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**MOVEMENT PATTERN,  
POWER GENERATION AND  
WELL-BEING IN CHILDREN  
AND YOUNG ADULTS WITH  
SPASTIC UNILATERAL  
CEREBRAL PALSY**

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To my family

## ABSTRACT

Patients with unilateral cerebral palsy (CP) all walk, most of them without great difficulty and without assistive devices. However, their walk tends to catch the eye because of abnormal, often asymmetrical movement in the upper and lower extremities. Although the impairment is limited and the involvement often is considered mild the deviation is apparent in both movement pattern and anatomy, with one leg being smaller and shorter.

Some of the patients have a more severe CP diagnosis which can include epilepsy, learning and psychological problems apart from the motor function impairment with spasticity, decreased motor control, co-contraction, muscle and joint contractures and muscle weakness and deformities. The expression of the static brain injury varies tremendously between individuals and changes with growth, which makes prognosis and setting of long term goals difficult.

Nevertheless many of the children, teenagers and young adults with unilateral CP manage well; for them treatment is seldom considered or recommended. On the other hand the knowledge about possibilities and available treatment options is limited. We consider these patients high functioning in relation to other groups with CP and have not paid much attention to their possible concerns and wish to normalize their movement pattern.

In these studies the goal was to explore different dynamic changes in movement pattern of the upper and lower extremities, explore anatomical changes in terms of muscle volume and leg length discrepancy and finally assess well-being with respect to self-esteem, sense of coherence and quality of life. Specifically we wanted to study variables that can help in the classification of movement pattern and help identify and separate between primary deviations caused by the brain injury and deviations arising from compensation mechanisms on both the involved and non-involved side.

The widely used Winters classification, using Gait analysis, was unable to classify 23% of children with a mean age of 8.1 years (3.6-19.8 years). For these patients we created a new complementary type to the Winters classification, type 0, describing mild impairment. Patients move between the classification types over time and with treatment. Concentric muscle work is redistributed from the ankles joints to the hip extensors in children on both the involved and non-involved side and in older patient groups, mean age 17.6 years (13.0-24.0 years) on the involved side. The amount of hip extensor work exceeds that in the control group. There was significant decrease of muscle volume on the involved side compared to the non-involved side, with more pronounced changes distally in the limb, on Magnetic resonance imaging assessment. There was a significant leg length discrepancy in the tibia, talus and calcaneus. Patients scored lower in self-esteem and quality of life assessment than the control group and there was a significant correlation between arm posture deviation and self-esteem.

In summary, with the new type 0 as a complement of the Winters sagittal plane kinematic classification, other variables can be useful when evaluating high functioning patients with unilateral CP. Advanced quantitative objective measurements such as Gait analysis and Magnetic resonance imaging may be useful to develop and evaluate rational treatment protocols.

The impact on self-esteem and quality of life should not be underestimated in teenagers and young adults with spastic unilateral CP.

## LIST OF PUBLICATIONS

The thesis is based on the original papers listed below. They will be referred to in the text by their Roman numerals.

- I. Classification of Spastic Hemiplegic Cerebral Palsy in Children.  
Jacques Riad, Yvonne Haglund-Akerlind, Freeman Miller.  
Journal of Pediatric Orthopedics October/November 2007; 27 (7), 758-764
- II. Power generation in children with spastic hemiplegic cerebral palsy.  
Jacques Riad, Yvonne Haglund-Akerlind, Freeman Miller.  
Gait and Posture 2008; 27, 641-647
- III. Leg Length Discrepancy in Spastic Hemiplegic Cerebral Palsy: A Magnetic Resonance Imaging Study.  
Jacques Riad, Thröstur Finnbogason, Eva Broström.  
Journal of Pediatric Orthopedics December 2010; 30 (8), 846-850
- IV. The impact of muscle volume differences on concentric muscle work during walking in spastic hemiplegic cerebral palsy.  
Jacques Riad, E.M. Gutierrez-Farewik, Christopher M. Modlesky, Freeman Miller, Eva Broström.  
Submitted.
- V. Self-esteem and sense of coherence in relation to upper- and lower extremity movement deviation during walking in teenagers and young adults with mild unilateral cerebral palsy.  
Jacques Riad, Eva Broström, Ann Langius-Eklöf.  
In manuscript.

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## ABBREVIATIONS AND EXPLANATIONS

GA	Three dimensional gait analysis
CP	Cerebral palsy
LLD	Leg length discrepancy
EMG	Electromyography
equinus	Plantar flexion (positioning of the foot in the ankle joint)
unilateral	One side
bilateral	Both sides
MRI	Magnetic resonance imaging
Rom	Range of motion
PODCI	Pediatric Outcome Data Collection Instrument
FAQ	Functional Assessment Questionnaire
FMS	Functional Mobility Scale
GMFCS	Gross Motor Function Classification System
GPS	Gait Profile Score
APS	Arm Posturing Score
QoL	Quality of Life
SOC	Sense of Coherence
ITIA	I think I am

# 1 INTRODUCTION

This thesis is focused on high functioning young patients with unilateral cerebral palsy. The main purpose – to explore their movement patterns during walking and their well-being – was prompted by the surprising scarcity of documentation and counseling of these individuals, as well as the lack of long term follow-up into adulthood.

The hypothesis was that the movement pattern during walking is important for more than just getting efficiently from point A to point B, and that compensation mechanisms often develop even in high functioning patients. There seems to be a strong, more or less conscious drive towards normality and symmetry in movement pattern. The reason could be functional aspects of movement (a plantigrade foot pointing forward is a prerequisite for efficient gait). It could also reflect a wish not to stick out in a crowd but rather be apprehended as normal, a wish especially pronounced in teenagers and young adults in their transition into adulthood. In patients with unilateral cerebral palsy the impairment is often mild but the movement pattern is nevertheless obviously deviant; this could affect self-esteem and well-being. Quantitative objective measurement tools make careful assessment of gait deviations possible and can prove useful to gain more knowledge and to develop rational treatment.

## 1.1 SPASTIC UNILATERAL CEREBRAL PALSY

### 1.1.1 Definition

Until a few years ago, unilateral cerebral palsy (CP) was called hemiplegic CP. The nomenclature has changed and CP is now described in terms of the type of motor disturbance (spastic, dyskinetic, ataxic, hypotonic and mixed) and further classified according to topographical distribution (unilateral or bilateral CP) [1-3].

By far the most common form of unilateral CP is spastic, the focus of the thesis. When the term hemiplegic has been used in quoted or cited text, it will usually be translated into unilateral CP. Nevertheless the term hemiplegic will be used occasionally in older titles and earlier produced images.

CP is a description of symptoms rather than a diagnosis [4]. CP is “an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of development” [1, 5]. The symptoms of the motor impairment that predominantly lead to disturbed motor function are spasticity, deficits in activation and co-activation with insufficient selective motor control, decreased passive motion with muscle contractures and joint deformities and muscle weakness [6-8].

Spasticity is defined as an increased, velocity-dependent, response to passive muscle stretch [9]. Motor control from the central nervous system is impaired, especially the counterbalancing inhibitory function, which gives rise to an increased muscle tone and uncontrollable movements.

Hyperactive reflexes and/or positive Babinski sign is sometimes also present on the contralateral side. Uvebrant states that it can be difficult to separate unilateral from bilateral CP, but also states that as long as the impairment on the “normal” side is not associated with any degree of disability, the diagnosis should be unilateral CP [10].

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However the cut off between unilateral and asymmetrical bilateral CP has been discussed [1]. In these studies any increased reflexes or increased muscle tone on the contralateral side has been an exclusion criterion.

### 1.1.2 History

In modern times, the physician William John Little (1810-1894) made observations on children with increased muscle tone after complicated deliveries. Little, who had suffered from polio as a child, had a residual deformity of his left foot (talipes equinovarus) as an adult. The foot deformity was much improved by an Achilles tendon lengthening performed by the German surgeon Stromeyer from whom Little learnt about the procedure. According to Mercer Rang, Little published his thesis in 1862 entitled [11].

“ON THE INFLUENCE OF ABNORMAL PARTURITION,  
DIFFICULT LABOURS, PREMATURE BIRTH, AND ASPHYXIA  
NEONATORUM, ON THE MENTAL AND PHYSICAL CONDITION  
OF THE CHILD, ESPECIALLY IN RELATION TO DEFORMITIES”

However the condition cerebral palsy had been noted long before, in ancient Egypt, and it may even be possible to distinguish unilateral CP in Egyptian archeological artifacts (Figure 1 and 2).

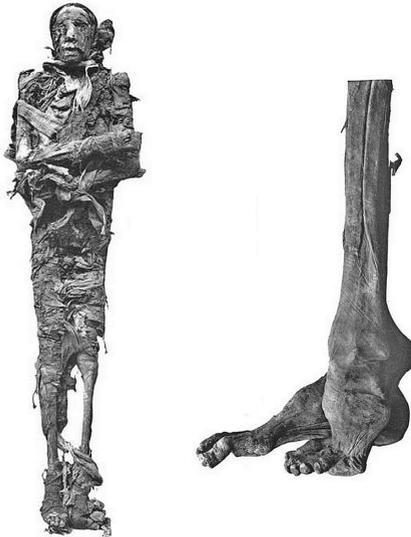


Figure 1. The pharaoh Siptah, who died young after reigning for only 6 years (1194-1188 BC), might have had spastic unilateral CP. Siptah’s mummy found in 1905 clearly shows the pharaoh’s left leg to be shorter than his right; his left foot was severely deformed with ankle equinus and the arm was in a “peculiar” position. The first proposed diagnosis of polio was challenged by Aufderheide and Rodriques-Martin in 1998 after reexamination. (See reference 11 page 282) [12]. The Egyptian Museum of Cairo.



Figure 2. The door keeper Roma lived sometime in the 18-20th Dynasty (1550-1080 BC). Judging from his short and thin leg he probably had polio or hemiplegic cerebral palsy. Despite his disability he achieved high status, acquired wealth and was married and had at least one child. Here he is seen sacrificing to Pharaoh along with his wife and son. Specimen 134. Courtesy of New Carlsberg Glyptotek, Copenhagen.

Gait analysis (GA), which is one of the tools used to measure outcome in the thesis, started in California in the 1870s. Leland Stanford bet 25 000 dollars (huge amount of money at the time) that a racehorse has all its legs off the ground at a time. Eadweard Muybridge used the time-sequence photography he had pioneered to help Stanford win the bet. Verne Inman (1905-1980), also working in California during the 1950s, introduced the concept “determinants of gait”. World War II left many soldiers and civilians as amputees and polio was still widespread. This prompted David Sutherland and Jacqueline Perry to continue to develop this concept, and their work forms the basis for modern gait analysis [11, 13].

### 1.1.3 Prevalence

Cerebral palsy (CP) constitutes the largest group of motor deficits in children, with prevalence around 2.3 of 1000 newborns in the western world [14, 15]. Unilateral CP is the most common CP syndrome among children born at term and second to bilateral CP in preterm children [16]. Unilateral CP occurs in around 6-7 of 10 000 births and accounted for 38% of all CP in a recent study [4, 14, 17].

### 1.1.4 Pathogenesis and etiology

The timing of adverse events in the developing brain determines the anatomical localization and type of lesion and thus the final neurological picture [18]. Three main pathogenic processes exist, each occurring at a different developmental stage. The first is malformations originating in early fetal life, the second is periventricular lesions mainly arising at 24-34 weeks of gestation and the third is cortical lesions that arise just before or at full term birth [18].

Different genetic and environmental risk factors render different pathogenesis. There are reports on families with an autosomal dominant inheritance and there are known risk factors such as preterm birth, small for gestational age, and also that the mother has experienced health problems during pregnancy [19-21]. Brain injuries seen in children born at term most often have an intrauterine origin [22, 23]. In preterm children with unilateral CP adverse perinatal complications have usually contributed to the brain injury [17, 24]. In other words, genetic and environmental risk factors influence which child will have unilateral CP. We do not know, however, to what degree and in what combinations these variables play a role. In the future some of them may have implications for prevention [20].

Advances in imaging of the brain with computer tomography and magnetic resonance imaging (MRI) allow documentation of the brain lesion and indicate at what timepoint it probably arose. The diagnosis can sometimes be confirmed, especially if there is malformation, and other pathology ruled out. The imaging may help physicians explain to parents how the injury arose, and provides epidemiological data [23, 25, 26]. Morphological findings in brain imaging of patients with CP are rarely an exact match with the clinical presentation and vice versa and so far imaging has limited clinical implications for functional mobility and certainly for movement pattern [26]. However, in unilateral CP, clinical manifestations can correlate with the brain image. Children with cerebral malformation had impairment of growth of arms and legs while periventricular lesions correlated only with impaired growth of the leg. Lesions close to the frontal horn correlated with arm dominance and patients with cortical and subcortical lesions had a more severe total disability [27].

### 1.1.5 Epilepsy, learning and psychological problems

Epilepsy is present in around 25 % of all patients with unilateral CP depending on the definition and age groups examined [17, 28]. Learning disability occurs in one of five individuals although several studies found no difference compared to controls on educational measures, including literacy and numeracy subtests [17, 29, 30]. Others report that more than one-third have specific learning difficulties [31].

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The London Hemiplegia Register study which included a large sample of children explored rate, type and persistence of psychological problems. Using a questionnaire they found that among 6 to 10 year olds, 61% had at least one diagnosable psychiatric disorder. Common types were anxiety or depressive disorders (in 25%), conduct disorder, characterized by disruptive and irritable behavior (in 24%), severe hyperactivity and inattention (in 10%) and autistic disorders (in 3%) [32]. According to Uvebrant “epilepsy and learning disability are the major accompanying impairments in unilateral CP” [10, 33].

### 1.1.6 Clinical characteristics

The signs of unilateral CP develop gradually during the second half of the first year. Sometimes early handedness when the child begins to develop grasping, around 3 to 6 months of age, is the first time parents suspect something is not right. A later manifestation is relatively late unsupported walking at around 18 months of age. All children with unilateral CP will walk, unless there is a severe additional developmental disorder or sequelae of grave epilepsy [33]. The inter-individual variability in the severity of the motor impairment is pronounced. One patient out of five has severe impairment; half have moderate and one third have mild impairment. Arm dominated unilateral CP is most common in children born at term and the leg is more impaired in children born preterm [17, 28, 29, 34].

The gait is characterized by asymmetry. There is difference in step length, and often the foot on the involved side is to a greater or lesser degree in equinus (plantar flexion) causing toe walking or early heel rise. Depending on the degree of spasticity and the level of involvement movement can be altered and decreased, also in the knee and hip joint [35, 36].

The arm is often in a flexed position in the elbow and wrist, the forearm in pronation and the thumb adducted [36].

The involved limb in spastic hemiplegic CP is usually smaller and shorter than the other limb. It is not clear whether the size difference arises because the brain injury has an impact on nerve innervations and growth factors or because the mechanical loading that normally stimulates growth is reduced [35-38]. Wilkins proposed in 1955 that decreased blood flow caused the difference in length and size [39]. The leg length and size discrepancy between the involved and non-involved side adds to the asymmetrical appearance and presumably, movement pattern.

