

Novelties in the management of children with cerebral palsy

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Introduction

Cerebral palsy (CP) is a group of permanent disorders of the development of movement and posture, along with the development of spasticity, dyskinesia, and ataxia [1]. “The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behavior, by epilepsy, and by secondary musculoskeletal problems” [2]. These disorders appear early on in life (often during the prenatal or perinatal period) and may be attributed to the cerebral lesion. The estimated prevalence is of 2 per 1,000 live births.

Although clinical signs may evolve during the child’s development, cerebral anomalies are non-progressive and permanent. CP is defined by a set of clinical descriptions rather than objective, biological, etiologic, or anatomical criteria. As such, CP is a rather generic term encompassing multiple pathologies or etiologic diagnoses benefitting from similar treatment strategies, such as middle cerebral artery infarction, congenital CMV infection, periventricular leukomalacia due to premature birth or lissencephaly. The earlier the diagnosis of CP is made, the more optimal the required treatment strategy. As a result, it is important to recognize the early signs of motor disorders, especially in children with high risk of CP, in preterm births, and children presenting with prenatal neurological signs [1].

The best approach includes a comprehensive assessment of the patient to guide management and early non-operative or operative treatment strategies is the best approach to accompany a child with CP into adulthood with a better quality of life. In this domain, there are continually new clinical and technical developments throughout the world. It is in the domain of pediatric orthopedic surgery where the most significant progress has been realized during the past twenty years.

As such, in this chapter consecrated to the actualities in the management of CP, we will firstly summarize the new developments in terms of evaluation to guide treatment, followed by the novelties in non-operative and then operative management of patients with CP.

Novelties in the evaluation of children with cerebral palsy

Developed by the World Health Organization and its collaborators, the International classification of functioning, disability and health: children and youth (ICF-CY) (figure 1) was recently completed by a “Core Set” for children with CP [3].

The CIF-CY-CP may be utilized as a tool to facilitate communication between clinicians of a multidisciplinary team in the management of children with CP. The CIF-CY-CP put forth a set of concepts which include the different aspects of a patient’s functioning in daily life and their personal ambitions and aspirations. This is particularly useful to establish a therapeutic program and evaluate its effectiveness. Moreover, the CIF-CY-CP may not only be used to assess the effectiveness of the treatment plan from a clinician’s point of view, but also, an aspect which is increasingly important in modern clinical management, from the patient’s or their families’ points of view (figure 2). The CIF’s new “Core Set” for children with CP facilitate the application of CIF in everyday practice. In order to carry out this evaluation, the clinician should select specific instruments that have been recognized in the literature which could help in qualifying each of the different categories of this “Core Set”.

	Part 1: Functioning and Disability		Part 2: Contextual Factors	
Components	Body Functions and Structures	Activities and Participation	Environmental Factors	Personal Factors
Domains	Body functions Body structures	Life areas (tasks, actions)	External influences on functioning and disability	Internal influences on functioning and disability
Constructs	Change in body functions (physiological) Change in body structures (anatomical)	Capacity Executing tasks in a standard environment Performance Executing tasks in the current environment	Facilitating or hindering impact of features of the physical, social, and attitudinal world	Impact of attributes of the person
Positive aspect	Functional and structural integrity	Activities Participation	Facilitators	not applicable
	Functioning			
Negative aspect	Impairment	Activity limitation Participation restriction	Barriers / hindrances	not applicable
	Disability			

Figure 1: Overview of the ICF-CY as a classification system (World Health Organization; 2007 [4]).

By applying the “Core Set”, we can ensure a comprehensive evaluation of functioning while adjusting the individual patient’s capabilities. Both positive and negative influences on the child’s capabilities have been described [5].

Body Structures, Body Functions, Activities and Participation		ICF Qualifier ^a										
		challenge					facilitator					
		0	1	2	3	4	0	1	2	3	4	
s110	Structure of brain											
b117	Intellectual functions											
b134	Sleep functions											
b167	Mental functions of language											
b210	Seeing functions											
b280	Sensation of pain											
b710	Mobility of joint functions											
b735	Muscle tone functions											
b760	Control of voluntary movement functions											
d415	Maintaining a body position											
d440	Fine hand use											
d450	Walking											
d460	Moving around in different locations											
d530	Toileting											
d550	Eating											
d710	Basic interpersonal interactions											
d760	Family relationships											
Influence of environmental factors on functioning		facilitator					barrier					
		+4	+3	+2	+1	0	1	2	3	4		
e115	Products and technology for personal use in daily living											
e120	Products/technology for personal indoor/outdoor mobility											
e125	Products and technology for communication											
e150	Design, construction and building products for public use											
e310	Immediate family											
e320	Friends											
e460	Societal attitudes											
e580	Health services, systems and policies											
Influence of personal factors on functioning ^b		positive					neutral					negative
		+					0					-
pf	Enjoyment of participation											
pf	Coping strategies in relation to pain											

Figure 2: Example of a control list summarizing the functional profile of a child or adolescent by using the short common ICF Core Set for children and adolescents with CP by Schiariti et al. [5].

Although the basics of the CIF underline the most important parameters to assess in children with CP, a proper description of the methods to obtain said measurements is lacking [3]. In the perspective of defining the “Core Set” of the CIF-CY-CP, a systematic review published by Schiariti et al. outlined the different areas that the CIF-CY’s outcome-measures tools target [6].

A typical gait analysis session includes the most frequently assessed areas by the outcome-measures. For example, the most frequently assessed areas in body functioning were control of voluntary movement and gait patterns (64% and 24%, respectively), multiple joint movements and muscle tone (40%, described in many cases by physical examination prior to a gait analysis session).

Similarly, the most frequently assessed areas for patient activity and participation were both gait and gait over short distances (78% and 62%, respectively), running, standing, and sitting, all of which could be assessed using movement or postural analysis. Clinical gait analysis is thus well suited as a useful evaluation tool in accordance with the core concepts of the CIF-CY-CP.

In addition, over the last ten years, outcome-measures have increasingly included the patient’s or family’s explicit perspectives. Financial institutions, advocacy groups, and expert groups have called for the active participation of patients, including those with developmental disorders, in the decision-making process of their management and of research in this area [7]. In some countries, the introduction of patient reported outcome measures (PROM) is even compulsory to improve the quality of healthcare.

The selection of the appropriate tool to guide therapy and evaluate clinical outcomes is paramount and should be integrated and confronted to the “CIF Core Set” where, for example,

health-related quality of life is not included in the theoretical structure. This could be tackled by contextual factors.

