Hand function and habilitation services among young children with unilateral or bilateral cerebral palsy

A cohort study of performance, development, and current practice

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Summary

Background: The performance of everyday activities is largely dependent on how we use our hands together. A large proportion of children with Cerebral Palsy (CP) experience limited hand function, which may restrict their participation and independence in everyday life situations. Whereas hand function in unilateral CP is thoroughly described, knowledge of their long-term development is limited. For children with bilateral CP, knowledge of both hand function and development is scarce. Although contemporary motor-learning-based interventions have been shown to improve hand function in children with unilateral CP, descriptions of such interventions for children with bilateral CP are almost absent and the effects are largely unknown. Characteristics of usual services that target hand function in children with CP have been scarcely described.

Aims: The overall aims of this thesis were to describe aspects of hand function and development of bimanual performance in young children with clinical signs of unilateral or bilateral CP, and to explore characteristics of current practice to enhance hand function.

Methods: The thesis includes three papers with population-based data gathered from the same cohort of young children who were newly recruited to the national CP follow-up program (CPOP) or the CP register of Norway (CPRN). Project co-workers in 17 out of the 21 regional pediatric rehabilitation units nationwide recruited the children and carried out the data collection. Data were retrieved from the CPOP, the CPRN, and through the research register entitled “The Habilitation Trajectories, Interventions, and Services for Preschool Children with CP” (CPHAB) - from which this thesis evolved. The total number of eligible children for the CPHAB was 202 children, with a participation rate of 53% for paper I, 63% for paper II, and 62% for paper III. Cross-sectional baseline data were used in papers I and II, whereas a longitudinal design was applied in paper III.

In all three papers, the children’s manual abilities were classified by the MACS or the Mini-MACS. In papers II and III, the bimanual performance was assessed by use of the Assisting Hand Assessment (AHA) for children with unilateral CP, and the Both Hands Assessment (BoHA) for children with bilateral CP. In paper I, descriptions of current practice were based on parent-reported data from the Habilitation Services Questionnaire (HabServ) and data reported by Occupational Therapists (OTs) in the CPOP. The mean age of the children was

approximately 30 months in all the papers.

Results: The distribution of Mini-MACS/MACS levels as found in paper II was level I = 23%, level II = 49%, level III = 16%, level IV = 6%, level V = 6%. Children with unilateral CP were classified at Mini-MACS/MACS levels I-III, and children with bilateral CP were classified at levels I-V. The results from papers II and III showed large variations in bimanual performance for both subgroups. The Mini-MACS/MACS levels and the AHA performance at 18 months of age could crudely predict various developmental trajectories on the AHA for the children with unilateral CP. For children with bilateral CP, the developmental change was very limited, although the Mini-MACS/MACS levels could discriminate between various levels of performance limits on the BoHA. In paper I, the parents reported that the vast majority of the children (80%) performed hand training, with large amounts of training, and training largely integrated into everyday activities. The OTs reported that a somewhat smaller proportion (62%) performed hand training. The only significant factor to predict which children performed hand training was the Mini-MACS/MACS levels ($p < 0.01$), showing most training for levels II and III. Although the parents reported large benefits of the training for >70% of the children, the only significant factor to predict large child benefits was the organizational form, with larger benefits for children who practiced in more than one organizational form ($p < 0.01$).

Conclusion: Despite knowledge of evidence-based interventions for children with unilateral CP, the children with bilateral CP were equally likely to perform hand training. Both children with unilateral and bilateral CP performed within a wide measure range on the AHA or the BoHA and displayed various developmental trajectories. The small developmental changes found among the children with bilateral CP need to be further explored, yet urges us to consider carefully the services provided to this subgroup. There is a need to develop and explore hand-function interventions for children with bilateral CP and to evaluate the effects of interventions on the long-term development of children with CP of all subgroups. The fact that parents utilized the children's everyday activities as opportunities for training and perceived larger child benefits from varied practice should be accounted for by carefully considering both the training intensity and the service organization in our current practice.

List of scientific papers

- Paper I: **Klevberg, G.L., Østensjø, S., Elkjær, S., Kjekken, I., and Jahnsen, R.B. (2017).**
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- Paper II: **Klevberg, G.L., Østensjø, S. Krumlinde-Sundholm, L., Elkjær, S., and Jahnsen, R.B. (2017).** *“Hand Function in a Population Based Sample of Young Children with Unilateral or Bilateral Cerebral Palsy”*. Physical and Occupational Therapy in Pediatrics (online). doi:10.1080/01942638.2017.1280873
- Paper III: **Klevberg, G.L., Elvrum, A-K., Zucknick, M., Elkjær, S., Østensjø, S., Krumlinde-Sundholm, I., Kjekken, I., and Jahnsen, R.B.** *“Development of bimanual performance in a population of young children with unilateral or bilateral cerebral palsy”*. Submitted: Developmental Medicine and Child Neurology.

Terms and abbreviations

ADL	Activities of Daily Living
Age-90	Observed and estimated ages at when participating children reach 90% of their limit on the AHA or BoHA.
AHA	Assisting Hand Assessment. A standardized assessment to measure and describe how children with unilateral CP use their affected hand during imanual activities.
AHA-18	Observed and estimated AHA outcome for participating children at 18 months of age.
ANOVA	Analysis of variance
BIMT	Bimanual Intensive Training
BoHA	Both Hands Assessment. A standardized assessment to measure and describe how children with bilateral CP use their hands together and separately during bimanual activities.
BoNT-A	Botulinum Neurotoxin A
CIMT	Constraint-Induced Movement Therapy
CNS	Central Nervous System
CP	Cerebral Palsy
CPHAB	The Habilitation Trajectories, Interventions and Services for Preschool children with Cerebral Palsy
CPOP	Cerebral Palsy Follow-up Program
CPOP/UE	CPOP protocol for upper extremities
CPRN	Cerebral Palsy Register of Norway
CT	Computer Tomography
FCP	Family-centered Practices
GMFCS	Gross Motor Function Classification System
HabServ	Habilitation Services Questionnaire
MACS	Manual Ability Classification System. Manual ability is classified at five functional levels, where level I describes the highest ability and level V describes the lowest ability. Used for children aged four years or older.

Mini-MACS	Mini Manual Ability Classification System. An adapted version of the MACS, used for children younger than four years of age.
MRI	Magnetic Resonance Imaging
OT	Occupational Therapist or Occupational Therapy
PT	Physical Therapist
SCPE	The Surveillance of Cerebral Palsy in Europe
SDC	Smallest Detectable Change

1 INTRODUCTION

My first encounter with Cerebral Palsy (CP) was when I met “Lisa” during my high-school years. With her nearsighted vision, impaired postural control, limited mobility, and limited control of arms and hands, we went to pick strawberries from my parents’ garden. In retrospect, I realize that this day was my “introductory course” to CP, to activity analysis, to the adaptation of activity and environment, to the complexity of hand function, and to the importance of hand function in performing everyday activities. Through participation in an enjoyable activity in the short Norwegian summer, I learned about “Lisa’s” struggle to keep her body stable, locate and *see* the strawberries, *grasp* them without squeezing them, *hold* and *manipulate* them between her hands, *transport* them to her mouth, and *release* them with precise timing. I also learned that adaptations of the activity provided her the opportunity to repeat the task successfully and gradually perform it more effectively, as I helped her to identify the berries and prepared so she could grasp them from an easier position. Despite her struggle, I most of all learned about the excitement she expressed by tasting the sweet and juicy berries picked in nature by her own hands.

Participation in everyday activities is the overall goal of pediatric rehabilitation. The example above demonstrates the important role of hand function for the performance of everyday activities and involvement in life situations. The interacting relationship between impaired body functions or structures, limited activity performance, and restricted participation, is illustrated in the International Classification of Disability and Health (ICF).¹ The role of the environment as a facilitator or a barrier to activity and participation is shown in my example above.

Activities of everyday life represent the core of Occupational Therapy (OT), and OTs have traditionally put much effort into understanding and promoting hand function through engagement in meaningful activity.^{2,3} As an OT in the Neuropediatric department at Oslo University Hospital, I saw many children with CP and limited hand function. I observed them in play situations, assessed them with standardized assessments, listened to their stories, provided specific hand training, and recommended treatment programs. I saw the success when the local therapists and families cooperated with the kindergarten staff to include structured practice in the children's everyday life, and I experienced the successful intensive training programs where children practiced hand skills in groups. I also saw the struggle by families and therapists to implement evidence-based training approaches into the usual services.

Traditionally, the focus of therapeutic management for children with CP has been to promote gross motor functioning and to normalize tone and movement patterns, as this was previously viewed as a prerequisite for hand function in the neurodevelopmental therapies.⁴ This focus has shifted due to new insights from contemporary motor learning theory and knowledge of brain plasticity, and throughout the last 15 years the knowledge of hand function in children with unilateral CP has vastly increased.⁵ Consequently, various intervention approaches have been developed and evaluated, and we now have evidence-based upper limb interventions for this subgroup.⁶ For children with bilateral CP, however, there is much less knowledge of hand function, limited descriptions of hand function interventions, and restricted evidence for the effect of hand training.⁷

In my clinical work, I saw the importance of knowing the children's resources and limitations before planning and recommending interventions. I, therefore, highly appreciate

standardized assessments of hand function, such as the Assisting Hand Assessment (AHA) for children with unilateral CP. I also truly welcome the Both Hands Assessment (BoHA) as a new tool for children with bilateral CP.⁷ The parents of children with CP often seek information on what the future will hold for their children,⁸ and when discussing such questions with parents, I realized that I did not have enough knowledge and found limited evidence in the research literature.

The awareness of lacking knowledge and gaps in the literature led me to this doctoral work. Thus, by utilizing the network of the national CP registers in this multicenter study, I have sought to increase the knowledge of hand function in children with unilateral or bilateral CP and to increase knowledge of the therapeutic management for these children.

2 BACKGROUND

2.1 Hand function and the performance of everyday activities

The ability to manipulate objects skillfully by the hands is characteristic of the human species and results from a refined interaction of visual, perceptual, cognitive, and motor functioning.⁴ The hand is viewed as “*the tool of the mind*”,⁹ and is a means by which we can respond to our environments through nonverbal gesticulation, human contact, and exploration and manipulation of objects.

Almost all activities of everyday life require the manipulation of objects by the hands, and the independent performance of self-care, school, work, and leisure activities is strongly associated with fine motor ability.^{10,11} Early childhood is a period of rapid motor development and of increasing mastery of self-care activities.¹² The independent performance of everyday tasks is intricately related to the development of hand motor skills, as the young child gradually progresses from an initial stage of static holding to the advanced stage of precise object transfer and manipulation by differentiated grasps and grip-lift synergies.¹⁰ Whereas most typically developing children independently finger-feed and drink from a bottle by one year of age, they may not tie their shoe laces until six to seven years of age, or use a knife and fork effectively until approximately eight years of age.¹²

Skillful manipulation of objects during everyday activities depends on the goal-directed reaching, precise grasping and releasing, accurate transferring of objects within or between the hands, and continuous adjustment of grasps and grip force to accommodate to the various natures of the objects.⁴ Although many activities may be accomplished one-handed, the most effective performance of most everyday activities is achieved by the use of both hands together in a bimanual synergy where the two hands play different roles.^{4,9} Children

with CP are less independent in everyday activities compared with their peers,¹³ they commonly experience limited manual abilities, and their typical role of the two hands is often disrupted.¹⁴

2.2 The International Classification of Functioning, Disability and Health

Hand function is a complex phenomenon, and CP is an umbrella term to describe a heterogeneous group of people with various degrees of disability. To understand the dynamic relationship between various factors that may influence functioning and disability, the ICF was developed as a universal framework.¹ The ICF is a biopsychosocial model and comprises three components of functioning: (1) body functions and body structures, (2) activity, and (3) participation, and two contextual components: (4) environmental factors and (5) personal factors. The ICF conceptualizes a person's functioning as a dynamic interaction between her or his health conditions, environmental factors, and personal factors. The ICF is also a relational model, which means that the severity of the disability emerges in the interaction between the functional and contextual components. There is no causal relationship between the components of the model, as they may all influence each other in any direction,¹⁵ as illustrated by the reciprocal arrows in Figure 1.

Figure 1. The International Classification of Functioning, Disability and Health (ICF).¹

CP is a disabling health condition that may cause impaired body functions/structures, such as impaired range of motion and joint contractures. This may lead to limited ability of the child to perform everyday activities, such as dressing oneself or feeding a doll during play activities, which again may restrict the participation of the child in social play. Changing the physical environment by toys that are easier to grasp, or alterations in the social environment by providing personal assistance or changing the rules of the game, may promote the child's activity performance and enhance participation. Furthermore, restricted participation and limited activity performance may cause inactivity of a child and hence lead to impaired body structures/functions, such as decreased range of motion or muscle strength, which again may influence the severity of the disability.

The ICF is accepted worldwide and across several disciplines as an important conceptual model that provides a common language to describe functioning and disability.¹⁶ The pediatric version of the ICF (ICF-CY) takes into account the developmental aspects of a child and the relationship to the context of personal and environmental factors throughout

childhood and adolescence.¹⁷ The ICF has been used as a conceptual frame of reference for the three substudies included in this thesis, and as an overarching model to understand child functioning, development, and service needs and provision in light of the interactions between the five components of the model.

2.3 Cerebral Palsy

CP is a complex and life-long disorder of movement and posture, caused by an early lesion to the developing brain that occurs before, during, or after birth.¹⁸ It is the most common cause of severe childhood motor disability and occurs in 2.2–2.5 per 1000 live births in the Western world,¹⁹⁻²¹ and results in approximately 160 new children with CP in Norway each year.²²

The causes of CP are multifaceted and often combined, and risk factors may include, e.g., intrauterine infections and intrauterine growth restrictions, placental pathology, multiple pregnancies, premature birth or low birth weight, neonatal stroke, or congenital malformations to the central nervous system (CNS).²³⁻²⁸

2.3.1 Definition

CP is a descriptive clinical term rather than an etiological diagnosis or a disease. Since the first known definition of CP in the early 19th century, several attempts to define CP more clearly have been made. Today's most widely accepted definition is the one proposed by a group of experts in the International Workshop on Definition and Classification of Cerebral Palsy in Bethesda, July 2004: *Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to nonprogressive 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 behavior, by epilepsy, and by secondary musculoskeletal problems.*²⁹

This definition explicitly addresses movement deficiencies as the common feature among people with CP, yet also highlights the functional consequences that may lead to activity limitations. Such limitations are also influenced by the additional nonmotor challenges that people with CP often experience,³⁰ hence the definition illustrates the complexity of the disorder and the heterogeneity of the CP population. Although the CP diagnosis is often not established until three to four years of age, signs of abnormal movement patterns that may predict CP can be recognized by specific assessments already during the first months of a child's life.³¹⁻³³ The mean age of the CP diagnosis in Norway is 23 months.²² Early detection is important for the initiation of early interventions that may enhance optimal child development.^{34,35}

2.3.2 Subtypes

The wide range of underlying causes, neurological pathology, and clinical manifestations calls for a system to classify CP into subgroups.²⁹ Such classification is important for precise clinical descriptions and prognostic purposes, as well as for research comparisons and evaluations of change within subgroups. The Surveillance of Cerebral Palsy in Europe (SCPE) has developed a system to classify subtypes according to their most predominant neurological symptoms, which is now internationally accepted.^{19,29,36} Following the SCPE classifications, CP is divided into three subgroups according to their predominant neuromotor disorder: spastic (unilateral or bilateral), dyskinetic, and ataxic.¹⁹ Spasticity is a common neurological phenomenon in central nervous systems lesions, and the group of individuals with spastic CP accounts for 80-90% of the CP population, whereas dyskinetic and ataxic CP are less common and occurs in approximately 7% and 4%, respectively.^{21,37-39}

Whereas the spastic type of CP is subdivided into unilateral CP when only one side of the body is affected, and bilateral CP when the motor impairments are evident on both sides, such subdivision is not established for dyskinetic and ataxic CP.¹⁹ In correspondence with clinical experience, studies have shown that dyskinetic and ataxic CP is most frequently bilaterally distributed.^{40,41} For this thesis, the term bilateral CP will, therefore, be used to include children with spastic, dyskinetic, or ataxic CP.

