DEPARTMENT OF WOMEN'S AND CHILDREN'S HEALTH
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GOAL-DIRECTED THERAPY FOR CHILDREN WITH CEREBRAL PALSY

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Nog finns det mål och mening
i vår färd – men det är vägen,
som är mödan värd.

Karin Boye 1927

ABSTRACT

The overall aims of the thesis were to study the effects of goal-directed therapy (GDT) in children with cerebral palsy (CP) in an ecological setting, using the ICF-CY as a frame of reference, and to determine the reliability of the selective motor control (SMC) scale. Measures included in this thesis were represented in the domains of body function/structure and activity and participation. A specific interest was directed towards the SMC scale, since associations with gross motor activity have previously been detected. During the past few decades, the aim with therapy for children with CP has gradually moved from normalisation to activity and participation in meaningful life situations. Modern theories of motor development, motor control and motor learning support a treatment philosophy in which children with CP are encouraged to actively search for optimal strategies to accomplish meaningful activities and are given optimal possibilities to practice in their everyday environments. Today emphasis is also made to actively include the family. There is now growing evidence in support of GDT, even though there is a variation in the therapy setting, the level of child/family's involvement in choosing the goals and the therapy duration. A specific interest were if GDT could give beneficial effects in comparison to Activity focused therapy (AT), what the long-term effects on gross motor capacity and goal attainment were and if body functions measures (SMC, Passive Range of Motion (PROM) and Modified Ashworth Scale (MAS)) were affected after GDT.

The inter-rater reliability of the SMC scale was evaluated by three assessors who simultaneously and independently scored the child's ability to dorsiflex the ankle. A consecutive sample of 40 children with CP, 3-16 years and in GMFCS I-V, was examined. The results revealed fair/good to strong inter-rater reliability. The test-retest reliability was achieved through a second examination of 29 children at a different occasion. Weighted Kappa statistics revealed strong test-retest reliability.

Comparison of GDT and AT was completed, with respect to everyday activities and gross motor capacity. Habilitation centres recruited 44 children, 1-6 years, 25 boys, GMFCS I-IV, MACS I-V, 27 bilateral CP and 17 unilateral CP. Focus with therapy in the GDT group (n=22) was directed towards individualized family selected goals with participation in group training, while focus in therapy in the AT group (n=22) was directed towards generalized aims with individualized therapy sessions. Evaluations after 12 weeks revealed significantly higher improvements in the GDT group in comparison to the AT group in both everyday activities and gross motor capacity.

Longitudinal investigation of gross motor capacity (GMFM-66) and goal attainment (GAS) in children receiving GDT (n=22) was accomplished by repeated assessments before, during and after a 12 week period of GDT. In addition, evaluations of SMC, PROM and MAS were completed before and after therapy. The assessments of gross motor capacity before GDT were stable. During and after the 12 weeks therapy, gross motor capacity improved significantly, whereas the long-term follow-up assessments did not reveal any further improvement. The goals were gradually reached to or above the expected level at the end of therapy. The long-

term follow-up assessments demonstrated a gradual progress towards higher performance, with no further change in gross motor capacity. No changes in SMC, PROM or MAS were detected.

Explorations of relationships between family selected goals and scores on standardized measures were performed using the ICF-CY as a classification system. The 110 goals from children participating in GDT were used. The meaningful concept of the expected level in the goals were coded and linked to the ICF-CY. The children's baseline assessments and change scores from standardized measures were used to explore the relationships. All GAS-goals were classified in the Activity and Participation domain within ICF-CY. The number of GAS-goals in the Mobility chapter and in the Self-Care chapter correlated to baseline scores in standardized measures. The change scores in standardized measures correlated to goal attainment in the Mobility chapter and in the Self-Care chapter.

Beneficial effects in gross motor capacity and everyday activity were detected after GDT. The SMC scale demonstrated moderate to strong reliability.

LIST OF PUBLICATIONS

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

I. Löwing K, Brogren Carlberg E. "Reliability of the selective motor control scale in children with cerebral palsy." Advances in Physiotherapy 2009; 11: 58-63

II. Löwing K, Bexelius A, Brogren Carlberg E. Activity focused and goal directed therapy for children with cerebral palsy – do goals make a difference? Disability and Rehabilitation, 2009; 31: 1808-16

III. Löwing K, Bexelius A, Brogren Carlberg E. Goal-directed functional therapy: A longitudinal study on gross motor function in children with cerebral palsy. Disability and Rehabilitation, 2010; 32: 908–916

IV. Löwing K, Hamer EG, Bexelius A, Brogren Carlberg E. Exploring the relationship of family-goals and scores on standardized measures in children with cerebral palsy, using the ICF- CY. Submitted

CONTENTS

LIST OF ABBREVIATIONS

B	Bilateral
CAS	Caregiver Assistance Scale
CI	Confidence Interval
CP	Cerebral Palsy
ES	Effect Size
FSS	Functional Skills Scale
GAS	Goal Attainment Scaling
GMFCS	Gross Motor Function Classification System
GMFM-66	Gross Motor Function Measure-66
GMFM-88	Gross Motor Function Measure-88
ICF	International Classifications of Functioning, Disability and Health
ICF-CY	International Classifications of Functioning, Disability and Health-Children and Youths version
MACS	Manual Ability Classification System
MAS	Modified Ashworth Scale
PEDI	Pediatric Evaluation of Disability Inventory
PROM	Passive Range Of Motion
RC	Relative Concentration
RP	Relative Position
RV	Relative Variance
SMC	Selective Motor Control
SMR	Standardized Response Mean
TA	Tibialis Anterior muscle
UL	Unilateral left
UR	Unilateral Right

INTRODUCTION

Throughout the years a large number of treatment options have been available for children with cerebral palsy (CP). During the last decades the mind-set towards CP has changed from focusing on the child's impairments and as the child as a passive recipient of treatment, to one with the child as an active participant in the treatment of a disability requiring a lifelong perspective. Today the ultimate aim with therapy is to promote activity and participation in everyday life according to the child's and family's priorities.

CEREBRAL PALSY

HISTORY

The first medical descriptions of cerebral palsy were made by the English surgeon William John Little (Morris, 2007). He held a series of lectures beginning in 1843, and described cerebral palsy as "a peculiar distortion which affects new born children which has never been elsewhere described,... the spasmodic tetanus-like rigidity and distortion of the limbs of new-born infants". William Little began linking the relationship of the disorder to premature and complicated births. Little proposed in his thesis that the causes for this disorder included; asphyxia at birth, premature birth and direct mechanical injury. Little further noted that the weakness of the fetus to nervous system damage depended on the stage of development. In the end of the 19th century, the disorder was commonly known as "Little's Disease". The name "cerebral palsy" was for the first time used in the late 1800's by Professor William Osler from Pennsylvania. Osler published his monograph 1889 in London, entitled "The cerebral palsies of children". Osler noted the relations between difficult delivery, asphyxia requiring prolonged resuscitation, seizures and arachnoid and subarachnoid bleeding. A few years later Sigmund Freud also described the disease. Freud agreed with William Little that difficult birth was common in children with cerebral palsy, but Freud suggested that a difficult birth might often be more of a symptom than a cause (Morris, 2007). Freud explained a relationship between lesions in the brain and the degree of paralysis and spasticity in the body. He noted that more superficial lesions caused more problems in the legs than in the arms. Freud's focus on associations between the clinical picture and neuropathology may have facilitated future research in the twentieth century (Morris, 2007).

DEFINITION

The definition of CP has varied through the years. CP is a term or a description, not a specific diagnosis (Hagberg et al., 1989). The lesions are as heterogeneous as the panorama of clinical symptoms (Hagberg et al., 1993;Krageloh-Mann et al., 1995). The disturbed motor function has been central in the various definitions of CP and all children with CP exhibit dysfunction in the control of movement and posture. Commonly occurring motor symptoms are spasticity, muscular weakness, decreased range of motion, dyskinesia, and problems with selective motor control of muscles.

A commonly used definition of CP during the last decades was formulated by Mutch and colleagues; " an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of development" (Mutch et al., 1992). As the cause of CP is a lesion to the immature brain, other aspect of functioning can also be affected, which in a lifespan perspective can extensively affect the individual. In the recently presented definition of CP the accompanying disturbances are now included and formulated: "Cerebral Palsy describes a group of disorders of the development of movement and posture, causing activity limitations, which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbance of sensation, cognition, communication, perception, and/or behaviour, and/or by a seizure disorder" (Rosenbaum et al., 2007). The new definition of CP covers a broad range of clinical presentations and degrees of activity limitation. It has therefore been proposed to further categorize individuals with CP into classes or groups (Rosenbaum et al., 2007).

PREVALENCE

The prevalence of CP in the Western world is reported to be approximately 2-3/1000 live births (Himmelmann et al., 2005;Bhasin et al., 2006;Westbom et al., 2007). Children with CP constitute the largest group of children with severe physical disabilities (Koman et al., 2004). In the western Swedish health care region, the prevalence of CP has been registered since 1954 by Hagberg and colleagues (Hagberg et al., 1996). The prevalence has varied through the years. During the 1960s the prevalence decreased due to advances in maternal health care and improved neonatal intensive care, but in the 1970s the prevalence increased, due to the increased fetal/newborn survival rate associated with improved obstetric and neonatal care. A recently published paper showed the prevalence in western Sweden to be 2.18 per 1000 live births (Himmelmann et al., 2010).

AETIOLOGY

CP is a term covering lesions occurring in the immature brain during pregnancy and at up to two years of age. The lesions are infrequently caused by a single factor, and several interacting factors have been discussed in order to explain the aetiology, such as infection and inflammation, asphyxia, infarctions, haemorrhage, environmental and genetic factors, growth restrictions and multiple pregnancies (Stanley et al., 2000). There are three factors of importance for the type of brain pathology: the maturity of the central nervous system at the time of the incidence, the size of the damage and the duration of the insult. The timing of the insult to the immature brain determines where the damage occurs due to selective vulnerability. An insult before the gestational age of week 20 predominantly results in migration disorders, between week 24-34 often results in white matter damage as a result of periventricular haemorrage (PVH) or periventricular leukomalacia (PVL), and an insult between week 38-42 commonly implies damage to the basal ganglia. A damage to the brain which occurs between week 35 and up to two years of age gives diffuse or focal damage to cortex (grey matter). Therefore children with CP have a large variety of symptoms.

Today advancement in imaging studies has contributed to growing knowledge of the underlying causes and timing of events (Bax et al., 2006;Krageloh-Mann and Horber, 2007b;van Haastert et al., 2008). In a review article, Krägeloh-Mann and Horber reported abnormal MRI in 86% of children with a diagnosis of CP. PVL was detected most frequently (56%), followed by cortical and grey matter lesions (18%). Brain maldevelopments were observed in only 9% of the children. In children with CP born preterm, PVL was detected in 90% in comparison to 20% in children carried to term (Krageloh-Mann and Horber, 2007a). In the European Cerebral Palsy study, whose aim was to find preventive strategies, 431 children were assessed clinically and parental questionnaires were collected. The results demonstrated a high rate of infections reported by mothers during pregnancy (Bax et al., 2006).

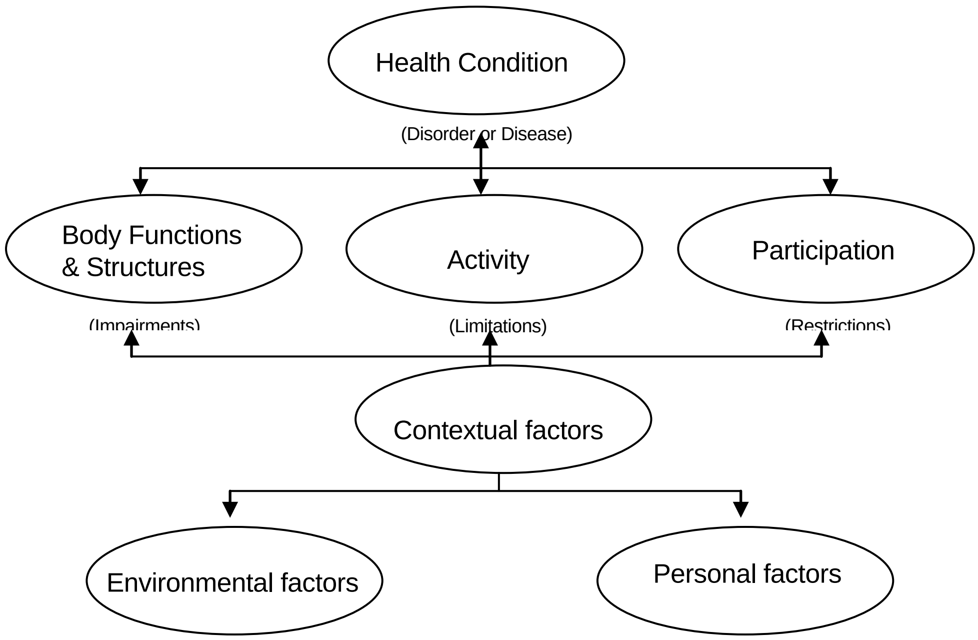

Figure 1. The International Classification of Functioning, Disability and Health model. Part one includes the components of Body Functions & Structures and Activity and Participation. Part two includes the components of contextual factors; Environmental factors and Personal factors.

INTERNATIONAL CLASSIFICATION OF FUNCTIONING, DISABILITY AND HEALTH

The International Classification of Functioning, Disability and Health (ICF) (Word Health Organization (WHO), 2001) can be used as a classification system to identify and sort the various motor disorders and accompanying disturbances that can be observed in children with cerebral palsy. As this group of children is heterogeneous and multifaceted, ICF can be used to produce a more complete picture of a child in the context of daily living, in which the physical environment and attitudes in society are also taken into account. ICF intends to cover the entire world population, not only people with disabilities or even more specifically CP, and recognizes disability as a universal human experience. The ICF shifts the focus from cause to impact by describing functioning and health conditions from various perspectives. Functioning is an umbrella term covering all body functions, activities and participation and analogously disability serves as an umbrella term for impairments, activity limitations and participation restrictions (Figure 1). In the ICF contextual factors represent the background of an individual's life and functioning and include environmental factors and personal factors. Environmental factors make up the physical, social and attitudinal environment that can influence a child's functioning. The umbrella terms ´functioning´ and ´disability´ are considered as a dynamic interaction between health conditions and contextual factors. In this light the ICF can be seen as a framework of human functioning (Figure 1). An important advantage is that the definitions are well described and today knowledge is growing about the various concepts. Body functions affected in children with cerebral palsy could for example be muscle strength, ability to selectively control muscles, coordination, sensibility, vision, muscle tone and range of motion. Activity is defined as an individual's execution of a task or action and could be dressing, walking, eating and reading. Participation is defined as involvement in a life situation, and examples are playing and participating in social and community roles. In this thesis no distinction between Activity and Participation will be made; Activity and Participation will be considered as a unit. Capacity is defined as an individual's ability to execute a task or an action within a standardized environment and performance is defined as what an individual "does do" in his or her current environment (Lollar and Simeonsson, 2005). There have been suggestions of including a third term; capability, which could be defined as an individual's capacity to execute a task in his or her current environment (Holsbeeke et al., 2009). In this thesis the distinctions between capacity and performance will be applied. The word "capability" will be used in the context of the Pediatric Evaluation of Disability Inventory (PEDI), in which the scoring criteria for capability is described as "capable of performing the item in most situations" in the everyday environment, which differs from the definition in ICF (Ostensjo et al., 2006).

