Effective use of the assisting hand in adolescents with cerebral palsy
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General Introduction
Cerebral palsy (CP) is a common cause of physical disability in early childhood, with a prevalence estimate of between 2 and 3 per 1000 live births. The often impaired upper limb function of children with unilateral CP is the main motor impairment that limits children’s ability to perform daily activities and restricts participation in the home, school, and community. Many children with CP regularly receive at least one form of treatment (e.g. physical or occupational therapy), with a view to improving hand use and the performance of activities. Compelling evidence from systematic reviews indicates that various interventions improve function at the activity level of the International Classification of Functioning, Disability and Health (ICF) in children with CP. In contrast, there is little information about the effect of upper extremity interventions in adolescents with CP or about the effect of hand orthoses and upper extremity surgery on hand use and performance in children or adolescents. One reason for this paucity of information is the lack of valid tools to assess arm-hand performance.

The aims of the studies of this thesis are to fill gaps in our knowledge by: (i) developing a tool to assess arm-hand performance in adolescents with CP; and (ii) evaluating the effect of two interventions (functional hand orthosis and upper extremity surgery) on hand use and performance of activities in children and adolescents with CP.

Cerebral palsy and the ability to handle objects

CP describes a group of permanent neurological disorders of the development of movement and posture that limit activity and which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The clinical manifestations of CP may change with time and as a result of development, learning, activities, therapies, and ageing. The severity of the disorder can be classified using the (i) Gross Motor Function Classification System (GMFCS) to describe patterns of gross motor severity in CP, (ii) Manual Ability Classification System (MACS) to describe how children with CP use their hands to handle objects in daily activities, and (iii) Communication Function Classification System (CFCS) to classify everyday communication.

Spastic CP is the most common type of CP and represents 70-80% of cases. Spasticity is a disorder of muscle control that is characterized by tight or stiff muscles and an inability to control these muscles and is associated with slow, effortful movement. The neurological dysfunction (muscle coordination, spasticity) and upper limb musculoskeletal impairments (contractures, hand posture) have a significant impact on the ability of children and adolescents with CP to use their hands to perform daily activities. Spastic CP reduces the ability to grasp and release objects and to use both hands together, which in turn adversely affects the ability of children to attain age appropriate independence and develop the autonomy and skills required to participate in important activities in the home, school, and community. The severity of the upper limb deformity is strongly correlated with the performance of activities.

Although CP is a life-long condition, some of the signs (for example, difficulties with maintaining posture and balance, poor coordination, and difficulties with fine motor control) may improve or worsen with time, as a result of development, therapies, and ageing.
Cerebral palsy in childhood and adolescence

Once children with CP leave primary school there is often a shift from direct one-to-one therapy to a more consultative adaptive approach. Studies show that adolescents’ participation in school, social, and recreational activities becomes more restricted. During the transition from childhood to adolescence, children with CP may experience many changes, such as (i) a growth spurt, which may increase muscle shortening or joint contractures and further limit activities and participation; (ii) an urge to become independent, which may create awareness of their problems in performing daily activities and the need to gain self-initiated goals; and (iii) their perceived competence in functioning, which may lead to a change in self-esteem, participation in leisure, school, and work.

With these changes in mind, it is important that professionals offer age-appropriate therapies to both children and adolescents, matching the self-initiated goals to improve hand use and performance. An intermittent consultative therapy and intervention may help the adolescent to optimize the performance of tasks relevant to that individual, thereby promoting autonomy, independent living, and employment.

CP instruments

Instruments measuring hand use and performance of activities

Children with CP

In order to evaluate the effect of different interventions, there is a need for multi-dimensional outcomes that involve the ICF levels “body function and structure” and “activity and participation”. Assessment on all ICF levels is needed to recognize the impact of CP on an individual's function and capability to engage fully in their lives. Most instruments commonly used to assess patients with CP focus on one ICF level. But a single level does not encompass that person’s functioning, which makes it necessary to use several instruments to cover all three ICF levels. According to the ICF, activity and participation can be subdivided into capacity (i.e. the highest possible level of functioning in a standardized controlled environment) and performance (i.e. the objectively detectable level of functioning in daily life). Both aspects are important to get a full picture of the possibilities and limitations to using the hand(s) while performing tasks. Previous studies suggest that in order to positively influence participation, interventions should focus on what a child actually does (performance) in day-to-day life regardless of what that child is capable of doing in a structured testing environment/clinic (capacity).

