# Measuring upper extremity muscle strength in children with Unilateral Spastic Cerebral Palsy

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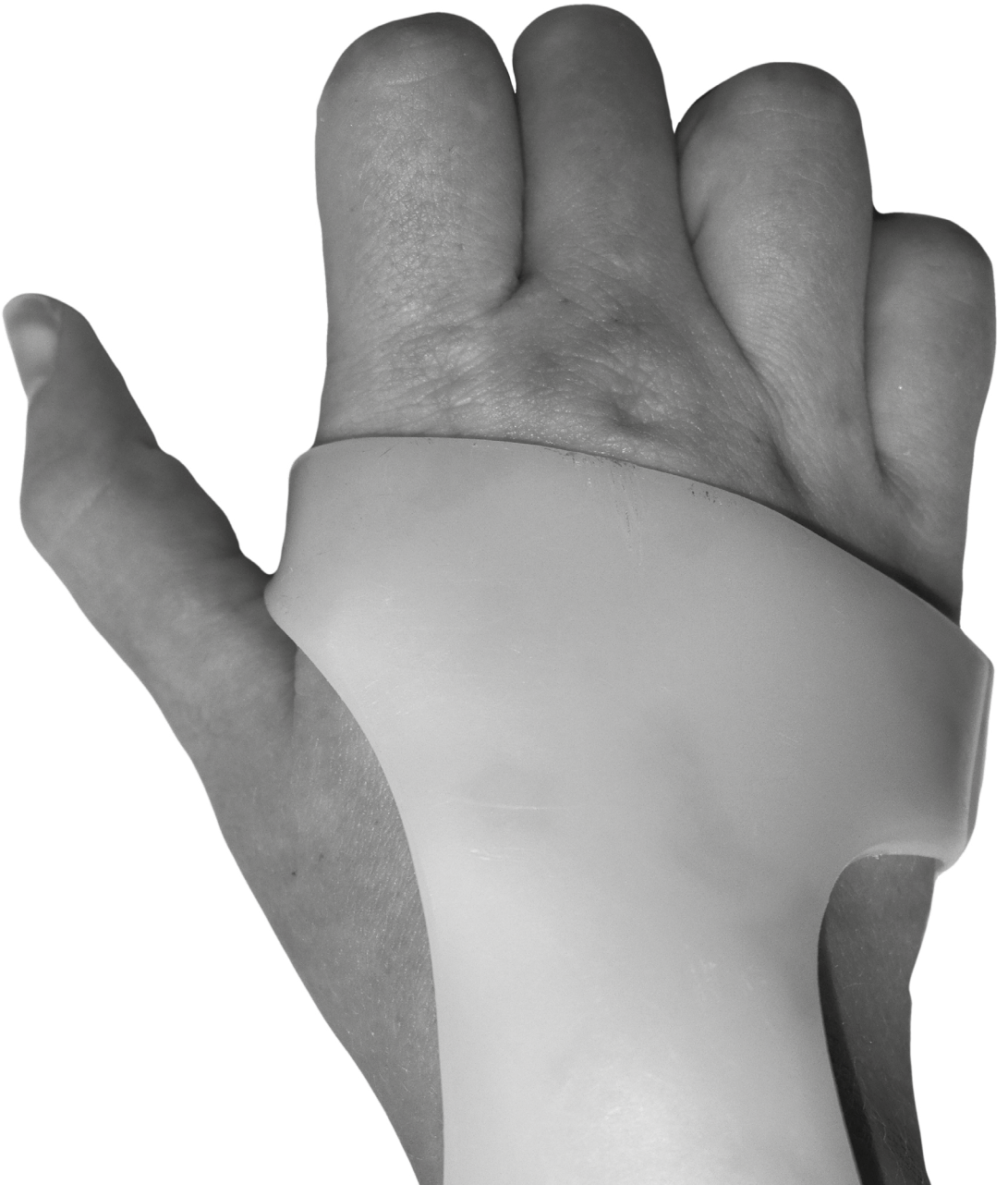

# Chapter 1

## General Introduction

"Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, behavior, by epilepsy, and by secondary musculoskeletal problems."[1]

Worldwide, CP is the most common motor disorder in childhood, with a prevalence in Europe of about 1.8–2.1 per 1,000 live births.[2–5] Within the Dutch pediatric rehabilitation, CP is the largest group (>32%) receiving interdisciplinary pediatric rehabilitation treatments.[6] Abnormal motor behavior (reflecting abnormal motor control) is the core feature of CP.[1]

The Surveillance of Cerebral Palsy in Europe (SCPE),[7] divides CP into three groups based on the predominant neuromotor abnormality; spastic, dyskinetic and ataxic CP.[7] By far the largest group (>85%) consists of children with spastic CP.[8] This group can be divided in bilateral and unilateral spastic CP. Almost 30% of all the children with CP is diagnosed with unilateral spastic CP (USCP).[8]

To distinguish on the basis of the severity of the motor impairments of the children with CP, several classification systems are used. The Gross Motor Function Classification System–Expanded & Revised (GMFCS- E&R) is a 5-level classification system that describes the gross motor function of children and youth with CP on the basis of their self-initiated movement with particular emphasis on sitting, walking, and wheeled mobility. Distinctions between levels are based on functional abilities, the need for assistive technology, including hand-held mobility devices (walkers, crutches, or canes) or wheeled mobility, and to a much lesser extent, quality of movement.[9,10]

To distinguish between the manual abilities of children with CP, the Manual Ability Classification System (MACS) is available. It describes how children with CP use their hands to handle objects in daily activities. The five levels are based on the children's self-initiated ability to handle objects and their need for assistance or adaptation to perform manual activities in everyday life.[11]

For both classification systems, a higher number in the classification level means more impairments.

## Unilateral Spastic Cerebral Palsy

USCP is characterized by motor impairments lateralized to one side of the body, resulting in an "affected" and a "non-affected" body side.[7,12,13]

In terms of gross- and fine motor functions, children with USCP have quite more possibilities compared to other types of CP. Mostly, children with USCP are classified with GMFCS level-I (walks without limitations) or level-II (walks with limitations). The difference between these levels is that children and youth in level-II have limitations in walking long distances and balancing; may need a hand-held mobility device when first learning to walk and may use wheeled mobility when traveling long distances outdoors and in the community. Children with GMFCS level-II further require the use of a railing to walk up and down stairs and are not as capable of running and jumping, compared to children with GMFCS level-I.[9]

Concerning the MACS-level, children with USCP are mostly classified with MACS level I (handles objects easily and successfully), MACS level-II (handles most objects but with somewhat reduced quality and/or speed of achievement) or MACS level-III (handles objects with difficulty; needs help to prepare and/or modify activities). In children with USCP, there is no relation between the GMFCS-classification and MACS-classification.[14]

## Arm hand functioning in children with USCP

Children with USCP are in the functional context, mostly more limited in manual ability than in gross motor function.[15] They usually have difficulty with grasping, reaching, releasing, and manipulating objects with the affected upper extremity (UE).[16,17]

Children with USCP seldom use their affected UE spontaneously in daily activities.[18,19] And if they use their affected hand, it will be as an assisting hand. Even with only minor impairment of the affected hand, they do not use it to its full potential in bimanual tasks. This lack of spontaneous use is referred to as 'developmental disregard' or 'learned non-use'.[17]

Although many basic activities can be performed with one UE, the performance of numerous complex activities in daily life requires the use of both arms and hands. However, impairments in bimanual fine motor function are found in about 60% of the children between the age of 4 and 16 years.[20,21] These impairments in bimanual fine motor function are characterized in particular by difficulties in the spatial and temporal aspects of bimanual coordination[22], and these difficulties in planning and performing bimanual activities negatively affect the independence, participation and Quality of Life of the children with USCP.[23] The reduced performance of the affected-UE in children with USCP is caused by several factors, like disturbances in passive and active range of motion (ROM), muscle tone, sensibility and muscle strength.[24–26] Muscle weakness appears to be one of the most important factors for the impairments in carrying out UE-activities.[24–26]

While the performance of the non-affected UE is often assumed to be within expected norms of children with typical development, several studies found reduced performance in hand function of the non-affected UE, compared to TD-children, e.g. on the Jebsen-Taylor Test of Hand Function, on a computerised version of the Peg Moving Task, on the Box and Blocks Test for dexterity and on the Tyneside Pegboard Test.[27–30]

However, whether as in the affected UE, limitation in muscle strength appears to be one of the most important factors in this reduced performance, is unknown.

## Upper extremity muscle strength in USCP

The muscle strength of the affected UE in children with USCP is typically lower compared to the strength of the non-affected UE (up to 64% less muscle strength).[31] Compared to the muscle strength of TD-children, the affected UE also has reduced muscle strength (up to 84% less muscle strength).[32] Furthermore, compared to their TD-peers, the affected UE of children with USCP 1) is slower in the application of force;[31,33–35] 2) shows sequential force generation;[33,36] and 3) has a reduced ability to adjust grasping forces to the object's physical properties.[37–40]

Regarding the non-affected UE, only two studies have investigated the muscle strength of the non-affected hand in children with USCP, with opposite conclusions. Rich et al. concluded that grip strength of the non-affected hand of the children with USCP is on average 12% less compared to TD-group.[27] However, Tomhave et al. concluded that there are no significant differences when the muscle strength of the non-affected hand was compared with norm-values.[29] It is therefore unclear whether muscle weakness in the non-affected UE exists and when it exists, whether this muscle weakness also exists in the other arm muscles.

## Measuring upper extremity muscle strength

The purpose of measuring UE strength can be either discriminative or evaluative. If the goal of the measurement is discriminative, the clinician wants to detect the possible existence of muscle weakness. If the goal of the measurement is evaluative, the therapist wants to measure whether changes in muscle strength occur, for example as a result of UE muscle strength training or due to the development of the child.

Muscle strength is assessed at the body functions level of the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY)[41] and several methods are available to measure muscle strength: dynamic (concentric, eccentric, isotonic and isokinetic) and static (isometric).[42] During dynamic muscle strength measurements the child needs to cooperate with the examiner and perform maximal contractions in one muscle group. Due to spasticity and/or cognitive limitations, this is a task that many children with CP find very difficult to perform.[43,44] In static/isometric muscle strength measurements, the ability of a muscle group to produce force without a change in overall muscle-tendon length is measured; therefore, limitations by spasticity (and range of motion) will not interfere with task performance during this measurement. Furthermore, in most daily activities the affected UE is used as an assisting hand (e.g. holding, stabilizing, carrying), and in these tasks a high percentage of maximum isometric muscle strength is needed.

For UE muscle strength measurements, the National Institutes of Health Common Data Elements (NIH-CDE) and the Dutch-guideline "spastic cerebral palsy in children" recommend Maximum Voluntary Isometric Contraction Testing, using Hand Held Dynamometers and grip/pinch dynamometer.[45,46] There are two methods to measure muscle strength with a Hand Held Dynamometer (HHD); the "make method" and the "break method". During the make method, the child applies maximum force against a fixed HHD. With the break method, the force of the examiner exceeds the maximum force of the child. In children with spastic CP, the make method is recommended.[47]

In most daily life manual activities, e.g. during carrying/moving a heavy box, not only a certain amount of muscle strength is required, but also the ability to maintain/regulate that strength for a certain time.

This ability is called functional strength and it needs to be measured during the performance of that specific task.

For children with CP, the "functional strength measurement" (FSM) test exists, measuring both lower extremity and UE muscle strength during the performance of functional tasks.[48] With this test, dynamic maximal muscle strength and submaximal strength during 30 second repetitive measure-

ments are recorded. Unfortunately, the ability to maintain the maximum functional muscle strength in a static sustained contraction cannot be measured with this test. Although the subtests of the FSM are functional based, they are also not tailored to relevant activities for children with USCP.

Because of the aforementioned reasons, two specific functional muscle-strength tests measuring unimanual and bimanual sustained contraction, i.e. the "Cup-Task" for determining maximal functional unimanual upper extremity strength, and the "Box-Task" for determining maximal functional bimanual upper extremity strength, were developed. In both tests, a combination of functional grip- and arm strength is measured by lifting the Box or Cup, which must be sustained for five seconds. In a pilot study, both tests were found to be feasible in children with USCP.[49] However, extensive research into the clinimetric properties has not been performed yet.

## Strength training in children with USCP

Although there is no minimum age requirement at which children can begin strength training, all participants must be mentally and physically ready to comply with coaching instructions and undergo and cope with the stress of a training program; in general this is possible if a child is ready for participation in sport activities (generally age 7 or 8 years).[50] Therefore, intensive strength training programs are often applied to children in elementary school.

About the effects of UE-muscle strength training in children with USCP, limited information is available. Rameckers et al. performed a critical review of the efficacy of upper limb strengthening in children with spastic CP.[51] Six upper limb strength training studies were found. Two RCTs [52,53] investigated the effect of stand-alone strength training (not combined with other interventions) of the upper limb in children with CP. In both studies small to large positive effects (2.7–58.9%) on muscle strength were reported whereas controls did not show any effects (5.3%). Four studies [54–57] were included in which a second intervention was added to the strength training (neuro muscular electro stimulation or Botulinum Toxin A). Overall an increase of muscle strength was shown, ranging from 0.1 to 105% directly after the intervention and from 41.8 to 84.7% at 3 months follow up [54,58], indicating a large variability.

Because of the large variability in the results of strength training (and especially in case of small improvements), measurement instruments with sound clinimetric properties are needed to properly interpret the outcome of such programs.

## Clinimetric properties of UE-muscle strength measurement instruments for children with USCP

In order to be able to make inferences about muscle strength, either in clinical practice or in research, strength has to be measured with an instrument that has sound clinimetric properties, related to reliability, validity and responsiveness.

To know to what extent the strength measurement instrument is suitable for discriminative and evaluative purposes in clinical practice, the clinimetric properties 'reliability' and 'responsiveness' of the measurement instrument need to be known. The reliability indicates the degree to which the measurement is free from the measurement error.[59] With the corresponding ICC-value it can be

determined whether the measurement instrument is usable for discriminative purposes. The related Standard Error of Measurement (SEM) is a measure of how far apart the outcomes of repeated measurements are; it is the standard deviation (SD) around a single measurement.[60]

Responsiveness indicates the ability of a measurement instrument to detect changes over time in the construct to be measured.[59]

For the interpretation of a change in score, the Smallest Detectable Change (SDC) and the minimally important change (MIC) are important. With the SDC it can be determined whether the difference between two (evaluative) measurements can be distinguished from a measurement error. In order to know whether a change score is also clinically important, the MIC is considered the most important value. The MIC is the smallest change score in the construct to be measured that patients, clinicians or relevant others perceive as important.[60]

The quality of each clinimetric property can be rated as positive, negative or indeterminate, according to international accepted criteria.[61]

Besides sound clinimetric properties, the methodological quality of the study design in which the clinimetric properties have been determined is of importance. The COnsensus-based Standards for the selection of health Measurement Instruments (COSMIN) is an initiative of an international multidisciplinary team of researchers with a background in epidemiology, psychometrics, medicine, qualitative research, and health care, who have expertise in the development and evaluation of outcome measurement instruments. By means of a Delphi study they developed a checklist with which the methodological quality of the study design of each clinimetric property can be determined.[62] When results of the methodological quality and rating of the statistical findings for the individual studies are combined, the overall level of evidence for the quality of the clinimetric properties of an instrument can be determined. The more studies of good methodological quality that report consistent clinimetric findings, the stronger the level of evidence of the investigated clinimetric property is considered to be.[61] In studies of excellent methodological quality, the results of clinimetric properties have a high level of evidence. In a studies of poor methodological quality, the results of the clinimetric properties have an unknow level of evidence rating.[63]

Due to the unique characteristics of children with USCP, such as disturbances in passive and active ROM, muscle tone, sensibility and muscle strength, the clinimetric properties of strength measurement instruments used for children with USCP should be studied specifically within this group.

## Scope and outline of this thesis

To get a detailed overview of the existing upper extremity muscle strength tests for children with CP, first a systematic review regarding their different clinimetric properties has been performed. Reported clinimetric properties were derived from the studies, but also the methodological quality of the studies was determined. A data-synthesis of both was performed to determine which measurement instruments are useable in clinical practice. (see Chapter 2)

Based on these results, only two instruments were identified as potential useful UE muscle strength measurement instruments. Next, the test-retest and interrater reliability of these measurement instruments in children with USCP was investigated, in a study designed using the guidelines of the COSMIN consortium (see Chapter 3).

Because none of the existing measurement instruments measures upper extremity strength in the context of functional activities, in which muscle strength must be maintained, we determined reliability and validity of two new functional muscle strength measurement instruments in children with USCP, also following the guidelines of the COSMIN consortium (see Chapter 4).

As only two studies compared the muscle strength of the non-affected UE of children with USCP to TD-children[27,29] and had opposite conclusions whether muscle weakness only occurs in the affected UE, we performed a study to compare the isometric muscle strength (measured with the HHD and E-link) of the non-affected UE of children with USCP to TD-children. (see Chapter 5)

In the last study, we present a critical perspective on how to interpret changes in muscle strength as measured with often used measurement instruments for the affected and the non-affected UE, taken the minimal important change and the measurement error of the measurement instruments into account. (see Chapter 6)

Chapter 7 includes the general discussion, evaluating and integrating the main results. Methodological considerations are discussed and implications for clinical practice and future research are presented.