The original ICF was considered to not sufficiently describe functioning in children and youth. The International Classification of Functioning, Disability and Health – Children and Youths version (ICF-CY) (Word Health Organization (WHO) and WHO Workgroup for Development of ICF for Children and Youth, 2007) is the extended version with details to describe the development of children and youth where special attention is given to aspects of learning, behaviour and development (Lollar and Simeonsson, 2005). Assessment of a child with cerebral palsy often exposes needs beyond the health condition and calls for the inclusion of other disciplines in treatment planning, including therapies, education, and social welfare (Lollar and Simeonsson, 2005). The aim of ICF and ICF-CY is to provide a unified and standard framework and language for description of health status (Cieza et al., 2005). Sharing a common conceptual approach and congruent terminology is important among multi-professional teams, across disciplines and across service systems (Lollar and Simeonsson, 2005). In order to optimize opportunities to practice in the daily environment, communicating with a shared language is even more central (Palisano and Murr, 2009). The use of ICF-CY also offers the child and family an opportunity to achieve an overall picture and thereby take part in decision making. Families often are exposed to various interventions suggested by specialists aiming to assist the child, based on their specific professions. Viewing the child in his or her context of everyday functioning and considering the needs of the child and family can facilitate realistic decision making in a respectful and mutual team together with the child and family. The ICF-CY model supports a vision with equal value on promoting activity and facilitating the child's participation in all aspects of life. The use of the ICF-CY framework provides an opportunity to work towards goals that address activity and social engagement (Beckung and Hagberg, 2002;Rosenbaum and Stewart, 2004).

BODY FUNCTIONS

Historically the focus in children with CP was the motor disorder, but gradually emphases directed towards accompanying disturbances have occurred. Such disturbances affect more than 50% of children with CP, both primarily but also by interacting with the motor disorders (Beckung et al., 2008;Enkelaar et al., 2008). Behaviour problems in children with CP have been found more frequently among children with epilepsy and learning disability, and affect both the child and the families' everyday life (Raina et al., 2005;Carlsson et al., 2008). In this thesis a few body functions related to the motor disorder will be discussed, namely, ability to selectively control muscles, passive range of motion and spasticity.

Selective motor control of muscles has been defined as "the ability to isolate the activation of muscles in a selected pattern in response to demands of a voluntary movement or posture" (Sanger et al., 2006). Selective motor control of muscles has, during the last decades, attracted increased attention, and interesting relations to gross motor activity were observed (Ostensjo et al., 2004;Fazzi et al., 2005;Desloovere et al., 2006;Voorman et al., 2007). A growing awareness of the importance of selective motor control of muscles has been observed both in research and in clinical practice (Kerr and Selber, 2003) and various scales has been developed (Boyd and Graham, 1999;Trost, 2004;Fowler et al., 2009). One of them explicitly assesses and grades which muscles that are primarily activated during dorsiflexion in the ankle, the Selective Motor Control scale (SMC scale) (Boyd and Graham, 1999). The scale is a five graded, ordinal scale and the scoring is made by observation. The objective of using the scale is to register whether the child can activate specific muscles in isolation. The best grade is given if the child can activate m. tibialis anterior, a muscle considered to rely on activation of motor cortex and transmission through the cortico-spinal tract to the target muscle (Petersen et al., 2003). The aetiology of loss of selective motor control of muscles is not yet fully understood (Rose, 2009;Dobson, 2010).

Monitoring passive range of motion (PROM) is of great interest in children with CP, as contractures and muscle stiffness occur in many children and the risk increases with age (Lin, 2003). In a recently published study, the range of motion was found to decrease in children with CP from the age of 2 years to 14 years (Nordmark et al., 2009). The explanations for the arising contractures have varied, but a main assumption has been that "contractures are caused by spasticity" (Mayer et al., 1997;Gormley, Jr. et al., 1997). During the last decades objections were raised to this statement and other theories were proposed. Lin stated that “contractures are posture dependent and arise through disuse and weakness” (Lin, 2003), whereas others have argued that activation pattern of motor units and fibre-types are important factors in muscle deformity and weakness (Gaough M 2009). Recent research has further questioned the influence of spasticity on contracture development, since reduced spasticity and decreased PROM were both observed in a longitudinal follow-up study after botulinum toxin treatment (Tedroff et al., 2009). A consensus has not been reached and the question calls for further research. Spasticity was, for a long time, considered as the main impairment in cerebral palsy, but was at that time often defined as a summation of various symptoms like asymmetry in posture, weakness, absence of selective motor control of muscles and clonus. In the definition by Lance (1980) spasticity was defined as “velocity-dependent increase in the tonic stretch reflexes with exaggerated tendon jerks, resulting from hyperexcitability of the stretch reflex” (Lance, 1980).

Spasticity is commonly assessed by a modified Ashworth scale (MAS) wherein the examiner grades the resistance to passive muscle stretch during high velocity motion (Peacock and Staudt, 1990). In a recently published study, plantarflexor spasticity was found to increase in children with CP up to four years and thereafter decrease up to twelve years of age (Hagglund and Wagner, 2008). However, questions have been raised concerning assessments of spasticity, as discrepancies have been observed between the signs of spasticity during activity and the signs of spasticity in a resting position during testing conditions (Crenna and Inverno, 1994;Poon and Hui-Chan, 2009). Lack of congruence between the definition and the methods of measuring spasticity have also been observed (Malhotra et al., 2009).

Activity and Participation

Since children with cerebral palsy comprise a very heterogeneous group, classifications systems have been developed. Children’s gross motor function can be classified by the use of the Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997). The classification is divided into five levels, spanning from children in level I who walk without restrictions to children in level V who have severely limited self-mobility (Figure 2). To accommodate the changing abilities at different ages, the system describes gross motor function for five age intervals: less than 2 years, 2 to 4 years, 4 to 6 years, 6 to12 years, and recently the interval 12 to 18 years was described and included in the expanded and revised version (GMFCS – E & R) (Palisano et al., 2008). The GMFCS has good reliability for children between 2-12 years of age and the classification is of prognostic value (Palisano et al., 1997;Beckung and Hagberg, 2002;Ostensjo et al., 2003;Hanna et al., 2008).

Activity can be expressed as either capacity or performance (ICF-CY). Among children with CP, assessment of gross motor capacity is frequently administered and evaluated by the use of the Gross Motor Function Measure (GMFM) (Russell et al., 2002). A prognosis of overall functioning is difficult to give to parents of a young child with cerebral palsy; there are still many developmental areas that must be investigated in a long-term perspective, and furthermore, there are many factors interacting with each other. The development of the gross motor function can however to some extent be predicted through the gross motor function curves (Rosenbaum et al., 2002). Repeated measures with GMFM served as a base to build the GMFCS (Palisano et al., 1997;Palisano et al., 2008) (Figure 2) as well as the gross motor growth curves. The gross motor growth curves serve an important purpose as they allow realistic expectations and thus can form a basis for goal-setting in therapy. Recently cross-sectional reference percentiles for GMFM-66 within the levels in GMFCS were developed. They allow a comparison with a normative score for each child's GMFCS level with respect to the age of the child (Hanna et al., 2008). A longitudinal follow-up of children's gross motor activity, measured by GMFM-66, has recently been presented in which a decline was observed in adolescents and young adults in GMFCS levels III, IV and V (Hanna et al., 2009). The results warrant attention and raise several questions concerning whether there are specific factors that could explain the decline and whether they can be prevented.

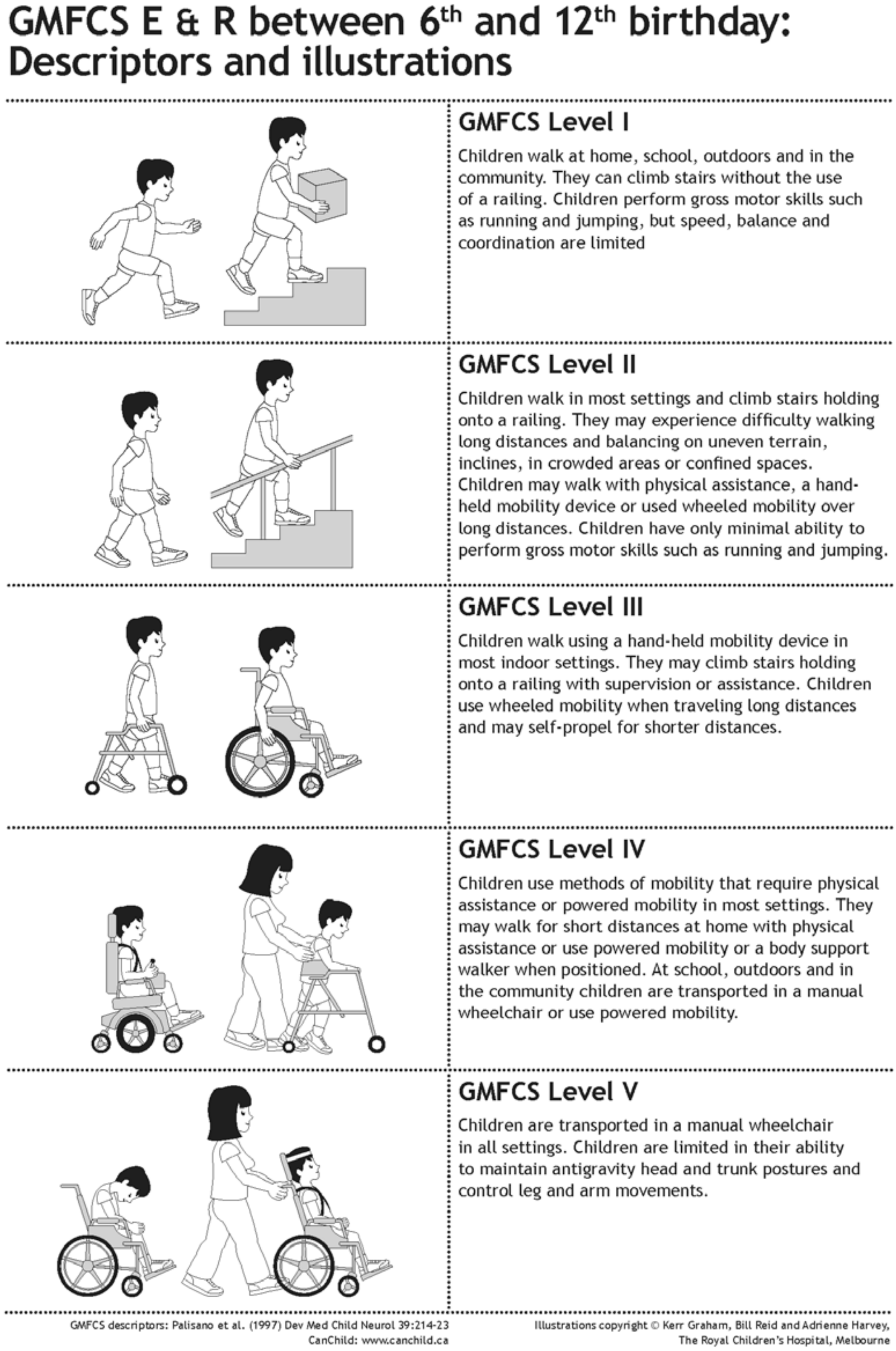

Figure 2. Gross Motor Function Classification System, GMFCS – E & R (Palisano et al 1997, 2007). The illustrations are reprinted with permission from Bill Reid and Kerr Graham.

The PEDI is commonly used to capture and evaluate the child's everyday performance. PEDI is a parental interview, in which parents are asked questions about their child's activities in everyday situations concerning self-care, mobility and social function and their requirements of caregiver assistance and modifications(Haley et al., 1992). PEDI was found to strongly reflect aspects of everyday functioning in young children with CP (Ostensjo et al., 2003). PEDI and GMFM are frequently used both in clinical practice and in research in children with CP as they give complementary information about gross motor capacity and mobility (Engelen et al., 2007). Used together they cover the child's capacity (what the child can do in a standardized environment, GMFM), capability (PEDI terminology; what the child can do in the everyday environment) and performance (what the child does do in the everyday environment) (Vos-Vromans et al., 2005;Engelen et al., 2007). The PEDI and the GMFM are also found to be sensitive enough to capture changes in mobility in especially children younger than four years (Vos-Vromans et al., 2005).

Manual ability has received relatively less attention in comparison to gross motor activity (v Meeteren et al 2008), somewhat surprisingly, as restrictions in manual ability have been observed in more than half of the group of children with CP (Arner et al 2008). The Manual Ability Classification System (MACS) for children with CP makes it possible to classify children's ability to handle objects in daily activities (Eliasson et al., 2006). MACS is divided into five levels, from level I in which the child handles objects easily and successfully, to level V in which the child does not handle objects (Table I). The knowledge about the child's actual performance in daily life is required when determining the level (Eliasson et al., 2006). MACS is reported to be a valid and reliable classification (Eliasson et al., 2006;Morris et al., 2006), even though caution is recommended when classifying children younger than two years (Plasschaert et al., 2009). Longitudinal data of hand-function in children with unilateral CP have been presented, in which children from 18 months of age were included and followed up to the age of 8 years. The participants were assessed with the Assisting Hand Assessment (AHA), a recently developed test aiming to measure and describe how effectively children with a unilateral disability in arm and hand use their assisting/affected hand during bimanual tasks (Krumlinde-Sundholm et al., 2007). The results demonstrated that children with relatively high scores at the 18 month examination reached a higher ability level at the final assessment, indicating that the scores in AHA at 18 month can be useful for prediction of development (Holmefur et al., 2010).

Table I. The five levels in the Manual Ability Classification System (MACS).

MACS	
I	Handles objects easily and successfully
II	Handles most objects but with somewhat reduced quality and/or speed of achievement
III	Handles objects with difficulty; needs help to prepare and/or modify activities
IV	Handles a limited selection of easily managed objects in adapted situations
V	Does not handle objects and has severely limited ability to perform even simple actions

RELATIONS BETWEEN BODY FUNCTIONS AND ACTIVITY AND PARTICIPATION

The ICF states that "an individual's functioning in a specific domain is an interaction or complex relationship between the health condition and contextual factors" (Word Health Organization (WHO), 2001). However, although there are bi-directional arrows linking the components in ICF (Figure 1), one can not assume a linear relationship (Darrah, 2008). Today there is a growing evidence concerning relationships between various domains within the ICF. The relationships between spasticity and gross motor activity and gait have been studied, and spasticity was reported to only explain a minor part of the variations in gross motor activity and gait, whereas strength was found to explain the predominant part (Ross and Engsberg 2007). In a longitudinal study of young children with CP, the relationship between spasticity and gross motor development were followed during one year. The results demonstrated that spasticity explained 8% of gross motor development, and the authors concluded that spasticity was only marginally related to gross motor development and that there were many other factors contributing to gross motor development (Gorter et al 2009).

Interesting relationships have also been observed between other body functions and structures. Beckung and collaborators found that intellectual disorder, epilepsy and severe visual and hearing disorder were significantly correlated to walking ability in children with CP (Beckung et al., 2008). Beckung and Hagberg studied associations between body functions, activity and participation in children with CP in ages 5-8 years. They discovered a strong relationship between learning disability, activity limitations and participation restrictions (Beckung and Hagberg, 2002). A recently published study reported limited manual ability in many adolescents with cerebral palsy and that limitations in manual ability were strongly related to limitations in everyday activities (van Eck et al., 2010). Others have explored the relationship between motor activity and mental functions in young children with CP and found a clear association. However, when there was dissimilarity it was always in favour of mental functions in relation to motor activity (Enkelaar et al., 2008). In a longitudinal study of gross motor activity in which a group of children aged between 11 and 13 years were followed during two years, selective motor control in the dorsiflexors of the ankle was found to be the most important predictor of the course in gross motor activity (Voorman et al., 2007). The result corroborates earlier findings by Østensjø and collaborators who also identified selective motor control of the dorsiflexors as the strongest predictor (in comparison to spasticity and range of motion) of variations in gross motor activity (Ostensjo et al., 2004).

