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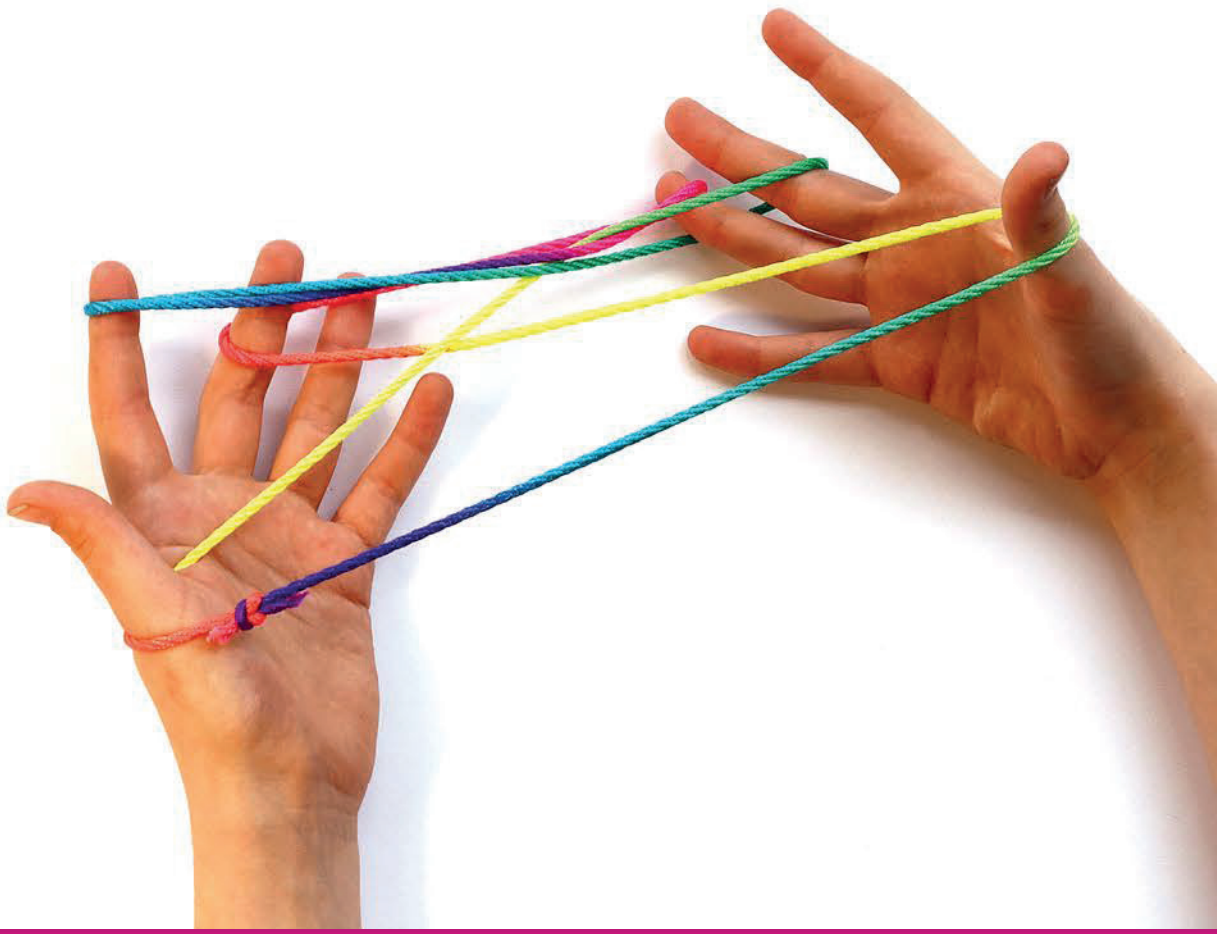

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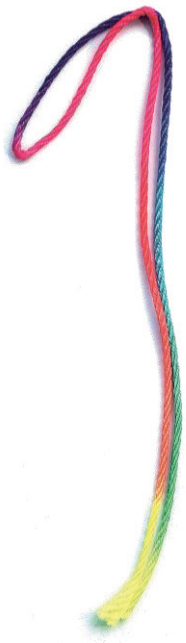
Contents



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.[1] 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.[2] Many children with CP regularly receive at least one form of treatment (e.g. physical or occupational therapy),[3] 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.[4] 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.[5] 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.[2] The clinical manifestations of CP may change with time and as a result of development, learning, activities, therapies, and ageing.[2] The severity of the disorder can be classified using the (i) Gross Motor Function Classification System (GMFCS)[6,7] to describe patterns of gross motor severity in CP, (ii) Manual Ability Classification System (MACS)[8] to describe how children with CP use their hands to handle objects in daily activities, and (iii) Communication Function Classification System (CFCS)[9] to classify everyday communication.

Spastic CP is the most common type of CP and represents 70-80% of cases.[10,11] 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.[2] Spastic CP reduces the ability to grasp and release objects and to use both hands together,[12,13] 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.[14] The severity of the upper limb deformity is strongly correlated with the performance of activities.[15]

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.[2]

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.[16,17] 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;[18,19] (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;[20,21] and (iii) their perceived competence in functioning, which may lead to a change in self-esteem, participation in leisure, school, and work.[16,22-25]

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.[26,27]

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.[4,28] 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).[4] Both aspects are important to get a full picture of the possibilities and limitations to using the hand(s) while performing tasks.[29] 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).[30]

Clinical observation-based measures, such as the Quality of Upper Extremity Skills Test (Quest)[31] and Melbourne Assessment 2,[32] 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[33] 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.[34-36] 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.[34-36] Two

questionnaires can be used to capture patient-reported perceived performance, the ABILHAND-Kids[37] and the Children's Hand-use Experience Questionnaire (CHEQ).[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.[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.[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.[42] The focus of interventions has shifted from the ICF[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.[5] Examples of activity-based interventions are bimanual training[43], context-focused therapy[44], goal-directed training,[45] and constraint-induced movement therapy.[46] These interventions help and teach the child / adolescent to use the affected hand as effectively as possible with or without compensation strategies.[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-

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.

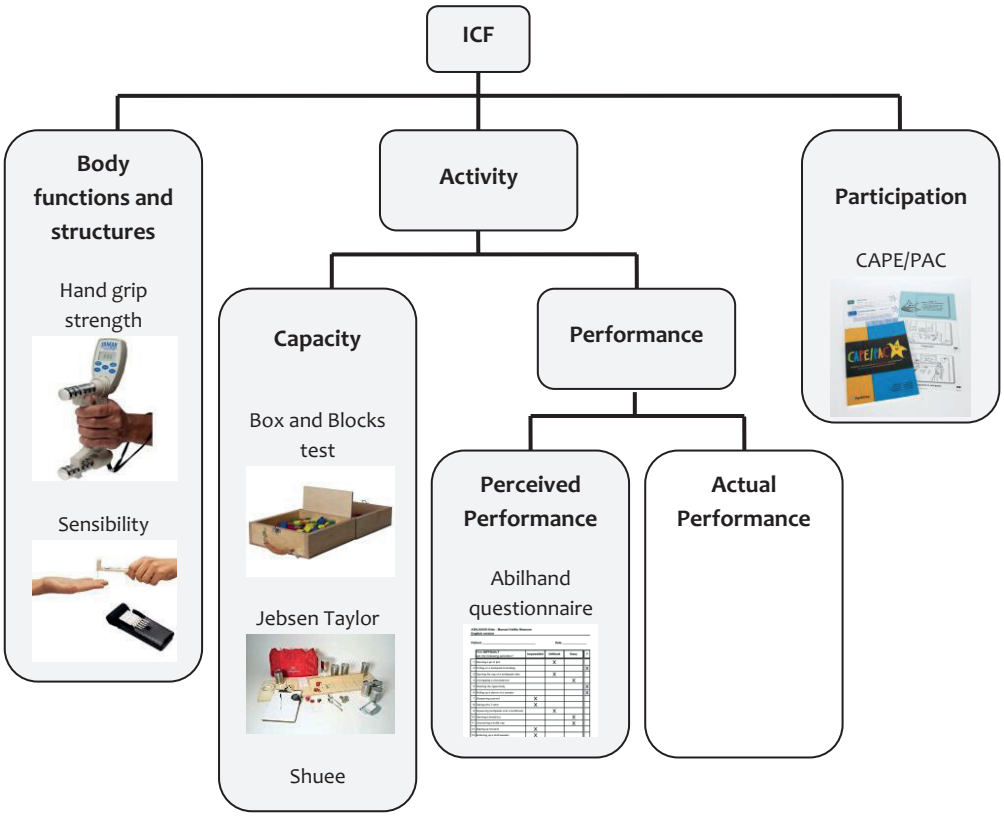


Figure 1. Instruments to assess the level of functioning in adolescents with cerebral palsy

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,[47,48] botulinum toxin and modified constraint-induced movement therapy[49], botulinum toxin and resistance training[50], or botulinum toxin and bimanual task-oriented therapy.[51] Botulinum toxin in combination with occupational therapy has been shown to improve hand function and the performance of functional hand activities.[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.[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.

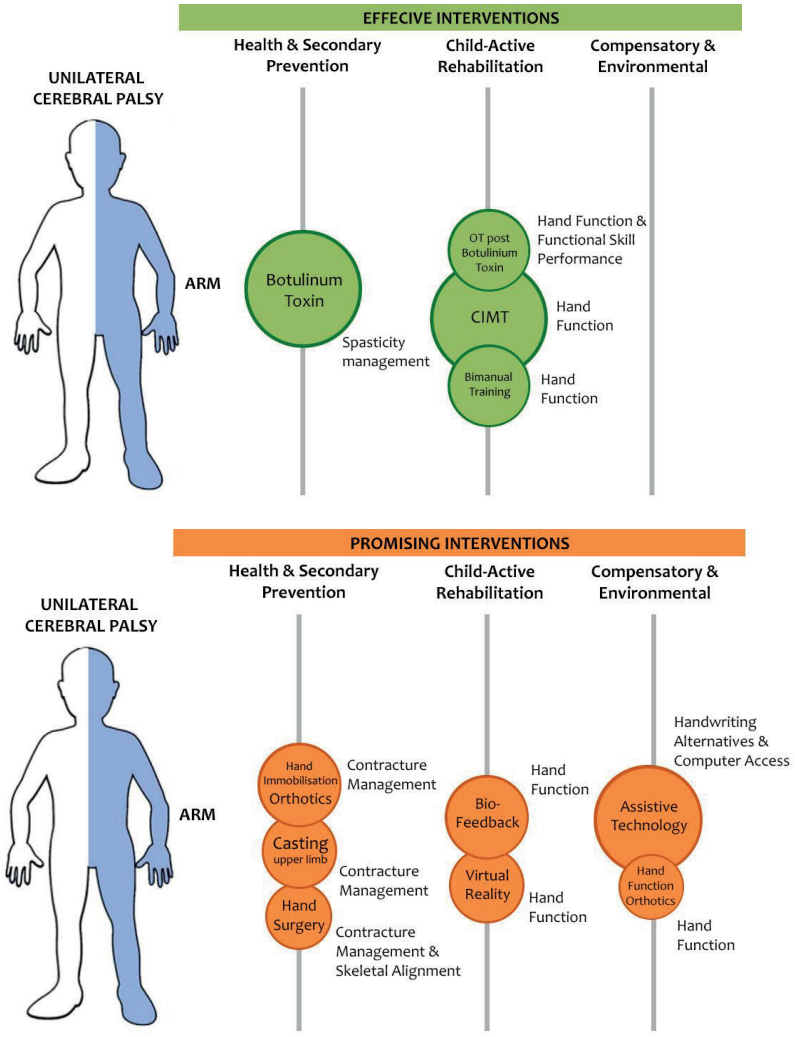


Figure 2. Effective interventions (Novak 2014)[56]

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.[57] 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.[58] 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.

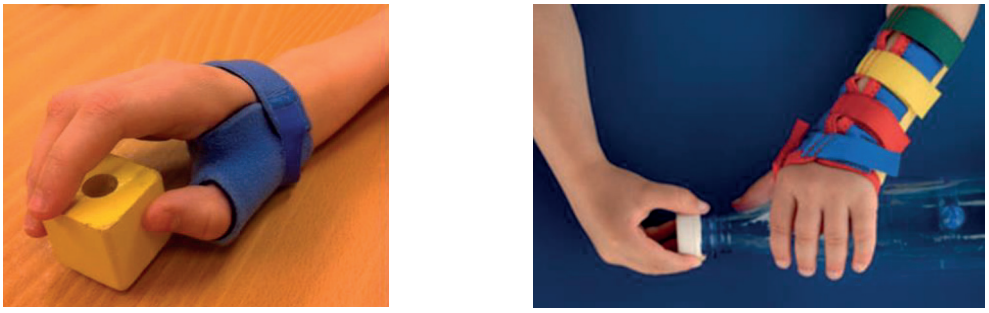


Figure 3. Functional hand orthosis

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[59] and correction of a thumb-in-palm deformity, preferably and if possible by adductor pollicis muscle slide[60] combined with extensor pollicis longus rerouting.[61]

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")[62,63] and/or outcomes describing body functions and structures, such as wrist/thumb positioning, muscle strength, ROM, and selective motor control.[64-67]

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.

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Development of the Assisting Hand Assessment for adolescents (Ad-AHA) and validation of the AHA from 18 months to 18 years

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Abstract

Aim

To develop and evaluate a test activity from which bimanual performance in adolescents with unilateral cerebral palsy (CP) can be observed and scored with the Assisting Hand Assessment (AHA), and to evaluate the construct validity of the AHA test items for the extended age range 18 months to 18 years.

Method

A new test activity was developed and evaluated for its ability to elicit bimanual actions in adolescents with (n=20) and without (n=10) unilateral CP. The AHA scores of 126 adolescents (mean age 14y 3mo, SD 2y 6mo; 71 males, 55 females) and 157 children with unilateral CP (mean age 6y 1mo, SD 2y 10mo; 102 males, 55 females) were analysed using the Rasch measurement model.

Results

The test activity elicited bimanual actions in 100% of typically developing adolescents and in 96.8% and 57.9% of adolescents with unilateral CP (moderately and severely limited hand function respectively). The scale demonstrated good construct validity; thus the same scoring criteria can be used for the age range studied.

Interpretation

The new Assisting Hand Assessment for adolescents (Ad-AHA) activity is valid for use with 13- to 18-year-olds to elicit bimanual performance in adolescents with unilateral CP. The same AHA scoring criteria can be used both for children and for adolescents within the age range 18 months to 18 years.

Introduction

Unilateral cerebral palsy (CP) is characterized by motor impairments on one side of the body, and the hand impairments in particular contribute to functional limitations. Current rehabilitation efforts predominantly aim to improve bimanual performance, but this requires valid and reliable performance-based instruments for evaluating the hand function of people of different ages with unilateral CP.

The Assisting Hand Assessment (AHA), which was introduced in 2003, has proved to be a valid performance-based instrument for children with CP. The AHA measures and describes how effectively children with unilateral disability use their affected hand to perform tasks requiring the use of both hands. This is in contrast to other tests in which people are asked to specifically use the affected hand, so that their best performance with that hand can be assessed. The AHA has been validated for children with unilateral CP aged 18 months to 12 years (Kids-AHA) and is sufficiently sensitive to monitor changes over time.[1–4] The AHA is often used to monitor the effect of interventions such as modified constraint-induced movement therapy,[4,5] botulinum neurotoxin injections,[6] hand and arm bimanual intensive training,[7] hand orthosis,[8] and hand surgery.[9]

There is currently no instrument specifically designed to assess how adolescents use the affected hand in bimanual activities. The Melbourne Assessment 2 and Shriners Hospital for Children Upper Extremity Evaluation (SHUEE) predominantly assess capacity, and the person-centered questionnaire ABILHAND-kids assesses perceived performance.[10–12] Therefore there is no test of bimanual hand use that can be used with both children and adolescents. Such an instrument would provide a unique opportunity to monitor development of hand use from childhood to young adulthood, and to evaluate effects of intervention at different age groups using the same scale. The Kids-AHA has proved its merits both in clinical practice and in research involving children with unilateral CP; the logical next step would be to expand the AHA for use in adolescents aged 13 to 18 years. Because the Kids-AHA play session is not appealing to adolescents – due to its somewhat 'childish' game objective, instructions, and objects used – a new test activity needs to be developed, involving a sequence of different bimanual actions that form a recognizable activity, to be able to observe and rate object-related actions of the affected hand. It is important that the test activity is appealing so that participants become engaged in the activity and will naturally adopt their habitual performance.

The aims of this study were (1) to develop a test activity from which bimanual performance in adolescents with unilateral CP can be observed, and to evaluate the validity of the content of this new activity, and (2) to determine whether the Kids-AHA scoring criteria can be used for children/adolescents aged 18 months to 18 years, by evaluating the construct validity of the AHA over this age range.

Methods

The study consisted of two phases: (1) develop a test activity for adolescents and evaluating the validity of the content of this new activity; (2) assess the construct validity of the AHA test items for the age range 18 months to 18 years.

Development of the new test activity

On the basis of experience gained with the successful use of the Kids-AHA play session and board games, as well as the toys and play session of the Mini-AHA for children 8 to 18 months,[13] we considered that the new activity should meet the following requirements. It should: (1) involve a sequence of bimanual actions or tasks that form a meaningful activity; (2) be semi-structured allowing therapist interaction to help tasks proceed if needed and to make it fun and engaging to elicit typical/habitual performance; (3) include objects that require bimanual handling and are age appropriate and familiar to most adolescents; (4) allow the observation of different aspects and levels of ability of bimanual performance; and (5) enable scoring of the AHA test items. For clinical practice, the activity should take approximately 15 minutes to complete and allow the same standardized video recording settings as the Kids-AHA activities, thereby allowing assessment in different settings.

Participants and recruitment

To evaluate the content of the new activity, two groups of adolescents were recruited (phase I). One group consisted of 10 typically developing adolescents, who were recruited through colleagues and friends. The other group consisted of 20 adolescents with unilateral CP who were recruited from the Department of Rehabilitation, Academic Medical Centre, Amsterdam, the Netherlands. The 20 adolescents with unilateral CP included 10 adolescents with moderately limited hand function who could use their affected hand for grasping and holding objects, but without individual finger movements, and 10 adolescents with severely limited hand function, who could not grasp or hold objects voluntarily with their affected hand.[14]

To evaluate the scale construct of the AHA for the age range 18 months to 18 years (phase II), a convenience sample of 126 participants (71 males, 55 females) aged 10 to 18 years was recruited from outpatient rehabilitation clinics and special schools in the Netherlands, Australia, and Sweden. Inclusion criteria were children and adolescents diagnosed with unilateral CP, with any severity level of affected hand function. Data for the adolescent assessments were added to those for the 164 assessments of 157 children (102 males, 55 females) who participated in the validation of the Kids-AHA version 5.0.[15] Demographic characteristics are presented in Table I.

Table I. Demographic characteristics

	n	Mean age, y:mo (SD)	Sex, male	Affected side of body, right
Phase I				
Typically developing adolescents	10	15:7 (2:4)	3	
Adolescent with unilateral CP, moderately limited hand function	10	16:9 (2:2)	4	7
Adolescent with unilateral CP, severely limited hand function	10	13:8 (3:4)	7	7
Phase II				
Children with unilateral CP	164	6:1 (2:10)	102	91
Adolescents with unilateral CP	126	14:3 (2:6)	71	56

SD, standard deviation.

Procedure and statistical analysis

To evaluate whether the test activity prompted the use of both hands (phase I), adolescents with and without unilateral CP played the new board game. Each session was videotaped and the use of both hands was scored as does or does not for the 19 subtasks of the game. The hypothesis was that typically developing adolescents would perform all actions bimanually. Adolescents with unilateral CP who could not grasp or hold objects with their affected hand were expected to perform the fewest bimanual actions. Descriptive data were analysed using Statistical Package for the Social Sciences (SPSS version 21.0; SPSS, Inc., Chicago, IL, USA).

To evaluate the construct validity of the AHA test items for the age range 18 months to 18 years (phase II), all 126 assessments of adolescents were scored using the Kids-AHA version 5.0 scoring criteria containing 20 items, each scored on a 4-point rating scale.[15] The adolescents and a combined sample of adolescents and children (n=290 assessments) were analysed with the Rasch model analysis using Winsteps version 3.80.1 software (Linacre JM, Beaverton, OR, USA).[16] Since all AHA test items have four response options, the polytomous rating scale Rasch model was used. A two-faceted (item and person) Rasch partial credit model was chosen because the metric distance between the thresholds separating rating scale categories are not the same across all 20 AHA items.

A stepwise procedure was used to investigate the construct validity of the AHA scale for the age range 18 months to 18 years, starting with investigating any existence of disordered thresholds between categories, which would indicate a malfunctioning rating scale.[16,17] Then item and person fit to the Rasch model assertions was evaluated.[18] Criteria for item misfit and removal were infit mean-square (MnSq) ≥1.4,[19] accompanied by Zstd-value ≥2.[20] Ninety-five per cent of the items and individual data should demonstrate acceptable goodness-of-fit values. The point measure correlation reflects how well the responses represent the item ability estimate. A value of 0.6 or more represents a good correlation.[16] A fundamental requirement of the Rasch model is that the scale has to measure a single construct only. Therefore, principal component analysis of the residuals was performed, and unidimensionality was confirmed if the first dimension (the AHA measures) explained at least 60% of the total variance, and if the second largest dimension explained less than 5% of the remaining variance of residuals.[16,21,22]

The Rasch model provides an indication of the match between the item difficulty and the ability of the individuals in the sample. A well-targeted scale (not too easy, not too hard) would have a person ability mean score of about zero.[17] To further determine how sensitive the test is in identifying a person's ability, the person-separation index was investigated. A person-separation index >2 and a reliability coefficient >0.80 indicates a high person reliability and that the test can distinguish between high and low performers.[22,23] The number of different levels of ability (strata) can be calculated using the following formula: (4xperson-separation index+1)/3.[23] Three to four strata is considered good, and more than 5 is excellent.[22]

The AHA scale consists of easier and more difficult items. To assess whether the hierarchy of item difficulty remained the same (invariant) across our two age groups (children and adolescents), differential item functioning (DIF) was evaluated using the Mantel–Haenszel statistic for polytomous scales. A test item is considered to show DIF when individuals of equal ability, but from different groups, have an unequal probability of item success, which will result in a different hierarchy of items. According to our sample sizes (164 and 126 respectively), we set our criteria for a significant 'moderate DIF' absolute value of Mantel–Haenszel DIF between 1.0 and 1.5 logits and a 'large DIF' absolute value ≥1.5 logits with the X^2 test of significance at the 0.05 level.[24,25]

To evaluate whether the same AHA scale can be used for both age groups, differential test functioning was performed to determine whether any identified DIF had a substantial impact. This was done by plotting the adolescents' ability measures on the basis of the item difficulty calibrations of the children against the adolescents' ability measures on the basis of their own item difficulty calibrations. The scale measures expressed in logits were transformed to a more user-friendly 0 to 100 scale (AHA-units).[26] There is no evidence of differential test functioning if the line of the paired ability measures is close to the identity line, within a 95% confidence interval.[27]

Ethics

For participating centres in the Netherlands, the Medical Ethics Committee of the Academic Medical Centre, Amsterdam, waived the need for ethical approval. For participating centres in Sweden and Australia, ethical approval for this study was granted by the respective institutional review board. All participants (>12y) and their primary caregivers gave informed consent, in compliance with research regulations and policies of the participating centres.

Results

Phase I: content validity

The new test activity for adolescents, the Ad-AHA board game 'Go with the Floe', consists of 25 specific objects that are presented in an attractive box. The objects require bimanual handling, for example reaching for and opening the game box, shuffling the cards, opening the pencil case, and cutting paper. The game is played according to rules and instructions printed on playing cards, and takes about 15 minutes. The adolescent is videotaped playing the game, to enable evaluation of their bimanual performance (while setting up, playing, and keeping count of the game). The administration procedure, therapist instructions, and interactions are similar to that of the original AHA, except that a larger table is used to allow observation of the adolescent's reaching ability.

Table II shows the number of bimanual actions performed when playing the 19 subtasks of the board game. All typically developing adolescents used both hands to perform the 19 subtasks. As expected, the proportion of the relatively able participants who used both hands was higher than that for the less able participants (96.8% vs 57.9%). The game could be played in a clinical setting, and the adolescents found it enjoyable. The average time taken to complete the test was 17 minutes (SD 3.5).

Phase II: construct validity

The AHA scale for the age range 10 to 18 years functioned as an unidimensional measure and met all criteria,[22] and therefore the combined sample of 290 assessments of adolescents and children was analysed as described below.

The rating scale functioned consistently across the 20 AHA 5.0 items and the average threshold calibrations were all ordered and increased monotonically. All items but one ('Orients objects') demonstrated acceptable goodness-of-fit to the Rasch model (95%). The point measure correlation for all 20 items was higher than 0.6 (range 0.73–0.90) (Table III), and principal component analysis revealed unidimensionality of the scale. The variance explained by the principal component was 82.1%. The unexplained variance in the first contrast was 2.0%.

Visual inspection of the person-item map (Fig. 1) indicated that AHA items were well distributed in the sample and generally appropriate for children and adolescents with unilateral CP, with no substantial gaps in person–item targeting, as confirmed by the mean ability score (0.9 logits) and the mean standard error (0.23 logits). None of the participants attained the minimum score and three participants (1%) attained the maximum score. The person separation index score was 6.30, indicating that the AHA can distinguish 8.7 strata. Thus there were nine levels of ability in this sample, showing a high person reliability (0.98).

Table II. Number of bimanual actions performed when playing the 19 sub-tasks of the board game

	Persons performing actions bimanually		
Actions required playing the board game	**Typical hand function[a] (*n*=10)**	**Moderately limited hand function[b] (*n*=10)**	**Severely limited hand function[c] (*n*=10)**
Reaching the game box	10	10	1
Opening the game box	10	10	5
Opening the game board	10	9	4
Opening plastic box	10	9	6
Tearing cello tape	10	10	6
Handling lock and key	10	10	7
Getting ice floes (game tokens) from stick	10	10	6
Handling plastic bag with marbles	10	10	5
Removing lid from bottle	10	10	6
Handling the stopwatch	10	9	6
Winding up the timer	10	10	6
Shuffling and handling the cards	10	9	6
Getting marble out of the bottle	10	10	6
Putting an ice floe back on the stick	10	10	7
Getting score form out of plastic sleeve	10	10	7
Opening the pencil case	10	10	7
Opening the marker	10	9	5
Cutting a paper	10	10	7
Handling the folder/envelop	10	9	7
Total *n* (%)	190 (100%)	184 (96.8%)	110 (57.9%)
Mean sum score AHA 5.0 (range)	80	55.70 (46–63)	31.50 (24–35)
Mean AHA-units score (range)	100	62 (49–70)	32.5 (19–37)

Ability levels were as follows; [a]Typically developing adolescents, [b]Adolescents with unilateral CP who could use their affected hand for grasping and holding objects without individual finger movements. [c]Adolescents with unilateral CP who could not grasp or hold objects with their affected hand but typically stabilizes objects by weight or support.

