From the Department of Woman and Child Health
Karolinska Institutet, Stockholm, Sweden

Performing bimanual activities in everyday life – experiences of children with unilateral cerebral palsy

Annika Sköld

Karolinska Institutet

Stockholm 2010

Cover photo: Stefan Thelin

Published by Karolinska Institutet. Printed by Reproprint AB

ISBN 978-91-7409-995-9

ABSTRACT

In everyday life, humans perform activities. Some activities demand the use of two hands and may be challenging to persons with reduced function in one hand. Alternative ways of performing such activities may then have to be used. However, when performing activities, humans also reveal who they are, as performance reflects individual preferences and values. There are many alternative ways of performing a given activity, and in manual activities, one aspect that may vary is how the hands are used, what role each hand is allocated, and how mobility and grasp forces are applied. People with unilateral cerebral palsy (CP) have reduced hand function due to an early brain lesion. This affects the hand and arm on one side of the body, reducing the range of possibilities to use the affected hand. This thesis therefore describes how activities are performed in the presence of the diagnosis of unilateral CP and how such performance is viewed by the affected children and adolescents themselves. To describe the complexity of hand use and activity performance, the preceding matters were considered in relation to changes experienced after upper extremity surgery (UES) and in relation to other groups with reduced hand function due to different diagnoses. Due to the lack of questionnaires focusing on activities usually performed using both hands, part of the work of this thesis research was to develop a new questionnaire.

The results indicated that children and adolescents with unilateral CP experience problems related to bimanual activities. Finding a suitable performance alternative might pose a dilemma while it is necessary to consider aspects in the activity, within oneself and in the environment. Compared with children and adolescents with obstetric brachial plexus palsy (OBPP) or upper limb reduction deficiency (ULRD), those with unilateral CP seem to experience more problems and perform fewer bimanual activities independently. Adolescents treated with UES experience improvements in activity performance and in appearance. As these experienced improvement were mostly related to changes in everyday life, and did not directly correspond to aspects that are measured objectively, it is important also to assess *qualitative* aspects of activity performance and hand use in everyday life. The Children's Hand-use Experience Questionnaire (CHEQ) includes activities that are frequently performed independently with two hands by children aged 6–18 years with the diagnoses ULRD, OBPP, and unilateral CP. CHEQ also displays good signs of validity in terms of test content and internal structure.

SVENSK SAMMANFATTNING

I vårt dagliga liv utför vi människor aktiviteter. Vissa aktiviteter kräver medverkan av båda händerna (bimanuella aktiviteter) och kan bli en utmaning för personer med nedsatt handfunktion. Det kan då bli nödvändigt att finna alternativa sätt att utföra aktiviteten. Aktivitetsutförande är emellertid inte bara att utföra en uppgift. När vi utför aktiviteter visar vi vilka vi är, utförandet återspeglar individens preferenser och värderingar. Aktivitetsutförande kan variera på många sätt och i bimanuella aktiviteter är en varierande aspekt hur man använder händerna; viken roll vardera handen tilldelas, hur man rör hand och arm och vilka krafter man använder. Personer med unilateral cerebral pares (CP) har nedsatt funktion i ena handen på grund av en hjärnskada tidigt i livet. Detta påverkar hand och arm på ena sidan av kroppen och minskar möjligheterna till variation i hand användande. Denna avhandling beskriver därför hur bimanuella aktiviteter utförs av barn med unilateral CP och hur barnen och ungdomarna själva uppfattar sitt aktivitetsutförande. För att kunna beskriva komplexiteten i handanvändande och aktivitetsutförande har det studerats också i förhållande till andra diagnosgrupper med liknande funktionsnedsättning och till förändringar i samband med övre extremitetskirurgi (UES). Det finns inga frågeformulär som fokuserar på bimanuella aktiviteter, utvecklandet av ett sådant frågeformulär har därför varit en del av detta avhandlingsarbete.

Resultatet visar att barn och ungdomar med unilateral CP kan uppleva problem med att utföra bimanuella aktiviteter. Det var inte alltid lätt att hitta ett alternativt sätt att utföra aktiviteten på och att välja strategi kunde ibland vara ett dilemma när hänsyn till aspekter i aktiviteten, hos sig själv och i omgivningen behövde tas. Jämfört med barn med OBPP och ULRD visades sig barnen med unilateral CP uppleva mer problem och utföra färre bimanuella aktiviteter självständigt. Ungdomar som genom gått behandling med övre extremitetskirurgi upplevde förbättringar både i användandet av handen, aktivitetsutförande och utseende. Dessa förbättringar beskrevs i förhållande till aktiviteter i dagliga livet och motsvarade inte direkt de aspekter som mättes i de objektiva mätningarna före och efter behandlingen. Det är därför viktigt att undersöka också kvalitativa aspekter av aktivitetsutförande och handanvändande i dagligt liv. Frågeformuläret Children’s Hand-use Experience Questionnaire (CHEQ) har visat sig inkludera aktiviteter som oftast utförs självständigt och med två händer av barn i åldersgruppen 6–18 år med diagnoserna ULRD, OBPP, och unilateral CP och har visat på god validitet.

List of Publications

I. Sköld, A., Josephsson, S., Eliasson, AC.Performing Bimanual Activities: The Experiences of Young Persons With Hemiplegic Cerebral Palsy. *American Journal of Occupational Therapy* 2004 Jul-Aug;58(4):416-25.

II. Sköld A, Josephsson S, Fitinghoff H, Eliasson AC. Experiences of Use of the Cerebral Palsy Hemiplegic Hand in Young Persons Treated with Upper Extremity Surgery. *Journal of Hand Therapy* 2007;20:262-73.

III. Sköld, A., Norling-Hermansson, L., Krumlinde-Sundholm, L., Eliasson, AC. Development and evidence of validity for the Children's Hand-use Experience Questionnaire – CHEQ. *Submitted.*

IV. Sköld, A., Eliasson, AC., Norling Hermansson, L. Bimanual Hand use in children with unilateral hand dysfunction – differences related to diagnosis. *In manuscript.*

V. Sköld A, Ekholm C, Eliasson AC. 'Improved Hand function remains after upper-limb tendon transfer and muscle release in children with hemiplegia'. *Developmental Medicine and Child Neurology* 1999;41(4):284-5.

CONTENTS

1 FOREWORD 1
2 INTRODUCTION 2
2.1 The importance of performing activities 3
2.1.1 Bimanual activity performance varies with activity, person and environment 4
2.1.2 Children's activity performance 6
2.2 Cerebral palsy 7
2.2.1 Unilateral cerebral palsy 7
2.3 To achieve knowledge of the performance of bimanual activities 11
2.4 Upper extremity surgery in unilateral CP 12
2.5 Other diagnoses involving unilateral dysfunction of hand and arm 13
3 RESEARCH AIMS 14
3.1 General Research Aim 14
3.2 Specific research aims: 14
4 MATERIAL AND METHODS 15
4.1 Design 15
4.2 Methods 16
4.2.1 Study I 16
4.2.2 Study II 17
4.2.3 Study III 18
4.2.4 Study IV 20
4.2.5 Study V 21
5 FINDINGS 22
5.1 Study I 22
5.2 Study II 22
5.3 Study III 23
5.3.1 Evidence for validity based on test content 23
5.3.2 Evidence for validity based on internal structure 23
5.4 Study IV 24
5.5 Study V 25
6 DISCUSSION 26
6.1 Hand use demands attention 26
6.2 Problems in activity performance need to be acknowledged 27
6.3 Changes in activity performance are seen after UES 29
6.4 A need to measure the subjective view of performance in bimanual activities. 30
6.4.1 Children as respondents or parents as proxy-raters? 31
6.5 Methodological considerations 32
6.6 Ethical considerations 33
7 CONCLUSION AND CLINICAL IMPLICATIONS 35
8 FUTURE RESEARCH 36
9 ACKNOWLEDGEMENTS 37
10 REFERENCES 38

List of abbreviations

AHA	Assisting hand assessment
ANOVA	One-way analyses of variance
CFUS	Caregiver Functional Use Survey
CHEQ	Children’s hand-use experience questionnaire
CP	Cerebral palsy
MACS	Manual Ability Classification System
OBPP	Obstetric brachial plexus palsy
PCA	Principal component analysis
PEDI	Pediatric Evaluation of Disability Inventory
PMAL	Pediatric Motor Activity Log
PUFI	Prosthetic Upper Extremity Functional Index
UES	Upper extremity surgery
ULRD	Upper limb reduction deficiency

1 FOREWORD

My research interest started when working as an occupational therapist in a Hand rehabilitation clinic. There I met many children with unilateral cerebral palsy (CP) undergoing upper extremity surgery (UES) and their families. The families often expressed that the intervention had resulted in important changes, making it easier to use the hand in everyday life. The demands on evidence based practice was increasing at this time, about 15 years ago, and in discussions with other professionals there were two questions which kept coming back; are the effects of the treatment remaining over time, and do the children use their hand more after the treatment? Encouraged by the hand surgeon at the hand rehabilitation department, I started off to investigate the issues. A quantitative study on objectively measured outcomes and a conjunct qualitative study on the experiences of a group of children and their families brought new information and understanding of the issue. However, while the interviews described experiences after UES, it also became apparent that they described a dilemma encountered by the participants of how to perform activities, in each situation. Children with unilateral CP are in literature generally described as being independent in everyday life activities to a high extent; it was therefore surprising that activity performance was described as a problem that constantly needed to be addressed in their daily life. This finding led me to go on with more studies, related to the performance of bimanual activities and to how the problems that they described can be acknowledged in clinical settings.

2 INTRODUCTION

This thesis takes as its starting point the body of occupational therapy knowledge. The focus is on how activity performance is affected in children and adolescents with unilateral cerebral palsy (CP). Knowledge from neuropediatrics will be used in seeking to understand how a person's particular condition can influence activity performance in unilateral CP. The aim is to create knowledge that is clinically usable and contributes to an understanding of how activity performance is formed in the presence of unilateral CP in children and adolescents.

Many everyday life activities are performed using both hands. Children and adolescents with unilateral CP, however, despite reduced function in one hand and arm, can often handle everyday life activities well and usually do not require the support of personal assistance, technical aids, or extensive interventions from children's habilitation services. Approximately 90% of children with unilateral CP have been found to independently handle most kinds of everyday life objects (Arner et al., 2008; Eliasson et al., 2006; Öhrvall et al. 2010), and in a study of participation, comparing CP subgroups (Hammal et al., 2004), children with unilateral lesions were found to manifest the highest level of participation. However, an early finding in this thesis research indicated another perspective: despite being able to handle most everyday life activities, adolescents with unilateral CP experience many problems in activity performance related to arm and hand dysfunction (study I). This apparent contradiction is the driving force of the present research.

The following introduction will describe in greater detail why activity performance is important as well as the concept of "bimanual activities" and how they may become problematic to children with unilateral CP. The work focuses on people aged 6–18 years, and study participants will collectively be referred to as "children". Though some of these participants are adolescents, they are also children in that their position in the family is that of a child. The hand with reduced function will be referred to as "the affected hand" and the other hand "the dominant hand". Two substantially different perspectives are presented in this work; the participants own description and the professional evaluation. Even though the professional evaluation might include an aspect of subjectivity, for sake of clarity, the participant's own description (as expressed in interviews or in questionnaires) will be referred to as subjective, in contrast to professional evaluations which will be referred to as objective.

2.1 THE IMPORTANCE OF PERFORMING ACTIVITIES

Occupations can simply be described as “what we do” (Christiansen 2005). “What we do” can however be viewed from several perspectives, showing a complex construct. From one view, the performance of occupations can be described as a concrete execution of tasks; “getting something done”. The routinely performance of daily activities may require low attention and may become more or less automatic (Kielhofner, 2008). However, another view is that occupational performance is essential to humans as a carrier of meaningful experiences (Jonsson & Josephsson, 2005). What we do and how we do it depends on individual interests, values, sense of competence and effectiveness (Christiansen, 1999; Kielhofner, 2008). Occupational performance therefore reflects who we are and becomes a presentation of oneself (Christiansen 1999). Further, “what we do” is also dependent on function in bodily systems and mental and cognitive abilities as well as physical and social environment including cultural and political structures (Kielhofner, 2008). Thus, human occupation and "what we do" is a complex interplay between individual and environment and when being in focus in research, this complexity needs attention.

Activity and occupation have been defined in various ways in the literature. In this thesis, *activity* performance will refer to simple “doing”, whereas *occupational* performance refers to the carrying out of an activity imbued with special personal and cultural values and meanings. Although the two cannot always be clearly distinguished, one distinguishing feature is that activity performance can differ in many ways within the same occupation. For example, the activity of “taking out money from a wallet” may be part of the occupation of “luxury shopping” or of “giving money to charity”. Even though the activity of taking out money is the same, these two occupations of which it is part may be associated with very different personal and cultural values and meanings.

Occupational performance has been described as dependent on the specific situation in which it occurs; a certain Activity is performed in a certain Environment by a certain Person, and the performance is dependent on variations in these three factors. Some theories emphasize that there is interaction between these three factors. Nelson (Nelson, 1988) describes occupation as the relationship between form (meaning the physical and socio–cultural dimensions) and performance, i.e., when someone “performs” an occupation, he or she goes through the form, making occupation the outcome of the joining of form and performance. According to Law and collaborators (1996), occupational performance “results from the dynamic interaction between people, their occupations and roles, and the environments in which they live, work and play”. This dynamic view, according to which occupation results from the interaction between the person, activity, and environment, has been chosen as a frame of reference in this thesis. However, since unilateral dysfunction of hand and arm mostly affects the performance in activities where both hands are needed, the focus in this thesis will be on such activities and specifically on how the hands are used. To my knowledge, no single theory or model describes variability in all these three aspects focusing on bimanual activities and hand use, thus, knowledge from various fields will be used to enlighten this.

2.1.1 Bimanual activity performance varies with activity, person and environment

The expression “bimanual activities” may give the impression that there is a clear distinction between activities performed using one or two hands, that is however not the case. A person makes choices about how to perform an activity and how to use the hands. For example, the activity “taking out money from a wallet” can be performed using one or two hands, and either hand can be selected to hold the wallet while the other picks up the money. The holding and picking up can also be done in several ways as regards the position of hand and fingers, forces applied, and time taken for the performance. In this thesis, the term “bimanual activities” refers to activities typically performed using both hands and difficult to perform using only one hand. The term “hand use” will in this thesis refer to whether one or two hands are used and how they are used. Bimanual activities are central to this thesis, because the performance of bimanual activities is often the crucial challenge for people with unilateral dysfunction of the hand and/or arm (Greaves et al., 2010).

Leconte and Fagard (2006) found that hand use in children aged 5–12 years varied with three types of factors: intrinsic, environmental, and task-related. Leconte and Fagard studied experimental situations, which explains their use of the word “task”; their view can however be taken as analogous to the view of activity, person, and environment jointly influencing activity performance, suggesting that this view is suitable also when focusing on hand use in bimanual activities. To understand how performance and hand use are formed in bimanual activities, the influence of these three aspects will be addressed.