Attention has also been directed towards the relationship between gross motor capacity and performance in everyday life (Tieman et al., 2004;Smits et al., 2009;van Eck et al., 2009). Smits and collaborators analyzed the GMFM-66 and PEDI mobility domain scores of 116 children with CP. They found that capacity, measured by GMFM-66, to a high extent could explain the everyday mobility in especially the group of children with bilateral CP (Smits et al., 2009). Similar results were demonstrated in a study in which 104 children, aged 11 -13 years, were followed during three years. The authors detected longitudinal relationships between motor performance and capacity. Their analysis revealed a relation of 1:4, meaning that to achieve 1 point change in performance required 4 points change in capacity (van Eck et al., 2009). Gross motor capacity and mobility performance were also compared across home, school and outdoors settings in 307 children with CP, and significant differences in performance across the various settings were discovered (Tieman et al., 2004). Furthermore Palisano and colleagues described common mobility methods at home, school and outdoors in a group of 636 children with CP. They discovered a discrepancy in the need for caregiver assistance in relation to the environmental setting, in which the outdoor setting required highest amount of caregiver support whereas less assistance was required at home (Palisano et al., 2003). The recent development of the Functional Mobility Scale (FMS) has made it possible to capture this variation across various environmental settings (Graham et al., 2004).

THEORETICAL FRAMEWORK

THEORIES OF MOTOR CONTROL AND MOTOR DEVELOPMENT

Motor control is defined as the ability to regulate or direct the mechanisms essential to movement (Shumway-Cook and Wollacott, 2007). A variety of theories to describe sensory-motor control and the control of aberrant sensory-motor function exists (Shumway-Cook and Wollacott, 2007). The theories put different weight on the various mechanisms or systems that regulate the control of movement, i.e. the central nervous system, the biomechanics of the body and factors related to the environment and how these systems interact. The prevailing theories can generate practical applications and thus have an impact on treatment approaches (Shumway-Cook and Wollacott, 2007). Bernstein’s theory, called the systems approach, became known to the Western world in 1967 when his book “The co-ordination and regulation of movement” was translated into English. His theory has had large influence on today’s therapeutic approaches in different movement disorders. In his theory the neural organization of sensory-motor control is in focus but he also stressed the importance of additional factors like inertia, reactive forces and degrees of freedom, contributing to the performance of motor control (Shumway-Cook and Wollacott, 2007).

Theories on motor development also span from directing an emphasis on genetic factors regulating e.g. the maturation of the central nervous system, to stressing the significance of the environment on the child’s development. Currently sensory-motor development is viewed as emerging from an interaction between genetically pre-determined neural networks and the environment represented by for example the neuronal group selection theory (NGST) (Sporns and Edelman, 1993). NGST proposes two phases of variability in motor development, a primary and a secondary phase. The primary variability is not tuned to specific details in the environment, meaning that identical afferent input produces a variable motor output. From these variable motor behaviours, a selection (based on afferent information) of the networks, generating the most favourable motor behaviours, takes place. A secondary variability phase appears later, in which the motor behaviours are based on situation-specific connections of neuronal groups to achieve an efficient motor control (Forssberg, 1999;Hadders-Algra, 2000a). The second phase occurs in a task specific time, like heel-strike in walking, which appears after the second year in children with typical development. The different theories on motor development influence therapeutic strategies. In NGST therapy focuses on producing and learning variable motor strategies optimally adapted to the child's environment (Hadders-Algra, 2000b).

By using a model, viewing the activities of the child as a result of an interaction between the child, the task and the environment, different theories of motor control and motor development can be applied and tested (Shumway-Cook and Wollacott, 2007) (Figure 3). This interaction can also serve as a framework during motor learning in which different parameters related to the task, the environment and the child can be adjusted to facilitate the child's learning process.

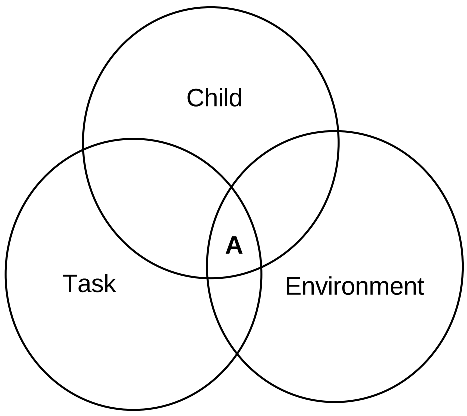

Figure 3. The activity (A) appears from interactions between the child, the task and the environment. (Modified from Shumway-Cook & Wollacott 2007)

THEORIES OF MOTOR LEARNING

Learning has been defined as the process of acquiring knowledge about the world, while motor learning is defined as a set of processes associated with practice or experience leading to relatively permanent changes in producing a skilled task (Shumway-Cook and Wollacott, 2007). The basis for learning is plasticity, defined as the ability to show neural modifiability, or changes in the efficiency or strength of synaptic connections towards long-term structural changes in the organization and numbers of neurons connected (Shumway-Cook and Wollacott, 2007). Learning can be considered as a process in which the outcome of this process is the memory of the knowledge or the ability (Kandel et al., 2000). There are several theories describing the process of learning new skills, one of them being the "three-stage" model of Fitts and Posner (Fitts and Posner, 1967). Learning, according to this model, starts with the cognitive stage, in which understanding of the task is in focus. The second stage is called the associative stage and contains refinement of the skill. The third stage is the autonomous stage, in which performance is achieved with low degree of attention and the performer can pay attention to other aspects simultaneously. Another model of learning connects to Bernstein's theory of motor control and is called the "systems three-stage theory" in which emphasis is directed to controlling the degrees of freedom during skill acquisition (Vereijken et al., 1992). The stages illustrate the learning procedure from the "novice stage", in which the degrees of freedom are few and the learner simplifies the task by coupling multiple joints, requiring high energy costs. The second stage is called the "advanced stage", during which the performer increases the degrees of freedom by controlling the joints independently in response to changes in the task or the environment. The final stage is the "expert stage", in which the performer engages all available degrees of freedom and the task is efficient, well-coordinated, and energy inexpensive (Vereijken et al., 1992). During skill acquisition, implicit and explicit processes operate in parallel. The implicit processes concern the dynamics of force generation, whereas the explicit processes are related to the performer and the environment and the mapping of the movements' shape-structure (Vereijken et al., 1992). These various characteristics of the learning process are important to have in mind during therapy, since the various stages require different support and feedback adapted to the child's strengths and difficulties.

Today there is evidence supporting a learning approach where practice should be performed in the everyday environment of the child (Newell, 1991;Campos et al., 2000). Learning in a challenging, engaging, motivational and supporting context also encourages the learning procedure (Biggs, 1999;Shepherd and Carr, 2006). The delivery, structure and amount of feedback have to be considered since interfering with the learning procedure has been observed to differ in children from that of adults (Thorpe and Valvano, 2002;Sullivan et al., 2008;Hemayattalab and Rostami, 2010). The effects of negative versus positive feedback also differ between children and adults. Children performed better after receiving positive feedback, while adults preferred information regarding their errors (negative feedback) (van Duijvenvoorde et al., 2008). The importance of expectations with respect to each child's actual capacity was pointed out a long time ago by Vygotsky (Chaiklin, 2003). He described, "*the distance between the actual developmental level as determined by independent problem solving and the level of potential development as determined through problem solving under adult guidance or in collaboration with more capable peers*", often referred to as "Zone of Proximal Development", in which he discussed the importance of supportive adults or peers in the process of learning new challenging skills (Chaiklin, 2003). This way of gradually increasing the expectations is in agreement with the goal-setting theory (Locke, 1968).

GOALS

To adjust the degree of difficulty according to a specific child's strengths and weaknesses, individual goals can be used. Goals can also help the child and family to prioritize learning new and meaningful tasks during specific time periods. For some children important gains observed by caregivers after interventions may not be possible to capture by standardized measures (McLaren and Rodger, 2003;Thomas-

Stonell et al., 2009) while scaling of individualized goals makes it possible to measure outcome (Hanna and Rodger, 2002).

The goal setting theory is based on the hypothesis that human behavior is purpose-driven and guided by an individual´s goals. The central point is that goals affect performance by focusing attention, directing effort, increasing motivation and enabling the development of strategies to achieve the goal (Locke, 1968). Goals should be Specific, Measurable, Achievable, Relevant and Time-limited (SMART) (Bovend'Eerdt et al., 2009). Siegert and Taylor emphasized the importance of the context in which goal setting takes place and the influence of factors relating to family background and social support network (Siegert et al., 2004). The authors also point out the close connection between goals and motivation (Siegert and Taylor, 2004). Today goal setting is commonly used in therapy with children with CP (Bower et al., 1996;Bower et al., 2001;Ketelaar et al., 2001;Ahl et al., 2005;Odman and Oberg, 2006;Law et al., 2007;Darrah et al., 2008;Nijhuis et al., 2008;Ostensjo et al., 2008;Wiart et al., 2010;Sorsdahl et al., 2010). Involvement of the child and family in collaboration with the multidisciplinary team to obtain the goals is required (Lin, 2003). However, the extent of involvement of the child and family demonstrates a large variation (Bower et al., 1996;Bower et al., 2001;Ketelaar et al., 2001;Ahl et al., 2005;Odman and Oberg, 2006;Law et al., 2007;Darrah et al., 2008;Nijhuis et al., 2008;Ostensjo et al., 2008;Wiart et al., 2010;Sorsdahl et al., 2010).

FAMILY-CENTERED SERVICE WITHIN AN ECOLOGICAL APPROACH

Family-centered service (FCS) is a service model in which parental needs and expectations are reflected upon (Dunst and Raab, 2004;Rosenbaum, 2004;Bamm and Rosenbaum, 2008;Palisano and Murr, 2009). The model includes an approach towards the family which is built on three principles - respect that parents know and want the best for their child, acknowledgment that every family is unique and recognition that optimal development occurs within a supportive family and a community context. A FSC approach offers a perspective in which the child and the biological dimensions of the child’s status are important but the needs of the parents and the family are central to incorporate. Scrutton states: “ It is now generally acknowledged that the efficacy of any physical treatment lies within the child´s day-to-day environment, and to pretend otherwise is a professional conceit” (Scrutton, 2004). Knowledge is also established about the fact that the well-being of families is essential to the well-being of the child (Rosenbaum, 2004). When the aim of therapy is to promote activity and participation in everyday life, it becomes apparent to include the context.

In the Ecological Systems theory the child’s development is perceived within the context of the system of relationships that form the child’s environment. Focus is on the interaction between various systems - the child’s maturing biology, the family, the community, and the societal environment that all are important for the child’s development (Bronfenbrenner, 2005). An ecological approach is in accordance with ICF-CY, where the child’s family and the environment are seen as important in supporting and challenging the child in the context of everyday situations. To optimally coordinate services, good team collaboration is needed. The key features in this collaborative process were considered to be communication, decision making, goal-setting, organization, team process and parent involvement (Nijhuis et al., 2007). Today many children with cerebral palsy receive extensive treatment which could create a “treatment dilemma”. Any treatment regime will in some way limit the child’s freedom and to some extent, take time from practicing in everyday situations (Scrutton, 2004). However considering this dilemma with respect forces professionels to observe the child’s overall situation and to reflect whether a new intervention really “adds” more than it “costs” for the child and the family (Gibson et al., 2009).

TREATMENT AND INTERVENTIONS

During the Second World War, rehabilitation of wounded soldiers started in England and demonstrated clear beneficial effects. Soon rehabilitation for adults developed and the ideas that evolved were also considered to suit children with cerebral palsy well. In the 1950s children with CP in Sweden had the right to use physical therapy and have, since the time period around 1960, been referred to habilitation units. Today habilitation centers offer a multi-professional service with a family-centered service approach with aims to facilitate everyday life for individuals with disabilities and to offer ample possibilities for participation in life situations (Handikapp och Habilitering, 2010). Previous treatment approaches were directed towards ’normalizing’ the child’s motor functions by reducing the child’s neurological deficits (Valvano, 2004). Focus in therapy was on the impairments in body functions and structures, and by treating them, the hypotheses was that the distance towards normality decreased. The approach is sometimes referred as a “bottom-up approach” (Gibson et al., 2009) and relied on the prevailing reflex/hierarchical theories. During the last decades focus has been moved and new treatment approaches have evolved (Bower et al., 1996;Bower et al., 2001;Ketelaar et al., 2001;Ahl et al., 2005;Law et al., 2007;Ostensjo et al., 2008;Sorsdahl et al., 2010).

A shift has gradually occurred in therapy, where the child now is given the possibility to be an active problem solver in the context of the day-to-day environment. Modern theories of motor development, motor control and motor learning support a treatment philosophy in which children with CP are encouraged to actively search for optimal strategies to accomplish meaningful activities and in which the children are given optimal possibilities to practice (Fitts and Posner, 1967;Vereijken et al., 1992;Gentile, 1998;Gibson and Pick, 2000;Hadders-Algra, 2000b;Brogren et al., 2001;Valvano, 2004;Shumway-Cook and Wollacott, 2007). The treatments are referred as being “top-down approaches” (Gibson et al., 2009). Studies in which the effect of a goal-directed approach has been investigated show promising results (Ketelaar et al., 2001;Ahl et al., 2005;Ostensjo et al., 2008;Wiart et al., 2010;Sorsdahl et al., 2010). The present studies however exhibit some dissimilarities concerning the setting in which therapies are conducted, the integration of practice in the child’s everyday environment and the extent to which families are involved in the goal-setting procedure. A specific interest was if GDT could give beneficial effects in comparison to AT and if there were long-term effects on gross motor capacity and goal attainment and if body functions measures (SMC, PROM, MAS) were affected after GDT.

AIMS

The overall aim of the thesis was to study the effects of goal-directed therapy in children with cerebral palsy in an ecological setting, using the ICF-CY as a frame of reference and to determine the reliability of the SMC scale

The specific aims of the studies were

To determine the inter-rater and the test-retest reliability of the SMC-scale in children with cerebral palsy in a clinical setting

To compare goal-directed functional therapy to activity-focused therapy with respect to achievement of everyday activities and gross motor function

To investigate gross motor function and goal attainment in children with cerebral palsy before, during and after goal-directed functional therapy, and to evaluate body functions, and explore relationships between gross motor function, goal attainment and body functions

To explore the relationships of family-selected GAS goals and scores on standardized measures using the ICF-CY as a classification system

METHODS

STUDY OUTLINES

STUDY I

A consecutive sample of 40 children with CP participated in the reliability study during May-December 2005. The children were assessed in conjunction with an ordinary visit at the physical therapy department at Astrid Lindgren Children's Hospital. The children's selective motor control of the ankle dorsiflexors was assessed by three physical therapists (PTs) simultaneously in the inter-rater reliability test. The scoring was performed independently and the PTs were blinded to each other's ratings throughhout the data collection. To evaluate the test-retest reliability, 29 of these children were assessed on an additional occasion by PT number 3. After the final data collection, all data were analyzed with Kappa statistics, percentage agreement and evaluations of the degree of random or systematic differences in disagreements (Table II).

STUDY II

Forty-four children with CP participated in the intervention study comparing Goal Directed Therapy (GDT, n=22) and Activity Focused Therapy (AT, n=22) during 2004-2006. All children were integrated in preschools at the local communities. Irrespective of which therapy approach the children received, the therapy was carried out by the local multi-professional team from their local habilitation center. Assessments were performed before and after a twelve-week intervention period by the same PTs (KL, AB), who did not participate in the therapy. The children's gross motor capacity was assessed by GMFM-66 and the children's everyday performance was assessed by PEDI. In addition goal attainment was evaluated in the GDT approach (Table II).

STUDY III

Twenty-two children with CP participated in the GDT approach and were followed longitudinally before, during and after the 12-week therapy period.

The therapy took place in the child's everyday environment and focused on the children's individual goals. Each child had five goals, graded according to GAS. Once a week the children participated in group meetings lead by multi-professional teams. Evaluation of gross motor capacity was performed at seven occasions. Goal attainment was evaluated during and after GDT. Furthermore, assessments of body functions (i.e. SMC-scale, PROM and spasticity) were performed before and after GDT (Table II).

