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Development and evaluation of an evidence-based and individually defined physical therapy approach for ambulant children with bilateral spastic cerebral palsy

Dissertation to obtain the degree of
Doctor in Rehabilitation Sciences and Physical Therapy

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General introduction

1. Background of the research project

Children with bilateral spastic Cerebral Palsy (CP) present with a varying degree of lower limb impairments, including increased muscle tone and weakness. These primary problems generally cause secondary impairments of muscle contractures and bony deformities and result in functional problems of gait and gross motor function.[1] Treatment of children with bilateral CP is usually situated within a multidisciplinary context, including physiotherapy as a key component. [2] Controlling primary problems and prevention of secondary problems are thereby overall treatment aims to improve, stimulate and optimize motor function.[3]

The International Classification of Functioning, Disability and Health (ICF) provided a useful framework to categorize assessment results and treatment approaches.[4,5] However, development of this framework has also confronted the paediatric physiotherapist with the heterogeneity and the complexity of the pathology. It thereby opened many discussions regarding the entry point in treatment. The interactive dynamics within the ICF suggests that a combination of interventions targeting the different levels of the model may be required.[5,6]

In the last decades, research in physiotherapy treatment of children with CP has increased remarkably. Many intervention studies so far have evaluated the effectiveness of individual techniques with promising results.[7,8] However, as highlighted by the ICF, daily clinical practice most likely requires a mixture of individual techniques, each targeting a specific problem. Still, there is lacking insight in the effectiveness of multifaceted physiotherapy approaches tailored to the individual needs of the child and thereby addressing more adequately the heterogeneity of the pathology.

Therefore, the current doctoral project will focus on an evidence-based, individually defined and targeted physiotherapy approach for children with bilateral CP.

The first part of the general introduction describes the definition and classification of CP with specific attention to the prevalence and clinical presentation of bilateral spastic CP. The second part of the introduction elaborates on problems of gait and gross motor function of these children and the rationale behind related treatment. Finally, the aims and outline of this doctoral thesis are described.

2. Bilateral spastic Cerebral Palsy

2.1 Definition, classification and prevalence

Cerebral Palsy (CP) describes a group of permanent disorders of the development of movement and posture causing activity limitations, which are attributed to non-progressive disturbances in the developing fetal or infant brain. [9,10] With an incidence of 2-4 per 1000 newborn children, it is the most common cause of physical disability in childhood.[1] As stated in the definition, CP is considered as an umbrella term, covering a wide range of clinical presentations and degrees of severity. Therefore, many research groups have attempted to categorize children with CP, with different purposes, such as description, prediction, comparison and evaluation of change.

In 2000, the Surveillance group of Cerebral Palsy in Europe (SCPE) reached a consensus on the definition, classification and description of children with CP.[11] The SCPE thereby identified wide variations in the use of terms such as spastic diplegia and quadriplegia and therefore, proposed a more reliable system (**Fig 1**). This innovative hierarchical classification system differentiated the subtypes of children with spasticity into only two categories, namely bilateral and unilateral involvement.

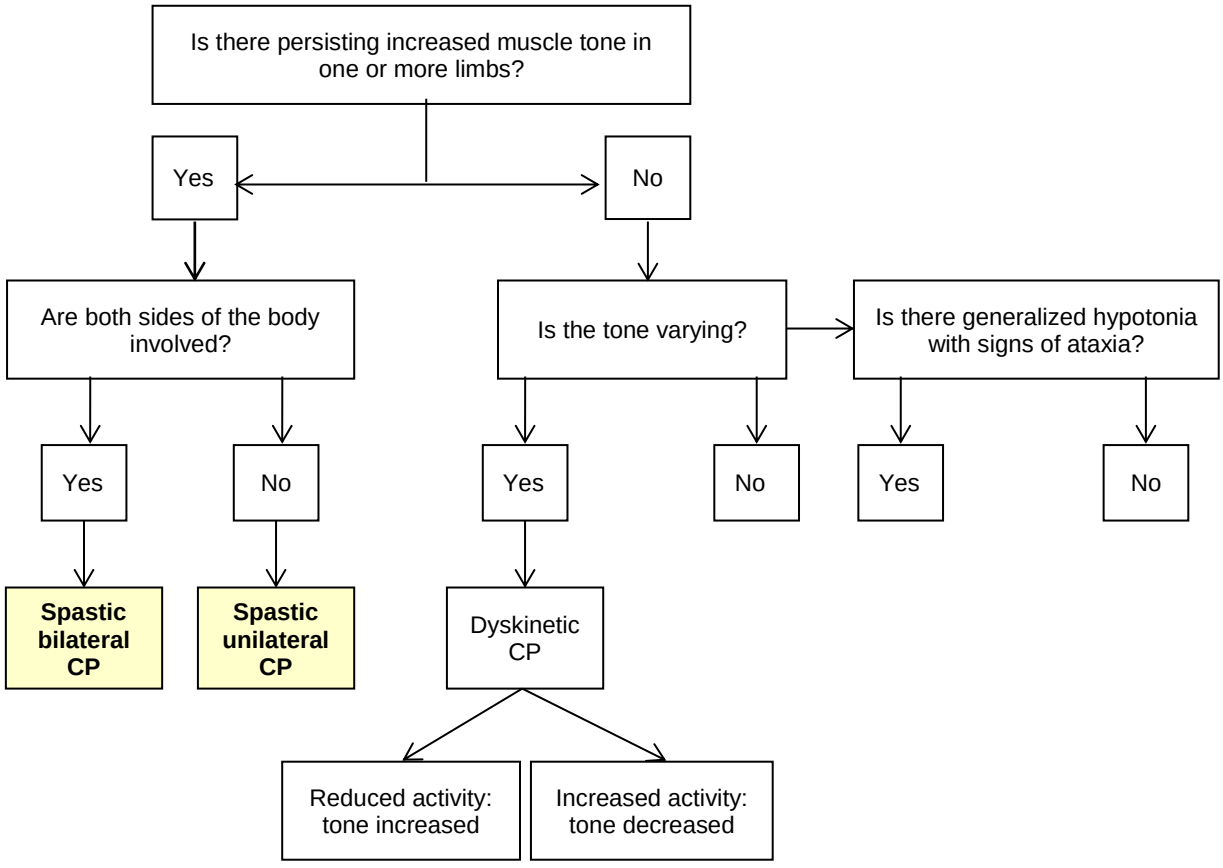

Figure 1. Hierarchical classification tree of Cerebral Palsy subtypes according to the Surveillance Group of Cerebral Palsy in Europe (Source: Cans et al. 2000[11])

In 2005, Bax et al. proposed four major dimensions of classification.[10]

The *first dimension* considers **the nature of their motor abnormalities**, including the observed tonal abnormalities and the diagnosed movement disorders. For the classification based on the observed tone abnormalities, the strategy of the SCPE has been adopted. This divides CP into the spastic, dyskinetic or ataxic type. Spastic CP is characterized by increased muscle tone and/or pathological reflex activation. Dyskinetic CP is characterized by involuntary, uncontrolled, recurring, occasionally, stereotyped movements and ataxia represents the loss of normal muscle coordination. For the key function of ambulation, the Gross Motor Function Classification System (GMFCS) is proposed, grouping individuals with CP into one of the five levels based on functional mobility.[12] A parallel classification for upper limb function is provided by the Bimanual Fine Motor Function (BFMF) Scale[13] or the Manual Ability Classification Scale (MACS)[14].

The *second component* considered other, **associated impairments** that interfere with the ability to function in daily life and may at times produce even greater activity limitation than the other impairments. Himmelmann et al. reported that 32% of the children with spastic diplegia showed more than two accompanying impairments.[15] Learning disabilities (49%), epilepsy (34%) and visual impairments (21%) were most commonly reported in this group of children.

As a *third component*, the **anatomic distribution and radiologic findings** are identified as the basis for classification.

As a *fourth component* of classification, Bax et al. identified **timing and causation** of the insult.[10]

The third and fourth component will be discussed in the section describing the pathogenesis of CP.

GMFCS	BFMF
Level I	**Level I**
Walks without restrictions	One hand manipulates without restrictions
Limitations in more advanced gross motor skills	The other hand manipulates with restrictions or limitations in more advanced fine motor skills
Level II	**Level II**
Walks without restrictions, limitations walking outdoors and in the community	(a) One hand manipulates without restrictions, Other hand is only able to grasp or hold (b) Both hands: Limitations in more advanced fine motor skills
Level III	**Level III**
Walks with assistive mobility devices, limitations walking outdoors and in the community	(a) One hand manipulates without restrictions. The other hand has no functional ability (b) One hand has limitations in more advanced fine motor skill. The other hand has only the ability to grasp or worse
Level IV	**Level IV**
Self-mobility with limitations, children are transported or use power mobility outdoors and in the community	(a) Both hands have only the ability to grasp (b) One hand has only the ability to hold or worse
Level V	**Level V**
Self-mobility is severely limited, even with the use of assistive technology	Both hands only have the ability to hold or worse

Figure 2. The Gross Motor Function Classification System (GMFCS)[12] and the Bimanual Fine Motor Function (BFMF)[13] (Source: Himmelman et al. 2006[15])

According to the SCPE, 85% of the children with CP can be classified as spastic.[16] Bilateral spastic CP thereby accounts for 46% of the children with CP. The population-based study by Himmelmann et al observed that children with spastic diplegia represented 39,2 % of the total population. The latter study also expressed gross motor function and bimanual fine motor function over the different CP types.[15] Of the 144 children with spastic diplegia, 54% were classified at GMFCS level I to II, and 15% at GMFCS level III, representing the ambulant population amongst the bilaterally involved children (**Fig 3**).

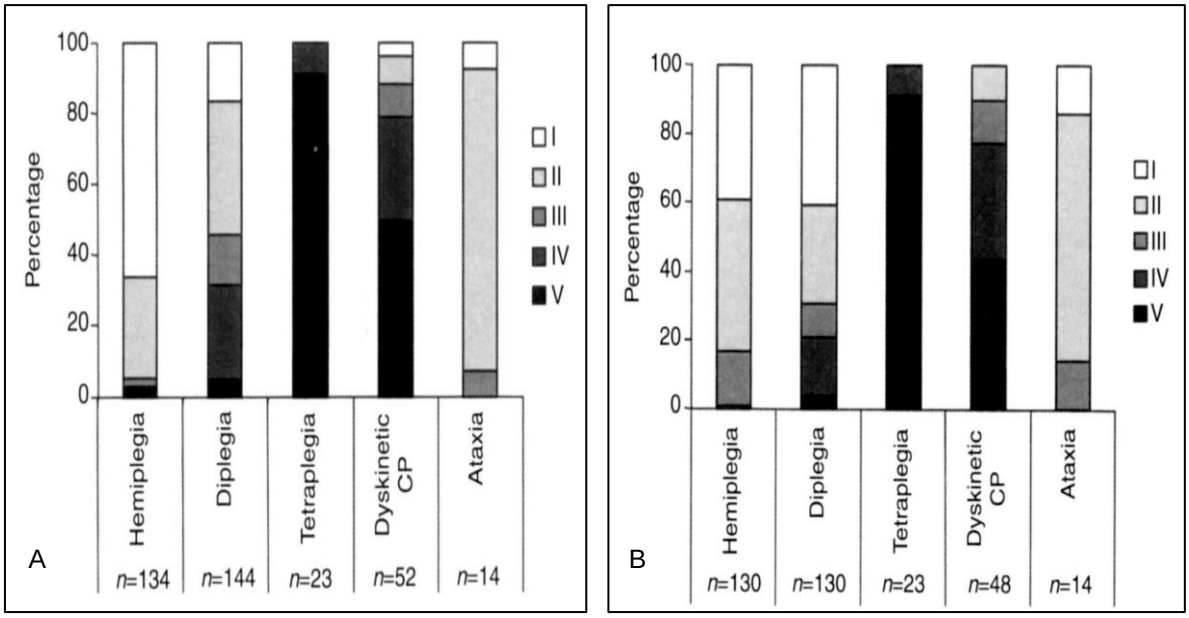

Figure 3. A. Distribution of the Gross Motor Function Classification System levels by type of CP in 367 children ; B. Distribution of the Bimanual Fine Motor Function levels by type of CP in 345 children with CP (Source: Himmelmann et al., 2006[15]).

2.2 Pathogenesis

CP results from an injury in the developing central nervous system, which can occur in utero, during delivery or in the first year of life. The patterns of brain lesions depend on the stage of brain development and can be divided into congenital or acquired brain lesions. Congenital lesions are usually caused by different types of events occurring the 28th day of life and can be categorized into malformations, periventricular lesions and cortical-subcortical lesions. **Malformations** usually develop during the first or second trimester of gestation and include predominantly cortical maldevelopments in bilateral distribution. In addition, different genetic conditions can also interfere with early brain development. **Periventricular lesions** generally occur during the late second and early third trimester. An insult during the early third trimester will mostly lead to periventricular white matter disease or leucomalacia (PVL). Insults at the end of the third trimester or around birth usually affect **the cortical and/or deep grey matter.**[17] **Acquired lesions** appear after the 28th day of life and before the age of three years. These lesions generally occur as a postnatal event like a trauma or an infection.

According to the SCPE however, abnormal MRI findings were found in only 90% of the children with bilateral spastic CP. Around 60% of the children showed PVL and only 10 to 15% showed grey matter lesions.[18] **Fig 4** provides a schematic and MRI view of PVL. Several authors have demonstrated the relationship between spastic diplegia and prematurity. [16,19,20] Himpens et al. observed significantly different distributions of the type of CP over the different groups of gestational age.[19] Spastic diplegia dropped down from 85% in the extremely preterm group to 23% in the term group.

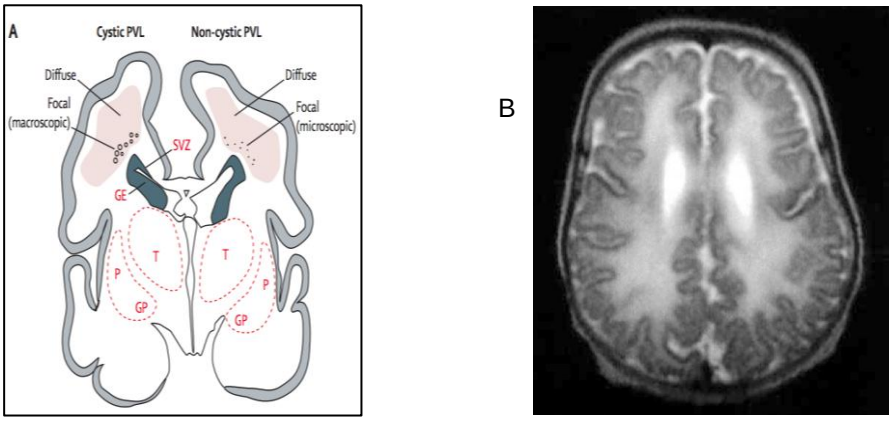

Figure 4. Periventricular leucomalacia (PVL): **A** Coronal sections from the brain of a 28-week-old premature infant. The dorsal cerebral subventricular zone (SVZ), the ventral germinative epithelium of the ganglionic eminence (GE), thalamus (T), and putamen (P)/globus pallidus (GP) are shown. The focal necrotic lesions in cystic PVL (small circles) are macroscopic in size and evolve to cysts. The focal necrotic lesions in non-cystic PVL (black dots) are microscopic in size and evolve to glial scars. The diffuse component of both cystic and non-cystic PVL (pink) is characterised by the cellular changes (Source: Volpe 2009)[21] **B** Weighted MRI scan of a 26 week premature infant. (Source: Blumenthal 2004)[22]

2.3 Clinical characteristics of children with bilateral Cerebral Palsy

2.3.1 The framework of the International Classification of Functioning, Disability and Health

To describe the clinical characteristics of bilateral spastic Cerebral Palsy, the International Classification of Functioning, Disability and Health (ICF) as defined by the World Health Classification (WHO) can be used (**Fig 5**).

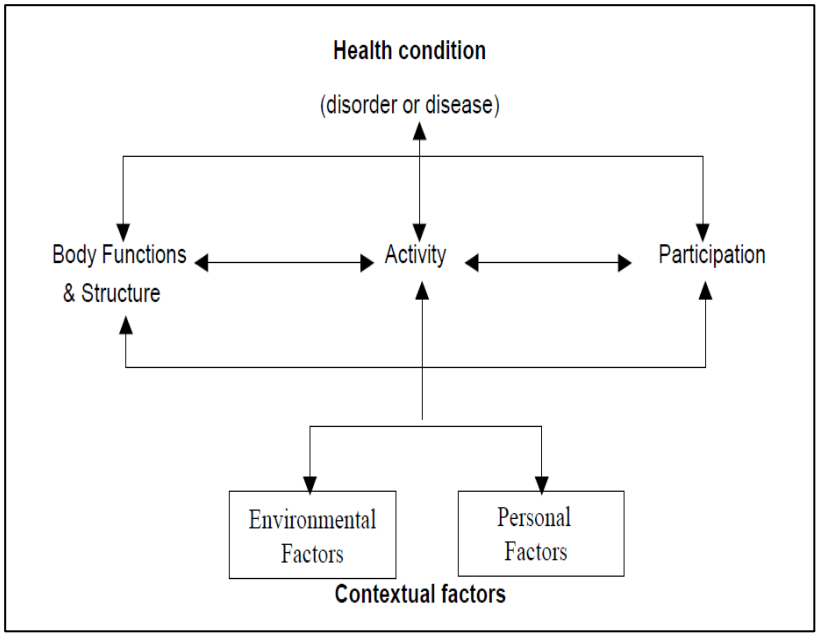

Figure 5. The conceptual framework of the International Classification of Functioning, Disability and Health (Source: WHO, 2001)

In the ICF, disability and functioning are viewed as outcomes of interactions between health conditions (diseases, disorders and injuries) and contextual factors.

Body functions are physiological functions of body systems (including psychological functions). **Body structures** are anatomical parts of the body such as organs, limbs and their components. **Impairments** are problems in body function or structure such as a significant deviation or loss.

Activity is the execution of a task or action by an individual. **Participation** is involvement in a life situation. Activity limitations are difficulties an individual may have in executing activities. Participation restrictions are problems an individual may experience in involvement in life situations.

Environmental Factors make up the physical, social and attitudinal environment in which people live and conduct their lives. (e.g. school and home environment). **Internal personal factors** influence how the individual experiences his/her disability (e.g. gender, age, coping styles, social background, education).

The International Classification of Functioning, Disability and Health – Children and Youth version (ICF-CY) is a WHO approved "derived" classification based on the ICF that includes further detailed information on the application of the ICF when documenting the relevant aspects of functioning and health in children and youth. The ICF-CY shares the same classification properties as its reference classification the ICF. However, in the interest of a streamlined, comprehensive ICF, which adequately addresses all aspects of functioning across the lifespan, the relevant stakeholders have recently agreed to merge the two classifications back into one while completing other updates and revisions.

As the overall conceptual framework of the ICF-CY is the same for both the ICF and the ICF-CY, this will further be referred to as 'ICF'.

2.3.2 Problems at the level of body structure and function

Spasticity

One of the main primary problems in children with bilateral spastic CP is spasticity. Following the definition by Lance (1980), spasticity is defined as *a motor disorder characterized by a velocity dependent increase in tonic stretch reflexes (muscle tone) with exaggerated tendon jerks, resulting from hyper-excitability of the stretch reflex, as one component of the upper motor neuron syndrome.* In children with bilateral involvement, spasticity is usually higher in the lower limbs compared to the upper limbs and most commonly affects m.Gastrocnemius, the Hamstrings, m.Rectus Femoris, mm.Adductores and m.Iliopsoas. In clinical practice, spasticity is usually evaluated using the Modified Ashworth Scale (MAS), complemented by the Tardieu Scale.[23-26]

Weakness

Muscle weakness has long been considered as a secondary problem in CP. It was considered as a result of co-activation of antagonistic muscles, reducing the net forces acting to rotate a joint. Although this lack of reciprocal inhibition causing excessive co-contractions has been confirmed[27,28], more recent investigations have identified muscle weakness as being more a primary problem in children CP. [26-30] Reduced muscle volumes throughout the lower limbs up to 50% were observed[31] and thereby, children with CP were less able to activate their target muscles maximally. [26,29,32] Although many questions regarding the pathophysiology of muscle weakness remain unanswered, muscular changes can probably not only be explained by classic interpretations of the effects of neural changes alone.[33] **Table 1** summarizes the facts regarding histological and physiological consequences regarding muscle strength problems in children with CP.

Table 1. Histological and physiological adaptations in the spastic muscle and consequences for children with Cerebral Palsy

Structural changes

- Altered distributions between type I (slow-twitch) muscle fibres and type II (fast twitch) fibres with generally, a predominance of type I fibres[34,35]
- Reduced muscle belly length and increased tendon length[26,33,36]
- Increased fibres size variability[30,35]
- Increased modulus of elasticity[37]
- Increased sarcomere length at the hamstrings[38]
- Reduced number of fibres, increased intramuscular fat and connective tissue[38]
- Inefficient recruitment of motor units[28]
- Reduced muscle cross-sectional area[39-42]
- Cross-bridge stiffness at muscular and collagen level[43]

Consequences

- Children with CP demonstrate a slower rate of maximal force production[28]
- Children with CP are less able to activate their muscles maximally[26,32]
- Children with CP show reduced half-relaxation time and disturbances in reciprocal inhibition[28,39]
- Children with CP show reduced muscle volumes[29,31,36,39,42,44-46]

In general, alterations at the spastic muscles cause the muscle to exert maximum force at a different point along the length-tension curve. Because of this biomechanical disadvantage, the muscle no longer works optimally for gross motor functions and walking. Muscle weakness thereby contributes for a great extend to functional capacity in these children.[27 47-54]

Muscle strength is most commonly evaluated using manual muscle testing. [55,56] Although manual muscle testing has limited sensitivity, no other evaluation tools provide an equal, easy-to-use screening tool. Controversial results are available regarding the use of handheld dynamometry.[57] Additionally, handheld dynamometry does not provide an outcome measure that can be used to define training-intensity. Isokinetic measurements show reasonable reliability, but are limited to measurements of the knee-flexors and extensors and are known to be expensive and time-consuming.[58] During the last years, the use of functional muscle testing has increased, as it is a useful and reliable means to measure muscle strength over multiple joints and muscles.[57]

Muscle contractures and skeletal deformations

As spasticity interferes with normal muscle stretch during growth, it is thought to contribute to the development of secondary muscle and soft tissue contractures and to skeletal deformations. Stiffening of the extra-cellular matrix with increased collagen, an increase of in vivo sarcomere length and reduced relaxation half-time can lead to higher passive stresses. The increased and unbalanced muscle tone sets off a cascade of bone and muscle responses. The combination of the changes in muscle architecture together with alterations in muscle use associated with spasticity results in stiffness. Combined with altered spontaneous limb movement, these cause reduced muscle length and limited joint function. The altered muscle forces cause abnormal forces on the growing bone, which as such can cause lever-arm dysfunctions.[59] Range of motion and muscle lengths are usually measured clinically, by goniometer. The measurements show moderate reliability.[25] In addition, as tone reduction became more successful in the last years, the role of muscle weakness in the development of bony deformities became more obvious. [60,61]

Selectivity

As a direct result from the brain lesion, children with CP are known to show typical movement patterns. These patterns, in combination with the spasticity and muscle weakness, attribute to the limited ability for the children to selectively activate specific muscles. Selective motor control involves isolating movements upon request, appropriate timing and maximal voluntary contraction without overflow movement. Different motor control tests are available for children with CP, most of them with limited reliability. [59,62-64]

Postural control

Impaired trunk control is one of the key features of children with CP. [65,66] Especially in children with PVL and children with diplegia, impaired trunk control is known as a major reason for postural problems.[67] **Fig 6** overviews the percentage of submaximal scores for the different items of the Trunk Control Measurement Scale, indicating trunk deficits for the different topographies. Literature on the evaluation of trunk control in children with CP is scarce and only a limited number of clinical tools are available to measure trunk control in children with CP. More static evaluations are the Seated Postural Control Measure (SPCM)[68] and the Spinal Alignment and Range of Motion Measure (SAROMM).[69] More recent and dynamic evaluation tools are the Segmental Assessment of Trunk Control (SATCo)[70] and the Trunk Control Measurement Scale (TCMS)[71].

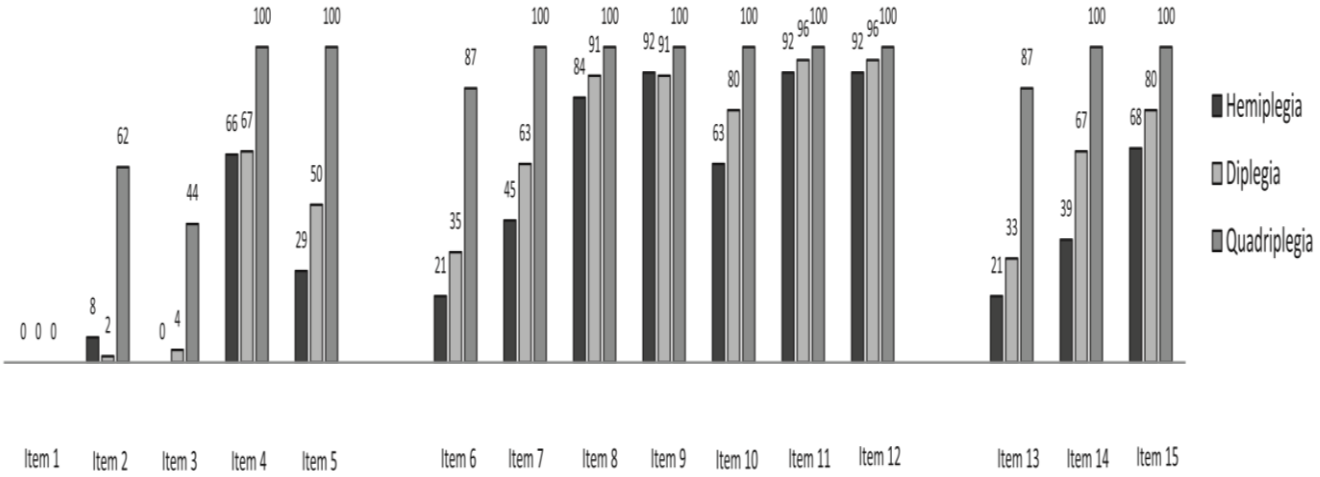

Figure 6. Percentage of submaximal scores for the different items of the Trunk Control Measurement Scale, indicating trunk deficits for the different topographies. (Source: Heyrman et al. 2013[71])

2.3.3 Problems at the level of activity and participation

Gait problems

The combination of pathological movement patterns, weakness and impaired muscle lengths in CP are known to cause a wide variation of pathological gait patterns. Children with CP usually walk independently but most have reduced speed, increased walking speed and increased energy expenditure. [72,73] The most commonly used classification in clinical practice and research is the classification of Rodda and Graham. It is based on a systematic review of literature and provides a description of the pathological patterns, as well as the potential underlying mechanism and strategies.[74]

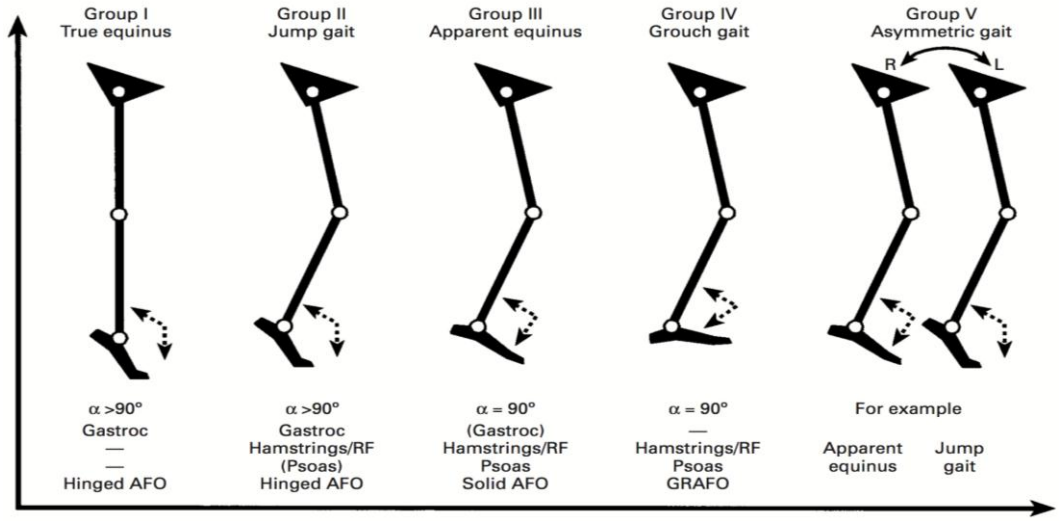

Figure 7. Visual representation of the sagittal gait patterns during mid-stance in spastic diplegia. Source: Rodda et al. 2004 [74]

Normal gait has several attributes. Because of the neuromuscular problems that occur in CP, all these attributes are lost in varying degrees. Pathological gait is a combination of cause and effect. The brain injury can interfere with gait in several ways. The primary effects of the brain injury (loss of selective motor control of muscles, difficulties in balance and abnormal muscle tone) arise from the brain as a direct result from the brain injury. These impose abnormal forces on the skeleton, causing secondary effects of musculoskeletal deformities. A child will cope with its primary and secondary problems using compensation strategies, which can be seen as tertiary problems. Pathological gait is always a results or a mixture of these primary, secondary and tertiary problems. Three-dimensional gait analysis usually includes joint kinematics (the quantitative three-dimensional measurement of motion), joint kinetics (the measurements of moments and power production occurring in the major articulations of the lower extremities) and dynamic electromyography (muscle activity patterns, frequently expressed as on-off signals of individual muscles and/or muscle groups). This allows integration of all these components and can thereby support the clinician in unravelling these problems and consequently, improve the understanding in the child's problems.[59]

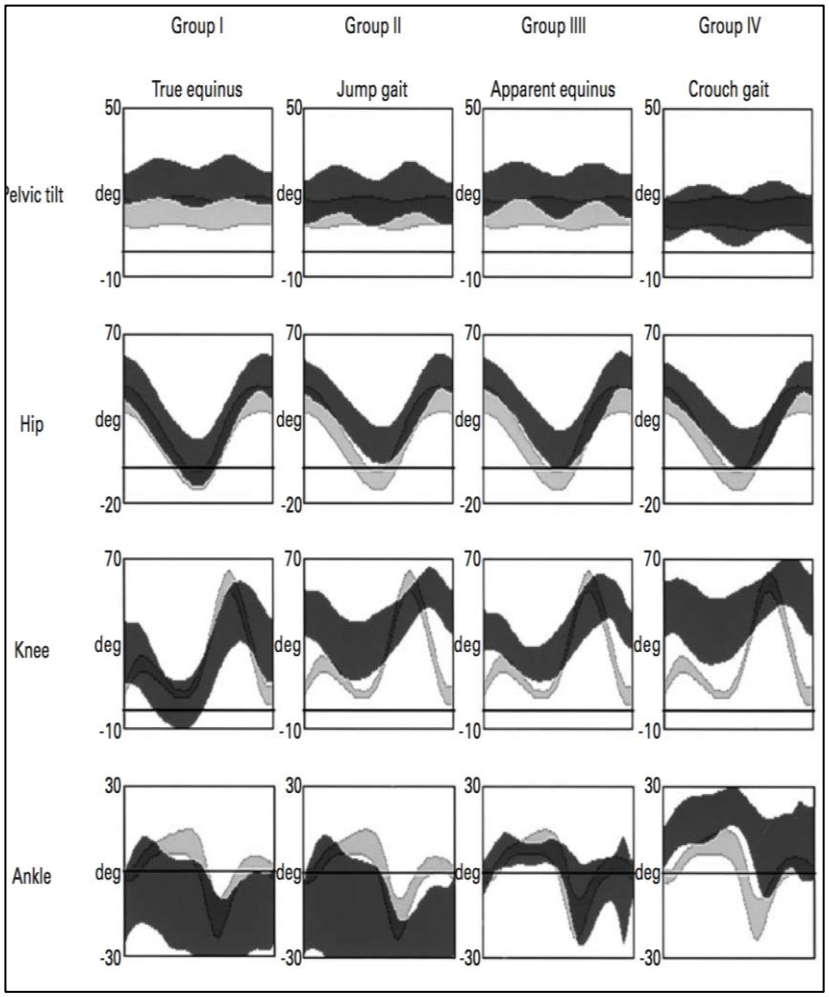

Figure 7. Summarized sagittal plane kinematic data for the typical gait patterns in spastic diplegia. The light-grey wave-forms represent the average gait pattern for a typically developing child; curves in dark-grey the typical gait patterns for children with spastic diplegia. (Source: Rodda et al. 2004[74])

The Movement Assessment Profiles (MAPS) and the Gait Profile Score (GPS) have been developed to summarize much of the complex information within the kinematic data.[75] Movement Analysis Profiles (MAPS) can be calculated as the root mean square error (RMSE) between the point-by-point comparison of the lower limb joint angle and the averaged joint angle of the reference group. Calculation of the average of all lower limb joint angles results in the GPS, which summarizes the overall severity of gait pathology. [59,75]

When analysing group data however, the heterogeneity of CP and the variety of gait patterns in children with spastic diplegia, might cause the phenomenon of 'regression to the mean'. As not all children show the same gait deviations, determining group improvements based on gait data is challenging. Z-scores represent a standardized deviation from a reference group.[76] Z-scores are non-dimensional and therefore, allow comparison of individual pathological parameters. The use of z-scores therefore provides the possibility to compare individual improvements within a group of children with different gait pathologies.

Limitations in gross-and fine motor function

The majority of children with bilateral spastic CP has moderate to severe problems in gross motor function and mobility. [1,13,15] The disorder in the development of gross motor function thereby represents the hallmark of these conditions. [52,77] Many different outcome measures are available to evaluate gross motor function in these children. A systematic review by Ketelaar et al.,[78] concluded that especially the Gross Motor Function Measure[79] and the Pediatric Evaluation of Disability Inventory[80] fulfilled the criteria of reliability and validity with respect to responsiveness to change.

The Gross Motor Function Measure 88 (GMFM-88) and 66 (GMFM-66) version are standardized observation measures to evaluate gross motor function changes in children with CP.[79] The tests as such have no age-limits, but the test items are selected with reference to a five-year-old with normal motor abilities, that can usually accomplish all items. The GMFM-88 is the oldest version, consisting of 88 items, divided into five dimension: lying and rolling (dimension A), sitting (dimension B) crawling and kneeling (dimension C), standing (dimension D) and walking, jumping and running (dimension E). Each item is scored on a four-point scale. Inter-and intra-reliability measures were high, even amongst un-experienced observers. [81-83] In addition, it has been validated by demonstrating its capability to detect change in gross motor function over time.[84]

The Pediatric Inventory of Disability Inventory (PEDI) is a standardized assessment instrument for chronically ill and disabled children from six months to 7,5 years. It consists of a questionnaire, designed to evaluate the child's functional ability along three scales: typical functional skill level, physical assistance and modifications or adaptive equipment used. Each scale is divided into three domains: self-care, mobility and social function. The PEDI has been developed for both discriminative as well as evaluative purposes. A number of studies report the PEDI as reliable and valid. [80,85] Responsiveness of the PEDI was confirmed over a six-month period.[86]

Rosenbaum et al. described the patterns of gross motor development of Canadian children with CP, using longitudinal observations.[77] Based on 2632 GMFM measures, five distinct motor development curves were created (**Figure 8**), describing the important and significant differences in the rates and limits of motor development among children with CP by severity. There was substantial variation in gross motor development among the different functional levels. Logically, children functioning at GMFCS I and II reach a higher maximum GMFM-66 scores than children functioning at level III, IV and V. The motor development curves also suggest a trend for children severe of impairment reach their limit in gross motor function level at a younger age than more functional children. In addition, children functioning at levels III through IV progress significantly faster that children with level I, but children in level II do not progress faster than level I.[77] In a similar Swedish study, Beckung et al. differentiated motor development amongst the different types of CP and found similar results for the subgroups of diplegia and hemiplegia.[87] Recently, Smits et al. confirmed the development trends seen in the Canadian study in a Dutch population.[88]

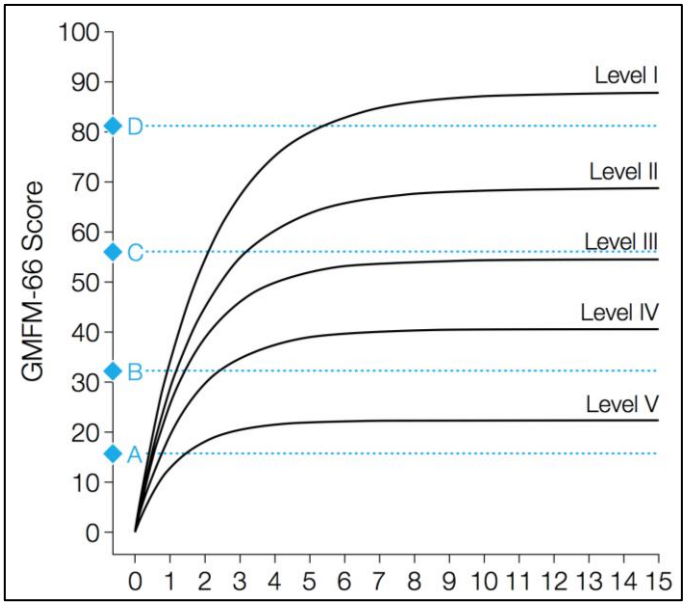

Figure 8. Predicted average development by the Gross Motor Function Classification System Levels. The diamonds on the vertical axis identify 4 Gross Motor Function Measure-66 items that predict when children are expected to have 50% chance of completing that item successfully. (Source: Rosenbaum et al. 2002[77])

Participation restrictions

Children with disabilities are less involved in leisure activities than their peers, participate less in family activities outdoors and often experience social experience at school.[89] Their recreational activities are therefore more passive, home-based and lack variety. Facilitators for participation include age, motivation, cognitive abilities, support, coping and environmental resources. Barriers are behavioural difficulties, activity limitations, parental stress and speech-and language impairments. [89-93] Other factors that influence the choice and intensity of participation are gender and family preferences. Children often report a significant higher presence of adults in their life and a reduced perception of independence and autonomy.[89] The School Function Assessment[94], the Children's Assessment of Participation and Enjoyment (CAPE), the Preference Activities for Children (PAC)[95] and the Life-H questionnaire[96] seem to show the best clinimetric properties to evaluate participation limitations. However, limited data is available to suggest that these measures are adequately sensitive to detect minimal clinically significant change. [97]

Evidence is provided that the Goal Attainment Scale is a more responsive measure of motor change and participation limitations in infants with motor delays than standardized motor assessment like the Peabody Developmental Motor Scales (PDMS) or the GMFM.[62,97,98] GAS has unique features that are advantageous for measurement of qualitative change and small but clinically important improvement in motor development and function. In addition, it can capture meaningful aspects of a person's progress and in that way, improve motivation. By convention, scores of -2 and -1 represent the two less favourable outcomes, 0 represents the expected level of goal attainment and +1 and +2 represent the two levels of goal attainment that exceed expectations. [98-100] Additionally, a change score can be calculated for multiple goals. The composite T-score, with a mean equal to 50 and a standard deviation of 10, provides a method to quantify change over time and across individuals.[100]Studies evaluating the clinimetric properties of the GAS in pediatric rehabilitation support the content validity and responsiveness to change. [98,101] However, evaluation of its concurrent validity suggests that GAS should not replace standardized idiosyncratic measures like the GMFM, the Peabody Developmental Gross Motor Scale and the PEDI but preferably used complementary. [98,99,101-104]

2.3.4 Interactive dynamics

The interactive dynamics the ICF suggests underlying correlations between the measurements at the different levels of the model. Chiarello et al. defined a multivariate model of determinants of change in motor abilities and participation in daily activities for children with cerebral palsy (**Fig 9**).[105] The model demonstrates the complexity of the pathology by scheduling the relationship between the different levels of the ICF. It demonstrates how aspects of the child, family, environment and community influence children's activity and participation.

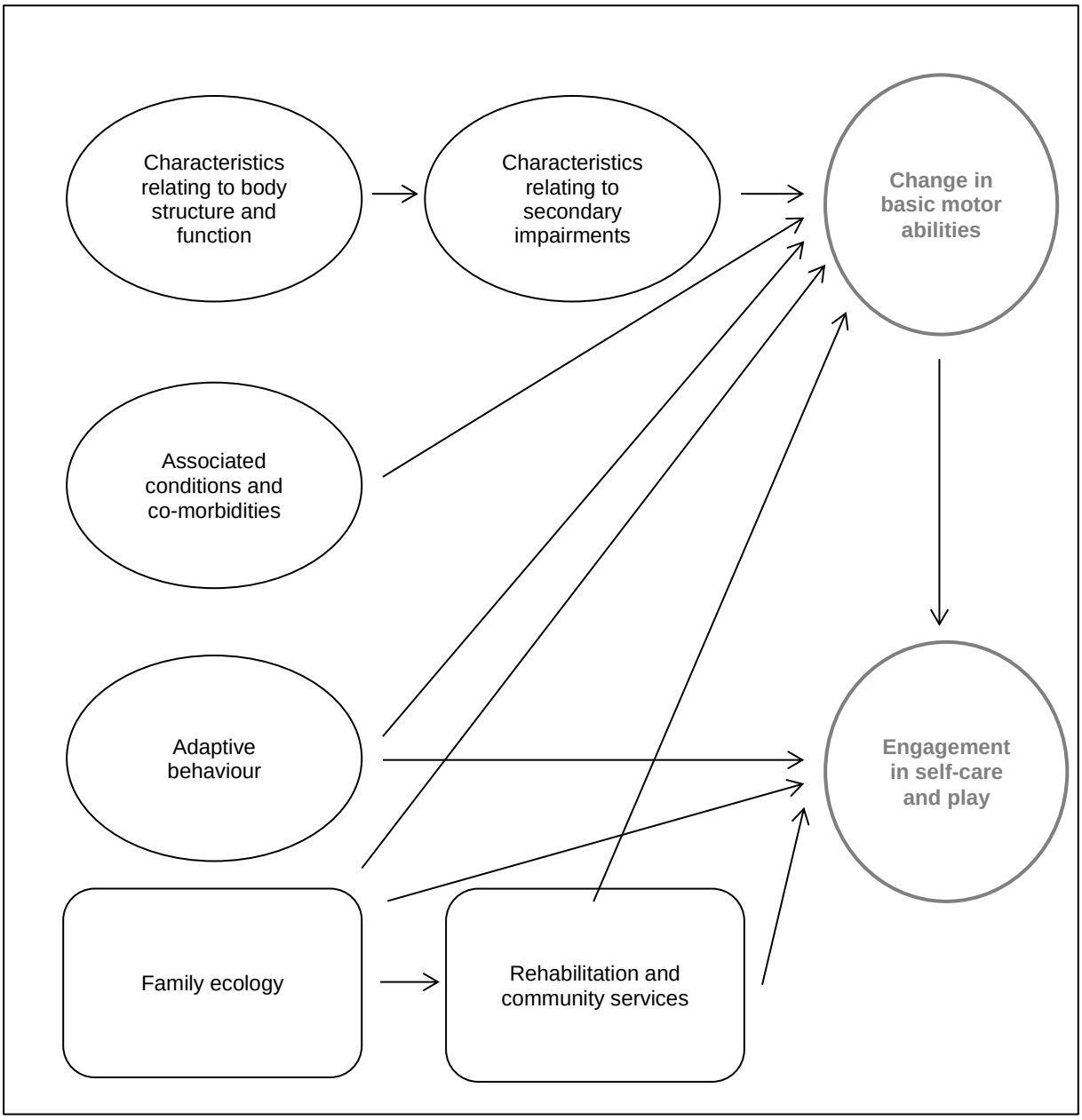

Fig. 9 Multivariate model of determinants of motor change in motor abilities and participation in daily activities for children with Cerebral Palsy. (Source: Chiarello et al.[105])

The relationship between the different levels of the ICF however, remains a complex und unravelled entity of direct and indirect interactions. Several researchers have therefore expressed the relationship between the different impairments underlying functional and participation limitations quantitatively and have confirmed inverse relationships between the different models.[47,52,53,106-109] Ostensjo et al. found moderate correlations between measurements of spasticity, strength and selective motor control and gross motor function, but weaker correlations of the impairments to mobility, self-care and social function of the PEDI evaluations.[52] Thereby, the selective motor control score was the strongest predictor explaining 15-45% of the variance in gross motor function. The GMFM-66 score accounted for 88% of the variations in mobility. Wright et al found poor relationships (r<40) between the change scores of measures of body functions and structure (spasticity), activity (GMFM and PEDI) before and after injections with botulinum toxin A.[53] In a more recent study including more functional children (GMFCS I and II), Bartlett et al. could only explain 58 % of the variance in gross motor function by the primary and secondary problems at impairment level.[47] Gorter et al. found only a very low correlation (-0,28) between spasticity and gross motor function.[106] Ross et al. found that, in children with spastic diplegia, strength explained more variation in gait and gross motor function than spasticity.[107] This positive relationship between strength and gait was confirmed by Eek et al.[108], who observed that in independently walking children showed more than 50% of the predicted strength values and by Damiano et al.[27], who demonstrated a direct relation between strength of the knee extensors, GMFM and gait velocity.

The relationship between gross motor function and participation restriction has been investigated by Kerr et al., indicating a moderate but negative relation between the GMFM-88 and the Lifestyle Assessment Questionnaire.[110] Beckung et al. showed that restriction in participation was best predicted by the GMFCS.[13] Jaspers et al. investigated the indirect relation between gait pathology and quality of life and observed an adverse impact of the severity of gait pathology on quality of life as reported by parents and children.[111] Both parents and children identified gait speed as an important factor contributing to their quality of life.

3. Treatment context of children with bilateral Cerebral Palsy

3.1 Multidisciplinary context

The pathology of CP is complex. The variety of clinical problems associated with the brain lesion all requires sufficient attention. This is at best offered by a multidisciplinary team surrounding the child and its family. [2,59]

Figure 9 describes the usual treatment time strived for at the University of Pellenberg. From birth until early childhood, management optimally consists of intensive physiotherapy to support motor development, and to maintain range of motion. Children with CP optimally receive conservative treatment until their motor patterns are matured, usually between 8 and 10 years. Delaying the often inevitable need of surgery is important because the results of early surgery are less predictable and have a higher risk of failure and relapse. [59,112] The consequences of persistent increased tone can be limited with the use of Botulinum Toxin A injections (BTX-A). BTX-A is a paralysing agent that inhibits the release of the neurotransmitter acetylcholine at the neuromuscular junction, resulting in a reduced muscle tone. Selective tone reduction by means of BTX-A, combined with different conservative treatment options, has the potential to achieve functional benefits. [113-115] An adequate follow-up treatment including orthotic management, serial casting and intensive physical therapy has been demonstrated of major importance for a maximal benefit of the injections. [2,113,114,116]

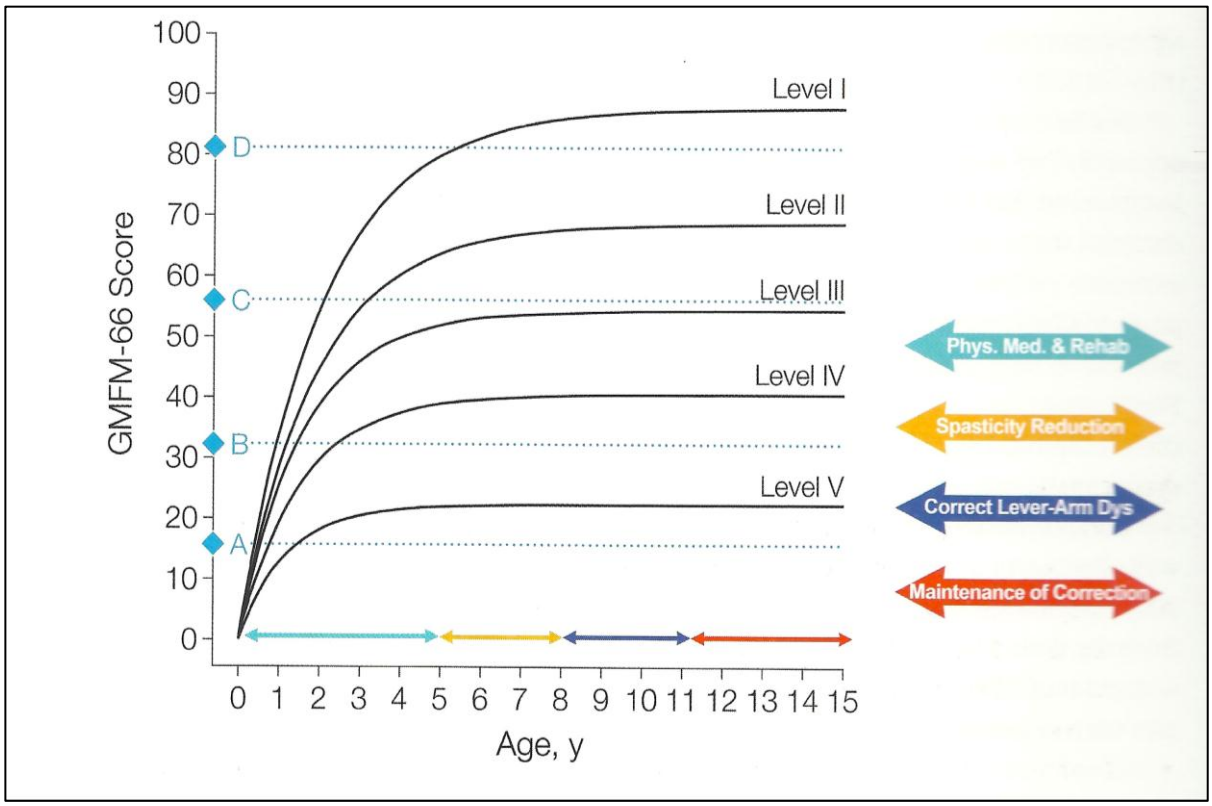

Fig 9. Usual treatment timeline at the Gilette Children's Speciality Hospital, Minnesota, US (Source: Gage et al.[59])

4. Physiotherapy in children with Cerebral Palsy

The motor impairments typically associated with CP are such that physiotherapy is often one of the first interventions recommended for a young child diagnosed with CP. Physiotherapy treatment in early childhood therefore mostly focuses on stimulation of motor development and learning new functional activities.[117,118] Thereby, stimulation by the parents and handling techniques are of major value to reassure treatment continuity. [119,120]

Although stimulation of motor development and functional activities remains a central aspect throughout the lifespan of children with CP, tone reduction becomes increasingly important, especially in the growing, school-aged child (**Fig 9**). Crucial in the success of BTX-A treatment is the adequate follow-up treatment with serial casting, appropriate orthotic management and intensive physical therapy. The selective tone reduction of overactive muscles provides a window of opportunity for therapeutic interventions by decreasing muscle tone, and thereby allowing increased range of motion, the potential to strengthen antagonist muscles and the opportunity to develop better motor control and balance. [8,116] Thereby, transferring the use of these improved joint-and muscle conditions into functional activities is an essential step.[121,122]

Nevertheless, treatment of children with CP is probably complicated by the fact that clinical manifestations are widely believed to change through life-span. In particular, after developmental gains in childhood, children with CP may decline in gross motor function as they move into adolescence and adulthood. Bottos et al. found that 13 of 29 children with CP who could walk independently before 18 years lost independent ambulation between the ages of 20 and 40 years.[123] In a survey of 766 Norwegian adults with CP, 88% of the participants were ambulant but 45% of this study group reported that their walking ability had deteriorated.[124] A large longitudinal study by Day et al on 7550 children with CP showed that 34% of the children who walked unsteadily and sometimes used a wheelchair as a child, lost their ambulatory mobility as young adults.[125]

During the last decades, the ICF has played a major role as an eye-opener for many physical therapists and researchers. [5,126] Using the ICF as a framework has confronted the therapist with the complexity of the pathology and the extensiveness of the problems, at all levels of the model. This concept has forced the therapist to consider the functional perspective of the pathology and to enable children to master important tasks and to participate in day-to-day activities.[1,127,128] Nevertheless, the core problem of CP implies the progressive nature of the pathology. Addressing primary impairments and prevention of secondary impairments therefore remains a difficult but inevitable task for the physiotherapist treating children with CP. [2,3,59] Therefore, treatment of children with CP is a balanced but challenging exercise between optimizing motor development, promoting independent functionality and participation but with sufficient attention to prevention of secondary problems that limit activities and participation.

In the last decades, research in physiotherapy treatment of children with CP has increased remarkably. Many intervention studies so far have evaluated the effectiveness of individual techniques and have shown promising results. [7,129] A variety of 'impairment-based' studies demonstrated positive effects of for example strength training on muscle strength, gait and gross motor in children with bilateral CP. [130-132] At the other side of the spectrum, there are 'participation-based' studies, demonstrating positive effects at children's participation and enjoyment.[6,133] Functional, task-oriented training programs demonstrate promising results on gross motor function and participation of children, but unfortunately, are not able to meet the problems regarding deteriorating muscle lengths and joint deformities.[134] The effectiveness of the individual techniques and specific approaches used in treatment of children with bilateral spastic CP are summarized in **chapter 1 and 2**.

Although the view of the ICF has brought a very positive change, it opened many discussions regarding the entry point in treatment. As a physiotherapist, it needs to be a well-balanced decision to aim for a focused treatment of an impairment, to modify the environment or maybe both.[5] In addition, children with CP and especially the weaker ones, are usually in need of assistive devices and consequently, adapting environmental aspects is an important task for the physiotherapist treating children with CP.[135,136] Adaptive orthopaedic materials and devices children are mainly used to improve the child's daily functioning, but can also be applied in positioning to prevent contractures and deformities.[137] In addition, the role of parents in the clinical decision process of the physical therapist is a factor that has been neglected in the past. It is however clearly demonstrated that enabling parents to participate more fully in their child's treatment regime, protects mothers from distress, increases self-efficacy and motivation and thereby therapy outcome.[138-140]

Addressing all these issues in a well-balanced therapy plan is a challenging task for the therapists. Therefore, several research studies have successfully introduced the principle of goal-setting in physiotherapy of children with CP. Goal-oriented treatment has the potential benefit from targeted interventions to enhance training specificity. Goal planning, in that context, can be used to identify the tasks and contexts of particular interest.[141] Several clinical reasoning strategies are available to guide the physical therapist in goal selection. Unfortunately, these clinical reasoning strategies are usually complex, extensive and are not developed specifically to guide the physiotherapist in priority setting.[142-144] Additionally, to our knowledge, none of these strategies has been validated. **Chapter 3** therefore introduces and validates a clinical decision framework, based on existing structures, to guide physiotherapists in defining the main problems for an ambulant child with bilateral spastic cerebral palsy. It thereby supports the selection of appropriate individual goals for the child.

There is lacking insight in the effectiveness of multifaceted physiotherapy approaches, tailored to the needs of the child and thereby, addressing more adequately the heterogeneity of the pathology and integrating all these aspects. Based on the goals selected in chapter 3, the evidence tables from chapter 1 and 2 can be used to select to the appropriate technique to reach the specific treatment goal. In this way, an individually defined, tailored treatment program can be defined.

As a next step, the effectiveness of the individually defined programs on gait and gross motor function will be evaluated in an intervention study. To be able to set-up a blinded and randomized controlled study, objective and blinded evaluation is required. Objective evaluation of gait can be performed independently at the University Hospital Pellenberg, by means of three-dimensional gait analysis. Evaluation of gross motor function can at best be performed using the Gross Motor Function Measure -88. However, an obstacle for blinded evaluation of the test is the need for the child to feel comfortable. An assessor unknown to the child might make the child insecure, frightened and less compliant. Therefore, a reliability study will be performed to evaluate the agreement between live scores of the GMFM-88, performed by a person familiar to the child, and the blinded scores of the video-registrations by independent assessors (**Chapter 4**).

Consequently, **in chapters 5 and 6**, the effectiveness of an individually defined treatment approach are compared to the effectiveness of a general program that is not based on individual problems of the child.

Even though only weak evidence is available for the majority of the physiotherapy interventions in lower limb treatment, physiotherapy still remains a key treatment for children with CP. This can mostly be supported by the fact that intervention studies in CP are limited by several factors and thereby face inevitable methodological difficulties. Although some researchers state that it is still very early for straightforward clinical guidelines, some key features need to be translated into practice. **Chapter 7** therefore evaluates evidence-based practice behaviour in usual care of ambulant children with bilateral CP.

5. Aims and outline of the project

5.1 Aims and objectives

The **overall aim** of the project is to validate and evaluate an individually defined targeted physiotherapy treatment approach for children with bilateral spastic cerebral palsy.

The **specific objectives** of the of doctoral project are

1. To define the effectiveness of the most commonly used techniques used in physiotherapy of children with CP, based on literature;
2. To develop a clinical decision framework for the identification of main problems and treatment goals for ambulant children with bilateral spastic CP;
3. To evaluate the validity and reliability of the Gross Motor Function Measure -88 by means of video-scoring;
4. To evaluate the effectiveness of an individually defined targeted physiotherapy program on gait and gross motor function of ambulant children with bilateral CP.
5. To evaluate evidence-based practice behaviour in usual care physical therapy in Flanders.

5.2 Outline

Part 1 (chapter 1 and 2) describe a systematic literature search evaluating the effectiveness of the most commonly used techniques and approaches in lower limb physiotherapy treatment of children with spastic CP. The results of the reviews are used to develop evidence tables summarizing the effectiveness of the different techniques on the most main outcome parameters in treatment of these children.

In part 2 (chapter 3), a clinical decision framework is developed to identify the main problems and treatment goals for ambulant children with bilateral spastic CP. This model is based on the International Classification of Functioning, Disability and Health and the Hypothesis Oriented Algorithm for Clinicians II.

In **part 3** (**chapter 4),** the agreement of the Gross Motor Function Measure-88 by means of blinded scoring of video-registrations is evaluated;

In part 4 (chapter 5, 6 and 7) the effectiveness of the individually defined physiotherapy treatment approach based on the evidence tables developed in chapter 1 and the clinical decision framework in chapter 3 and 4, are evaluated for children with bilateral spastic CP.

- Chapter 5 reports the results of a pilot study using a repeated measures design, comparing the effectiveness of the individually defined targeted treatment approach to the results of a general program and a period of usual care in ten children.
- Chapter 6 describes a randomized controlled trial comparing the effectiveness of the individually defined treatment approach to the effectiveness of the general approach using 60 interventions.
- Chapter 7 evaluates evidence-based practice behaviour in usual care physiotherapy in Flanders. This study compares the effectiveness of a predefined therapy program based on evidence-based guidelines to the effects of a period of usual care. It is hypothesized that supporting therapists by providing a predefined evidence-based intervention program, results in an improved treatment outcome compared to the usual care physiotherapy.

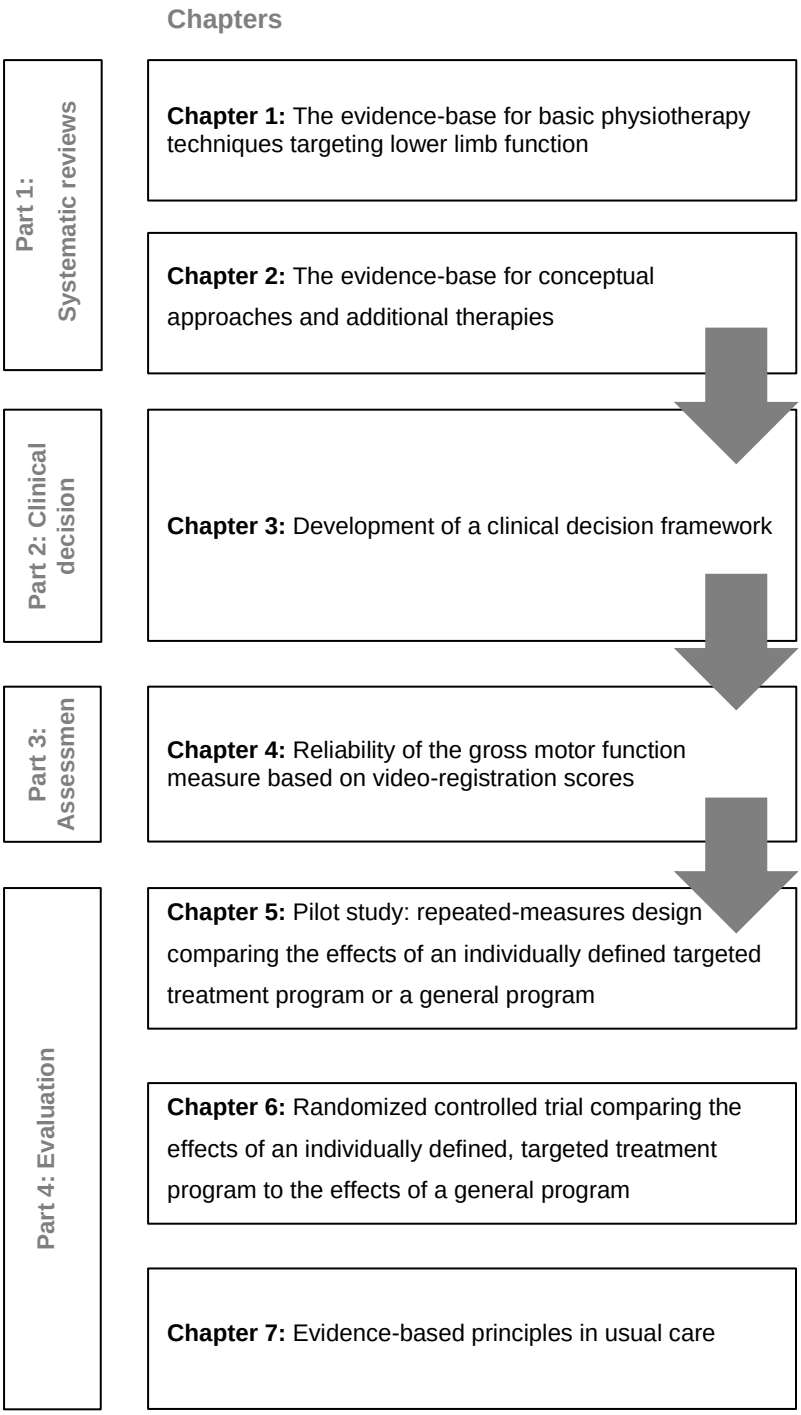

Fig 1. Outline of the project.

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Part 1
Systematic reviews

Chapter 1

The evidence-base for basic physical therapy techniques targeting lower limb function in children with cerebral palsy : a systematic review using the International Classification of Functioning, Disability and Health as a conceptual framework

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Abstract

Objective

This systematic review provides an overview of the effectiveness of basic techniques used in lower limb physical therapy of children with cerebral palsy. It aims to support the development of clinical guidelines for evidence-based physical therapy planning for these children.