2.1.1.1 Activity

Even though Leconte and Fagard (2006) found that the task influenced the hand used in performance, their use of experimental situations does not lend itself to drawing conclusions applicable to everyday life activities. However, the psychologist Guiard (Guiard, 1987) describes bimanual performance from the perspective of activities. Guiard categorises bimanual activities into three categories: unimanual (e.g., dart throwing), bimanual asymmetric (e.g., playing the violin), and bimanual symmetric, in which the two hands play the same role, either in phase (e.g., rope skipping) or out of phase (e.g., rope climbing). This classification has since been used in various contexts. However, Guiard suggests that no activity can be proven to be truly unimanual; for example, in dart throwing, the other hand may contribute to postural function, influencing the performance. Thus, some activities obviously demand the use of both hands, while in others, hand use varies and is not always obvious.

2.1.1.2 Person

As stated earlier, a person can choose how to perform an activity, based on personal preferences and in relation to the activity itself and the environment. Values and interests, as well as belief in what one can achieve, influence the choices made in activity performance (Kielhofner, 2008). It can be assumed that this is true for bimanual activities as well as other activities. However, no literature has been found as regards how the persons own values influence hand use. However, much knowledge has been generated as regards the neural control of the hand. Neural control of the hand involves several areas in the brain, working together in neural networks, selecting and planning movement performance (Forssberg, 1999; Pehoski, 2006). Without having to

pay attention to and plan every movement in a task, a person performs well known tasks in an efficient way. For example, when opening a drawer with one hand and manipulating something in the drawer with the other, the first hand starts, reaching out towards the drawer, preshaping the hand in relation to the size of the handle, and the second hand is starting before the first hand is finished, creating a temporal overlap between the movements of the two hands (Hung et al., 2004; Jeannerod, 1984). Neural control is guiding the movements also when grasping an object. When lifting an object, grip and load forces act in synergy, the simultaneous initiation and parallel increase of forces producing efficient and smooth movement. The demands of the task are anticipated rather than depending on sensory and proprioception feedback. An internal representation of an object's properties is built from previous knowledge and may be updated if afferent information indicates miscalculation of the forces needed (Flanagan et al., 2009). The way of grasping an object is also subconsciously influenced by how the object will be used; the grasp used when picking up an object may be depending on how the object will be used when handled in next stage of the performance (Steenbergen et al., 2007). Variation in both hand choice and grasping pattern is greater in younger than older children, reflecting the progressive refinement of ability during development (Forssberg, 1999; Leconte & Fagard, 2006).

Thus, whereas the general performance of the activity is influenced by values and personal preferences, humans are generally not making conscious choices about how to use the hands in well known activities, rather it is automatically formed by neural control. This is an important knowledge in this thesis, because if the neural control is impaired, hand use may not be automatic in the same way and the way of using the hands may become an issue which demand that the person make more conscious choices. Further, the functions of the musculoskeletal and somatosensory (related to sensibility) systems in the arm and hand, including range of motion, strength, and sensibility, are also crucial for how activity performance takes form (Eliasson, 2006).

2.1.1.3 Environment

The environment influences activity performance in various ways, as it may both demand particular behaviours and discourage or disallow others (Kielhofner, 2008). The environment includes both factors that influence all of society, such as cultural and political factors, and factors specific to the situation in which the person performs an activity, such as social and physical environments and the object to be handled. Being well aware of that environment comprises many aspects, this thesis will – due to the specific focus on hand use in bimanual activities – describe the more situation-specific aspects of the environment in terms of social aspects and object properties. Social aspects include the universal need for social acceptance as well as the need for practical, informational, and emotional support (Christiansen 2005). Objects are defined by Kielhofner (2007) as "naturally occurring or fabricated things with which people interact and whose properties influence what they do with them". Handling objects demands various degrees of hand function; the physical properties of objects, such as their size, friction, and weight, require forces of various strengths, and the grasping and lifting actions must be adjusted in relation to the object to produce smooth movement (Forssberg et al., 1995,). Previous experiences of handling objects are used to estimate what movements and forces are needed, new information rapidly contributing to updating the information base and adjusting the feed-forward strategy (Eliasson, et al., 1995; Forssberg, et al., 1992).

Summarising this paragraph, there is reason to believe that hand use and performance of bimanual activities is varying depending on variables in the three aspects activity,

person and environment. It can be assumed that a dynamical interaction between these three aspects form the activity performance but this has so far not been described specifically for the performance of bimanual activities when one hand has reduced function. Arnould and colleagues (Arnould et al., 2007) studied the relation between hand function and manual ability and found in a group of children with CP, aged 6-15, where about half of the participants had unilateral involvement, that hand function in terms of strength, dexterity and sensibility are not related to manual ability (as measured by the ABILHAND-Kids) in a predictable, straightforward way. In their study, gross manual dexterity in the dominant hand explained only 44% of the variance in manual ability while grip strength in the non dominant hand explained further 14%. The authors discuss that several factors may contribute to hand use both within person and environment, and suggest further research "to identify which contextual factors really contribute to the achievement of manual activities" (Arnould, et al., 2007)(p.713).

2.1.2 Children's activity performance

The activity performance of children differs from that of adults while children's activities are part of being in a certain developmental phase. Mandich and Rodger (Mandich & Rodger, 2006) describe children's activities as possibilities to develop abilities and to become social beings. Occupational skills can only be learned and mastered by doing. Successful doing can help children develop a healthy sense of who they are and what they can become. Therefore, the enabling of doing is important. Lawlor (Lawlor, 2003) emphasizes the importance of *doing with*, viewing the social aspects of childhood occupations as the most important. Play is central in children's lives, though the type of play differs according to age. In the ages of middle childhood (age 6–10 years), structured games and organised play predominate. Interacting with peers and following rules becomes more important. By 8-9 years children become more interested in crafts and hobbies as well as organized sports, it becomes more important to achieve something with the play. Social play increases in importance, e.g., belonging to groups and talking to friends (Case-Smith, 2005). Whereas earlier in life, play is more characterized by the qualities of exploring, participating, and imitating, the play of middle childhood is more characterized by expectations of certain behaviours. However, in the presence of restricted mobility, play can be different. An example is given by Tamm and Skär (Tamm & Skär, 2000). They found, based on interviews with children aged 6–12 years with restricted mobility due to various diagnosis, that children mostly played alone or with adults. Play with friends – either interactive or as an onlooker – was less common. During adolescence, the child takes on increased responsibility in activities of daily life and independence becomes more important. It is also an important aspect to fit in with peers and to be successful in obtaining a job (Shepherd 2005).

In conclusion, the view of activity performance as the outcome of dynamic interplay between activity, person, and environment is useful when describing the performance of bimanual activities, emphasizing the variability in performance arising from these three factors. Thus, when no dysfunction exists, performance choices are made by the person, both volitionally and by neural control. However, we lack knowledge of how the presence of unilateral CP affects this dynamic interaction and how this shapes activity performance. The following section will describe how the diagnosis of unilateral CP alters the prerequisites for activity performance.

2.2 CEREBRAL PALSY

CP occurs in 2–3 per 1000 live births in western countries and probably more frequently in developing countries (Krägeloh-Mann & Cans, 2009). A recent study of CP occurrence in western Sweden indicated a prevalence of 2.18 in 1000 live births (Surveillance of cerebral palsy in Europe, 2000). The diagnosis of cerebral palsy "describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that 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 disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems" (Rosenbaum et al. 2007, p. 9). Considering the focus of this thesis, it is worth noting that the definition points out that activity performance is limited. The group of CP is a heterogeneous group and it has been seen that independent performance in self care and mobility varies strongly between individuals (Öhrvall, et al., 2010).

2.2.1 Unilateral Cerebral Palsy

In the group of children with CP, about 30% have an involvement mainly on one side of the body (Staudt, 2008). In the 2007 updated classification of CP (Rosenbaum, et al., 2007), the term "unilateral CP" is recommended to replace the earlier used term for this condition, "hemiplegia". Before describing how activity performance and hand function are affected in unilateral CP, other aspects affected will be described to obtain an overview of the dysfunction. The leg on the affected side is often involved; (Uvebrant, 1988), in a study of 169 children with unilateral CP, found that 30% had a moderate limp but only 3% were unable to walk. Epilepsy was found in 34% and 18% had mental retardation. In the remaining subjects, with no mental retardation, 46% nevertheless had difficulties at school, mostly related to hyperkinetic behaviour or perceptual impairment (Uvebrant, 1988). In unilateral CP, mental retardation, epilepsy, and severe mental hemiplegia often coexist; according to Aicardi et al. (Aicardi et al, 2009), two subgroups can be discerned: "severe cases with multiple disabilities and a poor outlook for social and professional integration, and mild cases that interfere relatively little with everyday life". Psychiatric problems are common. Goodman and Graham (1996) studied a group of children with unilateral CP who attended a normal school, had an IQ over 90, had never had a seizure, and had a mild hemiparesis; they found that approximately 40% of the children manifested a psychiatric disorder, conduct, emotional, and hyperactivity disorders being predominant.

2.2.1.1 Hand function

In unilateral CP, hand use is limited by several factors related to disturbed hand function. The arm and hand are affected by various degrees of spasticity in some muscles combined with weakness in others, resulting in difficulties extending the wrist, supinating (outwardly rotating) the forearm, and straightening the thumb and fingers (Brown & EG, 2000). These movement restrictions result in slow performance, and sensibility is often impaired (Majnemer et al., 2008 a). In a study of 25 children with unilateral CP, Krumlinde-Sundholm and Eliasson (2002) found that 75% had decreased sensibility, having two-point discrimination of 3 mm, measures of sensibility being

highly correlated with dexterity. In everyday life, reduced hand function interferes with reaching and adjusting limb position in relation to the object to be grasped, while reduced sensibility makes it difficult to regulate the force in relation to the object and to use sensory information to update grasp quality. Even though the diagnosis indicates that the impairment is unilateral, it is now well known that limbs on both sides of the body are sometimes affected (Bax et al., 2005; Duque et al., 2003).

There are few studies of the development of hand function in unilateral CP. Fedrizzi et al. (2003) reported no significant change in grasp comparing children when they were 1–4 with when they were 11–18 years old. However, the test used by Fedrizzi et al., to evaluate what part of the hand (from whole hand to pincer grasp) was used when picking up three cubes, was said to display questionable reliability in a systematic review (Greaves, et al., 2010). Smits-Engelsman et al. (Smits-Engelsman et al., 2004) compared two age groups of children with unilateral CP, 5–9 and 10–15 years old, as regards force generation in finger flexion, and found that the older age group generated less force than did the younger one, indicating that strength may decline with time

2.2.1.2 Neural control of hand movements

In normally developing children, the synergy between grip and load forces, which enables smooth movements, is achieved at the age of approximately 6–8 years (Forssberg et al. 1991) and the ability to use information in an anticipatory way when grasping is achieved at the age of 8–11 years (Forssberg, et al., 1992). In children with CP aged 6–8 years, this synergy between grip and load force does not resemble that of typically developing children. In addition, children with CP rely on feedback to adjust the grip, which is time-consuming, and employ safety strategies in the form of using more grip force than needed (Forssberg et al. 1999). However, a study comparing 10 children with CP aged 6–8 and 19–21 years found improvement over time; synergy between grip and load force improved and there was less variation between trials, together with a decrease in movement sequentiation. Time to complete the grip-lift task decreased by 22%, and a parallel 45% decrease in time use was seen in the Jebsen-Taylor test (Eliasson et al., 2006). The ability to select a grasp that will be the most beneficial in the movement end-state is developed at the age of 5–6 years in typically developing children and assumes an adult-like pattern at the age of 10 (Crajé, Aarts et al., 2010; Thibaut & Toussaint, 2010). In a group of typically developing children aged 3–6 years, the proportion of comfortable end postures was found to increase with age; this was not the case in children with unilateral CP (Crajé, et al., 2010). In children with unilateral CP, coordination between limbs when performing bimanual activities is often difficult due to coupling, resulting in increased control of the movement but making asymmetrical bimanual movements (in which the two hands perform different movements) difficult (Kuhtz-Buschbeck et al., 2000; Utley & Steenbergen, 2006). In addition, coordination when holding an object in one hand and placing another object on top of it (requiring a simultaneous force increase in one hand and decrease in the other) has been found to be difficult; instead of increasing the grip force in relation to the expected weight deposited by the other hand, children with CP decrease the grip force (Islam et al., 2010).

2.2.1.3 Activity performance

Knowing that object handling may be difficult for children with unilateral CP leads to the question of how they can perform daily living activities. The MACS can be used to classify how children with CP handle manual activities in everyday life. Children without restricted independence due to manual ability limitations are on MACS level I or II (Eliasson et al., 2006; Morris et al., 2006). However, although independence is not

restricted, children in levels I and II may experience limitations. In level I, limitations may be present as regards the ease of performance of manual tasks that require speed and accuracy. In level II, a somewhat reduced quality and/or speed of performance may be present, and some tasks may be avoided or alternative ways of performance may be used. According to three studies, 87%, 92%, and 90% respectively of children with unilateral CP, were classified as level I or II (Arner et al., 2008; Eliasson et al., 2006; Öhrvall, 2010).

Pediatric Evaluation of Disability Inventory (PEDI) is a criterion-based norm-referenced instrument, assessing capability and independence in everyday life based on interviews with parents. Independence in the PEDI self-care domain – represented by a score of 100 – is typically achieved by the age of about 6 years (Haley et al.,1992). Öhrvall et al. (2010), in a study of children with CP aged 3-15 years, reported that children at MACS level I achieved scaled scores with a mean of 81.65 and from the age of 9, most children scaled 100. At MACS level II, the mean was 68.78, and only a few children scored 100, all of them aged 12 years or more. About half of the children at MACS levels I and II had unilateral CP. In line with this, Kerr et al. (Kerr et al., 2008), in a study of 94 children with unilateral CP, mean age 10.6 years, found a PEDI score of 75.2 in self-care. This means that still at the age of approximately 9–12 years, not all children had achieved the independence in self-care typically achieved by the age of about 6 years. However, Öhrvall et al. (2010) demonstrated that PEDI scores increased with age in children at MACS levels I and II, indicating that higher independence in self-care may be achieved, although later than in typically developing children.

2.2.1.4 Hand use

The actual use of the affected hand has been discussed from several perspectives. Greaves et al. (Greaves, et al., 2010) stated that people with unilateral CP often avoid using the affected hand. Taub et al. (Taub et al., 2004) suggested that "developmental disregard" or "learned non-use" could be the cause of not using the hand: repeated failure to use the hand leading to non-use may eventually result in an inability to use the affected hand. The efficiency of using the affected hand has been acknowledged in the development of the Assisting Hand Assessment (AHA) instrument, an objective measurement for children with unilateral hand dysfunction. In a study of the validity and reliability of the AHA it was seen in a sample of 37 children aged 1.5-5 years, that children with unilateral CP were more represented on the lower part of the scale, indicating less efficient hand use, compared to children with obstetric brachial plexus palsy (OBPP) and children with no dysfunction (Krumlinde-Sundholm & Eliasson, 2003). Even though this finding refers to younger children, no correlation was found with age, so the results may apply to older children as well. Thus it has been generally acknowledged that hand use is affected in unilateral CP, as regards both how often and how efficiently the affected hand is used. However, it has not been shown how the person experience this reduced ability to use the hand and how this influences the way the person chooses to perform bimanual activities. Given that hand use, when no disability is present, to a high degree is the result of neural control rather than of conscious choices, there is a need to find out how hand use is formed when disability is interfering with neural control as well as with musculoskeletal and somatosensory systems.