Based on these two areas, this chapter will focus on the most important recent developments in clinical gait and movement analysis in children with CP, followed by the best, most recent, and most recommended PROMs.

1 – Novel technologies in clinical gait analysis

The fundamental aspects of clinical gait analysis technology have slowly evolved since its inception in the 1980s-1990s. Practically, the basic equipment remain an optoelectronic system for movement capture based on markers, force plates to assess ground reaction forces, and electromyography (EMG) to record the electrical activity of the active muscles.

Nevertheless, a series of technological advancement for capturing similar parameters have since seen the light of day, but without the constraints of a gait analysis laboratory or the use of markers. These technologies are not new in their underlying concepts – as is noted by the dates of the references used in this section of the text – but remain accessible to users via ready-to-use commercial packages.

Inertial measurement units (IMU) combine an accelerometer, gyroscope, and magnetometer. Today, all these different capture systems may be placed on a single card, the size of which does not exceed a few centimeters. This combination of capture systems, if they are three-dimensional, allows the IMU to detect its absolute orientation relative to gravity and magnetic north. IMUs that integrate a long-range wireless communication system can function outdoors and in environments of daily life. When the IMU contains storage capacities, they function autonomously for long periods of time. IMUs may be used as a surveillance method for sporting activities (figure 3) or those of daily living, for example [8,9], to determine different gait events and calculate temporospatial parameters, for example [10], or to evaluate the orientation of the different segments of the body during gait or other activities providing kinematic data [11]. However, major technical challenges (capture system glitches, anatomical calibration) and a lack of robustness (subtle changes in the magnetic field during indoor use, in a hospital environment) actually prevent the use of IMUs during clinical gait analysis when the goal is to guide surgical decision-making.

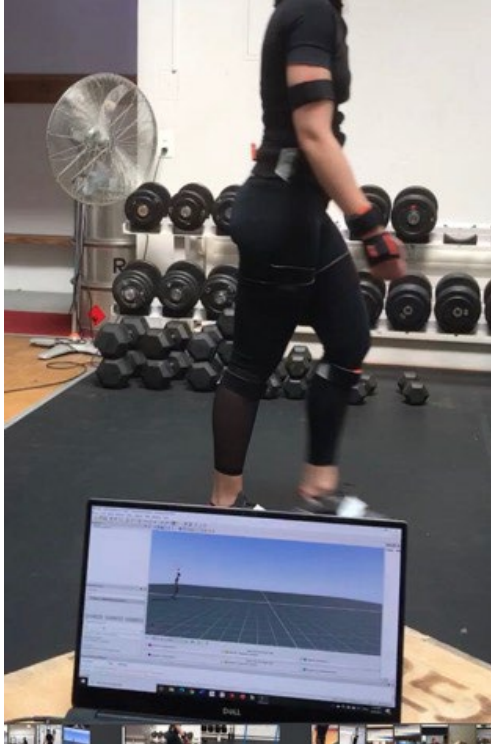


Figure 3: Data acquisition during physical activities by utilizing all parts of an IMU.

Similarly, motion detection technologies used in pressure plates can be used to estimate the vertical component of ground reaction forces required to describe the CP patient's kinetics, moments, and power, which are essential to describe both efficiency of and alterations in gait. This technology may be integrated in pressure carpets that are more practical for the recording of temporospatial gait data, for example [12] indoors, or as shoe insoles that are more practical to record data in the patient's daily environment or for a longer period, for example [13,14]. Furthermore, it is actually possible to combine these pressure detection technologies and an IMU within the same shoe insole, for example [15]. Shortcomings in pressure detection technologies include the lack of information on the components of the ground reaction forces the free moment, thus preventing calculations of inverse dynamics used in the clinical interpretation of gait for surgical planning.

Markerless motion analysis systems have progressed at a rapid speed during the last few years and may even become an adequate option for clinical gait analysis. Although the concept of markerless motion analysis is not new (for example [16,17]), the increase in calculation power, progress of conventional neural network algorithms, and the wide availability of training material, have allowed this technology to reach precision levels comparable to those of the marker-dependent optoelectronic systems [18,19]. Nevertheless, these developments are quite recent and other independent studies may be required in order to assess the precision and robustness of these systems for clinical use.

2 – Novelties in the clinical interpretation of gait

The process of interpreting clinical gait analysis data has also slowly evolved during the last few years, especially due to the development of Musculo-skeletal models allowing the estimation of biomechanical parameters that could not be directly measured [20], or the development of automated methods for the extraction of the characteristics and interpretation of gait data [21,22].

Gait kinematics, kinetics, and EMG data are at the basis of the evaluation and interpretation of clinical gait. An experienced gait analyst may extract a clear clinical image of the patient from such data, thus allowing them to determine the musculoskeletal deficiencies of the patient, that could be treated with conservative methods [23]. The objective assessment of gait improvement after treatment may also be realized through gait kinematics [24], or through a combination of temporospatial parameters and gait kinematics [25].

However, useful information remains inaccessible to direct measurement or are difficult to estimate directly from kinematic and kinetic data [26]. For example, the length of the muscle-tendon unit of biarticular muscles may be deduced from kinematic data, although this remains difficult [27].

The musculoskeletal models provide a direct estimation of muscle-tendon unit lengths based on the combination of a geometric musculoskeletal model and the patient's specific kinematic data. Ideally, the musculoskeletal model should be adapted to the patient's anatomy, but the parameters specific to muscle-tendon unit lengths are relatively impervious to missing patient-specific data, such as the precise trajectory of the muscle or torsion of a bone, while the same model and the same kinematic calculations are used [28,29]. With musculoskeletal models, the effectiveness of different treatment strategies may be quantified, such as botulinic toxin injections in the restoration of the length and velocity of a muscle while it is lengthened, as has been shown in a set of hemiparetic adults with spasticity of the right rectus femoris ambulating with a spastic knee gait [30].

The introduction of musculoskeletal modeling for the assessment of muscle-tendon unit lengths in a clinical context has led to a more conservative approach to hamstring lengthening surgeries in children presenting with a degree of crouch [31]. Data on the lengths and velocities of the different muscle-tendon units allow a better identification of the sub-groups of patients who might benefit most from certain surgical interventions such as hamstring lengthening [32,33].

Through the conventional analysis of inverse dynamics, we can calculate kinetic parameters, such as joint moments and power, based on kinematic data and subject-specific ground reaction forces. From these data, we may have an idea of the functions of the different muscle groups in both typically developing children and those with pathological gait patterns [34]. With the different approaches available today for musculoskeletal modelling, it is also possible to predict how load is shared between the different muscles during a given movement. The redundancy of the musculoskeletal system is resolved by assuming that recruitment of muscles is done in an optimal manner [26,35]. A deeper understanding of the mechanics and the roles of individual

muscles may lead to more efficient and personalized rehabilitation and treatment strategies [36,37].