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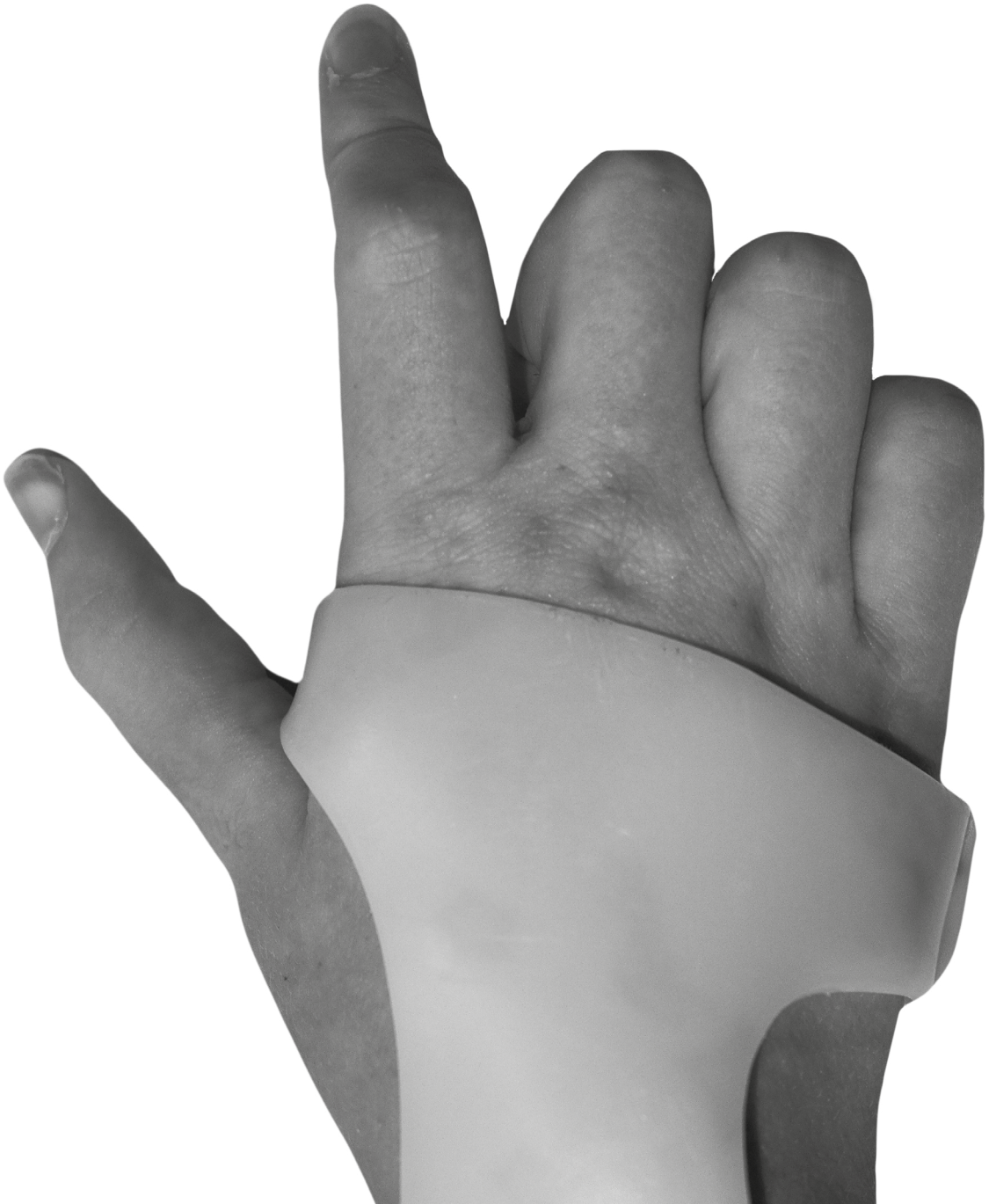

# Chapter 2

# Upper Extremity Strength Measurement for Children with Cerebral Palsy: A Systematic Review of Available Instruments

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## Abstract

**Background.** In order to make inferences about strength related to development or treatment interventions, it is important to use measurement instruments that have sound clinimetric properties.

**Purpose.** The objective of this review is to systematically evaluate the level of evidence of the clinimetric properties of instruments for measuring upper extremity muscle strength at the "body functions & structures" level of the International Classification of Functioning, Disability and Health for Children and Youth (ICF- CY) for children with cerebral palsy (CP).

**Data Sources.** A systematic search of the PubMed, EMBASE, OTseeker, CINAHL, PEDro, and MEDLINE databases up to November 2012 was performed.

**Study Selection.** Two independent raters identified and examined studies that reported the use of upper extremity strength measurement instruments and methods for children and adolescents with CP aged 0 to 18 years.

**Data Extraction.** The COSMIN (COnsensus-based Standards for the selection of health status Measurement INstruments) checklist with 4-point rating scale was used by 2 independent raters to evaluate the methodological quality of the included studies. Best evidence synthesis was performed using COSMIN outcomes and the quality of the clinimetric properties.

**Data Synthesis.** Six different measurement instruments or methods were identified. Test-retest, interrater, and intrarater reliability were investigated. Two test- retest reliability studies were rated as "fair" for the level of evidence. All other studies were rated as "unknown" for the level of evidence.

**Limitations.** The paucity of literature describing clinimetric properties, especially other than reliability, of upper limb strength measurement instruments for children with CP was a limitation of the study.

**Conclusions.** For measuring grip strength, the Jamar dynamometer is recommended. For other muscle groups, handheld dynamometry is recommended. Manual muscle testing (MMT) can be used in case of limited (below MMT grade 4) wrist strength or for total upper limb muscle strength. Based on lacking information regarding other clinimetric properties, caution is advised regarding interpretation of the results.

# Introduction

The term "cerebral palsy" (CP) describes a group of disorders of the development of movement and posture, causing activity limitations that are attributed to non- progressive impairments, that occur in the developing fetal or infant brain. Motor disorders in people with CP are often accompanied by disturbances of sensation, cognition, communication, perception, or behavior or a seizure disorder, or both.[1] Abnormal motor behavior (reflecting abnormal motor control) is the core feature of CP. It is characterized by various abnormal patterns of movement and posture related to defective coordination of movements or regulation of muscle tone.[2]

One of the effects of abnormal motor behavior is the loss of muscle strength. Children with CP have less strength in their affected side or sides compared with their peers who are developing typically.[3–5] Although some studies have focused on the loss of muscle strength in the lower extremities and evolving impairments of related activities,[3,6–8] the decrease in muscle strength of the upper extremities also may lead to limitations in activities of daily living, as grip strength is found to be a good predictor of use of the affected arm in bimanual performance in children with CP.[9,10] To determine whether strength is a limiting factor in the performance of activities of daily living, it is important to measure strength accurately.

Muscle strength is assessed at the body functions level of the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY)[11] and can be measured in 3 different ways: isometric, isotonic, and isokinetic.[12] In order to make inferences about strength, either in clinical practice or in research, strength has to be measured with an instrument that has sound clinimetric properties. Reliability, for example, is a very important property, among others such as validity and responsiveness. One needs to know the degree to which variations in results between repeated measurements occur. This so-called measurement error can arise from several sources: the measurement instrument itself, the person or people performing the measurement, the patient undergoing the measurement, and the circum- stances under which the measurement is performed.[13] The more studies of good methodological quality that report consistent clinimetric findings, the greater or stronger the level of evidence of the investigated clinimetric property is considered to be.[14]

Several studies have examined clinimetric properties of upper extremity strength measurement instruments for children who are developing typically. In most of these studies, test-retest, intrarater, and interrater reliability of handheld dynamometers measuring isometric muscle and grip strength in the upper extremities in children revealed excellent intraclass correlation coefficients (ICCs).[15–21] Moreover, evaluations of the validity of measurement instruments of isometric upper extremity muscle strength in children demonstrated excellent scores.[15,16,22] Studies that examined the clinimetric properties of lower extremity strength measurement instruments in children with CP revealed moderate to excellent intra- rater and interrater reliability.[21,23–27] Studies that examined strength measurement instruments for adults with brain damage showed excellent intrasession and intersession, test-retest, and intrarater reliability scores for the paretic side.[28–30] The nonparetic side showed moderate to excellent intrasession and intersession reliability scores.[28]

During isotonic and isokinetic muscle strength testing, the patient needs to be able to cooperate with the examiner and perform a maximum contraction of one muscle group. This is a task that many children with CP find very difficult to perform due to co-contraction of antagonists or agonists

or cognitive limitations, or both.[31,32] Furthermore, compared with their healthy peers, children with CP: (1) are slower in the application of force,[4,33–35] (2) show sequential force generation,[33,36] (3) have a reduced ability to adjust grasping forces to the object's physical properties,[37–40] and (4) have impaired motor planning.[33,34,41] Children with CP also have impairments in the spatial and temporal aspects of bimanual coordination.[42] Due to the unique characteristics of children with CP, the clinimetric properties of strength measurement instruments used for these children should be studied specifically in this group.

To our knowledge, no systematic review has been published regarding the different clinimetric properties of upper extremity strength measurement instruments for children with CP. The purposes of this article are: (1) to systematically review the clinimetric properties of instruments that measure upper extremity muscle strength at the "body functions & structures" level of the ICF-CY for children with CP and (2) to systematically assess the methodological quality of the clinimetric studies and the strength of the evidence provided regarding the clinimetric properties.

# Methods

## Data Sources and Searches

Electronic searches were conducted in the PubMed, EMBASE, OTseeker, CINAHL, PEDro and, MEDLINE databases from the inception of these databases until November 2012. The COSMIN (Consensus-based Standards for the Selection of Health Status Measurement Instruments) protocol for the systematic review of measurement properties was used to search the PubMed database. According to this protocol, the search strategy consisted of collections of search terms for the following characteristics: construct of interest, target population, instrument search, and psychometric properties.[43] For construct of interest, the following terms were used: Power OR Muscle strength OR Resistance OR Strength OR Contraction OR Lift OR "Isometric contraction" OR "Isotonic contraction" OR "Isokinetic contraction" OR Grip OR Pinch OR Grasp OR Functional OR Function OR Exercise OR Physical fitness OR Endurance OR Tolerance. Target population was defined as: Human AND Child AND ("Cerebral palsy" OR "Muscle spasticity" OR Diplegic OR Diplegia OR Monoplegic OR Monoplegia OR Quadriplegic OR Quadriplegia OR Spastic OR "Spastic Cerebral Palsies" OR "Unilateral Cerebral Palsy" OR Ataxia OR Atactic OR Distonia OR Distonic OR Hemiplegic OR Hemiplegia) AND ("Upper limb" OR Arm OR Fore-arm OR "Upper extremity" OR Shoulder OR Elbow OR Hand OR Wrist OR Finger OR Thumb OR Manual). Because this study did not focus on one specific measurement instrument but on all instruments that are used to measure upper extremity muscle strength, the instrument search was not defined. All search terms were combined with the filter for measurement properties.[43] Finally, the exclusion filter (stroke OR animals) was added. For the other databases, the above- mentioned words were combined.

## Study Selection

Studies of any design that evaluated reliability, validity, or responsiveness were eligible for inclusion. Other inclusion criteria were: (1) the study participants were children and adolescents (0–18 years of age) with CP; (2) the study examined a measurement instrument or measurement method for upper

extremity muscle strength (shoulder/elbow/ wrist/grip) at the "body functions & structures" level of the ICF-CY.11 No language restrictions were applied. Studies were excluded if adult patients or children without CP were included in the study sample.

After performing the search strategies (K.D.), 2 reviewers (K.D. and E.R.) independently screened titles and abstracts for relevance. In cases of no consensus, the opinion of a third reviewer (Y.J.) was decisive. Additionally, related articles and the references of the included articles were checked by one reviewer (K.D.) for relevance and potential inclusion. These potentially eligible articles were then independently screened by the 2 reviewers.

After consensus was reached, full- text reports of the included studies were retrieved and read by the 2 reviewers independently. They searched the articles for a clinimetric property of the instrument used to measure upper extremity muscle strength in children with CP.

## Data Extraction and Quality Assessment

The extraction and assessment consisted of several steps. First, the descriptive characteristics of the sample used in the studies, the procedures used, and the statistical outcomes reported in each study were extracted. Second, the methodological quality of the studies was assessed. Third, the quality of the clinimetric properties of the measurement instrument was evaluated. Finally, a best evidence synthesis was performed.

## Rating of Methodological Quality of Individual Studies

Two reviewers (K.D. and E.R.) independently assessed the methodological quality of the included studies using the COSMIN protocol. In case of disagreement, discussion with the third reviewer (Y.J.) followed until consensus was reached.

To assess methodological quality, the reviewers used the COSMIN checklist with the 4-point rating scale, which is recommended for use in systematic reviews of clinimetric properties (www.cosmin.nl).[44] This standardized and validated scoring system was developed based on discussions among experts.44 This scoring system allows the overall methodological quality of one clinimetric property per study to be calculated. The checklist consists of 9 boxes that each describe a measurement property (ie, internal consistency, reliability, measurement error, content validity, structural validity, hypothesis testing, cross-cultural validity, criterion validity and responsiveness) and 2 sub- checklists to determine the interpretability and generalizability of the study. Each box contains between 5 and 18 items detailing how each specific clinimetric property should be assessed (see Appendix for the example of the reliability box). Each item is scored on a 4-point rating scale (ie, "poor," "fair," "good," or "excellent").[44] A methodological quality score is obtained per box by taking the lowest rating of any item in that box ("worse score counts"). For our study, in accordance with the COSMIN protocol, only the boxes that corresponded to the investigated clinimetric properties were completed. Relevant items in the "Interpretability" box and the "Generalizability" box were used as a guide for extracting other relevant data from the included studies.

## Rating of Statistical Findings for Individual Studies

One reviewer (K.D.) assessed the quality of the clinimetric properties of the measurement instrument in each study by applying widely accepted quality assessment criteria to the statistical outcomes (Tab. 1).[14] The overall ratings are "good" (+), "negative" (—), and "indeterminate" (?).[14]

## Data Synthesis

One reviewer (K.D.) combined the results of the rating of the methodological quality and the rating of the statistical findings for the individual studies to determine the overall level of evidence for the quality of the clinimetric properties of the identified measurement instruments of upper limb muscle strength. This method of synthesizing evidence is similar to the method that is used to synthesize evidence from clinical trials.[45] The possible levels of evidence are: (1) strong, (2) moderate, (3) limited, (4) conflicting, and (5) unknown (Tab. 2).[46]

**Table 1.** Rating System for the Statistical Findings for Individual Studies[14]

| Measurement Property | Rating | Quality Criteria |
|---|---|---|
| Internal consistency | + | Cronbach alpha ≥.70 |
| | — | Cronbach alpha < .70 |
| | ? | Cronbach alpha not determined |
| Reliability | + | ICC ≥.70 OR n ≥.70 |
| | — | ICC < .70 OR n < .70 |
| | ? | Neither ICC nor weighted kappa |
| Measurement error | + | MIC > SDC OR MID > SDC OR MIC outside the LoA |
| | — | MIC ≤ SDC OR MID ≤ SDC OR MIC equals or inside LoA |
| | ? | MIC not defined |
| Content validity | + | The target population considers all items in the questionnaire to be relevant AND considers the questionnaire to be complete |
| | — | The target population considers items in the questionnaire to be irrelevant OR considers the questionnaire to be incomplete |
| | ? | No target population involvement |
| Structural validity | + | Factors should explain ≥50% of the variance |
| | — | Factors explain < 50% of the variance |
| | ? | Explained variance not mentioned |
| Hypothesis testing | + | Correlation with an instrument measuring the same construct ≥.50 OR ≥75% of the results were in accordance with the hypotheses AND correlation with related constructs was higher than with unrelated constructs |
| | — | Correlation with an instrument measuring the same construct < .50 OR < 75% of the results were in accordance with the hypotheses OR correlation with related constructs was lower than with unrelated constructs |
| | ? | Solely correlations determined with unrelated constructs |

| Measurement Property | Rating | Quality Criteria |
|---|---|---|
| Cross-cultural validity | + | Original factor structure confirmed OR no important DIF between language versions |
| | — | Original factor structure not confirmed OR important DIF found between language versions |
| | ? | Confirmatory factor analysis not applied and DIF not assessed |
| Criterion validity (predictive or concurrent) | + | Convincing arguments that gold standard is "gold" AND correlation with gold standard ≥.70 |
| | — | Correlation with gold standard < .70 despite adequate design and method |
| | ? | No convincing arguments that gold standard is "gold" OR doubtful design or method |
| Responsiveness | + | Correlation with an instrument measuring the same construct ≥.50 OR at least 75% of the results are in accordance with the hypotheses OR AUC ≥0.70 AND correlation with related constructs is higher than with unrelated constructs |
| | — | Correlation with an instrument measuring the same construct < .50 OR < 75% of the results are in accordance with the hypotheses or AUC < 0.70 OR correlation with related constructs is lower than with unrelated constructs |
| | ? | Solely correlations determined with unrelated constructs |

*ICC = intraclass correlation coefficient, n = Cohen (weighted) kappa, SDC = smallest detectable change, MIC = minimal important change, MID = minimal important difference, LoA = limits of agreement, DIF = differential item functioning, AUC = area under the receiver operating characteristic curve, + = positive rating, — = negative rating, ? = indeterminate rating*

**Table 2.** Synthesis of Study Quality and Findings [46]

| Level | Rating | Criteria |
|---|---|---|
| Strong evidence | + + + or ———— | Consistent findings in multiple studies of good methodological quality or in one study of excellent methodological quality |
| Moderate evidence | + + or —— | Consistent findings in multiple studies of fair methodological quality or in one study of good methodological quality |
| Limited evidence | + or — | One study of fair methodological quality |
| Conflicting evidence | ± | Conflicting findings |
| Unknown evidence | ? | Only studies of poor methodological quality |

*+ = positive rating, — = negative rating, ± = conflicting rating, ? = indeterminate rating.*

# Results

## Study Selection

The selection procedures are summarized in the Figure. Seven eligible studies were identified, and 3 types of reliability (ie, intrarater, interrater, and test-retest) in 6 different measurement instruments were studied. The measurement instruments and methods used were: (1) manual muscle testing (MMT), (2) the Jamar dynamometer, (3) a handheld dynamometer (HDD), (4) an instrument based on muscle strength torque sensors, (5) a computerized measurement tool using a strain gauge, and (6) a modified sphygmomanometer. A more detailed description of the included studies is given in Table 3.

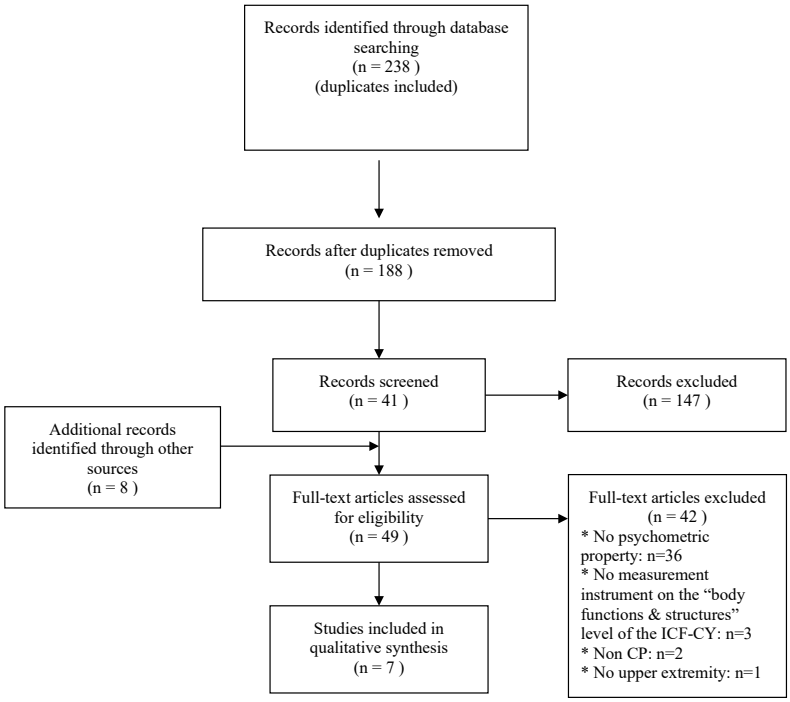


**Figure.** Flowchart of the search strategy and selection of articles.
*ICF-CY = International Classification of Functioning, Disability and Health for Children and Youth, CP = cerebral palsy*

*MMT.* Klingels et al.[47] investigated interrater reliability and test-retest reliability of MMT of shoulder flexion, abduction, and adduction; elbow flexion and extension; fore- arm supination and pronation; and wrist flexion and extension in children with CP. For test-retest reliability, ICC values between .88 and .96 were found. For interrater reliability, ICC values varied between .60 and .91.

*Jamar dynamometer.* Klingels et al.[47] investigated test-retest and inter- rater reliability of isometric grip strength of the upper extremity in children with CP using the Jamar dynamometer. Intraclass correlation coefficients of .96 and .95 were found for test-retest and interrater reliability, respectively.