2.3.3 Functional classification

The diagnosis of a child with CP, or the classification into a neuromotor subtype, does not give information on the child's functional abilities. Functional classification systems for CP have been developed to establish a common language that may enhance the communication between clinicians, researchers, and parents.⁴²

The functional abilities of children with CP are classified according to the Gross Motor Function Classification System (GMFCS) for gross motor abilities,⁴³ the Manual Ability Classification System (MACS) for manual abilities,⁴⁴ the Communication Function Classification System (CFCS) for communicative abilities,⁴⁵ and the Eating and Drinking Ability Classification System (EDACS) for eating and drinking abilities.⁴⁶ Each of these classifications describes the target abilities according to the child's usual performance by use of descriptive "word pictures" on five comprehensive levels, and the categories range from level I (highest ability) to level V (most limited ability).⁴² Although moderate correlations have been found between the different classification systems, the large number of "functional profiles" that may be found among children with CP highlights the heterogeneity of these children.^{46,47}

When the parents of a child with CP want to know "how bad" the situation is for their child, the word pictures from the various classification levels are useful to describe their child's

functional levels with other terms than the previously used “mild, moderate, or severe”.^{29,42}

For research purposes, clustering participants within functional classification levels may provide important insights into whether some specific characteristics are more prominent in some functional groups than in others, or whether some groups may benefit more from an intervention or develop differently than other groups. To investigate child development according to distinct functional levels may also give important predictive information for the planning of interventions, as well as for the communication between therapists and parents regarding realistic goals and expected development.⁴⁸⁻⁵⁰

2.4 Hand function in Cerebral Palsy

Population-based descriptions of hand function among young children with CP are scarce, yet register-based studies of children above four years of age have shown that more than 60% of the children are independently handling objects in age-relevant everyday activities (MACS levels I-II), while approximately 40% need adaptations or assistance (MACS levels III-V).^{38,51-56} The MACS levels are coarse categories of manual abilities; hence, the group classified as “independent” includes children who at the worst may experience some limitations to speed and accuracy (MACS level I) or children who may avoid certain activities or experience limited movement quality or speed that affects performance (MACS level II). Furthermore, being “in need of adaptations or assistance” may include children who are able to perform major parts of activities independently if the situation is well prepared or personal assistance is available (MACS level III). It may also include children who perform parts of selected and adapted activities with effort and limited success (MACS level IV), or children who are totally dependent on assistance, and at the best perform actions such as pushing a switch or passively holding an easy object (MACS level V). A description of the

MACS levels can be found in Section 4.4.1. and the MACS leaflet can be downloaded from www.macs.nu.

Assessment of hand function

The complexity of hand function and the heterogeneity of the performance among children with CP highlight the importance of describing various characteristics of hand function. To enable appropriate intervention planning and evaluation of change and development, it is important to describe aspects of hand function for children within the different neuromotor subtypes or functional levels, as well as the impairments, limitations, and restrictions within the various domains of the ICF.⁵⁷ Several assessment tools are available to describe, measure, and evaluate various aspects of hand function within the body function, activity, or participation components. Furthermore, the assessments may be used to differentiate between the *capacity* (i.e., the child's ability to use the arm or hand and execute a task under optimal circumstances and in a standardized setting) and the *performance* (i.e., the self-initiated use of the arm or hand in a natural environment).^{58,59} Whereas most of the available assessments measure the children's capacity, a combination of assessments describing capacity and performance is recommended, with a major emphasis on the latter qualifier.⁵⁹

For the purpose of this thesis, hand function will generally be described for the children with unilateral and bilateral CP separately, with bilateral CP including spastic, dyskinetic, and ataxic forms.

2.4.1 Hand function in unilateral CP

In register-based publications, the subgroup of children with unilateral CP is reported to account for approximately 30-40% of all children with CP.^{21,38} In the most recent annual

report from the two Norwegian CP registers, however, almost 50% of the children were found within this subtype.²² Children with unilateral CP experience sensory-motor impairments primarily on one side of the body, and generally have one well-functioning hand and one hand affected to various degrees. They most commonly walk without aids and usually present with manual abilities as classified at MACS levels I-III.^{60,61}

Hand function characteristics among children with unilateral CP have been extensively studied and a range of motor impairments have been identified.⁶² Such impairments may include increased muscle tone and reduced range of motion,^{53,55,63} muscle weakness,^{64,65} and decreased anticipatory grip-force control.⁶⁶⁻⁶⁹ Decreased fine motor speed, coordinative ability, and dexterity have also been documented,⁷⁰⁻⁷³ and the additional challenges of impaired vision,^{61,74} impaired tactile perception,^{75,76} reduced motor planning,^{77,78} and cognitive deficits^{79,80} may further complicate the performance of manual tasks.

Despite no direct relationship between motor or sensory impairments and activity limitations,^{4,81} close associations between hand impairments and the performance of fine motor activities have been identified in several studies.^{63,70} Whereas some children with unilateral CP may experience only slight clumsiness in their affected hand during high-precision tasks, others may lack the ability to use an active grasp and use their affected hand only for stabilization, or not at all.⁵ The most commonly used activity-based assessment tool in unilateral CP is the Assisting Hand Assessment (AHA), which is a measure of bimanual performance.⁸² The widespread use of the AHA has documented a wide performance range among convenience samples of children with unilateral CP.⁸²⁻⁸⁴ The complexity of impairments, activity limitations, and participation restrictions associated with poor hand function for children with CP is illustrated in Figure 2.

2.4.2 Hand function in bilateral CP

In register-based publications, the subgroup of children with bilateral CP accounts for approximately 60% of all children with CP,^{21,38} yet in the last annual report from the Norwegian CP registers the proportions of unilateral and bilateral CP are almost equal.²² The manual abilities among children with bilateral CP above four years of age range across all five levels of the MACS.^{53,60,61,85} With Swedish register-based data, more than 30% of the children with bilateral CP were classified as having very limited or no ability to manipulate objects (MACS levels IV-V), whereas approximately 70% of the children did manipulate objects in everyday activities (MACS levels I-III).⁵³ Despite the fact that almost 60% of children with bilateral CP experience difficulties handling objects in everyday activities (MACS levels II-V), knowledge of hand-function characteristics in this subgroup is scarce.⁵³

The characteristics of hand-function impairments among children with bilateral CP differ as a response to their underlying neuromotor pathology. Among the children with spastic CP, the movement pattern is recognized by a velocity-dependent increase in muscle tone following movement activation, together with decreased muscle strength, speed and range of motion, as well as slow and stiff movement patterns and impaired motor control.^{53,86,87} The movement pattern in dyskinetic CP is typically characterized by fluctuating muscle tone and slowness, along with involuntary, stereotyped, or uncontrolled movement patterns that are triggered by activity engagement.^{40,41,87-90} Among children with ataxic CP, poorly coordinated movements and poor precision when approaching a target are typical clinical manifestations of the motor disorder.^{19,87,91}

Hand-function impairments have been scarcely explored among children with bilateral CP,⁵³ yet impaired grip strength, impaired timing and force control, reduced range of motion, and

upper limb deformities have been reported.^{86,92-95} Additional challenges, such as tactile deficits, impaired vision, cognitive or behavioral problems, and epilepsy, are more commonly seen in bilateral CP compared with unilateral CP, and may add to the motor impairments and further complicate their performance of everyday activities.^{21,41,61,96,97}

Although both sides of the body are affected, the pattern of hand use among children with bilateral CP may be very diverse. Whereas some children may have two relatively well-functioning hands with active grasping ability in both, others may have two severely affected hands and no active grasping ability in either of the hands. A third category may present with an asymmetric pattern of hand use, more like what is seen in unilateral CP, with one well-functioning hand and one more affected.^{7,53} The bimanual performance of children with bilateral CP has not been described, as until recently there has been no test available to measure this.⁹⁸

Figure 2. The complexity of hand function in children with CP, illustrated according to the five components of the ICF (1) body functions and body structures, (2) activities, (3) participation, (4) environmental factors, and (5) personal factors. There are overlapping contents of the activity and participation components, as participation is defined as «*involvement in life situations*», and the activities describe the execution of tasks that are part of these life situations.

2.4.3 Development of hand function in CP

Whereas the development of gross motor functioning in children with CP has been extensively studied, and prediction of locomotor development can be made from levels of gross motor functioning at early ages,^{48,99-102} there is a scarcity of studies on the development of hand function.¹⁰³ Register-based data from the CP follow-up program in Norway (CPOP) show that impaired body functions (i.e., range of motion), may increase between 1 and 10 years of age,¹⁰⁴ yet data from the Swedish follow-up program (CPUP) suggest that contractures develop mainly in children with the most limited manual abilities.¹⁰⁵

Development of hand function in unilateral CP

In a study primarily investigating impairment-related changes of hand function during one-year follow-up of 5–15-year-old children, no significant changes in the range of motion, muscle tone, muscle strength, or quality of movement were found.¹⁰⁶ However, the results supported earlier findings that grip strength and speed in children with CP changes along a curve similar to typically developing children.⁹³ In a small study of five children with unilateral and five children with bilateral CP, decreased time during dexterity tasks, along with improved grip-lift synergies, were reported for the affected hand of the children with unilateral CP after a 13-year follow-up.⁹³ One of the first available studies of hand function development among children with unilateral CP used performance ratings on a four-point scale and reported small improvements in grip quality, yet no significant improvements in the spontaneous use of the affected hand between 2.5 and 12.5 years of age.¹⁰⁷

More recent descriptions of bimanual performance development, measured by the AHA, have shown that children with unilateral CP go through a period of rapid development prior to three to four years of age, before their development steadily levels off until adolescence.^{103,108} Children who perform most skillfully at 18 months of age seem to have a more rapid development and reach higher maximum performance compared with children with lower initial performance.¹⁰⁸ Furthermore, various developmental trajectories have been reported for children classified at different manual ability levels in convenience samples of children between 18 months and 12 years of age.¹⁰³

When looking at factors that may influence the development of bimanual performance in children with unilateral CP, the results are somewhat inconclusive. Even though there is evidence to support the long-term effects of early intervention or intensive training,¹⁰⁹ this

effect seems to be overruled by the more prominent effect of brain injury characteristics.^{110,111}

Development of hand function in bilateral CP

The development of hand function among children with bilateral CP is less explored than for unilateral CP, and only three studies were identified through the literature search.

The first study used data from a clinical intervention study where all the children received some type of intervention in a cross-over design.¹¹² The authors reported less change of hand function and quality of movement for children with bilateral CP compared with unilateral CP, and the initial improvements were followed by a decline already from three years of age in some children.¹¹² In the previously reported study on changes in hand function and grip control after a 13-year follow-up, the results were only partly reported for unilateral and bilateral CP separately.⁹³ Nevertheless, the graphs illustrated less improvement of movement speed and grip-lift efficacy among the children with a bilateral distribution, and only some of these children seemed to improve their performance.⁹³ The third study reported improvements in both grip strength and speed of grip-force change in the preferred hand for 28 children with spastic bilateral CP over a 12-month period.¹¹³ No study has described the development of bimanual performance among children with bilateral CP, presumably due to the lack of appropriate assessments for this purpose.⁹⁸

To my knowledge, the role of possible predictors for the development of hand function (e.g., brain injury characteristics and intensified periods of intervention) has not been explored in any published research study on children with bilateral CP. There is generally a lack of research on hand function among children with bilateral CP, and the effect of upper limb interventions for this subgroup has hardly been explored.

2.5 Theoretical perspectives of upper limb management

2.5.1 Motor learning theory and the acquisition of hand skills

Motor learning describes a set of internal processes associated with practice and experience that produces relatively permanent changes in the ability to perform motor activities through a specific skill.¹¹⁴ This may include the process of learning to perform e.g., the activity “getting dressed”, through practicing and learning the skills of tying shoelaces, or closing the zipper of a jacket. Contemporary motor learning theories are strongly influenced by the dynamic systems theory^{115,116} and the ecological approach,¹¹⁷ and emphasize the interactive processes that take place between the *person* who attempts to learn a new motor activity, the *activity* to be learned, and the *environment* in which the activity is performed.¹¹⁸ In the “neuromaturational theories”, which dominated the field of pediatric neurorehabilitation until only a decade ago, the objective was to promote motor learning through normalization of movement patterns or by working to change the underlying neuromotor or sensory impairments.¹¹⁹ A basic assumption was that changes in motor functions (e.g., grip strength and speed) would transfer to enhanced activity performance. Research summaries have shown that the effects of interventions that apply such former theoretical underpinnings are at the best inconclusive.^{120,121} Interventions that adhere to more current motor learning theories are often referred to as “top-down”, “activity-based”, or “functional therapies”, and are shown to be effective in promoting functional changes in hand function among children with CP.^{5,6,121} In simple terms, the contemporary motor learning theories argue “you learn what you practice”.⁴ Rather than “fixing” the underlying impairments, the objective of the activity-based interventions is to optimize the “functional and social independence” in everyday activities.^{120,122,123}

Figure 3. Summary of some key features of contemporary motor-learning-based interventions to enhance hand function and performance in everyday activities, as described in Section 2.5.1.

Different researchers emphasize various features of motor-learning-based interventions.

Some of these are presented in the following, and are illustrated in Figure 3. Polatajko and

colleagues have extensively focused on the application of *cognitive strategies* for problem

solving as essential to the motor learning processes, along with the importance of child-

chosen *activity goals* and *motivation*.^{124,125} The importance of the *natural environment* to

create everyday learning opportunities has been a major focus of work by Dunst and co-

workers.¹²⁶ Through a three-dimensional framework, they distinguish between practices that

are *contextually based* (i.e., taking place as part of the child's daily activity situations) or

noncontextually based (i.e., taking place in therapeutic settings), *child-initiated or adult-*

directed (i.e., type of learning opportunity) and *practitioner or nonpractitioner* implemented

(i.e., agent of provision).¹²⁶⁻¹²⁸

In addition, the importance of *experimentation* and *repeated practice* to create a stable

memory representation of successful strategies when learning a new motor skill has been a

major focus for contemporary hand-function interventions.^{4,129,130} Motor learning occurs as a journey through the cognitive, associative, and autonomous stages of learning,¹³¹ and according to Smidt and Lee the only way to progress from one stage to another is through experimentation and repetition of a task.¹¹⁴ Reaching adequate amounts of repetitions of a task to promote motor skill learning requires sufficient *intensity* or high *dosage* of practice, and this has been proposed as the most important factor to improve hand function in children with CP.^{130,132-134} There is inconclusive evidence of what is sufficient intensity to promote change, however, and the importance of making high-intensity training situations child-friendly and family-centered is a core component of pediatric upper limb management.^{4,130,135}

2.5.2 Family-centered practices

Repeated practice to promote motor skill learning may be facilitated by utilizing the children's natural learning opportunities.¹²⁶ The family context is an important arena for children's learning and exploration of social, emotional and motor skills, and home programs have traditionally been viewed as essential to improve performance within an OT approach to pediatric rehabilitation.^{2,3,136} OT home programs, where the therapists and families work in partnership following specific key criteria, have been shown to be effective in promoting functional upper limb changes in children with CP by enabling increased intensity of practice.^{6,134}

Family-centered practices (FCP) include the use of home programs as a means to making therapy child- and family-friendly, and is an approach to therapeutic management that *"honors and respects their [families] values and choices and which includes the provision of supports necessary to strengthen family functioning"*.¹³⁷ FCP are delivered as *relational*

practices, where the professional behavior is characterized by e.g., empathy, respect, and active listening, whereas the way in which professionals involve parents in decision-making, planning, and carrying-out of interventions are referred to as *participatory practices*.^{120,138,139}

FCP are found to be positively associated with greater family satisfaction, parent self-efficacy, competence, empowerment, and quality of life, and to larger functional improvements in the child's performance.^{138,140,141} The effects of FCP are primarily indirect and mediated by the parents' belief of self-efficacy and empowerment.¹⁴² The role of the family as an important contextual factor is illustrated in the environmental component of the ICF, and the family may be an essential facilitator to promote hand function in the management of young children with CP.¹²⁰

2.6 Evidence-based upper limb interventions

There is extensive evidence for the great plasticity of the young brain following injury, and environmental stimuli may strongly influence both functional and structural changes to the CNS.^{135,143,144} Such stimuli may be the various motor-learning-based interventions provided to facilitate hand motor skills in children with CP.¹⁴⁵ Different approaches are used for upper limb management, with the common overarching goal of promoting participation and independence in everyday activities. Systematic reviews and meta-analyses have identified various levels of evidence for their effectiveness to improve distinct aspects of hand function,^{5,6,121,134,146-149} and selected approaches are briefly presented in the following.