Data sources and study selection

A literature search in 5 electronic databases extracted literature published between January

1995 and December 2009. Studies were evaluated using the framework recommended by the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM), which classifies outcomes according to the International Classification of Functioning, Disability and Health.

Data extraction

Three independent evaluators rated the strength of evidence of the effects according to the AACPDM

levels of evidence classification, and the quality of the studies according to the AACPDM conduct score system.

Data synthesis

A total of 83 studies was selected and divided into categories (stretching, massage, strengthening, electrical stimulation, weight-bearing, balance-, treadmill- and endurance training). Interventions targeting problems at body function and structure level generally influenced this level

without significant overflow to activity level and vice versa.

Conclusion

The more recent studies evaluating strength training mainly demonstrated level II evidence for improved gait and gross motor function. There was limited evidence for specific information on intensity, duration and frequency of training.

Introduction

Cerebral palsy (CP) describes a group of disorders of movement and posture that cause activity limitations. CP is attributed to non-progressive disturbances occurring in the developing foetal or infant brain'[1,2] Physical therapy (PT) plays a major role in the treatment of children with CP. High intensities and frequencies of therapy are reported and varying approaches and techniques are used.[3] In lower limb treatment, basic techniques, such as passive stretching, massage, muscle strengthening and many others, are frequently used. These basic techniques usually target specific problems at the level of body structure and function, such as range of motion (ROM), strength and muscle tone. More complex treatment approaches, such as neurodevelopmental treatment (NDT), Vojta therapy and Petö therapy are generally based on different principles of motor learning and require specific, specialized training.

A structural basis for PT planning is provided by the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY).[4-7] The ICF-CY describes a conceptual framework to report the variety of health information (diagnosis and functioning) and thereby delivers a common language and terminology to describe the child's problems in relation to functions and anatomical properties, activity limitations and participation problems. Physical therapists can use the model to guide the selection of measurement tools, both in goal-setting and decision-making processes and to determine meaningful outcomes.

Limited evidence is available on the effectiveness of different PT interventions in CP. A high-quality review by Anttila et al. included both upper- and lower-limb treatment and included randomized controlled trials (RCTs) only.[8] This review found only limited evidence supporting strength training, constraint-induced movement therapy and hippo-therapy.

In general, little consistency is provided on the use of different techniques in clinical practice of lower limb treatment in CP. While interventions at any of the elements of the ICF model may be important, each using different PT strategies and techniques[9], only limited emphasis is put on differentiation of the outcome effects on the different levels of the ICF.

This systematic review aims to summarize the effectiveness of different basic PT techniques used in children with CP and to differentiate the outcome effects on the different levels of the ICF. It thereby aims to identify interaction effects between the different outcome levels of the ICF and to explore the possibilities to develop clinical guidelines in PT treatment of children with CP.

Method

Search strategy

A systematic, stepwise search of the literature on PT in CP was performed using the following electronic databases: Web of Science, PubMed, Cochrane Library, Physiotherapy Evidence Database (PEDro) and CINAHL. The general search terms used were: “cerebral palsy” and “physiotherapy”, “physical therapy”, “exercise” and “training”. More specific search terms

were: “stretching”, “electrical stimulation”, “electrostimulation”, “massage”, “strength”, “treadmill”, “balance” and “weight bearing”. Inclusion criteria were: original articles published in peer-reviewed journals between January 1995 and December 2009, focusing on PT interventions targeting lower limb treatment in children and adolescents (< 18 years) with CP. Only articles written in English were included. Articles including children with different pathologies or targeting upper limb or trunk only were excluded from the study. In addition, interventions using mixed approaches or techniques and post-surgical interventions were not included in the study.

Based on the title and abstracts of the resulting articles, a first selection resulted in 159 articles. In a subsequent step, all articles were then screened by the first author. Articles not fulfilling the inclusion criteria were withdrawn. If the title and abstract did not provide sufficient information to fulfill the inclusion criteria, the full article was checked. In addition, all case-studies, expert opinions and non-systematic reviews were excluded. As a final step, reference lists of all systematic reviews included in the study were searched and missing articles fitting the inclusion criteria were added. The inclusion of more doubtful articles was discussed with a second and third assessor.

A resulting total of 83 studies was included in the study. A flow diagram of the selection process is shown in **Fig 1**.

Data collection

The full texts of all selected articles were read. The following data were extracted: type of PT intervention, numbers of patients included, topographic distribution of cerebral palsy, age of patients, type, frequency and duration of intervention, duration of follow-up, evaluation method and timing, summary of the results and conclusion.

Grouping data

The selected articles were grouped according to the type of intervention used. Interventions at the level of body function and structure were stretching (n = 5), massage (n = 4), electrical stimulation (n = 13), strength training (n = 25), endurance and physical fitness training (n = 10) and weight-bearing (n = 7). Interventions at both body structure and function and activity level were balance training (n = 6) and treadmill training (n = 13). Interventions using treadmill training were considered as a separate group, since mixed training goals, such as gait function, endurance or strength, made these articles difficult to categorize in one of the previous groups.

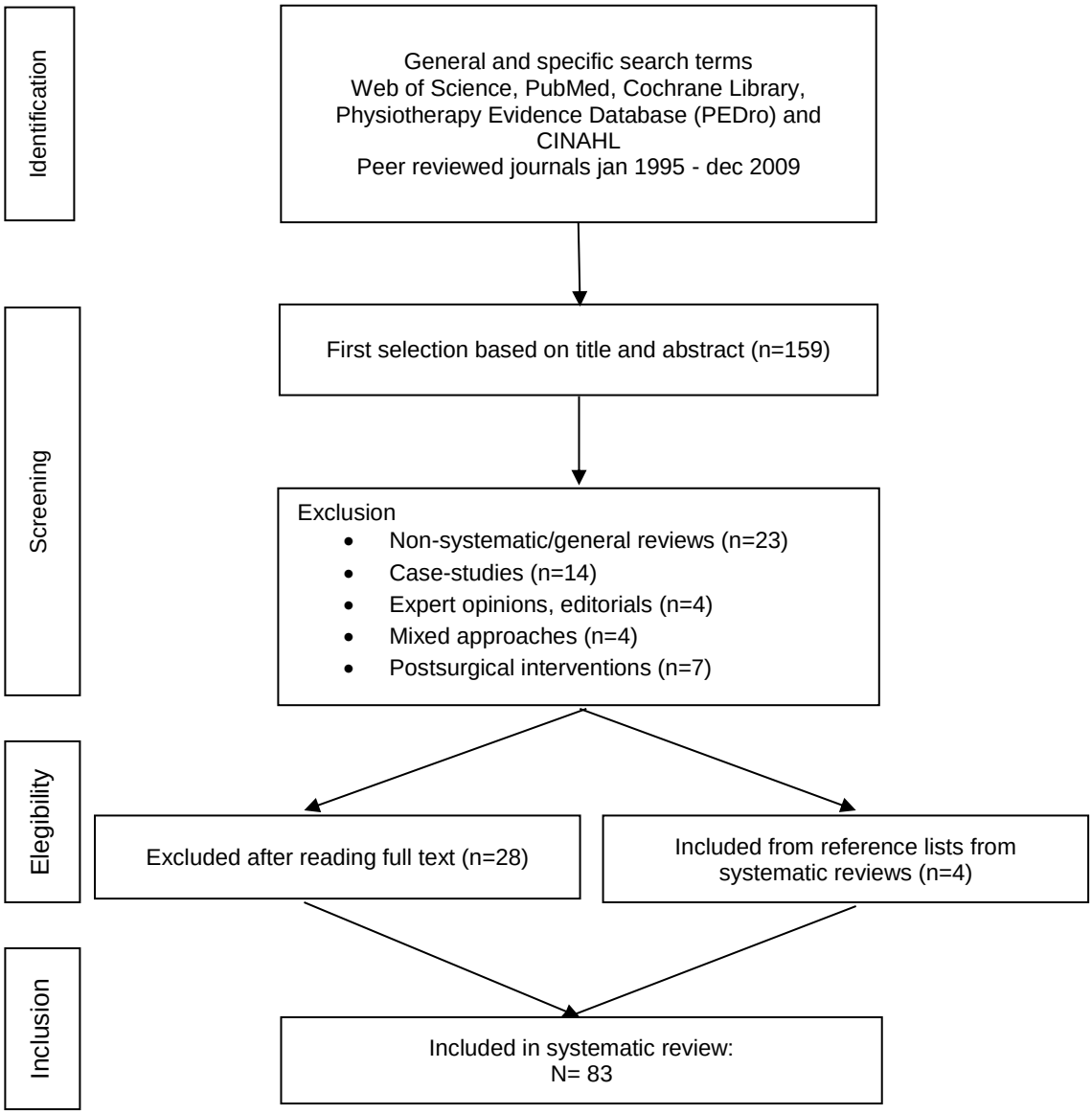

Fig 1. Flow-diagram describing the selection process

Classification and rating of the different outcome measures

Classification of outcome measures, rating of level of evidence and scoring of conduct scores was carried out by 3 independent evaluators: 1 PhD physical therapist (CVdB), 1 research physical therapist (IF) and 1 physical therapist MSc student (LVL).

Classification of the outcome measure

All 3 evaluators rated the outcome of the intervention on the level of the ICF: Body structure and function, Activities and Participation, Personal factors, and Environmental factors. Body structures are defined as the anatomical parts of the body, such as organs, limbs and their components. Body functions are the physiological functions of body systems (including psychological functions). Activity is the execution of a task or action by an individual. Participation is involvement in a life situation. Environmental factors make up the physical, social and attitudinal environment in which people live and conduct their lives. They can be viewed as facilitators (positive influence) or barriers (negative influence). Personal factors are the particular background of an individual's life and living, and compromise features of the individual that are not part of a health condition. These factors may include gender, race, age and other health conditions.[7]

Level of evidence

The same independent evaluators rated the studies according to their level of evidence using the rating system proposed by the American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) (**Table 1**).[10] Level of evidence describes the potential in a research study design to control for factors, other than the intervention, that may affect the observed outcome. In descending order, the designs decreasingly demonstrate that the intervention, and not something else, is responsible for the observed outcome. Level I evidence is the most definitive for establishing causality, with greatest reduction in bias. Level IV evidence can only hint at causality; level V evidence only suggests the possibility.[10]

Reliability between the grading of the levels of evidence assigned by the different evaluators was tested in pairs, using an un-weighted Kappa coefficient. The Kappa coefficient varied between 0.608 and 0.919. In a second step, all discrepancies were discussed. The raters argued the reasons for the score given. In case an agreement could not be found in this way, the score with the highest frequency was chosen (2 out of 3 raters scoring the same level of evidence). This final consensus score was used in the summary tables.

Table 1. Hierarchy of levels of evidence based on research design types (Amercan Academy of Cerebral Palsy and Developmental Medicine)

Level	Intervention (group) studies	Single Subject Research Design (SSRD)
I	Systematic review of randomized controlled trials (RCT's) Large RCT's with narrow confidence intervals (n>100)	Randomized controlled n-of-1 (RCT) Alternating treatment design Concurrent or non-concurrent multiple baseline Generalizability if the ATD is replicated across three or more subjects and the MBD consists of minimum of three subjects, behavior setting. These designs can provide causal interferences
II	Smaller RCT's with wider confidence intervals (n<100) Systematic reviews of cohort studies Outcomes research (large ecologic studies)	Non-randomized controlled concurrent MBD Generalizability if design consists of minimum three subjects, behaviours or settings Limited causal interferences
III	Cohort studies with concurrent control group Systematic reviews of case control studies	Non-randomized, non-concurrent, controlled MBD Generalizability if design consists of minimum of three subjects, behaviours or settings. Limited causal interferences.
IV	Case series Cohort studies without concurrent control group (eg historical control group) Case-control study	Non-randomized controlled SSRD with at least three phases (ABA, ABAB, BAB etc) Generalizability if replicated across three or more different subjects. Only hints at causal interferences.
V	Expert opinion Case study or report Bench research Expert opinion based on theory or physiologic research Common sense - anecdotes	Non-randomized controlled AB SSRD, generalizabiilty if replicated across three or more different subjects. Suggests causal interferences allowing testing of ideas.

Quality of the studies

The conduct of the study rating indicates the extent to which a study applied the controls possible within the research design. Quality assessment was again performed by the same 3 independent evaluators, using the conduct score system proposed by the AACPDM (**Table 2**). For group designs, the conduct of an individual study is judged as “Strong” (yes on 6–7 questions), “moderate” (score 4–5) or weak (≤ 3). For single subject designs, the conduct of an individual study is judged as strong (yes on 11–14 questions), moderate (scores 7–10) or weak (score < 7). Systematic reviews are also evaluated using this score system, reaching a maximum of 10 points. Inter-rater reliability of the conduct score system was tested resulting in an ICC score of 0.640 for group designs, 0.352 for single subject designs and 0.888 for systematic reviews.

Larger discrepancies were discussed in a similar way to the quality rating score. The answers of the different assessors to the questions were compared and the questions causing the disagreement were traced. Again, the reasons for the scores given were discussed until agreement was found and if no consensus could be found, the score with the highest frequency was chosen. After this discussion, the conduct score was recalculated. These consensus scores were used in the summary tables.

Table 2. Assessment of study conduct as proposed by the AACPDM

Conduct of group design studies
1. Were inclusion and exclusion criteria of the study population well described and followed?
2. Was the intervention well described and was there adherence to the intervention assignment? For 2-group designs, was the control exposure also well described?
3. Were the measures used clearly described, valid and reliable for measuring the outcomes of interest?
4. Was the outcome assessor unaware of the intervention status of the participants (i.e., were the assessors masked)?
5. Did the authors conduct and report appropriate statistical evaluation including power calculations? Both parts of the question need to be met to score 'yes'.
6. Were dropout/loss to follow-up reported and less than 20%? For 2-group designs, was dropout balanced?
7. Considering the potential within the study design, were appropriate methods for controlling confounding variables and limiting potential biases used?
Conduct questions for single subject designs
1. Was/were the participant(s) sufficiently well described to allow comparison with other studies or with the reader's own patient population?
2. Were the independent variables operationally defined to allow replication?
3. Were intervention conditions operationally defined to allow replication?
4. Were the dependent variables operationally defined as dependent measures?
5. Was inter-rater or intra-rater reliability of the dependent measures assessed before and during each phase of the study?
6. Was the outcome assessor unaware of the phase of the study (intervention vs. control) in which the participant was involved?
7. Was stability of the data demonstrated in baseline, namely lack of variability or a trend opposite to the direction one would expect after application of the intervention?
8. Was the type of SSRD clearly and correctly stated, for example, A-B, multiple baseline across subjects?
9. Were there an adequate number of data points in each phase (minimum of five) for each participant?
10. Were the effects of the intervention replicated across three or more subjects?
11. Did the authors conduct and report appropriate visual analysis, for example, level, trend and variability?
12. Did the graphs used for visual analysis follow standard conventions, for example x- and y- axes labeled clearly and logically, phases clearly labeled (A,B, etc.) and delineated with vertical lines, data paths separated between phases, consistency of scales?
13. Did the authors report tests of statistical analysis, for example celeration line approach, two-standard deviation band method, C- statistic, or other?
14. Were all criteria met for the statistical analyses used?
Conduct scores for systematic reviews
1. Were the search methods reported?
2. Was the search comprehensive?
3. Were the inclusion criteria reported?
4. Was selection bias avoided?
5. Were the validity criteria reported?
6. Was validity assessed properly?
7. Were the methods used to combine studies reported?
8. Were the findings combined appropriately?
9. Were the conclusions supported by the reported data?
10. What was the overall scientific quality of the overview?

Results

Stretching techniques

Two systematic reviews and 3 interventions studies were found that used stretching techniques **(Appendices, Table A).** The intervention studies included one RCT[12] and two single subject designs[11,13] ,all with moderate conduct scores. A total of 47 children with CP were included in these studies. Effectiveness was evaluated on the level of Body structure only and demonstrated level II evidence for the positive effect of passive stretching on ROM and spasticity.[12,13] Fragala et al. reported a significant deterioration in passive range of motion (pROM) after non-intervention periods longer than 5 weeks, but no significant change during the intervention periods.[11] A limited positive additional effect was found when stretching was combined with electrical stimulation[12] and heat application[13]. In addition, Lee & Ng indicated that sustained stretching of longer duration (minimum 30 s) was preferable to improve ROM and to reduce spasticity of the muscles around the targeted joints.[13]

The mean duration of treatment in the studies was 8.2 weeks (standard deviation (SD) 10.4), using a mean frequency of stretching of 4.5 times per week (SD 2.8). The 2 systematic reviews each included 7 studies and confirmed the weak evidence on the effectiveness of passive stretching.[14,15]

Massage

Four studies used different massage techniques in treatment of 125 children with CP (**Appendices, Table B**).

On body structure and function, one level II study found a significant positive effect on spasticity, ROM, fine-and gross motor function and cognitive behaviour.[16] However, similar effects were reported in the control group and the difference in progress between both groups was not evaluated. One level IV study found no significant effects on pROM and stretch reflexes[17], another study demonstrated significant improvements on Psychological well-being and Satisfaction with Life scales.[18] A similar study showed that children enjoyed the relaxing aspects of massage and reported a number of other improvements in their health, such as muscle relaxation, mobility and bowel movements and reduced pain.[19] On activity and participation level, Hernandez-Reiz[16] found significant positive effects on the Development Programming and Videotaped interactions, but Macgregor et al.[17] could not find significant improvements in gross motor function measurement (by the Gross Motor Function Measure; GMFM). On environmental factors, one level IV study demonstrated improved parent anxious behaviour and depressed mood, stress, self-efficacy and satisfaction with life, together with subjective feelings of their child's well-being and functioning after a 4-month training and support programme involving basic massage.[18]

The mean duration of the massage programme was 8.3 weeks (SD 2.9 weeks). Two studies reported a mean training frequency of twice per week, while in the two other studies the frequency was not specified.

Electrical stimulation

Of the 13 articles using electrical stimulation, 3 interventions used threshold electrical stimulation (TES, lower level electrical stimulation causing no visual contraction, < 10 mA)[22-22] and 8 intervention studies used neuromuscular electrical stimulation (NMES, higher level electrical stimulation causing visual contraction, > 10 mA)[23-32]. An overview of the studies using TES and NMES is given in **Appendices, Tables C & D**.

The studies using **TES** included a total of 59 children. The conduct of these studies varied from moderate[18] to weak[20-22]. On body structure and function, two level II studies based on a randomized controlled design found no significant long-term effect of TES on strength, ROM, deep tendon reflexes or muscle cross-sectional area of the m. quadriceps femoris and m. tibialis anterior[20,21]. Level IV evidence was found for the positive effect of 1 month of TES on ROM and selectivity.[22] On Activity level, only level IV evidence was found for standing/hopping on 1 foot.[22] Nevertheless, parents and children rated an important subjective feeling of improvement along with a high feeling of satisfaction.[20,21]

Seventy-eight children with CP were included in the 10 studies evaluating the effectiveness of **NMES**. On body structure and function, level II effectiveness was mainly demonstrated on strength[27], muscle cross-sectional area[27], ROM at the extremities[26,12,28] and the trunk[23] and on spasticity.[28,12,29] On activity level, two level II studies showed significant better effects of NMES on the quality of gait and walking velocity compared with the control group.[24,28] In a RCT design, Park et al. found significantly greater improvement on the sitting dimension of the GMFM in a group of children receiving electric stimulation of the abdomen and the back extensors compared with a control group.[23] This positive effect on gross motor function could not be demonstrated by van der Linden[25] and Kerr et al.[30] On participation level, Kerr et al.[30] measured statistically significant improvements on the Lifestyle Assessment Questionnaire and this provided level II evidence. The studies using TES were rather long-term and had a mean duration of 40.1 weeks (SD 23.9). The duration of the studies using NMES varied from 3 days to 12 weeks only, with a mean of 6.1 weeks (SD 4.7 weeks). The mean frequency of treatment was 4.95 and 9.37 times per week for TES and NMES, respectively.

One RCT compared TES with NMES, but showed no significant difference on body function and structure or on activity level.[30] One study compared the effectiveness of NMES with 15 s isometric voluntary contractions and reported improvements in strength and muscle morphology of the m. quadriceps femoris, but not in the triceps surae.[27] In contrast to NMES, volitional isometric strength training did not significantly influence walking speed.

Strength training

Twenty intervention studies and 6 systematic reviews evaluated the effect of strength training in children with CP. The studies were subdivided according to the specific modality of strength training used: isotonic strength training, functional strengthening exercises, isokinetic strength training, isometric strength training and mixed forms of strength training.

Appendices, Table E summarizes the 11 studies evaluating the effectiveness of **isotonic strength** training. A total of 128 children with CP was included. On the level of body function and structure, 7 out of 8 studies evaluating the effect of isotonic strength training on strength registered significant improvements[32,33,35,38,40-42], including one study demonstrating level II evidence. Other level II evidence was demonstrated on economy of movement.[39] Hints of causality or level IV evidence were found on muscle volume[42] , energy expenditure[36] and ROM[40]. A RCT by Dodd et al. demonstrated a significantly lower self-concept of scholastic competence and social acceptance in a group of 10 children with CP participating in a home based strength training programme compared with a control group.[37] However, another RCT by Unger et al. reported a significant improvement in the perception of body image of a group of children using circuit training.[39] On activity level, level II evidence was found for the effectiveness on gait[39,41] and gross motor function.[41] No studies evaluated the effect of isotonic strength training on participation level.

The studies using isotonic strength training reported only limited description of the resistance used during training. Three studies used a resistance of 65% of the one repetition maximum (RM) and one study used 80% of RM. The mean training frequency was 3.8 times per week, with a duration of 12.4 weeks. All studies using isotonic strength training scored moderate to weak based on conduct scores.

An overview of the different studies using functional, isokinetic, isometric and mixed strength training is provided in **Appendices, Table F&G**.

Four studies evaluated the effects of **functional strength training** and included a total of 40 children with CP. Functional strength training often consisted of a home-based training programme, including task-oriented strength training during functional activities. Effects of functional strength training were demonstrated on Activity level: gross motor function significantly improved in one level II study (46). A high-quality RCT by Dodd et al. demonstrated significant effects on muscle strength, but only a trend towards improved gross motor function.[44] Level IV evidence was also recognized for participation in school and leisure activities and self-perception.[45] The studies frequently used 8–10 repetitions and had a mean duration of 5.4 weeks at a frequency of 2.4 times per week.

Level II evidence was also found for **isokinetic strength training** and this on all levels of the ICF.[47,48]

None of the studies using any form of strength training demonstrated an increase in spasticity. Only 9 studies used a follow-up period, with an average duration of follow-up of 32 weeks. All these studies recognized a decrease in training results at all levels after a non-training follow-up period.

The studies demonstrate strong evidence of all forms of strength training on body structure and function, but to a lesser extent on activity and participation. **Appendices, Table H** provides an overview of the systematic review evaluating the effectiveness of strength training.[50-55] The results are confirmed by all except one meta-analytic review on strength training by Scianni et al. stating that muscle strengthening is not effective in children with CP.[55] However, since Scianni also included biofeedback and electrical stimulation under interventions of muscle strengthening the results cannot be compared with the results of this systematic review.

Endurance training

Eight intervention studies and two systematic reviews evaluated the effect of endurance training for children with CP, with a main focus on aerobic capacity **(Appendices, Table I).**[56-65] A total of 108 children were included in the intervention studies. Modalities used varied from walking and running to circuit training, cycling and mixed gymnastic exercises.

On body function and structure, significant level II effects were mainly found for aerobic capacity[56,60] , anaerobic capacity[60], muscle strength[60] , agility[60] and oxygen uptake[62]. On Activity level, level II significance was reported for improvements in gross motor function.[60,62] Improved Participation measured by use of the Children's Assessment of Participation and Enjoyment (CAPE) was reported by Verschuren et al.[60] Endurance training positively influenced quality of life (QoL) measured by the Health-Related Quality of Life (HRQoL) (level II evidence)[63] and self-perception of physical appearance (level IV evidence)[57].

The mean duration of the training programmes was 13.3 weeks (SD 13.1). Four studies used a follow-up period with a mean of 9.3 weeks.[57,59,60,63] Weak to moderate conduct scores were found for all interventions.

All studies reported little to no deterioration when comparing the measurements after follow-up with the immediate post-training results. The two systematic reviews on aerobic training both confirm the overall positive effects of endurance training in children with CP.[64,65] Nevertheless, both reviews report low methodological qualities of the studies and lack of information on the effectiveness on daily activity and participation level.

Weight-bearing

Of the selected studies, 6 intervention studies and 1 systematic review investigated the effectiveness of weight-bearing activities in children with CP **(Appendices, Table J)**.[66-72] A total of 89 children was included in the studies. Included modalities were standing programmes in different types of walking frames and PT programmes focusing on weight-bearing exercises. The studies were of very low to moderate quality.

On body structure and function, two level II studies demonstrated a positive effect of increased weight-bearing on bone mineral density.[66,69] However, level IV evidence was demonstrated for a positive effect on bone mineral density[68], ROM[67,70] and bowel activity[71]. A positive effect on the behavioural characteristics and personal feeling of improved daily functioning was demonstrated with level IV evidence.[68]

On activity and participation level, two level II studies evaluated the effectiveness of weight-bearing programmes on activities of daily living (ADL), functional performance and walking speed, but this did not yield statistical significance.[70,71]

The duration of the standing programmes was very variable and ranged from 2 weeks to 9 months, with a frequency ranging from 2 to 5 times per week.

Balance training

Five intervention studies and one systematic review evaluated the effectiveness of different forms of balance training for a total of 43 children with CP, each using different modalities **(Appendices, Table K**.[73-78] Three studies used a moveable platform to train balance, with one study using additional visual feedback.[74-76]

On body structure and function, one study targeting specific sitting balance found significant positive effects on trunk control, as well as in the extremities.[73] The studies mainly evaluated on Activity level and registered task-specific improvement meaning a reduced centre of pressure (CoP) area and a reduced stabilization time. One level II study also reported a more symmetrical walking pattern after a 6-week training programme and one level III study reported visible improvements in GMFM-D (no statistics) after a 5-day training programme. One study using multidimensional random perturbation also evaluated on activity level only and found level II evidence for significant improvement on gross motor function together with an improved mechanical efficiency.[77]

The systematic review including 12 studies, reported low quality experiments providing low levels of evidence on the effectiveness of studies targeting postural control and balance.[78]

Treadmill training

In the last years, treadmill training has become a very popular therapy method in CP. Three systematic reviews and 10 interventions studies (including 61 children) evaluated the effect of treadmill training in children with CP, with different objectives, such as gait rehabilitation, functional improvement and improvement of aerobic capacity **(Appendices, Table L)**.[79-91] Training modalities varied strongly among the different modalities, such as body weight support, treadmill speed (range 0.25 km/h up to 5 km/h or as fast as possible) and duration (10–30 min).

On the level of body structure and function, weak level IV evidence was reported for positive effects on energy expenditure[85] and H-reflex[87]. Effects of treadmill training in children with CP were mainly limited to activity level. Two level II[81,83] and 5 level IV studies[82,84,85,87,88] found significant effects on different gait parameters. One level II[84] and one level IV[88] reported a significant positive effect on gross motor function. The mean duration of the training periods was 6.4 weeks (SD 5.7 weeks) at a mean frequency of 6.7 times per week (SD 5.21). Three studies used a follow-up period, with a mean of 10.08 weeks. Results at follow-up are concurrent with the results of endurance training and also show very limited deterioration during the follow-up period.

Summary of the results

Table 3 provides an overview of the evidence of the different interventions. For this summary table, the most commonly used outcome parameters on the different levels of the ICF were selected.

It compares the number of studies finding statistical significant effects on this parameter to the total number of specific interventions evaluating this parameter. Some level II RCTs did not statistically compare between-group differences in progress. Therefore, in Table 3, these studies are assigned to the level IV studies. Although the design of these studies could be rated as level II, statistically significant differences within in the experimental or control group only can be compared with the analysis of a level IV case series.

On the level of body structure and function, the parameters spasticity, ROM, bone mineral density, energy expenditure, strength and muscle morphology were the most commonly used. As demonstrated in Table 3, level II evidence was most frequently demonstrated for the effectiveness of all forms of strength training on muscle strength. More conflicting evidence was found for the influence of NMES and endurance training on muscle strength. Weight-bearing positively influenced bone mineral density. On the level of activity and participation, level II evidence for positive effects on different gait parameters were also demonstrated by different forms of strength training. In addition, treadmill training, NMES and balance were methods that significantly improve gait. The same is valid for effectiveness on gross motor function. Limited evidence was available on the most useful methods to improve participation, but this is mainly because studies rarely used participation as an outcome measure.

Table 3a. Overview of the number of studies demonstrating level II evidence

	Level II evidence		
	Muscle tone	**Bone mineral density**	**Energy expenditure/movement efficiency**
	stretching (1/1)[12]	weight bearing (2/2)[69,72]	isotonic strength training (1/1)[39]
	NMES (1/2) [24,**28**]		functional strength training (1/1)[46]
	TES (0/2) [20,21]		balance training (1/1)[77]
Body structure and function	**ROM**	**Strength**	**Muscle morphology/cross sect area**
	stretching (1/1)[12]	NMES (1/3) [25,**27**,30]	isometric strength training (1/1)[27]
	NMES (1/4)[**12**,23,24,25]	TES (0/1)[20]	NMES (1/1)[27]
	TES (0/2)[20,21]	isotonic strength training (1/1)[41]	TES (0/1)[21]
		functional strength training (1/2)[**44**,46]	
		isometric strength training (1/1)[27]	
		endurance training (1/2)[56,**60**]	
	Gait	**Gross motor function**	**Participation**
	NMES (1/3)[**24**,25,27]	NMES (1/3)[23,25,30]	endurance training (1/1)[60]
	TES (0/1)[20]	TES (0/2)[20,21]	NMES (1/1)[30]
Activity and participation	isotonic strength training (2/2)[39,41]	isotonic strength training (2/2)[39,41]	
	isometric strength training (1/1)[27]	functional strength training (1/2)[**44**,46]	
	treadmill training (1/1)[83]	endurance training (2/2)[60,62]	
	balance training (1/1)[75]	balance training (0/1)[77]	

Abbreviations:

…/… indicates the number of studies reaching significant treatment effects versus the total number of studies evaluating the effect of the interventions on that specific parameter

In case of conflicting evidence, the reference demonstrating significant effects are in bold

ROM: Range Of Motion; **NMES:** Neuromuscular Electrical Stimulation; **TES:** Threshold Electrical Stimulation

Table 3b. Overview of the number of studies demonstrating level III and IV evidence

Level III and IV evidence

	Muscle tone	**Bone mineral density**	**Energy expenditure/movement efficiency**
	stretching (1/1)[13]	weight bearing (1/1)[74]	isotonic strength training (1/2)[34,**39**]
	NMES (2/2)[28,29]		treadmill training (1/1)[85]
	treadmill training (0/1)[81]		isokinetic strength training (0/1)[47]
	massage (1/2)[16,17]		
Body structure and function	**ROM**	**Strength**	**Muscle morphology/cross sect area**
	stretching (2/2)[11,13]	isotonic strength training (6/7)[**32,33**,34,**35,38,40,42**]	isotonic strenght training (1/1)[13]
	TES (1/1)[23]	functional strength training (1/1)[43]	
	NMES (2/2)[28,29]	isokinetic strength training (2/2)[47,48]	
	isotonic strength training (1/3)[35,40,42]	endurance training (1/1)[57]	
	massage (1/2)[**16**,17]		
	weight bearing (1/3)[70,**71,73**]		
Activity and participation	**Gait**	**Gross motor function**	**Participation**
	NMES (1/1)[28]	TES (0/2)[20,21]	massage (3/3)[16,18,19]
	isotonic strength training (5/7)[**32,34**,35,**36,38,40**,42]	isotonic strength training (3/4)[**34,38,40**,42]	treadmill training (1/2)[**84**,88]
	functional strength training (1/1)	functional strength training (1/1)[43]	**Environmental factors**
	endurance training (1/1)[63]	isokinetic strength training (2/2)[47,48]	massage (1/1)[18]
	treadmill training (6/7)[79,**81,82,84,85,87,88**]	endurance training (2/2)[61,63]	isokinetic strength training (1/1)[48]
	isokinetic strength training (1/1)[48]	treadmill training (2/5)[79,**81**,84,85,**88**]	weight bearing (1/1)[74]
		massage (1/2)[**16**,17]	**Quality of Life**
		balance training (1/2)[73,74]	treadmill training (1/1)[86]
			isokinetic strength training (1/1)[48]

Abbreviations:

…/… indicates the number of studies reaching significant treatment effects versus the total number of studies evaluating the effect of the interventions on that specific parameter

In case of conflicting evidence, the reference demonstrating significant effects are in bold

ROM: Range Of Motion; **NMES:** Neuromuscular Electrical Stimulation; **TES:** Threshold Electrical Stimulation

Discussion

This systematic review evaluated the effectiveness of different basic PT techniques used in the treatment of children with CP. Eighty-three articles were included, comprising 27 RCTs, 16 systematic reviews, 11 single-subject designs and 29 prospective, non-randomized trials. One study demonstrated level I evidence, 44 studies level II, 2 studies level III and 34 studies level IV. Since case-studies and non-systematic reviews were excluded, only 2 studies were included with level V evidence. These numbers demonstrate that, high-quality research is possible and being done. It should be noted that studies demonstrating high levels of evidence did not necessarily reach high conduct scores. even well-designed RCTs often showed methodological difficulties, such as a small number of participants, a lack of control over different confounding variables and an inappropriate description of the exercises used in the control group. Statistically, RCT designs often missed an appropriate comparison of between-group differences in treatment effects. on the other hand, some single-subject research designs (SSRDs) and prospective interventions used a relatively high number of participants and reached high conduct scores with an adequate control over confounding variables. For this reason, lower level of evidence designs were also included in this systematic review. Nevertheless, the authors support the AACPDM statement that level IV and V studies only hint at causality and the results of these studies should be interpreted very cautiously. Low to moderate agreement was found for the rating system on the conduct score system and this was specifically a problem for the SSRDs. The evaluators encountered difficulties when scoring the last questions regarding visual and statistical analysis. These questions were obviously more open for interpretation and therefore more subject to individual opinion.

Other weaknesses of the conduct score system was the fact that it did not consider the number of subjects in the studies and also did not take into account that some questions may be more critical than others. The conduct scoring system therefore was found to be valuable in giving a general score for the quality of the interventions, but should also be interpreted cautiously. Only one study reported adverse effects. Dodd et al.'s results suggested that participation in a relatively short home- based strength training programme may have an inhibitory effect on the self-concept of children with CP.[37] despite the inhibitory effect, self-concept in the experimental group remained positive after strength training, suggesting that clinicians should not be overly concerned about the psychological effects of the intervention. However, this study does demonstrate that we have to appreciate the fact that children with CP may show different values from their therapists.

A wide variety of frequencies and intensities were used in the intervention studies, and thus the optimal training modalities remain open for discussion. For strength training, many different types of resistive exercises were used and in physical fitness or endurance training, different training heart rates and durations of trainings were used. This indicates a need for structured research comparing different training intensities, which is necessary to develop more specific clinical guidelines.

Strength interventions often do not respect adequate training principles according to the training recommendations by the National Strength and Conditioning Association[92]: the age of the children is often not appropriate, the training intensity is often insufficient or the duration of training is too short. This might be another reason why Scianni et al.[55] and Verschuren et al.[93] did not find significant results regarding the effectiveness of strength training.

Another important issue is the fact that a lack of evidence does not always reflect a lack of effectiveness. In particular, the lack of control over confounding variables was often a major problem limiting the studies to demonstrate evidence. Careful interpretation is advised, considering the large numbers of studies evaluating, for example, strength training in comparison with a much lower number of studies evaluating the effectiveness of massage or stretching. In addition, a higher number of studies evaluated the effectiveness of the level of body structure and function in comparison with the number of studies using outcome parameters on activity and participation level. Older studies in particular often restricted their outcome parameters to the level of body structure and function.

None of the studies differentiated outcome effects according to the age or functional level of the participants. However, many studies use a wide age range and a large variety in functional level in their inclusion criteria. Nevertheless, a young child with mild unilateral CP might benefit from a completely different treatment approach with different techniques from a teenager with severe bilateral CP. Concerning effects on gross motor function, in particular, it is important to consider the expected outcome related to age and GMFCS level. A younger child with a GMFCS level I or II usually develops more in the natural course of development than an older child with a GMFCS level IV or V.[94-96]

A restriction of the ICF model was found in the scoring of the parameter QoL, a problem that was previously reported in the literature.[97] The World Health Organization (WHO) defines QoL as “an individual’s perception of their position in life in the context of the culture and value system in which they live and in relation to their goals, standards and concerns”.[98] WHO suggests that QoL can be broken down into different domains: physical, psychological, social relationships and environmental. Given the inclusion of personal and environmental factors in its model of functioning and disability, the ICF encompasses all domains that comprise human life and thus impact QoL. The components of functioning and contextual factors can be seen as the various manifestations of a person’s QoL. In this systematic review, not many interventions used QoL measures in their outcome parameters. However, to be able to give this discussion a place, the authors have scored these parameters under a separate category of QoL.

This systematic review did not include mixed or eclectic approaches. This decision was based mainly on the methodological limitations of mixed interventions to distinguish the causes of the different reported effects. Nevertheless, the results of this review indicate that, in clinical practice, one might have to combine different techniques and methods in order to have a meaningful outcome.

Summarizing the effectiveness of all these interventions demonstrates that the ICF model provides a good model to evaluate the effectiveness of different physiotherapy interventions for CP. The results reveal very limited interactions between the different levels of the ICF. In general, interventions targeting problems at body function and structure, mainly influence body structure and function with only limited overflow to activity level. Stretching only significantly improved ROM and spasticity, without evidence of improved ROM during walking or other functional activities. only for the different forms of strength training and for NMES, effectiveness was also demonstrated on activity level. Likewise, interventions at activity level directly influenced motor functions, but limited evidence was available for direct influence on impairment problems such as strength, ROM or spasticity. It was even more challenging to look for interaction effects of participation and other ICF levels.

This leads to the conclusion that a targeted treatment approach based on a complete and extensive evaluation on all levels of the ICF is necessary to create an appropriate treatment plan. In true efforts to increase independence and to prevent secondary disabilities, the child should be considered as a whole. The PT treatment plan should therefore comprise specific goals and exercises targeting the individual problems specified on each level of the ICF. Specific motor learning strategies might be necessary to integrate the different components of impairment level during functional activities. The effectiveness of different general conceptual approaches will therefore be investigated in a subsequent paper.[100] The aim of developing further specific clinical guidelines cannot yet be fulfilled with the present paper. Only a limited number of clinical messages could be deduced. Developing clinical guidelines should, however, be an important extra step, to be performed in a systematic way and based on both literature and expert opinion.

Conclusion

- Interventions targeting problems at body function and structure generally influenced this level without significant overflow to activity level and vice versa.
- Stretching can be useful to improve ROM in children with CP. The stretch should preferably be maintained for a minimum of 30 s.
- Conflicting evidence is available on the effectiveness of electrical stimulation strength. NMES (visual contraction, > 10mA) is preferable.
- Strength training is very effective in improving muscle strength and into a lesser extends to improve gait and motor function. Resistance of 65–80% of one RM seems to be well tolerated by the children. Effects seem to be lost relatively quickly after stopping the programme.
- Treadmill training is beneficial in improving gait and endurance in children with CP. With partial body weight support, treadmill training can be effective in very young children with CP.
- Endurance training is useful to improve aerobic endurance. effects seem to last minimally as long as the training programme itself. A training heart rate of 75% of the maximum heart rate seems to be well tolerated.
- Massage improves the feeling of well-being in children with CP and their parents.
- Balance training is optimally trained in a task-specific context.
- Weight-bearing is useful to improve bone mineral density in children with CP.

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Chapter 2

The evidence-base for conceptual approaches and additional therapies targeting lower limb function in children with cerebral palsy : A systematic review using the International Classification of Functioning, Disability and Health as a conceptual framework

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Abstract

Objective

This systematic review provides an overview of the effectiveness of conceptual approaches and additional therapies used in lower limb physical therapy of children with cerebral palsy and supports the development of clinical guidelines.

Data sources and study selection

A literature search in 5 electronic databases was performed, extracting literature published between 1995 and 2009. Studies were evaluated using the framework recommended by the American Academy for cerebral Palsy and Developmental Medicine (AACPDM), which classifies outcomes according to the International Classification of Functioning, Disability and Health (ICF). Data extraction: three evaluators rated the strength of evidence of the effects according to the AACPDM levels of evidence classification, and the quality of the studies according to the AACPDM conduct score system.

Data synthesis

A total of 37 studies used conceptual approaches (neurodevelopmental treatment (NDT), conductive education, Vojta therapy, sensory integration, functional training and goal-oriented therapy) and 21 studies focused on additional therapies (aquatic therapy and therapeutic horse-riding).

Conclusion

Level ii evidence was found for the effective- ness of therapeutic horse-riding on posture and for NDT and functional training on gross motor function. Goal-oriented therapy and functional training were effective on the attainment of functional goals and participation. With level IV evidence, NDT was effective on all levels of the ICF.

Introduction

Cerebral palsy (CP) describes a group of disorders of movement and posture that cause activity limitations. CP is attributed to non-progressive disturbances occuring in the developing foetal or infant brain.[1,2] The effectiveness of different basic physical therapy (PT) techniques in the treatment of children with CP has been described in a previous article.[3] The results of that analysis revealed limited interaction effects between the different levels of the International Classification of Function, Disability and Health (ICF) of the World Health Organization (WHO), thereby suggesting that specific neurological approaches and motor learning strategies might be necessary to facilitate these interaction effects.

Different principles of motor learning can be recognized. Neurophysiological and neuromaturational approaches are based largely on assumptions drawn from the neuromaturational theories of development. The main assumption of these theories is that the development of movements and motor skills result solely from the neurological maturation of the central nervous system (CNS): higher centres inhibit and control lower centres, thereby allowing voluntary movement.[4] That initial theory of motor development has evolved, with more recent theories of motor learning emphasizing that motor behaviour or developing behaviours should not be viewed as the unfolding of predetermined patterns represented in the CNS. These approaches favour a more heterarchical view, in which motor development and coordination are assumed to emerge from the dynamic interaction of many subsystems in a task-specific context. These approaches, therefore, are based on an active rather than a passive view of motor learning. People learn by actively attempting to solve the problems inherent to a functional task.[5,6]

In treatment of CP, various approaches are based on differ- ent theories of motor learning. Commonly used approaches in treatment of children with CP are neurodevelopmental treat- ment (NDT) or Bobath therapy, conductive education (CE) by Petö, reflex locomotion therapy by Vojta, and functional task-oriented training. Other methods used are the patterning method by Doman & Delacato, and sensory integration by Ayres.

NDT or Bobath therapy (Karl & Bertha Bobath, 1943) is an interdisciplinary problem-solving approach to the assessment, treatment and management of individuals with changes in sensorimotor, perceptual and cognitive function, tone and patterns of movement resulting from a CNS lesion.[7,8] In the last decade, especially, the use of NDT in treatment of children with CP has been controversial, with the most common concern that NDT insufficiently targets functional activities and participation by using only neuromaturational strategies in learning.[9]

CE (Andreas Petö, 1945) is based on an educational rather than on a medical model of intervention. CE integrates edu- cational and rehabilitation goals into a single programme to assist children with motor dysfunction to attain orthofunction, enabling them to attend school with maximum independence .[10] Repetitive learning is a central aspect within CE. In the same way as for NDT, several objections have been raised against CE, and cost-effectiveness, especially, is seen as a major concern.[11]

In the Vojta method (Vaclav Vojta, 1968) normal patterns of movement sequences, for example, reaching and grasping, standing up and walking, are not taught or trained as such. Vojta therapy rather stimulates the brain, activating “innate, stored movement patterns”, which are then “exported” as co- ordinated movements involving the musculature of the trunk and extremities.[12]

Functional and task-oriented training are based on learning motor abilities that are meaningful in the child’s environment and perceived as problematic by either the child or the parents.[5,6] The child has an active role in finding solutions to motor problems rather than the physical therapist providing a solution using handling. Functional goals are established with parents and children based on their priorities and functional activities are assumed to be learned by repetitive practice of goal-related tasks in functional situations.[5,6]

A closely related approach is the method of individual goal setting, as proposed by Bower et al..[13] Like functional and task-oriented training, this strategy uses the identification of individually defined tailor-made goals to structure the therapy process.[13] However, the approach is not necessarily based on repetitive learning, but sometimes uses an eclectic approach or a mixture of different techniques.

The patterning method (Doman & Delacato, 1955) derives from a phylogenetic interpretation of development. The therapeutic programme aims to recapitulate the physiological stages of motor development through exercises, by the involuntary imposition of patterns of activation or by having the child voluntarily practice, presumably missed, earlier stages of mastery.[14]

Sensory integration (Jean Ayres, 1960) refers to a theory and a neurological process that enables the individual to take in, interpret, integrate and use the spatiotemporal aspects of sensory integration from the body and the environment to plan and produce organized behaviour. It postulates that learning is dependent on the ability of normal children to take in sensory information derived from the environment and from movement of their bodies, to process and integrate these sensory inputs within the CNS and to use this sensory information to plan and produce organized behaviour.[15]

Besides these conceptual approaches, therapeutic horse- riding or hippotherapy and aquatic therapy or hydrotherapy are frequently used additional therapies for children with CP, with many different opinions regarding their effectiveness.[16,18]

Despite the frequent use of these treatment strategies, the effectiveness of these interventions is not clear and, especially, effectiveness on the different levels of the ICF has not been elucidated. Little consistency is provided on the use of these different approaches in clinical practice of lower limb treatment in CP. However, a differentiation of the effects supports a targeted treatment approach based on an adequate selection of intervention type adjusted to the specific therapy goal.

This systematic review aims to summarize the effectiveness of different conceptual approaches and additional therapies used in children with CP and to differentiate the outcome effects on the different levels of the ICF. It thereby aims to identify interaction effects between the different outcome levels of the ICF and to explore the possibilities of developing clinical guidelines in PT treatment of children with CP.

Method

Search strategy

A systematic, stepwise search of the literature on PT in CP was performed using the following electronic databases: Web of Science, PubMed, Cochrane Library, Physiotherapy Evidence Database (PEDro) and CINAHL. General search terms used were: “cerebral palsy” and “physiotherapy”, “physical therapy”, “exercise”, and “training”. More specific search terms were: “functional training”, “functional therapy”, “neurodevelopmental treatment”, “Bobath”, “Petö”, “conductive therapy”, “conductive education”, “Vojta”, “reflex locomotion”, “patterning”, “doman delacato”, “sensory integration”, “hydrotherapy”, “aquatic therapy”, “hippotherapy”, “horse-riding” and “goal-setting”.

Inclusion criteria were: original articles published in peer-reviewed journals between January 1995 and December 2009, focusing on PT interventions targeting lower limb and trunk treatment in children and adolescents (< 18 years) with CP. Only articles written in English were included. Articles including children with different pathologies or only targeting the upper limb were excluded from the study, as were interventions using mixed approaches or techniques.

Based on the title and abstracts of the articles, a first selection resulted in 127 articles. All articles were subsequently screened by the first author. Articles not meeting the inclusion criteria were withheld. If the title and abstract did not provide sufficient information to fulfil the inclusion criteria, the full article was checked. In addition, all case studies, expert opinions and non-systematic reviews were excluded. As a final step, the reference lists of all systematic reviews included in the study were searched, and missing articles meeting the inclusion criteria were added. The inclusion of doubtful articles was discussed with a second and third assessor.

A resulting total of 58 studies was included in the study. A flow diagram of the selection process is shown in **Fig 1.**

Data collection

The full text of all selected articles was read. The following data were extracted: type of intervention, numbers of patients included, topographic distribution of cerebral palsy, age of patients, type, frequency and duration of intervention, duration of follow-up, evaluation method and timing, summary of the results, and conclusion.

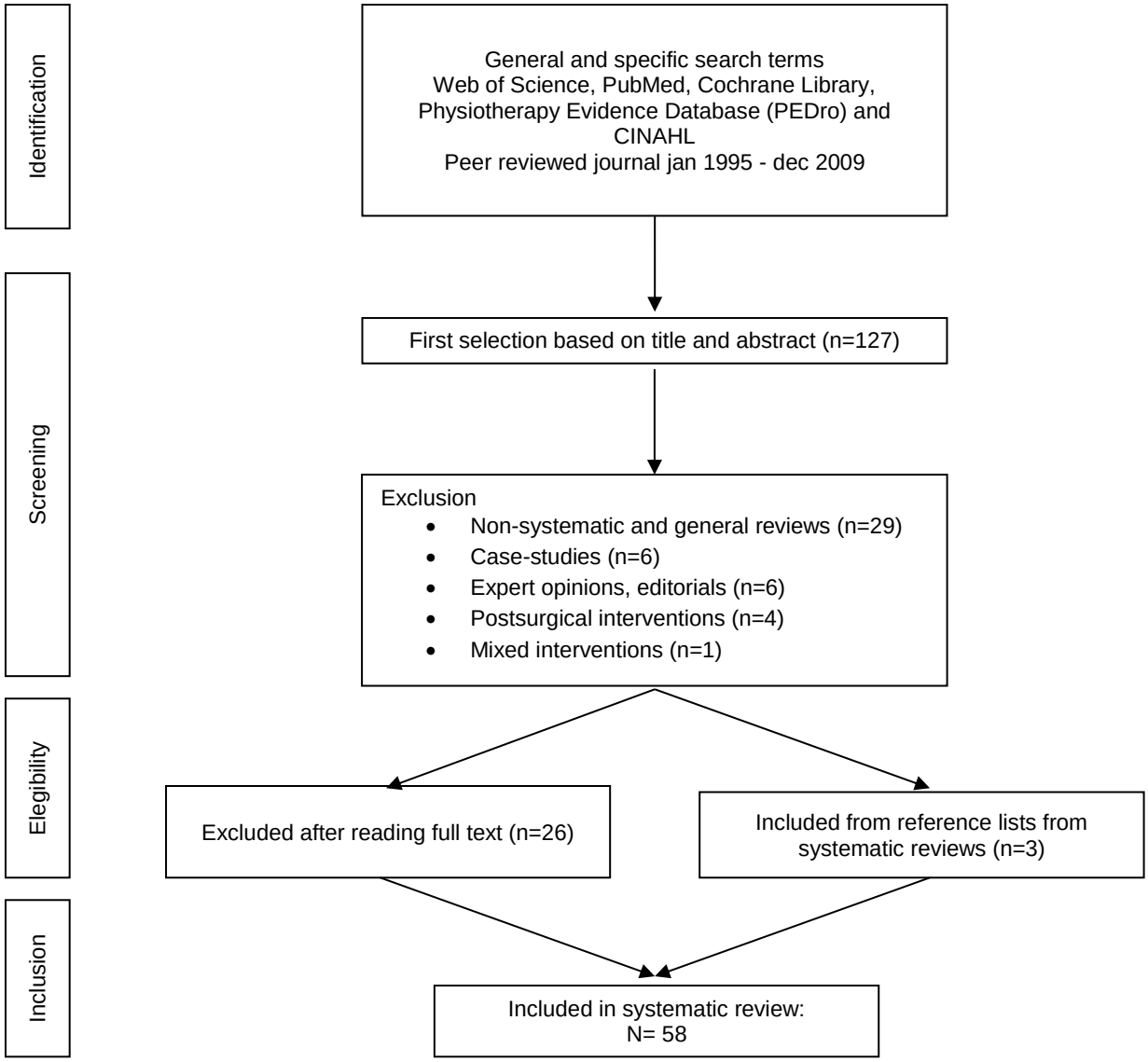

Fig 1. Flow chart describing the selection process

Grouping data

The selected articles were subdivided into two categories: a first group of articles covering the conceptual PT approaches (n = 37), and a second group of articles focusing on additional therapies (n = 21).

Classification and rating of the different outcome measures

Classification of outcome measures, rating of the level of evidence and scoring of conduct scores was carried out by 3 independent evaluators: 1 PhD physical therapist (CVdB), 1 research physical therapist (IF) and 1 physical therapist MSc student (CD).

Classification of the outcome

The evaluators classified the outcome of the intervention on the level of the ICF model: body structure and function, activities and participation, personal factors and environmental factors.

Body structures are defined as the anatomical parts of the body, such as organs, limbs and their components. Body functions are the physiological functions of body systems (including psychological functions). Activity is the execution of a task or action by an individual. Participation is involvement in a life situation. Environmental factors make up the physical, social and attitudinal environment in which people live and conduct their lives. They can be viewed as facilitators (positive influence) or barriers (negative influence). Personal factors are the particular background of an individual's life and living and comprise features of the individual that are not part of a health condition. These factors may include gender, race, age and other health conditions.[19]

Level of evidence

The same 3 independent evaluators rated the studies according to their level of evidence using the rating system proposed by the American Academy of Cerebral Palsy and Developmental Medicine (AACPDM).[20] Level of evidence describes the potential in a research study design to control for factors, other than the intervention, that may affect the observed outcome. In descending order, the levels of evidence decreasingly demonstrate that the intervention, and not something else, is responsible for the observed outcome. Level I evidence is the most definitive for establishing causality, with greatest reduction in bias. Level IV evidence can only hint at causality; level V evidence only suggests the possibility.[20] Any discrepancies were discussed and a final agreement score was used.

In a first step, agreement between the grading of the levels of evidence assigned by the different evaluators was tested in pairs, using a Kappa coefficient. The resulting agreement scores varied between 0.604 and 0.780.

As a second step, all discrepancies were discussed. The raters argued the reasons for the score given. If an agreement could not be found in this way, the score with the highest frequency was chosen (2 out of 3 raters scoring the same level of evidence). This final consensus score was used in the summary tables.

Quality of the studies

The conduct of the study rating indicates the extent to which a study applied the control possible within the research design. Quality assessment was performed using the conduct score system proposed by the AACPDM.[20] For group designs, the conduct of an individual study is judged as “strong” (yes on 6–7 questions), “moderate” (score 4–5) or weak (≤ 3). For single subject designs, the conduct of an individual study is judged as “strong” (yes on 11–14 questions), moderate (score 7–10) or weak (score < 7). Systematic reviews are also evaluated using a score system, reaching a maximum of 10 points. Validity assessment of the studies was performed by 3 independent evaluators. Similarly to the quality rating score, larger discrepancies were discussed and an agreement score was used. Inter-rater reliability of the validity assessment system was tested, resulting in an Intraclass Correlation Coefficient (ICC) score of 0.927 for group designs, 0.947 for single subject designs, and 0.906 for systematic reviews.

Similarly to the quality rating score, larger discrepancies were discussed. The answers of the different assessors to the questions were compared and the questions causing the disagreement were traced. Again, the reasons for the scores given was discussed until an agreement was found and if no consensus could be found, the score with the highest frequency was chosen. After this discussion, the conduct score was recalculated. These consensus scores were used in the results tables.

Results

Within the category of conceptual approaches, the included articles were grouped according to their named approach: Bobath or NDT (n = 13), Vojta or reflex locomotion therapy (n = 1), Petö or CE (n = 13), Doman Delacato or patterning (n = 0), a goal-oriented approach (n = 3), functional training (n = 6) and sensory integration according to Ayres theory (n = 1).

Neurodevelopmental treatment or Bobath therapy

Eleven intervention studies, which included a total of 181 children with CP, evaluated the effect of NDT (**Appendices, Table M**).[21,23] Five studies used a single-subject research design (SSRD), 3 studies were case series and 3 studies used a randomized control trial (RCT) design. The conduct scores thereby varied between weak (n = 2), moderate (n = 7) and strong (n = 2). The mean duration of the NDT treatment was 18.0 weeks (standard deviation (SD) 16.5 weeks) with a mean frequency of 4 times per week.

No level II evidence was found for the effectiveness of NDT on the level of body structure and function. Level IV evidence was found on posture[22], spasticity[25], range of motion (ROM)[25] and mechanical efficiency[29]. On activity level, level II and III evidence was found for the effectiveness of NDT on gross motor function.[26,28] Two other level II studies could not find significant effects on gross motor function: one study focused on treatment contexts[21] and one study on the frequency of intervention.[28] In these studies, however, significant improvements in gross motor function were registered in the intervention group (within- group differences), but no significant between-group differences were found when compared with the control group. One level III study found significant effects on participation and this on self-care and caregiver assistance.[26]

It should be noted that many of the studies were RCTs comparing different interventions, using NDT as a control intervention. Therefore, the results of these studies could not be scored as level II evidence and had to be scored as level IV evidence. Kerem compared a group of children receiving frequent NDT combined with Johnstone pressure splints (JPS) with a group of children receiving only NDT, and found a significantly higher improvement in ROM and spasticity in the combination group.[25] A high-quality RCT by Bar-Haim compared a group of children receiving Adeli Suit with a group of children receiving NDT.[29] The results demonstrated a superior effect of Adeli Suit treatment on mechanical efficiency during activities, while no differences were found when evaluated on activity level using the Gross Motor Function Measure (GMFM). In a complex SSRD comparing an AAB with an ABA design, Cherng compared the effectiveness of a NDT programme with a combined programme of NDT and body weight supported treadmill training.[30] The results of this study demonstrated no significant effects on gait parameters or gross motor function after the periods of only NDT.

Trahan & Malouin[27], Tsorlakis[28] and Christiansen[31] all focused on frequency of intervention. Where Tsorlakis underlined the importance of an intensive treatment regime of 5 times per week[28] Trahan & Malouin and Christiansen supported the feasible option of using intermittent periods with lower treatment frequencies. Both researchers demonstrated only a limited deterioration in gross motor function during these periods.[27,31]

There have been two previous systematic reviews evaluating the effectiveness of NDT in children with CP, which concluded that there was insufficient evidence to support NDT at the time of their review. Both reported the problems of evaluating and interpreting research results due to marked standardization problems in therapy, clinical and environment aspects.[32,33] The interventions evaluated by Butler & Darrah[32] and Brown & Burns[33] were much older and therefore there was only a limited overlap of 1 and 3 studies, respectively, with our review.

Conductive education according to Petö

Ten intervention studies evaluated the effectiveness of CE, including two RCTs and 8 case series and non-randomized controlled trials (**Appendices, Table N**).[10,34-45] A total of 185 children with CP were included in the intervention studies. The duration of the interventions was relatively long, with a mean of 30.1 weeks (SD 46.9) and high frequency of intervention of 4.6 times per week (SD 0.42). It was remarkable that 8 of the 10 interventions demonstrated weak conduct scores.

No level II evidence was found for the effectiveness of CE.

On body function and structure, the effectiveness of CE was only demonstrated with level IV evidence on language skills.[37] On activity level, the effectiveness of CE was demonstrated with level IV evidence on gross motor skills and individual motor goals.[36,40,44] Conflicting level IV evidence was available on participation[36,38,40,42] as well as on environmental factors parental coping and stress [34, 35, 38, 40]. The systematic reviews evaluating CE concluded positive effects of CE on motor function of children with CP, with effects comparable to the effects of different treatment approaches.[10,44,45] Training in a group probably had a significant social impact.

Sensory integration according to Ayers

One study was found evaluating the effectiveness of sensory integration (SI) in children with CP.[15] (**Appendices, Table O**) This study, based on a RCT, compared the effectiveness of sensory training, vestibular training, balance and postural reactions, bimanual activities and motor planning with the effectiveness of a home-based training programme. Effects were evaluated on Impairment and Activity level using the Ayers Southern Californian Sensory Integration test and on activity level using the Physical Ability Test. The results revealed significantly positive effects in favour of the children receiving sensory integration programme on both levels. Although this study had a level II design (RCT), the conduct score of this study was rated as “weak”. In addition, the study statistics were limited to within-group effect sizes.

Reflex locomotion therapy or Vojta therapy

One level II study evaluated the effectiveness of reflex locomotion according to Vojta.[46] **(Appendices, Table O)** and this only on activity level. Kanda evaluated a group of 5 children with CP receiving intensive Vojta therapy for 52 months. Comparing this group of children with 2 children re- ceiving no therapy and 3 children receiving insufficient therapy, revealed a significantly higher motor development level in the children following Vojta therapy. Taking into account the low quality (conduct score of 2/7) and the small number of participants in this study, no conclusions can be drawn on the effectiveness of Vojta therapy.

Patterning according to Doman Delacato

No study was found evaluating the effectiveness of patterning according to Delacato in children with CP.

Functional and task-oriented training

Six studies, including a total of 85 children, evaluated the effect of functional and task-oriented training, which mostly consisted of group training targeting specific activities of daily life.[4, 5, 47–50] (**Appendices, Table P**) These programmes were task-specific, with more attention on successful accomplishment of the specific tasks rather than quality of movement. Most of the studies used a variety of functional exercises and 1 study used a coordination dynamics board.[47]

On body structure and function, only weak evidence (level IV) was found for ROM, spasticity, and selective muscle activation.[50] No significant level II effects were found on strength. Effects of the functional training programmes were mainly found on activity level, with level II evidence on gross motor function and on different functional ambulation tests.[5,48,49] One level IV and 1 level II study reported improved Participation measured by Pediatric Evaluation of Disability Inventory (PEDI) self-care and mobility scores.[5,6]

The mean duration of training was 21.8 weeks (SD 23 weeks). Three studies included a follow-up, with a mean follow-up period of 10.3 weeks. The intensity of treatment ranged from very intense (2 times per day) to 3 times per month.

Goal-oriented approach

Three studies evaluated the effect of a goal-oriented approach in in total of 72 children with CP **Appendices, Table Q**).[13, 51, 52] Setting goals involves identify- ing and formulating standards of motor activity that are in advance of the child's current capacity or which slow down deterioration. Therefore, goals need to be formulated in such a way that there is no doubt about the extent to which they have been achieved when performance is reviewed.[13] In two RCTs (level II), Bower et al. compared the effect of therapy based on specific and SMART (Specific, Measurable, Attain- able, Realistic and

Timely) formulated goals with therapy based on general aims.[13,51] Significant effects were thereby highlighted on Activity level, with a positive effect on motor development evaluated by GMFM, but only in the short term. A more recent study by Löwing compared goal-directed functional therapy with activity-focused therapy.[52] This study demonstrated significantly clearer gains both on activity (gross motor function) and participation (measured with the PEDI) in the group of children receiving goal-directed functional therapy.

Within the category of additional therapies, a subdivision was made in a group of therapies using exercises in water (hydrotherapy, aquatic exercise) (n = 4) and horseback riding (therapeutic horseback riding, hippotherapy) (n = 14).