### 1.1.7 Changes with time and growth

In unilateral CP, as in other groups of CP, there are changes with time, although the brain injury underlying the spasticity is static. The changes are attributed to further growth, the maturation of the central nervous system, the development of secondary deformities and compensation mechanisms, together with the influences of various treatments [36, 40-45]. It is reported that some children deteriorate early and lose skills but on the other hand in some cases the symptoms of unilateral CP disappear between 3 and 10 years of age [46, 47]. In conjunction with the adolescent growth spurt some patients experience an increased muscle tone and spasticity, most probably due to the fact that soft tissue length and growth lag behind the skeletal growth [36].

### 1.1.8 Long term goal

After the diagnosis is set, the family needs to be informed and reassured that they are not to blame and should feel no guilt. Once this is done it becomes important to focus on the child's movement, walking, grasping and make a general assessment of what functional development can be anticipated [40].

To set a realistic long term goal, one must be able to make an accurate prognosis for the individual. From an orthopedic point of view, the long term goal is to optimize the person's ultimate independent mobility and consider psychosocial function and social integration into adulthood [36, 48]. Treatment involving orthopedic surgery should ideally be completed before the patient reaches 18 to 20 years of age, which is the time when the patients are most often discharged, and stop being seen by the pediatric orthopedic surgeon and the pediatric caring team [36].

### 1.1.9 Classification and functional mobility

Classification systems and scales can help in the communication between caregivers and make comparisons and evaluations of treatment possible. Classification systems can also be useful for treatment planning and in determining prognosis.

There are several classification systems and scales assessing physical function and walking ability in children and adults with disabilities.

Some systems are of a general character used in different patient groups, such as the Pediatric Outcome Data Collection Instrument (PODCI), Functional Assessment Questionnaire (FAQ) and the Functional Mobility Scale (FMS) [49-51]. There are also classification systems specific for CP, the Gross Motor Function Classification System (GMFCS), and for subgroups of CP (e.g., Rodda's system for gait patterns in spastic uni- and bilateral CP) and lastly different specific types within the subgroup of CP (e.g., Winters' classification of unilateral CP) [52-54].

The PODCI, developed by the North American Pediatric Orthopedic Society, measures outcomes that orthopedic treatment can affect, in different age groups and in different musculoskeletal disorders [51]. It includes scales assessing upper extremity function, transfers and mobility, physical function and sports, comfort (pain free), happiness and satisfaction, and expectations with treatment. The PODCI was explicitly designed to measure functional health focusing on musculoskeletal health.

The walking scale in the FAQ is a ten-point scale rating gait function regardless of pathology, developed mainly for children [50]. The parent ranks the child's ability to walk independently from the need of assistive devices, from the lowest function "Cannot take any steps at all" to the highest community function "Walks, runs and climbs on even and uneven terrain without difficulty or assistance".

Tervo et al used the PODCI, the FAQ, the Normalcy Index (a measure of amount of deviation in gait from normal), Energy expenditure and Gait analysis to measure improvement of physical function after intervention. They report that the PODCI scale discriminates between children with lesions of specific topographical classifications and (not surprisingly) that those with unilateral CP perform best and those with bilateral CP and more severe involvement perform worse [55].

The FMS has a six level rating scale and is used to assess functional mobility over three different distances, representing mobility at home (5 meters), in school (50 meters) and

## Introduction

in the wider society (500 meters) [49]. The FMS can discriminate between larger groups of children with varying levels of disability and functional mobility. In addition it is sensitive method to detect change after surgical intervention [49].

The GMFCS was developed for children with CP [53]. This five level classification scheme is based on abilities and limitations in gross motor function and discriminates between differences in self-initiated movement, with particular emphasis on sitting and walking (Figure 3). The GMFCS is applicable and classifies all patients with CP, from independent ambulators with very little impairment (GMFCS level I), to those that can hardly move even with wheelchair and assistive technology (GMFCS level V).

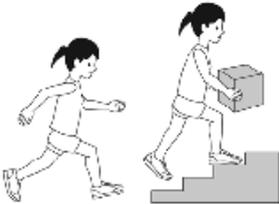
Winters' classification is uniquely used to classify gait pattern in patients with unilateral CP and is based on sagittal plane gait analysis data [52].

In addition the World Health Organization developed the International Classification of Function, Health and Disability (ICF) with the goal of a holistic and comprehensive view on function, health and disability [56]. According to Rosenbaum, this model offers many heretofore ignored "points of entry" for counseling and intervention in children and youth with CP [57]. This new tool de-emphasizes the patient's disease and impairment and focuses more on the components of health than on the consequences of disease. Participation and activity are two key concepts in the ICF.

Several of these classification systems and scales are not sensitive enough to capture deviations in the higher functioning groups; this is a potential problem. When used on patients with mild unilateral CP, the GMFCS, FMS and the PODCI are probably going to assign a majority of the patients the highest or close to highest classifications/scores available (ceiling effect).

In conclusion, the classification systems and scales used for high functioning groups need to be sensitive enough and appropriate (regardless of characteristics assessed). For that reason there was an interest to study the applicability of the Winters classification system developed for unilateral CP [52]. Obviously, if the Winters classification is to be used, the patient not only needs to have the diagnosis unilateral CP, but also be able to walk independently, without assistive devices, which excludes those with GMFCS level III, IV and V. In other words we are studying high functioning individuals with mild unilateral CP, with minor limitations in their ability to participate in society and with peers.

## GMFCS E & R Descriptors and Illustrations for Children between their 12<sup>th</sup> and 18<sup>th</sup> birthday

	<p><b>GMFCS Level I</b></p> <p>Youth walk at home, school, outdoors and in the community. Youth are able to climb curbs and stairs without physical assistance or a railing. They perform gross motor skills such as running and jumping but speed, balance and coordination are limited.</p>
	<p><b>GMFCS Level II</b></p> <p>Youth walk in most settings but environmental factors and personal choice influence mobility choices. At school or work they may require a hand held mobility device for safety and climb stairs holding onto a railing. Outdoors and in the community youth may use wheeled mobility when traveling long distances.</p>
	<p><b>GMFCS Level III</b></p> <p>Youth are capable of walking using a hand-held mobility device. Youth may climb stairs holding onto a railing with supervision or assistance. At school they may self-propel a manual wheelchair or use powered mobility. Outdoors and in the community youth are transported in a wheelchair or use powered mobility.</p>
	<p><b>GMFCS Level IV</b></p> <p>Youth use wheeled mobility in most settings. Physical assistance of 1-2 people is required for transfers. Indoors, youth may walk short distances with physical assistance, use wheeled mobility or a body support walker when positioned. They may operate a powered chair, otherwise are transported in a manual wheelchair.</p>
	<p><b>GMFCS Level V</b></p> <p>Youth are transported in a manual wheelchair in all settings. Youth are limited in their ability to maintain antigravity head and trunk postures and control leg and arm movements. Self-mobility is severely limited, even with the use of assistive technology.</p>

GMFCS descriptors copyright © Bakstano et al. (1999) Dev Med Child Neurol 39:214-21  
 CanChild: www.canchild.ca

Illustrations copyright © Kerr Graham, Jill Reel and Adrienne Harvey,  
 The Royal Children's Hospital, Melbourne

Figure 3. The Gross Motor Function Classification Scale between 12<sup>th</sup> and 18<sup>th</sup> birthday. With permission from Kerr Graham.

### 1.1.10 Rationale for these studies

Some high functioning patients with unilateral CP have deviations from the normal movement pattern that are so subtle that they are difficult to detect and treat. Clearly these patients differ from other CP patients in terms of abilities and treatment goals. The treatment team around these patients may tend to underestimate the possibilities and tone down the expectations of treatment. Not much attention has been directed toward this group of patients regarding ambulation and their possible concerns about their movement pattern and appearance.

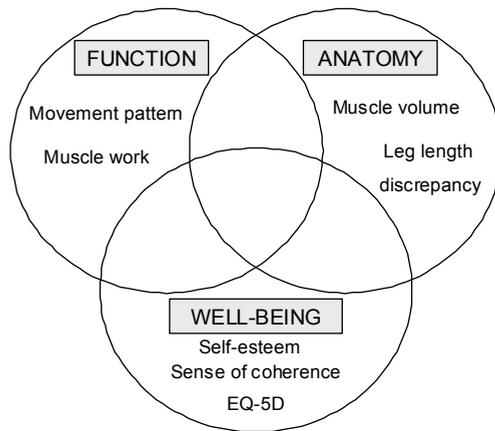


Figure 4. Multiple variables providing information from different domains were included in these studies, in an attempt to explore relationships between static anatomical changes, dynamic functional changes and influence on the individuals' well-being.

The basic idea underlying these studies is that subtle changes in movement pattern can only be appreciated if one uses suitable classification systems and bases the assessment on objective quantitative measurements. According to this line of thinking, use of the Winters classification system based on Gait analysis data and other variables such as muscle volume, muscle work and leg length differences might point to where effort should be directed to improve the patient's functional capacity. Primary, secondary and compensation mechanisms can be identified and rational treatment strategies developed. Finally knowledge about the influence of movement patterns on quality of life and well-being can provide an individual perspective and support the decision making. In this way treatment options can be presented and recommendations made with more informed patients and families (Figure 4).

## 1.2 MOVEMENT PATTERN DURING WALKING

Lack of neurological control gives rise to impaired motor function and there is a pronounced individual variety of movement patterns in ambulatory children with CP [40, 44]. Spastic unilateral CP constitutes the group of patients with CP with the best walking capacity. Nevertheless these high functioning individuals often have a noticeable asymmetry and deviant movement pattern during walking both in the gait pattern and the arm positioning and movement [58-60].

It can be difficult to separate primary deviations caused directly by the brain injury from secondary deviations that are consequences of the primary deviation [61]. In addition, one must consider the more or less voluntary compensation mechanisms that develop on both the involved and non-involved side [61]. Deviations depending on the impaired motor control and/or depending on structural changes such as leg length discrepancy adds to the difficulty of interpretation [62-64].

In the lower extremity the gait deviations mainly involve the foot and ankle but compensation mechanisms may lead to increased movement in the knee and hip joint. In more severe cases of unilateral CP the knee and hip are also directly involved, with impaired motor control, increased muscle tone and spasticity [52]. In the arm, the increased muscle tone leads to posturing and decreased motion during walking. The upper limb is often held flexed at elbow, wrist and fingers and is internally rotated [65]. The deviations often become more apparent with transitions between movements and during rapid movement, but also when the person is emotionally affected [66-68]. Arm posturing and gait deviations may be perceived as cosmetic and social impediments when the individual enters adolescence and becomes more self conscious [69, 70].

### 1.2.1 Three-dimensional motion analysis

“The purpose of gait analysis (GA) is to document the gait patterns and to help the clinician understand the causes of the gait abnormalities better by separating the primary from secondary deviations and compensation mechanisms” [61].

The gait is described for each limb separately. The gait cycle is divided into stance phase and swing phase. Stance phase starts with initial contact of the foot and ends with toe-off after approximately 60% of the gait cycle. With toe-off the stance phase ends and the swing phase starts. The swing phase makes up approximately 40% of the gait cycle and ends when that same foot that started the gait cycle gets contact with the floor again (initial contact). Double support time, when both feet have contact with the floor occurs twice in the gait cycle and lasts around 10 % each [13].

The main unique features of three dimensional motion analysis are the collection of data on the movement of different body segments in three dimensions (kinematics) and the analysis of the forces acting on the body segments (kinetics). The patient walks with several reflective markers on specific bony landmarks on the body which define the body segments. The markers' movement in the room is captured by several cameras, the different body segments are defined and their orientation in three dimensions during the gait cycle is collected. At the same time force plates in the floor register the position of the ground reaction force as the patient walks. From the

## Introduction

kinematic data and various models, the forces acting over different joints during the gait cycle can be calculated [71].

Beside the kinematic and kinetic data, temporal-spatial data are collected, mainly walking velocity, steps per minute, step length and step width [13].

Gait analysis (GA) helps identify and interpret different aspects of gait, but does not provide specific treatment protocols. The data provided need to be interpreted and weighed together with clinical findings, patient's concern, radiological assessments and other variables of importance before recommendations can be made [72, 73].

GA helps the physician identify problems that could be addressed through treatment. In this, GA differs from other, more general outcome tools, such as energy expenditure, walking velocity, functional mobility classifications, etc, which do not. These outcome tools can be used in the decision making and interpretation of the GA data (the kinematics and kinetics) but will not provide guidance in what measure/treatment to undertake. In this context modern GA combined with long clinical experience of this patient group is the ideal armory for physicians making recommendations to children, teenagers and young adults with CP and their families on how to improve functional mobility and gait pattern.

### **Winters' gait analysis criteria**

In mild unilateral CP the most frequent movement deviations are in the sagittal plane in the ankle joint, although the deviations can be very subtle [52, 74-76].

Winters' classification based on sagittal kinematics from the ankle, knee and hip is used to classify and describe gait patterns [52]. In study I the main aim was to assess the applicability of the classification. In studies II-V the system was also applied. The classification defines four gait patterns, where type 1 is the least involved and type 4 the most involved. Figure 5.

The type 1 deformity is caused by a weak or silent ankle dorsiflexor, a very rare finding in patients with cerebral palsy but common in adult patients after stroke or with a peripheral nerve injury. The gastrocnemius-soleus complex does not have excessively increased muscle tone. Type 1 has ankle plantar flexion in swing phase.

Type 2 is the most common among patients with CP and is characterized by a spastic or contracted gastro-soleus complex that causes plantar flexion with a more or less pronounced equinus gait. Type 2 has ankle plantar flexion in stance and swing phase.

In type 3 the knee is included in addition to the ankle deformity of type 2. The knee involvement is often an overactive and spastic hamstring muscle activity giving a short step and excessive knee flexion in stance. In the physical examination, hamstring contracture is noted and in some cases knee joint contracture develops.

Knee involvement may also include the stiff knee gait caused by a spastic rectus femoris muscle, with the typical toe drag in swing.

## Introduction

In the rare type 4 unilateral CP pattern, the hip joint is involved in addition to the ankle and knee joint. An increased muscle tone in the adductor and flexor muscles of the hip is noted in the clinical examination and there is decreased hip extension and range of motion during walking.

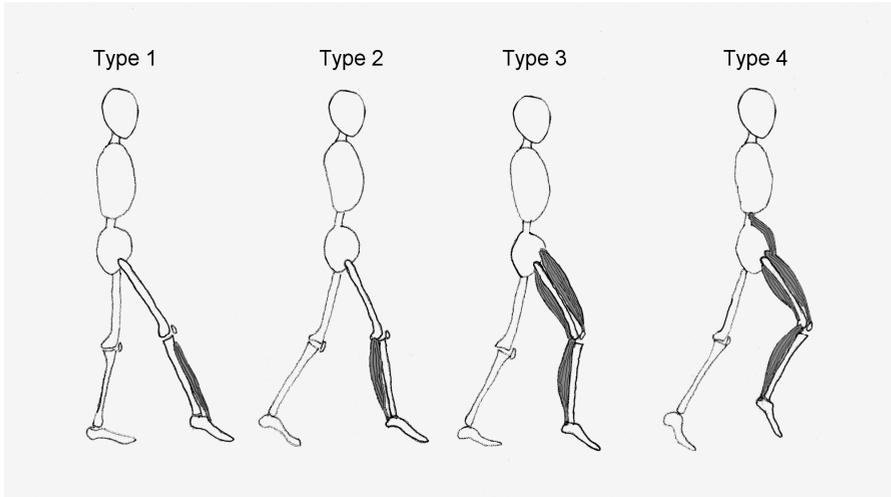


Figure 5. Winters' Classification – types of unilateral CP.