The more specific role of the different muscles during pathological gait is also better estimated by induced acceleration analysis, an algorithm allowing the assessment of a muscle's potential to increase the body's acceleration, or the specific joints within the kinematic chain, for example [38,40]. Although the geometry of patient-specific musculoskeletal models affects the results of induced acceleration, the obtained clinical data, the global functional roles of these muscles in sustaining body weight or increasing the body's acceleration during gait, are relatively impervious to missing patient-specific data [28].

The musculoskeletal modeling software available today, such as OpenSim® [41] and AnyBody® Modelling System [42], may be used to model the interaction between the human body and external devices, such as exoskeletons and orthoses [43,44]. In this concept, the effect of ankle-foot orthoses (AFO) on the effective length of the muscle [45] and muscle function during gait [46] in children with CP may also be better studied. Such data may be used to devise a more personalized treatment strategy for each patient.

Applying such methods of musculoskeletal analysis in children with CP may be difficult, since the muscle recruitment strategies considered as optimal in conventional musculoskeletal models may not be applicable in CP. A previous study on muscle activation using an EMG on typically developing volunteers imitating a pathological unilateral equinus gait showed similarities with the activated muscles in hemiplegic patients with CP. Pathological muscle activation of the gastrocnemius and the soleus during equinus gait could therefore be interpreted by the biomechanics of gait while walking in equinus [47].

Additionally, orthoses, such as ankle-foot orthoses, may be adjusted to meet the specific needs of each patient. However, a proper understanding of the biomechanical role of these orthoses during gait, particularly its effect on the energetic demand of the muscles, is still lacking. Musculoskeletal modelling allows a quantification of muscle activation and the required forces and moments across joints during gait. These methods are appropriate tools to better characterize the efficiency of these orthoses in terms of muscular energetic demands (figure 4).

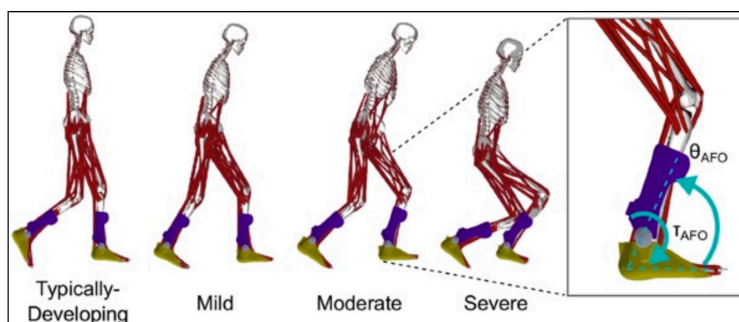


Figure 4: An understanding of the impact of an ankle-foot orthosis based on musculoskeletal modelling (image from [46])

These muscle activations, which are directly related to the measured kinematic pattern, may be predicted by conventional musculoskeletal modelling methods. Nevertheless, additional muscle activation that is not associated to a given kinematic model in optimal circumstances, such as muscle co-contractions due to apprehension and insecurity, joint instability, or pathological activations due to a suboptimal neuro-motor control, cannot be predicted with optimal motor recruitment models.

Recent advancements in modelling approaches have allowed further personalization of motor recruitment and the inclusion of an estimation of neuro-motor control specific to the patient, for example by using EMG data [48,49].

Moreover, novel approaches to predictive modelling are actually in development [50-52]. These simulations could generate new kinematic predictions without relying on experimental data, but purely on the basis of mathematical descriptions of the physiology and architecture of a muscle, the musculoskeletal geometry and the different neuromuscular control strategies. These novel approaches to modelling may allow a better understanding of the causal relationship of specific pathological mechanisms observed in patients with CP, such as muscle weakness and retractions [53,54].

The estimation of in vivo loads, such as forces acting across a joint, may also provide useful information for the orthopedic management of patients with CP. Although joint contact forces have previously been measured directly with instrumented joint prostheses [55], noninvasive options are still unavailable. Musculoskeletal modeling allows an estimation of total joint reaction forces resulting from the forces applied from adjacent segments and predicted strength of the muscles associated with a given movement [56,57] (figure 5).

For example, studies on the effects of different surgical interventions on joint loading have indicated that single-event multilevel surgery may restore normal musculoskeletal loads on the lower limbs of patients with CP [58], whereas temporary hemi-epiphysiodesis in patients with genu valgum reduces the load on the lateral compartment of the knee [59]. Furthermore, musculoskeletal modelling may provide a better understanding of the complex interactions between skeletal morphology and alignment, or muscle function and the resulting joint loads, such as in patients with patella alta presenting with crouch gait in whom the function of knee extensors may be restored by patellar tendon advancement [60,61].

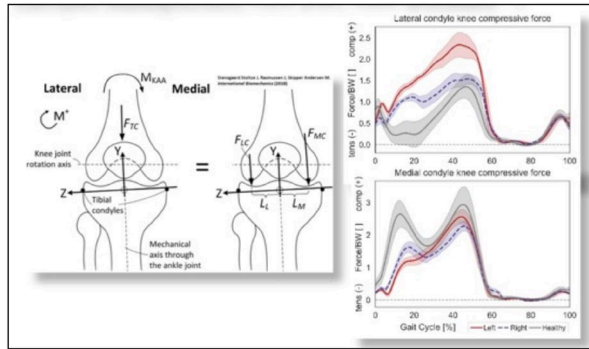


Figure 5: An example of asymmetrical articular loads as shown by personalized modelling in a patient with genu valgum (use of figure authorized by E. De Pieri).

Model geometry and muscle-tendon parameters considerably impact the estimated joint loads derived from musculoskeletal models [62,63]. The personalized geometry of these models may be achieved via different means. Many authors have shown that it is possible to create models that are entirely patient specific, for example by using an MRI [28,64,65]. However, the use of MRI in routine clinical use may not be cost-effective, although ongoing developments aim at decreasing its cost and complexity [66-68].

Other alternatives for musculoskeletal modelling do exist. Clinical gait analysis is often used in the surgical planning of patients with lower limb deformities, such as excessive internal femoral torsion or external tibial torsion [69]. In such cases, significant data may be obtained only through patient-specific bony modelling, by considering that the generic muscle trajectories follow the path of the bony deformity. For example, we have created a simplified process for patient-specific musculoskeletal modelling through the use of biplanar radiographs of the lower limbs, in order to study the effects of femoral and/or tibial torsion on hip and knee contact forces [70] (figure 6).