2.2.1.5 Social aspects

An important aspect of the social environment is the possibility of experiencing involvement in life situations, and this has been defined as participation by the World Health Organisation (WHO 2008). A low intensity of participation has been found in

children with CP in general, and informal activity is more common than are formal activities with friends in the broader community (Imms et al., 2009; Law et al., 2006). Children with unilateral CP have been reported to have fewer socially related problems than children with other forms of CP (Imms, et al., 2009; Majnemer et al., 2008, 2010; Orlin et al., 2010). Imms et al (2008) suggest that motor ability may not be a limiting factor until children are quite impaired. Therefore, compared with children with more severe forms of CP, children with unilateral involvement display fewer problems related to social factors. On the other hand, Lepage et al. (1998) stated that, even though children with unilateral CP were less disturbed in their life habits than were children with quadriplegia, "even children with mild hemiplegia can show significant disruption in recreational habits" (p. 270). It may thus be relevant also to study this group specifically, and to make comparisons to typically developing children, since many of them attend mainstream schools.

It may be fruitful to view the social participation for children with a mild disability from two different aspects. Pirpiris et al. (2006) found that, when wellbeing was divided into a psychosocial and a functional part, mild cerebral palsy had greater effects on children's psychosocial wellbeing than would be predicted by their functional disability; functional measurements were good at predicting functional wellbeing, but were weak at predicting the psychosocial aspects of wellbeing. Asbjørnslett & Hemmingsson (2008) also found a dual perspective in a study based on interviews with teenagers with disability due to various diagnoses, attending mainstream school. The teenagers experienced being just like their classmates, referring to the personality, but they were also aware of that others saw them as a person with problems, referring to capability. A recent study focusing specifically on unilateral CP (Moore et al., 2010) used interviews and thematic analysis with focus on quality of life. Findings are presented briefly, however it is concluded that participants (aged 5–17 years) did not focus on having CP on a daily basis and did not feel that it influenced their lives. In contrast to the two previous presented studies, this shows a more unitary view and it can be speculated then, whether the study by Moore et al captured the first, more positive, aspect. Negative aspects have however also been described in other studies. Yude et al. (1998) found that, compared with their classmates, children with unilateral CP were more often rejected, less often popular, had fewer friends, and were more often victimized. Children with unilateral CP have also been found to experience lower Quality of Life in the areas of physical, athletic, and scholastic competence (Russo et al., 2008; Schuengel et al., 2006).

In conclusion, studies demonstrate that children with unilateral CP have reduced hand function in terms of mobility, sensibility, and less efficient hand use, as described in experimental studies of neural control and from observations. In the personal element of the activity–person–environment interplay, the prerequisites for hand use on the part of children with unilateral CP differ from those of children with no dysfunction, though the activities that they are expected to perform are largely the same as those of children with no dysfunction. To a certain degree, children with unilateral CP are restricted in the performance of daily activities and in social participation, although less so than children with more severe CP. However, little is known about how the dynamic activity–person–environment interplay manifests itself in the presence of CP, nor are there any studies of how people with CP view this matter.

2.3 TO ACHIEVE KNOWLEDGE OF THE PERFORMANCE OF BIMANUAL ACTIVITIES

Reduced ability to use one hand might influence not only activity performance (the outcome of the dynamic activity–person–environment interplay), but also one's experience of how this dynamic interaction shapes activity performance. For example, if the activity "taking out money from a wallet" is performed when the social environment comprises people in hurry waiting in line behind a person with unilateral CP, and the wallet is difficult to handle, having only a small tab on the zipper, how does this affect how the person chooses to perform the activity, and what is it like to be in his/her position of having to perform the activity? In line with the strengthened position of the child in society today, bolstered by the United Nations Declaration of the Rights of the Child (General Assembly U.N., 1989), and by ideas of client- and family-centeredness in habilitation, there is good reason to learn about the subjective experiences of the person and his/her family. One method for this may be qualitative interviews, which can serve to yield an understanding of the person's experiences. Another method is to use quantitative questionnaires, which can yield information about *how much* of a certain dimension is acknowledged.

Little is known about how children with unilateral CP experience activity performance in everyday life. Qualitative studies have described experiences for the heterogenous group of cerebral palsy, for example with focus on participation and quality of life, but few studies are focused specifically on the subgroup of unilateral CP. Shikako et al. (2009) found that adolescents with higher motor functioning compared themselves to peers without reduced function whereas children with more severe motor dysfunction often compared themselves to other children with motor dysfunction. This supports the reasoning that it is important to describe experiences for each of these groups separately. One study focuses on students who attend main stream schools, although not being focused on children with unilateral CP it describes a situation which is typical for many of them (Asbjørnslett & Hemmingsson 2008). A recent study focusing specifically on unilateral CP (Moore, et al., 2010) used interviews and thematic analysis with focus on quality of life. Findings are presented briefly and only a small part concern activity performance. Thus there is a need for further knowledge on the experiences of activity performance for children with unilateral CP.

Questionnaires on hand use and manual ability are available, but no existing questionnaire focuses specifically on the performance of activities that are normally performed using two hands. The most suitable so far have been ABILHAND-Kids, the Paediatric Motor Activity Log (PMAL), and the Caregiver Functional Use Survey (CFUS). The ABILHAND-Kids questionnaire, developed for the parental rating of children with cerebral palsy, has displayed good signs of validity. However, three quarters of the activities it includes could be performed using only one hand (Arnould et al., 2004). PMAL was developed for children with unilateral CP and includes a mixture of unimanual and bimanual activities (Taub, et al., 2004; Wallen et al., 2009). CFUS asks about bimanual performance, though for activities not necessarily performed using both hands (Charles et al., 2006). The lack of assessment instruments focused on bimanual activities has hindered the creation of knowledge of how activities are performed and of subjective perceptions of activity performance. The only questionnaire with a clear focus on bimanual activities is the Prosthetic Upper Extremity Functional Index (PUFI), though this scale is only applicable to children who use upper limb prostheses (Wright et al., 2001). Since there is no questionnaire that

focuses on the performance of activities that require two hands, the Children's Hand-use Experience Questionnaire (CHEQ) was developed within the frame of this thesis.

So far in the introduction to this thesis, I have discussed how activity performance is important and how children with unilateral CP have specific problems in the performance of bimanual activities. There are however several intervention alternatives to address the ability to perform activities.

2.4 UPPER EXTREMITY SURGERY IN UNILATERAL CP

Various interventions affecting performance in bimanual activities are used for children with unilateral CP (Eliasson 2008). These can be directed towards both hand function and use as well as towards activity performance. One treatment focused on hand function is upper extremity surgery (UES), in which muscles are elongated or shortened, in combination with the transfer of muscles in order to support weak muscles to achieve a more functional movement pattern (Carlson 2005). Many reports on surgical techniques and outcomes have been published, though there are few outcome studies using reliable research methods and instruments. In a 2001 review, Boyd et al. (2001) stated that many positive results were reported subjectively, but no studies using a control group were found and most studies were retrospective. One prospective study using detailed objective outcome measurements was described, reporting improved range of motion of the wrist and forearm (Eliasson et al., 1998). In a later review, van Munster et al. (2009) found that studies of functional outcome after UES were heterogeneous, so meta-analysis could not be done. However, they concluded that data from the eight studies analysed indicated that surgery positively affected wrist supination and extension (dorsiflexion), improved grip strategy, enlarged the grip repertoire, and increased spontaneous hand use. The level of evidence for functional improvement is low; however, the authors reason that functional improvement may theoretically be expected given a better hand position. In the eight studies reviewed, postoperative assessment used either a broad range of follow-up time between individuals (e.g., ranging from 2 to 10 years) or a relatively brief time perspective (approximately 18 months), and the one study that includes a long-term follow-up (Hoffer et al., 1986) does not compare short-term and long-term results. However, a so far unpublished study by Pontén et al (Pontén et al., 2010) has used the AHA to assess the efficiency of the hand use. AHA shows good evidence of validity, reliability and sensitivity for change (Holmefur et al., 2007; Holmefur et al., 2009; Krumlinde-Sundholm et al., 2007). The result showed an improvement from 44.5 to 52.5 on a 0 – 100 scale. For 10 of the 18 children in the study, the change was larger than the least significant detectable difference (SDD) of the test. Improved results were also seen using the Zancolli classification (where 11 children improved), in the achievement of functional goals and in range of movement (Pontén, et al., 2010).

Summarising this paragraph, outcome studies on UES show good result but on a low level of evidence, the use of AHA for evaluation however has contributed with more reliable measurements. There is a lack of information about what happens over time, i.e., whether the results achieved by UES are long lasting. UES has mostly been evaluated and described from an objective perspective, and few studies include the perspective of the child or family. This is an important lack of information, since patient satisfaction is generally the aim of the treatment.

2.5 OTHER DIAGNOSES INVOLVING UNILATERAL DYSFUNCTION OF HAND AND ARM

This thesis considers two other diagnoses involving congenital unilateral hand dysfunction, namely, obstetric brachial plexus palsy (OBPP) and upper limb reduction deficiency (ULRD). In OBPP, loss of function varies with the level and severity of the nerve injury and ranges from weakness to severe paralysis in either the upper or the whole arm, and sensibility may be affected as well (Gilbert, 2001; Strombeck et al., 2007). In ULRD, the reduction is of either transverse (amputation-like) or longitudinal type, in which partial or intact fingers can be present on a more or less complete arm (International Organization for Standardization, 1989). Hence, people with longitudinal ULRD may experience practical limitations due to having a short arm or missing some fingers or parts of fingers (Brown et al., 1996; Hermansson, 2004). These two diagnoses share with unilateral CP the characteristic of having one properly functioning and one less functioning hand; however, these two diagnoses do not affect the brain and do not yield spasticity. Comparisons between the three groups may therefore serve to describe how bimanual activity performance can vary with and without the presence of brain lesion.

In conclusion, this introduction has identified that “bimanual activities” can be performed in many ways. By viewing activity performance as the outcome of dynamic interaction between activity, person, and environment, we can see that choices about how to perform an activity are influenced by variations in all three factors. Children with unilateral CP have reduced ability to use one hand, and to some degree have problems independently performing everyday life activities. However, it is not known how the dynamic interaction between activity, person, and environment forms the activity performance in the presence of unilateral CP, nor how the children’s own experience is related to this. The combination of lack of questionnaires focusing on the specific problems of children with unilateral CP and the use of comparisons with children having more severe dysfunctions has made it difficult to detect and describe problems, in both clinical settings and research. UES has demonstrated promise as a treatment to improve hand function, but so far there is poor knowledge of its long-term results and of patient experience on how it affects hand use in everyday life.

3 RESEARCH AIMS

3.1 GENERAL RESEARCH AIM

The aim of this thesis is to explore and describe experiences of activity performance in adolescents with unilateral Cerebral Palsy, with special focus on hand use in bimanual activities. A further aim was to evaluate long-term outcome after UES and to develop a questionnaire.

3.2 SPECIFIC RESEARCH AIMS:

1. to increase knowledge on how bimanual activities are performed and how the performance is experienced (Study I and IV)

2. to increase knowledge on how hand use and activity performance is experienced after UES (Study II)

3. to evaluate objective outcomes five years after UES (Study V).

4. to develop a questionnaire – CHEQ – and evaluate if the performance of bimanual activities can be rated in CHEQ in a valid way (Study III).

4 MATERIAL AND METHODS

4.1 DESIGN

The complex issue of activity performance required a combination of research methods. Objective data was gathered by quantitative methods in form of measurements. Subjective data was gathered by the use of qualitative methods in form of interviews and by the use of quantitative methods in form of ratings on a questionnaire.

Data collection was done on the same occasion for Study II and V; a quantitative and a qualitative study regarding UES. The number of participants was set to 10, due to practical reasons. However, the qualitative data in Study II turned out to describe rich descriptions of experiences of performing bimanual activities, apart from experiences related to UES, the same interviews were therefore also used for a new analysis in Study I. This new analysis was complemented with data from a focus group interview with 4 persons in Study I.

In Study III and IV, 86 new participants were selected, and were in Study IV complemented with 14 more participants, in order to allow comparisons on group level.

Table I. Type of studies and number of participants.

	Focus	Design	Data analysis	Number of participants	Participants taking part in more than one study
Study I	Experiences of daily life activities in the presence of unilateral CP	Qualitative	Comparative	10	-
Study II	Experiences in relation to upper extremity surgery	Qualitative	Comparative	14	10 of these also in study I
Study III	Development and validity of a questionnaire: CHEQ	Psychometric evaluation	Rasch analysis, Descriptive	86	-
Study IV	Description of groups of children with unilateral hand dysfunction	Quantitative	Descriptive	100	86 of these also in study III
Study V	Outcome of UES in children with unilateral Cerebral Palsy, 5 years follow up	Quantitative	Descriptive	Same participants as in study I	

Note: Additionally 27 persons representing 14 families took part in group interviews during the development of CHEQ and further 18 families took part in a field testing using a preliminary version of CHEQ in study III.

4.2 METHODS

4.2.1 Study I

Study I is a qualitative study based on interviews with adolescents regarding their experiences of performing activities in everyday life.

4.2.1.1 Participants

Ten participants taking part in Study V - long term follow up - were also invited to take part in the interview in Study I, on the same occasion. The selection procedure was therefore designed in relation to Study V. Candidates were selected among participants who had taken part in a previous study and undergone UES, 5 years earlier (Eliasson et al., 1998). Additional criteria were 1/ diagnosis of unilateral CP, 2/ attending or having attended regular education at the time of surgery. Participants were invited to take part in the study, following date of surgery consecutively, until 10 participants were selected. One male and nine females, 12-24 years of age, participated. Seven participants had dysfunction on the right side, three on the left side. According to the classification of Claeys and colleagues (1983), the hand function of the participants varied from mild dysfunction (pincer grasp and individual finger movements) to moderate dysfunction (global hand use). All participants had some kind of grip ability both before and after the treatment. Five of the participants (aged 12-19) were accompanied by a parent, and the parent was then invited to take part in the interview. Those who came alone (aged 17-24) were assumed to be old enough to respond themselves.

Additional participants in Study I were recruited for a focus group interview. Local occupational therapists were asked to suggest and invite candidates who were 1/ having a diagnosis of unilateral CP 2/ attending or having attended regular education and 3/ being willing and able to express themselves in the subject. Four participants took part in the focus group; one male and three females, aged 16 to 28 years of age.