*HHD.* Crowner and Racette[48] investigated test-retest reliability of the MicroFET HHD (Hoggan Health Industries Inc, Draper, Utah) for testing muscle strength of the shoulder and elbow muscles and the Baseline hydraulic HHD (FEI, Irving- ton, New York) for assessing grip strength. Vaz et al.[5] used a MicroFET2 HHD for investigating the test-retest reliability of muscle strength testing of the wrist flexors and extensors. In the study by Crowner and Racette,[48] a total score of .99 was mentioned without indicating what the score means or how it was calculated. In the study by Vaz et al,[5] ICC values between .93 and .98 were found for test-retest reliability of measurements of wrist flexion and extension.

*Muscle strength-torque sensors.* In the study by Bleyenheuft et al,[49] test-retest reliability of isometric fingertip grip muscle strength and load muscle strength was investigated. No significant difference was found between the first and second measurements (P = .935).

*Strain gauge technology.* Rameckers et al.[50] investigated test- retest reliability of maximum voluntary contraction (MVC) of the index flexor muscles. Intraclass correlation coefficient values of .99 were found for the finger and wrist flexors.

*Sphygmomanometry.* Glazier et al.[51] investigated test-retest reliability and Xu et al.[52] investigated intrarater reliability of grip strength measurements using a modified sphygmomanometer. Glazier et al[51] found a Pearson correlation coefficient of .97 for test-retest reliability and Xu et al[52] reported an ICC value of .919 for intrarater reliability of grip strength measurements.

## Rating of Methodological Quality of Individual Studies

For all 6 instruments, the COSMIN box for reliability could be completed to rate the methodological quality of the corresponding studies. Most studies were rated as "poor" for methodological quality. The studies of interrater reliability of MMT[47] and Jamar dynamometer[47] measurements were rated as "fair" for methodological quality (Tab. 4). According to the guidelines in the COSMIN manual,[53] the other boxes could not be completed.

## Rating of Statistical Findings for Individual Studies

The quality of the clinimetric properties was assessed for all 6 instruments (Tab. 4). Most clinimetric properties were rated as "good." Interrater reliability of shoulder and elbow measurements obtained with MMT was rated as "poor." Test-retest reliability of total upper extremity HHD measurements and of measurements of grip strength obtained with the modified sphygmomanometer were scored as "indeterminate" because no justified statistical method was used.

## Data Synthesis

The results of the methodological quality assessment and the quality assessment of the clinimetric proper- ties were combined and are presented in Table 5. For most of the instruments, the level of evidence was rated as "unknown." For upper extremity/wrist strength measurements with MMT and grip strength measurements with the Jamar dynamometer, the level of evidence was rated as "limited," with a positive rating of the clinical property. For shoulder/elbow muscle strength measurements with MMT, the level of evidence is rated as "limited," with a negative rating of the clinical property.

**Table 3.** Characteristics of Included Studies

| Measurement Instrument or Method | Description | Measurement Property/Study | Sample Characteristics | Time Interval Between Measurements | Test Protocol | Data Calculation |
|---|---|---|---|---|---|---|
| MMT | In MMT, the clinician applies a force to the target muscle or muscle group and rates the strength of the contraction according to an ordinal scale subjectively, as described by Daniels and Worthingham[59] | Interrater reliability Klingels et al[47] | N = 30 (13 girls, 17 boys) Age: $\bar{x}$ = 10.6 y, SD = 2.7, range = 5–15<br>18 children had right hemiplegia, 12 children had left hemiplegia<br>GMFCS[53]: level I = 22, level II = 8<br>Zancolli classification system[60]: pattern I = 23 children, pattern IIa = 3 children, pattern IIb = 1 child, pattern III = 3 children<br>17 children attended special school, 13 children attended mainstream school | 1 h | A detailed instruction manual was developed, and the assessors practiced together to standardize the test procedure | A total score for the upper extremity and subscores of all tested muscle groups were calculated |
| | | Test-retest reliability Klingels et al[47] | N = 23 | Around 2 wk | | |
| Jamar dynamometer | The Jamar dynamometer is an instrument for measuring isometric handgrip strength. It has 2 ergonomically designed metal handles stacked on top of each other that are separated by a hydraulic spring. | Interrater reliability Klingels et al[47] | N = 30 (13 girls, 17 boys) Age: $\bar{x}$ = 10.6 y, SD = 2.7, range = 5–15<br>18 children had right hemiplegia, 12 children had left hemiplegia GMFCS[53]: level I = 22, level II = 8<br>Zancolli classification system[60]: pattern I = 23 children, pattern IIa = 3 children, pattern IIb = 1 child, pattern III = 3 children<br>17 children attended special school, 13 children attended mainstream school | 1 h | The child was sitting with the arm adducted, the elbow flexed at 90°, and the forearm and wrist in the neutral position | Mean of 3 maximum voluntary contractions was recorded for each hand |
| | | Test-retest reliability Klingels et al[47] | N = 23 | Around 2 wk | | |

| Measurement Instrument or Method | Description | Measurement Property/Study | Sample Characteristics | Time Interval Between Measurements | Test Protocol | Data Calculation |
|---|---|---|---|---|---|---|
| HHD | The HHD is an electronic device that fits in the palm of a hand. A load cell (strain gauge technology) measures the isometric muscle strength applied to a transducer. | Test-retest reliability Crowner and Racette[48] | N = 2 | NR | The break method was used (the force of the examiner exceeds the force of the child). A detailed instruction manual was developed to standardize the test procedure. | NR |
| | | Test-retest reliability Vaz et al[5] | N = 11 (6 girls, 5 boys) Age: $\bar{x}$ = 8.5 y, SD = 1.8, range = 6–11 Zancolli classification system[60],[b] | 1 wk | The make method was used (the child applies force against a fixed HHD). A detailed instruction manual was developed to standardize the test procedure. The test order was randomized. | NR |
| Muscle strength-torque sensors | A handheld object, equipped with 2 parallel vertical grip surfaces of smooth brass (40 mm diameter, 30 mm apart) was developed. Two 3-dimensional muscle strength- torque sensors were used. | Test-retest reliability Bleyenheuft and Thonnard[49] | N = 12 (2 girls, 10 boys) Age range: 10–16 y<br>8 children had left hemiplegia,<br>4 children had right hemiplegia<br>GMFCS[53]: level I = 8 children, level II = 4 children | NR | Each child performed a sequence of 20 repeated trials under predictive conditions, followed by 10 trials under reactive conditions with each hand. The child gripped the handheld object so that it was stationary and then slowly released the thumb and index finger until the object slipped. | Four maneuvers preceded and followed the experiment to measure the coefficient of friction between the skin and the object |

| Measurement Instrument or Method | Description | Measurement Property/Study | Sample Characteristics | Time Interval Between Measurements | Test Protocol | Data Calculation |
|---|---|---|---|---|---|---|
| Strain gauge technology | A computerized measurement tool using a strain gauge | Test-retest reliability Rameckers et al[50] | N = 10 (6 girls, 4 boys) Age: $\bar{x}$– = 9.7 y, range = 4–16<br>7 children had left hemiplegia,<br>3 children had right hemiplegia<br>Zancolli classification system[60]:<br>pattern I = 5 children, pattern IIa = 1 child,<br>pattern IIb = 4 children | 1 mo | The child was asked to apply maximal muscle strength with the index and middle fingers positioned on the end of the lever. A detailed instruction manual was developed to standardize the test procedure. | NR |
| Modified sphygmomanometer | The sphygmomanometer was originally designed for measuring blood pressure. In the modified sphygmomanometer, the inflatable part is folded into 3 parts and contained in an elastic bag. It is an isotonic instrument that measures concentric contraction. The child squeezed the pouch, and the researcher read the millimeters of mercury displayed on the gauge to determine the core for each trial. | Test-retest reliability Glazier et al[51] | N = 19 (9 girls, 10 boys) Age: $\bar{x}$ = 4.5 y, range = 2.4–9.1<br>4 children had spastic quadriplegia,<br>1 child had triplegia,<br>14 children had spastic hemiplegia | 1 wk | NR | NR |
| | | Intrarater reliability Xu et al[52] | N = 10 | NR | Grip strength was measured in a standardized manner for all participants following the recommendations of Melvin and Barnes[61] | The authors suggested using the average of 3 trials to reduce the error factor that can occur with one reading |

*MMT = manual muscle testing, HHD = handheld dynamometer, GMFCS = Gross Motor Function Classification System, NR = not reported.*

[b] *Able to extend the wrist and fingers voluntarily but used the wrist predominantly in flexion during manual activities. The children could grasp and release objects with reduced quality and speed of movement; most could move their fingers in isolation and oppose one or more fingers. No child was receiving interventions for upper limb functions, showed deficits in passive wrist extension greater than 10 degrees, had undergone medical or surgical interventions for the upper limb, or had any associated pathologies.*

**Table 4.** Statistical Findings of Included Studies, Including Methodological Quality Scores

| Measurement Instrument or Method | Measurement Property | COSMIN Score | UE Total | Shoulder | Elbow | Wrist | Grip |
|---|---|---|---|---|---|---|---|
| MMT | Interrater reliability Klingels et al[47] | Fair | +<br>ICC = .90<br>95% CI = .80–.95 | —<br>ICC = .60<br>95% CI = .32–.79 | —<br>ICC = .62<br>95% CI = .34–.80 | +<br>ICC = .91<br>95% CI = .82–.96 | NA |
| | Test-retest reliability Klingels et al[47] | Poor | +<br>ICC = .96<br>95% CI = .91–.98 | +<br>ICC = .93<br>95% CI = .84–.96 | +<br>ICC = .88<br>95% CI = .74–.95 | +<br>ICC = .96<br>95% CI = .91–.98 | NA |
| Jamar dynamometer | Interrater reliability Klingels et al[47] | Fair | NA | NA | NA | NA | +<br>ICC = .95<br>95% CI = .89–.97 |
| | Test-retest reliability Klingels et al[47] | Poor | NA | NA | NA | NA | +<br>ICC = .96<br>95% CI = .90–.98 |
| HHD | Test-retest reliability | Poor | ? | NR | NR | NR | NA |
| | Crowner and Racette[48] Vaz et al[5] | Poor | .99<br>NR | NR | NR | +<br>ICC = .93–.98 | NA |
| Muscle strength–torque sensor | Test-retest reliability Bleyenheuft et al[49] | Poor | ?<br>No significant difference between measurements before and after testing (P = .935) | NR | NR | NR | NR |
| Strain gauge technology | Test-retest reliability Rameckers et al[50] | Poor | NA | NA | NA | NA | +<br>ICC = .99 |
| Modified sphygmomanometer | Test-retest reliability Glazier et al[51] | Poor | NA | NA | NA | NA | ?<br>r = .97 |
| | Intrarater reliability Xu et al[52] | Poor | NA | NA | NA | NA | +<br>ICC = .919 |

*MMT = manual muscle testing, ICC = intraclass correlation coefficient, 95% CI = 95% confidence interval, HHD = handheld dynamometer, NA = not applicable, NR = no measurement property reported, r = Pearson correlation coefficient, UE = upper extremity, + = ICC > .70 or n > .70, — = ICC ≤ .70 or n ≤ .70, ? = no justified statistical method*

**Table 5.** Levels of Evidence of Upper Extremity Strength Measurement Instruments

| Measurement Instrument or Method | Measurement Property | | |
|---|---|---|---|
| | Interrater Reliability | Test-Retest Reliability | Intrarater Reliability |
| MMT | + (upper extremity/wrist)<br>— (shoulder/elbow) | ? (upper extremity/shoulder/elbow/wrist) | 0 |
| Jamar dynamometer | + (grip) | ? (grip) | 0 |
| HHD | 0 | ? (upper extremity/wrist) | 0 |
| Muscle strength-torque sensor | 0 | ? (upper extremity) | 0 |
| Strain gauge technology | 0 | ? (index finger flexor strength) | 0 |
| Modified sphygmomanometer | 0 | ? (grip) | ? (grip) |

*MMT = manual muscle testing, HHD = handheld dynamometer, + = limited positive evidence, — = limited negative evidence, ? = unknown evidence, 0 = no evidence.*

# Discussion

The purpose of this systematic review was to study the clinimetric properties of upper extremity strength measurement instruments used for children with CP. This review clearly exposes the lack of adequate studies investigating clinimetric properties of upper extremity strength measurement instruments for children with CP.

In the few studies using measurement instruments of upper extremity strength, only test-retest, intrarater, and interrater reliability were investigated in a select group of age ranges, Manual Ability Classification system (MACS) levels,[54] and Gross Motor Function Classification System (GMFCS) levels.[55] No conclusions can be made regarding the possibility of determining changes over time (responsiveness), the smallest detectable change (SDC), or the standard error of measurement (SEM). Furthermore, it is not clear whether all of the measurement instruments specifically measure muscle strength, as validity has not been investigated. Therefore, more research on the other clinimetric properties must be done for all of the instruments before they are used in clinical practice or further studies.

Only 2 of the studies[47,51] were specifically designed to assess the clinimetric properties of the measurement instruments. All of the other studies were intervention studies, necessitating a reliability study of the outcome measurement. These findings may explain why only test- retest, interrater, and intrarater reliability are investigated in mostly small groups of children.

None of the measurement instruments were rated as "strong" or "moderate" for the level of evidence. According to the COSMIN standards, only the studies that reported on the interrater reliability of the MMT[47] and Jamar dynamometer[47] were rated as "fair" for methodological quality; therefore, the level of evidence was rated as "limited."

In MMT, interrater reliability of muscle strength measurements of the shoulder and elbow had poor statistical outcomes. Manual muscle testing, therefore, is not recommended for measuring muscle strength in these muscle groups. Only the total upper extremity MMT score and the score of the wrist muscles had good interrater reliability. Although MMT is commonly used in clinical practice, its use is dissuaded with other populations described in the literature,[56,57] despite the findings of

Klingels et al.[47] The studies by Noreau and Vachon[56] and Schwartz et al.[57] showed there is wide variability in grading values with MMT grades 4 and 5. Therefore, it is recommended that MMT be used in the positive-rated muscle groups (upper extremity total, wrist) in children with less muscle strength (≤ grade 3).

The Jamar dynamometer had good statistical outcomes and, therefore, is recommended for measuring grip strength in children with CP. The positive characteristics of the Jamar dynamometer are that it is a small device (handheld, lightweight [1.4 kg]) that is relatively inexpensive (retail price = $300) and easy to use. The negative characteristics of the Jamar dynamometer are that it can only be used to measure handgrip strength, and it cannot be used by children with very small hands. Moreover, the range (0–90 kg) and incremental 2-kg steps may not be suitable to measure minimal changes, especially for young or small children or for children with very poor muscle strength. Based on these results, the Jamar dynamometer appears to have good potential as a reliable and clinically useful instrument for measuring handgrip strength in children with CP. However, specific assessment of its clinimetric properties in children with CP is warranted.

According to the COSMIN standard, all of the other studies had poor methodological quality; therefore, the levels of evidence of all other studies were rated "unknown." The poor methodological quality is partly due to the fact that all of the studies used rather small sample sizes to investigate the clinimetric properties of the strength measurement instruments. Sample sizes varied between 2 and 30 people. The COSMIN manual[53] recommends a minimum sample size of 50, although a sample size of 100 would be better. Pooling data to achieve these sample sizes was not possible. Because of the unknown level of evidence, the outcomes of these clinimetric properties must be interpreted with extra care.

Although the levels of evidence for the other measurement instruments and methods (ie, HHD, muscle strength-torque sensors, strain gauge technology, and modified sphygmomanometer) were rated as "unknown," the clinimetric properties were rated as "good" or "indeterminate." Therefore, for some of these instruments, a sufficient level of evidence can be reached when the clinimetric properties are researched in studies of good or excellent methodological quality. In order to consider their clinical applicability, the positive and negative characteristics of these instruments will be described.

The positive characteristics of the MicroFET 2 dynamometer are that it is a small device (handheld, weighs less than 0.5 kg) that is relatively inexpensive (retail price = $1,095) and easy to use. Moreover, its ability to detect small changes might be good because of the small incremental steps of 1 N·m. The negative characteristics of the Microfet 2 dynamometer are that the assessor can have difficulty stabilizing the patient while using the device, the opposing strength of the examiner potentially contributes to the measured force, and inaccurate readings can be made when the force is not applied in a precise, perpendicular direction.[58] In addition, different protocols are used worldwide, and various articles have already identified the need for further research and development of standardized handheld dynamometry procedures in children with CP.[23–25] Studies researching the reliability of handheld dynamometry of the lower extremities in children with CP showed intrarater reliability (ICC) scores between .38 and .96 for the lower leg muscles in a sample of 10 to 25 children with CP.[21,23–27] Interrater reliability of handheld dynamometry of the lower extremities in children with CP varies between .39 and .94, depending on the muscle group and method of measuring.[21,23] The results of these studies are similar to those found in the studies on the upper extremities: low sample sizes and

mild to good reliability. When combined, these findings indicate that the MicroFET 2 dynamometer has potential as a reliable instrument for measuring upper limb muscle strength in children with CP. Future research should focus on all clinimetric properties of the HHD with regard to measuring the strength of the upper extremities of children with CP.

The positive characteristics of muscle strength-torque sensors and strain gauge technology are that the outcomes are computerized and show small incremental steps (because high-quality strain levers were used). Therefore, they can be very accurate. In addition, errors caused by inaccurate reading of the display by the examiner can be pre- vented by storing the outcome digitally, which can improve the reliability. Because of the small increments shown on the display, the outcomes of the modified sphygmomanometer are very accurate. A disadvantage of these instruments is that they are specially designed for research purposes. These instruments are not commercially or broadly available and, therefore, are more difficult to implement in daily clinical practice.

## Limitations

The COSMIN method is a strict method with stringent rules, and it sets high standards for methodological design of clinimetric studies and reporting. The COSMIN standards were originally developed for evaluating questionnaire-based measurement instruments. One of the stringent rules is a minimum of 50 included samples in the study to achieve good methodological quality. For most studies that focus on clinimetric properties of questionnaire-based measurement tools, it is easier to adhere to this standard compared with studies that focus on clinimetric properties of performance-based measurement tools. Therefore, it is possible that the COSMIN standards have limitations when evaluating measurement tools that are performance-based.

In most studies included in the present review, the lack of information on their design and other important items of the COSMIN checklist is highly remarkable. Information about the COSMIN items of "missing data" (the percentage of missing data), "how missing items were handled," and "independent administrations" (assessors blinded) often was absent. Also, the standard of the included sample size in the analysis often was not adequate. Because of this missing information and the small sample sizes, subitems automatically were given a low score in the COSMIN box. Furthermore, because of the limited number of studies that described clinimetric properties, it was not possible to compare studies and provide an overall conclusion of the best measurement instrument.

## Conclusion and Recommendations

Although several instruments for measuring upper extremity strength in children with CP are available, research on the clinimetric properties of these instruments is rarely done. To measure grip strength, it is recommended to use the Jamar dynamometer. For measuring other upper extremity muscle groups, it is recommended to use the HHD. Manual muscle testing can be used when measuring total upper extremity or wrist strength in children with CP who have very limited muscle strength (below grade 4). However, caution with interpretation of the test results is warranted because no information is available regarding the possibility of determining changes over time (responsiveness), the SDC, the SEM, and the validity of these instruments. Future studies should be

designed according to the COSMIN criteria; should go beyond interrater, intrarater, and test-retest reliability; and should be performed on children with CP from different age groups and all MACS levels,[54] according to a well-described protocol.