This process has added an additional 30 minutes to the patient's time (including transport to the medical imaging service and acquisition of the images at the service), and an additional 30 minutes for data assessment to create the patient-specific 3D bony models and identify the cutaneous landmarks in order to follow the segments of the body. Globally, this has increased the duration of gait analysis by almost a third, since clinical gait analysis previously requires around 2 hours for acquisition and 1 hour for data assessment.

An addition hour in total is extremely efficient considering the alternative being an MRI, which already requires the same amount of time only for data acquisition (considering the time required to prepare the patient and to gain access to an MRI). Furthermore, with biplanar radiographs, the motion capture markers may also be included, thus solving the problem of precisely registering the musculoskeletal model to the external markers [71].

For clinical use, this process obviously depends on the availability of biplanar radiography within proximity of the gait analysis laboratory, ideally in the same building.

Physical examination of the lower limb bony deformities may also be directly inputted to further improve the deformed generic musculoskeletal model [72]. Although there are some known discrepancies between the physical exam and the measurements that are based on medical imaging for femoral anteversion and tibial torsion [73], the global effect of these deformities on the required muscle strength and their consequences on joint contact forces provide useful information in clinical decision-making at a minimal cost.

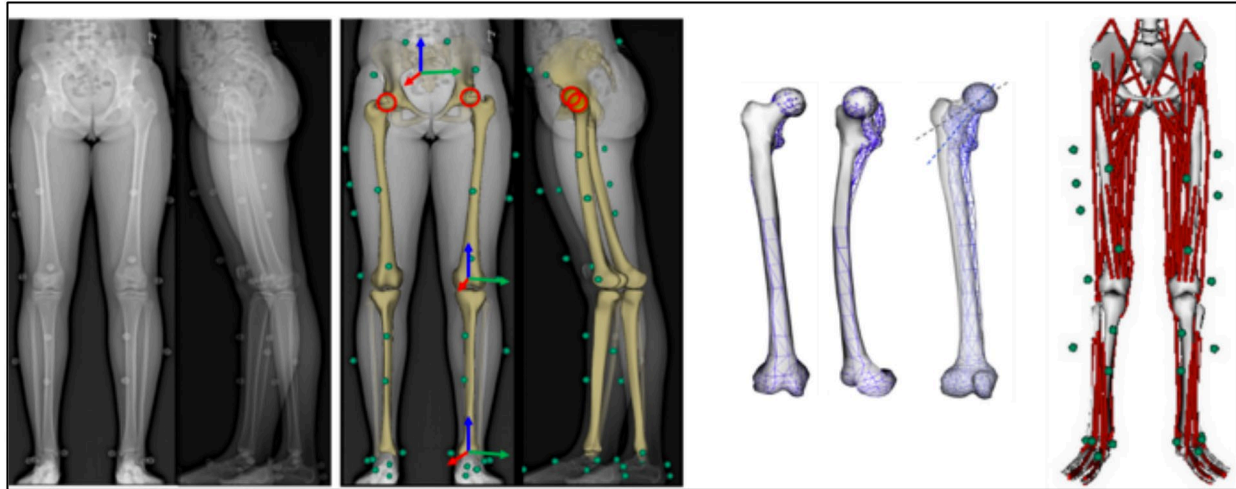


Figure 6: Pipeline to personalize the musculoskeletal model based on biplanar radiographs (adapted from [70] authorized by M. Sangeux).

Direct estimation of muscle-tendon lengths and their function during gait constitutes an undeniably significant addition to the information available for clinical decision making in patients with CP. Similarly, estimation of the in vivo musculoskeletal loads offers an innovative perspective in the orthopedic management of these patients.

The quantification of these biomechanical variables may provide clinicians with a better understanding of the pathological mechanism of an individual patient, as well as the potential effects different therapeutic strategies may have. Despite its apparent advantages, the use of musculoskeletal modelling in daily practice is still limited [20].

The actual employment of these methods is hardly simplified and requires close collaboration between physicians and engineers/modelers. The use of musculoskeletal modelling in clinical practice will thus depend on its evolution towards more simplified models with integrated patient-specific information/measurements (geometry, muscle and tendon properties, and motor control) to reliably estimate different biomechanical variables: For example, muscle activation, power, and joint loads.

3 – Patient Reported Outcome Measures (PROMs)

The assessment of quality of life integrates personal and environmental factors and has known a rapid development since the 2015 publication by the Medicare Access and Children's Health Insurance Program (CHIP) Reauthorization Act (MACRA).

Our healthcare systems should be based on values rather than volume. PROMs explore the manner with which a health issue may affect the quality of life of a patient and the extent to which treatment allows patients and their families to regain a desired level of functioning [74].

The assessment of quality of life is a part of that approach. For example, the European quality of life questionnaire KIDSCREEN may be provided to children and their parents. This could then be used to compare data between children with different syndromes and/or the general population. Furthermore, certain questionnaires such as the DISABKIDS is aimed at children with chronic diseases and includes a specific module for CP [75].

However, the noteworthy recent developments are new tools which help determine the therapeutic goals at middle and long term. Previously, the Goal Attainment Scaling method, which was adopted from the field of education, was used: the inconvenience was that the determined goals and their realization depended on the manner with which the criteria were constructed by the therapist. Whether or not said goals were reached could have been decided arbitrarily with a subject threshold that is either too high or too low.

Around two thirds of children with CP are ambulatory and undergo frequent interventions with the aim of modifying the natural history of their musculoskeletal pathology and to improve their global mobility and gait. In order to integrate the priorities and/or the expectations of children and their families, the multidisciplinary team at the Hospital for Sick Children in Toronto, Canada, has issued the Gait Outcomes Assessment List (GOAL), a novel results-evaluation method allowing the assessment of gait and functional mobility priorities of ambulating children with CP [76].

With the use of GOAL to guide treatment these children, objective clinical goals and the goals of the children's families may be aligned to improve both the outcomes and the satisfaction of interventions for children. Two versions of the questionnaire exist: Parent and child. GOAL evaluates the child's performances through 48 items (throughout the two versions) categorized into seven domains [76].

Throughout its development, and for the purposes of external validation, GOAL has been tested against multiple objective and subjective tools and scores: The Functional Mobility Scale (FMS), the Functional Ability Questionnaire (FAQ), the Gait Profile Score (GPS), and the criterion standard for measuring gait function, the Instrumented Gait Analysis (IGA), in order to cover all the domains of the CIF (figure 7).

