it should be noted that the changes in kinematical variables measured on a treadmill at preferred walking velocity might not be related to over ground functional capacity in people with CP (Gillett et al. 2019).

Daily gait training (á 30 minutes) with an inclined treadmill for six weeks showed positive results in ankle's active range of motion and gait speed in adults with CP. Intensive gait training might provide a way not only improve walking itself but also reduce the effect of contractures in adults with CP. (Lorentzen et al. 2017.) According to Willersley-Olsen et al. (2015), these changes in ankle range of motion could tell about plastic changes happening in the corticospinal tract. Willersley-Olsen et al. described that intramuscular coherence

measured from tibialis anterior could change positively because of the gait training done on the inclined treadmill. Positive changes in both beta and gamma frequency bands coherence were found, and this increase was also positively correlated with the subjects' capability to lift toes during the swing phase of gait. Many times, cerebral palsy patients cannot do toe-lift or heel strike because of motor deficits. Still, after the training, cerebral palsy subjects had significantly lower coherence values compared to typically developed controls. This tells the possibility and room for additional improvement after the 4-week treadmill training. (Willersley-Olsen et al. 2015.)

Recently, different kind of novel electrical, immersive and assistive technologies such as neuromuscular electrical stimulation (NMES), virtual reality (VR) and robotics (e.g., Lokomat and Hybrid Assistive Limb, HAL) have been used to influence gait recovery in children with cerebral palsy. Interventions including multichannel NMES-assisted gait demonstrate promising results in normalizing gait patterns, for example, applied technology may help children with spastic CP achieve a more upright gait. NMES is applied using wireless surface stimulators on lower-limb muscles, and stimulations are initiated by footsteps. In the future, 3D gait analysis accompanied by musculoskeletal modeling could be used to guide patient-specific NMES-assistance to facilitate even more appropriate muscle activations. (Rose et al. 2017.) VR-based training provides real-time feedback on multiple different sensory modalities to enrich the awareness of performance and result for a patient with a sensory deficit (e.g., CP). Multisensory feedback and improved engagement by VR-based training are suggested to have a positive influence on spatiotemporal parameters, i.e. walking velocity, cadence, and stride length in children with CP. (Booth et al. 2018; Ghai & Ghai 2019). Also, real-time feedback might be more valuable for children with worse initial gait. Still, the overall gait, measured as a composite score, might not improve similarly as the kinematic variables of attention. When focusing on either more complex feedback on kinematics or simpler spatiotemporal cueing the compensation movements from other parts of the body can take place during the gait. (van Gelder et al. 2017.)

HAL is a robotic device that can assist voluntary walking in response to the patient's intention measured by electromyography and force signals of steps and weight shifting. Immediate effects of gait training with wearable robots (HAL) have been promising in gait speed (0.71 to

0.83 m/s), cadence and mean step length (Matsuda et al. 2018; Takahashi et al. 2018). Furthermore, also on other robotic devices such as Lokomat improvements in walking speed and endurance have been discovered when training frequency has been over four times per week with a duration of ≥ 30 minutes (Carvalho et al. 2017). However, there is very limited data about interventions longer than four weeks in children with CP using these robotic technologies to enhance walking.

3 BIOMECHANICS OF TYPICAL AND CP GAIT

As simple as walking might look, the naturalness, efficiency and smooth coordination of human gait originate from the activity of several areas of the brain and involves the spinal cord, peripheral nerves, muscles, bones, and joints to cause the required motions. Bipedal walking comprises a repetitious sequence of limb motions to move the body forward while maintaining the posture. The rhythmic and coordinated pattern of nerve impulses needed for walking is produced by a central pattern generator, which consists of networks of neurons in the spinal cord. Still, neural input from higher levels and sensory feedback from the moving limbs is required for adapting to the environment and shaping the rhythmic motor output. (Guertin 2012.) However, neurodevelopmental disorders such as cerebral palsy can disturb the characteristics of normal walking which can be seen as biomechanical deviations from typical gait. (Whittle 2007, 1-30; Perry & Burnfield 2010, 3.) This chapter first describes the movements of lower extremities during walking and then secondly breaks down the typical gait dynamics and special cases of people with cerebral palsy.

3.1 Anatomy of lower extremities movements

In anatomy and movement analysis, the movement of the limbs is described using three basic reference planes: sagittal, frontal, and transverse plane (figure 5a). Most of the movement (i.e., forward and backward) in walking takes place in the sagittal plane, in any plane which divides the body into left and right sections. Flexion and extension joint movements and ankle's equivalent movements dorsiflexion and plantarflexion happen in the sagittal plane (figure 5b, 5c). The frontal plane divides the body into front and back sections and joint movements of adduction, abduction, inversion, and eversion of the foot occur in the frontal plane. Rotational movements such as internal and external rotation, pronation and supination of the hand or wrist are described in a transverse plane which divides the body into upper and lower sections. Pronation and supination of the foot are rotational movements about the long axis of the foot, which comprise of three components. Pronation involves forefoot eversion (sole pointing away from body's midline), ankle dorsiflexion and forefoot abduction while supination combines forefoot inversion (sole towards body's midline), adduction and ankle

plantarflexion. When investigating the movements of the lower limbs two important motion terms are valgus and varus. These terms are used especially in describing knee motion where a joint angulation is towards or away from the midline. Valgus deformity results in knock knees, whereas varus deformity results in a bowlegged presence. (Whittle 2007, 3.)

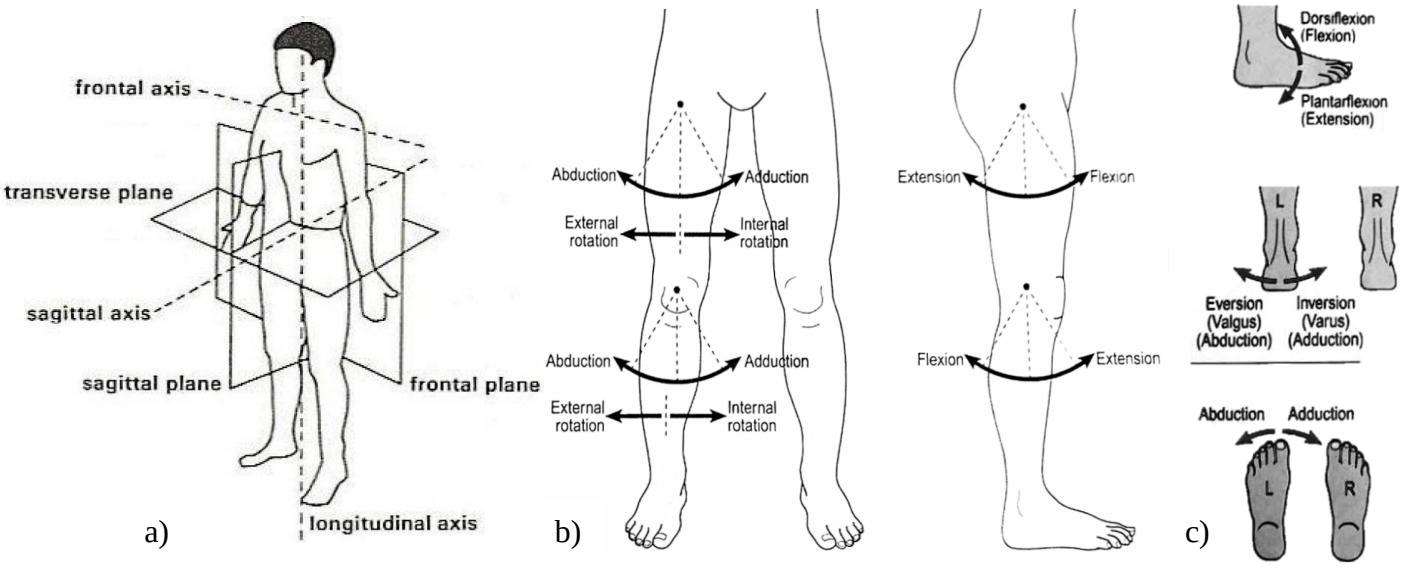

FIGURE 5. The anatomical position with three reference planes (a. de Oliveira et al. 2010) and movements about the hip, knee and ankle joint (b. & c. Whittle 2007, 4).

3.2 Gait cycle

Human gait consists of repeating movement patterns in each walking stride starting from the initial contact to the ground and ending with the same foot touching the ground again after the aerial phase. Each stride can be divided to stance (i.e., foot on the ground) and swing (i.e., foot in the air) periods and for analysis purposes gait cycle is defined to contain eight functional phases named in figure 6 and table 1. Each phase has its characteristics and objectives concerning posture, timing, and joint angles. Five of the phases take place during the stance period, which takes around 60% temporal gait cycle. Initial contact has been sometimes named *heel strike*, although in some individuals with cerebral palsy the heel may not touch the ground in any part of the gait cycle due to anatomical and functional

deformations. Likewise, the initial contact may happen with flat foot or forefoot first rather than with heel, for example, when the motor control of ankle dorsiflexion is weak. (Whittle 2007, 52-80; Perry & Burnfield 2010, 9-18.)

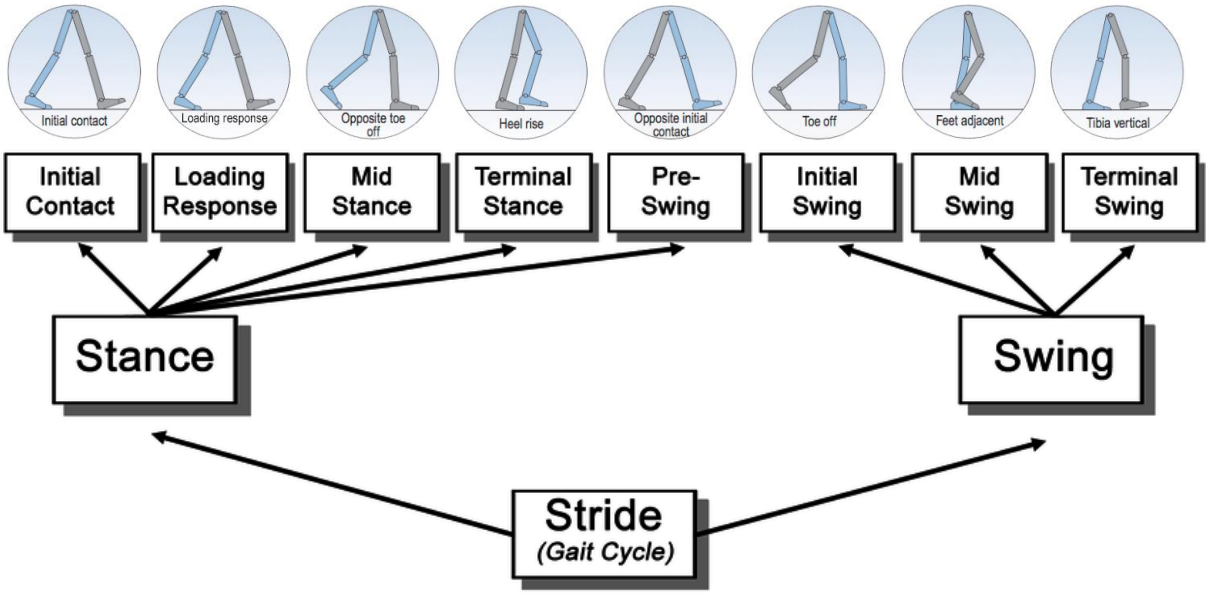

FIGURE 6. The functional division of the gait cycle. Stance and swing periods divide the gait cycle by foot contact. During stance, tasks of weight acceptance and single limb support must be accomplished while limb advancement is done during the swing phase. The eight phases are determined by limb postures. (upper part Whittle 2007, lower diagram Perry & Burnfield 2010.)

Perry & Burnfield (2010) divides the gait cycle to three types of tasks, weight acceptance, single limb support, and swing limb advancement. Weight acceptance task involves the first two gait phases, initial contact and loading response, so-called braking double support period. During these phases, there are three functional challenges to respond to, which are shock absorption, initial limb stability, and the preservation of progression. The swinging limb causes the rapid transfer of body weight, and the loading response requires stabilization in all lower body joints. Single limb support for the reference limb occurs during mid and terminal stance. The body weight progression over the stationary foot places demands in both sagittal and frontal plane for stabilization. The third task, swing limb advancement, starts already with preparation during the pre-swing phase and continues until the foot touches the ground again.

Ankle dorsiflexion and knee flexion are needed for sufficient toe clearance as well as hip flexion and knee extension to reach the adequate stride length. (Perry & Burnfield 2010.)

TABLE 1. Gait cycle. Double support (double limb stance) in light blue, single support (single limb support) phases in light green (Perry & Burnfield 2010). Each double support period lasts about 10% GC depending on walking speed as double support time decreases while speed increases (Whittle 2007, 52-80).

% GC	Phase	Movement	Objectives
0-2	Initial contact	Foot contacts the floor	Start stance with a heel rocker Impact deceleration
2-12	Loading response	Until to opposite limb toe-off	Shock absorption Weight-bearing stability Preservation of progression
12-31	Midstance	Opposite toe-off to heel rise of reference limb	Progression over the stationary foot Limb and trunk stability
31-50	Terminal stance	Heel rise to opposite initial contact	Progression of the body beyond the supporting foot; Limb and trunk stability
50-62	Pre-swing	Opposite initial contact to toe-off	Position the limb for swing Accelerate progression
62-75	Initial swing	Toe off to feet adjacent	Foot clearance of the floor Advancement of trailing limb
75-87	Mid-swing	Feet adjacent to tibia vertical	Limb advancement Foot clearance of the floor
87-100	Terminal swing	Tibia vertical to next initial contact	Complete swing of the limb Prepare the limb for stance

Gait cycle can be described with temporal parameters such as walking speed, step length, stride length, and cadence. Cadence is simply the number of steps taken in a given time, usually expressed as steps per minute. Step length is the distance between the heel contacts of the trailing leg and leading leg. Stride length contains one full gait cycle, thus combining two step lengths calculated as a sum of right and left step length. Walking speed is a product of

stride length and cadence (per 120 seconds), as the step lengths can differ between limbs in pathological gait populations. For example, in hemiplegic individuals might try to avoid putting weight on the affected side and hence having a significantly shortened step length on the affected limb. (Whittle 2007, 52-80.)

3.3 Muscles during walking

Muscles (figure 7) stabilize and propel the body forwards in the gait by generating the forces and moments allowing us to move. Muscle forces generate internal moments around the joint centers while external joint moments are created by ground reaction and inertial forces. Bipedal walking in human is thus a compromise between internal moments and external moments. Muscles are suitably designed for these kinds of activities and are sequentially activated in response to the gait phase demands to the limb posture (figure 8). (Gage & Schwartz 2009). Especially important muscles for walking are triceps surae (m. gastrocnemius and m. soleus), m. tibialis anterior, quadriceps, hamstrings, and gluteal muscles. Some of these muscles cross over one joint (monoarticular muscle) such as soleus, tibialis anterior and vastus lateralis/medialis while hamstring muscles, gastrocnemius and rectus femoris cross over two joints (biarticular muscles). (Stout et al. 2009.)

Hamstrings (biceps femoris, semimembranosus, and semitendinosus) function as hip extensors and knee flexors so during walking hamstrings work as limb stabilizers in contact and loading response and control hip flexion in the terminal swing. Anterior tibial group muscles, especially tibialis anterior, work as ankle dorsiflexors which are needed from pre-swing until terminal swing phases for ensuring toe clearance. Tibialis anterior has an important role also during the early contact to restrain ankle plantarflexion and partly absorb the force of impact. (Whittle 2007, 11-15; Perry & Burnfield 2010, 137-162). Hip extensor muscle, gluteus maximus, first controls the hip flexor moment caused by ground reaction forces and then accelerates the hip extension in early contact. During the loading response and mid stance gluteus maximus and medius are stabilizing in sagittal and frontal plane respectively. Three component vasti muscles (vastus intermedius, lateralis, and medialis) of quadriceps function as knee extensors. The fourth component of quadriceps, rectus femoris,

also flexes the hip due to originating from the anterior inferior iliac spine of the pelvis. Hip flexion and limb elevation are handled iliacus (iliopsoas), adductor longus and gracilis muscles primarily not by quadriceps. Rectus femoris controls excessive knee flexion during pre-swing and whole quadriceps secure the full knee extension before initial contact. This is needed for the intensive impact as knee becomes the main shock absorber during load response. (Gage & Schwartz 2009.)

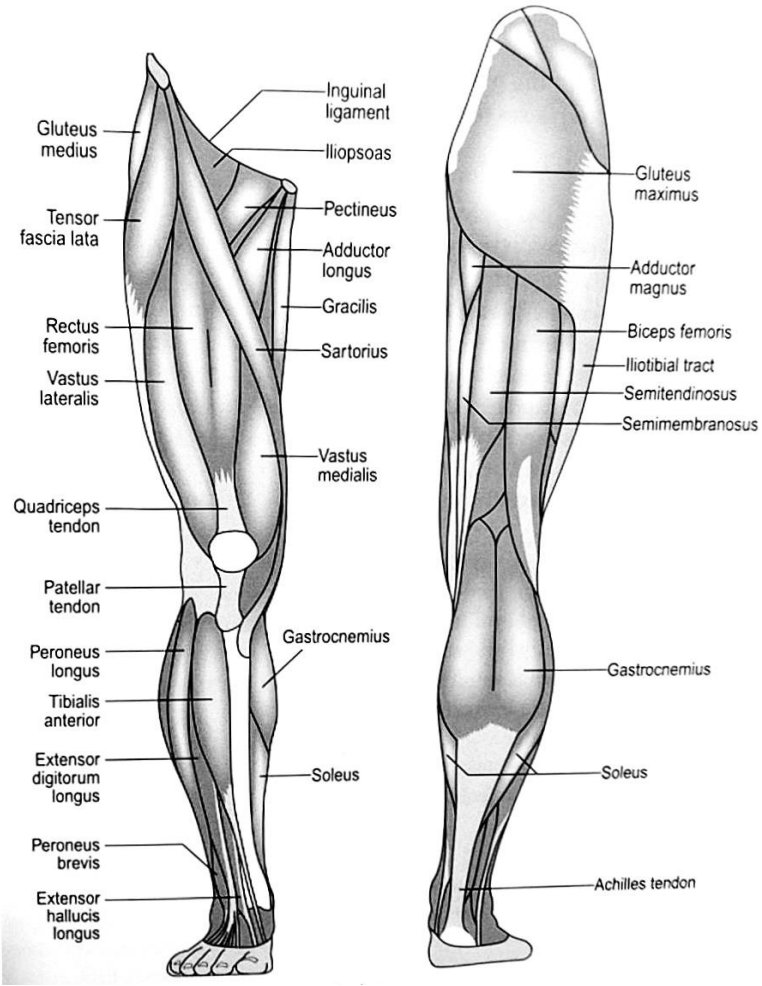

FIGURE 7. Superficial muscles of the right leg. (Whittle 2007, 13).

Triceps surae, a combination of soleus and gastrocnemius, the main function is ankle plantarflexion, but as gastrocnemius originates from the medial and lateral condyles of the femur, it also participates to knee flexion. Active range motion of the ankle during push-off phase is from 10° dorsiflexion to 20° plantarflexion, and at that phase ankle joint angular velocities can reach 300°/s in typically developed (Williams et al. 2019). Calf muscles are

vital for gait push-off as they peak in activity during terminal stance, and triceps surae generates about 50-60% total propulsion power required in walking (Gage & Schwartz 2009). During earlier stance phases triceps surae supports the ankle stability eccentrically enabling forward progression to forefoot without tibial collapse and making swing initiation possible. (Whittle 2007, 11-15; Perry & Burnfield 2010, 137-162.)

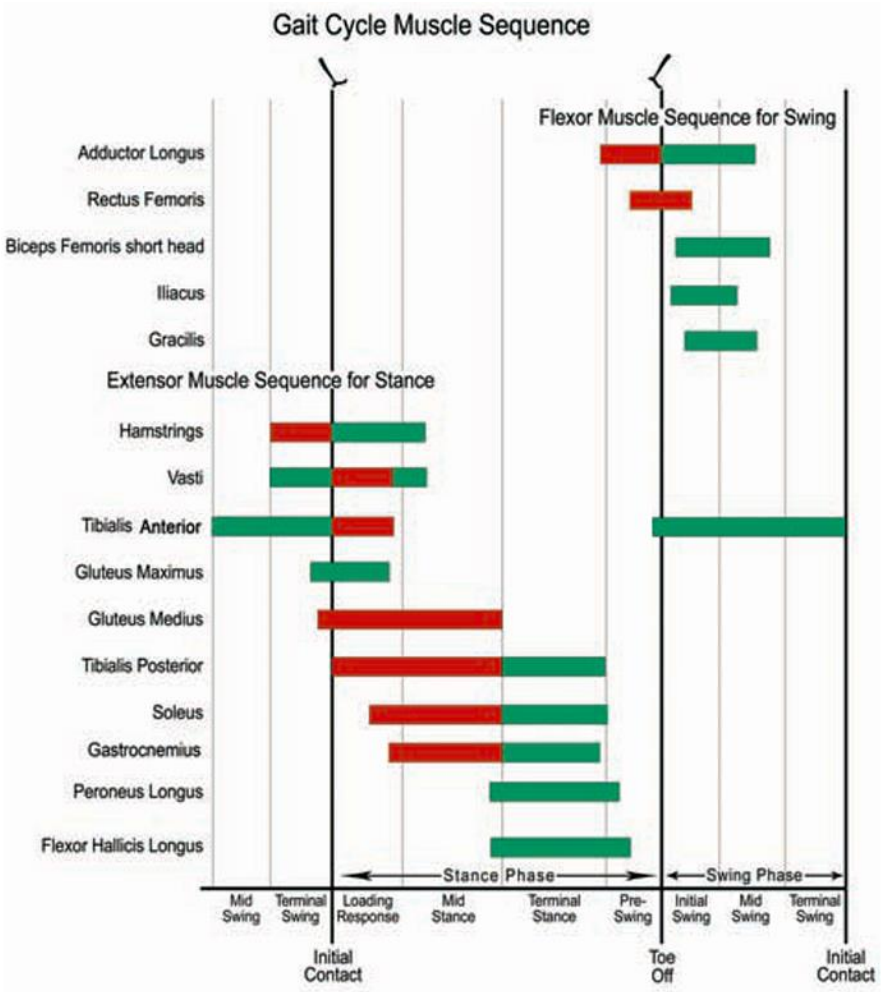

FIGURE 8. Muscle activation sequence during the gait cycle presented by Gage and Schwartz 2009. Eccentric muscle activity marked as red and concentric with green. While representing the typical activation pattern, it should be noted that the same movement can be achieved in multiple different ways depending on the individual, fatigue levels, or walking speed. For example, the neuromuscular system can maintain function and compensate the effects of muscle weakness to joint control by recruiting synergists or positioning center of mass in such a way as to limit the work needed by the weak muscle (Brunner & Rutz 2013).

3.4 Gait patterns in cerebral palsy

The effects of cerebral palsy on the musculoskeletal system are in general visible to the naked eye compared to typically developed individuals. The different gait abnormalities are augmented by progressively developing deformities throughout the individual's growth. For example, negative kinematical changes such as decreases in joint range of motion and declines both cadence and walking velocity have been demonstrated during the maturation of children with CP if no orthopaedical treatment is performed (Bell et al. 2002). The gait in children with CP is more unstable, have shorter step duration, and greater gait complexity compared to typically developed individuals (Piitulainen et al. 2018). Children with CP had significantly higher inter-stride variability in speed, stride length, swing, and stance, measured with foot-worn inertial sensors (Bregou Bourgeois et al. 2014). Most of the ambulatory cerebral palsy individuals can be divided into spastic hemiplegia and spastic diplegia gait deviation groups. However, the gait deviations of the individual rarely fit only one classification but are instead combining different patterns and the variety of deviations should be seen as a continuum. (Armand et al. 2016.) That is why it is important to, in addition to classification systems, identify patient-specific kinematic patterns defined by 3D motion analysis (figure 9).

Common gait patterns in spastic hemiplegia are drop foot and equinus with different knee stances (figure 10) while spastic diplegia group involves true equinus, jump knee, apparent equinus and crouch gait (figure 11). Clinical gait analysis using motion capture is used to quantify the complexity of pathological gait and identify the reasons behind the different gait deviations. Classifications of gait patterns for spastic hemiplegia and diplegia have been attempted by analyzing sagittal plane kinematics of the pelvis, hip, knee, and ankle. (Rodda & Graham 2001, Rodda et al. 2004.) Well defined classification helps the communication between professionals and subject's families both in the medical and research settings. With classification, homogenous subject groups can be defined, and certain treatments can be directed only to "right" groups. (Gage & Schwartz 2009.)

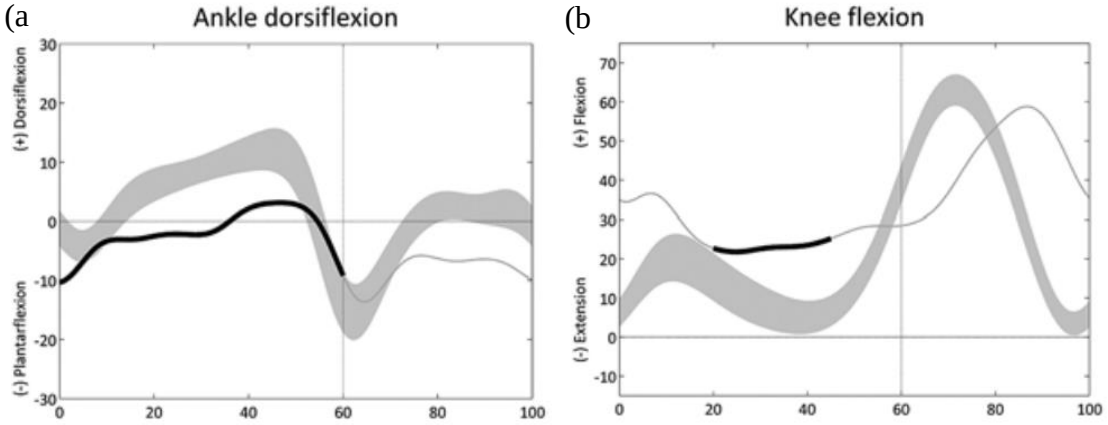

Figure 9. Measuring gait kinematics of lower body joints can reveal a more detailed picture of gait impairments. For example, picturing lack of ankle dorsiflexion (a) during stance (black curve) or limited knee extension during stance (b) compared with typical gait pattern (dark grey band) could help professionals to make better and more patient-specific decisions for gait deviation treatment. In this case, probable reasons could be plantarflexors' overactivity or contractures (a) and hamstring contractures or compensative excessive flexion on other joints (b). (Armand et al. 2016.)