The criteria used for Winters' classification are based on kinematic data (Figure 6) and to some extent on electromyography (EMG). The EMG changes are however discussed very little in the original article and are only sparsely discussed together with the classification system elsewhere [52].

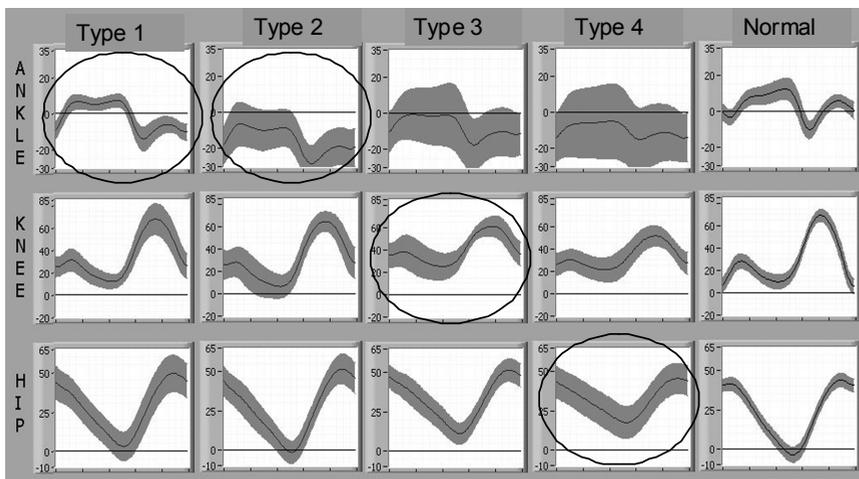


Figure 6. Kinematic changes in the sagittal plane. The ankle, knee and hip joint in the four types and in normals.

## Introduction

Type 1. Ankle joint involvement with plantar flexion at initial contact and no dorsiflexion above neutral in swing phase. The tibialis anterior muscle is silent or nearly silent on EMG.

Type 2. Ankle joint involvement with plantar flexion (equinus) during stance and swing phase. Secondary changes can be seen in the knee joint. Knee flexion at initial contact may be increased as well as knee extension in mid stance.

Type 3. Ankle involvement as type 2 and knee involvement with knee range of motion less than 45 degrees in the gait cycle and less than 50 degrees flexion in swing phase. Knee flexion will be increased during early and mid stance phase. EMG reveals early onset in swing phase and prolonged activity into stance phase.

The stiff knee pattern (not shown here) shows typical delayed and decreased flexion in swing phase in the knee joint and the EMG activity in the rectus femoris muscle is prolonged into swing phase.

Type 4. Ankle and knee involvement as above and hip involvement with hip range of motion less than 35 degrees.

Transverse and coronal changes are not included in the Winters classification. In some patients especially type 4 there is obvious rotational deformity in the hip with secondary changes in the pelvis kinematics. Increased movements in the pelvis and hips with abduction pattern to ensure foot clearance can be present in the coronal plane in type 1 and 2. Hip adduction can be noted in the coronal plane and is caused by increased spasticity in type 4 patients.

### **Deviation scores**

Recently, to obtain a measure of the overall movement deviation, scores and indices has evolved [50, 77, 78]. In the Gait Profile Score (GPS), the sum of deviations from several joints and dimensions from the lower extremity curves, collected with the GA is calculated. In a corresponding way the Arm Posture Score (APS) was described [58]. A movement deviation score for both gait pattern and arm posturing during walking is provided.

Even though the classification of Winters is widely used and has clinical implications, it has limitations. The classification published in 1987 was developed from observation of both children and adults and includes not only patients with cerebral palsy but also adult patients with stroke and traumatic brain injury.

The gait patterns in the different types of unilateral CP classified according to Winters are not always straightforward and clear. The age of the patient and the degree of maturation at the time of assessment, as well as previous treatment, can influence the results. A recent study on a population basis has shown that many patients cannot be classified according to Winters' criteria [76]. Several authors report problems using the classification and an update is warranted [54, 60].

Furthermore a more holistic and comprehensive approach to the entire movement pattern during walking would be of interest, especially an approach that included the often clear asymmetry in both the lower and upper extremities. It is reasonable to assume that the upper and lower extremity movement patterns influence each other.

### 1.3 CONCENTRIC MUSCLE WORK AND MUSCLE VOLUME

Muscle weakness is common in CP and is a part of the motor impairment [7]. Weakness is defined as failure or inability to produce or maintain an anticipated level of force [79]. The effectiveness of muscle contraction depends on several factors such as joint stability, lever arm length and positioning and coordination of muscle contraction. In addition intrinsic properties, such as fiber size and fiber type distribution determine the outcome of contraction [7, 35, 80, 81].

As discussed earlier in the text the involved limb in unilateral CP is usually smaller and shorter than the non-involved limb. It has been reported that both patients with unilateral and bilateral CP have smaller muscle volumes in involved lower limbs than normal controls. The measurements were performed with ultrasound, computer tomography and Magnetic resonance imaging [82-84].

It is known that muscle volume and muscle cross sectional area in healthy individuals is correlated with strength, maximum voluntary isometric contraction [6, 81, 85-91]. It is not clear whether the same principle applies in CP and if increased muscle strength influences gait. McNee and co-workers reported an increase of muscle volume in 13 ambulant children with spastic CP after plantar flexor strengthening training. These children gained in strength (number of heel rises) but not in function or gait [92]. In a comment, Damiano and Moreau mention unpublished data regarding muscle thickness in rectus femoris and vastus lateralis in relation to knee extensor maximal voluntary isometric contraction (MVIC) test in a group of children with CP (GMFCS II-IV) and age matched controls. Muscle thickness explained 50 to 70% of the variance in strength in the control group, but only about 22 to 32% for the group with CP [93]. The same authors state: “the potential for strength training to improve the gait kinematics of people with CP has been evaluated in only a few studies, and findings have been equivocal” and they cite references [88, 94, 95].

Muscles contribute differently to gait. Muscle contraction with lengthening (eccentric) and muscle contraction keeping the same length (isometric) act to stabilize. Muscle contraction with shortening (concentric) produces positive work responsible for propulsion [13] (see also Figure 7A and B). Modern computer-assisted modeling and simulations have made it possible to study muscle work during walking in more detail. Models and simulations can also be created to mimic clinical situations [96-98]. Kinetic data derived from the force plates in the Gait laboratory describe forces acting over the joints [35]. From the kinetic and kinematic data the timing and amount of muscle work over the different joints can be calculated.

Muscles responsible for propulsion through concentric muscle contraction are: Hip extensors (gluteus maximus), Hip flexors (medial hamstrings, the adductors and the iliopsoas), Knee extensors (quadriceps) and Ankle plantar flexors (gastrocnemius and soleus).

## Introduction

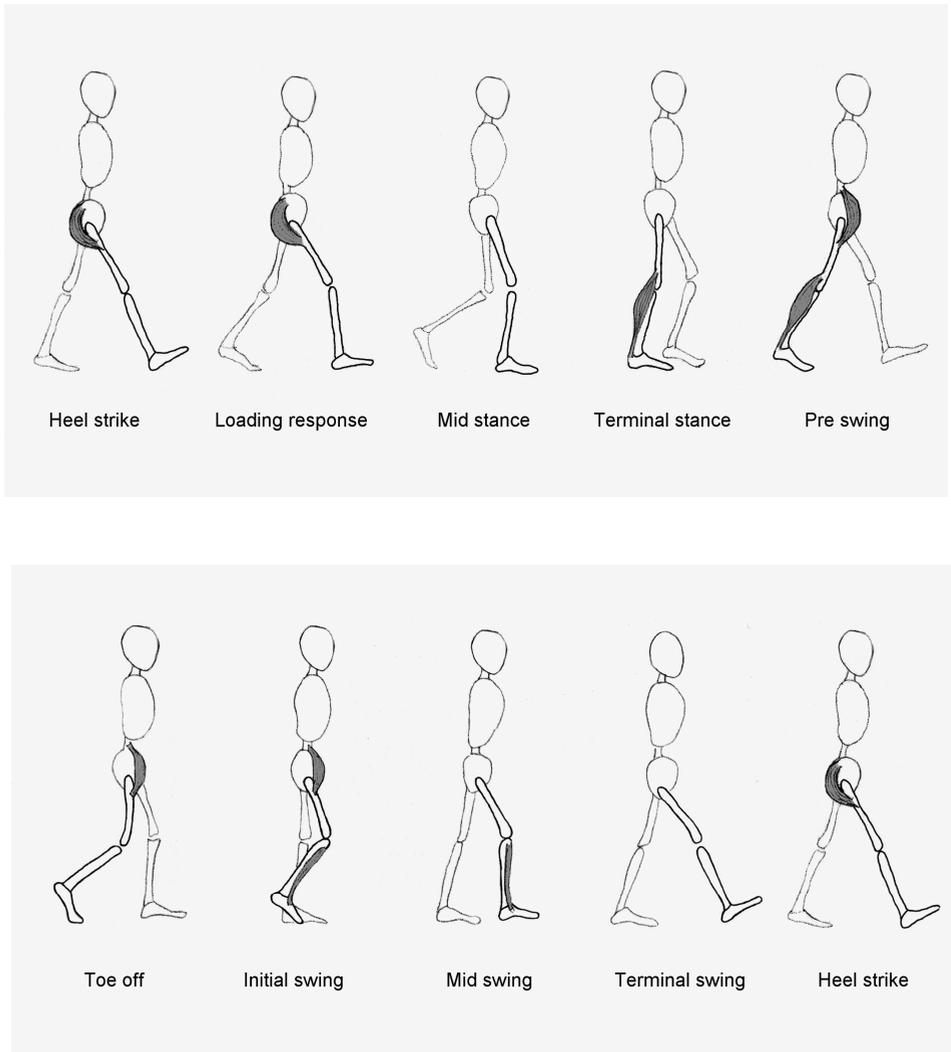


Figure 7 A and B. Muscles with concentric muscle contraction. Hip extensors contract in early stance, gastrocnemius in terminal stance and pre-swing, and hip flexors in pre-swing, at toe off and initial swing. Knee extensors, not drawn, contract during midstance.

There is a lack of understanding of the relation between muscle strength and muscle volume in separate muscle groups and the difference between the involved and non-involved side. The effect of these variables on gait in high functioning individuals with unilateral CP is not known.

Does difference in muscle volume correlate with muscle work during walking? Could alterations in the amount of work exerted in specific muscle groups help distinguish

compensation mechanisms from primary and secondary deviations [96, 97, 99-101]? Do patients with mild unilateral CP have sufficiently better motor control than more involved CP patients that muscle strengthening would have a gait-normalizing effect?

### 1.4 LEG LENGTH DISCREPANCY

Leg length discrepancy (LLD) is not uncommon and might occur in 50-70% of the general population [102, 103]. The effect in terms of chronic back pain and arthritis of the hip and knee and where a “cut-off” for when to treat has been long discussed [104, 105]. Compensation mechanisms for LLD, such as increased flexion in the hip and knee, and increased dorsiflexion in the ankle, have been observed in several groups of patients, including those with unilateral CP [60]. Having an anatomically shorter limb on the involved side might be an advantage, since limited dorsi flexion of the ankle in unilateral CP may cause a foot clearance problem in swing phase. In this situation the limb can be considered functionally optimal. However, a patient with more pronounced spasticity may experience increased plantar flexion and early heel rise during stance phase such that even an anatomically short limb may be functionally too long [35, 36].

It is difficult to separate the causes of deviations and to determine if deviations in movement pattern are primary or secondary (compensation mechanisms) in unilateral CP. The cause might be anatomical changes (including LLD), impaired motor control and compensation mechanisms. If treatment of LLD is being considered, it is important to know the degree and location of inequality.

### 1.5 WELL-BEING

The interest in assessing and measuring quality of life (QoL) has increased and new instruments have been developed. In clinical outcome studies after intervention and in observational studies one is more or less expected to include results on QoL together with other measurements of specific treatment or physiological effects. There are general (generic) and disease specific instruments. Generic instruments allow comparisons between different groups of patients, which can be of value in making policy decisions and for economical considerations. The disease specific instruments are useful to follow patient outcome over time and also to make comparisons with other treating units/regions.

It can be difficult to find age validated instruments for use on teenagers and young adults. This presents a problem since the transition from childhood into the teens and eventually into adulthood is a period of interest, involving many physiological changes and the development of personality and self-esteem [106].

Self-esteem defines how a person values him/herself and it plays a strong role for psychological and social health [107-109]. I think I am (ITIA) is a self-reported scale that has been developed in Sweden and has been used in clinical settings as well as in research measuring self-esteem [110]. The scale includes descriptions of a person's conception of himself/herself.

The sense of coherence (SOC) scale, implemented by Antvosky 1979, has been widely used among researchers in health and caring sciences, and has been translated to several languages [111, 112]. Antonovsky advocates a salutogenic view as opposed to a pathogenic view when health and disease are being discussed. A person's SOC is

## Introduction

considered to be a major determinant of maintaining his or her position on the health – ease - disease continuum and movement towards the healthy end. SOC is a concept frequently used to describe how a person deals with changes and crisis in life [111]. A high SOC usually goes hand in hand with a better quality of life and adaptation to disease and illness. According to Honkinen and co-workers a strong sense of coherence can be regarded as a readiness to mobilize personal resources and is assumed to compensate for a lack of resources [113].

The health related quality of life instrument EQ-5D is a generic standardized and validated instrument to describe and measure health status [114, 115]. EQ-5D was developed in Europe and is now used world wide as an instrument to measure health related quality of life. It can be used on an individual basis but also in a population, comparing patient groups for decision making and priorities in health care.

There are studies correlating gross motor function with QoL of patients with CP, however a more detailed assessment of motor function including movement pattern is rare [116, 117]. Pirpiris and co-workers report that the psychosocial well-being of children with mild CP was more strongly affected than that of children with more severe functional disability [118]. In addition Russo et al report that expectations on high functioning patients to perform at peer level in all aspects of functioning may leave young patients vulnerable to adverse consequences such as poor self-concept and low self-esteem [119].

It is not a trivial matter always to be last on sports day, or need help cutting up your food or be unable to carry your tray in the school canteen [120]. These sorts of things can make children and young adults feel uncomfortable, conspicuous, and make life at and outside school stressful and unrewarding. In addition many children have relatively “invisible” psychological problems, as discussed before/above [120]. Is there any teenager or young adult who would not prefer to move with a normal gait? In the outpatient clinic we have difficulties assessing and discussing these modalities. In addition it is not easy for teenagers and young adults to bring up new needs that have developed with age and maturity and to express new treatment goals.