Aquatic therapy or hydrotherapy

Four studies evaluated the effect of different aquatic therapy interventions in 66 children with CP, varying from swimming training sessions to individual aquatic exercises (**Appendices, Table R**). [53–56] The mean duration of the aquatic training programmes was 17.5 weeks (SD 1.91), with a mean frequency of 2.75 times per week. Three studies used a RCT design; however, they were rated with very weak conduct scores.[53,54,56] on the other hand, the single subject design was rated with a moderate conduct score.[55]

On body structure and function, level II effectiveness was demonstrated on vital capacity after 26 weeks of swimming sessions.[53] Nevertheless, this study was rated as weak, based on the conduct score. Furthermore, positive effects of aquatic therapy were demonstrated on self-perception, body awareness and child behaviour.[54,56] on Activity level, level IV evidence was demonstrated as significantly improved water-orientation skills, improved functional mobility and gross motor function .[53,56]

Therapeutic horse-riding or hippotherapy

Fourteen intervention studies including 217 children evaluated the effectiveness of hippotherapy in children with CP **(Appendices, Table S**).[57–73] The mean duration of therapy was 12.8 weeks (SD 5.7 weeks) with 4 studies, including a follow-up period of a mean duration of 11 weeks. Four RCTs, 5 case series and non-randomized controlled trials and 5 single-subject designs were selected. The majority of the studies had moderate conduct scores.

On body structure and function, level II evidence was demonstrated on trunk and pelvic posture and stability[58, 63] and child behaviour.[57] Level IV evidence confirmed improved posture[60,61,67] and controversial effects on spasticity.[64, 66] On activities, level II evidence was only demonstrated on upper limb function.[57] In addition, 4 level IV studies demonstrated significant improvements on gross motor function.[59,62,64,65] One level II study could not find a statistically significant increase in participation.[69] Controversial findings were found in the 2 III–IV studies.[61,65] One level II study showed improvement in Quality of Life (QoL), but the results were not statistically significant.[69]

The results did not concur with the findings of the 3 systematic reviews on horse-riding therapy, which concluded that evidence is available for the effectiveness of horse-riding therapy on muscle tone of children with CP. Our review found that therapeutic horse-riding was more effective to increase trunk and pelvic control and, to a lesser extent, to improve gross motor function.[71-73]

Summary

Table 1 provides an overview of the evidence of the different interventions. For this summary table, the most commonly used outcome parameters on the different levels of the ICF were selected. Similar as in the review describing basic techniques, **Table 1** considers the results of level II studies not statistically analysing between-group differences as level IV evidence.

On the level of body function and structure, **Table 1** demonstrates that level II evidence was only obvious for the effective- ness of horse-riding therapy on different measures of posture and postural control and for aquatic therapy on lung function.

No effectiveness could be demonstrated on muscle strength or muscle cross-sectional area for the different interventions. An indication of the effectiveness was demonstrated by the level IV studies highlighting significant effects of therapeutic horse-riding on spasticity and energy expenditure.

On activity level, NDT, functional training, goal-oriented approach, sensory integration as well as Vojta therapy significantly influenced gross motor function and this was graded as level II evidence. However, the effectiveness of sensory integration and Vojta therapy was only evaluated in one study, both with a low conduct score, indicating careful consideration of these results. The effectiveness of CE, therapeutic horse-riding and aquatic therapy on gross motor function was only supported by level IV evidence. NDT was the only intervention that significantly improved different gait measures; however, these only reached level IV evidence.

Of all the studies, 16 used different participation outcome measures. Only two level II studies found significant effects on participation, including one level II study indicating significant effects of goal-directed therapy on social function measured using the PEDI.[62] The effectiveness of NDT on self-care skills, caregiver assistance and subjective findings by the parents was demonstrated with level IV evidence.[21]

Table 1a. Overview of the number of studies demonstrating level II evidence

	Level II evidence		
Body structure and function	**Spasticity**	**Posture**	**Energy expenditure/movement efficiency**
		Therapeutic horse-riding (2/3)[57,**58,63**]	
	ROM	**Strength**	**Muscle morphology/cross sect area**
		Functional training (0/1)[48]	
Activity and participation	**Gait**	**Gross motor function**	**Participation**
	Functional training (1/2)[**48**,49]	NDT (2/4)[**27,28**,29,31]	Goal oriented approach (1/1)[52]
		Goal oriented approach (3/3)[13,61,62]	Functional training (1/1)[5]
		Functional training (2/3**)**[**5**,48,**49**]	Conductive education (0/1)[39]
		Conductive education (0/2)[37,39]	**Environmental factors**
		Therapeutic horseriding (0/2)[57,69]	Conductive education (0/1)[37]
		Vojta therapy (1/1)[46]	**Quality of Life**
			Therapeutic horse-riding (0/1)[69]

Abbreviations:

…/… indicates the number of studies reaching significant treatment effects versus the total number of studies evaluating the effect of the interventions on that specific parameter

In case of conflicting evidence, the reference demonstrating significant effects are in bold

ROM: Range Of Motion; **NDT:** Neurodevelopmental Treatment

Table 1b. Overview of the number of studies demonstrating level III and IV evidence

Level III and IV evidence

Body structure and function	**Spasticity**	**Posture**	**Energy expenditure/movement efficiency**
	Therapeutic horse-riding (1/2)[64,68]	NDT (1/1)[22]	Therapeutic horse-riding (1/1)[59]
	NDT (1/2)[25,30]	Therapeutic horse-riding (3/3)[60,61,67]	NDT (1/1)[29]
	Functional training (1/1)[49]		Aquatic therapy (0/1)[55]
	ROM	**Strength**	**Muscle morphology/cross sect area**
	NDT (1/1)[20]	Aquatic therapy (0/1)[55]	
	Functional training (1/1)[50]		
Activity and participation	**Gait**	**Gross motor function**	**Participation**
	NDT (2/2)[24,30]	NDT (4/4)[24,26,29,30]	NDT (1/1)[26]
	Functional training (2/2)[48,49]	Conductive education (3/4)[36,40,41,42]	Functional training (1/1)[6]
	Aquatic therapy (0/1)[55]	Functional training (3/4)[6,48,49,50]	Conductive education (2/4)[36,38,40,42]
		Aquatic therapy (1/1)[55]	Therapeutic horse-riding (1/2)[61,65]
		Therapeutic horse-riding (5/6)[59,61,62,64,65,68]	**Environmental factors**
		Sensory integration (1/1)[15]	Conductive education(1/4) [34,35,38,40]

Abbreviations:

…/… indicates the number of studies reaching significant treatment effects versus the total number of studies evaluating the effect of the interventions on that specific parameter

In case of conflicting evidence, the reference demonstrating significant effects are in bold

ROM: Range Of Motion; **NDT:** Neurodevelopmental Treatment

Discussion

This systematic review overviews the effectiveness of different conceptual approaches and additional therapies used in PT of children with CP. A total of 52 articles were included in this analysis. Thirty-four included articles were graded as level II, 3 as level III, and 21 as level IV. As for the basic techniques, these numbers demonstrate that high-quality research in PT is possible and is being done. However, the validity assessment demonstrates an overweight of moderate and weak conduct scores, highlighting limitations in methodological conduct, such as appropriate statistical or visual analysis and description of the control groups. In addition, the number of individuals overall who have been studies is small.

Kappa coefficients for the level of evidence scores were similar to the agreement scores reported in the previous article summarizing basic techniques and were acceptable.[3] The ICC scores calculated for the conduct scores system, however, were much higher in this part of the systematic review (0.927 for group designs, 0.947 for SSRDs and 0.906 for systematic reviews) as in the part evaluating basic techniques (0.640 for group designs, 0.352 for single subject designs and 0.888 for systematic reviews).[3] This may be explained by the experience of the raters, who scored and discussed the articles described in the previous article first. To increase inter-rater reliability of the scores, appropriate training and experience in training might therefore be recommendable.

No adverse effects were demonstrated in any of the studies.

As for the individual techniques, summarizing the effectiveness of all interventions demonstrates that the ICF provides a good model to evaluate the effectiveness of different physiotherapy interventions for CP. As recognized in the systematic review describing basic techniques, only specific measurements of QoL could not be scored by the ICF.[3] In clinical research trials, however, limited interventions measure outcome effects on all levels of the ICF and again, especially older studies only evaluated effectiveness on the level of body structure and function.

Only the studies using NDT appear to demonstrate a tendency of effectiveness on all levels of the ICF: impairment, activity and, to a lesser extent, participation measures, although the results were inconsistent. Surprisingly, in contrast to the recent critiques of NDT being too passive and not sufficiently targeting activity and participation level, 2 level II[27, 28], 1 level III[26] and 3 level IV studies[23,24,30] highlight a significant effect on gross motor function. These studies demonstrate the recent developments within the neurodevelopmental treatment concept, with an obvious and important component of integration of improved muscle tone and length into function. The results refute the arguments that NDT only targets problems at impairment level without sufficient attention to activity level and functionality of the child. From a motor learning point of view, PT in an NDT context is considered not to be a repetition of the functional task, but is carefully considered by task-analysis. In other words, the child is not taught to do these skills as best he or she can despite the presence of spasticity or fluctuating tone. Rather, there is specific preparation for specific functional skills to enable the child to function in the most efficient way possible. The aim is to perform postural and voluntary

tasks with the least possible interference from abnormal postural tone.

The results of this review are not confirmed by the conclusions of the two high-quality systematic reviews by Butler & Darrah[32] and Brown & Burns[33], evaluating the effectiveness of NDT. Butler and Brown did not include the same studies in their analysis.[27,28] The studies in their reviews were much older (pre-2001) and therefore, there was a limited overlap of 1 and 3 studies, respectively, with this review. In addition, the interventions available for their reviews were of lower quality.

CE claims to increase orthofunction, which means that, in spite of fundamental motor problems, an individual acquires strategies to be as independent as possible in activities of daily living and lives as normally as possible. Thus, CE is expected to be an intervention at the activity and participation dimensions of the ICF. Indeed, regarding CE, level IV evidence was demonstrated only on gross motor function, No studies evaluated the effectiveness of CE on body structure and function parameters, such as strength, ROM and spasticity, but only on cognition and language development.

From a motor learning perspective, the repetitive learning principles from CE lean to a certain extent towards the functional and task-oriented approach. It is possible that the additional impact of the goal-setting procedures and the active learning component in the functional and task-oriented approach, have a clearer and more obvious impact on gross motor function and participation.

The results of the goal-oriented approach by Bower et al.[13,51] and the functional and task-oriented approaches[5, 6, 50] demonstrate that individual goal-setting procedures can be very effective on the attainment of individual functional goals and gross motor function. Most of the individual goals are only defined on activity level. In the short term especially, the definition of measurable, specific treatment goals can be very motivating for therapist, parents and children. on participation level, only Ahl et al.[6] , Ketelaar et al.[5] and Löwing et al.[52] found significant effect of task-oriented training on the PEDI, but no other measures of participation, such as Children´s Assessment of Participitation and Enjoyment or Assessment of Life Habits for Children were used. However, in the Measures of Process Care Questionnaire, parents reported positive feelings about their involvement in the therapy and the goal-setting process, suggesting a positive impact on the child's environment.[6,51]

These approaches use a more neuromaturational learning component, and, therefore, Vojta therapy especially would be expected to work in the body structure and function dimension of the ICF. Nevertheless, there were no measures in this dimension. The 1 outcome of statistically significant gross motor function score in the activity dimension may represent greater coordination of purposeful movement.

It would be interesting and useful to compare the effectiveness of these more "passive forms" of learning with the more recent "action" approaches. Ketelaar et al.[5] compared a functional task-oriented approach with an approach that was based more on normalization of quality of movement, like that of NDT. The major difference between both approaches was defined as the active learning component. Normalization of quality of movement, however, might not necessarily mean a passive learning component, but may also be learned in an active way and in a functional, task-oriented context.

In this regard, another important aspect that needs more detailed investigation is age and severity of involvement. Children who are more mildly involved might benefit from a different approach from those children with severe involvement and, maybe, young children would also benefit from another approach than older children.

Home-based training was not included, since these programmes often include a mixture of approaches and techniques, and a mixture of occupational therapy and physical therapy. However, the authors would like to state that this does not mean that they underestimate the value of home-based therapy. As demonstrated by Karnish et al., children with disabilities have been found to demonstrate superior performance of skills in natural educational settings compared with in an isolated therapy room.[21]

A recent approach that could unfortunately not be included this review, is context therapy, as proposed by Darrah et al.[74] and Law et al. [75]. This approach is also built on the theoretical construct of dynamical systems theory, which posits that motor behaviours are organized around functional tasks or goals and that the specific motor solution is influenced by the spontaneous interaction between variables from the child, the task and the environmental influences. Context therapy emphasizes changing the parameters of the task or environment rather than focusing on remediation of a child's abilities. The assumption of this approach is that changes in the task and/or environment will enable the child to perform an activity that they were unable to do previously.

This is a review of the research evidence for the effectiveness of the most commonly used therapy approaches as a first step in supporting the therapist to build an evidence-based targeted therapy programme based on the main problems of the child. It is based on the AACPDM review process, which uses the ICF as a two-part conceptual framework: the ICF to identify the types of evidence currently available, and a level of evidence classification to rate the strength of that evidence. The types of outcomes that have been studied so far are very few; outcomes for which we have positive evidence have not yet been adequately replicated. The robustness of the evidence is still too weak and the number of peadiatric studies is still too few to provide conclusive evidence. Therefore, it is still very early to define specific clinical guidelines. However, a targeted treatment approach based on appropriate evaluation of all levels of the ICF is advised to create an appropriate, individually-defined treatment plan. Integrating improvements at body function and structure during functional activities and vice versa appears to be very challenging for children with CP. Therefore, continuing high-quality research focusing on the motor-learning aspect of integrating these components remains necessary for future research.

Conclusion

- The effects of NDT are demonstrated at all levels of the ICF.
- The use of CE has significant effects on gross motor function.
- Setting individual, measurable goals supports the achievement of functional PT goals.
- Functional training can be beneficial in learning new motor activities, but no studies demonstrate benefit on the level of body function and structure.
- Hippotherapy can be considered as an effective additional therapy method to improve posture and postural control.
- Except for a beneficial impact on lung function, hydrotherapy is not yet proven to be effective in children with CP.

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Part 2
Clinical decision framework

Chapter 3

A clinical decision framework for the identification of main problems and treatment goals for ambulant children with bilateral spastic cerebral palsy

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Abstract

The primary aim of the study was to investigate how a clinical decision process based on the International Classification of Function, Disability and Health (ICF) and the Hypothesis-Oriented Algorithm for Clinicians (HOAC-II) can contribute to a reliable identification of main problems in ambulant children with cerebral palsy (CP). As a secondary aim, to evaluate how the additional information from three-dimensional gait analysis (3DGA) can influence the reliability.

Twenty-two physical therapists individually defined the main problems and specific goals of eight children with bilateral spastic CP. In four children, the results of 3DGA were provided additionally to the results of the clinical examination and the GMFM-88 (gross motor function measure-88). Frequency analysis was used to evaluate the selected main problems and goals. For the main problems, pair-wise agreement was calculated by the number of corresponding problems between the different therapists and using positive and negative agreement per problem. Cluster analysis using Ward's method was used to evaluate correspondence between the main problems and specific goals.

The pair-wise agreement revealed frequencies of 47%, 32% and 3% for the identification of one, two or three corresponding main problems. The number of corresponding main problems was higher when additional information of 3DGA was provided. Most of the specific goals were targeting strength (34%), followed by range of motion (15.2%) and GMFM-D (11.8%). In 29.7% of the cases, therapists could not prioritize and exceeded the number of eight specific goals. Cluster analysis revealed a logic connection between the selection of strength as a main problem and as specific goal parameters. Alignment as a main problem was very often associated with specific parameters like ROM and muscle length and with hypertonia as a main problem.

The results show a moderate agreement for the selection of main problems. Therapists are able to use the proposed model for a logic and structured clinical reasoning. Setting priorities in the definition of specific goals is revealed as a remaining difficulty. Further research is required to investigate the additional value of 3DGA and to improve priority setting.

Introduction

Cerebral palsy (CP) describes a group of disorders of movement and posture, causing activity limitations. CP is attributed to non-progressive disturbances occurring in the developing foetal or infant brain[1,2] Several research studies have demonstrated the positive effects of goal-setting in physical therapy (PT) of children with CP. It is accepted that involving both parents and children in the definition of realistic and acceptable goals improves motivation and thereby therapy outcome. [3-6]

Another aspect in goal-oriented PT is the potential benefit from targeted interventions to enhance training specificity. Goal planning, in that context, is being used to identify the tasks and contexts of particular interest.[7] In children with CP however, this can be very challenging. Children with CP usually face a heterogeneous variety of problems. Primary problems like muscle tone, muscle weakness and many others usually have inevitable effects on activity and participation. Many research studies therefore praise the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) as a clinical framework and an important rehabilitation tool. [8,9] Physical therapists can use the model to organize the characteristics of children's functioning, to guide the selection of measurement tools and to determine meaningful outcomes.[10,11]

Still, recent research investigating the relationship between the different levels of the ICF do not demonstrate a clear and causal relationship between measures of impairments and functional outcome.[10,12,13] Several studies identify spasticity and weakness as causal factors in gross motor function. Still, the amount of explained variance suggests that other variables should be included to explain functional outcome more fully.[14] As a physical therapist, it might therefore be very challenging to determine factors that influence outcome of the PT program.[15] Setting priorities is an essential but complicated part of a targeted approach, requiring an adequate strategy for clinical reasoning. The hypothesis-oriented algorithm for Clinicians II (HOAC-II) provides a structured approach for science-based clinical practice. It is focused on functional deficits and how impairments relate to these deficits.[16] Unlike the ICF, the HOAC-II is directly strategy-oriented and is designed to guide the clinical reasoning process for the physical therapist. By generating a hypothesis as to why a problem exists, the HOAC-II supports the reduction of an often long problem list to a refined list of key problems.[16] The ICF and the HOAC-II can therefore be considered as complimentary to each other.

Three-dimensional gait analysis (3DGA) is a commonly used measurement tool that supports the identification of main problems in ambulant children with CP. Still, the use of the results of 3DGA is mostly limited to the identification of key problems for orthopaedic surgery and botulinum toxin treatment. It is considered as an important additional measure that supports the selection of target muscles.[17,18]

To the best of our knowledge, no studies have investigated the role of a combined strategic approach and the use of 3DGA in the identification of specific PT goals. However, it is hypothesized that the results of the 3DGA can provide additional information to support the paediatric physical therapist to identify the main and key problems of the child. In that way, combined with a complete evaluation at all levels of the ICF and the clinical reasoning of the HOAC-II, it could provide a better understanding in the relationship between the impairments and functional outcome of an ambulant child with CP.

This study aims to investigate how a clinical decision process based on the HOAC-II and the ICF can contribute to a reliable identification of main problems and specific PT goals in ambulant children with CP. Additionally, it aims to evaluate how the additional information of three-dimensional gait analysis can influence the reliability of this information.

Methodology

The study was organized in different phases, starting from development of the clinical reasoning tool and validation by expert consensus to reliability testing by means of a larger scale agreement study **(Fig. 1).**

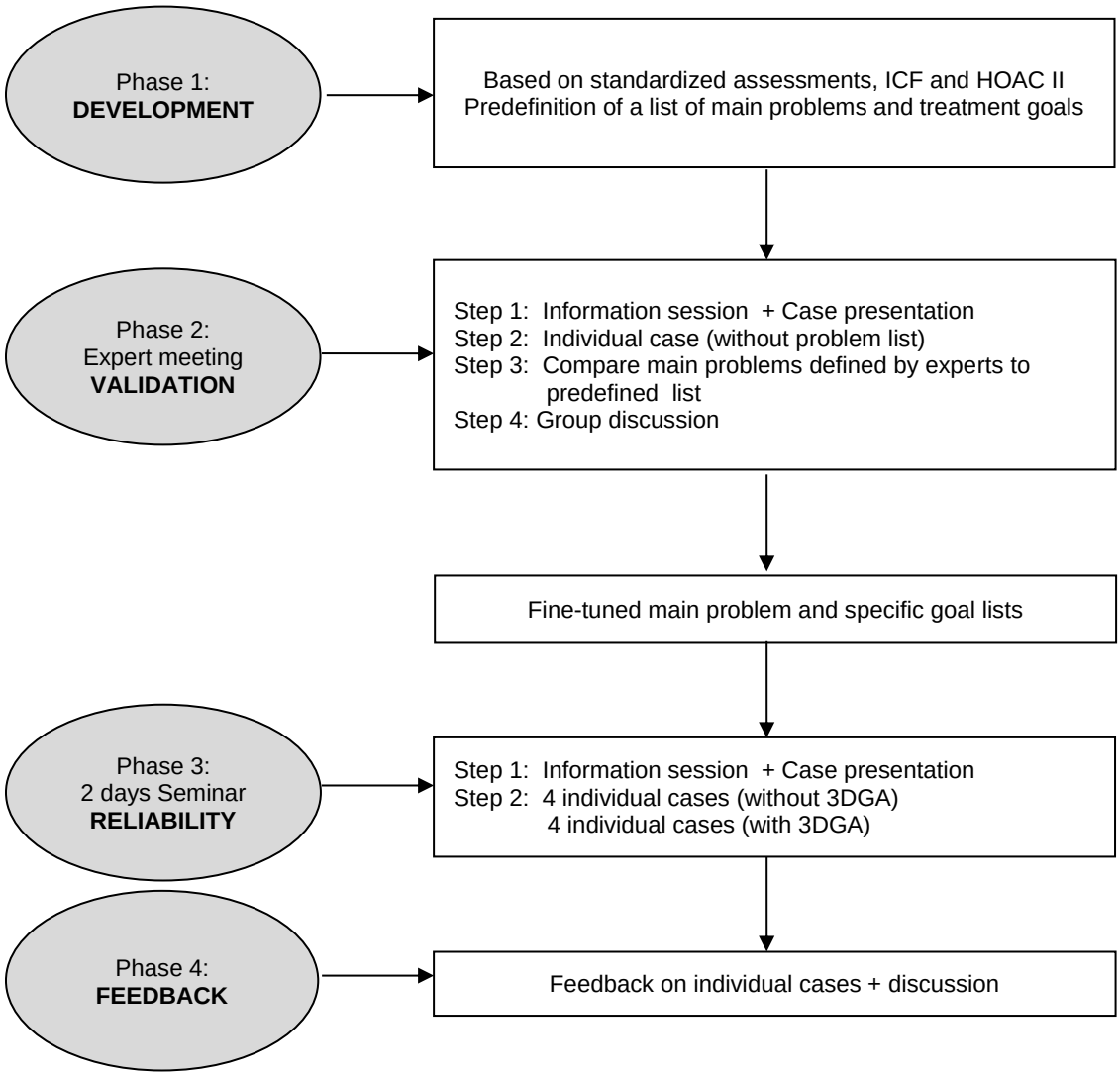

Fig 1. The different steps of validation and reliability testing

Abbreviations:

ICF: International Classification of Functioning, Disability and Health; HOAC-II: Hypothesis Oriented Algorithm for Clinicians II; 3DGA: Three Dimensional Gait Analysis

Phase 1 – Development of the clinical reasoning tool

The clinical reasoning tool was developed by the first authors of this manuscript (IF and JD) and was based both on clinical experience as on the existing validated structures of the ICF and the HOAC-II **(Fig. 2)**

As a first step, the results of the most routine practice assessments in ambulant children with CP were organized within the framework of the ICF.

At the level of body structure and function, clinical examination is commonly performed according to the standardized protocol used at the Laboratory of Clinical Motion Analysis and includes measurements of range of motion (goniometrical measurements), spasticity (Modified Ashworth Scale and Tardieu), selectivity (selective motor scale) and muscle strength (manual muscle testing). [19-21]

At activity level, the Gross Motor Function Measurement (GMFM-88) and three-dimensional gait analysis (3DGA) are used most frequently. The GMFM-88 is a standardized clinical instrument to evaluate gross motor function that fulfills the criteria of reliability and validity with respect to responsiveness to change.[22,23] It provides an overview of the gross motor capacities at different functional levels and organized within five dimensions. 3DGA is also performed according to the standardized protocol of the Laboratory of Clinical Motion Analysis of the University Hospital Pellenberg (Leuven). Results of 3DGA include measurement of joint movement during gait (kinematics), moment and power (kinetics) and muscle activity registered by EMG.

On participation level, structured interviews with the parents and children are often supplemented using the standardized questionnaire of the Children's Assessment of Participation and Enjoyment (CAPE).[24,25]

Organizing these results within the ICF provided a profound and complete overview of the child and resulted in an objective problem list of the child. In children with CP however, this problem list could be very extensive. Most children had problems at all levels of the ICF and as a therapist, one had to prioritize and select possible and realistic targets in treatment.

Therefore, as a next step, the HOAC-II was used to reduce this list to a selection of main problems: the key problems for the child. In this step, the therapist generated a hypothesis as to why a problem existed and the rationale behind this problem. The main problem was defined as the main underlying problem that can mostly explain the problems at all levels of the ICF. Consequently, when the main problems for the child were defined, the individual goals for the child could be deducted logically. Although not specified in this figure, during the treatment process, regular re-assessments and redefinitions are obviously necessary to test these hypotheses. In order to facilitate the next phase of the study, the authors prepared a list of main problems and a list of specific goals. The list of main problems was based on clinical experience and the most commonly reported main problems during the consultations at the CP reference centre of the U.Z. Pellenberg. The list of specific goals consisted of specific parameters from clinical examination, GMFM and 3DGA.

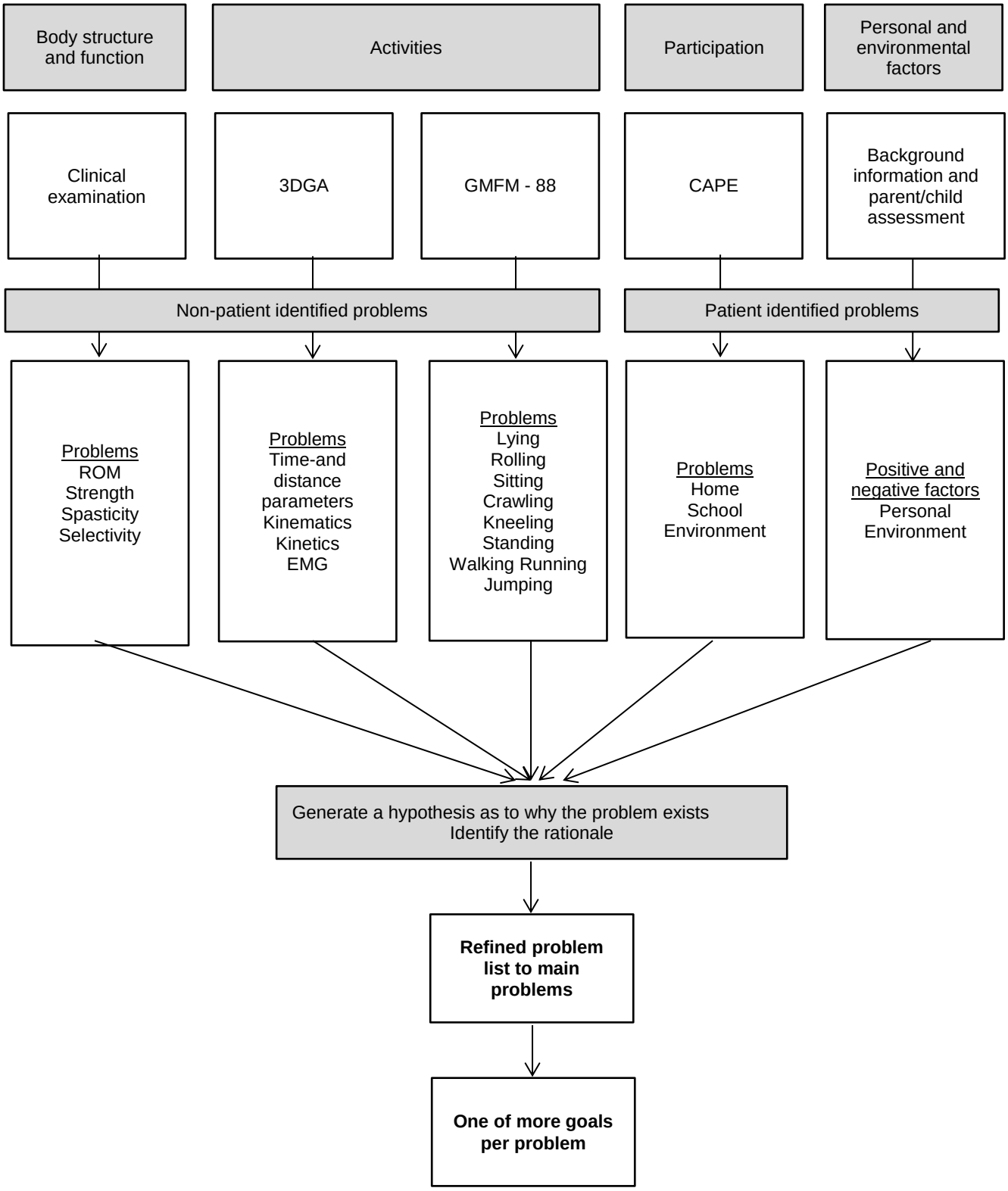

Fig 2. Structured approach of the ICF and the HOAC-II as a clinical reasoning tool in treatment of ambulant children with CP

Abbreviations:

GMFM-88: Gross Motor Function Measure; **3DGA**: Three-dimensional Gait Analysis; **CAPE**: Children's Assessment of Participation and Enjoyment, **ROM**: Range of Motion

Phase 2 – Validation of the clinical reasoning framework

For this part of the study, a small group of four expert paediatric physical therapists was recruited. The experts all had a minimum experience of 10 years in treatment of children with CP. A structured meeting was organized under the supervision and guidance of the first authors. During this structured meeting, the existing clinical reasoning process was presented and a case study was demonstrated and discussed as an example. Subsequently, the assessment results of another child were presented. The therapists were asked to individually use the presented approach to identify the main problems and goals for this child. When all four therapists had accomplished this task, the predefined list of main problems and the lists of treatment goals were introduced. The answers of the physical therapist were compared to the lists and it was checked if all answers were part of this predefined list. In case a main problem or a goal identified by one of the four therapists was not part this lists, this was discussed within the group and an agreement was searched whether or not this should be added to one of the lists. As a last part of the meeting, a group discussion was held to identify problems and goals that were not defined in these lists.

Phase 3 – Reliability testing by means of an agreement study

During the second phase of the study, 185 therapists were invited to a two-day seminar. All physical therapists had to be specialized in paediatric PT by means of an advanced paediatric neurology (masters) program or a Bobath certificate. A minimum of three years of experience in treatment of children with CP was required. Additionally, the therapists had to be familiar with 3DGA by following a specialized course in gait analysis. To insure an independent opinion, the selected therapists had no direct involvement with the University Hospital of Pellenberg. Twenty-five therapists agreed to participate.

Similar to the structured expert meeting, the seminar started with an information session on the clinical reasoning process. When all participants were familiar with the process, the assessment results of eight children were presented in the same structured way. All children presented during the study were children with bilateral spastic CP, aged between 4 and 9 years old. All children were ambulant and had a GMFCS level between I and III. Classification of the children's pathology according to the SCPE guidelines[1] as well as classification of the children's gait pattern according to Rodda (2004)[26] are listed in **Table 1**.

Background information regarding the child's environment and school was provided. A video selection of the GMFM and a short video of the child walking were showed and the results of the clinical examination were provided. In four of the eight cases, the results of the 3DGA were provided as supplementary information. The physical therapists worked individually. The cases were presented one by one. For every child, the therapists were asked to follow the structured approach. After the definition of a problem list, they were asked to select maximally three main problems from the list. Subsequently, they could select maximally eight goals for the different lists of treatment goals. In this way, three children were presented on the first day of the seminar and five children on the second day.

Table 1. Description of the children presented for the reliability study

	Age	GMFCS	Classification		Gait Pattern	3DGA provided
Child 1	4y3mo	I	Spastic	Bilateral	Asymmetric gait	no
Child 2	6y9mo	III	Spastic	Bilateral	Apparent equinus	yes
Child 3	4y11mo	II	Spastic	Bilateral	Asymmetric gait	no
Child 4	5y7mo	II	Spastic	Bilateral	True equinus	yes
Child 5	3y11mo	III	Spastic	Bilateral	Jump gait	yes
Child 6	4y5mo	II	Spastic	Bilateral	Jump gait	no
Child 7	6y1mo	III	Spastic	Bilateral	Asymmetric gait	yes
Child 8	6y6mo	I	Spastic	Bilateral	Jump gait	no

Abbreviations:

GMFCS: Gross Motor Function Classification System; **3DGA:** three-dimensional gait analysis; **Y:** years; **Mo:** months

The study was approved by the ethics committee of the University Hospital Leuven. The parents of the children agreed with the demonstration of their child's results and signed an informed consent accordingly.

Phase 4 – Feedback

Since no discussion amongst the therapists was allowed during the two-days seminar, a feedback-day was planned after completion of the seminar. During this day, the children were discussed one by one and feedback was provided on the results of the study. Results of these discussions are not part of this study.

Data analysis

For the main problems, initial data analysis consisted of a profound frequency analysis. Positive agreement was calculated as the percentage of therapists that selected the specific main problem, negative agreement was considered as the percentage of therapists that did not select this problem. Additionally, the frequencies of the three most frequently selected main problems were summed and then proportionally compared to the total number of possible selections (66). This percentage also provided a measure of agreement regarding the three most frequently selected main problems.

Another way to consider agreement was calculating the pair-wise agreement between the different therapists (231 combinations). An agreement score between 0 and 3 was noted when no corresponding goals were registered up to 3 when all main problems were selected accordingly. The frequencies of total agreement (score 3), 2 corresponding goals (score 2), 1 corresponding goals (score 1) and no agreement (score 0) were calculated per child. Subsequently, these frequencies were summed separately for the group of children with or without the results of the analysis.

Data analysis of the specific goals consisted mainly of an explorative frequency analysis. Goals were grouped into 14 categories. In case the therapist exceeded the number of eight goals, data was excluded.

To evaluate the additional value of 3DGA, these analyses were also performed separately for the with- and without gait analysis group.

To provide an impression of the clinical decision process of the therapists, the selection of main problems was compared to the selection of main goals. This linkage was further explored using hierarchical cluster analysis based on Ward's method using Euclidian distance as a dissimilarity index. This analysis was performed after rescaling the frequencies of the selected main problems and the specific goals: the frequency of the selected main problem or goal was divided by the total number of main problems or goals respectively for that child.

All statistics were performed in SPSS 21 IBM SPSS Statistics for Windows, Version 21.0. (Armonk, NY: IBM Corp).

Results

Phase 2: validation of the clinical reasoning framework

During the first session with the expert group, the proposed clinical reasoning process was introduced and feedback was asked. All four therapists agreed with the decision tree.

Regarding the list of main problems, two main problems were mentioned by the therapists but not separately in the list: muscle length and postural control. It was decided to add muscle length to the list and to use postural control as a subheading for static and dynamic alignment. The experts also proposed to keep the subdivisions in categories, to provide an easy structure. The list was finished with a total of 14 main problems. It was agreed that the number of main problems identified per child should have a maximum of three. The completed list is given in **Appendix T**.

The list of treatment goals was immediately accepted. This list consisted of 29 items from clinical examination, 43 goals derived from the 3DGA and 88 items from the GMFM-88. The complete list is provided in **Appendices V-W.** For the individual goals, it was agreed that a maximum number of eight goals could be selected.

For the reliability study, the experts proposed a more extensive explanation of the process and more time to introduce the goal of the study at the start of the two-days seminar. Additionally, the expert group proposed a more extensive introduction of the example case and a full-written copy of the example case.

Phase 3: reliability testing by means of an agreement study

Participants

Twenty-five therapists agreed to participate to the two-days seminar. However, at the first day, two therapists cancelled because of personal reasons. One therapist could not come back on the second day of the study and results of this therapist from the first day were excluded.

The results of 22 therapists were included. The average experience of the therapists was 16.1 years (range between 3 and 40 years). Seventeen therapists were certified Bobath therapists. Thirteen therapists had a physiotherapy degree at university level, comparable or equal to a Master's degree. Nine therapists had a professional physiotherapy degree, comparable or equal to a professional Bachelor's degree. Most therapists were full-time employed in paediatric rehabilitation (average percentage 94%) in a paediatric rehabilitation centre, special-needs school or private sector.

Main problems

The frequencies of the main problems identified by the therapists are provided in **Table 2**. For the total group, muscle strength (98 times or 18.56%), hypertonia (100 times or 18.94%) and static alignment (73 times or 13.83%) were most frequently identified as one of the main problems. Thirteen main problems (2.5% of all main problems) were registered that were not part of the list and nine times (1.7%) no problem was selected.

Table 2. Frequencies of the selected main problems

	Total		Total (with GA)		Total (without 3DGA)	
	n	%	n	%	n	%
Hypertonia	100	18,94	67	25,4	33	12,50
Hypotonia	20	3,79	6	2,27	14	5,30
Fluctuating muscle tone	1	0,19	1	0,38	0	0,00
Static alignment	73	13,83	31	11,7	42	15,91
Dynamic alignment	50	9,47	25	9,47	25	9,47
Lack of cocontration	34	6,44	18	6,82	16	6,06
Laxity	4	0,76	2	0,76	2	0,76
Extension pattern	7	1,33	3	1,14	4	1,52
Flexion pattern	23	4,36	17	6,44	6	2,27
ATNR	0	0,00	0	0	0	0,00
Other patroon	3	0,57	3	1,14	0	0,00
Muscle strength	98	18,56	47	17,8	51	19,32
Muscle length	52	9,85	25	9,47	27	10,23
Bony deformities	41	7,77	10	3,79	31	11,74
Others	13	2,46	7	2,65	6	2,27
No problem selected	9	1,70	2	0,76	7	2,65
	528	**100**	**264**	**100**	**264**	**100**

Abbreviations:

Total (without): total frequencies of child 1,3,6 and 8, (were the gait analysis information was not provided)

Total (with): total frequencies child 2,4,5 and 7 (were the results of the gait analysis was provided)

N: number

%: percentage of the frequency of the selected main problem to the total number of selected problems for the total group, the total group (without) and the total group with gait analysis information

ATNR: Aysmmetric Tonic Neck Reflex

Table 3 provides the frequencies of the selected main problems per child, together with the positive and the negative agreements. Child 2 and child 5 were children with the highest positive agreement values, with 95.9 and 90.5% respectively for the selection of hypertonia. Asymmetric Tonic Neck Reflex (ATNR) was never selected as a main problem.

Table 3. Positive and negative agreement values of the main problems per child

	Child 1			Child 2			Child 3			Child 4			Child 5			Child 6			Child 7			Child 8		
	N	**pos agr**	**neg agr**	**n**	**pos agr**	**neg agr**	**n**	**pos agr**	**neg agr**	**n**	**pos agr**	**neg agr**	**n**	**pos agr**	**neg agr**	**n**	**pos agr**	**neg agr**	**n**	**pos agr**	**neg agr**	**n**	**pos agr**	**neg agr**
Hypertonia	2	9,1	90,9	21	95,5	4,5	9	40,9	59,1	13	59,1	40,9	20	90,9	9,1	8	36,4	63,6	13	59,1	40,9	14	63,6	36,4
Hypotonia	2	9,1	90,9	2	9,1	90,9	5	22,7	77,3	3	13,6	86,4	0	0	100	5	22,7	77,3	1	4,5	95,5	2	9,1	90,9
Fluctuating muscle tone	0	0,0	100	0	0,0	100	0	0,0	100	1	4,5	95,5	0	0	100	0	0	100	0	0	100	0	0	100
Static alignment	10	45,5	54,5	5	22,7	77,3	15	68,2	31,8	7	31,8	68,2	5	22,7	77,3	12	54,5	45,5	14	63,6	36,4	5	22,7	77,3
Dynamic alignment	2	9,1	90,9	0	0,0	100,0	5	22,7	77,3	12	54,5	45,5	8	36,4	63,6	7	31,8	68,2	5	22,7	77,3	11	50,0	50,0
Lack of cocontraction	4	18,2	81,8	4	18,2	81,8	1	4,5	95,5	7	31,8	68,2	5	22,7	77,3	6	27,3	72,7	2	9,1	90,9	5	22,7	77,3
Laxity	0	0,0	100	0	0,0	100,0	0	0,0	100	2	9,1	90,9	0	0	100	2	9,1	90,9	0	0	100	0	0	100
Extension pattern	3	13,6	86,4	1	4,5	95,5	1	4,5	95,5	0	0	100	1	4,5	95,5	0	0	100	1	4,5	95,5	0	0	100
Flexion pattern	3	13,6	86,4	4	18,2	81,8	3	13,6	86,4	0	0	100	8	36,4	63,6	0	0	100	5	22,7	77,3	0	0	100
ATNR	0	0	100	0	0,0	100	0	0	100	0	0	100	0	0	100	0	0	100	0	0	100	0	0	100
Other pattern	0	0	100	0	0,0	100	0	0	100	0	0	100	0	0	100	0	0	100	3	13,6	86,4	0	0	100
Muscle strength	16	72,7	27,3	15	68,2	31,8	9	40,9	59,1	12	54,5	45,5	7	31,8	68,2	12	54,5	45,5	13	59,1	40,9	14	63,6	36,4
Muscle length	9	40,9	59,1	8	36,4	63,6	8	36,4	63,6	4	18,2	81,8	9	40,9	59,1	3	13,6	86,4	4	18,2	81,8	7	31,8	68,2
Bony deformities	11	50,0	50,0	3	13,6	86,4	5	22,7	77,3	2	9,1	90,9	2	9,1	90,9	9	40,9	59,1	3	13,6	86,4	6	27,3	72,7
Others	3	13,6	86,4	2	9,1	90,9	0	0	100	3	13,6	86,4	1	4,5	95,5	1	4,5	95,5	1	4,5	95,5	2	9,1	90,9
No problem selected	1	4,5	95,5	1	4,5	95,5	5	22,7	77,3	0	0	100	0	0	100	1	4,5	95,5	1	4,5	95,5	0	0	100
Sum 3 most frequently selected	37			44			33			37			37			33			40			39		
(sum/66)*100	56,1			66,7			50,0			56,1			56,1			50,0			60,6			59,1		

Abbreviations:
Pos agr: Positive Agreement or the percentage of therapists that selected the specific problem; **Neg agr:** Negative Agreement or the percentage of therapists that did not select the specific problem
N: number; **ATNR:** Asymmetric Tonic Neck Reflex

In this sample, the median percentage of the three most frequently selected main problems was higher when 3DGA was provided: 58.3 in comparison to 53.03% (**Fig. 3**).

As a next step in the agreement analysis, the pair-wise agreement between the different therapists was checked (231 combinations of therapists). The frequencies of the agreement scores were calculated per child (**Fig. 4**). Also here, a higher agreement was found for child 2 and 5.

The frequencies of the pair-wise agreements were summed. This analysis revealed that in 47% of the cases, therapists agreed on one of the main problems and in 32% of the cases, two main problems were selected similarly. Total agreement was only found in 3% of the cases and no agreement was found in 18% of the cases. In our sample, the proportion of no agreement (score 0) was higher when 3DGA was not provided (22% versus 14%). The proportion of moderate agreement (score 2) was higher when the results of the 3DGA were provided (37% versus 26%). The proportion of total agreement was equal between both groups.

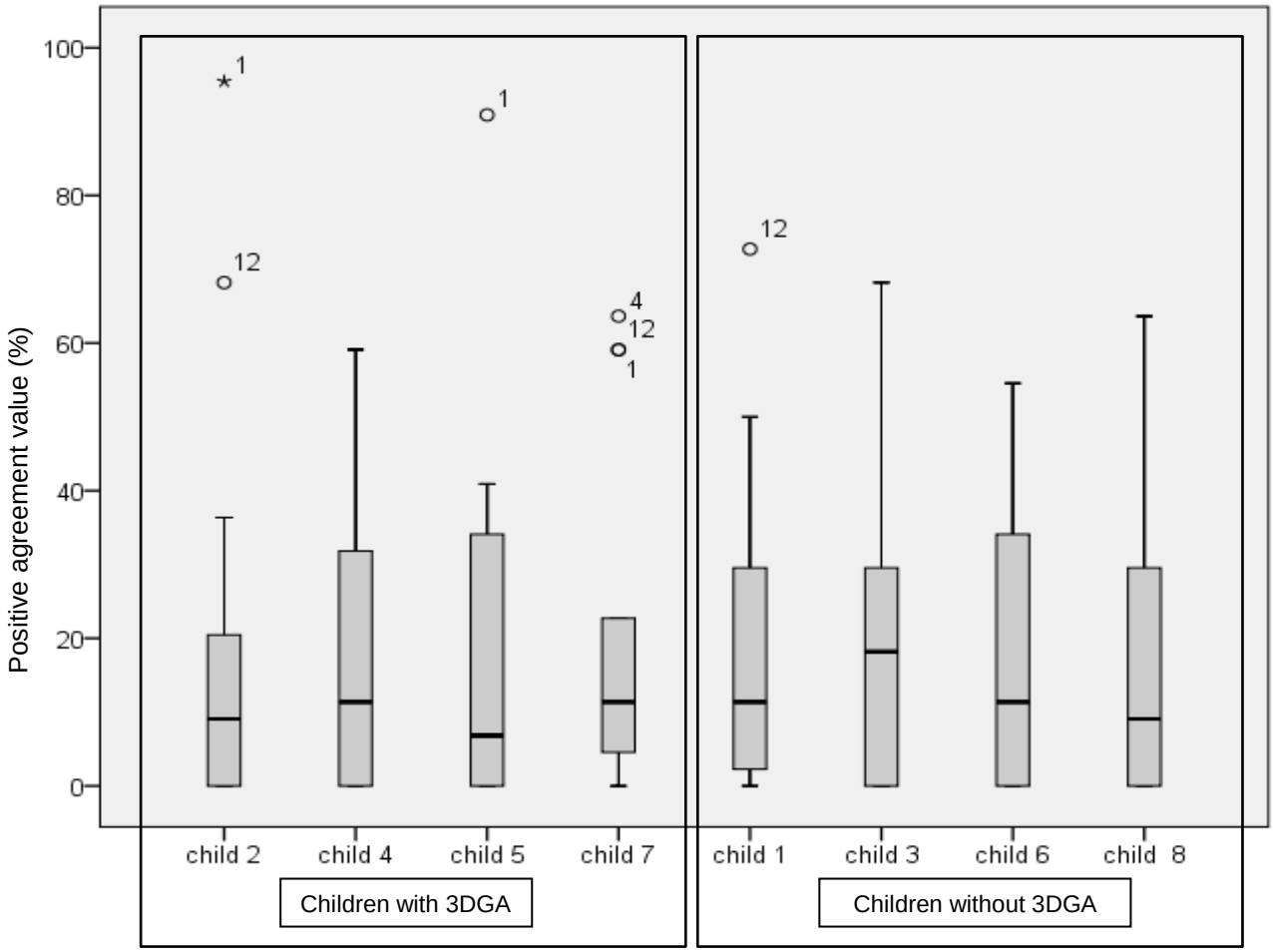

Fig 3. Distribution of the positive agreement values for the different main problems per child

Abbreviations:
° and * indicate the outliers with the corresponding number of main problem: 1 (hypertonia), 4 (static alignment) and 12 (strength)
3DGA: Three Dimensional Gait Analysis; **Positive agreement:** the percentage of therapists that selected the specific main problem

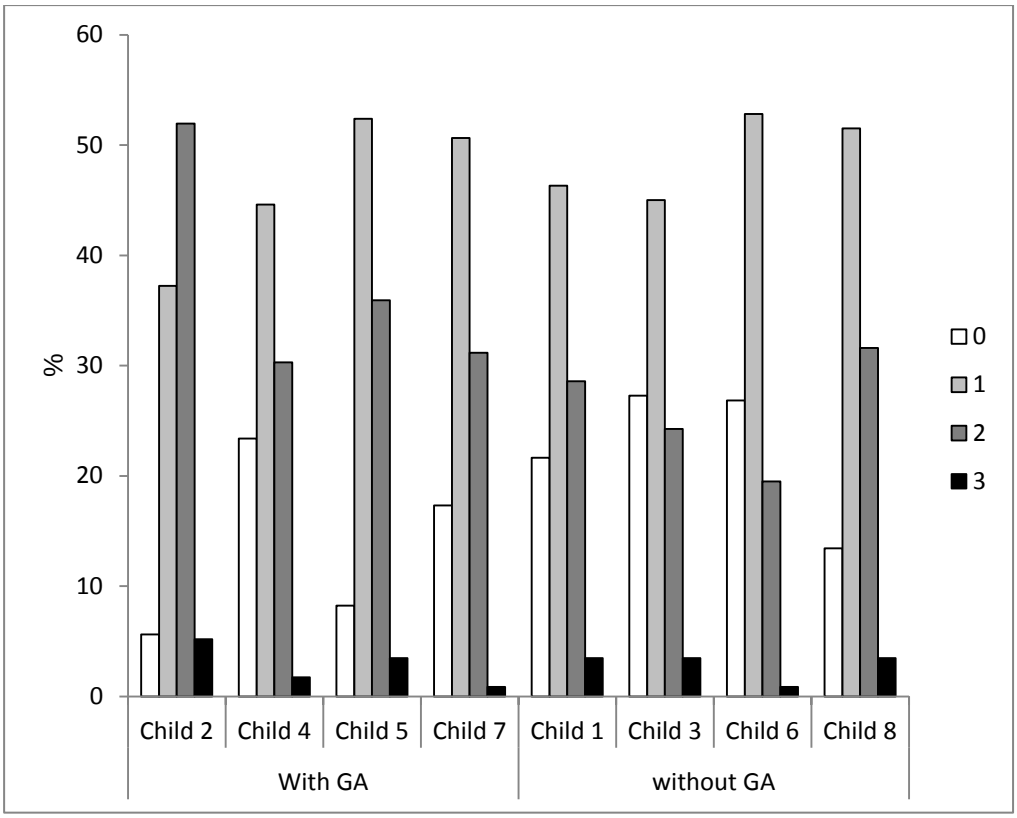

Fig 4. Proportional distribution of the number of corresponding main problems for the different children

0 no corresponding main problems
1 1 corresponding main problems
2 2 corresponding main problems
3 3 corresponding main problems
with GA children from whom the results of the three-dimensional gait analysis were provided
without GA children from whom the results of the three-dimensional gait analysis were not provided

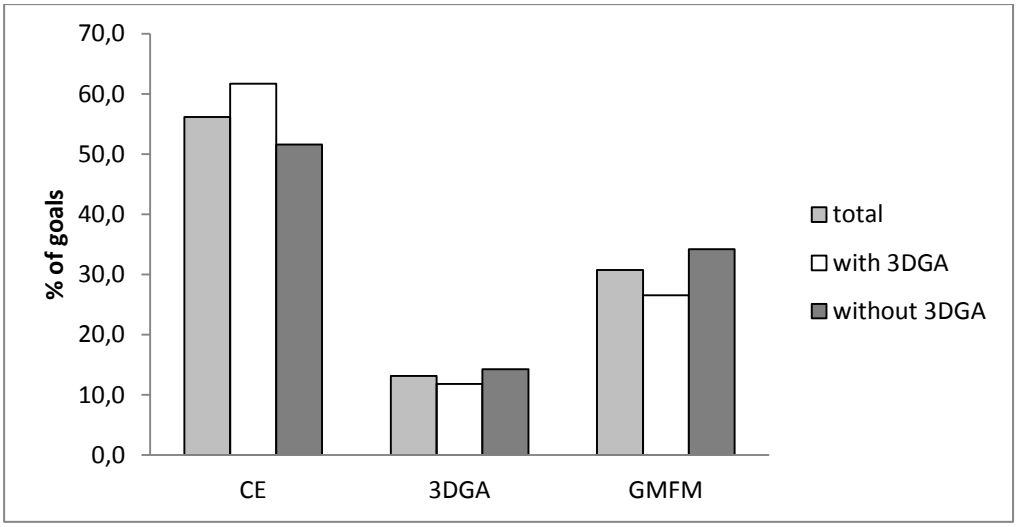

Fig 5. Proportion of selected goals based on clinical examination (CE), gait analysis (3DGA) or gross motor function (GMFM) for the total group (total), the group of children where 3DGA information was provided (with 3DGA) and not provided (without 3DGA)

Specific goals

A total number of 991 goals were selected. Since the total possible selection of goals was 1408 (22 therapists, eight children and eight goals), this means that 417 times or in 29.3% of the cases, the therapists could not limit the selection to eight goals.

Of all these goals, 556 (56.2%) were based on problems identified from clinical examination and targeted impairment level. 130 (13.1%) and 304 (30.7%) of the goals targeted gait and gross motor function parameters respectively **(Fig. 5**). When evaluating per child, this trend was similar. In all children, most of the goals were based on the results of the clinical evaluation.

Table 4 provides a more detailed overview of the selected goals. This demonstrates that most of the goals were targeting strength (34%), followed by range of motion (15.2%) and GMFM D (11.8%). For all children, different strength parameters were most frequently selected as treatment goals.

When the additional information of gait analysis was provided, the proportion of gait parameters selected as a specific goal was lower: 11.8 versus 14.2% (**Fig. 5**).

Overall clinical reasoning

To explore the clinical reasoning process of the therapists, the three most frequently selected main problems were compared to the three most frequently selected group of goals (**Table 5**). Logically, the selected goals were directly derived from the definition of main problems. In all children, a direct relation could be noticed between the main problems and the most frequently selected goals. Only for femoral anteversion and hypertonia, no specific goal could be derived. In all children except child 1, hypertonia was selected as one of the main problems. However, only in child 2, specific goals were based on spasticity parameters. In child 5, strength was not identified as a main problem. However, the most frequently selected goals for this child were based on strength parameters.

Fig. 6 demonstrates the proportion of goals from clinical examination and gait analysis in the different regions. For the goals based on clinical examination, in all children, the hip was obviously the most selected region. The comparison of the goals based on 3DGA with the goals based on clinical examination demonstrated a shift to goals based on ankle parameters.

Fig. 7 shows the dendrogram based on the rescaled frequencies of the selected main problems and goals. Three main clusters can be deducted. A first cluster combines the main problems and specific goals that were the least frequently selected: at the top of the dendrogram, from the specific goal bony deformities to the main problem of hypotonia. A second cluster can be divided into two subgroups. A first subgroup shows that muscle length and bony deformities as main problems are very often selected in combination with GMFM-D and E and dynamic alignment. The second subgroup shows association between flexion pattern, spasticity and GMFM-C. In the last cluster, strength as a specific goal, associates with the main problems of hypertonia, static alignment and strength. ROM and static alignment can be found together in a subgroup, also confirming obvious clinical reasoning.

Table 4. Frequencies of selected specific goals

	Child 1	Child 2	Child 3	Child 4	Child 5	Child 6	Child 7	Child 8	**Total**	Total (%)	Total (With)	%	Total (without)	%
ROM	14	12	27	16	26	8	26	22	**151**	15,21	80	17,7	71	13,1
Spasticity	0	10	5	3	11	1	10	10	**50**	5,04	34	7,5	16	3,0
Strength	52	27	37	38	41	58	46	45	**344**	34,64	152	33,6	192	35,5
Bony deformities	1	0	1	0	1	1	1	0	**5**	0,50	2	0,4	3	0,6
Pelvis	7	2	5	5	4	6	3	2	**34**	3,42	14	3,1	20	3,7
Hip	8	2	7	2	9	2	4	0	**34**	3,42	17	3,8	17	3,1
Knee	7	1	5	4	2	3	4	5	**31**	3,12	11	2,4	20	3,7
Ankle	4	2	1	4	1	1	3	5	**21**	2,11	10	2,2	11	2,0
TD parameters	1	0	1	1	0	1	2	1	**7**	0,70	3	0,7	4	0,7
GMFM-A	1	2	0	2	5	0	0	0	**10**	1,01	9	2,0	1	0,2
GMFM-B	0	4	2	0	4	2	0	0	**12**	1,21	8	1,8	4	0,7
GMFM-C	1	5	18	11	13	7	1	6	**62**	6,24	30	6,6	32	5,9
GMFM-D	20	7	15	5	13	17	20	20	**117**	11,78	45	10,0	72	13,3
GMFM-E	21	4	12	24	3	19	6	26	**115**	11,58	37	8,2	78	14,4

Abbreviations:
Total (with): total frequencies of child 1,3,6 and 8, (were the gait analysis information was provided); **%:** proportion of the frequency of the goal to the total number of goals in the with or without group; **Total (without):** total frequencies child 2,4,5 and 7 (were the results of the gait analysis was not provided); **ROM:** Range of Motion;
TD parameters: time and distance parameters derived from the 3DGA; **GMFM-A:** Gross Motor Function Measure dimension A; **GMFM-B:** Gross Motor Function Measure dimension B; **GMFM-C:** Gross Motor Function Measure dimension C; **GMFM-D:** Gross Motor Function Measure dimension D; **GMFM-E:** Gross Motor Function Measure dimension E

Table 5. Comparison of the most frequently selected main problems to the most frequently selected specific goals per child. In child 2,4,5 and 7 the results of the 3DGA were provided, in child 1,3,6 and 8 this information was not provided.

	Main problems	Selected goals
	strength	strength
Child 1	femoral anteversion	GMFM E
	static alignment	ROM
	hypertonia	strength
Child 2	strength	ROM
	muscle length	spasticity
	static alignment	strength
Child 3	strength	ROM
	hypertonia	GMFM C
	hypertonia	strength
Child 4	strength	GMFM E
	dynamic alignment	ROM
	hypertonia	strength
Child 5	muscle length	ROM
	dynamic alignment	GMFM C /D
	strength	strength
Child 6	static alignment	GMFM E
	hypertonia	GMFM D
	static alignment	strength
Child 7	strength	ROM
	hypertonia	GMFM D
	strength	strength
Child 8	hypertonia	GMFM E
	dynamic alignment	ROM

Abbreviations:
ROM: Range of Motion; **GMFM-C**: Gross Motor Function Measure – dimension C; **GMFM-D:** Gross Motor Function Measure – dimension D; **GMFM-E:** Gross Motor Function Measure Dimension E

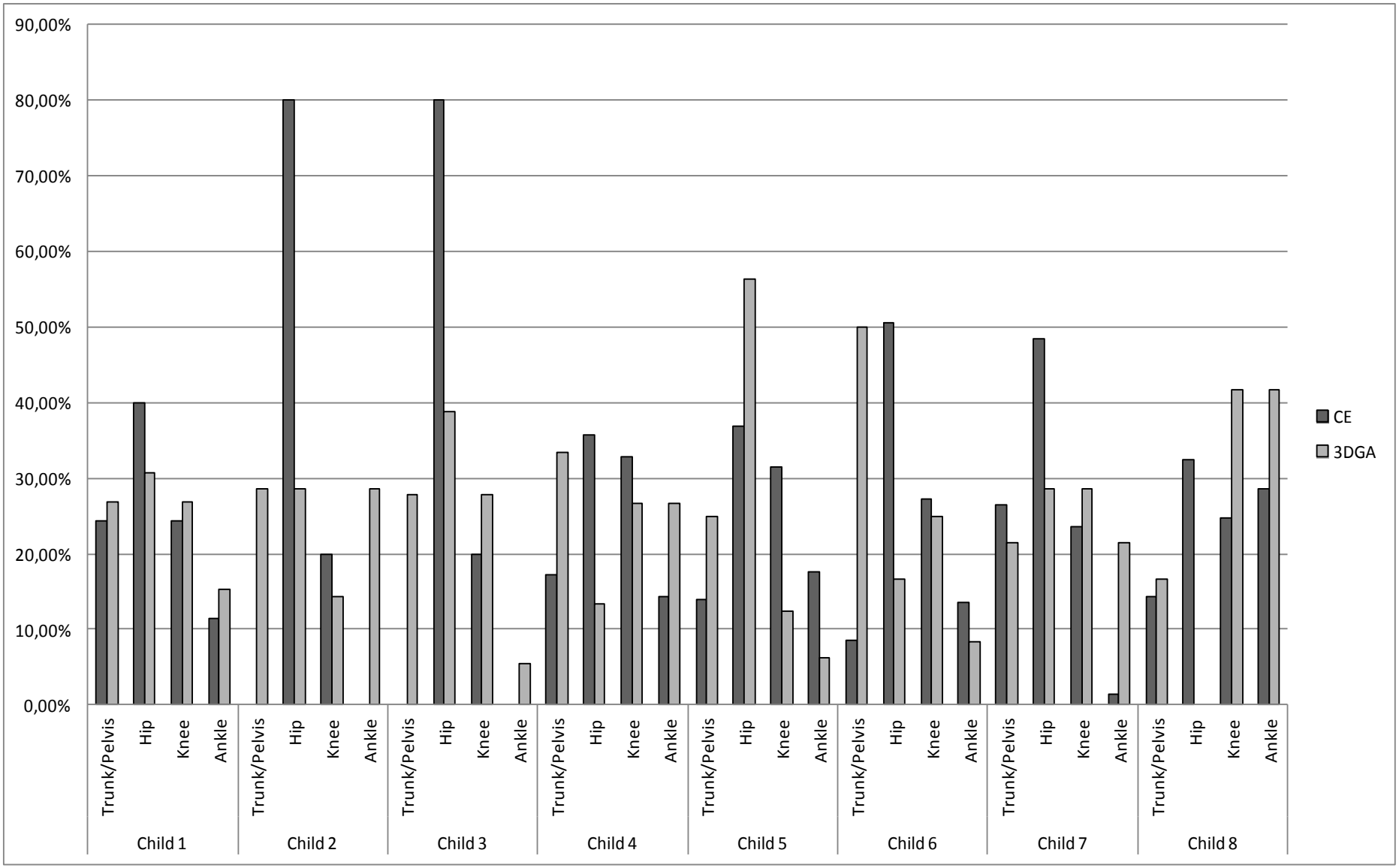

Fig 6. Proportion of the goals based on clinical examination (CE) and three-dimensional gait analysis (3DGA) per target region and per child

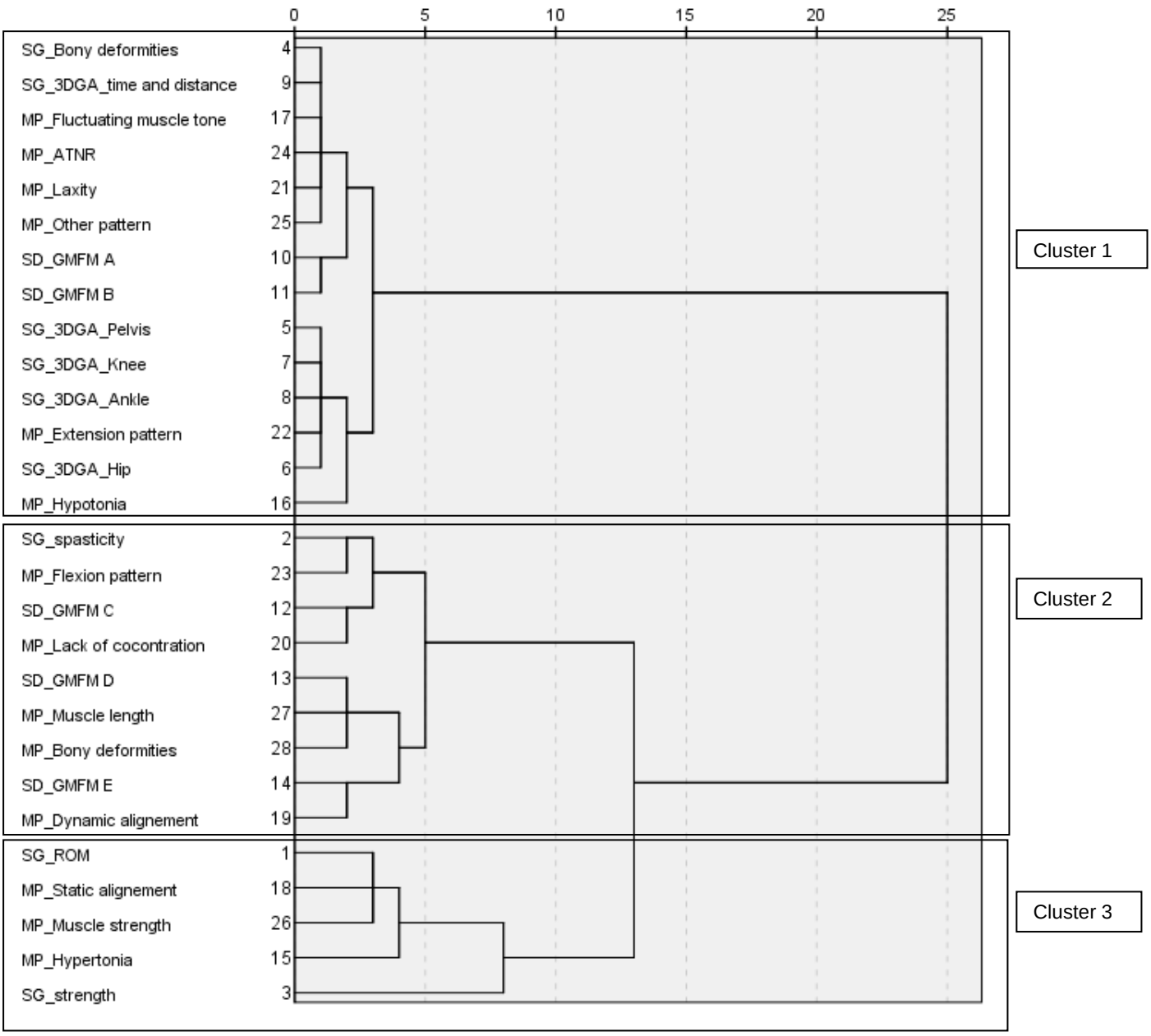

Fig 7. Dendrogram using Ward Linkage evaluating the Euclidian distance between the 14 main problems (MP) and the 16 categories of specific goals (SG)

Abbreviations
MP: Main Problem
SG: Specific goal
3DGA: three-dimensional gait analysis

Discussion

The primary aim of this study was to demonstrate how a clinical decision process based on the HOAC-II and the ICF can contribute to a reliable identification of main problems and specific goals for ambulant children with CP.