2.6.1 Impairment-directed interventions

Some upper limb interventions primarily aim to reduce impairments of body structures or body functions, such as spasticity, reduced motor control, and reduced range of motion. Impairment-directed upper limb interventions may include splinting and passive stretching,

hand surgery and spasticity-reducing medication (Botulinum neurotoxin A injections [BoNT-A]).¹²¹ There is inconclusive evidence of the effect of splinting and passive stretching to improve joint mobility and hand skills.¹⁵⁰⁻¹⁵³ The effects of hand surgery are also indecisive, yet studies indicate positive structural outcomes of joint alignment, grasping ability, bimanual performance, and satisfaction with hand appearance.¹⁵⁴⁻¹⁵⁹ Although BoNT-A injections are proven to be beneficial in reducing spasticity in target muscles,¹⁶⁰ there is limited evidence of an automatic transfer to improved motor function or activity performance.¹⁶¹ The recommended therapeutic approach is a combination of the various impairment-directed interventions and activity-based hand training, as reduced impairments may provide a window of opportunity to facilitate functional changes during activity performance.^{6,146,161,162}

2.6.2 Activity-based interventions

Constraint-Induced Movement Therapy

Constraint-induced movement therapy (CIMT) is the most widely documented treatment approach to improve hand function in children with unilateral CP.^{5,6,134,146,147,163,164} The key elements of this approach are (1) restricting the dominant hand by some constraint to enforce use of the affected hand and (2) intensive and structured practice of motivating and carefully selected activities.¹⁶⁵⁻¹⁶⁷ The basic assumption of the CIMT approach is to overcome the “learned nonuse” by children with unilateral CP, who may not have sufficient experience of using their affected hand successfully during activity performance.¹⁶⁸ Modified and child-friendly CIMT models have been developed.^{165,166,169} These are established as effective approaches to improve hand function for children with unilateral CP, across MACS levels and age groups.^{5,6,109,134,147,163,164,167,170}

Bimanual Intensive Training

To address the bimanual coordination impairments seen in children with unilateral CP, a highly structured model of bimanual training was developed, the Hand Arm Bimanual Intensive Training (HABIT).¹⁷¹ The HABIT is based on the same motor learning principles and framed in a similar context as the CIMT, yet the key elements are bimanual training and structured intensive practice of bimanual activities.^{133,172} The HABIT, and other variations of bimanual intensive movement therapy (BIMT), is shown to improve hand function in children with unilateral CP.^{121,172,173} When comparing the effects of CIMT and BIMT, the two distinct approaches are both found to be beneficial compared with the usual interventions.^{6,132,134,148,174} Hybrid models of CIMT and BIMT have been explored, following the idea that CIMT may be useful for “turning on” and promoting the unimanual capacity of the affected hand, followed by BIMT, which may facilitate transfer of the unimanual improvements into more skilled bimanual performance.^{5,133,148,175-178}

Functional and goal-directed training

A third activity-based approach to upper limb management in children with CP is functional training, also known as “activity-focused intervention” or “goal-directed therapy”.¹²⁰ This approach incorporates all the features of contemporary motor learning theory reported in Section 2.5.1. as it emphasizes the repetition of functional activities chosen by the child or parents as target goals, organized in a natural learning environment, to enable strategies for the solution of a child’s activity problem rather than promoting “normal” movement patterns.^{120,179,180} Results from studies on functional and goal-directed interventions lend promising evidence to this approach for the attainment of individualized and functional goals.^{121,123,179,181-187}

2.6.3 Context-focused interventions

Context-focused interventions aim to promote children's performance by changing constraints of the tasks or the environment, rather than trying to change the children's movement abilities.¹⁸² By this approach, the children are encouraged to find their own movement strategies as a solution to a motor problem, and practice of target activities or tasks takes place in natural environments by changing the demands of the environment rather than changing the capacity of the child.¹⁸⁸ To my knowledge, context-focused interventions have not been specifically explored with the aim of promoting hand skills. Nevertheless, it is adequate to mention context-focused intervention as an emerging approach to improving participation and independence in everyday activities, which is the ultimate goal also for upper limb interventions. Context-focused interventions target functional goals identified in collaboration between child, parents, and therapist within the scope of everyday activities and participation.¹⁸⁸ Promising results of context-focused interventions have been reported, with improved child performance, attainment of defined goals, and enhanced parent satisfaction.^{182,186,189-193}

2.6.4 Do some children with CP benefit more from upper limb interventions than others?

Large variability with respect to treatment effects of the aforementioned interventions has been reported, despite convincing results on a group level.^{5,121} Several factors have been explored as possible predictors to which children respond best to upper limb therapy, such as the age when performing the training, the ability level at baseline, the basic pattern of brain injury, or the corticospinal projection patterns.^{143,167,194-197} Such studies are few, however, and the findings are inconclusive.

It is likely not possible to find a “one-intervention-fits-all” approach to upper limb management, and combinations of various evidence-based intervention approaches have been explored with promising results.^{175,198-200} Limited knowledge exists, however, as to whether these approaches are included in the usual services to enhance hand function in children with CP, and the effects of the interventions are almost exclusively explored among the subgroup of children with unilateral CP.

2.7 The “Black box” of rehabilitation - what is current practice?

The current practice of upper limb management for children with CP should closely mirror the evidence base of up-to-date research to promote optimal development and participation. Although researchers commonly compare groups receiving target interventions with a control group that receives “usual therapy”, the content and intensity of such usual practice are rarely described in detail.²⁰¹ Whyte and Hart strongly argue for the need to describe the current practice, as they claim that the usual services and interventions may already include the intensity, repetitions, or specific active ingredients that are defined as key elements in an experimental intervention.²⁰² Without describing *what we do* in our current practice, we cannot draw conclusions on *what to change* according to evidence-based knowledge.

Two studies reported that pediatric OTs and PTs were aware of the key principles of general evidence based practice (e.g., intensified and contextual training), yet hardly included intensive models and home programs in their usual service delivery for children with CP.^{203,204} A third study reported that younger children and those with more severe motor limitations generally received larger amounts of therapeutic support.²⁰⁵ For upper limb management specifically, Northern Irish OTs and PTs reported that they did acknowledge

the hand motor limitations of children with CP, yet still provided low levels of evidence-based hand-function interventions.²⁰⁶ Specific descriptions of the usual services that target hand function are lacking in the literature.

The organization and content of the pediatric rehabilitation services vary between countries and regions, and the choice of therapy models is likely to reflect practical issues and differing social and health care systems between countries.⁵ The Norwegian health care is a public service system where all children and families have access to the same service delivery, provided through the municipalities and the specialist health care.²⁰⁷ The usual services for Norwegian children with CP include a standardized monitoring through the CPOP and the Cerebral Palsy Register of Norway (CPRN), yet these surveillance programs do not include standard recommendations for interventions. No international consensus guidelines for upper limb management exist,⁶ although national guidelines for the management of CP and spasticity are established in certain countries.^{208,209} It is unknown to what extent hand function in children with unilateral or bilateral CP is targeted through the usual Norwegian services, and whether evidence-based upper limb interventions and key principles of motor learning are reflected in the current practice.

2.8 Summary of literature and knowledge gaps

Based on the reviewed literature on hand function and interventions in children with CP, several knowledge gaps have been identified. Some of these are listed below.

- Population-based descriptions of hand function in children with CP is generally lacking, and knowledge of the manual ability levels for children below four years of age is scarce.
- Knowledge of hand function in children with bilateral CP is limited, and descriptions of bimanual performance are lacking.
- Knowledge of the development of bimanual performance in children with unilateral CP is limited and lacking for children with bilateral CP.
- Knowledge of interventions to promote hand function in children with bilateral CP is scarce.
- Descriptions of usual services and interventions to target hand function in children with CP is limited.
- Although national guidelines exist for the management of people with CP in certain countries, no international consensus exists for upper limb management.

3 AIMS

The overall aims of this thesis were to describe aspects of hand function and development of bimanual performance in a population of young children with clinical signs of CP and to explore characteristics of the current practice to enhance hand function. The overall aims were explored through three different substudies, reported in three separate papers, with the following specific aims and research questions.

Paper I

The aim was to describe characteristics of current practice to improve hand function in young children with clinical signs of CP and explore factors associated with the children who performed hand training and who had large parent-reported benefits of the training.

Research questions:

- a) What are the characteristics of hand training as reported by parents?
- b) What interventions do the OTs report?
- c) What child and family characteristics are associated with the children who performed hand training?
- d) What factors are associated with large parent-reported child benefits of the training?

Paper II

The aim was to describe aspects of hand function in young children with clinical signs of unilateral or bilateral CP.

Research questions:

- a) How are the manual ability levels distributed among young children with CP of all subtypes?

- b) How do children with unilateral or bilateral CP use their hands during bimanual performance?
- c) How are the manual ability levels of children with unilateral or bilateral CP associated with their assessed performance during bimanual activities?

Paper III

The aim was to describe developmental trajectories of bimanual performance in young children with clinical signs of unilateral or bilateral CP.

Research questions:

- a) How do children with unilateral or bilateral CP change their bimanual performance over time?
- b) Can manual ability levels predict the developmental trajectories of bimanual performance among children with unilateral or bilateral CP?
- c) Can bimanual performance at 18 months of age predict the developmental trajectories of children with unilateral CP?

A summary of the objectives for the three papers is presented in Table 1, along with a brief description of the participants, assessments, and statistical analyses included in the three substudies.

4 METHODS

4.1 Study design, study context, and recruitment procedure

This thesis consists of prospective, longitudinal and population-based data gathered between January 2012 and June 2016, through the larger research project entitled “The Habilitation Trajectories, Interventions, and Services for Preschool Children with Cerebral Palsy” (CPHAB). Clinical child-related data were collected through collaboration with the CPOP and CPRN. Parent-reported data in the CPHAB were gathered by use of questionnaires addressing the children’s health and participation, caregiver priorities, family empowerment and quality of life, and the parents’ perceptions of services and interventions as well as the child benefits of these. For this thesis, only selected data relevant for the description of hand function and hand-function interventions from CPHAB, CPOP, and CPRN were used.

The recruitment and data collection for the CPHAB took place in 17 out of the 21 regional pediatric rehabilitation units across Norway. Project co-workers provided oral and written project information to the parents, collected consent forms prior to the first assessment, and administered the data collection. The children were assessed at six-month intervals, with small deviations to this procedure for practical reasons. Baseline data were used in the cross-sectional papers I and II, whereas paper III had a longitudinal design and included data from all assessments (Table 1).

Paper	Design	Aims	Participants (% of population)	Classifications and Assessments	Statistical analyses
I	Cross-sectional descriptive	To describe characteristics of current practice to improve hand function in young children with clinical signs of unilateral or bilateral CP.	n=102 (53%) ¹	- Mini-MACS/MACS ⁴ - GMFCS E&R ⁵ - HabServ ⁶ - CPOP/UE protocol ⁷	- Descriptive - Fisher's exact test - Chi squared test - Multiple logistic regression
II	Cross-sectional descriptive	To describe aspects of hand function in young children with clinical signs of unilateral or bilateral CP.	n=128 (63%) ²	- Neuroimaging - GMFCS E&R - Mini-MACS/MACS - AHA ⁸ - BoHA ⁹	- Descriptive - Fisher's exact test - Chi squared test - Spearman's <i>rho</i> - Independent samples t-test - One-way ANOVA
III	Prospective longitudinal cohort	To describe developmental trajectories of bimanual performance in young children with clinical signs of unilateral or bilateral CP.	n=102 (62%) ³	- Mini-MACS/MACS - AHA - BoHA	- Descriptive - Intra-class correlation coefficient - Non-linear mixed effects models

¹Eligible participants: n=191. ²Eligible participants: n=202. ³Eligible participants: n=165.

⁴MACS: Manual Ability Classification System. ⁵GMFCS: Gross Motor Function Classification System Extended & Revised. ⁶HabServ: Habilitation Services Questionnaire.

⁷CPPOP/UE: Cerebral Palsy follow-up Program, protocol for the upper extremities. ⁸AHA: Assisting Hand Assessment. ⁹BoHA: Both Hands Assessment.

Table 1. Summary of the study outline for the three papers included in this thesis.

4.2 Inclusion and exclusion criteria

Children with clinical signs of CP, who were recruited to the CPOP or the CPRN from January 2012 through December 2014, were invited to participate in the CPHAB. Eligibility to participate in the CPHAB (hence for all three papers included in this thesis) required that the children were aged four years or younger when they were first registered in the CPOP or the CPRN. For paper I, an additional inclusion criterion was sufficient language skills among the parents to complete the questionnaire in either Norwegian or English. This latter criterion contributed to a smaller number both in the population and in the study sample of paper I compared with paper II. For paper II, all children included in the CPHAB were eligible for participation, whereas only children classified at MACS levels I-III and aged 18 months or older were eligible participants for paper III. Figure 4 gives an overview of the eligible and included children for all three sub-studies.

Figure 4. Participant flow for papers I, II, and III.

4.3 Participants

The participants in the three substudies were all recruited from the same population. All the children included in papers I or III were also included in paper II. Although the numbers of participants in papers I and III are identical, the included children in those substudies were not all the same because of the specific exclusion criteria. Seventy-eight children were included in all three papers.

4.4 Data collection

4.4.1 Functional classifications

Classification of manual abilities: MACS or Mini-MACS

The MACS was used to classify the children's manual abilities on a five-level ordinal scale, where level I indicates the highest ability and level V indicates the most limited ability. The MACS is a well-established system for classification of manual abilities both in clinical

practice and in research and provides a common language to describe the *performance* of how children with CP manipulate objects during everyday activities.⁴⁹ Psychometric testing of the MACS has shown high inter- and intrarater reliability, content validity, and stability over time when used for children above four years of age.^{44,210-212} The reliability of the MACS when used among children below four years of age is questionable.²¹³

During the planning of this Ph.D. project, we wanted to be able to classify the manual abilities of children who were younger than four years of age. The development of the Mini-MACS was in progress at that time, and we got permission to use the field version of the Mini-MACS for our study (E-mail correspondence with Eliasson, A.K., Jan. 29th 2013). During this study, the Mini-MACS was published and released for common use, with reports of high content validity and interrater reliability for parents and therapists.²¹⁴ The stability of the Mini-MACS over time remains unknown.

Table 2 is reprinted with permission from John Wiley and Sons Inc. (License number: 4119280066777) and illustrates the main difference between the Mini-MACS and the MACS.²¹⁴ The MACS was used for children ≥ 4 years, while the Mini-MACS was used for younger children. The MACS and the Mini-MACS were applied in all three papers and were retrieved from the CPOP. For practical reasons, the term MACS will be used throughout this thesis when referring to results where Mini-MACS and MACS levels are pooled together.

Table 2: Comparison of the content of the Manual Ability Classification System (MACS) for children four years and older and the Mini-MACS for children below four years of age. The complete MACS and Mini-MACS leaflets can be downloaded from www.macs.nu.

Table II: Comparison of Manual Ability Classification System (MACS) and Mini-MACS contents	
MACS	Mini-MACS
I. Handles objects easily and successfully. At most, limitations in the ease of performing manual tasks requiring speed and accuracy. However, any limitations in manual abilities do not restrict independence in daily activities.	Handles objects easily and successfully. The child may have a slight limitation in performing actions that require precision and coordination between the hands, but they can still perform them. The child may need somewhat more adult assistance when handling objects compared with other children of the same age.
II. Handles most objects but with somewhat reduced quality and/or speed of achievement. Certain activities may be avoided or be achieved with some difficulty; alternative ways of performance might be used but manual abilities do not usually restrict independence in daily activities.	Handles most objects, but with somewhat reduced quality and/or speed of achievement. Some actions can only be performed and accomplished with some difficulty and after practice. The child may try an alternative approach, such as using only one hand. The child needs adult assistance to handle objects more frequently compared with children of the same age.
III. Handles objects with difficulty; needs help to prepare and/or modify activities. The performance is slow and achieved with limited success regarding quality and quantity. Activities are performed independently if they have been set up or adapted.	Handles objects with difficulty. Performance is slow and with limited variation and quality. Easily managed objects are handled independently for short periods. The child often needs adult help and support to handle objects.
IV. Handles a limited selection of easily managed objects in adapted situations. Performs parts of activities with effort and with limited success. Requires continuous support and assistance and/or adapted equipment, for even partial achievement of the activity.	Handles a limited selection of easily managed objects in simple actions. The actions are performed slowly, with exertion, and/or with random precision. The child needs constant adult help and support to handle objects.
V. Does not handle objects and has severely limited ability to perform even simple actions. Requires total assistance.	Does not handle objects and has severely limited ability to perform even simple actions. At best, the child can push, touch, press, or hold on to a few items while in constant interaction with an adult.

*Table reprinted from Eliasson et al.²¹⁴ with permission from John Wiley and Sons Inc., May 31st 2017. License number: 4119280066777.