Clinical observation-based measures, such as the Quality of Upper Extremity Skills Test (Quest) and Melbourne Assessment capture the child’s upper limb capacity in a clinical environment or the child’s ability to use his/her upper limb. The Assisting Hand Assessment (AHA) is a clinical instrument to assess a child’s spontaneous use of the affected arm during play. A systematic review performed in 2012 concluded that the AHA is the only tool to measure bimanual performance using a standardized assessment of a spontaneous play session, and that it has sound psychometric properties in children with unilateral CP. The Kids-AHA has been validated for children with unilateral CP aged 18 months to 12 years and has proven reliable and sensitive enough to monitor changes over time.
questionnaires can be used to capture patient-reported perceived performance, the ABILHAND-Kids\textsuperscript{37} and the Children's Hand-use Experience Questionnaire (CHEQ).\textsuperscript{38} They both provide insight into children's upper limb function in daily life, especially with regard to the completion of activities of daily living. The CHEQ measures the perceived performance of bimanual activities of daily life and thus reflects children's experience of, and satisfaction with, their performance.

These instruments are only validated for children with CP. Yet it would be extremely helpful if the same scale could be used for all individuals with CP, as this would allow clinicians to monitor the development of hand use and the effectiveness of interventions at different ages over time.

**Adolescents with CP**

Several instruments can be used for adolescents with unilateral CP to assess the possibilities and limitations of using the hand(s) and how well activities are performed (Fig. 2). The Melbourne Assessment 2 and Shriners Hospital for Children Upper Extremity Evaluation (SHUEE) can be used to assess predominantly capacity, and the person-centred questionnaire ABILHAND-kids up to the age of 15 years to assess perceived performance.\textsuperscript{39-41} As yet, there is no valid and reliable tool for “actual performance” that assess (bimanual) arm-hand performance in adolescents (Fig. 1).

The AHA is a valid performance-based instrument that measures and describes how effectively children (9 months to 12 years of age) with unilateral CP use their affected hand to perform tasks requiring the use of both hands.\textsuperscript{34-36} The logical next step would be to extend the AHA for use in adolescents aged 13 to 18 years, which would provide an unique opportunity to monitor how hand use develops from childhood to young adulthood, and to evaluate the effects of an intervention, such as a hand orthosis and upper extremity surgery, at different ages, using the same scale.

**CP interventions**

**To improve hand use and performance of activities**

Different rehabilitation programmes (consultative, direct one-to-one, group- or home-based), targeting the upper extremity in individuals with CP, are used to improve hand use and the performance of patient-relevant tasks.\textsuperscript{42} The focus of interventions has shifted from the ICF\textsuperscript{4} component body functions (impairments) to the activities and participation component, in order to promote active use of the affected upper extremity. These interventions use principles of motor learning with intensive volitional practice of graded activities. Research has shown that specifically training activities of interest results in functional improvement.\textsuperscript{5} Examples of activity-based interventions are bimanual training\textsuperscript{43}, context-focused therapy\textsuperscript{44}, goal-directed training\textsuperscript{45} and constraint-induced movement therapy.\textsuperscript{46} These interventions help and teach the child / adolescent to use the affected hand as effectively as possible with or without compensation strategies.\textsuperscript{5}

There are situations in which it is not possible to start with an activity-based intervention, because the impairment itself (ICF body structures and functions) needs to be optimized first. For example, if a patient is not able to use the affected hand to fixate objects while performing bimanual tasks, it might be preferable to start with an impairment-
General introduction

A based intervention to reduce muscle spasticity and improve the passive range of motion (ROM), with a view to increasing the likelihood that the affected hand can be used more effectively. In turn, improving the underlying impairments in body functions and structures makes it more likely that the patient will use the hand during daily activities. This opens the way to introducing activity-based interventions.

**Figure 1.** Instruments to assess the level of functioning in adolescents with cerebral palsy

**Impairment-based CP interventions**

Examples of impairment-based CP interventions are botulinum toxin, hand orthoses, and upper extremity surgery. The aim of these interventions is to improve muscle balance and to create a neutral alignment of the wrist and thumb, which may improve the ability to grasp and release objects and to use both hands together. Botulinum toxin temporarily weakens the spastic muscle, whereas an orthosis and upper extremity surgery improve the ROM and change the alignment of the thumb and wrist joint, creating a more functional position of the hand. Upper extremity surgery involves releasing or lengthening spastic muscles, tendon transfer, and joint stabilization procedures. The combination of activity-based and impairment-based interventions is most likely the best approach to improve the patient’s functioning and independence in daily activities; for example, the combination
botulinum toxin and intensive therapy,\textsuperscript{47,48} botulinum toxin and modified constraint-induced movement therapy\textsuperscript{49}, botulinum toxin and resistance training\textsuperscript{50}, or botulinum toxin and bimanual task-oriented therapy.\textsuperscript{51} Botulinum toxin in combination with occupational therapy has been shown to improve hand function and the performance of functional hand activities.\textsuperscript{52-55}

Unfortunately, there is little evidence to support the use of a hand orthosis or upper extremity surgery in terms of improving hand use and performance of activities (Fig. 2), although there is promising supportive evidence.\textsuperscript{5,56} For this reason, more high-quality observational and comparative studies are needed to guide intervention planning and to enable therapists and patients make an objective decision about whether to proceed with one of these CP interventions.

