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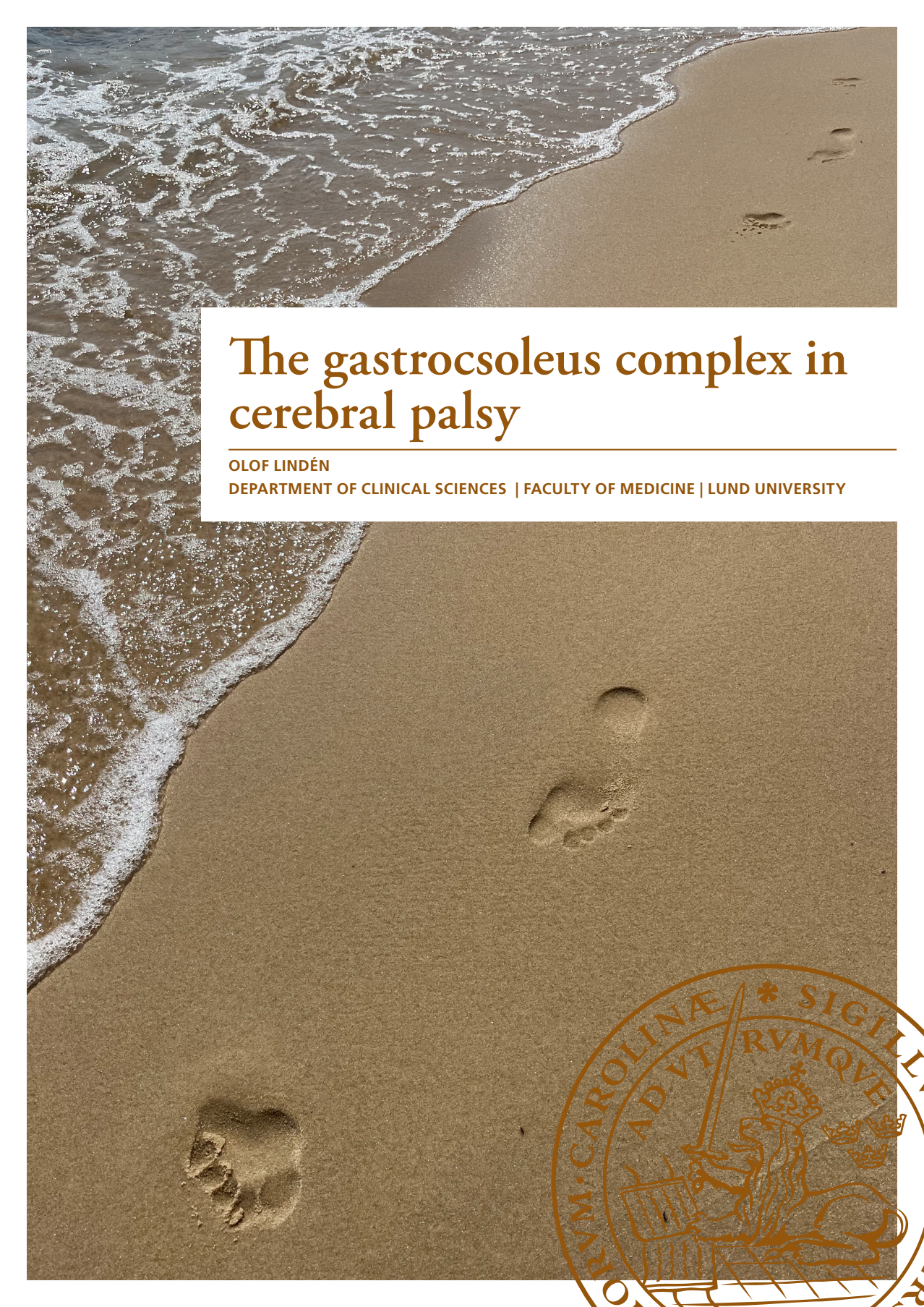
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The gastrocsoleus complex in cerebral palsy

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The gastrocnemius complex in cerebral palsy

The gastrocsoleus complex in cerebral palsy

Olof Lindén



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DOCTORAL DISSERTATION

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Abstract:

Background: Spasticity, contractures, and gait impairments are common challenges in children with cerebral palsy (CP). Spasticity of the gastrocsoleus muscle can lead to equinus deformity, which is often managed with surgical lengthening. In Sweden, the CPUP follow-up program monitors motor function, spasticity, and treatment interventions for nearly all children with CP nationwide.

Purpose: To increase knowledge on the development of spasticity and equinus deformity, the outcomes of gastrocsoleus lengthening, the reliability of range of motion (ROM) measurements, and regional variations in treatment practices.

Methods: Four studies were conducted using data from the Swedish CPUP registry. Study I analyzed longitudinal spasticity development in the gastrocsoleus muscle among children aged 0–15 years. Study II examined ankle and knee ROM after isolated percutaneous or open Achilles tendon lengthening, and gastrocnemius lengthening. Study III evaluated whether spasticity introduces bias in passive ROM measurements in children with unilateral spastic CP. Study IV explored regional differences in the use of botulinum toxin A, orthoses, casting, and surgery to manage equinus deformity.

Results: Spasticity increased until around age 5, then declined gradually, most notably in children with more severe motor impairment. Mean ankle ROM followed a similar pattern following surgery across all techniques, but was more favorable after percutaneous Achilles tendon lengthening. No side-related bias was found in ROM measurements. Significant regional variation in treatment strategies for equinus deformity was observed.

Conclusion: Spasticity follow a predictable age-related pattern in children with CP. Surgical outcomes vary with technique, and treatment practices differ between Regions. While CPUP enables detailed surveillance and quality improvement, further efforts are needed to ensure evidence-based, equal care across Sweden.

Key words: Cerebral Palsy, Spasticity, Range of Movement, Gastrocsoleus Lengthening

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The gastrocsoleus complex in cerebral palsy

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To children with cerebral palsy and their families.

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List of papers

The present thesis is based on the following original papers, which are referred to throughout the thesis by their Roman numerals:

- I. Lindén O, Hägglund G, Rodby-Bousquet E, Wagner P. The development of spasticity with age in 4,162 children with cerebral palsy: a register-based prospective cohort study. *Acta Orthop.* 2019 Jun;90(3):286-291. doi: 10.1080/17453674.2019.1590769. Epub 2019 Mar 25. PMID: 30907682; PMCID: PMC6534199.
- II. Lindén O, Lauge-Pedersen H, Hägglund G, Wagner P. Development of ankle and knee range of motion after isolated gastrocsoleus lengthening in children with cerebral palsy: a register-based longitudinal cohort study. *Acta Orthop.* 2025 Apr 17;96:331-338. doi: 10.2340/17453674.2025.43387. PMID: 40242883; PMCID: PMC12006035.
- III. Lindén O, Lauruschkus K, Wagner P, Hägglund G, Lauge-Pedersen H. The influence of spasticity on goniometric range of motion measurements in children with cerebral palsy. (Submitted)
- IV. Lindén O, Hägglund G, Lauge-Pedersen H. Regional Variation in the Management of Equinus Deformity in Children with Cerebral Palsy in Sweden. (Manuscript)

Abstract

The gastrocsoleus complex in cerebral palsy

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Background: Spasticity, contractures, and gait impairments are common challenges in children with cerebral palsy (CP). Spasticity of the gastrocsoleus muscle can lead to equinus deformity, which is often managed with surgical lengthening. In Sweden, the CPUP follow-up program monitors motor function, spasticity, and treatment interventions for nearly all children with CP nationwide.

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Abbreviations

AIC	Akaike information criterion
ARC	Annual rate of change
AS	Ashworth scale
BSCP	Bilateral spastic cerebral palsy
BTX-A	Botulinum toxin A
CI	95% Confidence interval
CP	Cerebral palsy
CPUP	Swedish surveillance program for cerebral palsy
GCL	Gastrocnemius lengthening
GMFCS	Gross motor function classification system
ICC	Intraclass correlation coefficient
ICF	International classification of functioning, disability and health
ITB	Intrathecal baclofen pump
MAS	Modified Ashworth scale
MEM	Mixed-Effects Model
OTAL	Open Achilles tendon lengthening
PTAL	Percutaneous Achilles tendon lengthening
ROM	Range of motion
SCPE	Surveillance of cerebral palsy in Europe
SDR	Selective dorsal rhizotomy
TAL	Achilles tendon lengthening
USCP	Unilateral spastic cerebral palsy

Thesis at a glance

Study	Aim	Methods	Results	Conclusions
I	To describe the development of spasticity in the gastrocnoleus muscle in children with CP.	Longitudinal data from CPUP were used. Ashworth Scale scores from ages 1–15 were analyzed for 4,162 children.	Spasticity followed a biphasic pattern with a peak in early childhood and gradual decline in adolescence. Most pronounced in children with GMFCS III–V.	Spasticity is dynamic over time. Age and GMFCS level should be considered in treatment planning and when evaluating interventions.
II	To compare outcomes after three surgical techniques for gastrocnoleus lengthening.	Children who had undergone PTAL, OTAL, or GCL were followed in CPUP. Ankle/knee ROM development and event-free survival were analyzed.	Mean ROM development was similar between groups. Event-free survival was higher after PTAL.	PTAL may be associated with more favorable long-term outcomes regarding ankle ROM.
III	To assess the agreement of passive ROM measurements in children with unilateral spastic CP.	Goniometric measurements of the knee and ankle were compared between sides and examiners. Agreement was assessed using mean differences and limits of agreement.	No systematic side-related bias found. Inter-examiner agreement was good to excellent.	Manual goniometry showed no systematic side-related bias, supporting its use in children with CP.
IV	To investigate regional variation in treatment of equinus in children with CP.	National CPUP data were analyzed for 4,551 children. Proportion treated with TAL, serial casting, or BTX-A was compared across Regions.	Significant regional differences were found for all three treatments. Patterns varied also by GMFCS level.	Treatment decisions are influenced by more than clinical factors. National coordination may be needed to improve consistency and equity in care.

Introduction

Cerebral palsy

Introduction

Overview of Cerebral Palsy

Cerebral palsy (CP) is the most common cause of motor disability in children, with a prevalence historically ranging between 2–3 per 1,000 live births(1) but declining in recent studies(2). It is a group of permanent disorders affecting, among other things, the development of posture and movement, caused by non-progressive disturbances to the developing fetal or infant brain(3).

Although the brain injury itself is non-progressive, the clinical presentation may change over time in relation to functional ability, muscle tone, and range of motion. CP is traditionally classified according to motor type (spastic, dyskinetic, ataxic, or mixed/unspecified) and topographical distribution (e.g., Unilateral Spastic Cerebral Palsy – USCP; Bilateral Spastic Cerebral Palsy – BSCP)(4, 5).

Spastic CP, the most common subtype (represent 75–80% of the population with CP(1)), is characterized by increased muscle tone and stiffness, but also muscle weakness, leading to difficulties with voluntary movement and a risk of developing contractures and skeletal deformities. Spasticity is typically asymmetric and variable in its presentation, and often coexists with other motor impairments, such as muscle weakness, impaired proprioception, loss of selective motor control, and co-contraction of agonist and antagonist muscles, which further impair voluntary movement(4, 6, 7). The clinical presentation varies widely and is influenced by the severity and distribution of the brain injury as well as by secondary musculoskeletal adaptations(8). The International Classification of Functioning, Disability and Health (ICF) provides a comprehensive framework to describe and understand these variations in functioning among children with CP(9).