Classification of gross motor function: GMFCS E&R

Because the Mini-MACS was not available for common use at the time of this data collection, and the manual abilities were thus not classified in many of the nonparticipating children, the children's levels on the Extended and Revised Gross Motor Function Classification System (GMFCS) was retrieved from the CPOP and used to enable a comparison of functional levels between the participants and nonparticipants in paper II. The GMFCS is the functional classification recommended to supplement the CP diagnosis according to the Surveillance of Cerebral Palsy in Europe (SCPE), and is standard procedure in the Norwegian CP register and follow-up program. Like for the MACS and Mini-MACS, the GMFCS is used to describe performance on a five-level ordinal scale, from level I, which indicates the highest gross motor ability (i.e., walks without limitations) to level V, which

indicates the most limited gross motor ability (i.e., transported in a manual wheelchair).²¹⁵

The GMFCS E&R is a valid and reliable classification system with sound predictive and discriminative properties that is shown to be stable over time.^{42,216}

4.4.2 Interventions and services

Parent-reported: The Habilitation Services Questionnaire

The parents completed the Habilitation Services Questionnaire (HabServ), which requests information about the services and interventions in which the child and family have taken part during the preceding six months. The questionnaire contains four parts: (1) training and stimulation, (2) assistive technology, (3) parent education, and (4) services and disability benefits.

Only data related to hand-function interventions were included in this thesis, and these were (a) the *training target* of hand skills or hand-related Activities of Daily Living (ADL) (dressing, eating, or playing), (b) *professional involvement* (OT, PT, kindergarten teacher, special education teacher, kindergarten assistant), (c) *degree of parental involvement* in training (not involved, observer, performer), (d) *intensity* of training (frequency and duration of the training period), (e) *setting* in which the training took place (e.g., home or kindergarten), (f) *organization* of training (individual, group, or integrated into daily activities), and (g) *goal directedness* of training (yes or no). Moreover, the parents reported their perceived child *benefits* of the training on a five-point Likert scale. The HabServ was used in paper I and the English version of the questionnaire can be seen in Appendix A.

OT-reported: The CPOP upper extremity protocol

Data on current interventions to improve hand function, as reported by the OTs, were retrieved from the CPOP upper extremity protocol (CPOP/UE). These data included

information on hand surgery, upper extremity Botulinum Neurotoxin A injections (BoNT-A), upper extremity orthoses, training of hand skills (CIMT, BMT, or goal-directed training) and ADL, and interventions provided to guide training. These data include the OT-reported training regardless of which professionals were involved, were used in papers I and II, and can be seen in Appendix B.

4.4.3 Assessments

The Assisting Hand Assessment (AHA)

The AHA is a standardized and criterion-referenced test used to measure and describe how children with unilateral CP and other childhood-onset unilateral disabilities use their affected hand and arm during bimanual activity.⁸² The child is seated at a table and is presented with selected standardized toys from a suitcase. Standardized scoring of how the affected hand is used together with the nonaffected hand during spontaneous play is made from video recordings, following criteria given in a detailed test manual. The AHA version 4.4 was used, in which performance is rated on a four-point scale for 22 test items. A total AHA score with the possible range from 22 to 88 is converted to a logit-based 0-100 scale (AHA units) based on Rasch analysis.²¹⁷ The AHA is proven to be a valid assessment tool, with good intra- and interrater reliability and sensitivity to change.^{82-84,218-220} A change of 5 AHA units is considered to be the smallest detectable change (SDC) of clinical value.^{108,217} Certified AHA raters in the regional pediatric rehabilitation units administered the AHA sessions and scored all the AHA videos, with the exception of 19 AHA videos for nine children that were scored by the Ph.D. candidate. The AHA was used in papers II and III.

In paper III, the observed and estimated AHA units at 18 months of age were grouped into three levels; high (63-100 units), moderate (39-62 units), and low (0-38 units). These levels

were based on consensus values as described in Nordstrand et al.,¹⁰³ and discriminate between three levels of performance according to the children's grasping ability. At the "high" level, children mostly use active grasps efficiently, at the "moderate" level, children mostly use active grasps yet sometimes not efficiently, and at the "low" level, children mostly do not use grasps or perform with a passive grasp.¹⁰³

The Both Hands Assessment (BoHA)

The BoHA is a new test, developed to measure and describe how children with bilateral CP spontaneously use their hands together and separately during bimanual activities.⁷ The BoHA is developed from the same concepts as the AHA and uses the same toys, but the test situation is adapted to provoke the spontaneous use of both hands. The BoHA includes 11 unimanual items to be scored for each hand separately and five bimanual items. The range of possible outcomes on the total bimanual score is 27-108, which is converted to a logit-based 0-100 scale (BoHA units) based on Rasch analysis. Psychometric testing has demonstrated good internal scale validity and item and person reliability.⁷ The interrater reliability and test-retest properties have not been investigated, and the SDC of clinical value is thus not known. Nevertheless, the low correlation that was observed between BoHA units and age during the test development may indicate that differences in performance represent distinct ability levels rather than developmental differences.⁷ For this thesis, a change of 5 BoHA units was considered a change of clinical value, based on the SDC for the AHA. Certified AHA raters in the regional pediatric rehabilitation units administered the BoHA sessions, except for sessions for five children that were administered by the Ph.D. candidate. All the BoHA videos were scored by the Ph.D. candidate, and one video for each of the children in the longitudinal study (paper III) was scored by a second rater with the testing sequence randomized by use of the Research Randomizer (www.randomizer.org).

The Intraclass Correlation Coefficient (ICC) for the interrater reliability between the two BoHA raters was 0.95 (CI 0.90-1.0, $p < 0.001$). The BoHA was used in papers II and III.

4.5 Statistical analysis

The statistical methods used in the three different papers were chosen based on the study aims and the distribution of the data. For all three papers, the data were approximately normally distributed; hence, parametric statistics were applied. All the analyses for papers I and II were computed using the statistical software program IBM SPSS Statistics version 22, whereas the statistical software R version 3.2.0 was used for the repeated measures analysis in paper III. The p -value of <0.05 was considered statistically significant for all the included analyses. Descriptive statistics for continuous variables were presented as means, standard deviations (SD), and 95% confidence intervals (CI). Categorical variables were presented as frequencies and proportions (%). A summary of the different statistical methods used in the three papers is shown in Table 1.

4.5.1 Exploring associations

Possible associations between MACS levels and age among the participants in paper I were explored by Fisher's exact test, with age split into four groups and treated as a categorical variable. In paper II, we explored the association between age as a continuous variable and the ordinal MACS levels by Spearman's rank order correlation coefficient (ρ).

In paper I, multiple logistic regression models were computed for two purposes: (1) to explore the role of various child- and family-related characteristics on the likelihood of children participating in training of hand or ADL skills and (2) to explore the impact of selected child and intervention characteristics on the likelihood of parents reporting large or

very large child benefits of the training. The models were based on theory and clinical reasoning, and all predictor variables were entered simultaneously in the models.

4.5.2 Comparison between groups

Two-way between groups analysis of variance (ANOVA) was conducted to explore the effect of age or MACS levels on the AHA or BoHA outcomes in paper II, and Dunnett's T3 and Tukey posthoc tests were used to explore differences in performance between the defined age groups or between the three included MACS levels. Furthermore, we used the independent samples *t*-test in paper II to compare outcomes on the BoHA between children with bilateral CP who had a symmetric or an asymmetric performance pattern on the test.

4.5.3 Exploration of change over time

Nonlinear mixed-effects models were used to explore the changes of AHA or BoHA outcomes over time, with separate models for AHA and BoHA data. An important advantage of the mixed-effects model is its ability to handle differing numbers of observations between the individuals, and it allows for various intervals between the assessments.²²¹ In paper III, mixed-effects models were thus fitted for children with a minimum of two AHA or BoHA assessments, as sensitivity analysis showed that the results were not substantially different when only children with a minimum of three assessments were included. The mixed-effects models were fitted according to the procedure described by Holmefur et al.¹⁰⁸ and Nordstrand et al.,¹⁰³ with the assumption that the performance on the tests would fit a "stable limit" model.⁹⁹ The model estimated the maximum performance (limit) on the AHA or the BoHA to be achieved and retained at older ages, and the rate parameter was estimated to describe how quickly the children increased their performance prior to reaching their limit. Because the rate parameter is difficult to interpret, transformations of

these values were made into a new parameter that describes the age at when the children reached 90% of their maximum performance (age-90) on the AHA or the BoHA. Both children with unilateral and bilateral CP were grouped according to their MACS levels, and children with unilateral CP were additionally grouped by their observed or estimated performance on the AHA at 18 months of age (AHA-18). Cut-off values for the three AHA-18 levels were previously presented in Section 4.4.3.

4.6 Ethical considerations

Parents of all the participating children signed consent forms prior to the first assessment, following written and oral information presented by the regional project co-workers. All the parents were informed of the right to withdraw their consent at any time, with no consequences for their children's future rehabilitation services (Appendix C). Families of children with disabilities experience extensive burdens and pressured life situations,²²² and the relatively large number of eligible children not invited to participate in the study should be viewed with this in mind. All the children eligible for participation were newly recruited to the national CP registers and were all thus recently identified as having CP. Although records were not taken of why children were not invited, several of the recruiters informally expressed that they hesitated to invite some parents in a vulnerable situation due to the additional assessments and questionnaires they would have to complete for the CPHAB study as a whole. Although the number of parents who declined the invitations was small, the decreasing number by each assessment for the longitudinal study may reflect an exhausting effect of the assessments for these families over time.²²³

The study was approved by the Regional Committee for Medical and Health Research Ethics of South-Eastern Norway (reg. nr. 2013/178) and the Data Protection Officer of Oslo

University Hospital (OUS). Authorization to use data from the CPOP and CPRN was granted by the joint publication board. Study permissions are presented in Appendices D-F.

5 RESULTS

A total of 202 children were eligible for participation in this study, but with specific exclusion criteria for papers I and III (see Section 4.2 and Figure 4). The total number of included children was, paper I, $n = 102$ (53% of eligible population); paper II, $n = 128$ (63% of eligible population); paper III, $n = 102$ (62% of eligible population). The mean age was approximately 30 months for all three substudies, and there was an equal proportion of children with clinical signs of unilateral and bilateral CP (Table 3).

The two main topics presented below contain a summary of the key findings from the three papers included in this thesis: Bimanual performance and development (papers II and III), and current practice to improve hand function (paper I).

5.1 Bimanual performance and development

The manual abilities of the 128 children included in this thesis were classified across all five MACS levels (paper II). As shown in Table 3, there was an equal proportion of children classified with unilateral CP ($n = 64$) and bilateral CP ($n = 64$; including spastic, dyskinetic, ataxic, and unclassified subtypes with bilateral clinical manifestations).

Table 3. Distribution of manual ability levels (MACS) according to CP subgroups (data from paper II)

MACS	Spastic unilateral CP, n (%)	Bilateral CP				
		Total, n (%)	Spastic	Dyskinetic	Ataxic	Unclassified ¹
Level I	15 (23%)	15 (23%)	15	0	0	0
Level II	41 (64%)	22 (34%)	16	1	1	4
Level III	8 (13%)	12 (19%)	11	0	1	0
Level IV	0 (0%)	7 (11%)	6	1	0	0
Level V	0 (0%)	8 (13%)	6	2	0	0
n	64	64	54	4	2	4

¹Clinical signs of bilateral CP

5.1.1 Bimanual performance and development in children with unilateral CP

The manual abilities of the children with unilateral CP were classified at MACS level I (23%), level II (64%), and level III (13%) (Table 3).

The cross-sectional description of bimanual performance among children with unilateral CP included baseline data from the longitudinal study, with analysis of AHA sessions for $n = 50$ children (paper II). The AHA units ranged from 12 to 100 (mean: 57.4, SD: 21.5), and the ANOVA revealed a significant effect of MACS levels on the AHA outcomes ($p < 0.001$). The difference was significant between all three MACS levels, and a better outcome was associated with MACS levels of higher ability. The overall effect of age on the AHA outcomes was also significant ($p < 0.001$), yet posthoc analysis showed that the difference was only significant between the two youngest age groups (18-23 or 24-35 months) and the oldest group (48-58 months), with highest outcomes for the oldest children.

As can be seen in Table 4, AHA assessments for 10 additional children were submitted to the register after completion of the analyses for paper II, hence the total number of children included in the longitudinal AHA analyses in paper III was $n = 60$. The longitudinal analysis of AHA data is based on 194 assessments, with a mean number of 3.2 (SD: 0.9, range: 2-6) assessments for each child.

On a group level, children with unilateral CP increased their performance on the AHA over time, with a rapid increase in performance up until approximately four years of age, followed by a slower improvement until the end of our data collection at approximately six years of age for the oldest child (Figure 5). Thirty-three children improved their observed AHA units more than the SDC. When exploring developmental trajectories according to the children's MACS levels or according to their observed or estimated AHA outcomes at 18

months of age, a pattern of more rapid development and higher limit of performance was found for the children classified at the more able MACS levels or with better initial AHA performance. The developmental limit of performance differed significantly between MACS levels I and II ($p < 0.001$), but not between levels II and III (Table 4 and Figure 5). When grouping the children into three AHA-18 groups (high, moderate, low), the difference in the limit of performance was only significant between the high and moderate groups ($p < 0.001$). For the time when the children reached 90% of their estimated maximum performance on the AHA, children at MACS level I reached their age-90 at 30 months of age, which was significantly earlier than the 57 months of age for children classified at MACS level II ($p < 0.001$). The age-90 did not differ significantly between MACS levels II and III.

Children with the highest performance on the AHA at 18 months of age had the most rapid developmental rate, as they reached their age-90 at 23 months of age, compared with 36 months and 112 months for the moderate and low groups, respectively. The difference was significant between all three AHA-18 groups ($p < 0.001$).

Table 4. Various features of the longitudinal data specified for each included manual ability level (MACS). Data from papers II and III.

Assisting Hand Assessment (AHA)						
Longitudinal study, n=60 (paper III)	Data from paper II n=50 ¹	Data from paper III n=60 ¹				
	AHA units at baseline	Age at baseline	Time of monitoring	Change in observed AHA units	Estimated maximum AHA limit	Estimated age at when 90% of limit is reached
	(n) Mean (SD)	Mean (SD) (range)	Mean (SD) (range)	Mean (SD) (range)	Mean (IQR)	Months (IQR)
MACS level I, n=14	(n=13) 81.0 (9.9)	35.0 (11.1) (18-58)	16.5 (7.6) (6-29)	3.4 (5.1) (- 1-18)	85.7 (78.8; 90.9)	29.5 (22.5; 34.1)
MACS level II, n=41	(n=32) 53.2 (14.8)	27.5 (9.3) (18-58)	16.4 (8.8) (5-51)	8.2 (9.9) (- 8-34)	70.2 (65.9; 74.2)	57.4 (27.9; 81.2)
MACS level III, n= 5	(n= 5) 23.0 (15.3)	20.8 (2.6) (18-25)	20.0 (8.2) (7-27)	14.0 (6.9) (+10-26)	69.1 (63.6; 74.6)	154.9 (107.2; 236.6)
Both Hands Assessment (BoHA)						
Longitudinal study, n=42 (paper III)	Data from paper II n=39 ²	Data from paper III n=42 ²				
	BoHA units at baseline	Age at baseline	Time of monitoring	Change in observed BoHA units	Estimated maximum BoHA limit	Estimated age at when 90% of limit is reached
	(n) Mean (SD)	Mean (SD) (range)	Mean (SD) (range)	Mean (SD) (range)	Mean (IQR)	Months (IQR)
MACS level I, n=13	(n=14) 68.6 (10.1)	38.4 (10.1) (20-56)	17.7 (8.7) (7-39)	6.0 (4.6) (- 1-15)	76.4 (72.8; 76.2)	29.5 (28.7; 30.3)
MACS level II, n=17	(n=15) 51.6 (12.6)	32.1 (10.2) (18-48)	18.8 (9.7) (5-36)	4.4 (3.0) (+ 1-12)	58.6 (50.9; 66.8)	29.5 (28.3; 30.3)
MACS level III, n=12	(n=10) 37.1 (9.8)	36.1 (12.8) (20-59)	15.6 (7.6) (6-33)	4.1 (4.8) (- 2-14)	47.3 (42.8; 51.0)	30.7 (29.9; 31.2)

¹ The number of participants with AHAs increased by n=10 between paper II and III, as these assessments were registered after the data analysis for paper II was finished.

² The number of participants with BoHAs increased by n=3 between paper II and III, for the same reason as stated above.

Figure 5. Observed and estimated outcomes on the Assisting Hand Assessment (AHA) for children with unilateral CP, grouped by levels of manual ability (MACS) (a+b) and levels of AHA performance at 18 months of age (c+d).

The mean curves for each group are presented by the colored dotted lines, with Confidence Intervals (CI) for the limits of performance illustrated by the solid grey vertical lines. The dotted vertical lines mark the age when the age-90 is reached for each group, with CI marked by the grey horizontal lines.

5.1.2 Bimanual performance and development in children with bilateral CP

The manual abilities of children with bilateral CP were classified across all five MACS levels (level I: 23%, level II: 34%, level III: 19%, level IV: 11%, level V: 13%), as illustrated in Table 3.