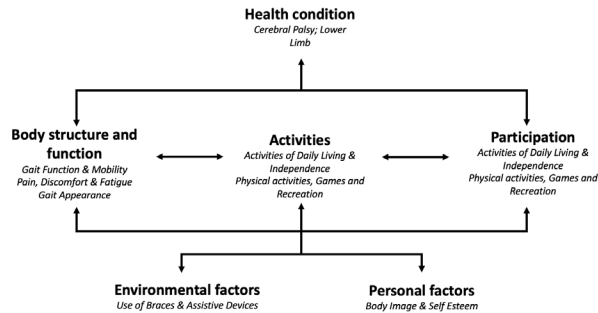


Figure 7: The Gait Outcomes Assessment List domains, according to the International Classification of Functioning, Disability and Health structure, from [76]

The Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) was developed by the same team in Toronto for the treatment of children with severe developmental disabilities and who are dependent on their parents or caregivers for a large portion of their daily needs. These children often present multiple comorbidities that may have a significant impact on their global health, comfort, functioning and quality of life.

These children undergo frequent interventions with the aim of preserving or improving their health, comfort, and quality of life and facilitate management. Evidence on the effectiveness of these interventions remain somewhat vague. As a result, this questionnaire was conceived in order to help better guide physicians when conceiving a management strategy for patients with more severe CP [77].

Validation studies have shown that the CCHILD is a reliable and valid measure of the physician's view of the patient's health, comfort and well-being. These studies have also shown the ease with which children with severe developmental disability may be treated, and that CCHILD constitutes a useful measurement of health-related quality of life in these children.

The CCHILD is made up of 37 items distributed among six sections representing the following domains: (1) Activities of daily living/personal care (nine items), (2) Positioning, transferring and mobility (eight items), (3) Comfort and emotions (nine items), (4) Communication and social interaction (seven items), (5) Health (three items), and (6) Overall quality of life (one item). A simplified version also exists for children suffering from hip disorders including 14 of the 37 original questions. Finally, in section 7, caregivers rate the importance of each of these items' contribution to their child's quality of life. The two PROMs developed for children with CP (GOAL and CCHILD) invert the traditional priorities of clinical care. Traditionally, the role of clinical care has been understood as a restoration, to the best of their abilities while considering the economical, technical, and social limitations, of an objective and externally judged "Normal function".

The role of patient-centered clinical care gives the priority to the restoration or the study of the necessary potential adaptations to allow the functions, and activities that matter most to the patient and their families. This change in philosophy allows the medical field, which is placed

under increasing economical constraints, to better align its priorities and investments on the most important domains according to the patients and their families.

Novelties in the therapeutic management of cerebral palsy

There is an increasing number of systematic reviews on the management of patients with CP. The works of Iona Novak et al. are essential for the therapeutic management of CP. A first publication in 2013, which was recently updated in 2020, constitutes the basis of evidence-based treatment of CP [78,79]. On the basis of the best available evidence, standard care of children with CP should include the following series of interventions [79]:

- Effective prevention strategies including prenatal corticosteroids, magnesium sulfate, caffeine, and hypothermia
- Effective strategies in terms allied health interventions including acceptance and commitment therapy, action observations, casting, constraint-induced movement therapy, environmental enrichment, fitness training, goal-directed training, hippotherapy, home programs, literacy interventions, mobility training, oral sensorimotor, oral sensorimotor plus electrical stimulation, pressure care, stepping stones triple P, strength training, task-specific training, treadmill training, partial body weight support treadmill training, and weight-bearing
- Effective medical and surgical strategies including anti-convulsant medication, intrathecal baclofen, bisphosphonates, botulinum toxin, botulinum toxin plus occupational therapy, botulinum toxin plus casting, diazepam, dental care, selective dorsal rhizotomy, scoliosis correction, hip surveillance, and umbilical cord blood cell therapy.

All therapists and physicians who are involved in the management of children with CP should be regularly updated on the latest evidence and developments. The management of CP is thus the most innovative and most advanced when compared to other domains of pediatric orthopedics.

1 – Novelties in non-operative management

Very recently, in December 2021, the Haute Autorité de Santé published a set of best-practice recommendations based on conclusive data in rehabilitation and readaptation of motor function in patients with CP. These recommendations should be well-known in order to better guide therapists treating patients with CP (figure 8).

These recommendations provide the proper indications and guide therapeutic management, identify the patients requiring specific treatments (age, severity, etc.), and specify the purpose of innovative interventions.

Evaluation of the clinical outcomes is recommended, and a table prioritizing interventions into three levels is suggested [80].

Rehabilitation interventions	Priority		
	Children 2-12 years	Adolescents 12-18 years	Adults >18 years
Conventional functional rehabilitation			
Passive mobility: passive nocturnal positions	2	2	3
Muscle reinforcement	1	1	2
Aerobic exercises or cardiopulmonary training	1	1	1
Biofeedback exercises	3	3	3
Walking exercises	1	1	1
Backward-walking exercises	2	2	3
Treadmill	2	2	2
Ankle foot orthosis for foot and ankle motor deficits	2	2	2
Ankle foot orthosis for equinus gait	1	1	2
Intensive rehabilitation programs			
Constraint Induced Movement Therapy (CIMT)	3	3	3
Modified Constraint Induced Movement Therapy (mCIMT)	3	3	3
Hand Arm Bimanual Intensive Therapy (HABIT)	1	1	3
Hand Arm Bimanual Intensive Therapy Including Lower Extremities (HABIT-ILE)	1	1	3
Adapted physical activity			
Physical activity	1	1	1
Sports	1	1	1
Balneotherapy	1	1	1
Hippotherapy	2	2	2
Robotics and/or computer assisted rehabilitation			
Interactive video games	2	2	2
Virtual Reality-based therapy	2	2	2
Rehabilitation based on other approaches			
Mirror therapy	3	3	3
Patient and family education	1	1	1

Figure 8: Prioritization of rehabilitation interventions; * 1 = priority; 2 = secondary priority; 3 = non-priority, from HAS 2021: Rehabilitation of musculoskeletal motor function in individuals diagnosed with cerebral palsy [81]

Concerning the non-operative management of CP, recent innovations are proposed in regular therapy, on the one hand by including intensive workout programs, on the other hand by extending the indications of robotics and device-assisted rehabilitation.

1.1 Functional training programs

For many years it was believed that muscle reinforcement increased spasticity and was contraindicated in children with CP. However, recent studies with a high level of evidence showed that physical training might increase the strength of these children with CP without increasing spasticity or other undesirable effects [82-84]. Today, rehabilitation is one of the primary pillars

in pediatric intervention and could modify the prognosis of these children and result in significant clinical differences in terms of invalidity and participation [85].