STUDY IV

Twenty-two children with CP participating in the GDT approach were included. GAS-goals (n=110) of the 22 children were coded and linked to the ICF-CY by two researchers independently (KL, EGH). The meaningful concept of the description of the expected level in the GAS goal was linked to the most suitable ICF-CY code. The children's levels in GMFCS and MACS, the baseline assessments and change scores from PEDI and GMFM-66, Goal attainment and Goal-chapter were used to explore the relationships (Table II).

Table II. Aims, study designs and main data analyses in the four studies in the thesis. Selective motor control scale (SMC), Relative rank Variance (RV), Relative Position (RP), Relative Concentration (RC), Effect size (ES), Goal directed therapy (GDT), Standardized response mean (SRM) and International classification of disability and Health (ICF-CY).

STUDY	AIM	STUDY DESIGN	DATA ANALYSIS
I	Examine the inter-rater and test-retest reliability of the SMC scale	Observational study	Descriptive statistics, Kappa statistics, RV, RP, RC
II	Compare two therapy approaches	Intervention study	Descriptive statistics, T-test, correlation, linear regression, ES
III	Evaluate gross motor activity and goal attainment before, during and after GDT and evaluate measures of body functions	Prospective longitudinal study	Descriptive statistics, Mixed Linear Model, T-test, Wilcoxon, SRM, correlation
IV	Explore the relationships of family-goals and standardized measures by the use of ICF-CY	Explorative study	Descriptive statistics, Kappa statistics, correlation

PARTICIPANTS

Study I. A consecutive selection of 40 children with cerebral palsy was recruited in conjunction to a regular consultation at Astrid Lindgren Children's Hospital. The children were from 3-16 years of age (mean 8 years 6 months, median age 7 years). They were classified in GMFCS level I-V, 18 children were girls and 22 were boys, 30 children had a bilateral CP and ten had a unilateral CP (Left:6/Right:4). Exclusion criteria were orthopedic surgery with muscle transfer around the ankle. Twenty-nine of the children had an additional consultation at the hospital during the time period of the study and were repeatedly assessed in the test-retest examination (Table III).

Study II. All 13 habilitation centers in Stockholm were invited to participate. They recruited 44 children with spastic CP from their catchments regions (Table III). The children were 1-6 years of age (mean 4 years 1 month, SD 1 year 5 months) and 19 were girls. They were classified in GMFCS levels I-IV and in MACS levels I-IV. A unilateral CP was present in 17 children (Left: 8, Right: 9), and a bilateral CP, in 27 children. All children were able to understand uncomplicated instructions and were integrated at pre-schools in the local districts. Exclusion criteria were orthopedic surgery or extensive treatment with other interventions during the time of the study. A randomization was not performed. The selection of therapy approach was a result of which habilitation centers that could recruit sufficient number of children from their catchments areas to arrange groups of children (7-8), as the GDT included group training. Hence, the child could remain in her ecological environment and avoid long distance traveling. In total three groups of children (n=22) were organized and participated in the GDT approach. The habilitation centers who not could arrange groups of children all together recruited 22 children, and they received the conventional Activity Focused therapy (AT) from their local habilitation centers. Consequently, these children could also remain in their ecological environment and avoid long distance traveling.

Studies III and IV. The 22 children from study II who participated in the GDT group were included. The children were 1-6 years of age (mean 3 years and 10 months, SD 1year 4 months), eleven girls, classified in GMFCS levels I-IV and MACS levels I-IV. A unilateral CP was present in eight children (Left: 3, Right: 5) and a bilateral CP, in 14 children (Table III). The inclusion and exclusion criteria were identical to those in study II.

Table III. The thesis covers four studies including children with cerebral palsy. Gender is presented as number of girls and boys. Age is presented as years. Gross Motor Classification System (GMFCS) and Manual Ability Classification System (MACS) are presented as number of children in each level. Distribution is presented as number of children with: Bilateral CP (B), Left unilateral CP (LU), Right unilateral CP (RU).

STUDY	n	GIRLS/ BOYS	AGE	GMFCS					MACS				B	LU	RU
				I	II	III	IV	V	I	II	III	IV			
I	40	18/22	3-16	13	12	10	3	2					30	6	4
II	44	19/25	1-6	19	10	8	7		16	15	10	3	27	8	9
III	22	11/11	1-6	10	5	3	4		7	9	5	1	14	3	5
IV	22	11/11	1-6	10	5	3	4		7	9	5	1	14	3	5

Overlapping: Three children from Study I were included in Studies II-IV. Twenty-two children from Study II were included in Studies III and IV.

CLASSIFICATIONS

The definition and classification of CP from 2006 (Rosenbaum et al., 2007) were applied, in which the classification system described in the Reference and Training Manual of the Surveillance of Cerebral Palsy in Europe (SCPE) was adopted. The predominant neuromotor abnormality is described as spastic, dyskinetic or ataxic, and spastic CP is further classified as Bilateral (B) when both sides are involved and as Unilateral Left (LU) when left side of the body is involved and as Unilateral Right (RU) when right side of the body is involved (Ashwal et al., 2004)(Studies I-IV).

The GMFCS was used to classify children's gross motor performance (Palisano et al., 1997) (Fig 2)(Study I-IV).

The MACS was used to classify children's manual ability in everyday situations (Eliasson et al., 2006) (Table I)(Study II-IV).

The ICF-CYwas used to link the meaningful concept of each goal to the most specific code available in Part one of ICF-CY (Fig 1) (Word Health Organization (WHO) and WHO Workgroup for Development of ICF for Children and Youth, 2007). In the ICF-CY coding system the letters refer to the component of the classification followed by a numeric code, starting with the chapter number (one digit) followed by the second level (two digits), the third level (one digit) and fourth level (one digit) (Study IV).

MEASURES AND DATA COLLECTION

BODY FUNCTIONS

In Studies I and III selective motor control of the dorsiflexors of the ankle was measured by the use of the five graded scale by Boyd and Graham (SMC) (Boyd and Graham, 1999). The scale is a criterion-referenced, observational measure used to estimate which muscles the child primarily activates. The scale ranges from 0, which indicates no movement, to 4, which indicates isolated selective motor control through available range of motion and with balanced activity in mm. tibialis anterior (Table IV). The child sits in a comfortable position with the legs in front. To our knowledge, no reports of the reliability and validity of the scale existed at the time of the study. In Study I three PTs simultaneously and independently graded each child's SMC, and PT 3 repeated the assessment at another occasion in 29 of the children (1-60 days before or after). In Study III the SMC was assessed collaboratively by the same PTs (KL, AB) before and after GDT in 22 children with CP (Figure 4).

Table IV. Selective Motor Control Scale of the dorsiflexion of the ankle (SMC) by Boyd and Graham. The scale is five graded ranging from 0 to 4. Muscle activity was evaluated in m. extensor hallucis longus (EHL), mm extensor digitorum longus (EDL) and m tibialis anterior (TA).

SCORE	DEFINITION
0	No movement
1	Limited dorsiflexion using EHL/EDL
2	Dorsiflexion using EHL, EDL & some TA activity
3	Dorsiflexion achieved mainly by TA + by hip & knee flexion
4	Isolated dorsiflexion through available range, balance of TA (without hip and knee flexion)

Passive range of motion (ROM) was assessed with goniometry in standardized positions. The intra-rater reliability has been reported to be high (Mutlu et al., 2007;Glanzman et al., 2008). The measurement error in children with CP was estimated to be 5° - 10°. In Study III PROM was assessed in the hip, knee and ankle in standardized positions, before and after GDT (Figure 4), by the same PTs (KL, AB) in collaboration.

Spasticity was estimated by the use of a modified Ashworth scale (MAS) (Peacock and Staudt, 1990). Several modifications of the original Ashworth scale exist. In the modification by Peacock & Staudt, the scale is six-graded, ranging from 0 (hypotonus) to 5 (extreme spasticity) (Table V). The reliability of this specific modification is not described, but moderate reliability has been reported for the Ashworth scale modified by

Bohannon and Smith (Clopton et al., 2005;Mutlu et al., 2008). The reliability varied in relation to the muscle tested and to the number of assessors. The highest reliability was demonstrated when the same assessor repeatedly tested the child (Fosang et al., 2003;Clopton et al., 2005;Platz et al., 2005). In Study III assessments were accomplished with MAS in the hip, knee and ankle muscles in standardized positions, before and after GDT(Figure 4), by the same PTs (KL, AB) in collaboration.

Table V. Modified Ashworth scale (MAS) by Peacock and Staudt.

SCORE	DEFINITION
0	Muscle tone is less than normal
1	No increase in muscle tone
2	Slight increase in tone; “catch” or minimal resistance to movement is felt during passive movement throughout less than half of the range of movement
3	Marked increase of muscle tone; resistance to movement is felt during passive movement through more than half of the range of movement. However, passive movement is easily performed
4	Considerable increase in muscle tone, passive movement is difficult to perform
5	Affected part is rigid in flexion or extension

ACTIVITY AND PARTICIPATION

Gross Motor Function Measure -66 (GMFM-66) was used to evaluate change in gross motor capacity (Russell et al., 2002). GMFM is the most frequently used assessment for evaluating gross motor capacity in children with CP (Harvey et al., 2008). GMFM is an observational, standardized and criterion-referenced measure, developed to evaluate change (Russell et al., 2002). The items cover gross motor capacity from lying and rolling, to walking, running and jumping. Two versions of the measure exist, GMFM-88 (Russell et al., 1989) and GMFM-66 (Russell et al., 2002). In GMFM-66 the 66 items are organized in increasing difficulty from 0 (low capacity) to 100 (high capacity) along an interval scale (Rasch analysis). Each item is scored on a four-point Likert scale (0-3), in which a score of 0 means that the child does not initiate the item, and a score of 3 means that the child completes the item in accordance to the GMFM-66 manual (Russell et al., 2002). A five year old typically-developing child is expected to achieve a score of 100. The GMFM-66 is reported to be valid, reliable and responsive to changes in gross motor capacity in children with CP (Vos-Vromans et al., 2005;Wang and Yang, 2006). GMFM-66 was administered by the same two PTs in collaboration (KL, AB), in a total number of 198 assessments. The Gross Motor Ability Estimator (GMAE) software was used to calculate a total score (1-100) for each assessment (Russell et al., 2002). In Study II the GMFM-66 assessments performed before and after 12 weeks of therapy in 44 children with CP were used (occasions 3 and 5, Figure 4), whereas the assessments performed at all seven occasions (Figure 4) were used in Study III. In Study IV the assessments of GMFM-66 before GDT (occasion 3) and the change scores after GDT (occasions 3-5) in the 22 children who received GDT were used to explore relationships between individual GAS-goals and standardized measures (Figure 4).

The PEDI (Haley et al., 1992) was used to register and evaluate children's everyday performance. PEDI evaluates the capability and performance in everyday activities in the domains of self-care, mobility and social function within two dimensions; functional skills and caregiver assistance. PEDI is a standardized interview with the parents containing 197 items reflecting the child's capability in his or her everyday environment and 20 items concerning the child's need for caregiver assistance (performance of everyday activities). A raw score is calculated which can be transformed either into a normative score which allows the possibility to compare the child with age-matched children with typical development, or into a scaled score (0-100) which provides an opportunity to compare with the child's own performance over time (Haley et al., 1992). In a recent systematic review, PEDI was found to display high internal consistency, high inter-rater and test-retest reliability, concurrent validity, discriminative validity and responsiveness to change (Harvey et al., 2008). In Studies II and IV the three functional skill scales (FSS) and the three caregiver assistance scales (CAS) were administered through interviews with the parents by the same PTs (KL, AB) in collaboration. In Study II the PEDI was performed before and after a twelve week intervention period in 44 children with CP (occasions 3 and 5, Figure 4). In Study IV the assessments before and the change scores were used from the 22 children who received GDT. Raw scores were transformed and presented as scaled scores. A Swedish version was used (Nordmark et al., 1999).

Goal Attainment Scaling (GAS) was used to define and grade the goals of therapy and to evaluate goal attainment (Kiresuk and Sherman, 1968;Kiresuk et al., 1994;King et al., 1999). GAS is a five graded scale and an individualized, criterion-referenced measure of change. The GAS scale ranges from -2, indicating what the child has performed at the time of goal-setting (baseline) through 0, indicating the expected performance, to +2, signifying a much better performance than expected (Table VI). When multiple goals are evaluated, the score can be converted into a summary score (T-score) (Kiresuk and Sherman, 1968). A T-score of 50 indicates all goals were attained to the expected level, and a score above 50 indicates attainment above the expected. GAS has high content validity (Palisano, 1993), but low concurrent validity (Palisano, 1993;Cusick et al., 2006). The inter-rater reliability was found to be high (Palisano, 1993;Steenbeek et al., 2010). The responsiveness to change was observed to be high (Ahl et al., 2005;Lowe et al., 2006;Steenbeek et al., 2007).

TableVI. Goal Attainment Scaling (GAS).

GRADE	**DEFINITION**
-2	Baseline
-1	Less than expected outcome
0	Expected outcome
+1	Greater than expected outcome
+2	Much greater than expected outcome

In Studies II, III and IV the GAS was used to define and grade the goals of therapy and to evaluate goal attainment in children receiving GDT (n=22). The parents of each child and sometimes the child chose five goals. The goals were graded according to GAS in collaboration with the same PTs (KL, AB). The evaluation of goal attainment was performed from the parent's perspective of the child's performance in everyday situations, in collaboration with the PTs. In Study II goal attainment after 12 weeks of GDT was evaluated. In Study III goal attainment was evaluated once during the GDT and at three occasions after the GDT (every sixth week, Figure 4). In Study IV the meaningful concept of the description of the expected level in each goal was linked to the most suitable code in ICF-CY. Goal attainment after 12 weeks of GDT was correlated to change scores in standardized measures.

Table VII. An overview of assessments used in the four studies. The outcome measures are classified within the ICF-CY.

MEASURE	STUDY	OBJECTIVE	BODY FUNCTIONS	ACTIVITY & PARTICIPATION	CAPACITY	PERFORMANCE
SMC	I,III	Selective motor control	x			
PROM	III	Passive range of motion	x			
MAS	III	Spasticity	x			
GMFM-66	II,III,IV	Gross motor activity		x	x	
PEDI	II,IV	Everyday activity		x		x
GAS	II,III,IV	Goal attainment		x		x
GMFCS	I,II,III,IV	Classification		x		x
MACS	II,III,IV	Classification		x		x

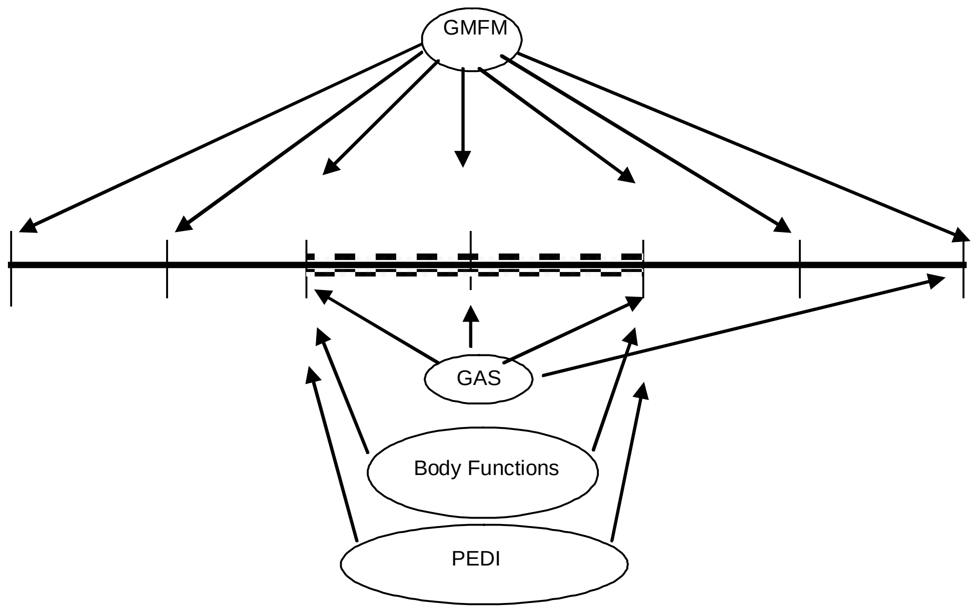

Figure 4. The figure illustrates the time line of the total data collection in Studies II-IV. The occasions when assessments were performed are indicated as "occasions 1 to 7". In Study II the assessments of the GMFM-66 and PEDI at occasions 3 and 5 were used for the AT group and the GDT group. The GAS was graded and evaluated at occasions 3 and 5 in the GDT group. In Study III the GMFM-66 were performed at seven occasions (occasions 1-7), GAS was evaluated at five occasions (occasions 3-7) and the assessments of body functions were performed at two occasions (occasions 3 and 5) in the GDT group. In Study IV the assessments of GMFM-66, PEDI and GAS at two occasions were used (occasions 3 and 5) in the GDT group.