## 1.6 TREATMENT

### 1.6.1 Treatment to improve gait

The treatment of children with CP generally involves early identification, with therapy and surgical intervention occurring during the growth years [121-124]. Different treatment modalities influence the ultimate ambulatory ability and efficiency, which sets the outcome for the individual’s final mobility and independence [36, 40, 41]. Treatment is seldom considered for high functional individuals with unilateral CP, partly because the impairment is mild and partly because effective treatment options are limited [94].

Although these may be reasonable explanations for the lack of treatment, the question of possible treatment arises. “Many children (and their families) or young adults wonder whether something can be done to improve their ambulatory function and/or normalize their gait pattern” (Novacheck 2000) [125].

## Introduction

A wide variety of treatment approaches and modalities are possible. Here we discuss the most common orthopedic treatment options for children and teenagers with unilateral CP.

In the young child, physical therapy and orthoses (static and dynamic) are used to maintain full range of motion by stretching muscles and joints to stimulate muscle growth and prevent deformities [36, 125]. Generally the physiotherapist supports the parents in the management of their child's improvement of motor skills. Muscle strengthening is also part of the physiotherapist's responsibility, usually introduced at a later stage [36, 125]. Botulinum toxin can be used in a specific problematic muscle, often the calf muscle in unilateral CP, to allow further maturation and delay surgical treatment until the child is older [126].

Novacheck states: "Generally surgical intervention is delayed until functional improvement has reached a plateau or gait pattern has matured. This is usually at age 5 years or later. The healing and recovery are faster and easier before the adolescent growth spurt. Therefore, if surgery is to be done, the optimal age is between 5 and 12 years" [125]. Another reason to wait is that it may become clear whether additional surgery might be beneficial and the procedures can be performed simultaneously [127]. The most common surgical procedure is the lengthening of the calf muscle complex, either through an Achilles tendon lengthening or a gastrocnemius recession. About half of the patients with unilateral CP will require a lengthening at 5 -8 years of age. Of these around a third will need another lengthening in adolescence [36].

In patients with Winters' classification type 1, the primary treatment is flexible leaf-spring orthosis. Another strategy is a transfer of the tibialis posterior through the interosseous membrane to the dorsum of the foot. This is rarely performed in children with CP but is more relevant in adults with type 1 pattern [128].

In type 2, early treatment includes first solid ankle-foot orthoses and then articulated orthoses. If the child has marked spasticity in the gastrocnemius-soleus complex botulinum toxin can be used a few times to make the orthoses fit. At age 4 to 7 years, a surgical lengthening of the gastrocnemius-soleus is often needed and this is done by lengthening either the Achilles tendon or gastrocnemius. A walking cast is applied below the knee for 4-5 weeks, after which usually no orthoses is needed. Foot deformity with addition of varus to the equinus may develop and warrant treatment. The cause may be an overactive tibialis posterior muscle and/or tibialis anterior muscle. Identifying the deforming forces and choosing appropriate surgical treatment may be a challenge. The physical examination, kinematics and EMG data are helpful.

Rotational deformities are not common in type 2. See below under type 4.

In type 3 the young child can benefit from botulinum toxin injection in the hamstring and gastrocnemius-soleus complex. Later when the hamstring contracture increases surgical lengthening of both muscles is often needed. Stiff knee gait can be rectified by transfer of the rectus femoris to the flexor side, for example to the sartorius muscle. Rotational deformities are common in type 3 and may motivate treatment. See below under type 4.

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Type 4 patients have the same need of surgery as those with type 2 and 3, along with additional procedures including the hip joint. In the sagittal plane, iliopsoas lengthening is sometimes needed, together with increased pelvic tilt so as to avoid excessive flexion in the hip. Adductor lengthening can sometimes be indicated.

Type 4 patients more often have rotational deformities.

As mentioned above, rotational deformities are uncommon in type 2 and need not necessarily be addressed before adolescence. In type 3 and 4 the deformities may cause apparent gait problems and can therefore need treatment earlier. Rotational osteotomy of the femur is most common and sometimes increased external tibial torsion needs to be addressed. Especially if an increased internal rotation/anteversion of the femur is corrected with an external rotational osteotomy, the tibial torsion becomes evident and needs to be addressed at the same time. In type 4 a slight varus and extension osteotomy together with the rotational osteotomy may be indicated [36, 125].

Limb length equalization is sometimes considered and may include shoe lift, epiphysiodes, shortening/lengthening of the femur or tibia [104, 129].

### **1.6.2 Treatment of the upper limb**

Abnormalities in the arm (e.g., elbow and wrist flexion, thumb-in-hand deformity, and forearm pronation) can be treated according to the same principles as in the lower limb. The treatment protocols differ depending on the treating physician. Night splints are commonly used, as is botulinum toxin. Occasionally muscle/tendon release surgery is performed on flexors, pronator muscles and adductor of the thumb at an early age. Tendon transfers around the wrist are frequently performed, with pronator release or rerouting. However, this is usually performed at a later stage, when the patient is more capable of participating in the postoperative regime including splints and occupational therapy [130, 131].



## **2 AIMS**

The overall aim was to study movement pattern in children and young adults with mild spastic unilateral cerebral palsy. The emphasis was on the dynamic muscle power generation during walking in relation to the static anatomical muscle volume and leg length measurements from both the involved and non-involved side. Additionally well-being in these high functioning individuals with unilateral cerebral palsy was explored.

Specific aims:

- I. To investigate how children with spastic unilateral cerebral palsy can be classified using Winters' criteria with sagittal kinematic data from three-dimensional gait analysis and to investigate if patients move between classifications over time and/or with surgical intervention.
  
- II. To investigate hip and ankle power generation on both the involved and non-involved side to assess asymmetries during walking in children with spastic unilateral cerebral palsy.
  
- III. To describe the degree and location of leg length discrepancy in teenagers and young adults with spastic unilateral cerebral palsy.
  
- IV. To measure the muscle volume differences between the involved and non-involved side and to study the correlation of power generation during walking with muscle volume differences in teenagers and young adults with spastic unilateral cerebral palsy.
  
- V. To explore the relationship between movement pattern in the upper and lower extremity during walking and self reported self-esteem, sense of coherence and quality of life in teenagers and young adults with mild spastic unilateral cerebral palsy.



### 3 MATERIAL AND METHODS

#### 3.1 STUDY OUTLINES AND RECRUITMENT

Study I and II were retrospective cross-sectional studies. From the medical records and the database, patients with spastic unilateral CP who had visited the Gait laboratory at AI DuPont Hospital for Children, Wilmington, Delaware, USA, with a complete GA between 1994 and 2006 were identified. In most cases of the 284 patients, GA was done as a preoperative assessment for surgical planning, or as part of a postoperative follow-up. The most common reason for exclusion in the first classification part of study I was prior lower extremity surgery. In the second part of study I, involving comparison over time of groups with vs without surgery, only patients with two GA were included. The main reason for exclusion in study II, in which power generation was calculated, was previous surgery (Figure 8).

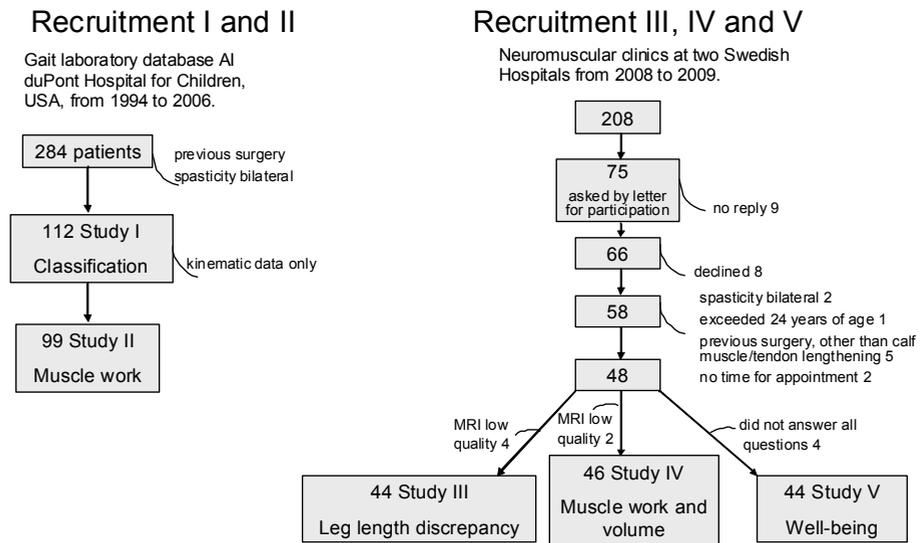


Figure 8. Recruitment of participants and reason for exclusion for study I and II to the left and study III, IV and V to the right.

## Material and Methods

In the prospective cross-sectional studies III-V, patients were recruited from the neuromuscular clinics of Astrid Lindgren Children's Hospital, in Stockholm and Skaraborg Hospital, in Skövde, Sweden between 2008 and 2009. In all, 208 patients were identified with unilateral CP. Inclusion criteria were 1) unilateral cerebral palsy diagnosed before the age of 2 years, 2) current age 13 to 24 years, 3) classification I or II according to the Gross Motor Function Classification Scale (GMFCS), 4) no previous lower extremity surgery other than calf muscle lengthening, 5) ability to cooperate during gait analysis (1.5 hours) and 6) ability to answer questionnaires. Exclusion criteria were any disease or previous injury affecting gait, and surgery or spasticity treatment (botulinum toxin) within the last year (Figure 8).

A letter of information and an invitation were sent out by mail to 75 patients who fulfilled the inclusion and exclusion criteria as far as could be determined from the medical records. Of these 75 patients, 66 replied. Of these, eight declined participation. Of the remaining 58 patients, 50 were eligible based on inclusion criteria; two had some spasticity on the contralateral side and would be classified as bilateral CP, one patient exceeded the age of 24, two had undergone soft tissue surgery other than calf muscle lengthening and three patients had had bone surgery. Further, two patients were lost because we were not able to find a suitable time for an appointment.

Those that agreed to participate visited the Gait Laboratory at Astrid Lindgren Children's Hospital once. A magnetic resonance imaging examination (MRI) was performed and the participants filled out the questionnaires at the same visit (Figure 8).

### 3.2 PARTICIPANTS

	Study I	II	III	IV	V
	Classification	Muscle work	Leg length discrepancy	Muscle work and volume	Well-being
	American cohort		Swedish cohort		
Number	112	99	44	46	44
Age	8.1 years (3.6-19.8)	8.4 years (4.0-19.8)	17.6 years* (13.0-23.0)	17.6 years (13.0-24.0)	17.6 years (13.0-24.0)
Females/Males	44/68	39/60	20/24	22/24	22/22
Method	GA	GA	MRI	GA/ MRI**	GA/ Questionnaires

Table 1. Outline of the studies regarding age, number of participants, and outcome measurements. GA=Gait Analysis. MRI=Magnetic resonance imaging. Study I and II including one cohort and study III, IV and V another cohort. (In study III \*median age is calculated. In study IV \*\* only 30 of the 46 patients had muscle volume measurements.)

In study I, 112 patients with spastic unilateral CP, from a total of 284 patients in the gait laboratory database, were identified and met the inclusion criteria. In study II, 99 patients with spastic unilateral cerebral palsy out of 284 patients, from the same gait

laboratory database as in study I, were identified and met the inclusion criteria. Study III included 44 patients.

In study IV, 46 patients were included and 30 patients had both MRI and GA performed. The remaining 16 subjects also had MRI and GA performed but the quality of the MRI image was poor and therefore muscle volumes could not be measured accurately. In study V, four patients did not answer all the items in the questionnaires and were therefore excluded. Thus, study V included 44 patients (Table 1).

Study IV and V also included 15 age and gender matched controls recruited from among researchers' friends and family, with a median age of 17.4 years (range 13.1-22.0 years).

In study I, II and III all participants were fully independent unassisted community ambulators primarily graded as GMFCS level 1. In study IV and V all were classified as GMFCS level 1, except two who were classified as GMFCS level 2.

Regarding the FMS assessed in study III-V, all patients but two had the highest possible score in all three distances. Those two were the same patients classified as GMFCS level 2, and they attained the second highest score, five, on both 50 and 500 meters in the FMS. Ten of the patients had attended or were currently attending special classes or schools because of cognitive limitations.

### **3.3 THREE-DIMENSIONAL GAIT ANALYSIS**

Three-dimensional gait analysis (GA) provides an objective quantitative dynamic measurement of gait, and is one of the main outcome tools in this thesis [35, 36].

In study I and II the gait analysis was performed with a Motion Analysis video capture system and all the data were reduced using Orthotrak with the Cleveland Clinic marker set (Motion Analysis Corp®; Santa Rosa, California). Multiple gait cycles were collected, typically between 10 and 25 cycles, and were time dilated, and a mean cycle with standard deviation was calculated. The patient walked at a self-selected speed. The Cleveland Clinic marker system was routinely used to collect the standard lower extremity joint kinematics and kinetics, on the hemiplegic and non-involved side. The kinetic data were collected using two forceplates (Advanced Mechanical Technology Inc. AMTI®, Watertown, MA). Generally three trials from each foot were collected. The kinetic and kinematic data were collected from the same trials. The ground reaction force vectors were collected and used together with kinematic data to calculate the moments and power generation/absorption. Normal reference values for different age groups were previously set in the laboratory.

In study IV and V, gait data were recorded at 100 Hz with an 8-camera three-dimensional motion capture system (Vicon Motion Systems®, Oxford, England). Retroreflective markers were placed on specific anatomical locations in accordance with the Vicon Plug-in-Gait Model. Multiple gait cycles were collected and a mean cycle with standard deviation was calculated. The patient walks at a self-selected speed on a 10-meter walkway. Two forceplates (9281CA, Kistler®) collect the ground reaction force vectors. Generally three trials from each foot were collected. In addition to the common lower extremity, the thorax, upper and lower arms, hands and the head were

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included. Patients walked at a self selected speed across the capture volume, which was 10 meters long. Variables of interest in the lower limb in these studies were; in the pelvis and hip flexion/extension, abduction/adduction and internal/external rotation. In the knee and ankle flexion/extension and finally foot progression was also noted. In the upper limb variables included shoulder flexion/extension, shoulder abduction/adduction, elbow flexion/extension, and wrist flexion/extension. Shoulder and elbow calculations were performed using Orthotrak, the clinical gait measurement and evaluation tool of Motion Analysis Corporation (Santa Rosa®, CA). Wrist flexion/extension is not included in Orthotrak, thus calculations were conducted using a custom LabView program. A set of Euler rotations were performed between the hand segment, consisting of the two wrist markers and a marker placed on the dorsum of the hand just proximal to the metacarpals, and the forearm segment, consisting of the wrist markers and the elbow marker. Time and distance variables were registered. The age and gender matched control group were used as normal references. Kinematics in upper and lower limbs was analyzed as was lower limb kinetics. Concentric muscle work/power generation in the sagittal plane was calculated.

### 3.3.1 Winters' classification

Winters' classification based on sagittal kinematics from the ankle, knee and hip was used to classify patients in all of the studies included in the thesis [52]. In study I the applicability of the classification was the main interest. Although Winters used EMG as a complement we did not in the classification.

The classification defines four gait patterns, where type 1 is the least involved and type 4 the most involved. The criteria are:

Type 1. Plantar flexion through swing phase.