4.2.1.2 Data collection

The aim of the individual interviews was to find out how each participant practically dealt with problems related to the reduced function of the hand. The interviews were semi structured and an interview guide was used (Bogdan & Biklen, 2003; Patton, 1986), based on a pilot interview made with an 18-year-old girl with unilateral CP. The participants were asked to describe how they performed activities that in the pilot interview were identified as being typically problematic, objects used in these activities were available to allow participants to handle the objects while describing their way of performing the activities. Thereafter, the participants were asked about situations where the use of the affected hand was avoided as well as activities that they were unable to perform. The knowledge acquired through the individual interview analysis revealed that further data on the reasoning employed when choosing strategies, rather than on strategies per se, and on how the participants themselves viewed their choices, was needed. In order to achieve richer descriptions, the method of focus group (Krueger & Casey 2000) was used. All interviews were tape recorded with the participant’s permission and transcribed verbatim by the interviewer

4.2.1.3 Data analysis

Data analysis was guided by a comparative method described by Bogdan and Biklen (2003) and inspired by Grounded Theory tradition (Strauss and Corbin 1998). The process of analysis started with reading and coding the transcripts to define the main ideas discussed in the interviews. All authors thoroughly read the transcripts to gain an overall sense of the content. In the second step, coding, the first author developed a detailed coding scheme which was discussed with the other authors for feedback. Alternative ways of understanding data were sought by consequently assessing if there might be more than one way to understand the text, and the same part of text could be assigned to more than one code. Each section of text related to a certain code was identified and brought together so that all parts of data relevant to the purpose of the study could be read according to the codes and serve as a background for further analysis. The third step involved describing the findings by organizing them into a coherent and meaningful whole, analysing similarities and differences between individuals. This way of working with data yielded a new understanding. We found that the way of using various strategies for activity performance could only be understood when experiences, feelings, and values of the individual as well as the environment in the specific situation, were considered. Based on this new understanding, data were organized into two main themes with nine subthemes.

4.2.1.3.1 Trustworthiness

Several methods were used to assure trustworthiness; peer debriefing through discussions between the authors (who had different experiences and fields of knowledge); reflective thinking about any possible bias, negative case analysis, and a drive to describe data as a whole without excluding any information related to the aim of the study (Gustavsson 2000, Lincoln & Guba 1985). Our ambition has also been to yield a rich description, with quotes from participants and descriptions of similarities and differences between participants to enable the reader to value transferability (Lincoln & Guba 1985).

4.2.2 Study II

Study II is a qualitative study focused on experiences after UES treatment.

4.2.2.1 Participants

The same group of 10 children as described in Study I constituted the group of participants also in Study II. All participants had undergone UES 5-7 year earlier, and all but one had been treated by the same surgeon. Both hand surgeons were certified and well-experienced in hand surgery. The surgery had been followed by a period of intensive training by an occupational therapist and a physiotherapist. Postoperative care included immobilization with a cast for the first six weeks, thereafter using a daytime wrist support for two weeks and a night splint for six weeks. Therapy was individual for each participant and included active and passive movements, training of grip and of activities of daily living (ADL). UES intervention as described in this thesis therefore includes two parts: surgery and postoperative care. Although how these aspects relate is open to discussion, they will here be viewed as a single unit and will be referred to as “UES”. At the time of interview, none of the participants had had any surgical revision. None of the authors, including the interviewer, had been involved in the treatment of the participants. In the surgery, multiple operative procedures were carried out in one session, according to individual needs.

4.2.2.2 Data collection

Data collection was described in Study I.

4.2.2.3 Data analysis

Data analysis was guided by a comparative method described by Bogdan and Biklen (2003) and inspired by Grounded Theory tradition (Strauss and Corbin 1998). Primarily, the material was read through and condensed to contain only data that corresponded to the aim of the study and coded according to content, by the first author (A.S.). Coding was discussed between A.S., A.-C.E. and H.F and data was described according to the coding. The second stage of the analysis was guided by methods described by Strauss and Corbin (1998). Whereas the first turn of analysis was based on ‘‘What are the participants describing?’’ and resulted in a descriptive analysis, the second turn of analysis was guided by asking ‘‘What do the findings express and how can they be understood?’’. Similarities and differences between individuals and between different parts of individual interviews were analysed, and a way to explain both the entirety of data and the individual parts through a few themes were sought. Alternative interpretations and explanations were discussed, and any inconsistent findings resulted in new discussions of the construct of themes, according to the method of ‘‘negative case analysis’’ described by Lincoln and Guba (1985 p. 309). The final construct resulted in three main themes.

4.2.2.3.1 Trustworthiness

Trustworthiness was addressed in the same manner as in Study I. In addition, the use of an interviewer not involved in the treatment and of triangulation with the objectively measured changes (described in Study V) strengthened the trustworthiness (Lincoln & Guba 1985).

4.2.3 Study III

In Study III, a new questionnaire, focused on bimanual activities and suitable for the age group 6-18 years was developed and evaluated psychometrically. The structure of the questions is presented in appendix 1, study III.

The item generation was based on study of literature, reasoning from clinical experience and interviews with children/adolescents and parents with any of the three diagnoses unilateral CP, OBPP or ULRD. 373 activities were examined against the following criteria: 1) requiring the use of two hands; 2) being frequently performed by many persons 3) possible for a wide age span to do; 4) not seasonal; 5) not too gender- or culture-specific; and 6) not strongly dependent on other functions, such as balance, gross motor function, or cognitive functions. Activities were reduced after field testing and additional reasoning between authors. A field version comprising 37 activities was tested by 18 families. The families were interviewed regarding the relevance of the activities and the questions. A final version with 29 activities was established and tested for evidence of validity.

4.2.3.1 Participants

Inclusion criteria in the validation study were 1/ diagnosis of unilateral CP, OBPP or ULRD (not using prosthesis) and 2/ age 6-18 years. A convenience sample of 96 families was recruited by local occupational therapists. 10 participants were excluded due to incorrect use of the questionnaire, resulting in 86 participants in the study. The families were recommended that children below the age of 13 should be assisted by an adult. In 32 families, the questionnaire was answered by the child alone, three of them were aged 11-12, the rest above the age of 13.

4.2.3.2 Data collection

The ratings in the questionnaire were done in three separate scales, using a 10-category rating scale with adjectives only in the ends. The three main scales in CHEQ describe to what degree the grasp is efficient (*Grasp efficacy*), whether the activity takes more time to perform compared to peers (*Time taken*), and whether the person feels bothered by the reduced hand function when performing the activity (*Feeling bothered*). In addition, for each activity it is reported whether the activity is performed independently, whether the affected hand is used and, if so, whether it is used for grasping the object or as a support without grasp.

4.2.3.3 Data analysis

The American Psychological Association (1999) recommend that evidence for the validity of an instrument should be assessed from various aspects. In this thesis, evidence for validity was evaluated from the aspects of 1/ test content 2/ internal structure, using Rasch analysis. All analysis was done on each scale separately. Analysis was conducted using Winsteps 3.65.0 Software (Linacre 2008).

Evidence for validity based on **test content** was evaluated by assessing how well the items corresponded to the concept "use of the affected hand in bimanual activities". Each item was analysed as regards to what degree it was performed independently and performed by the use of two hands.

Evidence for validity based on **internal structure** was evaluated in six different types of analysis: 1/ The rating scale structure was examined according to the recommendations by Linacre (2004) analyzing whether the categories were used consistently. Effectiveness was optimised according to the essential guidelines described by Linacre (2004): the number of observations in each category exceeding 10; the measure advancing monotonically by category; and an outfit mean-square of less than two. The impact on the validity and reliability of various alternatives of collapsing rating scale categories was also examined according to the recommendations of Bond & Fox (2007). 2/ Internal scale validity was examined by analysing goodness of fit; the degree of fit between the real rating and the expected rating for an item of a certain difficulty by a person with a certain ability. Criteria for misfit were set to infit MnSq > 1.4 in combination with a Z-value of > 2.0, which identifies items that show underfit to the expected hierarchy of item difficulty (Wright 1994). The data was considered to fit the model when 95% of the items show acceptable fit (Smith1991). 3/ Unidimensionality was analysed by examining values of principal component analysis (PCA) according to the "tentative guideline" by Lincare (2008); unidimensionality is supported if the measures explain >50% of the variance and the first contrast no more than 5%. 4/ Person separation was assessed by reliability coefficient and the number of person strata that persons could be separated into. The number of strata was calculated by the formula by Fisher (1992) [Strata = (4G+1)/3] where G is the separation ratio.

According to Fisher (1992), the number of strata must be at least two in order to separate individuals meaningfully. 5/ The precision of the sample mean was evaluated by the SE value. An SE below 0.385 yields a 99% confidence that the estimate lies within an interval of 1 logit (Linacre 1994) 6/ Evidence for validity based on response processes was also evaluated by visual inspection of the "Rasch-maps" (fig.1 Study III) – showing the distribution of persons graded by ability, together with the distribution of items, graded by difficulty- as well as by comparing the mean person ability measures to the mean item difficulty measure (Bond & Fox 2007).

4.2.4 Study IV

In study IV, CHEQ is used to study differences between diagnosis groups.

4.2.4.1 Participants

In Study IV, data from the 86 families in Study III was used and completed with data from 14 more families, using the same inclusion criteria and the same way of recruiting. In these 14 families, the questionnaire was answered by the child alone in 1 case where the child was aged 17, in 6 cases where the child was aged 6-10, the parents were respondents, and in the 7 remaining cases the child was 8-9 years of age and the ratings were done by the child and parent together.

4.2.4.2 Data collection

Whereas data for the 86 persons in Study III was collected in a paper-version of CHEQ, the additional 14 persons used a computer-version. Additional differences between the two versions was that in the paper version, a 1-10 rating scale was used, which in the analysis was collapsed into a 1-4 scale. In the computer-version, a 1-4 scale was used. In the computer version, the questions were presented in random order but in the paper version they were presented in a fixed order.

4.2.4.3 Data analysis

In Study IV, differences between diagnoses groups were studied.

In CHEQ, nominal data is generated on how activities are performed. This is presented in frequency for each individual. In order to enable comparisons between groups in Study IV, a percentage-value of the frequency for each person was calculated for three variables, these were named *Doing by oneself, Using the affected hand* and *Holding objects.* In each of these three variables, a higher score indicated more of the examined variable. In the three rating scales; *Grasp efficacy, Time taken* and *Feeling bothered*, ordinal data is transformed into interval data trough Rasch-analysis, generating log odds. In this study, the log odds were transformed into a 0-100 scale, and used for comparisons analysed by One-way analyses of variance (ANOVAs) and Scheffe's post hoc test. Alpha was set to $p<0.05$.

In order to see how diagnosis, gender and age influenced the degree of *Feeling Bothered* by the reduced hand function in the analysis, linear regression analysis was used (Altman, 1991). An unadjusted model was used to compare the three groups of diagnoses in *Feeling Bothered*, and an adjusted model was produced to control for the background variables gender and age.

Correlations between age and *Doing by oneself*, between *Grasp efficacy* and *Feeling bothered*, and between *Time taken* and *Feeling bothered* were calculated with

Pearson's r (Altman 1991) for each group of diagnoses, in order to examine how the variables relate depending on diagnoses.

4.2.5 Study V

Study V describes objectively measured outcomes of UES, 5 years after surgery.

4.2.5.1 Participants

The participants were the same as in Study II.

4.2.5.2 Data collection

Joint motion was tested with a goniometer; grip ability was measured using nine tasks requiring different grips (score 0 to 4 for each task; maximum score, 36); dexterity was measured by the time taken to move 10 cubes (1x1 cm); and strength was measured with the Martin vigorimeter (Gebrüder Martin, Postfach, D7200 Tuttlingen,Germany). For the sake of comparison, strength and dexterity were also measured in the non-affected hand. Preoperatively, the family, together with the surgeon and the therapist, proposed some functional objectives for surgery which were evaluated as fulfilled or not fulfilled by the family postoperatively.

4.2.5.3 Data analysis

Nonparametric statistic was used, due to the small size of the sample, reporting Median, min-max and significant differences at the level of $p<0.05$.

5 FINDINGS

5.1 STUDY I

In study I, it was found that there is not one single strategy used to perform a certain activity, but rather a range of strategies are used, depending on the circumstances in the situation. A main finding was that the situation of selecting strategy was often described as a dilemma. In order to select the strategy there was a need to estimate the feasibility of success, social aspects and personal aspects. It seemed that whatever strategy participants choose, the strategies have some negative consequences that require consideration. Estimating the feasibility f success include considering the match between the own ability and the demands of the task. Estimating social aspects may include finding a performance strategy that looks/appears “normal”; this may either lead to an increased use of the affected hand, in order to appear bimanual, or to avoiding the use if the affected hand, in order to conceal it. Asking for help may be a positive solution to the problems, however, asking for help could also interfere with a wish to be independent, to get the desired result and it could also influence the relationship with the other person in a negative way. Estimating personal aspects includes personal preferences in how activities should be performed.

A second main finding was that even though much effort may be given to find suitable strategy options, there are still consequences which have to be tolerated. Such consequences may be a need for planning, for taking extra time, for paying extra attention, for increasing the workload on the rest of the body and for limiting choice of task or task performance. The need for planning may involve selecting special garments or shoes which are not too difficult to button or tie. Extra time may be needed because of a need for changing hand-roles, when using both hands, and a need to perform the activity in several steps, when using only one hand. Due to the reduced hand function, the performance may be ineffective and sequences of activity performance may need to be repeated. Extra attention may be needed to control the affected hand and to compensate dysfunction of the hand, for example with vision. The rest of the body gets increased work load when compensating the decreased hand function, for example by extra strain on the other hand, on the teeth or when positioning the body in an awkward way in order to get in a position that facilitates the use of the affected hand. It may be necessary to omit activities or to restrict the choice to such options that are possible to perform. Another limitation is when activities can be performed, however not according to the implied rules, thus conflicting with personal values. The need to select suitable strategies for performance made it necessary to have a repertoire of strategies to choose from.

5.2 STUDY II

In Study II, experiences of hand use after Upper Extremity Surgery were described. It was found that changes were mostly described in aspects related to daily life; many daily activities had become easier to perform. The hand was now easier to use and it was therefore also used more. Changes in the appearance were of great importance, especially when reaching the teenage years. Generally, after treatment, the arm was described as having a more “striking” position and a more “natural” appearance. The

position of the arm was now straighter, with the forearm hanging down in a relaxed way. An improved appearance made it easier to show the arm. Also a negative change in appearance was described by some of the participants, they found the scars on the forearm to be disfiguring and sometimes wanted to hide the scars. The participants also described that the internal feeling of the arm was changed. Before treatment the arm was described as tense, stiff and hard, after the treatment the arm was generally described as looser and more relaxed. It was no longer necessary to stretch and to force the arm to straighten out.

5.3 STUDY III

In order to strengthen validity of the CHEQ, the development included interviews with parents and children, both before the constructing of the questionnaire and during the construction of the first, preliminary versions. Previous literature and clinical experience was also used. The final version was tested for evidence of validity as regards test content, internal structure and response processes.