As a first step in this process, validity of the clinical decision framework was evaluated. All experts agreed on the contents of the predefined lists and the structure of the clinical reasoning process. In addition, the results of the reliability study demonstrated that only 2.5% of the main problems identified by the therapists were not part of the list, which confirms good validity. However, one had to mention that the list of main problems was limitative and the authors decided not to include 'other problems'. The therapists were specifically asked to select items from the predefined list. Only in case they identified a main problem that was not part of the list, they were asked to note this down separately. The main problem that was not part of the list but was mentioned five times was endurance.

Considering the second step of the study, reliability was tested by means of the different agreement measures. The pair- wise agreement scores provided the main measure of agreement. This analysis revealed a low to moderate agreement, with 3% of agreement upon all three main problems. Still, in 32% of the pairs, an agreement of two out of three main problems was registered. A very logic association was demonstrated between the different main problems and specific goals. The therapists seemed to use the clinical reasoning in a way that the specific goals logically followed out of the selection of main problems.

However, for the specific goals, one has to mention that many therapists were not able to limit the number of goals to eight. Even though the therapists were instructed to select only eight goals, many therapists still reported more than the proposed number. In this case, the authors decided not to include the specific answer. This obviously influenced the results of our study.Nonetheless, the results of the study highlight that, even though the therapists were able to use the clinical reasoning structure in an adequate way, setting priorities was still a clear difficulty.

A possible explanation for the moderate agreement could be that the therapists were not sufficiently trained in using the structured clinical reasoning tool and the stepwise approach was fairly new. This can be confirmed by the feedback at the end of the study. All except one therapist reported that he or she had learned throughout participation of the study. The feedback from the therapists was very positive and the structure was appreciated as clear and user-friendly. Still, more time was required to complete the first case-study in comparison to the last one. Most likely, the therapists were much more familiar to the predefined lists by the end of the study. Nevertheless, a learning effect was not immediately visible throughout our results and the agreement was not higher in child 8 compared to child 1. More training time for the therapists might however, be advisable for future studies.

Another possible explanation could be the classification and pathology of the children. Although the selection of children was limited to ambulant children with bilateral spastic CP, all children had different functional abilities. The GMFCS levels of the children might have influenced the results of the

study. However, also this effect was not clearly visible throughout our results. Nevertheless, it is animportant future direction to test the agreement analysis on children with unilateral CP and/or fluctuating muscle tone.

Several studies have demonstrated significant differences in clinical reasoning between expert and novice physical therapists. [27,28] This aspect was not specifically reported in this study. However, it was explored in a frequency analysis. We could not find obvious differences between the expert and novice physical therapists. It is possible that younger physical therapists are more familiar with 3DGA, as in the last 10 years, gait analysis is part of the master's program in paediatric rehabilitation. Older therapists had to take additional courses in gait analysis, which might have been less extensive. Previous studies demonstrated that more experienced physical therapists rely more on observations and interactions with patients rather than medical records.[28] Additionally, due to our inclusion criteria, the youngest participating therapists in our study already had three years of experience, so no real non-experienced or novice therapists participated in our study.

The secondary aim of study was to demonstrate how the additional information of 3DGA can influence the reliability of this information. Different pair-wise agreement scores were found in both groups, in the advantage of the group where additional information of 3DGA was provided. This higher agreement might indicate a more profound insight into the child's main problems. However, the additional 3DGA information was only provided in four children. Since both groups consisted of different children, it was difficult to compare the choices of main problems and goals. The order in which the children were presented was decided randomly. There were a higher number of children with GMFCS level III in the group of children from whom the results of the 3DGA was provided. Hence, the results of this analysis should be interpreted carefully and more research is necessary to support or refute this statement.

Conclusion

This study confirms the proposed framework as a valid structure that supports logic clinical reasoning. The results show a moderate agreement for the selection of main problems but results highlight the difficulty of priority setting in the selection of specific goals. Further research is necessary to support the therapist in setting priorities and to investigate the additional value of 3DGA in this process.

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Part 3
Assessment

Chapter 4

A study of whether video scoring is a reliable alternative for blinded scoring of the Gross Motor Function Measure–88?

Under revision for Clinical Rehabilitation

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Abstract

Objective

To investigate the agreement between live- and video- scores of the Gross Motor Function Measure–88.

Design

Reliability study

Subjects

Forty children with bilateral spastic cerebral palsy

Interventions

Fifty evaluations were administered according to the test guidelines. The evaluation moments were videotaped. After a minimum interval of one month, the same assessor rated these video-recordings. Additionally, two physical therapy students scored the recordings twice, using a minimal interval of one month.

Main measures

Agreement between live- and video-scores as well as inter- and intra-rater agreement of the video-scores were assessed using intra-class correlation coefficients (ICC), standard error of measurements (SEM) and smallest detectable changes (SDC). Weighted kappa coefficients were used for individual item-analysis.

Results

Comparing the live- and video-scores from the same assessor, showed good to very good agreement for the total score (ICC 0,973; SEM 2,28; SDC 6,32), dimension B (ICC 0,938), D (ICC 0,965) and E (ICC 0.992) but lower for A (ICC 0,720) and C (ICC 0,667). Agreement for the total score was higher for live-versus-video compared to the inter-rater agreement by video (ICC 0,949; SEM 3,15; SDC 8,73) but lower than the intra-rater agreement by video (ICC 0,989; SEM 1,42; SDC 3,96).

Conclusion

Scoring the Gross Motor Function Measure-88 using video-recordings can be done reliably. Nevertheless, agreement between live- and video- scores is lower in comparison to the intra-rater reliability using video recordings only. In the future, the appropriate SEM and SDC values should be used to interpret the results of clinical trials.

Introduction

The Gross Motor Function Measure is a criterion-referenced and observational measure to evaluate gross motor function, validated for children with Cerebral Palsy, Down syndrome and traumatic brain injury.[1-3] The original Gross Motor Function Measure-88 (GMFM-88) consists of 88 items, divided into five dimensions.[4] Although, the Gross Motor Function Measure- 66 (GMFM-66) represents the more recent, shorter and more reliable version, it still has several disadvantages. It may be less descriptive for children functioning at lower ability levels and specific software is required to calculate the child's total score.[5] Therefore, the Gross Motor Function Measure-88 is still very commonly used.
A well-known challenge in pediatric research is the requirement of blinded assessment. Blinded evaluation has the possibility to reduce tester bias and thereby, to increase study quality. The difficulty in gross motor function evaluation however, is the need for a child to feel comfortable. Especially in young children, an assessor unknown to the child might make the child insecure, frightened and thereby, less compliant. This might reduce performance. Video-recordings can therefore be a useful tool to allow a qualitative and optimal testing situation. It does not only allow blinded evaluation by means of video scoring, but might also increase reliability since it allows the person scoring the test to pause scoring or to review items in case of doubt.
Several researchers have used video-recordings and have investigated inter-rater reliability of the Gross Motor Function Measure-88 by means of video-scoring.[1,6-8] However, to our knowledge, none of these studies evaluated agreement between the live- and video-scores. Nevertheless, scoring video-recordings as such might cause limitations. This is especially true for the Gross Motor Function Measure-88, as the test is known to be very vulnerable for missing items.[1, 4] It is important to verify whether scoring video-recordings increases the chance for missing items and whether this creates a systematic error.

The aim of the study was to examine the agreement in test results when comparing the scores of the Gross Motor Function Measure-88 live and by video. Therefore, the agreement between the live- and video-scores was compared to the inter-and intra-rater agreement of the raters.

Methodology

Children were recruited at the Cerebral Palsy Reference Centre of the University Hospital Pellenberg. Data from the participants of an ongoing intervention study was used. This study included ambulant children with bilateral spastic cerebral palsy, aged between four and nine years and gross motor function classification system level I to III. Children were excluded when they showed severe associated problems that could influence test results (blindness, deafness, severe cognitive limitations). All caretakers of the children signed an informed consent form.

All children were tested by the first author of the study, a pediatric physical therapist, acquainted with the test items and manual. The English version of the test manual was used and all evaluations were administered accordingly. The test was administered in the pediatric physical therapy room of the child's usual therapist so that the room was comfortable and familiar. During the tests, children were barefoot. No assistive devices were used. Each session took 40–60 minutes. The assessor tested all 88 items for each assessment. All assessments were videotaped for subsequent extra scoring. To register the child's performance without distraction, the camera was well positioned on a tripod and no additional person was used for filming. Video-recordings were not edited.
After a minimal interval of one month, the same physical therapist scored the video-recordings of the test. Additionally, the recordings were copied and distributed to two physical therapy students (MSc), specializing in pediatric physical therapy. Both students scored the video-recordings of the tests twice, again with a minimal interval of one month between both scoring moments. The students were also familiar to the test and the guidance manual and were trained by scoring five video recordings with the possibility to ask questions. It was allowed to pause the video-recordings and to review items as often as necessary.
According to the test guidelines, all items were tested and scored on an ordinal scale of zero (does not initiate) to three (completed item). Subsequently, the total dimension scores were calculated and expressed as percentage scores. Total scores were calculated as the average of all dimensions.

As a first step in the data-analysis, the agreement in test results between the live- and video- scores of the first evaluator was evaluated.
Secondly, intra-rater agreement of the video-scores was calculated using the first and second scores of both independent raters. Inter-rater agreement of the video-scores was calculated between the second scores of all three raters.
Association between the total and dimension percentage scores was evaluated using intra-class-correlation coefficients (ICC 3,1) for absolute agreement with 95% confidence intervals.[9] Although the ICC can be defined as a measure of association, it assesses the consistency or reproducibility of quantitative measurements made by different observers measuring the same quantity. Therefore, the ICC will further be described as an agreement measure.

To investigate systematic differences between the measurements and to identify possible outliers, Bland-Altman plots were used. The limits of agreement with 95% confidence interval (1,96 SD) were reported.[10]

In addition, the Standard Error of Measurement (SEM) and the smallest detectable change (SDC) were calculated based on the variance components.[11] The SEM provides an absolute index of reliability. It indicates the variability of the scores around the subject's true score and thereby provides a value for the measurement error in the same unit as the measurement itself. The SDC represents the amount by which a patient's score needs to change to ensure that the change is greater than the measurement error.

Agreement between the live-and video ratings was also assessed at item level. As items scores use an ordinal scale, Weighted Kappa coefficients (ƙ) are applied. However, when there is a limited range of scores, Weighted Kappa coefficients cannot be calculated. Therefore, the proportion of positive agreement was used as an additional measure for item-analysis.[12] Proportions of positive agreement are calculated as the proportion of scores with positive agreement divided by the total number of scores.

Finally, the number of items that could not be scored based on the video-recordings because of practical reasons (e.g. when the child was out of the video-range) was registered.

Statistical analysis was performed using the IBM SPSS 22 Statistics for Windows version 22.0 (Armonk, NY: IBM Corp). Weighted-kappa coefficients were obtained using MedCalc for Windows, version 12.5 (MedCalc Software, Ostend, Belgium).

Results

Forty children were included in the study. Mean (SD) age of the children at the time of testing was 5,8 (1,3) years. All children were diagnosed with bilateral spastic cerebral palsy. Twelve children were classified at gross motor function classification system level I, 18 at level II and 10 at level III. For ten children, two different measurements were used. When different measurements of the same child were used, a minimal interval of 10 weeks was applied. This resulted in a total number of 50 test results. Before the study, none of the children had previously been tested with the gross motor function measure-88.

Table 1 provides an overview of the results of the agreement measures between the live- and video-scores of the first evaluator. The results showed good to very good agreement for the total score (ICC 0,973), dimension B (ICC 0,938), D (ICC 0,965) and E (ICC 0,992) and good agreement for dimension A (ICC 0,720) and C (ICC 0,667).
The SEM for the total score when comparing the live and video-scores was 2,28% with an SDC of 6,32%. The limits of agreement at 95% CI as demonstrated in the Bland-Altman-plot (**Fig 1 and table 1**) for the total scores range from -5,28 to 7%. The mean value of the difference was 0,81%.
Weighted Kappa coefficients (**see appendix**) revealed a moderate to high agreement for all except four items: item 32 (attaining 4 point over left side) (ƙ=-0.101, positive agreement 0,96), item 38 (prone, forward creeping) (ƙ=-0.101, positive agreement 0,96), item 40 (4 point, attains sitting arms free) (ƙ=-0.204, positive agreement 0,92) and item 45 (crawling reciprocally (ƙ=0.310, positive agreement 0,96).

Table 2 provides the results of the intra-rater reliability for scores of video-recordings by the two independent evaluators. The ICC could be interpreted as very good for both the total score (ICC 0,989) and all dimension scores (ICC ranges from 0,894-0,993). The SEM for the total score was only 1,43% with an SDC of 3,96%. Overall, both independent assessors reported an average of 4,14 items per registration that could not be scored based on the video-recordings.

Table 3 represents the inter-rater agreement values for the second scores of all three raters. These results revealed a weak agreement for dimension A (ICC 0,374), a good agreement for dimension B (0,603) but a very good agreement for all other dimensions and the total score (ICC ranges from 0,836-0,996). The overall SEM was 3,15%, with an SDC of 8,73 %.

Fig 1. Bland-Altman plot representing the difference between the live- and the video-scoring of the total score of the gross motor function measure-88. The average difference and the 95% confidence interval (±1,96 SD) of the difference are indicated.

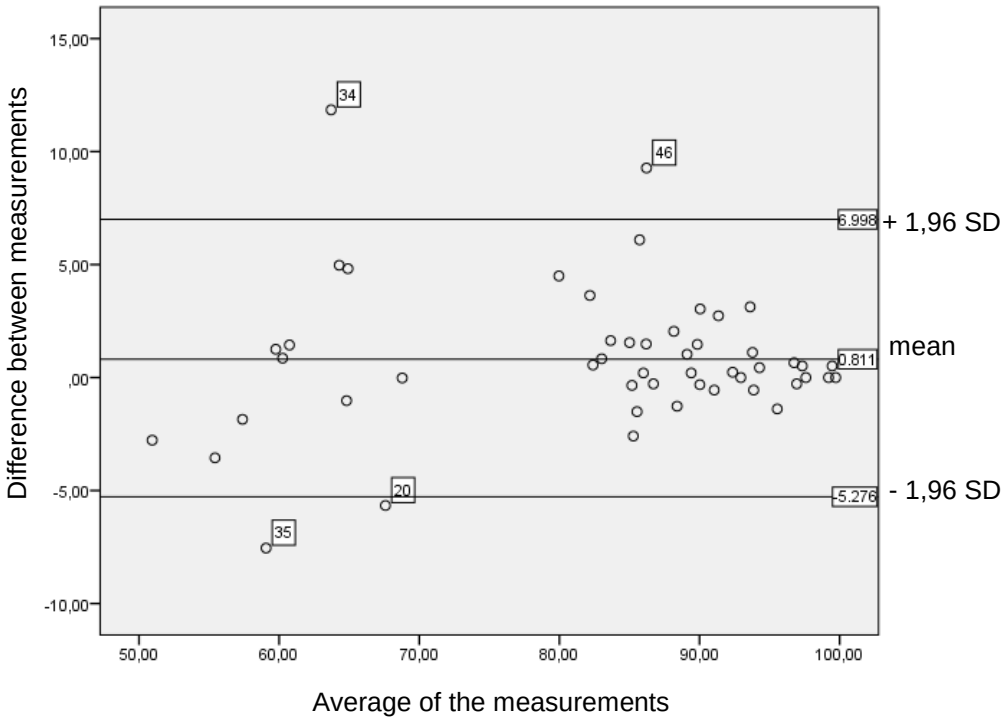

Table 1. Live-and video-scores of the GMFM-88 with corresponding agreement values (first rater)..

GMFM-88 parameter	Total score	Dimension A	Dimension B	Dimension C	Dimension D	Dimension E
Live scoring: Mean (SD) (%)	83,03 (14,01)	99,25 (2,13)	96,8 (6,26)	90,71 (12,46)	70,87 (25,97)	57,53 (28,12)
Video scoring: Mean (SD) (%)	82,22 (13,74)	99,25 (1,63)	97,3 (5,15)	88,00 (15,04)	69,28 (25,51)	57,28 (27,78)
Mean Diff (live-video) (SD diff)	0,81 (3,15)	0 (1,43)	-0,5 (1,97)	2,71 (11,11)	1,59 (6,65)	0,25 (3,61)
Limits of agreement (Bland Altman)	-5,28 - 7,00	-2,80 - 2,80	-4,36 - 3,36	-19,08 - 24,50	-11,44 - 14,63	-6,83 - 7,33
ICC agreement	0,973	0,72	0,938	0,667	0,965	0,992
(95%CI)	(0,952 - 0,985)	(0,553-0,831)	(0,893-0,965)	(0,481-0,796)	(0,939-0,980)	(0,986-0,995)
SEM (%)	2,28	1,00	1,42	8,01	4,79	2,53
SDC (%)	6,32	2,77	3,94	22,02	13,27	7,01

Abbreviations:
GMFM-88: Gross Motor Function Measure-88; **SD:** Standard Deviation; **Diff:** Difference as measured by Live-Video; **ICC** Intraclass correlation coefficient (ICC between 0,00-0,19 is considered as very weak, 0,20-0,39 as weak, 0,40-0,59 as moderate, 0,60-0,79 as strong and 0,80-1,00 as very strong agreement); **95%CI** : 95%confidence interval; **SEM**: t Standard Error of Measurement (SEM) calculated as **SEM** = $\sqrt{(\sigma^2_{PT}+\sigma^2_{residual})}$; **SDC:** Smallest detectable change calculated as 1,96*√2*SEM

Table 2. First and second rating of the two independent video-raters with corresponding agreement values.

	Total score		Dimension A		Dimension B	
Rater	**PT 2**	**PT 3**	**PT 2**	**PT 3**	**PT 2**	**PT 3**
Score 1: Mean (SD) (%)	78,75 (13,78)	77,89 (13,80)	97,53 (6,51)	95,41 (6,38)	94,17 (8,70)	93,17 (9,64)
Score 2: Mean (SD) (%)	77,82 (13,55)	78,78 (14,78)	96,55 (6,78)	96,51 (5,55)	92,57 (9,29)	93,57 (9,24)
Mean Difference (%)	0,93	-0,89	0,98	-1,10	1,60	-0,40
Limits of agreement (Bland Altman)	-2,63 - 4,50	-4,44 - 2,66	-4,01 - 5,97	-6,14 - 3,94	-7,96 - 11,16	-6,02 - 5,22
ICC agreement (95%CI)	0,989 (0,974-0,995)	0,989 (0,976-0,995)	0,918 (0,847-0,995)	0,894 (0,797-0,943)	0,842 (0,729-0,909)	0,954 (0,920-0,974)
SEM (%)	1,43	1,42	1,91	1,96	3,6	2,03
SDC (%)	3,96	3,93	5,29	5,43	9,97	5,62

	Dimension C		Dimension D		Dimension E	
Rater	**PT 2**	**PT3**	**PT 2**	**PT 3**	**PT 2**	**PT 3**
Score 1: Mean (SD) (%)	82,19 (16,68)	83,48 (15,71)	67,59 (25,91)	65,64 (24,87)	51,64 (26,47)	51,75 (26,58)
Score 2: Mean (SD) (%)	82,19 (16,41)	85,57 (14,79)	66,36 (25,22)	66,77 (24,81)	51,42 (26,23)	51,47 (26,80)
Mean Difference (%)	0,62	-2,10	1,23	-1,13	0,22	0,28
Limits of agreement (Bland Altman)	-11,95 - 13,19	-13,55 - 9,36	-6.91 - 9,36]	-9.81 - 7,56]	-6,03 - 6,48	-7,24 - 7,80
ICC agreement (95%CI)	0,926 (0,873-0,957)	0,919 (0,853-0,955)	0,986 (0,975-0,992)	0,983 (0,971-0,991)	0,993 (0,987-0,996)	0,990 (0,982-0,994)
SEM (%)	4,51	4,35	3,03	3,2	2,24	2,69
SDC (%)	12,49	12,05	8,39	8,86	6,2	7,45

Abbreviations:
GMFM-88: Gross Motor Function Measure-88; **SD:** Standard Deviation; **Diff:** Difference as measured by score1-score2; **ICC** Intraclass correlation coefficient (ICC between 0,00-0,19 is considered as very weak, 0,20-0,39 as weak, 0,40-0,59 as moderate, 0,60-0,79 as strong and 0,80-1,00 as very strong agreement); **95%CI** : 95% confidence interval; **SEM**: Standard Error of Measurement (SEM) calculated as **SEM** = $\sqrt{(\sigma^2_{PT}+\sigma^2_{residual})}$; **SDC:** Smallest detectable change calculated as $1,96*\sqrt{2}*SEM$

Table 3. Overall inter-rater agreement values for the second ratings of all 3 raters.

GMFM-88	Total score	Dimension A	Dimension B	Dimension C	Dimension D	Dimension E
ICC agreement	0,949	0,374	0,603	0,836	0,962	0,966
(95%CI)	(0,808-0,979)	(0,200-0,548)	(0,417-0,746)	(0,723-0,905)	(0,938-0,977)	(0,894-0,985)
SEM (%)	3,15	4,23	5,34	6,35	4,93	4,99
SDC (%)	8,73	11,72	14,8	17,6	13,67	13,83

Abbreviations:
GMFM-88: Gross Motor Function Measure-88; **SD:** Standard Deviation; **ICC** Intraclass correlation coefficient (ICC between 0,00-0,19 is considered as very weak, 0,20-0,39 as weak, 0,40-0,59 as moderate, 0,60-0,79 as strong and 0,80-1,00 as very strong agreement); **95%CI** : 95% confidence interval; **SEM**: Standard Error of Measurement (SEM) calculated as $\sqrt{(\sigma^2_{PT}+\sigma^2_{residual})}$; **SDC:** Smallest detectable change, calculated as $1,96*\sqrt{2}*SEM$

Discussion

This paper aimed to investigate the agreement between the live- and video-scores of the Gross Motor Function Measure-88.

The results showed a very good agreement between the live-and video-ratings for the total scores. The ICC of 0,973 demonstrated very good agreement. Additionally, the mean value of the difference (0,81%, demonstrated on the Bland Altman plots) did not significantly differ from 0, demonstrating no fixed or consistent bias.

Nevertheless, the ICC values comparing the association between the video-versus live scores were lower in comparison to the intra-rater assessment values (ICC 0,989 and 0,989) but higher in comparison to the inter-rater agreement as evaluated by video-ratings (ICC 0,949). Similarly, the SEM was lower for video versus live agreement (SEM 2,28) compared to the intra-rater values (SEM 3,32), but higher in the comparison to the inter-rater values (SEM 3,15).

In general, the lowest ICC scores were found for dimension A and C. The lower agreement of dimension A is consistent with previous results of other authors, as these items are more difficult to score and more prone to subjectivity.[4,6] The lower agreement in dimension C was probably caused by missing items, as most of the items that could not be scored based on the video-recordings, were items from dimension C. Items from dimension C usually require more active involvement and demonstration by the tester, who thereby often hinders the camera. These results suggest that it might be recommendable to use an additional person for filming. However, this might distract the child and therefore influence the test results negatively.

In general, our results showed lower inter-and intra-rater agreement values in comparison to previous studies using experienced raters.[5,6,13] Still, higher agreement values were observed compared to Nordmark et al., who investigated the agreement between un-experienced raters.[8] As the test manual can be used during the video-ratings, experience of the rater might of less importance regarding reliability. In our study, the master students were trained in the observation of children's motor functioning and familiar with the test but were not as experienced in rating the gross motor function measure as the first assessor.

Since only ambulant children with bilateral spastic cerebral palsy were included, our study population was relatively homogeneous. This limitation specifically concerns dimension A and B of the study, as very high scores were registered in these dimensions.

The SEM and SDC values were very variable for the different types of scoring. This requires carefulness when interpreting the results of clinical trials. For the total test score, the SEM of 2,28% should be considered as the measurement error when comparing live-to video-scores. When using video-scores, 1,43 and 3,15 % should be considered for intra-and- inter-rater reliability respectively. When interpreting changes in scores of the Gross Motor Function Measure-88 in future intervention studies, the appropriate SEM and SDC should be considered and this should depend on the type of scoring used.

Clinical messages

- There was a high agreement between the live- and video-scores of the gross motor function measure-88;
- Agreement between live- and video-scores was lower compared to the intra-rater reliability of video-scores;
- When using video-scores in a clinical trial, the appropriate reliability measures should be considered.

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Part 4
Evaluation

Chapter 5

A randomized, single-blind cross-over design evaluating the effectiveness of an individually defined, targeted physical therapy approach in treatment of children with cerebral palsy

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Abstract

Objective

A pilot study to compare the effectiveness of an individual therapy program with the effects of a general physical therapy program.

Design

A randomized, single-blind cross-over design.

Participants

Ten ambulant children with bilateral spastic cerebral palsy, age four to nine years.

Intervention

Participants were randomly assigned into a ten-week individually defined, targeted or a general program, followed by a cross-over.

Main outcome measures

Evaluation was performed using the Gross Motor Function Measure-88 and three-dimensional gait analysis. General outcome parameters were Gross Motor Function Measure-88 scores, time and distance parameters, gait profile score and movement analysis profiles. Individual goal achievement was evaluated using z-scores for gait parameters and Goal Attainment Scale for gross motor function.

Results

No significant changes were observed regarding gross motor function. Only after individualized therapy, step- and stride-length increased significantly (p = 0.022; p = 0.017). Change in step-length was higher after the individualized program (p = 0.045). Within-group effects were found for the pelvis in transversal plane after the individualized program (p = 0.047) and in coronal plane after the general program (p = 0.047). Between-program differences were found for changes in the knee in sagittal plane, in the advantage of the individual program (p = 0.047). A median difference in z-score of 0.279 and 0.419 was measured after the general and individualized program, respectively. Functional goal attainment was higher after the individual therapy program compared with the general program (48 to 43.5).

Conclusion

The results indicate slightly favorable effects towards the individualized program. To detect clinically significant changes, future studies require a minimal sample size of 72 to 90 participants.

Introduction

Many different techniques and approaches are available for physical therapy treatment of children with cerebral palsy.[1] Although promising results are available for isolated physical therapy techniques, evidence about the outcome of complete programs using a combination of different techniques is more limited.[2,3]

Several research studies have evaluated the effects of goal-setting in physical therapy of chil- dren with cerebral palsy.[4–9] The results of a recent systematic review, however, could not provide support for the effectiveness of goal-setting within an activity-focused approach on treatment outcome.[10] The authors of this review found that none of the included studies were designed specifically to determine the effect of goal-setting per se, as goal- setting was additional to guiding the interventions.

An important advantage of goal-oriented physical therapy however, is the potential benefit to enhance training specificity. Goal planning, in that context, can be used to identify the tasks and contexts of particular interest and thereby, to use a more targeted and focused approach.[11]

In a previous study, we developed and validated a clinical decision framework based on the hypothesis- oriented algorithm for clinicians[12] and the International Classification of Functioning, Disability and Health (ICF) for Children and Youth,[13,14] Organizing assessment results within the framework of the International Classification of Functioning provides a profound and complete overview of the child and results in an objective problem list for the child. In children with cerebral palsy, however, this list is generally very extensive. Therefore, as a therapist, one has to prioritize and select possible and realistic targets in treatment. The hypothesis-oriented algorithm for clinicians provided this strategy-oriented structure for clinical reasoning and was used to reduce the long problem list to a selection of main problems. The combined approach of both frameworks was demonstrated as a positive structure that supported clinical reasoning in pediatric physical therapists.[14]

As a next step, a pilot study was set up to use this clinical reasoning structure in a controlled intervention. This study, therefore, aims to investigate the effectiveness of an individually defined and targeted physical therapy approach using structured clinical reasoning on gait and gross motor function of ambulant children with bilateral spastic cerebral palsy. Using a randomized, repeated-measures, cross-over design, the outcome of this approach is compared with the outcome of a gen- eral program.

It is hypothesized that an individually defined targeted approach, based on structured clinical reasoning and including the results of three- dimensional gait analysis, provides better results than a general program.

Method

Participants were selected through the cerebral palsy reference center of the University Hospital of Leuven. Inclusion criteria were diagnosis of bilat- eral spastic cerebral palsy, Gross Motor Function Classification System Level I, II or III, age between four and nine years and sufficient cooperation to understand and execute simple verbal instructions. Patients were excluded when they showed severe muscle contractures or bony deformities, a history of multilevel orthopedic surgery or additional dis- orders that would make participation to the physi- cal therapy programs not feasible (e.g. severe cognitive disorders, deafness, blindness, etc.). The programs were planned at a time when children had not received botulinum-toxin A injections within the previous six months. A convenience sample of 10 children represented a manageable number to reassure an adequate follow-up for this pilot study. Children were recruited between January 2010 and January 2012.

The programs were executed by the child's personal physical therapist, who agreed to precisely follow the prescribed program. A physical therapy degree was required, no further specialization was mandatory. The therapist and parent or caretaker of the child signed an informed consent. The study was approved by the ethical committee of the University Hospital of Leuven.

At the start of the study, the selected group of 10 children was followed for a period of 10 weeks of usual care. During this period, usual physical therapy of the child was continued without a specific intervention. A diary was provided to document the content of the therapy sessions. The therapists were asked to fill in the diary after each session, to register how much of the therapy time targeted the impairment, activity and participation level.

Subsequently, the children were enrolled in the actual intervention study. For this part of the study, an alternating treatment design with rand- omization and cross-over was set up. A first treatment program, general or individualized, was presented to the participants based on an independent randomization procedure. Randomization was performed by a person blinded and independent to the study, and was based on the principles of minimization, as described by Altman and Bland.[15] After completing one program, a wash-out period of minimally 10 weeks usual care was continued. Consequently, each child participated in the remaining program. The children that originally received the individualized program, now received the general program and vice versa (**Figure 1**).

The general treatment program was based on the most common aims in children with bilateral spastic cerebral palsy of the involved age-range[16] and was the same for all children. The main aims were in accordance with the clinical management recommendations for children with spastic diplegia as stated by the task force of the American Physical Therapy Association.[16] The general considerations reported in this statement, are musculoskeletal and neuromuscular systems including strength, range of motion, posture, balance, motor function, muscle tone and movement patterns. The program, therefore, contained exercises to improve strength, selectivity and mobility and included a set of func- tional exercises to encourage mobility and play.

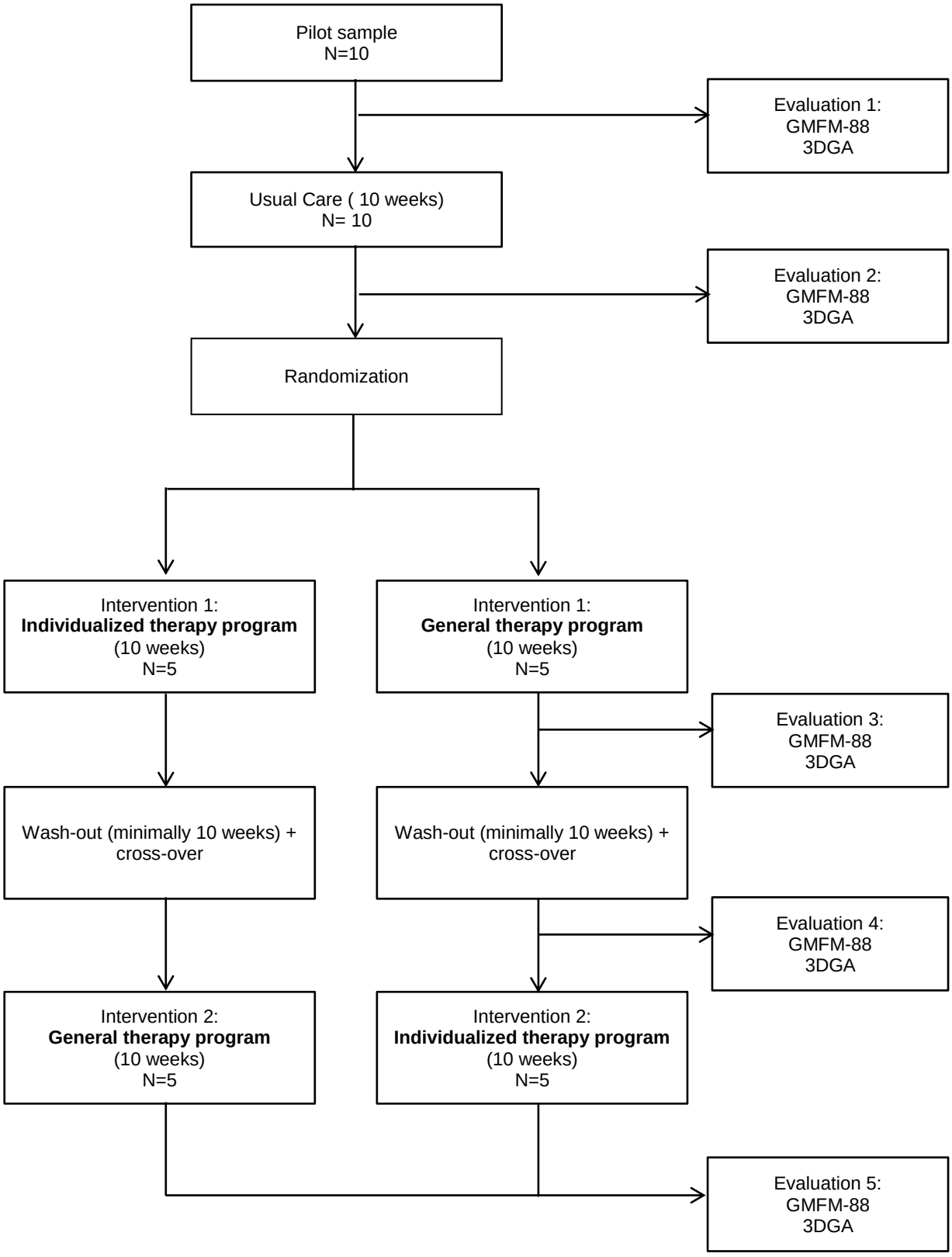

Fig 1: Study design

Abbreviations:
N:Number; **GMFM-88**: Gross Motor Function Measure 88; **3DGA:** Three-dimensional Gait Analysis

The individually defined, targeted programs (further referred to as “individualized programs”) were designed to target the specific main problems of the child, tailored to the needs of the child and specific problems at the different levels of the ICF at that particular moment. Definition of the individual problems was based on the results of all assessments. At the level of body structure and function, results of the clinical examination were considered. At activity level, three-dimensional gait analysis and gross motor function evaluation was used, complemented with a structured inter- view with the parents and child. The results of the gait analysis thereby played a major role in unrave- ling the motor problems of the child and the identification of the main problems. The programs were based on the clinical decision framework as described by Franki et al.,[14] which use the ICF and the Hypothesis-Oriented Algorithm for Clinicians (HOAC-II)[12] to identify a selection of main problems. This allowed goal-setting at all levels of the ICF, according to the needs of the child. The goals were discussed with the child’s parents or caretakers, as well as the child’s physical therapist. The designs of the programs thereby approached the reality of clinical practice, using a mixture of individual techniques, each targeting a specific problem of the child at that particular moment. The choice of the appropriate techniques to achieve a specific goal was performed according to the evidence-table designed by Franki et al. in previous systematic reviews.[2,3] **Table 1** provides an overview of the similarities and differences between the programs.

Table 1: Differences and similarities between the general, aim-oriented program and the individually defined targeted programs

	General program	**Individually defined, targeted program**
Differences	• The same program for all children; • Based on the most common problems of ambulant children with bilateral spastic CP; • Aspecific, based on general aims; • A predefined set of exercises to improve strength, selectivity and mobility and gross motor function; • The results of the three-dimensional gait analysis were not used to define of the program; • The program was not discussed with the therapist and parents.	• Different for all children; • Based on the results of the individual assessments; • Specific, tailored to the individual problems; • Goal-oriented; • Depending on the needs of the child, the exercises targeted the different levels of the ICF; • Based on structured clinical reasoning using the HOAC-II and the ICF; • The results of the three-dimensional gait analysis were included to define the program; • The program was discussed with the parents and the therapist.
Similarities	• Respected the basic principles for evidence-based PT regarding intensity and repetitions were respected. - Stretching: 3 repetitions of 30 seconds; - Strength: 3 series of 12 to 15 repetitions; - Functional exercises: not restricted. • Executed by the child's private physical therapist, familiar to the child; • At the child's usual frequency and duration of therapy.	• Respected the basic principles for evidence-based PT regarding intensity and repetitions were respected. - Stretching: 3 repetitions of 30 seconds; - Strength: 3 series of 12 to 15 repetitions; - Functional exercises: not restricted. • Executed by the child's private physical therapist, familiar to the child; • At the child's usual frequency and duration of therapy

Participants were evaluated before and after the period of usual care, as well as after each treatment program using the gross motor function measure,[17] standardized clinical examination and three-dimensional gait analysis.

The **Gross Motor Function Measurement-88** is a standardized clinical instrument to evaluate change in gross motor function in children with cerebral palsy. It fulfills the criteria of reliability and validity with respect to responsiveness to change.[18–20] The gross motor function evaluations were performed by the first author of this study, but video-taped. Two independent assessors, blinded to the set-up of the study, scored the video. The average of both video-scores was used.

Clinical examination and three-dimensional gait analysis were performed according to the standardized protocol used at the Laboratory of Clinical Motion Analysis of the hospital. An independent assessor, blinded to the study, performed both assessments. Clinical examination included measurements of range of motion (goniometrical measurements), spasticity (Modified Ashworth Scale and Tardieu), selectivity (selective motor scale) and muscle strength (manual muscle testing).[21–23] For gait analysis, information on kinematics, kinetics and spatial-temporal data was collected by a 16-camera VICON system (Nexus PluginGait marker set, Oxford Metrics, Oxford, UK), two ATMI forceplates (Advanced Medical Technology, Inc, Watertown, MA, USA) and a 16-channel EMG device (Zero-Wire, Cometa, Milano, Italy). Three representative trials were selected. Specific gait parameters were automatically extracted using a custom-made Matlab graphical user interface (Mathworks®, Natick, MA, USA). The results of the children were compared with the means and standard deviations of a reference group. Our reference data was based on the data obtained from a control group of 55 typi- cally developing children, with a mean age of 10.94 years, range between the ages of four years and 18 years. Outcome parameters were derived at two different levels (**Figure 2**).

At a **first, general level**, overall therapy success was evaluated using outcome parameters that provide a general outcome description. From the Gross Motor Function Measure, the total and dimensional percentage scores were extracted. Regarding gait analysis, time and distance parameters, the Movement Analysis Profiles and the Gait Profile Score were calculated.[24] The average scores from the left and right sides were used. The Movement Analysis Profiles were calculated as the root mean square error between the point-by-point comparison of the lower limb joint angle and the averaged joint angle of the reference group. Calculation of the average of all lower limb joint angles results in the Gait Profile Score, which summarizes the overall severity of gait pathology.[24]

Second, **individual goal achievement** was evaluated before and after the programs.
For the **individual, functional goals extracted from the Gross Motor Function Measure-88**, the original version of the Goal Attainment Scale (GAS) was used, as developed by Kirusek et al.[25] and used in several pediatric rehabilitation studies.[26,27] Before the start of the programs, individual goals based on specific goal items of the Gross Motor Function Measure-88, were formulated according to the SMART-principle.[28] A score between –2 and 2 was given depending on the goal achievement. Subsequently, a converted T-score and the percentage of achieved goals ($\geqslant 0$) were calculated.

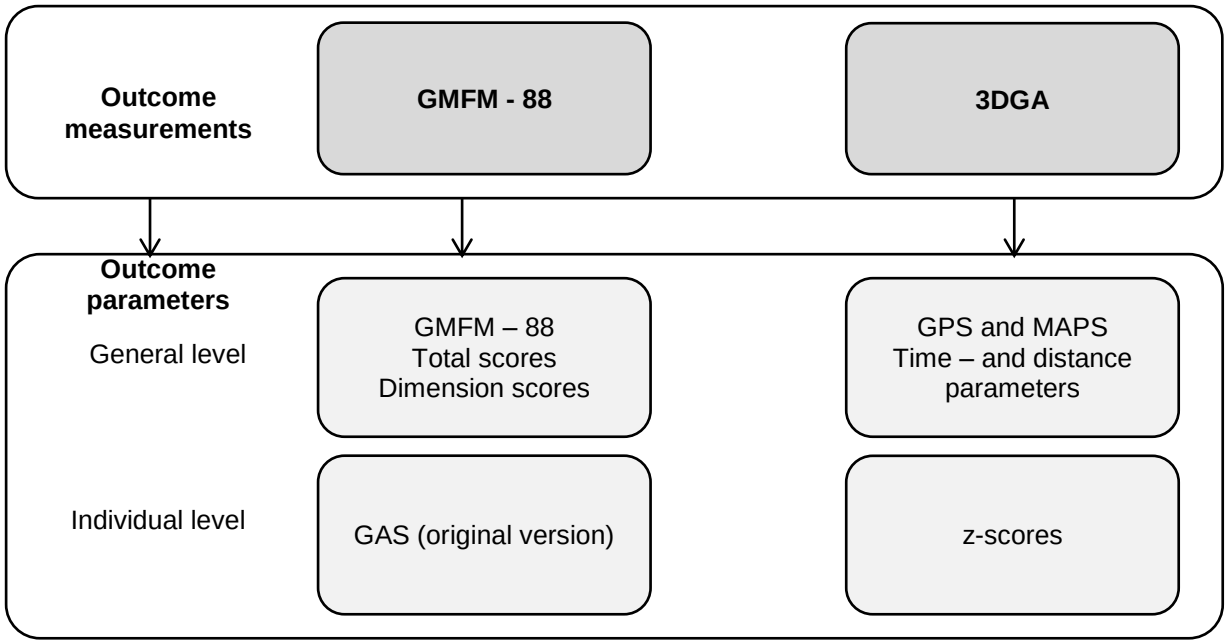

Fig 2. Outcome measurements and parameters

Abbreviations:

GMFM-88: Gross Motor Function Measure-88; **3DGA:** three-dimensional gait analysis; **GAS:** Goal Attainment Scale; **GPS:** Gait Profile Score; **MAPS:** Movement Assessment Profiles

Individual goal achievement of the specific goals derived from gait analysis was evaluated using z-scores and proportional changes. As a first step, two to eight specific deviated gait parameters were identified as specific goals for each child. Each goal was represented by a relevant data point that was expected to improve, such as maxima, minima, range of motion and angles at specific events in the gait cycle. These data points were defined based on literature and by clinical experts in gait analysis of children with cerebral palsy.[29,30] The parameters were converted into z-scores. A z-score represents the deviation from a reference group (z = (mean child – mean typical child)/standard deviation (SD) typical child). To evaluate the z-scores, the absolute difference to zero, in either direction, was considered as an equal deviation from typical gait. Following standard clinical practice, we assumed that a deviation greater than 1 could be considered as pathological. Therefore, all z-scores smaller than 1 were eliminated.[29] Consequently, the median z-scores of the specific goals were calcu- lated. Z-scores are non-dimensional and, therefore, allow the comparison of different parameters. Finally, the proportional change was calculated (proportional change = (|pre median z-score| – |postmedian z-score| / |premedian z-score|)). This percentage score allowed a description of therapy success. A positive proportional change described a successful therapy and a negative proportional change indicated an unsuccessful therapy. For the individually defined programs, these individual goals were taken into account to design the programs. For the general programs, the individual goals were only considered as outcome parameters, but not specifically taken into account to during the programs.

Statistical analyses were performed in different steps. First, other treatment modalities, including the use of orthosis, aquatic and horse-riding therapy, during the different intervention periods were compared using a McNemar test. Similarly, differences in the frequency and duration of the physical therapy sessions were tested using the related- samples Wilcoxon Signed-Rank test. Secondly, the median and interquartile range (IQR) of all parameters before and after the programs were calculated as appropriate. A related-samples Wilcoxon signed rank analysis was applied to calculate within-group differences of pre- and postvalues. In order to evaluate the effect between the different interventions, a related-samples Friedman's two-way analysis of variance by rank analysis with post-hoc related-samples Wilcoxon signed rank analysis was applied. This analysis compared the difference scores of the outcome parameters (difference score = Pre – Post), further described as between-program differences.

All statistics were performed in SPSS 21 IBM SPSS Statistics for Windows, Version 21.0. (IBM Corp, Armonk, NY, USA).

Results

Patient characteristics, as well as duration, frequency of therapy and the use of orthosis are reported in **Table 2** and were not significantly different between the different periods. The results of the diaries provided during the period of usual care show that 55.8% of therapy time was dedicated at impairment level. Thereby, 22.5% (interquartile range (IQR) 16.7–28.6) targeted joint mobility and muscle length and 33.3% (IQR 23.9–35.9) strength. A median of 48.1% (IQR 35.6–54.8%) of therapy time was used to train activities. Only 1.55% (IQR 0–6.5%) of therapy time addressed specific participation restrictions.

Table 2: Patient Characteristics

Patient characteristics			
Age (median + IQR)	6,2y (4,6 – 7,0 y)		
Gender (n) (male/female)	6/4		
GMFCS (n) (I/II)	5/5		
History of BTX-A injections (n)	4		
	UC	GT	IT
Use of day orthosis (n)			
>50%/day	8	7	7
<50%/day	0	2	2
no	2	1	1
Use of night orthosis (n)			
>50%/night	1	2	0
<50%/night	0	2	4
no	9	6	6
Frequency of PT (times/week) Med (IQR)	2 (2-3)	3 (2-3)	3 (2-4)
Duration of PT (minutes /session) Med (IQR)	40 (30-60)	40 (30-60)	40 (30-60)
Total PT (minutes/week)	105 (80-120)	120 (90-128)	120 (90-123)

Abbreviations:
UC: period of Usual Care; **GT:** period of general treatment; **IT:** period of Individualized, targeted treatment
GMFCS: Gross Motor Function Classification System; **PT:** Physical Therapy; **N:** number; **Y:** year

The median total percentage scores of the Gross Motor Function Measure-88 showed only very limited and no significant changes, but a slight increase after the individualized program, from 93.5 to 95.5% (**Table 3**).

Dimension D and E showed a slight decrease during the period of usual care, but an increase after the period of individualized therapy. Evaluating the between-program differences revealed no significance.

Considering time and distance parameters showed significant improvements on step and stride length ($p = 0.022$ and 0.017, respectively) after the period of individualized therapy. Between- program effects were, therefore, also significant for step length in the advantage of the individualized therapy program ($p = 0.045$). Changes in Gait Profile Score and Movement Assessment Profile Scores are provided in **Table 4**. Significant changes were found after the individualized program, for the pelvis at the transversal plane ($p = 0.047$), while for the general program differences were found for the pelvis in the coronal plane ($p = 0.047$). Between- program changes were only found for the knee in the sagittal plane and this in the advantage of the individualized therapy program ($p = 0.047$). The results of the Gait Profile Score showed slight progress after the individualized program, whereas deterioration was registered after the general program. No changes were measured after the period of usual care.

Functional goal attainment was higher after the individualized than after the general therapy pro-gram: 48.0 (IQR 43.3–59.3) compared with 43.5 (IQR 30.3–43.5) ($p = 0.285$). Additionally, the averaged percentage of achieved goals was 50.0 (IQR 25.0–100) after the individualized program, compared with 41.7 (IQR 0–75) for the general program ($p = 0.589$). Nevertheless, none of the programs caused treatment success (GAS $\geqslant$50) and the differences were not statistically significant. **Table 5** provides an overview of the changes in z-scores for the goals derived from the gait analysis after both training periods. This analysis revealed a median difference in z-score (pre– post) of 0.279 and 0.419 after the period of general and individualized therapy program, respectively. This was translated in a proportional change of 12% and 19%, respectively. These changes, how- ever, were not significantly different.

Table 3. Changes in GMFM-88 total and dimensional scores before and after the period of usual care, the general therapy program and the individualized therapy program.

	Usual Care			General Therapy			Individualized Therapy			p between		
	Pre	Post	p	Pre	Post	p	Pre	Post	p	UC vs	UC	GT
	Median (IQR)	Median (IQR)	(within)	Median (IQR)	Median (IQR)	(within)	Median (IQR)	Median (IQR)	(within)	GT	vs IT	vs IT
Total GMFM	94,5 (88,2-98,3)	94,5 (90,1-98,5)	0,173	92,2 (87,0-99,3)	92,5 (88,5-97,7)	0,333	93,2 (87,6-97,9)	95,5 (89,4-98,7)	0,065	0,173	0,917	0,441
GMFM-A%	100 (100-100)	100 (100-100)	1	100 (100-100)	100 (100-100)	1	100 (100-100)	100 (100-100)	1	1	1	1
GMFM-B%	100 (100-100)	100 (100-100)	1	100 (100-100)	100 (100-100)	1	100 (100-100)	100 (100-100)	1	1	1	1
GMFM-C%	100 (96,4-100)	100 (96,4-100)	1	100 (82,1-100)	100 (85,7-100)	0,068	100 (84,2-100)	100 (89,3-100)	0,18	0,317	1	1
GMFM-D%	89,1 (70,8-100)	87,8 (73,1-100)	0,581	90,4 (83,0-100)	90,4 (82,1-100)	0,917	89,1 (84,0-98,1)	94,9 (82,1-100)	0,667	0,581	0,465	0,735
GMFM-E%	86,5 (72,7-98,6)	83,7 (75,9-97,6)	0,893	77,1 (65,8-96,0)	80,9 (67,5-92,0)	0,572	78,8 (65,8-89,9)	82,6 (68,8-93,4)	0,675	0,344	0,752	0,932

Abbreviations:
GMFM: Gross Motor Function Measure-88; **GMFM-A:** Gross Motor Function Measure dimension A; **GMFM-B:** Gross Motor Function Measure Dimension B; **GMFM-C:** Gross Motor Function Measure Dimension C **GMFM-D:** Gross Motor Function Measure Dimension D **GMFM-E:** Gross Motor Function Measure Dimension E **IQR:** Interquartile Range; **Diff:** difference (pre-post); **p (within):** p value for the within-group effects provided by the Wilxocon Signed Ranks Test; **p value (between):** p value for the between-group effects provided by the Wilcoxon Signed Ranks Test

Table 4. The Movement Assessment Profiles (MAPS) and Gait Profile Score (GPS) before and after the period of usual care (UC), the GT program (general therapy) and the individually defined targeted therapy program (IT).

	Usual Care			General Therapy			Individualized therapy			p between		
	Pre	Post	p	Pre	Post	p (within)	Pre	Post	p	UC vs	UC	GT
	Median (IQR)	Median (IQR)	(within)	Median (IQR)	Median (IQR)		Median (IQR)	Median (IQR)	(within)	GT	vs IT	vs IT
MAPS Sag P	8,8 (3,4-12,2)	9,3 (4,3-12,9)	0,575	8,4 (4,0-15,1)	9,0 (5,6-14,0)	0,386	9,3 (6,1-12,9)	8,5 (4,7-14,7)	0,878	0,575	0,721	0,799
MAPS Sag H	9,9 (5,4-13,2)	9,8 (5,4-13,5)	0,508	8,6 (6,0-15,9)	10,2 (6,8-14,3)	0,575	10,8 (8,1-13,5)	10,2 (5,7-15,6)	0,799	0,386	0,878	0,575
MAPS Sag K	13,5 (7,6-19,3)	13,4 (10,2-16,7)	0,959	13,6 (8,3-16,1)	13,6 (8,7-19,8)	0,333	14,2 (9,2-20,9)	12,9 (7,3-18,9)	0,203	0,646	0,203	0,047
MAP Sag A	7,0 (5,5-12,3)	7,0 (4,9-13,1)	0,799	6,5 (4,5-11,1)	7,3 (5,5-10,4)	0,878	7,0 (4,9-14,7)	7,8 (5,6-12,5)	0,959	0,445	0,959	0,508
MAPS Cor P	2,9 (2,1-4,0)	2,8 (2,2-4,4)	0,721	3,4 (2,2-4,1)	3,3 (1,9-3,6)	0,047	3,4 (2,3-4,8)	3,3 (2,1-3,6)	0,241	0,721	0,878	0,799
MAPS Cor H	4,0 (3,6-5,3)	4,2 (3,4-5,1)	0,878	4,0 (3,7-5,9)	4,9 (3,6-6,4)	0,386	4,7 (4,0-5,3)	4,3 (4,0-5,9)	0,799	0,508	0,878	0,445
MAPS Trans P	8,4 (5,3-11,4)	7,3 (5,5-8,4)	0,241	6,5 (4,9-8,2)	5,1 (4,0-7,7)	0,508	6,9 (5,5-9,3)	5,7 (3,6-8,4)	0,047	0,333	0,241	0,139
MAPS Trans H	7,0 (6,4-12,4)	7,6 (6,1-11,5)	0,333	7,8 (6,3-12,2)	6,9 (5,2-9,3)	0,285	7,4 (5,9-10,0)	8,7 (6,1-10,47)	0,445	0,203	0,508	0,386
MAPS Trans F	10,4 (5,2-17,5)	9,0 (6,5-16,2)	0,445	10,9 (5,4-13,9)	9,1 (5,5-20,9)	0,508	9,1 (6,7-17,3)	10,0 (6,5-14,4)	0,878	0,139	0,799	0,799
GPS	10,0 (7,0-12,5)	9,7 (6,6-12,9)	0,445	9,9 (5,9-11,9)	9,4 (6,7-15,2)	0,386	10,5 (6,8-13,0)	10,9 (6,5-13,4)	0,721	0,139	0,646	0,333

Abbreviations
IQR: Interquartile Range; **p(within):** p value for the within-group effects of the z-scores as provided by the Wilcoxon Signed-Ranks test; **p (between):** p value for the evaluation of the between-program effects of the z-scores as provided by the Wilcoxon Signed-Ranks test; **Diff:** difference between the pre-and post value; **PC:** Proportional Change, calculated by (z pre- z post)/z pre; **UC:** Usual Care; **IT:** Individualized Therapy Program; **GT:** General Therapy Program; **MAPS:** Movement Assessment Profile; **Sag:** Sagittal plane; **Cor:** Coronal plane; **Trans:** Transversal plane; **P:** Pelvis; **H:** Hip; **K:** knee; **A:** Ankle

Table 5. Overview of the changes in z-scores and the converted proportional changes as derived from the individual goals based on the results of the three-dimension gait analysis

Child	General Program					Individualized program					p
	Pre	Post	Diff	PC	p (within)	Pre	Post	Diff	PC	p (within)	(between)
1	2,967	3,142	-0,175	-6%		2,718	2,656	0,062	2%		
2	1,646	1,184	0,462	28%		1,669	0,738	0,931	56%		
3	2,334	1,576	0,759	33%		2,271	1,629	0,641	28%		
4	3,093	2,475	0,618	20%		2,410	1,672	0,738	31%		
5	1,592	2,026	-0,434	-27%	0,368	2,449	2,536	-0,088	-4%	0,169	0,333
6	1,439	0,747	0,693	48%		3,349	3,003	0,347	10%		
7	1,485	0,741	0,744	50%		1,867	1,179	0,688	37%		
8	1,472	1,886	-0,414	-28%		1,453	0,961	0,492	34%		
9	2,134	2,038	0,096	4%		2,703	2,804	-0,101	-4%		
10	3,263	4,127	-0,864	-26%		2,788	3,681	-0,893	-32%		
50th (Median)	1,889	1,817	0,278	12%		2,429	2,104	0,419	19%		
25th	1,481	1,070	-0,419	-27%		1,817	1,124	-0,090	-4%		
75th	2,990	2,641	0,705	36%		2,735	2,853	0,700	35%		

Abbreviations
p(within): p value for the within-group effects of the z-scores as provided by the Wilcoxon Signed-Ranks test; **p (between):** p value for the evaluation of the between-program effects of the z-scores as provided by the Wilcoxon Signed-Ranks test; **Diff:** difference between the pre-and post value; **PC:** Proportional Change, calculated by (z pre- z post)/z pre

Discussion

The effects of the present study demonstrated a tendency in favor of the individually defined, targeted program on gait and gross motor function. On a general level, significant advantageous results were found for the individualized program on step length and for the movement assessment profiles of the knee in the sagittal plane. At an individual level, goal achievement was non-significantly higher after the individualized program compared with the general program, and this for both the functional goals as the goals based on gait analysis.

The applied approach was representative for the reality of clinical practice, which usually makes use of a mixture of individual techniques, each targeting a specific problem. It differed from more recent goal-oriented, activity-focused interventions that usually apply a 'top-down' approach within the IFC.[31] The clinical reasoning strategy used to define the individually tailored programs was based on a horizontal approach and, therefore, considered all dimensions of the model of equal impor- tance. It aimed to specifically address the thoughts behind the dynamic and interactive concept of the ICF and, thereby, also the multidimensional diffi- culties following cerebral palsy. Gait rehabilitation was an essential part of our programs and a large part of our outcome parameters consisted of gait measures. This was motivated by previous research that highlighted that both gait speed and the amount of gait pathology adversely affected quality of life, as reported by the child and their proxy.[32] This unique but realistic concept can be considered as a major strength of our study.

Another strength of the study was the randomized cross-over design, allowing comparison of individual changes. The length of the wash-out period, thereby, was the same as the duration of the program. Wash-out periods are required assuming that therapy effects are still present for a certain period after the intervention. In our recent systematic literature review, none of the studies using follow-up periods detected treatment effects lasting longer than the duration of the intervention itself.[2] Additionally, in the current study, only the contents of the programs were adjusted and the researchers did not interfere with the frequency and duration of the programs. These considerations allowed us to be confident that a 10 weeks wash- out period is sufficient to avoid overflow effects.

Nevertheless, only a limited number of parameters showed statistical significant changes. A first and major reason might be the small sample size. Although the set-up of the study was controlled and randomized, the sample size of 10 children provided low power. Post-hoc power calculation using SAS Power and Sample Size 3.1 (SAS insti- tute Inc, Cary, NC, USA) revealed the gait profile score as being most vulnerable for the limited power. Considering a change score of 3° on the gait profile score as clinically relevant, provided 8% power. To be able to detect this change, future studies should use between 72 (80% power) to 91 participants (90%). The Gross Motor Function Measure-88 was identified as least vulnerable for limited power. When using the Gross Motor Function Measure-88 as an outcome

measure, future studies should use between 12 (80% power) to 15 participants (90%) to detect a change of 3%.

A second restriction might be related to the fact that both programs were executed by the child's personal physical therapist. Besides the strict follow-up, influencing factors as cooperation of the therapist and correct execution of the programs still existed and were probably important confounding variables. The adequate follow-up could probably not completely control the correct appliance and execution of the prescribed programs. During the period of usual care, regular physical therapy sessions were continued. Therefore, the contents of the programs were dependent of the physical therapist responsible for the child and a large variation in therapy contents was possible. A diary was used to be able to monitor therapy contents without interfering with the quality and contents of therapy. Follow-up visits might have influenced therapy behavior and, therefore, diaries were preferred. Although the diary was a useful and objective tool, the physical therapists reported that filling the diary was time-consuming and caused a relatively large administrative burden. Therefore, diaries were avoided during the periods of interventions. The need for a diary during the periods of intervention was not strong as the programs were prede- fined using specific exercises. Therefore, follow-up during the interventions was performed using regular visits and phone-calls. Nevertheless, although the therapy programs were developed with more than 50% of the exercises targeting activity level, this did not necessary mean that therapy time was divided accordingly. Providing a diary during the periods of interventions might have provided more precise insight into the follow-up of the programs.