Understanding the neuropathology and functional development in CP is essential for designing appropriate assessment and treatment strategies during both childhood and adolescence.

Historical background

Although detailed medical documentation of CP is lacking before the 19th century, evidence suggesting the existence of CP in the Ancient World of Egypt and Greece can be found in art, literature, and paleopathological findings. However, due to limited historical records, it remains speculative. The earliest known medical description of a condition resembling CP was provided by Hippocrates in the *Corpus Hippocraticum*(10). The first detailed description was made by William Little in the 1840s, linking spasticity and joint deformities in children to early brain injury, particularly related to prematurity and birth complications. The condition was long referred to as Little's Disease. William John Little was a pioneering figure in orthopedic surgery and is often credited with introducing the technique of Achilles tendon tenotomy to England. After undergoing a successful tenotomy performed by Louis Stromeyer in Germany to correct his own clubfoot, Little was inspired to adopt and promote this surgical method. In 1837, he performed one of the first documented Achilles tendon tenotomies in England on a 15-year-old boy, marking a significant advancement in the treatment of neuromuscular deformities. Stromeyer referred to Little as the "Apostle of Tenotomy"(11). His work laid the foundation for modern orthopedic approaches to conditions like clubfoot and spastic CP(12). Over time, other key figures like William Osler and Sigmund Freud contributed to early classification systems. In the mid-20th century, the Little Club in the UK defined CP as a permanent but not unchanging motor disorder due to non-progressive damage to the developing brain. In 2006, an international expert group proposed a modern definition of CP as a group of permanent movement and posture disorders caused by non-progressive disturbances in the developing brain, often accompanied by sensory, cognitive, and behavioural impairments(4, 13).

Epidemiology and Prevalence

CP is the most common cause of impaired motor function in childhood, with a reported prevalence of approximately 1–3 per 1,000 live births in high-income countries(1, 2, 14). In low- and middle-income countries prevalence is uncertain, with estimates ranging from 0.8 to over 10 per 1,000 live births. This is likely due to limited access to maternal and neonatal healthcare, higher rates of perinatal risk factors, and lower survival of preterm infants with adequate neuroprotective care(15). Overall prevalence has declined over recent decades, possibly due to advances in neonatal care. Notably, the introduction of therapeutic hypothermia for neonates with hypoxic-ischemic encephalopathy has been associated with a reduced incidence of CP(2, 16). Initiatives such as the *Surveillance of Cerebral Palsy in Europe* (SCPE) and population-based CP registers like *Follow-Up Surveillance Programme for People with Cerebral Palsy* (CPUP) have been essential in tracking prevalence trends and forming public health strategies aimed at prevention and early intervention.

Etiology and Risk Factors

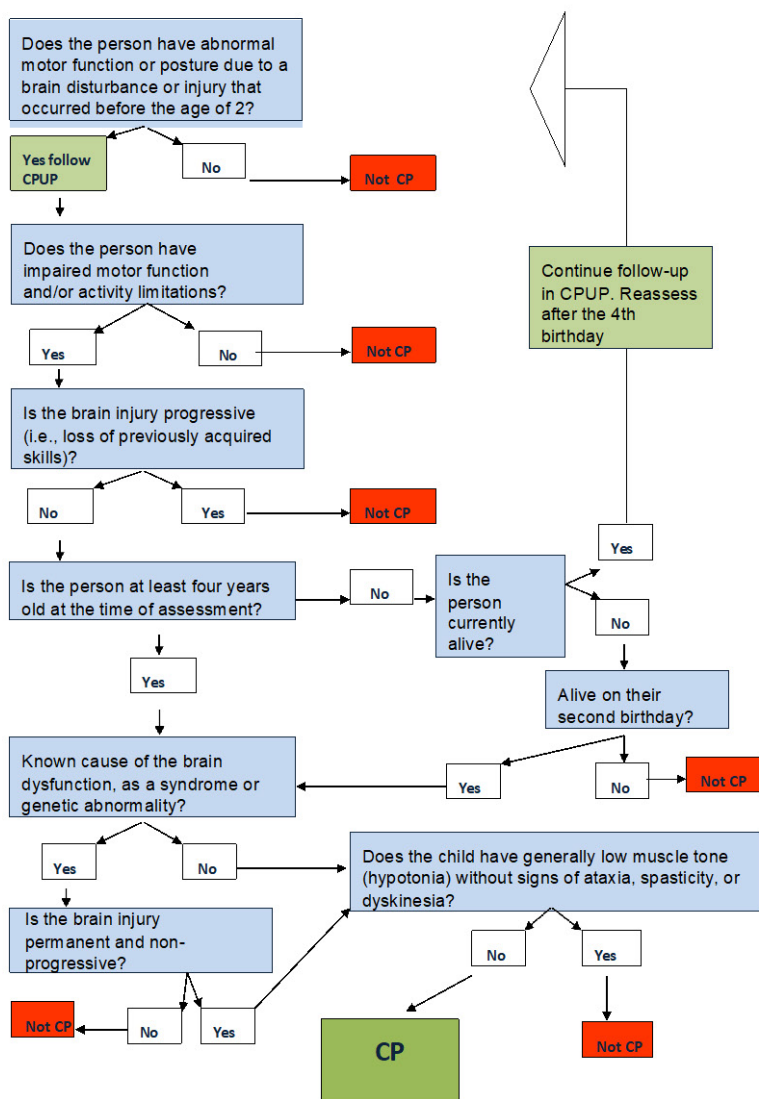
The etiological pathways can be broadly divided into prenatal, perinatal, and postnatal origins. Prenatal factors include congenital brain malformations, intrauterine infections, genetic abnormalities, and placental insufficiency(17). Perinatal causes are often related to birth complications such as asphyxia, preterm birth, low birth weight, and neonatal stroke(18). Postnatal contributors may include infections (e.g., meningitis, encephalitis), head trauma, or severe untreated neonatal jaundice(19).

Preterm birth is one of the most significant risk factors for CP, particularly when associated with intraventricular hemorrhage or periventricular leukomalacia(17). Other well-established risk factors include multiple births, maternal infections, and complications during delivery(17, 20). However, in many cases, the precise cause remains unknown, and it is increasingly recognized that a combination of vulnerabilities and events often contributes to the development of CP(20).

Classification of CP Subtypes

In CPUP, the diagnosis of CP is established by a pediatric neurologist, ensuring consistency and adherence to standardized criteria (Figure 1). CP is a clinically diverse condition, and classification plays an important role in diagnosis, communication, treatment planning, and research. To improve consistency across clinical and research settings, standardized classification systems such as the one developed by the SCPE have been adopted (Figure 2). This approach prioritizes neurological subtype (spastic, dyskinetic, or ataxic) and then describes the topographical distribution, e.g., BSCP or dystonic CP. This enhances comparability between studies and supports more precise descriptions of patient populations. The motor classification identifies the predominant movement disorder and includes three main subtypes: spastic, dyskinetic, and ataxic CP. *Spastic CP* is by far the most common and is characterized by increased muscle tone, spasticity. *Dyskinetic CP* involves involuntary movements and is further subdivided into dystonic and choreoathetotic forms. *Ataxic CP* is less common and primarily affects balance and coordination. When more than one motor type is present without a single dominant pattern, the term mixed CP may be used. Topographical classification refers to the distribution of motor impairment across the body. In spastic CP, this is typically described as USCP (sometimes referred to as spastic hemiplegia) or BSCP (which may present as diplegia or quadriplegia, depending on the extent of limb involvement). Dyskinetic and ataxic forms are generally considered bilateral.

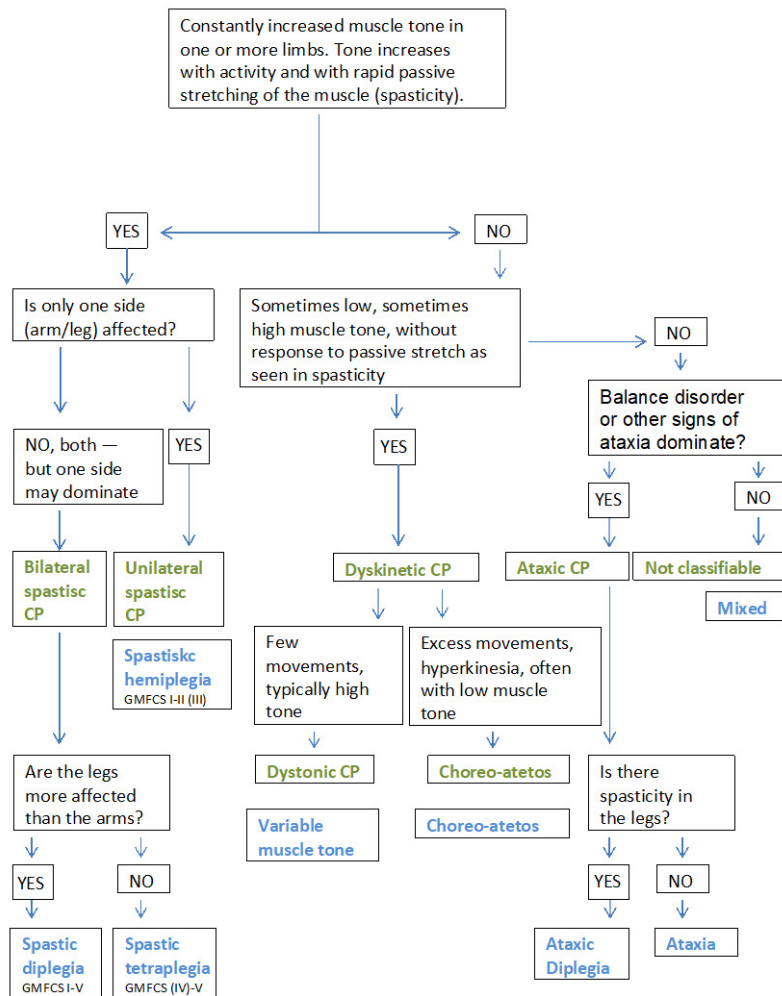
Decision Tree for the Diagnosis of Cerebral Palsy



Version for CPUP, based on SCPE 2000 and Rosenbaum et al 2007/lw 2014

Figure 1. Translated and reprinted with permission from CPUP.

Classification of CP Subtypes Between 4 and 8 Years of Age



Green = subtypes - SCPE
Blue = subtypes- Swedish Classification

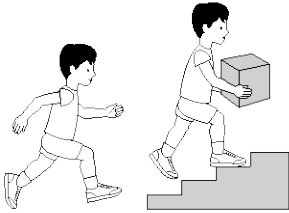
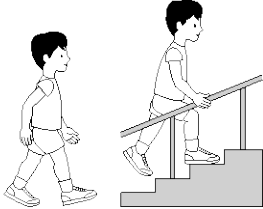
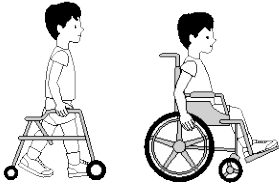
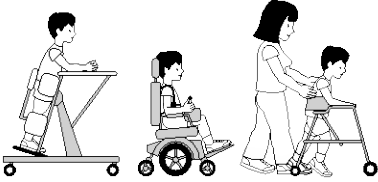
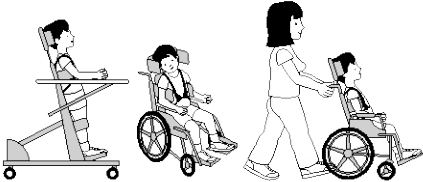
SCPE 2000, modified for CPUP

Figure 2. CP subtypes according to SCPE in green(5) and according to Hagberg in blue(1). Translated and reprinted with permission from CPUP.