5.3.1 Evidence for validity based on test content

According to the responses to the CHEQ questions, the final 29 activities were generally found to be relevant, commonly performed independently and involving the use of both hands. Each activity was regarded as not applicable by 0-13% of the persons. The percentage of persons who performed the activity independently varied between 43% and 100%, median value 92%. Each activity was performed using both hands by 70.7-97.5% of the respondents. Thus, the selected items in CHEQ were shown to fulfil the initial criteria of being performed independently and by using both hands to a high degree.

5.3.2 Evidence for validity based on internal structure

Analysis of the function of the 10 category rating scales showed an inconsistent order of thresholds in all three scales. This indicates that generally, the respondents did not use the categories of the rating scale in a consistent way, the ratings did not follow the expected hierarchy of the person's ability and the item's difficulty. When the scale was collapsed into four categories according to the guidelines by Linacre (2004), each category exclusively covered a certain proportion of the person's ability and the item's difficulty. The four-category scale, compared to the 10-category scale, also improved reliability and separation values. Further analyses were conducted applying the collapsed rating scale structure.

Rasch Analysis was used for item reduction, separately for each scale, during the development. Out of the final 29 items, 3 items showing misfit in the *Grasp Efficacy* scale and one of these also in the *Time taken* scale, were excluded from analysis (study III, table II). After this, only one item showed misfit in the scale *Grasp efficacy* ("Cut up a pancake") and one in the scale *Time taken* ("Cut meat"). This meets the recommendations by Smith (1991), that 95% of the items in each scale should show acceptable fit.

Analysis of the targeting of the difficulty of the items and the ability of the persons in the three scales showed that there was a fairly good distribution even though the mean ability measure of the persons was higher (3.03, 3.35 and 2.90 respectively) than the mean difficulty of the items (by default set to 0), indicating that the average person's ability was greater than the average difficulty of the items. This was also shown by the number of persons scoring a maximum on the scales: 15 persons on the *Grasp efficacy* scale, 13 persons on the *Time taken* scale and 18 persons on the *Feeling bothered* scale. None of these persons had a diagnosis of unilateral CP. However, considering the spread of the four rating scale categories, the Rasch maps showed a correlation between the distribution of item difficulty and person ability (study III, fig. 1). The person separation demonstrated that people can be separated into four or five groups with a reliability of 0.90 -0.94, varying between scales. The range of standard error was from 0.26 to 0.31, thus below the limit which according to Linacre (1994) yields a 99% confidence that the true estimate lies within an interval of 1 logit. The PCA varied between 57.4% and 64.1%, exceeding the limit of 50% and thus supported the assumption that each scale measures only one dimension (Linacre 2008).

5.4 STUDY IV

There was a significant difference between the groups in *Doing by oneself.* Participants with unilateral CP performed significantly fewer activities by themselves than children with OBPP and ULRD. Gender appeared to have no effect on this variable ($p=0.87$). *Doing by oneself* was moderately correlated with age for the whole group ($r=0.51$, $p<0.001$). Participants with unilateral CP had the weakest correlation ($r=0.48$, $p<0.001$) relationship between *Doing by oneself* and age, compared to participants with OBPP ($r=0.69$, $p<0.001$) and ULRD ($r=0.52$, $p<0.002$).

Analysis of the variable *Using the affected hand* showed that participants with ULRD used the affected hand more often (mean 96% of the activities performed independently) compared to the other two groups. The mean of *Holding objects* (if the hand was used for holding rather than as a support) was seen in 57% of the activities performed using two hands, describing the mean for the whole group, there were no statistical differences between the diagnosis groups. Gender did not appear to affect either *Using the affected hand* or *Holding objects*.

Unadjusted linear regression showed an association between the three diagnosis where *Feeling bothered* was on average higher (corresponding to individuals being less bothered) in both the OBPP group ($B=23.5$, $p<0.001$) and in the ULRD group ($B=21.2$, $p<0.001$) compared to the unilateral CP group (Table III, Study IV). The association was only mildly affected when adjusted for age and gender ($B=21.4$ and 21.0 respectively, $p<0.001$).

When Time taken and Grasp efficacy were included in the regression analysis, the difference in Feeling bothered between the diagnosis groups was eliminated; for OBPP the regression coefficients were $B=1.4$ (CI -3.85 – 6.65) and $B=4.5$ (-1.30 – 10.34) respectively, and for ULRD the regression coefficients were $B=0.8$ (-4.25 – 5.94) and $B=3.1$ (-2.65 – 8.74) respectively. This is mainly explained by the high correlation between these two variables and the main outcome variable, Feeling bothered. The correlation coefficient for Feeling bothered and Time taken was $r= 0.90$ ($p<0.001$), while that for Feeling bothered and Grasp efficacy was $r= 0.87$ ($p<0.001$).

Linear regression showed that *Feeling bothered* was explained by *Time taken* to a lower degree (69%) for children with unilateral CP and for children with OBPP (66%) than for children with ULRD (86%). *Feeling bothered* was explained by *Grasp efficacy* to 42% in children with unilateral CP, 69% children with OBPP and to 79% in children with ULRD. The relation between the variables explained 69%, 66% and 86% respectively of the variance in data.

5.5 STUDY V

The objective measurements in Study V show that there had been changes which could be related to the treatment and which to a certain extent were persistent five years later. The improvements in active wrist extension and supination were maintained (Wrist extension 20° preoperatively, 57.5° at 9 months and 60° at 5 years, Supination 35° preoperatively, 60° at 9 months and 60° at 5 years, Median values). The improvement in ability to grip objects also remained about the same (Grip scores 15.5/36 preoperatively, 20/36 at 9 moths and 20/36 at 5 years). Although not significant, the median value for strength and dexterity indicated that the results were maintained or even improved after 5 years (Strength 10 kPa preoperatively, 14 at 9 months and 20 at 5 years, dexterity 35 seconds preoperatively, 24 at 9 months and 20 at 5 years). The improvements in strength and dexterity may be due to normal development, therefore, these measurements were also assessed for the non-hemiplegic hand. The non-hemiplegic hand improved less than the hemiplegic hand, suggesting that the improvement was a result of surgery (table II Study V). The 10 participants altogether achieved the fulfilment of objectives to a degree of 73% at the 9-month follow-up and to 64% at the 5-year follow-up. Between one and six objectives were set up for each subject. The degree of fulfilment of objectives for each subject varied between 0 and 100%.

6 DISCUSSION

The aim of this thesis is to develop knowledge of activity performance, with a special focus on hands use in bimanual activities in children and adolescents with unilateral CP. The results of the constituent studies provide an interesting material to discuss in relation to the overall aim of the thesis. First, the need to pay attention to how the hand is used, described by adolescents with unilateral CP, will be discussed. Then, the experiences of performing bimanual activities will be discussed. The third section will discuss the possibility of improved activity performance by using UES. Finally, the use of the CHEQ questionnaire will be examined.

6.1 HAND USE DEMANDS ATTENTION

In the introduction to this thesis, hand use in well known activities was described to be directed on a subconscious level to a high degree. Normally, the person performing the activity is not focusing on the hand use, but on the result he/she wants to achieve. By neural control, the most suitable strategy in relation to the character of the activity and to the object, is selected and executed. In contrast to this, the findings of this thesis show that for children with unilateral CP, the choice of hand use was an issue which constantly demanded attention in the performance of bimanual activities. It was shown in the introduction that hand use varies with the kind of activity, the properties of the object and the physical environment. This was also seen in study I, but it was notable that the participants were also to a high degree aware of how their performance was depending with these aspects. This indicates that their choice of hand use demanded more attention. Also in contrast to what has earlier been described to influence hand use, it was in study I seen that the social environment also had an important influence. Depending on the social surrounding, the activity would be performed differently, for example using both hands or only one. Also more personal aspects which has so far not been described to influence hand use generally, was seen to influence hand use in study I; the person's own values and interest, the state of mood and how one wanted to present oneself in the eyes of others were personal aspects considered when choosing how to use the hand. Thus, from the aspect of hand use, the dynamical interaction between activity, person and environment seems to be substantially different in the presence of reduced function in one hand.

A minor finding in study I is also relevant to acknowledge; some participants expressed that they needed to concentrate on the performance of the hand, the use of the hand was thus not automatic but demanded awareness. One girl expressed this by saying that she had to "tell" her hand what to do. Although this was only mentioned by a few participants, it is interesting, since it has not been described earlier how the reduced neural control of hand use may be experienced by the person.

One aspect which might have been expected to affect hand use is age. This was however not found in this thesis; the CHEQ-variables *Using affected hand* and *Holding objects* did not correlate with age (unpublished result in Study IV). A longitudinal study has described development using AHA in children aged 1.5 to 8 years with unilateral CP (Holmefur et al. 2010). The study showed that children in MACS group I and II (where most of the children with unilateral CP are represented) reach 90% of their maximum AHA-level at the age of about 4 years, for MACS level III however, this

occurs later, at the age of 9-10 years. Fedrizzi (Fedrizzi, et al., 2003) studied older children and found similar results, although that study has been questioned as regards reliability, as described earlier. Thus, no studies have been found that show improved hand use by natural development in the period of 6-18 years, in children with unilateral CP.

In study II it was however found that changes in hand use were described after treatment. It was experienced to be easier to use the affected hand after UES, it was therefore also used more often. Improved hand use after UES has also been shown using the AHA; the variables "hold", "grip", stabilizes by grip" and "calibrate" improved most (Pontén, et al., 2010). Improved hand use has also been seen after other interventions, for example after Constraint Induced therapy, which is directly targeting the use of the hand (Hoare et al., 2007). The point here is however not to compare various alternatives but rather to describe how hand use may be affected by intervention.

As regards the activity-aspect, it was seen in study I that hand use was problematic almost only in bimanual activities. The only unimanual activity mentioned was shaking hands, since there is a social obligation to use the right hand in that activity. In study III, a variation in hand use was seen also within the group of bimanual activities; the degree of participants (including all three diagnosis) using both hands in each activity varied between 70.7% and 97.5% (study III table II). The influence of activity-aspects on hand use has so far been sparsely described. In order to tailor intervention in a suitable way, further knowledge on the demands of the activity is needed.

In conclusion, it was found in this thesis that from the aspect of hand use, variables in all three aspects - activity, person and environment - contributed to forming the performance in bimanual activities. This emphasizes the need to acknowledge how various aspects interact when describing hand use.

6.2 PROBLEMS IN ACTIVITY PERFORMANCE NEED TO BE ACKNOWLEDGED

This thesis has also acknowledged that there may be a need for children with unilateral CP to make choices about activity performance in a more general sense. Study I demonstrated that the participants repeatedly had to find ways to perform bimanual activities that were difficult to perform. This led to the dilemma of having to choose the best possible way of performing the activity, although negative consequences related to the performance still had to be tolerated. The dynamic nature of the situation, in which the demands of the activity, individual preferences and abilities, the qualities of the object involved in the activity and the expectations of people in the environment, were all considered, in line with how the interplay between activity, person and environment has been described previously (Law et al., 1996; Nelson 1988). However what has not been previously described is how this interplay is viewed by children with unilateral CP. Instead, children with unilateral CP have earlier been described as having a mild dysfunction and as capable of performing most activities in everyday life (Arner, et al., 2008; Hammal et al 2004). The findings of Study I thus seem to contradict this view. There may be several explanations for this apparent contradiction.

One way to understand this seemingly contradiction is that the view changes depending on the perspective chosen. When comparisons are made with children with more severe forms of CP, the group with unilateral CP may appear relatively well-functioning (van Eck et al., 2010; Imms et al., 2009; Majnemer et al., 2010; Orlin et al., 2010). When comparisons are made with children without dysfunction, problems related to having a mild dysfunction may appear (Yude et al., 1998). Considering that children with unilateral CP often attend mainstream schools and socialize with peers with no dysfunction, it is important to consider the latter perspective as well. An alternative might be to make comparisons with other groups with hand dysfunction. The diagnosis of OBPP and ULRD comprise dysfunction of one hand/arm, and children with these diagnoses usually attend mainstream schools, like many children with unilateral CP. In study IV it was seen that in almost all CHEQ variables, namely, in the *Grasp Efficacy*, *Time taken* and *Feeling bothered* scales as well as in the *Doing by oneself* variables, children with unilateral CP presented more problems than did children with OBPP or ULRD. Earlier studies comparing these groups have not been found, but Krumlinde-Sundholm and Eliasson (2003), who described the development of AHA, found that children with OBPP made more efficient use of the affected hand than did children with unilateral CP. Thus, comparisons with various groups sharing some commonalities with the group of unilateral CP may yield an additional view of the complexity of having a mild dysfunction. When making group comparisons, however, it must also be acknowledged that in the group with unilateral CP there is also great variation, so it cannot be assumed that the same differences are found on the individual level.

Another way to understand the seemingly contradiction is from the reasoning concerning occupational performance presented in the Introduction to this thesis: if occupational performance consists not only of practical “doing”, but also represents a way of expressing personal and cultural values and meanings (Christiansen et al., 2005), then it is important not only to achieve a result but also to be content with the performance and with how we can show others who we are through it. Goffman (1990) described the concept of “stigma management”, acknowledging that, when a dysfunction can be concealed, considerable effort may be expended to do so, so as not to stand out as different. According to Goffman (1990), this can also lead to excluding oneself from activities. This was also one strategy of activity performance identified in study I. It is important to identify not only what activities are found difficult but also whether activities have been excluded. A third way to understand the seemingly contradiction is to realize that the instruments used need to focus on the specific problem area for children with unilateral CP: the performance of bimanual activities. The development of CHEQ, described in study III, has made it possible to assess subjective views of the performance of bimanual activities.

This study has contributed to the knowledge on children with unilateral CP by showing that they may experience problems in activity performance that are related to the dynamic interplay between activity, person, and environment. Although this can - at a first view - seem to contradict earlier knowledge that has described this group as highly capable and satisfied in terms of health-related quality of life, it may rather be a complement. This new finding may relate to the fact that the present research considered and described the subject’s own experience as well as using an instrument focusing on the subjective view of bimanual activities and also comparing groups in which unilateral hand dysfunction is the common factor, rather than the diagnosis.

6.3 CHANGES IN ACTIVITY PERFORMANCE ARE SEEN AFTER UES

Both studies II and V describe changes five years after UES intervention, representing the objective and the subjective views of the changes. UES intervention as described in this thesis includes two parts: surgery and postoperative care, including the use of orthosis and training. Although it is open to discussion how these aspects relate, they are here viewed as a single unit. Generally, the intervention aims to improve range of movement, facilitate grasp ability, and improve appearance. As this research focused on describing changes occurring after treatment, an important aspect to discuss was whether natural development might have influenced any such changes.

Participants in study II described an interaction between treatment and natural development, i.e., it was easier to learn new things since the hand could be used in a better way after UES. An increase with age in capability and in the performance of daily activities for children with unilateral CP (aged 3-15 years) was shown by Öhrvall (2010). This was also supported by a finding of study IV; the number of CHEQ activities that were performed independently showed a fair correlation ($r = 0.48$) with age for children with unilateral CP, although lower than for children with ULRD or OBPP. Thus, improvements in activity performance may also partially be related to natural development.