Table III. Item statistics for the 20 items of the Assisting Hand Assessment 18–18

	Item	Measure	SE	Infit		Outfit		Point-Measure	
				MnSq	Zstd	MnSq	Zstd	Corr.	Exp.
	Chooses assisting hand when closer to objects	4.57	0.13	1.22	2.0	1.08	0.4	0.74	0.76
↑ More difficult	Manipulates	4.41	0.16	1.02	0.2	0.79	-0.8	0.80	0.80
	Grasps	2.57	0.15	0.78	-2.1	0.73	-1.3	0.84	0.82
	Moves forearm	2.30	0.14	1.35	3.2	2.07	4.2	0.80	0.84
	Varies type of grasp	1.66	0.14	0.79	-2.4	0.66	-2.1	0.88	0.85
	Reaches	1.60	0.13	1.09	1.1	1.30	1.7	0.85	0.86
	Readjust grasp	1.05	0.13	1.13	1.4	1.02	0.2	0.86	0.87
	Grip force regulation	0.97	0.13	0.65	-4.3	0.56	-3.3	0.90	0.86
	Releases	0.79	0.13	0.83	-2.0	0.76	-1.6	0.88	0.86
	Flow in bimanual task performance	0.50	0.13	0.60	-5.3	0.53	-4.1	0.88	0.83
	Stabilises with grasp	0.42	0.13	0.72	-3.4	0.63	-2.4	0.90	0.87
	Moves fingers	0.07	0.14	1.00	0.0	0.87	-0.9	0.84	0.83
	Moves upper arm	0.03	0.15	1.14	1.4	1.24	1.1	0.77	0.78
	Coordinates	-0.85	0.14	0.85	-1.8	0.97	-0.2	0.86	0.85
	Initiates use	-1.54	0.17	1.01	0.1	0.94	-0.3	0.82	0.82
	Orients objects	-1.83	0.14	1.46	4.2	1.39	1.5	0.80	0.85
Easier ↓	Proceeds	-2.81	0.14	1.27	2.7	1.74	2.8	0.79	0.83
	Amount of use	-4.09	0.15	1,12	1.3	1.46	1.3	0.77	0.80
	Stabilises by weight or support	-4.52	0.17	1.06	0.5	0.68	-0.7	0.73	0.73
	Holds	-5.30	0.18	0.68	-2.5	0.39	-1.7	0.74	0.72
	Mean	0.00	0.15	0.99	-0.3	1.00	-0.3		

SE=standard error, MnSq=mean square, Zstd=standardized as a z-score, Corr.= correlation, Exp=Expected

Two items demonstrated a large DIF between age groups ('Orients objects', 'Moves fingers') and five a moderate DIF ('Coordinates', 'Readjusts grasp', 'Manipulates', 'Moves forearm' and 'Stabilises by weight or support'). Thus, a difference in item difficulty order was found between children and adolescents. In adolescents, four items ('Readjusts grasp', 'Orients objects', 'Manipulates', and 'Stabilises by weight or support') were easier and three items ('Coordinates', 'Moves fingers', and 'Moves forearm') were more difficult than for children with the same level of ability.

The scatter plot for differential test functioning (Fig. 2) shows how well the test functioned in adolescents and children. The similarity of the measures of ability in children and adolescents means that any significant DIF had no impact on AHA 5.0 measurements; thus the scale can be used over the entire age range 18 months to 18 years.

Measure	Person	Item Score 1-2	Item Score 2-3	Item Score 3-4
	More able persons	*More difficult items*		
10	X			
9	X			X
8	X			
	X			
7	XXXX			XX
	XX			X
6	XXXX		X	XXXXX
	XXXXX			XXX
5	XX		X	X
	XXX		X	XX
4	XXXXX			
	XXXXXX		X	
3	XXXXXXXXX			
	XXXXX		X	XX
2	XXXXXXXXX		XX	
	XXXXXX	X	XX	
1	XXXXXXX		XX	
	XXXXXXXX		X	X
0	XXXXXXXXXX		XX	
	XXXXXXX			
-1	XXXXXX			
	XXXXXXXX		X	
-2	X	X		X
	XXXX	X	XX	X
-3	XX	XX		
	X	XXXX		
-4	XXX	X		
	XXX		X	
-5	XXX		XX	
	XX	X		
-6	XX			
	X	XX		
-7		X		
	XX	X		
-8	X	XX		
		XX		
-9				
		X		
-10	X			
-11				
	Less able persons	*Easier items*		

Figure 1. Distribution of the person ability measures for children and adolescents (n=290) and item difficulty calibrations (n=20 items, and a 4-point rating scale)

Left-hand column locates the person ability measures. Right-hand column locates the item difficulty measures. The items are positioned at their threshold values between score 1 (does not do) and 2 (ineffective); score 2 and 3 (somewhat effective); and score 3 and 4 (effective).

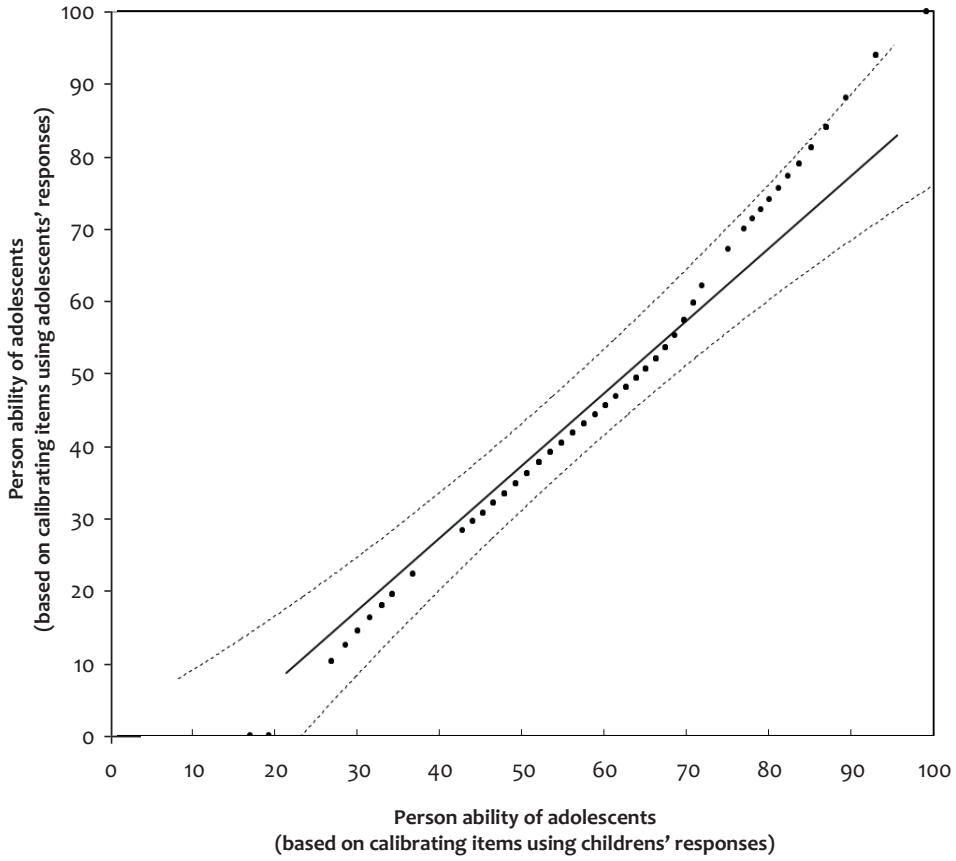

Figure 2. Differential test functioning

Discussion

This study provides evidence that the new test activity, the Ad-AHA board game, elicits the use of both hands, thus allowing observation of how adolescents actually use their affected hand to perform bimanual activities. The test activity was appropriate for 13- to 18-year-olds and the scale showed good construct validity within the whole age range of 18 months to 18 years. This means that the same AHA scoring criteria can be used both for children and for adolescents with unilateral CP.

The new board game involves a sequence of bimanual actions. Because players become engrossed in the game, it elicits spontaneous performance, as opposed to other tests that measure capacity. The therapist is allowed to interact and assist to help the tasks proceed if needed, which makes the game enjoyable for the adolescent and manageable regardless of severity of impairment.

The total sample of 290 assessments was large enough for Rasch analysis to yield reliable item calibrations and for measurements to be stable. Owing to its high person separation, the AHA scale can distinguish nine levels of effectiveness of affected hand use in children and adolescents with unilateral CP. The scale is, therefore, likely to be responsive to change. This provides strong evidence for construct validity and scale reliability. Only one item, 'Orients objects', did not demonstrate acceptable goodness-of-fit statistics and could be omitted. However, 95% of the items fitted well and this item was considered to be clinically important, providing relevant information about the ability of a person to make the overall bimanual actions smoothly and effectively. 'Orients objects' was easier to perform for adolescents than for children, as illustrated by a large DIF. This may be the reason for the misfit.

A key advantage of using Rasch analysis is the item- and person-map. It provides information about item and person hierarchy. In clinical practice, the ability of a person to use their affected hand can be matched to the difficulty of an item and can guide intervention planning. DIF analyses showed a difference in item hierarchy between children and adolescents. However, differential test functioning showed that any significant DIF had no impact on AHA 5.0 measurements. Thus the AHA scale can be used over the entire age range of 18 months to 18 years, but there is a need for two different item hierarchies, one for children and one for adolescents, when interpreting outcomes and planning treatment.

A possible limitation is that our sample of adolescents with unilateral CP might have differed from the total population of adolescents with unilateral CP. All adolescents in this study were recruited from rehabilitation departments / centres and were receiving therapy. Their hand function might have differed from adolescents with unilateral CP not receiving therapy. However, a large ability range was covered within the scale, from persons receiving a maximum score having no or minimal signs of affected hand function, to persons receiving a close to minimum score, actually not using the hand at all. Thus the scale appears to be appropriate for all ability levels.

Although this study demonstrated the validity of the new activity, the Ad-AHA board game, future research should focus on other test characteristics, such as test-retest and rater reliability. Because the activities used in the AHA depend on the age of the person performing the test, it may be necessary to determine the alternative-form reliability of different test activities (e.g. Ad-AHA board game vs School-kids AHA), especially when participants are of the transition age between two different test activities. Test–retest data

will also allow calculation of the smallest detectable change, which reflects the variation in scores due to measurement error.

The ability to use the AHA for individuals aged from 18 months to 18 years provides a unique possibility to monitor the development of hand use from toddler into adulthood, using the same scale. This would allow evaluation of the effectiveness of interventions at different ages as well as change over time. We propose that the AHA now be called the Assisting Hand Assessment 18–18 (AHA 18–18), because it includes the Kids-AHA versions (the Small-Kids AHA for 18mo–5y and the School-Kids AHA for 6–12y), as well as an adolescent version, the Ad-AHA for 13 to 18 years. The only difference between the age versions is the choice of test activity for observing bimanual performance. The use of an age-appropriate and engaging activity is essential for eliciting typical bimanual performance as opposed to unilateral capacity. The great advantage of all these versions is that the same scoring criteria can be used.[15]

Conclusions

The new test activity, the Ad-AHA board game 'Go with the Floe' has proved to be valid and can be used in clinical practice to elicit actual bimanual performance in adolescents with unilateral CP. In addition, the same AHA scoring criteria can be used both for children and for adolescents to measure how effectively the affected hand is used in bimanual performance. The AHA 18–18 covers the whole age range from 18 months to 18 years and is a valid performance-based instrument to monitor development from childhood to young adulthood, evaluate interventions, and guide intervention planning.

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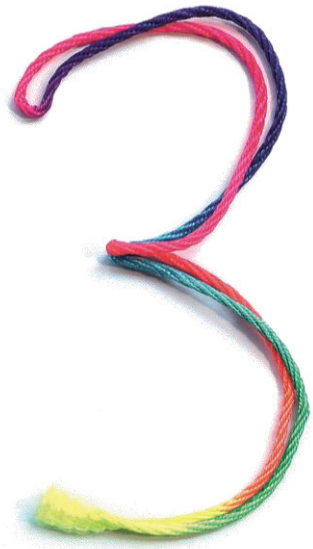

Reliability of the Assisting Hand Assessment in adolescents

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Abstract

Aim

To investigate the interrater and test–retest reliability of the Assisting Hand Assessment in adolescents (Ad-AHA) with cerebral palsy (CP) and to evaluate the alternate-form reliability of different test activities.

Method

Participants were 112 adolescents with unilateral CP (60 males, 52 females; mean age 14y 5mo [standard deviation {SD} 2y 8mo], Manual Ability Classification System levels I–III). Reliability was evaluated using intraclass correlation coefficients (ICC), smallest detectable change (SDC), and Bland–Altman plots.

Results

ICCs for interrater (n=38) and test–retest reliability (n=31) were excellent: 0.97 (95%CI 0.94–0.98) and 0.99 (95% CI 0.98–0.99) respectively. The alternate-form reliability of different test activities was excellent for children (age 10–12y, n=30) performing the School-Kids AHA and Ad-AHA Board Game 0.99 (95% CI 0.98–0.99) and for adolescents (age 13–18y) performing the Ad-AHA Board Game compared to the Ad-AHA Present (n=28) 0.99 (95% CI 0.95–0.98), or the Ad-AHA Sandwich (n=29) 0.99 (95% CI 0.98–0.99) tasks. SDC for test–retest was 4.5 AHA-units.

Interpretation

Ad-AHA scores are consistent across different raters and occasions. The good alternate-form reliability indicates that the different test activities can be used interchangeably in adolescents with unilateral CP. Differences greater than or equal to 5 AHA-units can be considered a change beyond measurement error. The use of logit based AHA-units makes change comparable for persons at different ability levels.

Introduction

The upper limb dysfunction of children and adolescents with unilateral cerebral palsy (CP) may make the performance of activities involving reaching, grasping, and manipulating objects difficult. A common goal of treatment for adolescents with unilateral CP is to improve bimanual performance, because the effective use of the affected arm and hand in combination with the well-functioning hand is important for independence. A valid and reliable tool to assess arm-hand performance is needed in order to plan interventions, determine the effectiveness of different interventions, and monitor the development of hand-use of individuals with unilateral CP.

Within the group of individuals with unilateral CP, assessments of hand-use have primarily been developed for younger children with unilateral CP. Currently only the Melbourne Assessment 2 (MA2), Shriners Hospital for Children Upper Extremity Evaluation (SHUEE), and ABILHAND-Kids can be used in adolescents. But the MA2 and SHUEE mainly assess capacity, and the person-centred questionnaire ABILHAND-Kids assesses perceived performance difficulties, which can be quite different from the observed performance.[1–3]

Recently, an adjusted and improved version of the Assisting Hand Assessment (AHA), the Kids-AHA 5.0, demonstrated strong evidence of internal construct validity,[4] and the same scoring criteria can also be used to assess the bimanual performance of adolescents in a newly developed test situation, the board game 'Go with the Floe' (Ad-AHA Board Game).[5] The AHA is commonly used to assess the quality of use of the affected hand during bimanual performance in individuals with unilateral CP. The Kids-AHA has been validated for children with unilateral CP aged 1 year 6 months to 12 years and has proven reliable and sensitive enough to monitor changes over time.[6–11] With the newly developed test situation of the Ad-AHA, there is strong evidence that the AHA 5.0 is a valid instrument for assessing the bimanual performance of individuals aged 18 months to 18 years (AHA 18–18).[5] The AHA is used to score object-related hand actions observed during a semi-structured test situation (10–20min) that elicits the use of both hands and which is suitable for different age groups. The AHA 18–18 includes different test activities: (1) with the 'Small-Kids AHA', children aged 18 months to 5 years engage in exploratory play with toys; (2) with the 'School-Kids AHA', children aged 6 to 12 years handle the same objects but within the context of two AHA adventure board games; and (3) with the Ad-AHA Board Game, adolescents aged 13 to 18 years handle age-appropriate objects while playing the board game 'Go with the Floe'. Furthermore, two test activities have been developed in an adult post-stroke version of the AHA (Ad-AHA Stroke) with unpublished evidence of validity (L. Krumlinde-Sundholm, B. Lindquist, J. Plantin & B. Hoare, manuscript in preparation): (1) (un-) wrapping a present (Ad-AHA Present) and (2) preparing a sandwich (Ad-AHA Sandwich), which may also be suitable for adolescents.

The Small-Kids AHA and the School-Kids AHA have excellent interrater, intrarater, test–retest, and alternate form reliability.[7,8] However, for the Ad-AHA Board Game, there is currently no evidence regarding inter- and intrarater agreement, test–retest stability, or interchangeability of test activities. This information is essential in order to be able to evaluate whether changes after treatment or follow up are real effects or the result of measurement error.

The aims of this study were to investigate different aspects of the reliability of the Ad-AHA for adolescents with unilateral CP concerning: (1) interrater, test–retest reliability and the smallest detectable change; (2) alternate-form reliability of the Ad-AHA Board Game

versus School-Kids AHA (age group 10–13y); (3) alternate-form reliability of the Ad-AHA Board Game versus Ad-AHA Present and the Ad-AHA Board Game versus Ad-AHA Sandwich (age group 13–18y).

Methods

Participants

A convenience sample of 112 children and adolescents with unilateral CP, aged 10 to 18 years, were recruited by therapists working in eight different rehabilitation centers in Australia, Sweden, and the Netherlands (see Acknowledgements at end of paper) between May 2012 and February 2016. Forty-two of the children and adolescents participated in more than one type of reliability evaluation. The demographic characteristics and distribution of the adolescents are presented in Table I.

All participants and their primary caregivers gave informed consent. Participation was voluntary, and the parents and adolescents were informed that they could withdraw from the study at any stage. The ethical review committees of the involved centers approved the study.

Instrumentation

The AHA 5.0 was used, which consists of 20 items that describe object-related hand actions scored on a four-point rating scale.[4] The same AHA scoring criteria can be used for both children and adolescents, but scored from different age-appropriate test activities, thereby covering the age range 18 months to 18 years (AHA 18–18).[5] The sum scores are converted to AHA-units (interval logit measures) ranging from 0 to 100.[4]

Procedure

All AHA assessments were videotaped according to the standard protocol described in the AHA manual and sent to one location for central scoring. The participants were tested once or twice depending on the type of reliability evaluated. None of the participants received any form of intensive therapy between the two test occasions. The examiners were blinded to the results of each other's and their own previous assessments. All videos were scored in random order.

To evaluate interrater reliability, the first 38 participants in the convenience sample played the Ad-AHA Board Game once and two raters independently scored the videotaped session. Both raters (occupational therapists) had at least 10 years of clinical experience and were trained in scoring the AHA 5.0.

To evaluate test–retest reliability, the adolescents played the Ad-AHA Board Game twice conducted by the same assessor, in the same environment with an interval of 1 to 2 weeks, and the videos were scored by the same assessor who was blind to previous obtained AHA scores. The test-retest condition thus includes stability of both the examined person's behavior and intrarater agreement. For that reason, a separate intrarater analysis was not performed.

The four different test activities (School-Kids AHA, Ad-AHA Board Game, Ad-AHA Present, and Ad-AHA Sandwich) elicit bimanual actions involving the same underlying trait.

Alternate-form reliability determines whether the scores for the outcome measures of these tests are consistent relative to each other. In this study, agreement between AHA scores was analysed by comparing scores on the Ad-AHA Board Game with scores on one of the other three test activities. Two out of 85 adolescents performed three different test activities (School-Kids AHA, Ad-AHA Board Game, and Ad-AHA Present). The maximum interval between two tests was 2 weeks.

Statistical analyses

Analyses were performed with SPSS software, version 21.0 (IBM Corp., Armonk, NY, USA).

Agreement between raters and stability across time were calculated using the intraclass correlation coefficient (ICC; type 2.1, a two-way random effects single measures model of absolute agreement) with associated 95% confidence intervals (CIs).[12] Agreement between alternate test forms was calculated using ICC type 3.1 (a two-way mixed effects single measures model of absolute agreement).[12] Commonly accepted minimal standards for reliability coefficients are 0.70 for group comparisons and 0.90 to 0.95 for making clinical decisions for individuals.[13] Portney and Watkins suggested that for many clinical measurements reliability should exceed 0.90 to ensure reasonable validity.[14] In this study, ICC values higher than 0.90 were considered excellent, between 0.75 and 0.90 good, between 0.60 and 0.75 moderate, and lower than 0.60 poor.[15]

The standard error of the measurement (SEM) is the amount of error that can be considered random measurement error. The SEM agreement was calculated using variance components from the ANOVA analysis as the square root of the pooled MeanSquare-time and MeanSquare-person 9 time (SEM = $\sqrt{[s^2t + s^2residual]}$).[16] The smallest detectable change (SDC) indicates a change beyond measurement error.[11] The SDC was calculated from the SEM of test–retest and alternate-form scores, using AHA-units (SDC = SEM x 1.96 x $\sqrt{2}$).[12] At an individual level, a difference equal to or greater than the SDC can be considered a real change.

The presence of heteroscedasticity (or nonuniformity of error) was visually inspected with Bland–Altman plots.[17]

In accordance with recommendations,[18] the outcome of the AHA is reported in the Rasch-derived logit-based 0 to 100 AHA-unit scale (AHA-units). The ICC for the total measure, the SEM, and the SDC are given in the same units as the outcome measure (i.e. AHA-units 0–100 scale). To explore the agreement on item level, item's raw scores were used.

Results

Interrater and test–retest reliability of the Ad-AHA Board Game

The ICC2.1 for interrater reliability was 0.97 (95% CI 0.94–0.98), indicating excellent agreement for the total AHA scores of 38 adolescents. With a mean interval of 9 days (range 7–14d), the test–retest ICC2.1 was also excellent, 0.99 (95% CI 0.98–0.99) (Table II).

The test–retest ICCs of all items (using raw scores) were good to excellent. Between two raters only 3 out of 20 items had moderate ICCs (Table II).

Table I. Demographic characteristics and distribution of participants to different reliability trials

Type of reliability	*n*	Mean age years and months (SD)	Sex Male (*n*)	Affected side, right (*n*)	MACS level (*n*)				GMFCS level (*n*)			
					I	II	III	N/a	I	II	III	N/a
Inter-rater reliability												
Ad-AHA Board game	38	13.1 (2.8)	19	14	5	19	7	7	25	6	0	7
Test-retest reliability												
Ad-AHA Board game	31	14.6 (2.2)	15	17	7	15	4	5	21	5	0	5
Alternate-form reliability												
Ad-AHA Board game vs. School-Kids AHA	30	11.8 (1.4)	22	17	6	14	5	5	22	2	1	5
Ad-AHA Board game vs. Ad-AHA Sandwich	29	16.1 (2.0)	16	18	5	20	1	3	23	3	0	3
Ad-AHA Board game vs. Ad-AHA Present	28	15.1 (2.2)	9	16	8	15	5	0	19	8	1	0

MACS, Manual Ability Classification System;[23] GMFCS, Gross Motor Function Classification System;[24] N/a, missing data; Ad-AHA, Assisting Hand Assessment in adolescents.

Alternate-form reliability

For all alternate-form conditions the ICCs for individual AHA items were good to excellent (Table II). The ICC3.1 for alternate-form reliability of the School-Kids AHA versus the Ad-AHA Board Game was 0.99 (95% CI 0.98-0.99). It was 0.99 (95% CI 0.95-0.99) for the Ad-AHA Board Game versus Ad-AHA Present, and 0.99 (95% CI 0.98–0.99) for the Ad-AHA Board Game versus Ad-AHA Sandwich (Table II). Visual inspection of the Bland–Altman plots (Fig. 1) showed no heteroscedasticity.

Measurement error

The SEM for interrater and test–retest trials was 2.3 and 1.6 AHA-units respectively. The SEM for test–retest for the Ad-AHA Board Game resulted in a SDC of 4.5 AHA-units. The SEM and SDC for the four alternate forms are presented in Table II. At an individual level, a difference in scores greater than or equal to 5 AHA-units can be regarded as being change beyond measurement error (i.e., exceeding the random variation caused by intraperson differences in behaviour and by rater inconsistency) when using one of the four test activities (Ad-AHA Board Game, School-Kids AHA, Ad-AHA Sandwich, or Ad-AHA Present) (Table II).

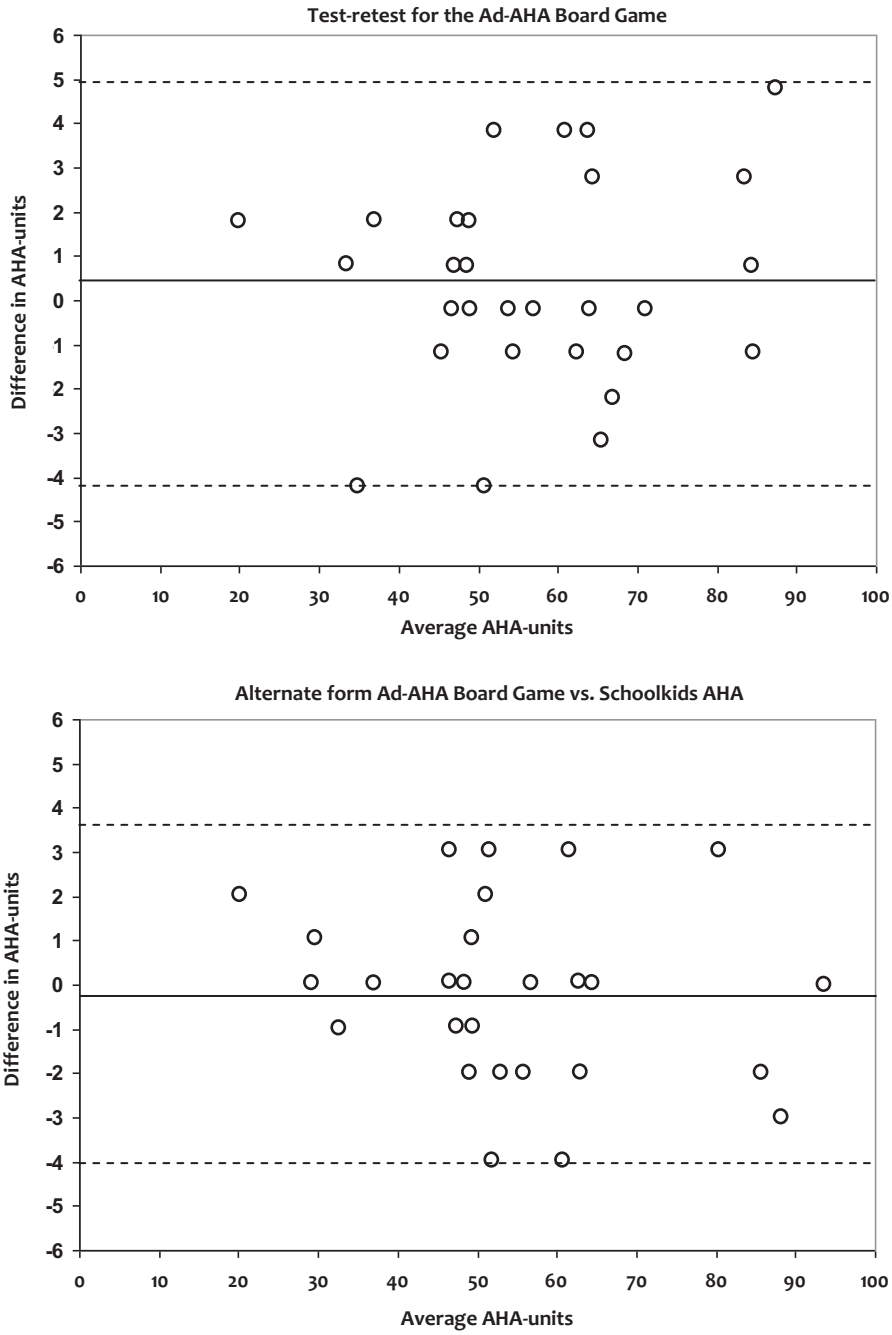

Figure I. Bland–Altman plots: difference against mean for Assisting Hand Assessment (AHA) measures (in AHA-units)

Solid line: group mean difference. Dotted line: limits of agreement.