Classification of gross motor function

The Gross Motor Function Classification System (GMFCS) was developed in the late 1990s to provide a standardized method for classifying gross motor function in children with CP (Figure 3)(21). Prior to its introduction, classification of motor severity was inconsistent, often relying on vague terms such as *mild*, *moderate*, or *severe*, which lacked precision and reproducibility. The GMFCS was designed to reflect a child's self-initiated movement, especially sitting and walking, with an emphasis on functional abilities, need for assistive devices, and limitations in mobility. The system includes five ordinal levels, where Level I represents the highest level of independence (walking without limitations), and Level V reflects the most severe limitations in motor function (requiring extensive support for mobility and posture). Classification is age-specific, originally spanning from infancy to 12 years, with a later addition of a band for ages 12–18 to account for changes in function during adolescence. Each level describes typical performance, not best capacity, and the GMFCS has shown excellent inter-rater reliability and stability over time(22). It is now widely used in both clinical and research settings as a reference framework for understanding functional status, planning interventions, and stratifying study populations.

GMFCS E & R between 6th and 12th birthday: Descriptors and illustrations

	GMFCS Level I Children walk at home, school, outdoors and in the community. They can climb stairs without the use of a railing. Children perform gross motor skills such as running and jumping, but speed, balance and coordination are limited.
	GMFCS Level II Children walk in most settings and climb stairs holding onto a railing. They may experience difficulty walking long distances and balancing on uneven terrain, inclines, in crowded areas or confined spaces. Children may walk with physical assistance, a hand-held mobility device or used wheeled mobility over long distances. Children have only minimal ability to perform gross motor skills such as running and jumping.
	GMFCS Level III Children walk using a hand-held mobility device in most indoor settings. They may climb stairs holding onto a railing with supervision or assistance. Children use wheeled mobility when traveling long distances and may self-propel for shorter distances.
	GMFCS Level IV Children use methods of mobility that require physical assistance or powered mobility in most settings. They may walk for short distances at home with physical assistance or use powered mobility or a body support walker when positioned. At school, outdoors and in the community children are transported in a manual wheelchair or use powered mobility.
	GMFCS Level V Children are transported in a manual wheelchair in all settings. Children are limited in their ability to maintain antigravity head and trunk postures and control leg and arm movements.

GMFCS descriptors: Palisano et al. (1997) *Dev Med Child Neurol* 39:214-23
CanChild: www.canchild.ca

Illustrations: Version 2 © BRL Reid, Kate Willoughby, Adrienne Harvey and Kerr Graham,
The Royal Children's Hospital Melbourne: EHC151050

Figure 3. The 5 levels of GMFCS (Gross Motor Function Classification System)(21). Reprinted with permission from prof. Kerr Graham, The Royal Children's Hospital, Melbourne, Australia.

Follow-Up Surveillance Programme for People with Cerebral Palsy - CPUP

CPUP is a Swedish national follow-up and surveillance programme for individuals with CP, developed to improve long-term outcomes and reduce preventable complications such as severe contractures, hip dislocations and severe scoliosis. The programme was initiated in southern Sweden in 1994 as a regional quality improvement initiative and has since expanded to cover the entire country, becoming an official national quality register in 2005 and since 2009 also including adults with CP. Children suspected of having CP are enrolled in the CPUP program as early as possible, typically before the age of 2 years. The diagnosis is confirmed by a neuropediatrician after 4 years of age, and children without CP are subsequently removed from the register. The proportion of children later found not to have CP is below 2%(1).

The aim of CPUP is to enable early detection and preventive intervention through regular, standardized assessments conducted by a multidisciplinary team. The programme includes systematic follow-up of motor function, range of motion, muscle tone, spinal alignment, and hip development, with results recorded in a centralized database. Children and adolescents with CP are assessed according to their GMFCS level, with the frequency of follow-up adjusted accordingly.

In addition to physiotherapeutic and orthopedic monitoring, CPUP also gathers data on interventions, assistive devices, and participation in daily life. Data are contributed not only by physiotherapists and orthopedic surgeons, but also by occupational therapists, orthotists, speech and language therapists, and psychologists. The programme also aims to increase knowledge about CP and to enhance collaboration between professionals involved in the care of individuals with CP. The programme has contributed significantly to improving clinical outcomes in Sweden and has been adopted as a model in several other countries.
<https://cpup.se/>

Functional Impairments and Clinical Presentation

Spasticity and Muscle Tone Abnormalities

Spasticity is the most prevalent motor abnormality in children with CP, particularly in the spastic subtype, which accounts for the majority of CP cases. It is classically defined by Lance as a “*velocity-dependent increase in tonic stretch reflexes (muscle tone) with exaggerated tendon jerks, resulting from hyperexcitability of the stretch reflex, as one component of the upper motor neuron syndrome*” (23).

The pathophysiology of spasticity involves disruption of descending inhibitory pathways, particularly the corticospinal and corticoreticular tracts, which normally modulate spinal reflex activity. This disinhibition leads to increased excitability of alpha motor neurons in the spinal cord, resulting in exaggerated stretch reflexes and

increased resistance to passive movement. In CP, spasticity often presents early in life and contributes to muscle shortening, joint contractures, and abnormal postural patterns over time. Spasticity is typically asymmetric and variable in its presentation, and often coexists with other motor impairments, such as muscle weakness, impaired proprioception, loss of selective motor control, and co-contraction of agonist and antagonist muscles, which further impair voluntary movement(4, 6, 7).

Understanding spasticity and its underlying mechanisms is essential for developing effective treatment strategies. Therapeutic approaches include physical therapy, pharmacologic agents (e.g., botulinum toxin A (BTX-A) injections and baclofen) and orthopedic or neurosurgical interventions, all of which aim to reduce tone, prevent contractures, and improve function.

Contracture Development

Children with CP, particularly those with spastic subtypes, are at high risk of developing muscle contractures over time. A contracture is a permanent shortening of muscles, tendons and/or joint capsule, leading to reduced joint range of motion (ROM). The development of contractures is influenced by several factors, including spasticity, muscle strength imbalance, growth-related imbalance between muscle and bone length, and limited mobility. The pathophysiology involves both neural and non-neural mechanisms. Loss of descending inhibitory control leads to increased stretch reflex activity, which—combined with muscle disuse and altered loading—results in progressive structural changes where the contract muscles display reduced fascicle length, increased stiffness, and alterations in connective tissue. These adaptations reduce the muscle’s ability to lengthen during growth, contributing to contracture formation(24). Contractures most commonly first affect biarticular muscles, such as the gastrocnemius, hamstrings, and hip flexors. Their functional impact depends on severity, joint involvement, and overall motor function level. Children classified in GMFCS levels III to V are at particular risk, as reduced active mobility and limited weight-bearing contribute to the development of muscle shortening(25). Early identification of reduced joint mobility and preventive strategies including stretching programs, orthoses, serial casting, treatment aimed at reducing spasticity, and, when indicated, orthopedic surgery are essential to limit the development and consequences of contractures(26).

Assessments Tools and Key Concepts

Spasticity Measurement: Modified Ashworth Scale (MAS)

The Ashworth Scale (AS)(27), introduced in 1964, is a clinical tool used to assess spasticity by measuring resistance during passive muscle stretch. In 1987, the scale was modified by Bohannon and Smith to improve its sensitivity, particularly for

detecting mild increases in muscle tone. This Modified Ashworth Scale (MAS, Table 1) introduced an additional grade (1+), resulting in a six-point scale (0, 1, 1+, 2, 3, 4)(28). The MAS is now more commonly used in both clinical and research settings, including within the CPUP program. While clinical evaluation of spasticity is most commonly performed using MAS, other instruments such as the Modified Tardieu Scale are also available(29). MAS is widely used due to its simplicity, but it primarily reflects passive resistance and has been criticized for its limited ability to distinguish spasticity from soft tissue stiffness or contracture. The Tardieu Scale, by incorporating movement velocity and angle of muscle reaction, provides additional information about the neural and mechanical contributions to increased tone(29). MAS has been used for several manuscripts in this thesis and is shown to be reliable(28, 30, 31).

Table 1. Modified Ashworth Scale (MAS)

Score	Description
0	No increase in muscle tone
1	Slight increase in tone, catch and release or minimal resistance at end of ROM
1+	Catch followed by minimal resistance through less than half of ROM
2	Marked increase in tone through most of ROM, but part easily moved
3	Considerable increase in tone, passive movement difficult
4	Affected part rigid in flexion or extension

*Passive Range of Motion Assessment: **Goniometry***

Goniometry refers to the systematic measurement of joint angles within specific planes of motion. The term derives from the Greek words *gonia* (angle) and *metron* (measure). The earliest known use of a goniometric concept dates to the 16th century, when the Dutch physician and mathematician Gemma Frisius employed a primitive angular measurement device to determine the position of celestial bodies in relation to the Earth. While originally developed for astronomical purposes, the principles of angle measurement later informed the design of instruments used in clinical assessment of human joint mobility(32).

Assessment of passive range of motion (ROM) is a key component in the clinical evaluation of children with CP, particularly for monitoring the development of muscle contractures and joint restrictions. It involves aligning a manual goniometer along anatomical landmarks to quantify joint angles during passive movement, e.g. to assess joint angles in the ankle, knee and hip. Accurate and repeated ROM assessment is essential for tracking musculoskeletal changes over time and for informing decisions about interventions(33).

Although goniometry is widely accepted, its inter-rater and intra-rater reliability might vary, particularly in populations with spasticity. Factors such as patient cooperation, muscle tone, positioning, and examiner technique can affect measurement consistency. Efforts to standardize procedures and use trained assessors can improve reliability(34, 35).

Radiographic assessment

Radiographic imaging plays a central role in the surveillance of musculoskeletal complications in children with CP, particularly in the monitoring of hip displacement and spinal deformities. Within CPUP, regular standardized hip radiographs are conducted to identify early signs of lateral displacement of the femoral head, often quantified using the migration percentage(36) (<https://cpup.se/>). This is crucial for timely intervention and prevention of hip dislocation, which can significantly impact pain, function, and quality of life(37). CPUP includes guidelines for radiographic hip screening based on age and GMFCS level, with increased frequency for children at higher risk (GMFCS levels III–V)(38). In addition, spinal radiographs are used to detect and follow the progression of scoliosis, especially in non-ambulant children, with assessments guided by Cobb angle measurements(39). These radiographic tools are essential complements to clinical examinations and functional assessments, providing objective data to guide orthopedic decision-making.

Gait analysis

Children with CP often present with complex and heterogeneous gait deviations due to both primary motor impairments and secondary musculoskeletal deformities. Gait analysis provides a comprehensive, objective assessment of gait often combining data from kinematics, kinetics, electromyography, plantar pressure, and video recordings. Gait analysis enables identification and understanding of individual gait deviations and facilitates clinical decision-making. It supports treatment planning by linking observed gait deviations to underlying impairments and helps evaluate outcomes following interventions(40).

Gait deviations in CP are commonly grouped into characteristic patterns. Rodda and Graham's classification provides a clinically useful framework for understanding common gait patterns in children with USCP and BSCP, linking observed postural patterns to underlying impairments and guiding treatment decisions(41).

In *USCP*, four types of gait patterns are described:

- *Type 1*: Isolated drop foot during swing phase; typically requires only orthotic support.
- *Type 2*: True equinus throughout stance phase; subdivided into 2a (neutral knee) and 2b (knee hyperextension); managed with orthoses, BTX-A, and possibly gastrocnemius or Achilles tendon lengthening.