The objective measurements made in study V show changes 9 months after intervention which were remaining five years later. Improvements were seen in range of movement, but no studies describing the normal development of range of motion were found to serve as a basis for comparison. No significant change in dexterity was found, even though the time needed for the task decreased more than in the dominant hand. This is in line with the findings by Eliasson et al. (2006) who found a decrease in time needed to complete a task, based on comparisons of measurements in a group of children at age 6–8 and again at age 19–21 years. The decrease in time was seen both in an experimental grip-lift task and in the Jebsen and Taylor test (Eliasson et al., 2006). Study V also found that strength improved, although not significantly so; however, strength increased in the dominant hand as well, indicating that a general age-related increase in strength may have occurred. There may however be reason to question whether an increase in strength over time would be expected without intervention. Smits-Engelsman et al. (2004) found that children aged 10–15 years had lower finger flexion strength than did children aged 5–9 years. This may indicate that a decrease in force generation could have been expected. The changes observed at the 9-month follow-up are in line with those observed in previous studies of outcome after UES (van Munster et al., 2007), though, to my knowledge, early and late results have not previously been compared.

Study V describes changes after UES from two different views; objective and subjective. It may be interesting to considerations how these two views coincide. It was seen that the particular objective measurements used, which are typical of outcome studies of UES, did not capture aspects viewed as important by the participants. The respondents described changes in everyday life activities, appearance, and the internal sensations of the arm. The objective measurements encompassed range of movement, speed, grip, and strength. Whereas speed and grip were also described in the interviews, many aspects touched on in the interviews were not captured in the measurements; these related more to activity performance in everyday life than to functional components of hand function. This predominance is in line with the findings of Nieuwenhuijsen et al. (2009); in ratings in the Canadian Occupational Performance

Measure for young adults with CP (about half of the participants had unilateral involvement), 98% of the reported problems were linked to activity and participation, and the remaining 2% to the body function component of the ICF classification.

The findings of study II show that changes experienced in everyday life were related both to an increase in the number of activities that could be performed and to the quality of activity performance. For example, decreased effort, better control of the hand, and ability to make more use of the involved hand were qualitative aspects that facilitated activity performance. Reports on qualitative aspects of performance are sparse. Whereas quantitative aspects of activity performance has been described after UES (Eliasson et al., 1998; Malizos et al., 2010; Matsuo et al., 1990; Tonkin et al., 2001), only two studies were found that measured qualitative aspects of performance. Johnstone et al. (2003) and Gong et al. (2010) reported improved ratings on a five-point scale ranging from "Very difficult" to "Completely independent" describing the ease in performing the activity "dressing". Thus, it would be valuable to assess activity performance after UES from the qualitative perspective as well.

McAuliffe (1999) saw a risk in not acknowledging the patient view: "The most worrisome indictment of current research strategy is that we are unable to determine whether the 'objective' results we are accustomed to measuring, such as postoperative strength and ROM, are at all meaningful to our patients''. The AHA makes it possible to assess the efficiency in the use of the hand in bimanual activities in an observation, which is suitable UES treatment in the case of unilateral CP (Pontén, et al., 2010). CHEQ now also makes it possible to describe the subjective view of the use of the hand in every day life. In order to be able to detect evidence for the efficacy of various treatment alternatives, it is however necessary also to use objective measurements. Hence, a combination of the two views would be optimal.

6.4 A NEED TO MEASURE THE SUBJECTIVE VIEW OF PERFORMANCE IN BIMANUAL ACTIVITIES.

This thesis has demonstrated that, for children with unilateral CP, activity performance may be problematic, specifically in bimanual activities. It has also demonstrated that CHEQ can be used to describe the subjective view of activity performance in bimanual activities; the activities included in CHEQ are frequently performed independently and with two hands, the scales are well targeted for children with unilateral CP, and there is evidence of validity of the ratings in each scale (study III). It may then be interesting to discuss how these two aspects relate: In what way does CHEQ capture the problems experienced by children with unilateral CP?

Whereas several other questionnaires ask about difficulty in general, two of the three questions on subjective perceptions in CHEQ are more specific. CHEQ first asks about how the activity is performed. The choice of questions in CHEQ was based on the findings of study I. In study I, avoiding activities and needing help were obviously seen as problematic, but problems were also encountered in activities performed independently. Certain activities take longer for children with unilateral CP than for their peers to perform, when using one or both hands, and grasp quality was central to the problem descriptions. These aspects were therefore the basis for two of the three scales in CHEQ. The third scale, describing whether the subject feels bothered by reduced hand function when executing activities, was intentionally formulated as a broad question. The findings of study I indicate that the underlying reason for feeling

bothered varied extensively. It is important for treatment planning that the underlying reason be identified, and it is not possible to capture this in a rating. Instead, the investigator must make further inquiries to determine the reasons why a subject feels bothered. This approach fits well with the top–down approach suggested for use in occupational therapy (Fisher, 2009). In addition, using CHEQ generates a report on how the activity is performed and how the hand is used; this provides additional information that is useful when determining how to address the problem.

6.4.1 Children as respondents or parents as proxy-raters?

CHEQ recommends that children under the age of 13 years should be assisted by an adult when rating; it is also possible to use the parent directly as a proxy-rater. It is thus germane to ask whether the use of a proxy-rater is suitable, or whether the child should be given the opportunity to answer for him/herself?

The issue of who should be the respondent when administering questionnaires to children has been discussed extensively in the literature, and most studies have focused on whether parents can be assumed to give the same ratings as their children would. Such studies unambiguously demonstrate that there may be a difference, though they vary in reporting how and to what degree ratings will differ. The difference mostly lies in the direction that parents rate more activities as problematic than their children do (Gates et al., 2010; Missiuna et al., 2006; Ylimäinen et al., 2010), though sometimes children rate more problems than do their parents (Dunn et al., 2009; Majnemer et al., 2008 b; Sheffler et al., 2009). Sheffler et al. (2009), however, mention that the differences found in their study were small, and perhaps not clinically significant. In a 2008 review, Upton et al. (2008) found that 90% of the reviewed studies (n = 19) found differences between children's and their parents' ratings, though only small ones; only two of the included studies identified significant differences, and these were in opposite directions.

Given that children's ratings and proxy ratings may differ, even if parents try to assume the child's perspective (Davis et al., 2007), the choice of respondent is more a question of which perspective is desired than of which respondent will give the most appropriate answers. Children's right to be heard has been advocated in the last decade; as the ones directly perceiving the problems in the present case, their perspective is unquestionably important to acknowledge, in line with the increased patient-centeredness of current healthcare. Family-centeredness – also a perspective that influences rehabilitation services – acknowledges that the child is part of a family in which the parents take responsibility and have the right to make decisions for the child. Thus, both perspectives may be important. Upton et al. (2008) emphasize that it is the parent's perception of their child's quality of life that is the principal determinant of health care service use. Sheffler et al. (2009) and Varni et al. (2002) recommend that both perspectives be measured.

To assure validity, measurements used for outcome evaluations should be made by the same person. Clinically, however, a questionnaire can also be used for goal setting: discussion between parents and child, and doing the ratings together, could be beneficial, giving parents new insight into how the child is thinking about the issues. On the other hand, separate ratings made by parents and child could also constitute a valuable basis for treatment planning discussions. That supports the current form of CHEQ, though a version for smaller children may be a useful complement.

6.5 METHODOLOGICAL CONSIDERATIONS

The qualitative studies (I and II) used a comparative analysis approach influenced by grounded theory. One way the approach of these studies differed from that of constant comparative analysis, commonly used in grounded theory (Strauss o Corbin 1998), was that the number of participants was set at the beginning of the study. In constant comparative analysis, data analysis is performed simultaneously with data collection, which ends when saturation is achieved, that is, when new material adds no new information to the analysis. According to Strauss and Corbin (1998), the risk of limited data collection is "that the theory might not be fully developed in terms of density and variation" (p. 292). It might thus be the case that more categories would have been discovered if the number of participants and the amount of data gathered had followed the principle of saturation. The influence of the number of participants is also a consideration with reference to study V. Ten participants is a small number for a quantitative study, and significant differences might have been evident in more variables before and five years after treatment had the sample been larger. However, given the poor knowledge of long-term prognoses after UES, even limited data must be considered valuable. In addition, the quantitative data, in combination with the findings of study IV, add to the value of the overall results.

In study III, Rasch analysis was used to support the item selection and to assess evidence of validity. Several choices made in the Rasch analysis can be discussed, for example, the choice not to exclude participants displaying misfit. In study III, 6–10% of participants displayed misfit to each scale, exceeding the 5% expected by chance (Smith, 1991). Misfit may have arisen because the questionnaire was incorrectly used. However, inspection of the ratings revealed no pattern suggesting that this would be the case. The participants also displayed variation as regards diagnoses, age, gender, respondent (child versus parent), and ability according to the scale in question. Thus, it was reasoned that this was a natural, unexpected variation in the target group and not a reason for excluding the participants from the analysis. This choice influenced the analysis, since it was seen that excluding these people would yield more unidimensionality as well as a higher reliability coefficient and separation value.

In study IV, data were collected using two versions of the CHEQ, a paper version and a computer version. Although it is more convenient and less time consuming to use a computer version, this was not expected to have influenced the results. However, even though the analysis was based on a four-category scale in both cases, the original ratings in the paper versions were derived from a 10-category scale that was collapsed in the analysis. According to Bond and Fox (2007), analysis of a collapsed scale will mostly improve the representation and interpretation of the measure. What is not known, however, is whether the respondents would have chosen the same categories if the scale were shorter. The 14 respondents who used the computer version, however, did not stand out in the analysis, suggesting that that version of the questionnaire did not significantly direct the responses. A limitation of study IV is that the stability of CHEQ has not been established; when this has been done, the findings of study IV can be strengthened. In addition, it is not known how CHEQ ratings differ between parents and children; information on this matter would support the strategy of using both kinds of respondents in the data collection. A further limitation in study IV might concern the generalisability of the findings. A representative sample is needed in order to make

estimations about the population under study. In study IV however a convenience sample was used. Thus the conclusions in study IV may be sample dependent. However, this is a clinical sample and therefore represent the group the occupational therapists meet in their every day praxis.

6.6 ETHICAL CONSIDERATIONS

The present research generated ethical considerations and dilemmas that needed attention. First, all studies in this thesis were approved by the ethics committee of Karolinska Institutet, Stockholm. All participants received verbal and written information describing the studies and gave informed consent to participate. The information stated that participation was voluntary and that participants could choose to withdraw at any time without further explanation.

Ten of the participants took part in interviews about five years after UES treatment. It may be the case that people who have had the opportunity to undergo treatment feel obliged to describe positive results, and do not feel free to criticize. Study II dealt with this possibility by using an interviewer who was not working at the same hospital where the patients had been treated. However, the setting was still a hospital and the interviews took place on the same occasion as they met with the occupational therapist who had been involved in the treatment, since the objective measurements were to be made by the same person as had made them previously. That negative considerations were raised in the interviews, however, confirms that the participants felt at least somewhat free to criticize.

A dilemma in the data collection was that the participants were 12–24 years old at the time of data collection. This is a time in life when adolescents become independent and their interaction with parents might become problematic; on the other hand, it may also be a time when support is needed in delicate situations. Thus, it was difficult to know whether or not it would be best for the adolescent to be accompanied by a parent. Given the present research interest, it would be useful to have parents as informants as well, as the time of intervention had been approximately five years earlier, when the youngest participants were 7 years old. It is probable then that a parent could add information that would otherwise be lost. When inviting the participants, following the surgeon's list, nothing was known about the families. Therefore the choice was made to let the families decide whether the child would be interviewed alone; when the child was accompanied by a parent, the parent was invited to take part in the interview. This was the case in five interviews in which the children were 12–19 years old. It was sometimes observed that the child seemed uneasy with the parent present, which could have been considered grounds for recommending that the interview be conducted with the child alone. However, having the parent accompany the child during the interview permits sharing of experience and gaining support if the interview gives rise to thoughts and worries. Material from the ten individual interviews was used in two studies, with two different analytical aims. Reusing the data in this way reduced need for data collection, which, from an ethical perspective, also reduced the burden on participants.

The four additional interviews were performed as group interviews. Ethical matters were considered beforehand, for example, whether participants would feel free to express anything that was on their minds or whether they would feel constrained by the fear that other participants would spread personal information. Another consideration was whether all participants would have the opportunity to express themselves, or whether the conversation would be dominated by a few participants. This proved not to be the case during the group interview, in which there was a feeling of joy at sharing experiences and listening to others. For several participants, it was the first time they had met someone else who shared their problems in everyday life, which was a positive experience.

One ethical consideration is to strive to reduce the effort involved in participation in the research. In study III, data was collected using a paper-based version of the CHEQ questionnaire. In study IV, an Internet version of CHEQ was available, which facilitated the completion of the questionnaire.

7 CONCLUSION AND CLINICAL IMPLICATIONS

In conclusion, this thesis has demonstrated that children and adolescents with unilateral CP experience problems in performing bimanual activities; the choice of how to perform the activity and of how to use the hand was often a dilemma where consideration was given to aspects in the activity, in oneself and in the environment. Compared with children with OBPP and ULRD, they experience more problems and perform fewer bimanual activities independently. To find out about such problems, it is necessary to consider what group is used for comparisons, to focus on bimanual activities, to consider how the interplay between activity, person, and environment affects the performance, and to consider both the subjective view of child and family as well as qualitative performance characteristics. Improvements in wrist extension, supination, and grip ability have been found to persist five years after UES treatment. Adolescents treated with UES have experienced improvements in everyday life and appearance. The described experiences were more related to changes in activity performance in everyday life, while objectively measured aspects were not described much. The CHEQ questionnaire has been shown to include activities that are frequently performed independently and with two hands by children aged 6–18 years diagnosed with ULRD, OBPP, or unilateral CP. CHEQ also displays good signs of validity in terms of test content and internal structure.

This thesis acknowledges certain issues that are important in clinical practice. One such issue is how intervention can be adapted to the needs of children with unilateral CP. It was found in study I that these children often wanted to hide their problems, which could lead to avoiding certain activities or performing activities in a way that was not satisfying to the individual. It may therefore be necessary to ask specifically and concretely about performance of bimanual activities, to help the individual describe any experienced problems. CHEQ may be a useful instrument in targeting such problems, constituting a good basis for treatment planning. Study I also demonstrated that the teenage years were difficult for children with unilateral CP, and how support is provided during this period of life merits careful consideration. Some participants found it difficult to find alternative strategies for the performance of certain tasks; one should therefore be aware that concrete guidance may be useful to some individuals. Children with unilateral CP constitute a heterogeneous group, so it is important to be aware that their individual support needs may differ.