Table II. Agreement between raters and alternate test forms, stability across time and smallest detectable change

	Interrater Ad-AHA Board Game	Test-retest Ad-AHA Board Game
	n=38	*n=31*
Measurement agreement		
For individual items in raw scores, ICC (95% CI)		
Initiates use	.80 (.64 - .89)	.94 (.88 - .97)
Amount of use	.63 (.39 - .78)	.79 (.61 - .89)
Chooses AH when closer to objects	.84 (.71 - .91)	.86 (.73 - .93)
Stabilizes by weight or support	.75 (.55 - .87)	.81 (.63 - .90)
Reaches	.84 (.70 - .92)	.93 (.85 - .96)
Moves upper arm	.66 (.43 - .81)	.92 (.85 - .96)
Moves forearm	.74 (.56 - .86)	.97 (.94 - .99)
Grasps	.80 (.65 - .89)	.95 (.90 - .98)
Holds	.86 (.75 - .93)	.94 (.87 - .97)
Stabilizes by grasp	.84 (.65 - .92)	.97 (.93 - .98)
Readjust grasp	.96 (.92 - .98)	.92 (.84 - .96)
Varies type of grasp	1.0	.94 (.88 - .97)
Releases	.79 (.53 - .90)	.96 (.91 - .98)
Moves fingers	.81 (.66 - .90)	.98 (.96 - .99)
Grip force regulation	.81 (.66 - .90)	.92 (.85 - .96)
Manipulates	.96 (.92 - .98)	.90 (.81 - .95)
Coordinates	.75 (.52 - .87)	.90 (.80 - .95)
Orients objects	.83 (.70 - .91)	.82 (.66 - .91)
Proceeds	.78 (.62 - .88)	.84 (.68 - .92)
Flow in bimanual task performance	.77 (.61 - .87)	.95 (.89 - .97)
For total measure in AHA-units, ICC (95% CI)	.97 (.94 - .98)	.99 (.98 -.99)
Measurement error		
SEM (AHA-units)	2.3	1.6
SDC (AHA-units)		4.5
Group mean difference (SD) (AHA-units)	-.47 (3.3)	-.55 (2.3)

Ad-AHA, Assisting Hand Assessment in adolescents; ICC, intraclass correlation coefficients; SEM, standard error of the measurement; SDC, smallest detectable change.

Alternate form Ad-AHA Board Game vs. School-Kids AHA *n=30*	Alternate form Ad-AHA Board Game vs. Ad-AHA Present *n=28*	Alternate form Ad-AHA Board Game vs. Ad-AHA Sandwich *n=29*
.90 (.81 - .95)	.88 (.76 - .94)	.81 (.62 - .91)
.89 (.78 - .95)	.78 (.58 - .89)	.91 (.82 - .96)
.90 (.79 - .95)	.96 (.92 - .98)	.91 (.81 - .96)
95 (.89 - .97)	1.0	.94 (.87 - .97)
.93 (.86 - .97)	.92 (.83 - .96)	.91 (.81 - .96)
.86 (.72 - .93)	.94 (.87 - .97)	.76 (.47 - .89)
.98 (.97 - .99)	.99 (.97 - .99)	.89 (.78 - .95)
.96 (.91 - .98)	.93 (.86 - .97)	1.0
.97 (.93 - .99)	.88 (.77 - .94)	.90 (.80 - .95)
1.0	.90 (.80 - .95)	.96 (.91 - .98)
.96 (.93 - .98)	.92 (.82 - .96)	.94 (.87 - .97)
.96 (.91 - .98)	.93 (.84 - .96)	.93 (.85 - .97)
.98 (.96 - .99)	.91 (.82 - .96)	.95 (.90 - .98)
.96 (.92 - .98)	.97 (.93 - .99)	.89 (.76 - .95)
1.0	.94 (.88 - .97)	.96 (.91 - .98)
.96 (.91 - .98)	1.0	.74 (.47 - .88)
.98 (.95 - .99)	1.0	.94 (.87 - .97)
1.0	.90 (.79 - .95)	.92 (.84 - .96)
.97 (.94 - .99)	.91 (.81 - .95)	.96 (.91 - .98)
.98 (.95 - .99)	1.0	.97 (.93 - .99)
.99 (.98 - .99)	.99 (.95-.99)	.99 (.98 - .99)
1.4	1.8	1.5
3.8	5.0	4.1
-.23 (1.9)	1.5 (2.1)	.59 (2.1)

Discussion

This study demonstrated excellent interrater and test–retest reliability of the AHA measures for adolescents with unilateral CP. The small test–retest SEM values indicate that measurement error due to random variance is acceptably low, taking into account not only the random error variance, but also possible rater and person inconsistency. Agreement between performance scores on the Ad-AHA Board Game and the School-Kids AHA was strong, which reflects excellent alternate-form reliability. Individual scores on the School-Kids AHA and the Ad-AHA Board Game were directly comparable, which makes it possible to use both test activities interchangeably for children aged 10 to 13 years. The alternate-form reliability was also good for adolescents (age 13–18y) performing three different activities. For interpretation of change knowledge about the size of the SDC is important. This study showed the SDC to be 4.5 AHA-units. Thus, a change of 5 AHA-units or more does (with 95% certainty) reflect a real change exceeding any random measurement error. This corresponds well with the SDC reported for the Kids-AHA version 4.4 (age 1y 6mo–12y).[18]

We calculated the SEM and SDC values of the alternate tests to determine whether the test activities can be used interchangeably. The higher the reliability, the lower the SEM, and the greater the confidence in the accuracy of an individual's test score. Because alternate-form reliability includes both the consistency of assisting hand use in different test activities as well as the consistency of performance across time, we expected that the estimated reliability coefficients would be somewhat lower and the SDC higher in comparison to test–retest values. We found a slightly higher SDC (5.0 AHA-units) for the Ad-AHA Board Game versus the Ad-AHA Present. The SDC for the Ad-AHA Board Game versus the School-Kids AHA was even lower (3.8 AHA-units), indicating that consistent performance is expected also when switching from the test activity appropriate for 10- to 12-year-old children (School-Kids Board Games) to the test activities appropriate for 13- to 18-year-old adolescents.

The different test activities of the Ad-AHA are important tools for observing and evaluating bimanual hand use in adolescents. The AHA 5.0 scoring system yielded consistent results even when the activities were performed in different ways due to cultural differences. For example, in the Sandwich task, some adolescents cut a slice of cheese with a knife (Australia) and some with a cheese slicer (the Netherlands and Sweden). There were also differences regarding the type of bread, spread, plastic bag closing devices, or wrapping paper and gift ribbons that were used in the different countries. However, these differences were discussed with participating therapists and it was ascertained that the tasks elicited bimanual actions sufficiently to make observation and scoring of AHA test items possible.

A limitation of this study might be the relatively small sample sizes to evaluate different types of reliability. It is generally accepted that at least 30 cases are needed for a reliable estimate of measurement error.19 Our samples for the alternate-form reliability in adolescents were somewhat smaller than the recommended sample size but it seems unlikely that this has affected the accuracy of the reliability indices. A limitation for the generalizability may be the fact that in our study, assessors had several years of experience with the AHA (one of them is an AHA teacher), which may have led to smaller measurement error. However, the test administration and AHA 5.0 scoring are highly standardized with precise and clear instructions specified in the AHA manual, which enhance test reliability. To evaluate test–retest reliability, the assessor was blind to previously obtained AHA scores

and none of the participants received any form of intensive therapy between the two test occasions. This study has demonstrated that the SDC of 5 AHA-units can be considered as a value of a change beyond measurement error, which is useful for interpreting effects of interventions. The highly acceptable SDC of 5 AHA-units (on the 0–100 scale) and the fact that the AHA 18–18 can distinguish nine significantly different levels of ability[5] indicate that the test has a high potential to be responsive to change. However, research is also needed to investigate the minimal clinically important difference. The minimal clinically important difference is the smallest change in scores that would be considered important or clinically meaningful to individuals.[16,20] This may be challenging for several reasons, for example small changes (even less than the SDC of 5 AHA-units) may be very meaningful for one child or adolescent but not for another, and the size of a meaningful change may vary across different levels of ability. For example, for a person with severely limited function of the affected hand, a change from a score of 1 ('does not do') to a score of 2 ('inefficient') on a few easy items like 'Amount of use', 'Holds', 'Stabilizes by weight or support', may imply a big difference making it possible to start using the two hands together at all. This small change may open up a possibility to perform some bimanual tasks, even if it is with difficulty. On the other end of the ability scale, a person already scoring 3 (somewhat effective/almost normal performance) on the more difficult items like 'Manipulates', 'Varies type of grasp', 'Readjusts grasp', may not encounter scores of 4 (effective) as a clinically important difference since already a score of 3 indicates a high ability and good assisting hand function.

To report outcomes and evaluate change, interval level measures are recommended.[21] Therefore the AHA raw-scores are converted into a logit-based 0 to 100 AHA-unit scale (AHA-units).[18] The logarithmic transformation 'stretches' the scale in the lowest and the highest ends and renders a scale with equal size of measure intervals along the whole range. Thus, this scale makes change comparable for persons at different starting points along the scale. It also implies that, for example, a change of 1 AHA raw-score in persons with an extreme low ability (22 raw scores) results in a change of 4 AHA-units. On the other hand, a change of 1 AHA raw-score in persons with an average ability (about 40 raw scores) may result in a change of only 1 AHA-unit. For clinical practice and research, it is important to know that a small change in raw-scores in individuals with extreme low or high ability may imply a big (significant) change in measures of the quality of use of the affected hand during bimanual performance, and the interval level AHA-units is the fairest measure of change.[22]

Conclusions

This study showed that the Ad-AHA has good-to-excellent interrater and test–retest reliability in adolescents with unilateral CP. Test scores for the alternate forms were comparable, showing that reliable AHA scores can be generated by using different age-appropriate tests situations for children and adolescents aged 18 months to 18 years. The excellent measurement agreement means that the different test activities can be used to evaluate change over time, with a score change of 5 AHA-units indicating a change beyond measurement error with 95% certainty, using the AHA 5.0 scale.

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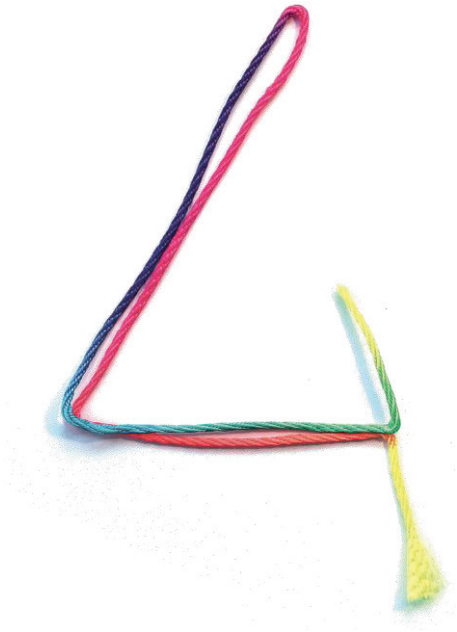

Immediate effect of a wrist and thumb brace on bimanual activities in children with hemiplegic cerebral palsy

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Abstract

Aim

The aim of this study was to determine the immediate effect of wearing a wrist and thumb brace on the performance of bimanual activities in children with spastic hemiplegic cerebral palsy.

Method

In a pre- and post-test cohort study of 25 children (age range 4 - 11y; mean age 8y 4mo [SD 2y 2mo]; 16 males, 9 females) with spastic hemiplegic cerebral palsy with a Zancolli classification hand score of I, IIA, or IIB (mild and moderate hand dysfunction; children with a Zancolli classification of III - severe hand dysfunction - were excluded from this study), performance of bimanual activities was evaluated with the Assisting Hand Assessment (AHA) on three occasions: one assessment with a static wrist and thumb brace placed on the affected hand and two other assessments without a brace. The differences between AHA scores obtained at the three assessments were evaluated using the repeated measures analysis of variance.

Results

Performance of bimanual activities while wearing the brace improved significantly compared to performance without the brace ($p<0.001$). With the brace, the mean AHA score increased by 3.2 (95% confidence interval 2.1–4.3) from 59.1 to 62.3. The scores of the two assessments without the brace did not differ significantly.

Interpretation

In children with spastic hemiplegic cerebral palsy, bracing of the wrist and thumb immediately improves spontaneous use of the affected upper limb in bimanual activities, possibly because bracing permits amore functional hand position.

Introduction

The impaired upper limb function of children with hemiplegic cerebral palsy (CP) is considered to be the main motor impairment that limits and restricts participation in activities of daily living.[1] Reduced hand function affects almost every aspect of daily life: self-care, school or work, and engagement in play or leisure activities.[2,3] Activities requiring bilateral manipulation are the most difficult to perform. It has been demonstrated that children with a unilateral motor disability, such as hemiplegic CP, have more difficulties with spontaneous manipulation during play or activities of daily living than during therapy sessions.[1] Therefore, Krumlinde-Sundholm et al.[4] developed the Assisting Hand Assessment (AHA), an observational instrument that evaluates spontaneous affected hand use in bilateral manipulation skills as well as any changes in hand use over time.

The aim of occupational therapy for children with hemiplegic CP is to optimize performance in tasks that require bilateral manipulation during play or activities of daily living. There is more need for randomized controlled studies to make evidence-based decisions in order to select the best treatment for a child with hemiplegic CP. The number of randomized controlled trials have increased over the past 10 years, especially for the investigation of the effect of botulinum toxin A therapy[5] and constraint-induced movement therapy.[6] Unfortunately, the effectiveness of a frequently used assistive device during therapy, a wrist and thumb brace, is unclear owing to a lack of randomized controlled studies in this area. A wrist and thumb brace used on the affected side aims to change the hemiplegic pattern of the upper limb (flexion of the wrist and fingers and thumb adduction), into a more functional position of the hand (a neutral position of the wrist and abduction/opposition of the thumb).

Preliminary findings of a small study suggest that wrist and thumb braces improve the hand function of children with hemiplegic CP.[7] Before embarking on a randomized controlled trial, this pre- and post-test cohort study was undertaken to investigate the immediate effects of a static wrist and thumb brace on the spontaneous use of the affected upper limb in bimanual activities in children with hemiplegic CP.

Methods

Participants and recruitment

From December 2008 to May 2009, all children with hemiplegic CP who were referred for occupational therapy to the Department of Rehabilitation at the Academic Medical Center, Amsterdam, or the Rehabilitation Center 'De Trappenberg', Huizen, were consecutively screened for inclusion and exclusion criteria by their rehabilitation physician. Figure 1 shows the flow diagram of the patient selection process. Children were included if they were aged between 4 and 12 years, diagnosed with spastic hemiplegic CP, and had a hand function with a classification of Zancolli I, IIA, or IIB.[8] Children who were classified as Zancolli III (severe impairment of hand function, with no voluntary extension of the fingers and wrist and no active extension of the fingers even with maximal wrist flexion) were excluded from this study.

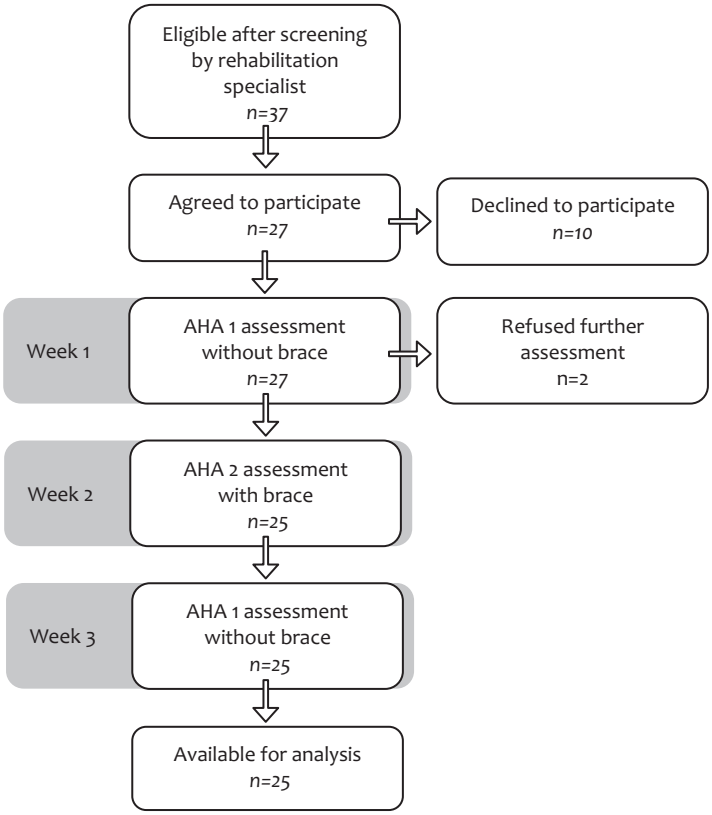

Figure 1. Flow diagram of patient selection process. AHA, Assisting Hand Assessment

AHA 1, without brace; AHA 2, with brace; AHA 3, without brace.

The sample size was based on a clinically relevant change score on the AHA of 0.97 logits, with a variance of the paired difference of 1.5 (based on a standard error of the mean of 0.35 in 18 children).[9] Twenty-five children were needed to detect this difference with 90% power and a two-sided 5% significance level. The Academic Medical Center and the Rehabilitation Center 'De Trappenberg' selected 37 eligible children; however, the parents of 10 did not consent to the study. Two children were not willing to complete the first AHA assessment and refused further participation. Thereafter, no children were lost to follow-up.

The mean age of the remaining 25 children with hemiplegic CP was 8 years and 4 months (SD 2y 2mo; age range 4–11y); 16 were male and 9 were female; and in 15 children the right side was affected, in 10 the left. Baseline demographics, clinical characteristics, and the results of the three assessments of all children are given in Table I.

The Medical Ethics Committee of the Academic Medical Center, Amsterdam, considered the procedures applied in this study to be part of standard treatment and thus waived the need for ethical approval. However, parental written consent was obtained from all parents.

Procedures/data collection

In a pre- and post-test cohort study, performance of bimanual activities by children with hemiplegic CP was evaluated with the AHA three times (AHA 1, AHA 2, AHA 3) with an interval of 1 week between tests: AHA 1 consisted of an assessment without a brace; AHA 2 consisted of an assessment with the brace placed on the affected hand; and AHA 3 was once again an assessment without a brace. For AHA 2, the brace was placed on the hand immediately before the assessment. After this assessment the brace was immediately removed and the children did not use the brace again during the study. All AHA assessments were randomly scored and the time period of 1 week between assessments was chosen in order to prevent recall effects. In between the assessments, children did not receive any additional treatment (such as constraint-induced movement therapy or botulinum toxin therapy) other than their usual therapy (bimanual training conducted, on average, two times a week). Before and during the first visit (AHA 1) the following data were collected: demographic and clinical variables, including the Gross Motor Function Classification System (http://www.netchild.nl),[10] Manual Ability Classification System,[11] Zancolli classification,[8] and House classification.[12] The AHA was administered and scored by a certified AHA rater (an occupational therapist not involved in the treatment of the children). During scoring of all AHA assessments of the 25 children, the rater was blinded to the scores from prior scored AHA assessments of each child.

Measures

Assisting Hand Assessment (AHA)

Hand use was measured with the AHA, which evaluates the spontaneous affected hand use in bilateral manipulation skills as well as the change in hand use over time. It measures a large range of skills, even those requiring very low ability levels.[4]

The AHA is a test kit with selected toys that stimulate spontaneous use of both hands. The way in which the children use the assisting hand is videotaped and their performance is scored on 22 items on a rating scale 1 to 4 (1, no use; 4, efficient use of the affected hand). The sum scores (range 22–88 points) constitute ordinal-level data. Logits (interval-level data) are obtained from a Rasch analysis (range 10.18 to 8.70 logits).

Rasch measurement analysis provides measures of equal intervals in the unit logits by converting the ordinal rating scale. Therefore, we used this scale for analysing the assessments. Sum scores are presented in the tables and figures for better interpretation.

The AHA can be performed only by certified AHA raters. The raw-score to logit-score conversions were obtained from the original authors of the AHA who have the large Rasch analysed database (http://www.ahanetwork.se).[4]

The AHA is a valid instrument for use in children with hemiplegic CP aged between 18 months and 12 years. Inter- and intrarater reliability are excellent.[13] Evidence supporting responsiveness to change was obtained in an intervention outcome study.[14] The current evidence of sound psychometric properties for the outcome measure is positive and indicates that the AHA can be useful for both clinical practice and research.[15]

Table I. Baseline demographic, clinical characteristics, and outcome of the three Assisting Hand Assessment (AHA) assessments. Mean age 8.4 years (SD 2.2y)

Child (*n*=25)	Age (mo)	Sex	Involved side	Zancolli class	GMFCS class	MACS class
19	56	F	L	I	2	3
13	90	M	L	I	1	2
22	68	M	R	I	1	2
1	136	F	R	I	1	1
24	123	M	L	I	1	2
11	78	M	R	I	1	2
2	103	M	R	I	1	1
25	124	F	L	I	1	1
23	118	M	R	I	1	1
4	96	F	R	I	1	1
6	126	F	R	I	2	1
5	116	M	R	IIA	1	2
9	91	M	R	IIA	1	2
10	110	M	L	IIA	1	2
12	104	F	R	IIA	1	2
15	64	M	R	IIA	1	3
14	135	F	R	IIA	1	2
3	123	F	R	IIA	2	2
17	135	M	R	IIA	1	2
7	128	M	L	IIA	1	2
16	79	M	L	IIB	2	3
8	55	F	L	IIB	2	3
18	101	M	R	IIB	2	3
20	68	M	L	IIB	2	3
21	81	M	L	IIB	2	3

M, male; F, female; School type 1, special school for children with physical disabilities; School type 2, primary school; AHA 1, without brace; AHA 2, with brace; AHA 3, without brace; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System.

School type	House class	AHA 1 sum score	AHA 2 sum score	AHA 3 sum score
0	1	61	65	60
0	0	62	67	61
1	0	63	66	63
1	3	64	68	65
1	1	64	66	64
0	0	68	72	68
0	0	73	73	74
0	0	76	78	76
1	0	78	82	79
0	0	81	81	81
0	0	86	86	86
0	3	48	57	48
0	1	48	55	49
0	1	55	57	56
1	1	56	58	56
0	3	56	56	56
0	1	57	62	59
0	1	59	63	58
1	3	61	67	61
0	1	64	64	64
0	2	31	35	31
0	2	33	33	33
1	1	40	44	41
0	2	43	44	43
0	1	51	59	52

Zancolli classification

The posture of the wrist, fingers, and thumb were evaluated and classified in accordance with the Zancolli classification.[8] The complete extension of fingers with the wrist in the neutral position or with less than 20° of flexion is classified as Zancolli I. The classification Zancolli IIA is used if active extension of the wrist is possible only when the fingers are flexed, and Zancolli IIB is used if no active extension of the wrist is possible even when the fingers are flexed. Zancolli III indicates severe impairment of hand function, with no voluntary extension of the fingers and wrist, and no active extension of the fingers even with maximal wrist flexion. Children in this category of hand/wrist function were excluded from this study.

Intervention

The intervention consisted of a static wrist and thumb brace, the 4068 Children's Wrist and Thumb Support by Otto Bock, placed on the affected hand (Fig. 2). This fabric brace is designed to provide support to both the wrist and the carpometacarpal joints. The anatomical design and small sizes enable a precise fit for a child's small hands. An adjustable thumb grip provides optimal thenar support, while an aluminium splint on the volar side supports the wrist.

Figure 2. Static wrist and thumb brace

Data analysis

Statistical Package for the Social Sciences (SPSS) version 16.0 (SPSS Inc., Chicago, IL, USA) was used for data analysis. The immediate effect of the wrist and thumb brace on the performance of bimanual activities was assessed by comparing the AHA scores of the three assessments (without, with, and without the brace) with repeated measures analysis of variance (ANOVA). Subgroup analyses were performed, based on Zancolli grade with the Friedman repeated measures analyses of variance by ranks, with post hoc comparisons using the Wilcoxon signed-rank test.

Table II. Outcome of three Assisting Hand Assessment (AHA) tests (total and Zancolli subgroups)

	Sum score (mean; 95% CI)			Mean change (95% CI)	
Group	**AHA 1**	**AHA 2**	**AHA 3**	**AHA 1 to 2**	**AHA 1 to 3**
Total (*n*=25)	59.1 (53.4 to 64.9)	62.3 (56.8 to 67.9)	59.4 (53.6 to 65.1)	3.2 (2.1 to 4.3)	0.2 (-0.1 to 0.5)
Zancolli I (*n*=11)	70.6 (64.7 to 76.4)	73.1 (68.1 to 78.1)	70.6 (64.6 to 76.7)	2.6 (1.3 to 3.8)	0.1 (-0.4 to 0.6)
Zancolli IIA (*n*=9)	56 (51.9 to 60.1)	59.9 (56.7 to 63.1)	56.3 (52.4 to 60.3)	3.9 (1.5 to 6.3)	0.3 (-0.3 to 1.0)
Zancolli IIB (*n*=5)	39.6 (29.6 to 49.6)	43.0 (30.3 to 55.8)	40.0 (29.5 to 50.5)	3.4 (-0.5 to 7.3)	0.4 (-0.3 to 1.1)

AHA 1, without brace; AHA 2, with brace; AHA 3, without brace.

Results

The performance of bimanual activities in children wearing a brace improved significantly compared with the performance of children not wearing a brace ($p<0.001$). When using the brace, the mean AHA score increased by 3.2 (95% confidence interval [CI] 2.1–4.3) from 59.1 to 62.3 (Table II). The two assessments without the brace did not differ significantly ($p=0.444$).