- *Type 3*: Equinus combined with stiff knee gait due to hamstring/quadriceps co-contraction; may require combined soft tissue surgery.
- *Type 4*: Severe proximal involvement with equinus, flexed knee and hip, hip adduction and internal rotation; treatment includes multilevel soft tissue and bony surgery.

In *BSCP*, four sagittal plane patterns are outlined:

- *True equinus*: Ankle plantarflexion with extended knees and hips.
- *Jump gait*: Equinus, knee and hip flexion, anterior pelvic tilt and lumbar lordosis.
- *Apparent equinus*: Normal ankle position but with flexed knees and hips.
- *Crouch gait*: Excessive dorsiflexion at the ankle combined with increased flexion at the knee and hip.

These patterns tend to evolve with age and intervention, and are influenced by factors such as muscle spasticity, contractures, and skeletal deformities. The plantarflexion–knee extension couple is a key biomechanical concept for understanding and managing these deviations. The classification serves as a basis for an algorithmic approach to intervention, incorporating spasticity management, soft tissue and bony surgery, and orthotic use(41).

Anatomy and Physiology of the Gastrosoleus Complex

The triceps surae complex or the gastrosoleus complex, comprising the gastrocnemius and soleus muscles and their shared distal tendon (the Achilles tendon), plays a central role in posture, gait, and propulsion (Figure 4). Anatomically, the gastrocnemius consists of two heads, medial and lateral, that originate from the posterior femur and cross three joints: the knee, ankle, and subtalar joints. In contrast, the soleus lies deep to the gastrocnemius and originates from the posterior tibia and fibula, crossing only the ankle and subtalar joints. Both the gastrocnemius and soleus muscles give rise to their own aponeuroses, from which individual tendons originate. These tendons glide independently for a distance before merging to form the Achilles tendon approximately 5 to 6 cm proximal to its insertion on the calcaneus(42). The tendon undergoes fiber rotation, with varying degrees of twist (30–150°)(43). Differentiation between gastrocnemius and soleus tightness can be attempted using the Silfverskiöld test, in which ankle dorsiflexion is assessed with the knee extended and then flexed; a greater restriction in extension indicates gastrocnemius involvement. (44).

Functionally, the gastrocnemius contributes primarily to dynamic movements such as push-off during gait, while the soleus serves more as a postural muscle. Muscle strength studies show that the soleus accounts for the majority of plantarflexion

force. This underscores the soleus' importance in sustained load-bearing tasks(45). The gastrocnemius complex plays a crucial role in stance phase control during gait. These muscles not only provide ankle plantarflexion, but also contribute significantly to knee extension through what is described as the plantarflexion–knee extension couple. This mechanism is load-dependent and allows the triceps surae to restrain tibial progression and promote knee stability, especially in late stance. The gastrocnemius contributes to knee extension under load despite its anatomical role as a knee flexor. Its function is altered depending on whether the limb is loaded (stance) or unloaded (swing). Soleus contributes purely to ankle plantarflexion, but also supports knee extension indirectly by stabilizing the tibia during mid-to-terminal stance. In normal gait, knee extension during stance is achieved in two phases: Initial phase is controlled by the knee extensors (e.g., quadriceps) and the terminal phases driven by the plantarflexion–knee extension couple, the soleus and gastrocnemius, supporting forward propulsion and energy-efficient gait. In CP, spasticity and weakness of the plantarflexors disrupt this mechanism. For example, weak or spastic gastrocnemius can lead to crouch gait (persistent knee and hip flexion). Excessive plantarflexor activity may cause knee hyperextension or limit ankle dorsiflexion, contributing to equinus deformity and gait instability. Patients with flexed knee gait lose the benefit of the plantarflexor–knee extension coupling, impairing stance phase stability(46).

Understanding the anatomy and physiology of the gastrocnemius complex is essential for interpreting gait abnormalities especially equinus and planning both conservative and surgical interventions in individuals with CP and other neuromuscular conditions. The impact of equinus vary across GMFCS levels. In ambulatory children (GMFCS levels I–III), equinus often presents as a dynamic deformity, leading to toe-walking and contributing to gait instability(47). Over time, if left unaddressed, it can progress to fixed contractures, further impairing mobility. In non-ambulatory children (GMFCS levels IV and V), equinus deformity can interfere with positioning, hygiene, and the ability to use orthotic devices, thereby affecting overall quality of life(48).

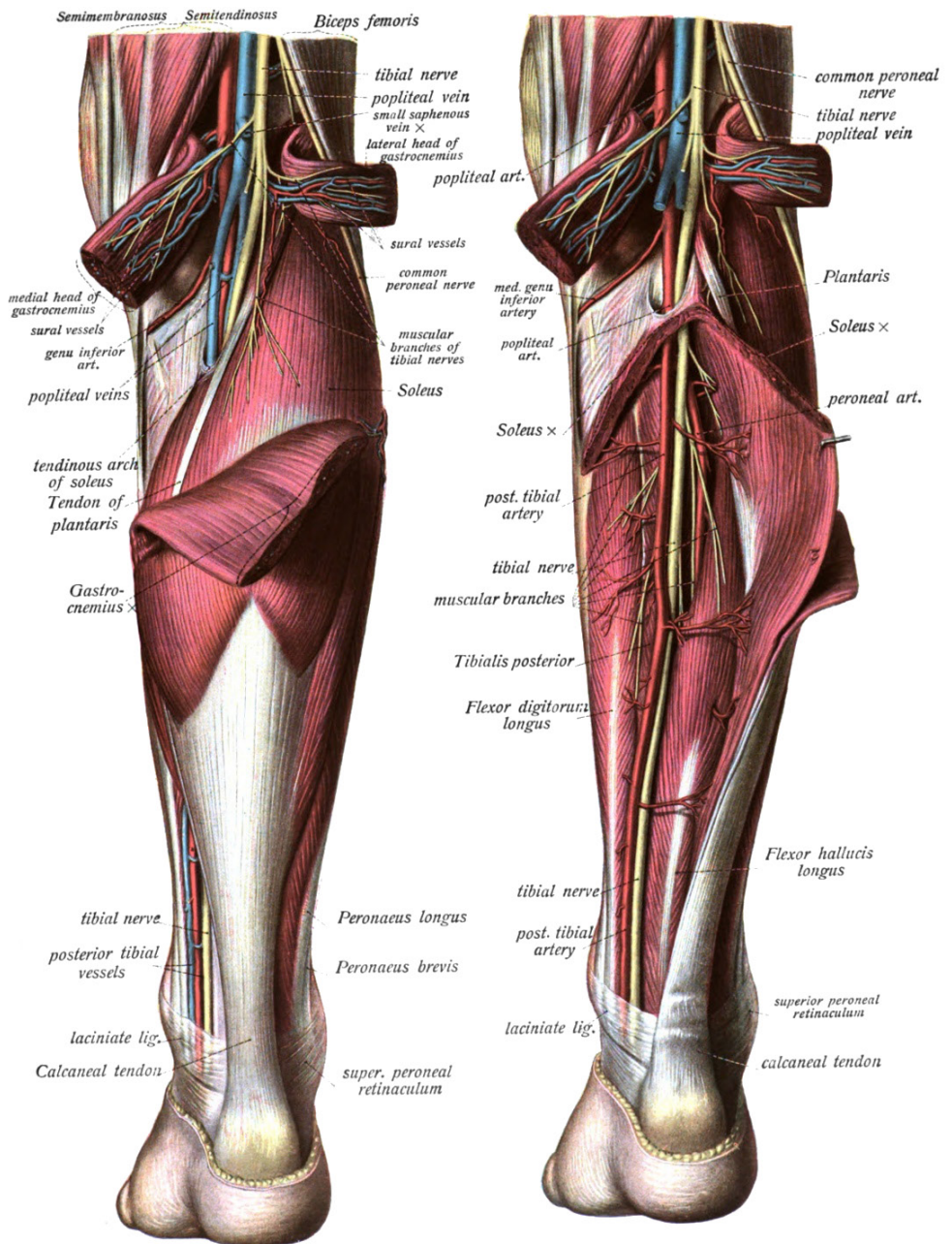


Figure 4. Anatomical illustration of the gastrocnemius complex. *Sobotta, Atlas and Text-book of Human Anatomy, 1909; Public domain.*

Treatment strategies

Physical therapy

Physical therapy is a central component in the management of CP, aiming to improve motor function and participation. Current evidence supports the use of goal-directed and functional training approaches, including gait training and constraint-induced movement therapy for upper limb function(49). Overall, the evidence base is mixed, and further research is needed to determine the most effective strategies for improving functional outcomes in children with CP(49). Passive stretching alone does not promote muscle growth or improve function in children with CP, though it may help slow contracture progression(50). The effectiveness of such interventions varies between individuals and long-term effects are unclear.

A strategy to prevent contracture development is the use of systematic positioning interventions throughout the day and night, often referred to as 24-hour postural management(51). It is demonstrated that prolonged muscle shortening, as in immobility, leads to sarcomere loss and structural muscle changes that predispose to contracture formation(52). The aims are to maintain joint ROM, promote symmetry, reduce pain and minimize prolonged positioning in deforming postures. Early and consistent use of postural support—such as standing frames, night-time splints, and adapted seating systems—to prevent contractures and improve body alignment in non-ambulant children with CP is important(53). More research is needed to clarify whether changes in muscle morphology, such as increased fascicle length, lead to functional improvements and delayed need for surgery(54).

Orthotics and serial casting

Serial casting is an intervention to gradually correct equinus deformity in children with CP most commonly using a below-knee cast but also targeting other joints. Treatment typically lasts between 2–6 weeks, with casts changed weekly to incrementally increase passive ankle dorsiflexion. A total of 2–6 cast changes is common. After casting, ankle–foot orthoses (AFOs) are often prescribed to maintain the achieved range of motion. Serial casting is effective in increasing passive ankle dorsiflexion in children with CP, when combined with BTX-A, the effect on ankle ROM may be enhanced(55, 56). While these interventions can improve gait function, potential adverse effects, such as altered knee kinematics and progression toward crouch gait, should be considered. (57). The use of AFOs is very common but varies widely between countries, despite similar healthcare systems(58). AFO use is most common in children with higher GMFCS levels and varies by CP subtype and age. There is however a lack of consensus and need for evidence-based guidelines to support more consistent prescribing practices(58).

Botulinum toxin type A(BTX-A)

BTX-A acts by blocking the release of acetylcholine at the neuromuscular junction, leading to a temporary chemical denervation of the injected muscle. This reduces muscle overactivity and spasticity, potentially improving mobility, decreasing pain, and facilitating functional movements in children with CP. The effect is transient, typically lasting 3–6 months, allowing time for concurrent rehabilitation and training to optimize treatment of contractures(59). BTX-A has become the most commonly used medical intervention for children with CP(59). However, improvements in gait function are generally small, short-lived, and vary depending on adjunctive treatments like serial casting and orthoses(59, 60). Most studies focus on tone reduction after a single injection, with limited data on functional or long-term outcomes. There are reports that BTX-A can cause muscle atrophy and replacement of contractile tissue with fat and connective tissue, raising concerns about long-term effects(61). However, when used judiciously and as part of a comprehensive treatment plan, the clinical benefits such as improved range of motion, reduced spasticity, and facilitation of functional gains often outweigh the potential risks when combined with careful monitoring and regular evaluation. Treatment patterns vary by sex, age, and gross motor function level, younger children and those with moderate to severe motor impairments are more likely to receive BTX-A(62). Injections in the gastrocnemius muscle are most common in younger children with better motor function, while treatment of the hamstrings and adductors is more frequent in older children with more severe impairments. The proportion of children treated with BTX-A to the gastrocnemius complex varies with age, peaking at the age of four to six years (Figure 5)(62).