3.4.1 Spastic hemiplegia

Every CP individual has their specific gait deviations, with differing degrees of involvement and combinations of impairments. Loss of selective motor control is more severe in the distal parts of the extremities (i.e., ankle and foot) than the proximal parts (i.e., hip) because the pyramidal tracts affected by cerebral palsy are distributed primarily to the distal ends of the limbs. Also, one-joint muscles are controlled better than biarticular muscles. (Gage & Schwartz 2009.) Gait patterns in spastic hemiplegia can be grouped into four patterns (figure 10), by their severity of impairments and impairment distribution from ankle to the pelvis. The differences in movement patterns between affected and non-affected limb are apparent. Type I group is characterized by affected side foot drop during swing phase resulting in initial contact not to be a heel rocker as normally but a toe strike. Drop foot is associated with the relative weakness of the tibial anterior muscle or overly active gastrocnemius and soleus muscles because in many cases the ankle has normal dorsiflexion capacity passively. The underlying cause of the weakness of the tibialis anterior might be the weaker central drive to

ankle dorsiflexors. Significantly lower common drive to the affected limb tibialis anterior has been found to produce foot drop leading to toe strike gait pattern. (Petersen et al. 2013.) Drop foot could be corrected with a simple ankle-foot orthosis. Due to foot's plantarflexed position compensations of the increased knee and hip flexion and lumbar lordosis happen during the foot clearance and early stance. (Whittle 2007, 195-218, Armand et al. 2016.)

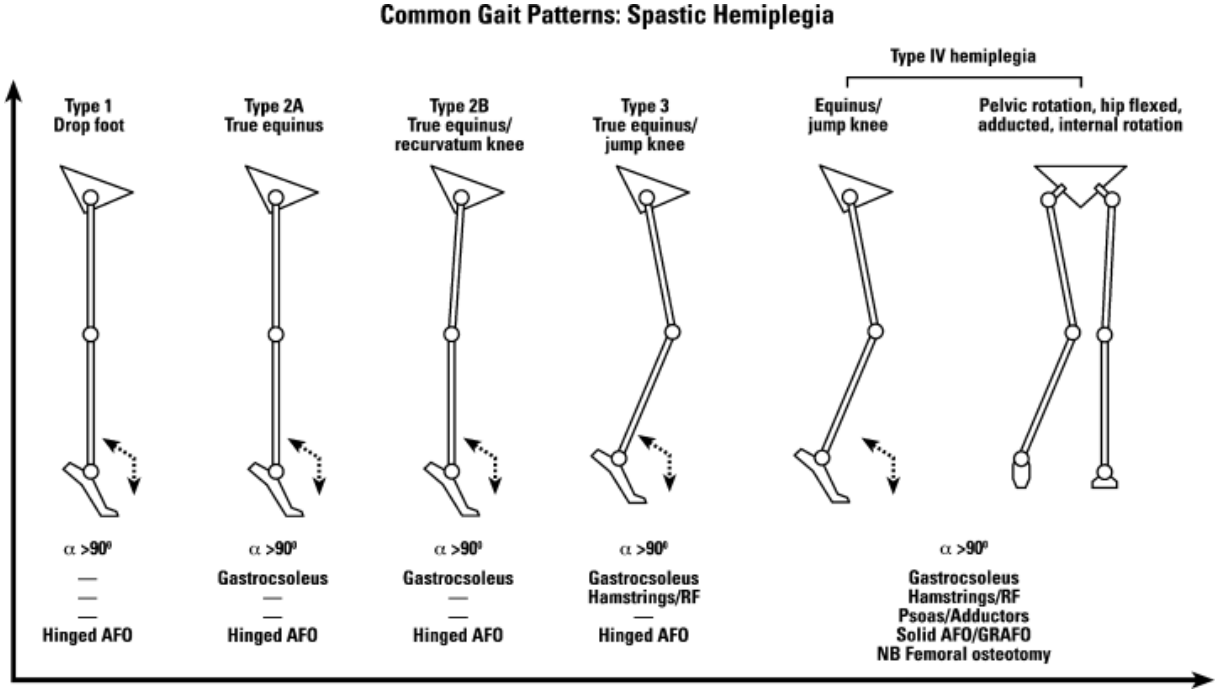

Figure 10. Common pathological gait patterns in spastic hemiplegia grouped into four different types. Rows under the stick figures indicate the ankle angle, contracture management areas, and orthotic management recommendations. (Rodda & Graham 2001.)

Type II hemiplegia gait pattern includes the foot drop in swing phase but also constant plantarflexion during stance phase because of contractures of the calf muscles, a deformity called as true equinus. Constant plantarflexion during mid-stance forces knee to hyperextension by producing external knee extensor moment (plantarflexion-knee extension couple) because of the ground reaction force vector shifts in front of the knee joint (Chang et al. 2010). Toe walking leads to smaller foot contact during stance and makes positioning of the foot harder before the initial contact in the late swing impairing the stability and fluidity of gait (Gage & Novacheck 2001). Usual surgical operation for type II hemiplegia contractures is Achilles tendon lengthening, while spasticity can be managed by BoNT-A injections with support of orthosis. For example, the hinged ankle-foot orthosis (HAFO) could enable heel-

toe gait and higher walking speeds, improved stride length, and decreased cadence. The orthosis can change ankle motion so that floor clearance by the toes is possible. Although some of the movement restrictions are crossed out, the EMG activity of tibialis anterior decreased overall as the HAFO substitutes the muscle making the tibialis anterior redundant. (Romkes et al. 2006.)

In group III, in addition to deviations from the first two, the impairments are spread even more to the knee joint. The knee joint has a limited flexion range of motion during swing phase because of overactive cocontraction of quadriceps and hamstrings, which is one of the main components contributing to gait inefficiency in CP (Unnithan et al. 1996). Cocontraction causes “stiff knee gait” where the step of the affected side can be seen sometimes as “a jump” back to the non-affected side. For the management of type 3 hemiplegia lengthening of hamstrings combined with the transfer of rectus femoris are instructed in addition to procedures mentioned above. (Rodda & Graham 2001, Whittle 2007, 195-218.) Type IV hemiplegia group has reduced motions also at the hip flexion and knee, which are compensated by anterior pelvic tilt and increasing pelvic lordosis in the terminal stance phase to maintain the stride length. Along with changes in sagittal plane motions, also pelvic rotation, hip adduction, and internal hip rotations might be apparent in type IV. Pronounced hip compensations and power production from hip and knee are present if ankle cannot withstand the power generating requirements of walking (Whittle 2007, 195-218). For a good overall management outcome both earlier distal and proximal impairments need to be handled, for example, hip flexors and adductors are lengthened and external rotation osteotomy of the femur is conducted. (Rodda & Graham 2001, Armand et al. 2016.)

3.4.2 Spastic diplegia

Spastic diplegia affects the gait patterns bilaterally, which leads to problems of stability in walking, and hence walking aids are commonly used. Spastic diplegia can be divided into four main gait pattern groups: true equinus, jump gait, apparent equinus, and crouch gait (figure 11). In true equinus, the most prominent characteristic is permanent ankle plantarflexion during stance leading to toe walking (equinus), but sometimes it is combined

with knee hyperextension (recurvatum). Jump knee gait pattern is affected by spasticity of thigh muscles in addition to equinus ankle. The knee is often stiff during the swing phase because of rectus femoris activity, and this shifts the hip to flexion and increases lumbar lordosis. BoNT-A therapy targeting gastrocnemius and hamstrings suits for younger children and surgical lengthening management of gastrocnemius, hamstrings, and iliopsoas muscles with rectus femoris transfer to semitendinosus are possibilities for older children. (Rodda & Graham 2001.) These operations should be done with SEMLS procedure, taking care of all contractures and lever arm dysfunction at one operation to avoid "birthday syndrome" (Armand et al. 2016).

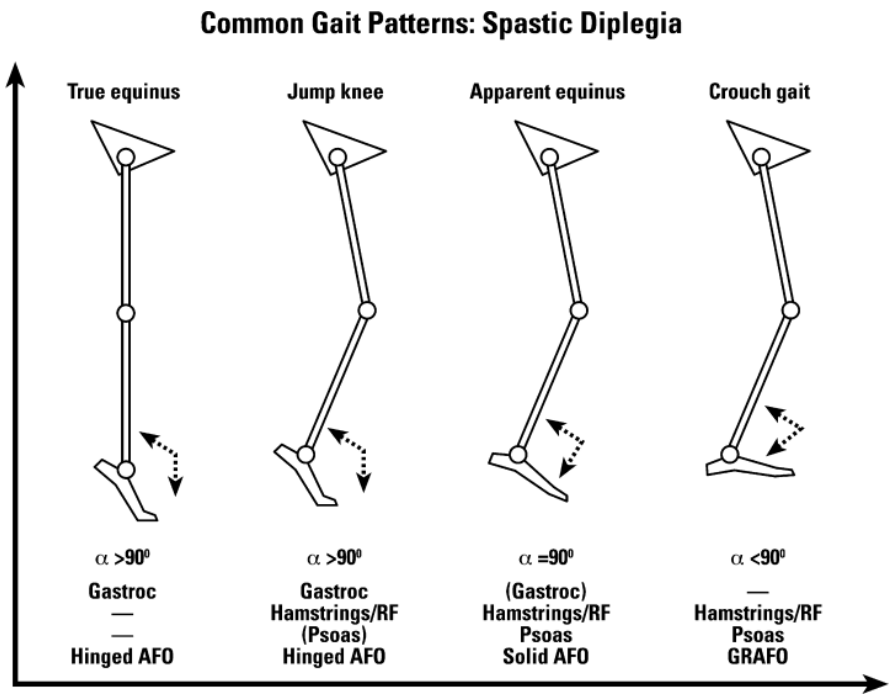

Figure 11. Common pathological gait patterns in spastic diplegia grouped into four different types. Rows under the stick figures indicate the ankle angle, contracture management areas, and orthotic management recommendations (AFO = ankle-foot orthosis, GRAFO = ground reaction ankle-foot orthosis). (Rodda & Graham 2001.)

Differing from the two earlier patterns, the apparent equinus shows normal range in the ankle, but the knee and hip are greatly flexed during the stance phase and will not reach full extension in any point of the gait cycle. Excessive flexion in knee and hip joint leads to walking on the toes, and that is why ankle in apparent equinus can be easily misinterpreted as equinus ankle. Thus, in apparent equinus applying BoNT-A to gastrocnemius or lengthening

it surgically would weaken the muscle and provoke the gait pattern more towards crouch gait. Management should be focusing on hamstrings and iliopsoas treatments. The fourth group spastic diplegia is crouch gait, which presents both excessive dorsiflexion at the ankle and flexion at the knee and hip joints. The pelvis is tilted posteriorly, different from the other gait patterns. Crouch gait can be an outcome of wrongly or inadequately addressed BoNT-A therapy or surgeries, but the energy expensive gait pattern can occur progressively due to spasticity of either hip flexors or the hamstrings. The crouch posture with excessive knee flexion is very demanding for quadriceps and patellar tendon as the ground force reaction vector direction is far off behind the flexed knee increasing the external moment. Constant knee flexion during stance blocks the normal plantarflexion-knee extension couple and hence increases energy consumption as one must produce the missing internal knee extension moment by contracting quadriceps. (Rodda & Graham 2001.) Apart from sagittal plane crouch posture, excessive hip adduction and internal rotation may be present in diplegic gait, resulting so called “scissor gait” where thighs and knees touch with each step (Perry & Burnfield 2010, 349-351).

The correct management of crouch gait is a complex situation including the lengthening of the hamstrings and iliopsoas, ground reaction ankle-foot orthosis and corrections of torsional bony problems. (Rodda & Graham 2001; Rodda et al. 2004; Whittle 2007, 195-218.) In diplegia, the gait pattern can also be asymmetric where the other affected limb displays different gait pattern or different level of involvement than the other limb (Rodda et al. 2004).

3.4.3 Walking with crutches

Stability, while walking and standing, together with mobility are big challenges in cerebral palsy population and walking aids such as crutches are used to increase the base of support, help with mobility limitations and improve one’s quality of life and autonomy. People with CP on GMFCS level II or III are the most probable users for crutches and other lighter mobility aids. Some at level II may be able to walk short distances without handheld mobility aid but prefer to use them constantly in their daily life. The most common model of crutches for long term usage in lifelong disabilities is forearm crutches, also called elbow, Lofstrand or

Canadian crutches. While the crutches admit of better ambulation capacity, walking with crutches may modify the gait pattern significantly. Supporting body weight partly through the arms may also lead to wrist and shoulder joint pain due to the vertical forces for which they are not originally designed to handle. Walking bipedally with two crutches can be described as four-point gait (i.e., reciprocal gait) where separate and alternate movements of legs and crutches go with a pattern, for example, right crutch – left leg – left crutch – right leg. This pattern is stable and leg movements resemble typical walking yet being slower than three-point step-through gait where legs move together as a single unit. (Whittle 2007, 122-128.)

Obviously, walking aids will change the walking biomechanics when compared to walking without walking aid. Krautwurst et al. (2016) investigated the effects of walking with crutches to lower body and trunk kinematics. Pelvis was significantly more anteriorly tilted, and less dorsiflexion was achieved while walking with walking aid. However, positively, with crutches hip and knees are better extended, but also the increased stability can be seen in significantly decreased step width and lesser cadence. Thus, walking with crutches could partially suppress or enlarge the effects gained through interventions or surgical operations, and hence assessing gait should be done with and without walking aids if possible. (Krautwurst et al. 2016.) Secondly, Yeung et al. (2011) made similar observations in walking cadence and increased anterior pelvic tilt but did not find significant changes in lower-limb kinematics.

4 GAIT ANALYSIS

Computerized gait analysis (CGA) is a way to analyze one's walking very precisely using methods such as 3D motion capture systems, accelerometry, force plates, and electromyography to measure human limb muscle functions, kinematics (gait patterns, figure 15) and kinetics (forces). In clinical populations such as CP gait analysis is used, for example to help managing gait problems, determining underlying causes of gait deviations and evaluating lower limb functionality before medical or surgical interventions. Gait analysis offers clinician "a snapshot" of a patient's pattern of walking, which can be compared to other moments of measurements or result databases (figure 12). (Armand et al. 2016.)

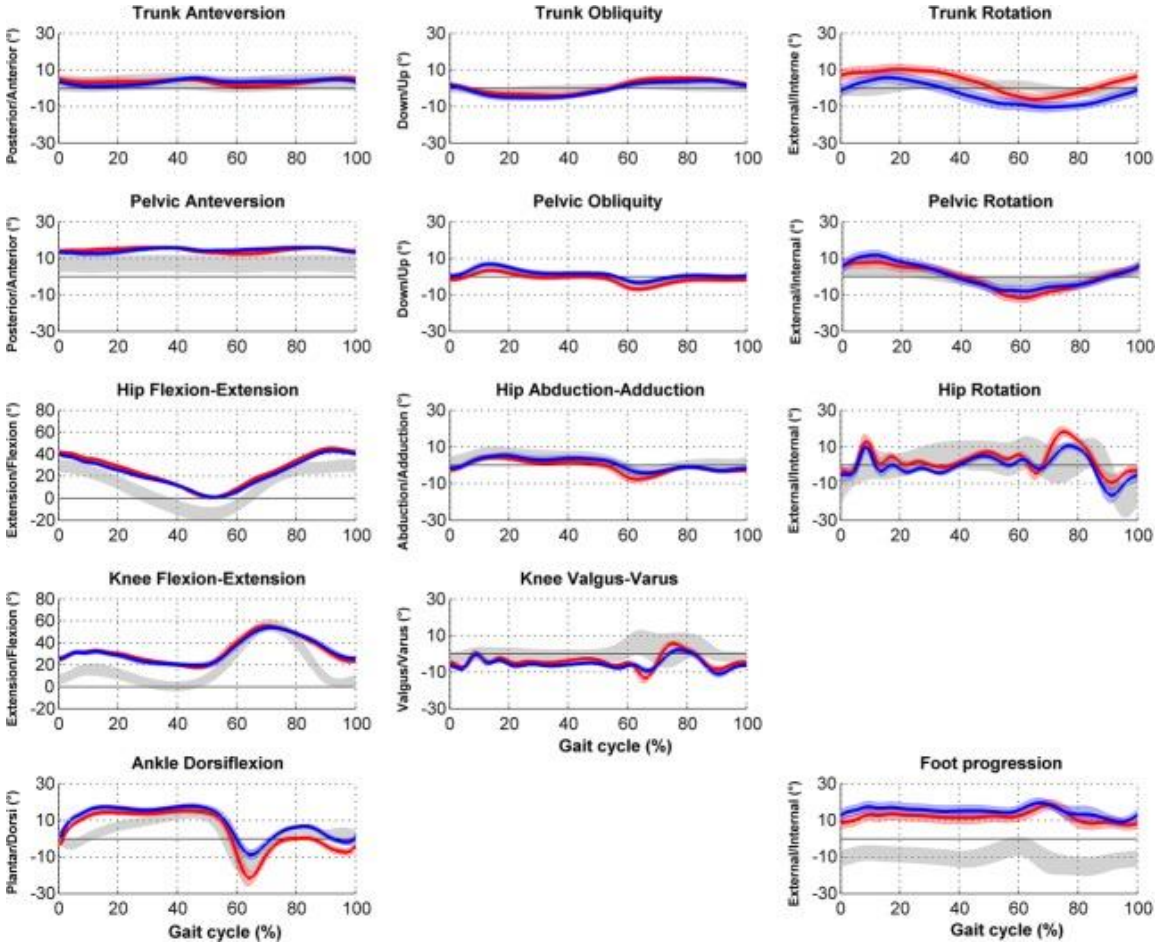

Figure 12. Example of data in clinical gait analysis report for bilateral spastic cerebral palsy: kinematics. The left side of the body is the red line, the right side is the blue line, and the grey band is normal reference kinematics of TD population. (Armand et al. 2016.)

As part of the standard of diagnosing pathological gait, CGA data can differentiate the primary gait abnormalities from the compensatory deformities. (Desloovere et al. 2005, Lofterod et al. 2007). For example, when conducting multiple procedures in one surgery, CGA is an important tool to guide the development of multilevel surgery plan as studies have shown that CGA data acquisition can result in changes in up to 80% of the plans (Khouri & Desailly 2017). To justify the importance of CGA, the gait functions have improved more when surgical recommendations acquired with CGA has been applied. After surgical or non-surgical treatments, CGA is an important part of decision-making for finding the optimal follow-up treatment and physical therapy post-operatively. (Wren et al. 2013.) Results from motion analysis laboratories have also been used in the development of new surgical techniques and orthoses (Chambers 2010).

4.1 Methodology for gait analysis

To fully capture the complexity of human walking, there is a need for multiple perspectives in the CGA. Motion capture provides the magnitude and timing of joint movements, force plates display the functional demands required during the stance phase, and electromyography reveals the muscle function during actions. (Perry & Burnfield 2010, 403-424.) Fusing these measurement systems, CGA improves our ability to objectively understand the gait dynamics beyond what our eyes can detect and secondly, it can provide real-time information while the patient is walking and record it for more detailed analysis. Common variables measured in gait analysis are kinematical (i.e., joint movements and angles, angular velocities and accelerations), kinetical (i.e., joint moments) and spatiotemporal variables such as gait velocity, cadence, stride length, and stride variability. (Chang et al. 2010.)

4.1.1 Motion analysis system

Nowadays, in the research and gait laboratory setting, there are several different three-dimensional kinematic motion analysis systems used for computerized gait analysis, which are more or less based on similar high-speed camera recording technologies. The three main approaches of motion capture systems differ mostly in their utilization of markers for

movement tracking. The most common marker technology used at the moment is optical-passive retroreflective markers which are captured with infrared cameras (e.g., Vicon and Qualisys systems). The two other common approaches are optical-active LED-markers (e.g., Optotrak) and markerless motion capture systems (e.g., Simi and Kinect). (Vicon Motion Systems 2019a.)

In optical-passive systems, the infrared light reflected off markers back into cameras are seen as bright spots. That is why first physically covering possible unwanted reflective spots in the laboratory and secondly software filtering of camera signal is needed to make automatic marker location detection more reliable. Automated multi-camera systems (e.g., figure 13) record each marker's two-dimensional coordinates (bright spot centroid) and by combining multi-camera information of marker required. However, passive markers do not have specific labels to which anatomical location they are related before they are labeled partly manually by the user and automatically by the software. The motion analysis softwares use static (e.g., T-pose or "motorbike" pose) or dynamic calibration trials where the user manually labels markers and software uses these relative marker locations to help the automatic marker labeling process in the walking trials. (Vicon Motion Systems 2019b.)

Two or more cameras are needed to reconstruct marker location in three-dimensional space and covering multiple gait strides in the gait lab usually 8-12 cameras are required. Comparing the camera count with the system's performance there was a significant improvement in accuracy when switching from 6 to 8 cameras. Adding two more cameras to have a total of 10 cameras did not further better the accuracy. (Eichelberger et al. 2016.) The number of cameras is not only dependent on the measurement volume but also the specific task measured. If markers are easily visible during walking trials fewer cameras are enough, but more cameras are needed if markers are likely to be blocked by walking aids such as crutches. Specified marker placements in every gait model are compromises between anatomical significance and marker stability during movements. (Perry & Burnfield 2010.)

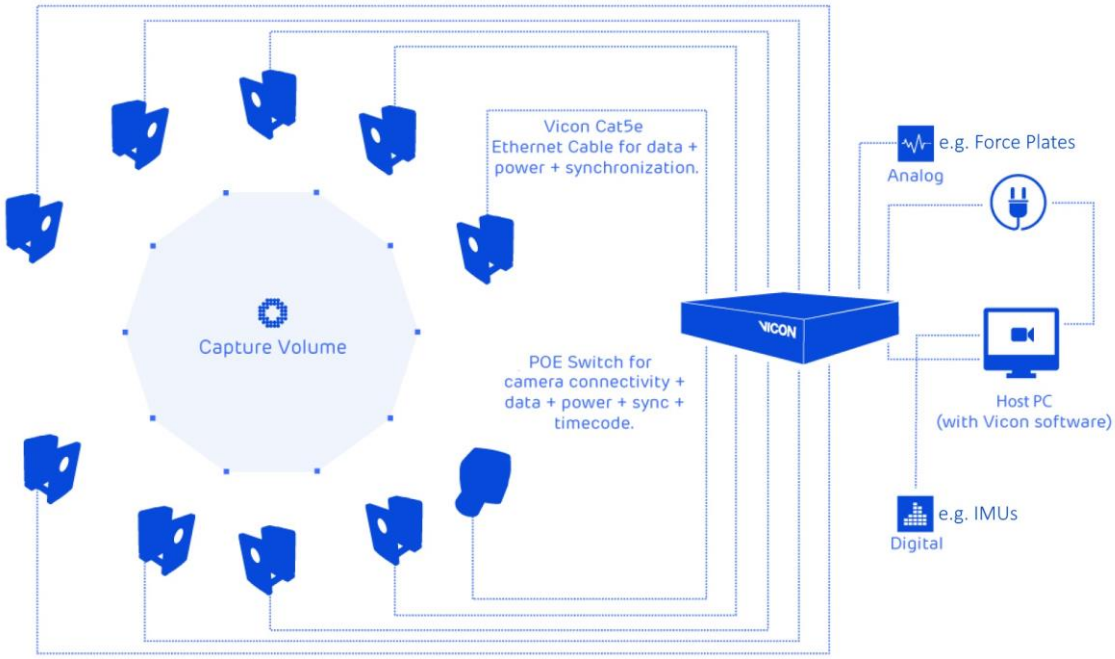

FIGURE 13. Example of kinematic motion capture system combined with analog devices (e.g., force plates) (Vicon Motion Systems, 2019a).

The precision and reliability of the measurements done on gait analysis systems is a priority in clinical and research practice. Camera recording frequency at 60 Hz is suitable for normal walking, but only higher frame rates (>100 Hz) give satisfactory results of capturing higher velocity movements (Perry & Burnfield 2010). While measuring in pre- and post-intervention settings, it is important to know if changes are because of treatment effects, measurement error, or a combination of both. Vicon system's marker error in dynamic experiments was found to be less than 2mm. By optimizing marker size and system sampling rate in reference to the measured movement's speed very high, 0.3 mm, precision can be acquired in Vicon. (Merriaux et al. 2017.) In addition to markers' skin movement artifacts, errors in gait kinematics calculations are usually due to erroneous palpation of anatomical landmarks leading to wrong marker placements and inaccurately defined joint coordination systems (Scheys et al. 2010). Calculating locations of joint centers even from erroneously placed surface markers can create a significant error in estimating joint angles and kinetics. Large propagation errors in hip moments and delayed flexion/extension timing were found when marker positions were changed, and hence hip joint center was estimated with ±30 mm

inaccuracy. (Stagni et al. 2000.) For joint angles over 5° errors may lead research and clinical decision-making to wrong conclusions. Highest reliability indices (within-assessor coefficient of multiple correlation) were found in the sagittal plane in the knee and hip joints while ankle joint was not charted, and pelvic tilt presented lower reliability. Overall transverse plane joint movements showed the least reliability and frontal plane reliability was between these two. The error in angles was reported being <4° for the sagittal plane, 2° for the frontal plane and highest errors were found in rotational movements of hip and knee. Less than 2° error is considered as an accepted range for most clinical decision-making, but the goal should always be providing measurements free of error. Most of the gait variables fell between 2° to 5° error range, which is still likely reasonable for clinical use depending on the situation, but then data interpretation needs to be done carefully. Additionally, in gait analysis of children with CP, it is important to pay attention to walking equipment (shoes, with or without orthosis or barefoot) as the marker placements could vary because of marker reapplication. (McGinley et al. 2008.) It seems that CGA is a reliable and precise method to measure clinically relevant changes in interventions when the gait analysis preparation is done with quality by a skilled operator. For example, goniometric measurement of the range of movement of the major lower limb joints has reported higher variabilities (>10°) (McDowell et al. 2000). Guidelines and standardization for clinical gait analysis specifically have been created at least in the USA (Commission for Motion Lab Accreditation) and UK (Clinical Movement Analysis Society) (Baker et al. 2016).

4.1.2 Kinematic analysis and variables

Describing the angular motion of the ankle, knee, hip joints, and pelvis is an essential part of the gait analysis. The kinematical analysis is based on the segment coordinate systems and has no reference to inertial properties. When performing the kinematic analysis part of CGA, one convention is to present the body via a biomechanical model, which is a collection of rigid segments representing skeletal structures. Segments' interaction with each other is described by joint constraints allowing zero to six degrees of freedom (DOF). DOF is a number of independent parameters (e.g., translational and rotational coordinates) necessary and sufficient to uniquely define both position and orientation of a rigid segment. For example, with at least three markers per segment a segment can be described using three position

coordinates (e.g., X, Y, Z) and three rotational variables (θx, θy, θz) describing rotations around the local coordinates system's main axes, thus these biomechanical models are called 6-DOF models (segment optimization). Segment optimization tracks each segment independently and hence does not have exact linkage connecting two segments. The method assumes that segments are linked firmly by the motion capture data as segments cannot come apart when the person did not come apart while performing motion analysis. Segment independency allows tracking real joint movements as many of the lower body joints are not a fixed axis but may include errors due to skin movement artifacts. Segment's three noncollinear points (markers or calculated virtual locations) define each segment's local coordination system (LCS) which moves correspondingly as the segment moves in the laboratory volume defined by global reference system (GCS). (Hamill et al. 2014.)