Another group of findings that is important for clinical practice concerns UES. The present findings indicate that improvements in hand function achieved nine months after surgery largely persisted after five years. This supports the view of UES as a treatment with long-lasting effects. On the individual level, however, some aspects of performance declined over time, so it may be important to follow up results to detect the need for complementary surgery. An important finding was that some participants found some of the surgical scars to be disfiguring. This is a question of surgical techniques and as such was not the focus of this thesis; however, it was found that the desire to hide the scar and the affected hand might lead to decreased use of the hand, influencing activity performance. This indicates a need to follow up the outcome of UES with regard to scars. Finally, the need to acknowledge both performance in everyday life and the subjective views of the child and family should be considered in relation to UES.

8 FUTURE RESEARCH

This thesis has drawn attention to research areas that merit further study. Objective and subjective changes in relation to UES were described here; however, it was difficult to make comparisons with the normal development of hand function, hand use, and activity performance in unilateral CP, since this has rarely been described for the studied age group. This thesis has also highlighted the need for stronger evidence supporting treatment with UES; earlier reviews have stated that good results have been described, but few reviewed studies are based on reliable methods (Boyd et al., 2001; van Munster, et al., 2007).

The CHEQ questionnaire has displayed good signs of validity, but stability has not yet been described, so test–retest validity needs to be studied. Furthermore, the number of activities performed independently was shown to increase with age (captured by the variable *Doing by oneself*). Knowledge of how this variable changes with age in typically developing children would increase our understanding of how development in unilateral CP differs from typical development, helping us interpret the findings of CHEQ.

9 ACKNOWLEDGEMENTS

I want to express my gratitude to those who made the work with this thesis possible in different ways:

- to the participants, for sharing experiences and for willingness to contribute to the development of CHEQ
- to my supervisors Ann-Christin Eliasson and Staffan Josephsson for sharing extensive knowledge from their specific fields of knowledge. Your inspiration and support has been highly valuable and appreciated!
- to the co-authors; Liselotte Hermansson, Lena Krumlinde-Sundholm, Katarina Lenke-Ekholm and Helen Fitinghoff for contributing with knowledge and for good cooperation.
- to the key persons in the research department; Hans Forssberg for making important research possible at the Neuropediatric unit and for support when needed, None-Mari Kemp for always being ready to give a helping hand and Mikael Dahn for good technical support.
- to PhD-students and researchers at the Neuropediatric unit for interesting discussions and a kind willingness to help in all situations; most of all to Ann-Marie, Marie H., Sermin, Kicki, Linda, Katarina, Marie B., Johan, Lena S., Momin, Kristina, Anna, Åsa H., Åsa B., Eva B., Örjan, Nellie….and many others that I have met during the years and shared so many things with.
- to members of "Kreativa konditoriet" for good discussions and to teachers at the occupational therapy unit for sharing knowledge.
- to all the occupational therapists who helped with the data collection
- to my children, Daniel & Line, the golden sunbeams in my life for putting up with so many "mmmm….jag kommer…..snart…" during the work with "my book", as well as to my companion in everyday life; Stefan.
- to my parents; Inga & Sigge, my brother Niklas, my sister Lena with family and all other close relatives as well as supporting friends and neighbours.

Finally, I want to express my sincere gratitude to the late Dr. Sorna Rajan who first encouraged me to do research in the area and who generously shared experiences from a long and devoted work with children with cerebral palsy in hand rehabilitation.

The work of this thesis was supported by grants from the Norrbacka-Eugeniastiftelsen, Stiftelsen Sunnerdahl Handikappfond, Sällskapet Barnavård, Groschinsky Memorial Fund, Eva and Oscar Ahréns Memorial Fund, The Swedish National Association for Disabled Children and Young People (RBU) and The Centre for Health Care Science at Karolinska Institutet.

10 REFERENCES

Aicardi, J., Bax, M., & Gillberg, C. (2009). *Diseases of the nervous system in childhood*. London: Mac Keith Press.

Altman, D. (1991). *Practical statistics for medical research*. London: Chapman & Hall.

American Educational Research Association, American Psychological Association National Council on Measurement in education. (1999). *Standards for educational and psychological testing*. Washington: American Educational Research Association.

Arner, M., Eliasson, A. C., Nicklasson, S., Sommerstein, K., & Hagglund, G. (2008). Hand Function in Cerebral Palsy. Report of 367 Children in a Population-Based Longitudinal Health Care Program. *Journal of Hand Surgery-American Volume, 33A*(8), 1337-1347.

Arnould, C., Penta, M., Renders, A., & Thonnard, J. L. (2004). ABILHAND-Kids - A measure of manual ability in children with cerebral palsy. *Neurology, 63*(6), 1045-1052.

Arnould, C., Penta, M., & Thonnard, J. L. (2007). Hand impairments and their relationship with manual ability in children with cerebral palsy. *Journal of Rehabilitation Medicine, 39*(9), 708-714.

Asbjornslett, M., & Hemmingsson, H. (2008). Participation at school as experienced by teenagers with physical disabilities. Scandinavian Journal of Occupational Therapy, 15(3), 153-161.

Bax, M., Goldstein, M., Rosenbaum, P., Leviton, A., & Paneth, N. (2005). Proposed definition and classification of cerebral palsy, April 2005 - Introduction. *Developmental Medicine and Child Neurology, 47*(8), 571-576.

Bogdan R.C., Biklen, S.K. *Qualitative Research for Education. An Introduction to Theories and Methods*. 4th ed. Boston: Allyn & Bacon.

Bond, T. G., & Fox, C. M. (2007). *Applying the Rasch model : fundamental measurement in the human sciences*. Mahwah, N.J.: L. Erlbaum.

Boyd, R. N., Morris, M. E., & Graham, H. K. (2001). Management of upper limb dysfunction in children with cerebral palsy: a systematic review. *European Journal of Neurology, 8*, 150-166.

Brown, J., & EG, W. (2000). Neurology of the upper limb. In B. Neville & R. Goodman (Eds.), *Congenital hemiplegia*. London: Mac Keith Press.

Brown, N., Lumley, J., Tickle, C., & Keene, J. (1996). *Congenital limb reduction defects. Clues from developmental biology, teratology and epidemiology*. London: The Stationery Office.

CarlssonMG. (2005) Cerebral Palsy. In Green´s Operative Hand Surgery. 5th ed. Philadelphia: Elsevier Churchill Livingstone.

Case-Smith, J. (2005). *Occupational therapy for children*. St. Louis: Elsevier Mosby.

Charles, J. R., Wolf, S. L., Schneider, J. A., & Gordon, A. M. (2006). Efficacy of a child-friendly form of constraint-induced movement therapy in hemiplegic cerebral palsy: a randomized control trial. *Developmental Medicine and Child Neurology, 48*(8), 635-642.

Christiansen, C., & Baum, C. (2005). The complexity of human occupation. In Christiansen, C., Baum, C. M., & Bass-Haugen, J. (2005). *Occupational therapy : performance, participation, and well-being*. Thorofare, NJ: Slack.

Christiansen, C. H. (1999). The 1999 Eleanor Clarke Slagle Lecture. Defining lives: occupation as identity: an essay on competence, coherence, and the creation of meaning. *American Journal of Occupational Therapy, 53*(6), 547-558.

Claeys,V., Deonna, Th., Chrzanowski, R (1983). Congenital hemiparesis: the spectrum of lesions. *Helvetica Paediatrica Acta.* 38, 439-55

Crajé, C., Aarts, P., Nijhuis-van der Sanden, M., & Steenbergen, B. (2010). Action planning in typically and atypically developing children (unilateral cerebral palsy). *Research in Developmental Disabilities, 31*(5), 1039-1046.

Davis, E., Nicolas, C., Waters, E., Cook, K., Gibbs, L., Gosch, A., et al. (2007). Parent-proxy and child self-reported health-related quality of life: using qualitative methods to explain the discordance. *Quality of Life Research, 16*(5), 863-871.

Dunn, N., Shields, N., Taylor, N. F., & Dodd, K. J. (2009). Comparing the self concept of children with cerebral palsy to the perceptions of their parents. *Disability and Rehabilitation, 31*(5), 387-393.

Duque, J., Thonnard, J. L., Vandermeeren, Y., Sebire, G., Cosnard, G., & Olivier, E. (2003). Correlation between impaired dexterity and corticospinal tract dysgenesis in congenital hemiplegia. *Brain, 126*, 732-747.

Eliasson, A.-C. (2006). Normal and impaired development of force control in precision grip. In A. Henderson & C. Pehoski (Eds.), *Hand function in the child* (2nd ed.). St. Louis, Missouri: Mosby Elsevier.

Eliasson, A.-C., & Burtner, P. A. (2008). *Improving hand function in cerebral palsy : theory, evidence and intervention*. London: Mac Keith Press.

Eliasson, A. C., Ekholm, C., & Carlstedt, T. (1998). Hand function in children with cerebral palsy after upper-limb tendon transfer and muscle release. *Developmental Medicine and Child Neurology, 40*(9), 612-621.

Eliasson, A. C., Forssberg, H., Hung, Y. C., & Gordon, A. M. (2006). Development of hand function and precision grip control in individuals with cerebral palsy: A 13-year follow-up study. *Pediatrics, 118*(4), E1226-E1236.

Eliasson, A. C., Forssberg, H., Ikuta, K., Apel, I., Westling, G., & Johansson, R. (1995). Development of human precision grip .5. anticipatory and triggered grip actions during sudden loading. *Experimental Brain Research, 106*(3), 425-433.

Eliasson, A. C., Krumlinde-Sundholm, L., Rosblad, B., Beckung, E., Arner, M., Ohrvall, A. M., et al. (2006). The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Developmental Medicine and Child Neurology, 48*(7), 549-554.

Fedrizzi, E., Pagliano, E., Andreucci, E., & Oleari, G. (2003). Hand function in children with hemiplegic cerebral palsy: prospective follow-up and functional outcome in adolescence. *Developmental Medicine and Child Neurology, 45*(2), 85-91.

Fisher, A.G. (2009). *Occupational Therapy Intervention Process Model: A model for planning and implementing top-down, client-centered, and occupation-based interventions*. Fort Collins, CO; Three Star Press.

Flanagan, J., Merritt, K., & RS, J. (2009). Predictive mechanisms and object representations used in object manipulation. In D. Nowak & J. Hermsdörfer (Eds.), *Sensorimotor control of grasping*. New York: Cambridge university press.

Forssberg, H. (1999). Neural control of human motor development. *Current Opinion in Neurobiology, 9*(6), 676-682.

Forssberg, H., Eliasson, A. C., Kinoshita, H., Johansson, R. S., & Westling, G. (1991). Development of human precision grip-i - basic coordination of force. *Experimental Brain Research, 85*(2), 451-457.

Forssberg, H., Eliasson, A. C., Kinoshita, H., Westling, G., & Johansson, R. S. (1995). Development of human precision grip .4. tactile adaptation of isometric finger forces to the frictional condition. *Experimental Brain Research, 104*(2), 323-330.

Forssberg, H., Eliasson, A. C., Redon-Zouitenn, C., Mercuri, E., & Dubowitz, L. (1999). Impaired grip-lift synergy in children with unilateral brain lesions. *Brain, 122*, 1157-1168.

Forssberg, H., Kinoshita, H., Eliasson, A. C., Johansson, R. S., Westling, G., & Gordon, A. M. (1992). Development of human precision grip .2. anticipatory control of isometric forces targeted for objects weight. *Experimental Brain Research, 90*(2), 393-398.

Gates, P., Otsuka, N., Sanders, J., & McGee-Brown, J. (2010). Functioning and health-related quality of life of adolescents with cerebral palsy: self versus parent perspectives. *Dev Med Child Neurol*.

Gilbert, A. (2001). *Brachial plexus palsy*. London: Martin Dunitz Ltd.

Goffman, E. (1990). *Stigma : notes on the management of spoiled identity*.[New ed.] Harmondsworth: Penguin Books.

Gong, H. S., Chung, C. Y., Park, M. S., Shin, H. I., Chung, M. S., & Baek, G. H. (2010). Functional Outcomes After Upper Extremity Surgery for Cerebral Palsy: Comparison of High and Low Manual Ability Classification System Levels. *Journal of Hand Surgery-American Volume, 35A*(2), 277-283.

Goodman, R., & Graham, P. (1996). Psychiatric problems in children with hemiplegia: cross sectional epidemiological survey. *British Medical Journal, 312*(7038), 1065-1069.

General Assembly., U. N. G. (1989). *Convention on the rights of the child.* Retrieved 100822, from http://www2.ohchr.org/english/law/crc.htm.

Greaves, S., Imms, C., Dodd, K., & Krumlinde-Sundholm, L. (2010). Assessing bimanual performance in young children with hemiplegic cerebral palsy: a systematic review. *Developmental Medicine and Child Neurology, 52*(5), 413-421.

Guiard, Y. (1987). Asymmetric division of labor in human skilled bimanual action - the kinematic chain as a model. *Journal of Motor Behavior, 19*(4), 486-517.

Gustavsson, A. (2000) *Tolkning och tolkningsteori 1: Introduktion* [Intrpretation and interpretation theory 1: and introduction]. Stockholm: Pedagogiska Institutionen, Stockholms universitet.

Haley, S., Coster, W., Ludlow, L., Haltiwanger, J., & Andrellos, P. (1992). *Pediatric Evaluation of Disability Inventory: development, standardization, and administration manual,* . Boston: Trustees of Boston University, Health and Disability Research Institute.

Hammal, D., Jarvis, S. N., & Colver, A. F. (2004). Participation of children with cerebral palsy is influenced by where they live. *Developmental Medicine and Child Neurology, 46*(5), 292-298.

Hermansson, L. (2004). *Upper limb reduction deficiencies in Swedish children : classification, prevalence and function with myoelectric prostheses.* [dissertation] Stockholm.

Hoare, B. J., Wallen, M. A., Imms, C., Villanueva, E., Rawicki, H. B., & Carey, L. (2010). Botulinum toxin A as an adjunct to treatment in the management of the upper limb in children with spastic cerebral palsy (UPDATE). *Cochrane Database of Systematic Reviews*.

Hoffer, M. M., Lehman, M., & Mitani, M. (1986). Long-term follow-up on tendon transfers to the extensors of the wrist and fingers in patients with cerebral-palsy. *Journal of Hand Surgery-American Volume, 11A*(6), 836-840.