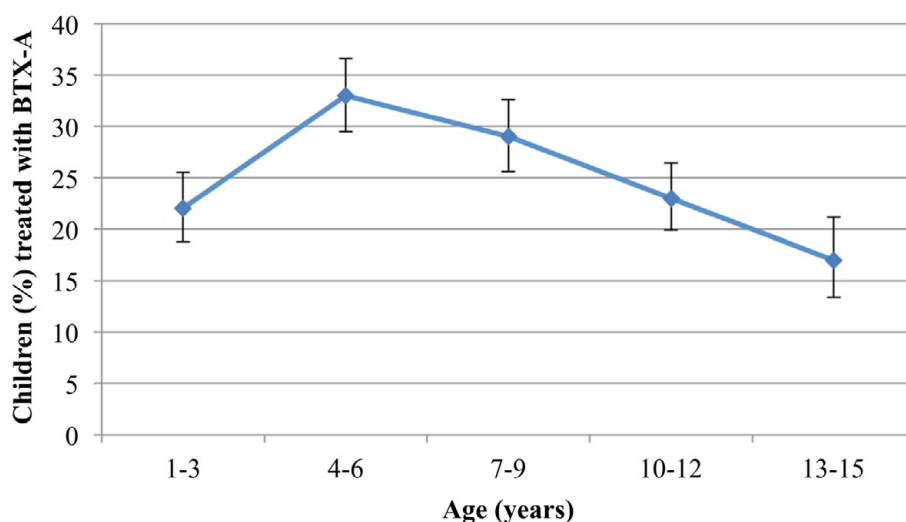


Figure 5. BTX-A treatment in relation to age. Proportion of children (%) treated with BTX-A based on age. The line segments represent the upper and lower bounds of the 95% confidence intervals. N = 3028(62).

SDR, ITB

Selective dorsal rhizotomy (SDR) is a surgical procedure used to reduce spasticity in individuals with CP, particularly when spasticity is seen as a major cause of functional limitation. Performed mostly in preschool-aged children, SDR involves selectively cutting sensory nerve rootlets in the dorsal spinal cord to decrease excessive excitatory input to motor neurons. The theoretical rationale is that spasticity results from impaired supraspinal control and increased excitability in spinal motor neurons. In CP, reduced reciprocal inhibition and altered sensory input contribute to the development of spasticity over time, as a possible physiological response to motor demands. SDR interrupts this process by cutting the overactive afferent fibers, thereby reducing spastic tone long-term. Rootlets are chosen for sectioning based on abnormal responses during intraoperative neurophysiological testing, though no standardized method exists. While some studies report positive long-term outcomes of SDR on function and activity(63), others highlight long-term limited benefits, complications, and the need for extensive rehabilitation and the risk of long-term adverse effects(64).

Intrathecal baclofen (ITB) therapy is another neurosurgical option for managing severe spasticity in individuals with CP, particularly in those with diffuse involvement and limited ambulation (GMFCS levels IV–V). ITB involves continuous delivery of baclofen directly into the intrathecal space via a subcutaneously implanted pump, enabling effective spasticity control with lower

systemic doses compared to oral administration. The treatment reduces muscle tone by enhancing GABA-B receptor activity in the spinal cord, thereby inhibiting excessive reflex activity. ITB has been shown to improve comfort, ease of care, and quality of life, though its effects on functional mobility are less consistent. As with SDR, ITB requires long-term follow-up and carries risks related to device complications and surgical procedures(65, 66)

Pharmacological treatment

Oral antispasticity medications, such as baclofen and diazepam are commonly used in children with CP to reduce generalized spasticity. Baclofen, a GABA-B receptor agonist, is the most widely used and works by inhibiting excitatory neurotransmission at the spinal level. While oral baclofen may provide relief from muscle tone and associated pain, its efficacy is often limited by systemic side effects such as drowsiness, hypotonia, and gastrointestinal discomfort. Diazepam, a benzodiazepine, enhances GABA-A receptor activity and is sometimes used for short-term relief of spasticity or spasms but carries a risk of sedation and dependency. Overall, oral medications are typically reserved for children with generalized spasticity, especially when more focal treatments like BTX-A or surgery are not appropriate or sufficient(67, 68).

Surgical gastrocnemius lengthening

Equinus deformity is often managed surgically when non – operative methods fail to achieve adequate dorsiflexion or prevent gait impairment or proper fitting and effective use of AFOs. A range of surgical techniques for gastrocnemius lengthening and tendo-Achilles lengthening (TAL) has been developed (48, 69). These procedures are commonly categorized into three anatomical zones along the posterior compartment of the lower leg, each with specific biomechanical and clinical implications (Figure 6)(70):

Zone 1 procedures, such as the Strayer and Baumann techniques, target the proximal gastrocnemius aponeurosis. These are selective to the gastrocnemius and provide modest, stable lengthening with low risk of overcorrection. They are typically indicated in cases with isolated gastrocnemius tightness (positive Silfverskiöld test) and aim to preserve plantarflexor strength(48, 71).

Zone 2 procedures, including the Baker and Vulpius techniques, address the distal part of the gastrocnemius aponeurosis combined with the underlying soleus fascia. These offer more substantial lengthening than Zone 1 but are non-selective. They are often used when contracture involves both the gastrocnemius and soleus (negative Silfverskiöld test) and are suitable for more advanced equinus deformity(70, 72).

Zone 3 procedures, such as Z-lengthening, White double hemisection(73) and Hoke triple hemisection, lengthen the Achilles tendon itself. These are non-selective, less

stable, but allow for the greatest degree of lengthening typically used in severe deformities. However, they are thought to carry a higher risk of complications such as over-lengthening, crouch gait, and recurrence(48, 74, 75).

The Silfverskiöld test is essential in preoperative decision-making, helping to determine whether the equinus deformity is due to isolated gastrocnemius contracture or involves the entire gastrocsoleus complex(44).

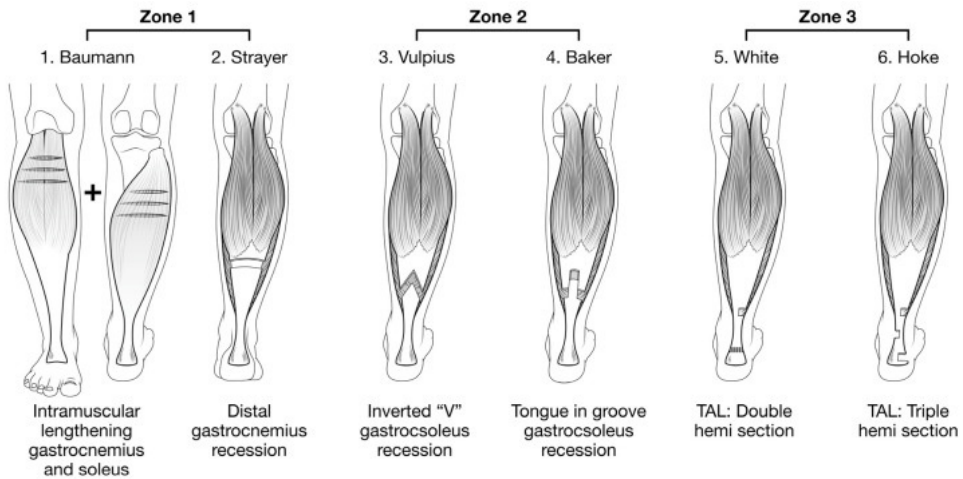


Figure 6. Surgical techniques for gastrocsoleus lengthening based on the Zonal classification of the gastrocsoleus muscle-tendon unit. Six of the more than 12 techniques for gastrocsoleus lengthening are illustrated, with two eponymous techniques shown in each Zone. Illustration reproduced by permission of Prof. Kerr Graham, The Royal Children's Hospital, Melbourne, Australia.

The purposes of this thesis

Background and aims

Study I: To monitor musculoskeletal development and guide interventions, Sweden established the CPUP surveillance program, which includes standardized assessments of spasticity using the Modified Ashworth Scale(28). Since 2005, CPUP has been a national quality register and currently includes over 95% of all individuals with CP born in Sweden from the year 2000 onward(76). Several studies have shown that spasticity seems to increase during early childhood and then gradually decline(3). A cross-sectional study of 547 children from southern Sweden showed that gastrocnemius spasticity peaked at around 4–5 years of age before declining through adolescence(77). While previous analyses of spasticity development were limited to cross-sectional data, we now performed a longitudinal analysis of the entire national CPUP - cohort, investigating the influence of age, sex, and gross motor function on the trajectory of spasticity.

Study II: Despite its widespread use, gastrocnemius lengthening carries risks, including recurrence of equinus or overcorrection leading to crouch gait(78, 79). Children with USCP are reported to be more prone to recurrence, whereas those with BSCP are at greater risk of crouch and calcaneal gait, particularly after tendinous lengthening(48, 78, 80). Additional factors reported to be associated with poorer outcomes include early surgical intervention and more severe preoperative equinus(81-84). Given the variability in outcomes and lack of consensus on optimal surgical approach, we aimed to investigate changes in ankle and knee ROM following isolated percutaneous tendo Achilles lengthening (PTAL), open tendo Achilles lengthening (OTAL), and gastrocnemius lengthening (GCL) using data from the CPUP register.

Study III: Assessing joint mobility is essential for physicians and physiotherapists when evaluating and treating these children. Early identification of a decreasing passive ROM is crucial for enabling successful treatment and optimizing long-term outcomes(26). The development of contractures can sometimes progress slowly, requiring a measurement method that is both valid and reliable. Previous studies indicate that traditional goniometry is generally reliable for measuring both ankle and knee ROM in individuals with and without spastic muscles(33, 35, 85-88), even

if some studies indicate that there is a considerable variability among children with spasticity(89). To our knowledge, no previous study has investigated children with USCP to evaluate the influence of spasticity on ROM measurement inter-examiner agreement. The aim of this study was to investigate the impact of spasticity on potential bias in goniometric ROM measurements of knee extension and ankle dorsiflexion in children with USCP by using each child as their own control, comparing measurements from the spastic limb to those of the contralateral limb.

Study IV: Although CPUP has contributed to standardized care and early identification of contracture development, annual reports from the registry have indicated persistent regional differences in how equinus deformity is managed (cpup.se). Variations may be influenced by factors such as local clinical routines, access to multidisciplinary teams, and differing interpretations of treatment indications. Our aim was to investigate regional differences in the management of equinus foot deformity in children with CP in Sweden, using data from the CPUP registry.

Methods

Study I: This longitudinal, register-based study used data from CPUP. We included all MAS assessments of the gastrocnemius muscle in children aged 0–15 years, born between January 1990 and March 2014, and reported to CPUP up to September 2016. MAS levels 1 and 1+ were combined due to the lack of evidence for an ordinal distinction between them, which meant that we used the original AS (90). Both legs were included in the analysis, except for children with unilateral spastic CP, where the side with highest AS score was selected. The analyses included all children, including those treated with BTX-A and oral baclofen, and were also repeated excluding children who had undergone SDR, ITB, or TAL.