One of the most common gait models used in gait laboratories is the conventional gait model, which has many variations such as Vicon's implementation called Plug-in Gait. This model has demonstrated good reliability for clinical decision-making (McGinley et al. 2008). Plug-in Gait uses Direct Kinematics (i.e., Direct Pose Estimation) for computing joint kinematics, position, and orientation of each segment, based on a set of three tracking markers (figure 14a). This offers outputs about three rotations in the pelvis segment, hip, and knee joints and two rotations for the ankle joint. For each time the model is scaled to the participant in respect of the relative marker positions. One of the weaknesses in Plug-in Gait is the modeling foot as one rigid body which limits foot modeling accuracy and clinical gait analysis decision making around the foot and ankle. This may induce wrong conclusions for example when foot's other joints (e.g., subtalar joint) motion is interpreted as whole ankle rotation (Bland et al. 2011) and possibly measured data in frontal and transverse planes cannot be trusted and therefore used (Davis 1991). The more accurate analysis would require more precise foot models (e.g., Oxford Foot Model) or additional measures with slow-motion video and pedobarography (plantar pressure) to help clinical decision-making. (Gage & Stout 2009.)

Other software may use, for example, Inverse Kinematics (i.e., Global Optimization) based musculoskeletal models (OpenSim). Musculoskeletal models may have the advantage to individualize the model better by adjusting the model parameters such as muscle-tendon lengths, bone lengths which might have been modified due to surgeries in CP population. This

might decrease the number of inherent errors related to anatomic variability (Simon 2004). The reliability of Inverse Kinematics has not yet researched enough in a diverse range of populations, but results of reliability have been promising from the small set of studies. The Inverse Kinematics (3-3-2-Degrees of Freedom) model showed <5° mean standard deviation in all lower body joint angles whereas Plug-in Gait model performed similarly in sagittal and frontal plane but was weaker in the transverse plane. (Kainz et al. 2017.) The three pose (position and orientation) estimation methods mentioned above differ mainly in mathematical complexity and their ability to handle artifacts in the motion data (Hamill et al. 2014).

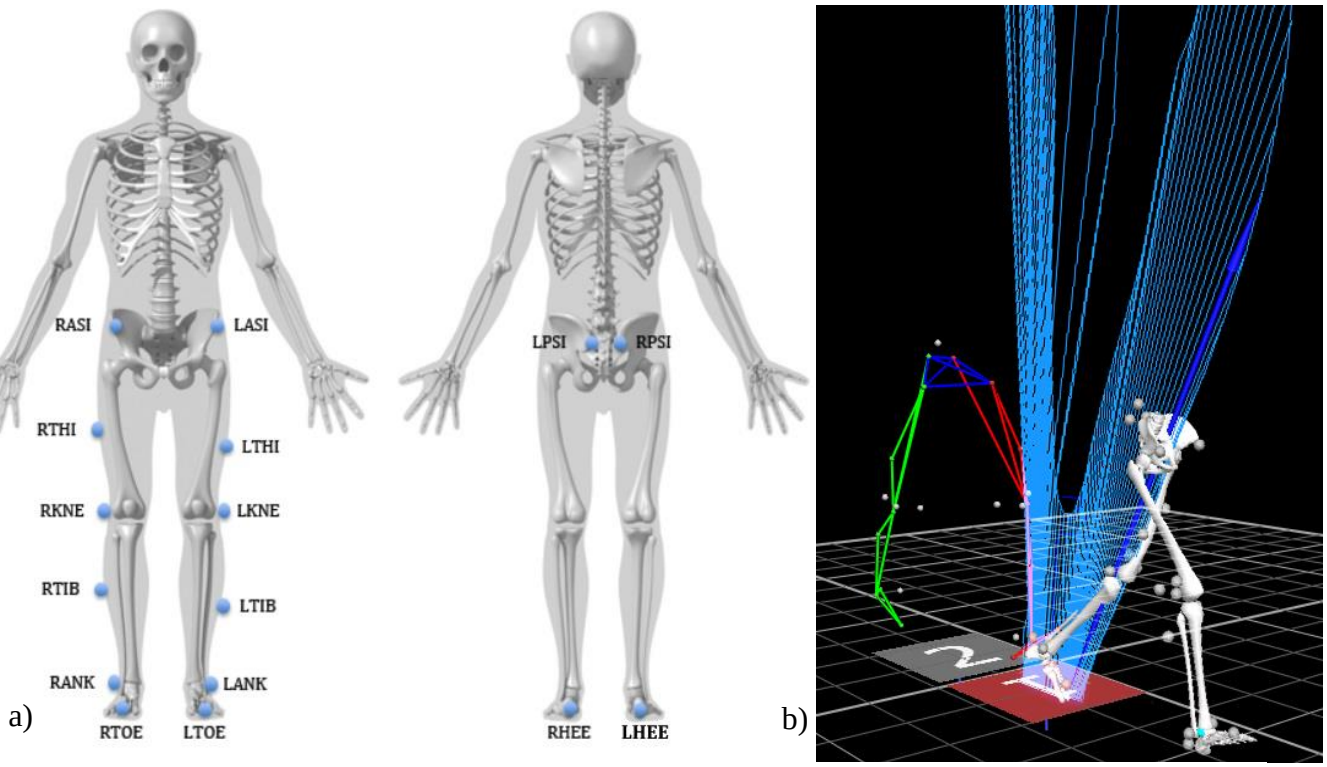

Figure 14. (a) Marker placement for Plug-in Gait lower body model and (b) example of labeled Plug-in Gait skeleton in Vicon Nexus software (left) and bone skeleton model (6-DOF) in Visual3D software based on Plug-in Gait model marker data (right).

Joint angles can be defined as relative orientations (3D rotation matrix) of one LCS with adjacent segment's LCS, for example, shank LCS relative to thigh LCS constitutes knee angles (figure 15). In this case, the Cardan-Euler approach is used for angle calculations. (Griffiths 2006.) Then, for example, knee angles can be presented as three anatomically and clinically relevant elements, flexion-extension, abduction-adduction (varus-valgus) and

internal-external rotation angles. To derive above-mentioned three joint angles for lower extremities, often Cardan sequence of three elementary rotations X-Y-Z is used in motion analysis software and hence these are called Cardan angles. In this sequence, first rotation is about the laterally directed axis (X), second rotation about an anteriorly directed axis (Y) and lastly rotation about the vertical axis (Z). Cardan angles describe the orientation of one coordinate system related to another coordinate system as ordered rotation sequences (e.g., X-Y-Z) starting from the original position of one coordinate system. (Hamill et al. 2014.) Hip angles are defined as the orientation of the thigh segment relative to the pelvis segment and ankle angle as foot segment relative to the shank. The distinction to other lower body angles, pelvic angles, and foot progression are absolute angles as they are referenced to the laboratory coordinate system. (Davis et al. 1991.)

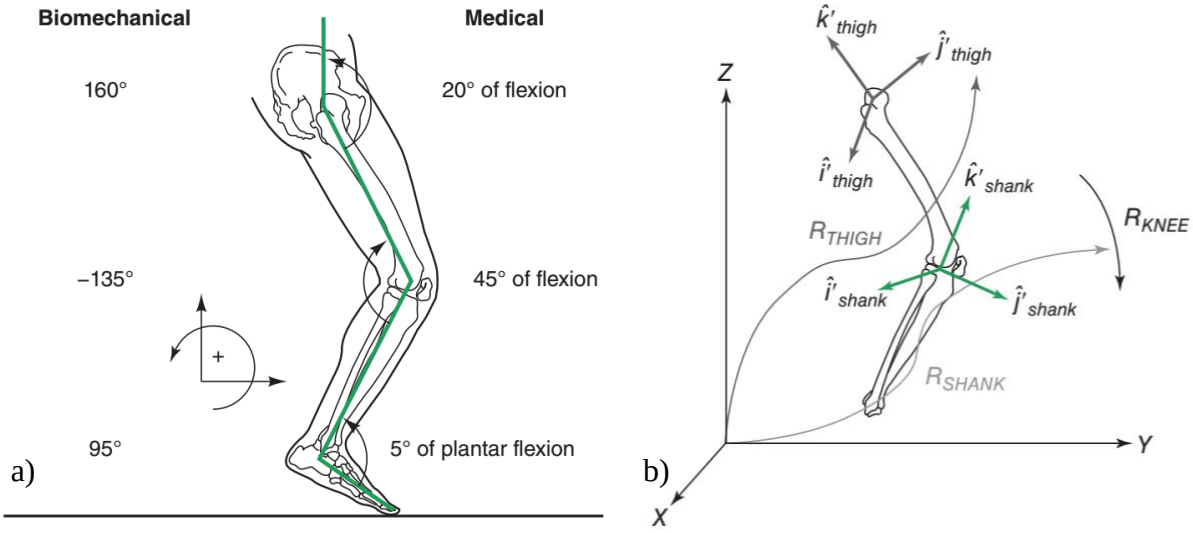

Figure 15. (a) Relative sagittal angles of the lower extremity in 2D (Robertson & Caldwell 2014). The knee joint angle in three dimensions is defined as the orientation of the shank segment relative to the thigh segment (Hamill et al. 2014).

4.1.3 Force plates

In gait analysis, we are interested in ground reaction forces (GRF) encountered by the participant during the stance phase. Force plates (figure 16a) are instruments for measuring the total force applied by the foot on the ground. (Whittle 2007, 80-84.) Applied force to the

force plate creates a proportional electrical signal due to length changes in its strain gauges, which is interpreted as a three-dimensional force vector. It is determined by the manufacturer of each force plate how many volts for every newton of force applied is measured. The GRF vector can be divided into its components: vertical, and two shear forces in anterior-posterior and medial-lateral directions. Location of the GRF vector initial point is called the center of pressure. Commonly, the force plates locate a force transducer in each of the four supporting corners, and by combining these four electrical signal changes the location and magnitude of force vector can be calculated. (Winter 2005.)

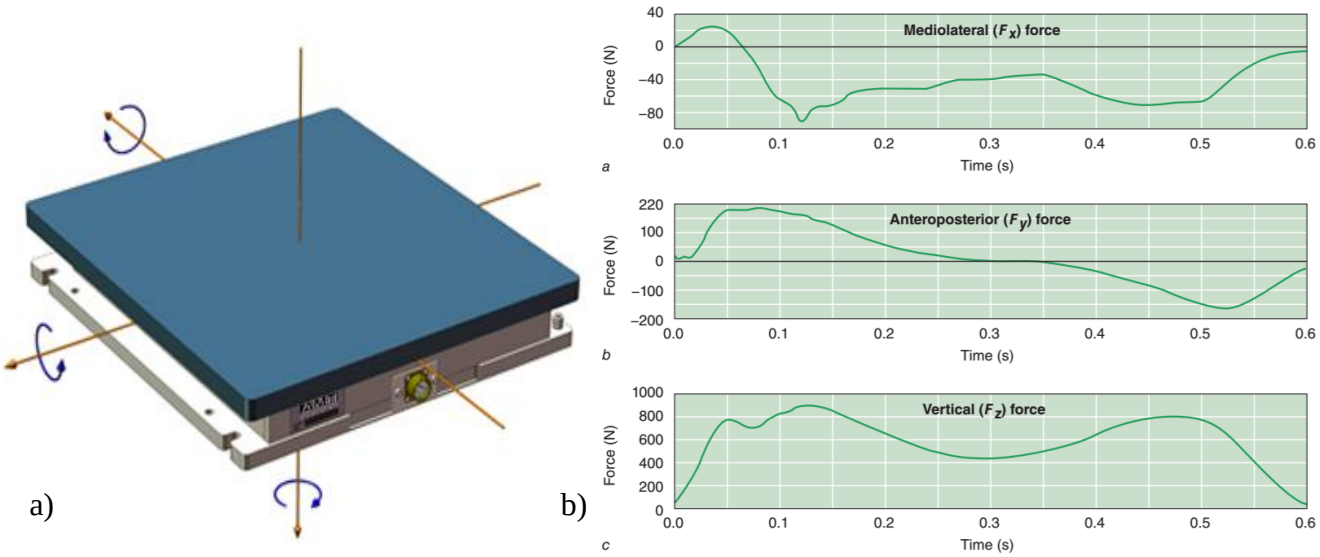

Figure 16. (a) Force plate demonstrating three force (F_x, F_y, F_z) and three moment (M_x, M_y, M_z) components measured during foot contact. Positive moments are defined by right hand rule. (AMTI). (b) Typical GRF during walking broken into mediolateral Fx (top), anteroposterior Fy and vertical force Fz (bottom). (Caldwell et al. 2014.)

Usually, the GRF vector is displayed as separate figures for each of its components (figure 16b). There is no general agreement on sign conventions (Whittle 2007, 80-84) but typically on global coordinate system Z-axis is in the vertical direction (positive upwards), Y-axis is horizontal in the direction of progression (positive anteriorly), and X-axis is perpendicular to these two axes (positive to the right). Though, each of the force plate manufacturers may have their own convention for plate reference system axes. The vertical component of the GRF (vGRF) during gait is characterized by two humps and many times a slight spike right after initial contact. (Caldwell et al. 2014.) The vGRF is one of the clinically relevant GRF

characteristic to walking accompanied by horizontal shear forces, vertical alignment of the force vector and COP. The two humps in vertical component (figure 16b) represent changes in the center of gravity (COG) during the early (full weight bearing) and late stance (plantarflexion push-off) while partial unloading (mid-stance valley) happens during mid-stance when body progresses over the stationary foot. Usually, gait also generates a sharp impact force (50-125% BW) at initial contact, a heel transient. In walking vertical forces oscillate around about ±30% of body weight level, but slower walking reduces the vertical acceleration resulting decrements of the peaks and depth of the mid-stance valley bringing the vGRF close to the bodyweight. Mediolateral forces are very small, and normally the force vector points most of the time towards the side of the swing leg to keep the balance. In anteroposterior direction braking phase consists of the first half and propulsion phase the latter half. (Whittle 2007, 80-84; Perry & Burnfield 2010, 459-463.) Pathology or crutches can limit the rate of the limb loading. As children with CP, many times have problems supporting their bodyweight, balancing at one leg stance and generating plantarflexion propulsion for push-off, reduced vertical and anteroposterior GRF in late stance and impaired ankle power has been reported in comparison with TD children (Williams et al. 2011; Lorenzo et al. 2018).

4.1.4 Electromyography

Electromyography (EMG) is a measurement technique for evaluating electrical activity signals produced by skeletal muscle fibers. Muscle activity graphed as EMG (figure 17) represents the summation of action potentials by multiple motor units under each electrode's detection area. In gait analysis it is common to include kinesiological (dynamic) electromyography by using surface electromyography (sEMG) technique, for example, to quantify the relative intensity of muscle function and timing of on-off signals of individual muscles or muscle groups indirectly. (Perry & Burnfield 2010, 425-456.) Other typical factors interpreted from EMG data for gait analysis are evidence of spasticity and hypertonia, selective muscle control and coordination. EMG interpretation contains various challenges starting from issues in signal recording and processing to the changing muscle function during complex movements. Also, the speed of walking plays a role in EMG interpretation for gait analysis. (Gage & Schwartz 2009.)

For sEMG, standardized electrode placement and measurement recommendations are given by SENIAM project (Surface ElectroMyoGraphy for the Non-Invasive Assessment of Muscles). Recommended inter-electrode distance for bipolar electrodes is 20 mm and sensor locations are described as a point on a line between two anatomical landmarks for each muscle. For example, sEMG electrode application for tibialis anterior muscle is at 1/3 on the line between the tip of the fibula and the tip of the ankle's medial malleolus. (SENIAM 2006.)

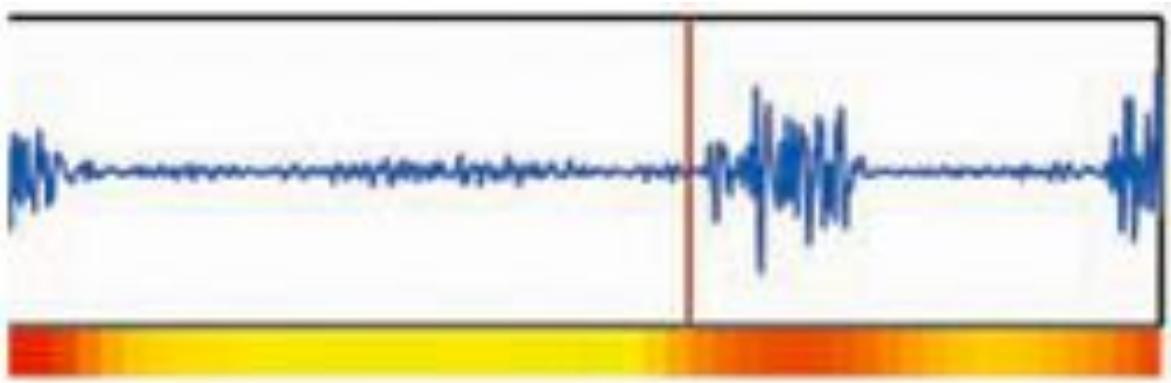

Figure 17. Example of a raw sEMG signal of tibialis anterior muscle during a gait cycle. The red vertical line represents toe off, and the bottom bar demonstrates muscle activation timing, lesser activity in yellow and stronger activity in red. (Gage & Schwartz 2009)

The timing of muscle actions during gait cycle is muscle specific as each muscle has its pattern of onset and cessation (table 2) which are rather consistent regardless of the speed of walking in children and adults. The onset timing can be deviant in people with neuromuscular deficits such as in CP, or it may be related to the mechanics or position of the joints. Timing of excitation could give insights for applications in musculoskeletal modeling and motor control in addition to understanding motor control and coordination in pathologies. (Gage & Schwartz 2009.) Perry and Burnfield (2010, 425-456) defined minimum criteria for the identification of onset and cessation points with computer analysis as minimal signal intensity being at least 5% of the maximum manual test EMG, and the minimum duration of muscle action is at least 5% of the gait cycle. Threshold-based methods have been easy to implement during computerized EMG signal analysis offering rapidity with large data sets and much better reliability compared with visual detection. Still, other more precise objective motor response onset detection algorithms have been developed, especially for measurements with participants having central motor disorders as the muscle activation profile can have larger

variability. Algorithms based on approximated generalized likelihood ratio (AGLR) decision rule method are found successfully detect events automatically in sEMG signals and being superior to threshold-based methods. (Staude & Wolf 1999; Solnik et al. 2010.) In addition to normal signal processing procedures (e.g., filtering and rectification) and statistical testing algorithms to detect onset events, the signal can also be conditioned with Teager-Kaiser energy operator (TKEO) to minimize erroneous onset detection and to improve the signal-to-noise ratio (Solnik et al. 2010).

Perry and Burnfield (2010, 425-456) described the seven-part classification of abnormal timing activity, including deviations: premature, prolonged, continuous, delayed, curtailed, absent, and out-of-phase. The additional muscle activity periods in premature or prolonged activation patterns may extend to another phase of the gait cycle and hence affect walking. It may hinder desired limb function adding muscle co-contraction to wrong gait phase or positively support for the abnormal joint position. Inhibiting the original motor action, the co-contraction may signal about poor motor control (Gage & Schwartz 2009). When some or all the desired muscle activity is missing the muscle activity timing is described as delayed, curtailed, or absent depending on the characteristics. Continuous muscle activity throughout the gait cycle is prevalent in the CP population and is always unwanted. Activity pattern being out-of-phase shows muscle activity when not expected and vice versa. It is important not to interpret EMG isolated from the kinematics of the limbs and joints to form a complete picture of the reasons behind the abnormal pattern. (Perry & Burnfield 2010, 425-456.)

TABLE 2. Muscle sequence controlling the foot joints (% gait cycle) (Perry & Burnfield 2010, 155).

Muscle	On	Off	Peak
Tibialis anterior	56	13	0
Soleus	7	52	43
Gastrocnemius	9	50	40

5 PURPOSE OF THE STUDY

According to Surveillance of Cerebral Palsy in Europe (2002), CP is the most common neurological disorder in children affecting about 2 of every 1000 children born in Europe. As there is currently no definitive cure for CP, it will persist and generate high treatment costs through one's lifespan (Rosenbaum et al. 2007). Even without a definitive cure for CP, the rehabilitation and medical care can minimize the deterioration effects to functional abilities during child- and adulthood to help people with CP survive to adulthood (Graham et al. 2016). Verschuren et al. (2016) published first exercise and physical activity recommendations specifically for people with cerebral palsy and messaged that there is a pressing need for more evidence-based research about adaptation mechanisms to training interventions in CP population. Prescription of resistance training exercises for individuals with cerebral palsy can be improved with knowledge of the biomechanics of walking, for example, via gait analysis (Williams et al. 2019). This study can show does the tailored exercise therapy intervention, based on the Verschuren et al. (2016) recommendations, consisting of two to three supervised sessions per week provide benefits in walking ability and related issues, first time for people with CP in Finland. This intervention description could be then used, modified, and adopted in clinical usage to possibly change practices even around the world. In addition to that, earlier exercise interventions have been demonstrated the gain of individual benefit in physical capacity and motor competence for each participant of the studies. CP has diverse clinical manifestations, and thus analyzing individual responses is also important. No studies have yet researched effects of three-month exercise therapy intervention, featuring multiple different types of training, to lower-body kinematics during gait in adolescents and young adults with CP.

The main research questions of this study are:

1) How tailored training intervention affects the lower body joint kinematics and vGRF during gait measured using CGA and walking performance measured using 6MWT?
   It is hypothesized that the intervention including both treadmill training and muscle strengthening will enhance walking speed in 6MWT (Moreau et al. 2016; Booth et al.

2018), improve gait kinematics towards values of typically developed and increase dorsiflexion at initial contact combined wider ankle ROM during gait (Willersley-Olsen et al. 2015; Kirk et al. 2016).

2) How tailored training intervention changes the motor control of the shank muscles measured as EMG onset timing?

   Erroneous antagonist co-contraction is shown to decrease the gait efficiency in CP (Unnithan et al. 1996) and always unwanted continuous muscle activity throughout gait cycle is many times prevalent in CP population (Perry & Burnfield 2010, 425-456). It is hypothesized that task-specific treadmill training would develop children's motor coordination (Graham et al. 2016) and hence get the shank muscles contract at the correct times during gait.

3) Does dorsiflexion/plantarflexion maximal torque production increase after the intervention?

   Walking ability is related to muscle strength in children with CP and the muscle weakness is the most noticeable in muscle groups around the ankle (Eek & Beckung 2008). Strength training has been proven to increase muscle strength and force production in people with CP (Dodd et al. 2002; Eek et al. 2008). Thus, it is hypothesized that intervention including regular strengthening exercises for shank muscles responsible for dorsiflexion or plantarflexion will increase the maximal torque measured with ankle dynamometry.

6 METHODS

6.1 Participants

The study was included within a broader project, the EXECP research project. A convenience sample of three male adolescents and young adults with spastic CP (hemiplegic or diplegic) aging from 16 to 21 years old participated in the study. Participants' GMFCS levels (I and III) were acceptable for the study (GMFCS I-III). Participants were recruited in Jyväskylä by public advertisements, information events, health center and hospital contacts and with the help of Central Finland CP association (Keski-Suomen CP-yhdistys ry). Exclusion criteria also included serial casting, surgical procedures of the lower limbs, dorsal rhizotomy, pharmacological treatments (e.g., botulinum toxin and intrathecal baclofen) in the last six months, dystonia, inability to understand verbal instructions, and inability to stand with the foot flat on the floor. Each participant is handled in the study as an individual case due to their differences in GMFCS and appearance of cerebral palsy.

Case 1 (C1). Participant C1 had spastic diplegia, and his functional capabilities were evaluated as GMFCS level III. Normally, he walks with knee ankle foot orthosis (KAFO) and crutches, but at longer distances (over 500 meters) a wheelchair is used. C1 could not stand-alone on a whole sole on the ground without KAFO and walking aid, so crutches were used during gait measurements. No orthosis was used during any of the laboratory measurements in the study. Visible flexed knee gait pattern was noticed at the first study visit while wearing no orthosis. Standing stance with crutches showed features of "lever arm disease" where out-toed stance and crouch gait pattern is present due to midfoot break and lateral tibial torsion. Torsional deformities of the long bones in the lower extremities are typically found together with contractures in spastic diplegia. Weakened triceps surae complex is thus unable to control the progression of the tibia during the mid-stance phase putting more demands on the quadriceps and possibly contributing to crouch pattern. (Rodda & Graham 2001). C1 did not have any recent surgical operations but had undergone hamstring tendon lengthening five years before measurements. Before and during the study he regularly participated in physical activities such as adapted ball games once a week and had physiotherapy once a week.

Case 2 (C2). Participant C2 had spastic left side hemiplegia. He had no recent surgeries, but Achilles tendon lengthening was done at the age of four. Cerebral palsy was combined with epilepsy and slight intellectual disability which did not cause any problems in communication with researchers. He was able to walk without walking aid in all situations during the measurements and could stand the whole sole fully on the ground. (GMFCS level I). Before the study, he had participated very actively to different organized physical activities such as adapted hockey, up to 5 times per week, and had physiotherapy once a week.

Case 3 (C3). Participant C3 had spastic diplegia combined with left leg peroneus paresis due to incomplete tendon surgery at the age of 11. Paresis has recovered to some degree in the last decade. He uses crutches for ambulating in his everyday life and can stand sole fully on the ground while supported with walking aid. Medication includes daily oral baclofen to reduce the effects of spasticity. C3 had earlier in the childhood undergone Achilles tendon lengthening and multiple tendon lengthening operations for knee and adductors. At the time of the study, he did not participate in organized physical activity, and hence his training frequency during the intervention was higher than in C1 and C2. Normally, he visited physiotherapist twice a week, while the visits included balance, strength training, and stretching.

6.2 Study protocol

The study was included within a broader project, the EXECP research project, which investigates the effects of 3-month individualized exercise therapy intervention on neuromechanical and metabolic characteristics of children and young adults with CP. In addition to the gait performance and muscle strength measurements in this study, the participants underwent measurements of corticospinal excitability, cortical proprioceptive processing measured with magnetoencephalography, joint range of motion, daily physical activity level, psychological questionnaires, and cardiometabolic risk factors during the project. Each CP participant had two pre-tests separated with three months, followed by a three-month intervention period, and an immediate post-test and a second post-test after the three-month maintenance period concluding the whole 9-month study period. (ISRCTN

2019.) Participants participated in the familiarization session one week before the pre-test 1. In the familiarization session, participants experienced the laboratory facilities, gait analysis procedure, strength, and flexibility tests. This longitudinal study focused on the time and measurements at pre-test 2, intervention period, and post-test 1 (figure 18). Studying the longitudinal effects of the exercise intervention, pre and post-tests included the same measurements of gait analysis, ankle dynamometry, and 6MWT. The ethical approval for the study was given from the Central Finland Hospital District ethics committee, and both participants and for children under 18 years also their legal guardians signed the informed consent to participate in the study. Participation to the study was entirely voluntary.