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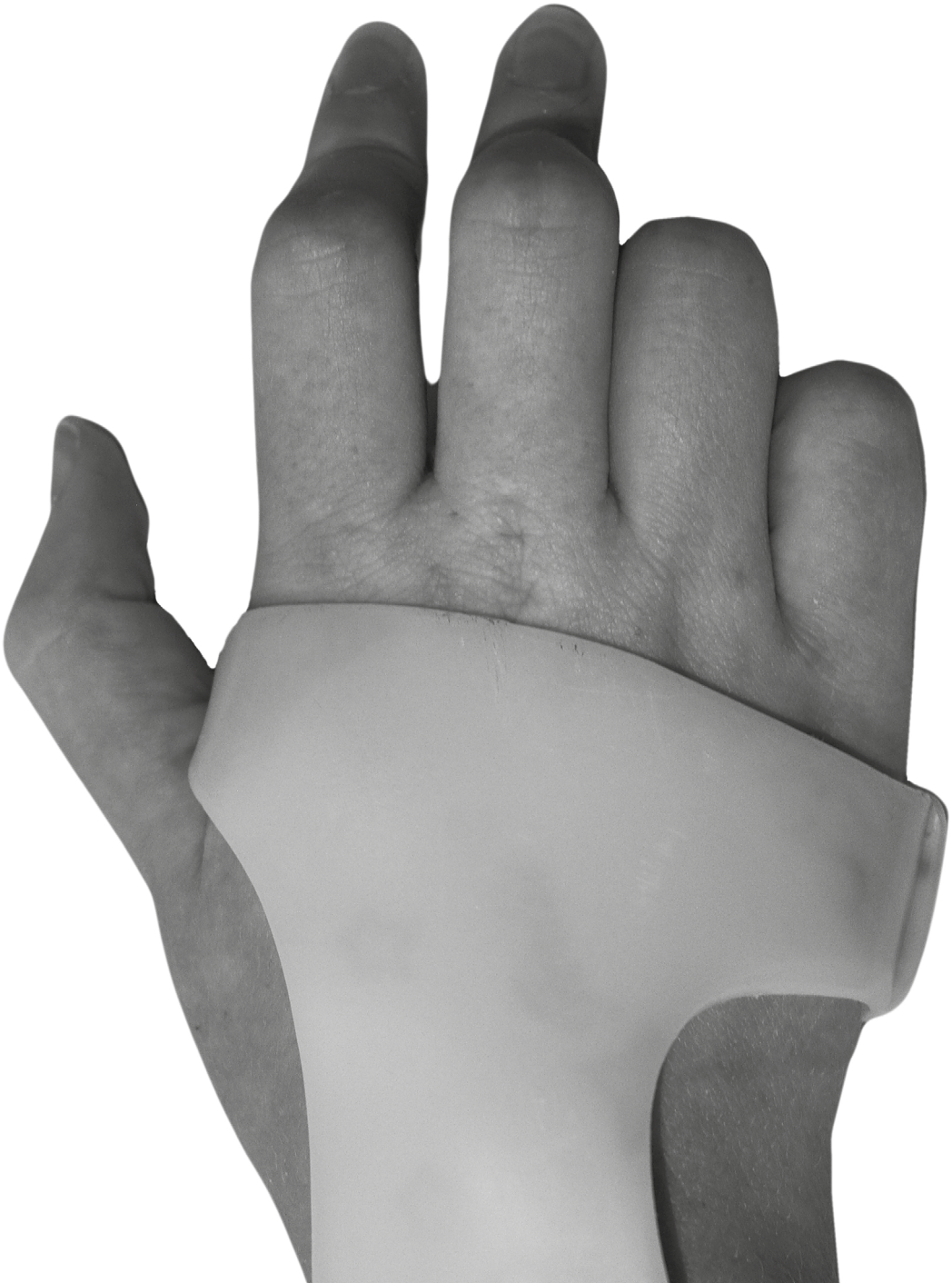
**Appendix.** Example of COSMIN (COnsensus-based Standards for the selection of health status Measurement INstruments) Measurement Property Box [57]

**Box B. Reliability: relative measures (including test-retest, interrater, and intrarater reliability)**

| | | Excellent | Good | Fair | Poor |
|---|---|---|---|---|---|
| **Design requirements** | | | | | |
| 1 | Was the percentage of missing items given? | Percentage of missing items described | Percentage of missing items not described | | |
| 2 | Was there a description of how missing items were handled? | Described how missing items were handled | Not described but how missing items were handled can be deduced | Not clear how missing items were handled | |
| 3 | Was the sample size included in the analysis adequate? | Adequate sample size (≥100) | Good sample size (50–99) | Moderate sample size (30–49) | Small sample size (< 30) |
| 4 | Were at least 2 measurements available? | At least 2 measurements | | | Only 1 measurement |
| 5 | Were the administrations independent? | Independent measurements | Assumable that the measurements were independent | Doubtful whether the measurements were independent | Measurements not independent |
| 6 | Was the time interval stated? | Time interval stated | | Time interval not stated | |
| 7 | Were patients stable in the interim period on the construct to be measured? | Patients were stable (evidence provided) | Assumable that patients were stable | Unclear whether patients were stable | Patients were not stable |
| 8 | Was the time interval appropriate? | Time interval appropriate | | Doubtful if time interval was appropriate | Time interval not appropriate |
| 9 | Were the test conditions (eg, type of administration, environment, instructions) similar for both measurements? | Test conditions were similar (evidence provided) | Assumable that test conditions were similar | Unclear if test conditions were similar | Test conditions were not similar |
| 10 | Were there any important flaws in the design or methods of the study? | No other important methodological flaws in the design or execution of the study | | Other minor methodological flaws in the design or execution of the study | Other important methodological flaws in the design or execution of the study |
| **Statistical methods** | | | | | |
| 11 | For continuous scores: Was an intraclass correlation coefficient (ICC) calculated? | ICC calculated and model or formula of the ICC is described | ICC calculated but model or formula of the ICC not described or not optimal. Pearson or Spearman correlation coefficient calculated with evidence provided that no systematic change has occurred. | Pearson or Spearman correlation coefficient calculated without evidence provided that no systematic change has occurred or with evidence that systematic change has occurred. | No ICC or Pearson or Spearman correlations calculated |
| 12 | For dichotomous/ nominal/ ordinal scores: Was kappa calculated? | Kappa calculated | | | Only percentage of agreement calculated |
| 13 | For ordinal scores: Was a weighted kappa calculated? | Weighted kappa calculated | | Unweighted kappa calculated | Only percentage agreement calculated |
| 14 | For ordinal scores: Was the weighting scheme (eg, linear, quadratic) described? | Weighting scheme described | Weighting scheme not described | | |



# Chapter 3

# Reliability of maximum isometric arm, grip and pinch strength measurements in children (7–12 years) with unilateral spastic cerebral palsy

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# Abstract

**Purpose:** To investigate test–retest and inter-rater reliability of maximum isometric arm muscle strength measurements using the hand-held dynamometer (HDD) and maximum isometric grip and pinch strength measurements using the Biometrics E-Link Evaluation System in children aged 7–12 years with unilateral spastic cerebral palsy.

**Materials and methods:** All data were obtained using a test–retest study design. The study met the conditions of the COSMIN criteria to achieve good methodological quality.

**Results**: For arm strength measurements, all test–retest reliability intraclass correlation coefficient (ICC) values and all but one inter-rater reliability ICC value indicated excellent reliability. For grip- and pinch strength measurements, all test–retest reliability and inter-rater reliability ICC values showed excellent reliability. The standard error of measurement values ranged from 4.97 to 11.36 N (HDD) and 0.37 to 1.81 kg (E-link). Smallest detectable change values ranged from 13.79 to 31.49 N (HDD) and 1.03 to 5.02 kg (E-link).

**Conclusions:** The HDD and E-link system are usable measurement instruments for cross-sectional muscle strength measurements in children with unilateral spastic cerebral palsy. It is not clear if both instruments are usable to measure changes in muscle strength within an individual, especially if a child with unilateral spastic cerebral palsy has low muscle strength. Caution in the interpretation of changes in muscle strength is therefore necessary.


*IMPLICATIONS FOR REHABILITATION*

- The hand-held dynamometer and E-Link Evaluation System are reliable measurement instruments to measure muscle strength of the arm and hand in children with unilateral spastic cerebral palsy, aged 7–12 years.
- Cross-sectional measurements; it is possible to measure upper extremity muscle strength in children with unilateral spastic cerebral palsy with the hand-held dynamometer and E-link system.
- Longitudinal measurements; changes in upper extremity muscle strength within one person should be interpreted with care, especially if a child with unilateral spastic cerebral palsy has low muscle strength.

# Introduction

Abnormal gross and fine motor functioning and organization (reflecting abnormal motor control) are the core features of unilateral spastic cerebral palsy (USCP).[1] In children with USCP, the muscles of the affected upper extremity (UE) are typically weaker than those of the contralateral UE and the strength of the UE of typically developing peers.[2–4] Muscle weakness of the UE may lead to limitations in daily activities, as grip strength has shown to be an important predictor of use of the affected arm in bimanual performance in children with USCP.[5,6]

Muscle strength in children with USCP could be measured for different reasons. Sometimes one needs to know if muscle strength could contribute to problems carrying out activities of daily living. Another reason could be if a therapist wants to know if muscle strength is gained after a strength training program. Strength training programs are recommended starting at 7 years of age.[7] Intensive strength training programs are often delivered to children in elementary school.

Several methods are available to measure muscle strength: dynamic (concentric, eccentric, isotonic, and isokinetic) and static (isometric). Dynamic muscle strength measurements may be influenced by spasticity as well as by limitations in the range of motion of the UE. In static/isometric muscle strength measurements, the ability of a muscle group to produce force without a change in overall muscle-tendon length is measured; therefore, limitations by spasticity and range of motion will not interfer with task performance during this measurement. Furthermore, in most daily activities the affected UE is used as an assisting hand (e.g., holding, stabilizing, carrying), and in these tasks a high percentage of maximum isometric muscle strength is needed.

(Isometric) muscle strength must be measured accurately; therefore, the instrument should have sound clinometric properties. A recent systematic review on the clinometric properties of measurement instruments for measuring UE strength in children with USCP concluded that research on clinometric properties is rarely conducted, and caution is needed regarding interpretation of the test results. Only reliability has been studied, and most of the available studies were of poor methodological quality.[8] Future studies should be designed according to the Consensus- based Standards for the selection of health Measurement INstruments (COSMIN) criteria and should use a well-described protocol. The hand dynamometer is recommended for measure- ment of grip strength. For measuring other UE muscle groups, it was recommended that the hand-held dynamometer (HDD) be used.[8] Reliability is an important clinometric property. One needs to know the degree to which variations in measurement appear when no changes in the disease or disorder have occurred. This so-called measurement error can arise from several sources: the measurement instrument itself, the person(s) assessing the measurement, the patient undergoing the measurement and the circumstances under which the measurement is performed.[9] Two important components of reliability are the standard error of the measurement (SEM) and smallest detectable change (SDC).[9] The SEM is a measure of how far apart the outcomes of repeated measurements are; it is the standard deviation (SD) around a single measurement.[9] The SDC is the smallest change in score that you can detect with the instrument, above measurement error in individual patients.[9] To date, no data on SEM and SDC of the hand dynamometer and HHD in children with USCP are available in literature.

The purpose of this paper is to investigate the test–retest and inter-rater reliability (including SEM and SDC) of maximum isometric arm strength (IAS) measurements using the HHD and maximum isometric grip and pinch strength (IGPS) measurements using the Biometrics E-Link Evaluation System (digitalized hand- dynamometer), in children with USCP in a study of good methodological quality according to the COSMIN criteria.[10]

# Materials and methods

## Study design

All data were obtained using a test–retest study design. Data were collected in the Netherlands and USA, from 2009 to 2016.



## Participants

Permission was granted by the Medical Ethical Board of the Maastricht University Medical Center and Maastricht University (METC azM/UM) in the Netherlands and at Teachers College, Columbia University in New York City, USA. In the Netherlands, the children were recruited from four different rehabilitation centres and related schools for special education, i.e., Adelante Rehabilitation Centre, Valkenburg, Libra Rehabilitation and Audiology, Tilburg, Revant Rehabilitation Centers, Breda and Goes, and Tolbrug Rehabilitation Centre, Den Bosch. In the USA, participants were a convenience sample of children participating in ongoing intensive UE studies at Teachers College, Columbia University.

This study focused on children with predominantly USCP between 7 and 12 years of age. The diagnosis USCP was based on the classification used by the child's neurologist or paediatrician. They were classified as Gross Motor Function Classification System (GMFCS)[11] I–II and Manual Ability Classification System (MACS)[12] levels I, II, or III. All participants were capable of following simple instructions. A child was excluded when he/she had undergone surgery or Botulinum Toxin-A treatment in the UE in the past 6 months. A child was excluded from the test–retest reliability measurements if he/she was participating in an intensive UE training program between the two measurements. There was no minimum muscle strength required to participate.

## Procedure

Children were tested at the location from where they were recruited. A standardized protocol, with detailed descriptions of all procedures and measurements, was used (see Supplementary material). Prior to testing, body weight, MACS[12] and GMFCS[11] level were determined.

In USCP it is stated that increased muscle tone and weakness are most pronounced in distal muscle groups.[6] Accordingly, we decided to only test elbow, wrist and hand (grip/pinch) strength. In each child, a pre-randomized mix of measurements, using the random.org app (mobile application software)[13]

was used. Each measurement was performed three consecutive times. Between each measurement, the child had at least 30 seconds of rest to allow for muscle recovery. In each test, both the affected hand (AH) and the non-affected hand (NAH) were measured. The NAH was tested before the AH.

For each measurement, the child was seated in an upright position in a chair with back support and armrests. For all measurements, the armrests of the chair were used to support the arms during testing. The initial posture was neutral position (0°) of the wrist joint and 90° flexion of the elbow joint.

With the HHD, isometric wrist extension with stretched fingers, wrist extension with flexed fingers, wrist flexion with stretched fingers and elbow flexion/extension were measured. With the E-Link Evaluation System, IGPS were measured. The handle position of the E-link handgrip was adapted to the child's hand size, according to the E-link guideline for positioning. The child was also asked whether the position of the handle felt the best. When there was doubt, other handle positions were tried.

Test scores were read by the therapist and registered by the same therapist on a test form. For the E-Link Evaluation System, test scores were also stored on the E-Link Evaluation System computer. Children were verbally encouraged by the therapist to produce maximum force, by saying "hard, harder, hardest" in a time span of 4–5 seconds.

To evaluate test–retest reliability, the standardized protocol was conducted two times by the same therapist within 2–4 weeks. This time interval was chosen because during normal development (without intensive UE training) no muscle strength loss or gain was expected, and the possible motivation/influence of the therapist/child to score the same result as during the first measurement (because the first result could be remembered by the child/therapist) was minimal. For the second time, test conditions were kept identical.

To evaluate inter-rater reliability, the standardized protocol was conducted two times on the same day, by two different therapists. There was approximately 30 minutes of rest between each assessment. This rest period was judged sufficient for the child to recover and limited the possibility for personal and environmental factors to change.

All measurements were performed by eight (paediatric) physical therapists, who had no direct professional connection with the participants. All the therapists performed the measurements in the Netherlands. Two of them also performed the measure- ments in the USA. They had four hours of training by an experienced paediatric physical therapist on how to use the standardized protocol in children with USCP.

## Measures

### *Isometric arm strength*

Maximum IAS was measured with the Microfet 2 HHD (Hoggan Scientific, LLC, Salt Lake City, UT). An HHD is an electronic device that fits in the palm of a hand. A load cell (strain gauge technology) measures the isometric muscle strength applied to a transducer. The "make method", in which the child applies force against a fixed HHD, was used.[14] The applied force was measured in Newtons.

### *Isometric grip and pinch strength*

Maximum IGPS of both AH and NAH were measured with the Biometric E-Link Evaluation System (Biometrics Ltd, Gwent, UK). As lateral pinch/key pinch is the easiest in children with USCP, this pinch position was chosen. The E-Link Evaluation System is a calibrated, computerized system incorporating a modified (digitalized) grip dynamometer and a pinch meter. The applied force was measured in 0.1 kg.

## Statistical analysis

For each test, the mean of three measurements was calculated. In this way, variability in muscle strength due to variations in placing the measurement instrument on a prescribed measurement spot (and therefore a smaller or larger torque arm) was minimized. Intraclass correlation coefficients (ICC), model two way random, type absolute agreement, with a 95% confidence interval (CI) were used to assess test–retest reliability and inter-rater reliability. An ICC > 0.80 reflects excellent reliability, while ICCs from 0.70 to 0.79 reflect good reliability.[15]

The SEM agreement was calculated as the square root of the error variance (including the systematic error).[9] The SDC was computed as 1.96 multiplied by the square of 2, multiplied by the SEM (SDC = 1.96 x √2 x SEM).[9]

# Results

A total of 86 children (53 boys, 33 girls; mean age 9 years, 3 months, SD 1 year 8 months) with USCP participated in this study. Their parents (and children aged 12 years) provided informed consent for participation. Due to the availability of the child and/ or measurement instrument or because the child met the exclusion criterion for the test–retest reliability study, some children only performed the measurements of the test–retest reliability and some only the measurements of the inter-rater reliability. Therefore, the sample sizes vary across the measurements. Participant characteristics per measurement are provided in Table 1.

## Test–retest reliability

For the IAS, 52 children performed all measurements. For the IGPS, the total number of participants was 65. There were no missing items. The test–retest reliability statistics of the IAS and IGPS are presented in Table 2.

### *Affected hand*

Test–retest ICC values for the IAS measurements varied between 0.887 (CI 0.799–0.936) for the elbow extension and 0.964 (CI 0.938–0.979) for the wrist extension. ICC values for the IGPS measurements were 0.940 (CI 0.896–0.965) for pinch strength and 0.948 (CI 0.914–0.968) for grip strength. For the IAS, the SDC was 13.79 N for wrist extension and 31.49 N for elbow extension. For the IGPS, the SDC was 1.03 kg for pinch strength and 3.47 kg for grip strength.

## *Non-affected hand*

ICC values for the IAS measurements varied between 0.888 (CI 0.806–0.936) for wrist extension with flexed fingers and 0.973 (CI 0.952–0.984) for elbow extension. For the IGPS measurements, ICC values were 0.937 (CI 0.895–0.962) for pinch strength and 0.942 (CI 0.904–0.964) for grip strength. The SDC for the IAS was 24.54 N for wrist extension with stretched fingers and 30.89 N for wrist flexion. For the IGPS, the SDC was 1.41 kg for pinch strength and 5.02 kg for grip strength.