The cross-sectional description of bimanual performance among children with bilateral CP in paper II included baseline data from the longitudinal study, with analysis of BoHA sessions for $n = 39$ children. The BoHA units ranged from 14-88 (mean: 54.0, SD: 16.5), and like the AHA outcomes, the ANOVA showed a significant effect of the MACS levels ($p < 0.001$), with better BoHA outcomes associated with MACS levels of higher ability. The overall effect of

age was also significant for the BoHA outcome ($p < 0.009$), yet the difference was only significant between the two youngest (18-23 or 24-35 months) and the oldest (48-58 months) groups. Moreover, the children with a symmetric performance pattern on the test ($n = 25$) achieved higher BoHA outcomes compared with those with an asymmetric ($n = 14$) hand use ($t = 3.616$, $p = 0.001$).

In addition, for children with bilateral CP, additional assessments were submitted to the register after the data analysis for paper II. The longitudinal analysis of BoHA data is thus based on 135 assessments for $n = 42$ children, with a mean number of 3.2 (SD: 1.0, range: 2-6) assessments for each child (Table 4).

On a group level, the children with bilateral CP slightly increased their performance on the BoHA over time, with the change of performance taking place primarily prior to the age of 30 months (Figure 6). Nineteen children improved their observed performance equal to or above 5 BoHA units during the study period. The estimated developmental limit on the BoHA was significantly different between all the MACS levels ($p < 0.001$), with the highest estimated limit for children classified at MACS level I. However, the rates of development were similar for all MACS levels, and the estimated age-90 was approximately 30 months for children in all three MACS groups.

Figure 6. Observed and estimated outcomes on the Both Hands Assessment (BoHA) for children with bilateral CP, grouped by levels of manual ability (MACS). The mean curves for each MACS level are presented by the colored dotted lines, with Confidence Intervals (CI) for the limits of performance illustrated by the solid grey vertical lines. The dotted vertical lines mark the age when the age-90 is reached for each group, with CI marked by the grey horizontal lines.

5.2 Current practice to improve hand function

Results from the analysis of data from the two separate data sources in paper I are presented below: (1) parent-reported characteristics of current practice from the HabServ Questionnaire and (2) OT-reported characteristics of current practice as reported in the CPOP/UE protocol.

5.2.1 Parent-reported training characteristics

The parents reported that almost 80% of the 102 children had performed training of hand skills and the selected hand-related ADL activities during the preceding six months and that more than 70% of these had defined goals for the training.

Intensity or dose of practice

According to the parents, the training had taken place for the whole six-month period or more in over 80% of the children, with a frequency of more than six times a week in 62% of

the children who performed hand training and in 80% of the children who performed ADL-training.

Practice context

More than 80% of the children performed training of hand skills or ADL in natural learning environments (i.e., home and/or kindergarten), whereas 12-21% of the children performed “noncontextual” training that took place primarily in a rehabilitation unit or at the PT clinic. The parents reported that training was organized in two or more forms for approximately 50% of the children, with more than 70% of the children performing the training as integrated into daily activities and/or as individual training sessions. The parents reported being actively engaged in the training of hand skills for 86% of the children, whereas the proportion who was actively involved in the training of ADL skills was 27%. Kindergarten staff were involved in the training of hand and ADL skills for 50-64% of the children, whereas OTs and PTs were involved in the training of hand skills for 26% and 64% of the children, and in ADL training for 13% and 28%, respectively.

5.2.2 OT-reported training characteristics

The OTs reported that 62% of the children ($n = 58$) performed training of hand skills and 30% ($n = 27$) performed training of the selected ADL-activities. In 54% of the children who performed training of hand skills and 64% of the children who performed ADL skills, the OTs reported documented treatment goals. The characteristics of training are reported with lesser detail in the CPOP/UE protocol compared with the HabServ, yet the OTs reported that 26% of the children with unilateral CP performed CIMT, and that hand and ADL-directed services were provided as guidance to training for 61% of the children.

5.2.3 Who was most likely to perform hand training?

Among the included child-related (age, CP subtype, MACS) and family-related (mother's employment and education) characteristics, the only statistically significant variable to predict the likelihood of parents ($p = 0.04$) and OTs ($p = 0.008$) reporting training of hand skills was the MACS level. The children classified at MACS levels II-III were more than five times more likely to perform hand training than children classified at level I ($p < 0.002$), yet there was no significant difference between level I and levels IV-V.

The logistic regression models to explain the likelihood of children who performed ADL skills were not significant, very limited variance was explained by the included predictors, and the results are hence not reported.

5.2.4 Who was most likely to benefit from hand training?

In the logistic regression model to predict the likelihood of parents reporting large or very large child benefits of the hand training, selected child-related (age, CP subtype, MACS) and intervention-related (training frequency, number of training settings, number of organizational forms, goals) characteristics were included. The only statistically significant predictor variable was the number of organizational forms, with more than seven times higher likelihood of large/very large child benefits when the training was organized in two or more forms ($p = 0.011$).

The regression model to explain parent-reported benefits of ADL training was not significant, very limited variance was explained by the included predictors, and the results are therefore not reported.

6 DISCUSSION

In this thesis, aspects of hand function and bimanual development has been described through the results of a cohort study of young children with clinical signs of unilateral or bilateral CP, along with descriptions of the characteristics of current practice to target hand function. The study is presented in three papers. The main findings will be summarized in the following section, preceding methodological reflections and discussions of the results considering clinical relevance and theoretical knowledge.

6.1 Main findings

Through paper II, we documented that the distribution of the manual ability levels in young children with CP is comparable to what has earlier been reported for older children yet with a somewhat reversed proportion of children in MACS levels I and II. We also confirmed that the manual ability levels of young children with unilateral CP ranges from MACS levels I to III, whereas bilateral CP includes children at all five MACS levels.

The results presented in papers II and III illustrate a wide range of bimanual performance on the AHA and BoHA for children with unilateral and bilateral CP, respectively. Significant associations between MACS levels and age to the bimanual performance outcomes on the two tests were found. The MACS levels crudely predicted the developmental limits of performance on both the AHA and BoHA, and for children with unilateral CP, the MACS levels also discriminated between the age when the children reached 90% of their performance limit. The AHA performance at an early age was also predictive of different developmental trajectories for children with unilateral CP. For children with bilateral CP, the changes of bimanual performance on the BoHA were very limited, and the age when the children reached their age-90 was the same for children classified at MACS levels I-III.

Through paper I, we showed that the parents reported the current practice of hand-function interventions to include high amounts of training for the vast proportion of the included children, and they perceived large child benefits from the training. The parents reported more training than the OTs, who on the other hand reported services to be provided as guidance to hand training for a large proportion of the children. The parents reported the training to be vastly activity-based (i.e., performed as integrated into everyday activities) and to take place in natural learning environments (i.e., home or kindergarten). Whereas children with unilateral and bilateral CP were equally likely to perform training, children at MACS levels II and III were more likely to perform hand training than children at MACS levels I, IV, and V. The parents were most likely to perceive large child benefits when the training was organized in various ways.

6.2 Methodological considerations

A thoughtful research design with careful planning of procedures, data collection, analysis, and interpretation of results are crucial to maximize the validity and generalizability of the results. In the following section, various aspects of internal and external validity will be discussed with respect to the three papers included in this thesis.

6.2.1 Internal validity

The internal validity relates to the validity of the results for the original study population, or whether there are systematic errors (bias) in the results.²²⁴ The internal validity of a study may be flawed at any stage of the research process, and relates to the choice of study design and the inclusion of participants, the data collection process and choice of assessments, and the analysis and interpretation of the results.

Study design

The participants included in all three papers were recruited from the same cohort of young children newly registered in the CPOP or the CPRN, and although the study as a whole had a longitudinal design, cross-sectional data from the baseline assessments were used in papers I and II. A cross-sectional design can only be used to describe a phenomenon or association between selected variables at single time points.²²⁵ The purpose of the first two papers was to describe characteristics of current practice (paper I), aspects of hand function (paper II), and to explore associations to selected person- or intervention-related characteristics. Cross-sectional designs were thus considered appropriate with respect to the research questions in the first two papers.

In paper III, the purpose was to describe the changes in bimanual performance over time with respect to selected grouping factors (MACS levels and performance at an early age), and a longitudinal design was therefore suitable. The benefit of using a longitudinal design to study development, compared with a cross-sectional design with assessments of different children at different time points, is the opportunity to describe the variability of performance both *within* the same individuals over time and *between* individuals or groups (e.g., MACS levels). Together with the statistical methods of mixed-models analysis, this is a recommended design to study the development of heterogeneous populations like children with CP.²²⁶ There are, however, some limitations to the longitudinal analysis in paper III. A larger number of children, a more even distribution of children in the different groups, and a higher number of assessments for each child would have strengthened the study. However, an important benefit to the mixed-models analysis is to handle differing numbers of assessments for each participant in a research study, as all data points in the data set are included to make estimates for the individual participants. The mixed-models analysis is thus

a well-suited statistical method to accommodate the common challenges of asymmetric designs, high dropout rates, or missing data in longitudinal studies.²²¹

Participants

Between 53% and 63% of the eligible children were included in the three papers. Standard recommendations for what is an “acceptable response rate” do not seem to exist, and the satisfactory rate should be viewed with respect to the study population, the research design, and the methods for data collection.²²⁷ If we had used only assessments that are part of the standard CPOP and CPRN protocols, the participatory rate would most likely have been much higher. However, when considering the burden of additional questionnaires and assessments for the CPHAB project to the vulnerable group of children with CP and their parents, the response rate for this thesis seems highly acceptable. A threat to internal validity is the risk that the nonparticipants share some common features that contribute to some bias in the results.²²⁸ There was no known systematic selection of participants to our study, however, and the distribution of CP subtypes and functional levels in the three papers indicate that our sample is representative of the population of children with CP.^{22,38,53} Nonresponse bias is thus not likely to influence our results. The small number of children with unilateral CP classified at MACS level III in paper III reflects the small proportion of children classified at this functional level in this cohort overall, which can be seen by comparing participants and nonparticipants in Table 1 of paper III.

A limitation to this thesis is that no formal record was taken of the reasons why eligible participants were not invited to participate, why they declined the invitation, and why they dropped out during the study. There is a possibility that some unknown selection bias in the inclusion of participants might have influenced our data and resulted in failure to detect

associations that in fact exist (Type II error), or that we might have detected associations that are in fact not real (Type I error).

Assessments

Data collection took place in 17 out of the 21 regional pediatric rehabilitation units. Data were partly collected specifically for this study (and for the larger CPHAB project) and partly as the standard assessments for the CPOP and CPRN. Included data were hence multicenter- and register-based, with several persons involved in the data collection. Excellent inter- and intrarater reliability of the AHA is documented,²¹⁹ the validity of the AHA outcomes in papers II and III is thus unlikely to be biased by use of register-based AHA data. For the BoHA, such psychometric testing has not yet been conducted. All the BoHA videos for papers II and III were scored by myself, however, and the risk of rater bias was controlled for by including a second rater who scored one video for each of the included children. As reported in paper III, the interrater reliability was excellent ($ICC = 0.95, p < 0.001$).

The sensitivity of the AHA to detect changes in bimanual performance has been documented in several studies.^{83,110} Such information is yet not available for the BoHA, however, and there is a possible risk that real changes that may have occurred in the children with bilateral CP have not been detected by the BoHA. Adding further assessment tools to the data collection in paper III, to identify possible changes in relevant impairments and activity limitations, would have strengthened the study and contributed to broader knowledge.

Blinding of the AHA and BoHA raters with respect to the test sequence was desirable for paper III, yet not possible to achieve with our register-based and multicenter approach. AHA data were mainly collected from the standard CPOP check-ups, and the OTs who followed the children on a regular basis scored the AHAs. Although I was blinded to the MACS levels

when scoring the BoHA videos and attempted to disregard the test sequence, it was often easily observable that the children got older. The second BoHA rater was fully blinded to both test sequence and MACS levels.

Questionnaire

The literature review that was conducted during the planning of the study revealed that there was no questionnaire available to gather specific parent-reported information about the services and interventions provided to children with CP and their families. An expert group, therefore, developed the HabServ questionnaire during the planning of the CPHAB, and selected parts were used in paper I. The HabServ was pilot-tested on the parents of 19 young children with CP in three rounds. Modifications were made after each round, by collapsing two related sections and clarifying some questions with examples. The construct validity of the questionnaire has not been investigated, and although test-retest data are available, the data have not been analyzed and thus the reliability is not yet known. Consequently, the parent-reported data are somewhat doubtful, as we do not know whether the parents interpreted the questions according to the intention. The HabServ data must hence be interpreted as a description of the target questions as written in the questionnaire.

6.2.2 External validity - generalizability of the results

The external validity refers to the generalizability of the results.²²⁸ The Norwegian CP registers (the CPOP and CPRN) include approximately 90% of the total population of children with CP.²²⁹ Because we were able to include more than 60% of the eligible children, and the participants closely resembled both the nonparticipants and the general CP population (as illustrated in Table 2 in paper II), some generalizations of the results may be made. The

somewhat distorted distribution of the MACS levels between participants and nonparticipants, illustrated in Table 1 of paper III, may reflect that we have been particularly successful to include the children classified at MACS level II, which may be considered a strength of the study.

For paper I, the parents of 11 children who could not complete the questionnaires due to language barriers were excluded from participation. This may have caused some selection bias, as there is a possibility that the excluded parents, who represent non-Western cultures, might potentially have responded differently to the questionnaire.

6.2.3 Summary of methodological considerations

To summarize, the study designs used in this thesis are considered appropriate to address the research questions raised in the three papers. A strength of the thesis is the use of population-based data with a representative distribution of subtypes and functional levels compared with the nonparticipants and to population-based descriptions of older children with CP, which allows for some generalization of our findings. In contrast, the numbers are small, which primarily reflects the generally low prevalence of CP and Norway being a small country. Together with the relatively large proportion of nonparticipating children (as illustrated in Figure 4), the small numbers, and the possibility of unknown selection bias may potentially have caused Type I and Type II errors in our results. For paper I, the parent-reported data must be interpreted as a response to the questions included in the questionnaire, and can only be generalized to the population of children with CP in a similar cultural context.

6.3 Discussion of the main results

In the following sections, the main findings regarding manual abilities and development of bimanual performance from papers II and III will first be discussed, preceding a discussion of the main results from paper I about the characteristics of current practice to target hand function.

6.3.1 Distribution of manual ability levels in a young population

This study is the first to report the distribution of manual ability levels in a cohort of children younger than four years of age, by use of the Mini-MACS, which was newly released for common use.²¹⁴ Although the distribution closely resembled what has earlier been reported in Scandinavian population-based studies of older children,^{22,38,51,53,56} the reversed proportion of children classified at Mini-MACS/MACS levels I (23%) and level II (49%) found in our study (reported in paper II) is similar to reports from two other international studies,^{54,55} and may be interpreted in two ways. First, the CP diagnosis is often not confirmed until five years of age,²³⁰ and young children with none or minor hand motor impairments may not have been identified and recruited to the national registers by the time of our data collection. Second, as children younger than four years of age may be in rapid development, the Mini-MACS levels may be less stable in the youngest children.²¹⁴ It may be expected that some of the children classified at Mini-MACS level II in this study will possibly be reclassified at level I (or level III) at a somewhat older age.

6.3.2 Bimanual performance and development

Another unique contribution of this thesis is the use of the BoHA to describe bimanual performance in children with bilateral CP for the first time. Whereas the AHA has been used to describe hand use and bimanual ability in children with unilateral CP for more than a

decade,⁸⁴ this study is the first to describe bimanual performance by use of either AHA or BoHA data in a population-based sample of children with CP. In papers II and III, we demonstrated large variations of bimanual performance both among children with unilateral and bilateral CP. The performance variations on the two tests reflect the known heterogeneity of children with CP and highlight the need to consider the unique resources, challenges, and service needs of each individual child.^{21,46,47}

Bimanual performance and development according to MACS levels

Because most everyday activities include the manipulation of objects and the use of both hands together, the close associations between the bimanual performance on the two assessments and the children's levels of manual ability when handling everyday objects were not surprising. Neither was it surprising to find that the MACS levels could separate between various developmental trajectories of bimanual performance on the AHA, as similar findings have been reported previously in Swedish convenience samples of children with unilateral CP.^{103,108} For children with bilateral CP, we had much less prior knowledge of their expected development. It was therefore interesting, albeit not surprising, that also among the children with bilateral CP, the MACS levels could discriminate between the estimated maximum limits of bimanual performance as measured by the BoHA.

The ability of the MACS to discriminate between various levels of bimanual performance limits over time on both the AHA and BoHA is clinically important, as it presents as a highly valuable tool for communicative purposes between professionals and parents regarding children's future development and realistic intervention goals.