\textbf{Figure 2.} Effective interventions (Novak 2014)\textsuperscript{56}
Functional hand orthosis

Hand orthoses, also known as braces or upper limb splints, are removable external devices designed to support a weak or ineffective joint or muscle. In children and adolescents with CP, a variety of orthoses made of various materials are used in clinical practice with two different purposes. A non-functional hand orthosis is used to reduce contractures or to improve muscle length during the night, so that the wrist achieves a more neutral position. This may improve the appearance of the hand during the day and make it possible to use the hand (for example, washing hands with more open hand or putting the arm through the sleeve). In contrast, a functional hand orthosis is worn during the day and helps promote an optimal upper limb position for performing activities. This leads to improvement in the activity and participation domains of the ICF, such holding and stabilizing objects while performing activities (Fig. 3). One study reported that functional hand orthosis dynamic activation at the wrist and increased compensatory shoulder muscle recruitment. Because of the lack of compelling evidence from different high-quality studies in this field, the effect of a functional hand orthosis on hand use and performance of activities is unclear.

Figure 3. Functional hand orthosis

Upper extremity surgery

The aim of upper extremity surgery is to facilitate the ability to grasp, release, and handle objects by: (a) weakening overactive spastic muscles, (b) strengthening weak muscles, and (c) stabilizing unstable joints. Common procedures include the release or transfer of the flexor carpi ulnaris tendon to increase wrist extension for functional grip purposes and correction of a thumb-in-palm deformity, preferably and if possible by adductor pollicis muscle slide combined with extensor pollicis longus rerouting.

There have been few studies of the effect of upper extremity surgery on hand use and performance. Of the available studies, most assessed the outcomes of upper extremity surgery using a functional classification scale (for example, the “House functional classification” and/or outcomes describing body functions and structures, such as wrist/thumb positioning, muscle strength, ROM, and selective motor control.

While adolescents constitute the largest group of patients who undergo upper extremity surgery, there is currently no instrument specifically designed to assess how adolescents use the affected hand in bimanual activities. This might be one of the reasons why little is known about the effect of upper extremity surgery on hand use and performance.
Aims of this thesis

The general aims of the studies described in this thesis are to develop and evaluate the validity and reliability of the AHA for adolescents with unilateral CP and to evaluate the effect of two different interventions, functional hand orthosis and upper extremity surgery, on hand use and daily performance in children and adolescents with CP.

Outline of this thesis

Part I:  Development of the Assisting Hand Assessment for Adolescents

Chapter 2 describes the development of a test to allow the observation of bimanual performance in adolescents with unilateral CP, the Assisting Hand Assessment for Adolescents (Ad-AHA). The validity of this test was evaluated first, and subsequently the construct validity of the test was evaluated in order to establish whether the Kids-AHA scoring criteria can be used for children/adolescents aged 18 months to 18 years. The study presented in Chapter 3 reports the different aspects of the reliability of the Ad-AHA for adolescents with unilateral CP, namely, (1) inter-rater, test–retest reliability, and the smallest detectable change; (2) agreement between performance scores on the Ad-AHA Board Game and School-Kids AHA (age group 10–13 years); and (3) agreement between performance scores on the Ad-AHA Board Game and Ad-AHA Present, and between the Ad-AHA Board Game and Ad-AHA Sandwich (age group 13–18 years).

Part II:  Effect of interventions on hand use and daily performance

The study described in Chapter 4 reports the immediate effects of a static wrist and thumb brace on the spontaneous use of the affected upper limb to perform bimanual activities in children with unilateral CP. The systematic review reported in Chapter 5 evidence on the effectiveness of upper extremity surgery on ICF activity outcomes in children and adolescents (aged < 20 years) with CP. The study presented in Chapter 6 describes the effects of upper extremity surgery on manual performance and patient-relevant outcomes in a consecutive series of children and adolescents with unilateral CP who were selected based on a multidisciplinary assessment and shared decision-making. Lastly, Chapter 7 discusses the main findings of the thesis in a broader perspective in relation to clinical practice and research, and reflects on methodological considerations. Suggestions for future research and clinical practice are given.
References


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