Study II: This longitudinal, register-based study also used data from CPUP. Children born between 2000 and 2011 who underwent isolated gastrocnemius-complex lengthening between 2000 and 2014 were eligible for inclusion. Surgical data were validated against operative records, and children were categorized into three treatment groups based on the type and level of surgery: GCL (primarily Baker, Vulpius, or Baumann procedures), OTAL or PTAL. Children were included if surgery occurred before December 31, 2014 and followed until their last available CPUP assessment or the end of the study period in 2021. The study evaluated changes in ankle and knee ROM before and after gastrocnemius lengthening. Primary outcomes were ankle dorsiflexion with the knee extended, and knee extension, both measured with the child supine and the hip extended. ROM assessments were conducted using a manual goniometer in standardized positions, as described in the CPUP manual. We also examined the association between outcomes and variables such as GMFCS level, age at surgery, and CP subtype. Limits for joint motion were defined as ankle dorsiflexion $\leq 0^\circ$ or $\geq 20^\circ$, and knee extension $< -10^\circ$. Children were excluded if they underwent other lower limb surgeries, if the type of gastrocnemius lengthening could not be verified, or if pre- and postoperative ROM data were missing.

Study III: Children with USCP, aged 2–17 years, were recruited from local habilitation units. Inclusion required a confirmed diagnosis of USCP by a pediatric neurologist. Children were excluded if they had undergone lower limb surgery within the past year or received BTX-A injections to the lower extremity within the last three months. Each participant underwent a single standardized assessment lasting approximately 30 minutes. Two experienced clinicians, one pediatric orthopedic surgeon and one pediatric physiotherapist independently assessed

passive ROM of both legs using a manual goniometer. The assessors were blinded to each other's measurements. Measurements included popliteal angle, knee extension, and ankle dorsiflexion with both flexed and extended knee. Patient positioning and goniometer placement followed CPUP guidelines. Spasticity in knee extensors, knee flexors, and plantar flexors was graded by consensus using the MAS. Additional recorded data included age, sex, and side of spasticity.

Study IV: This is a retrospective, registry-based cohort study. The aim was to investigate Swedish regional variations in the treatment of equinus foot deformity in children with CP, divided into six healthcare Regions. Using data from the CPUP registry, we analyzed treatment patterns over a ten-year period from 2013 to 2023. This time frame was chosen because it reflects relatively current clinical practices while ensuring that data from all Regions are complete and comparable. Treatment modalities analyzed included BTX-A injections, use of AFO, serial casting, and TAL (including all varieties of gastrocsoleus lengthening). Data were stratified by age, GMFCS level, and CP subtype to identify possible differences in treatment approaches.

Declaration of AI usage for this thesis: ChatGPT used for language editing. Prompts and results reviewed and revised by the author, all scientific content and interpretation solely by the author.

Statistical methods

Study I and II employed mixed effects modeling as the primary analytical framework, in order to appropriately handle the hierarchical and longitudinal structure of the CPUP data. In **Study I**, a two-level linear mixed effects model was used to analyze the development of spasticity in the gastrocsoleus muscle over time. One leg per child was randomly selected to reduce dependency between observations. Age-related changes in spasticity were modeled using a linear spline with a data-driven knot at 5.5 years, allowing for distinct annual rates of change (ARC) before and after this age. The model included random effects for the intercept and ARCs, with an unstructured covariance matrix. Interaction terms were used to test differences in spasticity trajectories by GMFCS level and between children with unilateral and bilateral CP. Model comparisons were conducted using likelihood ratio tests. Assumptions of normality were evaluated using residual diagnostics, and robustness checks were performed using Huber–White standard errors and sensitivity analyses with logistic models. In **study II**, linear mixed effects models were used to assess ankle and knee ROM following isolated gastrocsoleus lengthening. The models accounted for irregular timing of assessments, missing data, and individual-level variation. Time since surgery was modeled using polynomials, and interaction terms were included to capture differential treatment

effects at 0, 5, and 10 years postoperatively. Both crude and adjusted models were fitted, with adjustment for relevant covariates such as GMFCS level, CP subtype, age at surgery, and baseline ROM, selected using the modified disjunctive cause criterion. Model selection was guided by the Akaike Information Criterion (AIC), likelihood ratio tests, and residual plots. Additionally, Kaplan–Meier survival analysis was used to evaluate secondary outcomes such as surgical revisions, recurrence of contracture or overlengthening. Due to clustering, a Cox proportional hazards model with shared gamma frailty was applied, with hazard ratios estimated both unadjusted and adjusted for confounders. The validity of proportional hazards assumptions was examined using Martingale and Schoenfeld residuals. All statistical models were fitted using maximum likelihood estimation in Stata v.14 (StataCorp, College Station, TX), and all analyses adhered to current best practices for longitudinal data modeling in clinical research(91).

For **Study III** power analysis indicated that a minimum of 32 participants (64 legs) was required to detect a statistically significant bias in ROM measurements. To evaluate whether measurement variability differed between sides, the examiner differences for the spastic and contralateral limbs were compared within each participant. The mean difference between sides (i.e., the difference in examiner agreement) was analyzed using one-sample t-tests, with significance set at $p < 0.05$. Measurement agreement between the two examiners was also analyzed using Bland–Altman plots, generated separately for the spastic and contralateral limbs, to assess systematic bias and proportional differences. Although not a primary aim of the study, inter-rater reliability was estimated using the intraclass correlation coefficient (ICC), calculated separately for the spastic and contralateral limbs. Examiners were modeled as fixed effects and participants as random effects. ICC values were interpreted according to Koo et al.(92), where <0.5 indicates poor, $0.5–0.75$ moderate, and >0.75 good to excellent reliability. All analyses were conducted using IBM SPSS Statistics, version 28.0.

For **Study IV** we employed a descriptive design to investigate regional variations in the management of equinus foot deformity in children with CP in Sweden. Data from the national CPUP register was used, covering the period 2013–2023. The analysis included treatment modalities such as BTX-A injections, AFO, serial casting, and TAL. Data was stratified by geographic Region, GMFCS levels and CP subtypes. Descriptive statistics was used to summarize the prevalence and distribution of each treatment modality across Regions. To explore potential systematic variation, chi-squared tests was used to compare categorical treatment frequencies between Regions.

Methodological considerations

All four studies relied on well-defined samples and standardized assessment methods; Studies I, II, and IV used CPUP registry data, while Study III applied CPUP guidelines in a clinical study. The registry has high national coverage and long follow-up, ensuring representativeness and minimizing selection bias. The standardized measurement protocols reduce variability, although some degree of inter- and intra-rater variation is inevitable, particularly for goniometric ROM assessments and MAS grading. In Study I, the decision to merge MAS levels 1 and 1+ reduced misclassification risk but also limited the ability to capture finer distinctions in spasticity. In Studies I and II, mixed-effects models made it possible to handle the structure of the data, differences in follow-up times, and missing values, which improves the reliability of the results, although the observational design still limits conclusions about cause and effect. In Study II, surgical data were validated against operative records, which improved accuracy, but heterogeneity in surgical technique and postoperative management may have introduced residual confounding. Study III addressed measurement error directly, but the limited sample size and restriction to unilateral CP may reduce generalizability. Study IV, with its descriptive design, cannot determine underlying causes of regional treatment variation, but provides valuable insight into real-world clinical practice. Reliance on registry data meant that unmeasured confounding and variation in local practice routines could not be fully controlled for.

Ethical considerations

All studies were conducted in accordance with the Declaration of Helsinki and approved by the regional ethics review board in Lund, Sweden (Study I and II; LU-433-99) and the Swedish Ethical Review Authority (Ref. no. 2023-00195-01 and Ref. No. 2025-00145-01 for study III and IV respectively). For Studies I, II, and IV, only de-identified registry data were used, stored securely, and could not be traced to individual participants. Study III involved direct assessments of children with unilateral CP, and written informed consent was obtained from all parents or guardians, with assent from the children when appropriate. Because children with CP represent a vulnerable group, special attention was paid to minimizing burden and ensuring voluntary participation. The studies did not involve any intervention beyond routine clinical practice, and the potential benefits of improved knowledge for future care were considered to outweigh any risks. Study IV also highlights an ethical dimension of equity in healthcare, as regional differences in treatment may affect children's right to equal access to care.

Main results

Study I: We analyzed 57,953 assessments in 4,162 children with CP. The main finding was an age-related pattern in spasticity of the gastrocsoleus muscle: an increase during the first five years of life followed by a gradual decline up to age 15 (Figure 7 and 8). This pattern remained even when children treated with SDR, ITB, or TAL were excluded. The magnitude of change was greater in children with more severe motor impairment (GMFCS III–V), and girls and children born more recently tended to have slightly lower spasticity levels.

Statistical modeling confirmed the overall trends, with an average annual increase in AS of up to 0.07 points before age five and a decrease of up to 0.08 points annually after age five.

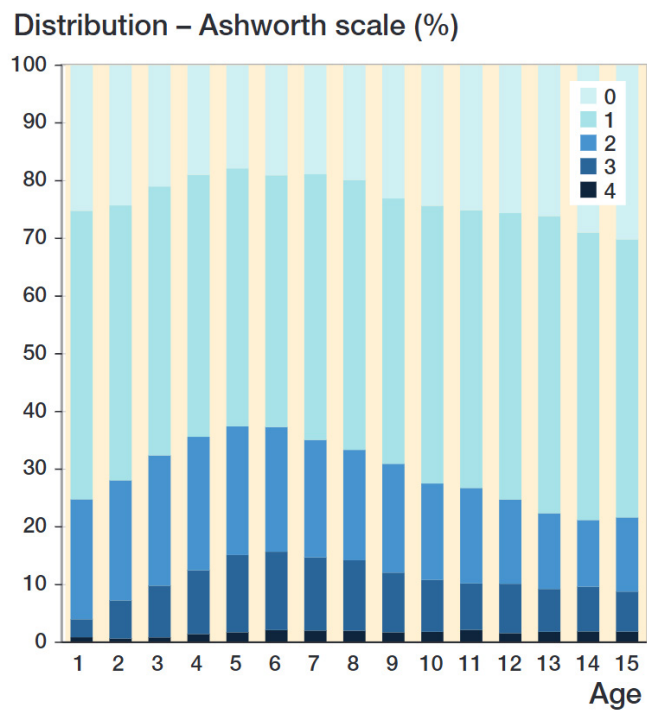


Figure 7. Degree of spasticity of the gastrocsoleus muscle according to the Ashworth scale related to age in the total sample of 57,953 measurements in 4,162 children

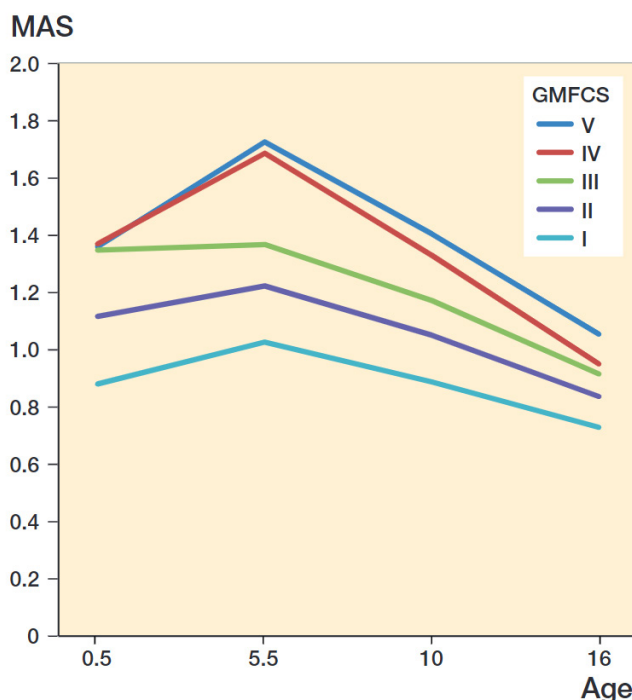


Figure 8. Prediction of the development of spasticity with age in relation to GMFCS level, using mixed-model analysis

Study II: This study included 184 children with CP who underwent isolated gastrocnemius lengthening between 2000 and 2014, with an average follow-up of 8.6 years. Most children were at GMFCS level I or II and underwent (PTAL).