In order to be efficient, resistance during strength training should be progressively increased. To this end, the most frequently used training method is progressive resistance training (PRT) [86]. Children exercise with individualized weights that are later increased progressively depending on the child's performance. Although PRT increases muscle strength in children with CP, studies on its functional advantages are contradictory [82,87].

Numerous daily activities require rapid movement of the limbs with contraction times of 50-200ms [88]. Knowing this, rapid strength development is clinically pertinent in children with CP, and it is essential to establish interventions designed to help the child produce quick movements rather than simply increasing strength, and to produce functional movements for daily living. One study by Schranz et al. (2018) [89] examined a home-based rehabilitation program in children with CP: a PRT and high intensity circuit training (HICT). In both groups, children showed functional improvements that were specific to the intervention, although strength was only significantly increased in the HICT group. The average training time was shorter in the HICT group. The authors found excellent and comparable observance in both groups and concluded that home-based HICT may be a preferable and efficient rehabilitation method in children with CP with high functional potential.

In order to increase the strength of the lower limbs and functional performances of children with CP, a pilot study was devised by Blundell et al. (2003) [90] including eight children aged 4 to 8 years old. The intervention consisted of four weeks of after-school exercises, two times per week for one hour and included circuit training in groups. Thus, every workstation was specifically equipped for intensive training (including a treadmill, step-ups, sit-to-stands, and a leg-press). This short program of task-specific rehabilitation exercises and training for children with CP has increased muscle strength and functional performances, which have shown lingering benefit over time.

In 2019, Guedin et al. [91] published a study on high intensity weight training in cerebral palsy. The authors studied 18 patients with CP aged between 8 and 20 years old. The experimental group underwent high intensity weight training of 20 minutes with functional exercises twice a week for 3 months. Parameters were measured (muscle endurance, walking speed, Gross Motor Function Measure [GMFM]) and the Kidscreen 27 quality of life questionnaire was obtained before and after high intensity training. After 3 months, a significant improvement in performances (muscle strength, endurance, and function) was reported compared to the control group. Similar tendencies were also found for quality of life.



Figure 9: Functional training in children with CP (use of image authorized by M. Kläusler).

In light of these results, functional strength training with elements of high intensity weight training in children and adolescents with CP seems to be a promising avenue to increase performances, quality of life, and participation (figure 9).

1.2 Robotics and device-assisted rehabilitation

The above-described exercises aim at improving the development and function while concentrating on the adaptation of the neural system [92]. However, the severity of the motor and neurological dysfunction in a portion of patients with CP, especially those with GMFCS IV and V, limits the applicability of these training modalities. These exercises usually require refined voluntary motor capabilities and a solid cognitive comprehension to follow instructions and execute the exercises [93].

Vibrotherapy (VT) is an easy and time-efficient alternative intervention that has recently gained popularity [94]. Mechanical oscillations act on the neuromuscular structures. The parameters of these vibrations depend on the frequency (number of completed cycles per second, 5-200Hz), the amplitude (vertical displacement of 0.5-10mm), and the type of VY (lateral and vertical sinusoidal) [93].

Focal vibrations act directly on the muscle belly or the tendon [95], and whole body vibration (WBV) are transmitted indirectly through the whole body [96].

The feasibility and effectiveness of these two methods do not rely on the subject's movement capabilities, health, or mental state. No motor prerequisites or cognitive abilities are necessary. In a systematic review, Ritzmann R. et al. showed that VT has several benefits at both the short and long terms [93]:

- A single acute session of VT reduced reflex excitability, muscle tone, spasticity, and coordination deficits and had a positive influence on GMFM, muscle strength, gait, and mobility in children and adults with CP.
- The chronic effects of VT based on a number of randomized controlled trials include significant advantages in terms of spasticity, GMFM, muscle strength, gait, mobility, and muscle mass.
- Certain interventions (weight training, stretching, and physiotherapy) may require previous VT sessions, and may provide better motor control for certain tasks (crouching, walking) undergone immediately after VT. It may also be used as a training method in and of itself, leading to better motor performances after a minimal intervention period of 3 weeks.

The included studies also had their limitations. There is significant variability between subjects with CP in terms of classification, affected brain areas, and handicapped body regions, as well as their responsiveness to nonpharmacological interventions. Their impact on spasticity and movement biomechanics, as well as adverse effects entailing seizures and hypotension, most probably influence the effects of VT exercises. Further research is required on the choice of VT parameters and the effects of VT on postural control.

Nevertheless, according to the research data, CP helps patients with CP by facilitating the quality of the movements and has a potential effect on bone mass [97].

2 – Novelties in operative management

Surgical techniques and their indications in CP are constantly evolving. The most recent developments concern treatment of CP children with both unilateral and bilateral equinus gait patterns, based on an analysis of the stability and pathophysiology of the increased muscle tone of the lower limbs.

Moreover, surgical management of the hips and the spine in children with severe CP has also changed in terms of integrating functional treatment.

2.1 Management of ambulation and instability of equinus gait

The most common deformity encountered in children with CP is pes equinus [98,99]. The primary mechanism of this deformity is due to the persistence of primitive reflexes, inducing hyperactivity of the calf muscles. This also leads to global instability in the standing position and during gait, with increased muscle tone of the entire lower limbs, and a disequilibrium between muscle growth and bone in which bones grow while muscle length remains the same. Particularly, in patients with spastic hemiplegia, ankle extensor weakness also plays an important role.

Concerning therapy, two different methods of treatment exist: conservative or surgical. Conservative treatment consists of physiotherapy, foot-ankle orthoses, casting, and botulin toxin A injections. A common problem in these patients is the development of internal rotation,

flexion, and adduction of the hips, as well as knee flexion contractures of the affected limb. A preliminary study on a retrospective cohort undergone by Brunner et al. [100] has evaluated the effects of restoring gait with a proper heel strike with functional bracing on the passive internal rotation of the hip. The affected foot was maintained in a correct anatomical position to realign the leg with the direction of walking. When a contracture of the gastro-soleus complex was found, a heel insole was inserted to compensate for the equinus and ensure better contact between the heel and the ground.

This study showed that patients who were able to correct their gait through insoles (patients with a proper heel strike [with antegrade balancing] and having worn their orthoses for at least eight hours a day for one year) showed an almost symmetrical internal rotation of the hip compared to patients still walking in equinus, who showed a more asymmetrical passive internal rotation of the hip.