The results of subgroup analyses, based on Zancolli grade, are also shown in Table II and Figure 3. Children with Zancolli grade I (n=11) wrist function showed a significant improvement on the AHA while wearing a brace ($p=0.001$). An increase of 2.6 from a mean AHA score of 70.6 without to 73.1 with a brace was found (95% CI 1.3–3.8). Also, in children classified as Zancolli grade IIA (n=9) wrist function showed a significant improvement on the AHA when the children were wearing a brace ($p=0.006$): an increase of 3.9 from 56 without to 59.9 with a brace (95% CI 1.5–6.3). In the subgroup of children classified as Zancolli grade IIB wrist function was too low to test for significance (n=5; $p=0.072$). However, when this subgroup used a brace, the mean AHA sum score increased by 3.4 from 39.6 to 43 (95% CI) 0.5 to 7.3). The differences in sum score on AHA tests 1 and 2 and AHA tests 1 and 3 against their mean are shown in Figure 3.

Group mean AHA scores were 3.2 points (sum score) higher when the children used a brace than when a brace was not used. For clinical practice it is important to compare the individual results of the AHA sum scores with and without the brace. Holmefur et al.[9] showed that an individual change in sum score on the AHA of 4 or more between two test sessions, with 95% certainty, can be considered a clinically relevant change. In our study, 13 children (52%) improved by at least 4 points on the AHA sum score when using a brace. These 13 children were distributed over the three subgroups: five children were classified as Zancolli I, five as Zancolli IIA, and three as Zancolli IIB (Fig. 3).

Detailed analysis of the scores for individual AHA items while wearing a brace revealed different patterns of change. A positive effect of bracing was found for 9 of 22 AHA items.

In six items ('stabilizes grip', 'readjust', 'calibrates', 'releases', 'grasps', and 'holds') the improvement was directly related to the supporting effect of bracing on grasping and releasing objects, whereas in the other three items ('moving fingers', 'reaches', and 'initiates use') this relation was less clear.

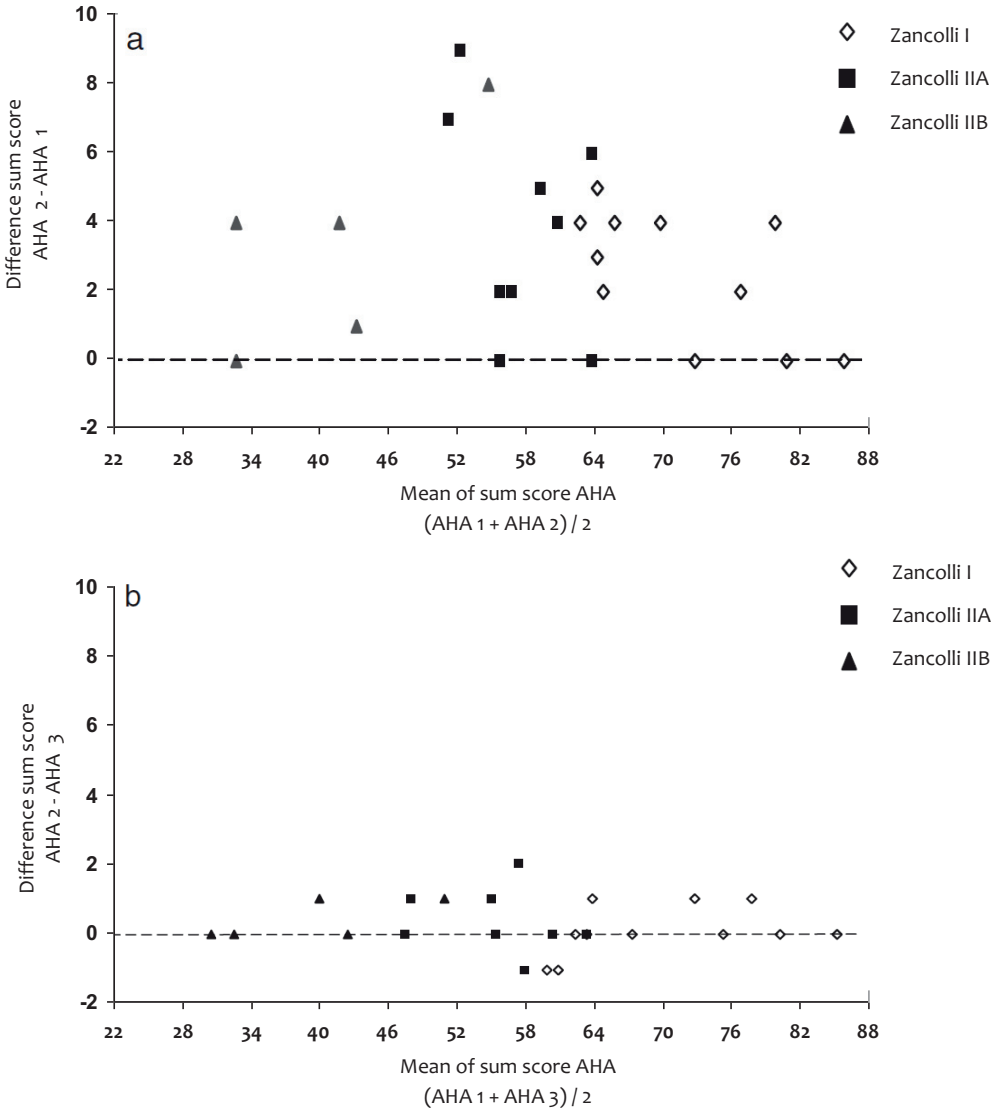

Figure 3. Difference in Assisting Hand Assessment (AHA) sum score between assessments against their mean, with different plotting symbols for the Zancolli grades

(a) Assessments week 1 (AHA 1, without brace) and week 2 (AHA 2, with brace). (b) Assessments week 1 and 3 (AHA 1, AHA 3, both without brace).

Discussion

This study shows that static bracing of the wrist and thumb may immediately improve the spontaneous use of the affected upper limb in bimanual activities in children with spastic hemiplegic CP measured with the AHA.

The positive effect on spontaneous use was seen only during the assessment while the child was wearing a brace. No learning effect of the test or carry-over effect from wearing a brace during this short period was found – the AHA sum scores of the last assessment without a brace did not differ from the first assessment without a brace. Impairments in hand function varied widely in our study population, and consequently a broad range of AHA sum scores were found. We performed subgroup analyses based on Zancolli grade. Despite the small sample sizes of the subgroups classified as Zancolli grades I and IIA (11 and 9 respectively), we found a significant improvement on the AHA in the performance of the children in these subgroups when wearing a brace. The subgroup classified as Zancolli IIB was too small (n=5) to draw conclusions about the effect of the brace.

Our results are in line with the few previous studies examining the effect of a brace in children with hemiplegic CP. These studies showed that wearing a wrist and thumb brace improved grip and manual dexterity in children with CP.[7] Also, increased grasp has been found in these studies.[16,17] However, to our knowledge, there are no studies reporting on the efficacy of a brace on spontaneous use of the affected upper limb in bimanual activities.

A plausible explanation for the immediate positive effect of a brace on spontaneous use is the support that the brace gives to both the wrist and the carpometacarpal joint. Without a brace, children classified as Zancolli I, IIA, and especially IIB flex their wrist while grasping objects. With a flexed wrist it is more difficult to close the hand properly and to hold objects firmly. The brace supports the wrist in a neutral position. Children classified as Zancolli I and IIA are still able to extend their fingers for grasping and releasing. Children classified as Zancolli IIB extend their fingers only with a flexed wrist. With a brace, some children were still able to extend their fingers but with more effort, and they needed more time to open their hand. The brace also supports the carpometacarpal joint and places the thumb in opposition and abduction, preventing the 'thumb-in-palm' posture. Therefore, children with spasticity of the thumb also benefit from the support for and positioning of the thumb that the brace offers.

A possible explanation for the positive immediate effect of the brace on the bimanual performance is that children are more aware of the assisting hand because of the sensation on the skin and the stretching sensation in the flexor muscles of the forearm, or simply because of the bright colours of the brace.

The difference of 3.2 points for the group mean AHA scores indicates that the child's usual performance in relevant and motivating activities was more effective with a brace. With a brace, children were able to grasp objects of different shapes without interaction of the dominant hand. In daily life this may optimize performance of bimanual tasks such as playing with construction material. Children were also able to open their hand and hold objects, such as a bottle or marbles, more easily when wearing a brace. Their grip was more powerful, their hand stabilized the objects, and more interaction between both hands was possible. These positive effects of bracing can be important for achieving a child's goal, for example holding the handlebar of a bicycle with two hands.

With evaluation on the level of individual AHA items, the therapist can make an evidence-based decision on the kind of activities in which the individual child will most likely be able to benefit most from the brace.

Our study has some limitations. The results should be interpreted cautiously because of the pre- and post-test cohort study design without a comparison group and the short follow- up time. It was impossible to blind the rater for the intervention. Another limitation is that the sample size was too small to perform a subgroup analysis based on Zancolli grade, which limits the ability to generalize the results to all children with spastic hemiplegic CP and specified Zancolli grade.

We studied the immediate effects of wearing a wrist and thumb brace, so no conclusion can be drawn about the long-term effects of prolonged use supported by training. Burtner et al.[7] observed decreased muscle activation in the forearm muscles during grip and suggested that this may result in muscle atrophy in the forearm muscles after a long period of using a wrist and thumb brace. So because long term use could result in muscle atrophy, intermittent use of a brace may be preferred. As randomized controlled studies about the long-term effects of bracing are lacking, it is not possible to give a clear recommendation for duration of wearing a brace.

In conclusion, bracing of the wrist and thumb immediately improved spontaneous use of the affected upper limb in bimanual activities in half of our sample of children with spastic hemiplegic CP distributed over Zancolli subgroups. The largest improvement was found in grasping and releasing objects. The effectiveness of daily use of a brace in combination with therapy to stimulate bimanual use compared with this therapy without a brace should be investigated in a randomized controlled trial with a sample size sufficient to identify subgroups that benefit most from bracing. Long-term follow-up is required to check for any side effects of bracing on hand use in bilateral activities.

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Effects of upper extremity surgery on hand use and activity performance in children and adolescents with cerebral palsy: a systematic review

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Submitted

Abstract

Aim

To evaluate and synthesize the evidence for the effectiveness of upper extremity surgical (UES) interventions on hand use and performance of activities in children and adolescents (aged<20 years) with cerebral palsy (CP).

Method

Searches were carried out in MEDLINE, Embase, psycINFO until May 2017. Studies were included if they were comparative studies with or without concurrent controls or case series with pre-test/post-test outcomes with a minimal sample size of 10; and report the effects of upper extremity surgery (UES) with a mean follow-up time of at least 5 months; and included patients diagnosed with CP aged up to 20 years; and used validated activity-based instrument to measure the effect of UES on hand use and performance. Risk of bias was assessed for the included individual studies using the ROBINS-I tool (Risk Of Bias In Non-randomized Studies - of Interventions) criteria for reviews. Quality assessment was performed for outcomes of interest, using Grading of Recommendations Assessment, Development, and Evaluation (GRADE).

Results

Twelve studies were included in the review, of which 1 was a comparative study on the effectiveness of UES, botulinum toxin injections and regular ongoing therapy. Only 2 out of the 12 studies were prospective. The ability to use the hand(s) and perform activities/tasks on request (capacity) or spontaneously and the perception of the patient's ability to use the hand(s) in task performance improved significantly after UES.

The quality of evidence was very low for each of the activity outcomes of interest due to heterogeneity of surgical interventions and outcome measures and the poor methodological quality of the studies with uncontrolled designs.

Interpretation

Current evidence for UES interventions to improve hand use and performance in children and adolescents with CP is of very low quality and thus prohibits recommendations to guide clinical practice. More high quality studies comparing different treatment options are needed to get more insight in the effectiveness of UES on hand use and performance of activities.

Introduction

Children and adolescents (patients) with cerebral palsy (CP) represent the largest diagnostic group treated in paediatric rehabilitation. The upper limbs are often affected with significant wrist and hand involvement.[1] The abnormal upper limb tone and hand posture, most frequently thumb adduction and/or flexion with limited wrist extension, have impact on the ability to use the affected hand and both hands together.[2]

Upper extremity surgery (UES) aims to improve muscle balance and hand posture, by releasing or lengthening spastic muscles, tendon transfers and joint stabilization procedures. The most common procedures in random order are: (I) release of pronator teres muscle to facilitate forearm supination[3,4] (II) transfer of the flexor carpi ulnaris tendon to facilitate or increase wrist extension for functional grip purposes[5] (III) correction of a thumb-in-palm deformity, preferably and if possible by adductor pollicis muscle slide[6] combined with extensor pollicis longus rerouting.[7] In contrast to activity-based interventions, UES is an invasive intervention followed by 5-6 weeks immobilisation in plaster after which an intensive activity-based therapy program may start.

There is a substantial emphasis in the medical literature reporting outcomes on the effect of UES in terms of "functions", i.e. motor outcomes, active and passive range of motion and reduce spasticity, position of the hand, appearance and grip function.[8] The effect of UES on hand use and performance is less clear and reported. The execution of activities can be assessed according to the patient's functional performance (what the patient usually does), by testing what a patient is able to do on request (capacity) or by evaluating the patient's self-perception of the ability to use affected arm.

The last decade, more studies have been performed using validated activity-based instruments to evaluate the effect of UES on activities, i.e. the use of the affected hand and perceived performance. To guide therapists in intervention planning and to inform patients on the risks, benefits and possible outcomes of UES on daily performance, more knowledge is needed for an overview of evidence about the effect of UES on hand use and performance.

The aim of this systematic review is to evaluate and synthesize the evidence for the effectiveness of UES interventions on hand use and performance of activities in children and adolescents (aged < 20 years) with CP. This review will focus on two types of activity-based outcome measures; (i) Functional performance outcome measures (clinician-based), which reflects the patient's performance on a quantifiable tasks, classification, interview or questionnaire, (ii) Perceived performance outcome measures (patient-/caregiver-based), which reflects the perception of the patient's ability to use the hand(s) in task performance.

Methods

This systematic review was conducted using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.[9] The review has been registered in the international prospective register of systematic reviews (PROSPERO #CRD42017058753).

Eligibility criteria

Published studies investigating the effect of UES in children and adolescents with CP were included if: (1) They were (pseudo-) randomized controlled trials, comparative studies with concurrent controls (including non-randomised experimental trial, prospective and retrospective cohort study, case-control study and interrupted time series with a control group), comparative studies without comparative controls (including historical control study, two or more single arm study and interrupted time series without a parallel control group), or case series with either post-test or pre-test/post-test outcomes with a minimal sample size of 10, and (2) They reported the effects of UES with a mean follow-up time of at least 5 months; and (3) The intervention UES was not combined with botulinum toxin injections 6 months prior to surgery; and (4) More than 75% of the participants were children/adolescents diagnosed with CP aged up to 20 years or if data on the children/adolescents with CP were reported separately and (5) A validated activity-based instrument was used to measure the effect of UES on hand use and performance. Studies were excluded if the full-text was not published in English, or a qualitative method was used to evaluate the outcome, i.e. when hand use and/or performance was evaluated as a qualitative measure of function.

Search strategy

The comprehensive literature search started with a reference set collected through Google scholar. References in the included articles were checked and used to complete the reference set up to 45 articles. The next literature search was conducted in May 2017 in three electronic databases indexing health-related journals: MEDLINE, Embase, psycINFO to identify clinical effectiveness of UES in patients with CP. The search strategy was designed with individual search terms for each database using title/abstract, controlled vocabulary (MeSH, EMtree, tiab) and key words ([cerebral palsy] or [upper limb spasticity]) and [relevant surgical interventions] (see appendix: search strategy). The search strategy was developed to maximize sensitivity of article identification. Key terms within the search strategy were mapped to medical subject headings (MeSH) in Medline and exploded to include subheadings and related terms. Review authors also searched reference lists of included studies and other narrative reviews, identifying no additional reference titles. All duplicates were removed. The search strategy was designed in collaboration with a clinical librarian according to the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.[9]

Study selection

Titles and abstracts were initially screened for study eligibility/relevance by the first author (AL) with the second author (AB) screening excluded titles and abstracts to ensure that no relevant papers were omitted. After the initial identification and screening, full-text articles reporting the effect of UES in children/adolescents with CP, were reviewed and independently assessed for eligibility according to the in- and exclusion criteria by two reviewers (AL and AB). Cases of disagreement were discussed until consensus was reached.

Data extraction

Two reviewers (AL and AB) extracted information from each included study independently and reported their findings on a data extraction sheet based on the "Oxford Centre for Evidence-Based Medicine Scale V2.1".[10] Disagreements were resolved by discussion between the two review authors. No authors were contacted to provide additional data.

Information was extracted from each included study on: (1) study design; (2) characteristics of the study sample (including age, types of CP); (3) surgical (contra-) indications (4) surgical technique; (5) length of follow up; (6) comparative subgroups; and (7) types of outcome measures (including functional performance outcome measures, evaluating patient's performance on a quantifiable task, classification or interview and patient reported outcomes evaluating the patient's activity level of using the hand(s).

Risk of bias in individual studies

The risk of bias for non-randomized observational interventional studies with validated outcome measures was evaluated by two reviewers (AL and AB) working independently using the ROBINS-1 measurement tool.[11] Each article was assessed as low, high, or unclear risk for six criteria and reported in the characteristics of included studies table and Risk of Bias table.

Assessment of the quality of evidence

The quality of the body of evidence for each effectiveness outcome was examined by two assessors (AB and AL) according to the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) Working Group criteria.[12] GRADE was used to summarize the body of evidence and to enable clinicians to more quickly interpret the findings of the review for clinical practice. The overall quality was determined to be high, moderate, low, or very low using a step-wise, structural methodology, for outcomes of interest across the studies and overall, as well as the strength of recommendation for use of the UES-intervention. The scores were compared and disagreements were resolved by discussion and consensus.

Data synthesis

Given the heterogeneity of UES-interventions (within and between studies) and reported outcomes a descriptive narrative synthesis of results is presented.

Results

Study selection

The electronic search provided a total of 8009 citations. After adjusting for duplicates 5656 remained. Of these, 5524 studies were discarded because, after reviewing the abstracts, it appeared that these papers were not published with a full text in English (n=562) or clearly did not meet the criteria (not about surgery and cerebral palsy and upper extremity) (4.962). The full text of the remaining 132 citations was examined in more detail. 108 studies did not meet the inclusion criteria as described, mainly because of a different outcome or population. Twelve studies were excluded because data was not presented separately or less than 75% of the participants were patients diagnosed with CP aged up to 20 years. Finally, a total of 12 studies were identified for inclusion in the review (Fig. 1).

Characteristics of included studies

Comparative effectiveness of UES was reported in 1 out of the 12 included studies, in which a (pseudo-) randomized controlled trial[13] evaluated differences in the effects of 3 interventions (UES, botulinum toxin injections and regular ongoing therapy) on functional improvement (Table 1.). One other study retrospectively compared outcomes between two subgroups based on surgical intervention.[15] Two out of the 12 studies had a prospective design[13,16] and the length of follow-up after UES varied greatly among the studies (range 4 months - 12 years). The two prospective studies state measurement time points in advance and 3 retrospective studies described the use of a standard protocol which included standardized follow-up time-points.[14,17,18] Four studies provided information about missing data and loss to follow-up.[13,17-19]

Inclusion criteria of study participation were generally described and in some studies descriptive information about participants was lacking. Eight studies included only children and adolescents with CP aged up to 24 years. [13-15,17,19-22] From the 3 studies including patients older than 20 years of age,[18,23,24] the percentage patients to include (>75% of the participants were aged up to 20 years) could be derived from the reported mean and range. Four studies included specific unilateral CP[13,17,19,20] of which 3 spastic CP.[17,19,20] Different surgical interventions were used for the most common deformities in children and adolescents with CP. Six studies evaluated the effect of multi-level upper extremity surgery.[13,14,16,17,19,20] The other studies evaluated more specific surgical interventions, in case of elbow deformity[14], wrist flexion deformity[18,21], thumb-in-palm deformity[22] and finger deformity.[23,24] Indications were based on presentation of pronation deformity, deficient active wrist extension with adequate digital control and wrist flexion deformity with the FCU as the primary deforming force and adduction of the thumb ray with flexion at the thumb (Table 1.).

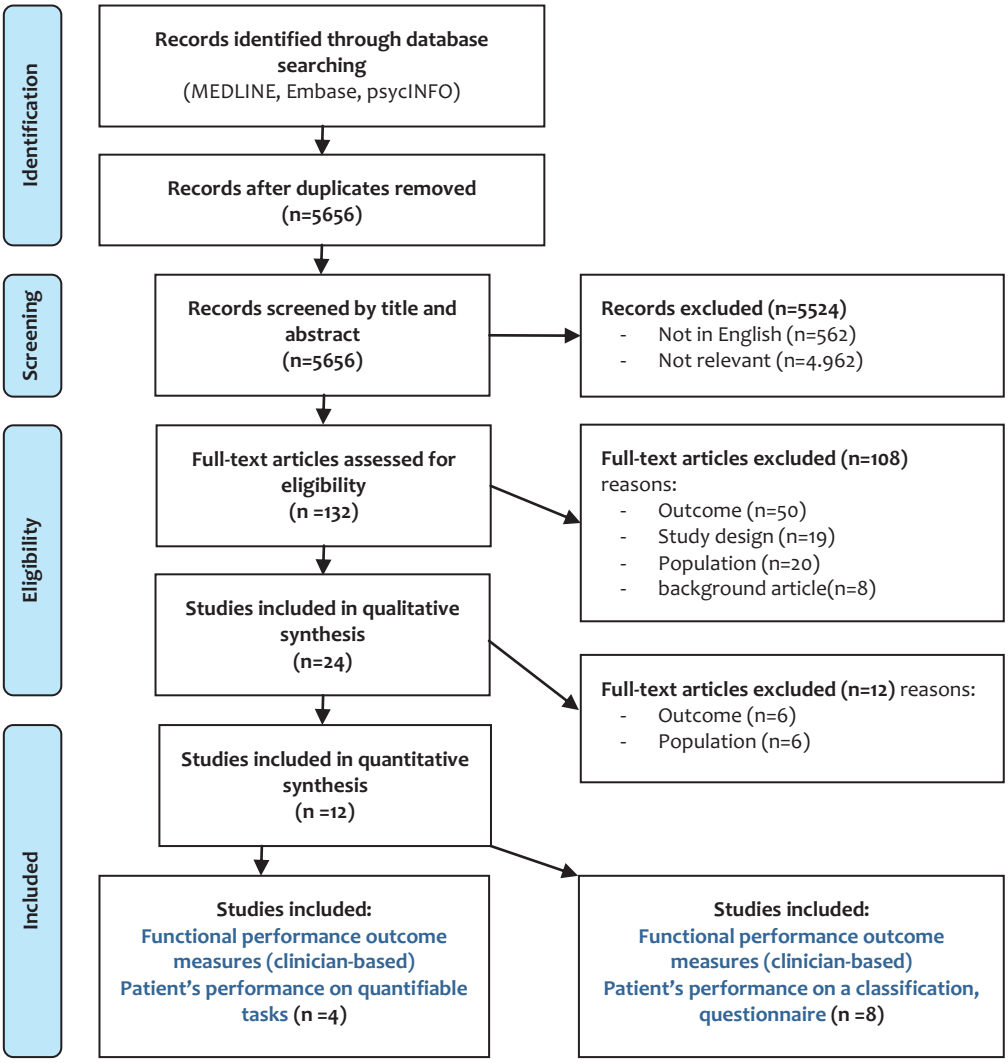

Figure 1. Flow diagram

Table 1. Summery included studies

Study	Study design	No of patients	% CP	Mean age (range)	Mean follow up (range)	Comparative sub-groups on the basis of:
Carlson 2012	Retrospective review	N=24 N=8	100%	10y (3-20y) 14y (5-20y)	22m (7-144m) 18m (6-51m)	Surgical intervention Fixed elbow deformity / contractures; less than 45° "dynamic", more than 45° "static"
Donadio 2016	Retrospective cohort study	N=20	100%	16.2y (12-17y)	22m (12-38m)	
House 1981	Retrospective review	N=56	100%	11.8y (4-20y)	2y-12y	
Libberecht 2011	Retrospective cohort study	N=15	75%	13.9y (4-30y)	23m (4-84m)	
Matsou 1990	Retrospective case series	N=19	100%	13y (6-23y)	4y (2-7y)	
Matsou 2001	Retrospective cohort study	N=26	100%	13y (9-35y)	4.5y (2-10y)	
Ponten 2011	Prospective cohort study	N=18	78%	11y (6-16y)	7m (5-14m)	
Roth 1993	Retrospective cohort study	N=17	100%	8.3y (3.3-15.5y)	2.6y (0.9-4.6y)	
Sanders 2006	Retrospective review	N=14 N=8	100%	9.9y 10.3 y	6m 12m	
Smitherman 2011	Retrospective case-control series	N=40	100%	13y1m (6.3-17.7y)	14m (7-24m)	
Van Heest 2008	Retrospective case series	N=13	100%	10.8y (7-24y)	3.6y (1-10y)	
Van Heest 2015	(pseudo) RCT	N=16	100%	4-16y	1y	Different interventions: Tendon transfer surgery, Botulinum toxin injections, Regular ongoing therapy.

Surgical (contra-) indication	Surgical technique
Indication: Dynamic elbow flexion deformities more than 30°. Contraindication: Deformity is unresponsive to therapy (botulinum A injections and occupational therapy with bracing and casting)	lengthening biceps, brachialis, BR z-lengthening biceps, sub-total myotomy of the brachialis, anterior elbow capsule release, and proximal release of the brachialis.
Indication: Contracture of the upper limb who had an arthrodesis of the wrist. Contraindication: Open growth plate	wrist arthrodesis
Indication: Deformity and imbalance of the thumb. No contraindications.	Lengthen AP, stripping 1 st DI, Lengthen FPL, PL to APL, PL+EPL, PL to EPB, BR+APL, BR+EPB, BR to EPL, FCR+APL, APL+EPL, ECRL+APL Tenodesis: APL+FCR, EPB+FCR, EPL+FCR, EPL to EPB, Fusion: MP joint, CMC joint, IP joint
No specific (contra) indications mentioned.	FCU to ECRB combined with: fractional lengthening of FL, PT rerouting, PT to EPL, PT release, FAR, FL+PT release
Indication: Deformity and rigidity of the fingers. No contraindications.	Combined release FDP + FDS + EDC
No specific (contra) indications mentioned.	Combined release FDP + FDS + intrinsic muscles.
No specific (contra) indications mentioned.	biceps lengthening, PT release, PT rerouting, FCU lengthening, FCU to EDC, FCU to ECRB, FCU to ECRL, FCR to ECRL, ECU to ECRB, BR to ECRB, PL release, finger flexor release / lengthening, thumb abduction (number of procedures) and swan neck procedures
No specific indications mentioned. Contraindications: Impaired vision and poor sensation together	pronator to supinator transfer, FCU to ECRB, BR to APL and EPB, Fusion MP
No specific (contra) indications mentioned.	No information
No specific (contra) indications mentioned, besides using the SHUEE.	FCU to ECRB, wrist and MCP arthrodesis, Thumb MCP sesamoid capsulodesis, AP release, EPL rerouting, PT lengthening, fractional lengthening FDS-FDP-FPL, BA lengthening
No specific (contra) indications mentioned.	FCU to ECRB, BR to ECRB, ECU to ECRB, FCU lengthening, flexor pronator slide, Wrist fusion
Indications: Standard for tendon transfer surgery: (1) pronation deformity; (2) deficient active wrist extension with adequate digital control, and wrist flexion deformity with the FCU as the primary deforming force; and (3) adduction of the thumb ray with flexion at the thumb MC) joint, with the inability to make a fist with the thumb outside the flexed digits. Contraindication: House score of 0, Previous upper-extremity surgery, Upper extremity botulinum toxin injections within the previous twelve months.	FCU to ECRB, PT release, EPL rerouting + AP release

Abbreviations: BR, brachioradialis; ECRB, extensor carpi radialis brevis; ECRL, extensor carpi radialis longus; ECU, extensor carpi ulnaris; EDC, extensor digitorum communis; FCR, flexor carpi radialis; FCU, flexor carpi ulnaris; PL, palmaris longus; PT, pronator teres; FDP, Flexor digitorum profundus; FDS, Flexor digitorum superficialis; FPB, Flexor pollicis brevis; FPL, Flexor pollicis longus; EPL, Extensor pollicis longus; EPB, Extensor pollicis brevis; APL, abductor pollicis longus; AP, abductor pollicis; EDC, extensor digitorum communis; FAR, flexor aponeurotic release; DI, digits; M(C)P, metacarpophalangeal; CMC, carpometacarpal; IP, interphalangeal.