Holmefur, M. (2009). *The assisting hand assessment : continued development, psychometrics and longitudinal use*. [dissertation] Stockholm.
Holmefur, M., Krumlinde-Sundholm, L., & Eliasson, A. C. (2007). Interrater and intrarater reliability of the assisting hand assessment. *American Journal of Occupational Therapy, 61*(1), 79-84.
Holmefur, M., Krumlinde-Sundholm, L., Bergstrom, J., & Eliasson, A. C. (2010). Longitudinal development of hand function in children with unilateral cerebral palsy. Develomental Medicine and Child Neurology, 52(4), 352-357.
Hung, Y. C., Charles, J., & Gordon, A. M. (2004). Bimanual coordination during a goal-directed task in children with hemiplegic cerebral palsy. [Article]. *Developmental Medicine and Child Neurology, 46*(11), 746-753.
Imms, C., Reilly, S., Carlin, J., & Dodd, K. (2008). Diversity of participation in children with cerebral palsy. *Developmental Medicine and Child Neurology, 50*(5), 363-369.
Imms, C., Reilly, S., Carlin, J., & Dodd, K. J. (2009). Characteristics influencing participation of Australian children with cerebral palsy. *Disability and Rehabilitation, 31*(26), 2204-2215.
International Organization for Standardization (1989). *Prosthetics and Orthotics - Limb Deficiencies - Part 1: Method of describing limb deficiencies present at birth.* (Vol. ISO 8548-1). Genève
Islam, M., Gordon, A., Forssberg, H., Sköld, A., & Eliasson, A.-C. (2010). Grip force coordination during a bimanual task in hemiplegic cerebral palsy.
Jeannerod, M. (1984). The timing of natural prehension movements. *Journal of Motor Behavior, 16*(3), 235-254.
Johnstone, B. R., Richardson, P. W. F., Coombs, C. J., & Duncan, J. A. (2003). Functional and cosmetic outcome of surgery for cerebral palsy in the upper limb. *Hand Clinics, 19*(4), 679-+.
Jonsson, H., & Josephsson, S. (2005). Occupation and Meaning. In C. Christiansen, C. M. Baum & J. Bass-Haugen (Eds.), *Occupational therapy : performance, participation, and well-being*. Thorofare, NJ: Slack.
Kerr, C., Parkes, J., Stevenson, M., Cosgrove, A. P., & McDowell, B. C. (2008). Energy efficiency in gait, activity, participation, and health status in children with cerebral palsy. *Developmental Medicine and Child Neurology, 50*(3), 204-210.
Kielhofner, G. (2008). *Model of human occupation : theory and application.* Baltimore, MD: Lippincott Williams & Wilkins.
Krägeloh-Mann, I., & Cans, C. (2009). Cerebral palsy update. *Brain & Development, 31*(7), 537-544.
Krueger, R.A., & Casey, M.A. (2000) Focus groups. 3rd ed. Thousand Oaks, CA: Sage.
Krumlinde-Sundholm, L., & Eliasson, A. (2003). Development of the Assisting Hand Assessment, a Rasch built measure intended for children with unilateral upper limb impairments. *Scandinavian Journal of Occupational Therapy, 10*, 16-26.
Krumlinde-Sundholm, L., & Eliasson, A. C. (2002). Comparing tests of tactile sensibility: aspects relevant to testing children with spastic hemiplegia. *Developmental Medicine and Child Neurology, 44*(9), 604-612.
Krumlinde-Sundholm, L., Holmefur, M., Kottorp, A., & Eliasson, A. C. (2007). The assisting hand assessment: current evidence of validity, reliability, and responsiveness to change. *Developmental Medicine and Child Neurology, 49*(4), 259-264.
Kuhtz-Buschbeck, J. P., Sundholm, L. K., Eliasson, A. C., & Forssberg, H. (2000). Quantitative assessment of mirror movements in children and adolescents with

hemiplegic cerebral palsy. *Developmental Medicine and Child Neurology, 42*(11), 728-736.

Law M, Cooper, B., Strong., S., Stewart, D., Rigby, P., Letts, L. (1996). The Person-Environment-Occupation-Model: A transactive approach to occupational performance. *Canadian Journal of Occupational Therapy, 63*, 9-23.

Law, M., King, G., King, S., Kertoy, M., Hurley, P., Rosenbaum, P., et al. (2006). Patterns of participation in recreational and leisure activities among children with complex physical disabilities. *Developmental Medicine and Child Neurology, 48*(5), 337-342.

Lawlor, M. C. (2003). The significance of being occupied: The social construction of childhood occupations. *American Journal of Occupational Therapy, 57*(4), 424-434.

Leconte, P., & Fagard, J. (2006). Which factors affect hand selection in children's grasping in hemispace? Combined effects of task demand and motor dominance. *Brain and Cognition, 60*(1), 88-93.

Lepage, C., Noreau, L., Bernard, P. M., & Fougeyrollas, P. (1998). Profile of handicap situations in children with cerebral palsy. *Scandinavian Journal of Rehabilitation Medicine, 30*(4), 263-272.

Linacre, J.M. (1994). Sample size and item calibration stability. *Rasch Measurement* Transactions, 7: 328.

Linacre, J.M. (2004) *Optimizing rating scale category Effectiveness*. In: Smith EV, Smith R, editors. Introduction to Rasch Measurement. Chicago: JAM Press.

Linacre, J.M. (2008). A User's Guide to WINSTEPS® MINISTEP Rasch-Model Computer Programs.

Lincoln, Y.S. & Guba, E.G. (1985). Naturalistic inquiry. California: Sage Publications.

Majnemer, A., Bourbonnais, D., & Frak, V. (2008 a). The role of sensation for hand function in children with cerebral palsy. In A.-C. Eliasson & P. Burtner (Eds.), *Improving hand function in children with cerebral palsy: theory, evidence and intervention*. London: Mac Keith Press.

Majnemer, A., Shevell, M., Law, M., Poulin, C., & Rosenbaum, P. (2008 b). Reliability in the ratings of quality of life between parents and their children of school age with cerebral palsy. *Quality of Life Research, 17*(9), 1163-1171.

Majnemer, A., Shikako-Thomas, K., Chokron, N., Law, M., Shevell, M., Chilingaryan, G., et al. (2010). Leisure activity preferences for 6-to 12-year-old children with cerebral palsy. *Developmental Medicine and Child Neurology, 52*(2), 167-173.

Malizos, K. N., Liantsis, A. K., Varitimidis, S. E., Dailiana, Z. H., & Rigopoulos, N. S. (2010). Functional gains after surgical procedures in spastic upper extremity: a comparative study between children and adults. *J Pediatr Orthop B, 19*(5), 446-453.

Mandich, A., & Rodger, S. (2006). Doing, being and becoming: their importance for children. In S. Rodger & J. Zivani (Eds.), *Occupational therapy with children*. Victoria, Australia: Blackwell publishing.

Matsuo, T., Lai, T., & Tayama, N. (1990). Combined flexor and extensor release for activation of voluntary movement of the fingers in patients with cerebral-palsy. *Clinical Orthopaedics and Related Research*(250), 185-193.

McAuliffe, J. A. (1999). Hand care in the new millennium: surgeon's perspective. *Journal of Hand Therapy, 12*(2), 178-181.

Missiuna, C., Pollock, N., Law, M., Walter, S., & Cavey, N. (2006). Examination of the perceived efficacy and goal setting system (PEGS) with children with disabilities, their parents, and teachers. *American Journal of Occupational Therapy, 60*(2), 204-214.

Moore, L. J. S., Allegrante, J. P., Palma, M., Lewin, J., & Carlson, M. G. (2010). Assessment of Quality of Life Needs of Children With Mild Hemiplegic Cerebral Palsy. *Childrens Health Care, 39*(2), 157-171.

Morris, C., Kurinczuk, J. J., Fitzpatrick, R., & Rosenbaum, P. L. (2006). Reliability of the manual ability classification system for children with cerebral palsy. *Developmental Medicine and Child Neurology, 48*(12), 950-953.

Nelson, D. L. (1988). Occupation: form and performance. *American Journal of Occupational Therapy, 42*(10), 633-641.

Nieuwenhuijsen, C., Donkervoort, M., Nieuwstraten, W., Stam, H. J., Roebroeck, M. E., & Transition Res Grp, S. W. N. (2009). Experienced Problems of Young Adults With Cerebral Palsy: Targets for Rehabilitation Care. *Archives of Physical Medicine and Rehabilitation, 90*(11), 1891-1897.

Orlin, M. N., Palisano, R. J., Chiarello, L. A., Kang, L. J., Polansky, M., Almasri, N., et al. (2010). Participation in home, extracurricular, and community activities among children and young people with cerebral palsy. *Developmental Medicine and Child Neurology, 52*(2), 160-166.

Pehoski, C. (2006). Cortical control of Hand-Object interaction. In A. Henderson & C. Pehoski (Eds.), *Hand funciton in the child* (2nd ed.). St. Louis, Missouri: Mosby Elsevier.

Pirpiris, M., Gates, P. E., McCarthy, J. J., D'Astous, J., Tylkowksi, C., Sanders, J. O., et al. (2006). Function and well-being in ambulatory children with cerebral palsy. *Journal of Pediatric Orthopaedics, 26*(1), 119-124.

Pontén, E., Lenke Ekholm, C., & Eliasson, A.-C. (2010). Hand surgery on children with early brain lesions improves bimanual function. Submitted.

Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., & Bax, M. (2007). A report: the definition and classification of cerebral palsy - April 2006. [Proceedings Paper]. *Developmental Medicine and Child Neurology, 49*, 8-14.

Russo, R. N., Goodwin, E. J., Miller, M. D., Haan, E. A., Connell, T. M., & Croty, M. (2008). Self-Esteem, Self-Concept, and Quality of Life in Children with Hemiplegic Cerebral Palsy. *Journal of Pediatrics, 153*(4), 473-477.

Schuengel, C., Voorman, J., Stolk, J., Dallmeijer, A., Vermeer, A., & Becher, J. (2006). Self-worth, perceived competence, and behaviour problems in children with cerebral palsy. *Disability and Rehabilitation, 28*(20), 1251-1258.

Sheffler, L. C., Hanley, C., Bagley, A., Molitor, F., & James, M. A. (2009). Comparison of self-reports and parent proxy-reports of function and quality of life of children with below-the-elbow deficiency. *Journal of Bone and Joint Surgery Am, 91*(12), 2852-2859.

Shepherd, J. (2005) Activities of daily living and adaptations for independent living. In. *Occupational therapy for children*. St. Louis: Elsevier Mosby.

Shikako-Thomas, K., Lach, L., Majnemer, A., Nimigon, J., Cameron, K., Shevell, M. (2009). Quality of life from the perspective of adolescents with cerebral palsy: "I just think I'm a normal kid, I just happen to have a disability". *Quality of Life Research*, 18(7),825-32.

Smith, R. M. (1991). The distributional properties of rasch item fit statistics. *Educational and Psychological Measurement, 51*(3), 541-565.

Smits-Engelsman, B. C. M., Rameckers, E. A. A., & Duysens, J. (2004). Late developmental deficits in force control in children with hemiplegia. *Neuroreport, 15*(12), 1931-1935.

Staudt, K.-M. (2008). Neurological classification and neuroradiology of cerebral palsy. In A.-C. Eliasson & P. A. Burtner (Eds.), *Improving hand function in cerebral palsy : theory, evidence and intervention* (pp. 61-78). London: Improving hand function in cerebral palsy : theory, evidence and intervention.

Steenbergen, B., Verrel, J., & Gordon, A. M. (2007). Motor planning in congenital hemiplegia. *Disability and Rehabilitation, 29*(1), 13-23.
Strauss, A. L., & Corbin, J. M. (1998). *Basics of qualitative research : techniques and procedures for developing grounded theory*. Thousand Oaks, Calif.: SAGE.
Strombeck, C., Remahl, S., Krumlinde-Sundholm, L., & Sejersen, T. (2007). Long-term follow-up of children with obstetric brachial plexus palsy II: neurophysiological aspects. *Dev Med Child Neurol, 49*(3), 204-209.
Surveillance of cerebral palsy in Europe (2000). Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. *Dev Med Child Neurol, 42*(12), 816-824.
Tamm, M., & Skär, L. (2000). How I play: roles and relations in the play situations of children with restricted mobility. *Scandinavian Journal of Occupational Therapy, 7*(4), 174-182.
Taub, E., Ramey, S. L., DeLuca, S., & Echols, K. (2004). Efficacy of constraint-induced movement therapy for children with cerebral palsy with asymmetric motor impairment. [Article]. *Pediatrics, 113*(2), 305-312.
Thibaut, J. P., & Toussaint, L. (2010). Developing motor planning over ages. *Journal of Experimental Child Psychology, 105*(1-2), 116-129.
Tonkin, M. A., Hatrick, N. C., Eckersley, J. R. T., & Couzens, G. (2001). Surgery for cerebral palsy part 3: Classification and operative procedures for thumb deformity. *Journal of Hand Surgery-British and European Volume, 26*(5), 465-470.
Upton, P., Lawford, J., & Eiser, C. (2008). Parent-child agreement across child health-related quality of life instruments: a review of the literature. *Quality of Life Research, 17*(6), 895-913.
Utley, A., & Steenbergen, B. (2006). Discrete bimanual co-ordination in children and young adolescents with hemiparetic cerebral palsy: recent findings, implications and future research directions. *Pediatr Rehabil, 9*(2), 127-136.
Uvebrant, P. (1988). *Hemiplegic cerebral palsy : aetiology and outcome*. Stockholm: Almqvist & Wiksell Periodical Co.
van Eck, M., Dallmeijer, A. J., van Lith, I. S., Voorman, J. M., & Becher, J. G. (2010). Manual ability and its relationship with daily activities in adolescents with cerebral palsy. *Journal of Rehabilitation Medicine, 42*(5), 493-498.
van Munster, J. C., Maathuis, K. G. B., Haga, N., Verheij, N. P., Nicolai, J. P. A., & Hadders-Algra, M. (2007). Does surgical management of the hand in children with spastic unilateral cerebral palsy affect functional outcome? *Developmental Medicine and Child Neurology, 49*(5), 385-389.
Varni, J. W., Seid, M., Knight, T. S., Uzark, K., & Szer, I. S. (2002). The PedsQL 4.0 Generic Core Scales: sensitivity, responsiveness, and impact on clinical decision-making. *Journal of Behavioral Medicine, 25*(2), 175-193.
Wallen, M., Bundy, A., Pont, K., & Ziviani, J. (2009). Psychometric properties of the Pediatric Motor Activity Log used for children with cerebral palsy. *Developmental Medicine and Child Neurology, 51*(3), 200-208.
WHO. (2008). *International classification of functioning, disability and health*. Geneva: World Health Organization.
Wright BD, Linacre JM. (1994). Reasonable mean-square fit values. *Rasch Measurement Transactions* 8: 370.
Wright, F. V., Hubbard, S., Jutai, J., & Naumann, S. (2001). The Prosthetic Upper Extremity Functional Index: development and reliability testing of a new functional status questionnaire for children who use upper extremity prostheses. *Journal of Hand Therapy, 14*(2), 91-104.

Ylimäinen, K., Nachemson, A., Sommerstein, K., Stockselius, A., & Norling Hermansson, L. (2010). Health-related quality of life in Swedish children and adolescents with limb reduction deficiency. *Acta Paediatrica*. 2010 May 3. [Epub ahead of print]

Yude, C., Goodman, R., & McConachie, H. (1998). Peer problems of children with hemiplegia in mainstream primary schools. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*(4), 533-541.

Öhrvall, A.-M., Eliasson, A.-C., Löwing, K., P, Ö., & Krumlinde-Sundholm, L. (2010). Self-care and mobility skills in children with cerebral palsy, related to their manual ability and gross motor function classifications. *Developmental medicine & child neurology, online*.