Distinct developmental trajectories between unilateral and bilateral CP

The AHA and BoHA are similar tests, yet the outcomes may not be directly compared because the scoring items and scoring criteria are somewhat different. Nevertheless, as both tests produce Rasch-based measures on a 0-100 scale, share many common features, and are developed from the same concepts, some coarse comparisons may be made.

The two subgroups of children with unilateral and bilateral CP presented with rather distinct developmental patterns. The change in performance over time was very limited for the children with bilateral CP, whereas the mean change in observed performance for the children with unilateral CP presented as clinically relevant differences. Careful scrutiny of the longitudinal data yields no explanation of the differences with respect to, e.g., age differences, number of assessments, or time of monitoring between the two groups (see Table 4). For children with unilateral CP, the MACS levels could crudely discriminate between the rates of development, as illustrated by the age when they reached 90% of their estimated maximum AHA limit (MACS I: 30 months, MACS II: 57 months, MACS III: 155 months). This was not evident for the children with bilateral CP, who reached the age-90 for the BoHA outcomes at roughly 30 months among all three MACS levels.

The distinct developmental trajectories for the two subgroups may be explained in different ways. For the AHA, the psychometric properties are thoroughly investigated, and the test-retest reliability and responsiveness to change have been documented in several studies.^{82,83,217,218,231,232} For the BoHA, the psychometric properties are less explored, and although the sensitivity of the test to differentiate between various performance levels is shown, the responsiveness to change is not known.⁷ There might thus be a risk that changes in the bimanual performance of the children with bilateral CP in paper III occurred without

being detected by the BoHA. Nevertheless, the limited development demonstrated in our study somewhat confirms earlier reports by use of different assessments,^{93,112} yet needs to be further explored.

The underlying nature of the motor impairments in unilateral and bilateral CP may also contribute to their distinct development. Having both hands affected, but one hand more affected than the other, may be particularly challenging – as illustrated by the significant difference between the BoHA outcomes for children with a symmetric and an asymmetric performance pattern in paper II. On the other hand, high ability to use the dominant hand may be the key factor for more proficient bimanual performance, and better grasping ability in the affected hand at an early age may predict higher levels of performance and more rapid development, as illustrated in paper III.

The role of additional challenges (e.g., cognitive, perceptual, and sensory impairments) to the motor impairments may be important for bimanual performance and development.

Although such added challenges are most common in the more severely impaired children,³⁰ we also know that they are most frequently seen among children with bilateral CP.^{21,41,61,96,97}

The development of bimanual performance in typically developing children progresses tremendously during the second year of life, as the child by two years of age is able to differentiate between various grasps and adjust the grip force to the nature of the objects.²³³ At the same time, the typically developing child repeatedly practices everyday activities, such as putting on socks and shoes, or willingly sets the table for a meal.¹² Being able to complete successfully increasingly more complex activities as the child gets older depends on previous experience and the exploration of objects and activities that are gradually more advanced, yet within what Vygotsky called “the proximal zone of

development”.²³⁴ The movement patterns of most children with CP are usually characterized by slow and poorly coordinated motor performance, which in turn may lead to less object exploration and slower development.⁴ The distinct developmental limits and rates between functional levels and subgroups in our study may thus partly reflect various degrees of experience from object manipulation. Whereas the children with at least one “good” hand may have successfully explored various objects independently from an early age, the children who have more limited grasping ability, limited coordinative ability, or two hands that are more severely impaired, may not have experienced successful object manipulation to the same extent.

More research is needed to learn more about the bimanual performance and development in both children with unilateral and bilateral CP. The development of hand function should be described by use of other activity-based assessments, and by use of assessments that measure functioning within the body functions and participation components of the ICF. Furthermore, the influence of cognitive, sensory, and perceptual impairments for hand-function development should be investigated among both children with unilateral and bilateral CP.

Development of bimanual performance and the role of early ability

In paper III, our results also confirmed the findings of the two previous Swedish studies on children with unilateral CP,^{103,108} that the level of AHA performance at 18 months of age could crudely discriminate both between estimated limits of performance and the age at which the children reached 90% of their limit. The AHA-18 levels were categorized according to the children’s performance by use of grasps, and the findings suggest that the children who efficiently use active grasps at 18 months of age have a more rapid development and

reach higher maximum performance levels than the children with less efficient or no active grasps. This indication is of significant clinical relevance. Being able to grasp, hold, release, turn, and manipulate different objects between the hands may be of crucial importance for the 18-month old child to explore new opportunities, and thus learn and develop new hand motor skills required in everyday activities.^{11,14,54} Although the role of early grasping ability for bimanual performance development needs to be further explored, the results urge us to reflect on how we provide interventions that promote active grasping in the youngest children with unilateral CP.

The large performance variations over time for both children with unilateral and bilateral CP is important, as it underlines the need to consider the individual child's distinct performance and development, rather than using mean curves to draw firm conclusions on the expected development. Nevertheless, the mean curves are important to illustrate the most likely developmental trajectory for children classified within distinct functional groups, when communicating with parents regarding prognosis, goal setting, and intervention planning for their child.

Contradictory to the validity studies of the two assessments,^{7,82} we found a significant association between age and the bimanual performance outcome for both the AHA and BOHA in paper II. One explanation may be the young age of our children, as we have learned from paper III, and from previous research, that the most rapid change in performance seems to occur prior to three to four years of age. After this age, the curves tend to flatten out, particularly for children with unilateral CP classified at MACS levels I and II.^{103,108} The significant association between AHA performance and age, identified in the young population included in paper II, may reflect the rapid development of typically developing

young children who at 18 months of age have just recently started to manipulate objects bimanually.⁹

Motor-learning-based interventions are shown to improve hand function in children with unilateral CP and may possibly impact the long-term development of bimanual performance among children with CP.^{6,110,121,170} The effect of hand-function interventions for children with bilateral CP is hardly described in the literature, and descriptions of hand-function interventions for this subgroup are lacking. Whether the distinct developmental curves for the two subgroups may in part reflect the extent to which hand function is targeted differently for the two subgroups remains to be explored.

6.3.3 Current practice to improve hand function

As presented in paper I, the parent-reported data showed that the vast proportion of the children performed hand training and training of the selected ADL activities (dressing, eating, and playing), while the OTs reported training for a somewhat smaller proportion.

Why did parents and OTs report different proportions of children to perform training?

Data on the characteristics of current practice come from two separate sources. Whereas the parents responded to a questionnaire specifically developed for the CPHAB project, requesting what target areas their child had “been practicing” during the preceding six months, the OTs’ responses were retrieved from the regular CPOP protocol as part of the ordinary follow-up program and refer to the therapeutic management by various professionals. It was hence not expected that the responses by the parents and the OTs would correspond. The large proportion of children reported by the parents to practice hand and ADL skills could possibly mirror the practice of any typically developing preschool child

who repeatedly rehearses the same tasks over and over again to be independent and skillful in everyday life.¹² Nevertheless, the fact that the parents also reported professionals to be largely involved in the training, and that training was organized as a combination of group and individual sessions, as well as integrated into everyday activities, implies that this practice must be something additional to what is common for typically developing children. The rather large proportion of children reported by the OTs to receive services as guidance to training may reflect the philosophy of family-centered practice and integrated training by use of home programs.¹³⁶

Who performed training?

The only significant factor in the logistic regression models to predict the likelihood of performing hand training was the MACS levels, where children classified at MACS levels II and III were more likely than children at levels I, IV, and V to perform hand training, as reported by both the parents and the OTs. Neither the age, the CP subtype, nor family-related factors were significant predictors, which was somewhat surprising. Evidence-based interventions primarily exist with documented effects for the sub-group of children with unilateral CP,^{6,121} who were therefore expected to participate more in hand training. Furthermore, the importance of early intervention has been highlighted over the last decade,^{109,235-237} which we hypothesized would be reflected in our data. However, in accordance with previous studies of current pediatric rehabilitation practice,^{204,238} also in the Norwegian upper limb management, there seem to be gaps between what is recommended practice and what is usually provided. Nevertheless, in the National Institute of Care Excellence (NICE) guidelines for the management of CP and spasticity, there are no specific recommendations supporting more extensive interventions for specific age groups or subgroups.^{208,209}

Who benefits most from training?

The results from paper I furthermore showed that the parents perceived large child benefits from the training for the vast proportion of the children, and the logistic regression analysis showed that the only significant factor associated with large or very large child benefits was when the training was organized in various ways (e.g., individual sessions and integrated into everyday activities). This supports the recommendation of utilizing natural learning environments to achieve repetitions and goal-directed practice.¹²⁶

Clinically meaningful effects of hand-function interventions have been suggested to result primarily from the *dose* and *organization* of therapy, rather than from the treatment ingredients per se.^{5,130,132,133} When we consider the two intensity-related factors in our parent-reported data, the *duration* (for how long had the training taken place?) and the *frequency* (how many hours or days of the week did the child practice?), the training intensity seems high. The finding that the frequency factor was not significant for the reported child benefits was particularly surprising. Comparisons of similar interventions at various intensities have indicated that more training is better,^{5,130,133} and the intensity of therapy may be described as intensive “blocks” of short duration/high intensity, or as “distributed” with long duration/less intensity.¹³⁰ In the NICE guidelines for spasticity management, intensive blocks of activity-focused interventions are recommended.²⁰⁹ It appears that the current practice in our study included a distributed management approach. Almost all the children had performed training for the whole of the preceding six-month period, and approximately 60% practiced daily or several times a day. We do not know, however, what this training in fact included, rather than primarily being organized individually and integrated into everyday activities, taking place mostly at home and in kindergarten, and with defined goals. The training was thus largely *contextualized*, which is

viewed as an important way to create meaningful learning opportunities.²³⁹ Whether the training was *specific* enough, however, and whether it included sufficient *repetitions* of target activities that were at the “*just right challenge*” to promote change and motivation, cannot be concluded from our data. These latter principles of motor learning theory (see Figure 3) are important features of evidence-based interventions and should be included in the current upper limb management. As stated by Novak in a description of OT home programs; “garbage in - garbage out”, referring to the acknowledgment that the mode of intervention is not as important as what we put into the therapy.¹³⁹ Structured OT home programs have been described as a “window of opportunity” to achieve a higher dosage of therapy and may be a useful approach to more structured contextualized interventions if they are carefully planned according to the defined principles.^{136,139,240}

Our findings that the training was largely contextualized and integrated into the children’s natural activity settings may indicate that the biopsychosocial way of understanding disability and hand-function limitations is reflected in the current upper limb management for children with CP. An important feature of the ICF model is the reciprocal relationship between the various components (Figure 1), and the role of environmental factors. The overarching goal of hand-function interventions is independence and participation in everyday activities, and such contextualized and integrated training largely identified in this thesis may thus be viewed as a way of “enhancing participation by participation”, instead of providing therapy primarily in noncontextualized therapeutic environments hoping to achieve transfer of skills to the everyday situations.

Careful planning of hand-function interventions around motivating and attainable target activities that are important to the child, not too easy to perform, yet is still within reach of

what the child is able to accomplish by structured practice (i.e., activities at the “just right challenge”⁴ or within the “proximal zone of development”²³⁴) is also an important asset of motor-learning-based interventions.⁴ This highlights the importance of thorough assessment of children’s functioning within various ICF components, to achieve meaningful and individualized activity goals. Given the documented effects of goal-directed and activity-based therapy to improve motor performance,^{180,182,184,185,241} we were also somewhat surprised to find no significant effect of the “goals for training” factor on the perceived child benefits. Both the parents and the OTs reported defined goals for the training for a large proportion of the children. Nevertheless, having individualized goals set for interventions does not necessarily mean that those goals are set in collaboration between parents and therapists, or implemented as specific components of the training as such, which are core features of goal-directed functional therapy.^{4,120,180} Although defined goals may increase awareness of certain target areas,²⁴² a previous study demonstrated that having goals written, yet not specifically practiced, does not cause changes in functional skills or promote goal attainment.^{243,244}

The parent-reported child benefits represent an essential perspective of the upper limb management of children with CP, yet must not be confused with the objective measures of performance outcomes of interventions.²⁴⁵ Important insights have been described through self-reported measures of hand use in a previous study of upper extremity surgery, yet without a direct relationship to what was detected by quantitative objective measures of capacity or performance.²⁴⁶ The objective evaluation of training effects for the children included in this thesis cannot be performed by our applied study design. Nevertheless, the parents’ perceptions of large child benefits provide indications that there are significant advantages to utilizing the children’s everyday activity settings and the people naturally

taking part in these, in a varied and goal-directed way, to promote hand function and activity performance.

Is the current practice evidence based?

Whether the Norwegian current practice, as reported by parents and OTs in paper I, reflects what ought to be included in the usual services and interventions for upper limb management in young children with CP can only partly be answered through this thesis.

When looking at the key factors identified through the overview of motor learning theory in Section 2.5.1 (Figure 3), it appears that many principles are incorporated in the current practice, such as activity-based interventions, goals for training, interventions utilizing the natural learning opportunities with both parents and professionals as agents of provision, and high amounts of training. Nevertheless, as already mentioned, many questions remain unanswered regarding the specificity and goal-directedness of the training, the repeated practice of target activities and the intensity concerning the number of training hours.

From the OT-reported data in paper I, we found that 26% of the children with unilateral CP performed CIMT training, which is the most highly documented training approach to improving hand function.^{121,247} With more children included for paper III, and data retrieved from the CPOP register at a later age, we found that the proportion of children with unilateral CP who had performed CIMT had increased to 41%, and the proportion for BIMT was 31% (see Table I of paper III). This trend is interesting, yet contradictory to the idea of early intervention, and may indicate that the Norwegian children with CP receive more structured and evidence-based upper limb training as they approach school age. Previous research has provided evidence that CIMT may be beneficial in promoting changes in hand function during both childhood and adolescence.¹⁶⁷ Hence, rather than interpreting the low

amounts of CIMT training for the children in paper I as a failure to provide evidence-based practice, we may view the increase of this training approach between the analysis for paper I and paper III as a promising trend for more evidence-based upper limb therapies. The NICE guidelines recommend CIMT training as an important approach to activity-focused interventions.²⁰⁹ The parent-reported descriptions of the current practice may imply an eclectic approach to a distributed intervention model, where the OTs and PTs collaborate and supervise parents and kindergarten personnel in how to integrate principles from evidence-based practice into everyday learning situations.²⁰⁶

We cannot interpret from our data whether the current practice of upper limb management is family centered. From paper I, we know that 10-23% of the parents reported participating in parent education programs targeting goal setting or motor training, more than 80% of the parents were actively involved in the hand training, and the training was organized within everyday activities for approximately 70% of the children. Nevertheless, we do not know whether this reflects the participatory practices where the goals are set in collaboration between the child, family, and professionals, or whether the integrated training is planned and individualized according to the family needs and wishes.^{120,139} In a previous study investigating the extent to which parents of young Norwegian children with CP experienced the general pediatric rehabilitation practices to be family centered, Myrhaug and co-authors concluded that the services generally seemed to be satisfactorily delivered as relational practices, yet to a lesser extent organized as participatory practices.²⁴⁸

Our findings that a large proportion of the training took place at home, integrated into everyday activities, with defined goals, and with parents actively involved, might indicate that the services are partly provided as home programs. Increasing research evidence exists

of the successful implementation of CIMT, BIMT, or other upper limb therapies delivered in home-based settings,^{166,249-258} yet parents are shown to comply more strongly with home programs when they encourage partnership between parents and therapists rather than implementing predefined programs into the family's everyday life.^{136,139,259,260}

To enhance the specificity, goal directedness, intensity, and functional effects of the current upper limb management, an important step may be to organize the large amounts of contextualized training in a more structured way. By establishing mutually agreed-upon home programs that are based on assessments within all the ICF components, the services may be carefully individualized to meet the child and family needs.^{136,139} In addition, by structuring the large amounts of training as more intensive blocks of goal-directed therapy,²⁶¹ the children may experience functional changes recognized as child benefits by the parents, yet also captured by objective assessments. In turn, such evidence-based current practice may contribute to improved hand function, promote independence in everyday activities, and most importantly stimulate participation in various life situations in a long-term perspective.

7 CONCLUSIONS AND CLINICAL IMPLICATIONS

The results from this thesis show that both children with unilateral and bilateral CP present with limited hand function, perform within a large performance range on the two included assessments of bimanual performance, and display various developmental trajectories.

Children with unilateral and bilateral CP were equally likely to perform training, and the vast majority among both groups performed hand training.

Our findings raise some questions about the evidence-based practice in the upper limb management of young children with CP in Norway. The research literature mainly describes hand-function interventions for children with unilateral CP, and even for this subgroup, there seems to be a gap between what we know about effective approaches to intervention and the extent to which this is included in the usual services. For children with bilateral CP, there are limited evidence-based interventions, and we do not know what the best practice is. Still, children in this subgroup seem to perform training to the same extent as children with unilateral CP.