Type 2. Plantar flexion through stance and swing phase.

Type 3. Ankle involvement as type 2 and knee involvement with knee range of motion less than 45 degrees in the gait cycle and less than 50 degrees flexion in swing phase.

Type 4. Ankle and knee involvement as above and hip involvement with hip range of motion less than 35 degrees.

### 3.3.2 Gait profile score and arm posture score

From the movement data (kinematics) scores of deviation from normal of the upper and lower extremity during walking were calculated and the amount of deviation quantified. The Gait Profile Score (GPS; Figure 9) is a measurement of gait deviation (lower extremity) developed by Baker et al [77]. It consists of nine different kinematic curves; curves in three dimensions from the pelvis and hip, curves in the sagittal plane from the knee and ankle and finally the foot progression curve.

In the upper extremity the corresponding measurement for arm movement deviation, the Arm Posturing Score (APS; Figure 9), is calculated using the kinematic data on shoulder flexion, shoulder abduction, elbow flexion and wrist flexion [58].

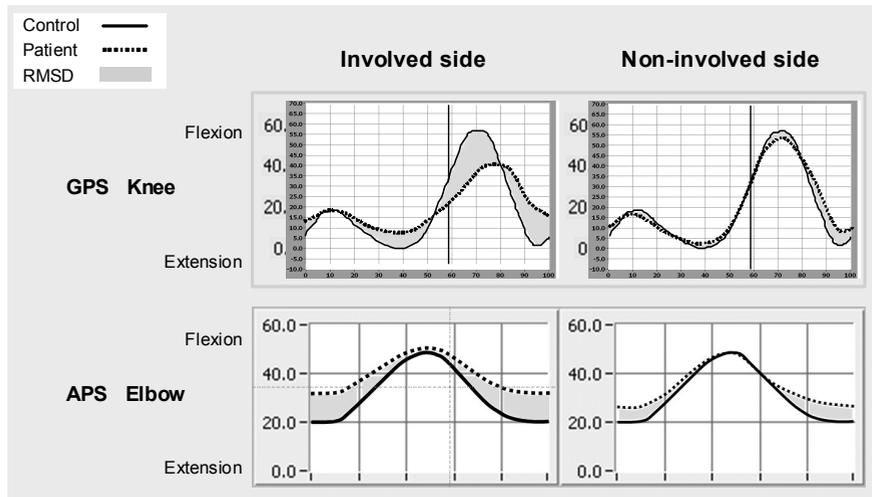


Figure 9. GPS and APS. The deviation is illustrated on the involved and non-involved side with the RMSD, i.e., the mean amount of deviation from the control group is calculated in each time frame of the gait cycle for all trials included. Here we illustrate the knee and elbow joint in the sagittal plane. Figure from own not published data.

Each variable's deviation was calculated as the root mean square deviation (RMSD) from normal. In each of the 100 frames a gait cycle consist of, the degree of deviation from the normal curve was calculated. The means in each frame for several trials were then calculated and all the values from each of the 100 frames were added to provide one value for deviation in that specific dimension in that specific joint. By using the root of the square, the negative values were converted into positive values, thus providing a true value of deviation regardless of direction. Finally the deviations from several joint and dimensions were summed, nine for the GPS and four for the APS. For both the GPS and the APS a higher score means a greater deviation from normal. The measurement of the amount of deviation is in the unit degrees.

### 3.3.3 Concentric muscle work / power generation

The definition of the terms forces, power and work is often unclear and the words are sometimes used interchangeably. The main interest in these studies is on concentric muscle work. Muscles contract either with shortening (concentric), keep the same length at contraction (isometric) or contract with lengthening (eccentric). The concentric muscle contraction produces work (positive work). Concentric muscle contraction is responsible for propulsion when walking and occurs mainly in the sagittal plane in the hip, knee and ankle. Eccentric and isometric muscle contraction is responsible for absorption of forces controlling range of motion and stabilizing joints. For an object with a certain mass to move, a force needs to be applied. A force applied over a distance is called work (measured in the unit Joules). Concentric muscle work is the same as the sum of power generation over time. The kinematic data provide the

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angular velocity in a joint and the kinetic data provide the moment. Power generation is the product of angular velocity and joint moment.

In three dimensional gait analysis (GA) the power in a joint is divided into three different anatomical planes. Concentric muscle work (positive work) is calculated as the cumulative sum of power generation in each plane. Work is the time integral of power and thus the area above the zero line represents the amount of concentric muscle work performed. The area below the zero line represents the power absorption, mainly controlled by eccentric muscle contraction (Figure 10).

In the hip joint the hip extensors (mainly gluteus maximus) produce work in early stance phase and hip flexors (medial hamstrings, the adductors and the iliopsoas muscles and to some extent the gluteus medius muscle) at the end of stance and beginning of swing phase (Figure 10A). The knee extensors (quadriceps muscle) generate work during several parts of stance phase (Figure 10B). Ankle plantar flexors (gastrocnemius and soleus muscle and the smaller flexors including tibialis posterior and toe flexors muscles) produce work in late stance phase (Figure 10C).

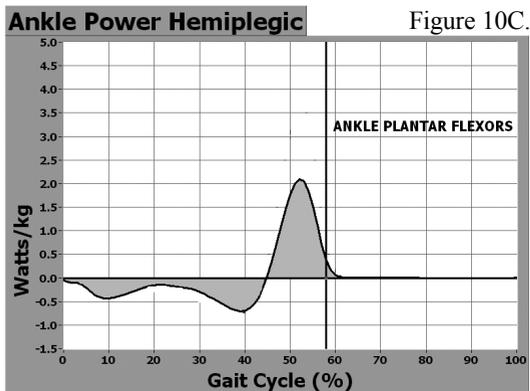
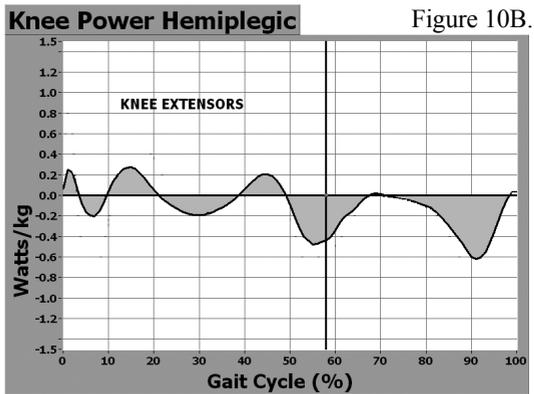
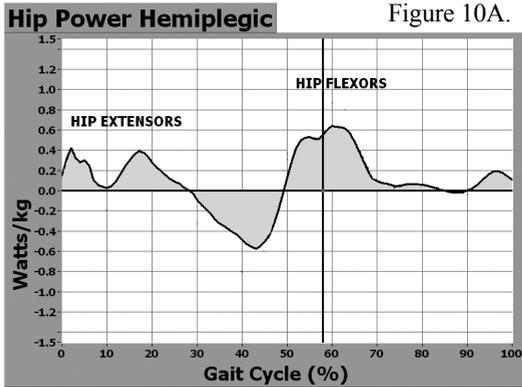


Figure 10 A-C. The concentric muscle work is calculated as the area under the curve, above the zero line. The area under the zero line is the power absorption. The illustration is from the involved side in an individual with unilateral cerebral palsy.

### 3.3.4 Physical examination

All participants were examined by an experienced physiotherapist. Assessment included passive range of motion of the upper and lower extremity using standardized positions and using a goniometer. Spasticity was assessed in elbow flexors and extensors and in the rectus, hamstrings, gastrocnemius and tibialis posterior muscles, according to the modified Ashworth scale. Manual test of muscle strength and motor control was done in the lower extremity. Measurements of weight and height were obtained.

## 3.4 MAGNETIC RESONANCE IMAGING

The MRI system used in study III and IV was a Philips Achieva 1.5 T (Philips Medical Systems®, Best, The Netherlands) located at Astrid Lindgren Hospital for Children, Stockholm, Sweden. Magnetic resonance imaging (MRI) is non-invasive, generates no radiation and can be used to visualize both soft tissue and bone structure.

### 3.4.1 Muscle volume measurements

The patient was examined supine with both lower limbs fully extended and parallel to the long axis of the body. In order to measure muscle volume, consecutive axial images, T2 weighted, were acquired from the highest insertion of the psoas muscle (mid thorax) to the sole of the feet. The slice thickness was 5 mm with 10 mm gaps between slices. The volume of the iliopsoas, the individual muscles of the hip and thigh and the dorsi and plantar flexors of the ankle on the shank were analyzed on a computer using custom software developed with Interactive Data Language (IDL; Research Systems®, Inc, Boulder, CO). The software separates muscle from other tissues, such as bone, adipose and skin [132]. See Figure 11.

Briefly, the individual muscles in each raw image were traced over the muscle boundary and labelled appropriately. The images were filtered and image segmentation was performed with a fuzzy clustering algorithm [133]. The cross-sectional area was determined for each muscle in each image. The volume of each muscle was determined by multiplying the cross-sectional areas by 1.5 to account for the thickness of the image (0.5 cm) and the space between images (1.0 cm).



Figure 11. Muscle outline.

### 3.4.2 Leg length measurements

The patient was examined supine with both lower limbs extended and parallel to the long axis of the body. Sagittal T1 weighted gradient echo images of the lower extremity were obtained, covering an area that included both the iliac crest and the sole of the foot. The slice thickness was 10 mm with 1 mm gaps between slices. To cover the whole length of the lower extremity three or four stacks of sagittal images were acquired. These were automatically fused after acquisition using MobiView software (Philips Medical Systems®, Best, NL) to display the entire lower limb in one image. Using specific anatomical landmarks on the sagittal view, length was calculated separately for the pelvis, femur, tibia, talus and the calcaneus (Figure 12).

All measurements were performed independently by two experienced observers trained in interpretation of musculoskeletal MRI. To minimize inconsistencies and to standardize the method the two observers agreed on common reference points for the measurements.

The pelvic height, the femur and tibial length and the talus and calcaneus height were assessed on the right and left side for all subjects.

To assess the reliability of the measurements we examined reproducibility (intra-observer comparison) and degree of agreement (inter-observer comparison).

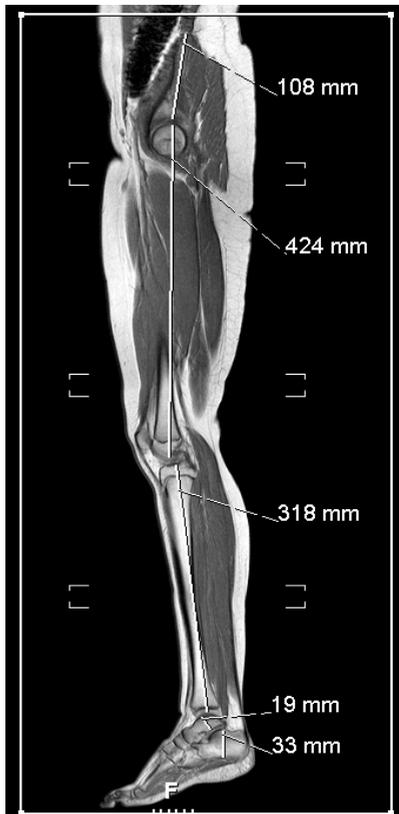


Figure 12. Leg length measurement with the height of the pelvis, length of femur and tibia and height of talus and calcaneus. Study III. With permission JPO.

### **3.5 WELL-BEING QUESTIONNAIRES**

In study V self report questionnaires were given to the participants to respond to at the time of the GA. Parents and friends of the participant were not present in the room at the time. The participants sat alone unless the physiotherapist who had just performed the GA thought the individual needed support. If that was the case the physiotherapist would sit beside the participant and help by reading the questions. If more support was needed the questionnaire would not be completed. The questionnaire for assessing self-esteem, the “I think I am” takes the longest to fill in and was for that reason the last the participant was asked to answer. In that way the physiotherapist could leave it out if she found that the two previous short questionnaires were difficult for the participant.

#### **3.5.1 Self esteem**

The self-esteem questionnaire I think I am (ITIA) consists of five subscales, Physical characteristics (14 items, example “I have a nice face”), Talents and skills (14 items, example “Other people do things better than I do”), Psychological well-being (16 items, example “I can easily make up my mind”), Relations with family and Relation with others (14 items each, example “My family would always help me” and “I feel that I am different from others” respectively). The response alternatives range from “Just like me” (+2 points) to “Not at all like me” (-2 points). The total score ranges from minus 144 to plus 144. A higher score reflects higher self-esteem. ITIA was originally developed from items included in internationally validated and well established instruments of self-esteem and self-image. The reliability and validity of ITIA has been shown to be acceptable and in the present study Cronbach’s alpha was 0.92 for the total score and between 0.73 and 0.89 for the subscales.

#### **3.5.2 Sense of coherence**

The sense of coherence (SOC) scale includes the subject’s perceptions of three components: comprehensibility, manageability and meaningfulness. The SOC instrument has been used on subjects from 13 years of age to adulthood; it has been shown to have good reliability and validity and to be cross-culturally applicable [112]. The SOC scale includes 13 items each having a 7-point response scale from “strongly agree” to “strongly disagree”. Example of a question: “Do you have the feeling that you’re being treated unfairly?” Total score ranges from minimum 13 to maximum 91. A higher score means a higher SOC. In this study Cronbach’s alpha was 0.80.

#### **3.5.3 Quality of life**

In the first part of the health related quality of life instrument EQ-5D, the individual classifies his/her health in five dimensions (Physical movement, Hygiene, Main activities, Pain, and Anxiety and depression [114]. A three graded scale is used. For example the participant chooses between “I have no problems looking after myself”, “I have some problems looking after myself” and “I have a lot of problems looking after myself”.

The second part of the EQ-5D is a Visual Assessment Scale (VAS) labeled from “best imaginable health state” (endpoint 100) to “worst imaginable health state” (endpoint zero) where the participant makes a mark along the scale to indicate his/her health state

at present. In the third part, which is the EQ-5D index, the participant is assigns an index from a weighted reference/tariff. This part of the index was not used in this study.

### **3.6 STATISTICAL METHODS**

SPSS version 12, 15 and 18 was used for all data analyses (SPSS, Chicago, Illinois, USA). In study I-III independent samples t-test and paired t-test were used to compare groups. Data were normally distributed. In study II all comparisons between groups were in consecutive/severity order, which means that type 0 was compared to type 1, type 1 was compared to type 2, type 2 to type 3 and type 3 to type 4. In study III the intra- and inter-observer comparisons were performed using Intra class correlations (ICC). In study IV descriptive statistics were presented as mean and standard deviations and ratio of the involved and non-involved side for numerical variables, or as percentages for categorical variables. A paired t-test was used to test the null hypothesis that there was no difference in mean between the sides. The Wilcoxon signed rank test was applied when distribution deviated from normality. To measure the strength of linear association between the ratio of muscle work and muscle volume, the Pearson correlation coefficient was calculated. Multiple linear regression analysis was employed to estimate the relationship between the ratio of muscle work and muscle volume of the involved and non-involved sides while controlling for other demographic and clinical variables. The standard or simultaneous method was used and variables with known clinical importance were included in the model.