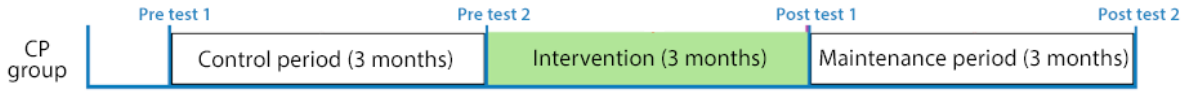

FIGURE 18. Whole 9-month experimental protocol design for each participant. This study focused on the time and measurements between Pre-test 2 and Post-test 1.

6.2.1 Training period

Recent research on positive effects of strength and aerobic training in people with CP has brought up multiple research projects with different kind of training protocols trying to bridge the gap between basic science and clinical relevance (Gillett et al. 2015; Ryan et al. 2016). In this study, the exercise therapy intervention of twelve weeks consisted of two to three supervised sessions (á 90 minutes) per week, and each session was interspaced by at least 48 hours. C1 and C2 had two supervised training sessions as they were engaged actively to physiotherapy and other organized physical activity during their free time. C3 did not participate in other organized physical activity than weekly physiotherapy visit, thus having three supervised training sessions per week in this study. The intervention was individualized to take care of deficits recognized by instrumented gait analysis, flexibility, and strength tests. Each training session (table 3) during intervention started with 5-10 minutes of gait training on a non-motorized 6° or 7.3° incline treadmill (Vida XL, Venlo, Netherlands) with hands supported, was followed by strength (60-75 minutes) and flexibility training (10-20 minutes) for main lower limb muscles. Participants were instructed and given constant verbal feedback during gait training to walk at a comfortable speed, trying their best to achieve heel strike and

thus avoiding the toe walking pattern. To maintain walking quality, the 10-minute gait training was performed in multiple parts to give enough rest for the participant. Strength training was performed using both weight stack machines and free weights for finding the best modification and progression for the participant on each training session. A total of 10 different exercises were selected for the intervention, half of them being single-joint exercises. The strength training program had two protocols performed alternating training sessions, each protocol consisting of 8 to 10 exercises for lower limbs and trunk muscles. Strength training prescription respected the American College of Sports Medicine and National Strength and Conditioning Association guidelines (Faigenbaum et al. 2009; Garber et al. 2011) and guidelines of physical activity for cerebral palsy (Verschuren et al. 2016). Participant's abilities and limitations were taken into account while designing and performing the training sessions. Participants received the non-motorized incline treadmill also at their homes and were encouraged to walk and train the toe clearance phase of gait a minimum of 10 minutes per day during the intervention.

TABLE 3. Detailed prescription of the training types during the sessions in the intervention period.

Type	Task	Training load	Details
Strength	Isokinetic, Isometric, concentric and eccentric	7 to 10 exercises, 2-4 sets of 6-15 repetitions. 1-2 minutes of rest. 2-3 times a week. Movement tempo: concentric: 1-3 s, eccentric: 2-3 s Duration: 60-75 minutes.	Multi and single joint exercises. Biofeedback may be utilized. Progression by increasing intensity and decreasing volume of repetitions.
Flexibility	Passive-static Stretching	4 x 45s per muscle, maximal tolerable intensity. 2-3 times a week. Duration: 10-20 minutes.	Target muscles: triceps surae, hamstrings, quadriceps and hip flexors.
Gait training	Walking	Focusing on the quality of walking, especially on the toe clearance. Every training session and daily at home Duration: 5 to 10 minutes.	Five minutes warm-up and cool down will be performed.

6.2.2 Testing protocol

Gait evaluation testing session of two to three hours involved the instrumented gait analysis measurement, six-minute walking test (6MWT), but also blood and body composition measurements. The strength measurements done in the ankle dynamometry were done at a different day interspaced of 48 hours with other testing sessions. The goal of the 6MWT is that the participant attempts to walk the longest distance possible in 6 minutes (Maher et al. 2008). All equipment related to the gait analysis (i.e., EMG, reflective markers, footswitch) were removed before the test.

Testing session protocol:

- Blood sampling (participants are under fasting condition).
- Body composition tests (Inbody, height), arterial stiffness.
- The meal is served. Participants can rest quietly for about 10 minutes.
- Participant preparation at the biomechanics laboratory (reflective markers, EMG electrodes, heel switch).
- Warm-up protocol and ensuring measurement quality (walking in submaximal and then in maximal speed).
- 6x1-minute instrumented gait analysis.
- Rest of 15 minutes.
- 6-minute walking test (6MWT) in a 30-meter-long indoor corridor.
- Session end.

6.3 Gait analysis data collection and Visual3D analysis

In the gait analysis session, participants performed six times one-minute walking trials with one-minute seated rest between trials. Work-to-rest ratio 1:1 was chosen to avoid fatigue and its effect on changing walking mechanics. Work-to-rest ratio was modified with more rest periods, if it was necessary for the participant, nevertheless a total of 6 minutes of walking was always recorded. Trials were performed by walking back and forth on a 7,41-meter indoor walking path of rubber matting marked with tape (figure 19). The three-minute warm-

up consisted of comfortable submaximal walking bouts to get familiar with the walking path, turns between walking bouts and wearing the instrumentation during walking. At the same time, the motion capture, EMG, and foot switch function were tested. During the measurement trials, participants walked at a self-selected maximum walking speed while being constantly encouraged by researchers to walk at their maximal speed. To study onset timing of three shank muscles during walking, bipolar self-adhesive EMG electrodes (Blue Sensor N, Ag/AgCl, Ambu, Ballerup, Denmark) were placed in tibialis anterior (two pairs, distal and proximal electrodes at least 10 cm apart), soleus and medial gastrocnemius, and connected via wireless EMG system (TELEmyo 2400T G2, Noraxon, Scottsdale, AZ, USA) at 1.5 kHz sampling frequency. Electrode placement and skin preparation were performed according to SENIAM recommendations (Seniam 2006). For detecting heel strike and toe-off events and determining gait cycle in relation to EMG onset measurement, participants wore foot switch sensors (Noraxon, Scottsdale, AZ, USA) attached to their sole under or over the socks inside the shoes on the heel and forefoot. Synchronized foot switch and EMG data were recorded with the Spike2 software (CED, Cambridge, UK). Lower limb 3D kinematics were acquired with an eight-camera motion capture system (Vicon Motion Systems, Oxford, UK) at 200 Hz and forces were simultaneously measured with two mounted force plates at 1 kHz sampling frequency (AMTI OR6-6-2000, AMTI Inc., Watertown, USA) (figure 19). Force plates were connected via Strain Gage Amplifier (AMTI MiniAmp MSA-6) and analog-digital-converter to motion capture core unit (MX Giganet) where the force and motion capture signals were synchronized. The participant was asked to walk normally and not to try intentionally step always on the force plate. If the participant did not feel fatigued while walking the couple first minute-long trials, the total of six minutes of walking could be divided to longer than minute bouts. System calibration was done with the Vicon 5-marker passive calibration wand.

Anthropometrics (body mass, height, leg length, ankle and knee width) were measured before the gait analysis trials, and participant's information was updated to the motion capture software for scaling the model correctly using marker locations at every measurement session. Total of 25 retroreflective markers (13mm diameter) were attached to the trunk and lower body, but 16 of them at anatomical landmarks defined by Plug-in Gait lower body model were used in the motion analysis and are described in table 4 and figure 14a. Additional markers in

the medial ankle malleolus and medial femoral epicondyle, two-marker thigh cluster and sacral marker for accelerometer were not used in this study. Participant wearing markers and instrumentation was calibrated for the motion analysis with the static standing trial.

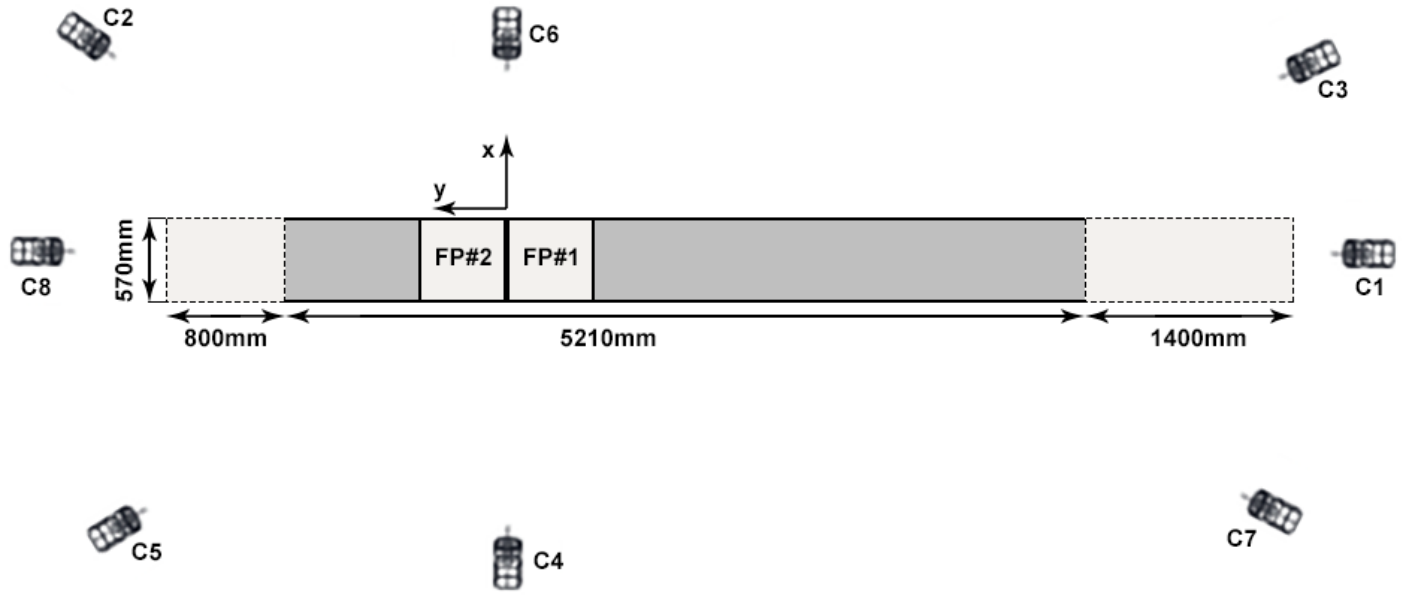

FIGURE 19. Motion capture setup (Vicon Motion Systems Ltd., Oxford, UK) in the biomechanics laboratory with eight Vicon MX T40(-S) infra-red LED cameras pictured from above. Cameras were mounted at different heights to optimize capture when measuring walking with crutches. Dashed lines indicate the turn areas, and the grey fill the main measurement volume. Two force plates (FP#1 & #2, 51 cm x 46 cm, AMTI OR6-6-2000, AMTI Inc., Watertown, USA) provided ground reaction force data, and initial contact and toe-off events to teach the gait events algorithm in Visual3D.

This study utilized the Vicon Plug-in Gait lower body model, which is the implementation of the Conventional Gait Model and is based on the Newington-Helen Hayes gait model. After recording the motion capture data in the Vicon Nexus 2.5 software the camera data of marker locations were reconstructed, the trial was cropped to the length of interest, markers were labeled with accurate marker names, and the marker data was gap filled if needed. Then the data was exported to the Visual3D biomechanics analysis tool (C-Motion Inc. Germantown, MD, USA) where the kinematic modeling and analysis were performed. Using the Plug-in Gait marker set, the Plug-in Gait model was implemented in Visual3D by using segment optimization (6-DOF method) instead of Vicon Nexus Direct Pose Estimation (Direct Kinematics). The results of implementation may differ slightly from Vicon's Plug-in Gait

implementation, but the differences are small and not clinically significant. To calculate ankle, knee and hip joint, and pelvis angles (figure 20), in segment optimization, the body is presented via a biomechanical model, which is a collection of rigid segments representing skeletal structures. Segment optimization tracks each segment independently and hence does not have exact linkage connecting two segments. This provides a possibility to track real joint movements as many of the lower body joints are not correctly presented as a fixed axis. With three markers per segment, a segment can be described using three position coordinates (e.g., X, Y, Z) and three rotational variables (θx, θy, θz) describing rotations around the local coordinates system's main axes. (C-Motion Inc. 2017a.) Hip joint centers were calculated using Davis regression equations (Davis et al. 1991), and other joint centers were calculated using measured ankle or knee width, marker size, and position.

TABLE 4. Markers and their locations in the Plug-in Gait lower body model (Vicon, 2019c), see also figure 14a.

Marker	Location	Segment
LASI & RASI	Left & right anterior superior iliac spine	Pelvis
LPSI & RPSI	Left & right posterior superior iliac spine	Pelvis
LTHI & RTHI	Over the lower (L) or upper (R) lateral 1/3 surface of the left thigh	Femur
LKNE & RKNE	On the flexion-extension axis of the knee on the lateral side	Femur
LTIB & RTIB	Over the lower (L) or upper (R) 1/3 surface of the right shank	Tibia
LANK & RANK	On the lateral malleolus on the line of the transmalleolar axis	Tibia
LHEE & RHEE	On the calcaneus at the same height above the plantar surface of the foot as the toe marker, on the shoe surface.	Foot
LTOE & RTOE	Over the second metatarsal head, on the mid-foot side of the equinus break between forefoot and mid-foot, on the shoe surface.	Foot

The output joint angles expressed as Cardan angles were defined as relative orientations (3D rotation matrix) of one LCS with adjacent segment's LCS, for example, shank LCS

("moving" distal segment) relative to thigh LCS ("fixed" proximal segment) constitutes knee angles (figure 15b). To derive the three anatomically and clinically relevant joint angles (flexion-extension, abduction-adduction, and rotation) for lower extremities, Cardan sequence of three elementary rotations X-Y-Z was used to calculate Cardan angles (Hamill et al. 2014). Similarly, ankle angles were defined as foot segment relative to the shank and hip angles as the orientation of the thigh segment relative to the pelvis segment. Pelvic angles were calculated as Cardan angles between the pelvis LCS and (virtual) laboratory coordinate system using the axial rotation-obliquity-tilt sequence (Z-Y-X Cardan sequence).

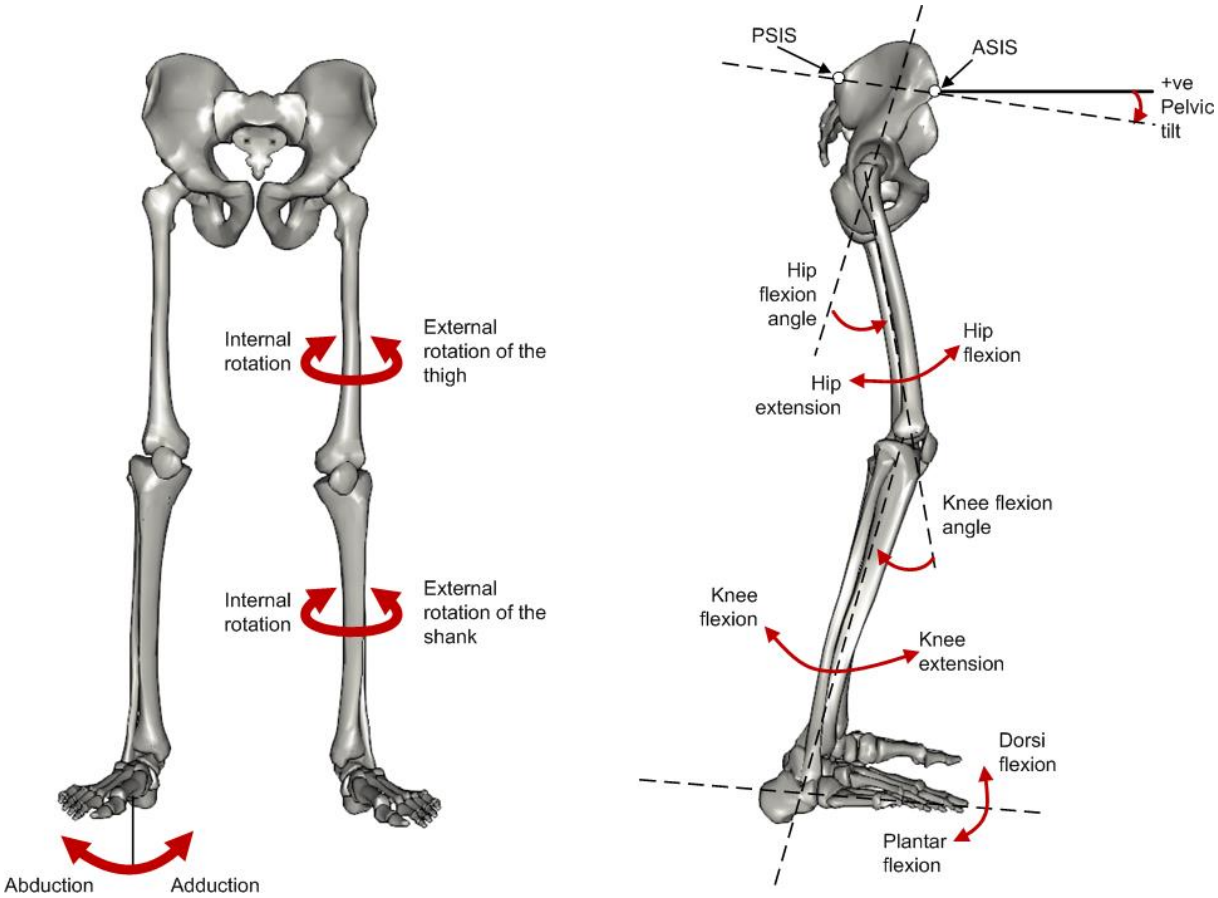

FIGURE 20. Joint kinematic variables provided by Plug-in Gait modeling. (Vicon 2017).

6.4 Dynamometry

Unilateral ankle plantarflexion and dorsiflexion force measurements in both isometric and isokinetic (14°/s) situations were performed in a custom-built ankle dynamometer (figure 21)

(University of Jyväskylä, Finland). Participants were seated in the chair with the knee fully extended and foot pedal in the 0° position where the sole is perpendicular to the shank, or 28° into plantarflexion. The angle of the back of a chair was determined at 60° hip angle. Participant's both legs were tested, and both dorsiflexion and plantarflexion were measured, if it was possible for the participant. Three to five trials with 1-2 minutes of rest was measured in each test. Both plantarflexion and dorsiflexion tests started with a 2-second maximal isometric muscle action, followed by isokinetic effort (14°/s). Visual feedback and strong verbal encouragement were provided to a participant, and the highest attained torque values of three maximum voluntary contraction (MVC) trials were recorded with two minutes rest in between the trials.

The measured leg was tightly attached to the foot pedal with straps, where a force transducer (Precision TB5-C1, Raute, Nastola, Finland) measured the applied force. Heel rise movement was measured during the trials using the heel sensor under the heel. Foot reaction force and heel position data were collected with 16-bit AD board (CED 1401, Cambridge Electronic Design, Cambridge, UK) at 1 kHz and Spike2 software. The best trial in each test session was analyzed, and torques at 0°, 5°, 10°, 15°, 20°, 25°, and 28° were reported.

FIGURE 21. Measuring CP participant's right ankle force production in ankle dynamometer.

6.5 Data and statistical analysis

Motion and force plate data were captured simultaneously in Vicon Nexus 2.5 software. For each trial of each participant the marker data was first reconstructed, cropped to wanted length, labeled, and marker gaps were interpolated given the data had good quality. Shorter marker gaps were filled automatically with spline fill and longer gaps were filled with the best fill option, but only when the trial included similar motion pattern stride before or after. Even though part of the gap filling was done automatically, the kinematical data was checked manually afterwards. If the marker was missing for the most part of step sequence, the trial was discarded. Kinematic and vGRF data were then exported, and their analysis was performed in Visual3D v6 software. Three-dimensional marker trajectories and GRF data were low-pass filtered using a fourth-order Butterworth filter with a cutoff frequency set at 8 Hz and 50 Hz respectively. The cutoff frequency for gait analysis marker trajectory data was defined after residual analysis (Winter 2005) using the data from the first measurement trials. Frequency spectrum analysis using Fast Fourier transform was done in MATLAB. The decision for 8 Hz cutoff was made as it appeared that for more rapidly moving markers (foot and shank segment markers) higher cutoff frequency (8 Hz) was recommended compared with thigh and pelvis segment markers (6 Hz). For unfiltered vGRF data, it was seen that 99% of the signal included under 44 Hz and thus 50 Hz was a safe choice for cutoff frequency for the low-pass filter. Kinematic data was time-normalized to gait cycle length (0-100%) and averaged over the cycle using the gait events created with force plate assignments and target pattern recognition (TPR) algorithm (Stanhope et al. 1990). Later analysis and graph processing were done in MATLAB using the exported kinematical data in ASCII format. Mean and standard deviations were calculated for each joint angle, kinematical parameter, and spatiotemporal variable.

Concerning the spatiotemporal parameters, walking speed was calculated using the actual stride length divided by actual stride length, as using only fully completed strides in the measurement trials. Step frequency was computed separately for the left and right limb using step time divided by 60 seconds. Step time for left side was determined from right heel strike to left heel strike and for right side the other way around. Stride length is the distance in the walking direction between consecutive proximal end positions (heel strike) of ipsilateral foot

and step length is measured similarly as a distance between heel strikes of the alternating feet. Stride width is the perpendicular (mediolateral) distance from the opposing heel strike position to a stride vector. Double limb support is a sum of initial and terminal double limb support. Initial double limb support begins with the heel strike of the foot in question and ends when contralateral foot toe-offs. Terminal double limb support phase begins with contralateral foot heel strike and ends when the foot in question toe-offs. (C-motion Inc. 2017b.)

Onset timing analysis of the EMG signal from gait analysis trials was performed in MATLAB to give information about neuromuscular coordination. The recorded EMG signal included synchronization signal from Vicon Nexus software indicating live recording. Only walking strides containing appropriate footswitch data and EMG quality during recording were included in the analysis, totaling at least 50 analyzed strides for each participant from one measurement session. First EMG data was bandpass filtered (10-500 Hz) using a fourth-order Butterworth filter. MATLAB based scripts for detection of the muscle activation intervals from surface EMG signals used algorithms based on AGLR decision rule method and was conditioned with TKEO to minimize erroneous onset detection and to improve the signal-to-noise ratio (Staude & Wolf 1999; Solnik et al. 2010). Onset timing frames were then compared with foot switch onset frames and position in the gait cycle was calculated. Means and standard deviations of the onset timings were calculated to formulate muscle on-off graphs.

7 RESULTS

7.1 Spatiotemporal variables

Spatiotemporal data from instrumented gait analysis revealed very different behaviors after exercise intervention on different participants (table 5). C1 (spastic diplegia) had slower walking speed accompanied by radically lower step frequency (-17.4%, -12.4%, left and right step frequency, respectively) and slightly longer steps (1.6% and 6.6%). Due to slower walking speed, both contact and swing times increased, but the proportion of stance mildly decreased (-2.7%, -0.7%) as swing time increased more than contact time. This was also seen in decreased double limb support (-13.1%). Also, the stride width increased a little (6.7%) at a lower walking speed.

Participant C2 (spastic left side hemiplegia) had the highest walking speed of the participants in the PRE measurements, but it decreased a lot (-22%) in the POST measurements. C2 had a distinct disparity between limbs in step frequency, step length, contact and swing times pointing out the left side hemiplegia with the shorter stance (-6.5% POST), shorter steps (-17.6%) and lower step frequency (-12.9%) in the left side. C1 and C2 slowed their walking differently, C1 increased especially his swing portion of the gait cycle possibly due to using the crutches while C2 stayed more time on the ground and swing time stayed similar PRE and POST.

Participant C3 (spastic diplegia combined with left leg peroneus paresis) increased his walking speed after the exercise intervention (4.8%) accompanied by increased step frequency (6.7% and 0.3% left and right, respectively). Larger increase on left step frequency brought the step frequency to be symmetrical between the limbs in POST measurements (133.2 and 133.8 steps/minute, left and right, respectively). Stride width was narrowest of all three participants and decreased to 7 cm in POST measurements. Large differences between left and right step length were seen (-31.4% shorter in the right side) although the difference reduced a little in POST measurements. C3 bore weight on the right side also relatively shorter duration as stance period lasted 58.7% of the gait cycle compared to 60.5% on the left side. Also, double limb support phases decreased -12.4%.

TABLE 5. Spatiotemporal parameters from PRE and POST gait analysis measurements. Results are presented as mean ± standard deviation and difference between PRE and POST measurements (Diff.). L = Left leg, R = Right leg.

	C1			C2			C3		
	PRE	POST	Diff. (%)	PRE	POST	Diff. (%)	PRE	POST	Diff. (%)
Speed (m/s)	1,23 ± 0,05	1,09 ± 0,08	-11,4	1,64 ± 0,10	1,28 ± 0,12	-22,0	1,25 ± 0,11	1,31 ± 0,08	4,8
Step frequency (steps/minute)	L: 117,6 ± 7,3 R: 119,2 ± 8,2	L: 97,1 ± 8,5 R: 104,4 ± 7,5	L: -17,4 R: -12,4	L: 114,2 ± 5,9 R: 135,9 ± 5,5	L: 107,1 ± 7,1 R: 123,0 ± 7,4	L: -6,2 R: -9,5	L: 124,8 ± 8,9 R: 133,4 ± 11,7	L: 133,2 ± 6,7 R: 133,8 ± 5,9	L: 6,7 R: 0,3
Stride length (m)	1,25 ± 0,05	1,29 ± 0,05	3,2	1,58 ± 0,05	1,35 ± 0,08	-14,6	1,17 ± 0,09	1,18 ± 0,05	0,9
Stride width (m)	0,15 ± 0,02	0,16 ± 0,03	6,7	0,13 ± 0,03	0,15 ± 0,02	15,4	0,09 ± 0,03	0,07 ± 0,02	-22,2
Step length (m)	L: 0,64 ± 0,03 R: 0,61 ± 0,03	L: 0,65 ± 0,03 R: 0,65 ± 0,03	L: 1,6 R: 6,6	L: 0,76 ± 0,03 R: 0,82 ± 0,03	L: 0,61 ± 0,05 R: 0,74 ± 0,04	L: -19,7 R: -9,8	L: 0,72 ± 0,06 R: 0,45 ± 0,05	L: 0,70 ± 0,04 R: 0,48 ± 0,02	L: -2,8 R: 6,7
Contact time (ms)	L: 601 ± 51 R: 623 ± 51	L: 688 ± 60 R: 724 ± 70	L: 14,5 R: 16,2	L: 563 ± 24 R: 617 ± 36	L: 643 ± 53 R: 681 ± 43	L: 14,2 R: 10,4	L: 578 ± 68 R: 562 ± 65	L: 545 ± 28 R: 534 ± 36	L: -5,7 R: -5,0
Swing time (ms)	L: 411 ± 34 R: 396 ± 25	L: 503 ± 61 R: 469 ± 27	L: 22,4 R: 18,4	L: 409 ± 21 R: 348 ± 18	L: 413 ± 27 R: 363 ± 17	L: 1,0 R: 4,3	L: 361 ± 21 R: 372 ± 23	L: 355 ± 16 R: 375 ± 18	L: -1,7 R. 0,8
Stance (%)	L: 59,4 ± 5,0 R: 61,1 ± 5,0	L: 57,8 ± 5,0 R: 60,7 ± 7,2	L: -2,7 R: -0,7	L: 57,9 ± 2,5 R: 63,9 ± 3,7	L: 60,9 ± 5,0 R: 65,1 ± 4,1	L: 5,2 R: 1,9	L: 61,5 ± 7,2 R: 60,1 ± 6,9	L: 60,5 ± 3,1 R: 58,7 ± 3,9	L: -1,6 R. -2,3
Double limb support (%)	21,3 ± 6,5	18,5 ± 5,2	-13,1	22,1 ± 3,2	25,6 ± 3,5	15,8	21,8 ± 7,4	19,1 ± 3,1	-12,4
Initial double limb support (ms)	L: 112 ± 32 R: 106 ± 35	L: 108 ± 24 R: 112 ± 38	-3,6 5,7	L: 92 ± 15 R: 123 ± 16	L: 120 ± 18 R: 148 ± 19	L: 30,4 R: 20,3	L: 80 ± 29 R: 124 ± 40	L: 82 ± 28 R: 102 ± 31	L: 2,5 R: -17,7

7.2 Kinematics

Lower body kinematics on the ankle, knee, and hip from PRE- and POST-measurements for three participants are graphed in figures 22-30. Kinematics is normalized to a whole gait cycle (0-100%). Means and standard deviations of the major sagittal kinematical parameters of interest in this study are demonstrated in table 6. Due to errors in C1 data ankle data, only sagittal data is presented in that joint. There was no increase in dorsiflexion in the terminal swing phase on C1 after the intervention. The dorsiflexion was decreased in IC (L and R) and in TO in right. Also, the ROM of the ankle flexion was less in POST (-8.31° and -12.55°) but then higher knee flexion ROM was found (+7.97° and +5.93°). No effect of second toe-lift in the terminal swing phase after intervention was seen in foot inclination and TOE marker excursion as the increases in TOE marker excursion amplitude during swing can be related to increased hip flexion in a swing phase. For C1, knee has significantly more flexion (40.98-44.57° at IC) in the stance phase (figure 23) than in the typically developed gait, showing the typical effect of crouch gait. Shank rotation moved greatly from the internal rotation towards a slight external rotation on both sides. Hip flexion/extension in C1 follows closely the typically developed gait near the IC but hip is extended more than normally in region of the TO (figure 24).