This retrospective cohort gives new and interesting insights into conservative treatment for patients with equinus gait. The primary goal of treatment should not be to obtain a plantigrade foot, but to restore a proper heel strike during gait, by compensating for equinus if necessary, and accepting a later correction of the equinus deformity.

As such, deformities at the level of the hip and, in the best of cases, the knee, are avoided and global muscle tone is decreased. As a result, the child could train toward a developing a normal gait pattern.

The risks of surgical lengthening of equinus are recurrence and, even worse, loss of functional performance due to the weakening of important muscle groups, such as the gastro-soleus complex for propulsion and hamstrings for hip extension. The consequences of such complications may be a talus deformity and the development of crouch gait.

As a result, in standard clinical practice, in order to assess the risk deterioration, gait analysis should be utilized preoperatively, the eligible muscles be injected with botulinic toxin A, and, 6 weeks after injections, gait analysis be repeated.

Muscle weakening from botulinic toxin injections may reveal the limited stability and global muscle weakness that are compensated by increased muscle tone in the injected muscle groups (figure 10).

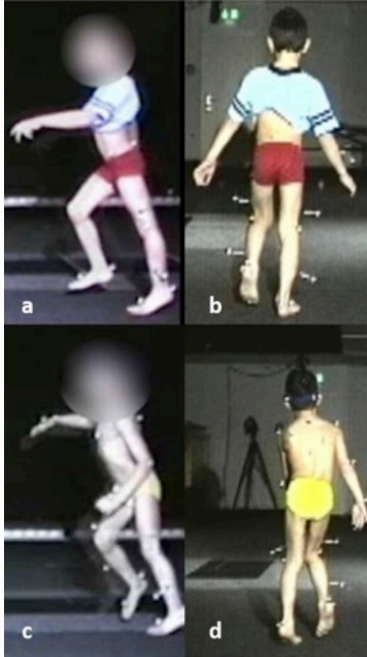


Figure 10: Destabilization of the gait pattern after preoperative Botox injection at the level of the hamstrings. a+b before injection, c+d after injection showing increased knee flexion and internal rotation due to instability (use of image authorized by R. Brunner)

Surgical management includes different types of interventions aiming at resolving the problem of spastic equinus. In most cases, surgical treatment primarily consists of lengthening the gastro-soleus muscle-tendon complex [101-104]. In their systematic review, Shore et al. [105] gathered the results from ten procedures that differed by anatomical zone. Zone 1 is from the gastrocnemius origin and ends at the most distal fibers of the medial belly of the gastrocnemius. The operative procedures in Zone 1 include the Baumann and the Strayer distal gastrocnemius recession. Zone 2 is from the distal gastrocnemius belly to the end of the soleus muscle fibers and includes the Baker and Vulpius gastro-soleus aponeurotic lengthening. Zone 3 is the Achilles tendon and includes all forms of lengthening of the Achilles tendon, including open Z tendo-Achilles lengthening (TAL) and heel cord advancement (HCA).

The primary goal of the systematic review by Shore et al. was to examine the evidence related to the surgical management of the equinus deformity in CP by comparing 35 articles. The study suggested that the two most important factors in determining the outcomes of surgery in an equinus foot in CP were age and CP subtype (hemiplegia or diplegia [105]).

However, this study also revealed that the literature was full of retrospective case series, without any control groups and without randomized control trials, and with heterogeneous patient groups with a short-term follow-up. As such, it was not possible to determine the actual rate of equinus or talus recurrence, which lead to the authors concluding that insufficient evidence existed to prefer one type of surgical intervention over the other. Nonetheless, a higher incidence of recurrence of equinus was found in children with hemiplegia, and a higher incidence of altered

gait patterns and crouch gait in children with diplegia, especially after interventions targeting the Achilles tendon [105].

The study by Rutz et al. [106] showed that spasticity of the gastro-soleus complex in spastic equinus may be reduced significantly by tibialis anterior tendon shortening (TATS) along with a TAL. Reduced spasticity of the tibialis anterior muscle was found in patients with hemiplegia. During clinical evaluation utilizing manual muscle testing, no differences were seen in muscle strength of the gastro-soleus and tibialis anterior muscles in the short term. In addition, postoperative, 27 of the 29 patients showed an active dorsiflexion of the ankle that was absent preoperatively [106]. This was the first study that utilized the Movement Analysis Profile (MAP) and the Gait Profile Score (GPS) [24] to measure surgical outcome in the treatment of pes equinus in children with CP. The most significant limitation of this study was the limited follow-up period (only 14 months). In 2016, Tsjang et al. evaluated the results of associating TATS and musculo-tendinous lengthening of the calf in patients with spastic equinus. This procedure showed favorable outcomes in terms of foot positioning during gait [107]. They concluded that TATS, combined with triceps surae lengthening, is recommended for the treatment of spastic equinus in CP. They also recognized the limitations of the study, since it included only a short-term follow-up of a heterogeneous patient population without a control group, and all 19 subjects had undergone additional surgery [107].

A very recent retrospective study by Dussa et al. (2021) [108] evaluated the efficacy of combining TATS and triceps surae lengthening on ankle dorsiflexion in the swing phase and the position of the foot in the sagittal plane during gait by comparing it to only triceps surae lengthening. The study did not show advantages for the combined procedure compared to lengthening only the calf muscles. Triceps surae lengthening included aponeurotic lengthening of gastrocnemius muscle, or a Strayer or Z-lengthening of the Achilles (figure 11), depending on the fixed degree of equinus contracture. If a TATS had been done, it was decided without being based on pre-established criteria, while the TAS (figure 12) in the patient group from Rutz et al. or Kläusler et al. were realized in combination with TAL.

Kläusler et al. published a long-term retrospective study (5.8 years) to verify the effectiveness of combining TATS with TAL in CP patients with GMFCS levels I and II [109]. The study showed that, after the intervention, all patients were capable of walking without an AFO, and that all patients were able to actively dorsiflex the ankle after surgery. The study had multiple limitations: A small sample size (23 patients), lack of a control group (non-operative patients or patients operated only with a TAL), loss to follow-up of 3 patients (excluded due to recurrence), the results were significant only for one sub-type of CP, and no measures of quality of life or function for these patients was included [109].

Further research is paramount including more homogeneous patient groups, even though the results are promising, especially considering this requires only little supplemental effort on the physician's behalf (additional 20 minutes of surgical time, without significant cost increases for the implants) and an identical postoperative treatment (6 weeks of casting, followed by 6 weeks of rigid orthoses, and 3 to 6 months of hinged orthoses), such as in isolated Achilles lengthening.