In study V independent t-test and Pearson correlation coefficient were used. For internal consistency of the questionnaires, Cronbach's alpha was calculated. A Cronbach's alpha coefficient equal to or greater than 0.70 was considered satisfactory. The level of significance was specified at 0.05.



## 4 RESULTS

### 4.1 CLASSIFICATION (I)

#### 4.1.1 Winters classification at baseline

In the first part of study I the gait pattern of 112 children with spastic unilateral CP without previous surgery, mean age 8.1 years (range 3.6-19.8 years) was classified. We used the Winters classification system based on sagittal kinematics from the hip, knee and ankle on the involved side. Twenty-six (23%) of the 112 could not be classified. Their deviation was detectable, but too small to classify as type 1. This group is named type 0 (Figure 13).

Type	N (%)	Hip Rom	Knee Rom	Ankle variables
0	26 (23)			↕
1	31 (28)			↕↕
2	22 (20)	↕	↕	↕
3	19 (17)	↕	↕	
4	14 (12)	↕		

Figure 13. Distribution of the different Winters' classification types with the added type zero (0). The number and percent of patients. The variables hip range of motion, knee range of motion and six different ankle variables (see table 2). Arrow ↕, indicates where differences were found comparing types of unilateral CP.

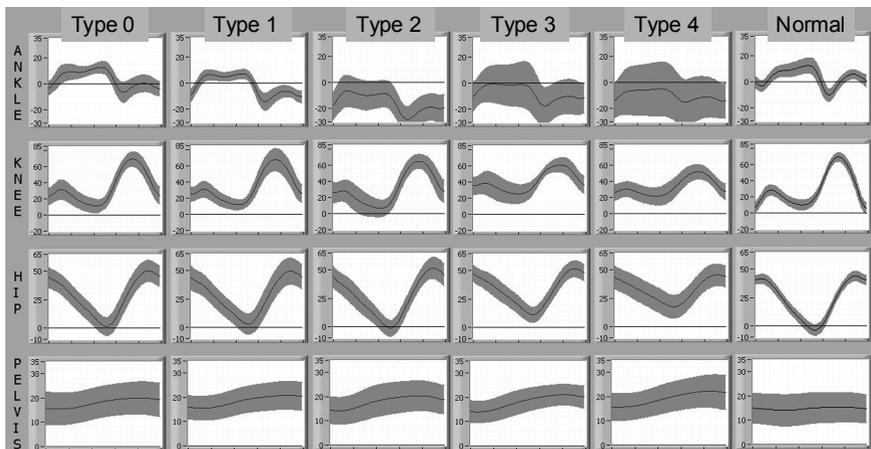


Figure 14. The different Winters classifications, including “type 0”, for the 112 participants. Mean, black line; standard deviation, gray band. Kinematic sagittal data from the pelvis, hip, knee and ankle. With permission JPO.

## Results

In line with the classification criteria we found significant differences in lower limb range of motion (Rom) in the type 4 group compared to the type 3 group ( $p<0.001$ ). Knee Rom was significantly lower in the type 3 group compared with type 2 group ( $p<0.001$ ) (Figure 14).

There were several significant differences between ankle variables between the type 0 group and type 1 group and also between the type 1 group and the type 2 group. (Table 2 and figure 14).

Ankle						
Type	Initial contact	Max plantar flexion 0%-30%	Max dorsiflexion 0%-30%	Max dorsiflexion 30%-60%	Max plantar flexion	Max dorsiflexion in swing
0 (n=26)	-3.0	-3.5	10.5	13.5	-7.9	5.4
1 (n=31)	-9.0	-9.0	7.2	7.4	-15.8	-4.0
2 (n=22)	-17.8	-18.2	-6.1	-8.7	-29.8	-16.8
3 (n=19)	-12.5	-13.2	-1.8	-2.9	-20.9	-9.8
4 (n=14)	-11.0	-11.3	-0.9	-0.1	-14.9	-5.5
Comparison						
0-1	<0.001	<0.001	0.009	<0.001	<0.001	<0.001
1-2	<0.001	0.001	<0.001	<0.001	<0.001	<0.001
0-2	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
2-3	-	-	-	-	-	-
3-4	-	-	-	-	-	-

Table 2. Ankle kinematics on the involved side in the sagittal plane for different Winters types. Comparison between groups (p value). With permission JPO.

### 4.1.2 Classification changes with time

In the second part of the classification study (study I) the purpose was to investigate if patients move between classification types over time or with surgical intervention. The re-classification (GA2) was performed after a mean of 3.1 years (1.2-7.0 years) for patients who had no operation and a mean of 3.0 years (1.2-6.9 years) for those who had surgery. Among the non-operated patients 47% (7/15) kept the same level and 53% (8/15) worsened over time. In the patients who had surgery 26% (8/31) kept the same level, 61% (19/31) improved and 13% (4/31) got worse (Table 3).

## Results

Patient category	No surgery	Surgery
Age GA 1 (years)	5.8 (3.6-9.3)	7.1 (4.0-13.2)
Time GA1-GA2 (years)	3.1 (1.2-7.0)	3.0 (1.2-6.9)
Total number of patients	15	31
Kept the same group	7	8
Moved up 1 group (worse)	3	2
Moved up 2 groups (worse)	1	1
Moved up 3 groups (worse)	4	1
Moved down 1 group (improved)		11
Moved down 2 groups (improved)		6
Moved down 3 groups (improved)		2

Table 3. Re-classified patients. Age, in years, at first gait analysis (GA 1), time between gait analysis 1 and 2, patient's classification type/group and the change into different groups over time for patients that had no surgery and for those that had surgery.

With permission JPO.

### 4.2 CONCENTRIC MUSCLE WORK / POWER GENERATION (II AND IV)

The calculations of concentric muscle work / power generation were done the same way in study II and IV even though the nomenclature is somewhat different. The scientifically relevant difference between the studies is that in study II the patients' mean age was 8.4 years (range 4.0-19.8 years) and in study IV 17.6 years (range 13.0-24.0 years). In study IV knee extensor work was calculated in addition to hip and ankle work, but not in study II. Furthermore, study IV included other variables such as muscle volume, which will be discussed later.

Both studies showed a significant shift in concentric muscle work from the ankle joint to the hip joint compared to controls, in study II on both sides and in study IV on the involved side. Concentric muscle work was lower on the involved side than on the non-involved side in both studies, although no statistical significance was reached in study II, where the patients had been classified according to the modified Winters classification. The difference in muscle work between the involved and non-involved side was significantly greater distally (in the ankle joint) than proximally (in the hip joint). In study IV knee extensor work in the involved limb was calculated and found to be significantly decreased compared to both the non-involved side and the control group ( $p < 0.001$ ; Table 4).

## Results

Mean Concentric muscle work (Joule/kg)			
	Cerebral palsy group n=46		Control group n=14
	Involved side	Non-involved side	average right and left side
Hip extensors	0.090 *	0.111 **	0.051
Hip flexors	0.150 *	0.209	0.191
Knee extensors	0.075 **	0.154	0.144
Ankle plantar flexors	0.211 **	0.350	0.347

Table 4. Concentric muscle work in the hip, knee and ankle on the involved side, the non-involved side and in the control group. Significant difference compared with the control group: \* ( $p<0.05$ ) and \*\* ( $p<0.001$ ).

Increased maximum plantar flexion moment was found in early stance in type 1 and 2 compared to type 0 in study II, ( $p=0.019$ ) and ( $p=0.001$ ) respectively. In the second half of stance the moment was significant lower in type 2 compared to type 1 on the involved side ( $p=0.034$ ). On the non-involved side it was increased ( $p=0.025$ ).

### 4.3 MUSCLE VOLUME (IV)

All muscles on the involved side, from the iliopsoas muscle to the plantar and dorsi flexors of the shank, had a significantly smaller volume than their counterparts on the non-involved side ( $p<0.001$ ) except the muscle gracilis ( $p=0.176$ ). The muscle volume difference was greater distally, with proximal muscles being more equal in size (Figure 15 and Table 5).

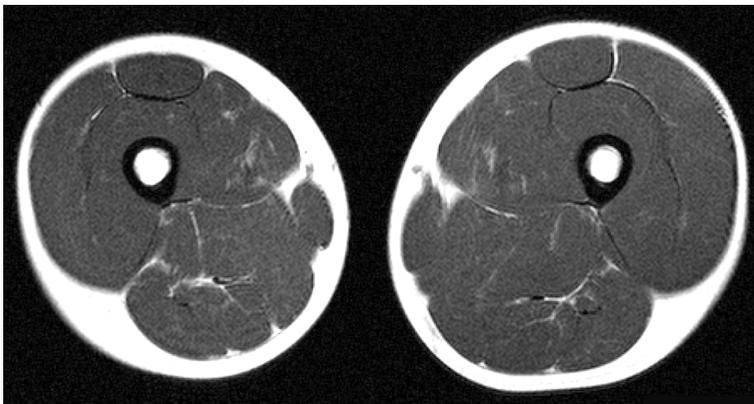


Figure 15. Cross sectional image of the mid thigh and shank to demonstrate muscle size difference. Involved side to the left and non-involved side to the right.

## Results

Muscle volume (cm <sup>3</sup> )      N=30				
	Involved side	Non-involved side	Percent of non-involved side	P value
	mean and standard deviations			
Dorsi flexors	151 (35)	212 (57)	71.2	<.001
Plantar flexors	782 (233)	1070 (299)	73.1	<.001
Rectus Femoris	194 (58)	246 (82)	78.9	<.001
Quadriceps	1306 (312)	1675 (470)	78.0	<.001
Biceps	208 (53)	255 (71)	81.6	<.001
Medial Hamstrings	492 (132)	581 (161)	84.7	<.001
Semimembranosus	156 (47)	182 (50)	85.7	.001
Semitendinosus	128 (46)	144 (48)	88.9	<.001
Sartorius	134 (41)	147 (49)	91.2	<.001
Gracilis	84 (28)	90 (23)	93.3	.176
Adductors	537 (163)	683 (207)	78.6	<.001
Adductor Longus	102 (40)	116 (44)	87.9	<.001
Gluteus Maximus	597 (168)	709 (200)	84.2	.021
Gluteus Medius	245 (61)	254 (64)	96.5	<.001
Iliopsoas	341 (92)	360 (99)	94.7	<.001

Table 5. Muscle volume measurements and differences assessed with Magnetic resonance imaging. Muscle volume difference in percent (involved/non-involved side) x100.

## Results

### 4.4 CONCENTRIC MUSCLE WORK AND MUSCLE VOLUME (IV)

Concentric work and volumes of muscles acting over the hip, knee and ankle joints were quantified in the sagittal plane. Muscle groups responsible for propulsion acting over each joint were examined on the involved and non-involved side (Table 6).

Concentric muscle work (Joule/kg) N=46				
Muscle group	Involved side	Non-involved side	Ratio	P value
Hip extensors: <i>Gluteus max</i>	0.09	0.11	0.811	0.028
Hip flexors: <i>Adductors, Medial hamstrings, Iliopsoas</i>	0.15	0.21	0.718	<0.001
Knee extensors: <i>Quadriceps</i>	0.08	0.15	0.487	<0.001
Ankle: <i>Plantar flexors</i>	0.21	0.35	0.603	<0.001

Muscle volume (cm <sup>3</sup> ) N=30				
Muscle group	Involved side	Non-involved side	Ratio	P value
Hip extensors: <i>Gluteus max</i>	597	709	0.842	<0.001
Hip flexors: <i>Adductors, Medial hamstrings, Iliopsoas</i>	1370	1624	0.844	<0.001
Knee extensors: <i>Quadriceps</i>	1306	1675	0.780	<0.001
Ankle: <i>Plantar flexors</i>	782	1070	0.731	<0.001

Table 6. Concentric muscle work during walking in the sagittal plane, and muscle volume. Measurements from the hip, knee and ankle on the involved and non-involved side. Ratios between the sides were calculated (involved/non-involved).

To examine the relationship between the difference in muscle volume and the difference in concentric muscle work, ratios were used. The ratios were calculated by dividing the value for involved side by that for the non-involved side (Table 7). Correlations between these ratios, muscle work and muscle volume, were calculated (Figure 14). Pearson's correlation coefficient for ratios in the ankle, knee and hip were: Ankle plantar flexor volume and work 0.403 ( $p < 0.05$ )  
 Knee extensor muscle volume and work 0.655 ( $p < 0.01$ )  
 Medial hamstring muscle volume ratio and hip flexor work ratio 0.481 ( $p < 0.05$ )  
 Adductor muscle volume ratio and hip flexor work ratio 0.578 ( $p < 0.05$ )  
 There was no significant relationship between iliopsoas muscle volume ratio and hip flexor work ratio or between gluteus maximus muscle volume ratio and hip extensor work ratio.

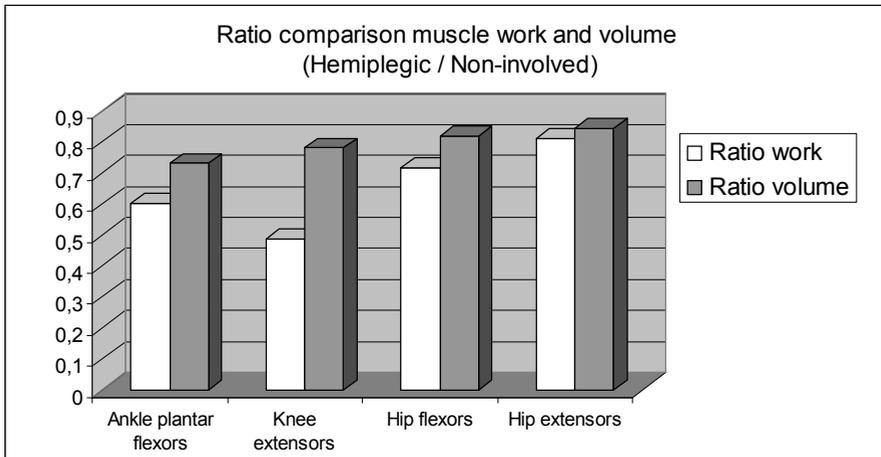


Figure 16. The ratio of the muscle volume and the ratio of concentric muscle work, (involved side divided by the non-involved side) reveal disparity between muscle groups at different joints.

#### 4.4.1 Influence of other variables

In a multiple linear regression analysis we explored the influence of muscle volume ratios at each joint level and other variables such as age, previous calf muscle surgery, Winters' classification, walking speed and tibia- and femur leg length discrepancy on each muscle group's work ratio.

Walking speed normalized to leg length was 1.36 m/sec (SD 0.28) and had no significant influence, neither had age, tibial or femoral length discrepancy.

The result indicated that ratio of plantar flexor muscle volume and previous calf muscle surgery explained 35% of ankle plantar flexor work ratio variance.

The ratio of quadriceps muscle volume explained 39% of the variance of knee extensor work ratio. Winters' classification and previous calf muscle surgery were significant predictors and explained 40% of the variance in hip extensor work ratio.

The medial hamstring muscle volume ratio explained 28% of variance of hip flexor work ratio.