Mixed effects models (MEM) showed that the mean ankle dorsiflexion improved from about -5° preoperatively to 15° immediately postoperatively, followed by a gradual decline to $0-5^{\circ}$ at 10-year follow-up across all three surgical techniques (PTAL, OTAL, GCL)(Figure 9). Differences in mean ROM between the groups were small, and confidence intervals excluded any statistically significant differences.

In contrast, survival analysis using Kaplan–Meier (K-M) estimates revealed statistically significant differences in event rates (defined as ankle dorsiflexion $\leq 0^{\circ}$ or $\geq 20^{\circ}$), with PTAL showing a lower event risk compared to OTAL and GCL. Likelihood ratio test revealed a statistically significant difference between the three surgical groups when the event was defined as ankle dorsiflexion $> 20^{\circ}$, with the lowest event rate observed in the PTAL group, followed by OTAL and then GCL.

When event was defined as dorsiflexion $< 0^\circ$, few events occurred, and no statistical comparison could be made.

Regarding knee extension, mean values declined in all groups postoperatively, particularly in children with higher GMFCS levels. Although the mixed effects model indicated a more pronounced decline in PTAL and GCL groups, survival analysis using Cox regression did not detect any statistically significant differences between the surgical techniques. Wide confidence intervals reflected low statistical precision, and a fully adjusted model was not feasible due to the limited number of events.

Both analytical approaches, MEM (mean trends) and K-M (event-based risk), highlight different aspects of the outcome. While mean ankle ROM was comparable between groups, the event-based analysis revealed relevant differences in risk, particularly favoring PTAL.

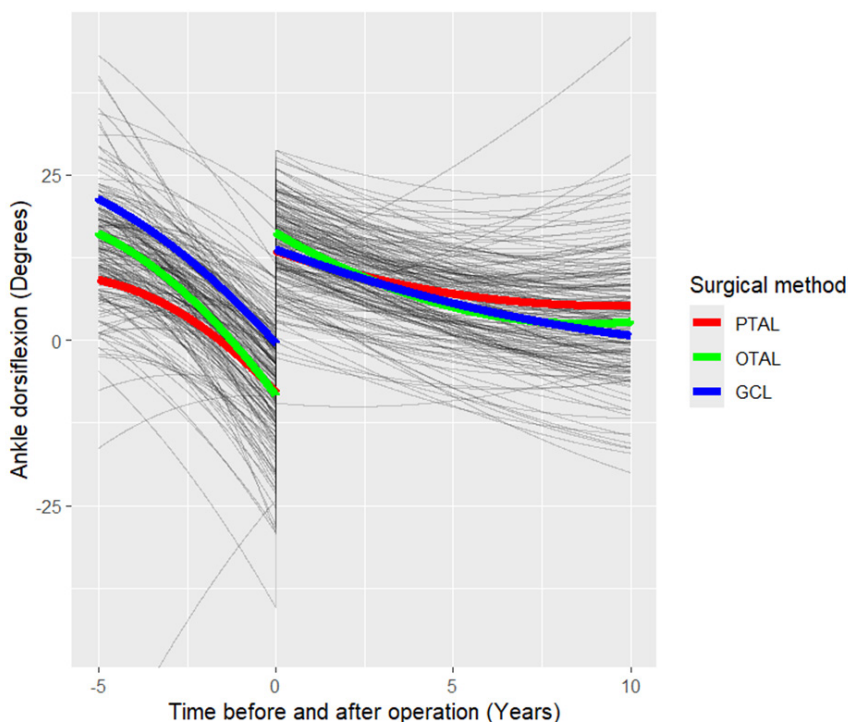


Figure 9. Development of individual and population mean ankle dorsiflexion before and after surgery produced using mixed effects regression modeling.

Study III: This study included 32 children. No statistically significant side-related bias was detected for any measurement. For example, the between-side mean difference was -0.22° (95% CI -2.53 to 2.09) for ankle dorsiflexion with the knee

extended and 0.31° (95% CI -1.63 to 1.01) for knee extension; in both cases the confidence intervals include zero, indicating no evidence of systematic side-related bias and equivalent examiner agreement.

Bland-Altman plots demonstrated small mean differences between examiners for all joint measurements, confirming good overall agreement. However, the limits of agreement varied depending on joint and limb side. The largest variation was observed for the popliteal angle and for ankle dorsiflexion with the knee flexed on the contralateral side, suggesting slightly greater variability in these less affected limbs. Although not part of the original study aim, inter-rater reliability was assessed using ICC. Across all joint measurements, ICC values indicated good to excellent agreement. The highest reliability was observed for knee extension on the spastic side (ICC = 0.911), and lowest reliability was seen for ankle dorsiflexion with a flexed knee on the contralateral side (ICC = 0.653). All ICC values were statistically significant.

Study IV: A total of 4,551 children with CP were included in the study, representing Sweden divided into 6 healthcare Regions. Nationally, 13.7 % had received TAL, 62% AFO, 9.0 % serial casting, and 31.5 % BTX-A (Figure 10). The proportion of children who had received these treatments varied statistically significantly across Regions. The proportion of children operated with TAL ranged from 10 % to 17.8 % ($\chi^2(6) = 27.3$, $p < 0.001$), AFO use from 56 % to 70 % ($\chi^2(6) = 42.4$, $p < 0.001$), serial casting from 2.5 % to 17.5 % SC ($\chi^2(6) = 126.2$, $p < 0.001$), and BTX-A treatment from 20.5 % to 39.0 % ($\chi^2(6) = 92.7$, $p < 0.001$). TAL was most commonly performed in children with GMFCS levels II–IV. The use of AFOs was highest among children with GMFCS levels IV and V. Serial casting was primarily used in children GMFCS levels I–III. BTX-A was most frequently administered to children with GMFCS levels II–IV, peaking at level III. The average age at first TAL procedure was similar across all Regions.

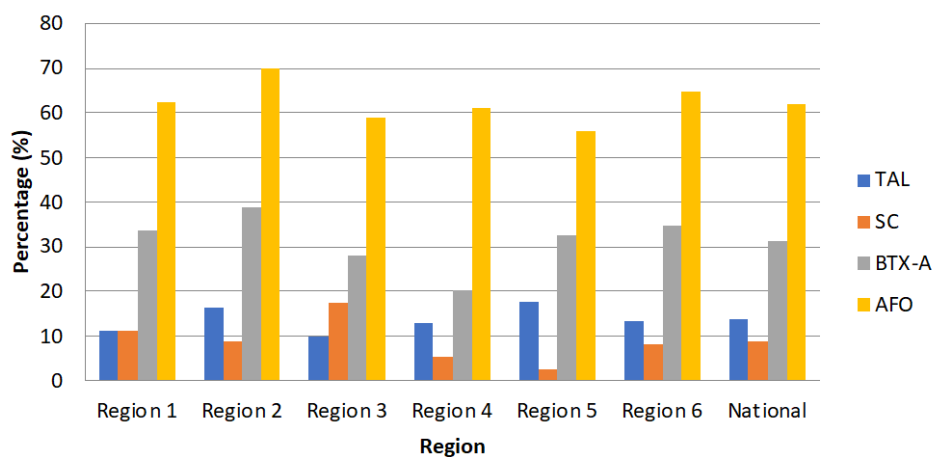


Figure 10. Proportion of children treated by Region. TAL = surgical lengthening of the gastrocnemius complex, SC = serial casting, BTX-A = botulinum toxin A injection to the gastrocnemius complex, AFO = ankle foot orthosis.

Discussion

Study I: This longitudinal study establishes that children with CP often present with normal or low muscle tone in infancy, followed by an increase in spasticity during early childhood(3, 93). The subsequent decrease in spasticity is consistent with earlier regional findings from southern Sweden(77) and resembles patterns seen after brain injury in adults, where initial spasticity peaks and then resolves as part of neurological recovery(94-96). This study demonstrate that the findings are not due to a cohort effect, but reflect the age-related development of spasticity in children with CP. A major strength of the study is the use of mixed effects modelling and its ability to account for both individual variability and overall trends in the data, making it particularly well suited for analyzing longitudinal changes in children with repeated measurements over time. Another strength is the nationwide, population-based design, comprehensive register coverage, and long follow-up.

While BTX-A was used in some children during the study period, often around the age of peak spasticity(62), its potential effect on AS measurements is likely to reinforce rather than confound the overall pattern of rising and falling degree of spasticity.

The study acknowledges limitations in the use of AS, including challenges distinguishing spasticity from contracture—particularly at higher AS scores(97) and variability in inter-rater reliability(35). However, systematic reviews have concluded that both inter- and intra-rater reliability of the AS is generally satisfactory(31), especially for the original version of the scale used in this study.

Spasticity, while potentially limiting joint range and contributing to contractures may also provide compensatory function in the presence of muscle weakness. With age and increased body weight, a reduction in spasticity may result in gait deterioration e.g., progression from toe walking to crouch gait which is consistent with longitudinal observations of declining ambulatory function in adults with CP(98).

In summary, spasticity of the gastrocsoleus muscle in children with CP tends to increase during early childhood and then decline, emphasizing the importance of age-specific treatment planning and the need for appropriate control groups when evaluating spasticity-reducing interventions.

Study II: The mean ankle and knee ROM developed similarly across all three surgical groups (PTAL, OTAL, and GCL). The largest estimated differences were approximately 5° at 5 years, close to the known measurement error for goniometry(85, 99) and not considered clinically relevant. Survival analysis showed a statistically significantly lower event rate for ankle ROM after PTAL compared to OTAL, and a non-significant trend toward lower risk compared to GCL. For knee extension, no firm conclusions could be drawn due to statistical uncertainty.

Thresholds for event definitions were based on literature describing normative values and clinical relevance(25, 78, 100-102). The 14% equinus recurrence rate in our cohort aligns with recent meta-analyses(103). No statistically significant associations were found between event rates and age at surgery, preoperative ROM, or CP subtype potentially due to limited statistical power. Prior studies show mixed results on these associations and a key limitation in the included studies was the lack of adjustment for confounding, which reduces the interpretability of observed associations. Furthermore, the common reliance on P values without reporting effect sizes or confidence intervals limits the ability to assess the precision and consistency of results(48, 81-83, 104).

Despite opinions advising against tendinous surgery in BSCP due to the risk of overlengthening and crouch gait(80), no clinically relevant differences in ankle ROM development related to surgical technique were observed. However, passive ROM alone shall not fully justify surgical selection. Decision regarding surgery should be based on a comprehensive individual assessment that includes, but is not limited to, passive ROM.

Knee ROM followed a similar trajectory as previously reported in a population-based cross sectional study from Sweden(105), suggesting that its development may be relatively unaffected by the surgical method used on the gastrocnemius-soleus complex.