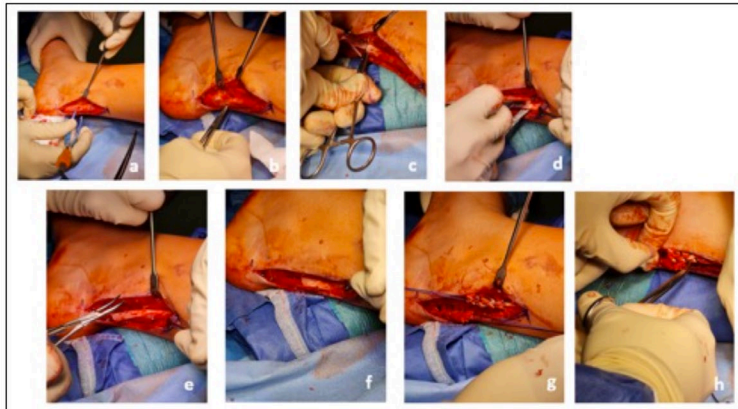


Figure 11: Modified Achilles tendon lengthening technique: a- tendon dissection, b- tendon separation, c- distal incision, d- proximal incision, e- level of lengthening, f- regulation of length, g- preparation for reinforced suture, h- suturing under tension with the ankle at 90 degrees (authorized by E. Viehweger).

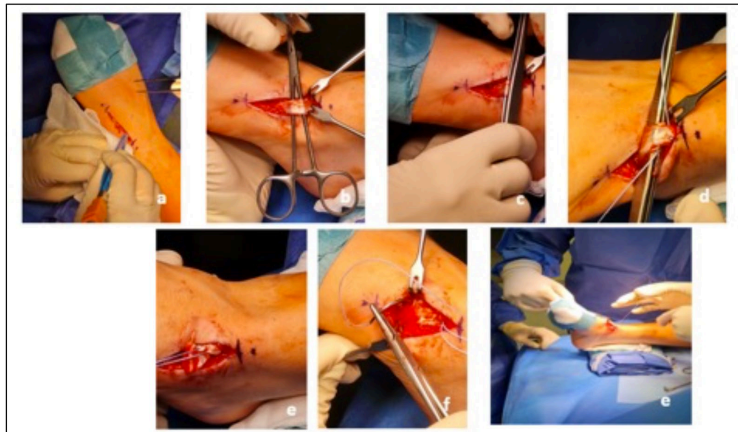


Figure 12: Modified tibialis anterior shortening technique: a- level of approach, b- tibialis anterior isolation, c- tendon rolling on clamp, d- insertion of fibertape-style non-absorbable suture, e- tendon plication, f- transosseous suture, g- tibialis anterior tendon shortening with free foot with the aim of maintaining the ankle at 90° (authorized by E. Viehweger).

2.2 Surgical treatment strategies in hip dislocation and spinal deformity in patients with severe CP

On the basis of integrating goal-oriented management strategies, surgical management of hip dysplasia and of spinal deformities in children with severe CP was recently updated and is based on functional treatment.

In young children, hip dysplasia must be diagnosed early on, either by a validated surveillance program, or regular and comprehensive follow-ups integrating neuro-orthopedic surgeons early on, at least in order to establish a common therapeutic project.

The goal is to maintain the pelvis horizontal and the hips mobile to allow a comfortable and painless sitting position in adulthood. This position also implies a decrease in global muscle tone and easier access to daily care.

The treatment of spasticity with targeted botulinic toxin (Btx) injections should be suggested early on, around the age of 24 months [110]. Similarly, in case of subluxated and unbalanced hips, with muscle contractions in muscles groups that play a significant role in the pathophysiology of the CP hip, such as the psoas, the hip adductors, or the hamstrings, selective tenotomies and Botox injections may be suggested and have shown good results when associated with prolonged positioning in adapted orthoses. The results are even better if the hips are initially mobile, notably with good external rotation.

Nevertheless, once underlying structural deformities take place, and the hips become stiff in internal rotation, the association of Btx and soft tissue procedures will become insufficient [111]. If hip reconstruction is indicated, a femoral varus and derotation osteotomy, with or without a pelvic osteotomy should be performed. In early cases with isolate femoral dysplasia (excessive anteversion, coxa valga), isolated femoral corrective procedures may be sufficient, associated with soft tissue procedures and Botox injections aimed at balancing the pelvis. Pre-operative CT-scans may help in describing the type of dysplasia [74].

Intra-operative arthrography may also help in describing the acetabular dysplasia and the position of the labrum once the femoral head is repositioned. If the femoral head is not properly positioned, capsulotomy and acetabular revision surgery may be required.

To correct acetabular dysplasia, the actual gold standard in treatment consists of a Dega or Pemberton acetabuloplasty. At the end of the procedure, the hip should be inherently stable. Postoperative immobilization should be ensured in abduction with a soft lower-limb brace (figure 13) for three weeks, followed by night-time braces, along with a wheelchair for at least one year. Immobilization by casting is only indicated in rare cases with osteoporotic instability. Particularly, immobilization in extreme abduction is no longer recommended. When increased muscle tone disrupts pelvic equilibrium and the symmetry of the sitting position, the muscle tone should also be addressed in the postoperative setting.



Figure 13: Postoperative immobilization after CP hip reconstruction

Even in case of late painful and severe dislocations, palliative surgery, such as femoral head resection, should be avoided and late reconstruction of the hip, or hip arthroplasty should be considered. Hip reconstruction may improve pain and function, even if the hip is incongruent in the immediate postoperative period. Long term studies have shown considerable plasticity of the deformed femoral head until adolescence [112].

Palliative management strategies have shown good outcomes only in hips that remain mobile without impingement on the pelvis. Periarticular soft tissue procedures associated with two or three Botox injections may decrease pain and increase quality of life in patients with a subluxated hip in the mid-term.

In case of a significant scoliotic deformity found in conjuncture with a dislocated hip, there is actually a standardized reasoning protocol on the orders of surgical treatment. If the dislocated hip is relatively mobile and pain free, spine surgery should be performed before the hip is treated, thus balancing the pelvis. The family should be informed that hip degeneration may take place in the coming months due to the redistribution of the strains on the hip [113].

However, in the case of a fixed, non-mobile, and anteriorly dislocated hip, or those with a windswept deformity, correcting the spine first with fixation of the pelvis may render the seated position either impossible or may require a very asymmetrical sitting position. Operative management includes first reconstructing the hip to improve mobility and stability, to treat contractures, and to correct the anatomic deformity.

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