There were clear differences discovered between the affected and non-affected limb in the C2 lower body kinematics. The non-affected right leg represents a distinct second toe lift (figure 25) seen as increased TOE marker excursion and foot inclination in the terminal swing. At the same time, left foot inclination angle shows a flat-footed initial contact on the affected leg and no clear effect after the intervention (-1.76°, 2.04°, PRE, and POST respectively). Both knees are flexed (30.97-39.38° at IC) significantly more than in typical gait from terminal swing to midstance (figure 26). Left knee is also heavily adducted after the IC and during the pre-swing and swing phase, being present also after the intervention with slightly less adduction (figure 27). Other clear differences compared with the typical gait were left thigh internal rotation during the whole gait cycle and right hip abduction from the TO until midstance. The internal rotation on the left side was decreased after the intervention (figure 28).

More affected left leg displayed different kinematic patterns especially in ankle and knee joint compared with the right leg in the participant C3. C3 walked with crutches during the gait analysis session. The biggest difference in the ankle kinematic patterns is the premature plantarflexion on the left ankle during the midstance (figure 28). At the same time during the midstance, the left knee extends rapidly while the knee was significantly flexed at IC (45.69°, 37.70°, PRE, and POST respectively) (figure 29). Also, the ankle is simultaneously externally rotated during the midstance. Right ankle shows second toe lift and the initial contact is made with the heel as the ankle is in slight dorsiflexion (11.70° and 15.89°) and upwards inclined (9.83° and 4.83°) at IC. Left side improved the toe lift a little in the terminal swing phase when examining the TOE marker excursion, foot inclination angle, and ankle sagittal angle. While the knee and hip sagittal flexion angles decreased in the swing phase on the left leg, most of the increase in toe lift should come from the increased ankle dorsiflexion. Deviations from the typical walking pattern were found on hip abduction on the left side during the swing and hip adduction during the stance on the right side (figure 30).

7.2.1 C1 kinematics

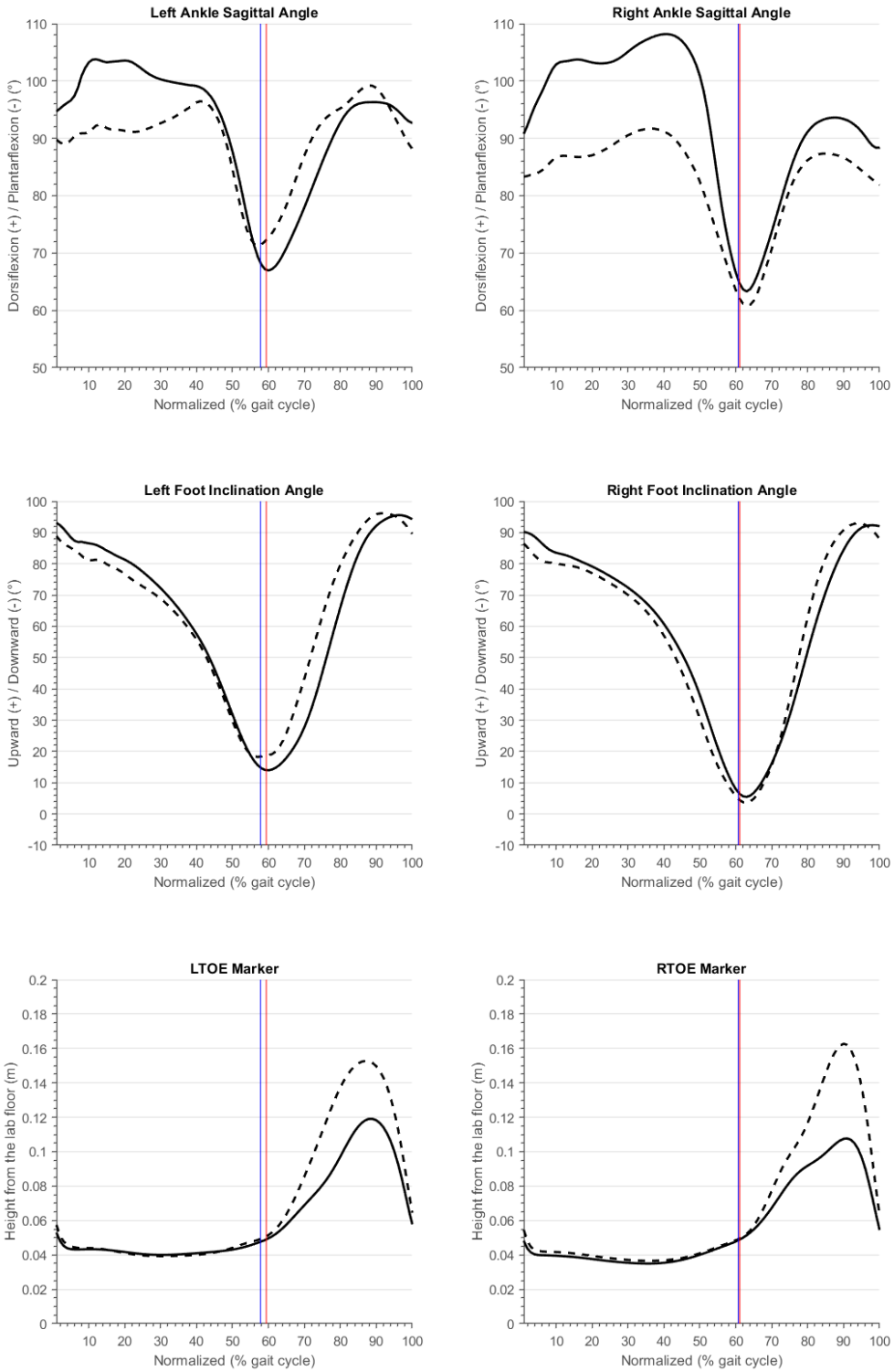

FIGURE 22. C1 Ankle Sagittal kinematics. Solid line and dashed line represent PRE and POST gait, respectively. Red vertical line toe-off in PRE and blue vertical line in POST.

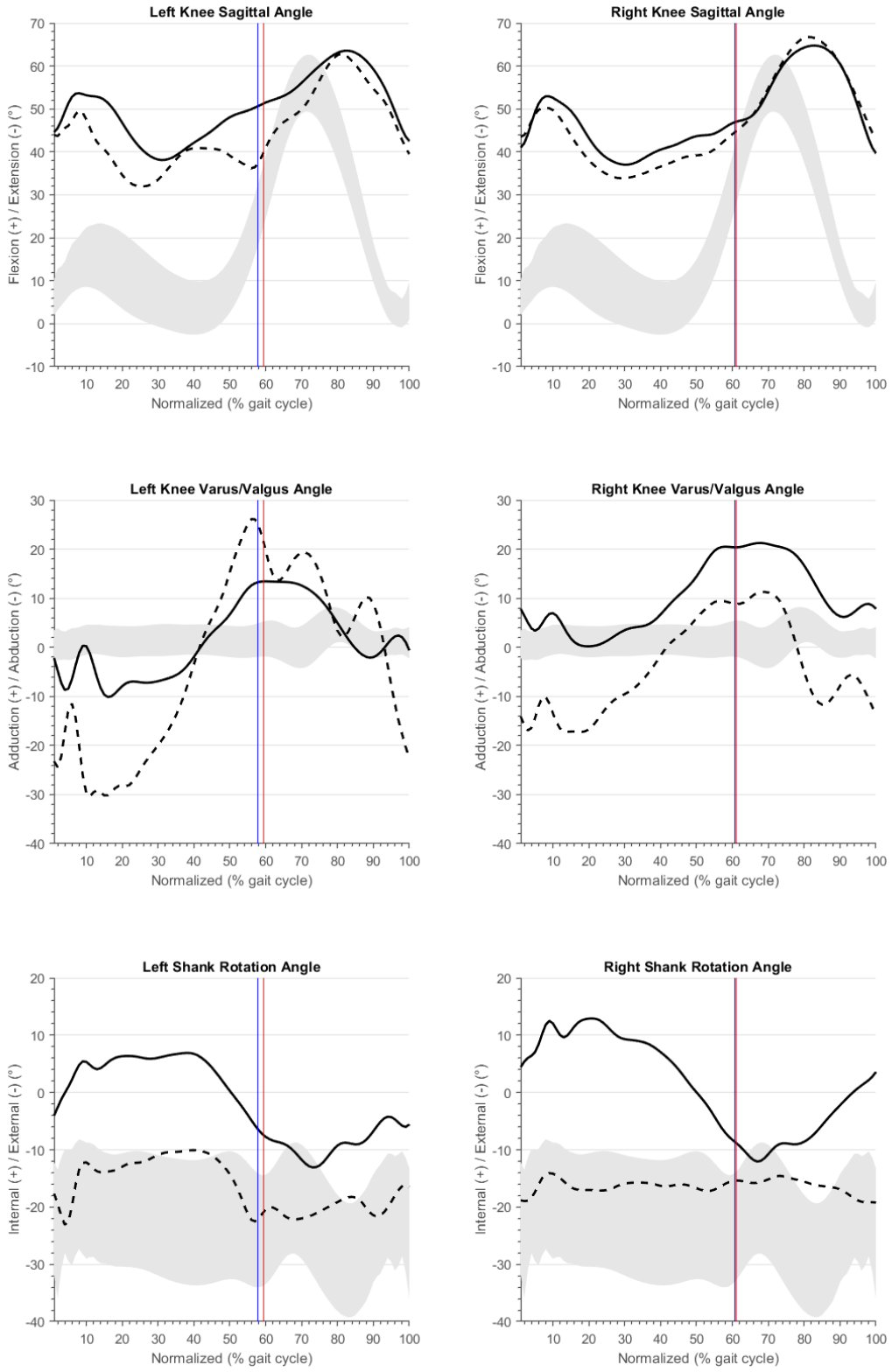

FIGURE 23. C1 Knee angles. Solid line, dashed line and shaded represent PRE, POST and typically developed gait, respectively. Red vertical line toe-off in PRE and blue vertical line in POST.

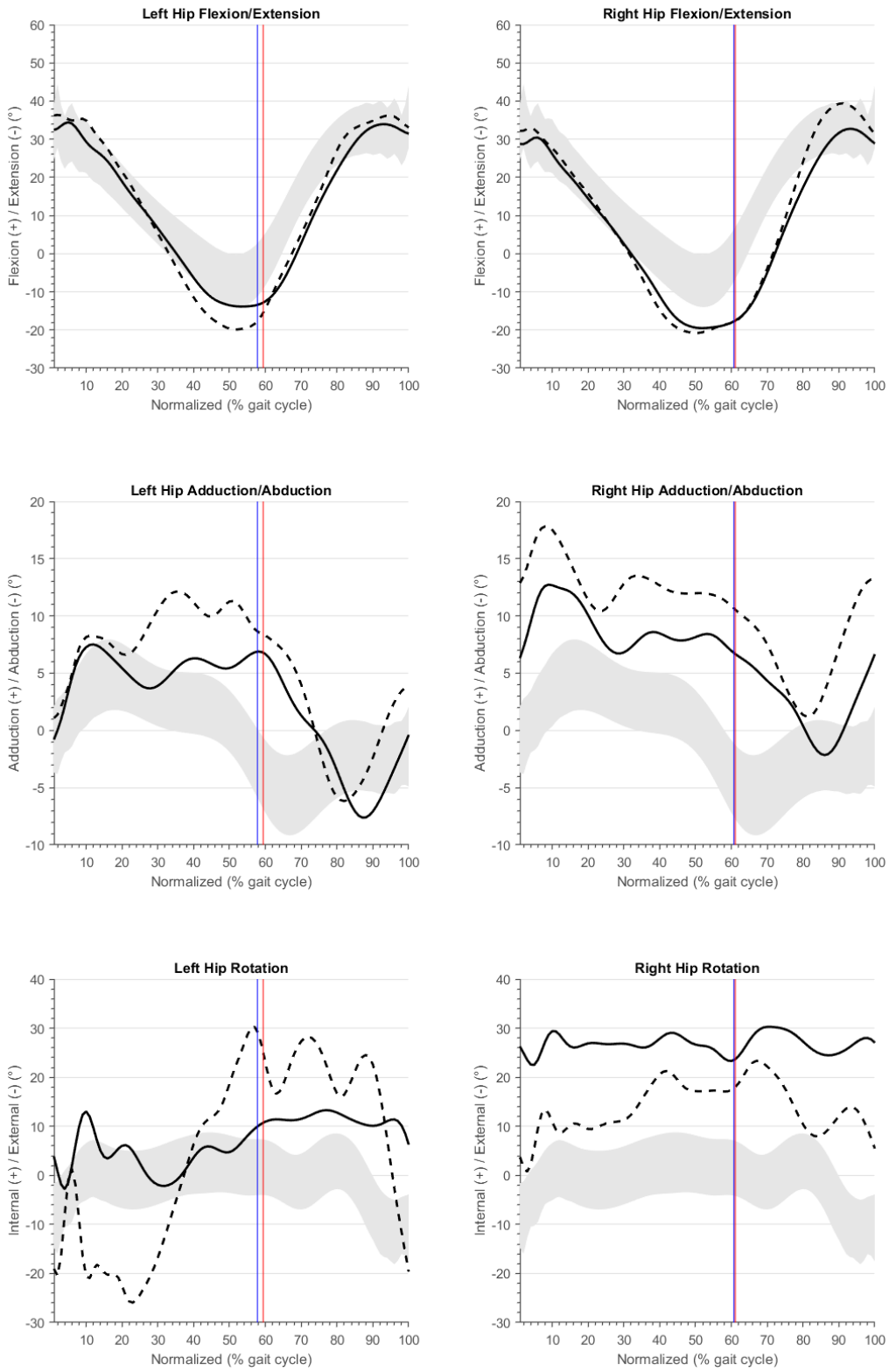

FIGURE 24. C1 Hip Kinematics. Solid line, dashed line and shaded represent PRE, POST and typically developed gait, respectively. Red vertical line toe-off in PRE and blue vertical line in POST.

7.2.2 C2 kinematics

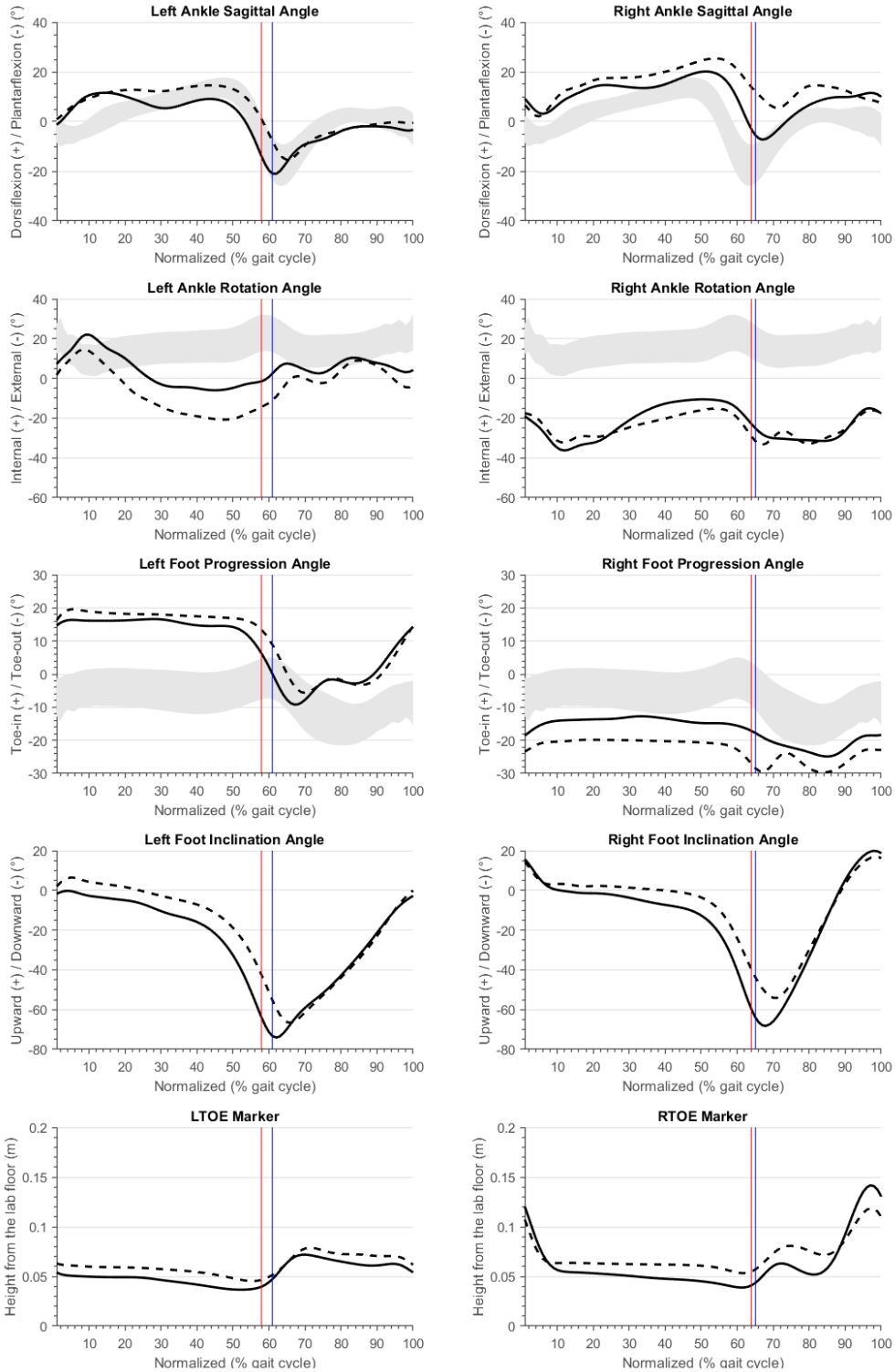

FIGURE 25. C2 Ankle kinematics. Solid line, dashed line and shaded represent PRE, POST and typically developed gait, respectively. Red vertical line is toe-off in PRE and blue vertical line in POST.

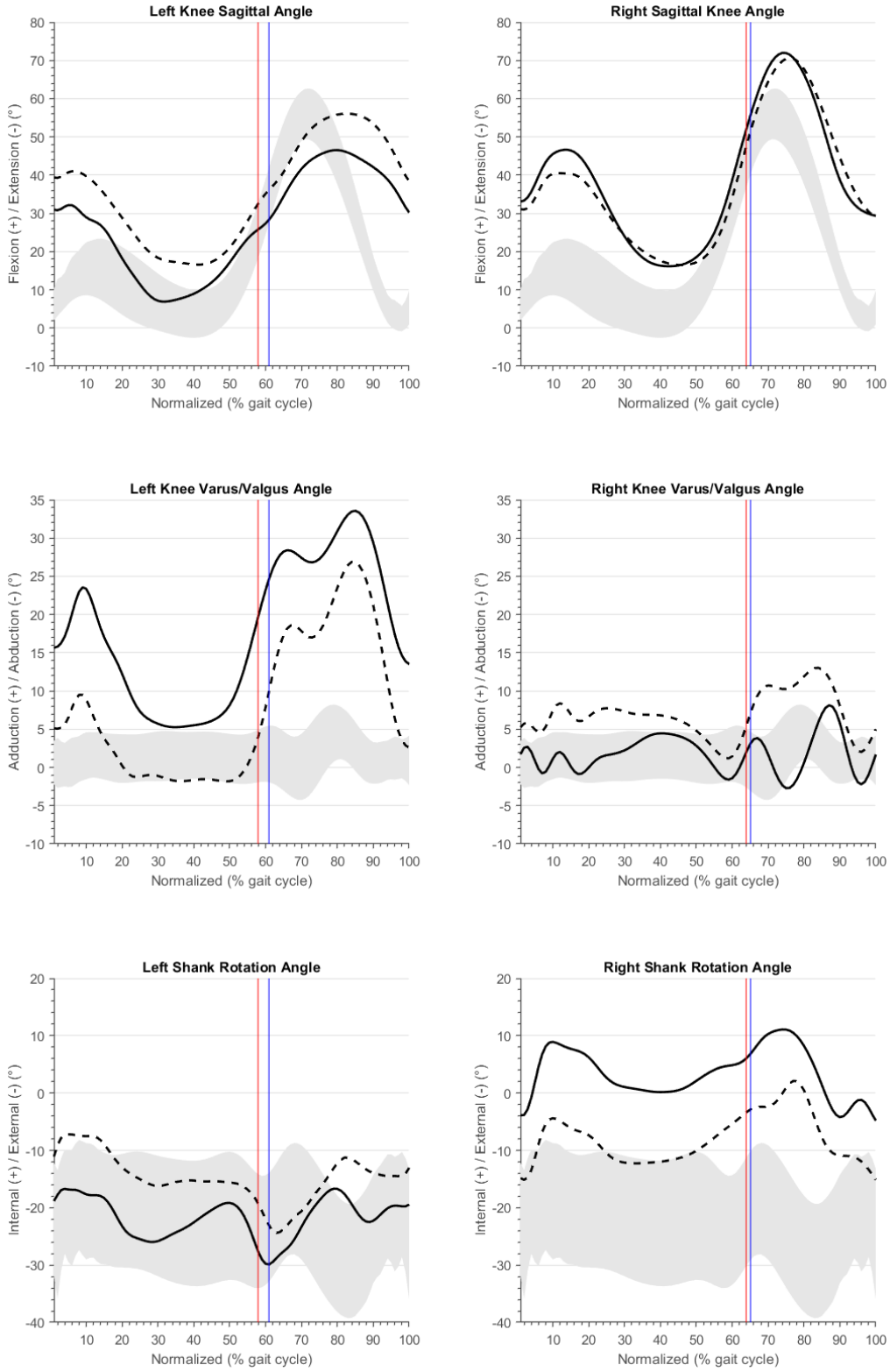

FIGURE 26. C2 Knee kinematics. Solid line, dashed line and shaded represent PRE, POST and typically developed gait, respectively. Red vertical line is toe-off in PRE and blue vertical line in POST.

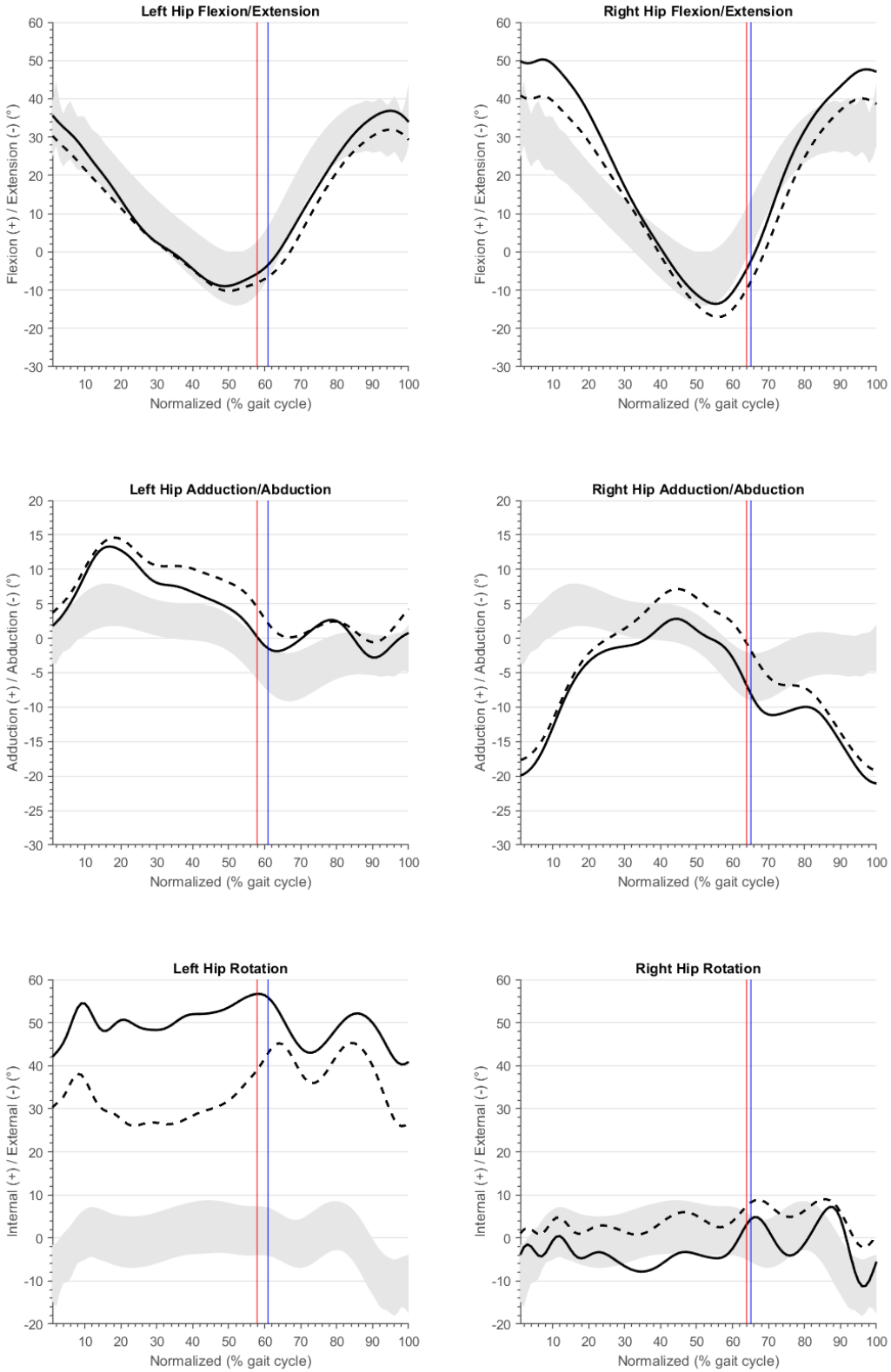

FIGURE 27. C2 Hip kinematics. Solid line, dashed line and shaded represent PRE, POST and typically developed gait, respectively. Red vertical line toe-off in PRE and blue vertical line in POST.