INTERVENTIONS

HABILITATION CENTRES

At the time of the study (2004 – 2006), there were 13 habilitation centres in Stockholm County. The centres were situated in a broad geographic area around Stockholm County from Norrtälje in the north to Södertälje in the south, over a total distance of 112 kilometres. Children were referred to the local Habilitation centre according to where the family lives. The Habilitation centres coordinate their practise into multi-professional teams, in which combined interventions from medical, pedagogical, psychological and social perspectives are applied with respect to the needs of the child and the family (Handikapp och Habilitering, 2010).

ACTIVITY FOCUSED THERAPY

In activity focused therapy the child receives treatment according to an individually written habilitation plan. The plan is written after a joint meeting with the family/child and the multi-professional team during which general aims for the child's therapy are formulated. The child and family can be assisted by a multi-professional team consisting of a special needs teacher, a speech and language therapist, an occupational therapist, a social worker, a psychologist and a physical therapist The general aims and the child's/family's strengths and needs provide the basis for the interventions from the diverse professionals, often using an activity focused approach. An activity focused approach has a family-centred attitude, wherein the needs from both the child and the family are incorporated. Emphasis is on ample and variable opportunities for playful practise in ecological settings as the child is actively involved in learning everyday activities. The treatment frequencies occurred in agreement to the plan (1-2 times a week) and took place at the rehabilitation centre, at the preschool and at the child's home. The child received individually-tailored treatment sessions, the environment was modified with respect to the child's strengths and needs, and the parents and preschool teachers received support and instructions to facilitate the child's learning.

GOAL DIRECTED THERAPY

The definition of GDT was: A therapy that emphasizes the learning of meaningful activities (expressed as goals) in the child's environment, wherein the activities are regarded as important by the child, the parents and others in the child's environment. The goals are established based on the parents' and children's priorities. Learning takes place in individually-tailored interventions in the child´s natural environment by repetitive practice of the everyday goal activities, in a motivated, challenging and playful way, and in combination with impairment-focused interventions. The overall aim of the therapy is to improve everyday performance in activities and participation.

In GDT, emphasis is directed towards the child and family in the goal-setting process with the aim to select goals that are meaningful in the lives of the child and family. The selected goals are thoroughly analyzed with respect to the actual performance considering facilitating and hindering factors. Furthermore attention is on involving all people engaged in the child's everyday environment, to achieve a joint plan for therapy. The intervention is directed towards 1) the child by supporting motor learning, towards 2) the everyday environment by facilitating activity and participation and towards 3) the task by adjustments in relation to the child's strength and difficulties (Fitts and Posner, 1967;Newell, 1991;Vereijken et al., 1992;Gentile, 1998;Biggs, 1999;Campos et al., 2000;Valvano, 2004;Shepherd and Carr, 2006;Shumway-Cook and Wollacott, 2007). In the GDT the families and people in the children's environment initially participated in a one-day education concerning the diagnosis CP, the principles of motor learning and the importance of motivation and play as a foundation in therapy. Subsequently the 12 week therapy started, and was integrated into the child's everyday environment (i.e. preschool, home and surroundings) and once a week at the habilitation centre together with 7-8 other children who participated in the GDT. The parents, the preschool teachers and the multi-professional teams at the corresponding habilitation center delivered the therapy collaboratively.

The multi-professional team consisted of speech and language therapists, occupational therapists, special needs teachers and physical therapists. The therapy was multifaceted as focus was the children's individual goals. At the group meetings the children participated in everyday activities pertaining to eating, dressing, playing, communication and mobility.

STATISTICAL ANALYSES

A variety of statistical methods were used in this thesis (Table VIII). The reliability analysis of the SMC scale in Study I was performed by weighted Kappa statistics and percentages agreements, as the SMC scale represents ordinal data (Lantz, 1997). The Kappa coefficient provides a measure of agreement that takes into account the agreement expected purely by chance (Cohen J, 1960;Cohen, 1968;Sim and Wright, 2005). Weighted Kappa has been recommended to use for ordinal scales, as it offers the possibility to reflect the degree of disagreement, emphasizing the size of differences between raters (Cohen, 1968). As the Kappa coefficient in itself does not indicate whether the disagreement is related to random or systematic differences, additional analyses were performed. These calculations were: the Relative rank Variance (RV), which empirically measures the random part of disagreement ($0 \leq RV \leq 1$), the Relative Position (RP), which empirically measures the systematic shift in position between two raters ($-1 \leq RP \leq 1$), and the Relative Concentration (RC), which empirically measures the systematic shift in concentration ($-1 \leq RC \leq 1$)(Svensson, 1998). A value of RP value close to zero indicates no systematic change in position between the two raters. An RC value close to zero indicates no systematic change in concentration between the two raters. $RC > 0$ indicates that the classifications of rater 2 are more concentrated in the central categories than the classifications of rater 1. $RC < 0$ indicates that the classifications of rater 1 are more concentrated in the central categories than the classifications of rater 2 (Svensson, 1998).

In Study IV unweighted Kappa statistic was used to calculate agreement between the coding of GAS goals in the ICF-CY linking procedure, as the chapters represents nominal data. In addition percentage agreement was calculated according to all digits in the codes. Kappa values were defined according to Fleiss' definitions - values less than 0.40 imply poor agreement, values between 0.40 and 0.75 indicate fair to good agreement, and values above 0.75 indicate strong agreement (Fleiss J and Cohen J, 1973).

In Study III Wilcoxon´s test was used for within-group comparison of the MAS scores and the SMC scale (ordinal data). The most affected leg was analysed for each child, or in case of symmetry, the right leg was used (Sutherland et al., 1999). GMFM-66 scores for assessment at the occasions 3 and 5 were converted to percentiles, which were further analysed (Russell et al., 2002;Hanna et al., 2008). Parametric statistics for within-group comparisons (t-test) were used for analysis of GMFM-66 percentiles and PROM.

In Study II the changes in interval data (PEDI, GMFM-66) were analysed using paired students t-test for within-group comparison and the independent student t-test was used for comparisons between the two therapy groups.

The Kruskal Wallis test was used for group comparisons within the GMFCS, MACS and age groups in Study III.

Multiple linear regressions using a model consisting of age, sex, distribution of CP, level of GMFCS and MACS were utilized to explain the improvement in PEDI outcome in the GDT group in Study II.

A Mixed Linear Model with unstructured means of GMFM-66 and with time, GMFCS and age-group used as factors was calculated to follow gross motor development during the seven assessments in Study III.

Differences were considered significant with a p-value < 0.01.

The number of goals within the chapters in ICF-CY was calculated in Study IV. The number of goals achieved at various levels in GAS in each GMFCS level was calculated in Study III. The sum of all 110 goals was converted to a T-score by the use of tables for summary T-score (Kiresuk et al., 1994) (Study III, IV). Summation of all goals belonging to the ICF-CY chapters in Mobility and in Self-Care respectively was converted to T-scores in Study IV.

Non-parametric correlations (Spearman) were calculated between SMC, PROM and MAS scores in Study III. Non-parametric correlations were also calculated between improvements in GMFM-66 (occasions 3 and 5) and the final GAS T-score in Study III. Furthermore, Spearman's correlation coefficients were calculated between baseline scores (occasion 3) of GMFM-66 and PEDI and number of GAS-goals in the Mobility and Self-care chapters in ICF-CY, between GMFCS, MACS and number of GAS-goals in the Mobility and Self-care chapters in ICF-CY respectively, and between goal attainment (T-score) in the Mobility chapter and in the Self-Care chapter and change scores in GMFM-66 and PEDI. Parametric correlations (Pearson) were performed between change scores in PEDI (FSS and CAS) mobility and GMFM-66 in the GDT group and the AT group in Study II. The interpretation according to Cohen was used (Cohen J, 1988). Correlations were considered significant if they reached both a correlation coefficient > 0.44 and a p-value < 0.05.

Effect sizes (ES) were calculated in two ways. In the study comparing two treatment approaches (GDT and AT) the ES was calculated by dividing the difference between the two groups' mean change score (the mean value from the GDT minus the mean value from the AT), with the SD from the baseline assessment in the AT group (Cohen J, 1988). In the longitudinal study ES was calculated by dividing the mean change score by the SD of the change score, which is often referred to as Standardized Response Mean (SRM) (Wright and Young, 1997).

The interpretation of Cohen suggests that a small effect represents a value around 0.20 and is not directly identified, a medium effect has a value around 0.50 and is large enough to be identified, and a large effect has a value of 0.80 or above and is very obvious to identify (Cohen J, 1988).

Table VIII. An overview of statistical methods used in the four studies in the thesis. Student's t-test for within- and between-group comparison, Wilcoxon signed rank test for within-group test, Effect Size (ES), Standardized Response Mean (SRM).

STATISTICAL METHODS	**STUDY I**	**STUDY II**	**STUDY III**	**STUDY IV**
Descriptive statistics	X	X	X	X
Students t-test, independent		X		
Students t-test, paired		X	X	
Mixed Linear Model			X	
Linear Regression		X		
Pearson correlation		X		
ES		X		
SRM			X	
Wilcoxon signed rank test			X	
Kruskal-Wallis			X	
Spearman correlation			X	X
Unweighted Kappa				X
Weighted Kappa	X			
Relative Variance	X			
Relative Position	X			
Relative Concentration	X			

RESULTS

BODY FUNCTIONS

All 40 children accomplished the inter-rater reliability examination in Study I. The inter-rater reliability revealed fair/good to strong agreement between pairs of assessors (Kw =0.58-0.77). The percentage agreements (PA) varied between 55-72.5%. The RV (0.000 to 0.030), RP (-0.09 to 0.11) and RC (-0.26 to 0.16) were all low (Table IX).

Table IX. Inter-rater reliability of the SMC scale assessed in Right and Left foot in 40 children with CP. Simultaneous assessments were performed by three assessors (PT1, PT2 and PT3) and the results were analysed between pairs of assessors. The analyses included weighted Kappa (Kw), percentage agreement (PA), Relative Variance (RV), Relative Position (RP) and Relative Concentration (RC).

TEST	RIGHT PT1 - PT2	RIGHT PT1 - PT3	RIGHT PT2 - PT3	LEFT PT1 - PT2	LEFT PT1 - PT3	LEFT PT2 - PT3
Kw	0.58	0.69	0.73	0.64	0.75	0.77
PA %	55	70	72.5	62.5	67	67.5
RV	0.005	0.003	0.001	0.030	0.000	0.014
RP	0.11	0.03	-0.09	-0.08	0.07	0.03
RC	-0.26	-0.08	0.16	-0.17	-0.10	0.10

In total the three PTs made 240 assessments, and out of them, 159 showed identical values, 76 assessments differed one level and 5 assessments differed two levels. Most scores were assigned to level three and four (Table X).

Table X. A total of 240 assessments/scorings were performed (40 children, three PTs, left and right foot). The table shows the number of times the scores were given in the various levels of the SMC scale.

LEVEL	NUMBER OF TIMES
0	12
1	26
2	36
3	89
4	77

The test-retest reliability was performed at two separate occasions in 29 children (those who had a regularly-planned visit at the hospital during the time period of the study), by PT3. The results of the test-retest reliability showed strong agreement between assessment instances (Kw = 0.88 – 1). The PA was 90-100 %. The RV (0 - 0.005), the RP (0 - -0.11) and RC (0 – 0.05) were all low (Table XI).

Table XI. Test-retest reliability was evaluated using repeated tests made by PT3. The first examination (PT3) and the second examination (PT3X) of Right and Left foot in 29 children with CP were used for analyses. The analyses included weighted Kappa (Kw), percentage agreement (PA), Relative Variance (RV), Relative Position (RP) and Relative Concentration (RC).

TEST	RIGHT PT3 - PT3X	LEFT PT3 - PT3X
Kw	1	0.88
PA %	100	90
RV	0	0.005
RP	0	-0.11
RC	0	0.05

Body functions measures were evaluated before and after GDT in 22 children with CP (Study III). The evaluations of SMC in ankle dorsiflexors revealed no significant changes 12 weeks after therapy. The children's PROM was evaluated by goniometry in the hip, knee and ankle. Improvement was detected in ankle dorsiflexion, with a mean improvement of 9° (95% CI: 5°-13°, p<0.001). No other significant changes were detected in PROM. MAS scores were evaluated in muscles around the hip, knee and ankle. There were no significant changes observed 12 weeks after therapy.

ACTIVITY AND PARTICIPATION

Everyday activities (capability and performance) were evaluated by PEDI before and after a period of 12 weeks therapy in 44 children with CP (Study II). Half of the group (n=22) received AT and the other half (n=22) received GDT (Results also used in Study IV). Comparison of the two groups with respect to age and to levels in GMFCS and MACS did not reveal any differences. The baseline assessments in PEDI caregiver assistance scale (CAS) in self care was higher in the AT group at the start (p<0.01). No other significant differences between the two groups could be observed (Table XII).

Table XII. Comparisons of baseline assessments, presented as mean and standard deviation (SD), in PEDI Functional Skills Scales (FSS) and in Caregiver Assistance Scales (CAS) in the Activity Focused Therapy (AT) group (n=22) and in the Goal Directed Therapy (GDT) group (n=22).

PEDI	AT GROUP	GDT GROUP	p
FSS SELF CARE	58.6(10.6)	52.5 (9.4)	0.049
FSS MOBILITY	62.2 (19.4)	55.6 (15.5)	0.223
FSS SOCIAL FUNCTION	62.0 (8.0)	56.0 (11.0)	0.043
CAS SELF CARE	54.9 (14.4)	43.2 (13.3)	0.007
CAS MOBILITY	63.6 (17.6)	54.0 (17.4)	0.076
CAS SOCIAL FUNCTION	60.3 (16.0)	50.7 (19.8)	0.684

Evaluations 12 weeks after the therapy demonstrated significant improvements in the GDT group in comparison to the AT group with respect to all three Caregiver Assistance Scales (CAS) and to the Functional Skills Scales (FSS) in Self Care and Mobility. No significant differences were observed in the FSS in Social Function between the two groups (Table XIII). The effect sizes (ES) in the GDT group were calculated in the FSS/CAS: in Self Care 1.7/1.9, Mobility 1.1/1.4 and Social Function 0.7/1.5.

Table XIII . Comparisons of the improvements after 12 weeks of therapy (Independent t-test) between the Goal Directed Therapy (GDT) group and the Activity Focused Therapy (AT) group. Assessments included six scales in the Pediatric Evaluation of Disability Inventory (PEDI); the Functional Skills Scales (FSS) and the Caregiver Assistance Scales (CAS) in Self-Care, Mobility and Social Function. Data are presented as mean of the differences between the two groups and 95% confidence interval (95% CI). The number of participants in each group was 22.