A third limitation was the duration of the applied interventions. Ten weeks was a relatively short period to reach significant improvements. Guidelines by the National Strength and Conditioning Association stated that strength training should be applied 2–4 times a week on non-consecutive days for at least 8–20 weeks in order to find an increase in muscle strength.[33] Therefore, a study of longer duration could have resulted in greater benefits. Additionally, longer-term studies allow an evaluation at participation level.[34] As the study period of 10 weeks was too short to expect significant changes at participation level, outcome evaluation did not include participation measures.[34] Nonetheless, the design of the individualized integrated discussions with parents, children and therapists. In that way, individual participation limitations were considered during the development of the programs and were considered of equal value among the assessment measures when designing the programs.

Additionally, while the inclusion criteria allowed inclusion of children with Gross Motor Function Classification Level III, no children func- tioning at this level could be included for this inter- vention study. Children with Gross Motor Functional Classification Level III are more fre- quently in need of tone-reduction by means of bot- ulinum toxin and, therefore, we were not able to include these children during the predefined win- dow of six months post-botulinum toxin.

The results demonstrated very limited difference but advantageous results of the interventions compared with the period of usual care. This might be explained by the variation in therapy as reported by the

different therapists. Remarkably however, as all children were ambulant and thus very func- tional, the percentage of time spent on impairment level (range between 40.3% and 70.3% of therapy time) was relatively high during the period of usual care. During the periods of intervention, the number of exercises targeting activity level was usually higher than 50%. Although this implies that during the interventions the average therapy time spent at activity level was probably higher than 50%, it might be a future recommendation to monitor therapy time more accurately during the periods of intervention as well.

The proportional change scores allowed us to describe therapy success by explaining the gain or loss in performance. However, one needs to be careful with the interpretation of a proportional change score because it is easily influenced by extraneous factors (such as the standard deviation of the control group), and it may therefore overes timate outcome results.

The study results of the present study are concur rent with the previous experience of our research group, in which we evaluated the effectiveness of an individually defined approach.[9] This study showed fundamental differences with the present study: the order of the programs was not randomized and they were of shorter duration (6 weeks). Additionally, the present structured clinical reasoning was not yet the basis for the individual programs.

The recent review by Brogren et al. concluded that the scientific evidence regarding goal-setting is inconclusive.[10] Also, Law et al. could not find significant differences between a context-focused therapy containing goal-setting, and child and activity focused therapy without goal-setting.[35] However, these studies used goal-setting within an activity-focused approach only. This again supports our hypothesis that a successful treatment approach is probably multifactorial and depends on several aspects like correct appliance of techniques, adequate goal and priority setting, frequency and duration of therapy, timely tone-reduction, attention for posture and positioning and maybe many others. Future research stud- ies should probably continue to look for the effectiveness of individually tailored approaches addressing the heterogeneous nature of cerebral palsy, not only in individual goal-setting, but also in the choice of techniques and approaches.

In **conclusion**, the results can only provide an indication of the additional value of goal-setting within an integrative approach. To confirm our hypothesis however, future studies should include sample sizes of minimally 72 participants, evaluate long-term effects and use individual goal-achievement measures.

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Chapter 6

Therapy effects and prognostic factors in a general and an individually defined physical therapy program for ambulant children with bilateral spastic Cerebral Palsy

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Abstract

The first aim of this study was to compare the effectiveness of an individually-defined therapy program (IT) and a general therapy program (GT) on gait and gross motor function in children with bilateral Cerebral Palsy (CP). The second aim was to evaluate interaction effects, time-effects, treatment with Botulinum Toxin A, age, gross motor function classification scale (GMFCS), therapy frequency, and treatment quality as factors influencing outcome.

Forty children with spastic bilateral CP (mean age 6 years 1month, GMFCS I-III) were included in an evaluator-blinded, randomized controlled-trial to receive either IT or GT over a 10 week period. Nineteen of these children were enrolled into a second and/or third program, resulting in 60 interventions. Outcome was evaluated using the Gross Motor Function Measure–88 (GMFM-88), three-dimensional gait analysis, the Five-Times-Sit-To-Stand-Test (FTSTS) and the Timed-Up-and-Go (TUG). Primary outcome evaluated individual goal achievement using the Goal Attainment Scale (GAS) for gross motor function goals and z-scores for goals based on specific gait parameters. Secondary outcome included total- and dimensional scores of the GMFM-88, time- and distance gait parameters, Gait Profile Score, Movement Analysis Profiles and time needed to complete the TUG and FTSTS.

There was a clear tendency for a higher individual goal achievement after the IT program compared to the GT program. There were higher, but non-significant GAS and z-score changes following the IT program compared to the GT program (GAS: 46.2 for the IT versus 42.2 for the GT group, $p=0.332$, ES 0.15; z-score: 0.135 for the IT compared to 0.072 for the GT group, $p=0.669$, ES 0.05).

Significant time-effects could be found on GAS-T score ($p<0.001$) and the GMFM-88 total score ($p<0.001$). Age was identified as a predictor for GAS and GMFM-88 improvement ($p=0.023$ and $p=0.044$).

In conclusion, no significant differences could be registered between the effects of an IT and a GT approach. Future research should use longer intervention periods, and improved evaluation of treatment quality. Comparison with a non-intervention group is necessary to re-evaluate the benefits of an IT approach.

Introduction

Cerebral palsy (CP) describes a group of disorders of movement and posture, causing activity limitations, attributed to non-progressive disturbances occurring in the developing fetal or infant brain.[1,2] Thereby, 45% of the children with CP show bilateral involvement with increased muscle tone or spasticity.[1] Ninety percent of the children with bilateral spastic CP show abnormal MRI findings, with periventricular leucomalacia or white matter disease associated with prematurity observed most frequently.[3] Although the brain lesion in CP is known to be non-progressive, the consequences of the injury are not. These often lead to secondary problems such as muscle contractures, bone deformities and thereby, loss of gross motor function and mobility.[2] CP is seen as a chronic disability, with physical therapy (PT) as a major life-long intervention. Over the past 10 years, research into PT interventions for children with CP has grown exponentially. However, literature describes a large variety of frequencies and intensities of PT.[4,5] Also regarding the applied techniques and therapy approaches, different outcome studies report diverse and contradicting results. [6-8]

A first and major methodological problem in the evaluation of the outcome of PT programs is the heterogeneous nature of CP.[2] Children with CP usually present with a variety of problems, distributed among all levels of the International Classification of Functioning (ICF).[9] Interventions at any of the elements of the ICF may be important and appropriate.[7,9] Many interventions studies so far have evaluated the effectiveness of individual techniques and have shown promising results.[7,8,10] However, the use of isolated techniques might not be satisfactory to reach clinically significant treatment effects for this group of children. Also the reality of clinical practice usually requires a mixture of individual techniques, each targeting a specific problem. In a previous study, we therefore developed and validated a clinical framework to identify the main problems and specific goals for ambulant children with CP.[11] We thereby hypothesized that individually tailored PT treatments more specifically meet the needs of the individual child and therefore also provide better treatment results. This hypothesis was tested in a smaller pilot study that indicated non-significant but advantageous results in favor of an individually defined, targeted approach.[12] To the best of our knowledge, no other studies have evaluated the effects of an individually defined PT approach for children with CP.

A second methodological hindrance to outcome evaluation is the variety of factors influencing interventions. Many other confounding factors hamper even the most optimally designed interventions: the difficulty to control the frequency of therapy[5], treatment quality and fidelity[13] and the fact children from different age groups[14] and children with different functional mobility levels[15] might require different therapy approaches. Additionally, children with spasticity are in regular need of Botulinum-Toxin A (BTX-A). Selective tone reduction by means of BTX-A treatments in children with CP is usually recommended because of a lack of progression in motor development, development of muscle contractures, intolerance of splinting and / or decrease in functionality. There is now evidence that the consequences of persistent

increased muscle tone can be limited when conservative therapies such as PT and orthotics are complemented by a selective treatment for spasticity by BTX-A.[16-18] This selective tone reduction thereby provides a window of opportunity for therapeutic interventions as the decreased muscle tone allows increased range of motion, the potential to strengthen the antagonist muscle and the opportunity to develop better motor control and balance. [19,20]

Several studies reported these variables as confounding factors or attempted to control for these variables by optimizing the quality of their study designs. However, to the best of our knowledge, no intervention studies have controlled for the combination of these variables either design-wise or statistically.

The aim of this paper is twofold. The first aim is to compare the effectiveness of an individually defined therapy approach to the results of a general therapy program. We thereby hypothesize that an individually defined treatment aim provides better treatment results than a general treatment approach.

As a second aim, we evaluate the extent to which time-effects, Botulinum Toxin A (BTX-A), age, gross motor function classification (GMFCS), frequency of therapy and quality of the programs influence treatment outcome. It is hypothesized that these factors can be identified as predictors in treatment outcome.

Methodology

Participants

Participants were selected through the Cerebral Palsy Reference Centre of the University Hospital Pellenberg, between January 2010 and September 2013**.** Inclusion criteria were diagnosis of bilateral spastic CP, age between 4 and 9 years and sufficient cooperation to understand and execute simple verbal instructions and Gross Motor Function Classification System (GMFCS) level I, II or III. The GMFCS classifies children into five levels based on functional mobility.[21] Children functioning at GMFCS I to III are ambulant, with GMFCS level I referring to children able to walk indoors without restrictions, children functioning at level II have difficulties walking outdoors and children with GMFCS III use assisting devices for functional mobility. Children were excluded when they showed severe muscle contractures or bony deformities, a history of multilevel orthopedic surgery or additional disorders which would influence the execution of the PT programs (e.g. severe cognitive disorders, deafness, blindness etc.). The parents or caretakers as well as the physical therapist of all children that were eligible for inclusion, were contacted. When the parent or caretaker as well as the physical therapist agreed to participate, both signed an informed consent form. Fifty-seven children and their physical therapists were invited to participate. Although all physical therapists agreed to participate, seventeen parents declined because of personal and practical reasons. (**Fig.1**) The study was approved by the ethical committee of the University Hospital Leuven.

Study Design

To fulfill the **first aim,** the study was designed as a randomized, controlled and evaluator-blinded trial. A flow-cart of the study design is provided in **Fig.1**. Randomization was performed by an independent person who was not involved in the selection procedure and did not have access to clinical information about the children. Children were randomly assigned into a 10 weeks individually defined (IT) or a general (GT) PT program. However, as timing of BTX-A was decided by the multidisciplinary team and not influenced by the study, some children were in need for BTX-A at the time of inclusion in the study and others were in a BTX-A free period. Therefore, to obtain comparable groups, stratification was performed according to the BTX situation. This resulted in two strata: BTX-A immediately prior to the intervention or no BTX-A prior to the intervention. Additionally, within each BTX-stratum, a minimization procedure was applied for GMFCS and age according to the procedure described by Altman & Bland.[22]

Subsequently, after a wash-out period of minimally 10 weeks, children, parents and physical therapists willing and able to continue, were enrolled into a second and/or third program. The children were newly randomized into an IT or GT program, according to the same procedure. Of the 40 children eligible for inclusion, 19 children were enrolled into a second program and 1 child continued a second and a third program. However, within the different BTX-A strata, participation to the same type of program was avoided. For example, if a child started with an IT program in a post-BTX-A situation, the child would receive a GT program as a second post-BTX program. If the second program for this child was planned in BTX-free situation, the child was newly randomized into a GT or IT program. Although participation to multiple programs was possible, the wash-out period allowed us to consider each participation to a program as a new observation. From that perspective, all analyses were performed on the total number of 60 interventions. However, the presence of correlation induced by multiple participation was taken into account in the statistical approach (see section 2.7 statistical analysis).

To fulfill the **second aim** of the study, patient characteristics as well as therapy frequency and quality were registered accurately (see section treatment quality).

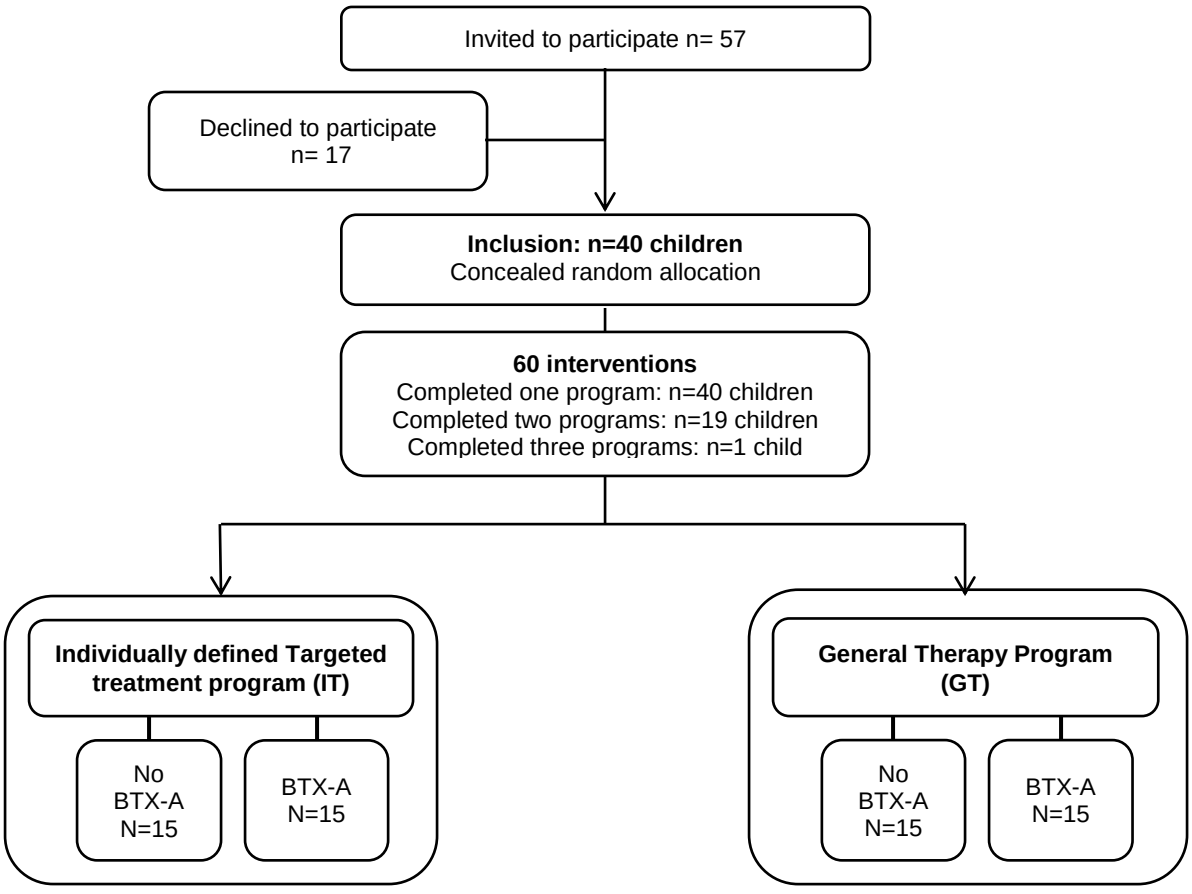


Fig 1. Study flowchart

Abbreviations:N: number of programs; n: number of children; BTX-A: botulinum toxin A

Interventions

The **individually defined targeted programs (IT)** were designed to target the specific main problems of the child. These were, tailored to the needs of the child and specific problems at the different levels of the ICF at that particular moment. Definition of the individual problems was based on the results of all assessments. At the level of body structure and function, results of a clinical examination were considered. At activity level, three-dimensional gait analysis (3DGA) and gross motor function evaluation was used, complemented with a structured interview with the parents and child. The results of the 3DGA thereby played a major role in unraveling the motor problems of the child and the identification of the main problems. The programs were based on the clinical decision framework as described by Franki et al (2014), which use the ICF and the Hypothesis-Oriented Algorithm for Clinicians (HOAC-II) to identify a selection of main problems. [11,23] This allowed goal setting at all levels of the ICF, according to the needs of the child. The goals were discussed with the child's parents or caretakers as well as the child's physical therapist. The designs of the programs thereby approached the reality of clinical practice, using a mixture of individual techniques, each targeting a specific problem of the child at that particular moment. The choice of the appropriate techniques to achieve a specific goal was performed according to the evidence-table designed by Franki et al in previous systematic reviews. [7,8]

The **general treatment program (GT)** was not based on the individual assessment results of the child. Instead, it targeted the most common aims in children with bilateral spastic CP of the involved age-range and was the same for all children. The program consisted of a predefined set of exercises to improve strength, selectivity and mobility and included a set of functional exercises.

Both the IT and GT programs were executed by the child's personal physical therapist, who agreed to precisely follow the prescribed program. A PT degree was required but no further specialization was mandatory. As only the content of the programs were influenced by the researchers, the frequency and duration of the therapy remained unchanged during the study and followed the usual intensity as recommended by the multidisciplinary team. Following the basic principles for evidence-based PT, stretching was performed in 3 repetitions of 30 seconds, strength was trained in series of 3 times 12 to 15 repetitions and functional exercises were not restricted to a specific repetition number.[7]

Table 1 of chapter 5 provides an overview of the similarities and differences between the IT and GT programs. The general program is provided in **Appendix Y**, an example of an individually defined provided in **Appendix Z**.

Compliance, Follow-up and Evaluation of Treatment Quality

The participating therapists were contacted prior to the start of the study. During this conversation, the physical therapist received an extensive explanation of the study and the importance of the programs. The physical therapist had the opportunity to refuse or agree upon participation. However, all physical therapists agreed and were very motivated to participate. For the individual programs, the first author of the study discussed the main problems of the child with the physical therapist before the start of the study. Based on this commonly defined main problems, the first and third author of the study (IF and JD) proposed a first draft of the program. This draft was again discussed with the therapists and if necessary, changes were made. Actively involving the therapists in designing the program, increased awareness and thereby motivation of the participating therapists. When the child was enrolled into the general program, the program was proposed to the therapist but when necessary, minor changes were allowed. During the second week of the program, the therapist was contacted by phone or e-mail to discuss the progress of the program and again, the possible needs to change and adjust exercises. Minor adjustments to specific exercises were allowed, major changes in treatment goals were avoided.

A follow-up visit was organized around five weeks after the start of the program. During that visit, a discussion was held with the therapist to evaluate the progress of the program. Additionally, in teh presence of teh researcher, the therapist performed a regular therapy session using the specified program. The quality of execution of this therapy session was scored using a custum-made quality assessment form (**appendix AA**). To the best of our knowledge, no specific and validated evaluation forms are available to score the quality of exercises. Therefore,, the authors based their quality assessment form on the main parameters used in physical therapy education.[24-26] The form evaluated the position of the child, number of repetitions, intensity, feedback and attention for compensation (50% of the score). The other 50% of the quality score was used to evaluate the distribution of the session time in more analytical (impairment level) and more functional exercises (activity and participation level), resulting in a possible maximum quality score of 100%.

Assessment

Participants were evaluated before and after each treatment program. To evaluate the child at the level of body structure and function, clinical examination was used. At activity level, three-dimensional gait analysis (3DGA), gross motor function measure (GMFM-88), the timed-up-and-go (TUG) and five-times-sit-to-stand-test (FTSTT) were used.

Clinical examination included measurements of range of motion (goniometrical measurements)[27], spasticity (Modified Ashworth Scale and Tardieu) [28-30], selectivity (selective motor scale) [31] and muscle strength (manual muscle testing).[24,27]

For **3DGA,** information on kinematics, kinetics and spatial-temporal data was collected by a 15-camera VICON system (Nexus PluginGait marker set, Oxford Metrics, Oxford, UK), two ATMI force plates (Advanced Medical Technology, Inc, Watertown, Massachussets) and a 16 channel electromyography device (Zero-Wire, Cometa, Milano, Italy). Three representative walking trials were selected. Specific gait parameters were automatically extracted using custom-made software designed in Matlab (Mathworks®, Natick, MA, USA). Clinical examination and 3DGA were performed according to the standardized protocol used at the Laboratory of Clinical Motion Analysis of the hospital. An independent assessor, blinded to the study, performed both assessments.

The **GMFM-88** is a standardized clinical instrument to evaluate change in gross motor function in children with CP. It fulfills the criteria of reliability and validity with respect to responsiveness to change. [32,33] GMFM-88 measurements were collected by the 1st author of this study and video-taped. An independent assessor, blinded to the set-up of the study, scored the video-registrations.

The **timed up and go (TUG) test and the five-times-sit-to-stand test (FTSTS)** are measures to assess functional mobility. The TUG test is a test used for assessing functional ambulatory mobility and dynamic balance and has a high intra-rater reliability.[34] The FTSST is a reliable assessment tool that is representative for functional performance.[35]

Outcome Parameters

Outcome parameters were derived at two different levels **(Fig 2).**

Primary outcome parameters were based on achievement of the individual goals defined using the results of the GMFM-88 and 3DGA.

For the **individual, functional goals extracted from the GMFM-88**, the original version of the Goal Attainment Scale (GAS) was applied, as developed by Kirusek et al.[36] and used in several pediatric rehabilitation studies.[37,38] Before the start of the programs, individual goals based on specific goal items of the GMFM-88, were formulated according to the SMART-principle meaning that goals are Specific, Measurable, Assignable, Realistic & Time-related.[39] A score between -2 and 2 was given depending on the goal achievement. Subsequently, a converted T-score was calculated. Converted GAS scores under 50 are usually considered non-successful treatments. Scores of 50.or more represent successful treatments.

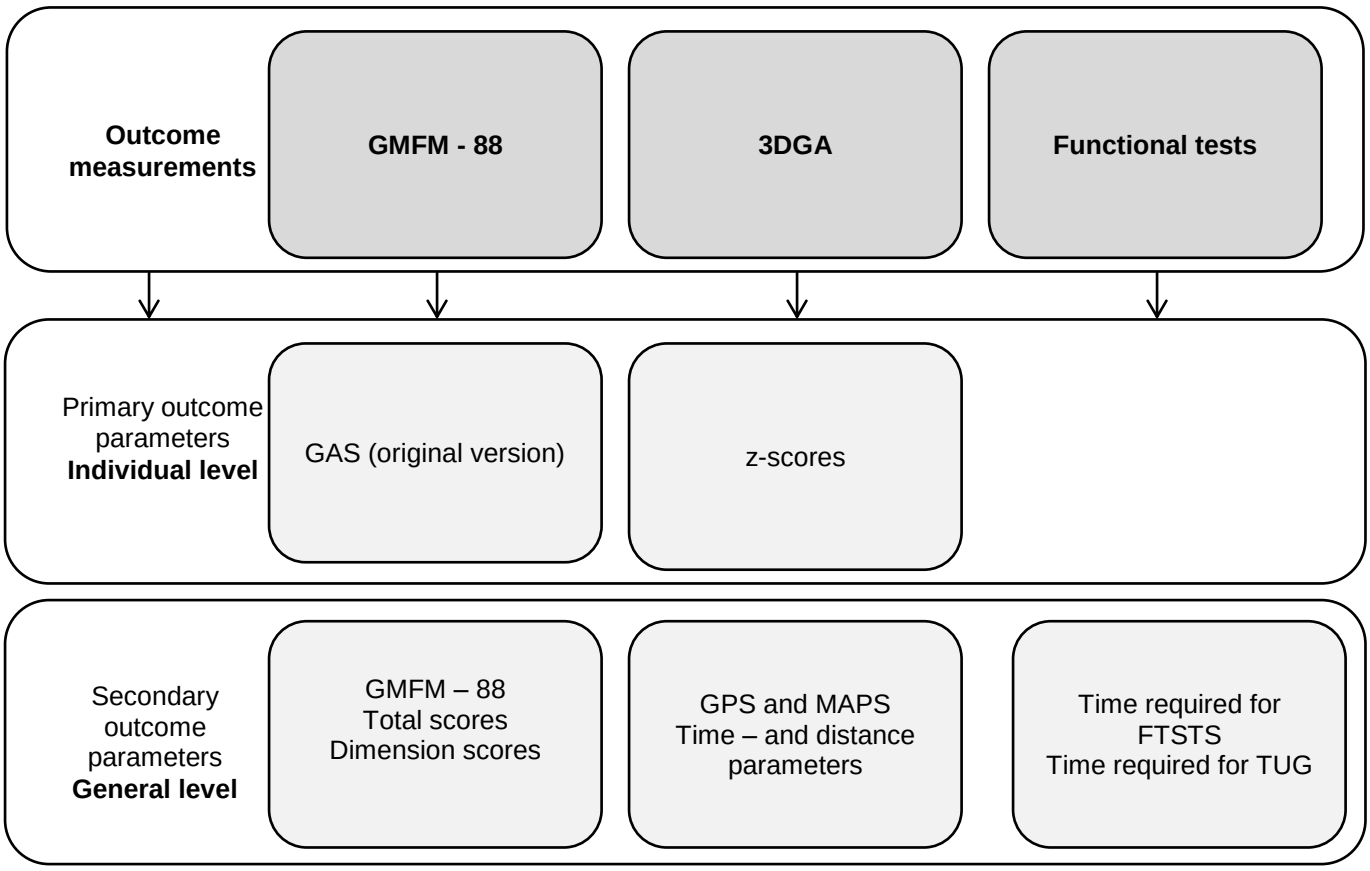


Fig 2. Outcome measurements and parameters

Abbreviations: GMFM-88: Gross Motor Function Measurement; **3DGA:** Three-Dimensional Gait Analysis; **GAS:** Goal Attainment Scale; **GPS:** Gait Profile Score; **MAPS:** Movement Assessment Profiles; **FTSTS:** Five-Times-Sit-To-Stand; **TUG:** Timed-Up-and Go

Individual goal-achievement of the specific goals derived from 3DGA, was evaluated using z-scores. As a first step, 2 to 8 specific deviated gait parameters were identified as specific goals for each child. Each goal was represented by a relevant discrete value that was expected to improve, such as maxima, minima, range of motion and angles at specific events in the gait cycle. These discrete values were defined based on literature and by clinical experts in 3DGA of children with CP.[40] The values were converted into z-scores. This z-score represented standardized deviation from a reference group and was calculated by the following formula: (mean child-mean typical children)/ SD typical children. Our reference data was based on the data obtained from a control group of 55 typically developing children, with a mean age of 10.94 years, range between the ages of 4 years and 18 years. Z-scores are non-dimensional and therefore allow the comparison of different parameters. To evaluate the z-scores, the absolute difference to zero, in either direction, was considered as an equal deviation from typical gait. Consequently, individual changes were calculated as the difference between the pre and post value of the z-scores. The median value of the z- change scores represented the individual progress on gait analysis goals.

Secondary outcome parameters were defined at a general level, using outcome parameters that provide a general outcome description and thereby represent a more overall therapy success.

For the **GMFM-88,** the total and dimensional scores were extracted and expressed as percentages. For the **functional tests**, the average of three trials was calculated.

From the 3DGA data, **Movement Analysis Profiles (MAPS), the Gait Profile Score (GPS) and time-and distance parameters** were calculated.[40] The MAPS were calculated as the root mean square error (RMSE) between the point-by-point comparison of the lower limb joint angle and the averaged joint angle of the reference group. Calculation of the average of all lower limb joint angles results in the GPS, which summarizes the overall severity of gait pathology.[40] The average scores from three left and three right trials were used. **Time-and distance parameters** were corrected for normal age development, by normalizing the data for step length.[41] Subsequently, the normalized time-and distance parameters were also transferred into z-scores. To allow comparison of individual progress, pre-post values were calculated using the absolute values.

Statistical Analysis

As a **preparatory step**, observations were summarized using means and SDs as appropriate. Baseline characteristics were compared using Pearson Chi-square tests for the use of orthotics and independent t-tests for age, therapy hours per week and treatment quality.

To evaluate the **first goal of the study**, the changes in outcome after both programs were compared, using a linear model with 'program' (IT or GT) as factor of interest and 'BTX', 'GMFCS' and 'age' as additional predictors. To take into account the correlation between the changes from the same child, a random child effect was added yielding a so-called linear mixed model. The Estimated Marginal Means (EMMeans) of the change scores were calculated, providing the mean response for that factor, adjusted for the variables in the model. Additionally, effect sizes (ES) were calculated using the Cohen's *d* formula. According to Cohen, effect sizes of 0,2 to 0,5 are considered as small, 0,5 to 0,8 are medium and >0,8 are large.[42]

To evaluate the second goal of the study, a specific set of analysis aimed to identify which children benefited most from the interventions. First, interaction effects between the 'type of program' and BTX, GMFCS and age were evaluated to verify if the effect of the specific program depends on these characteristics. Second, this analysis was also performed regardless of the type of program they were enrolled. Therefore, a linear mixed model analysis with random effect child was used for each potential predictor separately (BTX, GMFCS, age, time or within-group effects, therapy hours per week and treatment quality). For all analysis, the changes calculated as pre-post value were used. P-values smaller than 0,05 were considered significant. A Bonferroni Holms procedure was applied to correct for multiple testing for the two primary outcome parameters. All statistics were performed in SPSS 22 IBM SPSS Statistics for Windows, Version 22.0. (Armonk, NY: IBM Corp).

Results

Patient and therapist characteristics

Forty children participated in the study, resulting in a total of 60 interventions. Thirty interventions were performed in a post-BTX-A situation, 30 interventions in a BTX-A-free situation. (**Fig 1**)

Table 2 provides a detailed description of the patient characteristics. Both groups were comparable regarding age, GMFCS and prior BTX-A injection. The total therapy time per week was higher in the GT group compared to the IT group, with 184,2 (SD 82,3) and 158,8 (SD 79,03) minutes per week respectively. This difference was not statistically significant. Also regarding treatment quality, no relevant differences were observed between both programs: 61,19 % (SD 13,86) for the GT and 64,91 % (SD 16,78) for the IT group.

Thirty-two different therapists participated in the study. On average, the therapists had 13,8 years of experience (SD 9,5). Twenty-seven therapists had a PT degree at university level, 17 were specialized in pediatric rehabilitation and 21 were certified Bobath therapists.

Table 2. Patient Characteristics

	GT	IT
Gender (n)		
Male	19	18
Female	11	12
Age (mean + SD, in months)	77,16 (15,23)	68,35 (16,72)
GMFCS (n)		
Level I	9	10
Level II	13	11
Level III	8	9
Use of day orthosis (n)		
>50%/day	24	25
<50%/day	3	3
no	3	2
Use of night orthosis (n)		
>50%/night	11	12
<50%/night	12	12
no	7	6
Total PT (minutes/week)	184,2 (82,3)	158,8 (79,03)
Sessions/week	3,9 (1,5)	3,7 (1,5)
Minutes/session	48,5 (13,3)	43,3 (14,6)
Program in a post-BTX situation	30	30
Prior BTX (before study)		
No prior BTX	8	9
1 time	2	7
2 times	5	6
3 times	3	4
more than 3 times	12	8

Abbreviations

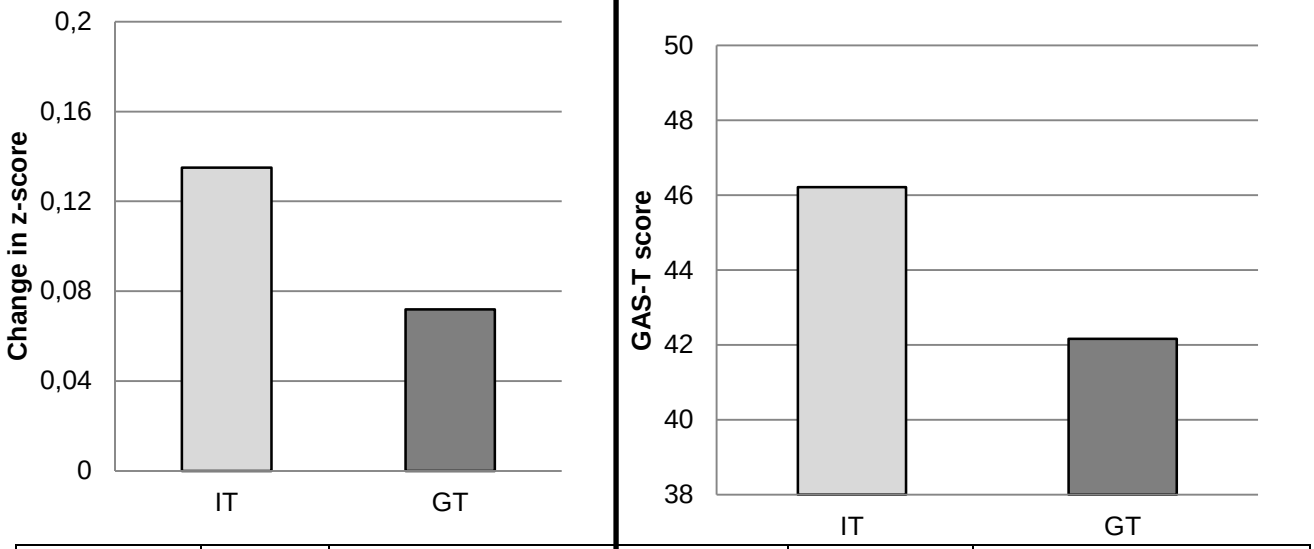
UC: period of Usual Care; **GT:** period of general treatment; **IT:** period of Individualized, targeted treatment

GMFCS: Gross Motor Function Classification System; **PT:** Physical Therapy; **N:** number

First analysis: General versus individually defined therapy

Primary outcome parameters – individual goal achievement

Fig. 3 demonstrates the EMMeans after correction for BTX-A, GMFCS and age for the changes in z-scores and the GAS. This figures demonstrates that both the change in z-scores and the GAS-T score were non-significantly higher after the IT compared to the GT program (p=0,669; ES=0,05 and p=0,322; ES=0,15).



EMMean	Std. Error	95% Confidence Interval		EMMean	Std. Error	95% Confidence Interval	
		Lower Bound	Upper Bound			Lower Bound	Upper Bound
0,135	0,103	-0,071	0,341	46,208	2,91	40,354	52,063
0,072	0,105	-0,138	0,282	42,166	2,799	36,542	47,79

Fig 3. Estimated marginal means (after correction for GMFCS, BTX-A and age) for the changes in z-scores (change score = pre-post value) and the Goal Attainment Scale (GAS-T score) for the Individually Defined Therapy Program (IT) and the General Therapy program (GT).

Secondary outcome parameters – general parameters

Fig.4 provides the distribution of the observed GMFM-88 total scores before and after the programs for the different treatment groups. No significantly different changes in outcome could be registered between both groups. The estimated mean change score was -1,65% (95% CI -2,37 to -0,917%) for the IT group and -1,27% (95%CI -1,98 to -0,565) for GT group (p=0,463; ES =0,29).

Regarding the **GMFM dimensions**, no significant differences between both intervention groups could be registered. For dimension D, the EMMean change (pre-post) was -5,28% (95% CI -7,81 to 2,75) for the IT group and only -1,8 % (95% CI -4,27 to 0,674) for the GT group, (p=0,056, ES=0,64). For dimension E, the EMMean change showed no change after the IT program, but a slight improvement after the GT program: 0,83% (95%CI -2,70 to 1,04) versus -2,74 (95% CI -4,52 to -0,96) (p=0,086; ES=-0,25).

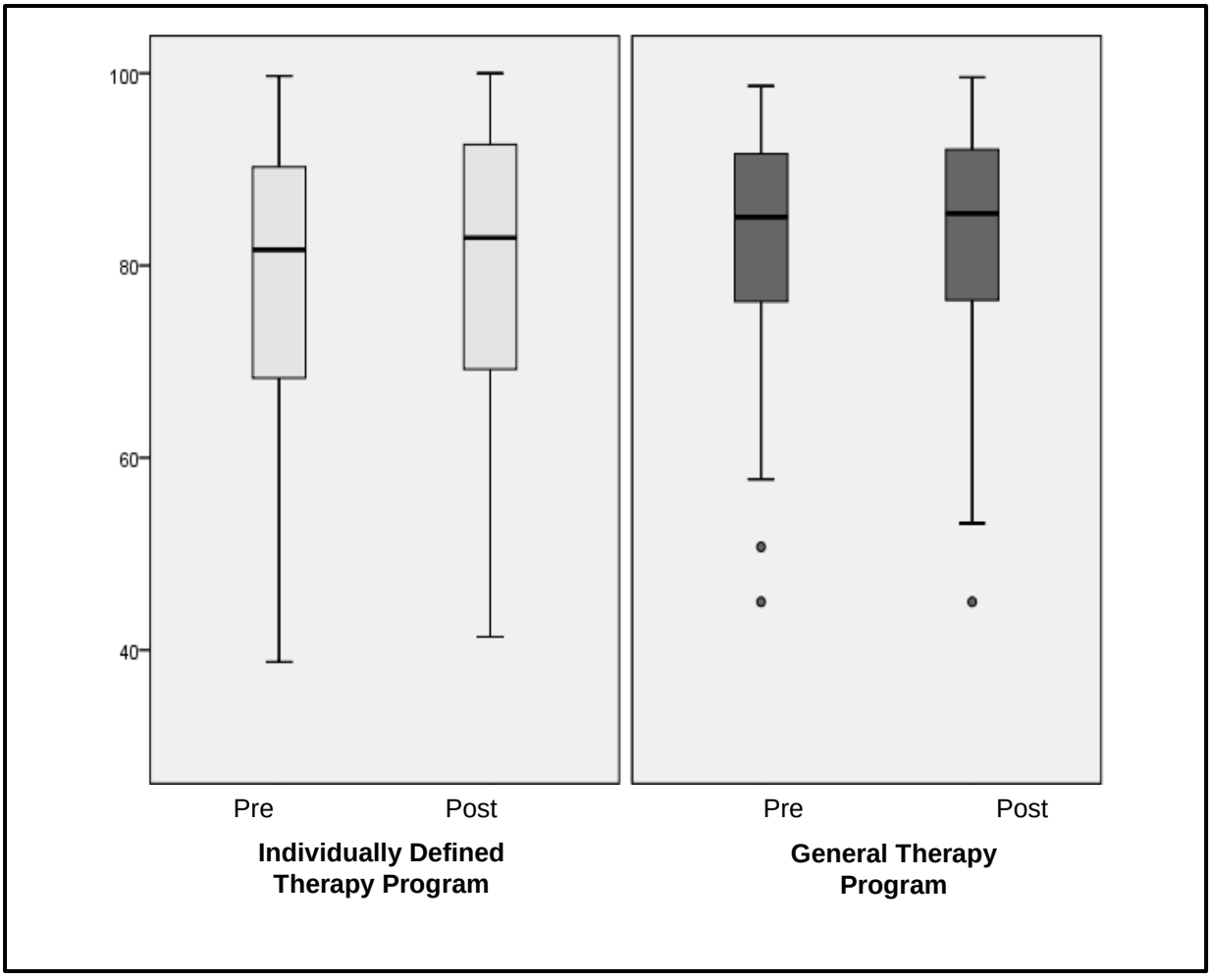


Fig 4. Scores of the Gross Motor Function Measurement (GMFM-88) before and after the programs.

The EMMean change score for the **GPS** showed limited changes for both groups: -0,66° (95%CI -3,2 to 1,86) in the IT group and 0,28° (95%CI -2,29 to 2,84) in the GT group (p=0,607; ES=-0,13). Fig.5 provides an overview of the changes in **MAPS** in the different planes for both treatment groups. Although generally more deterioration was found for the GT group, especially in the sagittal plane, no significant differences were found in response to the programs. (**Fig 5**).

The means and standard deviations of the **normalized time-and distance parameters** are reported in **table 3**. This shows improved cadence in the IT group but slight deterioration in the GT group (p=0,053, ES=0,57). The increased single support phase in the IT group shows improved stability in the IT group but not in the GT group (p=0,034; ES=0,6).

No significant different effects were obtained for the time needed for the **FTSTS and the TUG test**: p=0,478; ES=-0,10 and p=0,887; ES=0,23 respectively.

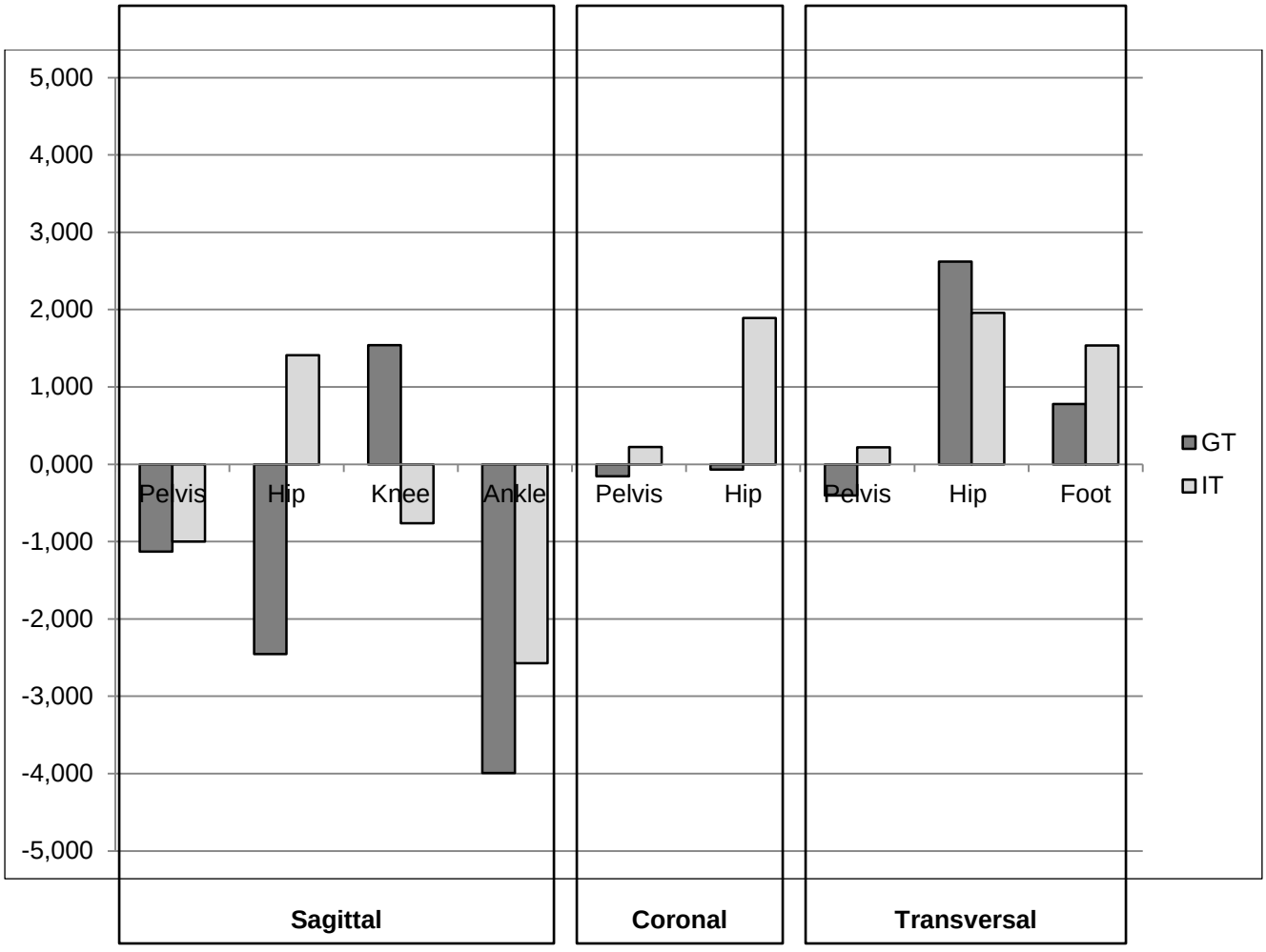


Fig 5. Changes in the movement assessment profile scores (MAPS) for the Individually Defined (IT) and the General (GT) therapy group. Change scores are calculated as the pre-post values (positive value = improvement, negative value=worsening)

Table 3. Means and standard deviations for the normalized time-and distance parameters

	IT			GT					
	Pre	**Post**	**Diff**	**Pre**	**Post**	**Diff**	**F**	**p value**	**ES**
Cadence	-4,04 (5,04)	-3,99 (4,20)	0,67 (2,14)	3,20 (3,93)	-3,55 (3,92)	0,64 (2,46)	3,914	0,053	0,57
Step Length	-0,15 (0,23)	-0,76 (0,07)	-0,56 (0,18)	-0,14 (0,19)	-0,75 (0,07)	-0,52 (0,25)	0,101	0,755	-0,15
Step Time	1,26 (1,81)	1,10 (1,51)	0,21 (0,72)	0,87 (1,57)	0,99 (1,44)	-0,13 (1,16)	1,357	0,249	0,36
Walking velocity	-0,65 (0,76)	-0,61 (0,65)	0,07 (0,31)	-0,48 (0,75)	-0,50 (0,75)	-0,02 (0,36)	0,451	0,507	0,26
Stride Length	-0,16 (0,24)	-0,13 (0,22)	0,02 (0,11)	-0,15 (0,20)	-0,14 (0,18)	0,01 (0,14)	0,016	0,9	0,09
Stride Time	1,35 (1,96)	1,17 (1,59)	0,27 (0,72)	0,98 (1,70)	0,98 (1,49)	-0,02 (1,29)	0,788	0,379	0,28
Single Support	0,62 (1,02)	0,48 (0,66)	0,19 (0,53)	0,37 (0,54)	0,42 (0,61)	-0,10 (0,42)	4,729	0,034	0,6
Double Support	1,75 (2,82)	1,51 (2,29)	0,32 (1,33)	1,31 (2,96)	1,26 (2,08)	0,21 (2,49)	0,002	0,963	0,06
Foot Off	2,60 (1,63)	2,44 (1,44)	0,20 (0,83)	2,17 (1,40)	2,15 (1,12)	0,08 (0,70)	0,007	0,931	0,15

Abbreviations:

IT: Individually defined targeted treatment group; **GT:** General Therapy group; **Diff:** difference as defined by pre-post value; **ES:** effect size

Second Analysis: Prognostic Factors

Interaction effects

For neither for the primary, nor for the secondary outcome parameters, significant **interaction** were registered between the type of program and BTX, age or GMFCS.

Time or within-group effects

For individual goal achievement, significant time-effects could be found on GAS-T score (p<0,001) but not on z-scores (p=0,142). Almost all children showed improved GMFM-88 scores, with an EMMean change of -1,46% (95% CI -1,96 to 0,96; p=0,000). For dimension D and E, mean changes of -3,42 % (95% CI -5,25 to -1,60; p=0,000) and -2,07% (95% CI -3,30 to -0,85; p=0,006) were registered. With a mean change of -0,36°(95% CI -2,10 to 1,38), the GPS showed no time-effects (p=0,683). Furthermore, significant time-effects were found for stride length (p<0,001).

BTX-A

The GAS was lower for children in the BTX-A situation: 40,5 (95%CI 34,63 to 46,44) versus 47,8 (95%CI 41,7 to 53,92) for the children not receiving BTX-A. This difference however, was not significant (p=0,087). In contrast, for the individual goals based on the 3DGA, children who received BTX showed a mean positive change of 0,215 (95% CI 0,017 to 0,413) on z-scores. Children who did not receive BTX-A, showed a limited change of -0,006 (95% CI -0,208-0,195). Again, this difference was not significant (p=0,122). For none of the other outcome parameters however, the influence of BTX-A however, was observed.

Age

Significant and relevant relations between age and changes were found on the attainment of functional goals (p= 0,023), the GMFM D (p=0,047) and walking velocity (p=0,044), timing of foot off (p=0,030) and the STS test (p=0,041). Older children showed a higher achievement of functional goals and change in GMFM-D score, but younger children showed a larger change in walking velocity, timing of foot-off and time needed for FTSTS.

GMFCS

No significant effects could be found for GMFCS in relation to gait – or gross motor changes.

Remarkably, functional goal achievement seemed to decrease with a decreased level of functionality. GAS was 48,8 (95% CI 41,2 to 56,37) for the children with GMFCS I, 43,6 (95% CI 36,5 to 50,6) for children with GMFCS II and 38,8 (95% CI 30,7 to 48,9) for children with GMFCS III. However, due to the wide variability seen in the large CI, these differences were not significant (p=0,203) and should be interpreted carefully.

An opposite trend was visible for the goals derived from 3DGA: children with GMFCS III seemed to have a non-significantly higher improvement on their z-scores (change 0,144 95% CI -0,13-0,419) than children with GMFCS II (change 0,121, 95% CI -0,116-0,358) and GMFCS I (change 0,059, 95%CI -0,203-0,321). Again, the wide variability in resulted in a non-significant p-value (p=0,893).

Treatment quality

No significant relations were found between treatment quality and changes in outcomes.

Treatment intensity per week

The number of minutes of therapy per week showed a significant influence on the MAPS of the foot in transversal plane (p=0,003).

Discussion

The first goal of this study was to evaluate differences between the effects of an individually defined therapy and a general training program on gait and gross motor function. The results of our study could not confirm the first hypothesis that an individually defined approach provides better treatment results. No significant differences could be registered between the intervention groups. Nevertheless, the GAS-T scores suggest a higher degree of individual goal achievement after the IT program compared to the GT program. This could be seen in the achievement of higher GAS scores (46,2 versus 42,2) as well as slightly greater changes in the z-scores based on 3DGA (0,135 vs. 0,072) in the advantage of the IT group. A similar trend was visible on GMFM dimension D, the Movement Assessment Profiles, cadence and the duration of single support phase.

A major reason for the lack of significant differences could be the content of the GT program. The GT program was designed to control for the individual differences of therapists during their usual care. It thereby primarily aimed to bring all therapy interventions during the control phase to the same level. As a result, the GT program was not only a standardized, but also still a highly qualitative program with a wide set of exercises targeting the major problems in children with bilateral CP from this age-range. Therefore, the difference between the IT and the GT program may not have been sufficient to reach significant differences in outcome. Additionally, even the GT program was often very different compared to the usual exercises the child received prior to the intervention study. In a subsequent research project, we compared the effectiveness of a period of usual care to the effects of a predefined intervention program. (Franki et al., 2014c). No changes to a slight deterioration in gross motor function and a similar trend on gait analysis parameters was found after the period of usual care. These results are in contrast with the results of both intervention programs, as a trend of non-significant improvements is observed. Registering the contents of usual care physical therapy (no intervention) using a diary, demonstrated a very large focus of usual physical therapy on problems at impairment level (stretching & analytical strength training). Only 41,7% of therapy time was spent on functional, activity-related exercises. Our intervention programs always contained more than 50% of exercises targeting the activity level. This might suggest that simply a change in therapy from usual care to GT or IT might be an important factor influencing the outcome.

As such, it is hopeful to see that all children responded well to the therapy programs in terms of their gross motor function. Although sometimes with small changes, almost all children showed improved GMFM-88 scores, resulting in significant time-effects for the GMFM-88. Only three children showed a decrease in GMFM-88 scores. This was also reflected in individual functional goal achievement based on the GMFM, as significant time-effects were also found for the GAS. The mean GAS for the total group was 44,26 with an average percentage of achieved goals of 48,2%. Results were less pronounced for gait analysis parameters. As for the GPS, only 50% of the children showed an improved gait pattern after the programs. On gait analysis goals, only 33% of the program responses were positive.

The lack of significant effects of the training programs may also be related to the duration of the applied interventions. Ten weeks was a relatively short training time. Guidelines by the National Strength and Conditioning Association (NSCA), stated that strength training should be applied 2 to 4 times a week, on non-consecutive days and for at least 8 to 20 weeks in order to affect muscle strength.[44] Since strength training was an essential part of both programs, a study of longer duration could have resulted in greater benefits. Also for functional changes, a period of 10 weeks might have been too short. In a recent review evaluating the effectiveness of upper limb training, Sakzweski et al. concluded that 40 hours of therapy was adequate to yield meaningful clinical changes on the upper limb and individualized outcome.[45] In another review evaluating dosing parameters, Gannotti et al. concluded that thresholds for time to produce changes in performance warrant further investigation.[4] Studies evaluating the effectiveness regarding BTX-A however, demonstrated positive effects up to one year after injection.[46]

Additionally, as some children participated in multiple programs, a learning effect might have been created for the therapists. Therapists receiving the IT program first, were more aware of and therefore probably more attentive to the individual problems of the child. Subconsciously and instinctively, they might have incorporated these aspects during the general program. Twenty-three children received the IT program as a first program and 17 children received the GT program first. However, separate analysis of the results of the 40 first interventions (excluding the repeated interventions in the same child) showed no major differences to the results of all 60 interventions. This allows us to reject our hypothesis of indirect carry-over effects.

The second analysis attempted to evaluate the influence of the factors BTX-A regime, age, GMFCS, treatment quality and therapy time per week on the outcome of the intervention. This analysis revealed a non-significantly but relevant lower GAS for the children receiving BTX-A prior to their program (GAS=40,5) compared to children not receiving BTX (GAS=47,8). The children receiving BTX-A however, were mostly children with a lower functional level. This might therefore coincide with the trend that was visible on GMFCS levels, where children with lower functional levels also seemed to achieve less functional goals. In contrast, when the focus was on goals based on gait analysis, children with lower functional levels and children receiving BTX-A seemed to reach their goals more easily.

The results revealed age as a relevant predictor for GAS-T scores and change in GMFM-88 total dimension D scores. When considering individual functional goals, older children seemed to benefit more from the period of intervention. Therefore, it is useful to involve older children more actively in the process of goal-setting, as this might cause greater therapy-effects.

Our study evaluated the effectiveness of a general program that was the same for all children. Nevertheless, supervision and guidance of a physical therapist reassured correct execution of the exercises. Especially, for the younger children, this was still crucial. As group training is known to have positive effects on participation, it might however be a recommendation for future studies to compare the

influence a predefined program using group sessions to the effects of individual training using the same program.

Yi et al evaluated contributing factors for the change of gross motor function in children with spastic CP after physical therapy.[47] Initial GMFM score and degree of spasticity were identified as major predictors in the responsiveness to therapy: children with mid-range of initial GMFM-88 scores showed more improvements on the GMFM-88 score compared to lower-and higher range of initial GMFM-88 scores. The degree of spasticity was identified as a negative predictor for functional changes. A similar trend is visible in the results of our study, as children with GMFCS III show lower GMFM-88 scores and generally show a higher degree of spasticity.

No relationship was found between the effects of the program and treatment quality. Treatment quality was evaluated using a self-developed quality assessment form. The assessment form evaluated the basic factors of position, repetition, intensity and feedback and thereby focused on the correct technical and practical execution of the exercises. Other factors like context, used materials and play are not included in the assessment form and might have influenced our results. Although the specific exercises were prescribed, the time to invest in certain exercises was not prescribed. Our assessment form did not capture adequately the amount of time that was spent for the more analytical exercises versus the time that was spent on more functional exercises, a factor that also may have influenced the outcome. The Paediatric Rehabilitation Observational measure of Fidelity (PROF) is a new generic measure evaluating treatment fidelity and quality, developed and published by Di Rezze et al. in 2012.[48] At the start of our study in 2011, unfortunately, this measure was not yet available. However, the PROF focuses on the comparison of child and context therapy and may therefore not fully capture the quality of performance of the exercises as such. A combination of our quality assessment form with the PROF may address this issue in future studies.

In a systematic review examining dosing parameters for children with CP, Gannotti et al. registered a large variability in frequency, intensity and time of training.[4] Our study also could not find a relationship between the amount of therapy per week with the progress on gait and gross motor function. This confirms the conclusion of Gannotti et al. that identified multiple gaps in knowledge and the need for conceptualizing dosing thresholds for individual children. In our study, total therapy time was not influenced by the researcher and children remained at their usual therapy frequency. Therefore, total therapy time per week was higher in the GT group compared to the IT group, with 184,2 (SD 82,3) and 158,8 (SD 79,03) minutes per week respectively. Nevertheless, testing the difference in therapy per week using an independent t-test revealed no statistical difference. In addition, therapy hours per was evaluated as a prognostic factor in the second part of the study and revealed no statistically significant influence. Therefore, the authors hypothesize that the differences in the amount of therapy per week in this study, can be excluded as a causal factor for the observed differences between both groups.

In **conclusion**, no significant differences could be registered between an individually defined and a general physical therapy treatment approach for children with bilateral spastic CP. Only slight, non-significant trends in the advantage of the IT program were visible. Age was identified as a predictor for the attainment of functional goals. Future research using longer intervention periods, improved evaluation of treatment quality and comparison with a non-intervention group is necessary to confirm trends of the current study.

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Chapter 7

Evidence-based practice and its effectiveness in physical therapy treatment of children with bilateral spastic cerebral palsy

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Abstract

Aim

This study aims to identify the contents of usual care physical therapy and to compare the effectiveness of 10 weeks usual care (UC) to the effects of a 10 weeks intervention of a predefined evidence-based physical therapy program (IP).

Method

Sixteen ambulant children with bilateral spastic CP (age 5y11m, GMFCS I-III) started with UC, in which the content of their therapy sessions was registered using a diary. Subsequently, all children enrolled into the IP, applying a predefined evidence-based program with feedback and support to the therapists. Outcome was evaluated using the Gross Motor Function Measurement (GMFM-88) and the Gait Profile Score (GPS), Movement Assessment Profiles (MAPS) and time-and distance parameters, extracted from three-dimensional gait analysis.

Results

During UC, 22,5% (IQR 16,7-32,5%) of therapy time addressed muscle length and 33,3% (IQR 30,2-33,3%) strength. 41,7% (IQR 33,3-56,2%) of therapy time was used for functional exercises and 0,1% (IQR 0-6%) addressed participation limitations. The applied resistance in strength training was not quantified or graded. In 67,5% of the sessions, therapists performed gait training.

We found a significant increase in GMFM-88 score after the IP (p=0,002), but not after UC (p=0,56). Improved sagittal MAPS were only observed after the IP.

Conclusion

UC physiotherapy in CP is predominantly dedicated to problems at impairment level and characterized by insufficient dosing. An evidence-based predefined intervention provides more advantageous effects on gross motor function compared to UC.

Introduction

The use of the International Classification of Functioning, Health and Disability (ICF) has provided an opportunity to integrate several perspectives in children with Cerebral Palsy (CP).[1] Nevertheless, the use of the ICF in rehabilitation has confronted many physical therapists with the complexity of the pathology of CP and the variety of problems at all levels of the model.[2] Additionally, as several researchers have demonstrated the relationships in between the different ICF levels [3,4], the ICF has opened different points of entry in treatment, causing several different opinions regarding physical therapy treatment for these children.[5] As up today, systematic reviews evaluating different physical therapy programs in children with CP, have demonstrated promising results regarding the effectiveness of individual techniques.[5-7] However, these studies also identify the need to evaluate the effectiveness of well-balanced therapies, using a mixture of techniques, addressing all levels of the ICF. To be able to address the individual problems of a child, the physical therapist therefore, nowadays, faces the difficult and challenging task of knowledge translation.[8]

In two recent intervention studies, the effectiveness of an individually defined, targeted physical therapy approach was compared to the effects of a general physical therapy program.[9,10] Overall, the effects of both programs were not found to be significantly different and the authors could not provide evidence for the individually defined, targeted approach. Nevertheless, the individually defined as well as the general program demonstrated effectiveness. In both studies, almost all participating children demonstrated improved Gross Motor Function Measure (GMFM-88) scores and improved gait characteristics evaluated using three-dimensional gait analysis. Although the limited power of both studies also limited the possibility to draw clear and definite conclusions, we identified therapy support based on guidelines from literature as a potential factor influencing our treatment results. This raised the question to define the influence of the evidence-based intervention program as such. It may be possible that guiding and supporting the participating therapists by providing guidelines built on evidence based treatment algorithms, might already have caused a significant impact on treatment quality and outcome.

In a large randomized controlled trial, Campbell et al. evaluated the effectiveness of an evidence alert system on evidence-practice behavior of 135 allied health professionals.[8] The results demonstrated that the group of therapists receiving a three days skills training workshop and multiple faceted workplace support significantly improved their evidence based knowledge. However, this knowledge was not translated into significant evidence-based practice behavior in treatment. Campbell et al. therefore identified the need for tailored knowledge translation strategies. Unfortunately, so far, no other studies focused on the impact of stimulation of evidence-practice behavior and workplace support on the treatment outcome.

Therefore, this study aims to evaluate evidence-based practice behavior in usual care physical therapy in Flanders. Secondly, it aims to compare the effectiveness of a predefined physical therapy program based on evidence-based guidelines to the effects of a period of usual care. It is hypothesized that supporting therapists by providing a predefined evidence-based intervention program, will result in an improved treatment outcome compared to the usual care physical therapy.

Methodology

Participants

A convenience sample of 16 children was recruited at the Cerebral Reference Centre of the University Hospital Pellenberg. Children were included when they were diagnosed with bilateral spastic CP, aged between 4 and 9 years and a GMFCS level between I and III. Children were excluded when they received Botulinum Toxin A injections within the past six months, orthopedic surgery in the past or had severe associated problems that limited participation to therapy (blindness, deafness, severe cognitive limitations or autism).

Study design

All children started the study with a registration period. During this period, the children's usual physical therapy (UC) was registered using a diary. Immediately following the UC period, children were enrolled into a predefined intervention program (IP).

Both the period of UC and IP had a duration of 10 weeks. Before and after both periods, children were evaluated using the Gross Motor Function Measurement (GMFM-88) and three-dimensional gait analysis (3DGA).

Usual Care

During the period of UC, no specific intervention was provided. Children received the frequency, intensity and contents of therapy as they usually received by their own private physical therapist. A diary was provided to the child's physical therapist. The diary contained session forms, designed for the therapist to carefully register the contents of therapy after each session. A template session form is provided in **Appendix AB**.

In the session forms, the therapist noted down how much **time of the therapy session** targeted problems at the different levels of the ICF. At the **level of body structure and function**, the time spent on muscle length and muscle strength was registered. For muscle length**,** the therapists were asked to report the targeted muscles, the number of repetitions, the duration of stretch and treatment modality (activo, activopassivo or passivo).[11] About muscle strength, therapists noted the specific muscle that was trained, the intensity (number of repetitions and resistance) and whether the exercise was performed as analytical (single-joint) or functional (multiple-joint) muscle work. Concerning **activity level**, the specific functional activity that was practiced and the averaged position in which this was performed, was registered. Finally, the therapist reported whether any problems **at participation level** were specifically addressed.

During the period of UC, the therapist was not contacted and no instructions or advice was provided regarding physical therapy treatment of the child.

Intervention period

Immediately following the UC period, children were enrolled into a predefined intervention program (IP), designed by the first and third author of the study (IF and JD). The programs were executed by the child's personal physical therapist, who agreed to precisely follow the prescribed program. Similar as for the UC program, the children received the frequency of therapy as they usually received by their own private physical therapist.

Two different types of programs were designed. Ten children received an individually defined, targeted program tailored to the individual needs of the child and six children received a general program based on general age-appropriate treatment aims for children with bilateral spastic CP.[12] For an extensive description of the differences and similarities between both types of intervention programs, the authors refer to our previous intervention studies.[9,10] All intervention programs had two major common features.