7.2.3 C3 kinematics

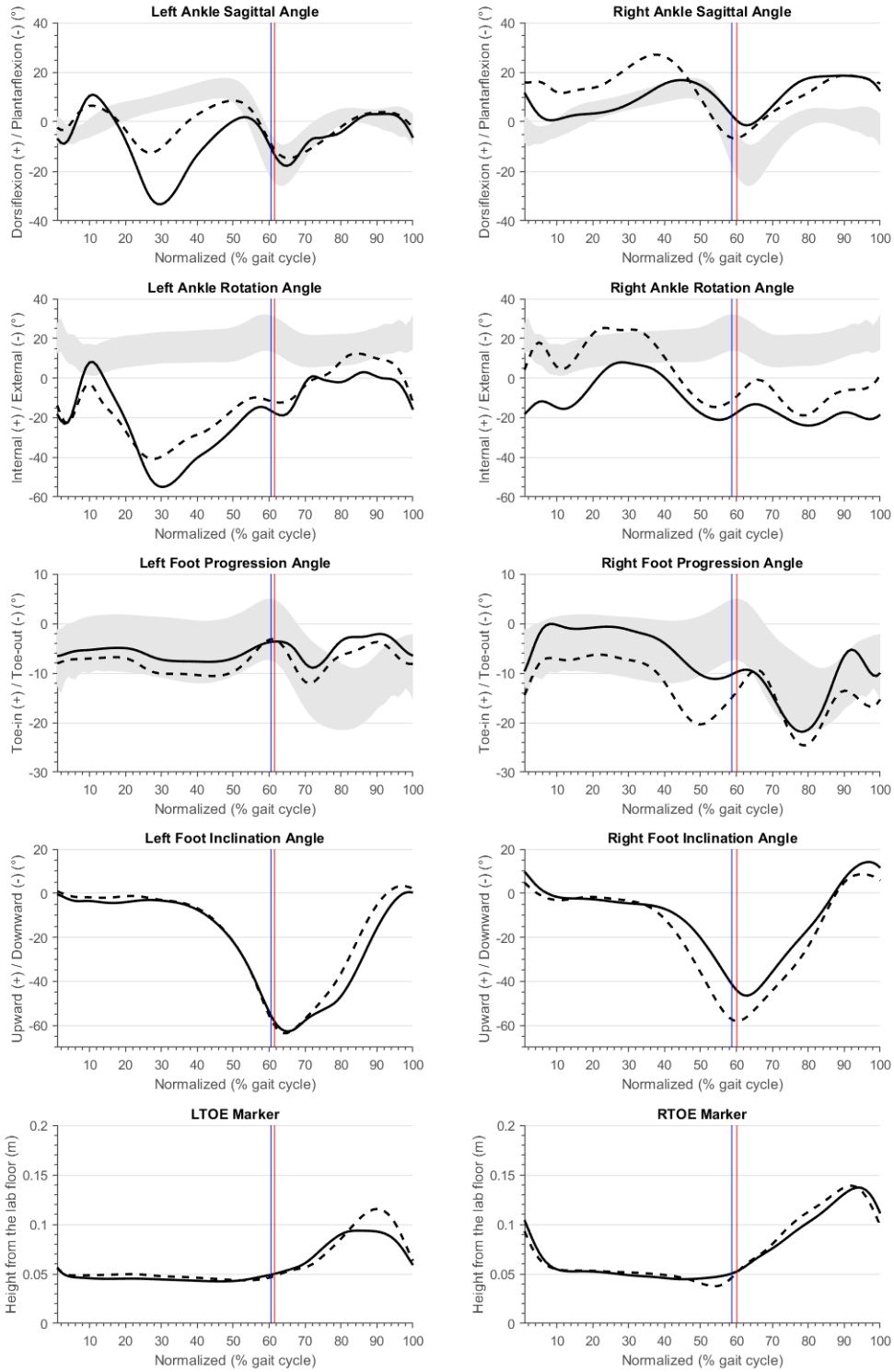

FIGURE 28. C3 Ankle kinematics. Solid line, dashed line and shaded represent PRE, POST and typically developed gait, respectively. Red vertical line toe-off in PRE and blue vertical line in POST.

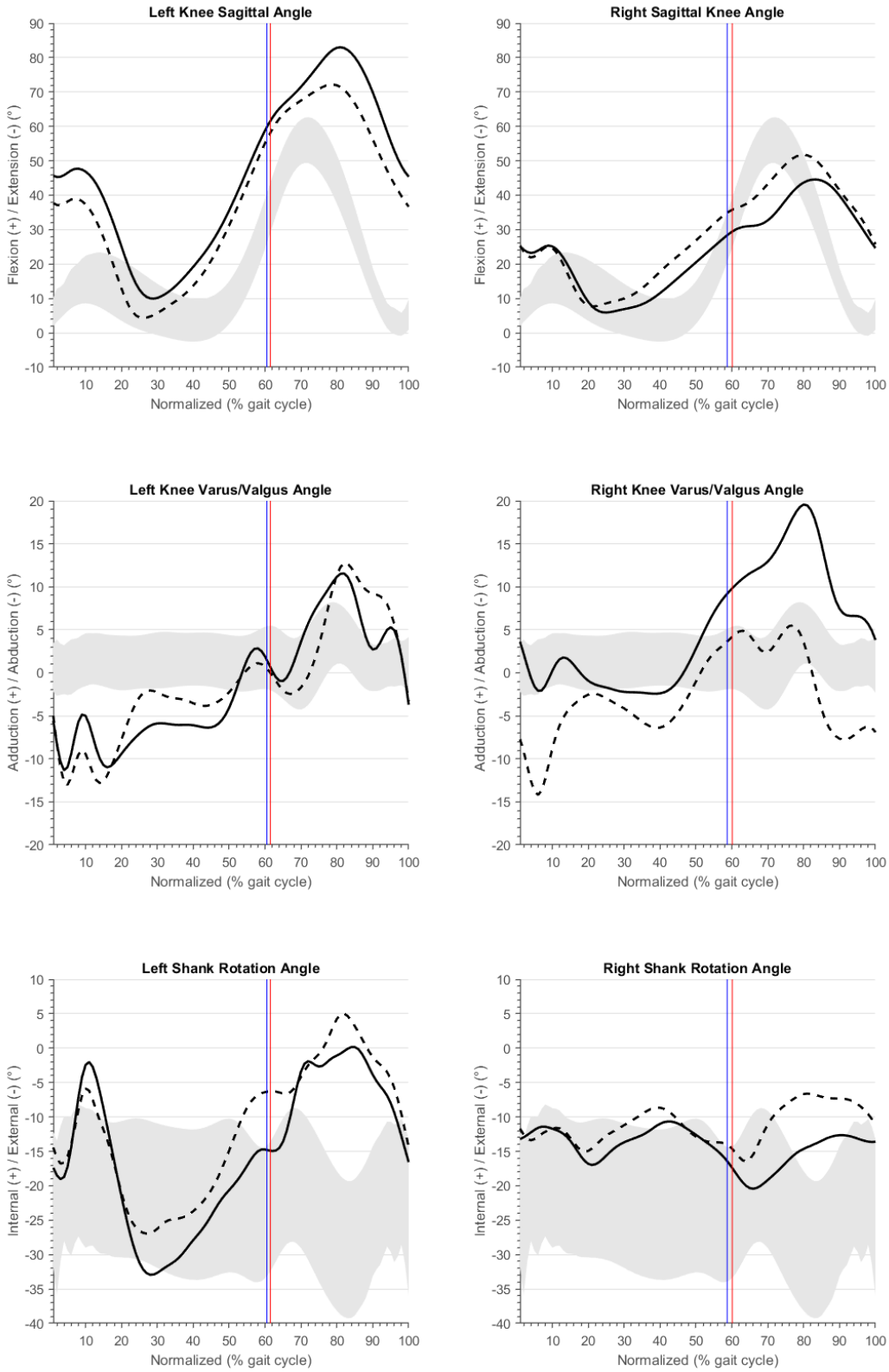

FIGURE 29. C3 Knee kinematics. Solid line, dashed line and shaded represent PRE, POST and typically developed gait, respectively. Red vertical line toe-off in PRE and blue vertical line in POST.

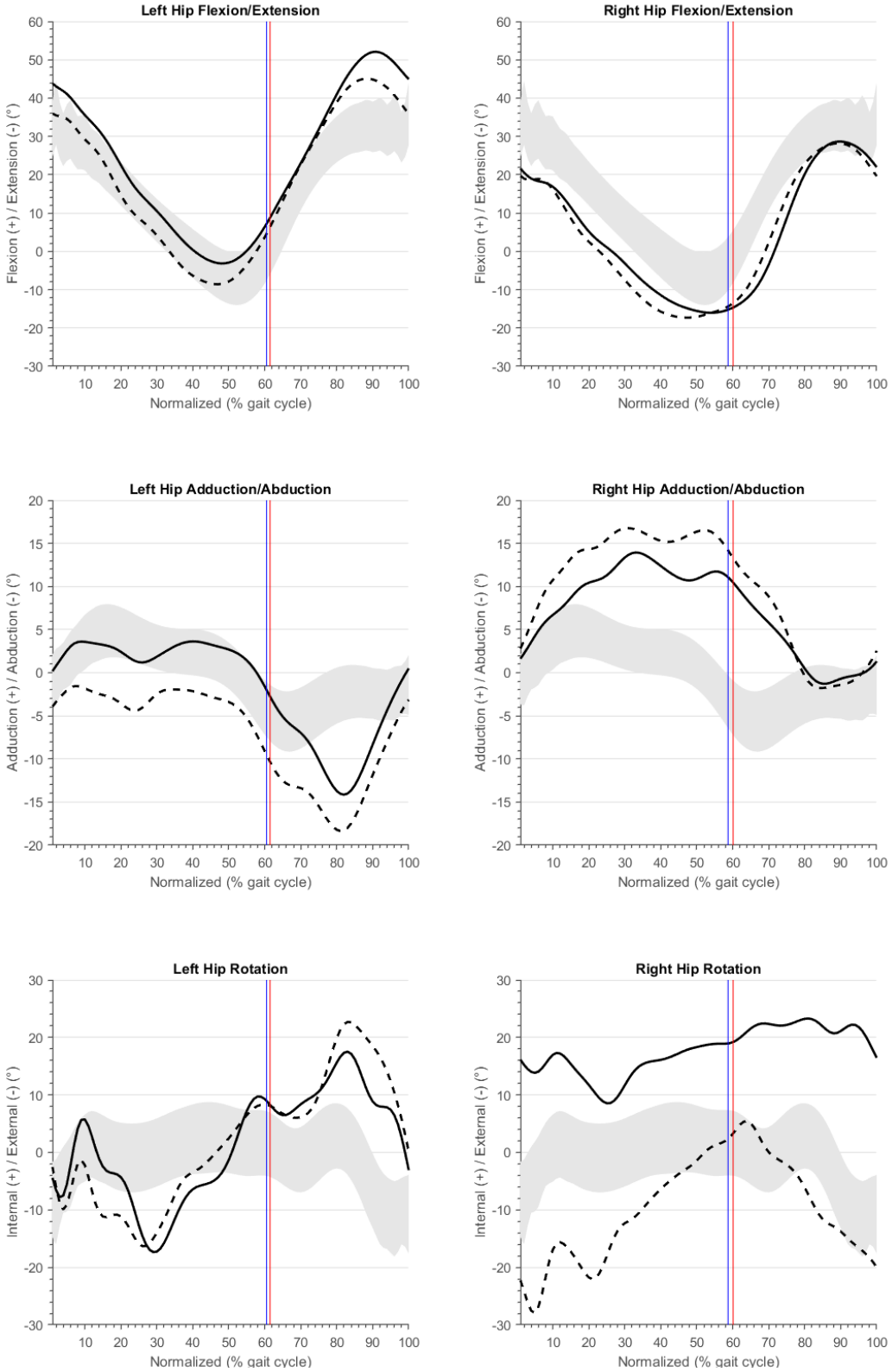

FIGURE 30. C3 Hip kinematics. Solid line, dashed line and shaded represent PRE, POST and typically developed gait, respectively. Red vertical line toe-off in PRE and blue vertical line in POST.

TABLE 6. Sagittal kinematical parameters from PRE and POST gait analysis measurements. Results are presented as mean ± standard deviation, in degrees.

	C1		C2		C3	
	PRE	POST	PRE	POST	PRE	POST
Dorsiflexion at IC	L: 94.69 ± 2.87 R: 90.78 ± 2.12	89.80 ± 3.22 83.30 ± 1.47	L: -1.32 ± 0.81 R: 9.24 ± 2.10	0.89 ± 1.35 6.67 ± 1.88	L: -6.74 ± 6.73 R: 11.70 ± 3.91	-2.34 ± 3.83 15.89 ± 1.93
Dorsiflexion at TO	L: 67.19 ± 3.72 R: 64.78 ± 5.69	71.47 ± 4.56 62.11 ± 5.34	L: -14.32 ± 3.75 R: -3.24 ± 4.31	-8.05 ± 4.11 12.21 ± 3.19	L: -12.05 ± 9.50 R: 0.63 ± 5.10	-7.75 ± 5.78 -6.63 ± 5.54
Peak dorsiflexion in swing	L: 96.31 ± 2.80 R: 93.63 ± 1.34	99.23 ± 4.95 87.34 ± 1.24	L: -1.92 ± 1.11 R: 11.53 ± 1.27	0.17 ± 0.99 14.64 ± 1.83	L: 3.17 ± 3.35 R: 18.59 ± 1.77	3.93 ± 3.07 18.70 ± 1.20
Peak dorsiflexion in stance	L: 103.79 ± 3.67 R: 108.18 ± 3.19	96.47 ± 4.22 91.72 ± 2.09	L: 11.56 ± 1.12 R: 20.13 ± 1.13	14.56 ± 1.58 25.42 ± 1.21	L: 10.94 ± 8.77 R: 16.81 ± 2.57	8.49 ± 5.33 27.04 ± 1.87
Ankle flexion ROM	L: 39.73 ± 6.21 R: 45.77 ± 8.25	31.42 ± 7.84 33.22 ± 5.10	L: 33.70 ± 2.18 R: 28.47 ± 1.68	31.11 ± 2.95 24.26 ± 2.15	L: 47.41 ± 7.82 R: 23.52 ± 3.88	26.02 ± 3.69 35.47 ± 4.78
Mean foot progression angle	-	-	L: 8.59 ± 8.80 R: -16.91 ± 3.81	10.70 ± 9.11 -22.85 ± 3.29	L: -5.58 ± 1.80 R: -7.88 ± 6.35	-7.76 ± 2.30 -13.28 ± 5.46
Foot inclination at IC	L: 93.12 ± 1.60 R: 90.23 ± 1.61	88.88 ± 3.27 86.48 ± 1.79	L: -1.76 ± 0.99 R: 15.70 ± 4.02	2.04 ± 3.09 14.34 ± 1.91	L: -0.42 ± 1.91 R: 9.83 ± 4.34	0.84 ± 1.48 4.83 ± 2.02
Knee flexion at IC	L: 44.57 ± 2.44 R: 40.98 ± 2.35	43.97 ± 5.84 43.49 ± 3.15	L: 30.97 ± 1.79 R: 33.08 ± 1.64	39.38 ± 2.95 31.08 ± 2.32	L: 45.69 ± 2.63 R: 25.16 ± 4.31	37.70 ± 3.56 25.37 ± 3.73
Knee flexion at TO	L: 51.12 ± 3.55 R: 47.01 ± 2.62	37.53 ± 5.58 44.77 ± 2.93	L: 25.85 ± 1.12 R: 52.25 ± 3.84	36.20 ± 3.18 51.19 ± 5.41	L: 60.75 ± 7.14 R: 29.36 ± 3.82	55.14 ± 8.29 35.13 ± 4.78
Knee flexion ROM	L: 26.11 ± 2.90 R: 28.58 ± 2.68	34.08 ± 5.79 34.51 ± 4.01	L: 39.95 ± 1.65 R: 56.22 ± 0.97	40.54 ± 1.64 54.81 ± 2.58	L: 74.17 ± 5.36 R: 39.71 ± 2.27	69.19 ± 3.81 45.34 ± 3.08
Hip flexion at IC	L: 32.40 ± 2.08 R: 28.75 ± 1.16	36.21 ± 6.25 32.16 ± 2.09	L: 35.67 ± 0.74 R: 49.81 ± 1.37	30.35 ± 2.51 40.84 ± 3.02	L: 43.83 ± 3.03 R: 21.61 ± 3.84	35.96 ± 1.95 19.73 ± 2.73
Hip flexion at TO	L: -13.02 ± 2.64 R: -17.59 ± 2.56	-17.46 ± 2.65 -17.75 ± 2.87	L: -5.62 ± 0.82 R: -4.39 ± 1.77	-6.50 ± 2.55 -7.92 ± 3.07	L: 7.99 ± 4.59 R: -14.70 ± 3.65	3.71 ± 5.11 -14.21 ± 1.67

IC = initial contact, TO = toe-off, ROM = range of motion, L = left, R = right.

7.3 Muscle activation patterns

Muscle onset timing during the gait cycle for muscles medial gastrocnemius (MG), soleus (SOL), and tibialis anterior (TA) were averaged from at least 50 complete strides per measurement session and muscle. On C1 both MG and SOL are activated in the end of the loading response and transfer to the midstance phase (figure 31). SOL slightly ahead (10.1, 10.8% GC and 8.6, 9.9% GC, PRE and POST respectively, left mentioned first) of MG (12.6, 11.4% GC and 9.3, 9.6% GC). SOL ceased a bit before the toe-off while MG was still active

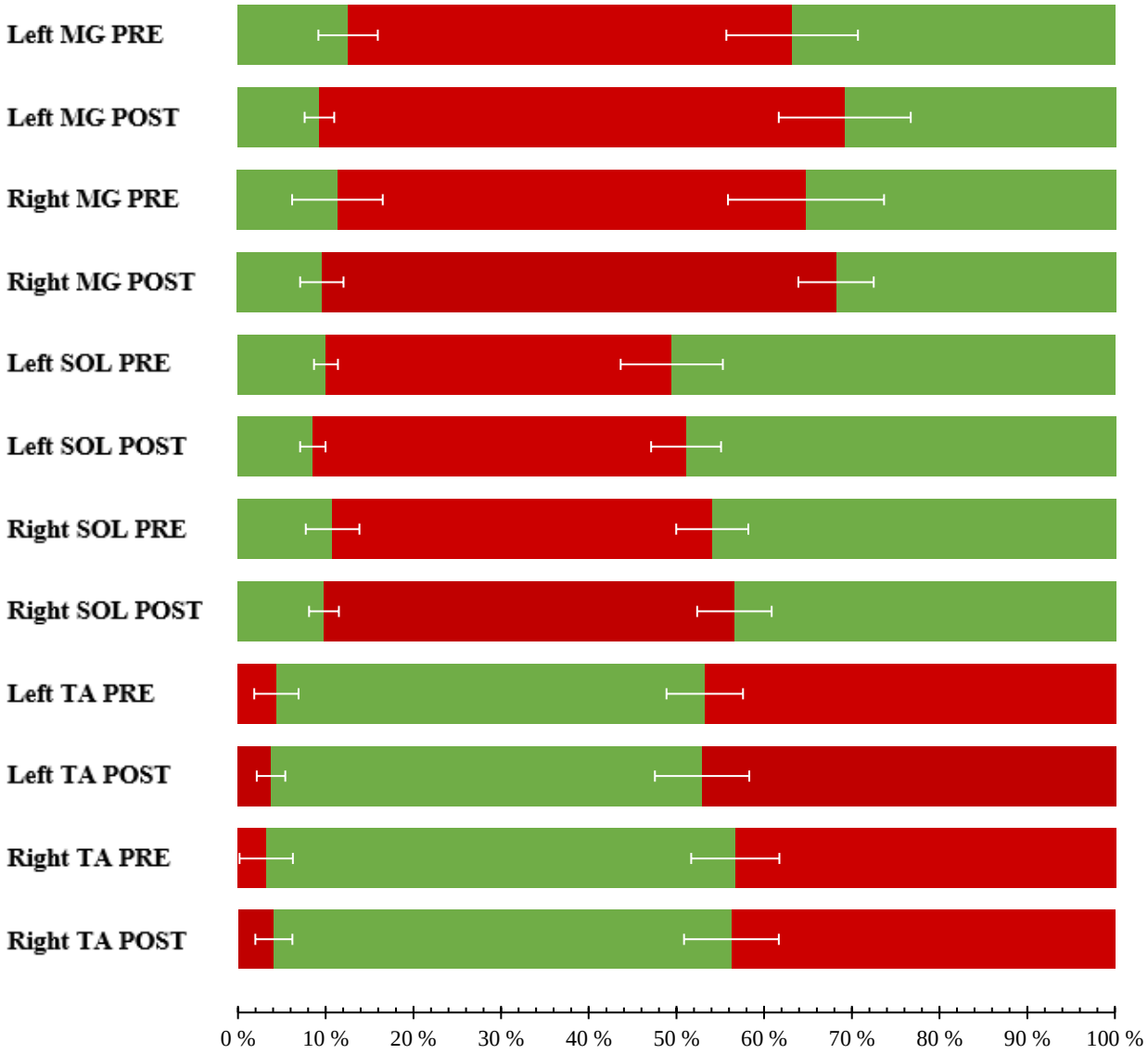

FIGURE 31. C1 muscle onset timing during the gait cycle (%) for muscles medial gastrocnemius, tibialis anterior, and soleus. Red indicating active muscle and green no muscle activation. White error bars indicate the standard deviation of the on/offset moments.

until initial swing phase. In both SOL and MG onset happened earlier and ceased later in POST compared with PRE. TA onset occurred in the pre-swing phase (53.2, 56.7% GC and 52.9, 56.3% GC) overlapping the diminishing activity of SOL. TA relaxation took place right after the initial contact both before and after intervention.

Compared with C1, C2 had clearly a more diverse muscle onset pattern varying also between the limbs (figure 32). First MG onset occurred already in the late terminal swing phase in left and on right (POST) continuing until terminal stance. On a left leg MG was also active in the middle of the relaxation in 10% of the analyzed strides (light red, figure 32). The affected left

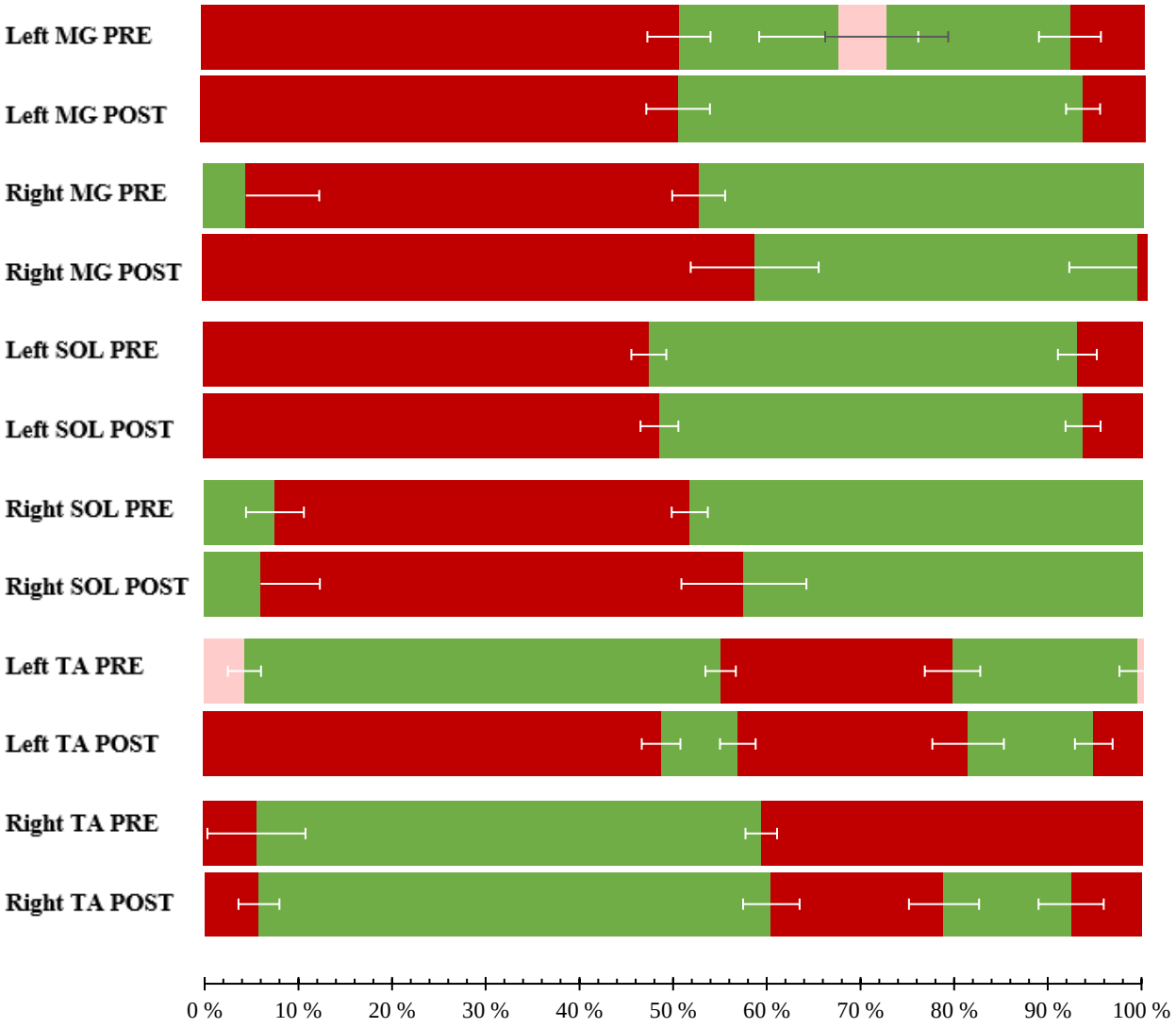

FIGURE 32. C2 muscle onset timing during gait cycle (%) for muscles medial gastrocnemius, tibialis anterior, and soleus. Red and light red indicating active muscle and green no muscle activation. White error bars indicate the standard deviation of the on/offset moments.

leg had also SOL activated significantly before initial contact but on the right leg the SOL activation pattern was similar to typically developed. TA pattern changed a lot after the intervention, on the affected side the TA was almost constantly active and co-contracting at the same time as the triceps surae muscle group. On the right leg similar effect wasn't noticed, but the continuous EMG activity from toe-off to loading response was broken to two separate bursts with relaxation period during the mid- and terminal swing phase.