Basic principles of motor learning appear to be generally incorporated into the current practice of upper limb management as described in this thesis. However, we do not know the specific ingredients of this training, although the organization with respect to activity-focused and contextualized learning situations seems promising. The current practice seems to be largely organized with distributed intensity, in contrast to the recommended blocks of intensified and structured training periods. Our data do not provide evidence as to whether the intensity includes sufficient repetition of target activities within the “just right challenge” to promote functional change.

In a meta-analysis of upper limb interventions for children with unilateral CP, the authors argued that upper limb interventions should adhere to the following clinical guidelines⁶ (1) contemporary motor learning theory should be the basis for activity-based interventions, (2) interventions should be goal-directed with child- [or parent] chosen goals, (3) goal attainment should be objectively evaluated, (4) valid and reliable objective measures of upper limb outcomes should be used, and (5) the dose of therapy must be sufficient to promote change.

More unified international guidelines may be beneficial to ensure more structured and evidence-based interventions, and more systematic knowledge translation is important to promote knowledge and awareness of how to implement the evidence-based approaches to upper limb management into current practice.

8 FUTURE RESEARCH

Through the process of this doctoral work, new research questions have emerged that need attention in future studies. For children with bilateral CP, the list of unanswered questions is extensive. There is a need to identify hand-function challenges systematically within all the ICF components, and to investigate associations between various cognitive and motor impairments to hand-function limitations and the performance of everyday activities and participation. Associations between gross motor function and hand function should also be explored, as children with bilateral CP represent a largely diverse group, and the role of e.g., postural stability during seating may be of particular interest. Furthermore, the limited development of bimanual performance in our study needs to be confirmed in larger studies with various outcome measures, and importantly, the responsiveness to change and the test-retest reliability of the BoHA need to be established.

For the development of bimanual performance in children with unilateral CP, there is still a need for larger population-based studies in which the children are followed across a longer time span. The role of various factors in the development of bimanual performance should be explored, particularly the role of early grasping ability and the impact of early and intensified interventions.

With respect to upper limb management, several evidence-based approaches exist for children with unilateral CP. Knowledge of these approaches should be used to describe and explore possible effects also among children with bilateral CP. An important future issue will be to identify what interventions contribute to changes in hand function, what factors influence the treatment effects, and how to put evidence-based interventions into current

practice to promote the long-term development of hand function in both children with unilateral and bilateral CP.

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Paper I

Paper II

Paper III

Appendices

The Habilitation Services Questionnaire (HabServ)

The purpose of this questionnaire is to gain information on habilitation services provided to the child and its family during the **last 6 months**. Habilitation services include all extra interventions and benefits that may be offered to children with CP in support of the child and its family.

The questionnaire covers 4 groups of interventions and services:

1. **Training and stimulation** (motor-, communicative- and everyday skills, etc.)
2. **Assistive technology** (mobility, communication, training, daily care, transportation, environmental adaptations, etc.)
3. **Courses and parent training** (organized by the municipality, the specialist health care, user organizations, etc.)
4. **Services and benefits** (physiotherapy, special education, individual plan, respite service, financial support, etc.)

On the next page you will find an example of how to complete the questionnaire. Please consider the example carefully before you start.

This is an example of how to complete the questionnaire. Please consider this carefully before starting.

Training and stimulation

What has the child been practicing during the <u>last 6 months?</u>	What professionals took part in the training?	How were you involved in the training?	For how long was the training carried out?	How often did the training take place?	Where did the training take place?	How was the training organized?	Were goals set for the training?	How much did the child benefit from the training?
Please mark with an "x" next to the areas that were targeted during training. Then, fill in the appropriate numbers in each of the following columns.	1= physiotherapist 2= preschool teacher 3= special education teacher 4= assistant 5= occupational therapist 6= others 7= do not know	1= not involved 2= observing 3= performing	1= 1-2 weeks 2= 3-4 weeks 3= 1-2 months 4= 3-4 months 5= the entire period 6= do not know	1= once/week 2= twice/ week 3= 3-5 times/ week 4= 6-7 times/ week 5= several times daily 6= do not know Please fill in <u>only one</u> number	1= home 2= preschool 3= school 4=physiotherapist's 5= habilitation center 6= swimming pool 7=riding center 8= other sites	1= individually 2= group 3= part of everyday activities 4= do not know	1= yes 2= no 3= do not know Please fill in <u>only one</u> number	1= no/small benefits 2= some benefits 3= large benefits 4= very large benefits 5= unsure Please fill in <u>only one</u> number

Usual training								
Gross motor skills (i.e. sitting, rising, standing, walking)	X	1,4		4	2	3	1	2
Stretching								
Hand motor skills (i.e. grasping, releasing, cutting)								
Language and speech	X	4		5	2	2	1,2	3
Alternative communication (when speech problems)								
Dressing and undressing	X	1,5		4	3	1,2	3	1
Toilet training								
Social skills (i.e. playing with children/adults)	X	4,5		2	4	2	2	1
Others, please comment								
Did the child participate in particular training-/habilitation programs (i.e. Conductive education, Family Hope Center, Vojta) during the last 6 months?								
No	☐	Yes	☐	Please indicate program:				

1. Training and stimulation

What has the child been practicing during the last 6 months?	What professionals took part in the training?	How were you involved in the training?	For how long was the training carried out?	How often did the training take place?	Where did the training take place?	How was the training organized?	Were goals set for the training?	How much did the child benefit from the training?
Please mark with an “x” next to the areas that were targeted during training. Then, fill in the appropriate numbers in each of the following columns.	1= physiotherapist 2= preschool teacher 3= special education teacher 4= assistant 5= occupational therapist 6= others 7= do not know	1= not involved 2= observing 3= performing	1= 1-2 weeks 2= 3-4 weeks 3= 1-2 months 4= 3-4 months 5= the entire period 6= do not know	1= once/week 2= twice/ week 3= 3-5 times/ week 4= 6-7 times/ week 5= several times daily 6= do not know	1= home 2= preschool 3= school 4=physiotherapist’s center 5= habilitation center 6= swimming pool 7=ridding center 8= other sites	1= individually 2= group 3= part of everyday activities 4= do not know	1= yes 2= no 3= do not know	1= no/small benefits 2= some benefits 3= large benefits 4= very large benefits 5= unsure Please fill in only one number
Usual training								
Gross motor skills (i.e. sitting, standing, walking)								
Stretching								
Hand motor skills (i.e. grasping, releasing, cutting)								
Language and speech								
Alternative communication (when speech problems)								
Eating and drinking								
Dressing and undressing								
Toilet training								
Play skills (i.e. building, puzzling, playing with dolls)								
Social skills (i.e. playing with children/adults)								
Physical activities (i.e. swimming, horse riding, biking, riding, sledding)								
Others, please comment								
Did your child participate in particular training-/habilitation programs (i.e. Conductive education, Family Hope Center, Vojta) during the last 6 months?								
No	☐	Yes	☐	Please indicate program:				

2. Assistive technology

What type of assistive technology has been used during the <u>last 6 months</u> ?	Where is the aid/device used?	How often, or for how long, is the aid/device used?	For what purpose is the aid/device used?	How much does the child, or you, benefit from using the aid/device?
Please <u>mark</u> with an “x” next to the assistive technology that has been used. Then, fill in the appropriate numbers in each of the following columns.	1=home 2=kindergarten 3=school 4=respite home 5=leisure activities 6=transportation 7=others 8=do not know	1=<once / week 2=1-2 times / week 3=3-4 times/ week 4=5-7 times/ week 5=several times daily 6=most of the day 7=nightly 8=do not know	1=prevent deformities 2=improve functioning 3=participation in activities 4=ease daily care	1=no/small benefits 2=some benefits 3=large benefits 4=very large benefits 5=unsure
	Walking aid			
	Manual wheelchair			
	Powered wheelchair			
	Standing aid			
	Adapted bike			
	Special stroller			
	Bathing-/showering aid			
	Toilet aid			
	Eating aid			
	Speech generating device	*LT		
	Communication book	*LT		
	Assistive devices for play/stimulation			
	Special chair			
	Adapted bed			
	Exercise aid			
	Technical aid to lift the child			
	Doorstep eliminator			
	Automatic door opener			
	Ramp			
Assistive technology in car				
Orthoses				
Others, please comment				
Others, please comment				

*HT= High-tech /computer based
*LT=Low-tech / simple conversation equipment

3. Courses and parent training

What were the topics of the course/parent training in which you participated during the last 6 months?	Who was responsible for the course/training?	How often did the course/training take place?	For how long did the course/training last?	How was the course/training organized?	How much did you benefit from the course/training?
<p>If you have participated, please mark with an "x" next to the topics of the course/training.</p> <p>Then, fill in the appropriate numbers in each of the following columns.</p>	1=municipality 2=specialist health care 3=Learning and Mastery Center 4=user organizations 5=others (comment) <u>Please fill in only one number</u>	1=once 2=twice 3=3-4 times 4=> 4 times <u>Please fill in only one number</u>	1=1-7 hours 2=8-15 hours 3=16-25 hours 4=26-35 hours 5=>35 hours <u>Please fill in only one number</u>	1=lecture 2=demonstration 3=practical training 4=group discussion 5=web-based 6=conversation 7=others, please comment <u>Please fill in only one number</u>	1= no/small benefits 2= some benefits 3=large benefits 4=very large benefits 5= unsure <u>Please fill in only one number</u>
	The CP diagnosis				
Parenting a child with disabilities					
Legal rights					
NGO peer work					
Individual plan					
Goal setting					
Motor training					
Play and stimulation					
Alternative / augmentative communication					
Epilepsy					
Eating and nutrition					
Leisure activities					
Computer technology					
Others, please comment					
Others, please comment					

4. Services and benefits

What economical or other benefits have the child and the family received during the <u>last 6 months</u> ?	How satisfied are you by the volume or size of the benefits?
Please <u>mark</u> with an “x” next to the benefits that you, or your child, is granted due to the child’s CP diagnosis.	1=not satisfied 2=quite satisfied 3=well satisfied 4=very well satisfied
Then, fill in the appropriate number in the following column.	Fill in only <i>one</i> number
Physiotherapy	
Special education	
Occupational therapy	
Additional preschool assistant	
Professional psychological support	
Physician follow-up	
Public Child Health Care Center	
Individual plan	
Personal coordinator	
Multidisciplinary team	
Basic benefits	
Supplementary benefits	
Caregiver benefits for child’s illness	
Caregiver benefits	
Respite service	
Leisure assistant	
User controlled personal assistant	
Car benefits	
Housing benefits	
Training benefits	
Others, please comment	
Others, please comment	

Are there any services or benefits that you have been missing or asked for, yet not been granted?
Here you can write anything you may find important to communicate regarding interventions, services and benefits provided to the child and family.

Oppfølgingsprogram for barn med cerebral parese Ergoterapiprotokoll

Versjon 01.01.2015.

Personnummer:	Dato undersøkelse:
Etternavn:	Fornavn:
Navn ergoterapeut i habiliteringstjenesten:	
Navn ergoterapeut i 1.linje:	

Diagnose

SCPE Diagnose			
Spastisk	Unilateral	Hemiplegi, høyre	☐
		Hemiplegi, venstre	☐
	Bilateral	Diplegi	☐
		Kvadriplegi	☐
Dyskinetisk		Choreoathetose	☐
		Dystoni	☐
Ataktisk			☐
Ikke klassifiserbar CP			☐

Klassifisering- og kartleggingsinstrument

MACS Manual Ability Classification System							
Klassifikasjon av håndfunksjon							
	I	II	III	IV	V	Ikke klassifisert	Kommentar
MACS	☐	☐	☐	☐	☐	☐	

CFCS Communication Function Classification System						
	I	II	III	IV	V	Ikke klassifisert
CFCS	☐	☐	☐	☐	☐	☐
Barnet bruker følgende kommunikasjonsformer:						
☐	Tale					
☐	Lyder (som "aaaah" for å påkalle samtalepartnerens oppmerksomhet)					
☐	Blikkpeking, mimikk, gester og/eller peking (med en kroppsdelt, pekepinne eller laser)					
☐	Manuelle tegn (håndtegn)					
☐	Kommunikasjonsbok, -tavler og/eller bilder					
☐	Lav- eller høyteknologiske talemaskiner					
☐	Annet					

HOUSE funksjonsklassifisering av hver hånd	Hø	Ve
House 0 - 8	_____	_____
	Ja	Nei
Har barnet tohåndsfunksjon	☐	☐

AHA Assisting Hand Assessment
Dato utført: _____ Sumpoeng: _____ Skalpoeng %: _____ Logitbased 0-100 AHA-unit: _____

PEDI Pediatric Evaluation of disability Inventory			Dato utført _____	
Område:		Skalert skår	Standard feil	
Egenomsorg	Funksjonelle ferdigheter			
Forflytning	Funksjonelle ferdigheter			
Sosial fungering	Funksjonelle ferdigheter			
Egenomsorg	Hjelpebehov			
Forflytning	Hjelpebehov			
Sosial fungering	Hjelpebehov			
Antall tilpasninger:	Ingen	Barneutstyr	Tekn. hjelp.	Omfattende
Egenomsorg				
Forflytning				
Sosial fungering				

Spastisitet

SPASTISITET		
etter "Modified Ashwort" skala (0, 1, 1+, 2, 3, 4)		
	Hø	Ve
Albufleksorer	_____	_____
Underarmspronatorer	_____	_____
Håndleddsflexorer	_____	_____
Tommelfleksorer /adduktorer	_____	_____
Fingerflexorer	_____	_____

Leddbevegelse

LEDDBEVEGELSE passiv og aktiv		Hø	Ve
Skulder	Passiv fleksjon (>160°)	_____°	_____°
	Aktiv fleksjon	_____°	_____°
Albu	Passiv ekstensjon (0°)	_____°	_____°
	Aktiv Ekstensjon	_____°	_____°
Underarm	Passiv supinasjon (90°)	_____°	_____°
	Aktiv Supinasjon	_____°	_____°
Håndledd	Passiv ekstensjon m/flekt.fingre (70°)	_____°	_____°
	Passiv ekstensjon m/ekst.fingre (70°)	_____°	_____°
	Passiv ulnardeviasjon (30°)	_____°	_____°
	Passiv radialdeviasjon (20°)	_____°	_____°

Viser tegn på smerte ved undersøkelsen	Ja	Nei	
	☐	☐	

HOUSE Klassifikasjon av tommelens stilling ved aktivt grep				
		Hø	Ve	
Thumb-in-palm Type 0 - 4				
Ingen thumb-in-palm 0
	1	2	3	4

ZANCOLLI Klassifikasjon av håndledd og fingres evne til aktiv ekstensjon				
		Hø	Ve	
Zancolli Type 0, 1, 2A, 2B, 3				
Uten anmerkning 0
	1	2A	2B	3

Ergoterapi

ERGOTERAPI

Har barnet hatt kontakt med ergoterapeut i tillegg til CPOP-vurderingen:

Ja Nei
☐ ☐

Ergoterapi:

- ☐ Trening håndfunksjon
☐ ADL-trening
☐ Hjelpemiddeltilpasning
☐ Stoltilpasning
☐ Tilrettelegging og veiledning

TRENING AV HÅNDFUNKSJON

Har barnet hatt trening for å bedre håndfunksjon siden forrige CPOP vurdering:

Ja Nei
☐ ☐

Type håndfunksjonstrening:

- ☐ Råd og veiledning for håndfunksjon
☐ Funksjonell håndtrening
☐ Modifisert CI-terapi, CIMT,
☐ Bimanuell intensiv trening, BIMT,

Antall ganger i uken				Uker, tidsperiode			
<1	1-2	3-5	6-7	<2	2-6	7-12	>12
☐	☐	☐	☐	☐	☐	☐	☐
☐	☐	☐	☐	☐	☐	☐	☐
☐	☐	☐	☐	☐	☐	☐	☐

Håndfunksjonstreningen er utført av:

- ☐ Ergoterapeut ☐ Fysioterapeut ☐ Foreldre ☐ Pedagog , andre

		Ja	Nei				
Er det formulert mål for tiltaket	☐	☐	☐	Er målet oppnådd	Ja	Nei	Delvis
					☐	☐	☐

ADL-TRENING

Har barnet hatt ADL-trening

Ja Nei
☐ ☐

Type ADL-trening:

- ☐ Spise/ drikke-situasjonen
☐ Toalett-situasjonen/ personlig hygiene
☐ Av- og påkledning
☐ Leke eller skolesituasjoner
☐ Tilrettelegging/veiledning av ADL-funksjon