Outcome measures for hand use and performance of activities

Table 2 presents the different activity-based outcome measures used to evaluate the effect of UES on the patient's hand use and performance. Clinicians evaluated the patient's performance using a "functional performance outcome measure", including quantifiable tasks in 4 studies[13,16,17,19] and a classification, interview or questionnaire in 8 studies.[14,15,18,20,21-24] The perception of the patient's ability to use the hand(s) in task performance was evaluated with the Canadian Occupational Performance Measure (COPM) in 2 studies.[13,16]

Risk of bias within studies

There was an overall high risk of bias within the included studies as shown in Table 3. In mostly all retrospective studies the risk of bias was high for the items on selection (due to lack of allocation concealment, loss to follow-up), confounding (inherently to not controllable studies, different baseline characteristics) and measurement of outcomes (blinding of outcome assessment). The two prospective studies are judged to be at serious risk of bias in at least one domain, but not at critical risk of bias in any domain.

GRADE assessment of the quality of the evidence

The quality of evidence was very low for each postoperative outcome after UES in children and adolescents with CP (Table 4.) The scarcity of comparative studies, the selection of participants and lack of blinding in outcome assessment have introduced a high risk of bias, which has been accounted for in the GRADE analysis.

Functional performance

The patient's ability to handle objects on request (capacity) was evaluated with the Shriners Hospital Upper Extremity Evaluation (SHUEE)[28] dynamic positional analysis (DPA), the Box and Block Test (BBT),[29] and the Jebsen Taylor Hand Function Test (JHFT).[30] The 3 studies using the SHUEE-DPA showed a significant improvement on the ability to perform functional tasks on request. Scoring of the Jebsen Taylor and the BBT is based on the time it takes the patient to perform the uni-manual tasks. No change after surgery was found in the speed of performing a uni-manual task performed with the affected hand. For interpretation of change knowledge about the the minimal clinically important difference (MCID) is important. Unfortunately, MCIDs are unknown for the SHUEE, JHFT and BBT. (Table 4)

The patient's performance of activities (what the patient usually does) was evaluated with the SHUEE spontaneous functional analysis (SFA) and the Assisting Hand Assessment (AHA)[25-27] and significant improvement on the spontaneous functional use was found in all 3 studies. For the AHA, a change of 5 AHA-units or more does (with 95% certainty) reflect a real change exceeding any random measurement error[37], which was seen in one study for 10 of the 18 children.[16] The individual scores of the second study were not presented.[13] (Table 4)

One study showed a improvement in functional abilities 1-year after surgery in patients with CP, evaluated with the Functional Independence Measure for Children (WEEFIM).[31,32] Hand function was evaluated before and after surgery with the House Functional Classification (HFC)[22] in 7 studies, with a range from 0 (no use of the hand) to 8 (complete

spontaneous use of the hand). The mean HFC score changed significantly in the studies suggesting that the affected arm/hand changed from a (fair) passive assist before surgery to a (fair) active assist after surgery.

Perceived performance

One study presented the outcomes based on the COPM[33-36] (performance) and showed that the children's perception of the performance of selected goals improved from 2.6 (range 1–8) to 6.4 (3–10) after surgery.[16] The other study did not report the change in COPM performance for the surgical intervention group, but reported no difference between the three intervention groups.[13]

Table 2. Activity-based outcome measures evaluating the effect of UES

Functional performance, based on:
Quantifiable tasks:
Assisting Hand Assessment (AHA)[13,16]
Shriners Hospitals Upper Extremity Evaluation (SHUEE)[13,17,19]
Box and Block Test (BBT)[13]
Jebsen Taylor Hand Function Test (JTHF)[19]
Classification, interview, questionnaire:
Functional Independence Measure for Children* (WeeFim)[14]
House Functional classification (HFC)[15,18,20-24]
Perceived performance, based on:
Canadian Occupational Performance Measure (COPM)[13,16]

Table 3. ROBINS-I risk of bias assessment

Study	Pre-intervention		At intervention
	Confounding	Selection of participants	Classification of intervention
Carslon 2012	🔴	🔴	🟠
Donadio 2016	🔴	🔴	🟠
House 1981	🔴	🔴	🟠
Libberecht 2011	🔴	🔴	🟠
Matsou 1990	🔴	🔴	🟠
Matsou 2001	🔴	🔴	🟠
Ponten 2011	🔴	🔴	🟠
Roth 1993	🔴	🔴	🟠
Sanders 2006	🔴	🔴	🟠
Smitherman 2011	🟠	🟠	🟠
Van Heest 2008	🔴	🔴	🟠
Van Heest 2015	🟠	🟠	🟢

Each domain is determined to exhibit low, moderate, serious, or critical risk of bias.

- 🟢 **Low** risk indicates that the study is "comparable to a well-performed randomized trial" in the domain being evaluated.
- 🟡 **Moderate** risk of bias indicates the study is "sound for a non-randomized study" but not comparable to a rigorous randomized trial.
- 🟠 **Serious** risk of bias indicates the presence of "important problems,"
- 🔴 **Critical** risk of bias indicates the study is "too problematic to provide any useful evidence on the effects of intervention".

The overall risk of bias of each study was equal to the most severe level of bias found of any domain

Post-intervention				
Deviation from intended interventions	Missing data	Measurement of outcomes	Selection of reported result	Overall risk of bias assessment
● red	● orange	● orange	● orange	● red
● red	● orange	● orange	● orange	● red
● red	● orange	● orange	● orange	● red
● red	● orange	● yellow	● orange	● red
● red	● orange	● orange	● orange	● red
● red	● orange	● orange	● orange	● red
● orange	● yellow	● orange	● yellow	● red
● red	● orange	● orange	● orange	● red
● red	● orange	● orange	● orange	● red
● orange	● yellow	● yellow	● orange	● orange
● orange	● orange	● yellow	● orange	● red
● green	● yellow	● yellow	● yellow	● orange

Table 4. Summary of findings: Outcomes of upper extremity surgery on hand use and performance in children and adolescents with cerebral palsy

Outcome measurement	*Studies (participants)*	Mean change in score (SD)	95% confidence interval of the mean	Statistical information provided by author	Quality of evidence*
SHUEE DPA					Very low
	40[17]	17.6 (-) (24%)	-	p<0.0001	
	13[19]	-	-	p<0.02	
	16[13]	21.6 (9.4)	15.91 to 27.32	p<0.05	
Jebsen-Taylor					Very low
	13[19]	-	-	p=0.81	
BBT					Very low
	16[13]	2.0 (5.9)	-1.85 to 4.65	p>0.05	
SHUEE SFA					Very low
	40[17]	4.0 (9%)	-	p<0.0001	
	16[13]	6.5 (11.0)	0.37 to 12.56	p<0.05	
AHA					Very low
	18[16]	6.2 (3.7)	4.34 to 8.0	p<0.0001	
	16[13]	3.1 (5.4)	0.21 to 5.92	p<0.05	
WEEFIM					Very low
	14[14]	2.8 (1.4)	-	P>0.05	
HFC					Very low
	24[15]	2 (2)	0 to 2	P<0.01	
	8[15]	2 (2)	0 to 4	P<0.01	
	20[21]	1.95 (1.4)	1.32 to 2.58	P<0.001	
	56[22]	2.7 (1.7)	0.22 to 2.23	P<0.001	
	15[18]	3.0 (-)	-	P=0.002	
	26[24]	1.7(1.7)	0.98 to 2.35	P<0.001	
	19[23]	3.2 (1.2)	2.59 tot 3.70	P<0.001	
	17[20]	1.8 (-)	-	P<0.001	

***** Grades of Recommendation, Assessment, Development and Evaluation

Discussion

This systematic review showed that there is very low quality evidence for the effectiveness of UES on hand use and performance of activities in children and adolescents with CP. The methodological quality of the 12 included studies was poor and evidence for comparative effectiveness is limited.

Previously performed reviews demonstrated positive effects of UES in children and adolescents with CP on upper limb function (i.e. dexterity, range of motion at the wrist and forearm and grip strength). [38-40] Our review was performed to evaluate the effect on patients' functional and perceived performance measured with validated activity-based outcome measurements rated by clinicians or patients /caregivers. The patient's functional performance was measured in 4 studies on quantifiable tasks with the SHUEE, AHA, JHFT or BBT and in 9 studies by classification, interview or questionnaire, with the WEEFIM or HFC. Four outcome measures (SHUEE, AHA, WEEFIM and HFC) showed significant improvement in all included studies. The HFC was the most used functional performance outcome measure to evaluate the effect of UES. The JHFT and BBT showed no significant improvement after UES, evaluating the ability to perform uni-manual standardised tasks by recording the time needed to perform the tasks. The results of the JHFT and BBT may suggest that speed and/or the ability to perform activities with one hand (the affected hand), do not change after surgery.

Limitations

The findings of this systematic review should be interpreted in the context of its limitations. There was substantial heterogeneity of UES-interventions ranging from a multi-level surgery to a single release of the palmaris longus and of activity-based outcome measures. Then, the retrospective case series design used in most studies means the evidence is initially rated low, and has a high risk of bias, like recall and publication bias which may have limited the available evidence.

Many studies were excluded on the used outcome measure as they used unique and individualized, often qualitative outcome measures. Most of these studies used a qualitative method to evaluate the outcome. For example, daily activities were evaluated as a qualitative measure of function or a questionnaire was used in which subjects were asked about the change in hand use after UES. A lack of reported statistical precision (e.g. no estimates of effects with confidence intervals) was found within the included studies, making pooled analyses impossible.

Implications for practice and research

Considering the overall very low quality of the evidence, it is not possible to make clinical recommendations on the effect of UES. However, the positive results of the effect of UES on hand use and performance presented in all included studies certainly justify the performance of controlled studies.

Different validated activity-based instruments were used in the included studies and both clinicians and patient/caregivers were positive about the effect on hand use and performance of activities after UES. To enhance the comparability of studies and perform pooled analysis, the use of a core set of instruments to gather and share data in international databases is recommended. This information will be of importance to improve

selection criteria, to indicate specific surgical procedures and to predict outcomes for UES. Based on the findings of this review it is suggested to add at least the 3 activity-based outcome measures to the core set when evaluating the effect of UES; including AHA, SHUEE DPA and COPM. The SHUEE DPA and AHA because they evaluate the use of both hands together, the SHUEE DPA has more components of measuring capacity and AHA of measuring performance. Future studies should also focus on the perceived performance when evaluating the effect of UES on hand use and performance. The COPM can be used to measure patient related outcomes on both perceived performance and satisfaction of the ability to perform the activity. Besides these activity-based outcome measures, more assessments on all ICF levels ("body function and structure", "activity and participation") are needed to recognize the impact of the changes after UES on patients' function and possibilities to engage fully in their lives.

Details on the post-operative therapy are reported in only a few studies. To be able to interpret the effect of UES it is necessary to have consensus about the post-operative therapy, because the use of different post-operative therapies is likely to influence the outcomes after UES on hand use. Future studies should compare post-operative therapy to identify the optimal frequency, duration, intensity and focus (improve function and/or performance of activities). A period of post-operative casting will always follow after UES. However, there is a lack of knowledge about the effect of casting on the results after UES. More knowledge is needed about aspects like the optimal (over-corrected) position of the arm/hand and time of cast removal.

Furthermore, future studies should evaluate long-term outcomes. It is not well known if the UES interventions continue to add benefit over years or whether the gains are lost when aging. More high quality comparative effectiveness studies are needed to be able to determine optimal patient selection criteria and indications for specific UES-procedures.

Conclusions

This systematic review found very-low-quality evidence that UES can improve the patient's hand use and performance of activities of children and adolescents with CP. More high quality studies are needed to obtain more insight in the comparative effectiveness of UES on hand use and performance in children and adolescents with CP.

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Appendix 1. Search strategy

Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily and Ovid MEDLINE(R) <1946 to Present>

Search date: 3 April 2017

#	Searches	Results	After deduplication
1	cerebral palsy/ or (exp upper extremity/ and (paralysis/ or hemiplegia/))	20752	
2	(cerebral palsy or upper limb spasticity or (brain damage and (hand adj3 function*)) or spastic hemiplegia).ab,kf,ti.	19623	
3	1 or 2	26584	
4	(lengthening or slide? or release or rerout* or stabilization or capsulodesis).ab,kf,ti.	628218	
5	(surgery or surgical* or treatment or therapy or therapeut*).ab,hw,kf,ti. or th.fs.	7171856	
6	(upper extremit* or upper limb* or hand? or shoulder? or elbow? or wrist? or thumb? or finger?).ab,hw,kf,ti.	602756	
7	5 and 6	206257	
8	4 or 7	825483	
9	3 and 8	2930	
10	remove duplicates from 9	**2849**	**2847**

Embase Classic+Embase <1947 to 2017 March 31> (Ovid)

Search date: 3 April 2017

#	Searches	Results	After deduplication
1	cerebral palsy/ or (exp *upper limb/ and (*paralysis/ or *hemiplegia/))	35752	
2	(cerebral palsy or upper limb spasticity or (brain damage and (hand adj3 function*)) or spastic hemiplegia).ab,kw,ti.	28025	
3	1 or 2	38247	
4	(lengthening or slide? or release or rerout* or stabilization or capsulodesis).ab,kw,ti.	799955	
5	(surgery or surgical* or treatment or therapy or therapeut*).ab,hw,kw,ti.	10004692	
6	("7" or "8" or "9" or "33" or "34").ec.	4489958	
7	(upper extremit* or upper limb* or hand? or shoulder? or elbow? or wrist? or thumb? or finger?).ab,hw,kw,ti.	848640	
8	(5 or 6) and 7	445588	
9	4 or 8	1229066	
10	3 and 9	4886	
11	remove duplicates from 10	**4693**	**2664**

PsycINFO <1806 to March Week 4 2017> (Ovid)

Search date: 3 April 2017

#	Searches	Results	After deduplication
1	cerebral palsy/ or ((arm/ or shoulder/ or wrist/ or elbow/) and (paralysis/ or hemiplegia/ or hemiparesis/))	4586	
2	(cerebral palsy or upper limb spasticity or (brain damage and (hand adj3 function*)) or hemiplegia).ab,id,ti.	7615	
3	1 or 2	7893	
4	(lengthening or slide? or release or rerout* or stabilization or capsulodesis).ab,id,ti.	45392	
5	(surgery or surgical* or treatment or therapy or therapeut*).ab,hw,id,ti.	814942	
6	(upper extremit* or upper limb* or hand? or shoulder? or elbow? or wrist? or thumb? or finger?).ab,hw,id,ti.	97731	
7	5 and 6	15450	
8	4 or 7	60536	
9	3 and 8	467	
10	remove duplicates from 9	**467**	**148**

Effects of upper extremity surgery on manual performance in children and adolescents with cerebral palsy: a multidisciplinary approach using shared-decision making

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Submitted

Abstract

Background

Little is known about the effects of upper extremity surgery (UES) on the manual performance of children and adolescents with cerebral palsy (CP). This clinical cohort study describes our experience with patient selection based on multidisciplinary assessment and shared decision-making (SDM) and the effects of UES on manual performance and patient-relevant outcomes.

Methods

All patients (up to 20 years of age) with CP referred to our multidisciplinary team for UES evaluation between July 2011 to May 2017 were included. Suitability for UES was assessed based on a comprehensive, multidisciplinary screening and the decision to proceed with surgery was made together with the patient. Individual patient-relevant goals were identified with the Canadian Occupational Performance Measure (COPM); perceived independence in performing bimanual activities at home was assessed with the ABILHAND (-kids), and perceived quality of use of the affected hand during daily activities was assessed with a Visual Analog scale (VAS). The quality of use of the affected hand during bimanual performance was measured with the Assisting Hand Assessment (AHA) and gross manual dexterity with the Box and Block Test (BBT). All baseline assessments were repeated 9 months after-surgery.

Results

Of 66 patients assessed by the multidisciplinary UES team, 44 were considered eligible for UES. Of these patients, 39 underwent UES and were evaluated in the pre-post study (mean age 14y9m [SD 2y10m], 87% unilateral CP, 72% MACS level II). All outcomes improved significantly after UES: COPM-Performance +3.2 (SD 1.6) ($p<0.001$), COPM-Satisfaction +3.3 (SD 2.1) ($p<0.001$), ABILHAND + 1.5 (SD 1.2) logits ($p<0.001$), AHA +6.7 (SD 4.2) units ($p<0.001$), and BBT +2.3 blocks/minute (SD 4.9) ($p=0.021$). The improvement in COPM-P, COPM-S, ABILHAND, AHA, and BBT performance was clinically meaningful in 80%, 77%, 55%, 71%, and 31% of patients, respectively.

Conclusion

Careful assessment of eligibility for UES, based on multidisciplinary screening and SDM, resulted in a clinically relevant improvement in patient-specific functional and/or cosmetic goals and manual performance after UES in most patients with CP.

Introduction

Spastic cerebral palsy (CP) results in typical disabling deformities of the upper limb and abnormal hand posture that affect the ability to grasp and release objects and to use both hands together.[1,2] Affected individuals often need assistance to perform everyday activities.[3] Different surgical and nonsurgical interventions are available to improve the ability to use the affected hand effectively during daily activities. Yet their effectiveness is not clear because studies have involved heterogeneous patient populations, few or no patient-relevant outcomes, and ambiguous selection criteria for surgical treatment.

The aim of upper extremity surgery (UES) is to improve muscle balance and hand posture in order 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. The most common procedures for this are: (I) release of pronator teres muscle to facilitate forearm supination[4,5], (II) release or transfer of the flexor carpi ulnaris tendon to increase wrist extension for functional grip purposes[6], (III) correction of a thumb-in-palm deformity, preferably and if possible by adductor pollicis muscle slide[7] combined with extensor pollicis longus rerouting.[8]

Most studies have assessed UES outcomes using a functional classification scale (for example the "House functional classification"[9,10] and/or instruments assessing wrist/thumb positioning, muscle strength, range of motion, and selective motor control.[11-14] However, improved functions has not been shown to be associated with a higher ability to perform daily activities.[15-17] In order to provide patients and professionals with relevant information to enable them to understand the risks, benefits, and outcomes of the various treatment options, more needs to be known about the pros and cons of UES and whether surgery improves the ability to handle objects and perform patient-relevant everyday activities. Appropriate patient selection is important and is preferably done by a multidisciplinary team of professionals and with shared-decision making (SDM).[18] A SDM approach will help the patient and UES team use evidence-based information to come to the best possible treatment decision. This approach encourages patients to express their values and preferences.

This clinical cohort study describes our experience with patient selection for UES, based on a multidisciplinary assessment and SDM, and the effects of UES on manual performance and patient-relevant outcomes in a consecutive series of children and adolescents with unilateral CP.

Methods

Multidisciplinary approach and shared decision-making

The clinical cohort study was performed in a tertiary referral center for UES for patients with CP in the Netherlands and included all patients (up to 20 years of age) with CP who were consecutively referred to our UES team for UES consideration between July 2011 and May 2017. The multidisciplinary UES team consisted of a hand surgeon, pediatric rehabilitation physician and occupational therapist. Each patient was assessed and discussed by the UES team during a single visit in order to reach a shared-decision about whether or not to proceed with surgery.

First, the occupational therapist collected information about patient-specific goals, perceived independence to execute bimanual activities at home, perceived quality of use of the affected hand during daily activities, spontaneous use of both hands, and gross manual dexterity. Then the patients were seen by the hand surgeon and pediatric rehabilitation physician for a medical examination, which included assessment of active and passive range of motion, grip and pinch strength, voluntary and selective motor control, spasticity, and the presence of involuntary movements. To make the best possible treatment decision, the following criteria were assessed and discussed with the patient and their parents (Fig. 1): (I) absence of dystonia or athetosis, because in these types of movement disorders the imbalance of muscles is variable and the outcome of surgery is unpredictable; (II) achievable patient-specified goals (functional and/or cosmetic); and (III) absence of developmental disregard, i.e. a large discrepancy between the ability to use and the actual use of the affected hand.[19,20] The UES team helped the patient compare treatment options and expected outcomes and reached a shared decision with the patient. If UES was not indicated because criterion II or III was not met, alternative conservative treatment options were discussed with the patient, with the possibility for future re-assessment by the UES team.

Assessment of activity limitations

Patients were asked to set five patient-specific "hand-use oriented" goals based on the problems they identified on the Canadian Occupational Performance Measure (COPM) and rated their performance and satisfaction on a scale from 1 to 10 [21-24], with a minimum clinically important difference [MCID] of >2.[25] The perceived independence in performing bimanual activities at home was assessed using the patient/parent-reported questionnaire of unilateral and bilateral activities, ABILHAND(-kids).[26,27] In this study, we considered the smallest detectable change of 1.18 logits to reflect a moderate clinically significant change.[28,29] The self-perceived "ease of use" of the affected hand when performing daily activities was scored on a visual analog scale (0–10 cm) of 0 to 10). The VAS is sensitive for measuring changes associated with treatment or time and has a MCID of 1.37 cm.[30]

The quality of use of the affected hand during bimanual performance was measured using the Assisting Hand Assessment (AHA). Object-related hand actions were scored during a semi-structured test situation that elicits the use of both hands. Two different age-related test activities were used to score the AHA 5.0 [31] , namely, the 'School-Kids AHA' and 'Ad-AHA Board Game'.[32,33] A score change of 5 AHA units or more (with 95% certainty) reflects a real change exceeding any random measurement error.[33,34] The ability to use the affected hand was assessed with the Box and Block Test (BBT), in which the patient is asked to transfer, with the affected hand, as many blocks as possible from one box to another in 60 seconds.[35,36] A change exceeding at least 6 blocks/minute (measured by the same rater) can be interpreted as a true change (with 95% certainty)[37,38]

Manual performance was categorized using the Manual Ability Classification System (MACS).[39]

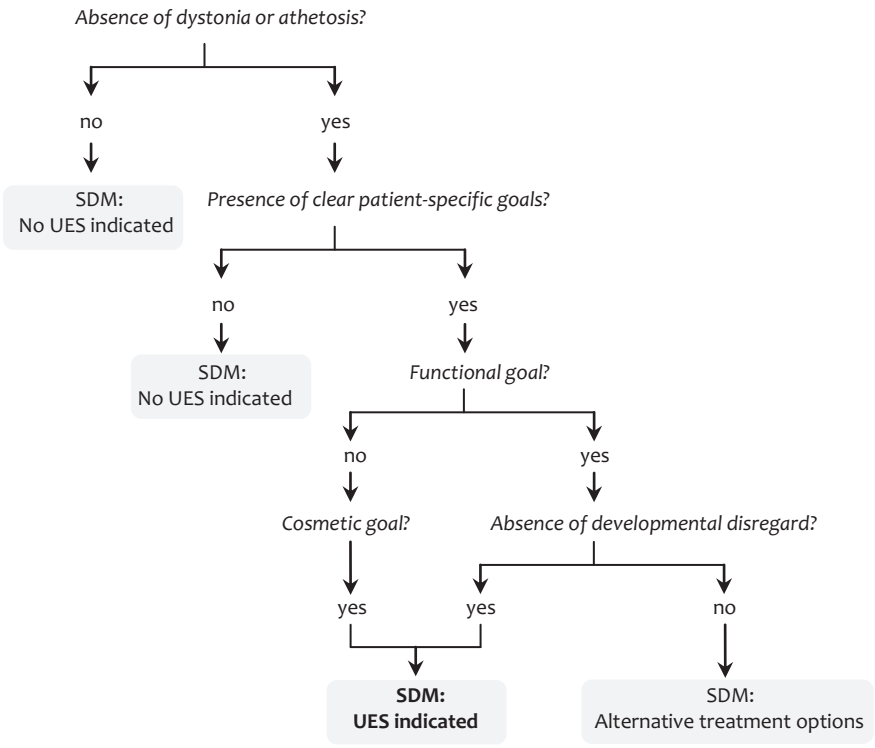

Figure 1. Decision-tree for upper extremity surgery (UES) in patients with cerebral palsy, based on multidisciplinary assessment and shared decision-making (SDM)

Intervention and postoperative therapy

Different UES procedures were executed (Table 2). After UES, the upper limb was immobilized for 5-6 weeks. After cast removal, day/night resting hand orthoses were used in conjunction with therapy to maintain the increase in muscle length achieved with UES and to facilitate improved motor control. The postoperative therapy was supervised by the patient's own rehabilitation physician and therapist. All baseline assessments were repeated 9 months after surgery.

Statistical Analyses

Paired samples t-tests were performed to compare pre- and postoperative scores on all measures. The level of significance was set at 5% ($P<0.05$). All analyses were performed with SPSS software, version 21.0 (IBM Corp., Armonk, NY, USA).

Ethics

Before study entry, all patients (>12 years of age) and/or their primary caregivers gave informed consent for the use of their medical data. Participation was voluntary, and all participants were informed that they could withdraw from the study at any stage. The Medical Ethics Committee of our hospital waived the need for ethical approval, because all assessments and interventions were an integral part of standard care.