On C3 both the MG and SOL on the right side were activated already during the terminal swing but on the left side (more affected) the MG was activated as in typically developed (figure 33).The activation pattern for MG and SOL shifted to a bit earlier phase after the intervention, as the onset occurred at 89.9% GC in POST and 99.1% GC in PRE for MG

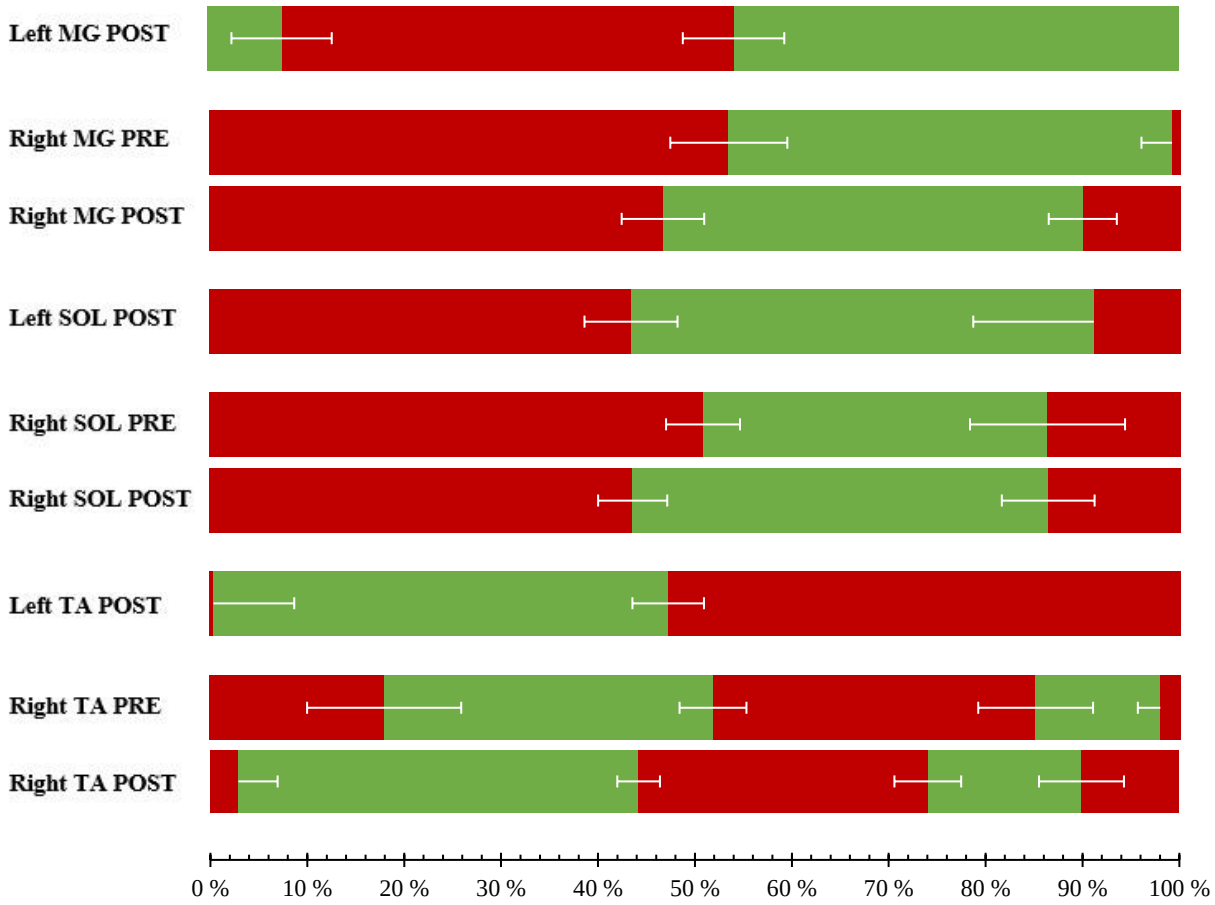

FIGURE 33. C3 muscle onset timing during the gait cycle (%) for muscles medial gastrocnemius, tibialis anterior, and soleus. Red indicating active muscle and green no muscle activation. White error bars indicate the standard deviation of the on/offset moments. Left side PRE values were discarded due to EMG and foot switch quality.

while the cease of the MG and SOL activity appeared seven percentage points earlier than GC. Similar shifting effect was seen also in right TA. C3 had the earliest TA muscle onset of all three participant at 44.2% GC (right leg POST).

7.4 Vertical ground reaction forces

Vertical GRF was sampled each percentage of the ground contact (GC), on average, on 15,9 completed steps on the force plates per leg on each measurement session. C1 (with crutches) vGRF pattern and values were not dramatically changed by the intervention (figure 34). On the left side the mean maximum loading response peak was 573 N (100% BW) and 602 N (105% BW) and on the right side 624 N (111% BW) and 609 N (108% BW), PRE and POST respectively on both sides. The vGRF pattern has a sharp impact force at initial contact following a distinct loading response during the transition from loading response to midstance. However, the pattern is missing the second peak partly at terminal stance corresponding with the push of ankle plantarflexor muscles and downward acceleration of COG. This may be linked up to walking with crutches as the toe-off is compensated with a push from the crutches.

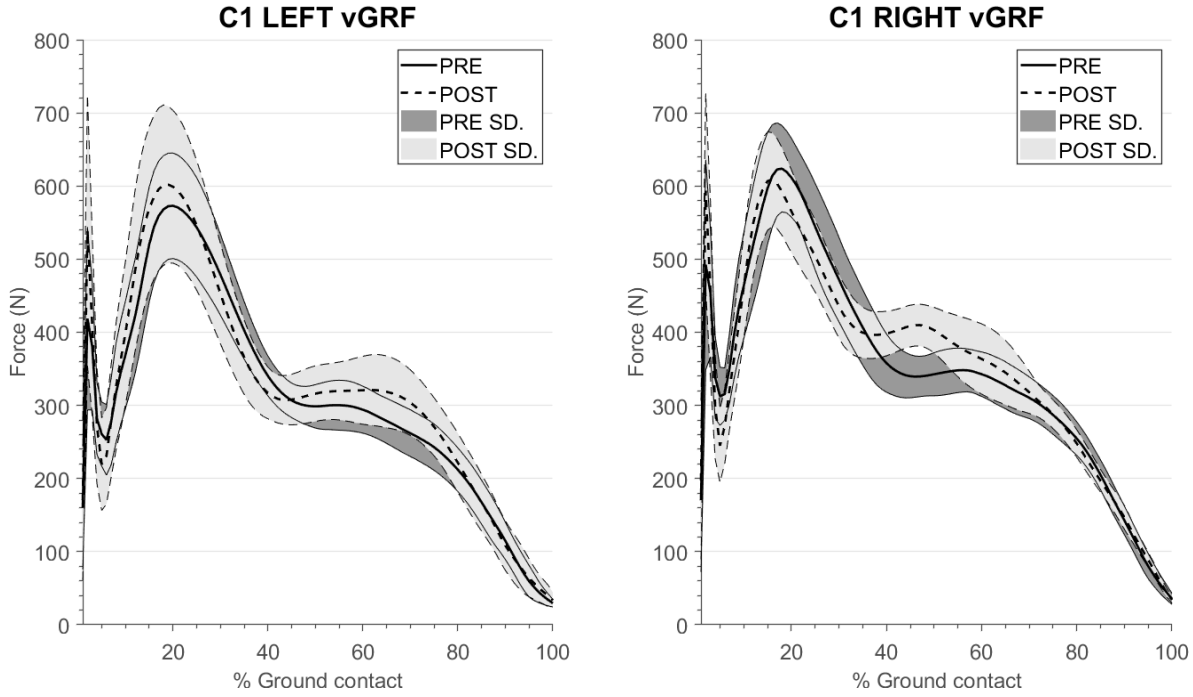

FIGURE 34. C1 Left and right vGRF. Mean vGRF values plotted as bolded solid (PRE) and dashed (POST) lines, and shaded areas representing the standard deviation.

C2 vGRF pattern and values demonstrated some differences in the pattern due to the hemiplegia and changes because of the decreases in walking speed (figure 35). On the PRE measurement there was no difference in the mean loading response peak between legs (973 N, 137% BW and 985 N, 139% BW) but the terminal stance peak was clearly less on the left leg (712 N, 101% BW) than on the right leg (837 N, 118% BW). Similar differences between affected and non-affected side were also seen in the spatiotemporal parameters, showing slightly limited use of left leg for stance. The vGRF values decrease on the POST measurement due to slower walking speeds, which is seen as dropped peak humps and increased mid stance valley. After the intervention, with slower walking speeds the affected left side has higher loading response (875 N, 122% BW vs. 806 N, 113% BW) and terminal stance peak (732 N, 102% BW vs. 715 N, 100% BW) than the non-affected right side.

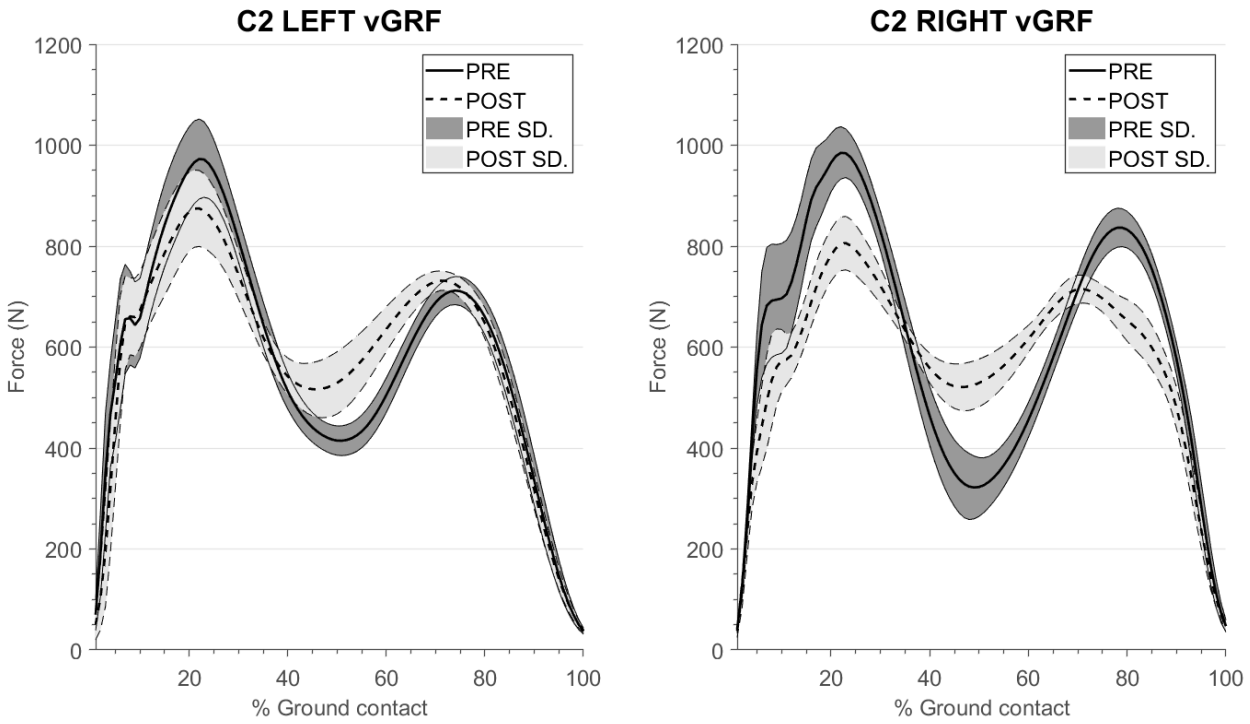

FIGURE 35. C2 Left and right vGRF. Mean vGRF values plotted as bolded solid (PRE) and dashed (POST) lines and shaded areas representing the standard deviation.

C3 (crutches) vGRF pattern was found the most different between the two limbs in these three cases. On the more affected side (left leg with peroneus paresis) the values were significantly lower and pattern very different compared with the right side (figure 36). The mean loading

response peak was 392 N (85% BW) and 434 N (92% BW) on the left, while 645 N (139% BW) and 753 N (159% BW) on the right, PRE and POST respectively. Increases in the values on both legs from PRE to POST are partly due to the increased walking speed on the POST measurements. One distinct characteristic on the left side is also the depth of the midstance valley (94 N, 20% BW and 108 N, 23% BW), PRE and POST, respectively. This demonstrates a characteristic "hop" for C3 on the left mid stance where with support of crutches the left foot slides slightly backward while the knee extends fully during the midstance.

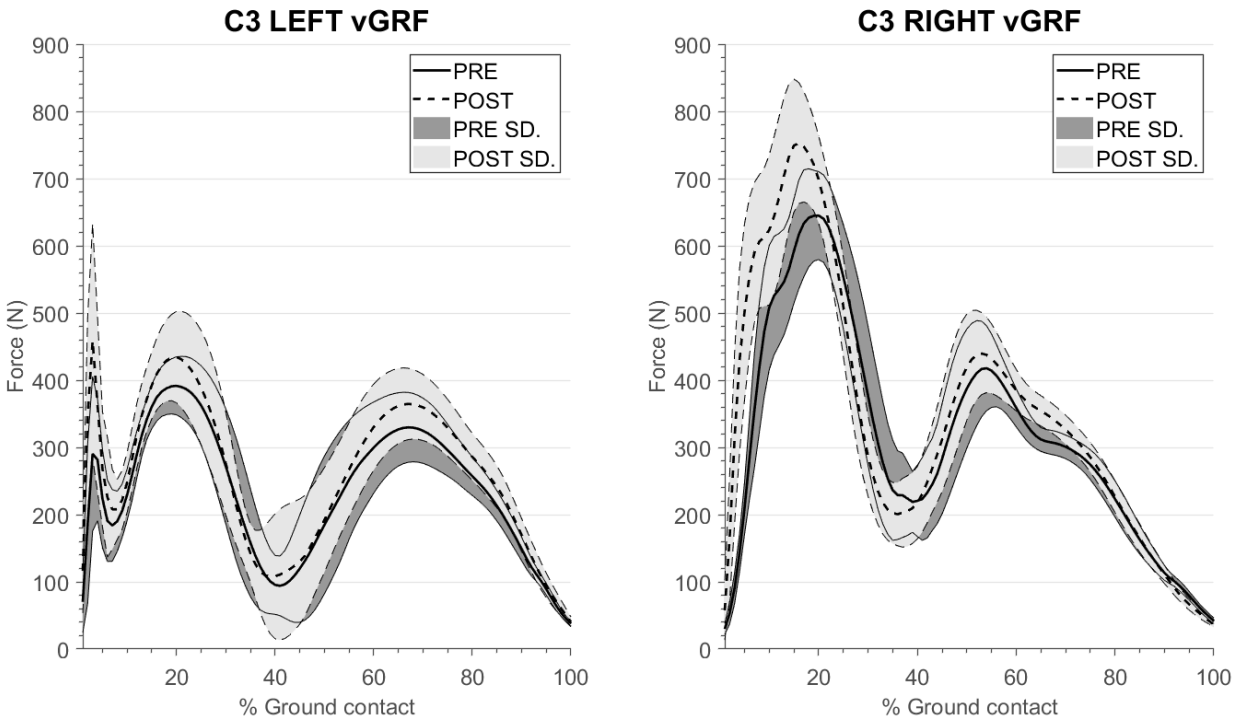

FIGURE 36. C3 Left and right vGRF. Mean vGRF values plotted as bolded solid (PRE) and dashed (POST) lines and shaded areas representing the standard deviation.

7.5 Ankle plantarflexion and dorsiflexion strength

Maximal voluntary isometric plantarflexion torque production measured in custom-built ankle dynamometer with all three case subjects. C1 demonstrated a clear decrease from PRE measurements for both left (-58.0%) and right (-18.2%) torque production in POST measurements (table 7). At the POST session, the C1 was frustrated with the inability to

perform the plantarflexion at the level of PRE measurement. As when C1 was not pushing hard, the torque levels were relatively high but right away after pushing maximum the torque did not increase similarly. Thus, the frustration and inability to perform plantarflexion hampered the results.

C2 had a significant difference between the affected side (left, 37.64 Nm) and non-affected side (right, 134.52 Nm) in PRE measurement. After the intervention the right ankle plantarflexion stayed on relatively high level although it decreased (29.1%), but the left ankle plantarflexion torque levels increased 75.4% to 66.02 Nm. C3 increased maximal isometric ankle plantarflexion torque production on both legs, left 59.2% and right 29.9%.

TABLE 7. Maximal isometric plantarflexion at ankle 90° (sole of the foot perpendicular to the shank).

	C1			C2			C3		
	PRE	POST	Diff. (%)	PRE	POST	Diff. (%)	PRE	POST	Diff. (%)
Left (Nm)	62.85	26.43	-58.0	37.64	66.02	75.4	29.94	47.67	59.2
Right (Nm)	48.35	39.54	-18.2	134.52	95.34	-29.1	25.35	32.93	29.9

For C2 and C3 concentric ankle torque production was measured in both PRE and POST the intervention for both legs. Isokinetic measurement for plantarflexion started with two-second isometric while the participant pushed as hard he could. After the isometric part, the pedal started to move at 14°/s to dorsiflexion until stopped at 28°of dorsiflexion and the participant tried to push the pedal hard during the whole movement. Dorsiflexion was measured on the right leg for C2 and C3 in both PRE and POST, while the dorsiflexion was not possible on the left leg at the PRE. C2 increased the affected leg torque production in isometric and in 5-10 degrees in plantarflexion, but at larger plantarflexion angles, the results were similar (figure 37). For right leg, the initial results were much higher than the affected leg, but the force production was decreased in isometric and 5-10 degrees in plantarflexion.

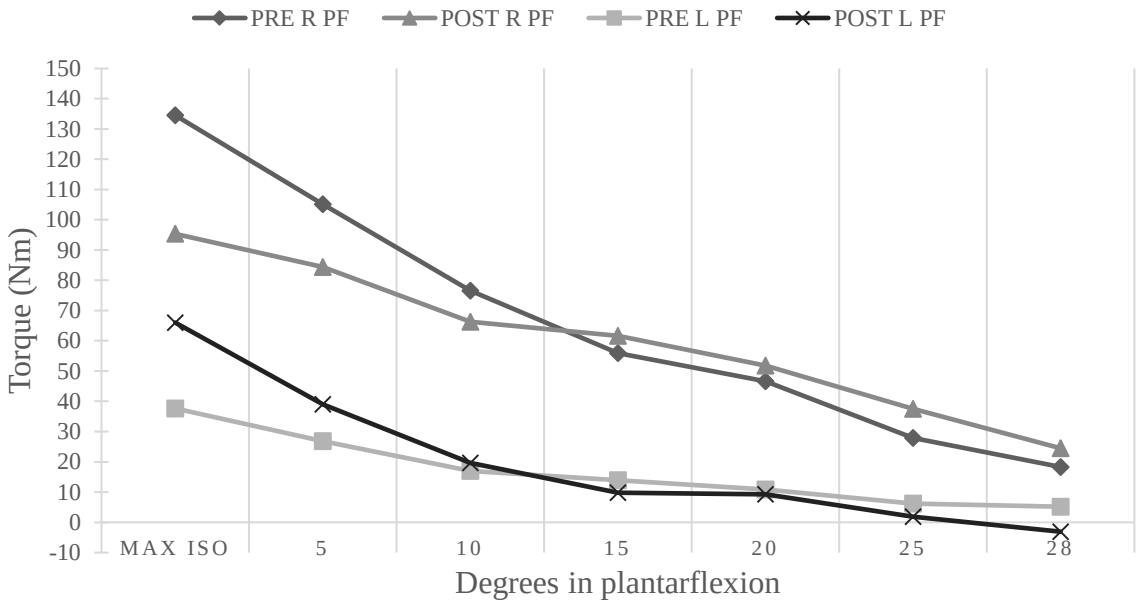

FIGURE 37. C2 - Plantarflexion in isokinetic (14°/s) conditions.

C3 increased the ankle isometric plantarflexion torque production for both legs and at 5, 25, and 28 degrees of plantarflexion on the left leg (figure 38). At POST intervention isokinetic trials for right leg, the C3 could only produce torque until 10 degrees in plantarflexion, and after that, the ankle resisted the motion (dorsiflexion). Dorsiflexion was recorded for both C2 and C3 with right leg on PRE and POST intervention (figure 39). Dorsiflexion trial was started at 28° plantarflexion with maximal isometric dorsiflexion of 2 seconds and continued with isokinetic 14°/s dynamic phase, the participant continuing the dorsiflexion strongly. C2 could produce the highest isometric dorsiflexion force in PRE, but in POST measurement C2 produced higher dorsiflexion torques in later phases (20-0 degrees in dorsiflexion) of the isokinetic measurement. The changes between PRE and POST were other way around for C3 as the force production at isometric and at 25° in plantarflexion timepoints were higher than in PRE, but C3 could not produce dorsiflexion in later angles (15-0° in plantarflexion).

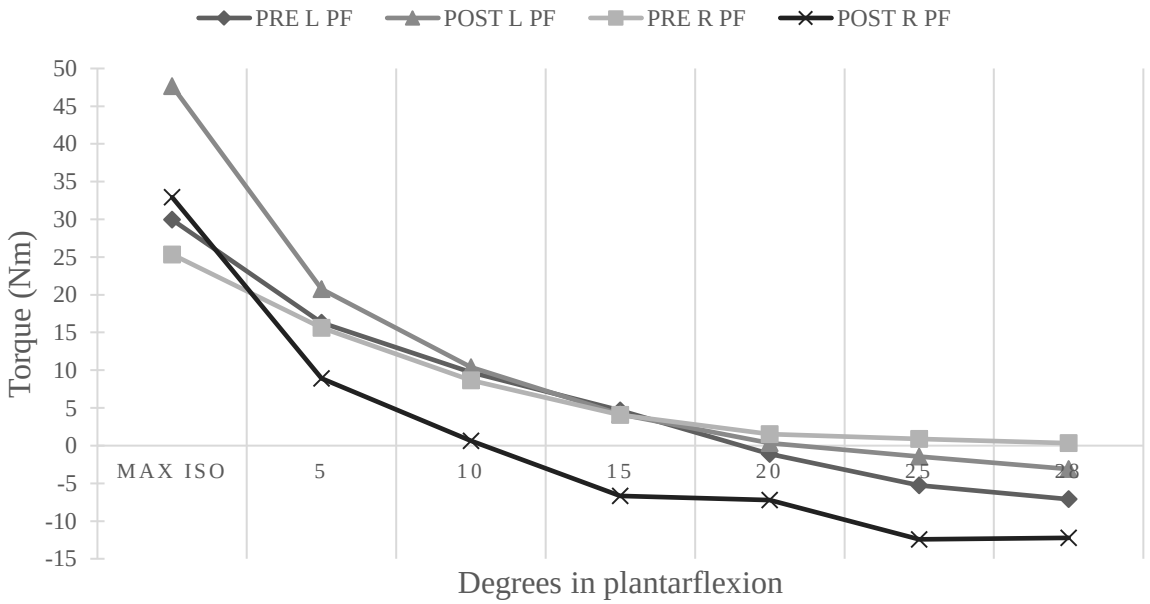

FIGURE 38. C3 - Plantarflexion in isokinetic (14°/s) conditions.

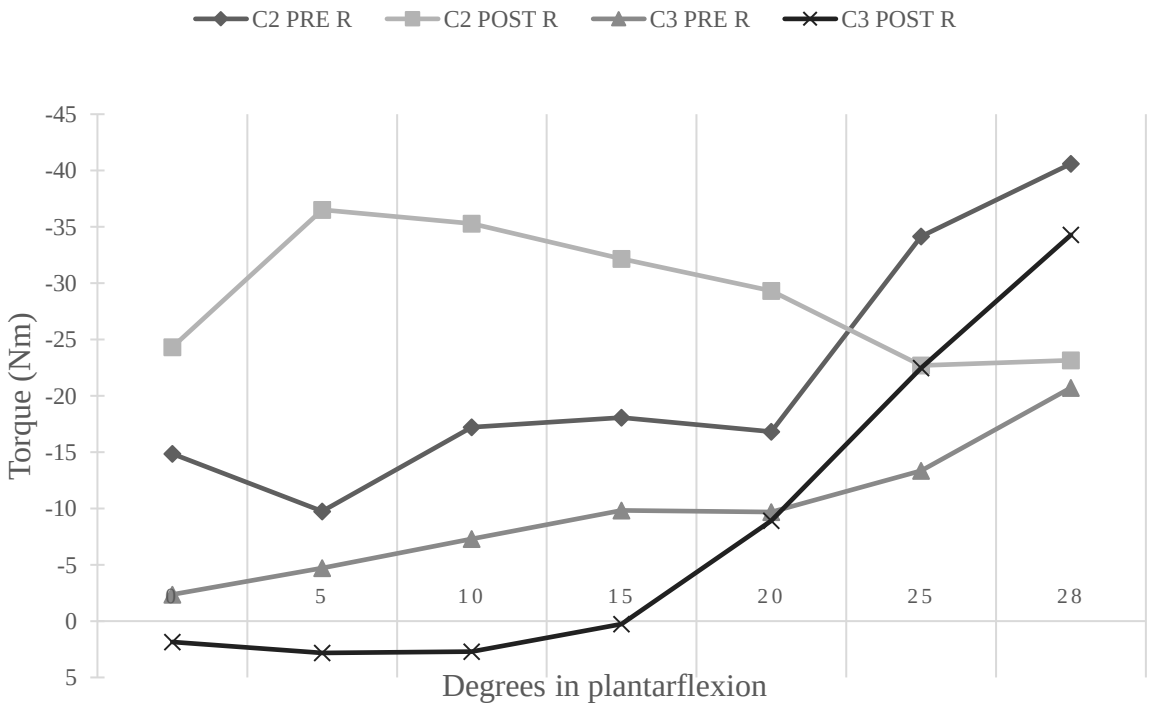

FIGURE 39. Concentric dorsiflexion (14°/s) from 28 degrees of plantarflexion to 0 degrees of dorsiflexion. Right leg C2 & C3. 28 degrees is the maximal isometric dorsiflexion value.

7.6 Six-minute walk test

Six-minute walk test (6MWT) reflecting the functional capacity and gait performance of the CP participants showed increases in C1 (5.8%) and C3 (8.1%) but not in C2 (-9.3%) after the intervention (table 8). C1 and C2 decreased their walking speed in the gait analysis measurement, but only C2 walked less in 6MWT in the same testing day. In C2 decrement to PRE test was not seen in POST2 6MWT, three months after intervention ending at the end of the maintenance period, where a 103-meter improvement to PRE was seen. This indicates a low motivation for C2 during POST measurements.