We applied two statistical approaches: mixed-effect models and survival analysis, which yielded different results. While the former showed no group differences in mean ROM, the latter detected significant differences in event risk. This underscores how different methods capture distinct aspects of the outcome distribution and the importance of using multiple analytic strategies(106, 107).

Strengths of the study include the nationwide, population-based design, comprehensive surgical register coverage, and long follow-up. Limitations include the lack of functional outcome data (e.g., gait analysis), potential residual confounding, incomplete data on soleus-specific ROM, and variation in follow-up timing. These were partly addressed through modeling strategies and adjustment for GMFCS level.

Study III: The main finding was that spasticity did not introduce any clinically relevant bias in ROM measurements between the spastic and contralateral limbs in children with USCP. Examiner agreement was consistent across all measured joints, despite expected differences in muscle tone. These findings are in line with previous studies demonstrating good reliability of goniometric ROM assessments in children with CP when performed by trained examiners(35).

Bland-Altman analyses showed small mean differences between examiners across all ROM measures, although limits of agreement varied somewhat depending on joint and limb side. The greatest variability was observed in measurements of ankle dorsiflexion with a flexed knee and the popliteal angle, particularly on the contralateral side, possibly reflecting challenges in standardizing end range under less constrained conditions.

Although the study was not originally powered to evaluate inter-rater reliability, ICC values were generally good to excellent for all joint measurements. Slightly higher reliability was observed for measurements on the spastic side, consistent with previous studies evaluating joints affected by spasticity(35, 108).

A strength of the study design was the within-subject comparison, reducing confounding from age, growth, and gross motor function. However, some children showed mild spasticity in the contralateral limb, raising the possibility of undiagnosed bilateral CP in a few cases. Nonetheless, low spasticity levels in the rectus femoris and hamstrings likely contributed to high agreement in knee-related measures.

To our knowledge, this is the first study to compare examiner agreement of ROM measurements between the spastic and contralateral sides within individuals with USCP. The absence of significant side-related bias and the overall high inter-rater reliability support the use of goniometry as a valid and robust tool for clinical and research assessment of joint mobility in this population.

Study IV: Statistically significant regional variation was observed in the use of the four treatments studied— TAL, AFO, serial casting and BTX-A injections to the gastrocnemius. A reciprocal pattern appeared between the use of serial casting and TAL, suggesting differing regional preferences regarding non-operative versus surgical management of equinus deformity. This variability in clinical decision-making warrants further investigation, particularly regarding treatment indications and long-term outcomes.

The use of BTX-A did not correlate with the use of either TAL or serial casting. Despite similar overall levels of motor impairment across Regions, large differences in the use of BTX-A were observed, suggesting that access, clinical routines, or treatment culture may also play a role. The variation cannot be explained by functional level.

A limitation of the study is that treatment exposure was defined as having received the intervention during the 10 year follow up, without information on timing, dosage, or frequency. Also differences in data completeness or registration routines between Regions cannot be excluded.

A major strength of the study is the use of population-based data from the national CPUP registry with a large sample size, national coverage, and standardized classification systems (GMFCS) that enhance the generalizability and reliability of the findings. By including all children regardless of functional level or treatment status, the study provides a comprehensive overview of current clinical practice in lower limb contracture and spasticity management.

In summary the findings indicate substantial regional variation in the treatment of children with CP in Sweden. The results highlight the need for further evaluation of clinical guidelines, treatment equity, and the impact of regional healthcare structures on intervention patterns.

General discussion

The findings of this thesis reflect the complex and evolving nature of spasticity and its management in children with CP. The observed trajectory of increasing spasticity in early childhood followed by a gradual decline raises important questions about how interventions are timed and evaluated. As spasticity tends to diminish naturally with age, it becomes challenging to determine whether treatment effects—pharmacological or surgical—are causal or coincidental.

No clinically or statistically significant differences in mean ROM were found between surgical techniques. This reflects a broader challenge in CP research: many outcomes assessed in large register-based studies, including this one, primarily address the *Body Structure and Function* level of the ICF framework. While such measures are valuable, they do not fully capture what matters most to patients and families—such as mobility, independence, and participation in daily life. Function, gait, and participation outcomes, which align with the *Activity* and *Participation* components of the ICF, are often overlooked or remain unmeasured in register studies. To bridge this gap, the use of patient-reported outcome measures (PROMs) is essential. PROMs can provide critical insight into the lived experience of individuals with CP, including perceived function, quality of life, and goal attainment, and should be integrated into future research and clinical follow-up. The high reliability of goniometric measurements, even in spastic limbs, is encouraging and supports continued use in clinical follow-up. However, the precision of these tools does not solve the more fundamental issue of how to interpret change in a condition with non-linear natural history and multiple sources of variation.

Finally, the pronounced regional variation in treatment patterns cannot be explained by differences in motor function. This suggests that access to care, local expertise, and treatment culture strongly influence intervention choices. While this diversity may reflect adaptability to local conditions, it also raises concerns about equity and standardization. To ensure that children with CP receive evidence-based and timely care, there is a need for clearer national guidelines.

General conclusions

This thesis explores different aspects of the lower limb in children with CP, drawing on comprehensive data from the Swedish CPUP surveillance program. The studies collectively provide new insights into spasticity development, outcomes after gastrocnoleus lengthening, measurement reliability in clinical practice, and treatment variability across Regions.

Spasticity of the gastrocnoleus muscle typically follows a biphasic trajectory, with increasing tone during early childhood followed by a gradual decline in adolescence (Study I). This pattern, mirrors post-injury spasticity dynamics seen in adults. The increase and decline in tone were most pronounced in children with severe motor impairment (GMFCS III–V). These findings highlight the importance of age-specific treatment planning and underscore the necessity of long-term follow-up and appropriate control groups in treatment evaluations.

The average development of ankle and knee ROM was comparable across three common surgical methods—PTAL, OTAL, and GCL. Although mixed effects models did not reveal clinically relevant differences in mean ROM, survival analyses demonstrated a lower event rate ($\text{ROM} \leq 0^\circ$ or $\geq 20^\circ$) following PTAL, suggesting a potentially more favorable long-term outcome regarding ROM for this technique. This is notable, especially given traditional concerns about overcorrection in BSCP following tendinous procedures. Nevertheless, passive ROM alone is not sufficient for surgical decision-making, as functional outcomes such as gait are not captured by ROM measures alone.

No side-related bias was found in goniometric measurements between the spastic and contralateral limbs in children with USCP. Inter-examiner agreement was consistent across all joints, and ICC values confirmed good to excellent reliability, in line with previous findings. These results validate the continued use of manual goniometry in both clinical and research contexts, supporting its role in surveillance programs like CPUP.

Substantial regional variation was demonstrated in the use of four common interventions—TAL, AFO, serial casting, and BTX-A injections—despite a relatively consistent distribution of motor function levels across the country. These findings suggest that treatment decisions are influenced not only by clinical characteristics but also by local practices, access to resources, and possibly differing interpretations of guidelines. The observed variation highlights a need for further

exploration of clinical decision-making pathways and treatment outcomes across settings. Ensuring equitable and evidence-based care for children with CP requires both national coordination and transparency regarding how treatment practices are implemented regionally.

Together, these findings emphasize the value of high-quality registry data in evaluating longitudinal musculoskeletal outcomes in children with CP and to identify regional differences. They also point to the need for individualized, multifactorial decision-making in treatment planning, balancing spasticity, joint ROM, functional capacity, and child-specific goals.

Future perspectives

The studies in this thesis underscore the importance of individualized and data-driven care for children with CP. As the CPUP registry continues to expand and mature, future research can leverage increasingly detailed longitudinal data to further refine our understanding of musculoskeletal development, treatment outcomes, and variation in clinical practice.

A key area for future work is the integration of functional outcomes, particularly gait analysis, into national surveillance frameworks. Passive range of motion provides only a partial view of a child's musculoskeletal function. Objective assessments of movement, including instrumented gait analysis or wearable sensor technology, would enhance the ability to tailor interventions and evaluate their real-world impact.

The use of advanced statistical methods, including machine learning, holds promise for identifying subgroups of children most likely to benefit from specific interventions. This could support more precise treatment selection and reduce the risk of both under- and overtreatment. Furthermore, continued efforts to standardize outcome reporting, particularly with regard to thresholds for contracture, recurrence, and functional goals, will improve comparability across studies and Regions.

International collaboration across CP registries may also provide opportunities to explore variability in practice and outcomes beyond national borders. By harmonizing data definitions and sharing analytical frameworks, researchers and clinicians can address larger, more diverse populations and generate stronger evidence for best practices.

Finally, future work should explore family-centered outcomes, such as participation, independence, and quality of life. These dimensions are crucial for understanding the full impact of musculoskeletal interventions and aligning treatment decisions with the goals and priorities of children and their families.

In summary, the future of CP care lies in multidimensional, longitudinal, and collaborative approaches that combine high-quality data with meaningful outcomes, both clinical and functional.

Summary in Swedish

Cerebral pares (CP) är den vanligaste motoriska funktionsnedsättningen hos barn och innebär en livslång påverkan på muskelkontroll och rörelseförmåga. Många barn med CP utvecklar stramhet i muskler och leder, så kallad spasticitet och kontrakturer, vilket kan påverka gångförmåga och vardagsfunktion. För att följa barnens utveckling och förbättra behandlingen används i Sverige uppföljningsprogrammet CPUP, där bland andra fysioterapeuter regelbundet bedömer barnens muskeltonus och rörlighet.

Syftet med avhandlingen har varit att bättre förstå hur spasticitet och rörlighet i benens leder utvecklas över tid hos barn med CP, samt att utvärdera effekter av kirurgiska ingrepp och undersökningsmetoder.

I den första delstudien analyserades över 57 000 bedömningar från barn med CP i hela Sverige. Resultaten visade att spasticiteten i vadmuskeln oftast ökar fram till cirka fem års ålder, för att därefter oftast minska gradvis fram till tonåren. Mönstret var tydligast hos barn med mer omfattande rörelsehinder. Dessa resultat är viktiga för att bättre kunna planera rätt insatser vid rätt tidpunkt.

Den andra studien följde barn som opererats för s.k. spetsfot, där vadmuskeln förlängs för att förbättra fotledens rörlighet. Tre olika kirurgiska metoder jämfördes, och man fann att den mest skonsamma metoden – perkutant (via små hudsnitt) – var förknippad med mest önskvärd rörelseutveckling.

I den tredje studien undersöktes om spasticitet påverkar hur noggrant rörligheten kan mätas med goniometer (vinkelmätare). Resultaten visade att skillnader mellan undersökare var små, oavsett om mätningen gjordes på den spastiska eller den icke-spastiska sidan. Det stärker tilltron till denna metod även vid spasticitet.

I den fjärde studien analyserades hur behandling av spetsfot varierar mellan olika regioner i Sverige. Resultaten visade att det finns betydande skillnader i hur ofta olika behandlingsmetoder, som ankel/fot - ortoser, botulinumtoxin A - injektioner, seriegipsning eller kirurgi används – även när barnen har liknande förutsättningar. Det pekar på behovet av tydligare riktlinjer för mer jämlik vård.

Sammanfattningsvis visar avhandlingen att spasticitet hos barn med CP ofta följer ett typiskt mönster över tid, att effekten av kirurgi varierar beroende på metod, att vanliga mätmetoder fungerar väl även vid spasticitet – och att det finns regionala

skillnader i vården som behöver minska. Genom förbättrad kunskap kan behandlingen av barn med CP bli mer träffsäker, jämlik och effektiv.

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