Antall ganger i uken				Uker, tidsperiode			
<1	1-2	3-5	6-7	<2	2-6	7-12	>12
☐	☐	☐	☐	☐	☐	☐	☐
☐	☐	☐	☐	☐	☐	☐	☐
☐	☐	☐	☐	☐	☐	☐	☐
☐	☐	☐	☐	☐	☐	☐	☐

ADL treningen er utført av:

- ☐ Ergoterapeut ☐ Fysioterapeut ☐ Foreldre ☐ Pedagog , andre

		Ja	Nei				
Er det formulert mål for tiltaket	☐	☐	☐	Er målet oppnådd	Ja	Nei	Delvis
					☐	☐	☐

ORTOSER

	Ja	Nei		Ja	Nei
Har barnet fått en håndortose	☐	☐	Bruker barnet håndortosen	☐	☐
	Ortose for å forbedre funksjon		Ortose for passiv tøyning		
	Hø	Ve		Hø	Ve
Albu	☐	☐	Albu	☐	☐
Underarm	☐	☐	Underarm	☐	☐
Håndledd	☐	☐	Håndledd	☐	☐
Tommel	☐	☐	Tommel	☐	☐
Fingre	☐	☐	Fingre	☐	☐
Funksjonsortosens materiale er	☐ Rigid	☐ Fleksibelt	☐ Lycra		
Tøyningsortosens materiale er	☐ Rigid	☐ Fleksibelt	☐ Lycra		
Anvendingstid tøyningsortose	☐ ≤ 6 t/døgn	☐ ≥ 6 t/døgn			
Har ortosen ønsket effekt	☐ Ja	☐ Nei			

Kirurgi og spastisitetsreduserende behandling

Har barnet siden forrige innsendte protokoll (vurdering) gjennomgått:

Håndkirurgisk behandling Ja ☐ Nei ☐ Dato: _____

Trening etter håndkirurgi Ja ☐ Nei ☐

Har barnet siden forrige innsendte protokoll (vurdering) gjennomgått:

Botulinum toxin injeksjon BoNT-A Ja ☐ Nei ☐ Dato: _____

Trening i forbindelse med BoNT-A Ja ☐ Nei ☐

Injeksjon er satt i:

- ☐ Skuldermuskulatur
- ☐ Albumuskulatur
- ☐ Pronatormuskler
- ☐ Håndleddsmuskler
- ☐ Fingerfleksorer
- ☐ Tommelmuskler

Konklusjon

15.03.2013

Rehabilitation and habilitation trajectories, intervention and services for preschool children with cerebral palsy (CPHAB)

This is a request for you to participate in the thematic research registry *Rehabilitation and habilitation trajectories, intervention and services for preschool children with cerebral palsy (CPHAB)*. This inquiry goes to all children registered in the Cerebral Palsy Registry of Norway (CPRN) and the national motor follow-up programme for Cerebral Palsy, CPOP. CPHAB is included as one out of five model projects in the newly created Research Centre for Habilitation and Rehabilitation Models & Services (CHARM), funded by the National Research Council (NRC) and located at the University of Oslo, Institute for Health and Society. CPHAB includes all children below the age of three years, registered in the CPOP during 2012, 2013 and 2014.

The key goals of the CPHAB are to:

- offer children and parents a systematic assessment during preschool age, also pertaining to areas other than physical functioning
- acquire knowledge regarding how parents experience the services and interventions directed towards the child and its family
- acquire knowledge regarding the influence of the services and interventions on children's development of skills and participation in everyday activities, and for parents' perceptions of their situation
- contribute to the further development of the CPRN and the CPOP, hence also assure the quality of the follow-up of children with CP

Participation in the CPHAB includes assessment of the child twice a year for a maximum of six times (over a period of three years). Beyond the CPOP registrations, the child's health and quality of life, coping, functional skills and participation in everyday activities will be assessed following an established protocol. Parents will be interviewed and requested to complete questionnaires together with project co-workers at the habilitation centers, in addition to separate questionnaires regarding the family situation, parents' quality of life and services and interventions in which the child and family participates. The entire assessment, including CPOP registrations and questionnaires, will take approximately three hours.

How will the information about you and your child be used?

Project co-workers in the habilitation centers forward all collected information to the person responsible for the registry in CPHAB, who is also a leader for the CPOP at Oslo University Hospital (OUS), Rikshospitalet. All information will be stored in a separate database within the security systems of the OUS. The registry will be de-identified with a code list that is stored separately. Only the person responsible for the registry has access to information identifiable to the individuals. You will have the right to access the registered information on you and your child, and to correct mistakes or have data deleted - unless when data is already included in publications and scientific analyses. All data will be deleted within the year of 2030.

The collected information will be used in research. All research projects using data from the CPHAB will seek approval from the Regional Committee for Medical and Health Research Ethics (REK). If information is to be treated by other institutions, it will be monitored by the OUS.

Voluntary participation

Participation in the CPHAB is voluntary. If you do not wish to participate, you can decline without any reason. You may also withdraw from the registry at a later point in time, with no consequence for the services that your child and family receive.

If you would like to participate, please sign the consent form on the next page.

If you have questions regarding the CPHAB, please contact:

Sigrid Østensjø
Prosjekt leader
Oslo and Akershus University College (HIOA)
Cell: 48143821
E-mail: sigrid.ostensjo@hioa.no

Reidun Jahnsen
Registry responsible
Oslo University Hospital (OUS)
Cell: 95738379
E-mail: reijah@ous-hf.no

Consent for participation in the CPHAB

I consent to my child's participation in the CPHAB, and to the storing of the collected information about my child and family in a separate database at Oslo University Hospital.

Child's name _____

(Signed by project participant, date)

(Signed by project participant, date)

I confirm that I have provided information about the registry

(Signed, role in the project, date)

Region:	Saksbehandler:	Telefon:	Vår dato:	Vår referanse:
REK sør-øst	Gjøril Bergva	22845529	11.03.2013	2013/178/REK sør-øst D
			Deres dato:	Deres referanse:
			05.03.2013	

Vår referanse må oppgis ved alle henvendelser

Til: Reidun Jahnsen

2013/178 Håndfunksjon og habiliteringstjenester for barn med cerebral parese

Vi viser til tilbakemelding fra prosjektleder, mottatt 05.03.2013, i forbindelse med ovennevnte søknad. Tilbakemeldingen er behandlet av komiteens leder på delegert fullmakt.

Forskningsansvarlig: Oslo universitetssykehus

Prosjektleder: Reidun Jahnsen

Prosjektomtale

Formålet med prosjektet er å generere kunnskap om utviklingen av håndfunksjon hos barn med CP gjennom en populasjonsbasert standardisert kartlegging fra barna blir registrert i det nasjonale CP-registeret (CPRN) og CP-oppfølgings-programmet (CPOP). Målet er også å identifisere karakteristika ved habiliteringstjenestene som ytes og determinanter for utviklingen av håndfunksjon. Prosjektet er tilknyttet det tematiske forskningsregisteret, CP-habilitering, som har som formål å kartlegge livssituasjon og tjenestetilbud til barn med CP og deres familier i et bredere perspektiv enn CP-registeret og CP-oppfølgingsprogrammet. Data vil samles inn over to år, og barna vil bli undersøkt minimum 3 ganger med 6 måneders mellomrom. Kunnskapen vil ha betydning for planlegging og gjennomføring av en kunnskapsbasert og helhetlig habilitering for alle barn med CP, og det vil dermed bidra til målrettede tiltak fra tidlig alder. Helseopplysninger skal overføres til land innenfor EU/EØS.

Saksgang

Søknaden ble første gang behandlet i møtet 07.02.2013, hvor komiteen utsatte vedtak i saken. Komiteen hadde ingen innvendinger til studien som sådan, men etterlyste et eget informasjonsskriv for den aktuelle studien.

Prosjektleders tilbakemelding ble mottatt 05.03.2013.

Komiteens vurdering

I tilbakemeldingen gjør prosjektleder rede for at det ikke skal samles inn andre opplysninger utover det som allerede samles inn i CPHAB-registeret. For CPHAB-registeret foreligger det samtykke hvor det fremgår at innsamlede data vil bli brukt til forskning.

Basert på tilbakemeldingen fra prosjektleder, slutter komiteen seg til at det foreliggende samtykke er dekkende for den aktuelle studien av håndfunksjon. Det er ikke nødvendig å innehente nytt samtykke.

Vedtak

Med hjemmel i helseforskningsloven § 9 jf. 33 godkjenner komiteen at prosjektet gjennomføres.

Godkjenningen er gitt under forutsetning av at prosjektet gjennomføres slik det er beskrevet i søknad, protokoll, tilbakemelding fra prosjektleder, og de bestemmelser som følger av helseforskningsloven med forskrifter.

Tillatelsen gjelder til 16.06.2016. Av dokumentasjonshensyn skal opplysningene likevel bevares inntil 16.06.2021. Opplysningene skal lagres avidentifisert, dvs. atskilt i en nøkkel- og en opplysningsfil. Opplysningene skal deretter slettes eller anonymiseres, senest innen et halvt år fra denne dato.

Forskningsprosjektets data skal oppbevares forsvarlig, se personopplysningsforskriften kapittel 2, og Helsedirektoratets veileder for «Personvern og informasjonssikkerhet i forskningsprosjekter innenfor helse og omsorgssektoren».

Dersom det skal gjøres vesentlige endringer i prosjektet i forhold til de opplysninger som er gitt i søknaden, må prosjektleder sende endringsmelding til REK.

Prosjektet skal sende sluttmelding på eget skjema, senest et halvt år etter prosjektslutt.

Du kan klage på komiteens vedtak, jf. forvaltningslovens § 28 flg. Klagen sendes til REK sør-øst D. Klagefristen er tre uker fra du mottar dette brevet. Dersom vedtaket opprettholdes av REK sør-øst D, sendes klagen videre til Den nasjonale forskningsetiske komité for medisin og helsefag for endelig vurdering.

Vi ber om at alle henvendelser sendes inn på korrekt skjema via vår saksportal: <http://helseforskning.etikkom.no>. Dersom det ikke finnes passende skjema kan henvendelsen rettes på e-post til: post@helseforskning.etikkom.no.

Vennligst oppgi vårt referansenummer i korrespondansen.

Med vennlig hilsen

Stein A. Evensen
Professor dr. med.
Leder

Gjøril Bergva
Rådgiver

Kopi til: terje.rootwelt@ous-hf.no; oushfdlgodkjenning@ous-hf.no

Oslo universitetssykehus HF

Postadresse:
Trondheimsveien 235
0514 OsloSentralbord:
02770Org.nr:
NO 993 467 049 MVA

www.oslo-universitetssykehus.no

PERSONVERNOMBUDETS TILRÅDING

Til: Reidun Jahnsen, registeransvarlig
Sigrid Østensjø, prosjektleder

Kopi: Avdelingsleder
Forskningsleder i klinikk

Fra: Personvernombudet ved Oslo universitetssykehus

Saksbehandler: Helge Grimnes

Dato: 21.03.2012

Offentlighet: Ikke unntatt offentlighet

Sak: Personvernombudets tilråding til innsamling og databehandling av personopplysninger

Saksnummer/
ePhortennummer: 2012/5429

Personvernombudets tilråding til innsamling og behandling av personopplysninger for forskningsregisteret ”Habiliteringsforløp, tiltak og tjenester for førskolebarn med cerebral parese (CPHAB)”

Viser til innsendt melding om behandling av personopplysninger / helseopplysninger. Det følgende er personvernombudets tilråding av forskningsregisteret.

Med hjemmel i Personopplysningsforskriftens § 7-12 jf. Helseregisterlovens § 36 har Datatilsynet, ved oppnevning av personvernombud ved Oslo Universitetssykehus (OUS), fritatt sykehuset fra meldeplikten til Datatilsynet. Behandling og utlevering av person-/helseopplysninger meldes derfor til sykehusets personvernombud.

Databehandlingen tilfredsstiller forutsetningene for melding gitt i personopplysningsforskriften § 7-27 og er derfor unntatt konsesjon.

Personvernombudet tilrår at forskningsregisteret gjennomføres under forutsetning av følgende:

1. Databehandlingsansvarlig er Oslo universitetssykehus HF ved adm. dir.
2. Behandling av personopplysningene / helseopplysninger i forskningsregisteret skjer i samsvar med og innenfor det formål som er oppgitt i meldingen.
3. Data lagres som oppgitt i meldingen (vedlagt). Annen lagringsform forutsetter gjennomføring av en risikovurdering som må godkjennes av personvernombudet ved OUS.
4. Vedlagte samtykke benyttes, inklusive markerte tillegg og endringer foretatt av personvernombudet. Eventuelle fremtidige endringer som berører formålet, utvalget inkluderte eller databehandlingen må forevises personvernombudet før de tas i bruk.
5. Det innhentes nytt samtykke fra de inkluderte i registeret ved fylte 16 år.
6. Inkluderte i registeret må jevnlig informeres skriftlig om hvilke delstudier registeropplysningene benyttes i, minimum en gang pr. år.

7. Kryssliste som kobler aidentifiserte data med personopplysninger lagres som angitt i meldingen og oppbevares separat på forskningsregisterleders avlåste kontor.
8. Hver delstudie som benytter opplysninger i registeret håndteres som følger:
 - a. Innhenter REK-godkjenning, med OUS som forskningsansvarlig.
 - b. Meldes til personvernombudet på vedlagte meldeskjema og avventer personvernombudets tilråding. Dette gjelder uavhengig av om REK skal godkjenne prosjektet eller ikke.
 - c. Meldes internt ihht. gjeldende rutiner for internkontroll ved OUS.
 - d. Delstudiers bruk av opplysninger i registret behandles som angitt i meldingen.
 - e. Hver delstudie godkjennes av styringsgruppen for registeret.
 - f. Opplysninger utleveres ikke til studier med annen forsknings- og databehandlingsansvarlig enn OUS.
9. Kobling mot CPRN-registeret ved Sykehuset i Vestfold skjer i samsvar med føringer fra CPRNs styringsgruppe og ihht. CPRNs konsesjonsvilkår. Overføring av opplysninger fra CPRN forutsettes å følge gjeldende retningslinjer for håndtering av sensitive personopplysninger ved OUS, jfr. styrende dokumenter i eHåndbok.
10. Dersom formålet eller databehandlingen endres må personvernombudet informeres.
11. Dersom antall inkludert i registeret vesentlig overstiger det forhåndsmeldte antallet, må personvernombudet informeres.
12. Dersom forskningsregisteret endres i forhold til det som avdelingsleder og forskningsleder har godkjent, må ny godkjenning innhentes.
13. Kontaktperson for forskningsregisteret skal hvert tredje år sende personvernombudet ny melding som bekrefter at databehandlingen skjer i overensstemmelse med opprinnelig formål og helseregisterlovens regler.
14. Data slettes eller anonymiseres ved forskningsregisterslutt **31.12.2030** ved at krysslisten slettes og eventuelle andre identifikasjonsmuligheter i registeret fjernes. Når formålet med registeret er oppfylt sendes melding om bekreftet sletting til personvernombudet.

Forskningsregisteret er registrert i sykehusets offentlig tilgjengelig database over forsknings- og kvalitetsstudier.

Lykke til!

Med vennlig hilsen
for Personvernombudet

Helge Grimnes
Personvernrådgiver
Oslo universitetssykehus HF
Stab fag & pasientsikkerhet
Seksjon for personvern og informasjonssikkerhet

Epost: personvern@oslo-universitetssykehus.no
Web: www.oslo-universitetssykehus.no/personvern

Oslo universitetssykehus HF

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Vår ref./dir.tlf.: Reidun Jahnsen/23070207

Dato: 21.01.2013

Søknad om tilgang data fra CPRN og CPOP-databasen

Viser til søknaden om tilgang til data fra CPRN og CPOP i forbindelse med studien

Hand function and habilitation services in preschool children with cerebral palsy – a population based study

Studien har vært forelagt publikasjonsutvalget for CPRN/CPOP som består av professor Gunnar Hägglund, Universitetet i Lund, professor Lorentz Irgens, Medisinsk fødselsregister, professor Torstein Vik og professor Jon Skranes, NTNU, førsteamanuensis Marie Berg, Sunnaas sykehus/Høgskolen i Oslo og Akershus, professor Irene Elgen UiB og undertegnede, som denne gangen var inhabil og derfor ikke deltok i vurderingen. Utvalget for øvrig uttrykker enstemmig at prosjektet er viktig og at det bør få tilgang til de ønskede data.

Tilråding fra Personvernombud og REK må framlegges, eventuelt dokumentasjon for at søknad til REK ikke er nødvendig.

Publikasjonsutvalget ønsker lykke til med prosjektet!

Vennlig hilsen for publikasjonsutvalget

Reidun Jahnsen
Leder CPOP