**Table 1.** Participants characteristics.

| **Measurement** | **Characteristics** | |
|---|---|---|
| Test-retest reliability study | | |
| Total group | Age group | Gender mix |
| Isometric Arm Strength measurements (HHD)<br>n = 52. 33♂. 19♀.<br>Mean age 9.3 years. SD 1.9 years.<br>35 right side affected. 17 left side affected<br>MACS level; I: n = 16. II: n = 28. III: n = 4.<br>missing n = 4 | Age 7. n = 13 | 6♂. 7♀ |
| | Age 8. n = 10 | 4♂. 6♀ |
| | Age 9. n = 7 | 5♂. 2♀ |
| | Age 10. n = 5 | 4♂. 1♀ |
| | Age 11. n = 6 | 5♂. 1♀ |
| | Age 12. n = 11 | 9♂. 2♀ |
| Isometric Grip and pinch strength measurements (E-link system)<br>n = 65. 41♂. 24♀.<br>Mean age 9.2 years. SD 1.8 years.<br>40 right side affected. 25 left side affected<br>MACS level; I: n = 21. II: n = 33. III: n = 7.<br>missing n = 4 | Age 7. n = 15; | 7♂. 8♀ |
| | Age 8. n = 12 | 5♂. 7♀ |
| | Age 9. n = 12 | 7♂. 5♀ |
| | Age 10. n = 5 | 4♂. 1♀ |
| | Age 11. n = 10 | 9♂. 1♀ |
| | Age 12. n = 11 | 9♂. 2♀ |
| Inter-rater reliability study | | |
| Total group | Age group | Gender mix |
| Isometric Arm Strength measurements (HHD)<br>n = 53. 31♂. 22 ♀.<br>Mean age 9.0 years. SD 1.7 years.<br>31 right side affected. 22 left side affected<br>MACS level; I: n = 15. II: n = 29. III: n = 4.<br>missing n = 5 | Age 7. n = 14 | 8♂. 6♀ |
| | Age 8. n = 11 | 4♂. 7♀ |
| | Age 9. n = 9 | 7♂. 2♀ |
| | Age 10. n = 8 | 5♂. 3♀ |
| | Age 11. n = 3 | 2♂. 1♀ |
| | Age 12. n = 8 | 5♂. 3♀ |
| Isometric Grip and Pinch Strength measurements (E-link system)<br>n = 54. 31♂. 23 ♀.<br>Mean age 9.0 years. SD 1.7 years<br>31 right side affected. 22 left side affected<br>MACS level; I: n = 15. II: n = 29. III: n = 5.<br>missing n = 5 | Age 7. n = 14 | 8♂. 6♀ |
| | Age 8. n = 11 | 4♂. 7♀ |
| | Age 9. n = 10 | 7♂. 3♀ |
| | Age 10. n = 8 | 5♂. 3♀ |
| | Age 11. n = 3 | 2♂. 1♀ |
| | Age 12. n = 8 | 5♂. 3♀ |

*Abbreviations: HHD = hand held dynamometer; ♂ = male; ♀ = female; SD = standard deviation; n = population size; MACS = Manual Ability Classification System*

**Table 2.** Test–retest reliability of isometric arm strength measurements and isometric grip and pinch strength measurements in children (7–12 years) with unilateral spastic cerebral palsy.

| Movement | n | Mean T0 (SD) | Mean T1 (SD) | Diff T0–T1 | ICC | 95% | CI | SEM | SDC |
|---|---|---|---|---|---|---|---|---|---|
| Isometric arm strength measurements (HHD) | | | | | | | | | |
| Wrist-extension AH | 52 | 28.6 N (19.7 N) | 29.7 N (18.1 N) | 1.1 N | 0.964 | 0.938 | 0.979 | 4.97 N | 13.79 N |
| Wrist-extension (flexion fingers) AH | 52 | 32.1 N (20.5 N) | 34.0 N (19.6 N) | 1.9 N | 0.948 | 0.909 | 0.970 | 6.32 N | 17.51 N |
| Wrist-flexion AH | 52 | 34.4 N (15.2 N) | 36.9 N (15.9 N) | 2.5 N | 0.894 | 0.814 | 0.940 | 6.84 N | 18.96 N |
| Elbow-flexion AH | 52 | 73.3 N (30.0 N) | 78.8 N (30.2 N) | 5.5 N | 0.930 | 0.868 | 0.962 | 10.98 N | 30.45 N |
| Elbow-extension AH | 52 | 65.5 N (25.8 N) | 70.3 N (24.3 N) | 4.8 N | 0.887 | 0.799 | 0.936 | 11.36 N | 31.49 N |
| Wrist-extension NAH | 52 | 57.7 N (20.4 N) | 61.2 N (19.5 N) | 3.5 N | 0.905 | 0.830 | 0.946 | 8.38 N | 23.22 N |
| Wrist-extension (flexion fingers) NAH | 52 | 64.0 N (26.2 N) | 66.8 N (23.3 N) | 2.8 N | 0.888 | 0.806 | 0.936 | 11.14 N | 30.89 N |
| Wrist-flexion NAH | 52 | 68.2 N (24.8 N) | 64.5 N (24.6 N) | 3.7 N | 0.932 | 0.878 | 0.962 | 8.85 N | 24.54 N |
| Elbow-flexion NAH | 52 | 104.4 N (40.1 N) | 108.2 N (38.3 N) | 3.8 N | 0.958 | 0.927 | 0.976 | 11.12 N | 30.81 N |
| Elbow-extension NAH | 52 | 94.8 N (41.1 N) | 97.8 N (40.1 N) | 3.0 N | 0.973 | 0.952 | 0.984 | 9.36 N | 25.93 N |
| Isometric grip and pinch strength (E-link system) | | | | | | | | | |
| Grip strength AH | 65 | 5.2 kg (4.4 kg) | 4.8 kg (3.5 kg) | 0.4 kg | 0.948 | 0.914 | 0.968 | 1.25 kg | 3.47 kg |
| Pinch strength AH | 65 | 1.7 kg (1.2 kg) | 1.9 kg (1.1 kg) | 0.2 kg | 0.940 | 0.896 | 0.965 | 0.37 kg | 1.03 kg |
| Grip strength NAH | 65 | 12.6 kg (5.6 kg) | 12.2 kg (5.3 kg) | 0.4 kg | 0.942 | 0.904 | 0.964 | 1.81 kg | 5.02 kg |
| Pinch strength NAH | 65 | 3.7 kg (1.4 kg) | 3.9 kg (1.5 kg) | 0.2 kg | 0.937 | 0.895 | 0.962 | 0.51 kg | 1.41 kg |

*AH: affected hand; CI: confidence interval; Diff: difference; HHD: hand-held dynamometer; ICC: interclass correlation coefficient; kg: kilograms; n: population size; N: Newton; NAH: non-affected hand; SEM: standard error of the measurement; SD: standard deviation; SDC: smallest detectable change; sig: level of significance.*

**Table 3.** Inter-rater reliability of isometric arm strength measurements and isometric grip and pinch strength measurements in children (7–12 years) with unilateral spastic cerebral palsy.

| Movement | n | Mean T0 (SD) | Mean T1 (SD) | Diff T0–T1 | ICC | 95% | CI | SEM | SDC |
|---|---|---|---|---|---|---|---|---|---|
| Isometric arm strength measurements (HHD) | | | | | | | | | |
| Wrist-extension AH | 53 | 25.4 N (19.5 N) | 27.9 N (19.6 N) | 2.5 N | 0.963 | 0.932 | 0.980 | 5.26 N | 14.59 N |
| Wrist-extension (flexion fingers) AH | 53 | 28.9 N (19.3 N) | 31.0 N (18.3 N) | 2.1 N | 0.919 | 0.860 | 0.953 | 7.31 N | 20.25 N |
| Wrist-flexion AH | 53 | 34.2 N (14.5 N) | 35.9 N (16.0 N) | 1.7 N | 0.840 | 0.724 | 0.908 | 8.03 N | 22.25 N |
| Elbow-flexion AH | 53 | 64.1 N (24.4 N) | 70.0 N (25.5 N) | 5.9 N | 0.882 | 0.784 | 0.934 | 11.62 N | 32.21 N |
| Elbow-extension AH | 53 | 58.7 N (20.4 N) | 60.8 N (19.6 N) | 2.1 N | 0.799 | 0.652 | 0.883 | 11.46 N | 31.76 N |
| Wrist-extension NAH | 53 | 50.2 N (19.9 N) | 53.3 N (20.6 N) | 3.1 N | 0.878 | 0.789 | 0.930 | 9.02 N | 25.01 N |
| Wrist-extension (flexion fingers) NAH | 53 | 53.3 N (24.5 N) | 55.8 N (23.1 N) | 2.5 N | 0.897 | 0.822 | 0.941 | 10.20 N | 28.26 N |
| Wrist-flexion NAH | 53 | 56.8 N (20.6 N) | 55.3 N (18.2 N) | 1.5 N | 0.886 | 0.803 | 0.934 | 8.59 N | 23.80 N |
| Elbow-flexion NAH | 53 | 90.4 N (27.2 N) | 92.8 N (30.1 N) | 2.4 N | 0.913 | 0.850 | 0.950 | 11.59 N | 32.13 N |
| Elbow-extension NAH | 53 | 82.2 N (25.8 N) | 82.8 N (26.2 N) | 0.6 N | 0.942 | 0.899 | 0.967 | 8.48 N | 23.49 N |
| Isometric grip and pinch strength (E-link system) | | | | | | | | | |
| Grip strength AH | 54 | 5.9 kg (5.3 kg) | 6.1 kg (5.1 kg) | 0.2 kg | 0.976 | 0.959 | 0.986 | 1.32 kg | 3.65 kg |
| Pinch strength AH | 54 | 1.9 kg (1.6 kg) | 2.0 kg (1.5 kg) | 0.1 kg | 0.964 | 0.938 | 0.979 | 0.43 kg | 1.19 kg |
| Grip strength NAH | 54 | 13.2 kg (7.3 kg) | 13.3 kg (7.3 kg) | 0.1 kg | 0.960 | 0.932 | 0.977 | 1.90 kg | 5.28 kg |
| Pinch strength NAH | 54 | 3.8 kg (2.1 kg) | 3.8 kg (2.1 kg) | 0 kg | 0.967 | 0.943 | 0.981 | 0.54 kg | 1.51 kg |

*AH: affected hand; CI: confidence interval; Diff: difference; HHD: hand-held dynamometer; ICC: interclass correlation coefficient; kg: kilograms; n: population size; N: Newton; NAH: non-affected hand; SEM: standard error of the measurement; SD: standard deviation; SDC: smallest detectable change; sig: level of significance.*

## Inter-rater reliability

For the IAS, 53 children performed all measurements. For the IGPS, the total number of participants was 54. The inter-rater reliability statistics of the IAS and IGPS are presented in Table 3. There were no missing items.

### *Affected hand*

Inter-rater ICC values for the IAS measurements varied between 0.799 (CI 0.652–0.883) for elbow extension and 0.963 (CI 0.932–0.980) for wrist extension. For the IGPS measurements, ICC values were 0.964 (CI 0.938–0.979) for pinch strength and 0.976 (CI 0.959–0.986) for grip strength. The SDC for the IAS was 14.59 N for wrist extension and 32.21 N for elbow flexion. The SDC for the IGPS was 1.19 kg for pinch strength and 3.65 kg for grip strength.



### *Non-affected hand*

ICC values for the IAS measurements varied between 0.878 (CI 0.789–0.930) for wrist extension and 0.942 (CI 0.899–0.967) for elbow extension. ICC values for the IGPS measurements were 0.960 (CI 0.932–0.977) for grip strength and 0.967 (CI 0.943–0.981) for pinch strength. The SDC for the IAS was 23.80 N for wrist flex- ion and 32.13 N for elbow flexion. For the IGPS, the SDC was 1.51 kg for pinch strength and 5.28 kg for grip strength.

# Discussion

The purpose of this study was to investigate the reliability of maximum isometric UE strength measurements in children aged between 7 and 12 years with USCP using the HHD, and maximum IGPS measurements using the Biometric E-Link Evaluation System, in a high-quality study designed according to the COSMIN criteria.[10]

For the IAS measurements in this study, all test–retest reliability ICC values and all inter-rater reliability ICC values, except elbow flexion of the AH, indicated excellent reliability, which is in line with those previously reported by Crowner and Racette[16] and Vaz et al.[4] However, assessment of the clinometric properties of the HDD was not the main goal of these studies. This could explain their small sample sizes and the limited description of the design of the reliability study. In both studies, information about GMFCS[11] and MACS[12] level was missing. The study of Vaz et al. was performed before the MACS levels were published. In the study of Crowner and Racette,[16] only two children were included for the reliability part of the study and in the study of Vaz et al., 11 children. Also, other information about COSMIN design requirements, such as the time interval, was not described. Moreover, for example, in the study of Vaz et al.[4] Important information about test conditions and the independence of measurements was not described.

For the IGPS measurements in this study, all test–retest reliability ICC values and all inter-rater reliability ICC values showed excellent reliability. No previous study has reported clinometric properties of the E-link Evaluation system in children with USCP. Nevertheless, the results are in line with

those reported in another study involving healthy participants (adults, 18–25 years).[17] Furthermore, the part of the E-link Evaluation system which was used for determining grip strength shows similarities with the Jamar dynamometer. The reliability of the Jamar dynamometer was reported in the study of Klingels et al., and both test–retest (ICC 0.96) and inter-rater (ICC 0.95) reliability were excellent. This study was rated moderate according to the COSMIN criteria [8]. In terms of appearance, size and function, both instruments are more or less the same. The differences between the Jamar and the E-link are that they have different manufacturers, and the Jamar has incremental steps of 0.45 kg (1 lb), whereas the E-link Evaluation system has incremental steps of 0.1 kg.

As the present study fulfils the COSMIN criteria for a study with good methodological quality, it can be concluded that almost all arm/hand strength measurements have excellent test–retest reliability and excellent inter-rater reliability in the group of children with USCP, aged 7–12 years. Only the inter-rater reliability of the elbow flexion of the AH was classified as "good", meaning there was more variability in the performance of this measurement. A possible explanation for this variability in the performance of this measurement could be the higher muscle strength values. With higher muscle strength values, it is more difficult for the therapist to check/control that there is no movement in the joint. Reviewing the above-mentioned results, it can be concluded that both methods can be used reliably for cross-sectional measurements, for example, as a screening instrument or to determine UE strength in children with USCP.

However, besides test–retest reliability and inter-rater reliability, the SEM and SDC-values are important components of reliability.[9] SEM and SDC values for these measurement instruments have not been reported before. With the SEM and SDC values, the usability in patients in clinical practice can be determined, certainly when these instruments are used to determine changes over time in individual patients. In order to correctly interpret changes, for example, after a strength training program, one needs to know how much improvement is necessary to be sure that this improvement is not due to error. So, the change needs at least to be larger than the SDC. Unfortunately, so far, no clear information is available on how much improvement a child with USCP can achieve after a strength-training program. However, in cases where the muscle strength at baseline of a child is already less than the SDC-value, it will be very unlikely that by any intervention one can achieve an improvement (i.e., more than double the strength) above the SDC threshold. So, the utility of both instruments for measuring changes in muscle strength can there- fore be a problem in children with USCP with low muscle strength (below the SDC-value). Therefore, currently it is not possible to draw firm conclusions about the usability of both measurement instruments to measure changes over time. Additional research on the effects of strengthening and other interventions of the upper extremities in children with USCP is recommended to draw firm conclusions about the usability of the instruments in clinical practice, especially in children with USCP with low muscle strength.

## Limitations

Age, gender and MACS level were not ideally distributed, and therefore, some of these variables could have influenced the results. As this study population has an age range of 7–12 years, caution is advised when testing children of different age groups. The population size was too small to calculate separate ICC/SEM/ SDC values for each MACS level.[12] Caution is advised when testing children with USCP with

MACS-level III, because of the small population of included children. Unfortunately, group size was also too small to determine the specific characteristics of children with a muscle strength below the SDC value. Another limitation of the study could be the high number of measurement therapists, as this could have resulted in higher measurement errors. However, the high number of therapists involved in the measurements resembles clinical practice.

## Conclusions

The HDD and E-link system are usable measurement instruments in cross-sectional measurements of UE muscle strength in children with USCP. It is not clear if both instruments are usable for measuring changes in UE muscle strength within one person, especially if a child with USCP has low muscle strength. Therefore, caution in the interpretation of changes in UE muscle strength is necessary. More research on the effects of strengthening interventions of the upper extremities in children with USCP is recommended. Once the effects of strengthening interventions of the upper extremities in children with USCP have been examined, the usability of all instruments for longitudinal measurements will require reconsideration.



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No potential conflict of interest was reported by the authors

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# Chapter 4

# Psychometric Evaluation of 2 New Upper Extremity Functional Strength Tests in Children with Cerebral Palsy

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Dekkers, K. J., Smeets, R. J., Janssen-Potten, Y. J., Gordon, A. M., Speth, L. A., & Rameckers, E. A. (2019). Psychometric Evaluation of 2 New Upper Extremity Functional Strength Tests in Children With Cerebral Palsy. *Physical therapy, 99*(8), 1107–1115.

# Abstract

**Background.** For children with unilateral spastic cerebral palsy (USCP), reduced muscle strength can lead to activity limitations. However, none of the existing measures of upper extremity strength measure strength in the context of functional activities in which strength must be maintained for several seconds.

**Objective.** The objective of this study was to evaluate the psychometric properties of 2 newly developed functional hand and upper extremity muscle-strength tests (Cup-Task and Box-Task) in children aged 7 to 12 years with USCP.

**Design.** A longitudinal study design was used.

**Methods.** A standardized protocol with detailed descriptions of all procedures and measurements was used to determine test-retest reliability, interrater reliability, and criterion validity.

**Results.** A total of 86 children (53 males, 33 females, mean age = 9.3 years) with USCP participated in this study, with a subset performing each measurement. Only the results of children who were able to perform the measurement were included for analysis. Excellent test-retest reliability (intraclass correlation coefficients = 0.887–0.944; 95% confidence intervals = 0.713–0.969) and interrater reliability (intraclass correlation coefficients = 0.896–0.960; 95% confidence intervals = 0.813–0.980) were observed. The Cup-Task Affected-Hand and Box-Task were moderately correlated with maximum isometric grip strength. The Cup-Task Nonaffected-Hand had a low correlation with maximum isometric grip strength.

**Limitations.** Age, sex, and manual ability were not normally distributed, which could have influenced the results.

**Conclusions.** For children with USCP who can perform the tasks, the Cup-Task and BoxTask are reliable and valid instruments for measuring functional upper extremity muscle strength.

# Introduction

Impaired performance of upper extremity activities is reported in ~50% of children with unilateral spastic cerebral palsy (USCP), and these impairments affect the children's independence and quality of life.[1,2] Brændvik et al concluded that muscle strength strongly correlates with activity and that reduced strength can result in activity limitations.[3] Upper extremity activities are assessed using a number of clinical tests and questionnaires. The National Institutes of Health Common Data Elements recommends a variety of supplemental tests for assessing upper extremity motor function for children with cerebral palsy.[4] These tests include the ABILHAND-kids,[5] the Assisting

Hand Assessment,[6] the Melbourne Assessment-v2,[7] the Quality of Upper Extremity Skills Test,[8] and the Shriners Hospital Upper Extremity Evaluation.[9] Recommended exploratory measures of dexterity include the Box and Blocks Test of Manual Dexterity,[10] the Jebsen-Taylor Test of Hand Function,[11] the Nine-Hole Peg Test,[12] and the Purdue Pegboard.[13] All of these instruments measure function (active and passive range of motion, tone, segmental alignment of the extremity) or quantify the capacity (upper extremity use in a standardized, controlled environment), capability (upper extremity function in the daily environment), or performance (actual use of the upper extremity in the daily environment) of the upper extremity. However, none of these measures directly consider the role of strength in upper extremity activities.