PEDI	MEAN DIFFERENCE	95 % CI	p
FSS SELF-CARE	4.8	3.1 - 6.6	0.001
FSS MOBILITY	5.5	2.6 - 8.4	0.001
FSS SOCIAL FUNCTION	3.6	0.5 - 6.8	0.026
CAS SELF-CARE	10.4	7.1 - 13.8	0.001
CAS MOBILITY	8.9	4.9 - 12.9	0.001
CAS SOCIAL FUNCTION	10.5	6.1 - 14.8	0.001

Within-group comparisons revealed significant improvements in the GDT group in all measured aspects of PEDI (Table XIV) in contrast to the AT group, in which no significant improvements were found 12 weeks after therapy (Table XV).

Table XIV. Baseline assessments and evaluation after 12 weeks in the GDT group, presented as mean and standard deviation (SD). The change scores are presented as mean and 95% confidence interval (95% CI) in the GDT group (n=22).

GDT GROUP PEDI	BASELINE MEAN (SD)	AFTER MEAN (SD)	P	MEAN CHANGE	95%CI
FSS SELF CAR	52.5 (9.4)	57.35 (9.4)	<0.001	4.9	3.3 – 6.4
FSS MOBILITY	55.6 (15.5)	61.44 (13.9)	<0.001	5.8	3.5 – 8.2
FSS SOCIAL FUNCTION	56.0 (10.9)	61.22 (8.9)	<0.001	5.3	3.3 – 7.2
CAS SELF CARE	43.2 (13.3)	54.14 (13.2)	<0.001	11.0	9.0 – 13.0
CAS MOBILITY	54.0 (17.4)	63.41 (14.6)	<0.001	9.4	5.8 – 13.0
CAS SOCIAL FUNCTION	50.7 (19.8)	61.26 (17.3)	<0.001	10.6	6.8 – 14.4

Table XV. Baseline assessments and evaluation after 12 weeks in the AT group, presented as mean and standard deviation (SD).The change scores are presented as mean and 95% confidence interval (95% CI) in the AT group (n=22).

AT GROUP **PEDI**	BASELINE MEAN (SD)	AFTER MEAN (SD)	P	CHANGE MEAN	95%CI
FSS SELF CAR	58.6 (10.6)	58.7 (11.6)	0.946	0.0	-0.9 – 1.0
FSS MOBILITY	62.2 (19.4)	62.5 (17.8)	0.718	0.3	-1.5 – 2.2
FSS SOCIAL FUNCTION	62.0 (8.0)	63.6 (10.3)	0.209	1.6	-1.0 – 4.3
CAS SELF CARE	54.9 (14.4)	55.9 (17.2)	0.688	0.6	-2.3 – 3.4
CAS MOBILITY	63.6 (17.6)	64.1 (16.5)	0.625	0.5	-1.6 – 2.5
CAS SOCIAL FUNCTION	60.3 (13.1)	60.8 (11.9)	0.904	0.1	-2.2 – 2.5

Gross motor capacity was evaluated by GMFM-66 before and after 12 weeks of therapy in the AT group and in the GDT group (Study II). Comparison at the baseline did not reveal any significant difference (AT group: 63.1 SD 16.0 and GDT group: 58.5 SD 13.6, p=0.318). Comparison between the two groups after 12 weeks of intervention revealed significant improvements in the GDT group with a mean difference of 4.0 points (95% CI: 2.2 – 5.8, p<0.001). The ES in the GDT group was 1.4.

In Study III the gross motor capacity was evaluated at seven occasions in the group receiving GDT. Three baseline assessments were performed, as well as one assessment during the therapy and three assessments of GMFM-66 after the end of therapy (six weeks between each occasion, Figure 4). The baseline assessments before therapy did not demonstrate any significant changes (occasions 1-2: 0.15, p=0.799, occasions 2-3: 1.13, p=0.054), nor did the assessments after the end of therapy (occasions 5-6: 0.73, p=0.225, occasions 6-7: -0.78, p=0.207). The assessments during the therapy revealed significant improvements (occasions 3-4: 2.92, p<0.001 and occasions 4-5: 2.19, p<0.001). The total improvement in GMFM-66 during the 12 week intervention was 5.07 (95% CI: 3.77-6.38, p<0.001) (Results also used in Study IV) (Table and Figure). No significant differences were observed in the extent of improvements with respect to the children's age, gender, level in GMFCS or MACS or distribution of CP. A mean value of the effect of therapy (occasions 3 –5) was calculated - the standardized response mean (SRM) - for each GMFCS level, which showed GMFCS I: 1.27, GMFCS II: 3.36, GMFCS III: 2.38, GMFCS IV: 2.02.

Table XVI. GMFM-66 performed (every sixth week) at seven occasions in the GDT group. GMFM-66 scores presented as mean values and 95% confidence interval (95% CI). Pairwise comparisons between the times GMFM-66 were performed, presented as mean change scores.

OCCASION	GMFM-66	95%CI	PAIRWISE COMPARISONS OCCASION	CHANGE SCORE	P
1	54.59	51.66 – 57.51			
2	54.74	51.81 – 57.66	1-2	0.15	=0.799
3	55.87	52.95 – 58.79	2-3	1.13	=0.054
4	58.79	55.86 – 61.71	3-4	2.92	<0.000
5	60.98	58.06 – 63.91	4-5	2.19	<0.000
6	61.71	58.78 – 64.64	5-6	0.73	=0.225
7	60.93	57.99 – 63.86	6-7	-0.78	=0.207

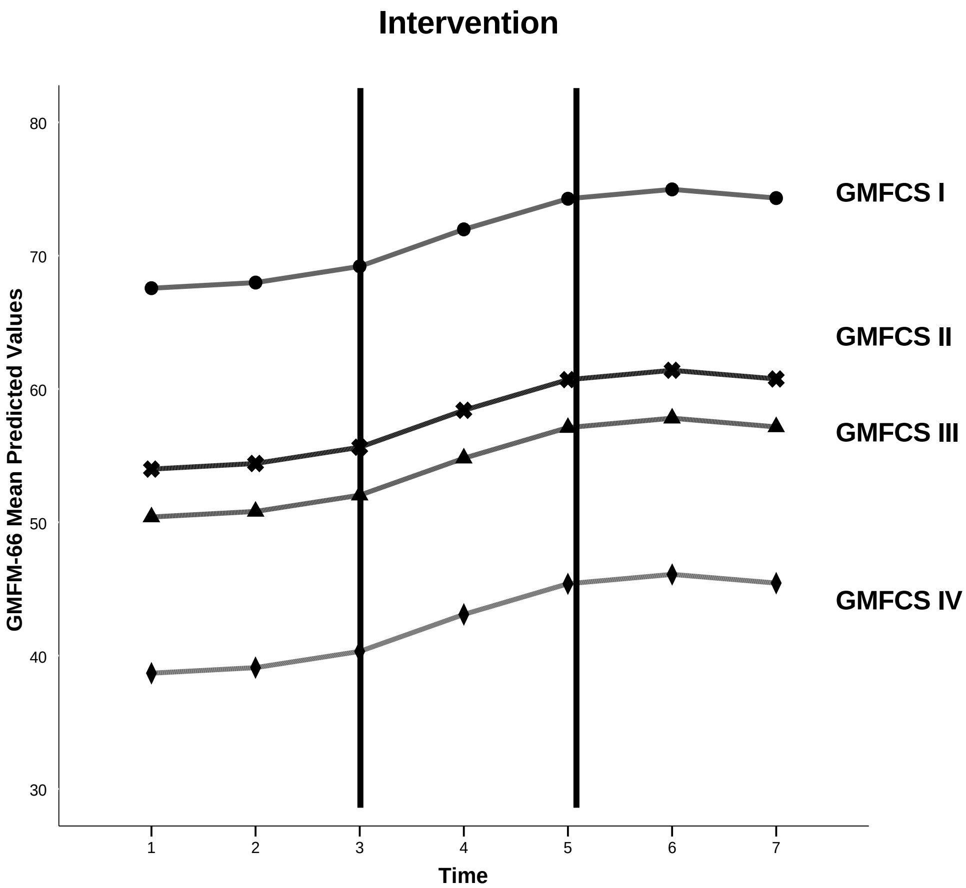

Figure 5. GMFM-66 scores (y-axis) at the seven occasions (x-axis) in the GDT group, in groups according to the four GMFCS levels. The intervention period was performed during occasion 3 and occasion 5 (solid vertical lines). Calculations were performed by Mixed Model linear using means of GMFM-66 and with GMFCS, time and age-group used as factors. Significant improvements were observed between occasions: 3-5 (therapy period: mean 5.07:p<0.001). No significant improvements were detected between the assessments that took place before therapy, nor between the ones that took place after therapy (n=22).

References percentiles (R.P.) of the GMFM-66 assessments before (occasion 3) and after (occasion 5) GDT and the change scores were presented for all 22 children in the GDT group (Study III). The mean change score in R.P was 18 (95% CI: 12 – 25, p<0.001). The median change score was 14 (and the 25th and 75th percentiles: 5 – 26, p<0.001). No significant differences were detected between children in the various levels in GMFCS (Figure 5). An increased percentile was observed in 19 out of 22 children, and the three children who did not change remained in the percentile observed at occasion 3. These three children were all in GMFCS level I and their ages were: 2 years 6 months, 3 years 2 months and 4 years respectively (Table XVII).

Table XVII. The GDT groups scores in GMFM-66 and the References percentiles (R.P) before and after therapy. The level in GMFCS, the age (months) of the child, the score in GMFM-66 and the R.P for the GMFM-66 assessments before (occasion 3) and after (occasion 5) GDT are presented. The R.P. change scores were calculated and presented. (n= 22 children).

GMFCS	AGE	GMFM OCCASION 3	R.P.	AGE	GMFM OCCASION 5	R.P.	CHANGE SCORE
I	30	65.33	65	33	66.69	65	0
I	32	58.09	30	35	69.22	75	45
I	32	68.86	80	35	73.63	90	10
I	37	64.27	45	40	66.33	50	5
I	38	78.28	95	41	79.99	95	0
I	42	70.39	60	45	73.1	65	5
I	48	71.22	50	51	81.93	85	35
I	48	68.86	35	51	69.63	35	0
I	62	71.69	25	65	79.11	50	25
I	71	79.11	45	74	85.23	70	25
II	16	35.26	3	19	42.85	25	22
II	28	48.09	50	31	54.15	75	25
II	40	64.98	95	43	69.22	97	2
II	55	60.92	75	58	65.33	85	10
II	66	64.98	75	69	68.86	85	10
III	44	47.68	45	47	50.09	60	15
III	56	56.62	85	59	62.39	97	12
III	69	51.09	35	72	55.39	65	30
IV	27	35.26	65	30	41.79	95	30
IV	51	35.67	30	54	45.56	80	50
IV	61	44.97	70	64	49.85	90	20
IV	68	45.91	75	71	48.73	85	10

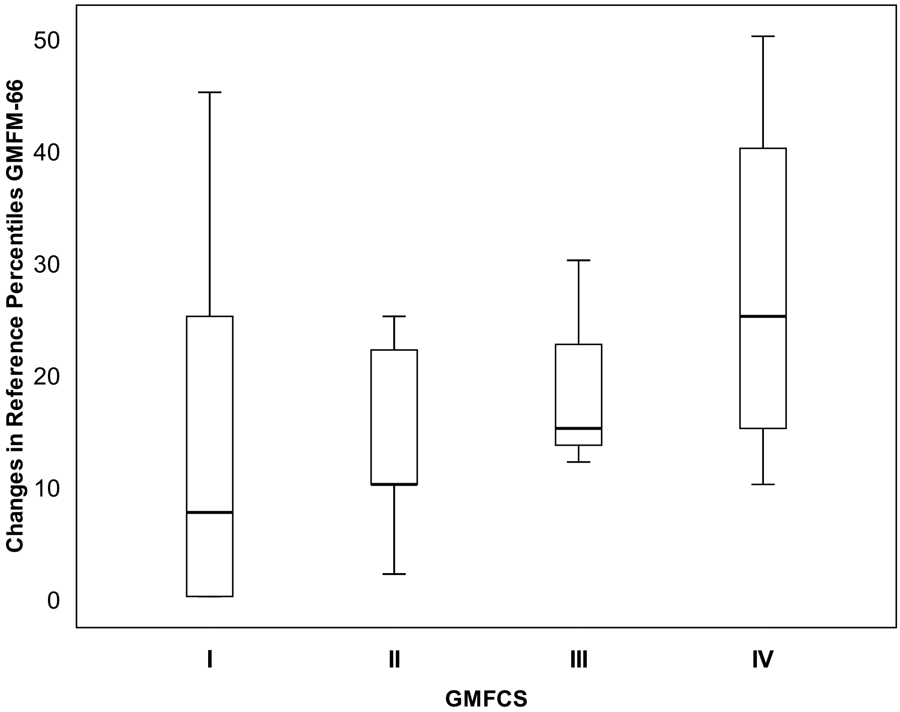

Figure 6. Box plots illustrating the median changes in GMFM-66 reference percentiles 12 weeks after GDT (y-axis) in the GMFCS levels I – IV respectively (x-axis). The boxes represent the median value and the 25th and 75th percentile (n=22).

Each family/child chose five goals (Studies II, III, IV), which were graded according to goal attainment scaling before GDT (occasion 3) and evaluated at four occasions every sixth week (occasions 4,5,6,7)(Study III). The evaluation in the middle of the GDT period (after six weeks, occasion 4) demonstrated that eight goals remained at -2, 39 goals had progressed to -1, 40 goals had reached the expected outcome, and 23 goals had attained a higher level than expected (Table XVIII). At the end of the intervention (after 12 weeks, occasion 5) none of the goals remained at -2, 17 of the goals had reached -1 and 93 of the goals had attained the expected level or higher (Table XVIII). Analysis of the extent of goal attainment at the end of GDT (occasion 5), with respect to gender, age, MACS and GMFCS levels and distribution of CP did not reveal any significant differences (Figure 7, Study III).

At the long-time follow-up six weeks after the intervention (occasion 6), none of the goals remained at -2, 12 goals were at -1 and 98 of the goals had attained the expected level or higher (Table XVIII). At the final long-time follow-up (occasion 7), 6 goals were still at -1 and 104 of the goals had attained the expected level or higher and of those goals there were 50 goals which had attained +2 (Table XVIII). No significant differences in goal attainments were observed with respect to the children's gender, age, level in GMFCS or MACS or distribution of CP.

Table XVIII. Goal Attainment Scaling (GAS) was used to grade the goals. Goals were set before the intervention (occasion 3) and evaluated during and after therapy (occasions 4,5,6,7). The number of GAS-goals attained at the various GAS scores from -2 to +2 are presented (n=110 goals, from 22 children).

OCCASION / GAS	3	4	5	6	7
-2	110	8	0	0	0
-1	0	39	17	12	6
0	0	40	27	23	16
+1	0	17	38	42	38
+2	0	6	28	33	50

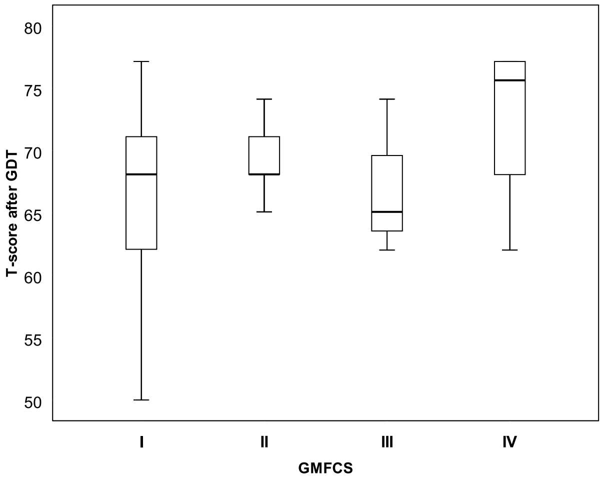

Figure 7. Box plots illustrating the median total t-score for all goals (n=110)12 weeks after GDT (y-axis) in the GMFCS levels I – IV respectively (x-axis). The boxes represent the median value and the 25th and 75th percentile.