The first central and common aspect in the intervention programs was that basic principles regarding evidence-based training were respected. All programs consisted of a predefined set of exercises to improve strength, selectivity and mobility and a set of functional exercises. Thereby, stretching was prescribed as three repetitions of 30 seconds and strength training was prescribed in series of three times 12 to 15 repetitions. Functional exercises were not restricted to a specific repetition number. As appropriate, all intervention programs contained more than 50% of exercises at activity and participation level.[13-15]

The second major common feature in the intervention programs was the support and follow-up to the therapist. At the start of the intervention study, therapists were visited and the prescribed program was thoroughly explained. All exercises were provided in a clear and structured way, with specific instructions regarding anatomical position, sets and repetitions. The specific exercises were discussed and demonstrated if necessary. During the second or third week, the therapist was contacted by phone or email to discuss the progress of the program and the possible needs to change and adjust exercises. Additionally, a follow-up visit was organized around halfway through the program. During this visit, the program was again discussed and the therapist performed a regular therapy session using the program, in attendance of the first author of the study. Difficulties in performance were discussed and if necessary, some exercises were demonstrated. The opportunity for feedback on handling or position was thereby provided.

Due to these common aspects in our programs, and because previous data analysis revealed that the overall effects of both programs were not found to be significantly different, we could test our hypothesis regarding the influence of evidence-based support on the pooled data-set.

An overview of the differences and similarities between the intervention programs and the period of usual care is provided in **Table 1**.

Table 1. Differences and similarities between the period of usual care and the interventions

	Intervention programs	Usual care

Differences	• The program was predefined and designed by the first and third author of the study;
	• The predefined program was thourougly explained and discussed with the therapist at the start of the 10 weeks;
	• One follow-up phone-call and one follow-up visit was made during the program;
	• The therapist had the opportunity to ask questions and to discuss problems regarding the child with the first and third author of the study;
	• If necessary, exercises were explained and demonstrated;
	• Based on the most common problems or specific goals for ambulant children with bilateral spastic CP;
	• A predefined set of exercises to improve strength, selectivity, range of motion, gait and gross motor function and if necessary addressing participation limitations;
	• The exercises targeted the different levels of the ICF;
	• Minimally 50% of the exercises were functional activities;
	• Basic principles for evidence-based PT regarding intensity and repetitions were provided to the therapist
	  o Stretching: 3 repetitions of 30 seconds;
	  o Strength was trained in series of 3 times 12 to 15 repetitions;
	  o Functional exercises were not restricted in repetition number.
		• The physical therapist was free in decision making about the goals and specific exercises for the child;
		• No intervention, a continuation of therapy as usual;
		• No follow-up phone-calls or therapy visits;
		• No guidance or advice regarding the goals and specific exercises in therapy;
		• Therapist were not given the opportunity to ask questions or to discuss problems;
		• No specific evidence-based guidelines were provided;
		• The contents of the therapy and the use of therapy time registered using a diary;
		• The therapist decided how much therapy time and how many exercises targeted the different levels of the ICF.

Similarities	• Executed by the child's private physical therapist, familiar to the child; • At the child's usual frequency and duration of therapy. • A duration of 10 weeks.	• Executed by the child's private physical therapist, familiar to the child; • At the child's usual frequency and duration of therapy. • A duration of 10 weeks.

Evaluations

Before and after the period of usual care and the interventions, all children were evaluated using the gross motor function measure (GMFM-88) and three-dimensional gait-analysis (3DGA).

The **GMFM-88** is a standardized clinical instrument to evaluate change in gross motor function in children with CP.[16] It fulfills the criteria of reliability and validity with respect to responsiveness to change.[17,18] GMFM-88 measurements were collected by the 1st author of this study and video-taped. Two independent assessors, blinded to the set-up of the study and without access to information about the children, scored the video-registrations. The independent assessors were pediatric physical therapy students (MSc). The average of both scores was used. The total and dimensional scores were extracted and expressed as percentages.

For **3DGA**, information on kinematics, kinetics and spatial-temporal data was collected by a 15-camera VICON system (Nexus PluginGait marker set, Oxford Metrics, Oxford, UK), two ATMI force plates (Advanced Medical Technology, Inc, Watertown, Massachussets) and a 16 channel EMG device (Zero-Wire, Cometa, Milano, Italy). Three representative trials were selected. Specific gait parameters were automatically extracted using a custom-made Matlab graphical user interface. (Mathworks®, Natick, MA, (USA). Specific parameters extracted from the 3DGA data were the Movement Analysis Profiles (MAPS), the Gait Profile Score (GPS) and time-and distance parameters. The MAPS were calculated as the root mean square error (RMSE) between the point-by-point comparison of the lower limb joint angle and the averaged joint angle of the reference group. Calculation of the average of all lower limb joint angles results in the GPS, which summarizes the overall severity of gait pathology.[5] The average scores from three left and three right trials were used as outcome parameters. Time-and distance parameters were transformed into non-dimensional parameters as reported by At Hof (1996).[5]

Statistical Analysis

As a preparatory test, the frequency of therapy as well as the use of orthosis during the period of UC and IP were compared using the Wilcoxon Signed Ranks test and the Mc Nemar test respectively.

To evaluate the first goal of the study, data from the diaries were extracted and summarized using Medians (Me), interquartile ranges (IQR) and frequencies as appropriate.

The median duration and frequency of therapy as well as the total therapy time per week was calculated. The registered time spent on impairment level (muscle length and muscle strength), activity and participation was analyzed and expressed as percentage of total therapy time.

For stretching, the median parameters of intensity (number of sets and repetitions and the duration of stretch) were calculated for each target muscle. The applied resistance was registered and grouped as no resistance or manual/external resistance. In addition, the frequency of functional strength training was compared to the total frequency of muscle training.

Regarding the specific exercises at activity level, frequencies were registered for the position (lying, sitting, kneeling/high-kneeling or standing) and type of activity (walking, jumping, stairs or more complex activities as climbing, hopping or jumping). Subsequently, the frequency of the applied position or activity was expressed as a percentage or relative frequency by dividing it by the total number of therapy sessions. The median percentage for all children was calculated.

For participation training, frequencies were calculated for sport, home-based or school-based specific activities. The relative frequency was calculated similarly to the activities.

To evaluate the second goal of the study, the median change scores (pre-post) of the GMFM-88, MAPS, GPS and time- and distance parameters were compared between the UC and IP using the Wilcoxon-Signed Ranks Test. Changes within the period of UC and IP were evaluated based on the pre-and post values, using the Wilcoxon-Signed Ranks test as well.

Results

Participants

Sixteen children were included in the study. The median age of the children was 5y11months (IQR 5y1m – 6y10m). Five children were classified as GMFCS I, four children GMFCS II and two children GMFCS III. Details of the therapy frequency and the use of orthosis are provided in **table 2**. There were no significant differences between both periods regarding therapy frequency (p=0,888) and the use of orthosis (p=0,135). All children except one were included in mainstream education. All children received physical therapy by a private physical therapist. One child was included in a special education program and received physical therapy by the physical therapist at school and two extra sessions per week by a private therapist.

Fourteen different physical therapists participated in the study. The median number of experience was 8 years (IQR 5-11 years). Eleven therapists were specialized in pediatric rehabilitation and 12 therapists had a basic or advanced Bobath certificate.

As three therapists reported not having enough time to complete the diaries, three diaries were not returned and usual care data was not available for these children. Nevertheless, as the therapist still fully participated in the use of the programs and the children underwent all analysis, they were not excluded from the study.

Evidence-based practice behavior in usual care physical therapy

Fig 1 provides the distribution of therapy time over the different problems on the ICF. Based on the diaries, therapists reported that more than 50% of therapy time targeted impairment level, with 22,5% (IQR 16,7- 32,5%) of therapy time spent on stretching and 33,3% (IQR 30,2-33,3%) of therapy time targeting muscle strength.

Regarding activities and participation level, 41,7% (IQR 33,3-56,2%) of therapy time was spent on specific functional exercises and only 0,1% (IQR 0-6%) was used to address specific participation issues.

Table 2. Treatment characteristics during the period of usual care and interventions

	Usual Care	Intervention
Frequency (times/week)	2 (1 - 5)	2,5 (1-4)
Duration (minutes of session time)	45 (30-60)	35 (30-60)
Therapy per week (minutes/week)	105 (30-300)	120 (30-240)
Use of day orthosis		
Not used	3	3
Not Frequently (<50%/day)	2	2
Frequently used (>50%/day)	6	6
Use of night orthosis		
Not used	8	6
Not Frequently (<50%/night)	1	4
Frequently used(>50%/night)	2	1

Note:

Frequency, duration and therapy per week are provided using medians (IQR), for the use of orthosis, the number of patients is provided

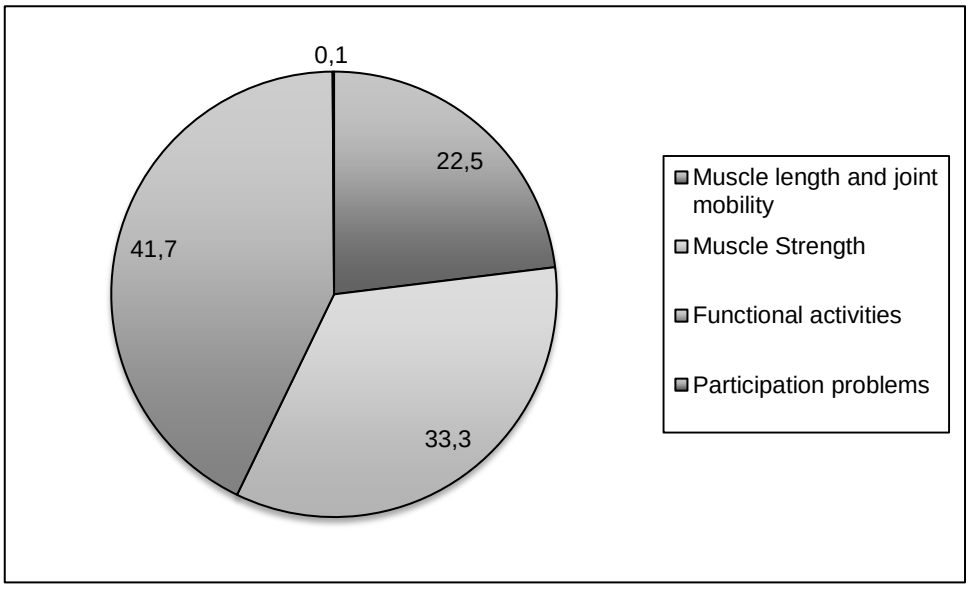

Figure 1. Proportion of therapy time spent on the different problems of the ICF based on registration in the diaries during the period of usual care.

Table 3 summarizes the characteristics and the treatment modalities applied for muscle length and muscle strength. Regarding muscle length, M.Triceps Surae, hamstrings and M.Psoas were most frequently stretched. All therapists applying stretching techniques, targeted these muscles. An active or dynamic component of stretch was most frequently used, except for hamstrings, where only 44,4% of the stretching time applied an active component. The number of repetitions varied between 2 to 6 times and the stretch was mostly maintained for 10-15 sec.

Strength training was most commonly performed for the hip extensors, hip abductors and ankle dorsiflexors (90,8; 90,8 and 80% respectively). For the hip extensors and ankle dorsiflexors, exercises were mostly performed as analytical exercises, while no functional strength exercises were used. Three sets of 10 repetitions were used most commonly. In the majority of the exercises, therapists reported that they applied external or manual resistance. However, none of the therapists quantified the applied resistance and none of the therapist registered or mentioned an increase in intensity or resistance.

Table 3. Characteristics and use of the treatment modalities used for muscle length and muscle strength as registered in the diaries during the period of usual care.

	Muscle length			
	Frequency (%)	Active (%)	Duration of stretch (sec)	Repetitions (N)
M.Triceps Surae	100 (86,7-100)	61,1 (0-90)	10 (10-17)	3 (2-5)
Hamstrings	100 (84,2-100)	42,4 (0-91)	10 (10-16)	5 (3-6)
M.Psoas	100 (66,2-100)	89,7 (17,9-100)	10 (10-15)	3 (2-5)
Adductors	65 (30-100)	100 (87,5-100)	15 (8-19)	4 (2-6)
m.Rectus Femoris	57,5 (20-95)	100 (100-100)	12 (10-15)	2 (2-3)

	Muscle strength				
	Frequency (%)	Functional (%)	Sets (N)	Repetitions (N)	Resistance (%)
Hip extensors	90,8 (67,5-100)	0 (0-19,4)	3 (2-3)	10 (10-12)	100 (81,9-100)
Hip abductors	90,8 (62,1-100)	10,8 (0,5-8,3)	3 (2-3)	10 (10-13,8)	100 (84,0-100)
Ankle dorsiflexors	80,0 (30-100)	0 (0-3)	3 (2-3)	10 (10-10)	100 (45,8-100)
Abdominal muscles	70 (52,5-100)	1,7 (0-77,8)	3 (2-3)	10 (10-14)	100 (52,4-100)
Knee extensors	67,5 (38-98)	18,1 (0-48,2)	3 (2-3)	10 (10-10)	100 (94,6-100)
Ankle Plantar Flexors	10,9 (6,2-25,4)	50 (12,5-87,5)	2 (1-3)	10 (10-11)	75 (12,5-100)

Notes:

Medians and interquartile ranges are provided

Active: proportion of total time targeting muscle length using an active/dynamic stretch

Functional: percentage of total time targeting muscle strength using functional strength training

Resistance: percentage of total time targeting muscle strength applying external or manual resistance

Functional exercises were mainly performed in standing (63,3 %), followed by kneeling or high-kneeling (28,3 %) and sitting (10,0%). In 67,5 % of the sessions, therapists performed gait training and transfers were practiced in 39,2 % of the sessions. **(table 4).** Regarding participation, therapists most frequently reported that they address limitations at school, followed by sports and problems at home. As participation issues were not often addressed, the median frequencies were 0 (range 0-29%), 0 (range 0-62%) and 0 (range 0-12%) respectively.

Effects of evidence-based practice behavior on outcome

Comparing the GMFM-88 changes during the period of UC to the changes during the IT, showed no significant differences (p=0,595). **Table 5** provides the **GMFM-88 scores** before and after the programs. However, a significant improvement on the GMFM-88 total score was observed after the IT (p=0,002) but not after the period of UC (p=0,508). Similar results were found for GMFM dimension E that showed significant improvement after the IT (p=0,042) but not after the period of UC (p=0,139).

On **gait analysis measures**, no significant between-group differences were observed for the GPS and MAPS. Nevertheless, the median change score for the GPS showed a slight improvement after the period of IT (change 0,26°; p=0,33) and deterioration after the period of UC (change -0,48°, p=0,50). These changes were not significant. Overall, slight improvements were registered after the IP for all MAPS except the MAPS for the ankle in sagittal plane and the hip in coronal plane. After the UC, slight improvements were only registered for the MAPS in the coronal and transversal plane (**Fig 2)**. **Table 6** represents the normalized time-and distance parameters. Significant deterioration was observed for step length during the period of IP. However, as these values ranged within normal values, these differences may clinically be interpreted with limited relevance. Otherwise, no significant changes could be observed.

Table 4. Frequencies of the positions and specific activities targeted in the functional exercises as registered in the diaries during the period of usual care.

Position	Frequency Med(IQR)
Lying	7,5 (0-43,7)
Sitting	10,0 (0-52,1)
Kneeling or high-kneeling	28,3 (1,3-64,3)
Standing	63,3 (41,3-100)
Target function	**Frequency Med(IQR)**
Trunk control	7,5 (0 - 19,2)
Transfers	39,2 (5,0 - 53,8)
Gait	67,5 (50 - 100)
Arm & hand function	12,1 (1,3 - 48,5)
Jumping, hopping or climbing	9,6 (0 - 20,0)
Stair climbing	0 (0 - 15,0)

Note Median frequencies represent the number of times using the specific position or targeting the specific function to the total number of therapy sessions.

Table 5. Results for the Gross Motor Function Measure before and after the period of usual care and the period of intervention

	Usual Care					Interventions					
	Pre		Post			Pre		Post			
	Median	IQR	Median	IQR	p (within)	Median	IQR	Median	IQR	p (within)	p (between)
GMFM total	92,09	87,91; 98,20	93,21	86,22; 98,01	0,508	88,67	85,69; 97,34	91,35	85,46; 98,30	0,002	0,594
GMFM-A	100,00	100; 100	100,00	100; 100	0,317	100,00	100; 100	100,00	100; 100	1,000	0,317
GMFM-B	100,00	100; 100	100,00	100; 100	0,317	100,00	100; 100	100,00	100; 100	1,000	0,317
GMFM-C	100,00	85,71; 100	100,00	89,29; 100	0,655	100,00	84,82; 100	100,00	85,12; 100	0,686	0,180
GMFM-D	87,15	70,84; 98,08	86,54	75,00; 99,04	0,779	84,62	77,24; 98,08	89,10	81,73; 100	0,214	0,237
GMFM-E	74,30	60,26; 90,28	80,91	61,28; 96,88	0,139	71,88	58,68; 87,33	75,69	60,41; 91,49	0,042	0,515

Abbreviations: GMFM-88: Gross Motor Function Measure–88; IQR Interquartile range

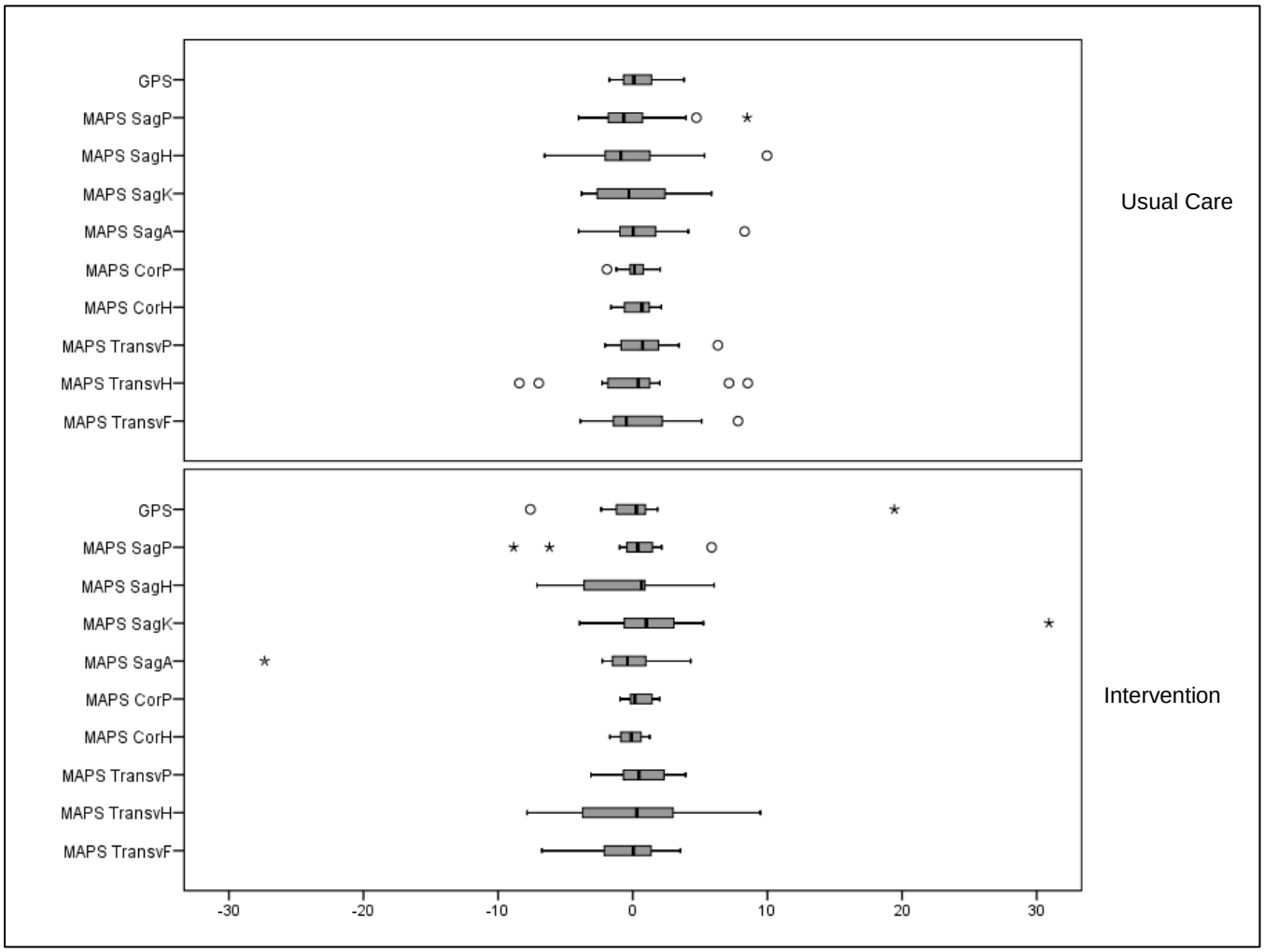

Figure 2. Median, interquartile ranges, minimal and maximal values of the changes in the Movement Assessment Profiles (MAPS) and the Gait Profile Score (GPS) after the period of Usual Care and the Intervention Period. (change score = pre-post)

Abbreviations:

Sag: Sagittal; **Cor:** Coronal; **Trans:** Transversal; **P:** Pelvis; **H:** Hip; **K**: Knee; **A:** Ankle; * and ° represent the outliers

Table 6 Normalized time-and distance parameters before-and after the period of Usual Care and Intervention

	Usual Care					Interventions					
	Pre		Post			Pre		Post			
	Median	IQR	Median	IQR	p (within)	Median	IQR	Median	IQR	p (within)	p (between)
Cadence	0,55	0,45;0,61	0,54	0,46;0,61	0,955	0,54	0,46;0,60	0,53	0,50;0,60	0,550	0,438
Step Lenght	0,80	0,60;0,87	0,7565	0,65;0,83	0,187	0,76	0,66;0,83	0,17	0,15;0,23	<0,001	<0,001
Step Time	1,82	1,62;2,12	1,86	1,63;2,19	0,816	1,86	1,65;2,18	1,88	1,66;2,06	0,796	0,438
Stride Lenght	1,59	1,20;1,78	1,52	1,34;1,68	0,179	1,52	1,37;1,69	1,63	1,28;1,67	0,293	0,121
Stride Time	3,64	3,29;4,52	3,76	3,28;4,39	0,918	3,76	3,33;4,40	3,78	3,35;4,0	0,501	0,393
Walking Velocity	0,42	0,29;0,53	0,40	0,36;0,49	0,379	0,41	0,34;0,49	0,43	0,36;0,45	0,421	0,326
Single Support	0,34	0,32;0,36	0,33	0,32;0,38	0,737	0,34	0,32;0,38	0,34	0,31;0,38	0,456	0,756
Double Support	0,17	0,14;0,28	0,18	0,14;0,24	0,605	0,18	0,13;0,25	0,20	0,16;0,28	0,798	0,609
Foot Off	0,60	0,59;0,64	0,60	0,58;0,63	0,569	0,60	0,58-0,63	60,84	0,59;0,63	0,91	0,91

Note
Time and distance parameters are presented as non-dimensionless parameters, according to Hof

Discussion

The **first aim of the stud**y was to evaluate the contents and effects of usual care physical therapy for children with bilateral spastic CP in Flanders.

The results revealed a dominant focus on impairment level, with 22,5% of therapy time spent on muscle length and 33,3% of therapy time targeting muscle strength. Although the time per session spent on functional activities was very limited, the contents of the functional exercises seemed more appropriate. In 67,5% of the sessions, gait training was part of therapy and in 63,3% of the therapy sessions, functional exercises were performed in and from a standing position. This can be considered as clinically relevant and appropriate in relation to the classification and pathology of the participating children.[12]

Exercises on muscle length followed the evidence-based recommendations using three repetitions. However, the applied stretches were of relative short duration (10 sec) in comparison to the findings of Lee et al, demonstrating more advantageous results for a 30 seconds stretch.[19]

Regarding muscle strength, the number of three sets of 10 repetitions that was most commonly reported, was very close to the recommendations by the NSCA guidelines, that recommended three sets of 12 to 15 repetitions.[5] Although the physical therapists registered a very high frequency of using external of manual resistance, dosing of the applied resistance was not registered. In 95% of the exercises, therapists reported the use of manual resistance and only in 5% of the cases, therapists applied an external resistance. The sessions forms in the diaries, specifically asked to specify the applied resistance. However, none of the therapists quantified the applied resistance nor mentioned an increase in intensity or resistance throughout the ten weeks training period. Therefore, the median applied resistance could not be objectified nor quantified. These results correspond to the findings of Verschuren et al., who identified the common problem of insufficient dosing as a major limitation to achieve treatment effects.[20]

In a large cohort study, Palisano et al. reported parent ratings on the focus and process for occupational and physical therapy.[21] The authors thereby highlighted that a great extent of therapy was addressed to primary impairments. However, it is difficult to compare the present results adequately to the results of Palisano et al., as our study questioned therapist instead of parents about the therapy focus and content. The use of diaries in our study aimed to find more objective figures to evaluate therapy contents. To our knowledge, no other studies have monitored therapy contents. Still, monitoring therapy using a diary as such might be a more specific and precise tool to evaluate therapy contents. However, it is also possible that it gives therapists the feeling of being evaluated. In general, therapists also experienced the diary as time-consuming and therefore, causing a large additional administrative burden. Additionally, although one must thrust the therapists in their honesty, there may also be a discrepancy between what was registered in the diary and what was actually performed.

The **second goal of this research paper** was to compare the effectiveness of the period of UC to the effects of an evidence-based physical therapy program used as an intervention. It was hypothesized that supporting therapists and providing an evidence-based predefined intervention program, would cause better treatment results than usual care physical therapy.

Effects the interventions were mainly visible on gross motor function. Children showed a significant change in GMFM-88 total and dimension E score after the period of intervention but not after the period of usual care. On gait measures, a non-significant tendency was visible showing improved MAPS in sagittal plane after the period of intervention but not after the period of UC. However, the reason behind these differences remains unclear. The diaries demonstrated a rather low ratio of therapy time addressing functional activities during the period of UC (41,7%). The predefined intervention programs consisted of more than 50% functional exercises. This might, besides the support to the therapists, be an important factor influencing our results. A limitation of our study was that we did not monitor the execution of the IP using a diary. This could have provided us more precise information on the specific time spent on the different exercises of the program. The differences in effects may also be related to the impact of the intervention as such. Providing therapist a framework of support might break routine therapy behavior and might as such make therapists more aware of the contents and impact of their treatment. Finally, a fundamental change in the content of the routine therapy might have an effect on the motivation of the children, who might have been triggered by new tasks and exercises.

It is therefore promising to see the positive effects of the evidence-based predefined intervention. This suggests the need for future studies to understand the mechanisms behind the effects of an therapeutic intervention and further investigate knowledge translation strategies to improve evidence-base practice behavior.

Conclusion

In physical therapy of ambulant children with bilateral CP, regular therapy sessions during a period of usual care are dedicated into a large extent to problems of muscle length and muscle strength. Thereby, especially muscle strength exercises seemed to have insufficient dosing compared to evidence-based guidelines. Functional exercises mainly involved exercises in standing and specifically gait training. A period of evidence-based intervention providing support to the therapists, demonstrated more advantageous effects on gross motor function compared to a period of usual care.

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General discussion

The scope of this doctoral project was to develop and evaluate an evidence-based, individually defined and targeted physiotherapy approach for ambulant children with bilateral spastic CP.

Children with bilateral CP face a variety of problems at all levels of the ICF. Therefore, the physiotherapist treating these children stands for the challenging task of priority setting and selecting appropriate treatment goals. A variety of physiotherapy treatment tools are available to reach those treatment goals, ranging from basic techniques (stretching, strengthening etc.) to more complex treatment concepts like neurodevelopmental treatment, functional training and conductive education, possibly complemented with additional therapies like aquatic therapy and horse-riding therapy.

The first part of this project (chapter 1 & 2) provided an overview of the available evidence for these techniques. An extensive literature review was performed and evidence tables were developed to overview the possible use of these different techniques and approaches that can be applied to reach the most frequently selected treatment goals for ambulant children with bilateral CP. In the second part (chapter 3), a framework was developed and validated to support the physiotherapist in clinical decision making. This framework facilitates the selection of individual treatment goals based on an appropriate definition of the main problems of the child. In part 3, the reliability of the GMFM-88 by means of video-scoring was assessed. The last part (chapter 5, 6 & 7) evaluated the effectiveness of an evidence-based, individually defined targeted treatment approach by comparing it to the effectiveness of a general program and a period of usual care.

Fig 1 summarizes the different chapters and highlights the main conclusions of each study.

This general discussion will summarize the main findings of the doctoral thesis and will provide critical considerations and recommendations for future research.

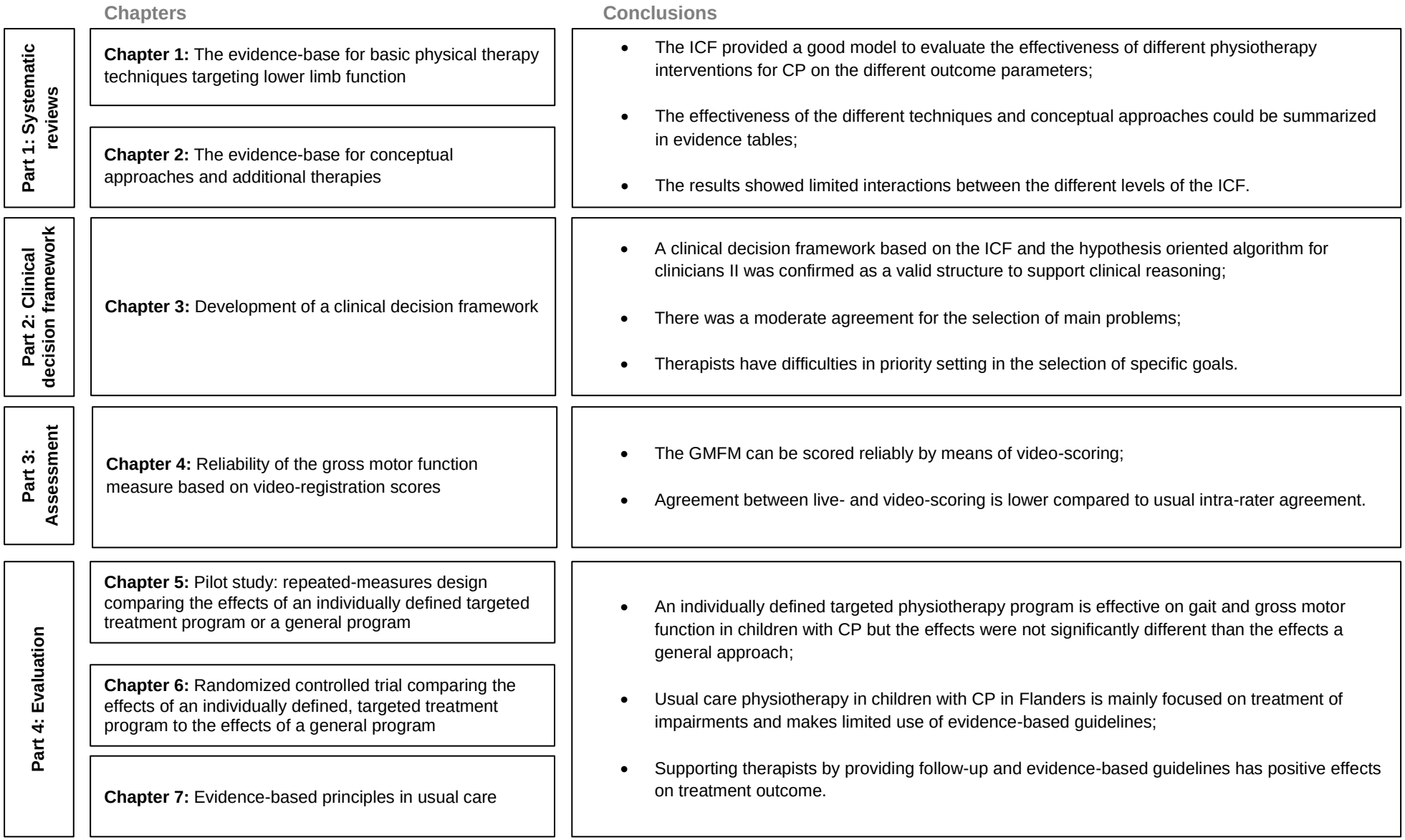

Fig 1. Outline and conclusions of the different studies.

1. Summary of the results

As a **first step** in this doctoral study, a **systematic literature review** was performed to describe and discuss the effectiveness of the different physiotherapy techniques and approaches targeting the lower limbs in children with CP. Between 1995 and 2010, 87 studies could be selected evaluating the effectiveness of **basic techniques (study 1)**.[1] Evidence-tables were developed to facilitate the selection of the appropriate techniques to reach specific treatment goals. The most frequently selected outcome parameters in the interventions were situated at impairment level. To improve range of motion, the use of stretching could be supported with level II evidence. Strong evidence was found for the effects of isometric and isotonic strength training on muscle strength. On the other hand, conflicting evidence was found for the use of electrical stimulation and endurance training on different strength measurements. Energy expenditure and movement efficiency were positively influenced following functional and isotonic strength training and treadmill training. On activity measures, level II evidence supported the use of treadmill training, isotonic or isometric strength training and balance training to improve different gait measures. Gross motor function was found to be effectively influenced by endurance training and strength training. Regarding participation and quality of life, level IV evidence was established for the effectiveness of massage on self-efficacy and on reduced parental anxious behaviour and depression.

Furthermore, 37 studies evaluated the effectiveness of **conceptual approaches (study 2)**.[2] At impairment level, only level IV evidence was found for the effects of NDT on posture. At activity level, level II evidence was found for the effect of functional training, NDT and goal-oriented approaches on gait and gross motor function. To improve children's participation, functional training using a goal-oriented approach was confirmed as being effective.

Overall we could conclude that limited interactions were found between the different levels of the ICF. Interventions targeting problems at body structure and function mainly influenced this level without significant overflow to activity level and vice-versa. An important clinical message following this conclusion was that a targeted treatment approach based on a complete and extensive evaluation at all levels of the ICF is necessary to create an appropriate treatment plan.

Therefore, **part two** aimed to develop a **clinical reasoning strategy** to support the paediatric physiotherapist in selecting the appropriate treatment goals **(study 3).**[3] The clinical reasoning framework integrated two existing but complementing frameworks: the **ICF and the Hypothesis Oriented Algorithm for Clinicians – II (HOAC-II).** The ICF provided the perfect structure to organise assessment results. The HOAC-II was used as a structured strategy-oriented approach for evidence-based clinical reasoning and to reduce the problem list to a refined shortlist of main problems. Within this structure, three-dimensional gait analysis was used as a supportive tool to unravel the complex interaction between the different components and problems. The framework was validated using a group of 22 physiotherapists, who identified the main problems and specific treatment goals for eight ambulant children with bilateral CP. Cluster analysis revealed a logic connection between the

selection of main problems and specific goals, confirming the algorithm as being a valid structure that supports logic clinical reasoning. Additionally, the results of this study revealed a very high agreement between the therapists concerning the identification of main problems. Agreement regarding the selection of specific treatment goals was lower and in 29% of the cases, therapists could not reduce their selection of goals to a number lower than eight. Most of the goals however, targeted muscle strength, range of motion and items from the GMFM-88 dimension D.

As a next step, in **part 3** of the project, the evidence-based knowledge and the clinical reasoning strategy were used to develop an individually defined, targeted treatment approach. The clinical reasoning strategy was applied to identify the main problems of the child. The corresponding specific treatment goals were selected based on the discussion with the child, the child's therapist and parents. The evidence tables from the systematic literature review were used to select the appropriate treatment technique to reach the goals. To be able to evaluate the effectiveness of the individually defined targeted treatment approach, an objective, blinded assessment of the children was necessary. Thereby, both clinical gait analysis and the GMFM-88 were identified as valid and reliable measures to evaluate gait and gross motor function in children with cerebral palsy. As the Clinical Motion Analysis Laboratory at the University Hospital Pellenberg has built up extensive experience in clinical gait analysis with several efforts to achieve high-standard quality measurements, these evaluations could be considered as being reliable. Additionally, the measurements could be performed by the experienced but independent staff of the laboratory, blinded to the set-up of the study.

To be able to use the GMFM-88 in an evaluator- blinded trial, the GMFM-88 had to be tested in a new reliability study assessing the agreement between a live assessment and an evaluator-blinded assessment by video-scoring **(study 4**).[4] The results revealed a high agreement between the live-and video scores but lower agreement compared to regular intra-rater reliability. Blinded video-scorings of the GMFM-88 were reliable, but when interpreting the results of clinical trials, we concluded that the changes should be interpreted using the adjusted responsiveness measures.

Consequently, **part 4** of this dissertation represented the intervention studies evaluating the effectiveness of the individually defined targeted treatment approach. **Study 5** was designed as a randomized, single-blind repeated measures study.[5] After a period of 10 weeks usual care with no intervention, ten children with bilateral spastic CP were randomly assigned into an individually defined, targeted treatment program or a general program, followed by a cross-over. Evaluation was made using the GMFM-88 and three-dimensional gait analysis with outcome measures evaluating both the general progress of the child as well as the attainment of the individual goals of the child. The results of this study only indicated slightly favourable effects towards the individually defined program but could not confirm statistically significant differences between the outcomes of both programs. Limited changes however, were observed after the period of usual care. **Study 6** compared the effectiveness of the individually defined approach to the effectiveness of a general therapy program in a larger, randomized trial using the same outcome measures.[6] Although this study included 60 interventions, the results did also not demonstrate statistically different results between an individually defined treatment approach and a general program on gait and gross motor function of these children.

However, analysis of the entire intervention group highlighted positive treatment effects for all children, regardless of the type of program they were enrolled.

Hence, we hypothesized that supporting therapists by providing a predefined evidence-based intervention program, may have contributed to the improved treatment outcome compared to the usual care physiotherapy. Consequently, **study 7** aimed to evaluate evidence-based practice behaviour in usual care physiotherapy in Flanders and to compare the effectiveness of the predefined physiotherapy programs based on evidence-based guidelines to the effects of a period of usual care.[7] The results of this study indeed revealed that the predefined intervention programs provided better treatment results compared to the period of usual care, with significant differences in changes on gross motor function. Looking into the contents of usual care physiotherapy in Flanders showed that physiotherapy still mainly focuses on problems at impairment level and that exercises were characterized by insufficient dosing.

2. Critical considerations

2.1 Systematic reviews

2.1.1 Basic techniques

In 2008, Anttila et al. performed two high-quality systematic reviews evaluating the effectiveness of physiotherapy and conductive education.[8] Although these reviews only included randomized trials and systematic reviews respectively, the authors still identified severe limitations in methodological quality because of the large variations in population, interventions and outcome parameters of the trials. In our systematic reviews, we also observed a large discrepancy between the study designs (providing the level of evidence, Sacket) and the study quality scores (conduct scores, AACPDM recommendations). Our findings confirm that the randomized controlled trials, valued as providing a high level of evidence, generally showed conspicuously low quality scores. This conclusion led to our main motivation to include all types of intervention studies regardless of their level of evidence.

Comparing the specific conclusions, Anttila et al. only concluded moderate evidence of ineffectiveness of strength training on gross motor function. Our systematic reviews found level II evidence for the effectiveness of strength training on both strength measurement and gross motor function. These differences in results can probably be explained by the fact that Anttila et al. applied different study inclusion criteria. In addition, Anttila et al. could not yet include the trials published between 2007 and 2010, a period during which several intervention studies on strength training were published.

A more recent meta-analysis by Scianni et al. reported inconclusive results regarding the effectiveness of strength training in children with CP.[9] These conflicting results may be explained by the fact that Scianni et al. considered neuromuscular electrical stimulation as a direct strength intervention and therefore, included different studies using strength training in the meta-analysis. Nevertheless, all reviews described the findings regarding the use of strength training as promising and identified the weak trial qualities as factors limiting the development of specific evidence-based guidelines. Verschuren et al. summarized the contents of the strength interventions and thereby identified insufficient dosing used in the majority of the studies as a major contributor for limited results.[10]

For the remaining basic techniques, our conclusions regarding stretching, balance training, endurance training correspond with the results of Antilla et al.

2.1.2 Conceptual approaches

Regarding the effectiveness of the different approaches, different systematic reviews report even more conflicting results.

Regarding **Neurodevelopment Treatment (NDT) or Bobath therapy**, our review found level IV evidence for the effectiveness of NDT but identified the concept as the only treatment concept causing effects at all levels of the ICF. In an AACPDM evidence report, Butler & Darrah found no advantage of NDT over the alternatives to which is was compared.[11] With the exception of immediate improvement of dynamic range of motion, there was no consistent evidence that NDT demonstrated positive effects on the development of contractures, abnormal motor responses or motor development. Brown & Burns reported similar findings.[12] The conclusions of Anttila et al. were more positive and reported high-level evidence of NDT on the developmental status of very young infants.[8,13] A very recent systematic review by Novak et al. however, concluded that there was no superior value of Bobath therapy compared to the more cognitive motor learning strategies and therefore, graded the therapy approach as 'red light'.[14] Red light was defined as ineffective, indicating that it should not be used.

Nevertheless, Bobath therapy is one of the oldest treatment approaches for children with CP and probably still one of the most frequently used principles worldwide. In order to make evidence based guidelines, it is important to evaluate the available evidence and the contents of the concept more critically.

An important reason for the controversial conclusions might again be the different study inclusion criteria applied by different reviews. Our study included level IV studies, but all other studies only included RCT's and/or high level of evidence studies (I and II only). Many of the studies evaluating the effectiveness of NDT were not randomized controlled trials or had lower levels of evidence. Therefore, these studies were not included in the systematic reviews by Antilla et al., Novak et al., Butler & Darrah and Brown & Burns.[8,11-13,15] Additionally, the review by Butler & Darrah and Brown & Burns are much less recent and therefore, included older studies (until 2001).

Limited and controversial evidence is available regarding the use of Bobath therapy. Still, acknowledging the limitations of clinical trials of the involved RCT studies, it might be a precipitated decision to refute the usefulness of the concept. Additionally, when an intervention does not provide different treatment results compared to an alternative, it might more reflect the equal value of both therapies instead of its ineffectiveness.

Evaluating the content of the Bobath concept more critically is a very challenging experience. Worldwide, different interpretations are made regarding the concept and unfortunately, some of these have drifted away from the original concept as defined by Dr and Mrs Bobath. Additionally, Bobath is a concept that has evolved a lot.

Looking into literature regarding the content of Bobath therapy highlighted several reported concerns regarding the concept. It is worthwhile to critically consider these concerns in the view of the original ideas and more recent evolutions.

A first major concern is the statement that Bobath therapy is rather passive, using only neuromaturational principles of motor learning.[15-17]

Motor learning is described as a set of (internal) processes associated with practice or experience leading to relatively permanent changes in the capability for responding.[18] Motor learning can therefore be seen as a very broad term. Over the years, different principles of motor learning have been developed, all based on both explicit and implicit assumptions, associated with an underlying theory of motor development or motor control. Neurophysiological and neuromaturational approaches are largely based on assumptions that the development of movements and motor skills result solely from the neurological maturation of the central nervous system. This maturation involves that higher centres inhibit and control lower centres, thereby allowing voluntary movement. More recent theories of motor learning emphasize that motor or developing behaviour should not be viewed as the unfolding or predetermined or prescribed patterns represented in the central nervous system. These approaches favour a more heterarchical view in which motor development and coordination are assumed to emerge from the dynamical interaction of many subsystems in a task-specific context. This approach, therefore, is based on an active rather than a passive view of motor learning. People learn by actively attempting to solve the problems inherent to a functional task.[19,20] Looking back into to the Bobath concept, brings us to the following original text by Dr and Mrs Bobath (**Appendix AC**). The basic idea is that the child is assisted in problem-solving and that therapist enables him or her to experience the patterns of movement and success in achieving the task. For example, through facilitation, the Bobath clinician intends to provide key components of the spatial and temporal aspects of a specific movement or task. The therapist thereby enables the child to have an experience of movement that is not passive but he or she cannot yet do alone. The Bobath concept tries to progress from a facilitated movement to a hands-off, active learning position for the child. From that perspective, the active learning component applied within the Bobath concept probably reflects more its heterarchical and active view on motor learning.

A second important concern is that the Bobath concept is considered to address impairments (muscle tone and range of motion) only and insufficiently targets activities and participation.[15,21]The Bobath approach aims to modify the patterns of abnormal postural tone and to facilitate more normal motor patterns as a preparation for a specific functional skill such as dressing, eating, drinking, writing, walking etc. Treatment is not considered as a repetition of the functional task but is carefully considered by task-analysis. There is stepwise preparation for a specific functional skill to enable the child to function in the most possible efficient way. The aim is to perform postural and voluntary tasks with the least possible interference from abnormal postural tone. Secondly, parent-training and home-based training are an inherent part of the Bobath concept. The Bobaths emphasized that learning requires meaningful goals, relevant to the child. (**Appendix AB**). While the Bobath concept maybe based on a different strategy compared to functional, task-oriented training, the integrative approach

of the Bobath concept attempts to target the multifaceted problems of the child, thereby approaching the recent developments and ideas regarding child-and family involvement and participation. It does not only take into account the impairments of the child, but beyond that, prepares the child for specific activities and participation limitations. In that way, Bobath therapy integrates and even fully addresses the different components of the ICF.[22]

The third important concern regarding the interpretation of the Bobath concept is that it fails to allow the incorporation of other treatments (e.g. strength training) that have been shown to be equally effective.[23] The Bobaths viewed strength more as a secondary problem in children with CP and have thereby probably overemphasised the neural control of movement.[24] This may have caused them to neglect the importance of muscle strength, force production and movement velocity. This can probably be explained by the fact that the Bobaths pioneered their work over 60 years ago, long before the recent developments in imaging technologies. Recent publications regarding Bobath therapy have reported the need to integrate strength training.[22,24] Nevertheless, integration of strength training is probably not yet fully dispersed amongst all Bobath-clinicians.

In summary, due to the limited available evidence regarding the Bobath-concept, evidence-based decision making about the Bobath concept is difficult. A high degree of confusion and different opinions exist regarding the content of the concept. Nevertheless, the basis of the concept meets the current evidence-based needs formulated by the dynamic concept of the ICF. To be applied in an adequate way, the concept needs to be evolving, with a dynamic and opened view on new knowledge.

Functional training is often considered as the opposite concept of Bobath therapy. Functional training is based on a task-specific repetition of activities. Movement exploration, a (cognitive) selection of solutions to new task demands and the adaptation to changes in the environmental context are assumed to be critical parts of motor learning.[20,25,26] Functional training prefers hands-off treatment and focuses on learning the child to perform a task within naturally occurring restraints. Nevertheless, level II evidence for the effectiveness of functional training was only demonstrated on gait and gross motor function.

To our knowledge, no systematic review specifically summarizes the intervention studies evaluating the effectiveness of functional training.

A first major difference between functional training and Bobath therapy is the specific preparation for function. Bobath therapy uses a more stepwise approach, starting from a specific preparation for function with attention for compensations. Functional training immediately practices the repetition of the specific functional task. Secondly, the cognitive component that is more often applied within functional training is much less pronounced within the Bobath approach, which is more based on movement experience.

Despite these difference, as both approaches aim to improve the child's daily functioning with attention to family involvement, supporting materials and environmental aspects, they probably lie closer to each other than they claim to.

Nevertheless, the heterogeneity of the pathology of CP requires individually defined treatment goals. Choosing the appropriate technique to reach the treatment goals is challenging and requires a balanced but evidence-based decision by the physiotherapist treating the child. Based on the available evidence, functional training seems to be very useful for the child to reach short-term functional goals. [20,25,27] Including children in functional goal-setting thereby clearly had a major additional value.[28-30]

The effectiveness of physiotherapy in children with CP probably lies more in an adequate combination of techniques than the selection of an individual technique or approach. Discussions regarding the so-called named approaches are often biased, theoretical and transcend the contents of the approaches as such. Much more important than the discussion in which technique is more valuable than another, is probably an adequate and evidence-based integration of different approaches. Evidence-based physiotherapy may therefore rather focus on the strengths of the different approaches and the effectiveness that already has been proven. Such thinking strategy allows integration of different aspects from different approaches into a comprehensive, multi-layer treatment approach that can address the individual problems of a child.

The developed evidence tables can support the physiotherapist in selecting an appropriate technique to reach a specific goal. Nevertheless, this approach requires an appropriate selection of individual treatment goals, which can be very challenging for the physiotherapist, seen the heterogeneous, complex and evolving nature of the pathology of cerebral palsy. An adequate clinical reasoning structure is still necessary to support the therapist in its evidence-based decision-making and priority-setting.

2.2 Clinical decision framework

In part two of this dissertation, a clinical framework was developed to support the therapist in evidence-based decision making. The ICF was thereby used as a structure to organize the assessment results, complemented with the Hypothesis-Oriented Algorithm for Clinicians – II (HOAC-II), to integrate a strategy-oriented component. This first goal of the study was to investigate how clinical framework could contribute to a reliable identification of main problems and specific PT goals in ambulant children with CP. There was moderate agreement in the selection of main problems. The agreement between the selection of specific goals was lower and the physiotherapists had difficulties in limiting their selection of specific goals to eight treatment goals. It is possible that specific individual goal-setting is still relatively new or unknown to the therapists and therefore maybe an aspect that requires additional training. However, it is also possible that selecting specific individual goals, is more subject to individual physiotherapist differences. Still, a logic connection was observed between the

identification of main problems and the selection of specific treatment goals. The first goal of this study was thereby achieved.

The second goal of this study was to evaluate how the additional information of three-dimensional gait analysis could influence the reliability of this information. Unfortunately, this second goal could not be achieved because of methodological restrictions in our study design. The gross motor function levels of the participating children were not equally divided over the groups with and without gait-analysis information and the number of children in both groups was too small. Although the agreement in the selection of main problems was higher in the group of children where gait analysis information was provided, we have to be careful to link these observed differences to the additional value of gait analyses only. Nevertheless, as gait analysis is a frequently used tool to evaluate ambulant children with CP, the use of this information to improve the understanding of the child's functional problems remains an important topic that requires further research.

2.3 Agreement video-scoring GMFM-88

The ICC values for the live-and video scores from the same assessor were good, but were lower compared to ICC values for the intra-rater agreement of the MSc students scoring the video-registrations. One might conclude that video-scores provided very good inter-and intra-rater-reliability values, but that the video-registrations as such created a small, systematic error that lowers concurrent validity of the GMFM-88 measurements. To evaluate the reasons behind this lower ICC values, it is recommended to compare the repeated live-scorings as well as the repeated video-scores all of the same assessor. Nevertheless, the reliability results were acceptable and allowed us to evaluate the within-person change values for the GMFM-88. **Fig 2** provides an overview of the different values of the within-person changes as reported in study 4.

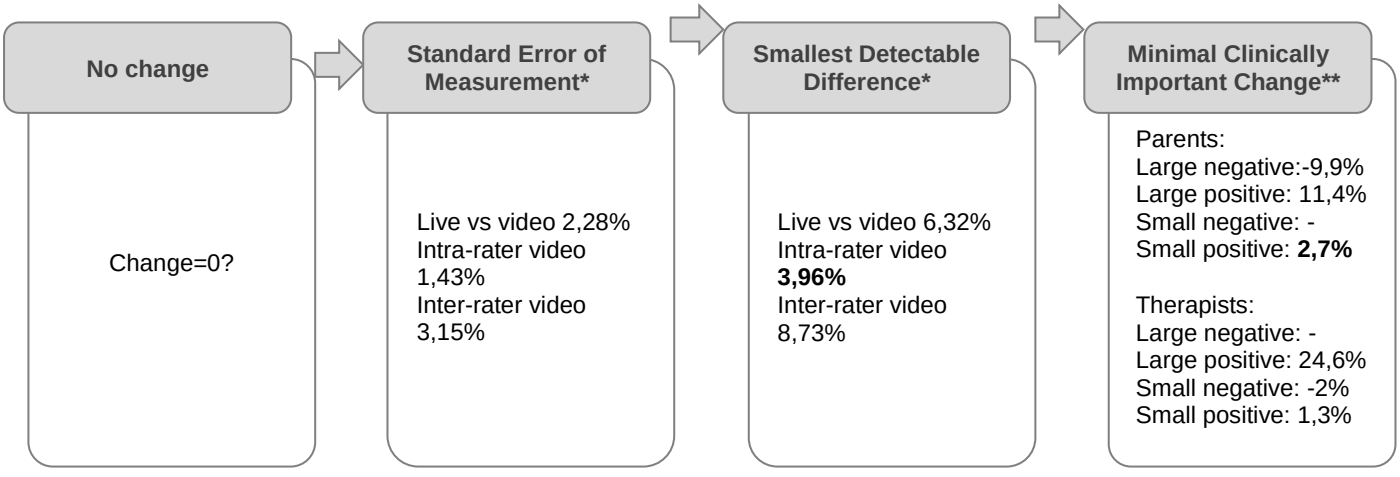

Fig 2. Within-person changes for the Gross Motor Function Measure-88

* Results from the agreement study (study 2) **Results of minimal clinical important change from Russell et al.[34] **-** not rated

Thereby, the **Standard Error of Measurement (SEM)** provides an absolute index of reliability. It indicates the variability of the scores around the subject's true score and thereby provides a value for the measurement error in the same unit as the measurement itself. The SEM allows the calculation of the **Smallest Detectable Difference (SDD) or the Minimal Detectable Change (MDC),** which is usually calculated as (SEM*1,96*√2). The SDD represents the amount by which a patient's score needs to change to ensure that the change is greater than the measurement error. The **Minimal Clinical Important Change/Difference (MCID)** is referred to as the smallest change in measurement that is considered to be meaningful based on clinicians ratings.[33]

The SEM and SDD could be calculated based on our agreement study. The MCID represented in **Fig 2** is based on the results of the study of Russell et al, who compared the subjective ratings of progress by parents and therapists to the actual change in GMFM-88 scores after a period of 6 months of physiotherapy.[34] When parents rated 'a small improvement' in gross motor functioning, an actual change of 2,7% was registered on the GMFM-88 score. Therapists seemed to capture these differences somehow easier, rating a 'small positive' progress already when an actual change of 1,3% change on the GMFM-88 was registered.

As relevant conclusions from this study, one can state that the intra-rater reliability of the GMFM-88 scores based on the video-scores provided a very good agreement and video-scores can be used reliably to evaluate the effects of an intervention. Provided that the same rater scores the video-registrations, one can define **3,96%** as a change that provides 95% certainty that the change is larger than a measurement error. (**Fig 2.)** When evaluating clinical relevance, on might use the **2,7%** of GMFM-88 change, representing the small positive change in gross motor functioning noticed by the parents, as MCID. **(Fig.2)**

2.4 Intervention studies

The first interventions (study 5&6) compared the effectiveness of the individually defined approach to a general program. Outcome was evaluated using the GMFM-88 and three-dimensional gait analysis, using parameters measuring general progress as well as progress at individual goals. Study 7 evaluated the effectiveness of a predefined, evidence-based treatment program to the effects of usual care physiotherapy. **Table 1** summarizes the results for the main outcome measures in the different intervention studies.

Regarding **gross motor function**, the results demonstrate overall positive effects for both intervention programs **(Table 1**). Still, the changes in GMFM were usually quite small. In study 6, the estimated change (pre-post) in GMFM-88 score was only -1,65% and -1,27% for the IT and the GT group respectively. These differences were not only smaller than the minimally clinically detectable difference reported by Russell et al. (2,7%)[34], but even smaller compared to the smallest detectable differences for the intra-rater reliability (3,96%) as calculated in by the reliability study.[4] This statistical significance can probably be explained by the observed trends in the groups, as almost all children

tended to improve their GMFM-88 scores. When all observed scores change in the same positive direction, smaller changes can already cause statistical significance effects. Comparable overall positive changes in GMFM-88 scores were found in study 5. However, due to the small number of participants in this study, no statistical significance could be observed in this study.

Trends in the outcome parameters of the **three-dimensional gait analysis** were different (**Table 1**). In none of the studies, significant changes could be found for the GPS. Only in the intervention group of study 7, changes in GPS were larger than the reported minimal clinical important difference as reported by Baker et al.[35] Positive changes in time-and distance parameters were only found for step- and stride length and this after the IT program in study 5 and 6.

Looking into the **parameters at individual level,** children also seem to reach their functional, activity-based goals more easily compared to the individual gait-analysis goals (**Table 1**). The motivational aspect of individual functional goals might have a significant impact on these results. Several studies have demonstrated the positive impact of goal-setting[25,27,36] and logically, children are probably more motivated for functional activities than specific corrections of their gait patterns. Nevertheless, the results in study 6 demonstrate non-significant but positive evolution of both interventions on gait analysis goals.

Table 1. Within-group changes for the main outcome measures for the intervention studies

		IT program			**General program**			**Usual Care**		
		Study 5	Study 6	Study 7*	Study 5	Study 6	Study 7*	Study 5	Study 6	Study 7
Individual goals	GAS	✓	✓	N/A	=	✓	*	N/A	N/A	N/A
	z-scores	=	✓	N/A	=	✓	*	N/A	N/A	N/A
General parameters	GMFM-88	✓	☑	☑	=	☑	*	=	N/A	=
	GMFM-88 - D	✓	☑	=	=	☑	*	=	N/A	=
	GMFM-88 - E	✓	☑	☑	✓	✓	*	=	N/A	=
	GPS	=	=	✓	=	=	*	=	N/A	=
	Time & distance parameters	☑	✓	=	=	=	*	=	N/A	=

Abbreviations:

☑ significant improvement; ✓ non-significant improvement (>smallest detectable difference); ☒ significant deterioration; ✗ non-significant deterioration (>smallest detectable difference); = no change (<smallest detectable difference); * pooled results with general program; GAS Goal Attainment Scale; GMFM-88 Gross Motor Function Measure 88; GPS Gait Profile Score; IT Individually defined, targeted; N/A Not Applicable

Based on these overall results, one might conclude that problems in gross motor function are more responsive to change compared to gait patterns. It may also be possible that the period of 10 weeks was too short to cause significant effects in gait. Additionally, although several exercises from the programs aimed at improving gait, it is very probable that the actual therapy time spent on gait problems was too short. The results of the analysis of usual care demonstrated that generally, therapists practice gait in 67% of their therapy sessions. Nevertheless, the time per session that was spent on functional activities was usually very short. It might be possible that although the prescribed programs contained more than 50% exercises at activity level (including gait), the therapy time spent on different gait problems was still too short. A last reason might be the well-known problem of regression to the mean. The heterogeneous nature of the pathology causes children to have problems at different joints and levels, which makes it very difficult to detect individual changes using general outcome parameters.[37]

Overall, although the within-group changes indicate advantageous results for the individually defined, targeted program, no statistical between-group differences could be found. The hypothesis that an individually defined, targeted treatment approach provides better treatment results than a general, aim-oriented program could thereby not be confirmed. In the discussions of the concerning papers, several causes could be identified to explain these results. A first and major reason was probably the relatively low number of participants, providing low statistical power. This was confirmed in the power-analysis of study 5. Additionally, the time required for the different exercises was not indicated by the researchers. Therefore, therapists may still spent too much time on the preparatory exercises of tone reduction, range of motion and analytical muscle strength and time left for functional exercises may have been too limited. This was clearly reflected in the treatment quality scores. A last important reason might be that differences between the general and the individually defined program might have been too limited to detect differences in treatment outcome. The general program was still a well-balanced program addressing the variety of problems seen in children with bilateral spastic cerebral palsy of this age-range and thereby, often came too close to the individually programs. Also, by making very small adjustments and by providing appropriate feedback, well-experienced therapists often still managed to approach the exercises from the general program in an ‘individual way’. Nonetheless, both intervention programs provided better treatment results than a period of usual care, which was confirmed in our last study.

3. Recommendations for future research

This doctoral project aimed to develop and evaluate an evidence-based, individually defined and targeted treatment approach for ambulant children with bilateral spastic CP. The majority of physiotherapy intervention studies evaluate the effectiveness of individual techniques. These studies thereby contrast with the reality of clinical practice. As the pathology of CP is characterized by a variety of problems at all levels of the ICF and the interaction effects between the different levels are limited, there is a need for individually tailored and multi-layer approaches. It might also be possible that individual techniques complement each other in combined approaches. For example, limited level II evidence is available for the effectiveness of stretching on muscle length and range of motion. In clinical practice however, stretching is usually combined with a dynamic component of activation or strengthening of the antagonist. It might be that the use of strengthening the antagonist reinforces the effectiveness of stretching. However, interventions evaluating these combined approaches are scarce. These interventions are probably more complex to organise and to evaluate. Still, they reflect the reality of clinical practice and can thereby provide more opening for evidence-based practical guidelines. In addition, a clear and expedient clinical framework is needed to structure such interventions. Our clinical reasoning was only developed and validated for ambulant children with bilateral spasticity, aged 4 to 9 years. Further research is necessary to evaluate its use in children from different age-categories, typologies and functional levels.

Additionally, the use of gait analysis in identifying main problems and treatment goals needs to be further explored. Gait analysis has proved its effectiveness in the identification of goals for orthopaedic surgery and BTX-A.[38-40] At the University Hospital of Pellenberg many children receive frequent gait analysis during the course of their treatment follow-up. Therefore, the information is usually available, allowing large retrospective cohort studies. Study 3 gave an indication that the results can support physiotherapists in identifying the main problems of the child, but the way this can be used in clinical practice, needs to be further investigated.

A weakness in our study was that we did not include strength measurements in our outcome parameters. The use of our strength measurements was limited to the analysis of the manual muscle testing scores and the visual interpretation of kinematic and kinetic parameters in the gait analysis results that reflect strength issues. Several researchers have observed significant increases in strength measurements after progressive resistance training, but identified smaller and non-significant mean increases in kinematic and functional outcomes.[41-45] Some researchers thereby refer to the need for analytical approaches to identify individuals who are most likely to benefit from strength training and to identify the specific muscles that require strengthening.[41] In addition, insufficient loading was identified as a reason for strengthening interventions to be ineffective. This might as well be related to the limited possibility to evaluate muscle strength and thereby, to identify weaker muscles. Although the use of handheld-dynamometry is a great progress compared to manual muscle testing, it still shows limited reliability and does not reflect functional, multiple joint muscle work.[46-48]

Instrumented three-dimensional gait analysis may thereby be a promising tool to contribute to the identification of the impact of muscle weakness on ambulatory function.