For upper extremity strength measurements, the National Institutes of Health Common Data Elements recommends maximum voluntary isometric contraction testing using hand-held dynamometers and a grip/pinch dynamometer, and manual muscle testing using the Medical Research Council Muscle Grading Scale[14] to determine different grades of muscle strength. Within manual muscle testing, grades 4 and 5 seem insufficiently sensitive to assess muscle strength or to detect small to moderate increases of strength.[15]

In most manual activities, not only is a certain amount of muscle strength required, but also the ability to maintain that strength for a short time (2–5 seconds), for example, during carrying/moving a heavy box. Therefore, a certain level of coordination is also necessary to perform the tests adequately. Measuring muscle strength during a functional task in which it is expected that muscle strength plays a major role (ie, "functional strength" measurements) enables measurement not only of the strength of different simultaneously working muscles but also the task-specific generation of the strength.

For children with USCP, various *functional* muscle-strength tests for the lower extremity are available, including the "Lateral Step-up," "Sit-to-Stand," and the "Attain stand through half kneel, without using arms" tests.[16] For the upper extremities, the "functional strength measurement" test is feasible in children with CP.[17] With this test, maximum explosive muscle strength and 30-second repetitive measurements are recorded. Unfortunately, the ability to maintain the functional strength in a sustained contraction is not measured with the currently available muscle-strength tests. Thus, there are no functional upper extremity strength measures that quantify strength when sustained contractions are required. Therefore, we have developed 2 specific functional muscle-strength tests in the context

of unimanual and bimanual activity: the "Cup-Task" for determining maximal functional unimanual upper extremity strength, and the "Box-Task" for determining maximal functional bimanual upper extremity strength. Both tests measure a combination of functional grip and arm strength which must be sustained for 5 seconds. In a pilot study, both tests were found to be feasible in children with USCP.[18]

We used the COSMIN (COnsensus-based Standards for the Selection of health Measurement INstruments) checklist (www.cosmin.nl) as guidance for designing and reporting our study on the clinimetric properties of this new instrument. The COSMIN checklist is a consensus-based checklist that can be used for selecting a measurement instrument, peer reviewing a manuscript, and designing or reporting a study on measurement properties.[19] With the checklist, the methodological quality of a study can be classified as poor, moderate, good, or excellent.[20]

The objective of our study was to investigate test-retest and interrater reliability of the Cup-Task and Box-Task for children aged 7 to 12 years with USCP. A secondary objective was to assess the criterion validity of the Cup-Task and Box-Task.

# Methods

## Study Design

A longitudinal study design was used. Ethics approval was granted by the Medical Ethical Board of the Maastricht University Medical Center and Maastricht University (METC azM/UM, trial number NL45430.068.1) in the Netherlands and at Teachers College, Columbia University, New York City, New York (USA). Data were collected in the Netherlands and United States from 2009 to 2016.

## Participants

In the Netherlands, children were recruited from 4 rehabilitation centers and related special education schools, namely the Adelante Rehabilitation Centre in Valkenburg, Libra Rehabilitation and Audiology in Tilburg,

Revant Rehabilitation Centers in Breda and Goes, and Tolbrug Rehabilitation Centre in Den Bosch. In the United States, children were recruited while they were participating in ongoing intensive upper extremity studies at Teachers College, Columbia University.

To be included in this study, children were required to be diagnosed with USCP and aged between 7 and 12 years.

Furthermore, children had to be classified as GMFCS level I to II (Gross Motor Function Classification System[21]), MACS-level I, II, or III (Manual Ability Classification System[2]), and be able to follow simple instructions. Children were excluded if within the past 6 months they had undergone surgery or received botulinum toxin-A treatment of the upper extremity. Children were also excluded for the test-retest reliability measurements if they were participating in an intensive upper extremity training program between the 2 measurements. The aim was to include more than 50 children per measurement.[20]

## Procedure

Measurements took place at the rehabilitation center where the children were being treated. A standardized protocol with detailed descriptions of all procedures and measurements was used. Prior to testing, the participants' body weight and MACS and GMFCS levels were determined.

One set of strength measurements consisted of the Cup-Task, the Box-Task, and the E-LINK system. The sequence of the tasks was randomized. For the Cup-Task and E-LINK system, both the nonaffected hand (NAH) and the affected hand (AH) were measured.

To evaluate test-retest reliability, the set of strength measurements was conducted twice by the same assessor with a 2-week interval. This time interval was chosen because during normal development (without intensive upper extremity training), no loss or gain in muscle strength was expected and memory of the first results would be limited. For the second session, test conditions were kept identical. For some children, the results of the second set of strength measurements were used for the premeasurements of intensive upper extremity studies, and the time span was sometimes extended to 4 weeks for these children due to scheduling difficulties.

To evaluate interrater reliability, 2 sets of strength measurements were conducted by 2 different assessors on the same day. There was at least 30 minutes of rest between each assessment. This amount of rest time was judged sufficient for the child to recover and limited the possibility for personal and environmental factors to change. If a child was measured for test-retest reliability and interrater reliability, the results of the first interrater reliability measurements were used for the test-retest reliability.

To evaluate criterion validity, a maximum isometric grip strength (IGS) measurement was performed with the Biometrics E-LINK evaluation and exercise hand kit (Biometrics Ltd, Newport, Gwent, UK). The IGS measurement was performed 3 consecutive times, and the mean of the 3 measurements was calculated. Thus, variation in muscle strength, due to variations in grasping/handling of the measurement instrument, was minimized. Between each measurement, the child had at least 30 seconds of rest to allow recovery.

Test scores were determined by the assessor and registered by the same assessor on a test form. In case of IGS measurements, test scores were also stored on the E-LINK evaluation system computer.

All measurements were performed by 8 different assessors, all (pediatric) physical therapists, who had no direct professional connection with the participants. Two of the assessors (K.D. and E.R.) are experienced pediatric research physical therapists and were involved during the entire project. The other 6 assessors were physical therapists who are studying for their advanced degree in pediatric physical therapy. They were involved in the study for 6 consecutive months. Each assessor received 4 hours of training from K.D. or E.R. regarding the use of the standardized protocol in children with USCP. Before each measurement, each assessor had to read/practice the measurements protocol.

## Measures

Both tasks were developed based on an expert's opinion and the identification of the most frequently reported needs of the children with USCP in our Bimanual Intensive Movement Treatment. Goals involving lifting a box, tablet, or cup were most frequently reported by the children.

## *Cup-Task: unimanual functional muscle-strength test.*

The goal of the Cup-Task was to test maximum unilateral functional muscle strength by determining the weight (in grams) the child could lift and hold for 5 seconds with 1 hand, using a measuring cup filled with adjustable weight. The equipment included an adjustable table, small weights, a water jug filled with 1000 cc water, and a measuring cup (322 g) with a maximum content of 1000 cc and a handle that could be held with the cylinder grip (see Fig. 1). The table was set at the height of the iliac crest of the child. The measuring cup was placed on the table. The NAH was tested prior to the AH. Between each attempt, 90 seconds of rest was provided. The number of attempts needed to determine the maximal weight could vary.

The child was instructed to lift the cup horizontally by flexing the elbow and fixating the wrist with the cup stable in the horizontal plane for 5 seconds without lowering it. The wrist was in the neutral position or in slight radial deviation. Ulnar deviation was only allowed when there was no other possible way to lift the cup and to keep it horizontal. After 5 seconds, the cup was replaced on the table.



Cup and Box

| | Length | Width | Height |
|---|---|---|---|
| Cup | 21.5 cm | 15 cm | 13.5 cm |
| Box | 42 cm | 34 cm | 24 cm |


Weights

| | Length | Width | Height |
|---|---|---|---|
| Weight 50 gram | 6.5 cm | 6.5 cm | 0.2 cm |
| Weight 500 gram | 6.5 cm | 6.5 cm | 2 cm |
| Weight bag 1 kg | 12 cm | 9 cm | 0-5 - 1.0 cm |
| Weight bag 1,5 kg | 14 cm | 10 cm | 0.5 - 1.5 cm |
| Weight bag 2 kg | 14 cm | 11 cm | 0.5 - 2.0 cm |
| Weight bag 3 kg | 15 cm | 12 cm | 0.5 - 2.5 cm |


**Figure 1.** Cup and box used for the Cup-Task and Box-Task.
*cm = centimeter; g = gram; kg = kilograms.*

*Nonaffected hand.* Each participant started the Cup-Task NAH with a starting weight of 500 g. After each successful attempt, a weight of 100 to 500 g was added until an attempt was unsuccessful. The assessor was instructed to gradually build up to the maximum weight. If an attempt was unsuccessful, the weight was reduced in increments of 100 g until the child could perform the task as described.

*Affected hand.* Because the abilities of the AH differ considerably between children with USCP, it is very likely that functional muscle strength of the AH also differs between children. Therefore, before the Cup-Task AH started, the starting weight was determined using water. First, the child was instructed to lift and hold the empty cup with a flexed elbow and fixated wrist and to keep it steady in the horizontal plane. Next, the assessor filled the cup with water, using a fluent movement, until the child could no longer hold the measuring cup horizontally. The amount of water was measured, and this amount was used as the starting weight for the first attempt. When, during the first attempt, the task was not performed as described, the weight was reduced by 100 cc of water for the second attempt. When the task was performed as described, in each attempt the weight was increased by increments of 100 cc until an attempt was no longer successful. Above 1000 cc the water was replaced by weights.

### *Box-Task: bimanual functional muscle-strength test.*

The goal of the Box-Task was to test maximum bilateral functional muscle strength by determining the weight (in kilograms) a child can lift and hold for 5 seconds with both hands using a box filled with weight bags. The equipment included an adjustable table and a plastic box (0.8 kg) with handles (Fig. 1).

The box was situated on a table with the height adjusted so that the top of the box was at the height of the child's iliac crest. The child was instructed to lift and hold the box horizontally for 5 seconds by flexing the elbows and fixating the wrists. After 5 seconds, the box was replaced on the table. The first attempt involved lifting the box without weight. If the weight of the empty box could be held according to the criteria, 0.5 to 2 kg (depending on how easy it was for the child to lift the box) of weight was supplemented. The assessor was instructed to gradually build up to the maximum weight. Between each attempt, 90 seconds of rest was provided. The number of attempts needed to determine the maximal weight could vary.

### *Isometric grip strength.*

Maximum IGS strength was measured with the Biometrics E-LINK evaluation and exercise hand kit, both in the AH and NAH. The E-LINK evaluation system is a calibrated, computerized system incorporating a modified (digitalized) grip dynamometer. The applied force was measured in 100-g increments. Children were seated in an upright position in a chair with back support and armrests. The initial posture was a neutral position (0˚) of the wrist joint and 90˚ flexion of the elbow joint. The handle position of the E-LINK handgrip was adapted to the child's hand size, according to E-LINK guidelines. The child was also asked whether the position of the handle felt comfortable. When there was doubt, other handle positions were attempted.

In children with USCP, a previous study showed excellent test-retest reliability (intraclass correlation coefficient [ICC] values of 0.948 (95% confidence interval [CI] = 0.914–0.968) for the AH, and 0.942 (95% CI = 0.904–0.964) for the NAH. Interrater reliability was excellent for the AH (ICC = 0.976; 95% CI = 0.959–0.986) and for the NAH (ICC = 0.960; 95% CI = 0.932–0.977).[22]

## Statistical Analysis

Before determining reliability and validity of the Cup-Task and Box-Task, the percentage of participants who could not perform each test was determined. The measurements of these participants were excluded for the reliability and validity analyses.

ICC, model 2-way random, type absolute agreement, with 95% CI were used to assess test-retest reliability and interrater reliability. An ICC greater than 0.80 reflects excellent reliability, whereas ICCs from 0.70 to 0.79 reflect good reliability.[23]

The standard error of measurement (SEM) agreement was calculated as the square root of the error variance (including the systematic error).[24] The smallest detectable change (SDC) was computed as 1.96 multiplied by the square root of 2, multiplied by the SEM ($SDC = 1.96 \times \sqrt{2} \times SEM$).[24]

A simple and widely used method to interpret the SDC values is the Bland-Altman limits of agreement.[25] An assumption of the limits of agreement is that the differences between 2 measurements are normally distributed. When differences are not normally distributed, log transformation can be attempted.[26] However, in log-transformed data, the antilog of the difference between 2 values on a log scale is a dimensionless ratio.[25] For that reason, we only calculated limits of agreement when all differences were normally distributed (determined by the Shapiro-Wilk test).

To assess criterion validity, a Pearson correlation coefficient between the functional upper extremity strength measurements and the maximum IGS was calculated. For the Cup-Task AH and the Box-Task, a comparison with the IGS AH was made. For the Cup-Task NAH, a comparison with the IGS NAH was made. In all measurements, a significant moderate Pearson correlation coefficient[27] between 0.50 and 0.70 was hypothesized, because muscle strength seems to be the most important component of the functional strength measurement, but coordination and some endurance are also important components. The hypothesized values are also in line with previously reported validity values for functional strength measurement in children with CP.[17] Values were considered statistically significant at $P < .05$.



## Role of the Funding Source

This study was funded by the Forward for Children With Disabilities Foundation, Valkenburg, the Netherlands; the Revant Innovation Foundation, Breda, the Netherlands; and the Johanna Children's Foundation, Arnhem, the Netherlands. The funders had no role in the design, data collection, analysis, interpretation, or reporting of this work, or the decision to submit the work for publication.

# Results

A total of 86 children with USCP participated in this study. Because not every child performed every measurement (due to unavailability and/or measurement instrument), a different number of children was available for each test. For the statistical analyses of the reliability and validity values, only the children who could perform the specific measurement were included. See Figure 2 for a detailed description of the participant characteristics.

The results of the reliability studies for all tasks at different time points are shown in Table 1 (outcomes of the measurements) and Table 2 (reliability and validity).

## Cup-Task AH

### *Test-retest reliability.*

Of the 54 children tested, 9 children (16.9%) could not (adequately) perform 1 or more measurements with the empty cup. See Figure 2 for details.

Test-retest reliability was excellent (N = 45; ICC = 0.887; 95% CI = 0.713–0.948), with an SEM value of 284 g and an SDC value of 787 g.

### *Interrater reliability.*

Of the 54 children, 11 (20.3%) could not (adequately) perform the measurement with the empty cup in one of the attempts. See Figure 2 for details.

Interrater reliability was excellent (N = 43; ICC = 0.960; 95% CI = 0.918–0.980), with an SEM value of 142 g and an SDC value of 393 g.

### *Criterion validity.*

Of the 84 participants, 23 children (27.4%) could not perform the measurement with an empty cup and the IGS measurement. See Figure 2 for details. The Pearson correlation coefficient (N = 61) between the Cup-Task AH and IGS of the AH was moderate ($r = 0.638$; $P \leq .001$).

## Cup-Task NAH

### *Test-retest reliability*

All 54 children were able to perform the measurements on repeated occasions (Fig. 2). Test-retest reliability was excellent (ICC = 0.944; 95% CI = 0.895–0.969), with an SEM value of 272 g and an SDC value of 755 g. As seen in the Bland-Altman plot (Fig. 3), there were several outliers, but these are included in the calculations.

### *Interrater reliability*

All 54 children were able to perform the measurements during both tests (Fig. 2). Interrater reliability was excellent (ICC = 0.898; 95% CI = 0.825–0.941), with an SEM value of 421 g and an SDC value of 1166 g.

### *Criterion validity*

All 75 participants were able to perform both measurements. The Pearson correlation coefficient between the Cup-Task NAH and IGS NAH was low ($r = 0.489$; $P \leq .001$).

## Box-Task

### *Test-retest reliability*

Sixty-five children performed both measurements, and 3 children (4.8%) were not able to perform 1 or 2 measurements adequately. Test-retest reliability was excellent (N = 62; ICC = 0.934; 95% CI = 0.875–0.963), with an SEM value of 1.38 kg and an SDC value of 3.82 kg.

### *Interrater reliability*

All 54 children who participated in this part of the study were able adequately to perform the measurements twice. Interrater reliability was excellent (ICC = 0.896; 95% CI = 0.813–0.941), with an SEM value of 1.82 kg and an SDC value of 5.05 kg.

### *Criterion validity*

All 85 participants could lift the box and also perform the IGS adequately. For 1 child, an assessor determined that the empty box was the maximum weight capable of being lifted. The Pearson correlation coefficient between the Box-Task and maximum IGS AH was moderate ($r = 0.555$; $P \leq .001$).