#### 4.5 LEG LENGTH DISCREPANCY (III)

The mean total leg length discrepancy (LLD), when adding the calcaneus, talus, tibia, femur and pelvis was 9.15 mm (SD 10.8) with the involved side being shorter. The mean height of the calcaneus and the talus and the mean length of the tibia were significantly shorter on the involved side compared to the non-involved side (Table 7).

## Results

	Length hemiplegic side (mm)	Length Non-involved side (mm)	Relative difference (%)	Mean difference (mm)	95% Confidence Interval	P value
Calcaneus	40.5	41.4	97.8	0.9	(0.3 - 1.5)	0.003
Talus	29.6	30.9	95.8	1.3	(0.4 - 2.2)	0.006
Tibia	358.7	366.8	97.8	8.1	(6.1 - 10.1)	0.000
Femur	452.9	453.3	99.9	0.4	(-1.2 - 2.0)	0.624
Pelvis	114.8	114.2	100.5	-0.6	(-2.3 - 1.0)	0.409

Table 7. Mean length/height of the calcaneus, talus, tibia, femur and pelvis on the involved and non-involved side, relative difference in percent, mean absolute difference, confidence interval and p-values for differences in length. N=44.

With permission JPO.

In 7/44 (16%) of subjects the LLD was between 10-15 mm and in 14/44 (32%) of subjects the involved side was shorter with more than 15 mm. Six patients had a pronounced discrepancy of more than 20 mm. The distribution of the total LLD is illustrated in Figure 17.

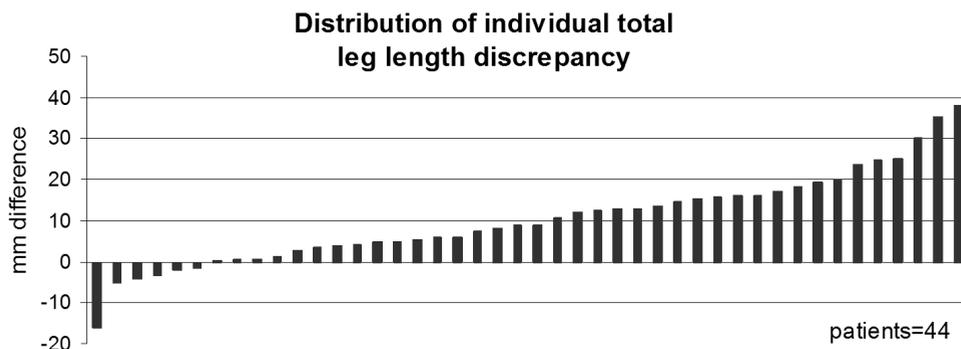


Figure 17. Distribution of the total leg length discrepancy for each patient individually including the calcaneus, talus, tibia, femur and pelvis. With permission JPO.

#### 4.6 SELF-ESTEEM, SENSE OF COHERENCE AND QUALITY OF LIFE (V)

Patients with mild unilateral CP showed no significant difference in sense of coherence (SOC) or quality of life (EQ-5D) compared to the control group, but there was a difference in terms of self-esteem (ITIA). Lower extremity movement deviation (GPS) and arm posturing deviation (APS) was significantly greater in the patient group compared to the controls (Table 8).

There was a significant correlation ( $r=0.719$ ,  $p<0.001$ ) between the SOC and ITIA scores. Thus, a strong SOC is correlated to high self-esteem.

APS correlated with SOC (coefficient  $-0.375$ ,  $p=0.05$ ), ITIA (coefficient  $-0.397$ ,  $p=0.001$ ) and EQ-5D VAS (coefficient  $-0.335$ ,  $p=0.05$ ). Thus, better arm movement (less deviation) was related to higher SOC, self-esteem (ITIA) and quality of life (EQ-5D VAS). There were no significant correlations between the SOC, ITIA and EQ-5D VAS scores and leg movement deviation measured as GPS.

	Patient group	Control group	95 % Confidence interval of the difference		Significance p value
	n=44	n=15	lower	upper	
	mean and SD				
Sense of coherence	62.8 (12.1)	64.6 (9.8)	-8.80	5.12	0.595
“I think I am”	63.4 (31.1)	84.7 (26.1)	-39.7	-2.8	0.025
<i>Physical index</i>	10.9 (7.6)	17.4 (6.2)	-10.6	-2.4	0.005
<i>Skills</i>	8.2 (8.5)	17.5 (4.9)	-13.0	-5.6	<0.001
<i>Psychological well-being</i>	13.4 (9.2)	16.4 (7.0)	-8.4	2.3	0.260
<i>Relation to family</i>	17.9 (8.3)	18.2 (10.7)	-6.8	6.3	0.925
<i>Relation to others</i>	13.0 (7.4)	15.1 (6.7)	-6.5	2.2	0.332
EQ-5D VAS	78.3 (17.0)	81.1 (13.7)	-6.9	12.4	0.573
Gait profile score	6.9 (2.1)	4.1 (1.2)	1.64	3.92	<0.001
Arm posture score	10.5 (5.0)	5.7 (2.1)	3.02	6.66	<0.001

Table 8. Patient and control group mean and standard deviations (SD), confidence interval for differences and p-values. Variables are Sense of coherence (SOC), I think I am (ITIA) with the five subscales, the EQ-5D Visual Assessment Scale (EQ-5D VAS), Gait profile score (GPS) and Arm posture score (APS). In SOC, ITIA and EQ-5D VAS a high score indicates good health. In GPS and APS a high score indicates more pronounced deviation from normal.



## 5 DISCUSSION

### 5.1 CLASSIFICATION (I)

Twenty three percent of the patients referred to the gait laboratory had a deviation too small to classify as type 1. We name this group, type 0.

In Winters classification, adults and patients with a diagnosis other than CP were included, which can explain why type 1, generally caused by a peripheral nerve injury, rarely is found in the CP population [52]. In our patients, type 1 pattern with no dorsiflexion in swing is probably due to spasticity in the calf muscle and an inability to selectively activate the counterbalancing tibialis anterior muscle.

The paper by Rodda et al suggests no spasticity treatment in the calf muscle of type 1 patients but recommends treatment for type 2 patients with stance phase equinus due to spasticity or contracture [54]. Again we are not convinced that patients with type 1 pattern, or type 0 pattern, necessarily lack spasticity entirely. With time these patients might develop more spasticity and/or a contracture that diminishes the ability of the already “weak” tibialis anterior to function. Additionally, results on kinetics from study II revealed that early plantar flexion moments increased from type 0 to type 1 and to type 2 pattern. Most likely in unilateral CP type 0, 1 and 2, patients experience different degrees of spasticity.

In the Winters study EMG was used, but it is difficult from the report to understand how [52]. In type 1 pattern, EMG should show little or no activity in the tibialis anterior muscle. Stebbins and co-workers found several different EMG patterns in the gastrocnemius and tibialis anterior muscles of patients with unilateral CP and proposed that EMG data might be a valuable addition for classification of unilateral CP in the future [134]. We did not find the EMG data to contribute in the classification and agree with Hullin et al that these data are difficult to interpret and quantify [45].

Our findings are in line with a recent population based study where McDowell and colleagues report that as many as 38 of 94 (42%) children, slightly older than our group, could not be classified according to the Winters criteria [52]. Obviously the distribution among Winters types will vary depending on the population studied.

The theory that a patient with spastic unilateral CP can move between Winters classification types with time is supported by our data, even though our sample is small. Patients move between groups, and those who do not receive surgical treatment mostly move into more severe groups as they age. To gain knowledge regarding the natural progression and to evaluate the degree of deterioration without surgical treatment, a multi-center study with a large group of patients would be required. By adding type 0 all patients can be classified and consequently also followed over time with reclassification providing information on the development in specific patients.

In addition to the sagittal kinematic data, Rodda 2001, Aminian 2003, and Dobson and Graham 2005 draw attention to the transverse plane changes with internal rotation of

the hip and retraction of the pelvis in Winters' classification type 4 [54, 127, 135]. After the primary deformity of the femur (the primary deviation) is corrected with rotational osteotomy, pelvic retraction (the secondary deviation) disappears [54, 127, 135]. Our results (data not shown) also revealed significant changes in the femur on the involved side compared to the non-involved side, with an increase of femoral torsion, in the Swedish cohort [136].

### **5.2 CONCENTRIC MUSCLE WORK / POWER GENERATION (II AND IV)**

In study II and IV we noted lower muscle work on the involved side compared to the non-involved side, with a more pronounced difference distally in the limbs. In addition a shift of work from the ankle joints to the hip joints was found.

Boyd et al and Zurcher et al report on improvement in power generation after botulinum toxin treatment of the spastic calf muscle in unilateral CP [137, 138]. A better positioning of the foot with less spasticity causing early heel rise most probably explains the improvement. Our results from study IV also indicate that previous Achilles tendon lengthening partly contributes to normalization of the ratio of concentric muscle contraction of the plantar flexions (ratio involved/non-involved side).

Olney and co-workers measured power generation/muscle work on the involved side only, in children with unilateral CP, age range 5.0 to 11.8 years old, and found that the ankle plantar flexors produced approximately a third of the concentric muscle work instead of the normal two thirds [101]. This is in line with our findings and in addition the same trend was noted on the non-involved side. A potential power generation from the non-involved ankle is not fully used and optimized. This might be because symmetry of movement is preferred or it may be a sign that the young patient with unilateral CP finds it difficult to simultaneously control the involved and non-involved side at different joint levels.

Hip extensor work in patients of all ages with unilateral CP is increased on both sides compared to matched controls. The fact that this persists at least up to young adulthood motivates further reflection and consideration of treatment possibilities. The increased hip extensor work by the proximal muscles is most probably a way to compensate for impaired motor control and function in the distal muscles that have more severe CP involvement.

Why do we not see a corresponding increase in iliopsoas muscle concentric contraction? Our perception is that the iliopsoas muscle should have equally good motor control as the gluteus maximus.

We speculate that compensation mechanisms do not necessarily occur merely to substitute for lost function, but preferably in a manner that will also reinforce symmetry. The iliopsoas muscle could possibly increase work on the involved side around toe off phase when the opposite non-involved stable foot is on the ground in end of loading response and in mid stance. However the opposite occurs when the non-involved side is preparing for swing. The difference is the situation with a much less stable and controllable foot on the involved side. At the time for iliopsoas concentric

muscle contraction on the non-involved side the instable foot of the involved limb is struggling to support the body, dealing with the ground reaction force, during and just after loading response. There is no room for compensation mechanisms at this stage and hence an asymmetrical recruitment would be the result.

The hip extensor muscles on the other hand can and are used for both propulsion and stability and are mobilized in a similar manner on both sides. This leaves the iliopsoas muscle useful for unilateral increase of hip flexion if needed to help clear the involved limb in swing phase.

Gage states that kinetic data contribute towards a better understanding and can help in preoperative decision-making [35]. In our study the early increased plantar flexor power generation and decreased moment in group 2 in study II, is interpreted as being explained by increased tone and plantar flexion in the younger patients. The non-involved side showed increased plantar flexion moment early in stance, interpreted as a way to create vaulting for clearance of the involved limb as a compensation mechanism.

### **5.3 MUSCLE VOLUME (IV)**

We found decreased muscle volumes on the involved side compared to the non-involved side with greater differences distally in the lower limb.

Our findings are in line with previous reports by Lampe et al and Elder et al [6, 83]. The degree of difference between the involved and non-involved side and the muscles examined was overall the same with the exception that we were not able to outline individual muscles in the shank since the quality of the MRI was not ideal. We used a semi automated segmentation method which needed a fair amount of manual muscle contouring. The manual contouring is the most common method, but it is time-consuming which limits its use [139].

Oberhofer et al report on normalized muscle volume and length (normalized to body mass and segmental length) in children with bilateral and unilateral CP compared to a control group [89]. Using MRI measurements they found a difference of 22% in the calf and 26 and 22% in the hamstrings and quadriceps muscle volume respectively, between children with CP and the control group. They conclude that other properties of muscle architecture than muscle volume could be of interest, such as muscle fiber length and pennation angles, to evaluate treatment [89].

Damiano and Moreau tested the maximal voluntary isometric contraction and found the correlation of muscle thickness significantly lower in CP patients compared to healthy controls [93]. Other variables than muscle volume seem to play a role [7, 80, 89].

### **5.4 CONCENTRIC MUSCLE WORK AND MUSCLE VOLUME (IV)**

There was a correlation between muscle volume and concentric muscle work during walking in these patients with unilateral CP.

## Discussion

Muscle strengthening programs in children with CP, have yielded some improvement in strength but not necessarily in gait function [88, 94, 95]. These published studies are few and vary greatly in terms of outcomes; thus it has been difficult to compare the effects of different muscle strengthening strategies. McNee et al could report on both increase in muscle volume and strength after strength training program, however gait did not improve [92].

Again we have to bear in mind that these patients have relatively good motor control and it is difficult to interpret the subtle influences on muscle function and the individual variability. However we discuss issues that could cause concern in a clinical situation when surgical muscle lengthening is being considered.

Spasticity in the adductor longus muscle (in the adductor muscle complex) can cause excessive adduction and flexion in the hip joint.

Spasticity in the semitendinosus muscle (the medial hamstring muscles) often reduces the patient's ability to extend the knee, which shortens the step. Restricted knee extension also has a negative impact on pre-positioning of the foot which is a prerequisite for normal gait and can be of special importance in the hemiplegic CP patient whose capacity for dorsiflexion in the ankle joint is often insufficient. There could be a concern that lengthening of either muscle in these two cases might decrease concentric muscle work. According to our reasoning, the adductor longus and the semitendinosus are relatively small compared to the total muscle volume of the adductors and the medial hamstrings, respectively. In addition the comparison to the non-involved side, both the volume ratio and the work ratio, renders essentially equal numbers. From this we conclude that lengthening of the adductor longus or the semitendinosus would most likely not cause major loss of strength in these situations.

Interestingly, previous surgical lengthening of the calf muscle increased the ratio of muscle work in the ankle plantar flexors. One interpretation is that the foot was better positioned in the surgically corrected patients and could generate more work, which partially normalizes the ratio. It could be considered a confirmation of one of the important prerequisites for efficient gait [35]. In other words, patients with unilateral CP are often in need of an Achilles tendon or calf muscle lengthening to be able to position the foot at initial contact; correct positioning makes concentric muscle work possible. More importantly it provides a plantigrade foot, which is essential for compensatory hip extensor work. This assumption is in line with the idea that power loss at the ankle after tendon Achilles lengthening is of minor concern in patients with unilateral CP [35, 137, 138].

When we classified the 99 patients in Study II according to the modified Winters' classification system, all groups had higher hip extensor work on both the involved and non-involved side compared to the age matched control group, including the type 0 patients, whose gait deviation was very mild. We could confirm these findings in the older patient group in study IV on the involved side.

In addition, muscles were smaller on the involved side throughout the limb, albeit with individual variability. In our study the ratio, the difference, between the involved and non-involved side showed greater differences (lower ratios) for concentric muscle work

than for muscle volume, which hints that other variables than muscle size matter in patients with CP, which is in line with earlier reports [6, 7, 80, 81, 89].

### **5.5 LEG LENGTH DISCREPANCY (III)**

There was a difference in lower limb length in this high functioning group of patients with spastic unilateral CP. The difference was significant in the calcaneus, talus and tibia.