Regarding the use of the different therapy concepts, Bobath currently stands in the line of fire. Although some comments regarding the concept are correct, others are probably based on misinterpretations. The reason behind these misinterpretations might well be caused by the limited available information about the concept. The writings from Dr and Mrs Bobath are scarcely published and seem to be very fragmented. The concept is used all over the world, but with different ideas and interpretations. A clear and concise definition based on a common view is probably needed for the concept to survive. It might be important to emphasize the preparation for function more clearly as a core aspect of the concept together with the existing clear structured, clinical reasoning approach. Furthermore, as most of the available information regarding the effectiveness of Bobath therapy is available through level IV evidence, high-quality interventions are urgently necessary. Finally, the Bobaths have identified their concept as being dynamic and opened to new evidence-based knowledge and techniques. The progressive view of the concept has been confirmed in Flanders, where the paediatric Bobath therapy course was integrated into a fully academic paediatric postgraduate course. This was a big step forward for the program and might be an example for other countries applying the Bobath concept.

The basis of the individually defined therapy approach applied in this PhD was constructed on a wide set of principles, selected from different concepts. The approach thereby addressed the horizontal and dynamic structure of the ICF. Evidence-based guidelines and structured clinical reasoning were applied and attempted to transcend the on-going discussions between the theoretical concepts. Physiotherapy of children with cerebral palsy requires a dynamic physiotherapy approach, based on the balanced decision between the need to control impairments with sufficient attention to activity and participation, both on short-and long term. Goal-setting was a central aspect of the individually defined, targeted approach. This was not only a means to motivate the children, but also a means to structure the programs, to prioritize treatment goals and to address the individuality of a child's problems.

Future interventions should have a longer duration. In that way, the use of participation goals can be integrated more optimally. As ten weeks was too short to expect significant improvement at participation level, participation problems were only taken into account to design the individually defined programs. Extending the duration of the program will thereby allow an evaluation of the effectiveness of the programs at participation level. In addition, it may be encouraged to organize future interventions as multicentre studies. This allows larger treatment groups. Additionally, using international multicentred studies may allow the comparison of the effects of the different treatment strategies used in the different centres without the need to change the usual therapy.

The last study of this dissertation showed the positive impact of providing evidence-based guidelines and support to paediatric physiotherapists on treatment outcome. A fundamental change in the content of the routine therapy might have caused an effect on the motivation of the children, who might have been triggered by new tasks and exercises. It is therefore promising to see the positive

effects of the evidence-based predefined intervention. This suggests the need for future studies to understand the mechanisms behind the effects of a therapeutic intervention and further investigate knowledge translation strategies to improve evidence-base practice behaviour.

4. Conclusion

This doctoral project made a contribution to the development of an evidence-based individually defined, targeted treatment approach for ambulant children with bilateral cerebral palsy. A systematic review summarizing the effectiveness of the most commonly used physiotherapy techniques highlighted limited interaction effects between the different levels of the ICF. As the outcomes were differentiated at the different levels of the ICF, these results can support the physiotherapist in selecting the appropriate treatment technique. Secondly, an innovative and validated clinical decision framework allowed an adequate selection of the main problems and treatment goals. Comparing the individually defined treatment approach to a general physiotherapy approach demonstrated promising results that need to be confirmed on larger groups, using programs of longer duration. Supporting physiotherapist by providing evidence-based guidelines and follow-up has beneficial effects on treatment outcome. Future research should take a next step to promote evidence-based behaviour in clinical practice.

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Summary

Cerebral Palsy (CP) is defined as a group of disorders of the development of movement and posture, causing activity limitations that are attributed to non-progressive disturbances occurring in the developing fetal or infant brain. With an incidence of 2 to 4 in 1000 children, CP is the most common physical disability in childhood. It is characterized by a neurodevelopmental condition that begins in early childhood and persists throughout lifespan. It is estimated that around 46% of the children with CP show bilateral spasticity. In these children generally the lower limb is more involved than the upper limb.

Besides the increased muscle tone, impairments in the lower limb include muscle weakness, reduced selective muscle use, sensory deficits and secondary risk of muscle contractures and deformities. Gait and gross motor function are thereby often compromised, restricting the child's possibilities to participate in educational or leisure activities. In addition, the pathology of CP is often complicated by the fact that clinical manifestations are widely believed to change throughout lifespan.

The complexity of the problems seen in children with CP requires a multidisciplinary treatment. Thereby, the physiotherapist plays a key role. Physiotherapy treatment of children with CP is a balanced but challenging exercise between optimizing motor development, promoting independent functionality and participation and prevention of secondary problems that limit this daily functioning.

Nevertheless, although limited but promising results are available for the effectiveness of individual techniques, studies evaluating the effectiveness of integrative physiotherapy programs are scarce. However, a combination of different techniques may address he heterogeneity of the pathology in a more optimal way. This doctoral thesis therefore aimed to develop and evaluate the effectiveness of an evidence-based individually defined treatment approach for ambulant children with bilateral cerebral palsy.

In **part 1** of this doctoral study **(chapter 1 & 2),** a systematic literature review was performed to describe and discuss the effectiveness of the different physiotherapy techniques and approaches targeting the lower limbs in children with CP. The evidence of the most commonly used physiotherapy techniques was evaluated by differentiating the effects on the different levels of the ICF. Evidence-tables were developed to facilitate the selection of the appropriate techniques to reach specific treatment goals. Overall, limited interactions were found between the different levels of the ICF. Interventions targeting problems at body structure and function mainly influenced this level without significant overflow to activity level and vice-versa. An important clinical message following this conclusion was that a complete and extensive evaluation at all levels of the ICF is necessary to create an appropriate treatment plan for an individual child.

Part 2 (chapter 3) aimed to develop a clinical reasoning strategy to support the paediatric physiotherapist in selecting the appropriate treatment goals. The clinical reasoning framework integrated two existing but complementing frameworks: the ICF and the Hypothesis Oriented Algorithm for Clinicians – II (HOAC-II). The ICF provided the perfect structure to organise assessment results. The HOAC-II was used as a structured strategy-oriented approach for evidence-based clinical reasoning and to reduce the problem list to a refined shortlist of main problems. Within this structure, three-dimensional gait analysis was used as a supportive tool to unravel the complex interaction

between the different components and problems. The framework was validated with the support of a group of 22 physiotherapists, who identified the main problems and specific treatment goals for eight ambulant children with bilateral CP. Cluster analysis revealed a logic connection between the selection of main problems and specific goals, confirming the algorithm as a valid structure that supports logic clinical reasoning. Additionally, the results of this study revealed good agreement between the therapists concerning the identification of main problems. Agreement regarding the selection of specific treatment goals was lower and in 29% of the cases, therapists could not reduce their selection of goals to a number lower than eight. The majority of the goals however, targeted muscle strength, range of motion and items from the GMFM-88 dimension D.

As a next step, the evidence-based knowledge and the clinical reasoning strategy were used to develop an individually defined, targeted treatment approach. The clinical reasoning strategy could be applied to identify the main problems of the child. The corresponding specific treatment goals were discussed with the child, the child's therapist and parents. The evidence tables from the systematic literature review allowed us to select the appropriate treatment technique to reach these treatment goals.

To be able to evaluate the effectiveness of the individually defined targeted treatment approach, an objective, blinded assessment of the children was necessary. Thereby, both clinical gait analysis and the GMFM-88 were identified as valid and reliable measures to evaluate gait and gross motor function in children with cerebral palsy. Three-dimensional gait analysis could be performed reliably and independently at the clinical motion analysis laboratory of the University Hospital Pellenberg. However, to be able to use the GMFM-88 in an evaluator- blinded trial, the reliability of GMFM-88 by means of video-scoring had to be tested. Therefore, **part 3 (chapter 4**) described a reliability study assessing the agreement between a live assessment and an evaluator-blinded assessment by video-scoring. These results revealed a high agreement between the live-and video scores but lower agreement compared to regular intra-rater reliability. Blinded video-scorings of the GMFM-88 were reliable, but probably caused a systematic error compared to live-scoring. When evaluating the results of clinical trials, we concluded that the changes should be interpreted using the adjusted responsiveness measures.

Consequently, **part 4** (**chapter 5, 6 & 7**) of this dissertation represented the intervention studies evaluating the effectiveness of the individually defined targeted treatment approach. **Study 5** was designed as a randomized, single blind repeated measures study. After a period of 10 weeks usual care with no intervention, ten children with bilateral spastic CP were randomly assigned into an individually defined, targeted treatment program or a general program, followed by a cross-over. Evaluation was made using the GMFM-88 and three-dimensional gait analysis with outcome measures evaluating both the general progress of the child as well as the attainment of the individual goals of the child. As both programs seemed to be equally effective, the results of this study only indicated slightly favourable effects towards the individually defined program. No statistically significant differences between the outcomes of both programs could be found. However, limited to no changes were observed after the period of usual care. **Study 6** compared the effectiveness of the

individually defined approach to the effectiveness of a general therapy program in a larger, randomized controlled trial using the same outcome measures. Although this study included 60 interventions, the results did also not demonstrate statistically different effects on gait and gross motor function of these children between an individually defined treatment approach and a general program. However, again, analysis of the entire intervention group highlighted positive treatment effects for all children regardless of the type of program they were enrolled.

Hence, we hypothesized that supporting therapists by providing a predefined evidence-based intervention program may have contributed to the improved treatment outcome compared to the usual care physiotherapy. Consequently, **study 7** aimed to evaluate evidence-based practice behaviour in usual care physiotherapy in Flanders and to compare the effectiveness of the predefined physiotherapy programs based on evidence-based guidelines to the effects of a period of usual care. The results of this study indeed revealed that the predefined intervention programs provided better treatment results compared to the period of usual care, with significant differences in changes of gross motor function. Looking into the contents of usual care physiotherapy in Flanders showed that physiotherapy still mainly focuses on problems at impairment level and that exercises on strength and range of motion were generally characterized by insufficient dosing.

In **conclusion,** this doctoral project made a contribution to evidence-based physiotherapy of ambulant children with bilateral spastic CP by developing an individually defined, targeted treatment approach. Promising results are demonstrated regarding the effectiveness of the approach. However, the results need to be confirmed on larger groups, using programs of longer duration. Supporting physiotherapists by providing evidence-based guidelines and follow-up has beneficial effects on treatment outcome. Therefore, future research should take a next step to promote evidence-based behaviour in clinical practice.

Samenvatting

Cerebrale Parese (CP) is een overkoepelende term voor een groep blijvende stoornissen in de ontwikkeling van houding en beweging die kan worden toegeschreven aan niet-progressieve hersenafwijkingen in de vroege ontwikkeling van het kind. Deze kunnen leiden tot beperkingen in de activiteiten. Met een prevalentie van 2 à 4 per 1000, is het de meest voorkomende fysieke aandoening bij kinderen. CP manifesteert zich in een grote diversiteit aan neurologische stoornissen en klinische beelden. Ongeveer 46% van de kinderen met CP vertoont bilaterale spasticiteit, waarbij het onderste lidmaat over het algemeen meer aangedaan is dan het bovenste. Naast deze toegenomen spiertonus, vertonen deze kinderen ook vaak spierzwakte, beperkingen in selectiviteit, sensorische problemen en een verhoogd risico op spiercontracturen en botdeformiteiten. Deze problemen leiden dan vaak tot moeilijkheden bij het stappen en het uitvoeren van grofmotorische activiteiten, waardoor zij beperkt zijn in deelname aan sociale en schoolse activiteiten. Daarenboven wordt de problematiek van CP nog bemoeilijkt door het progressieve karakter van het klinisch beeld doorheen de jaren.
De complexiteit van de problemen van kinderen met CP vereist een multidisciplinaire aanpak. Daarbij speelt de kinesitherapeut een belangrijke rol. De kinesitherapeutische behandeling van deze kinderen vraagt om een complexe balans tussen het optimaliseren van de motorische ontwikkeling, het stimuleren van functionaliteit en onafhankelijkheid en de preventie van secundaire problemen die deelname aan dagdagelijkse activiteiten kunnen beperken.

De meeste kinesitherapeutische interventiestudies bij kinderen met CP evalueren de effectiviteit van individuele technieken. Hoewel de resultaten van deze studies veelbelovend zijn, is er nog steeds geen evidentie over de effectiviteit van integratieve programma's die een combinatie maken van verschillende technieken. De heterogeniteit in de pathologie van CP doet echter vermoeden dat een totaalaanpak, bestaande uit verschillende technieken en gericht op de specifieke problemen van het kind, meer tegemoet zou komen aan de individuele problemen van dit kind.
Dit doctoraatsonderzoek had als doel het ontwikkelen en evalueren van een evidence-based, individueel aangepaste behandelingsstrategie voor stappende kinderen met bilaterale spastische CP.

In **deel 1** van dit onderzoek **(hoofdstuk 1 & 2),** werd een systematisch literatuuronderzoek uitgevoerd, waarin de effecten van de individuele kinesitherapeutische technieken en concepten bij de behandeling van problemen in de onderste ledematen bij kinderen met CP werden nagegaan. De evidentie van deze technieken werd geëvalueerd en de effecten werden gedifferentieerd op de verschillende niveaus van het ICF. Er werden evidentietabellen gemaakt die de systematische keuze van bepaalde technieken om een specifieke doelstelling te kunnen bereiken kunnen vergemakkelijken. In het algemeen werden beperkte interactie-effecten gevonden tussen de verschillende niveaus van het ICF. Interventies die gericht zijn op het herstellen van problemen op het niveau van lichaamsstructuur- en functie hadden meestal alleen effect op dit niveau en vertoonden geen over-flow effecten naar het activiteitenniveau. Omgekeerd kon geen evidentie worden gevonden dat technieken die gericht zijn op het verbeteren van specifieke functionele activiteiten effectief zijn om problemen op lichaamsstructuur en functie te verbeteren. Een belangrijke klinische boodschap die uit dit onderzoek kon afgeleid worden, hield in dat een adequaat kinesitherapeutisch behandelplan best

vertrekt vanuit een totaalevaluatie van het kind, waarbij, bij voorkeur,alle niveaus van het ICF geëvalueerd worden.

Deel 2 (hoofdstuk 3) van deze studie had als doelstelling het ontwikkelen van een strategie die het klinisch redeneren van de pediatrische kinesitherapeut kan ondersteunen bij het selecteren van specifieke doelstellingen. Het framework dat daarbij ontwikkeld werd steunde op twee gekende, complementaire structuren, met name, het International Classification of Functioning, Disability and Health (ICF) en het Hypothesis-Oriented Algorythm for Clinicians (HOAC-II). Het ICF kon daarbij gebruikt worden als een perfecte structuur om de onderzoeksresultaten logisch en overzichtelijk op te lijsten. Het HOAC-II gaf ons een gestructureerde, strategische aanpak om het klinisch redeneren te ondersteunen. Op deze manier kon het ontwikkelde klinisch redeneermodel de kinesitherapeut ondersteunen. Hierbij kon op een correcte manier de lange probleemlijst van een kind gereduceerd worden tot een verkorte lijst van hoofdproblemen. Binnen dit model werden de resultaten van de drie-dimensionele ganganalyse gebruikt om een beter inzicht te krijgen in de complexe interacties tussen de verschillende problemen op de verschillende niveaus. Het klinisch redeneermodel kon gevalideerd worden bij een groep van 22 therapeuten, die ieder afzonderlijk de hoofdproblemen en behandeldoelen van 8 kinderen met bilaterale CP identificeerden. Clusteranalyse toonde een logische relatie tussen de selectie van hoofdproblemen en behandeldoelen. Dit bevestigde dat ons klinisch redeneermodel een valide structuur was dat logisch en gestructureerd klinisch redeneren kan faciliteren. Bovendien zagen we een hoge overeenkomst tussen de verschillende therapeuten bij het identificeren van hoofdproblemen. Bij het selecteren van specifieke doelstellingen was de overeenkomst tussen de therapeuten lager. Bovendien konden de therapeuten zich in 29% van de gevallen niet beperken tot een selectie van maximaal 8 behandeldoelen. De meerderheid van de geselecteerd behandeldoelen was gericht op spierkracht, spierlengte- en gewrichtsmobiliteit en items uit de Gross Motor Function Measure-88.

In een volgende stap werd de kennis uit de evidentietabellen en het gevalideerde klinisch redeneermodel gebruikt om een individuele en doelgerichte therapie-aanpak te ontwikkelen. Het klinisch redeneermodel kon gebruikt worden om de individuele problemen van een kind te identificeren. Vervolgens werden de specifieke behandeldoelen geselecteerd en besproken met de therapeut, de ouders en het kind zelf. De evidentietabellen uit het systematisch literatuuroverzicht konden worden aangewend om de correcte techniek te kiezen voor het bereiken van deze specifieke behandeldoelstellingen.

Om de effectiviteit van deze individuele en doelgerichte therapie-aanpak te evalueren, was een objectieve en onafhankelijke evaluatie van het stappen en de grofmotorische functie van de kinderen noodzakelijk. Daarbij werden drie-dimensionele ganganalyse en de gross motor function measure-88 (GMFM-88) geïdentificeerd als valide en betrouwbare metingen om de gang en grove motoriek van deze kinderen te evalueren. Drie-dimensionele ganganalyse kon betrouwbaar en onafhankelijk worden uitgevoerd in het laboratorium voor klinische bewegingsanalyse van het U.Z.Pellenberg. Om de GMFM-88 te kunnen gebruiken in een geblindeerd onderzoek, moest de betrouwbaarheid en validiteit van de GMFM-88 scores via videoregistratie worden nagegaan. In **deel 3 (hoofdstuk 4)** van

deze doctoraatsstudie werd de overeenkomst nagegaan tussen de live- en de video-scores van de GMFM-88. Hoewel deze resultaten een hoge overeenkomst tussen beide scores toonden was de intra-rater betrouwbaarheid tussen de live en video-score lager dan wanneer ze herhaald op video werd gescoord. De GMFM-88 kon dus betrouwbaar worden gescoord via videobeelden, maar men moet rekening houden met een systematische fout tussen de live- en videoscores. Dit kan gedaan worden door de aangepaste betrouwbaarheidsmaten te gebruiken.

Vervolgens werd in deel 4 (**hoofdstuk 5, 6 en 7**) van dit doctoraatsonderzoek de effectiviteit van een individuele en doelgerichte behandelstrategie geëvalueerd. **Studie 5** was daarbij opgezet als een gerandomiseerd, blinde studie met herhaalde metingen. Na een registratieperiode van 10 weken (zonder interventie), werden tien kinderen met bilaterale spastische CP via een gerandomiseerde procedure verdeeld over twee groepen: de helft van de kinderen kreeg een 10 weken durend individueel aangepast doelgericht programma, de andere helft kreeg een algemeen, aspecifiek oefenprogramma. Na dit programma werden een cross-over doorgevoerd: de kinderen die in een eerste fase een individueel programma hadden gevolgd kregen nu een algemeen programma en omgekeerd.

Voor en na de programma's werden de kinderen geëvalueerd door middel van een drie-dimensionele ganganalyse en de GMFM-88. De geselecteerde outcome parameters evalueerden daarbij zowel de algemene progressie van het kind als het al dan niet behalen van individuele doelstellingen. De resultaten van deze studie toonden slechts kleine, niet-significante verschillen aan tussen de effecten van beide programma's. De verschillen waren wel in het voordeel van het individuele programma. Weinig therapie-effecten konden echter geregistreerd worden na de registratieperiode.

Studie 6 vergeleek de effecten van een individueel aangepast doelgericht therapieprogramma met de effecten van een algemeen programma door middel van een groter, gerandomiseerd gecontroleerd onderzoek met dezelfde outcome parameters. Hoewel in deze studie 60 interventies werden gebruikt, toonden ook deze resultaten geen statistisch significante verschillen op de gang en de grove motoriek van de kinderen tussen de effecten van beide programma's. Analyse van de resultaten van de totale interventiegroep toonde echter wel positieve behandeleffecten aan voor alle kinderen, onafhankelijk van het type programma.

Bijgevolg stelden we de hypothese dat het ondersteunen van de therapeuten door een gestructureerd evidence-based programma aan te bieden kan bijgedragen tot de verschillen in outcome tussen de programma's en de registratieperiode. **Studie 7** had daarom als doel om het evidence-based therapiegedrag in de reguliere kinderpraktijk in Vlaanderen te evalueren en de effectiviteit hiervan te vergelijken met de effecten van de gestructureerde programma's. De resultaten van deze studie bevestigden onze hypothese. Er werd een significant verbetering in de grove motoriek vastgesteld bij deze kinderen na de interventie, die niet vastgesteld werd na de registratieperiode. Inhoudelijk studie van de therapie tijdens de registratieperiode toonde aan dat de therapeuten zich spontaan meestal toelegden op problemen op stoornisniveau en dat er vaak onvoldoende gedoseerd en opgebouwd werd bij het uitvoeren van deze oefeningen.

Samenvattend kunnen we stellen dat dit doctoraatsonderzoek een bijdrage heeft geleverd tot een verbeterd inzicht in evidence-based kinesitherapie bij stappende kinderen met bilaterale spastische CP. De effecten van de individueel aangepaste en doelgerichte behandelstrategie, die ontwikkeld werd in dit onderzoek, lijken veelbelovend. De resultaten moeten echter bevestigd worden op grotere groepen en moeten gebruik maken van programma's met een langere duur. Het ondersteunen van therapeuten in de klinische praktijk door hen intensief op te volgen en evidence-based richtlijnen mee te geven had positieve effecten op de outcome van de behandeling. Toekomstig onderzoek richt zich nu best op een volgende stap, met name op het promoten van evidence-based therapiegedrag in de klinische praktijk.

Appendices

Table A. Overview of the studies using stretching – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs.contr	ICF	LE (CS)
2003, Fragala[11]	prospective cross over	7	-	4y2mo-18y2mo	GMFCS V	Phase B: intervention phase with PT+ passive stretching Phase A: non-intervention phase	20	6	1,5	pROM hip (goniometer)	=	↓		I	IV (9/14)
	SSRD *ABAB*					A (4 weeks)- B (14 weeks) – A (2 weeks) -B (6 weeks) stretching 3*, 40-60sec,30min				pROM knee (goniometer)	=	↓		I	
2008, Khalili[12]	Within patient-control	11 (22 legs)	11	X=13y SD=1y	Diplegia Knee flexor spasticity	Exp (one leg): 30min ES Qcps (30 Hz; pulse width 0,4msec,4s/on 4sec off, ramp 0,5sec) + stretching hamstrings	4	-	5	MAS hamstrings pROM knee ext (goniometer)			↓ ↑	I I	II (5/7)
	Smaller RCT				Non-ambulant	Contr: contralateral leg from same child: stretching									
2008, Lee[13]	randomized	29	-	4-13y	dystonia	4 sessions, random order: A 5*10 sec stretching, B 5*30 sec stretching, C hotpack+5*10sec stretching, D hotpack+5*30sec stretching	0,5	-	7	act hamstrings stretch (EMG)			↓ (BD vs AC)	I	III (9/14)
	SSRD Cross-over				hypertonia	24h interval				extensibility hamstrings			↑ (AB vs CD)	I	
2006, Pin[14]	*syst review*	7 studies	-	-	-	Medline, CINAHL, PsychINFO, Embase, Cochrane Library, PEDRO ...-April 2006 keywords: cerebral palsy, muscle spasticity, stretching, physical therapy, range of movement				AACPDM guidelines Pedro scale				-	II (7)
2008, Wiart[15]	*syst review*	7 studies	-	-	-	Cinahl, Embase,Medline, PsycINFO, Scopus keywords: cerebral palsy, range of motion, stretching, contracture, positioning …-2007				level of evidence AACPDM guidelines				-	II (7)

Abbreviations:

N: Number; Exp: Experimental group period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE Level of Evidence, CS Conduct Score; y: years; mo: months; GMFCS: Gross Motor Function Classification; PT: Physical Therapy; sec: seconds, min: minutes; pROM: passive range of motion; X: Mean; SD: Standard Deviation; HZ: Hertz; ES: Electrical Stimulation; Qceps: m.Quadriceps Femoris; MAS Modified Ashworth Scale; Ext: extension; h: hours; act: activity; syst review: systematic review; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine; SSRD: Single Subject Research Design

Table B. Overview of the studies using massage – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs.contr	ICF	LE (CS)
2005, Hernandez-Reif[16]	RCT	10	10	Exp: X=29mo SD=8mo	ambulant	Exp: massage, 30 minutes	12	-	2	muscle tone flexors and extensors (MAS,ALT)	↓,↓	=,↓		I	**II (3/10)**
					nonambulant	Contr: reading				pROM hip abductors and extensors (goniometer)	↑	=		I	
				Contr:X=33mo SD=10mo	spastic										
	smaller RCT				ataxia					Developmental Programming for Infants And Young Children	↑	↑		A	
					athetosis										
										Videotaped Interactions	↑	↑		A/P	
2007, Macgregor[17]	prospective intervention	5	-	12-15y	spastic diplegia	massage therapy calf muscles, 14 min (stretching transversely)	5	-	2	pROM ankle dorsiflexion (goniometer)	=			I	**IV (3/10)**
										gross motor function (GMFM)	=			A	
	Case series					+slow passive stretching				stretch reflexes (EMG)	=			I	
2007, Barlow[18]	prospective intervention	70 parents 67 children	-	not reported	not reported	Training and Support Programme for parents	8	17,3	not specified	psychological well-being	↑			I	**IV (5/10)**
										Satisfaction with Life Scale	↑			I	
						(providing parents the basic skills in massage, 8 sessions)				Perceived stress scales	↓			E	
	case series									Parents' and child selfefficacy scale	↑			E	
										parental health status	↑			E	
										Child functioning	↑			I/A/P	
2010, Powell[19]	prospective intervention	43	-	8-15y	not reported	Training and Support Programme for parents	8	17,3	not specified	enjoyment (qualitative interviews)	↑			P	**IV (2/10)**
	Case series					(8 weekly sessions, 1h)									

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS Conduct score; y: years; mo: months; GMFCS: Gross Motor Function Classification; PT: Physical Therapy; sec: seconds, min: minutes; pROM: passive range of motion; X: Mean; SD: Standard Deviation; GMFM: Gross Motor Function Measure; h: hours

Table C. Overview of the studies using Threshold Electrical Stimulation (TES) – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method				Results					
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (* /week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
2001, Sommerfelt[20]	*RCT* cross-over matched groups ***smaller RCT***	6	6	4-12y X=8y8mo	diplegia GMFCS II-III	Exp: TES year 1 (40Hz, <10mA, 300µS, 5h/night, 6 nights)+PT; usual PT year 2 Contr: TES year 2 (40Hz, <10MA, 300µS, 5h/night, 6 nights)+PT; usual PT year 1	52	52	6	muscle strength (MMT) ankle ROM (goniometer) deep tendon reflexes speed of standardized movements gross and fine motor function (PDMS) walking speed (6 min & 6mwalk test) Parent subjective feeling (questionnaire)			= = = = = = ↑	I I I I A A I	II (5/7)
2002, Dali[21]	*RCT* Multi-centre ***smaller RCT***	36	21	5-18y X=10y11mo	hemi (25) diplegia (32) walking	Exp: TES, 1-5µA, 35Hz, 0,46µA/mm², Qceps and Tib Ant, 6h/night Contr: placebo (inactive stimulators) ! All children continued usual PT program	52	-	6	Gross motor function (set tests) ROM legs and arms (goniometer) spasticity legs and arms (MAS) cross sectional area Qceps/ Tib Ant (CT) parent questionnaire on motor skills			= = = = =	A I I I A	II (5/7)
2004, Mäenpää [22]	Pro-spective ***case-series***	17	-	3,8 - 8,9y X=6,4y	hemiplegia (11) diplegia (6)	TES of Tib Ant,1,8 times/week; 20-60min; 10-20Hz, 4-20mA, 300µsec, on/off 1/1 During ordinary PT sessions	4,3	39	1,8	active dorsiflexion, toe flex/ext, in/eversion aROM and pROM dorsiflexion standing on one foot and hopping	↑ ↑ ↑			I I A	IV (3/7)

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention group or period; Vs: Versus; ICF: International Classification of Functioning; LE Level of Evidence; CS Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFCS: Gross Motor Function Classification; TES: Threshold Electrical Stimulation; HZ: Hertz; h: hours; MMT: Manual Muscle Testing; ROM: Range of Motion; PDMS: Peabody Developmental Motor Scales m: meters; Qcps: m.Quadriceps Femoris; Tib Ant: m.Tibialis Anterior; MAS:Modified Ashworth Scale; CT: Computerized Axial Tomography; flex: flexion; ext: extension; aROM: active Range of Motion; pROM: passive Range of Motion

Table D. Overview of the studies using Neuromuscular Electrical Stimulation (NMES) – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
2001, Park[23]	*RCT* ***smaller RCT***	14	12	8-16y Exp: X=16,6y, SD=4.4y Contr: X=12,5y, SD=3.7y	spastic diplegia	Exp: PT (NDT) + NMES abdomen +posterior back muscles; 30 min/day; 25-30mA, 250µsec pulse width, 35Hz; 10sec on/12 sec off Contr: PT (NDT) only	6	-	6	Cobb (Xray spine) kyphotic angle (Xray) Gross Motor Function (GMFM)	↓ ↓ ↑	= ↓ ↑	= ↓ ↑	I I A	II (3/7)
2002; Detrembleur[24]	*RCT* ***smaller RCT***	6	6	4,75y-6y X=5y	diplegia (3) hemi (9)	Exp: BTX-A + NMES calf muscles (20Hz, 0,2msec, 50-90mA, 30min, 6*/day, 3days followed by PT) Contr: BTX-A + no adjuvant NMES, PT 2-3*/week post BTX-A PT:strength ankle dorsiflexors, stretch calf muscles and hamstrings, gait rehabilitation	0,5	26	42	gait (3DGA +EMG, Physicians Rating Scale) spasticity triceps surae (MAS) measurement of muscle stiffness ROM ankle (goniometer)			↑ = = =	A I I I	II (6/7)
2003, van der Linden[25]	*RCT* matched groups ***smaller RCT***	11	11	5-14y X=8y6mo SD=2y9mo	diplegia (14) hemi (7) quadri (1) independent walkers	Exp: NMES Glut Max, 1h/day, 10 Hz, 75µs (week 1); 2*30 min (week 2); 1 h 30Hz, 100 µsec (week 3-6) Contr: no electrical stimulation, usual PT ! All children continued usual PT program	8	-	6	Passive hip rotation (goniometer) gait (3DGA) gross motor function (GMFM) strength Glut Max (myometer)			= = = =	I A A I	II (4/7))
2004, Maenpaa[26]	Intervention prospective ***SSRD AB***	12	-	4,5-16y	hemiplegia moderate myocontracture triceps	Phase 1: Baseline (no intervention) Phase 2:NMES gastroc, 300 µA, 30 Hz, 5*/week, 1h	4	-	5	a+pROM dorsiflexion (goniometer) ROM popliteal angle (goniometer)	↑ ↑	= =		I I	V (7/14)

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFCS: Gross Motor Function Classification System; PT: physical therapy; NDT: neurodevelopmental treatment; NMES: Neuromuscular Electrical Stimulation; HZ: Hertz; GMFM: Gross Motor Function Measure; BTX-A: botulinum toxin type A; 3DGA: threedimensional gait analysis; EMG: electromyography; MAS: Modified Ashworth Scale; ROM: Range of Motion; Glut Max: m Gluteus Maximus; h: hours; a ROM: active Range of Motion; pROM: passive Range of Motion; MVC: maximum voluntary contraction; triceps s: mm.Triceps Surae; gastroc: m.Gastrocnemius; MRI: Magnetic Resonance Imaging; Unilat: unilateral; CMAP: Compound Muscle Action Potential; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine; SSRD: Single Subject Research Design

Table D (continued). Overview of the studies using Neuromuscular Electrical Stimulation– subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
2007, Stackhouse[27]	*RCT*	6	5	8 - 12y	spastic diplegia	Exp: NMES percutaneous; pulse duration 5-200 µs, amplitude 25 mA, pulse freq 50pps, 15 min	12	-	3	MVC Qceps (dynamometer)	↑	↑	↑	I	II (4/7)
	smaller RCT			Exp:X=10y7mo	GMFCS II-III					MVC Triceps S (dynamometer)	=	=	↑	I	
				Contr:X=10y5mo						walking speed (3DGA)	↑	=	=	A	
						Contr: 15 MVC				Qcps cross sectional area (MRI)	=	=	↑	I	
						Qcps and triceps s				Triceps S cross sectional area (MRI)	=	=	=	I	
2007, Kang[28]	*RCT*	7	11	16m-10y	spastic diplegia	Exp: BTX-A + ES gastroc (40 Hz, 0,3 msec, 10-25mA, 30 min)	13	-	2	gait (Physicians Rating Scale)	↑	=		A	II (5/7)
	smaller RCT			X=45mo	GMFCS I-IV					Spasticity (MAS)	↓	↓		I	
						Contr: BTX-A gastroc				pROM ankle and knee (goniometer)	↑	↑		I	
						! PT for all children, 2*/week									
2008, Khalili[12]	*RCT*	11	11	X=13y	diplegia	Exp (one leg): 30min NMES Qcps (30 Hz; pulse width 0,4ms,4s/on 4soff, ramp 0,5s) + stretching hamstrings	4	-	5	spasticity hamstrings (MAS)			↓	I	II (5/7)
	smaller RCT	(22 legs)		SD=1y	non-ambulant knee-flexor spasticity					pROM knee ext (goniometer)			↑	I	
						Contr (contralateral leg): 5*/week stretching									
2008, Rha[29]	*RCT*	11	12	X=46 mo	diplegia (18)	NMES post BTX-A injection	1	4,3	6	CMAP gastroc (EMG)	↓	↓		I	II (3/7)
	smaller RCT			SD=18,1mo	quadriplegia (5) GMFCS I-IV	Unilat: ES 25Hz(11 children) or 4Hz (12 children) m.Gastrocnemius				spasticity m.Gastrocnemius (MAS)	↓	↓		I	
						Unilat: Sham stimulation (7 days, 30 min/day)									
						+ 6*week usual PT									
2006, Kerr[30]	*RCT*	38	22	5-16y	diplegia (55)	Exp 1(n=18):NMES, 1h/day, 35Hz,300m	-	-	5	peak torque Qcps (isokinetic dynamometer)	=	=	=	I	II (7/7)
	placebo			X=11y	quadriplegia (1)	Exp 2 (n=20): TES, 8h/d, 5d/week, 35Hz, 300 msec, <10mA				gross motor function (GMFM)	=	=	=	A	
					distonia (1)					Lifestyle assessment questionnaire	↑	=	↑	P	
	smaller RCT			SD=3y6mo	not class (2) ambulant	Contr (n=22): placebo									
2004, Kerr[31]	***Syst Review***	18 studies	-	-		Pedro, Cinhal, Medline Amed using 'electrical stimulation and CP'	-	-	-	AACPDM levels of evidence	-	-	-		II (8)
	RCT's														

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFCS: Gross Motor Function Classification System; PT: physical therapy; NDT: neurodevelopmental treatment; NMES: Neuromuscular Electrical Stimulation; HZ: Hertz; GMFM: Gross Motor Function Measure; BTX-A: botulinum toxin type A; 3DGA: threedimensional gait analysis; EMG: electromyography; MAS: Modified Ashworth Scale; ROM: Range of Motion; Glut Max: m Gluteus Maximus; h: hours; a ROM: active Range of Motion; pROM: passive Range of Motion; MVC: maximum voluntary contraction; triceps s: mm.Triceps Surae; gastroc: m.Gastrocnemius; MRI: Magnetic Resonance Imaging; Unilat: unilateral; CMAP: Compound Muscle Action Potential; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine

Table E. Overview of the studies using isotonic strength training – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
1995a, Damiano[32]	pre-post	14	-	6-14y	spastic diplegia	training Qcps	6	-	3	MVC quadriceps (handheld dynamometer)	↑			I	IV (5/7)
	case series			X=9,1y	knee flexion contracture	ankle load 65% f 1RM, 4*5 repetitions				crouch gait (3DGA)	↓			A	
				SD=2,5y	ambulant	isotonic, concentric and excentric				stride lenght (3DGA)	↑			A	
1995b, Damiano[33]	prospective	14	25	6-14y	contr: non CP	exp: 65%max Qcps	6	-	3	strength m.Qcps (handheld dynamometer)	↑			I	IV (5/7)
	non randomized CT		no CP	X=9,1y	exp: spastic diplegia	contr (NL): 65% max Qcps									
				SD=2,5y											
1998, Damiano[34]	pre-post	11	-	6-12y	diplegia (6)	weakest lower limb muscles	6	-	3	MVC (isometric, handheld dynamometer), 8 muscles	↑			I	IV (5/7)
	prospective			X=8,,81y	hemiplegia (5)	velcro attached free-weights, 65% of max isometric strength value				gross motor function (GMFM)	↑			A	
	case series			SD=2,32y	limited community ambulators					gait (3DGA)	↑			A	
						4*5 repetitions				energy expenditure (heart telemetry)	=			I	
2004, Johnson[35]	case-series	5	-	3y10mo-9y11mo	diplegia (1)	2*/day, concentric and eccentric	3	3	14	Gait (video and footprint analysis)	=			A	IV (2/7)
	AB-design			X=6,06y	hemiplegia (3)	trampoline jumping, slope walking, toe raises, sit-to-stand, stairclimbing				ROM ankle during midstance (goniometer and markers)	=			I	
					ataxia (2)					strength plantar flexors (sphygomometer)	=			I	
2004, Eagleton[36]	pre-post	7	-	12-20y	not reported	training program (school gym or local fitness centre)	6	-	3	gait velocity, cadence, step length (10m walk test)	↑			A	IV (2/7)
	prospective				independently ambulant										
	case series					free weights and theraband exercises for trunk, hip, knee and ankle flexors and extensors, hip abductors				3 minute walk test	↑			A	
										energy expenditure index (heart rate pre-and post walking test)	↓			I	
						40-60 min, 8 to 10 repetitions at 80% 1 RM									

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFCS: Gross Motor Function Classification System; Qceps: M.Quadriceps Femoris; RM: Repetition Maximum; MVC: Maximum Voluntary Contraction; 3DGA: threedimensional gait analysis; GMFM: Gross Motor Function Measure; ROM: Range of Motion; min: minutes; MAS: Modified Ashworth Scale; MMT: manual muscle testing, GFAQ: Gilette Functional Assessment Questionnaire; FMS: Functional Mobility Scale; MMT: Manual Muscle Testing; TUG: Timed Up and Go; pROM: passive Range of Motion ; NDT: Neurodevelopmental Treatment

Table E. (continued) Overview of the studies using isotonic strength training – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
2004, Dodd[37]	RCT ***smaller RCT***	10	7	8-16y X=12,1y SD=2,5y	spastic diplegia	Exp: home based, hip extensor, ankle plantar flexor, knee extensor (heel rises, half squats, step-ups) Contr: normal daily activities	6	12	3	self-concept perception of scholastic competence perception of social acceptance perception of athletic competence			= ↓ ↓ ↓	I I I I	II (5/7)
2005, Morton[38]	repeated measures prospective ***SSRD AB***	8	-	6y10mo-11y2mo X=8y5m	hypertone GMFCS III	progressive strengthening programme, hamstrings and Qcps free-weight(65% of max isometric strength)	6	4	3	muscle strength Qceps and hamstrings (hand-held dynamometer) resistance to passive stretch (myometer) motor development (GMFM) 10m timed walking test and walking speed	↑ ↓ ↑ ↑			I I A A	IV (5/7)
2006, Unger[39]	RCT ***smaller RCT***	21	10	13-18y X exp=15,9y X contr=16,28y	hemi (16) diplegia (15) independently ambulant	Exp: circuit training, free weights, 1-3*/week,40-60min; individually designed programmes, 8-12 exercises Contr: no additional intervention	8	-	2	measure of crouch (3DGA) economy of movement (3DGA) perception of functional competence (Self Perception Questionnaire) perception of body image			↑ ↑ = ↑	I/A I I	II (5/7)
2008, Eek[40]	pre-postdesign ***Case series***	16	-	X=12y6mo range 9y4mo-15y4mo	GMFCS I - II spastic diplegia	free weights, rubber bands, body weight, 3*10 repetitions (easy, medium and heavy)	8	-	3	muscle strength hip, knee and ankle (handheld dynamometer) gross motor function (GMFM) ROM hip, knee, ankle (goniometer) gait kinematics and kinetics (3DGA) spasticity hip adductors, hamstrings, plantar fl and rectus femoris (MAS)	↑ ↑ ↑ ↑ =			I A I A I	IV (4/7)
2008, Lee[41]	RCT ***smaller RCT***	9	8	Exp: X=6,3y, SD=2,1y Contr: X=6,3y, SD=2,9y	spastic diplegia (9) spastic hemiplegia (8) GMFCS II-III	Exp:strengthening program, 60 min (warm up, functional strengthening exercises, isotonic exercises using weight cuffs, 2-10 repetitions) Contr : usual care (NDT, ROM exercises, gait)	5	6	3	muscle tone hip and knee (MAS) gross motor function (GMFM) strength hip, knee, ankle (MMT) gait (3DGA) Lateral step-up, squat to stand			= ↑ ↑ ↑ ↑	I A I I/A A	II (4/7)
2009, McNee[42]	pre-postdesign ***Case series***	13	-	6y11mo-16y11mo X=10y11mo SD=3y	diplegia (8) hemiplegia (5) GMFCS I - III	plantar flexion strengthening (thera band and heel rises)	10	13	4	muscle volume m.Gastrocnemius (3D US) gait (3DGA) gross motor function (GFAQ, FMS, TUG) unilateral heel rises pROM ankle (goniometer)	↑ = = ↑ =			I I/A A I I	IV (4/7)

Abbreviations:N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFCS: Gross Motor Function Classification System; Qceps: M.Quadriceps Femoris; RM: Repetition Maximum; MVC: Maximum Voluntary Contraction; 3DGA: threedimensional gait analysis; GMFM: Gross Motor Function Measure; ROM: Range of Motion; min: minutes; MAS: Modified Ashworth Scale; MMT: manual muscle testing, GFAQ: Gilette Functional Assessment Questionnaire; FMS: Functional Mobility Scale; MMT: Manual Muscle Testing; TUG: Timed Up and Go; pROM: passive Range of Motion ; NDT: Neurodevelopmental Treatment

Table F. Overview of the studies using functional strength training – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
2003, Blundell[43]	prospective repeated mesures ***SSRD ABA***	8	-	X = 6,3y range 4-8y SD=1,3y	diplegia (7) quadriplegia (1) ambulatory	Phase 1: 2 weeks follow-up Phase 2: one hour circuit training (functional strength : treadmill, steps, sit to stand, leg pre Phase 3: 8 weeks follow-up ss)	4	8	2	strength hip, knee and ankle flexors and extensors (dynamometer) Motor Assessment Scale:sit-to-stand, lateral step-up Minimum Chair Height Test walking speed (2 minutes walk test) walking speed (timed 10 m test)	↑ ↑ ↑ = ↑			**I** **A**	**IV (9/14)**
2003, Dodd[44]	RCT ***smaller RCT***	11	10	X=13y1mo SD=3y1mo range 8-18y	spastic diplegia GMFCS I -III	home based training Contr: usual care Exp: LL training : heel squats, heel raises and step-ups (3 sets of 8-12 repetitions, 20-30 min)	6	12	2	strength ankle plantar flexors, knee + hip extensors, (hand-held dynamometer) gross motor function (GMFM) timed-stair test self-selected walking speed (10 metres walking test)			↑ = = =	**I** **A**	**II (7/7)**
2003, McBurney[45]	prospective no control ***case series***	11	-	X=12y9mo SD 2y10mo range 8-18y	spastic diplegia GMFCS I-III	home based training, 8-10 repetitions each heel squat, heel raise and step-up (plantar flexors, knee extensors, hip extensors)	6	-	3	perception of body image perception of functional performance perception of social participation	↑ ↑ ↑			**I**	**IV (3/7)**
2007, Liao[46]	RCT ***smaller RCT***	10	10	range 5-12y Exp: X=85,6mo SD=20,8 Contr: X=91,3mo SD=17,5mo	spastic diplegia GMFCS I and II	Exp: regular PT + sit-to-stand exercises Contr: regular PT only max 25 repetitions, 1 session	6	-	3	gross motor function (GMFM) gait speed (timed 10 metres walking test) isometric strenght Qcps (Nicholas Manual Muscle tester) sit to stand Physiologic Cost Index (heart rate walking - rest/walking speed)			↑ = = ↑ ↑	**I** **A**	**II (6/7)**

Abbreviations

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; SSRD: Single Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; LL: Lower Limb; PT: Physical Therapy; percut: percutaneous; MVC: Maximum Voluntary Contraction; Qceps: m.Quadriceps Femoris; triceps s: m.Triceps Surae; MVIC: Maximum Voluntary Isometric Contraction, HR: Heart Rate; GMFM: Gross Motor Function Measure; GMAE: Gross Motor Activity Estimator; MRI: magnetic resonance imaging; HR: Heart Rate; QoL: Quality of Life; 3DGA: threedimensional gait analysis

Table G. Overview of the studies using isokinetic, isometric and mixed forms of strength training – subjects, interventions, evaluation, results, level of evidence and conduct scores

Author	Design	Subjects				Method				Results					
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
Isokinetic strength training															
1995, Mac Phail[47]	pre-post Prospective ***case series***	17	-	12-20y	quadriplegia (1) hemiplegia (9) diplegia (7) ambulant , without aids	min 45 minutes 3 submaximal warm-up efforts, 15 concentric and 15 eccentric MVC of knee extensors and flexors	8	13	3	peak torque knee flexors and extensors (dynamometer) spasticity (MAS + ankle clonus) gross motor function (GMFM) walking efficiency (Energy Expenditure Index, HR)	↑ = ↑ =			I A	IV (2/7)
2006, Engsberg[48]	RCT ***smaller RCT***	12	-	X=9,7 SD=3,3y	spastic diplegia GMFCS I-III	Exp 1(n=3): dorsiflexor group Exp 2 (n=3): plantar flexor group Exp 3 (n=3): dorsi+plantarflexor group Exp 4 (n=3): control group	12	13	3	gross motor function (GMAE-GMFM) pROM ankle (goniometer) strength ankle plantar and dorsiflexors (KinCom dynamometer) gait kinematics and gait speed (3DGA) spasticity (KinCom dynamometer) quality of life parents+children (Peds QOL)	=, ↑ = ↑ ↑,= ↓ ↑, =	= = = = = ↓		I A P E QoL	II (4/7)
Isometric strength training															
2007, Stackhouse[27]	RCT ***smaller RCT***	6	5	8 - 12y Exp:X=10y 7mo Contr:X=10y5mo	spastic diplegia GMFCS II-III	Contr: NMES percut, pulse duration 5-200 µs, amplitude 25 mA, pulse freq 50pps, 15 min Exp: volitional max effort contractions, 15 MVC Qcps and triceps s	12	-	3	MVIC Qcps (dynamometer) MVIC M.Triceps Surae (dynamometer) walking speed (3DGA) Qcps cross sectional area (MRI) Triceps S cross sectional area (MRI)	↑ = = = =	↑ = ↑ = =	↑ ↑ = ↑ =	I A	II (4/7)
Mixed strength training															
2001, Fowler[49]	Prospective ***non randomized CT***	24	12 no CP	7-17y X=11,4 SD =3y	Exp:spastic diplegia Contr: typically developing	isometric Qcps (kinCOM) isotonic Qcps (cuff weights) isokinetic Qcps (60°/sec) max 25 repetitions, 1 session	0,1	-	-	stretch reflexes pendulum test (spasticity KinCom; EMG + goniometer)	=			I	IV (4/7)

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; LL: Lower Limb; PT: Physical Therapy; percut: percutaneous; MVC: Maximum Voluntary Contraction; Qceps: m.Quadriceps Femoris; triceps s: m.Triceps Surae; MVIC: Maximum Voluntary Isometric Contraction, HR: Heart Rate; GMFM: Gross Motor Function Measure; GMAE: Gross Motor Activity Estimator; MRI: magnetic resonance imaging; HR: Heart Rate; QoL: Quality of Life; 3DGA: threedimensional gait analysis

Table H. Overview of systematic reviews evaluating strength training

Author	type	N	Intervention	Evaluation	LE (CS)
1997, Darrah[50]	syst review	7 studies	Medline, CINAHL, Eric, Psychinfo, Sport DISCUS keywords cerebral palsy, exercise, strength and physical training 1966-1997		II (7)
1998, Haney[51]	syst review	8 studies	search strategy not reported	-	**II (1)**
2002, Dodd[52]	syst review	23 studies	Medline, PubMed, EMBASE, CINAHL, Sports Discus DARE, Psychinfo, ERIC, AusportMed, AMI, Cochrane, Pedro; 1966-2000 keywords: exercise, strength and physical training	Pedro scale	**II (8)**
2008, Verschuren[53]	syst review	20 studies	Medline, PubMed, EMBASE, CINAHL, Sports Discus, Cochrane, PEDro, untill sept 2006 keywords:CP,exercise, strength, working capacity,(an)aerobic power, endurance, cardiorespiratory physical training	Pedro Scale Outcome on ICF	**II (8)**
2008, Mockford[54]	syst review	13 articles	Medline, AMED, CINAHL, Cochrance Library, EMBASE, PEDro, PsychInfo, SPORTdiscus ,,,-March 2007 keywords: CP, strength exercise, weight training and lifting, resisted exercise, resistance exercise, resisted training, resistance training	Amsterdam-maastricht List	**II (8)**
2009, Scianni[55]	syst review meta-analysis	6 RCT's	CINAHL, MEDLINE, EMBASE and Pedro no language restrictions, RCT's only children with spastic CP upto 20y	Pedro Meta-analysis ICF	**I (8)**

Abbreviations:

N: Number of studies, LE: Level of Evidence; CS: Conduct Scores; syst revie: systematic review, RCT; Randomized Controlled Trial; ICF: International Classification of Functioning

Table I. Overview of the studies using endurance and physical fitness training – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
1998, van den Berg - Emons[56]	RCT ***Smaller RCT***	10	10	X=9,2 y SD = 1,4y	spastic CP diplegia (16) tetraplegic (4) ambulant non-ambulant	Group 1:2*/week gymnastics + 4*/week physical training + usual PT Group 2: 2*/week gymnastics+usual PT sports programme=wheelchair driving, cycling, running, swimming, flying saucer exercises, mat exercises	39	-	2	Physical Activity Ratio (energy expenditure/sleeping meth rate) Fat mass (anthropometry) Peak aerobic power (ergometer) Anaerobic power (ergometer) Isokinetic muscle strength (Cybex II)	↑ = ↑ ↑ ↑	= ↑ = ↑ =	= = ↑ = =	A I I I I	II (3/7)
1999, Darrah[57]	Pro-spective ***Case series***	23	-	X=14.2y	hemiplegia (13) diplegia (5) quadriplegia (2) ataxia (2) dystonia (1) ambulant (without aids)	warm-up (10min), aerobic exercise (10-30min), strength training (30 min), stretching and cooling-down(20 min) group training	10	10	3	Energy Expenditure Index (EEI) and heart rate strength shoulders flexors, knee extensors, hip extensors and abductors (handheld dynamometer) flexibility(sit-and-reach+behind the back reach) Self Perception Profile fo Adolescents and Children	= ↑ ↑ ↑			I I I I	IV (3/7)
2002, Shinohara[58]	Pro-spective ***non-randomized CT***	6	5	Exp: X=14,6y SD=0,9y range 13,3-15,8y Contr X =14,2y SD=10,7y range 11,8-16,3y	ambulant non-ambulant	Exp: leg exercises, 20 minutes at AT point 8-20,6 weeks, 1,1 to 2,3 */week) Contr: arm cranking ergometer, 20 min, AT point 5-19,9 weeks at 1-2,3 times/week work rate 7 or 10 W per minute	13	-	1,8	Oxygen Uptake (respirometer/gasometer)	↑	=		I	IV (3/7)
2005, Schlough[59]	***ABAB*** ***SSRD***	3	-	17-20y	spastic CP hemiplegia (1) diplegia (2) GMFCS I and III	A1: no intervention B1: initial intervention: treadmill, stepper, elliptical machine A2: no intervention B2: additional 15 weeks, treadmill, stepper, elliptical machine	6 B1 6 weeks B2 7 weeks	2 A1 2 weeks A2 2 weeks	3	Energy Expenditure Index (EEI) muscle strength Qcps, hamstrings, ankle plantar and dorsiflexors (handheld dynamometer) gross motor function (GMFM) Self-Perception Profile for College Students (SPCS) Rate of Perceived Exertion (RPE)	not stats			I I A I I	IV (9/14)

Abbeviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; AT point: Anaerobic Thresshold Point; W: watt; Qceps: m.Quadriceps Femoris; RM: Repetition Maximum; GMFM: Gross Motor Function Measure; HRQoL: Health Related Quality of Life; CAPE: Children's Assessment of Participation and Enjoyment; MoVra: Mobiliteitsvragenlijst/mobility questionnaire; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine; SSRD: Single Subject Research Design

Table I (continued). Overview of the studies using endurance and physical fitness training – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Authors	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
2007, Verschuren[60]	RCT Multi-center ***Smaller RCT***	32	33	7-18y Exp X =11,6y SD 2,5 Contr 2 X=12,7y SD 2,7	GMFCS I-II unilateral (45) bilateral (23)	Exp: usual care + 45min circuit Contr: usual care Circuit training: 5 min warm-up, 25-35 min functional aerobic exercises, anaerobic exercises and muscle strengthening in circuit, 5 minutes cool-down	35	17,3	2	anaerobic capacity (power sprint test) aerobic capacity (10 min shuttle run test) strength lower extremities (30 sec RM) agility (10*5m sprint test) gross motor function (GMFM) participation (HRQoL, CAPE)			↑ ↑ ↑ ↑ ↑ ↑,↑	I I I I A A/P	II (5/7)
2007, Williams[61]	Pro-spective ***single subject*** ***ABA***	11	-	11-15y X=12y 7mo SD 1y 4mo	GMFCS IV-V spastic (8) dyskinetic (3) diplegia (1) quadriplegia (7)	Phase A: baseline, no intervention Phase B: training (75% of max endurance, 100% for speed/power) Phase C: follow-up, no intervention	6	6	3	gross motor function (GMFM) cycling ability (pedalling resistance, overload)	↑ ↑,↑	= =		A I/A	IV (10 /14)
2007, Uninithan[62]	RCT ***Smaller RCT***	7	6	X exp : 15,9y range 14-18y X contr=15,7y range 14-17y	spastic diplegia GMFCS II-III	Exp: 70min/session, aerobic interval training and strength (handweights, 20 repetitions UL and 4*10 LL+usual PT (2*/week NDT) Contr: usual PT NDT 2*/week, no additional training	12	-	3	gross motor function (GMFM) VO2 (open circuit spirometer) %VO2max (open circuit spirometer) VE peak	↑ ↓ ↓ ↑	= = = =	↑ ↓ ↑ ↑	A I I I	II (4/7)
2009, Gorter[63]	repeated measures ***SSRD***	13	-	8-13y	GMFCS I-II	A circuit with 4 stations of functional exercises on aerobic endurance, walking distance, walking velocity and ambulation 30 minutes, 60-70%Hrmax	9	11	2	max aerobic capacity (Bruce Test) walking distance and velocity (6 min run) functional mobility (Timed Up&Down Stairs) Ambulation Questionnaire (MoVra)	↑ ↑ ↑ ↑			A A A A	IV (4/14)
2008, Rogers[64]	***Syst Review***	13 studies	-	-	-	Medline, Embase, Cinahl, Pascal, Cochrane Library, CSA Neuroscience Abstracts, Pedro and Sports Discus 1960-2006 keywords: cerebral palsy, athetoid, ataxic, spastic diplegia, hemiplegia, quadriplegia, aerobic exercise, training, physical activity, aquatic/pool/water therapy, exercise training, continuous exercise				AACPDM guidelines				-	II (8)
2009, Verschuren[65]	***Syst Review***	20 studies	-	-	-	Medline, PubMed, EMBASE, CINAHL, Sports Discus, Cochrane, PEDro until september 2006 keywords cerebral palsy, exercise, strength, working capacity, aerobic/anaerobic power, endurance, cardiorespiratory physical training				AACPDM guidelines				-	II (8)

Abbreviations:N: Number; Exp: Experimental group or period; Contr: Control group or l period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE:Level of Evidence:; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; AT point: Anaerobic Thresshold Point; W: watt; Qceps: m.Quadriceps Femoris; RM: Repetition Maximum; GMFM: Gross Motor Function Measure; HRQoL: Health Related Quality of Life; CAPE: Children's Assessment of Participation and Enjoyment; MoVra: Mobiliteitsvragenlijst/mobility questionnaire; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine.

Table J. Overview of the studies using weight-bearing exercises – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
1999, Chad[66]	RCT	9	9	Exp:X=9y, SD=2,9y Contr: X=9y, SD=2.7y	dependent and independent walkers	Exp: weight bearing physical activity, 2*/week (2 months) and 3*/week (6 weeks), 60 min (20 min UL, 20 min LL, 20 min trunk, facilitation of normal movement with weight bearing) Contr: usual lifestyle habits	14	-	2,5	Bone Mineral Content proximal femur and femoral neck % Bone Mineral Content proximal femur and femoral neck (dual energy Xray absorptiometry)			↑ ↑	I I	II (2/7)
2000, Katz[67]	prospective intervention ***case series***	36	-	X=5y 4-7y	diplegia (14) hemi (15) ambulatory	Achilles tendon lengthening surgery + 2 weeks casting and early weight-bearing (after 2 days) and gait training	2	260	not reported	pROM ankle (goniometer) visual gait analysis (heel/toe contact)	* no stats			I I/A	IV (1/7)
2002, Gudjonsdottir[68]	prospective intervention ***smaller RCT***	2	2	4y5mo-5y11mo	non-ambulant	Phase 1(8 weeks): group 1 dynamic standing, group 2 static stander (5*/week, 30 min) Phase 2 (3 sessions): all children standing in the different standing frames	8	-	5	ROM hip and knee ext, ankle dorsiflex (goniometer) BMC femoral neck, great trochanter, intertrochanteric) (Dual energy x-ray absorptiometry behavioral characteristics (CRIB)	* no stats			I I I	II (3/7)
2004, Caulton[69]	RCT ***smaller RCT***	13	13	4,3y -10,8y	non-ambulant	Exp: 50% longer standing Contr: normal duration of standing varying from 14min to 365 min standing/week	39	-	7	vertebral tibial volumetric trabecular bone density (vTBMD) (quantitative computed tomography) proximal tibial volumetric trabecular bone density (vTBMD) (quantitative computed tomography)			↑ =	I I	II (5/7)
2009, Gibson[70]	prospective intervention ***SSRD***	5	-	5-9y X=7y2mo SD=1y4mo	non-ambulant	phase A: standing in a standing frame, 1h/day phase B: no intervention ABAB design	6	-	5	Popliteal angle (goniometer and angle finder) ADL (feedback form)	↑ =	↓ =	↑ =	I A	IV (9/14)
2009, Eisenberg[71]	prospective intervention ***Non-randomized CT***	11	11	Exp:X=6,2y, SD=2,1 Contr:X=6,7y, SD=1,6y	spastic quadriplegia GMFCS IV-V	Exp: Heart Walker Contr: passive standing programme	26	-	7	bowel activity (diary) functional performance (PEDI) bone mineral density (ultrasound) walking speed (2 min walking test)	↑ ↑ ↑ ↑	= ↑ ↑ =	↑ = = =	I A/P I A	III (4/7)
2007, Pin[72]	***syst review***		-	-	-	Medline, CINAHL, psychINFO, Embase, full Cochrane library, PEDro start - 2006 keywords: child, cerebral palsy, bone density, hip dysplasia, contracture, range of motion, stretching, muscle spasticity, bowel and urinary function, morale, communication, hand function, feeding			-	PEDro scale					II (7)

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE:Level of Evidence:; CS: Conduct Score; RCT: Randomized Controlled Design; SSRD: Signel Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; pROM: passive Range of Motion; BMC: Bone Mineral Content; ADL: Activities of Daily Living; CRIB: Carolina Record of Individual Behavior; PEDI: Pediatric Evaluation of Disability Inventory

Table K. Overview of the studies using balance training – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
1995, Myhr[73]	retrospective cohort	10	-	2,1y-5,8y X=3,6y	mild/moderate/ severe	5 y functional sitting position (pelvis forward, upper body anterior to the fulcrum, hip belt and abduction orthosis)	260		no specified	sitting position (Sitting Assessment Scale)	↑			I/A	IV (3/7)
	case series				spastic diplegia					head/trunk control	↑			I/A	
2003, Shumway-Cook[74]	prospective intervention	6	-	9y2mo-12y11mo	hemiplegia (2)	phase A: usual therapy only	0,8	1	7	area and time to stabilization from CoP (moving forceplate)			↓	I	III (9/14)
					diplegia (4)	phase B: balance training on moving platform (100 perturb/day for 5 days) + usual PT 30min-1h/week							↓	A	
	SSRD			X=9y2mo	GMFCS I and II					gross motor function (GMFM)			=	A	
	ABA			SD=2y		ABA design									
2005, Ledebt[75]	RCT	5	5	5-11y	hemiplegia	Group 1: balance training with visual feedback	6	4	3	CoP displacement standing	↓	=	=	I/A	II (4/7)
	smaller RCT					Group 2: control group				CoP displacement during dynamic standing	↓	=	↓	I/A	
										average step length asymmetry	↓	=	↓	A	
2005, Woollacott[76]	prospective intervention	6	-	X=9y4mo	hemiplegia (2)	5 days of intensive reactive balance training	0,8	4,3	7	muscle cocontractions (EMG):					IV (4/7)
					diplegia (4)	(100 perturbations/day on a moveable platform)				timing of muscle contraction	↓			I	
	case series				GMFCS I and II					distal-proximal muscle sequence	↑			I/A	
										agonist	↑			I	
										antagonist	↓			I	
2008, Bar-Haim[77]	RCT	10	10	8,9-12,9y X=9,2	GMFCS II-IV	Contr: structured intensive treatment Exp: structured intensive treatment + random perturbation	4,3		7	gross motor function (GMFM)	↑	=	=	A	II (4/7)
	smaller RCT					1 month, daily treatment of 1,5h, 20 sessions physio: stretching, functional weight-bearing, walking activities random perturbation: engine induced passive cycling, 10 min				Mechanical Efficiency during stair climbing (rate oxygen consumption)	=	↑	↑	I	
2005, Harris[78]	syst review	12 studies	-	-	-	search on Medline, CINAHL, EMBASE, PsychINFOR, SportDiscus, Cochrane Database of Syst Review, Cochrane Controlled Trial Register, PEDro, DARE, Dissertation Abstracts 1990-2004 Cerebral palsy, Balance, Posture, Postural Control			-	AACPDM guidelines					II (8)
	syst review														

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct score; RCT: Randomized Controlled Design; SSRD: Single Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; pROM: passive Range of Motion; CoP: Centre of Pressure; GMFM: Gross Motor Function Measure; EMG: Electroymyography; syst review: systematic review; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine

Table L. Overview of the studies using treadmill training – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/ week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
1997, Richards[79]	prospective intervention	4	-	1.7-2.3y	nonambulant	treadmill training+ NDT based PT	17,3	-	4	gross motor function (GMFM)	=			A	IV (3/7)
						treadmill on low speed (7cm/sec), progressive weight-bearing				gait (videographic test +3DGA)	=			I/A	
	case series									Supported Walker Ambulation Performance Scale	=			A	
2000, Schindl[80]	prospective intervention	10	-	6-18y	6 nonambulatory	Phase B : BWSTT , 25 min/session	13	-	3	gross motor function (GMFM)	↑	=		A	V (8/14)
				X=11,5y	4 req cont phys assistance	Body weight support at start: X=14% (0-40%) + usual PT				Functional ambulation category	↑	=		A	
	SSRD AB					Mean treadmill speed 0,23 m/s (start of the study) to 0,40 (end of the study)									
						Phase A: usual PT only (30 min, 2-3 sessions per week)									
2007, Cherng[81]	ABA or AAB	4	4	3,5-6,3y	GMFCS I-III	Exp:ABA	12	6	2,5	gross motor function (GMFM)	↑	=		A	II (9/14)
	SSRD			X=4y	spastic diplegia	Contr:AAB				time-and distance parameters (GaitRite)	↑	=		I/A	
						A=regular physio,NDT, 30 min/session									
						B=BWSTT, 20min/session+regular physio				muscle tone (MAS)	=	=		I	
										selective motor control (SMC)	=	=			
2007, Philips[82]	prospective intervention	6	-	6-14y	hemiplegia (4)	2*/day BWSTT	2	-	14	fMRI (3 tasks): (1) active ankle dorsiflexion (2) finger tapping of the uninvolved hand; and (3) active ankle dorsiflexion of the involved ankle	=			I	IV (3/7)
				X=10y5mo	diplegia (2)	Body support decreased from 30% initially to 0%									
					GMFCS I	Treadmill speeds ranged from 2.4 to 3.1kmph initially and increased to 3.7 to 5.0kmph with training.									
	case-series									walking speed	↑			A	
										distance walked for 6 minutes	=			A	
2007, Dodd[83]	prospective	7	7	5-14y	GMFCS III-IV	Exp: BWSTT (0.40 km/h upto 0,60km/h)	6	-	2	10 min walk test			=	A	II (4/7)
	matched			X=8y10m	athetoid quadri (6)	Untill child stopped or was tired (med 12min(start) to 21 min (end)				self-selected walking speed in 10 min			↑	A	
	non randomized CT			SD=2y6m	spastic quadri (6)										
					spastic diplegia (2)	Contr: usual PT (content not reported)									
2007, Begnoche[84]	prospective intervention	5	-	2,3-9,7y	quadriplegia (1)	2h/session treadmill+NDT	4	-	3,5	gross motor function (GMFM)	=			A	IV (3/7)
					diplegia (4)	15-35 min/session partial body weight treadmill training				functional performance (PEDI-FS)	=			A/P	
	case series				GMFCS I-IV					gait (pedography)	↑			I/A	
										walking speed (timed 10m walk test)	=			A	

Abbreviations: N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct score;RCT: Randomized Controlled Design; SSRD: Single Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; NDT: neurodevelopmental treatment; 3DGA: threedimensional gait analysis; GMFM: Gross Motor Function Measure; BWSTT: Body Weight Supported Treadmill Training; QoL: Quality of Life; PEDI: Pediatric Evaluation of Disability Inventory; syst review: systematic review; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine

Table L. (continued) Overview of the studies using treadmill training – subjects, interventions, evaluation, results, level of evidence and conduct scores.