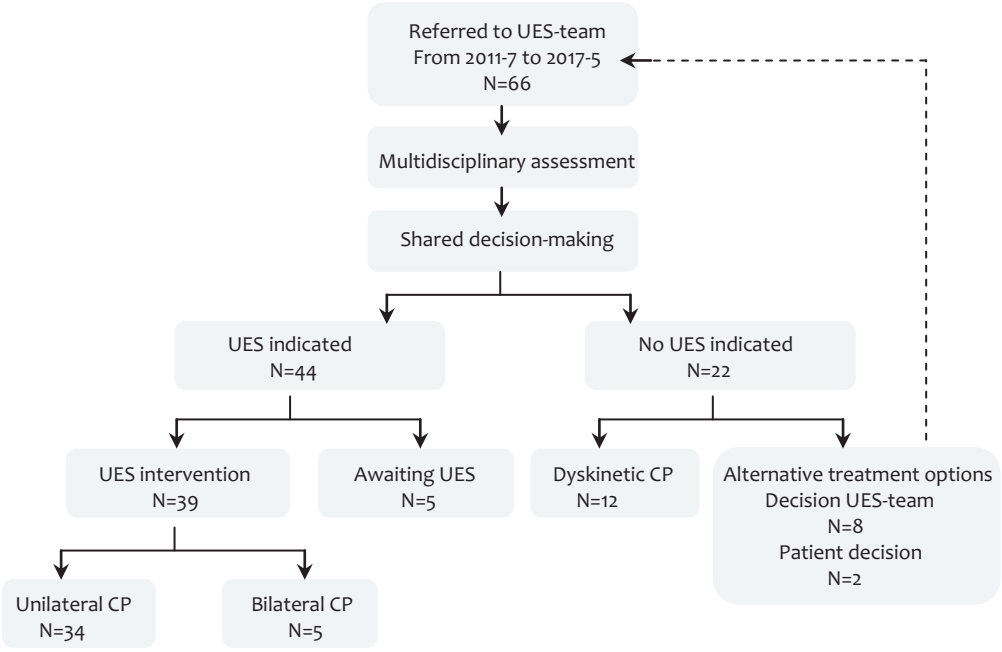

Figure 2. Flow diagram for assessment of patients with cerebral palsy (CP) eligible for upper extremity surgery (UES)

Results

Between July 2011 and May 2017, 66 patients with CP were consecutively referred to our multidisciplinary UES team, 44 of whom decided to undergo UES (Fig. 2). The results of 5 patients were not available at the time of the study, so the data of 39 patients were used. They were examined on average, 4 months (range 0 - 15 months) before surgery and 9 months (range 6 - 11 months) after surgery. The age of the included patients ranged between 7 years 10 months and 19 years 7 months. Twenty-two patients were not selected for UES after multidisciplinary assessment and SDM, 2 of whom preferred a non-surgical intervention despite being considered eligible for UES by the UES team (Table 1).

Eligibility for UES was based on the extensive multidisciplinary assessment. The COPM identified patient-specific "hand-use oriented" goal(s), which were linked to the different surgical procedures (Table 2). Information from the Abilhand, AHA, and BBT was considered to obtain insight into the ability to use the affected hand and the likelihood that COPM goals could be achieved. For example, the goal of one of the adolescents was to be

able to move and position coins with the affected hand without the help of the other hand. The UES team explained that the patient probably would not be able to achieve this goal because the pre-surgery score on the AHA showed that the affected hand could function as assisting hand but would not be able to stabilize objects effectively. This complex movement requires the independent movement of fingers, which would not be achieved with UES. Together with the UES team, the patient refined his/her goals and expectations. The UES team advised 8 patients to start preoperative therapy in order to stimulate the actual use of the hand during daily activities. This advice was given to 2 patients because there was a discrepancy between the actual use of the affected hand (measured by the Abilhand and AHA) and the ability to use the affected hand (measured by the BBT), and to 6 patients because alternative, non-surgical treatments had not been tried previously (Fig. 2).

Table 3 presents the mean differences between pre-and postoperative assessments and the proportion of patients achieving clinically meaningful improvement in the self-reported outcomes (COPM, ABILHAND, and VAS), unimanual capacity (BBT), and in the case of unilateral CP in manual performance (AHA). All outcomes showed a statistically significant improvement at 9 months. 80% and 77% of the participants had a clinically significant improvement in COPM performance and satisfaction, respectively and 71% on manual performance (AHA) (Table 3). Patients whose primary goal was to improve the appearance of the affected upper extremity also showed improved manual performance and patient-specific functional goals (Table 4).

Table 1. Characteristics of all 66 children/adolescents with cerebral palsy assessed for upper extremity surgery (UES)

		No-UES indicated	**No-UES indicated *(yet)***	**No-UES *Patient-decision***		**UES indicated**	
Characteristics		**n=12**	**n=8**	**n=2**	**n=5**	**n=34**	**n=5**
Gender	Male	5	3	2	1	16	4
	Female	7	5	0	4	18	1
Mean age		15y4m	13y1m	16y1m	14y9m	14y8m	15y2m
Bilateral		6	5	1	-	-	4
Unilateral		6	3	1	5	34	-
Affected side	Left	3	1	1	1	18	-
	Right	3	2	-	4	16	-
Spastic		3	8	2	5	34	4
Dyskinetic		9	-	-	-	-	-
MACS	1	2	3	-	-	2	-
	2	3	2	-	3	27	1
	3	2	1	2	2	5	1
	4	1	1	-	-	-	2
	5	1	1	-	-	-	1
	missing	3	-	-	-	-	-

Table 2. Patient-specific "hand-use oriented" goals linked to the surgical options

Examples of patient-specific "hand-use oriented" goals	To facilitate:		Type of surgery performed	No of patients
Combing long hair	Elbow extension	⇨	Z-lengthening of the biceps tendon and/or brachialis muscle slide[22]	2
Washing oneself (in the shower)	Forearm supination	⇨	Pronator teres release[23,24]	14
Holding a plate	Wrist extension	⇨	Release or transfer of the FCU tendon[25]	30
Making the bed	Thumb abduction and opposition	⇨	Adductor pollicis muscle slide[26] combined with an EPL rerouting[27] (correction of a thumb-in-palm deformity)	30
Carrying a box (with 2 hands)				
Holding paper while writing	Finger extension (while wrist is extended)	⇨	Fractional lengthening of the extrinsic finger flexor muscles	8
Washing and drying dishes	Joint stabilization of thumb MCP	⇨	Capsulodesis procedure thumb MCP[28] (by hyperextension)	13
Holding game controller				
	Stabilization of the fingers	⇨	Stabilization of Swanneck deformities in the fingers[29]	10

Table 3. Performance on different measures of outcome pre-intervention and 9 months post-intervention

	N	Pre-intervention	Post-intervention 9-month	Δ Mean difference (SD)	p-Value	Clinically meaningful improvement (% of the participants)	Above smallest detectable change (% of the participants)
COPM-P (SD)	35	3.4 (1.3)	6.5 (1.7)	3.2 (1.6)	<0.001	80%	
COPM-S (SD)	35	3.5 (1.7)	6.6 (1.7)	3.3 (2.1)	<0.001	77%	
ABILHAND-logits (SD)	34	0.9 (1.9)	2.4 (1.8)	1.5 (1.2)	<0.001		55%
VAS (SD)	29	3.3 (2.0)	5.7 (1.9)	2.4 (1.9)	<0.001	62%	
AHA-units (SD)	31	49.7 (10.9)	56.4 (11.2)	6.7 (4.2)	<0.001		71%
BBT (SD)	29	19.8 (9.9)	22.1 (10.4)	2.3 (4.9)	0.021		31%

SD=standard deviation; COPM-P: Canadian Occupational Performance Measure (COPM) performance, COPM-S: Canadian Occupational Performance Measure (COPM) satisfaction, VAS: Visual Analog scale, AHA; Assisting Hand Assessment , BBT: Box and Block Test

Table 4. Patient-specific functional and cosmetic goals

Order of importance to participant:	n	MACS level I	II	III	Clinically meaningful improvement n (%) of the participants: COPM-P	COPM-S	BBT	AHA	ABILHAND	VAS	Satisfied about cosmetic result
1. Functional goals 2. no cosmetic goal	15	0	10	5	10 77%	10 77%	5 39%	10 77%	9 69%	5 39%	N.A.
1. Functional goals 2. Cosmetic goal	14	1	10	3	8 89%	8 89%	1 11%	4 44%	2 22%	4 44%	77.8%
1. Cosmetic goal 2. Functional goals	8	1	7	0	6 86%	6 86%	2 29%	7 100%	4 57%	4 57%	100%

Discussion

The present study shows that a multidisciplinary approach using SDM to select patients likely to benefit from UES resulted in clinically relevant improvements in manual performance and patient-relevant outcomes after UES in more than 80% of the assessed children and adolescents with CP. Thus this approach appears to be effective in selecting patients who may benefit from upper extremity surgery.

The comprehensive multidisciplinary assessment and strict selection criteria contributed to the effectiveness of surgery in this patient population (87% unilateral spastic CP, 72% MACS level II), based on the improvement in manual performance. Given the high proportion of successful outcomes, it can be questioned whether our selection criteria for UES were too strict and more patients might benefit from surgery. For instance, manual performance may also improve after UES in patients with a low ability to handle objects (MACS level III, IV) or only a few limitations (MACS level I). In particular, patients with MACS level III or IV may benefit from the improved muscle balance and more normal hand posture achieved with UES, followed by rehabilitation interventions, such as intensive bimanual training. Even small changes in muscle balance may change a non-functional hand into a hand that is able to hold objects passively or stabilize objects, as measured with the AHA.[31,33] In the current study, as most patients showed a clinically significant improvement, it was not possible to refine selection criteria based on the characteristics of patients who did not show such improvement. Future research should evaluate whether adapting the UES selection criteria leads to improvement across all MACS levels.

Although it was not possible to distinguish the specific effects of SDM on UES outcomes, this study showed that involving patients in treatment decisions was beneficial. This enabled the UES team to align surgical and postoperative treatment plans and to achieve the best outcome in terms of patient-specific goals. Only two patients preferred nonsurgical treatment. In both cases, the choice was based on a lack of motivation for the intensive postoperative rehabilitation. Prior to UES, the feasibility of achieving patient-specific goals was carefully discussed, so that patients had clear and realistic expectations about what could be achieved, which resulted in a high level of satisfaction with the functional and cosmetic outcomes. Our findings show that selection of the appropriate procedure is linked to the achievement of patient-relevant goals.

A high proportion of patients achieved clinically meaningful improvements in self-reported outcomes (COPM, ABILHAND and VAS), unimanual capacity (BBT), and in the case of unilateral CP in manual performance (AHA). However, it should be appreciated that results for different outcome measures may vary, because of differences in patient-specific goals, choice of surgical procedure, and manual performance before surgery. For example, while manual performance did not improve in some patients, these patients experienced that they could use the affected hand more effectively in terms of their patient-relevant outcomes and vice versa. These findings confirm previous evidence that the evaluation of UES should be based on different outcomes.[16,17] The instruments used in this study measured different constructs and were complementary, which makes them suitable for selecting patients eligible for UES and for evaluating the effect of surgery.

In our study, 59% of the patients (22 out of 37) had a cosmetic goal for UES, such as "hope my hand looks more normal and/or less noticeably different", no flexed wrist, thumb out of the palm, and correction of Swanneck finger deformities. These patients were embarrassed by the appearance of the spastic hand and often used to hide their hand

behind the back, in pockets, or under the table, with as consequence that the affected hand was not used to perform daily activities. This might explain why, in this study, all patients who were pleased with the cosmetic appearance of their hands after surgery also showed improved manual performance and patient-specific functional goals. This is one of the reasons it is important not to underestimate the psychosocial impact of a hand deformity[12] and the effect on manual performance.

On the basis of these findings, it would be advisable to use a core set of instruments to assess the effect of UES, to predict outcomes, and to improve selection criteria. Use of a standard set of instruments would also make it easier to compare the results of different studies. In addition to the instruments used in this study (COPM, AHA, ABILHAND and BBT), a validated instrument measuring patient satisfaction with cosmetic appearance will add important information to the patient-specific goals. A limitation of this study is that instruments that could link function and activity were not used. Therefore, as a last addition, the "Shriners Hospitals Upper Extremity Evaluation" dynamic positional analysis (SHUEE-DPA)[40] should also be included. The SHUEE-DPA analyzes body function (in five different segments: thumb, finger, wrist, forearm, and elbow) by describing the position of the segment during 16 different activities. This would make it possible to measure and link "function" and "activity" when evaluating the result of UES.[16, 41-43]

A limitation of this study is its observational design and lack of control group when evaluating the effect of UES. Although a randomized controlled trial (RCT) is the best trial design for determining treatment effectiveness, different factors, such as the permanence and invasive character of the surgery and patient preferences, may limit the feasibility of RCTs in children and adolescents. Only a few trials of varying quality have reported the effect of UES on manual performance. [16, 41-44] Instead of RCTs, sound observational studies may be the next best method to evaluate the effect of UES. "Real-life" studies may help intervention planning, by describing the selection process, the personalized intervention, and patient-relevant outcomes. Additional high-quality observational studies of the effectiveness of UES are needed. Another limitation of this study was that different pediatric rehabilitation physician and therapists carried out the intensive postoperative functional program and its content was not monitored. A standardized postoperative hand therapy program that can be customized to patient-specific goals is needed in order to optimize the outcome of UES. Because of the small number and heterogeneous characteristics of the children and adolescents with CP referred to each UES team, preference should be given to a multicentre study. With a larger study, preferably a randomized controlled trial with a waiting-list control group and a longer follow-up, it should be possible to define patient selection criteria (based on e.g. age, severity) and to predict and achieve optimal outcomes that match patient-specific goals and expectations.

Conclusions

This study showed that careful patient selection, based on multidisciplinary assessment and SDM, results in clinically relevant improvements in patient-specific functional and/or cosmetic goals and manual performance after UES in children and adolescents with CP.

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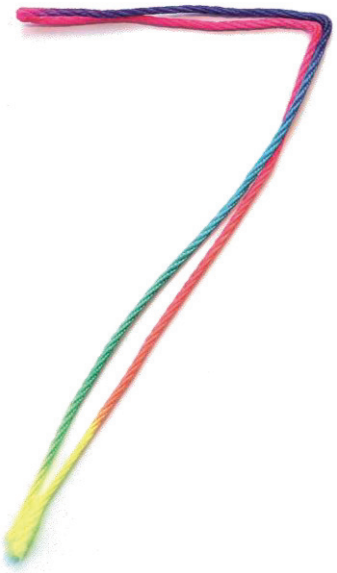

General discussion

Cerebral Palsy (CP) influences the acquisition of age-appropriate independence and may negatively impact the development of the skills that children and adolescents with CP need in order to participate in activities that are important to them in the home, school and community. Different rehabilitation programmes targeting the upper extremity in CP are used to improve hand use and the performance of patient-relevant tasks.[1,2] The general aim of this thesis was to improve knowledge regarding the assessment and management of upper extremity functioning in children and adolescents with CP. The studies involved children and adolescents whose neurological dysfunction and upper limb musculoskeletal impairments significantly affected their ability to use their hands to perform daily activities.

In this last chapter, the main findings of the studies are critically discussed, and implications for clinical practice and future research are given.

Main findings

Assisting Hand Assessment for Adolescents: evidence of validity and reliability

The Assisting Hand Assessment (AHA) measures and describes how effectively children with unilateral disability use their affected hand to perform tasks requiring the use of both hands. The first part of this thesis showed that the newly developed test activity, the Ad-AHA board game 'Go with the Floe' (Fig. 1), can be used in clinical practice to elicit bimanual performance in adolescents with unilateral CP.[3] To evaluate the construct validity of the AHA test items, all 126 assessments of adolescents were scored using the Kids-AHA version 5.0 scoring criteria containing 20 items (AHA 5.0 scale).[4] The AHA scale for the age range of our sample (10 to 18 years) functioned as a unidimensional measure that met quality criteria, as assed using Rasch measurement model,[5] and therefore we combined our data with data from an earlier AHA study involving children aged 18 months to 12 years with unilateral CP.[4] With the combined sample (290 assessments) the scale demonstrated good construct validity, which means that the same AHA scoring criteria can be used for children and adolescents aged 18 months to 18 years (AHA 18-18) with unilateral CP (chapter 2). Four different test activities can be used (School-Kids AHA, Ad-AHA Board Game, Ad-AHA Present, and Ad-AHA Sandwich) to elicit bimanual actions involving the same underlying trait. Chapter 3 showed that the test scores for different test activities were comparable, demonstrating that reliable AHA scores can be generated when using different age-appropriate tests situations for children and adolescents aged 18 months to 18 years.[6] The excellent measurement agreement implies that the Ad-AHA can be used to evaluate change over time, with a score change of 5 AHA units indicating a change beyond measurement error with 95% certainty, using the AHA 5.0 scale (chapter 3).

With the newly developed Ad-AHA, we fulfilled the need for a measurement tool to assess how effectively adolescents use their affected hand in bimanual activities.[3] According to the 'International Classification of Functioning, Disability and Health' (ICF), assessment of functioning in the domain "activities and participation" should address capacity as well as performance.[7] Capacity refers to a person's functioning in a standardized environment (can do) whereas performance refers to the performance in a real life environment (does do).[7]

So far, tools for assessing capacity and perceived performance were only available for use in adolescents with CP. Capacity can be measured with the Melbourne Assessment, the Box and Block Test, the Jebsen-Taylor Hand Function Test and the Shriners Hospital for

Children Upper Extremity Evaluation (SHUEE), whereas perceived performance can be measured with the Abilhand questionnaire, the Canadian Occupational Performance Measure (COPM), and the Children's Hand-use Experience Questionnaire (CHEQ).[8-11] Studies have shown that the SHUEE[12] can be used to assess predominantly capacity, but it also measures the performance of activities.[8-11] The SHUEE was developed to assist in targeting and measuring the outcome of spasticity and surgical management.[13-15] Besides analysing grasp and release, it also assesses the spontaneous functional use of the upper extremity during standardized simple tasks (e.g. taking money from a wallet, tearing paper).[16] The added value of the (Ad-)AHA is that it measures and describes how effectively patients use their affected hand in bimanual performance.[4,3,17-19] To ensure that the person's spontaneous use is measured, the new Ad-AHA presents the test activity as a board game, which requires bimanual handling of objects and is age appropriate and familiar to most adolescents. While the adolescent is playing the game, the therapist is allowed to interact and help if needed to keep and make it fun and engaging so as to elicit typical/habitual performance. This way of presenting an activity ensures that functioning is observed in an objective manner, without the subject even realizing that the activity is being assessed.[3] Our study in chapter 3 showed that the different test activities are age appropriate and can be used interchangeably in adolescents with unilateral CP, which makes it more attractive to adolescents and therapists, which in turn contributes to actual (spontaneous/habitual) performance.[3] The excellent agreement between scores obtained with different test activities (School-Kids AHA, Ad-AHA Board Game, Ad-AHA Present, and Ad-AHA Sandwich) indicates that different test activities can be used to evaluate change over time (Chapter 3). It should be kept in mind that only changes ≥5 AHA units can be interpreted as real changes beyond measurement error, which suggests that the AHA may not be sensitive enough to detect clinically relevant changes at the individual level.[6] Information on the minimal clinically important difference for the AHA is lacking but from clinical experience it is known that smaller changes can be very meaningful to the child and that the size of a meaningful change may vary across different levels of ability. The minimal clinically important difference needs further study.

Functional hand orthosis

The second part of this thesis showed that a functional hand orthosis (static bracing of the wrist and thumb) immediately improved the spontaneous use of the affected upper limb for bimanual activities in selected children with spastic unilateral CP (chapter 4).

Since this study, two studies with higher levels of evidence that evaluated the effect of functional hand orthoses have been published.[21] These two randomized controlled trails (RCTs)[22-24] found a functional hand orthosis not to have an effect on capacity, assessed using the "Melbourne Assessment quality of upper limb movement"[22] and the Box and Blocks Test (BBT).[21] These findings seem to be in contrast with our findings, but are not directly comparable because we assessed predominately the typical/habitual performance of activities and not the capability to execute activities. In agreement with the findings of these RCTs, we found large individual differences in the effects of using the orthosis. This stresses the need for individualized tailored assessment and therapy. The item hierarchy of the items of the AHA provide insight into the type of activities for which the individual child will most likely benefit from using the orthosis (chapter 4). Our study and the two RCTs evaluated the effects of intermittent use of a functional hand orthosis, so no conclusion can

be drawn about the effects of intensive use (during the whole day) of an hand orthosis in the short and long term. Little is known about the long-term effects of a hand orthosis because of the scarcity of relevant studies, but positive and negative results of long-term use can be anticipated. One study observed decreased activation of the forearm muscles during grip and suggested that bracing for a long period may lead to atrophy of the forearm muscles.[25] However, this adverse long-term effect was less apparent when a functional hand orthosis was used intermittently. It is expected that long-term intermittent use a functional hand orthosis will result in an increase in hand use and performance, when combined with goal-directed training, the value of which has been demonstrated by Elliot et al.[23] However, further studies are needed to obtain insight into the long-term effectiveness of wearing a functional hand orthosis in combination with training and, in possible side effects.

Upper extremity surgery

Our review (chapter 5) showed that there is insufficient evidence to recommend upper extremity surgery to improve hand use and performance in children and adolescents with CP. The quality of evidence was very low for each postoperative outcome after upper extremity surgery, mainly because of heterogeneous surgical interventions, different activity-based outcome measures, and poor-quality studies with uncontrolled designs. However, the consistent positive results of upper extremity surgery presented in the 12 included studies justify the initiation of additional, preferably controlled, studies. These studies showed that the ability to use the hand(s) to perform tasks on request (capacity) or spontaneously (performance) and the perception of the patient's ability to use the hand(s) improved significantly after upper extremity surgery. An RCT is the best study design for determing treatment effectiveness. However, different factors, such as the invasive and irreversible character of surgery, and patient preferences, may limit the feasibility of RCTs for studies on upper extremity surgery in children and adolescents. This might be the reason that only 1 of the 12 included studies in our review evaluated the effectiveness of upper extremity surgery in a RCT.[14] This comparative study showed that tendon transfer surgery was more effective than botulinum toxin injections or regular, ongoing therapy in children with upper-extremity CP. The next best design to evaluate the effects of upper extremity surgery may be a cohort study design with a long follow-up, in which patient selection criteria are described in detail and pre-treatment characteristics are studied to identify predictors of (long-term) treatment outcome. A potential predictor of treatment success may be the age at which upper extremity surgery is performed. Currently, there is no evidence available about the timing of upper extremity surgery and the optimal age range for surgical intervention in patients with CP. This requires careful long-term follow-up and monitoring into adulthood.

Our clinical cohort study (chapter 6) showed that a comprehensive multidisciplinary assessment and the use of strict selection criteria contributed to the effectiveness of surgery in our patient population. From this study, we can conclude that children and adolescents with unilateral CP who are able to handle most objects with somewhat reduced quality and/or speed (MACS level II) will probably benefit from surgery, based on assessments evaluating hand use and performance. Given our high proportion of successful outcomes, it can be questioned whether our selection criteria for upper extremity surgery were too strict and more patients might have benefited from surgery. To establish better

criteria for patient selection, larger cohort studies with patients of different ages, CP subtypes, and CP severity are needed. However, some selection criteria are in common use. For example patients with dystonia or athetosis are frequently excluded from surgery, because the outcome of surgery is unpredictable owing to the variable muscle imbalance.

The first selection of patients who are referred to a multidisciplinary upper extremity surgical team is made by the child's own physician. Medical professionals and therapists working with patients with CP have the possibility to choose from a range of different interventions. Selecting the intervention that is most appropriate for the individual patient is based on best research evidence, clinical expertise/experience and patients values and preferences. Surgery was once one of the interventions of first choice. In the last decades, more studies have compared the effect of non-surgical interventions, such as constraint-induced movement therapy (CIMT), hand-arm intensive bimanual training (HABIT), intramuscular injections of botulinum toxin A (BoNT-A), and intensive occupational therapy (OT).[26] Because of the high-level evidence supporting these interventions, they are the first to be chosen by the medical professionals, and only a select group of children are referred to an upper extremity surgery team. It can be questioned whether this pre-selection for upper extremity surgery by the referring physician contributes to the exclusion of patients who might also benefit from surgery. We expect that this pre-selection influenced our outcomes, because we may have assessed only those children and adolescents in whom one of the other interventions did not provide (long-term) improvement in hand use and performance.

Methodological considerations

Interpretation of Assisting Hand Assessment scores

At the time we evaluated the effect of a functional hand orthosis (chapter 4) AHA scores were mostly reported as raw scores, and changes in raw scores (sum of item scores) were presented rather than logit-based scores.[20] A change on the AHA of 4 or more points between two test sessions (with 95% certainty) was considered a clinically relevant change.[19] In 2012, Krumlinde recommended the use of AHA units instead of raw scores when interpreting change after interventions at an individual level.[27] She concluded that simply adding up raw scores of unknown intervals might misrepresent results. For raw scores, changes at the lower or upper ends of a scale may underestimate the real increase in ability as compared with changes in the middle of the scale. When applying these new insights we found that we had overestimated the effect of a functional hand orthosis - now 32% instead of the reported 52% of the children had a significant clinically relevant change. (chapter 3)[6] Thus one in three children benefited from static bracing of the wrist and thumb when it came to using their affected hand to perform tasks requiring the use of both hands.

Different constructs

To obtain full insight into the performance of activities of a child/adolescent, two different performance-based assessments are needed, reflecting two different constructs. In chapter 6, we used outcome measures for perceived performance (COPM, ABILHAND, and VAS) as well as the AHA measuring spontaneous (habitual/typical) use of the affected hand during bimanual task performance. Although one might expect perceived and typical performance

to be correlated, we could not demonstrate this in our sample – we did not find an association between the Abilhand and AHA (r = 0.012, p = 0.95). The lack of correlation indicates that different aspects of the constructs are measured when evaluating performance of activities with these two instruments. The Abilhand measures perceived performance in which the patient or parents use a questionnaire to indicate how daily activities are performed (with ease, with difficulty, or not possible to perform at all). How the patient performs the activity, i.e. with one or two hands or by using compensation strategies, is not evaluated. In contrast, with the AHA a therapist observes and scores how effectively the affected hand is used to perform activities requiring the use of both hands. A difference may occur because the AHA provides an objective measure of functioning, so that hand use may be considered less effective with the AHA than with the Abilhand. This difference highlights the need to evaluate the performance of activities with outcome measures that evaluate both perceived and typical/habitual performance, in order to refine treatment.

Generalizability

Both effect studies (chapters 4 and 6) included a selected group of patients with CP, which limits the generalizability of findings to the overall CP population. We selected a specific age range in both studies. The first study (chapter 4) included children up to 12 years of age.[20] This age range was chosen because at the start of this study, the AHA had been validated in children aged 18 months to 12 years only, and thus the effect of a functional hand orthosis could not be ascertained in adolescents with CP older than 12 years. The other study (chapter 6) included a high proportion of adolescent (79%, age range 13 to 19 years). The positive effects of upper extremity surgery on hand use and performance found in this study were mostly based on outcomes measured with the Ad-AHA in this older age group. Therefore, we are not able to draw conclusions about the effect of surgery in young children, who also might benefit from surgery.

A substantially higher proportion of children and adolescents with unilateral CP were included in studies of chapter 4 (100%)[20] and chapter 6 (87%) than would be expected on the basis of the distribution of CP subtypes in Europe, in which just over a quarter of all children with CP (29.2%: 95% CI 27.9 to 30.4) have unilateral spastic CP.[28] Due to the low number of children with bilateral CP no conclusions can be drawn about the effect of a functional hand orthosis or upper extremity surgery in this population, in which more than 60% of the children have decreased hand function.[29] This suggests that there is a need for more evidence about the effects of these interventions in patients with bilateral CP.