GA can be used to discriminate between functional and structural LLD, which can help in decision-making regarding LLD treatment [62]. Kaufman and Walsh report that gait asymmetries in ground reaction force were observed at LLD of more than 20 mm [62, 64]. They state that the main reason for compensation by the long or the short limb, is energy conservation. It is not clear how this statement is supported although it is a reasonable assumption. However, their study included patients with various diagnoses and causes of LLD although patients with neuromuscular conditions were excluded. Therefore it is difficult to know if this applies to the high functioning patients with unilateral CP included in our studies.

Allen and co-workers examined kinematics of the involved and non-involved side in children with unilateral CP [60]. They report that patients with LLD have a similar compensatory movement pattern to those without LLD, but that it is more pronounced, with increased flexion on the non-involved side. The degree of involvement and/or the classification of these participants is not stated, and the leg length measurements were performed manually supine with a tape measure which makes it difficult to assess the reliability.

Saraph 2006 could show the same kinematic changes as Allen et al on the non-involved side, and also that kinematics normalized with surgical leg length equalization, in children 9.5-17.2 years old [60, 129].

Our results not only confirm the LLD between the involved and the non-involved side, but also provide information on the anatomical location of the difference, which is of importance when considering treatment. It is reasonable to assume that proximal tibial epiphysiodesis would be the limb equalization procedure of choice, as it is relatively well tolerated and safe.

In spastic unilateral CP it can be difficult to clearly define whether deviations in gait pattern result from the impaired motor control, LLD or are a consequence of secondary compensatory mechanisms [62-64]. GA can help to assess if gait pattern deviations are present in patients with and without LLD in unilateral CP, in other words if the patient can compensate functionally for the LLD or not. If there is an anatomical LLD and the patient compensates with increased flexion in the hip and knee and increased dorsiflexion in the ankle on the non-involved side, limb equalization surgery would probably be successful and the compensation mechanisms would disappear. On the other hand in patients whose compensation mechanisms consist of vaulting and circumduction, equalization would not be suitable.

Our results support the idea of length equalization at the tibia level, since this is where the main difference is located. In addition we agree with Allen and co-workers that the compensation mechanisms with increased flexion on the long non-involved side in the hip, knee and ankle joint dorsiflexion can be treated with the relatively safe surgical procedure, proximal epiphysiodesis.

Some patients with unilateral CP would benefit from close monitoring of LLD development with continued growth into adolescence and assessment with GA [60]. In this way the development of LLD could be better understood and a more accurate assessment to reach expected goals could be made.

### **5.6 WELL-BEING (V)**

This study shows that in teenagers and young adults with mild unilateral CP the individuals' self-esteem, but not sense of coherence, is significantly lower than in a control group. Additionally there are correlations between movement deviations in the upper extremity during walking and both self-esteem and sense of coherence. No such correlations were seen with deviations in gait.

Our findings suggest that there is an impact on self-esteem in teenagers and young adults with mild unilateral CP. We draw the same conclusion as Russo et al [119], namely that self-esteem and self-concept can be affected by physical impairment in high functioning patients with unilateral CP, which was clearly reflected in the subscales "physical appearance" and "skills".

A few studies have examined quality of life in relation to severity of involvement in cerebral palsy defined according to the GMFCS. Most report decreased quality of life with increased severity, increased GMFCS [140-143], but some report an impact on the quality of life of patients with mild involvement [69, 118].

Pirpiris and co-workers report that in patients with CP the psychosocial well-being of children with mild cerebral palsy was more strongly affected than that of children with more severe functional disability [118].

Our results confirm an association with arm posturing and can be interpreted as showing that the value of treatment for a person's appearance while moving should not be underestimated.

Furthermore we found a correlation, although moderate, between arm deviation (APS) and ITIA, SOC and EQ-5D. Elbow flexion is the main contributor to the APS. Corry et al reported good results after spasticity reduction with botulinum toxin in elbow flexors: patients and parents alike expressed satisfaction with the spasticity reduction even though there was no objective improvement in terms of normalization of elbow motion [144].

In a retrospective study Vitale and co-workers correlated the general "Child Health Questionnaire" with LLD measured by scanogram in 76 patients between 5.1 and 17.4 years of age [145]. They found a modest correlation with self-esteem, family cohesion and psychosocial summary scores. In addition those with LLD more than 20 mm had significantly lower scores (were worse), in all subscales, compared to normal controls,

## Discussion

which those with LLD smaller than 20 mm did not. We had only six patients with a discrepancy of more than 20 mm and therefore cannot draw any conclusion from this data.

All the studies mentioned above include children; it would be of interest to know if LLD increases with further growth and if the changes in gait pattern persist into adulthood.

Assessment of self-esteem and SOC might provide broader insight, reveal a patient's expectations and constitute a foundation for a more comprehensive understanding of the individual's perspective. Furthermore few studies follow patients into young adulthood which constitutes a time of transition and development [146].



## 6 GENERAL DISCUSSION

Patients with spastic unilateral CP often show differences in leg size and deviations in movement pattern during walking in both upper and lower extremities, although their impairment is mild. The deviations are subtle and difficult to define. The primary deviations can be hard to distinguish from compensation mechanisms that have developed. Ideally the compensatory mechanisms should not be treated, or at least not corrected, but rather the primary deviations, even though the brain injury itself cannot be rectified.

The Gross Motor Function Classification Scale (GMFCS) and the Functional Mobility Scale (FMS) revealed that almost all the patients in our studies attained the highest (i.e. best) scores. It is possible that not only our patients, but all patients with unilateral CP would be classified as GMFCS I and II, with a few exceptions, making the usefulness of the scale very limited in this context. Consequently these classification scales do not help in the management of high functioning individuals. However, the GMFCS and FMS contribute to some extent by describing the groups of patients investigated in these studies [49, 53].

In teenagers and young adults treatment is seldom considered partly because the impairment is mild and partly because few effective treatment options are available. Targeted muscle strengthening and coordination exercise programs combined with focal muscle lengthening and bone realignment in specific cases could provide improvement.

We are of the opinion that the goal for most high functioning teenagers and young adults with motor impairment will be to normalize their movement pattern, and we challenge the assumption that activity and participation is their ultimate goal, or can be used as a measure of successful management and treatment.

Movement pattern and symmetry are of importance. “As a social animal, a human’s survival depends upon its ability to identify, to interpret and to predict the actions of others” [70]. Enquist and Arak published a paper with the title “Symmetry, beauty and evolution” in *Nature* 1994, where they discuss the influence of symmetry in recognizing objects, especially human motion, but also in a biological signaling context [147].

The common abnormalities of arm and hand positioning caused by spasticity may be socially stigmatizing for the child and cause parents anxiety. Arm posturing and gait deviations may be perceived as cosmetic and social impediments when the individual enters adolescence and becomes more self conscious [58, 59, 69].

For these reasons data was collected on both upper and lower extremity deviations during walking, and well-being was assessed.

One needs to bear in mind that treating children and teenagers with CP – regardless of level of function – is one of the most difficult and challenging tasks in pediatric orthopedic surgery. It is the professional treatment team around these patients that have the knowledge and responsibility to provide the children/teenagers/young adults and

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their families with the best recommendations and the best care possible. The counseling needs to contain information, recommendations, support and guidance in the direction towards the long term goal ahead.

For these reasons we were interested in studying not only young children but also the experiences and changes in teenagers and young adults with unilateral CP. Youth and young adulthood is the time for establishment in the grown up world and the degree of success in this venture is in some ways an evaluation of whether the long term goal has been reached.

It can be difficult to detect deviations based solely on sagittal kinematic data from Gait analysis (GA). Other kinematic variables, kinetic data, anatomical differences in muscle volume and leg length might have impact on gait and movement and reveal changes in this high functioning group. In addition the well-being of these individuals is important to assess so as to learn about their concerns with movement patterns. Of interest would be to complement the Winters classification with not only the type 0, but also with other variables from the GA. In conjunction with possible future Magnetic resonance imaging (MRI) classification mirroring the time and extent of the brain lesion, a more comprehensive classification system could be developed.

Dynamic quantitative and objective assessment with GA including various variables and MRI muscle volume measurements are advanced diagnostic and outcome instruments that could help in developing adequate treatment options even for the high functioning individual with unilateral CP.

Targeted muscle strengthening programs could be developed using MRI assessments of muscle volume before and after physiotherapy muscle strengthening programs. These high functioning patients may be in better position with milder motor impairment than patients in previous studies, and therefore benefit more.

### **6.1 THEORY ABOUT MUSCLE AND LEG LENGTH DIFFERENCE**

We cannot conclude whether the differences in muscle volume and leg length arise because the brain injury alters patterns of innervations and growth factor release, or because of disuse of the involved limb, with less muscle activity and less loading. It is reasonable to believe that mechanical load differences between the involved and non-involved side is not the only explanation to differences.

Even if the support time (time of loading) during walking is shorter on the involved limb than on the non-involved side, the muscle forces acting over the joints for stabilization also influence the growth plates. One would expect that in the knee joint for example, both the proximal growth plate of the tibia and the distal growth plate of the femur would be influenced, and in that way affect the length of both bones. However, that is not the case, and in addition the MRI studies revealed that differences in both muscle volume and leg length are more pronounced distally in the limb than proximally.

The distal location of changes in the limb coincides with the areas most strongly affected by the cerebral palsy and spasticity. Even if the underlying cause of the muscle

volume differences and leg length discrepancy cannot be determined, we speculate that in unilateral CP, the brain injury itself directly influences the growth of the limb.

### **6.2 METHODOLOGICAL CONSIDERATIONS AND LIMITATIONS**

One of the limitations of these studies was the selection and recruitment of patients, which is often the case in cross-sectional studies and retrospective studies. There was no randomized or otherwise controlled recruitment of patients, and it is not possible to assess skewness in the selection.

The patients were not assessed by a neurologist to evaluate and confirm the diagnosis of unilateral CP at the time of inclusion. On the other hand two experienced physiotherapists saw all the patients and performed the physical examination.

#### **Three dimensional gait analysis**

Capturing and quantifying motion with existing three dimensional techniques involve several problems regarding accuracy. Models used are derived from normal adults and are hardly subject specific. There are several reports on subject specific models and Scheys et al. used MRI based kinematic models and traditional generic models and could show differences in patients with CP [148]. Consistent marker placement is another problem, that was minimized by having few well trained persons placing the markers in the same studies [149].

In study IV a regression analysis was performed to assess if walking speed had any influence on muscle work ratios between the involved and non-involved side. However there are several earlier reports with musculoskeletal modeling and simulations calculating the effect of walking speed on muscle function and describing muscles contributing to stability and progression [97, 150, 151]. In these studies walking speed was not controlled for other than in relation to influence on muscle work ratio (study IV).

One limitation in study II was the use of the term power generation over time. The values of these measurements cannot be compared with the values in study IV. The reason is that in study II, power, the rate at which work is performed (Watts) is calculated on the time unit, percent of gait cycle. However the results can be used for comparisons with the same report or for side to side comparisons within the same patient.

#### **Magnetic resonance imaging**

Muscle volume measurements with MRI are time consuming and not yet automated, since muscle distinction is difficult or impossible with current methods [139]. In these studies the quality of the imaging was in some cases low causing difficulties with the measurements of muscle volumes. The task was too time consuming and we could not include more than 30 of the total 46 patients.

Imaging methods for measuring leg length are standard radiographic techniques, computed tomography, ultrasound and MRI scans [104, 152]. Leitzes et al compared results obtained with MRI, conventional radiography, and computer tomography

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regarding length on cadaver femur and found that all three techniques gave excellent reliability [153]. MRI has won increasing acceptance as a technique for bone assessment in musculoskeletal imaging with no harmful ionizing radiation. There are limitations regarding accessibility and a relatively long imaging time though. Additionally the method is very dependant on patient cooperation which might be a problem in children [154]. Sabharwal and Kumar concluded in a recent review article, “at present there is no single imaging method that can be considered ideal”[154].

Another limitation of the muscle volume study is that the MRI measurement technique has not been tested for reliability. Inter- and intra-observer comparisons are ongoing that will show how valid and reliable these measurements are.

### **Self reporting questionnaires**

The high correlation between sense of coherence (SOC) and self-esteem (ITIA) questionnaires reflects that they to a high extent measure the same thing. From this perspective the SOC has fewer questions and is much faster to answer.

The EQ-5D was developed for adults but has been used down to the age group 14-15 years in previous studies. A child-friendly version in Swedish has recently been developed [155]. However there are still challenges using the child-friendly version since it has not been used in large groups of patients and there are until now no weighted scores, as in the adult version.

The lack of an in-depth exploration of self-esteem and quality of life is also a limitation; these issues could have been followed up with more thorough interviews by professionals in the field. In addition an assessment of the patients’ participation and degree of activity was not performed, although this would have provided useful information on the group.

## **6.3 ETHICAL CONSIDERATIONS**

In study I and II the patients included were performing a GA as part of their medical care. In study III, IV and V patients were participating after they had agreed to (informed consent) and the investigations were performed exclusively for the study. Neither GA nor MRI investigations are harmful or painful. Even though the questions in the self reporting questionnaires were rather neutral and not provocative or intimate, they could of course generate reflections and thoughts that the patient might find uncomfortable. For this reason the mothers of two young patients, who showed signs of distress were contacted afterwards to confirm that they were feeling well and were in their habitual mode. One of these children had showed some concern when filling in the questionnaire and the other one when performing the MRI investigation. The examination with GA and MRI together with filling out the questionnaires was time-consuming. For the purpose of these studies this was considered clearly acceptable from an ethical point of view.

## 7 CONCLUSIONS

Twenty-six of 112 (23%) patients with unilateral CP had mild involvement and could not be classified according to the Winters' classification. The classification is complemented with a type 0 which allows all patients to be classified.

Patients move between classification types with time and treatment. The new modified Winters classification scheme helps to make comparisons and prognoses.

There is bilateral shift of concentric muscle work from the plantar flexors of the ankles to the hip extensors in children and teenagers with unilateral CP compared to controls.

Power loss at the ankle following Achilles tendon lengthening may not influence the gait pattern significantly, since most of the power comes from the hip muscles.

In 25% of the patients the leg length discrepancy exceeded 15 mm, even in those with mild unilateral CP. The discrepancy is mainly located in the tibia, talus and calcaneus, which has clinical implications for where treatment should be targeted at limb equalization.

This limited study suggests a relationship between the degree of involvement in CP and the location of the leg length discrepancy.

Muscle volumes on the involved side in unilateral CP are smaller than corresponding volumes on the non-involved side, with more pronounced differences distally in the limb.

Deviations from normal are recognizable in kinetic data: there are concentric muscle work changes even in mild unilateral CP in young children up into adult ages.

Kinetic data can contribute to identify and separate compensation mechanisms from primary deviations.

Teenagers and young adults with mild unilateral CP score lower on self-esteem assessments than healthy controls and increased deviation in arm movement is associated with lower self-esteem.

When evaluating teenagers and young adults with unilateral CP the movement pattern should not be assessed solely from a functional perspective. Possible concerns about appearance and influence on self-esteem should be considered.



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