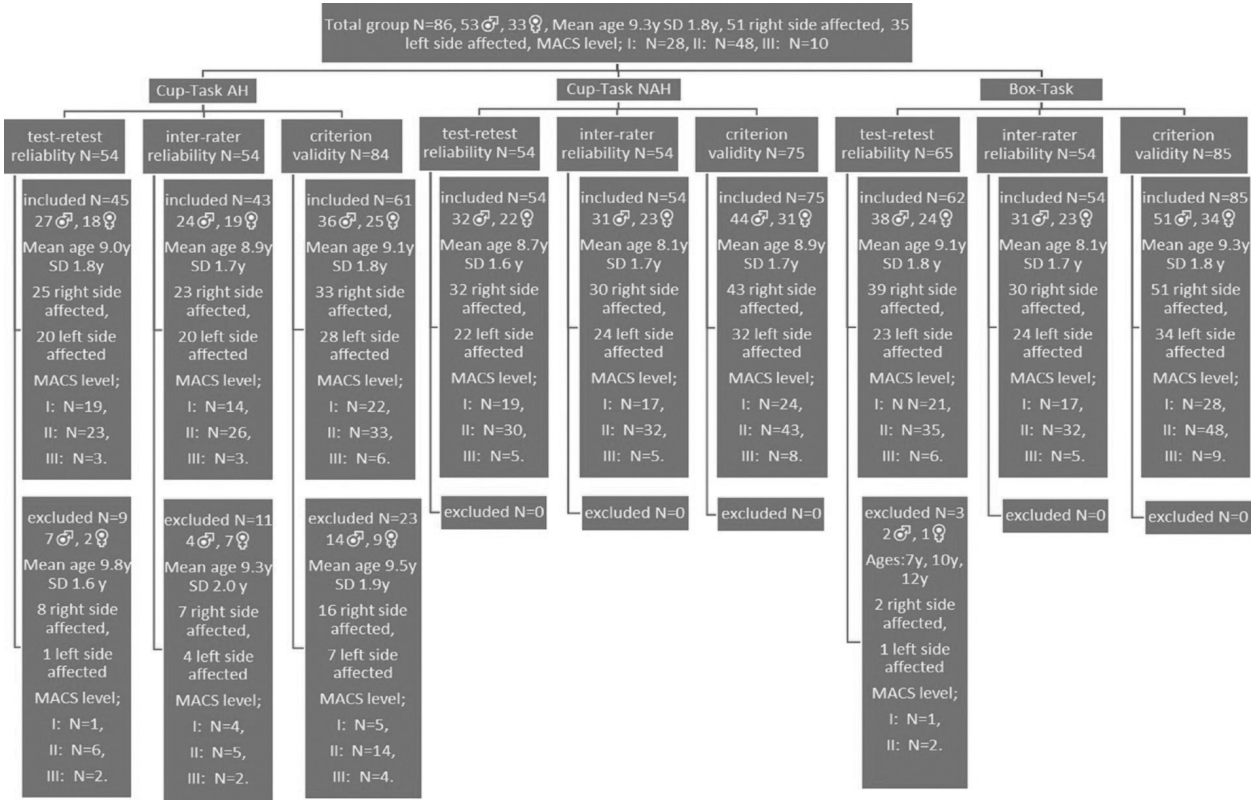


**Figure 2.** Participant characteristics.

*AH = affected hand; MACS = Manual Ability Classification System; NAH = nonaffected hand.*

**Table 1.** Outcomes

| | Test-Retest Reliability | | | | Interrater Reliability | | | |
|---|---|---|---|---|---|---|---|---|
| Task | Mean T0 (SD) | Mean T1 (SD) | Difference Between T0 and T1 | Shapiro-Wilk Test (*P* value) | Mean T0 (SD) | Mean T1 (SD) | Difference Between T0 and T1 | Shapiro-Wilk Test (*P* value) |
| Cup-Task AH | 568 g (535 g) | 787 g (687 g) | 210 g (342 g) | ≤ .001 | 602 g (475 g) | 679 g (545 g) | 77.3 g (187 g) | ≤ .001 |
| Cup-Task NAH | 1500 g (836 g) | 1637 g (823 g) | 137 g (363 g) | .09 | 1622 g (1068 g) | 1580 g (881 g) | 42.7 g (599 g) | .01 |
| Box-Task | 5.79 kg (3.74 kg) | 6.53 kg (3.99 kg) | 0.74 kg (1.81 kg) | ≤ .001 | 5.42 kg (4.03 kg) | 6.26 kg (4.30 kg) | 0.84 kg (2.46 kg) | ≤ .001 |

*AH = affected hand; Diff = difference; NAH = nonaffected hand.*

**Table 2.** Psychometric Properties of the Measurement Instruments

| | Test-Retest Reliability | | | | | Interrater Reliability | | | | | Criterion Validity | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Movement | N | ICC | 95% CI | SEM | SDC | N | ICC | 95% CI | SEM | SDC | N | Pcc with IGS | *P* value |
| Cup-Task AH | 45 | 0.887 | 0.713–0.948 | 284 g | 787 g | 43 | 0.960 | 0.918–0.980 | 142 g | 393 g | 61 | 0.638 | ≤ .001 |
| Cup-Task NAH | 54 | 0.944 | 0.895–0.969 | 272 g | 755 g | 54 | 0.898 | 0.825–0.941 | 421 g | 1166 g | 75 | 0.489 | ≤ .001 |
| Box-Task | 62 | 0.934 | 0.875–0.963 | 1.38 kg | 3.82 kg | 54 | 0.896 | 0.813–0.941 | 1.82 kg | 5.05 kg | 85 | 0.555 | .001 |

*AH = affected hand; CI = confidence interval; ICC = intraclass correlation coefficient; IGS = isometric grip strength; NAH = nonaffected hand; Pcc = Pearson correlation coefficient; SDC = smallest detectable change; SEM = standard error of measurement*

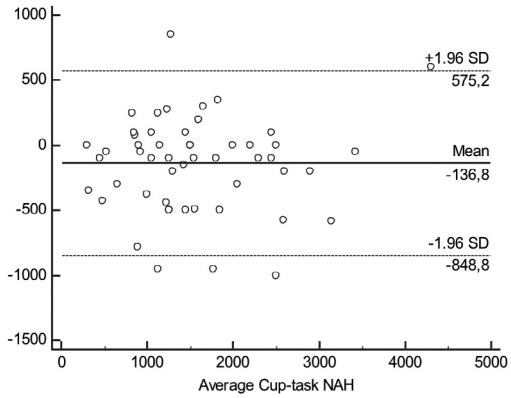


**Figure 3.** Limits of agreement:

*Cup-Task NAH test-retest reliability. NAH = nonaffected hand.*

# Discussion

The purpose of this study was to evaluate the psychometric properties of 2 newly developed functional hand and upper extremity muscle-strength tests in children aged 7 to 12 years with USCP.

## Reliability

Not all the children were able to perform the Cup-Task AH sufficiently. Therefore, the number of included children for the reliability part of the study was lower than intended. For the Cup-Task NAH and Box-Task, the target population size was achieved.

All test-retest reliability ICC values and all interrater reliability ICC values for all measurement instruments showed excellent reliability. These results are in line with the previously performed pilot study.[18] Because of the skewed distribution of the data of the Cup-Task AH and Box-Task, only the limits of agreement for the test-retest reliability of the Cup-Task NAH could be calculated. The Bland-Altman plot showed good agreement, as reflected by the narrow limits of agreement.

Because of the excellent test-retest reliability and excellent interrater reliability, all instruments can reliably be used for cross-sectional measurements, for example, to determine the muscle strength of the AH and NAH in children with USCP.

Beside test-retest reliability and interrater reliability, the SEM and SDC values are important components of reliability.[24] SEM and SDC values for these measurement instruments have not been previously reported. With the SEM and SDC values, the usability in patients in clinical practice can be determined, certainly when these instruments are used to determine changes over time in individual patients. By using these values, it can be determined whether the changes in functional strength after a strength training program are larger than the changes that can occur due to variability between 2 measurements. Unfortunately, no information is available on how much functional upper extremity strength improvement a child with USCP can achieve after such a training program. Therefore, it is not possible to draw clear conclusions about the usability of the measurement instruments to measure real and clinically important changes.

## Criterion Validity

In more than 25% of the participants, the assessor judged that the child was not able to perform the Cup-Task AH. Therefore, this measurement instrument is not suitable for a substantial proportion of children with USCP. Subanalyses of the participants who could not perform the task showed a wide distribution of age, sex, and MACS level; they included 5 children with MACS level I, 14 children with MACS level II, and 4 children with MACS level III. Comparing these MACS levels with those of the children included in the validity study (MACS level I, n = 22; level II, n = 33; level III, n = 6; see Fig. 2), it might be concluded that this task is more difficult to perform for children with MACS level III. Within the MACS level III group, most children were unable to lift the cup horizontally with the AH because they could not grasp the handle of the cup. For the children who were able to perform the Cup-Task AH, the Pearson correlation coefficient met the expectations. Therefore, it can be concluded that the

Cup-Task AH is valid for the children who are able to perform the task. For the children who could not perform the Cup-Task AH, another measurement instrument is needed to measure functional hand and upper extremity muscle strength.

The Cup-Task NAH and Box-Task were feasible for all participants, irrespective of the MACS level. The Pearson correlation coefficient of the Box-Task also met the expectations. However, the Pearson correlation coefficient of the Cup-Task NAH ($r = 0.489$) was just below the expected range (0.50–0.70).

Because these functional strength instruments have not been studied previously with children with USCP, no direct comparison with other studies can be made. Of the 11 supplemental tests recommended by the National Institutes of Health Common Data Elements for assessing upper extremity motor function in children with CP, only maximum voluntary isometric contraction testing and manual muscle testing measure muscle strength, and none measure upper extremity functional strength.[4] Thus the current tests have the potential to expand the number of reliable and valid tests for this population, and fill an important gap in our understanding of strength in the context of function.

## Strengths and Limitations

The strength of this study is its methodological quality. The COSMIN criteria were important for the design of this study, which resulted in scientifically valuable results.

Unfortunately, age, sex, and MACS level were not ideally distributed, which could have influenced the results.

Because this study focused on the age range of 7 to 12 years, caution is advised when testing children in other age groups. The population size was too small to calculate separate ICC/SEM/SDC values for each MACS level. The small number of children included in this study with MACS level III means that caution is advised when testing such children. Moreover, the large number of assessors could be a limitation of the study because this could have resulted in more measurement errors. However, the large number of assessors more closely resembles clinical practice.

# Conclusion

The Cup-Task and Box-Task are reliable and valid measurement instruments for measuring functional hand and upper extremity muscle strength in children with USCP who can perform such tasks. However, in most cases, for children with USCP and MACS level III the Cup-Task AH will not be suitable. To determine the usability of both instruments in children with USCP in longitudinal measurements, more research on the effects of increasing functional upper extremity strength in children with USCP is recommended.

## Acknowledgments

Special thanks to all children who participated in this study. Also, special thanks to the schools and/or rehabilitation centres for their cooperation.

## Ethics Approval

Ethics approval was granted by the Medical Ethical Board of Maastricht University Medical Center and Maastricht University (METC azM/UM, ref. no. NL45430.068.1), Maastricht, the Netherlands, and Teachers College, Columbia University, New York City, New York, USA. Data were collected in the Netherlands and United States from 2009 to 2016.

## Funding

This study was funded by the Forward for Children With Disabilities Foundation, Valkenburg, the Netherlands; the Revant Innovation Foundation, Breda, the Netherlands; and the Johanna Children's Foundation, Arnhem, the Netherlands.

## Study Registration

This study was registered at METC azM/UM (ref. no. NL45430.068.1).

## Disclosures

The authors completed the ICJME Form for Disclosure of Potential Conflicts of Interest and reported no conflicts of interest.

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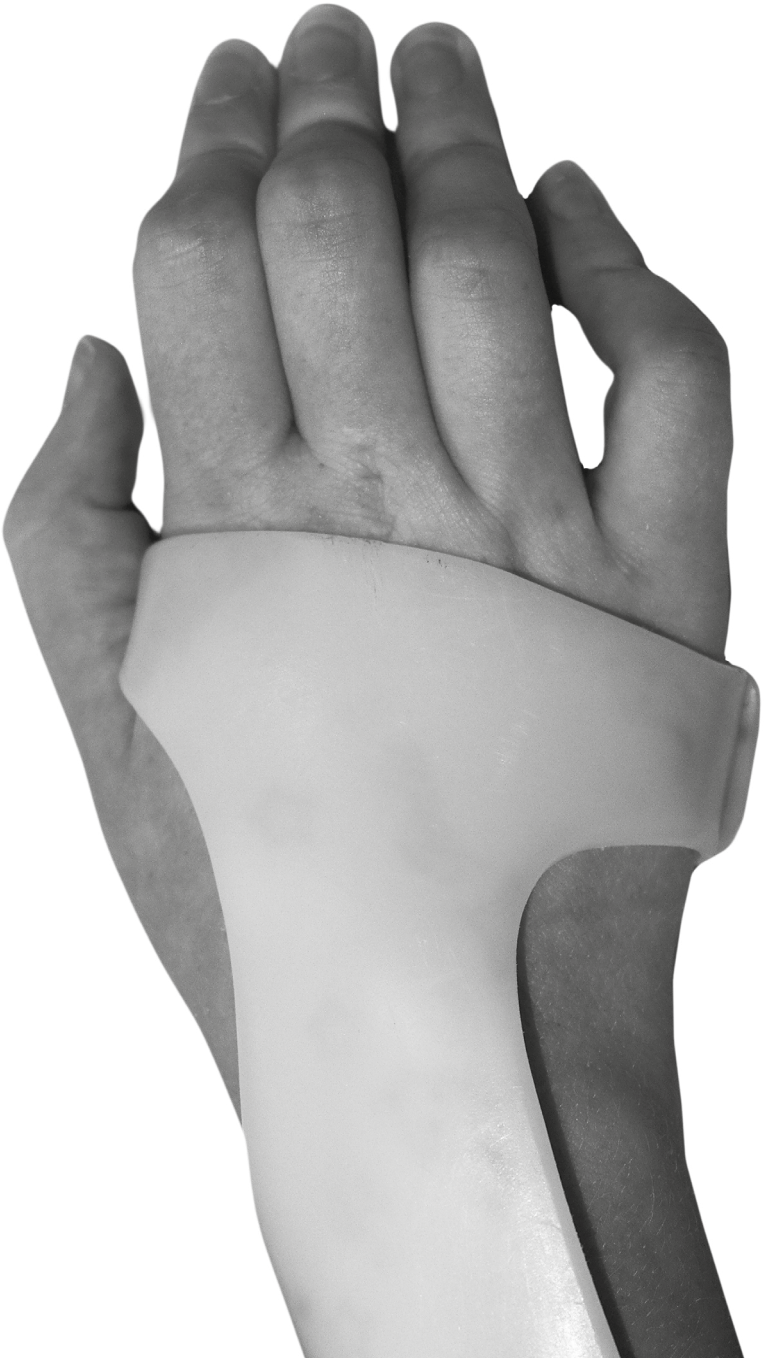

# Chapter 5

# Upper extremity muscle strength in children with unilateral spastic cerebral palsy: A bilateral problem?

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## Abstract

**Objective***:* The objective was to investigate whether muscle strength in the non-affected and affected upper extremity (UE) in children (7–12 years) with Unilateral Spastic Cerebral Palsy (USCP) differs from that in children with typical development (TD).

**Methods:** A cross-sectional study design was used. In children with USCP, isometric arm strength (wrist flexion, wrist extension with flexed and extended fingers, elbow flexion/extension) was assessed in 72 children (mean [SD] age 9.3 [1.9] years) and isometric grip/pinch strength was assessed in 86 children (mean [SD] age 9.3 [1.8] years). For children with TD, arm/grip/pinch strength was assessed in 120 children (mean [SD] age 9.5 [1.7] years). Arm strength was measured with the MicroFET2 hand-held dynamometer and grip/pinch strength with the Biometric E-Link evaluation system. The non-affected UE of children with USCP was compared with the preferred UE of children with TD, because both sides represent the preferred UE. The affected UE was compared with the non-preferred UE of children with TD, as both sides represent the non-preferred UE.

**Results:** In all measurements except for grip strength of the preferred UE, children with USCP were weaker than children with TD.

**Conclusions:** In children with USCP, muscle strength weakness exists in both upper extremities.

*Impact statement:* When uni- or bimanual ability limitations are present in children with unilateral cerebral palsy, investigation of the muscle strength of the non-affected UE should be part of the assessment.

# Introduction

Unilateral spastic cerebral palsy (USCP) is characterized by motor impairments lateralized to one body side, resulting in an "affected" body side and a "non-affected" body side.[1–3] Several studies have shown that muscle strength of the affected upper extremity (UE) is considerably impaired compared to the non-affected UE and compared to the UE strength of children with typical development (TD).[4–6] Muscle strength weakness of the affected UE is one of the motor impairments affecting manual abilities.[6,7]

Several magnetic resonance imaging (MRI) studies in children with supposed USCP have shown bilateral brain lesions.[8–11] Also, many clinicians perceive problems in the "non-affected" body side, and often this described possible impairments of the "non-affected" body side or found reduced performance of the "non-affected" UE in children with cerebral palsy compared to children with TD.[12–15]

Whether muscle strength weakness in the more-affected UE is the cause of this reduced performance is unclear, and one should keep in mind that these differences can also be attributed to problems with speed and/or coordination. So far, only two studies have investigated the muscle strength of the non-affected hand in children with USCP, with opposite conclusions. One study showed that grip strength of the non-affected hand of the children with USCP was, on average, 12% weaker compared to a group of children with TD.[12] Another study found no significant differences between grip/pinch strength of the non-affected hand of children with USCP compared to norm values of children with TD.[14] Both studies only studied grip/pinch strength and studies on the strength of the non-affected forearm and upper arm muscles are lacking. Hand strength is important for executing fine motor activities, but also the strength of upper and lower arm muscles is important during gross motor UE activities, such as lifting and carrying objects. It is therefore important to assess whether muscle weakness in the non-affected UE is present or not, as this might have consequences when selecting the proper assessment/treatment to map/improve UE function.

Research on this topic has been done among adults after unilateral stroke. Several studies have reported motor impairments in the non-affected UE in adults after unilateral stroke.[16–23] Although these motor impairments are substantially less severe than in the affected UE, they can produce significantly limiting (bilateral) functional impairments, including problems performing the activities of daily living.[24–26]

In more than 50 papers, most of which focused on activities involving the lower limb of adults with unilateral stroke, muscle strength appeared to be related to functional activity performance.[27] Such a relationship was also demonstrated for the upper limb[28] and it has been proven that strengthening interventions do not only improve muscle strength but also activity after stroke.[29]

Although the cause for both USCP and stroke originates in the brain, the body functions in children with USCP have hardly developed at the time the brain injury/malformation occurs. Because children with USCP only use their non-affected UE spontaneously in daily activities,[30,31] the non-affected UE is maximally stimulated during development. Adults who have had a stroke used both hands normally before the stroke occurred. Therefore, it cannot simply be concluded that the findings in adults with stroke also apply to children with cerebral palsy.

The aim of this study was to investigate whether muscle strength in the "non-affected" UE in children with USCP differs from children with TD. As prior studies that assessed the strength of the affected side used small sample sizes and/or only studied hand strength and/or used a measurement instrument that shows wide grading values when applied in children with moderate to good muscle strength,[4–6] the muscle strength of the affected UE is also examined within this study.

# Methods

## Study design

All data were obtained according to a cross-sectional study design. Data were collected in The Netherlands and the USA from 2009 to 2017.

The non-affected UE of children with USCP was compared with the preferred UE of children with TD, because both sides represent the preferred UE. The affected UE was compared with the non-preferred UE of children with TD, as both sides represent the non-preferred UE.

## Participants

Permission was granted by the Medical Ethical Board of the Maastricht University Medical Center and Maastricht University (METC azM/UM; trial number NL45430.068.1) in The Netherlands and by Teachers College, Columbia University in New York City, USA.

For the children with USCP, muscle strength values were obtained from a study on the reliability of maximum isometric arm and grip/pinch strength measurements.[32] Within this study population the sample sizes for arm and grip/pinch strength varied due to the availability of the children and/or measurement instruments at the facilities. Children were recruited from four different Dutch rehabilitation centers and related schools for special education: Adelante Rehabilitation Center, Valkenburg; Libra Rehabilitation and Audiology, Tilburg; Revant Rehabilitation Centers, Breda and Goes; and Tolbrug Rehabilitation Center, Den Bosch. In the USA, the children with USCP were a convenience sample of children participating in an ongoing intensive UE training program at Teachers College, Columbia University. To be included, the child had to be diagnosed with USCP and aged between 7 and 12 years. The child also had to be classified as Gross Motor Function Classification System (GMFCS)[33] level I or II and Manual Ability Classification System (MACS)[34] level I, II or III. All participants were capable of following simple instructions. A child was excluded when he/she had surgery or botulinum toxin A treatment in the UE in the past six months.

The children with TD were recruited in The Netherlands. Primary schools in different regions, both in cities and the countryside, were approached to participate in this research. After informed consent of the management of the school, children were selected at random and invited to participate in this study. After informed consent had been received from their parents (and from the children aged 12 years), the children were invited for the measurements.

## Procedure

The measurements took place in the child's own environment: the rehabilitation center for the children with USCP; and primary school for the children with TD.

A standardized protocol with detailed descriptions of all procedures and measurements was used. Prior to testing, body weight and height, MACS and GMFCS level (for the children with USCP) were determined. All children performed one set of strength measurements, consisting of isometric arm strength (IAS) with the hand-held dynamometer (HHD) and isometric grip/pinch strength (IGPS) with the E-Link evaluation system (see Measures for a description). The sequence of strength measurements was randomized. Both upper extremities were measured successively, with the preferred UE being tested before the non-preferred UE.