After the end of the intervention study period, the meaningful concept of the expected levels of 110 GAS-goals was coded and linked to ICF-CY by two independent researchers (Study IV). Agreement of the coding was examined by calculation of unweighted Kappa, according to the letter and first digit, and revealed high agreement (K=0.94, 95% CI: 0.88-1.00). Furthermore percentage agreement was performed, according to all digits in the codes, and the result demonstrated 85% agreement. In those codes in which disagreement was present, consensus was reached by discussions with two other experts (AB, EBC). All goals were linked to the domain “Activity and Participation” in ICF-CY. The 110 GAS-goals were assigned codes which belonged to the following chapters: Chapter 1 “Learning and applying knowledge” (n=4), Chapter 2 “General tasks and demands”(n=1), Chapter 3 “Communication”(n=4), Chapter 4 “Mobility”(n=50), Chapter 5 “Self-Care”(n=50) and Chapter 9 “Community, social and civic life”(n=1) (Figure 8). The most frequently occurring codes belonged to the “Self-Care” and “Mobility” chapters, and were: **5501** Carrying out eating appropriately (n=14), **5400** Putting on clothes (n=12) and **4551** Moving around/ climbing (n=9).

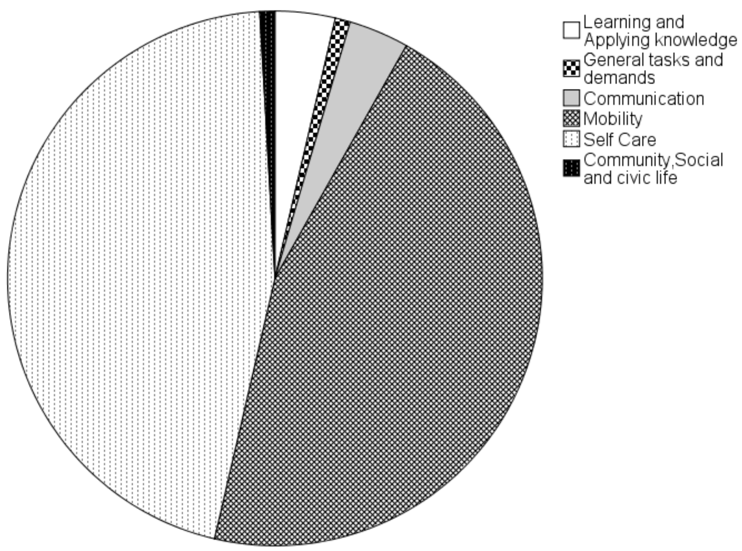

Figure 8. The 110 GAS-goals were coded and linked to the Activity and Participation chapters in ICF-CY. The following chapters were represented and the number of goals in each chapter is presented: ‘Learning and applying knowledge’ (n=4), ‘General tasks and demands’ (n=1), ‘Communication’ (n=4), ‘Mobility’ (n=50), ‘Self-care’ (n=50) and ‘Community, social and civic life’ (n=1).

RELATIONS BETWEEN BODY FUNCTIONS AND ACTIVITY AND PARTICIPATION

Analyses of relationships between and within various body function measurements and activity and participation measurements were performed in the group of 22 children receiving GDT (Study III). A relationship was demonstrated between the children’s ability to selectively dorsiflex the ankle (SMC scores) and the level in GMFCS (r_s = -0.59 $p<0.01$).The MAS scores in hip adductors and PROM in hip abduction displayed a strong correlation (r_s = -0.75, $p<0.001$). Furthermore a moderate correlation was observed between PROM in ankle dorsiflexion and MAS in plantarflexor (r_s = 0.51, $p<0.05$). At the end of the intervention, a strong correlation was observed between MAS in knee flexors and popliteal angle (r_s = 0.70, $p<0.001$).

In Study IV the analysis of the families'/children's choice of goals revealed relationships between the number of GAS-goals linked to the Mobility chapter in ICF-CY, and the baseline scores in both the PEDI functional skills scales (FSS) in mobility (r_s = -0.55, p=0.008) and the GMFM-66 scores at baseline (r_s = -0.57, p=0.006). A moderate relationship was also observed between the number of GAS-goals linked to codes in the Self-care chapter in ICF-CY and the baseline scores in PEDI caregiver assistance scale (CAS) in self-care (r_s = 0.44, p=0.041). The children's level in GMFCS correlated to the number of goals selected in the ICF-CY chapters of "Mobility" (r_s = 0.58, p=0.005) and the level in MACS correlated to the "Self-Care" chapter (r_s = -0.46, p=0.032).

The total goal attainment for all GAS-goals in the "Mobility" chapter correlated to the change scores in GMFM-66 (r_s = 0.48, p=0.026) and PEDI FSS in mobility (r_s = 0.58, p=0.006), while no significant correlation was observed to PEDI CAS in mobility (r_s = 0.37, p=0.10). The total goal attainment for all GAS-goals in the "Self-Care" chapter correlated to the change scores in PEDI CAS in self-care (r_s =0.57, p=0.007), while no significant correlation was observed to PEDI FSS in self-care (r_s = 0.39, p=0.08).

DISCUSSION

The overall aim of the thesis was to study the effects of goal directed therapy in children with cerebral palsy with respect to performance of everyday activities, gross motor capacity and body functions measures, i.e., SMC in the dorsiflexors in the ankle, passive range of motion and MAS scores. Thus the main findings concerned various effects and relationships between and within the ICF-CY domains "Body functions and structures" and "Activity and Participation" before, during and after goal directed therapy in children with CP.

GENERAL DISCUSSION OF THE RESULTS

The GDT was integrated in the children's everyday environment, and in addition the children participated in group sessions once a week at the habilitation centre in their neighbourhood. The focus in therapy was the individualized goals. The AT therapy was also integrated in the children's everyday environment. In addition the children received individualized therapy once or twice a week at the habilitation centre, pre-school or at home according to the habilitation plan. The focus in therapy was related to general aims. In both approaches the therapy was predominantly performed in the children's ecological environment. Members of the multi-professional team at the local habilitation centre, in collaboration with the family and people in the child's environment, accomplished therapy in both approaches. The dissimilarity of the approaches concerned principally the presence of individual goals and group therapy (7-8 children) in the GDT approach versus general aims and individual therapy in the AT approach. The intensity, frequency and the amount of time spent in the two therapy approaches can be a question of discussion, as the distinction between intensity of therapy and practice in the everyday environment must be carefully considered (Palisano and Murr, 2009).

Assessing children with CP requires a comprehensive range of tools. The selection of assessments should therefore be tailored to the children involved, the purpose of measuring and the setting (Harvey et al., 2008). Today there are several studies reporting evidence of activity focused and goal directed therapy with respect to assessments of activity and participation (Ketelaar et al., 2001;Ahl et al., 2005;Ostensjo et al., 2008;Sorsdahl et al., 2010). Simultaneously, a great deal of research concerns impairments such as lack of selective motor control of muscles, reduced PROM and presence of spasticity. Our aim was to investigate both components in the ICF-CY, i.e. Body functions/structures and Activity and participation. A discussion of results in the various domains will be followed by a methodological discussion.

BODY FUNCTIONS

During the last decade a growing realization of the importance of SMC has been demonstrated (Ostensjo et al., 2004;Vos-Vromans et al., 2005;Fazzi et al., 2005;Desloovere et al., 2006). A hypothesis in the planning of this research was that children's ability to selectively dorsiflex their ankle could be a predictor of gross motor capacity outcome after GDT. The SMC scale published by Boyd and Graham (Boyd and Graham, 1999) is used in clinical practice and was intended to be used to evaluate the intervention. Therefore, its reliability was examined. The results of the inter-rater reliability demonstrated fair/good to strong agreement between pairs of raters. This may seem unsatisfying, even though the scores rarely displayed a difference of more than one grade. One factor that perhaps to some extent can explain the lack of better reliability was that the study was completed in a clinical setting in conjunction with a regular visit to a PT; the description in the scale states that to achieve a grade of 4, one should observe: "Isolated dorsiflexion through available range of motion…", and only the examining PT knew the range of motion. In addition the descriptions of the various grades also caused some ambiguity. The inter-rater reliability was recently also reported by Smits and colleagues, and their results corroborated our findings (Smits et al., 2010).

The test-retest reliability was good, indicating that the scale was stable during the period tested. Recently a new assessment of selective motor control of muscles in the lower extremities has been presented, the selective control scale of the lower extremity (SCALE), demonstrating both strong reliability and validity (Fowler et al., 2009).

The SMC was assessed before and after GDT and no changes were observed. These results are not very surprising as the intervention was directed towards children's goals in everyday activities and not specifically directed towards improving ankle dorsiflexion. PROM was assessed in the hip, knee and ankle before and after GDT. The only change observed in PROM was an increase in ankle dorsiflexion, but the improvement was within the range of measurement error. Spasticity was assessed in muscles around the hip, knee and ankle, and no changes were detected after the therapy. Thus no deterioration of the measured body functions occurred although they were not specifically addressed during therapy.

Activity and Participation

The comparisons of everyday performance in the two groups at the start of the interventions revealed no differences with respect to age, levels in GMFCS and MACS. The only difference in baseline assessments was detected in performance of self-care (CAS), where the AT group was more independent at the start of therapy. Gross motor capacity, everyday capability and performance improved more in the GDT group in comparison to the AT group in all measured scales except in capability (FSS) in social function. Within the group of children receiving GDT, improvements were seen in all aspects of everyday activities measured by the six PEDI scales whereas no within group differences were detected after 12 weeks of AT therapy. GDT especially induced high improvements in the caregiver assistance scales, CAS. This finding contrasts to some extent the findings of Ahl and collaborators (Ahl et al., 2005). They evaluated a group of pre-school children before and after five months of functional, goal directed therapy, and found the largest improvements in the capability scales (FSS). The effects of functional therapy were also compared to neurodevelopmental treatment (NDT) in a group of children 2-7 years of age (Ketelaar et al., 2001). The children in the functional therapy group improved in both capability and performance (FSS and CAS). These results were corroborated by our findings. Recently Sørsdahl and co-workers presented the effect of goal-directed, activity-focused physiotherapy in a group of children 2-9 years of age (Sorsdahl et al., 2010). The therapy was accomplished in a group setting at the habilitation centre, with training five days a week during three weeks. Improved capability (FSS) in self-care and improved performance (CAS) in self-care and mobility was found. The discrepancy between various study outcomes can possibly be explained by several factors such as the age of the children, the GMFCS and MACS levels at the commencement of the intervention and baseline status with respect to ceiling effect for children with high performance initially (Ketelaar et al., 2001;Vos-Vromans et al., 2005). When interpreting results, it is vital to consider what constitutes a clinical meaningful change. Responsiveness and sensitivity are sometimes used interchangeably, but there is an important distinction. Responsiveness means that clinically important changes are possible to capture, whereas sensitivity implies the ability of a measure to identify changes regardless of whether the changes are meaningful or not (Liang, 2000;Iyer et al., 2003). PEDI has demonstrated responsiveness (Vos-Vromans et al., 2005). In our study the children's capability increased around 5 points in all scales and the performance increased almost 11 scores in all three scales. The meaningful change in PEDI has been identified by clinicians to be 11 scaled scores across all six scales (Iyer et al., 2003;Haley et al., 2010). However, a question to be raised is whether an investigation of the parent's opinion of a meaningful change had received similar results.

GROSS MOTOR CAPACITY

The evaluation of gross motor capacity was accomplished by GMFM-66, and comparison between the groups demonstrated beneficial effects in the GDT group. The improved gross motor capacity reached a level which could be considered a clinically meaningful change (5.07 scores). Wang and Yang described an improvement of 3.71 scores during a time period of 3.5 months as a "great improvement", and identified a clinically meaningful improvement to be 1.58 scores (Wang and Yang, 2006). Gross motor capacity was followed in the GDT group before, during and after the intervention. The baseline assessments were stable, whereas the assessment in the middle of the period of therapy and immediately after the end of therapy demonstrated improved capacity. During the long-term follow-up no further improvements occurred. No differences between GMFCS levels related to the extent of improvements during the intervention could be detected in this study. Comparisons to other studies using a goal-directed approach revealed similar results in gross motor capacity (Ketelaar et al., 2001;Ahl et al., 2005;Sorsdahl et al., 2010). To this date it is difficult to draw any clear conclusions about which elements of therapy that were more important than others for significantly increased gross motor capacity. The GMFM-66 scores were also presented in comparison to reference percentiles, which make it possible to compare a child's score with the usual outcome of other children in the child's age and GMFCS level (Hanna et al., 2008). In our study most children improved more than expected during a three month period, but the results require careful interpretation as the variability in change can be large among children with CP (Hanna et al., 2008).

INTENSITY

The impact of the intensity of therapy is an issue investigated by several authors. In these studies a NDT approach was mostly used (Tsorlakis et al., 2004;Christiansen and Lange, 2008;Ustad et al., 2009;Van den et al., 2010;Arpino et al., 2010). Some of them evaluated the therapy per se, while others aimed to compare intensive versus non-intensive NDT. A recent study examined five children, aged 6-12 months, who received blocks of four weeks of intensive therapy and eight weeks of 'physiotherapy as usual'. The intervention was NDT and evaluations were performed by GMFM-66 every forth week. The children's gross motor capacity improved, but no differences were observed between the intensive and 'therapy as usual' periods (Ustad et al., 2009). The result corroborates the findings by Christensen and Lange, who compared the effects of intensive therapy with pause periods versus continuous therapy in two groups. The children were 1-8 years of age, GMFCS I-V, and the therapy was NDT during 30 weeks. Both groups improved in gross motor capacity, as evaluated by GMFM-66, but no difference was observed between the two approaches (Christiansen and Lange, 2008). The mean change scores in the two groups were 3.3 and 4.6 respectively after 30 weeks, which can be compared to the change score in our study of 5.07 after 12 weeks. Similar results were observed by Bower and collaborators, who studied children 3-12 years, GMFCS III-V. They compared intensive versus routine amount of therapy during six months and no differences were detected between the approaches (Bower et al., 2001). In contrast to these results, Tsorlakis and colleagues reported significant differences between two groups of children, receiving either intensive or continuous NDT. The children were in ages of 3-14 years and in GMFCS I-III. After 16 weeks the GMFM-66 mean change scores in the groups were 1.18 and 2.36 respectively, and the mean change score between the groups was 1.18. The authors concluded that the effectiveness of NDT and the need for intensive treatment were supported (Tsorlakis et al., 2004). Efficacy of intensive versus non-intensive NDT therapy in randomized controlled studies was reviewed in a recent paper (Arpino et al., 2010). The conclusion from this meta-analysis was that intensive NDT may improve gross motor capacity, but the effect was considered modest. Only four studies were included due to the low number of available randomized controlled trials.

The question of frequency and intervals of intensive versus continuous treatment episodes remains a challenging area that warrants further exploration. The answers most likely vary as much as the children/families and the contexts do. Thus, a flexible approach, in which therapy can be adjusted to changing needs and contexts of the child and family during different stages in their lives, is most likely the answer (Palisano and Murr, 2009;Wiart et al., 2010). When therapy is flexibly integrated into the everyday context, it becomes more difficult to explicitly measure the intensity (Simeonsson et al., 1991;Ketelaar et al., 2001;Ahl et al., 2005;Law et al., 2007;Palisano and Murr, 2009).