We included mainly patients with a relatively high ability to handle objects in important daily activities, as classified according to the MACS.[30] (chapter 6) However, patients with lower or even higher ability level might also benefit from upper extremity surgery. Our study showed that the proportion of patients classified as MACS level II (patient able to handle most objects but with somewhat reduced quality and/or speed of achievement) was 72%, which is not consistent with the earlier reported proportion of 20-25%.[31]

These differences in patient population (age range, type of CP and MACS level) may have caused an overestimation of the effects of upper extremity surgery or use of a functional hand orthosis.

Measuring performance in a clinical setting

Both intervention studies (chapters 4 and 6) investigated the impact of CP on an individual's ability to perform activities, as assessed with the AHA.[20]

In the ICF, performance is understood as "involvement in a life situation" or "the lived experience" of people in the actual context in which they live.[7] Thus assessing performance in a clinical setting seems contradictory. The use of (semistructured) test kits and the presence of a therapist violates the social context. Therefore, in strict sence capacity is evaluated.

That said, it is not always practical or feasible for therapists to observe and assess patients in their own environment.[32] With the (Ad-)AHA it is possible to assess spontaneous use of both hands in a semistructured context, using different test activities (Ad-AHA Board game, Ad-AHA Sandwich and Ad-AHA Parcel) of which the adolescent may choose. In the case of the AHA, the best context is a room with a table, chair, and an activity which evokes the spontaneous use of both hands and which can be assessed afterwards, using a video recording of the activity. The setting itself, the clinic, is of less influence because the activity involves a play session with a board game allowing interaction with family members. The Ad-AHA board game ensures that functioning is observed objectively, without the subject even realizing that the activity is being assessed.[3] All these aspects together makes the (Ad-) AHA an appropriate instrument to capture how well the affected hand is spontaneously used as an assisting hand during bimanual task performance, this in contrast to instruments measuring a person's best capacity. [4,3,17-19]

Implications for clinical practice

Assisting Hand Assessment for Adolescents

The newly developed and validated test activity, the Ad-AHA board game (Chapter 2 and 3), is already available and in use in clinical practice to elicit bimanual performance of activities in adolescents with unilateral CP. [3,6] With the AHA 18-18 it is possible to evaluate interventions, monitor development from childhood to young adulthood, and guide intervention planning.[3] (Fig. 1)

Figure 1. Ad-AHA Board Game

The Ad-AHA has proven its merits as outcome measure for adolescents with unilateral CP in clinical practice, and in the study evaluating the effect of upper extremity surgery on hand use and the performance of daily activities (Chapter 6).[3] The AHA 18-18 covers the age range 18 months to 18 years, which makes it possible to follow the development of the effective use of the affected hand while performing bimanual tasks and provides information about the possible improvement or deterioration of hand skills with age.[3] In clinical practice, it is important to monitor how hand skills change from infancy to adulthood, because hand skills develop rapidly during the first few years of life, followed by a refinement of these skills until adolescence, after which further refinement occurs at a lower rate until adulthood.[34] Skills are refined when children/adolescents learn new age-related skills that are important for daily life or after an intervention at older ages.[34] In addition to this age-related refinement, hand skills may also deteriorate with age. Both fine and gross motor development has been reported to decrease in children with low ability after 11 years of age.[35,36] The impaired function may persist into adulthood, and the ability to use the affected hand may deteriorate further in later life, as a result of contractures, muscle weakness, and atrophy, all of which may increase in adulthood. [37-40] Being able to measure this development (improvement or deterioration) in hand skills will help professionals to plan appropriate interventions.

Functional hand orthosis

Static bracing of the wrist and thumb improves hand position for performing manual tasks, and thus braces should be considered as a treatment option to improve functional performance (Chapter 4). It should be realized that not all children with unilateral CP will immediately benefit from a functional hand orthosis. In our study, some children were less able to extend their fingers and to grasp and release objects while wearing the functional hand orthosis, and this should be taken into account when a functional hand orthosis is considered as a means to improve hand use and the performance of activities. Some children will need time to get used to an orthosis. By evaluating performance in terms of individual AHA items, the therapist wil be able to suggest specific activities for which the functional hand orthosis will be beneficial.[20] For example, a more neutral position of the wrist may help the child to fixate paper more effectively while writing, or when combined with a thumb out of the palm the orthosis will enable the child to grip bicycle handlebars more effectively with both hands.

Upper extremity surgery

Eligibility for upper extremity surgery should be based on extensive multidisciplinary assessment. Different instruments should align surgical and postoperative treatment plans in order to achieve the best outcome in terms of patient-specific goals related to the performance of activities in daily life. As the heterogeneity of study outcomes precludes meaningful meta-analysis of data,, it would be advisable to use a core set of instruments to improve selection criteria, to support the choice of surgical intervention, to assess the effect of surgery, and to predict outcomes.

Use of a core set of instruments would also make it easier to compare the results of different studies. In addition to the instruments used in our study (COPM, AHA, ABILHAND, and BBT, chapter 6), a validated instrument measuring patient satisfaction with cosmetic appearance would provide important information about patient-specific goals.

Furthermore, adding the "Shriners Hospitals Upper Extremity Evaluation" dynamic positional analysis (SHUEE-DPA) allows to measure and link "function" and "activity" when evaluating the result of surgery, by analysing function and describing the position of the thumb, fingers, wrist, forearm, and elbow during 16 different activities. The COPM can be used to identify patient-specific "hand-use oriented" goals, which may be linked to the different surgical procedures. Information from the Abilhand, AHA, and BBT should be collected to obtain insight into the ability of the patient to use the affected hand and the likelihood that COPM goals will be achieved. If the results from the Abilhand, AHA, and BBT show a very low ability to use the affected hand effectively (i.e., not able to fixate or hold objects), some goals might be very difficult to achieve. For example, when the goal of an adolescent with no forearm supination is to hold a plate with drinks and to shake hands, the patient should refine his/her goals and expectations with the help of the surgical team.

The same core set (COPM, AHA, ABILHAND, BBT, SHUEE-DPA) can be used to guide postsurgical treatment. Activity-based therapy should be used to improve hand function and the performance of functional hand activities after surgery, and may include intensive therapy, modified constraint-induced movement therapy, or bimanual task-oriented therapy.

Implications for future research

Assisting Hand Assessment for Adolescents

In children with CP, several interventions have proven to be effective in improving hand use and performance. The next step would be to use the Ad-AHA when evaluating these interventions in adolescents and extend study participations to patients up to the age of 18 years. The continuous and large age range of the AHA 18-18 makes it possible to use the same scale to monitor the development or possible deterioration of hand use from early childhood to young adulthood.

Future research should focus on possible changes in adolescents with CP, in order to improve interventions that can prevent a deterioration of hand skills with age and which can facilitate further refinement of acquired skills. Information about improvement or deterioration will also aid the professional when planning interventions to improve hand use and performance. Furthermore, studies of the long-term effect of interventions from childhood to adulthood, and of changes in hand function during the childhood-to-adolescence-to adulthood transitions are needed. The logical next step would be to extend the AHA for use in adults aged 18 years and older, in order to evaluate the effects of interventions in adults with CP (Fig. 2).

In the study presented in chapter 6, 13% of the included patients were diagnosed with bilateral CP. Regrettably, the (Ad-)AHA could not be used in this group to evaluate the effect of upper extremity surgery on (bimanual) arm-hand performance because the instrument is not valid and reliable for use with children and adolescents with bilateral CP. This is particularly unfortunate because more than 60% of children with bilateral CP have decreased hand function.[29] In the past, there was a lack of appropriate outcome measures for different ICF levels validated for use in children with bilateral CP.[41] Recently, the Both Hands Assessment (BoHA) was developed by adapting the AHA.[42] New evaluation studies should use the BoHA to measure bimanual performance in children with bilateral CP and to quantify a possible asymmetry between hands. Besides the Kid's-AHA, Ad-AHA, and BoHA,

more versions have been developed within the "AHA family", based on age groups and diagnoses (bilateral and unilateral CP). (Fig. 3)

In order to evaluate hand use and performance in the entire CP population, it is necessary to further extend the AHA and to investigate the possibility of using the same AHA scale for a broader age range. The principal researchers involved in the development of the AHA family joined in the recently set up "AHA research network" to continue these studies, to collaborate with other investigators, and to contribute to future AHA research (www.ahanetwork.se). The ultimate goal is to develop an AHA for patients with unilateral and bilateral CP who are able to handle objects, although with varying difficulty (Manual Ability Classification System (MACS) levels I-III), in order to improve intervention planning, evaluate (new) interventions, determine their effectiveness, and monitor the development of hand use.

Small-Kids AHA
18 months to 5 years

School-Kids AHA
6 to 12 years

Ad-AHA
13 to 18 years

Figure 2. The AHA 18-18

Intervention studies that aim to optimize hand use and performance of activities

Future studies should evaluate the effect of a functional hand orthosis, using appropriate and valid outcome measures, in order to assess benefits (more effective us of the assisting hand) as well as potential harms (such as muscle atrophy).

Furthermore, there is a need for studies evaluating the immediate and long term effect of a hand orthosis in different age groups (including adolescents). The findings of various studies suggest that a functional hand orthosis may be effective for performing certain daily tasks, which highlights the need for research into the effect of intermittent use of a functional hand orthosis in combination with training.

There is still insufficient evidence to recommend upper extremity surgery as a treatment option for improving the hand use and performance of children and adolescents with CP. To make clinical recommendations, future research should include patients of different ages and ability. In order to predict and achieve optimal outcomes after upper extremity surgery that match patient-specific goals and expectations related to the performance of activities important to the patient and to define patient selection criteria (based on e.g. age, severity), larger studies are needed, preferably RCTs comparing the effects of surgical and non-surgical intervention, possibly with a waiting-list control group and monitoring into adulthood.

Besides the choice of surgical intervention, the postoperative hand rehabilitation programme has a major impact on hand use and performance. More research is needed into

the effect of a postoperative hand therapy. Such therapy should be activity-based with a focus on patient-relevant goals in order to optimize the outcome of upper extremity surgery.

Lastly, in order or evaluate the cosmetic results of upper extremity surgery, outcome measures evaluating the psychosocial impact of a hand deformity and cosmetic appearance should be developed and validated.

The findings of this thesis have contributed to knowledge relevant to the assessment and management of upper extremity functioning in children and adolescents with CP. The newly developed and validated instrument, Ad-AHA, makes it possible to assess the actual performance of adolescents with CP, which in turn makes it possible to evaluate the effect of interventions. The continuous and large age range of the AHA 18-18 will enable investigators to monitor the development of, or changes in, hand use from childhood to young adulthood. We found that the results achieved with functional hand orthosis and upper extremity surgery were promising in terms of the improvement in hand use and the performance of activities of a selected group of patients with CP.

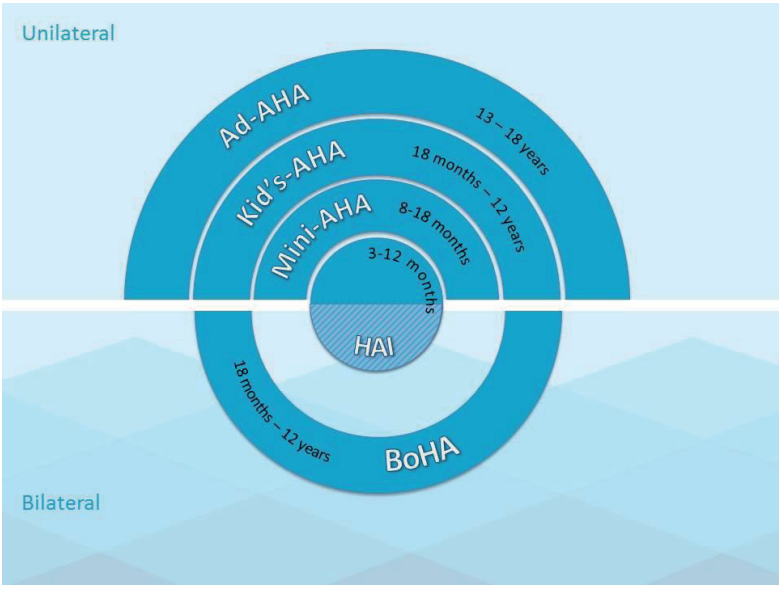

Figure 3. AHA family

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Summary

Effective use of the assisting hand in adolescents with cerebral palsy

This thesis aims to enhance the knowledge on assessment and management of upper extremity functioning in children and adolescents with CP, by developing and evaluating the validity and reliability of the Assisting Hand Assessment for adolescents (Ad-AHA) with CP (Part I) and by evaluating 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 (Part II). **Chapter 1** provides a general introduction objectives and outline of this thesis.

Part I: Development of the Assisting Hand Assessment for adolescents (Ad-AHA)

Chapter 2 demonstrates the development and the validation of the new test activity, the Ad-AHA board game, to observe how adolescents with CP actually use their affected hand to perform bimanual activities. The study consisted of two phases: (1) development of the test activity for adolescents and evaluation of the validity of the content of this new activity; (2) assessing the construct validity of the AHA test items for the age range 18 months to 18 years using Rasch measurement model.

To be able to measure how well the affected hand is spontaneously used as an assisting hand during bimanual task performance, the new test activity should meet several requirements. It should involve a sequence of bimanual actions or tasks that form a meaningful activity include objects that require bimanual handling, are age appropriate and familiar to most adolescents. The activity should be semi-structured allowing therapist interaction to help tasks proceed if needed and to make it fun and engaging to elicit typical/habitual performance. Finally, the new activity should allow observation of different aspects and levels of ability of bimanual performance and enable scoring of the AHA test items.

To evaluate the construct validity of the AHA test items for the age range 18 months to 18 years, a combined sample of adolescents and children (n=290 assessments) were analysed with the Rasch model analysis. Differential test functioning was performed to evaluate whether the same AHA scale can be used for both age groups

The study showed that the new test activity, the Ad-AHA board game, can be used in clinical practice to elicit actual bimanual performance in adolescents with unilateral CP. The scale showed good construct validity within the whole age range of 18 months to 18 years, meaning that the same AHA scoring criteria can be used both for children and for adolescents with unilateral CP. The AHA 18-18 covers the whole age range from 18 months to 18 years and is a valid performance-based instrument to monitor development from childhood to young adulthood, evaluate interventions, and guide intervention planning.

Chapter 3 focuses on the different aspects of the reliability of the Assisting Hand Assessment in adolescents (Ad-AHA). A convenience sample of 112 children and adolescents with unilateral CP, aged 10 to 18 years, were recruited by therapists working in eight different rehabilitation centres in Australia, Sweden, and the Netherlands. All AHA assessments were videotaped according to the standard protocol described in the AHA manual and sent to one location for central scoring. The participants were tested once or twice depending on the type of reliability evaluated.

To evaluate interrater reliability, the first 38 participants in the convenience sample played the Ad-AHA Board Game once and two raters independently scored the videotaped session. Thirty one adolescents played the Ad-AHA Board Game twice with an interval of 1 to 2 weeks to evaluate the test-retest reliability. The videos were scored by the same assessor who was blind to previous obtained AHA scores. The four different test activities (School-Kids AHA, Ad-AHA Board Game, Ad-AHA Present, and Ad-AHA Sandwich) elicit bimanual actions involving the same underlying trait. Alternate-form reliability was evaluated to show whether the scores for the outcome measures of these tests are consistent relative to each other. Agreement between AHA scores was analysed by comparing scores on the Ad-AHA Board Game with scores on one of the other three test activities.

This study showed that the Ad-AHA has good-to-excellent interrater and test-retest reliability in adolescents with unilateral CP. Ad-AHA scores are consistent across different raters and occasions. Individual scores on the School-Kids AHA and the Ad-AHA Board Game were directly comparable, which makes it possible to use both test activities interchangeably for children aged 10 to 13 years. Test scores for all different Ad-AHA test activities are comparable, showing that reliable AHA scores can be generated by using different age-appropriate tests activities for children and adolescents aged 18 months to 18 years. The excellent measurement agreement means that the different test activities can be used to evaluate change over time, with a score change of 5 AHA-units indicating a change beyond measurement error with 95% certainty, using the same AHA 5.0 scale.

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

In Chapter 4 - 6, the effect of two different interventions on the ability to perform daily activities was evaluated. Both interventions aim to achieve improvement of muscle balance and a more normal hand posture, which may affect the ability to grasp and release objects and to use both hands together.

Chapter 4 presents the immediate effect of wearing a functional hand orthosis while performing tasks. All children with unilateral CP who were referred for occupational therapy to the Department of Rehabilitation at the Academic Medical Center, Amsterdam, or the Rehabilitation Center 'De Trappenberg', Huizen, were consecutively screened for inclusion and exclusion criteria by their rehabilitation physician. Children were included if they were aged between 4 and 12 years, diagnosed with spastic unilateral CP and classified as Zancolli I, IIA, or IIB. Performance of bimanual activities by children with unilateral CP was evaluated with the Assisting Hand Assessment (AHA) three times (AHA 1, AHA 2, AHA 3) with an interval of 1 week between tests: AHA 1 consisted of an assessment without an functional hand orthosis; AHA 2 consisted of an assessment with the orthosis placed on the affected hand immediately before the assessment; and AHA 3 was once again an assessment without an orthosis. All AHA assessments were randomly scored and the time period of 1 week between assessments was chosen in order to prevent recall effects. In between the assessments, children did not receive any additional treatment other than their usual therapy. The immediate effect of a functional hand orthosis on the performance of bimanual activities was assessed by comparing the AHA scores of the three assessments (without, with, and without the functional hand orthosis) with repeated measures analysis of variance (ANOVA). Subgroup analyses were performed, based on Zancolli grade with the

Friedman repeated measures analyses of variance by ranks, with post hoc comparisons using the Wilcoxon signed-rank test.

The study showed that the performance of bimanual activities in children wearing the orthosis improved significantly compared with the performance of children not wearing the orthosis. No learning carry-over effect from wearing an orthosis during this short period was found. With evaluation on the level of individual AHA items, the therapist can decide in which activities the individual child will most likely be able to benefit most from the functional hand orthosis.

Chapter 5 provides a systematic review summarizing the clinical postoperative outcomes of upper extremity surgery on hand use in children and adolescents (aged < 20 years) with CP. Searches were carried out in MEDLINE, Embase, psycINFO until May 2017. Studies were included if they investigated the effect of upper extremity surgery on hand use and activity performance with or without concurrent controls or in case series with pre-test/post-test outcomes with a minimal sample size of 10; and report the effects of upper extremity surgery at least 5 months postoperative; and included more than 75% of the participants with CP aged up to 20 years; and used a validated activity-based instrument for hand use and activity performance. Risk of bias was assessed for the included individual studies using the ROBINS-I tool (Risk Of Bias In Non-randomized Studies - of Interventions) criteria for reviews. Quality assessment was performed for outcomes of interest, using Grading of Recommendations Assessment, Development, and Evaluation (GRADE).

The electronic search provided a total of 8009 citations, after adjusting for duplicates and reviewing the abstracts, the full text of 132 citations was examined in more detail. Finally, a total of 12 studies were identified for inclusion in the review. Comparative effectiveness of upper extremity surgery was reported in only 1 out of the 12 selected studies. Two out of the 12 studies were prospective; the other studies evaluated the effect of upper extremity surgery retrospectively.

The patient's functional performance was measured in 4 studies on quantifiable tasks with the Shriners Hospital Upper Extremity Evaluation (SHUEE), Assisting Hand Assessment (AHA), Jebsen Taylor Hand Function Test (JHFT) or Box and Block Test (BBT). Nine studies used a classification, interview or questionnaire to evaluate the functional performance using the Functional Independence Measure for Children (WEEFIM) or House Functional Classification (HFC). All included studies evaluating the ability to perform functional tasks on request (capacity), the spontaneous functional use (performance) and the self-perception of the patient's performance presented a significant improvement.

There was an overall high risk of bias within the included, predominantly retrospective studies. The risk of bias was high for the items on selection, confounding and measurement of outcomes. The quality of evidence, evaluated with "The Grading of Recommendations Assessment, Development and Evaluation" (GRADE), was very low for each postoperative outcome after upper extremity surgery in children and adolescents with CP due to heterogeneous surgical interventions, different activity-based outcome measures and poor-quality studies with uncontrolled designs.

Considering the overall very low quality of the evidence, it is not possible to make clinical recommendations on the effect of upper extremity surgery. However, the positive results of the effect of upper extremity surgery on hand use and activity performance presented in the included studies certainly justify the performance of more high-quality comparative studies.

In **Chapter 6** the results of a clinical cohort study are presented. The study describes our experience with patient selection based on multidisciplinary assessment and shared decision-making (SDM) and the effects of upper extremity surgery on manual performance and patient-relevant outcomes.

The study was performed in a tertiary referral centre for upper extremity surgery for patients with CP in the Netherlands and included all patients (up to 20 years of age) with CP who were consecutively referred to our team for upper extremity surgery consideration between July 2011 and May 2017. The multidisciplinary team consisted of a hand surgeon, paediatric rehabilitation physician and occupational therapist. Each patient was assessed and discussed by the team during a single visit in order to reach a shared-decision about whether or not to proceed with surgery.

Eligibility for upper extremity surgery was based on the extensive multidisciplinary assessment. The occupational therapist collected information about patient-specific goals identified on the Canadian Occupational Performance Measure (COPM), perceived independence to execute bimanual activities at home using the ABILHAND-kids questionnaire, perceived quality of use of the affected hand during daily activities scored on a visual analog scale (VAS), spontaneous use of both hands using the Assisting Hand Assessment (AHA) and the Ad-AHA Board Game (**Chapter 2 and 3**), and gross manual dexterity assessed with the Box and Block Test (BBT). Then the patients were seen by the hand surgeon and pediatric rehabilitation physician for a medical examination. To make the best possible treatment decision, the following criteria were assessed and discussed with the patient and their parents: (I) absence of dystonia or athetosis, because in these types of movement disorders the imbalance of muscles is variable and the outcome of surgery is unpredictable; (II) achievable patient-specified goals (functional and/or cosmetic); and (III) absence of developmental disregard, i.e. a large discrepancy between the ability to use and the actual use of the affected hand. The team helped the patient compare treatment options and expected outcomes and reached a shared decision with the patient. If upper extremity surgery was not indicated because criterion II or III was not met, alternative conservative treatment options were discussed with the patient, with the possibility for future re-assessment by the multidisciplinary team.

Between July 2011 and May 2017, 66 patients with CP were consecutively referred to our multidisciplinary team of whom XX received UES. Eventually, the effect of upper extremity surgery on hand use and activity performance was evaluated in 39 patients, 4 months (range 0 - 15 months) before surgery and 9 months (range 6 - 11 months) after surgery. The age of the included patients ranged between 7 years 10 months and 19 years 7 months. The results of 5 patients were not available at the time of the study. Twenty-two patients were not selected for upper extremity surgery after multidisciplinary assessment and shared decision-making. Two out of these 22 patients preferred a non-surgical intervention despite being considered eligible for upper extremity surgery by the upper extremity surgery team.

All outcomes showed a statistically significant improvement at 9 months. 80% and 77% of the participants had a clinically significant improvement in COPM performance and satisfaction, respectively and 71% on manual performance (AHA). Patients whose primary goal was to improve the appearance of the affected upper extremity also showed improved manual performance and patient-specific functional goals.

The study showed that careful patient selection, based on multidisciplinary assessment and SDM, results in clinically relevant improvements in patient-specific

functional and/or cosmetic goals and manual ability after upper extremity surgery in more than 80% of the assessed children and adolescents with CP.

Chapter 7 provides a general discussion of the results of the individual studies in this thesis placed in a broader perspective. Four topics are discussed: interpretation of Assisting Hand Assessment-scores; actual and perceived performance-based assessments reflecting two different constructs; differences in patient population and consequences for generalizability; and measuring performance in a clinical setting. In addition, implications for further research and clinical practice are given.

Samenvatting

Effectief gebruik van de assisterende hand door adolescenten met een cerebrale parese

Dit proefschrift beoogt de zorg voor kinderen en adolescenten met een cerebrale parese (CP) te verbeteren door de ontwikkeling en validering van de Assisting Hand Assessment voor adolescenten (Ad-AHA) (Deel I). Met de Ad-AHA is het mogelijk om bij adolescenten het gebruik van de aangedane hand tijdens de uitvoering van tweehandige activiteiten te meten en het effect van interventies te evalueren. Daarnaast vergroot dit proefschrift de kennis die beschikbaar is over het effect van twee interventies (functionele handorthese en chirurgie van de bovenste extremiteit) op het gebruik van beide handen tijdens het uitvoeren van activiteiten door kinderen en adolescenten met cerebrale parese (CP) (Deel II). **Hoofdstuk 1** bevat een algemene introductie inclusief achtergrond, doelen en korte beschrijving van de verschillende studies.

Deel I: Ontwikkeling van de Assisting Hand Assessment voor adolescenten

Hoofdstuk 2 beschrijft de ontwikkeling en validatie van de Assisting Hand Assessment voor adolescenten met CP (Ad-AHA). Deze studie bestaat uit twee delen. Het eerste deel betreft de ontwikkeling van een nieuwe activiteit (Ad-AHA bordspel) waarmee tweehandigheid wordt uitgelokt bij adolescenten met CP. Het tweede deel bestaat uit evalueren van de constructvaliditeit van de AHA score items voor kinderen en adolescenten met CP binnen de leeftijdsrange van 18 maanden tot en met 18 jaar op basis van Rasch analyse.

De nieuwe testactiviteit van de AHA, het Ad-AHA bordspel, voldoet aan de vooraf opgestelde voorwaarden voor het spontaan uitlokken van tweehandigheid. De activiteit bestaat uit verschillende objecten en een reeks bimanuele acties of taken, die vervolgens samen een zinvolle activiteit vormen. De objecten zijn aantrekkelijk voor de adolescenten en lokken tweehandigheid uit. De activiteit is semigestructureerd, waardoor interactie door therapeut mogelijk is om de deelhandelingen succesvol te laten verlopen en de activiteit (het bordspel) leuk en aantrekkelijk te maken. Dit alles met als doel om zoveel mogelijk spontaan gebruik van beide handen uit te lokken. Bovendien bestaat de activiteit uit zowel eenvoudige als meer complexe deelhandelingen, waardoor de mate van effectiviteit van het gebruik van de assisterende hand met de AHA score items beoordeeld kan worden.

Voor de evaluatie van de constructvaliditeit van de AHA score items voor de leeftijd van 18 maanden tot 18 jaar, is een gecombineerde steekproef van adolescenten en kinderen (n=290) geanalyseerd met behulp van het Rasch-model waarmee tevens bepaald kon worden of voor beide leeftijdsgroepen dezelfde AHA-schaal kan worden gebruikt.

De resultaten van deze studie laten zien dat het Ad-AHA bordspel valide is en in de klinische praktijk kan worden ingezet om spontaan gebruik van beide handen uit te lokken bij adolescenten met unilaterale CP. Dezelfde AHA-schaal en scoringscriteria kunnen worden gebruikt voor kinderen en adolescenten met unilaterale CP binnen de leeftijdsrange van 18 maanden tot 18 jaar (AHA 18-18). We kunnen concluderen dat de AHA 18-18 een valide instrument is om de ontwikkeling van kindertijd tot jongvolwassenheid te volgen, interventies te evalueren en interventieplanning te begeleiden.