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs contr	ICF	LE (CS)
2007, Provost[85]	prospective intervention ***case series***	6	-	6-14y	diplegia (4) hemiplegia (4) ambulatory GMFCS I	body weight supported treadmill training 2*/day 30 min	2	-	14	gross motor function (GMFM) Energy Expenditure 6min walking test Ten-Meter Walking Velocity Single Leg Balance Test	= ↓ ↑ ↑ =			**A** **I** **A** **A** **A**	**IV (4/7)**
2009, Dieruf[86]	prospective intervention *Case-series*	6	-	6-14y	GMFCS I diplegia (2) hemiplegia (4)	2*30min/day BWSTT (30 min: 10 min walking, with 5 min rest)	2	-	14	Quality of life (Ped QOL Inventory) fatigue (PedsQL multidisciplinary Fatique Scale)	= =			***QoL*** ***QoL***	**IV (3/7)**
2009, Hodapp[87]	prospective intervention ***case series***	7	-	5-15y X=9,7y	GMFCS I-III	treadmill training, without body weight support; 1,90km/h to 3,10km/h	1,5		7	soleus H reflex walking speed (ground and treadmill)	↓ ↑			**I** **A**	**IV (4/7)**
2009, Mattern-Baxter[88]	prospective intervention ***case series***	6	-	2,5-3.9y	GMFCS I - IV spastic diplegia (3) spastic quadri (1) hypotonia (1) dystonia (1)	2 individualized treadmill walks 1h, gait speed as fast as possible	4	4,3	3	gross motor function (GMFM) functional performance (PEDI) walking speed (6 minute walk test) walking speed (10m walk test)	↑ ↑ ↑ ↑			**A** **A/P** **A** **A**	**IV (5/7)**
2009, Willoughy[89]	***syst review***	5 studies	-	-	-	Cinahl, Cochrane, Pedro, ERIC, PsychINFO, AMED, PubMed, Ausport Medical and Sports Discus until july 2008 keywords: cerebral palsy, child, treadmill training				effect sizes					**II (9)**
2009, Mattern-Baxter[90]	***syst review***	10 studies	-	-	-	Academic Search Complete, Blackwell Synergy, Cochrane Library,Google Scholar, Health Source, Nursing Academic, PubMed,Science Direct,SCOPUS and SPORTDiscus keywords: adolescent, child, gait, physical endurance, fitness, spastic CP, treadmill training and walking; 1997 - 2008				Sackett's levels of evidence					**II (6)**
2009, Mutlu[91]	***syst review***	-	-	-	-	Medline, PubMed, Google, Embase, Ovid Medline, Galter Health Sciences Library, Pedro Cochrane, Cinahl, APTA 1950-2007 keywords: CP, treadmill training, PBWSTT, locomotor therapy, gait, walking, physial therapy methods				AACPDM guidelines					**II (8)**

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; NDT: neurodevelopmental treatment; 3DGA: threedimensional gait analysis; GMFM: Gross Motor Function Measure; BWSTT: Body Weight Supported Treadmill Training; QoL: Quality of Life; PEDI: Pediatric Evaluation of Disability Inventory; syst review: systematic review; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine

Table M. Overview of the studies using Neurodevelopmental Treatment of Bobath therapy – subjects, interventions, evaluation, results, level of evidence and conduct scores

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (CS)
1995, Karnish[21]	prospective intervention	3	-	4,14 and 6y	spastic quadri	Training 1: PT in an isolated therapy room	1,5	-	14	motor skill tasking (video):					**II (7/14)**
						Training 2: PT in a natural education setting				quality of performance	=	=	=	**A**	
	ABABAB					2 sessions/day, each of the two settings each day, 10 sessions				speed of completion	=	=	=	**A**	
	SSRD					inhibition/facilitation, transfer, standing balance, motor skill training						* no stats			
1997, Jonsdottir[22]	Prosp intervention	8	-	10-15y	spastic quadri	Phase A: 1 week no treatment (control phase	1	-	7	postural assessment scale (Bertoti):					**III (6/14)**
	Alternationg treatments					Phase B: 1 week NDT (daily, 35 min; focus on reaching) (exp)				postural control (Seated Postural Control Measure)	↑	=		**I/A**	
	SSRD ABAC					Phase C:1 week practice (repetition, no focus on quality) (control)				Total Displacement of the Head and Shoulder	↑	=		**I**	
1999, Trahan[23]	prospective intervention	50	-	12-79mo	quadri (24)	PT as NDT, 45min/session	35	-	2	gross motor function (GMFM)	↑			**A**	**IV (4/7)**
	Cases series				hemi (16)										
					diplegia (10)										
2000, Adams[24]	prospective intervention	40	-	X=6y	hemi (11)	6 weeks intensive NDT	6	-	2	Stride and step length, foot angle, base of support, cadence, velocity (pedograph)	↑			**I/A**	**IV (3/7)**
	Cases series			2,6y-10,2y	diplegia (18)	1h individually defined training									
					triplegia (3)										
					ataxia (5) athetoid (3)										
2001, Kerem[25]	prospective intervention	17	17	X group 1 =48,82mo	spastic diplegia	Exp: NDT +Johnstone Pressure Splints	13	-	5	spasticity (MAS)	↓	↓	↓	**I**	**III (3/7)**
	Non-randomized			X group 2= 47,52mo	moderate	Control: NDT				Somatosensory Evoked Potentials	↓	↓	↓	**I**	
										ROM (goniometer)	↑	↑	↑	**I**	

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct score; RCT: Randomized Controlled Design; SSRD: Single Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; NDT: Neurodevelopmental Treatment; GMFM: Gross Motor Function Measure; MAS: Modified Ashworth Scale; PEDI: Pediatric Evaluation of Disability Inventory; ROM: Range of Motion; syst review: systematic review; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine

Table M (continued). Studies using Neurodevelopmental Treatment or Bobath therapy – subjects, interventions, evaluation, results, level of evidence and conduct scores

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (CS)
2002, Knox[26]	repeated measures	15	-	X=7y4mo	quadriplegia (9)	Exp phase(phase B): NDT,75 min/session	6	18	3	self care skills/caregiver assistance (PEDI)	↑	=	↑	A/P	III (8/14)
	ABA			range 2-12y	diplegia (4)										
	SSRD				athetoid (1)	Control phase (phase A): usual therapy				gross motor function (GMFM)	↑	=	↑	A	
					ataxia (1)					Parent Questionnaire (individual goals)	↑	=	↑	A	
					GMFCS I - V										
2002, Trahan[27]	Pro-spective	5	-	X=33mo	GMFCS IV-V	Phase A: baseline, conv PT, 2*/week, 8 to 20 weeks	4	-	4	gross motor function (GMFM)	↑	↑	↑	A	II (10/14)
	SSRD ABAB			range10-37mo	spastic quadriplegia	Phase B:PT4 weeks, 4*/week altered with 8 weeks no PT(total 24 weeks, ABAB design)									
2004, Tsorlakis[28]	RCT ***Smaller RCT***	17	17	X=7y3mo range 3-14y	hemiplegia (10) diplegia (12) tetraplegia (12) GMFCS I-III	Exp: 16 weeks NDT, 2*/week Contr: 16 weeks NDT, 5*/week	16	-	3	gross motor function (GMFM)	↑	↑	↑	A	II (5/7)
2006, Bar-Haim[29]	RCT ***Smaller RCT***	12	12	group 1:X=8,3y group 2:8,1y	GMFCS II-IV diplegia (11) Quadri (12)	Exp: Adeli suit Contr: NDT 4 weeks, 2h/day, 5*/week	4	39	5	gross motor function (GMFM)	↑	↑	=	A	II (6/7)
										Mechanical Efficiency Index during stairclimbing	↑	↑	↑	I	
2007, Cherng[30]	ABA or AAB	4	4	3,5-6,3y	GMFCS I-III	Group 1: ABA, Group 2:AAB	12	6	2,5	gait (GaitRite)	↑	=		I/A	II (8/14)
				X=4y	spastic diplegia	Contr: Reg PT, 2-3*/week NDT				Gross Motor Function (GMFM)	↑	=		I/A	
	SSRD					Exp: BWST, 2-3*/week+regular PT				muscle tone (MAS)	=	=		I	
										selective motor control (SMC)	=	=			
2008, Christansen[31]	RCT ***smaller RCT***	10	14	med 3 y range 1y-8y1mo	spastic CP GMFCS I -V	Exp: intermittent 4 weeks 4*/week followed by 6 weeks no therapy	30	-	3	gross motor function (GMFM)	↑	↑	=	A	II (5/7)
						Contr: continuous training, 1*/week									
2001, Butler[32]	***Syst Review***	21 studies	-	NDT studies	-	Medline, Healthstar, ClinPSYCH, CINAHL, Cochrane; until 2000/2001 keywords: neurodevelopmental treatment, NDT, Cerebral Palsy				AACPDM level of evidence				-	II (8)
2001, Brown[33]	***Syst Review***	17 studies	-	NDT studies	-	Medline, CINAHL, Cochrane, Cochrane Library, EMBASE, Eric, Healthstar, PsychInfo, Medline, Sociofile keywords: neuromuscular fascilitation, NDT, Bobath, motion therapy, exercise therapy, therapeutic exercises, kinetic chain exercises, psychomotor and therapeutic touch				Jadad scale Sacket levels of evidence				-	II (7)

Abbreviations:N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; SSRD: Single Subject Research DesginX: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; NDT: Neurodevelopmental Treatment; GMFM: Gross Motor Function Measure; MAS: Modified Ashworth Scale; PEDI: Pediatric Evaluation of Disability Inventory; ROM: Range of Motion; syst review: systematic review; AACPDM: American Academy of Cerebral Palsy and Developmental Medicine

Table N. Overview of the studies using conductive education – subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (CS)
1995, Coleman[34]	prospective intervention ***non-randomized CT***	11	9	19-69 mo X=45 mo	quadriplegia (11) diplegia (7) athetoid (1) hemiplegia (1)	Exp: conductive education Contr: traditional early intervention 6 months	26		not reported	parental perception and coping (questionnaire on Resources and Stress(QRS-F) areas of development (Vulpe Assessment Battery)			= =	**I/A** **I/E**	**III (2/7)**
1995, Catanese[35]	prospective intervention ***non-randomized CT***	17	17 matched	4 y - 7y1mo	mild (10) moderate (18) severe (6)	Exp: conductive education Contr: traditional early intervention 6 months	26		no reported	areas of development (Vulpe Assessment Battery) parental perception and coping (questionnaire on Resources and Stress (QRS-F) standardized cognitive test			= = =	**I/A** **I/E** **I**	**III (3/7)**
1997, Hur[36]	prospective intervention ***non-randomized CT***	19	17	3,5-4,5y	severe mild moderate	Exp: Conductive education Contr: British Special Education Program	156			skills for independence (Vineland Adaptive Behavior Scales) child functional level (Development Profile 2)	↑ ↑	↑ ↑	= =	**I/A/P** **A**	**III (2/7)**
1998, Reddihough[37]	RCT + matching ***Smaller RCT***	32	34	12-36 mo X=22mo3 wks	diplegia (11) quadriplegia (42) ataxia (2)	Group 1(randomized): CE (2,8h/week) Group 2(randomized): NDT (2,9h/week) Group 3(non-randomized): CE (3.2h/week) Group 4(non-randomized):NDT (2.2h/week)	26			areas of development (Vulpe Assessment Battery) gross motor function (GMFM) language development (Reynell Dev Lang Scale parental coping and stress (Parent Stress Index)	↑ ↑ ↑ =	↑ ↑ ↑ =	= = = =	**I/A** **A** **I** **E**	**II (4/7)**
1999, Woolfson[38]	prospective intervention ***Case-series***					conductive education+NDT	52		not reported	Semistructured interviews with the parents on perception of child remediation, parental re-education and redefinition	=			**I** **E** **P**	**IV (2/7)**

Abbreviations;

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; OT: Occupational Therapy; ST: Speech Therapy; GMFM: Gross Motor Function Measure; PEDI: Pediatric Evaluation of Disability Inventory; PDMS: Peabody Developmental Motor Scales; GAS: Goal Attainment Scale; SRIGM: Self-Reported Individualized Goal Measure; ROM: Range of Motion

Table N (continued) Overview of the studies using conductive education – subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (CS)
2003, Stiller[39]	RCT	7	12	2y5mo-9y2mo	hemi (2)	Exp: CE(6h/day, 5*/week)	5		6h/day	gross motor function (GMFM)	=	↑ (1)	=	A	II (5/7)
				Group 1: X=76mo SD=21	diplegia (22)	Contr 1: intensive therapy (PT,OT,ST) 5h/day, 5*/week)				fine motor function (PDMS)	=	=	=	A	
										functional abilities (PEDI)	=	↑(1)	=	A/P	
	Smaller RCT			Group 2=X=48mo SD21mo	quadri (14)	Contr 2: special education(6h/day, 5*/week)									
				Group 3:X=47mo SD 24mo											
2005, Wright[40]	Pro-spective	9	-	Year 1 X:6,5y, SD=0,8	GMFCS III-V	8 months intensive CE class	8		5*/week	gross/fine motor function (GMFM, PDMS, QUEST, PEDI, GAS)	↑			A/P	IV (3/7)
					diplegia 3										
				Year 2:X=4y6mo SD=1	quadri 1					self concept	↑			A	
	Case series									participation at school	↑			P	
										Family stress	↑			E	
2005, Odman[41]	Pro-spective	30	24	3-16y	GMFCS I-V	Exp: CE(move &walk)	2	52	Exp:4-5*/week	gross motor function (GMFM)	↑	↑	↓	A	III (3/7)
	repeated measures				diplegia 30	2-4h/day, 4-5*/week, 15 days				functional activities (PEDI-Functional Measures)	=	↑	=	A	
	control group				hemiplegia 4	Contr: Traditional Health Care (learning motor skills)			Contr:4*/week						
					tetraplegia 5										
					dyskinetic 13	3h/day, 4*/week;14 days									
					ataxic 2										
2006, Odman[42]	Pro-spective	30	24	3 - 16 y	GMFCS I-V	Exp: CE (move&walk), 2-4h/day, 4-5 days/week	2		Goup 1: 4-5*/week	gross motor function (GMFM)	↑	↑	↓	I	III (3/7)
	repeated measures				diplegia 30					functional activities (Pedi-Functional Measures)	=	↑	=	A	
	control group				hemiplegia 4	Contr: Traditional Health Care (Lemo), 3h/day, 4*/week									
					tetraplegia 5				Group 2: 4*/week	individualized goals (SRIGM)	↑	↑	=	A/P	
					dyskinetic 13										
					ataxic 2										
2009, Odman[43]	Pro-spective	15	-	4-17y	GMFCS I-V	intensive group training, 4 weeks, 2-4h/day, 3-5*/week	4		3-5*/week	semistructured interview with the parents	* no stats			I/E	IV (1/7)
	Case series				di (2) tetra (2)										
					hemi (6)										

Abbreviations: N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; CT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; PT: Physical Therapy; OT: Occupational Therapy; ST: Speech Therapy; CE: Conductive education; GMFM: Gross Motor Function Measure; PEDI: Pediatric Evaluation of Disability Inventory; PDMS: Peabody Developmental Motor Scales; GAS: Goal Attainment Scale; SRIGM: Self-Reported Individualized Goal Measure; ROM: Range of Motion

Table N (continued)

Author	type	N	Intervention	Evaluation	LE (CS)
1999, Woolfson[44]	***Syst Review***	10 studies	Medline, ERIC, PsychLit, Social Science Citation Index No time limitatons	-	**II (4)**
2000, Pedersen[45]	***Syst Review***	9 studies	Medline, ERIC, PsychINFO no time limitations, studies using control groups keywords: conductive education	-	**II (3)**
2004, Darrah[10]	***Syst Review***	88 studies	Medline (1966-2001), HealthSTAR (1975-2000), Cinahl (1982-2001), EMBASE (1988-2001), ERIC (1966-2001), AMED (1985-2001), Psychinfo (1984-2001)	-	**II (8)**

Abbreviations:

LE: Level of Evidence; CS: Conduct Score; Syst Review: Systematic Review

Table O. Overview of the studies evaluating the effectiveness of sensory integration and Votja therapy - subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (Cs)
Sensory integration															
2001, Bumin[15]	RCT	32	9	Group 1&2:X=7.06y	spastic diplegia	Group 1 (n=16):SMP Group 2 (n=16):SMP, group training	14	no follow-up	3	Ayers Southern California Sensory Integration Test	↑ (1+2)	= (3)		I/A	II (3)
	Smaller RCT			Group 3:X=7y		Group 3 (n=9): home programme SMP: 3*/week, 1,5h/session, 3 months SMP= sensory training, vestibular training, balance and postural reactions, bimanual activities and motor planning				Physical Ability Test	↑ (1+2)	= (3)		A	
Vojta therapy															
2004, Kanda[46]	RCT ***smaller RCT***	5	5	1m-3m	spastic diplegia	Group 1(n=5): Vojta 52 months, 30min/session, 90-120 sessions/month Group 2 (n=2): no treatment Group 3 (n=3): insufficient therapy	208	3y	90-120 sessions/ month	Highest motor developmental level			↑	I/A	II (2)

Abbreviations

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; SMP: Sensory Perceptual Motor Training

Table P: Overview of the studies using functional and task-oriented training- subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (CS)
2001, Ketelaar[5]	RCT ***Smaller RCT***	28	27	Group 2: X=54, SD=20 Group 1: X=56, SD=20 2-7y	mild or moderate hemiplegia (32) di plegia (11) quadri plegia(12)	Exp: Practicing functional activities Contr: Training based on normalization and quality of movement	78	-	Exp: 3,4 Contr 3,8	gross motorfunction (GMFM) functional performance (PEDI)	↑ ↑	↑ ↑	= ↑	A A/P	II (6/7)
2005, Ekström[6]	prospective No control ***Case-series***	14	-	1y6mo-6y X=3y7mo	GMFCS II-V	functional, goal oriented training 2*/day to 25*/day (varying)	21,7	13	2-25	individual goals (GAS) gross motor function (GMFM) functional perfomance (PEDI) Measure of Process Care	↑ ↑ ↑ ↑			A A A/P E	IV (2/7)
2005, Schalow[47]	prospective intervention *case-series*	8	-	X=15y 7-27y	not reported	Low intensity coordination dynamics therapy: crawling, treadmill walking, jumping, exercising on a special board)	13	-	4	motor programs (EMG) coordination	= =			I A	IV (1/7)
2007, Crompton[48]	RCT ***Smaller RCT***	8	7	6-14y group 1: X=9,9y SD=2,5y group 2:X=11,2y SD=1,9	spastic diplegia GMFCS I-III	Exp: LL training (circuit trainig: closed kinetic chain, strength, balance,coordination, stretching; 70% of max work rate) Contr :UL dexterity training (stretching, games and task requiering manipulation and dexterity, inhand-manipulation) + usual therapy (0-2*/week)	6	6	2	gross motor function (GMFM) Timed Up and Go Uptime (mean uptime hours) 10 min walk test strength LL/UL (dynamometer) hand tasks (BOT) gross manual dexterity Handwriting Speed test rapid hand manipulation	= ↓ ↑ = =,= = = ↑ ↑	= = ↑ = =, ↑ ↑ ↑ = =	= = = = = ↓ = = ↓	A A A A I A A A A	II (5/7)
2009, Salem[49]	RCT ***Smaller RCT***	5	5	range 4,9-10,2y X=6,53y, SD=1,8Y	GMFCS I - III diplegia (8) quadri (2)	Contr: walking and balance; facilitation and normalization of movement patterns. Exp: task-oriented training;	5	-	2	Timed Up and Go gross motor function (GMFM)	↑ ↑	= =	↑ ↑	A A	II (5/7)
2010, Löwing[50]	prospective intervention *ABA design SSRD*	22	-	X=46 mo SD=16mo	GMFCS I-IV	phase B: goal directed functional acitivites (learning new skills in daily life) Phase A: Follow-up	12	12	in ADL 7	Gross Motor Function (GMFM) individual functional goals (GAS) pROM (goniometer) selectivity (selective motor control) spasticity (MAS)	↑ ↑ ↑ ↑ ↓	= = = = =		A A I I I	IV (8/14)

Abbreviations: N: Number; Exp: experimental group/period; Contr: control group/period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; SSRD: Single Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFCS: Gross Motor Classification System; GMFM: Gross Motor Function Measure; PEDI: Pediatric Evaluation of Disability Inventory; GAS: Goal Attainment Scale; LL: Lower Limb, UL: Upper Limb; BOT: Bruininks-Oseretsky Test of Motor Proficiency; MAS: Modified Ashworth Scale, pROM: Passive Range of Motion

Table Q: Overview of the studies using goal-setting procedures- subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method					Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (CS)
1996, Bower [13]	RCT 2*2 factorial ***smaller RCT***	22	22	X group 1=6.3y X group 2=5.5y Xgroup 3=5.8y Xgroup 4=5.6y	spastic quadriplegia	Group 1: usual PT based on general aims Group 2: intensive PT (1h/day) based on general aims Group 3: usual PT based on specific goals Group 4: intensive PT (1h/day) based on specific goals 2 weeks selection of goals/aims based on GMFM treatment type was mixed (eclectic)	2	-	7	gross motor function (GMFM)			↑ (3+4)	**A**	**II (4/7)**
2001, Bower [51]	RCT 2*2 fact ABA ***smaller RCT***	28	28	X=5.9 range 3-12y	GMFCS III-V spastic diplegia spastic quadriplegia	Group 1(n=15): PT based on general aims (12h/6 months) Group 2 (n=13): intensive PT (1h/day) based on general aims Group 3 (n=13): PT based on specific goals(12h/6months) Group 4 (n=15): intensive PT (1h/day) based on specific goals ABA design: 6 mo baseline observation, 6 mo intervention, 6 mo follow-up	26	26	7	gross motor function (GMFM and GMPM) Measure of Process Care Questionnaire			= = 	**A** **A** **E**	**II (7/7)**
2009, Löwing [52]	Pro-spective Multi-centre ***non-random-ized CT***	22	22	X=4y1mo SD =1y5mo	GMFCS I-IV unilateral 17 bilateral 27	Exp: goal directed therapy (group training and day-to-day practice, 1*/week, individual goals) Contr: activity directed therapy (1*/week, based on general aims)	12	-	1	functional abilities (PEDI) gross motor function (GMFM)	↑ ↑	= =	↑ ↑	**A/P** **A**	**II (4/7)**

Abbreviations

N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Int: Intervention group; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFCS: Gross Motor Classification System; GMFM: Gross Motor Function Measure; GMPM: Gross Motor Performance Measure; PEDI: Pediatric Evaluation of Disability Inventory

Table R: Overview of the studies using hydrotherapy or aquatic therapy- subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method				Evaluation	Results			ICF	LE (CS)
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Follow-up (weeks)	Freq (*/week)		Exp	Contr	Exp vs cont		
1998a, Hutzler[53]	RCT *smaller RCT*	23	23	5-7y Group 1: X=5,7y SD 1 Group 2: X=5,5 SD 0,9	hemiplegia (17) diplegia (19) quadriplegia (6) ataxia/athetosis (4)	Group 1: swimming sessions+ physical activity at gym Exp: NDT; 30 min, 4*/week	26	-	2	Lung function (spirometer) Water Orientation Score	↑		↑	I I/A	II (1/7)
1998 b, Hutzler[54]	RCT *smaller RCT*	23	23	5-7y x=507y	diplegia (19) hemiplegia (17) quadriplegia (6) ataxia/athetosis (4)	Exp: movement and swimming program (3*/week,30min) + group movement+NDT Contr: NDT 30 min, 4*/ week	26	-	3	Water Orientation Score Selfperception (Martinek-Zaichkowsky Self Concept Scale)	↑	= =		I/A I	II (2/7)
2005, Thorpe[55]	Pro-spective intervention *SSRD AB*	7	-	7-13y X=9,7y SD=1,8y	spastic diplegia (6) spastic hemiplegia (1)	phase A: Individual aquatic exercise sessions, 45 min phase B: usual therapy	10	11	3	leg strength (handheld dynamometer) gait velocity(3min walking test) energy expenditure (Resting Heart Rate) gross motor function (GMFM) functional mobility (TUG)	= = = ↑ ↓	= = = = =		I A I A A	IV (8/14)
2007, Ozer[56]	prospective intervention ***smaller RCT***	13	10	Exp: X=8,1y SD 1,5y Contr:X=8,9y SD 1,5y	not described	Exp: 14 weeks swimming training + trad PT Contr : traditional PT only	14	26	3	Child Behaviour Check List: body awareness competence problem behaviour	 ↑ = =			I	II (3/7)

Abbreviations:

N: Number; Exp: Experimental group or experimental period; Contr: Control group or control period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; SSRD: Single Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; NDT: Neurodevelopmental Treatment; VC: Vital Capacity; TUG: Timed Up and Go test; GMFM: Gross Motor Function Measure; GMPM: Gross Motor Performance Measure; PEDI: Pediatric Evaluation of Disability Inventory

Table S: Overview of the studies using therapeutic horse-riding subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (* /week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (CS)
1995 a, Mac Kinnon[57]	RCT	10	9	4 - 12y	mild	Exp: usual PT+ 1h/week riding classes	26	-	1	gross and fine motor function (GMFM, PDMS)	=		=	A	II (5/7)
				X=6,5y	moderate					posture (bertoti)	=		=	I/A	
	smaller RCT			SD=6,5y	independent sitting	functional riding skills, basic horse and stable knowledge, games on horseback)				BOT - UL speed and dexterity	↑		↑	A	
										ADL (Vineland Adaptive Behaviour Scales)	=		=	I	
										self perception (Harter Self Perception Scale)	↑		=	I	
										child behaviour (Child Behaviour Checklist)	↑		↑	I	
						Contr: waiting list + usual PT				Qualitative questionnaire of performance	↑		=	A	
1998, Quint[58]	prospective matched	13	13	9-16y	spastic quadriplegia	Exp: BABS (powered saddle imitating a walking horse) 10 times, 10 min	4	-	2	pelvic tilt in sitting (photographic measurement)	↑	=	↑	I/A	II (5/7)
	Non-random-ized CT					Contr: static saddle, 10 times, 10 min									
1998, Mc Gibbon[59]	prospective repeated measures	5	-	X=9y6m	independent walking	Phase A: usual PT	8	-	2	gross motor function (GMFM)	↑	=		A	IV (9/14)
					diplegia (4)	Phase B: usual PT + 30 min hippotherapy (muscle elongation and relaxation, optimal postural alignment and independent sitting)				energy expenditure (heart rate)	↓	=		I	
					hemiplegia (1)					stride length, cadence and velocity	=,=,=	=,=,=		I/A	
	SSRD														
	AB design														
1999, Kuczynski[60]	prospective intervention	25	-	3-10y	tetra (12)	20 min microprocessor controlled saddle riding	13	-	2	postural sway (centre of pressure measurement)	↓			I/A	IV (1/7)
				X=6,3y	diplegia (4)										
	case series			SD=1,7y	hemiplegia (9)					functional abilities (PEDI)	=			A/P	
1999, Haehl[61]	prospective intervention	2	-	subject 1:9,6y	Spastic/athetoid quadriplegia	hippotherapy: slow to medium walking speed; movements anw walking, forward sitting, minimum hands-on	12	-	1	posture and postural control (markers)	↑			I	IV (3/7)
	case series			subject 2: 4y	spastic diplegia										

Abbreviations:

N: Number; Exp: Experimental group or period; Contr: Control group period; Freq: Frequency; Int: Intervention; Vs: Versus; ICF: International Classification of Functioning; RCT: LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; SSRD: Single Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFM: Gross Motor Function Measure; BOT: Bruininks-Oseretsky Test of Motor Proficiency; MAS: Modified Ashworth Scale; PEDI: Pediatric Evaluation of Disability Inventory

Table S (continued): Overview of the studies using therapeutic horseriding- subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method				Results				
		N (exp)	N (contr)	Age	Type	Intervention	Int (weeks)	Follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	Exp vs cont	ICF	LE (CS)
2002, Sterba[62]	prospective repeated measures ***SSRD*** ***AB design***	17	-	X=9y10m SD = 10 mo	diplegia (12) quadriplegia (3) hemiplegia (2) GMFCS I-V	phase A : horse back riding therapy (exercises during riding, prone lying,…) Phase B: usual PT	18	6	1	gross motor function (GMFM) functional independence (WeeFIM)	↑ =	= =	↑ =	A A	IV (10/14)
2003, Benda[63]	prospective intervention control group ***smaller RCT***	7	8	4-12y	spastic independent sitting, standing	Exp: 8 min of hippotherapy (rythmic multidimensional movement of the horse) Contr: 8 min stationary barrel (neutral warmth for a fleece saddle,symmetrical forward sitting posture	1 session	-	-	EMG of trunk, upper leg muscles during sitting, standing and walking asymmetry score			 ↓	I/A I	II (4/7)
2004, Cherng[64]	prospective intervention repeated measures ***AB design***	14	-	3y1m-11y5m group 1: X=92,3 m group 2:X=93 m	quadriplegia (5) diplegia (7) hemiplegia (2) ambulant non-ambulant	Exp: AB Contr: BA phase A= PT only phase B: usual PT + horseback riding therapy 40 min, 2*/week	16	16	2	gross motor function (GMFM) muscle tone of hip adductors (MAS)	↑ =	= =	↑ =	A I	IV (10/14)
2004, Casady[65]	prospective ***SSRD*** ***ABA design***	10	-	2,3-6,8y X=4,1y SD=1,7	spastic quadri (2) spastic dipl (1) hemiplegia (3) athetosis (1) non-specified (11)	Phase B: 10 weeks, 1*/week hippotherapy Phase A: 20 weeks usual PT	10	10	1	functional abilities (PEDI) gross motor function (GMFM)	↑ ↑	= =	↑ ↑	A/P A	IV (6/14)
2008, Zurek[66]	prospective intervention ***case series***	16	-	14-16y X=9,3y SD=3,8y	spastic dipl (7) spastic hem (5) other (4)	Hippotherapy: 15-35 min on saddle (1 session)	-		-	Limb skin surface temperature	↑			I	IV (2/7)
2009, Shurtleff[67]	prospective intervention ***SSRD*** ***AB design***	11 CP	8 (non-CP)	5-13y X=8y	diplegia	Phase A: hippotherapy,45 min, + usual PT Phase B: wash-out, usual PTonly AB design	12	12	1	3D analysis of head/trunk stability reaching/grasping	↑ ↑			A	IV (4/7)

Abbreviations: N: Number; Exp: Experimental group or period; Contr: Control group or period; Freq: Frequency; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; SSRD: Single Subject Research Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFM: Gross Motor Function Measure; BOT: Bruininks-Oseretsky Test of Motor Proficiency; MAS: Modified Ashworth Scale; PEDI: Pediatric Evaluation of Disability Inventory

Table S (continued): Overview of the studies using hydrotherapy or aquatic therapy- subjects, interventions, evaluation, results, levels of evidence and conduct scores

Author	Design	Subjects				Method				Results				
	type	N (exp)	N (contr)	Age	Type	Intervention	Duration int (weeks)	Duration follow-up (weeks)	Freq (*/week)	Evaluation	Exp	Contr	exp vs contr	ICF	LE (CS)
2009, McGibbon[68]	RCT ***smaller RCT***	25	22	Group 1: X=8y5mo Group 2: X=8y8mo	GMFCS I-IV diplegia (25) quadriplegia (9) hemiplegia (7) mixed (6)	Phase 1: 10 minutes hippo (group 1) /10minutes barrel (group 2) Phase 2: 12 weekly hippotherapy sessions	12		1	adductor spasticity (EMG) Gross motor function (GMFM) self-perception (self-perception profiles)	↓ ↑ =,=,=	= = =,=,=		**I** **A** **I**	**II (6/7)**
2009, Davis[69]	RCT ***smaller RCT***	50	49	4 to 12y Exp: X=7y8mo Contr: X=8y2mo	GMFCS I-III	Exp: therapeutic horseriding, 30-40 min Contr: usual activities	10		1	gross motor function (GMFM) health status (CHQ) quality of life (CP QoL, KIDSCREEN)	 =	 ***QoL***	= =	**A** **I/P**	**II (6/7)**
2009, Debuse[70]	retrospective qualitative multicenter ***case-series***	17	-	4-63y	GMFCS I-V	6 weeks to several years hippotherapy	not reported	not reported	not reported	Semistructured interviews				**I** **A** **P** **E**	**IV (1/7)**
1995b, Mac Kinnon[71]	***syst review***	11 studies	**-**	**-**	**-**	-	-	-	-	**-**					**II (0)**
2007, Sterba[72]	***syst review***	11 studies	-	-	-	Cochrane library, Dare, Medline, CINAHL keywords: HBRT, hippotherapy, developmental riding therapy, equine-movement therapy, riding for disabled, therapeutic horse-riding therapy, therapeutic riding, cerebral palsy, exercise therapy, horrseback riding, horses, physical therapy techniques, recreational therapy, rehabilitation, therapeutic exercise				Critical Review Form (Law et al 1998)					**II (8)**
2007, Snider[73]	***syst review***	9 studies	-	-	-	Pedro, Medline, CINAHL, ERIC, HealthSTAR keywords horse, riding, hippotherapy,horseback riding equine movement therapy ,,,,-2005				levels of evidence according to Sacket Pedro PICO format					**II (8)**

Abbreviations;

N: Number; Exp: Experimental group or experimental period; Contr: Control group or control period; Int: intervention; Freq: Frequency; Vs: Versus; ICF: International Classification of Functioning; LE: Level of Evidence; CS: Conduct Score; RCT: Randomized Controlled Design; X: mean; SD: Standard Deviation; y: years; mo: months; GMFCS: Gross Motor Classification System; GMFM: Gross Motor Function Measure; CHQ: Child Health Questionaire; MAS: Modified Ashworth Scale; PEDI: Pediatric Evaluation of Disability Inventory

Appendix T. List of main problems

Primary problems	Problem nr
Muscle tone	
Hypertonia	1
Hypotonia	2
Fluctuating muscle tone	3
Postural control	
Static alignment	4
Dynamic alignment	5
Insufficient co-activation	6
Laxity	7
Movement patterns	
Extension pattern	8
Flexion pattern	9
Asymmetric Tonic Neck Reflex (ATNR)	10
Other pattern	11
Muscle strength	12
Secondary problems	
Muscle lenght	13
Bony deformities	14

Appendix U: Lists of individual goals from clinical examination

ROM and muscle length	
ROM1	Hip extension deficit (Thomas test)
ROM3	ROM hip abduction (Knee and hip flexed)
ROM4	ROM hip abduction (Knee and hip extended)
ROM5	Knee extension deficit
ROM6	Popliteal angle
ROM7	Ankle ROM dorsiflexion (knee flexed)
ROM8	Ankle ROM dorsiflexion (knee extended)
ROM9	Varus deformity
ROM10	Valgus deformity
ROM11	Rectus femoris length
ROM12	Length of tensor fasciae latae
Spasticity	
SP1	Spasticity of hip flexors (Modified Ashworth)
SP2	Spasticity of hip adductors (Modified Ashworth)
SP3	Spasticity of hamstrings (Modified Ashworth)
SP4	Spasticity of Rectus Femoris (Duncan Ely)
SP5	Spasticity of soleus (Modified Ashworth)
SP6	Spasticity of gastrocnemius (Modified Ashworth)
SP7	Spasticity of tibialis posterior (Modified Ashworth)
Strength and selectivity	
STR1	Strength abdominal muscles
STR2	Strength hip abductors
STR3	Strength hip extensors (manual testing)
STR4	Strength knee extensors (manual testing)
STR5	Strength knee flexors (manual testing)
STR6	Strength ankle dorsiflexors (manual testing)
STR7	Strength ankle plantar flexors (manual testing)
Bony deformities	
BD 1	Patella alta
BD 2	Femoral Anteversion
BD 3	Tibio-femoral angle
BD 4	Bimalleolar Angle

Appendix V: List of goals from three-dimensional gait analyis

Time and distance parameters	
TD1	Walking speed
TD1	Timing of toe off
TD2	Cadence
TD3	Step length
TD4	Endurance
TD5	Step width
TD6	Energy consumption
Pelvis	
P1	Mean pelvic anterior tilt
P2	Range of pelvic motion in sagittal plane
P3	Range of pelvic motion in coronal plane
P4	Range of pelvic motion in transverse plane
P5	Pelvis asymmetry
Hip	
H1	Maximum hip extension in stance
H2	Total sagittal hip range of motion
H3	Maximum hip flexion in swing
H4	Peak hip abduction in swing
H5	Mean hip rotation in stance
H6	Maximum hip rotation in stance
H7	Timing of '0' sagittal hip moment in stance
H8	Maximum hip extension moment in stance
H9	Maximum hip flexion moment in stance
H10	Hip adduction in stance
H11	Hip angle at IC
Knee	
K1	Knee angle at initial contact
K2	Maximum knee extension in stance
K3	Range of knee motion in stance
K4	Maximum knee flexion in swing
K5	Timing of maximum knee flexion in swing
K6	Maximum knee extension moment in stance
K7	Maximum knee flexion moment in stance
K8	Phasic activity of rectus femoris
Ankle	
A1	Ankle angle at initial contact
A2	Maximum dorsiflexion in stance
A3	Ankle angle at midswing
A4	Ankle ROM during push-off
A5	Ankle moment at loading response
A6	Maximum ankle plantar flexion moment at preswing
A7	Maximum ankle power absorption in loading response and midstance
A8	Maximum ankle power generation at preswing
A9	Phasic activity of tibialis anterior
A10	Phasic activity of gastrocnemius
A11	2nd ankle rocker
A12	Max. DF in swing
A13	Ankle rotation during stance
A14	1st ankle rocker
Foot	
F1	Mean foot progression angle

Appendix W: List of goals from Gross Motor Function Measure (GMFM-88)

Dimension A Lying and rolling	
GMFM 1	Supine, head in middle, turns head with extremities symmetrical
GMFM 2	Supine, brings hands to midline, fingers one with the other
GMFM 3	Supine, lifts head 45°
GMFM 4	Supine, flexes right hip and knee through full range
GMFM 5	Supine, flexes left hip and knee through full range
GMFM 6	Supine, reaches out with right arm, hand crosses midline
GMFM 7	Supine, reaches out with left arm, hand crosses midline
GMFM 8	Supine, rolls to prone over right side
GMFM 9	Supine, rolls to prone over left side
GMFM 10	Prone, lift head upright
GMFM 11	Prone on forearms, lifts head upright, elbows extended, chest raised
GMFM 12	Prone on forearms, weight on right forearm, fully extends opposite arm forward
GMFM 13	Prone on forearms, weight on left forearm, fully extends opposite arm forward
GMFM 14	Prone, rolls to supine over right side
GMFM 15	Prone, rolls to supine over left side
GMFM 16	Prone, pivots to the right 90°, using extremities
GMFM 17	Prone, pivots to the left 90°, using extremities
Dimension B Sitting	
GMFM 18	Supine, hands grasped by examiner, pulls self to sitting with head control
GMFM 19	Supine, rolls to right side, attains sitting
GMFM 20	Supine, rolls to left side, attains sitting
GMFM 21	Sit on mat, supported at thorax by therapist, lifts head in midline, maintains 3 sec
GMFM 22	Sit on mat, supported at thorax by therapist, lifts head in midline, maintains 10 sec
GMFM 23	Sit on mat, arms propping, maintains 5 seconds
GMFM 24	Sit on mat, maintains arms free, 3 seconds
GMFM 25	Sit on mat with small toy in front, leans forward, touches toy, re-erects without arms propping
GMFM 26	Sit on mat, touches toy placed 45° behind child's right side, returns to start
GMFM 27	Sit on mat, touches toy placed 45° behind child's left side, returns to start
GMFM 28	Right side sit, maintains arms free, 5 seconds
GMFM 29	Left side sit, maintains arms free, 5 seconds
GMFM 30	Sit on mat, lowers to prone with control
GMFM 31	Sit on mat with feet in front, attains 4 point over right side
GMFM 32	Sit on mat with feet in front, attains 4 point over left side
GMFM 33	Sit on mat, pivots 90° without arms assisting
GMFM 34	Sit on bench, maintains arms and feet free, 10 seconds
GMFM 35	Standing, attains sit on a small bench
GMFM 36	On the floor, attains sit on a small bench
GMFM 37	On the floor, attains sit on a large bench
Dimension C Crawling and kneeling	
GMFM 38	Prone, creeps forward 1.8 m (6')
GMFM 39	4 point, maintains weight on hands and knees, 10 seconds
GMFM 40	4 point, attains sit arms free
GMFM 41	Prone, attains 4 point, weight on hands and knees
GMFM 42	4 point, reaches forward with right arm hand above shoulder
GMFM 43	4 point, reaches forward with left arm hand above shoulder
GMFM 44	4 point, crawls or hitches forward 1.8 m (6')
GMFM 45	4 point, crawls reciprocally forward, 1.8 m (6')
GMFM 46	4 point, crawls up 4 steps on hands and knees or feet
GMFM 47	4 point, crawls backwards down 4 steps on hands and knees or feet
GMFM 48	Sit on mat, attains high kneeling using arms, maintains, arms free, 10 seconds
GMFM 49	High kneeling, attains half kneeling on right knee using arms, maintains arms free, 10 seconds
GMFM 50	High kneeling, attains half kneeling on left knee using arms, maintains arms free, 10 seconds
GMFM 51	High kneeling, walks forward on knees 10 steps, arms free

Appendix W (continued): List of goals from Gross Motor Function Measure (GMFM-88)

Dimension D Standing	
GMFM 52	On the floor, pulls to stand at large bench
GMFM 53	Standing, maintains arms free, 3 seconds
GMFM 54	Standing, holding on to large bench with one hand, lifts right foot, 3 seconds
GMFM 55	Standing, holding on to large bench with one hand, lifts right foot, 3 seconds
GMFM 56	Standing, maintains arms free, 20 seconds
GMFM 57	Standing, lifts left foot, arms free, 10 seconds
GMFM 58	Standing, lifts right foot, arms free, 10 seconds
GMFM 59	Sit on small bench, attains standing without using arms
GMFM 60	High kneeling, attains standing through half kneeling on right knee, without using arms
GMFM 61	High kneeling, attains standing through half kneeling on left knee, without using arms
GMFM 62	Standing, lowers to sit on the floor with control, arms free
GMFM 63	Standing, attains squat, arms free
GMFM 64	Standing, picks up object from floor, arms free, returns to stand
Dimension E: Walking, running, and jumping	
GMFM 65	Standing, 2 hands on large bench, cruises 5 steps to the right
GMFM 66	Standing, 2 hands on large bench, cruises 5 steps to the left
GMFM 67	Standing, 2 hands held, walks forwards 10 steps
GMFM 68	Standing, 1 hand held, walks forwards 10 steps
GMFM 69	Standing, walks forward 10 steps
GMFM 70	Standing, walks forward 10 steps, stops, turns 180°, returns
GMFM 71	Standing, walks backward 10 steps
GMFM 72	Standing, walks forward 10 steps, carrying a large object with 2 hands
GMFM 73	Standing, walks forward 10 consecutive steps between parallel lines 20 cm (8” apart)
GMFM 74	Standing, walks forward 10 consecutive steps on a straight line 2 cm (3/4”) wide
GMFM 75	Standing, steps over stick at knee level, right foot leading
GMFM 76	Standing, steps over stick at knee level, left foot leading
GMFM 77	Standing, runs 4.5 m (15’), stops and returns
GMFM 78	Standing, kicks ball with right foot
GMFM 79	Standing, kicks ball with left foot
GMFM 80	Standing, jumps 30 cm (12”) high, both feet simultaneously
GMFM 81	Standing, jumps forward 30 cm (12”) high, both feet simultaneously
GMFM 82	Standing on right foot, hops on right foot 10 times with a 60 cm (24”) circle
GMFM 83	Standing on left foot, hops on left foot 10 times with a 60 cm (24”) circle
GMFM 84	Standing, holding 1 rail, walks up 4 steps, alternating feet
GMFM 85	Standing, holding 1 rail, walks down 4 steps, alternating feet
GMFM 86	Standing, walks up 4 steps, alternating feet
GMFM 87	Standing, walks up 4 steps, alternating feet
GMFM 88	Standing on 15 cm (6”) step, jumps off, both feet simultaneously

Appendix X: Item analysis evaluating the agreement between the live-and video-ratings rated by PT1

Item GMFM		K	PA
Dimension A: Lying and rolling			
Item 1	Supine, head in middle, turns head with extremities symmetrical	1,000	1,00
Item 2	Supine, brings hands to midline, fingers one with the other	1,000	1,00
Item 3	Supine, lifts head 45°	0,490	0,96
Item 4	Supine, flexes right hip and knee through full range	0,703	0,92
Item 5	Supine, flexes left hip and knee through full range	0,611	0,90
Item 6	Supine, reaches out with right arm, hand crosses midline	1,000	1,00
Item 7	Supine, reaches out with left arm, hand crosses midline	1,000	1,00
Item 8	Supine, rolls to prone over right side	1,000	1,00
Item 9	Supine, rolls to prone over left side	1,000	1,00
Item 10	Prone, lift head upright	1,000	1,00
Item 11	Prone on forearms, lifts head upright, elbows extended, chest raised		0,98
Item 12	Prone on forearms, weight on right forearm, fully extends opposite arm forward	1,000	1,00
Item 13	Prone on forearms, weight on left forearm, fully extends opposite arm forward	1,000	1,00
Item 14	Prone, rolls to supine over right side	1,000	1,00
Item 15	Prone, rolls to supine over left side	1,000	1,00
Item 16	Prone, pivots to the right 90°, using extremities		0,98
Item 17	Prone, pivots to the left 90°, using extremities		0,98
Dimension B: Sitting			
Item 18	Supine, hands grasped by examiner, pulls self to sitting with head control	1,000	1,00
Item 19	Supine, rolls to right side, attains sitting	1,000	1,00
Item 20	Supine, rolls to left side, attains sitting	0,851	0,98
Item 21	Sit on mat, supported at thorax by therapist, lifts head in midline, maintains 3 sec	1,000	1,00
Item 22	Sit on mat, supported at thorax by therapist, lifts head in midline, maintains 10 sec	1,000	1,00
Item 23	Sit on mat, arms propping, maintains 5 seconds	1,000	1,00
Item 24	Sit on mat, maintains arms free, 3 seconds	1,000	1,00
Item 25	Sit on mat, small toy in front, leans forward, touches toy, re-erects without arms propping	1,000	1,00
Item 26	Sit on mat, touches toy placed 45° behind child's right side, returns to start	1,000	1,00
Item 27	Sit on mat, touches toy placed 45° behind child's left side, returns to start		0,98
Item 28	Right side sit, maintains arms free, 5 seconds	0,587	0,84
Item 29	Left side sit, maintains arms free, 5 seconds	0,545	0,84
Item 30	Sit on mat, lowers to prone with control	1,000	1,00
Item 31	Sit on mat with feet in front, attains 4 point over right side		0,98
Item 32	Sit on mat with feet in front, attains 4 point over left side	-0,0101	0,96
Item 33	Sit on mat, pivots 90° without arms assisting	0,714	0,86
Item 34	Sit on bench, maintains arms and feet free, 10 seconds	1,000	1,00
Item 35	Standing, attains sit on a small bench	0,686	0,90
Item 36	On the floor, attains sit on a small bench	0,879	0,96
Item 37	On the floor, attains sit on a large bench	0,798	0,92

Appendix X (continued): Item analysis evaluating the agreement between the live-and video-ratings rated by PT1

Dimension C: Crawling and kneeling			
Item 38	Prone, creeps forward 1.8 m (6')	-0,010	0,96
Item 39	4 point, maintains weight on hands and knees, 10 seconds		0,98
Item 40	4 point, attains sit arms free	-0,020	0,92
Item 41	Prone, attains 4 point, weight on hands and knees		0,96
Item 42	4 point, reaches forward with right arm hand above shoulder		0,94
Item 43	4 point, reaches forward with left arm hand above shoulder		0,94
Item 44	4 point, crawls or hitches forward 1.8 m (6')		0,98
Item 45	4 point, crawls reciprocally forward, 1.8 m (6')	0,310	0,96
Item 46	4 point, crawls up 4 steps on hands and knees or feet	0,601	0,82
Item 47	4 point, crawls backwards down 4 steps on hands and knees or feet	0,593	0,82
Item 48	Sit on mat, attains high kneeling using arms, maintains, arms free, 10 seconds	0,527	0,84
Item 49	High kneeling, attains half kneeling on right knee using arms, maintains arms free, 10 sec	0,627	0,68
Item 50	High kneeling, attains half kneeling on left knee using arms, maintains arms free, 10 sec	0,641	0,68
Item 51	High kneeling, walks forward on knees 10 steps, arms free	0,694	0,86
Dimension D: Standing			
Item 52	On the floor, pulls to stand at large bench	0,740	0,96
Item 53	Standing, maintains arms free, 3 seconds	0,704	0,86
Item 54	Standing, holding on to large bench with one hand, lifts right foot, 3 seconds	0,440	0,82
Item 55	Standing, holding on to large bench with one hand, lifts right foot, 3 seconds	0,564	0,86
Item 56	Standing, maintains arms free, 20 seconds	0,811	0,88
Item 57	Standing, lifts left foot, arms free, 10 seconds	0,747	0,84
Item 58	Standing, lifts right foot, arms free, 10 seconds	0,792	0,86
Item 59	Sit on small bench, attains standing without using arms	0,692	0,76
Item 60	High kneeling, attains standing through half kneeling on right knee, without using arms	0,745	0,68
Item 61	High kneeling, attains standing through half kneeling on left knee, without using arms	0,813	0,74
Item 62	Standing, lowers to sit on the floor with control, arms free	0,799	0,76
Item 63	Standing, attains squat, arms free	0,774	0,80
Item 64	Standing, picks up object from floor, arms free, returns to stand	0,974	0,98

Appendix X (continued): Item analysis evaluating the agreement between the live-and video-ratings rated by PT1

Dimension E: Walking, jumping and running			
Item 65	Standing, 2 hands on large bench, cruises 5 steps to the right	0,490	0,96
Item 66	Standing, 2 hands on large bench, cruises 5 steps to the left	0,490	0,.96
Item 67	Standing, 2 hands held, walks forwards 10 steps		0,98
Item 68	Standing, 1 hand held, walks forwards 10 steps	0,556	0,96
Item 69	Standing, walks forward 10 steps	0,824	0,90
Item 70	Standing, walks forward 10 steps, stops, turns 180°, returns	1,000	1.00
Item 71	Standing, walks backward 10 steps	1,000	1.00
Item 72	Standing, walks forward 10 steps, carrying a large object with 2 hands	0,855	0,94
Item 73	Standing, walks forward 10 consecutive steps between parallel lines 20 cm (8" apart)	0,892	0,86
Item 74	Standing, walks forward 10 consecutive steps on a straight line 2 cm (3/4") wide	0,825	0,82
Item 75	Standing, steps over stick at knee level, right foot leading	0,799	0,74
Item 76	Standing, steps over stick at knee level, left foot leading	0,855	0,80
Item 77	Standing, runs 4.5 m (15'), stops and returns	0,739	0,68
Item 78	Standing, kicks ball with right foot	0,951	0,96
Item 79	Standing, kicks ball with left foot	0,936	0,94
Item 80	Standing, jumps 30 cm (12") high, both feet simultaneously	0,815	0,82
Item 81	Standing, jumps forward 30 cm (12") high, both feet simultaneously	0,824	0,78
Item 82	Standing on right foot, hops on right foot 10 times with a 60 cm (24") circle	0,911	0,94
Item 83	Standing on left foot, hops on left foot 10 times with a 60 cm (24") circle	0,769	0,88
Item 84	Standing, holding 1 rail, walks up 4 steps, alternating feet	0,657	0,66
Item 85	Standing, holding 1 rail, walks down 4 steps, alternating feet	0,695	0,70
Item 86	Standing, walks up 4 steps, alternating feet	0,709	0,72
Item 87	Standing, walks up 4 steps, alternating feet	0,731	0,74
Item 88	Standing on 15 cm (6") step, jumps off, both feet simultaneously	0,813	0,78

Abbreviations:
K: Weighted Kappa Coefficients of 0,00-0,39 were considered as weak, 0,40-0,75 as moderate and 0,76-1,00 as very strong.
PA: proportion of positive agreement calculated as the proportion of scores with positive agreement divided by the total number of scores.
The blank cells represent the cells where K could not be calculated.
The green cells represent the items with very good agreement. (K >0,81)
The orange cells represent the items with moderate to good agreement. (K > 0,61 < 0,81)
The red cells represent the items with poor to fair agreement. (K < 0,40)

Appendix Y. The general program

Aim	Position	Repetitions
Stretching		
mm.hamstrings m.Gastrocnemius m.Psoas	Long-sitting Standing Prone lying	3*(30 sec)
Pelvic & trunk control		
hip extension (bridging)	supine, knee-extension with hip adduction	3*20
hip extension (briding)	supine, knee-extension with hip abduction	3*20
sidewards sitting	knee-standing	2*20
trunk rotation	prone lying, hips extended, thighs on bal	5*10
pelvis anteversion/retroversion*	hands&knees	20
Strengthening		
plantar flexor muscles	toe standing, knees extended	
m.Obliq Abd	supine, rolling against resistance	
m.Glut Max	prone lying, hip extension	
m.Rectus Femoris&Vastii	supine, knee extension squatting	3*20
m.Glut Max	prone lying, hip extension	
Sit to stand	Sitting, 90° hip, knee & ankle flexion	20
Standing balance		
anteroposterior direction	standing, feet closed (throwing &catching balls)	20
	standing, hips abducted (idem)	20
lateral	standing, feet together (resistance against pelvis)	20
one leg standing	one-leg standing	20
Gait training		
sidewards walking	sidewards walking against a wall	20 steps
backwards walking	backwards walking	20 steps
pelvic control	shoulders externally rotated, walking between lines	20 steps
Jumping*		
forwards	standing, feet closed	5
backwards	standing, feet closed	5
sidewards	standing, feet closed	5

* More difficult, only for children with GMFCS I

Appendix Z. Example of an individually defined training program

Goal 1: E. has improved knee extension at initial contact	Position	Rep
Exercise 1: Length mm.Hamstrings	Supine lying	
	Dynamic stretch in standing	3*20 s
Exercise 2: Strengthening m.Rectus Femoris &Vastii	Long sitting, knees in extension	3*15
Exercise 3: Active hip extension	Long sitting, hips abducted, knees extended	20*
	Standing, hips and knees extended	5 min
Exercise 4: Walking exercises to improve knee ext	Walking	
Goal 2: E. has improved hip extension during gait	Position	Rep
Exercise 1: Stretching m.Psoas	Prone lying, knees flexed, hip extension	20*5s
Exercise 2: Active hip extension	Prone lying, knees flexed, hip extension	20*5
Exercise 3: Hip extension during gait	Backwards walking, guiding hip extension	5 min
Goal 3: E. can stand on one leg (L&R) for 4s	Position	Rep
Exercise 1: Weightshift	Standing	
Exercise 2: Pelvic stability exercises		
Exercise 3: Single-leg stance exercises		
Goal 4: E. can climb15 steps of the stairs, carying backpack (school)	Position	Rep
Exercises 1-3: Preparatory exercises: goal 1+2		
Exercise 4: Small step-up exercises (sidewards, backwards)		
Exercise 5: Guided stair climbing, progression towards climbing with backpack (for school) using support		

Appendix AA. Quality assessment form

Type		Position	Repetitions	Duration/intensity	Feedback Hands on/off	Attention to compensations	
Exercise 1		/2	/2	/2	/2	/2	
Exercise 2		/2	/2	/2	/2	/2	
Exercise 3		/2	/2	/2	/2	/2	
Exercise 4		/2	/2	/2	/2	/2	
Exercise 5		/2	/2	/2	/2	/2	
Exercise 6		/2	/2	/2	/2	/2	
Exercise 7		/2	/2	/2	/2	/2	
Exercise 8		/2	/2	/2	/2	/2	
Exercise 9		/2	/2	/2	/2	/2	
Exercise 10		/2	/2	/2	/2	/2	
Total		…/…	…/…	…/…	…/…	…/…	**…./….** **= …/50**
Analytical versus functional training:							**…/50**
Total quality score							**…/100**

Appendix AB. Session form

Session n° …

Date: …… from …… to ……. (therapy time: ……. minutes)

IMPAIRMENT LEVEL

Joint mobility and muscle length (total …. minutes)

Muscle group	Position	Duration of stretch	Repetitions	
M. Gastrocnemius	stance/prone/supine/sitting	*12 sec*	*3*	Active/Passive/Activopassivo/Inhibition
	stance/prone/supine/sitting			Active/Passive/Activopassivo/Inhibition
	stance/prone/supine/sitting			Active/Passive/Activopassivo/Inhibition
	stance/prone/supine/sitting			Active/Passive/Activopassivo/Inhibition
	stance/prone/supine/sitting			Active/Passive/Activopassivo/Inhibition
	stance/prone/supine/sitting			Active/Passive/Activopassivo/Inhibition

Strength: (total …. minutes)

Muscle group	Position	Resistance	Repetitions	
M. Gastrocnemius	stance/prone/supine/sitting	*Body weight*	3*15	Analytical/ Global
	stance/prone/supine/sitting			Analytical/ Global
	stance/prone/supine/sitting			Analytical/ Global
	stance/prone/supine/sitting			Analytical/ Global
	stance/prone/supine/sitting			Analytical/ Global
	stance/prone/supine/sitting			Analytical/ Global

ACTIVITY LEVEL

Functional exercises (total ….. minutes)

Describe the exercise and specific points of attention.

	Description	Attention points
Lying		
Sitting		
Standing		
Gait		
Transfer		
Trunk control		
Arm-and hand function		
Others		

PARTICIPATION LEVEL: participation to social activities (total time ….minutes)

Specific preparation for social activities:

- Home:
- School:
- Sports:
- Others:

REMARKS

Appendix AC Citations from Mr and Mrs Bobath

'*The Bobath concept is a problem-solving approach to the assessment and treatment of individuals with disturbances of function, movement and postural control due to a lesion of the central nervous system. The concept provides a way of observing, analysing and interpreting task performance. It takes into account that Cerebral Palsy and allied conditions comprise a group of symptoms in which there is a large variety. Therefore, treatment needs to be flexible and needs to be adapted to the many needs and the variety of the individual child. No standardized set of exercises will be adequate to the needs of all children.'*

' … *We learned that the background for normal children's growing ability to move against gravity and for their great variety of movement and skills was the development of righting and equilibrium reactions. This helped us towards a more dynamic treatment, i.e. facilitation of sequences of automatic movements. We found a way of using key points of control from where inhibiting of abnormal activity could be done simultaneously with facilitation of normal activity. We started to use 'reflex inhibiting' dynamic patterns rather than 'static reflex inhibiting postures'.*

(Mrs Bobath, Development of neurodevelopmental treatment, 1992)

'There needs to be opportunity for practice for learning/ re-learning to occur, either by the individual or with the help of carers. '

'Goals need to be realistic according to the client's potential and appropriate to the environment encountered during daily life. These principles integrate with the main ideas of motor learning theory, which requires the active participation of the client. This is not new. Bobath in the 1960s stated that "unless you stimulate or activate your patient in the way in which new activities are possible, you have done nothing at all. So the handling techniques as such are only the very first step in treatment, though they are very important'

(Bobath B. Bobath K. 1975. Motor development in the different types of cerebral palsy. Heinneman Medical Books, London).