GOALS

All goals were coded and linked to the ICF-CY domain Activity and Participation. A finding that possibly could be explained by the fact that the families/children were responsible for choosing the goals (McDougall and Wright, 2009). The linking procedure of all goals made by two independent researchers demonstrated high agreement, which corroborates other studies (Ogonowski et al., 2004;Kronk et al., 2005;Nijhuis et al., 2008). Most goals were linked to the Mobility chapter and the Self-Care chapter, while only a few goals were linked to the chapters of Communication, Learning and applying knowledge, General task and demands and the Community, social and civic life. The finding corroborates results from Ödman and collaborators, who investigated two intensive training programs in which family-selected goals were also used for evaluation. Most goals were coded and linked to the Mobility chapter but in contrast to our study, the second largest number of goals was linked to the Body Function component, while the third largest number of goals was observed in the Self-Care chapter (Odman and Oberg, 2006). The preponderance of goals in the Mobility and Self-Care chapters could probably to some extent be explained by the children's young age and thereby the parents focus of interest in these areas (McConachie et al., 2006).

The evaluation of the children's goal attainment displayed a gradually rising level of performance towards and above the expected level. The evaluation in the middle of the intervention period revealed that many goals remained below the expected level, emphasizing that the timeframe of 12 weeks was often adequate for the set goals. At the end of the intervention most goals were reached to the expected level or above. In the long-term follow-up the children continued to progress. No differences were observed in the level of goal attainment with respect to the children's age, GMFCS or MACS levels or distribution of CP. In comparison to recent research evaluating goal attainment, our group attained a rather high level of goal attainment (Ketelaar et al., 2001;Ahl et al., 2005;Odman and Oberg, 2006;Ostensjo et al., 2008;Sorsdahl et al., 2010). The explanations are probably multifaceted, and some reflections will be discussed. GAS was used to evaluate individual goal attainment, but the most important aspect was probably the creation of an individual plan for each child. The process included a thorough analysis of the child's present performance in various environments, followed by a collaborative discussion with people in the child's environment, and subsequently a mutual agreement was made (Simeonsson et al., 1991;McLaren and Rodger, 2003). The practice towards achieving the goals was integrated into the child's everyday environment, a motivating context in which the child was given support, but also was challenged (Fitts and Posner, 1967;Vereijken et al., 1992;Gentile, 1998;Biggs, 1999;Shepherd and Carr, 2006). The difficulty gradually increased according to the predefined levels in each goal scale, providing an individually-tailored way to reach the goal (Chaiklin, 2003;Locke and Latham, 2006). The goals were attained through an interaction of the child's own performance, the task and the environment (Shumway-Cook and Wollacott, 2007). In some goals the focus was to succeed with less support from people/equipment whereas other goals involved an increased quantity or speed. The fact that the family and sometimes the child were responsible for selecting the goals could also have had an impact on the compliance for practice and on the outcome. As the therapy was integrated into the child's everyday environment, the structure and practice became immediately integrated in the context in which the child needed to accomplish the activity. This fact could possibly explain the progress toward higher performance even after the end of the intervention.

Another aspect reported by Raina and collaborators (2005), which also could be considered in relation to family health, is the fact that children's behaviour is one of the most important predictors of caregiver's well-being (Raina et al., 2005). The GDT approach, which is built on structured practice in everyday situations, could thus influence the child's behaviours and parental stress by providing a predictable environment and clear frames (Bamm and Rosenbaum, 2008;Dunst and Trivette, 2009). However, in this research the experiences of the parents were not investigated.

When children are offered some kind of intervention it is important to consider the 'treatment dilemma' (Scrutton, 2004) and consider whether the proposal of a new intervention is in accordance with the interest of the child and the family (Gibson et al., 2009). A family-centred service approach, which is a part of GDT, emphasizes a collaborative, decision-making process between professionals and parents wherein the child's needs are in focus. It is thus easier to avoid limiting the child's freedom by proposing interventions that "cost" more then they "add" (Gibson et al., 2009).

POSSIBLE FACTORS CONTRIBUTING TO THE OVERALL POSITIVE RESULTS

Already in 1978 the positive results of a goal directed intervention was reported (Maloney et al., 1978) and today there is a growing body of evidence concerning the effectiveness of goal directed therapy (Simeonsson et al., 1991;Ketelaar et al., 2001;Ahl et al., 2005;Ostensjo et al., 2008;Sorsdahl et al., 2010). Some differences exist in the therapeutic setting, the length of the therapy, the presence of goals and the family involvement in the goal-setting and in the therapy. However the questions concerning what ingredients in the therapy that makes the effectivness are not fully answered yet. Proably several factors interact and some of them will be summarized. Meaningful goals, selected by the child/family increases the child's motivation and the family's motivation to support the child's learning (Majnemer et al., 2010). As the goals are practised in everyday life the occasions for learning are often many, ensuring a certain intensity which is important when learning new skills and furtermore the practice is implemented in the everyday environment both at home and at the pre-scool (Campos et al., 2000;Valvano, 2004;Ketelaar et al., 2008). The initial education of parents and pre-school staff is also one aspect which might contribute to good results. The education helps the parent understand the sensory-motor problems the child might face and thereby supports the parents as teachers of everyday activities. During regular pre-scool and home visits by members from the multi-professional team, the parents and pre-school staff learn that the child needs to be challanged to actively find efficient strategies and also that the child requires appropriate feed-back in this process (van Duijvenvoorde et al., 2008). The group sessions could also contribute to the child's learning as children often enjoy being together doing activities they take pleasure in and by inspiration of viewing peers practicing. Finally the multidisciplinary involvement in the planning of the intervention in collaboration with the family, could also contribute to the results (Nijhuis et al., 2007). Further studies will hopefully contribute to a better understanding of what are the most effective ingredients in goal-directed therapy.

RELATIONS BETWEEN BODY FUNCTIONS AND ACTIVITY AND PARTICIPATION

Within the domain of Body function in the ICF-CY, relationships were observed between relatively high MAS scores and limited PROM around the hip, knee and ankle. Similar associations have been detected earlier in cross-sectional research (Abel et al., 2003;Ostensjo et al., 2004;Wright et al., 2008). Wright and colleagues explored relationships in change scores after an intervention, and even though positive outcome in most measured aspects were detected, the associations in change scores after intervention were no more than fair (Wright et al., 2008). Relationships between domains, e.g. between MAS and gross motor capacity, have been investigated in a number of studies. Only small or moderate relationships have been observed (Abel et al., 2003;Ostensjo et al., 2004;Ross and Engsberg, 2007;Wright et al., 2008). These relationships, as well as the influence of spasticity on other outcomes, are still not fully understood and warrant further research (Gorter et al., 2009). In a recently published study by Bartlett and colleagues, the aim was to assess decline in gross motor capacity in children in GMFCS III-V. Explorations of relationships revealed that limitation in range of motion was an important factor related to a decrease in gross motor capacity over time (Bartlett et al., 2010). A relationship between different domains in ICF-CY, was observed in our study, between the children's SMC scores and level in GMFCS, which is in agreement with the recent findings by Fowler and collaborators, even though they used another scale, the SCALE (Fowler et al., 2009).

However, our hypothesis of the SMC as a possible predictor of gross motor improvement was not confirmed. In fact, no detectable factors were found that could explain improvements in everyday performance or in gross motor capacity. Evaluating a multifaceted treatment approach is challenging, but explaining all the underlying mechanisms calls for extensive future research (Damiano, 2006;Sussman, 2010;Rosenbaum, 2010). For formal program evaluation purposes, the use of both standardized and individual measures has been recommended (King et al., 1999). The exploration of relationships between standardized and individual measures used in our intervention study revealed findings which corroborate the recommendation of King and colleagues.

The families' choice of GAS-goals was related to baseline assessments in standardized measures, and in addition the goal attainment displayed interesting relationships towards standardized measures. Most goals were linked to the Mobility and Self-Care chapters within the ICF-CY and the explorations revealed a somewhat contrasting pattern. To thoroughly explain these relationships would require further research, but some hypotheses can be made. Mobility goals were selected when the child showed relatively extensive limitations in mobility. In contrast Self-Care goals were selected when the child displayed relatively high performance in self-care. The explanation for choosing Mobility goals could possibly be that self-care activities to some extent require gross motor activity, i.e., putting on a pair of trousers by oneself demands gross motor activity. When the family was aware of the child's limited gross motor activity, this was prioritized, as they possibly presumed Self-Care goals being too difficult and not realistic. In contrast children displaying high performance in self-care were assigned a relatively high number of goals in Self-Care, implying that the family may have prioritized Self-Care goals when they considered them realistic (Locke and Latham, 2002;Chaiklin, 2003). Another dissimilarity was detected between goals in the Mobility and Self-Care chapters. Goal attainment in Mobility goals was associated to improvements in gross motor capacity and capability (FSS) in mobility, whereas goal attainment in Self-Care goals was associated to improved performance (CAS) in self-care. As the goals were attained through an interaction of the child's own performance, the task and the environment (Shumway-Cook and Wollacott, 2007) one can assume that Mobility goals were to a higher extent achieved by the child who learned the activity, whereas the Self-Care goals were possibly more affected by the environment, i.e., caregivers who reduced their amount of help. The results confirm the use of both standardized and individual measures and emphasize the inclusion of families in goal-setting.

METHODOLOGICAL CONSIDERATIONS

The reliability study was intentionally performed in a clinical setting since the SMC scale is frequently used in clinical practice (van Genderen et al., 2010). The weighted Kappa statistics were used to analyse the agreements since the SMC scale represents data in an ordinal data level (Cohen, 1968;Sim and Wright, 2005). The inter-rater reliability was fair/good to strong between pairs of assessors, while the stability of the SMC scale was observed to be strong, demonstrated by the test-retest reliability. The distinction between test-retest and intra-rater reliability in the literature can sometimes cause confusion. The objective in test-retest reliability is to test the stability in the feature being tested over time, and the objective in intra-rater reliability investigation is to test the stability of the rater over time. However, achieving strong test-retest reliability requires the stability of both the rater as well as the feature being tested. In the reliability study, we had considered performing the inter-rater reliability testing from video recordings, but as our intention was to capture assessment in clinical reality, this was ultimately not done (van Genderen et al., 2010).

The intervention study was performed in the children's everyday environment; the inclusion of children in the two groups was performed by the habilitation centres. A randomization was not possible to perform due to travel distances and limited numbers of children in the target inclusion criteria at each centre. The examinations of the children were completed at each child's local habilitation centre and the assessors were not blinded to the intervention the child received. The assessors did not participate in training of the children. However, the assessors had long experiences of the selected measurements, i.e., PEDI, GMFM-66, GAS, SMC, ROM and MAS. All in all, these factors can simultaneously be considered as weaknesses and as strengths. In a recent systematic review of physical therapy interventions for children with cerebral palsy, the authors concluded that there was some moderate, but mostly limited evidence, of the various PT interventions. The lack of evidence was due to limitations in methodological quality and to variations in population, interventions and outcomes (Anttila et al., 2008). Well-designed studies are needed, but there are many difficulties to bridge. The population of children with CP is heterogeneous and the child's/family's need of therapy throughout life varies. Furthermore, in an intervention study, the outcome measures are selected based on the investigated variables (Manns and Darrah, 2006). All these factors imply a huge variation among studies. Although randomized controlled trials are often referred to as the "gold standard" for outcome research, Grossman and Mac-Kenzie contend that, depending on the internal validity of data

obtained during a randomized controlled trial and the clinical question, the results of non-experimental designs may provide stronger evidence (Grossman and Mackenzie, 2005;Palisano, 2006;Sussman, 2010;Rosenbaum, 2010). Determining the most efficient treatment options requires careful assessments and evaluations of treatments effects.
However, the evaluation of treatment effects could be difficult to relate to just one factor due to the combination of treatments (Gormley, Jr., 2001). Today the phrase “ecological validity” sometimes is used, which means that the results from research are possible to generalize in the natural environment.

The choice of evaluation measures was based on the age of the participants, the context and the aim of the research. GMFM-66 and PEDI are frequently used in the target group of children with CP and the psychometric issues have been reported to be good (Haley et al., 2010;Russell et al., 2010). GAS was chosen as a measure of individualized change since research identified GAS as more flexible to capture family-selected goals than the Canadian occupational performance measure (COPM) (Cusick et al., 2006). The combined use of GAS and ICF-CY has also been recommended to coordinate, simplify and standardise assessment and outcome evaluation (McDougall and Wright, 2009).The possibility to assign weights to the goals was not used, as the families selected goals they considered equally important for their child to achieve (Bovend'Eerdt et al., 2009). The available studies reporting effects after goal directed therapy/functional therapy have not included assessments of body function measures. Using the ICF-CY as a frame of reference offers a possibility to both observe aspects of limitations in activity and participation as well as impairments in body function and structure (such as SMC, PROM and MAS) in children with CP. The SMC was assessed since research demonstrated interesting relationships between SMC and gross motor activity (Ostensjo et al., 2004;Fazzi et al., 2005;Desloovere et al., 2006;Voorman et al., 2007) and the scale was going to be used in evaluation of the intervention. The reason for assessing PROM and using MAS was the fact that they measure aspects of body functions frequently affected in children with CP (Hagglund and Wagner, 2008;Nordmark et al., 2009). Today the aim of treatment is to improve activity and participation, but body functions are affected and must be addressed in therapy. By investigating the relationships between domains of the ICF-CY, we might reach a better understanding of how this best is done.

LIMITATIONS OF THE STUDIES

Two aspects in the study design which contribute to a lack of control for confounding variables and bias were that the two groups were not randomized, and that the researchers were not blinded to which of the two interventions the children underwent. The GDT group also received a more extensive examination than the AT group. The studies lack information on the children’s magnetic resonance imaging (MRI) findings and the comorbidity which might have been of value to further clarify the study results. The ethical application did not include a request to collect such data from the journals, and MRI data is not always present in such young children. Recruitment of a larger sample could possibly have permitted more advanced statistical calculations, particularly in Study III. The results from the intervention study are not possible to generalize since no children in GMFCS V participated and since there were small sample sizes in GMFCS III and IV.

Conclusions and clinical implications

The investigation of the SMC scale demonstrated that inter-rater reliability was fair/ good to strong and that the test-retest reliability was strong. Clinical implications of this study are that a careful observation of the specific muscles activated is important, and that assessment of selective motor control should be performed after examination of range of motion and spasticity, to obtain knowledge of the available range of movement for the specific child. The results imply that repeated testing should preferably be completed by the same assessor.

A group of children with CP in pre-school ages and GMFCS I-IV demonstrated clear gains in everyday activities and gross motor capacity as a result of goal-directed therapy in comparison to children receiving activity focused therapy. The main differences were the formulation of individualized goals and the group meetings in the group receiving goal directed therapy while the activity focused group received generalized aims and individual therapy. The clinical implication is that individualized goals and possibly also group training and parental education give beneficial effects.

Gross motor capacity improved in a group of children receiving goal directed therapy and was maintained 12 weeks later. The individualized goals were reached to a high extent, and the children gradually progressed towards higher performance after the end of the intervention. Assessments of SMC in the ankle, passive range of motion and MAS scores did not reveal any clinical changes after therapy. The clinical implication is that therapy focusing on family-selected goals with practice integrated in the everyday environment can contribute to performance of everyday activities but possibly also support gross motor capacity.

All goals of a group of children with CP in preschool ages were classified in the Activity and Participation domain in ICF-CY. Most goals were linked to codes belonging to the Mobility and Self-Care chapters. The families' choice of goals was associated with children's scores in standardized measures at the time of goal-setting. The goal attainment observed after therapy was associated to improvement in standardized measures. The results confirm the use of both standardized and individual measures, and emphasize the inclusion of families/children in goal-setting.

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