In **hoofdstuk 3** zijn de verschillende aspecten van de betrouwbaarheid van de Assisting Hand Assessment voor adolescenten (Ad-AHA) geëvalueerd. Honderdtwaalf kinderen en adolescenten met unilaterale CP (10 tot 18 jaar) vanuit acht verschillende revalidatiecentra in Australië, Zweden en Nederland, hebben aan dit onderzoek meegedaan. De AHA

activiteiten werden op video opgenomen volgens het standaardprotocol zoals beschreven in de AHA-handleiding en naar één locatie gestuurd voor centrale scoring. De deelnemers werden een of twee keer getest, afhankelijk van het type betrouwbaarheid dat werd geëvalueerd.

Om de mate van overeenstemming tussen verschillende beoordelaars te evalueren, speelden de eerste 38 deelnemers het Ad-AHA bordspel één keer, waarna de twee beoordelaars de sessie aan de hand van de video-opnamen onafhankelijk scoorden. Eenendertig adolescenten speelden het Ad-AHA bordspel tweemaal met een interval van 1 tot 2 weken om de test-hertest betrouwbaarheid te evalueren. De video's werden beoordeeld door dezelfde beoordelaar voor wie de eerder behaalde AHA-scores onbekend waren. Vervolgens werd de overeenstemming tussen de vier verschillende AHA test activiteiten geëvalueerd (School-Kids AHA, Ad-AHA Bordspel, Ad-AHA Present en Ad-AHA Sandwich).

Deze studie toonde aan dat de Ad-AHA een betrouwbaar instrument is om het gebruik van de assisterende hand door adolescenten met unilaterale CP te kunnen beoordelen. De Ad-AHA-scores van verschillende beoordelaars en bij herhaalde testafname zijn consistent. Individuele scores op de School-Kids AHA en de Ad-AHA Board Game van kinderen in de leeftijd van 10-13 jaar kwamen sterk overeen, wat inhoudt dat het mogelijk is om beide testactiviteiten te gebruiken voor kinderen van 10 tot 13 jaar. Ook de testscores verkregen met de 3 Ad-AHA-testactiviteiten (Ad-AHA Bordspel, Ad-AHA Present en Ad-AHA Sandwich) voor adolescenten zijn vergelijkbaar. Dit houdt in dat er betrouwbare AHA-scores kunnen worden gegenereerd aan de hand van verschillende leeftijdsgebonden testactiviteiten voor kinderen en adolescenten van 18 maanden tot 18 jaar. De uitstekende overeenkomst tussen de metingen betekent dat de verschillende testactiviteiten kunnen worden gebruikt om veranderingen in de tijd te evalueren. Daarnaast laat deze studie zien dat een scoreverschil van 5 AHA-units een verandering betekent die groter is dan de meetfout en daarom gezien kan worden als een echte verandering.

Deel II: Effect van interventies op gebruik van de hand en uitvoering van activiteiten

In Hoofdstuk 4 - 6 van dit proefschrift worden de effecten beschreven van twee verschillende interventies bij kinderen en adolescenten met CP op het gebruik van beide handen tijdens het uitvoeren dagelijkse activiteiten. Beide interventies zijn gericht op het verbeteren van de spierbalans en het plaatsen van de hand in een meer functionele stand, en beogen het pakken en loslaten van voorwerpen en het gebruik van beide handen tijdens het uitvoeren van tweehandige activiteiten te bevorderen.

In **Hoofdstuk 4** is het directe effect van het dragen van een functionele handorthese tijdens het uitvoeren van activiteiten beschreven. Alle kinderen met een unilaterale CP in de leeftijd tussen 4 en 12 jaar, die voor ergotherapie waren verwezen naar de afdeling revalidatie in het Academisch Medisch Centrum in Amsterdam, of het revalidatiecentrum 'De Trappenberg' in Huizen, werden achtereenvolgens gescreend op geschiktheid voor deelname door hun revalidatiearts. Kinderen met de diagnose spastisch unilateraal CP werden geïncludeerd als het grijp- en loslaatpatroon was geclassificeerd als Zancolli I, IIA of IIB.

Bij alle deelnemers werd de uitvoering van tweehandige activiteiten drie keer, met een interval van 1 week tussen tests, geëvalueerd met behulp van de Assisting Hand Assessment (AHA 1, AHA 2, AHA 3). AHA 1 bestond uit een beoordeling zonder orthese, waarvan de

score werd vergeleken met de AHA 2 (meetmoment met orthese die vlak voor de beoordeling werd omgedaan bij de aangedane hand) en AHA 3 opnieuw zonder orthese, waarvan de score met AHA 1 werd vergeleken. De video-opnames van de AHA afnames werden in willekeurige volgorde gescoord met een tijdsperiode van 1 week tussen de beoordelingen om "herinneringsbias" te voorkomen. Tussen de beoordelingen door kregen de kinderen geen andere therapie dan hun gebruikelijke therapie. Het direct meetbare effect van de functionele handorthese op de gebruikt van de assisterende hand tijdens de uitvoering van tweehandige activiteiten werd geëvalueerd met de AHA. Subgroep analyses werden uitgevoerd op basis van de Zancolli-classificatie.

De resultaten laten zien dat 52% van de kinderen de assisterende hand effectiever konden gebruiken op het moment dat ze de orthese om hadden. Er was geen sprake van een leer effect van het dragen van de functionele handorthese (geen verschil tussen de twee metingen AHA 1 en AHA 3). Uit de studie kwam naar voren dat de therapeut met behulp van evaluatie op het niveau van AHA-items, het individuele kind kan adviseren bij welke activiteiten de functionele handorthese mogelijk bijdraagt aan het effectiever kunnen gebruiken van de assisterende hand.

Hoofdstuk 5 laat de resultaten zien van een systematische review die het effect van een chirurgische ingreep aan de bovenste extremiteit op het gebruik van de hand tijdens de uitvoering van activiteiten bij kinderen en adolescenten met CP (leeftijd <20 jaar) samenvat. Voor deze beoordeling werden tot mei 2017 zoekstrategieën uitgevoerd in de databases MEDLINE, Embase en psycINFO. Studies werden geïncludeerd als ze; (i) het effect van een chirurgische ingreep aan de bovenste extremiteit evalueerden, met of zonder een controle groep en bij een minimale steekproef van 10 patiënten; (ii) ze de effecten evalueerden met een follow-up-tijd van tenminste 5 maanden; (iii) de deelnemers voor meer dan 75% uit kinderen en/of adolescenten met CP (leeftijd ≤20 jaar) bestonden; (iv) ze gebruik maakten van een gevalideerd meetinstrument om het effect van chirurgie aan de bovenste extremiteit te meten op het gebruik van de hand tijdens de uitvoering van activiteiten. Het risico op bias werd voor de geïncludeerde studies beoordeeld met behulp van de ROBINS-I-tool (Risk of Bias In Non-randomized Studies- of Interventions). De kwaliteitsbeoordeling van het geaggregeerde bewijs werd uitgevoerd met behulp van "Grading of Recommendations Assessment, Development and Evaluation" (GRADE).

De elektronische zoekactie leverde in totaal 8009 citaties op. Na het verwijderen van de duplicaten en het beoordelen van de abstracts, werd de volledige tekst van 132 citaties beoordeeld op inclusiecriteria. Ten slotte werden in totaal 12 studies geïncludeerd voor deze studie. De effectiviteit van chirurgie aan de bovenste extremiteit werd in slechts 1 van de 12 geselecteerde onderzoeken vergeleken met andere interventies. Tien van de twaalf studies evalueerden het effect van chirurgie aan de bovenste extremiteit retrospectief. De studies beschreven verschillende chirurgische ingrepen voor de meest voorkomende problemen bij kinderen en adolescenten met CP. Het gebruik van de arm/hand tijdens de uitvoering van activiteiten werd gemeten in 4 studies door middel van het observeren van kwantificeerbare taken met behulp van de Shriners Hospital Upper Extremity Evaluation (SHUEE), Assisting Hand Assessment (AHA), Jebsen Taylor Hand Function Test (JHFT) of Box and Block Test (BBT). Acht studies gebruikten de House Functional Classification (HFC) en 1 studie gebruikte een interview/vragenlijst, de Functional Independence Measure for Children (WEEFIM). Alle geïncludeerde studies lieten een significante verbetering zien als gevolg van de chirurgische ingreep op het gebruik van de arm/hand op verzoek (capacity)

en tijdens spontaan uitvoeren van activiteiten gebruik. Daarnaast gaven de patiënten aan tevreden te zijn over het effect van de ingreep op het gebruik van de arm en hand.

De kwaliteit van de studies was laag, met name vanwege risico op vertekening van de resultaten. De kwaliteit van het bewijs geëvalueerd met GRADE werd als zeer laag beoordeeld voor elk van de uitkomst categorieën. De categorieën werden onderverdeeld op basis van meetinstrumenten die kwantificeerbare taken evalueerden en meetinstrumenten die met behulp van een interview of vragenlijst de uitvoering van activiteiten evalueerden. De kwaliteit van bewijs werd laag gescoord als gevolg van heterogene chirurgische ingrepen, verschillende uitkomstmaten en de opzet van de voornamelijk ongecontroleerde studies die een hoog risico op vertekening van resultaten geeft.

Gezien de zeer lage kwaliteit van het bewijs, is het niet mogelijk om klinische aanbevelingen te doen over het effect van chirurgie aan de bovenste extremiteit bij deze doelgroep. De positieve resultaten van het effect van chirurgie aan de bovenste extremiteit op handgebruik en de uitvoering van activiteiten in alle geïncludeerde studies rechtvaardigen echter wel de aanbeveling om meer vergelijkende studies uit te voeren.

In **Hoofdstuk 6** wordt in een klinische cohortstudie het effect van chirurgie aan de bovenste extremiteit op het gebruik van de hand onderzocht. Ten eerste wordt het proces beschreven dat gebaseerd is op een multidisciplinaire beoordeling, uitgebreide onderzoeken en shared decision-making. Vervolgens worden de effecten van de chirurgische ingreep op de uitvoering van activiteiten en patiënt relevante uitkomsten gepresenteerd.

De studie werd uitgevoerd in een tertiair verwijzingscentrum voor chirurgie aan de bovenste extremiteit voor patiënten met CP in Nederland en omvatte alle patiënten (tot 20 jaar) met CP die tussen juli 2011 en mei 2017 werden verwezen naar het multidisciplinaire spastisch handenteam. Dit team bestaat uit een handchirurg, kinderrevalidatiearts en ergotherapeut. Door de patiënt zorgvuldig te boordelen, in het multidisciplinair handenteam te bespreken en de uitkomsten te delen met patiënt, werd tot een gezamenlijk besluit gekomen om wel of niet te opereren.

Tijdens de uitgebreide multidisciplinaire beoordeling werd bekeken of de patiënt aan de criteria voldeed voor een chirurgische ingreep. De ergotherapeut verzamelde informatie over: (i) patiënt-specifieke doelen, geïdentificeerd met behulp van de Canadian Occupational Performance Measure (COPM); (ii) ervaring van de patiënt ten aanzien van het zelfstandig uitvoeren van tweehandige activiteiten in de thuissituatie, uitgevraagd met behulp van de ABILHAND-kids-vragenlijst; (iii) ervaring van de patiënt ten aanzien van de kwaliteit van gebruik van de aangedane hand tijdens het uitvoeren van dagelijkse activiteiten, gescoord op de visuele analoge schaal (VAS); (iv) spontaan gebruik van beide handen tijdens het uitvoeren van tweehandige taken, geëvalueerd met behulp van de Assisting Hand Assessment (AHA en Ad-AHA **hoofdstuk 2 en 3**); en (v) eenhandige vaardigheid, beoordeeld met de Box and Block Test (BBT).

Vervolgens werden de patiënten gezien door de handchirurg en kinderrevalidatiearts voor een uitgebreid medisch onderzoek. Om de beslissing ten aanzien van de chirurgische behandeling te kunnen nemen, werden de volgende criteria beoordeeld en besproken met patiënt en ouders: (I) afwezigheid van dystonie of athetose, omdat bij dit soort bewegingsstoornissen de disbalans van spieren variabel is en het resultaat van een operatie onvoorspelbaar; (II) haalbare door de patiënt gespecificeerde doelen (functioneel en / of

cosmetisch); en (III) afwezigheid van "vergeten de hand te gebruiken", d.w.z. een grote discrepantie tussen het vermogen om de hand te kunnen gebruiken en het spontaan gebruik van de aangedane hand. Mogelijke behandelingsopties en verwachte resultaten werden vervolgens met de patiënt besproken. Indien niet aan criteria II of III werd voldaan, werden alternatieve behandelopties met de patiënt besproken, met de mogelijkheid voor toekomstige herbeoordeling door het multidisciplinair handenteam.

Van juli 2011 tot en met mei 2017 werden 66 patiënten met CP (in de leeftijd van 7-20 jaar) doorverwezen naar het multidisciplinaire handenteam. Het effect van chirurgie aan de bovenste extremiteit werd bij 39 patiënten 4 maanden vóór de operatie en 9 maanden na de operatie geëvalueerd. Vijf patiënten hadden nog geen operatie gehad op het moment dat de effecten werden geëvalueerd. Tweeëntwintig patiënten werden na de multidisciplinaire beoordeling en shared decision-making niet geselecteerd voor chirurgie, waaronder 2 kinderen die zelf de voorkeur gaven aan een niet-chirurgische interventie, ondanks een positief oordeel van het multidisciplinaire handenteam over de geschiktheid voor operatie.

De resultaten van deze studie lieten een statistisch significante verbetering zien 9 maanden na de operatie vergeleken met voor de operatie. Respectievelijk 80% en 77% van de patiënten scoorden klinisch significant beter op de COPM-uitvoering en -tevredenheid. Het gebruik van de aangedane hand gemeten met de (Ad-)AHA gaf bij 71% een klinische relevante verbetering. Bij patiënten voor wie het primaire doel was om het uiterlijk van de aangedane arm en hand te verbeteren, werd tevens een verbetering op het gebruik van de hand(en) en patiënt-specifieke functionele doelen gemeten.

Selectie voor operatie vond plaats op basis van zorgvuldige multidisciplinaire beoordeling en bespreking en shared-decision-making. We kunnen concluderen dat een chirurgische ingreep aan de bovenste extremiteit waarvoor de indicatiestelling op deze manier tot stand is gekomen bij meer dan 80% van de geselecteerde kinderen en adolescenten met CP tot klinisch relevante verbeteringen leidt van zowel de patiënt specifieke functionele en / of cosmetische doelen als de handvaardigheid.

In **Hoofdstuk 7** worden de resultaten van de individuele studies in dit proefschrift beschreven en in een breder perspectief geplaatst. Hierin wordt op vier onderwerpen kritisch gereflecteerd: (i) interpretatie van de Assisting Hand Assessment scores; (ii) meetinstrumenten en beoordeling van verschillende constructen; (iii) verschillen in patiëntenpopulaties en de consequentie voor generaliseerbaarheid; en (iv) het meten van spontaan gebruik van de arm en hand in een klinische setting. Tot slot worden klinische implicaties besproken en worden aanbevelingen gedaan voor toekomstig onderzoek.

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•

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•

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•

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•

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•

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Leden van de promotiecommissie: prof. dr. C.M.A.M. van der Horst, prof. dr. R.H.H. Engelbert, prof. dr. J.B. van Goudoever, Dr. A.I. Buizer, prof. dr. M.W.G. Nijhuis-van der Sanden en prof. dr. C.K. van der Sluis, hartelijk dank voor de bereidheid mijn proefschrift kritisch te lezen en zitting te nemen in de promotiecommissie.

•

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•

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•

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Mijn paranimfen, Marianne Immink en Karin Boeschoten. Marianne, we zien elkaar chronisch te weinig, maar altijd als we elkaar zien of spreken is het gezellig en goed en de band die is gegroeid tijdens onze opleiding ergotherapie in Amsterdam blijft bestaan. Dank voor je onvoorwaardelijke vriendschap, ik ben blij dat je mij als paranimf terzijde wilt staan. Karin, dat jij mij als paranimf bijstaat vind ik een buitengewoon geruststellend idee, de passie voor het vak (kinder-) ergotherapie en de AHA is wat ons onder meer bindt. Ik leerde je kennen als organisator en docent van mijn eerste AHA cursus en vond in jou een fijn maatje om te sparren over mijn onderzoeksplannen. Dank voor je fijne vriendschap!

•

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•

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•

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Curriculum Vitae
Portfolio
Publications

Curriculum Vitae

Annoek Louwers (1972) graduated from Sint Lucas Creative Arts Academy in Boxtel in 1994 and then did her Bachelors in occupational therapy at Amsterdam University of Applied Sciences (Hogeschool van Amsterdam). She began as an occupational therapist at Eemland Hospital (now: Meander Medisch Centrum) in Amersfoort in 1998 where she initially worked with adults before specializing in paediatric occupational therapy. In 2000, she moved to De Trappenberg Rehabilitation Centre in Huizen.

Her interest in science combined with occupational therapy led her to enrol in a Masters in Evidence-Based Practice (EBP) at the Academic Medical Center (AMC) at the University of Amsterdam in 2006. She had the opportunity there to combine research and patients, especially children's, care in the Department of Rehabilitation and, since graduating in 2009, has coordinated EBP projects for allied health professionals in this department.

Annoek has specialized in the treatment of children with hand problems due to spasticity (cerebral palsy) and congenital hand deformities for the past fifteen years. In addition to her work as an occupational therapist, she has participated in various research projects. She began her PhD research entitled 'Effective Use of the Assisting Hand in Adolescents with Cerebral Palsy' in 2012. This included developing the Assisting Hand Assessment for adolescents with unilateral CP and evaluating the effect of two different interventions, namely functional hand orthosis and upper extremity surgery, on hand use and daily performance in children and adolescents with cerebral palsy. Her research was supervised by Prof. dr. Frans Nollet and Dr. Anita Beelen from the AMC in Amsterdam and Dr. Lena Krumlinde-Sundholm from the Karolinska Institutet in Stockholm.

Portfolio

Name PhD candidate: Annoek Marleen Louwers
PhD period: June 2012 – December 2017
Name PhD supervisor: Prof. dr. F. Nollet

PhD training

Courses (9.5 ECTS)	Year	Hours	ECTS
AHA course Amsterdam	2012	32	1.0
Scientific Writing in English for Publication. AMC Graduate School	2013	42	1.5
Practical Rasch Measurement - Core Topics. Statistics.com USA	2013	60	2.1
Oral presentation. AMC Graduate School	2014	22	0.8
Rasch workshop (Karolinska Institutet, Stockholm)	2014	14	0.5
"AHA-teaching" course	2014	32	1.0
e-BROK ('Basiscursus Regelgeving Klinisch Onderzoek')	2015	28	1.0
Clinical Epidemiology: Systematic Reviews. AMC Graduate School	2016	20	0.7
AHA and refresher course Amsterdam	2016	16	0.5
ENDNOTE X7. AMC Graduate School	2017	2.5	0.1
GRADE. AMC Graduate School	2017	7.0	0.3

Attended conferences (5.0 ECTS)			
4th International Cerebral Palsy Conference (ICPC) Pisa, Italy	2012	32	1.0
World Congenital Hand Symposium, Rotterdam, The Netherlands	2015	32	1.0
The 27th annual meeting of the European Academy of Childhood Disability (EACD), Copenhagen, Denmark.	2015	32	1.0
International Conference on Cerebral Palsy and other Childhood-onset Disabilities (5th International Conference of Cerebral Palsy (ICPC), 28th Annual Meeting of the European Academy of Childhood Disability (EACD) 1st Biennial Meeting of the International Alliance of Academies of Childhood Disability (IAACD) EACD Stockholm	2016	32	1.0
European Academy of Childhood Disability (EACD), Amsterdam	2017	32	1.0

Presentations at conferences (4.0 ECTS)			
Poster presentations			
COTEC, Stockholm: "Efficacy of a wrist and thumb brace on bimanual activities in children with hemiplegic cerebral palsy"	2012	14	0.5
EACD Stockholm: "Reliability of the adolescent version of the Ad-AHA and alternate-form reliability of different test activities of the AHA in adolescents with unilateral CP"	2016	14	0.5
7th MOVE meeting (Faculty of Behavioural and Movement Sciences)	2016	14	0.5
Oral presentations			
World Congenital Hand Symposium, Rotterdam, The Netherlands: "Assisting Hand Assessment 18 months to 18 years: development and validation".	2015	14	0.5
EACD Kopenhagen	2015	14	0.5
EACD Stockholm: "Development of the Assisting Hand Assessment for adolescents (Ad-AHA) and validation of the AHA for the whole age range 18 months to 18 years"	2016	14	0.5
EACD Stockholm: Instructional course "Measuring bimanual hand use across the ages in children with cerebral palsy".	2016	14	0.5
EACD Amsterdam: Effect UES	2017	14	0.5

Teaching (3.0 ECTS)			
Workshop AHA-meeting, Stockholm	2012	14	0.5
(Ad-)AHA course Amsterdam	2012	32	1.0
Ad-AHA course Amsterdam	2016	16	0.5
AHA instructors meeting (long-term vision)	2016	14	0.5
1e Symposium Nederlands Verwijsnet CP Handchirurgie	2017	14	0.5

List of publications

Louwers A, Warnink-Kavelaars J, Daams J, Beelen A. Effects of upper extremity surgery on hand use and activity performance in children and adolescents with cerebral palsy: a systematic review. Submitted

Louwers A, Warnink-Kavelaars J, Obdeijn M, Kreulen M, Nollet F, Beelen A. Effects of upper extremity surgery on manual performance in children and adolescents with cerebral palsy: a multidisciplinary approach using shared-decision making. Submitted

Louwers A, Meester-Delver A, Folmer K, Nollet F, Beelen A, Immediate effect of a wrist and thumb brace on bimanual activities in children with hemiplegic cerebral palsy. Developmental Medicine & Child Neurology, 2011; 53(4), 321-326.

Louwers A, Beelen A, Holmefur M, Krumlinde-Sundholm L. Development of the Assisting Hand Assessment for adolescents (Ad-AHA) and validation of the AHA from 18 months to 18 years. Developmental Medicine & Child Neurology. 2016; 58(12), 1303-1309.

Louwers A, Krumlinde-Sundholm L, Boeschoten K, Beelen A. Reliability of the Assisting Hand Assessment in adolescents. Developmental Medicine & Child Neurology. 2017

Kreulen M, Meester-Delver A, Burger-Brouwer CMJ, Louwers AM, Chirurgische behandeling van de spastische hand bij patiënten met cerebrale parese. NED TIJDSCHR HANDTHER 2008;17 (1):24-27

Meester-Delver A, Kreulen M, Burger-Brouwer CMJ, Louwers AM, Het handenteam voor patiënten met spasticiteit van arm en hand. NED TIJDSCHR HANDTHER 2008;17 (1):20-23

Verkerk GJQ, Wolf MJMAG, Louwers AM, Meester-Delver A, Nollet F, De reproduceerbaarheid en validiteit van de Canadian Occupational Performance Measure bij ouders van kinderen met beperkingen. NED TIJDSCHR ERGOTHER 2007;28:28-34

Verkerk GJQ, Wolf MJMAG, Louwers AM, Meester-Delver A, Nollet F, The reproducibility and validity of the Canadian Occupational Performance Measure in parents of children with disabilities. CLIN REHABIL 2006;20 (11):980-988

Contribution of authors

Chapter 2: Louwers AM, Beelen A, Holmefur M, Krumlinde-Sundholm L. Development of the Assisting Hand Assessment for adolescents (Ad-AHA) and validation of the AHA from 18 months to 18 years. Developmental Medicine & Child Neurology 2016, 58: 1303–1309. AL was responsible for the conduct of the study, the design of the (statistical) analytic plan, the (statistical) analysis, interpretation of the data, drafting and revision of the manuscript. AB contributed to the conceptualization of the study, interpretation of the data, and the revision of the manuscript. MH and KS contributed to the conceptualization of the study, (statistical) analysis, interpretation of the data, and the revision of the manuscript.

Chapter 3: Louwers AM, Krumlinde-Sundholm L, Boeschoten K, Beelen A. Reliability of the Assisting Hand Assessment in adolescents. Developmental Medicine & Child Neurology 2017. AL was responsible for the conduct of the study, the design of the statistical analytic plan, the statistical analysis, interpretation of the data, drafting and revision of the manuscript. KB contributed to the conduct of the study, and revision of the manuscript. LKS contributed to conceptualization of the study, the design of the statistical analytic plan, the statistical analysis, interpretation of the data, and revision of the manuscript. AB contributed to conceptualization of the study, the design of the statistical analytic plan, the statistical analysis, interpretation of the data, and revision of the manuscript.

Chapter 4: Louwers AM, Meester-Delver A, Folmer K, Nollet F, Beelen A. Immediate effect of a wrist and thumb brace on bimanual activities in children with hemiplegic cerebral palsy. Developmental Medicine & Child Neurology 2011, 53: 321–326. AL was responsible for the conduct of the study, the design of the statistical analytic plan, the statistical analysis, interpretation of the data, drafting and revision of the manuscript. AMD and KF contributed to the conduct of the study and revision of the manuscript. FN contributed to conceptualization of the study, interpretation of the data, and revision of the manuscript. AB contributed to the conceptualization and conduct of the study, interpretation of the data, and revision of the manuscript.

Chapter 5: Louwers AM, Warnink-Kavelaars J, Daams J, Beelen A. Effects of upper extremity surgery on hand use and activity performance in children and adolescents with cerebral palsy: a systematic review. AL was responsible for writing of the protocol and review, screening of titles and abstracts, assessment for inclusion, 'Risk of bias' assessment, data extraction, data entry into RevMan, data analysis, assessment of quality of evidence, and interpretation of results. AB contributed to writing of the protocol and review, screening of titles and abstracts, assessment for inclusion, 'Risk of bias' assessment, data extraction, data analysis, assessment of quality of evidence, and interpretation of results. JD contributed to the revision of the manuscript and was responsible for search strategy. JW contributed to writing of the protocol and revision of the manuscript.

Chapter 6: Louwers AM, Warnink-Kavelaars J, Obdeijn M, Kreulen M, Nollet F, Beelen A. Effects of upper extremity surgery on manual performance in children and adolescents with cerebral palsy: a multidisciplinary approach using shared decision-making. Submitted. AL was responsible for the conduct of the study, the design of the statistical analytic plan, the statistical analysis, interpretation of the data, drafting and revision of the manuscript. JW

was responsible for the conduct of the study, and contributed to the revision of the manuscript. AB contributed to the conceptualization of the study, oversaw all aspects of the conduct of the study, oversaw analyses, interpreted data, and contributed to the revision of the manuscript. JW, MO and MK contributed to the conceptualization of the study, interpretation of the data, and the revision of the manuscript. FN contributed to the conceptualization and conduct of the study, interpretation of the data, and revision of the manuscript.

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STICHTING Vrienden van De Trappenberg

Handfast

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