TABLE 8. 6MWT performed PRE and POST intervention in a 30-meter corridor and results presented as meters walked.

	C1			C2			C3		
	PRE	POST	Diff. (%)	PRE	POST	Diff. (%)	PRE	POST	Diff. (%)
6MWT (m)	424.5	449	5.8	551	500	-9.3	388.5	420	8.1

Note: C2 at POST was not motivated, he stopped for a few seconds on almost all transitions. C2 result in 6MWT at POST2 (3 months after the intervention, at the end of maintenance period) was 653 m (+18.5%).

8 DISCUSSION

The purpose of this study was to determine if a 3-month exercise intervention has a positive effect on lower body joint kinematics and muscle activation during gait, gait performance and lower limb strength in three young case subjects with CP. CP has a diverse clinical manifestation, and thus analyzing individual responses of these three case subjects with distinctly different backgrounds is important. Generally, the analysis provided likely evidence to improved gait performance in 6MWT after the intervention in all participants (5.8, 8.1 and 18.5%). Secondly, two participants improved plantar and dorsiflexor strength either isometrically or concentrically, measured as torque in an ankle dynamometer. Still, differences in strength between the affected and non-affected or less-affected limb were evident after the intervention. Finally, the gait analysis results, joint kinematics, and muscle activation patterns, in general, were not improved after the intervention. One of the main targets during the intervention was to improve toe clearance utilizing ankle dorsiflexion during the swing phase of gait. Only one of the participants slightly improved his affected side's toe lift after the intervention. Mostly the same compensations and pathological gait patterns were present also after the intervention. A similar discrepancy between overground measures of functional capacity and gait kinematics have also been found in earlier research (Gillett et al. 2019).

8.1 C1

The walking speed was slower in POST measurement which affected naturally also multiple other spatiotemporal parameters such as step frequency. Longer steps were seen as swing time increases and the aim for quality walking may have influenced the walking speed, even though the participant was encouraged to walk as fast as he could. Stride width was in the upper end of the normal values when compared to Armand et al. (2016) reported normal values. Wider steps than in other case participants originate from the scissor-type crouch gait where the wide base of support is needed, especially when walking without the crutches' support.

Absent of an ankle first rocker was evident in the sagittal ankle joint kinematics. This is associated with weak ankle dorsiflexors, which was one of the key targets of the training during the intervention. Increased dorsiflexion during stance phase in PRE measurements might signal soleus weakness, dorsiflexion during stance was less in POST measurements. Increased dorsiflexion and soleus weakness are two of the elements influencing the increased knee flexion during the stance phase in C1. (Armand et al. 2016.) TOE marker excursion was used to track the toe lift during swing to describe the foot clearance development. After the intervention there was an increase of toe lift in the mid-swing and terminal swing, but it appears to result from other joint articulations, especially due to increased hip flexion but also increased right knee flexion.

Excessive knee flexion is present during the whole gait cycle which is a clear indicator of a crouch gait pattern. This possibly arises from the hamstring contracture or overactivity in which the weaker knee extensors try to respond. This crouch gait pattern was accompanied by scissor gait pattern. Knee went rapid movement from abduction to adduction during midstance and late stance phases, during the single-limb stance. Shank rotation was mostly in the normal gait values after the intervention, slightly externally rotated. Hip flexion mostly keeps the normal pattern, but the hip hyperextended during the pre-swing and initial swing. At the same time hip is also adducted and internally rotated more than normally. Contralateral pelvic drop increased femoral anteversion especially on the right side (femoral anteversion: an inward twisting of the thigh bone), might lead to “pigeon toes”, or in-toeing and excessive external tibial torsion might be one of the impairments behind this pattern. Additionally, Steele et al. (2012) found out in musculoskeletal simulations of CP crouch gait that less strength is required from the hip abductors and ankle plantarflexors during crouch gait. This suggests that weakness of these muscle groups may be partly responsible to crouch gait and that these muscle groups should be targets of strength training. (Steele et al. 2012.) Crouch gait can root in variable combinations of muscular, neurologic, and bony deformities. Also, rapid bone growth and weight gain during puberty may lead to progressive problems in gait at multiple joints and planes and loss of mobility. (Galey et al. 2017.)

When assessing gait patterns while the participant is using walking aids such as crutches here, it should be noted that the assistive devices might in negative cases hide or even exaggerate

the therapeutic effects. Krautwurst et al. 2016 showed that the kinematics were worse while walking with crutches, enhancing erroneous limb positioning present without a device. In this intervention the training was done mainly without crutches, but the walking measurements were done mainly with crutches. Higher terminal vGRF peak after the intervention demonstrated higher weight-bearing and possibly better ankle push-off. This can be seen also that before the intervention crutches assisted more than post-intervention. The ankle push-off and toe-off are compensated with using the crutches.

There was a clear decrease in the maximal isometric plantarflexion torque results after the intervention. C1 was frustrated with the inability to perform the plantarflexion at the level of PRE measurement. As when C1 was not pushing hard, the force levels were relatively high but right away after pushing maximum the force did not increase similarly. Thus, the frustration and inability to perform plantarflexion hampered the results. This demonstrates well the problems CP has in the selective motor control and possible cocontraction of the shank muscles. Still, the ankle kinematics during the gait didn't show distinct changes in the ankle dorsiflexion during swing and initial contact which might indicate that the TA strength and contraction shouldn't restrict the plantarflexor strength. Especially in this kind of constrained measurement circumstance.

C1 has quite typical EMG onset pattern, MG active in pre- and initial swing. Triceps surae muscles were activated longer, even though the swing period was longer in the POST, stance period shortened (-0.7, -2.7% right and left respectively). TA activity didn't change after the intervention. What is interesting about the absence of dorsiflexion in the initial contact, is that the TA was active during the initial contact and it seems that there was no cocontraction preventing the toe lift. It might be that the TA muscle coordination and strength are not enough for this type of activity.

8.2 C2

Walking speed in PRE measurements proved that C2 is capable to walk at similar velocities as his typically developed peers. After intervention, many of the changes seen in the

spatiotemporal parameters are rooting from the slower walking speed. When the walking speed increases to fast walking speeds, both the step frequency and step length tend to increase significantly (Öberg et al. 1993; Fukuchi et al. 2019). There were clear differences between the affected (left) and non-affected limb (right) in pre-tests. The affected side of the body stepped on a slower frequency, took shorter steps, spent more time in swing phase and had a shorter contact time than the non-affected side. It is common that people with hemiplegia have clear differences in weight-bearing and stride parameters between the sides, for example due to the altered neural control and muscle weakness on the affected side (Li et al. 2018). Also, the disparity between limbs was seen in the isometric plantarflexion torque and vGRF terminal stance peak. Differences between affected and non-affected limb decreased after the intervention in swing-stance relation and step frequency. Still, after the intervention there were distinct differences between the limbs in these parameters and the step length differences were even larger than before the intervention. The affected side had clearly incomplete push-off capabilities (terminal stance vGRF peak) during the gait but the slower walking speed in the post measurements hid the difference as the non-affected side ankle was not needed to produce power as near the maximum as in faster walking speeds. This was also in line with the results from Fukuchi et al. (2019) comparing the gait parameters on different walking speeds. Therefore, strength improvements in the plantarflexors may not be reflected at slower walking speeds where ankle power generation requirements are less. Evaluation of a range of walking speeds around typical walking speed would be useful in future studies (Gillett et al. 2019).

Joint kinematics presented deviations between the two sides of the body and the deviations also mostly remained after the intervention. Toe lift was very clear on the non-affected side but the affected side missed the second toe lift which would set the foot ready for the heel contact. EMG onset timing revealed that triceps surae muscle on the left limb was activated already significantly before the initial contact. This led to cocontraction with tibialis anterior at the terminal swing in POST or even to a plantarflexion dominance in PRE. Premature gastrocnemius activity around the footstrike is connected with muscle weakness (Schweizer et al. 2013). TA presented on both sides a two-interval burst with silent period between, indicating the pick up of the foot at the initial swing and terminal swing to control the heel rocker. If both the triceps surae and tibialis anterior are activated during the terminal swing

the foot initial contact is likely flat-footed. Additionally, in C2 on the affected side the EMG intensity was lower in the terminal swing compared with initial swing, telling about problems in active ankle dorsiflexion capabilities. The prolonged moderate TA activity into mid and terminal stance may tell on prolonged inversion during the stance and TA activity has been seen at full foot in flatfooted participants. (Gray & Basmajian 1968.)

In dynamometry, the non-affected side was markedly stronger in plantarflexion at all joint angles compared with the affected side while the affected side got stronger in isometric conditions compared with pre-intervention. The dorsiflexion torque during movement was enhanced on the non-affected side after the intervention and C2 was able to produce dorsiflexion torque for whole angle spectrum from 28° plantarflexion until to ankle in 90°. This suggests there might have been some positive changes also on the affected side. It should be noted that the strength level would still have been low as the participant couldn't produce dorsiflexion in the familiarization sessions and the TA activity was seen mainly during initial swing. Even though, the TA would be relatively strong the participant might lack the ability to voluntary activate the muscle during the swing phase of gait and thus continue the compensation from knee flexion for the toe clearance. Weak ankle dorsiflexion strength and plantarflexion activity during the late swing and initial contact are possibly the root causes leading to a flat-footed initial contact on the left, affected, side.

Secondly, the distinctly increased knee flexion during stance on both left and right side influence the increased ankle dorsiflexion during stance, especially on the right side. Knee flexion at the initial contact and through the stance phase could tell on hamstring overactivity in addition to plantarflexor overactivity. On the left side (affected) the knee flexion at the loading response is accompanied with typical type IV hemiplegia gait pattern (Rodda & Graham 2001) with large knee and hip adduction movement likely showing hip abductor weakness, but the hip internal rotation might be a confounding factor (Armand et al. 2016). This deviation and compensation with contralateral pelvic drop might be one reason the participant feels the affected side uncomfortable which shortened the step parameters on the affected side. Generally, the joint kinematics did not change for the better, some positive changes in lesser left knee adduction, right shank rotation, and hip extension. The most important training goal with the toe lift did not improve on the affected side and the same

compensation strategy with knee flexion and adduction and hip flexion were still present after the intervention.

Ma et al. (2019) also showed that diplegic CP had greater knee flexion and ankle dorsiflexion during the stance phase compared with TD children. They suggested that uphill walking, such as the training on the treadmill was done in this study would increase the severity of the pathological gait as inclined walking encourages to use more knee flexion and ankle dorsiflexion. In our study in every supervised session of treadmill walking, the participants were told to focus on toe clearance using the ankle dorsiflexion and to avoid excessive knee flexion.

Walking speed in PRE gait analysis and 6MWT demonstrated C2's good gait performance. After the intervention, walking speed decreased in both situations and was likely due to low motivation despite the encouragement throughout the measurement session. Especially when the POST2 6MWT, after the three-month maintenance period, was significantly better than the earlier test results (POST 2: 653 m, average speed: 1,81 m/s). This indicates that the gait performance may have been better right after the intervention, but the motivation hid the development. It is notable that the severity of CP can change due to participant's alertness and emotional state, which can complicate objective evaluation of young children as seen here in post-tests (Bonow et al. 2018).

8.3 C3

C3 was the only case subject of the three who increased his walking speed also in the gait analysis. The increased walking speed was seen as shorter contact time on both sides and higher cadence on the more affected left limb leading to symmetrical cadence between limbs after the intervention. Step length did not increase similarly and the asymmetric nature of stride was present also after the intervention. Significantly shorter steps on the right side are resulting from the knee hyperextension (recurvatum) of the contralateral limb during midstance limiting the forward progression and delaying preparation for single limb advancement tasks. Thus, the ipsilateral knee flexion is delayed and reduced and initial

contact is taken with flexed knee earlier than normally. In joint kinematics the effect of the more affected, left knee hyperextension can be seen as a rapid plantarflexion, ankle external rotation because the tibia does not advance normally during the stance. Commonly, knee recurvatum during stance arises from plantarflexor spasticity or contractures and compensates work usually done by quadriceps (Perry & Burnfield 2010, 353-354). In this situation, more likely, the hyperextension is due to the multiple tendon lengthening operations for knee and adductors during childhood. These surgeries may have hampered the hamstrings' ability to produce restraining static and dynamic forces during stance. The hyperextension was still present after the intervention but in slightly lesser form.

C3 places significantly less weight on the more affected side limb and utilizes the crutches heavily while the knee hyperextension unloads the limb momentarily during midstance. From the vGRF, it can be seen the lack of terminal stance peak on both sides indicating more reliance on crutches and less from natural limb advancement or plantarflexor muscles during push-off. It may be that the crutches hide some of the effects of the intervention by supporting the habits and old gait patterns. Regardless of the changes in the kinematics, C3 improved his gait performance significantly in 6MWT (+8.2%) after the intervention and the walking speed similar to the walking speed in POST gait analysis when taking account the time spent in turns in 6MWT.

Before the intervention, C3 had a distinct problem as the inadequate toe lift was ruining quickly the tip of all of his left foot shoes. The asymmetry between the less and more affected side in the motor control and dorsiflexion strength can be seen in the parameters defining the toe lift. The right foot presents a clear heel rocker at the initial contact while the left foot lacks the ankle first rocker contact with a flatter foot. However, small improvement was seen in the swing phase toe clearance on the more affected side when taking account the other joint articulations such as knee and hip flexion which both decreased after the intervention at the initial contact. Still, a compensation C3 is demonstrating for toe clearance is excessive knee adduction and ankle external rotation as the because the knee is less flexed than in typical gait. The increased hip adduction during mid and terminal stance may indicate hip abductor weakness but increased adduction might also appear due to pelvic level confounding factors not analyzed here.

Some differences between limbs were noticed in the muscle activation patterns. While the more-affected left side MG was activated quite normally during the single-limb stance and push-off, the left leg SOL and right leg MG and SOL have activated already before the initial contact and even shifted earlier in the terminal swing after the intervention. Thus, the right leg triceps surae was activated at the same time as the right leg TA but it seems that it did not lead to lesser toe lift as in C2. The difference between SOL and MG activity patterns on the left side may arise from bent knee initial contact which favors soleus force production because MG is originating from femur and bending the knee lessens its capabilities. In C3 the TA activation on the right leg was split to two intervals, similarly as in C2, indicating the pick up of the foot at the initial swing and terminal swing to control the heel rocker. However, the more affected left leg had constant TA activity already from terminal stance until initial contact. Normally, the TA has constant activity from pre-swing until the end of loading response and the peak activity happening during initial swing and loading response. During the mid-swing the activity usually diminishes momentarily compared with peak activity phases. (Gray & Basmajian 1968; Perry & Burnfield 2010, 57.)

C3 was the only participant to increase maximal plantarflexion isometric torque for both legs and isometric dorsiflexion torque on the measured right leg, although it should be acknowledged at the same time that C3 was the weakest participant of the three. Even though the strength capabilities increased especially at isometric conditions after the intervention, On the less affected leg, C3 struggled in the higher plantarflexion angles and angles towards neutral ankle in dorsiflexion direction when compared with before intervention. Particularly, the weak plantarflexion torque production, or even turning into resistive torque already in early plantarflexion angles may describe the deficiency in motor control in dynamic situations in that measurement session.

8.4 Limitations of the study

Instrumented gait analysis gives very detailed information about participant’s walking but the method imposes also great demands for the researcher and measurement session design specifically when dealing with a special population such as CP. First the extensive and

expensive measurement equipment with cameras and force plates needs to be calibrated correctly. The researcher needs to be trained with marker placement to attain consistent marker application as marker-based motion analysis system's reliability relies heavily on the skill of assessors (Tsushima et al. 2003; McGinley et al. 2008; Leigh et al. 2013). Also, CP population with bone deformities and other anatomical deviations creates a more challenging environment for marker positioning. Despite the pilot measurements, application of the markers was a slight learning process after all during the first measurements, which were not analyzed in the thesis. Posterior pelvic (RPSI & LPSI) and foot markers turned out to be the most difficult to apply correctly. The consistency in marker application was tried to maximize using the same person to apply the markers for each measurement session. The anthropometrical measurements were repeated to attain reliable results and after dropped markers the participant calibration was repeated. All erroneous data were tried to exclude to the best as the motion capture data was consistently checked also manually. A rather short walkway and enough but limited camera count constricted the amount of quality gait data. Analyzing walking trials to both directions of the walkway provided a great amount of kinematical data to safely quantify the joint kinematics. There would have been a possibility to analyze only trials with higher walking speed (from right to left in figure 19) but it would have limited vastly the kinematical data. Also, in C1 and C2 the walking speed was low enough that they could accelerate to the walking speed already until the force plates when coming from the shorter direction.

While the Plug-in Gait model used for joint kinematics calculations is validated also in clinical decision-making (McGinley et al 2008) it still possesses some limitations, e.g. modeling the foot and measuring reliable movement in frontal and transverse plane (Davis 1991). The main limitations of every marker-based motion analysis system are that estimating the movement of internal structure is linked external measurements. Errors can cumulate from marker placement on anatomical landmarks, soft tissue artifacts, definitions of joint centers and anatomical deformities like in many times in CP population. Thus, some carefulness is also needed when interpreting results of motion analysis using subject-specific models, which are created just by scaling generic models, for participants with anatomical deformities. In the measurements two of the three Plug-in Gait model's markers representing the foot had to be

placed on the shoes. They were placed as correctly as possible, but it is likely that measurement error is greater compared to marker placement on the skin.

Compliance and participant's motivational state during measurements and the intervention were possible confounding factors to the results after the intervention. In addition to the training at the supervised sessions, the participant was instructed for home training with the non-motorized treadmill and continue their normal physical activity similarly as during the control period. However, treadmill training was not completed perfectly at home, only C1 did it successfully, both C2 and C3 did not train as much as it was instructed. This may have caused an inferior intervention effect, especially in the gait-related parameters. Severity of CP can vary with the patient's alertness and emotional state (Bonow et al. 2018), which can complicate objective evaluation in young children as seen here in post-tests. Especially important this is when studying children and adolescents with cerebral palsy as data quality can be affected by the emotional, mental and motivational state of a child. The gait analysis and 6MWT sessions were conducted on one day per participant which may have affected the condition and concentration even though enough rest, fluid and food was tried to provide between different measurements.

Self-preferred speed was hypothesized to lead to more natural gait compared with paced gait e.g. on a treadmill. Paced gait would have standardized the walking speed but it remains as a question would a better intervention effect be seen in a more forced situation such that. However, also self-selected gait can mask underlying impairments and allow for compensations (van Gelder et al. 2017). Naturally, with paced gait some of the walking speed-dependent changes in kinematics would have been minimized. However, the aim of the intervention was to bring positive changes to normal, natural gait of the CP participants, so the measurement with self-preferred walking speed and under encouragement of researchers was used to investigate the natural gait pattern. Due to the encouragement the walking speed was slightly outside of their comfortable self-preferred speed to challenge the participant to reach better gait pattern.

EMG onset timing analysis of MG, SOL and TA muscles binary excitation provides information about the temporal activation patterns during the gait cycle and is one of the most valid applications of sEMG signal. Still, one cannot draw conclusions e.g. about the activation intensity based on this data as it was not analyzed. Also, it may be that also other shank muscles such as extensor hallucis longus and extensor digitorum longus also have attributed to the dorsiflexion capability in addition to the measured TA muscle. In this analysis the proximal TA sEMG electrode placement was used and it might have influenced the activation pattern slightly. Vigotsky et al. (2018) showed that some parts of a muscle may be electromyographically "silent" while the other parts are not. Also, Peter et al. (2019) concluded that SOL sEMG activity may be easily affected by crosstalk if not sufficiently far from medial and lateral gastrocnemius. and onset/offset during walking should be interpreted with caution. Furthermore, EMG onset timing was normalized to a gait cycle measured with the footswitch. Although the footswitch was adjusted for every measurement session to capture the gait events correctly the stance and swing phase results may vary from the motion capture data and interchangeableness should be examined with caution.

This study observed three case subjects with different ages, backgrounds, and functional abilities. No averages were generated using all three participants' data as the participants were so heterogeneous. Thus, the individual case subject results are not generalizable across the spectrum of different CP impairment levels. However, the gait performance was improved in all participants in the end, which implies some association with the training during the intervention. Therefore, rehabilitation of gait deviations should be treated individually but some general principles for exercise prescription could be derived from the results of this study.

8.5 Future research

Designing successful interventions that promote functioning and well-being of people with CP is extremely important, for example to prevent secondary musculoskeletal impairments. Because CP affects individuals throughout their lifetime it is important that interventions try not only promoting functional changes but also behavioral changes where exercise is

understood as part of a lifestyle. Moreover, just improving strength is not enough anymore, also motor pattern must change. There were promising results using gait training to stimulate the toe lift capability from Willerslev-Olsen et al. (2015) but similar results were not consistently found with these three cases. Therefore, it would be important to study the underlying neuromechanical and cortical mechanisms to understand better the changes that are induced due to the rehabilitation or training. Ultimately, developing and studying effective ways to improve CP patients' walking and other functional abilities would help their everyday life and increase their overall health.

In this study lower body joint kinematics, vGRF, muscle activation patterns, gait performance, and lower limb strength were evaluated before and after the exercise intervention for three case subjects. A higher number of participants could have made possible to generalize and statistically analyze changes achieved due to the intervention. Extending the lower body model to full body model would have brought interesting insights about upper body compensations during the gait and made possible to account the usage of crutches. In addition, using inverse dynamics, kinetics could have provided information about joint moments and joint forces. In future, developing forward dynamics simulations using musculoskeletal modeling would possibly enhance the success of orthopedic interventions by predicting post-operational gait performance already before the operation (Pitto et al. 2019). Also, developing valid markerless multi-purpose motion or gait analysis applications would enable lighter and quicker clinical measurement sessions or open possibilities for remote rehabilitation and more regular follow-ups.

8.6 Conclusions

The tailored twelve-week exercise therapy intervention may likely improve the gait performance and strength in adolescents and young adults with CP. However, improvements do not happen hand in hand with gait quality. Generally, the same compensations and pathological gait patterns were present also after the intervention. While gait analysis data can present the direct effects of intervention, kinematics have only a weak relationship with functional ability measures. However, gait analysis allows identification and further

understanding of gait deviations. The intervention period may not be long enough to induce changes in motor patterns and major gait deviations such as crouch gait but may improve gait performance. Therefore, rehabilitation of gait deviations should be treated individually but some general principles for exercise prescription could be derived from the results of this study. Even though there were no clear signs of improvement along the whole spectrum of measures, it should be noted that even keeping the same performance and functionality level would be good in CP population as the disorder weakens the body in course of time. Possible positive changes in muscle strength or gait efficiency would be still beneficial to better ambulatory abilities for daily living, albeit kinematics does not change.

This study provides more means and considerations to individualize the training or treatment for adolescents and young adults with CP. Provided are also examples of interpreting the gait analysis data and identifying the targets for the intervention. Instrumented gait analysis could be an important part of rehabilitation design and accompanied by strength and functional measures give an extensive view of the functional and muscular capabilities of people with CP. Finally, one of the most important takeaways of the results is that there is no need to be afraid of typical strength training principles when dealing with CP population.

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APPENDIX

Appendix A: Training tables

TABLE 1. Description of the strength training exercises.

	Seated Calf Raise	Standing Calf Raise	Seated Dorsiflexion	Seated Machine Knee Extension	Seated Machine Knee Flexion	Seated Horizontal Leg Press	Squat	Hip Flexion	Roman Chair Trunk Extension	Isometric Hollow Rock
Muscles targeted	Soleus, Gastrocnemius	Soleus, Gastrocnemius	Tibialis Anterior	Quadriceps femoris	Hamstrings	Gluteus maximus, quadriceps femoris, hamstrings, triceps surae	Gluteus maximus, quadriceps femoris, hamstrings, triceps surae	Iliopsoas, rectus femoris, sartorius, tensor fasciae latae, tibialis anterior	Erector spinae, multifidus	Trunk flexors, hip flexors, transversus abdominis
Inicial Position	Seated with knees at 90°. Forefoot on a 5cm step, ankle in maximal attainable dorsiflexion. Weight over the distal thigh of training leg.	Standing with hips and knees at 0°, and the forefoot on a 10 cm step in maximal attainable dorsiflexion. Holding on parallel bars for balance*.	Hips at 70-90°, knees at 0-20°, ankle in full plantarflexion. An elastic band on the forefoot resists the dorsiflexion movement.	Hips at 80°, knees at 115°. Knee and machine's lever arm center of rotation aligned. Lever arm positioned at the distal shank.	Hips at 80°, knees at 0-5°. Knee and machine's lever arm center of rotation aligned. Lever arm positioned at the distal shank.	Hips at 110-90°, knees at 90-100°. Feet and knees at hip width. 0-20° of hip external rotation is allowed.	Standing with hips and knees at 0°. Holding an adjustable support with both hands. Feet and knees at hip width. 10-20° of hip external rotation is allowed.	Supine position, arms laying by the side and both legs touching the mat. An elastic band on the forefoot resists the hip flexion and ankle dorsiflexion.	Hips and knees at 0°, the chair is 30° inclined. Padded support at pelvis height. Distal posterior part of the shank locked against a padded support.	Supine position, arms laying by the side and both legs touching the mat.
Kinesiologic Description	Unilateral or bilateral ankle plantarflexion.	Unilateral or bilateral ankle plantarflexion	Unilateral or bilateral ankle dorsiflexion.	Unilateral or bilateral knee extension.	Unilateral or bilateral knee flexion.	Unilateral or bilateral hip and knee extension, and ankle plantarflexion.	Bilateral hip and knee extension, and ankle plantarflexion	Unilateral hip flexion and isometric ankle dorsiflexion.	Isometric trunk and hip extension.	Isometric trunk and hip flexion. Knee extension.

Hip 0° = anatomical position (positive values = flexion), Knee 0° = fully extended. Ankle 90° = sole of the foot at right angles with tibial axis, > 90° = plantarflexion.

*If the exercise is too hard, leaning on the bars and helping with the arms will be allowed. If it becomes too easy, it will be done at the leg press machine.

TABLE 2. Training progression (left) and example exercise list per training session (right)

Week	Volume	Load	Movement Duration (s)	Rest (s)	Session A*	Session B*
1 - 4	3 sets of 8 repetitions	8 RM	3 concentric 3 eccentric	60	1 – Seated calf raise 2 – Seated dorsiflexion	1 – Seated machine knee flexion 2 – Seated machine knee extension
5 - 8	3 sets of 8 repetitions	8 RM	1 concentric 3 eccentric	90	3 – Standing calf raise 4 – Hip flexion	3 – Hip flexion 4 – Standing calf raise
9 - 12	4 sets of 6 repetitions	6 RM	! concentric 2 eccentric	90	5 – Seated horizontal leg press 6 – Roman chair trunk extension 7 – Squat	5 – Seated horizontal leg press 6 – Isometric hollow rocks 7 – Squat

! = ballistic muscle action; RM = repetition maximum; * = each session has a minimum of 7 exercises and a maximum of 10 (i.e. all exercises).