The HHD and E-Link measurements were performed three consecutive times and the mean of the three measurements was calculated. In this way, variation in muscle strength due to variations in placing the measurement instrument near the described measurement spot was minimized. Between each measurement, the child had at least 30 seconds of rest, leaving sufficient time for the muscles to recover.

Test scores were read by the therapist and registered by the same therapist on a test form. For the E-Link evaluation system, test scores were also stored on the E-Link evaluation system computer.

All measurements were performed by ten different assessors having no direct professional relationship with the participants. Two of the assessors (K.D., E.R.) involved during the entire project were experienced pediatric research physical therapists. The other eight assessors involved in the study for 6 months were master's students in pediatric physical therapy. Each assessor received 4 hours of training from K.D. or E.R. regarding use of the standardized protocol.

## Measurements

The child was seated in an upright position on a chair with back support and armrests. The armrests of the chair were used to support the arms during testing. The initial posture was a neutral position (0**°**) of the wrist joint and 90**°** flexion of the elbow joint. For elbow extension, the initial posture was adjusted so that the lower arm could move and elbow extension was possible. If a child was unable to perform the test, the result of the measurement was not used in the analysis.

### *Isometric arm strength (IAS)*

Maximum isometric muscle strength of the wrist extension with extended fingers, wrist extension with flexed fingers, wrist flexion and elbow flexion/extension were measured with the MicroFET2 HHD (Hoggan Scientific LLC, Salt Lake City, UT, USA). The HHD is an electronic device that fits in the palm of the hand of the assessor. A load cell (strain gauge technology) measures the isometric muscle strength applied to a transducer. The "make method", in which the child applies force against a fixed HHD, was used.[35] The applied force was measured in Newtons. Children were encouraged by the therapist to produce maximum force. Reliability of the measurements for children with USCP is excellent.[32]

### *Isometric grip/pinch strength (IGPS)*

The IGPS was measured with the Biometric E-Link evaluation system (Biometrics Ltd, Gwent, UK), a calibrated, computerized system that incorporates a modified (digitized) grip dynamometer and a pinch meter. The applied force was measured in 0.1 kilograms. The handle position of the E-Link handgrip was adapted to the child's hand size according to the E-Link guidelines for positioning. The child was also asked where the position of the handle felt the best. When there was uncertainty, other handle positions were tried. Children were encouraged by the therapist to produce maximum force. Reliability of the measurements in children with USCP is excellent.[32]

## Statistical analysis

All statistical analyses were performed using R.

### *Participant Characteristics*

Descriptive statistics including means, standard deviations, and confidence intervals were used to summarize participant characteristics and strength measurements by age and group (children with USCP or children with TD). Independent samples t-tests were used to compare baseline characteristics.

### *Isometric Arm Strength (HHD)*

Exploratory analyses revealed positive correlations between measures of arm-strength in the five different positions (wrist extension with extended fingers, wrist extension with flexed fingers, wrist flexion, elbow flexion, elbow extension). Rather than analyzing each outcome separately, and in order to avoid Type I errors, a multivariate analysis of variance (MANOVA) was performed on the five variables (as a matrix of dependent variables), with age, sex, group, and an age × group interaction as independent variables. A separate MANOVA was performed for the preferred UE and the non-preferred UE.

Given that MANOVA does not permit specification of how the combination of dependent variables differ between groups, linear discriminant analysis (LDA) was performed as a follow-up. LDA is a dimensionality reduction technique that can be used to characterize two or more classes/categories. For these analyses, we used LDA to examine which variables best distinguished between the two groups (children with USCP and children with TD).

### *Isometric Grip/Pinch Strength (E-Link)*

The analysis for grip and pinch strength involved 4 generalized linear models (GLM) that were fit using a Gaussian distribution with an identity link. The models were separately fit to evaluate the difference between groups for 1) grip strength of the preferred UE, 2) pinch strength of the preferred UE, 3) grip strength of the non-preferred UE, and 4) pinch strength of the non-preferred UE. Age, sex, and an age × group interaction were also included in the models to adjust for their potential impact, with

the associations between age and strength assumed to be linear. In our specific analysis, the group children with USCP was set as the reference group. Thus, the parameter estimate for group is the difference between the group of children with TD relative to the group of children with USCP, when adjusted for age, sex, and age × group interaction. Given the possibility of heteroscedasticity, robust standard errors were computed for the parameter estimates using generalized estimating equations with an independent correlation structure. P-values were computed using a Wald statistic.

# Results

Participant characteristics for gender, age, height, weight, preferred/non-preferred side and MACS-levels (for children with USCP) are provided in Table 1.

All children were able to perform the measurements. For children with USCP, muscle strength values of 72 children for the IAS measurements and 86 children for the IGPS measurements were taken. For the children with TD, a total of 120 children were included in all measurements. There were no significant differences between the groups in age (USCP-IAS/TD: p = 0.53; USCP-IGPS/TD: p = 0.56), height (USCP-IAS/TD: p = 0.07; USCP-IGPS/TD: p = 0.06) or weight (USCP-IAS/TD: p = 0.93; USCP-IGPS/TD: p = 0.88).

**Table 1:** Descriptives of the participants.

| **USCP** | **Age-groups** | |
|---|---|---|
| Isometric Arm Strength measurements (HHD)<br>n = 72; 45♂, 27♀<br>Mean age 9 years 3 months. SD 1 year 9 months.<br>45 right side affected. 27 left side affected<br>MACS level; I: n = 23. II: n = 42. III: n = 7<br>Mean height = 139cm; SD 12.3cm<br>Mean weight = 37.2kg; SD 10.2kg | Age 7; n = 17 | 9♂, 8♀ |
| | Age 8; n = 12 | 5♂, 7♀ |
| | Age 9; n = 12 | 9♂, 3♀ |
| | Age 10; n = 8 | 5♂, 3♀ |
| | Age 11; n = 7 | 5♂, 2♀ |
| | Age 12; n = 16 | 12♂, 4♀ |
| Isometric Grip and pinch strength measurements (e-link system)<br>n = 86. 53♂, 33♀<br>Mean age 9 years 3 months. SD 1 year 8 months.<br>51 right side affected. 35 left side affected<br>MACS level; I: n = 29. II: n = 47. III: n = 10<br>Mean height = 139cm; SD 12.2cm<br>Mean weight = 37.0kg; SD 10.1kg | Age 7; n = 19 | 10♂, 9♀ |
| | Age 8; n = 14 | 6♂, 8♀ |
| | Age 9; n = 17 | 11♂, 6♀ |
| | Age 10; n = 9 | 5♂, 4♀ |
| | Age 11; n = 11 | 9♂, 2♀ |
| | Age 12; n = 16 | 12♂, 4♀ |
| **TD** | **Age-groups** | |
| All measurements<br>n = 120. 60♂, 60♀<br>Mean age 9 years 5 months. SD 1 year 7 months.<br>Preferred right side: n = 106; preferred left side n = 14<br>Mean height = 143cm; SD 12.6cm<br>Mean weight = 37.4kg; SD 11.6kg | Age 7; n = 20 | 10♂, 10♀ |
| | Age 8; n = 20 | 10♂, 10♀ |
| | Age 9; n = 20 | 10♂, 10♀ |
| | Age 10; n = 20 | 10♂, 10♀ |
| | Age 11; n = 20 | 10♂, 10♀ |
| | Age 12; n = 20 | 10♂, 10♀ |

*♂ = male; ♀ = female; cm = centimeter; HHD = hand held dynamometer; kg = kilogram; MACS = Manual Ability Classification; n = population size; SD = standard deviation; TD = typically developing; USCP = Unilateral Spastic Cerebral Palsy.*

## Isometric Arm Strength

### *Preferred UE*

The means and differences in arm muscle strength are presented in Table 2. Table 3 provides the parameter estimates for the MANOVA. For the preferred UE, children with USCP were consistently weaker than children with TD. There was a significant interaction between age and group suggesting that differences between the two groups are not constant across the age groups for the combined dependent variables $F(5, 183) = 2.49$, $p < .05$, Pillai's Trace = 0.063. Across the age groups, the difference in muscle strength of the elbow flexors and elbow extensors appear to change most between children with USCP and children with TD. It is remarkable that at the age of 7, the group of children with USCP is stronger in elbow flexion and elbow extension than the group of children with TD. At the age of 12, the group of children with TD is stronger in these muscle groups compared to the group of children with USCP.

Discriminant analysis was used to determine if the five measurements of IAS differentiated between children with USCP and children with TD. Table 3 provides a summary of the linear discriminant function coefficients associated with each measurement. Wrist extension, wrist extension with fingers flexed, and elbow extension provided the greatest contribution to group separation. Figure 1 demonstrates the group separation using the values of the discriminant function for the group of children with USCP and group of children with TD. Despite differences between the two groups, there is some degree of overlap in the distributions.

### *Non-preferred UE*

For the non-preferred UE, children with USCP were consistently weaker than children with TD. Table 3 provides the parameter estimates for the MANOVA. There was a significant interaction between age and group suggesting that differences between the two groups are not constant across the age groups for the combined dependent variables $F(5, 183) = 5.14$, $p < .001$, Pillai's Trace = 0.12. Across the age groups (young to old), the difference between the two groups changes the most for elbow flexion and elbow extension.

Table 3 provides a summary of the linear discriminant function coefficients associated with each measurement for the non-preferred UE. Wrist extension with fingers flexed and wrist flexion provided the greatest contribution to group separation. Figure 1 demonstrates the group separation using the values of the discriminant function for the group children with USCP and group children with TD. For most cases, there appears to be clear separation between the two groups.

**Table 2:** muscle strength values, between groups and between ages

| | USCP | | | TD | | | Between groups (TD-USCP) | | Difference in muscle strength per age; USCP vs TD, in percent (%) | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Mean | SD | Min-max | Mean | SD | Min-max | Diff mean (95% CI) | Diff % (95%CI) | 7 y | 8 y | 9 y | 10 y | 11 y | 12 y |
| *preferred UE* | | | | | | | | | | | | | | |
| Wrist extension, extended fingers | 55.2N | 20.4N | 5.3–100.7N | 69.8N | 19.8N | 26.4–119.2N | 14.6N (8.7–20,4N) | 21% (12–29%) | 13 | 15 | 17 | 14 | 17 | 33 |
| Wrist extension, flexed fingers | 60.1N | 25.4N | 18.3–139.8N | 76.4N | 21.3N | 33.0–141.9N | 16.3N (9.6–23,1N) | 21% (13–30%) | 16 | 20 | 20 | 9 | 17 | 31 |
| Wrist flexion | 63.1N | 25.1N | 21.7–147.3N | 70.7N | 20.0N | 22.0–126.3N | 7.6N (1.2–14,1N) | 11% (2–20%) | 5 | 7 | 5 | -8* | 26 | 18 |
| Elbow flexion | 102.0N | 37.8N | 34.0–217.7N | 106.9N | 30.6N | 41.2–202.2N | 4.9N (-4.9–14,8N) | 5% (-5–14%) | -17* | 3 | -9* | -3* | 7 | 24 |
| Elbow extension | 93.0N | 37.3N | 38.7–257.3N | 93.6N | 26.2N | 18.7–156.7N | 0.6N (-8.5–9,7N) | 1% (-9–10%) | -21* | 3 | -14* | -9* | 9 | 17 |
| Grip strength | 13.5kg | 6.5kg | 2.5–38.2kg | 16.0kg | 4.9kg | 6.7–28.2kg | 2.5kg (0.8–4.1kg) | 15% (5–25%) | 14 | 14 | 14 | 8 | 17 | 15 |
| Pinch strength | 3.8kg | 1.5kg | 0.8–8.2kg | 4.3kg | 1.3kg | 1.7–10.4kg | 0.6kg (0.2–1,0kg) | 13% (5–22%) | 25 | 18 | 10 | 4 | 13 | 5 |
| *non-preferred UE* | | | | | | | | | | | | | | |
| Wrist extension, extended fingers | 25.7N | 18.2N | 0.1–93.1N | 69.8N | 19.3N | 25.0–119.2N | 44.1N (38.5- 49.6N) | 63% (55–71%) | 61 | 53 | 68 | 57 | 64 | 68 |
| Wrist extension. flexed fingers | 29.7N | 18.5N | 0.1–98.7N | 76.7N | 20.7N | 34.2–138.0N | 46.9N (41.1- 52.8N) | 61% (54–69%) | 55 | 57 | 62 | 59 | 71 | 62 |
| Wrist flexion | 34.2N | 14.1N | 5.1–82.0N | 69.1N | 19.3N | 16.3–122.6N | 35.0N (30.2–39.7N) | 51% (44–57%) | 42 | 46 | 46 | 49 | 59 | 58 |
| Elbow flexion | 70.8N | 28.4N | 22.7–144.0N | 104.9N | 29.7N | 50.3–203.1N | 34.2N * (25.6–42.8N) | 33% (24–41%) | 14 | 35 | 25 | 24 | 35 | 47 |
| Elbow extension | 63.6N | 23.0N | 27.0–125.4N | 92.5N | 25.4N | 21.4–160.4N | 29.0N (21.7–36.2N) | 31% (24–39%) | 14 | 33 | 18 | 32 | 40 | 43 |
| Grip strength | 5.2kg | 4.3kg | 0.1–26.2kg | 15.1kg | 4.8kg | 4.5–28.3kg | 9.9kg (8.7–11.2kg) | 66% (57–74%) | 57 | 73 | 67 | 63 | 71 | 61 |
| Pinch strength | 1.6kg | 1.1kg | 0.1–5.5kg | 4.1kg | 1.2kg | 1.6–9.1kg | 2.5kg (2.2–2,8kg) | 60% (53–68%) | 61 | 63 | 67 | 50 | 63 | 53 |

* Negative value: the mean of the USCP-group is higher than the mean of the TD-group

*95%CI = 95% confidence interval; diff = difference; kg = kilogram; max = maximum; min = minimum; n = population size; N = Newton; SD = standard deviation; TD = typically developing; USCP = Unilateral Spastic Cerebral Palsy; y = year*

**Table 3.** Results from MANOVA and LDA of Isometric Arm Strength

| **Preferred Upper Extremity** | | | |
|---|---|---|---|
| **Variable** | **Pillai's trace** | **Approx F** | **p*** |
| USCP vs. TD (Group) | 0.23 | 10.83 | < 0.001 |
| Age | 0.30 | 15.53 | < 0.001 |
| Sex | 0.02 | 0.75 | 0.586 |
| Age × Group interaction | 0.06 | 2.49 | < 0.05 |
| **Non-preferred Upper Extremity** | | | |
| **Variable** | **Pillai's trace** | **Approx F** | **p*** |
| USCP vs. TD (Group) | 0.68 | 78.92 | < 0.001 |
| Age | 0.30 | 15.80 | < 0.001 |
| Sex | 0.01 | 0.34 | 0.888 |
| Age × Group interaction | 0.12 | 5.14 | < 0.001 |

| **Preferred Upper Extremity** | |
|---|---|
| **Variable** | **Discriminant Function Coefficients** |
| Wrist Extension | 0.031 |
| Wrist Extension (fingers flexed) | 0.033 |
| Wrist Flexion | 0.013 |
| Elbow Flexion | -0.008 |
| Elbow Extension | -0.030 |

| **Non-preferred Upper Extremity** | |
|---|---|
| **Variable** | **Discriminant Function Coefficients** |
| Wrist Extension | 0.019 |
| Wrist Extension (fingers flexed) | 0.029 |
| Wrist Flexion | 0.032 |
| Elbow Flexion | -0.009 |
| Elbow Extension | -0.012 |

*TD = typically developing; USCP = Unilateral Spastic Cerebral Palsy;*

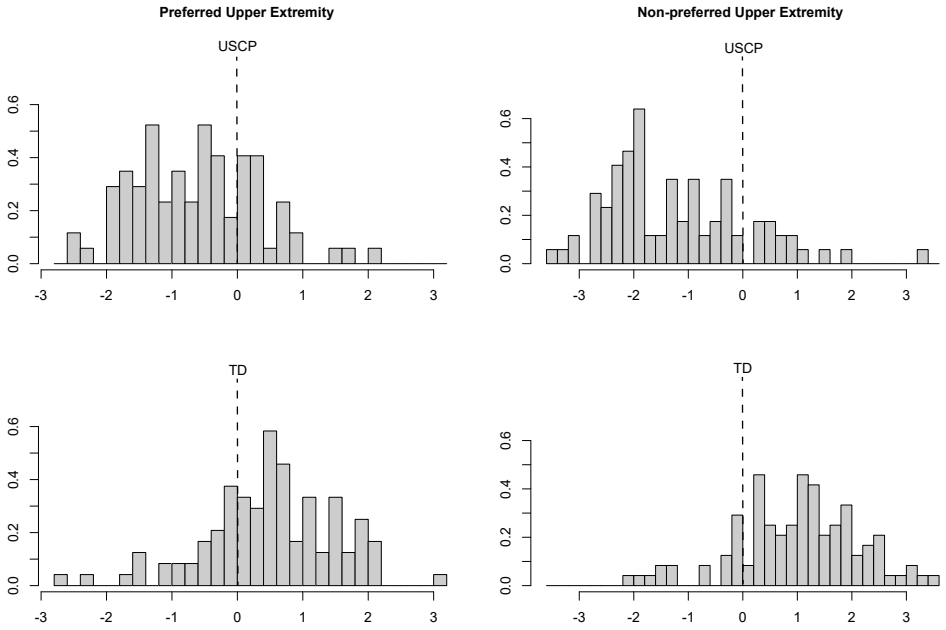


**Figure 1.** Group separation using the values of the discriminant function for the group of children with USCP and group of children with TD

## Isometric Grip/Pinch Strength

The means and differences in grip and pinch strength are presented in Table 2. For three out of the 4 measurements, analysis of grip/pinch strength resulted in statistically significant group differences. Table 4 summarizes the results for the fitted models. For pinch strength of the preferred UE, pinch strength of the non-preferred UE, and grip strength of the non-preferred UE, children with TD on average showed higher scores when controlling for age and sex. In addition, for grip strength of the non-preferred UE, there was a significant age by group interaction with children with TD showing greater gains in strength over time (Figure 2).

**Table 4.** Results from GLM of Grip and Pinch Strength

| | Estimate | SE | p* |
|---|---|---|---|
| **Preferred Upper Extremity** | | | |
| *Pinch Strength, kg* | | | |
| Intercept** | 2.88 | 0.23 | < 0.001 |
| TD relative to USCP | 0.76 | 0.29 | < 0.01 |
| One year increase in age | 0.43 | 0.07 | < 0.001 |
| Female relative to Male | -0.28 | 0.17 | 0.08 |
| Age × Group interaction | -0.09 | 0.10 | 0.41 |
| *Grip Strength, kg* | | | |
| Intercept** | 9.74 | 0.85 | < 0.001 |
| TD relative to USCP | 1.35 | 1.07 | 0.21 |
| One year increase in age | 1.76 | 0.27 | < 0.001 |
| Female relative to Male | -0.68 | 0.64 | 0.29 |
| Age × Group interaction | 0.38 | 0.36 | 0.30 |
| **Non-preferred Upper Extremity** | | | |
| *Pinch Strength, kg* | | | |
| Intercept** | 1.23 | 0.20 | < 0.001 |
| TD relative to USCP | 2.14 | 0.25 | < 0.001 |
| One year increase in age | 0.19 | 0.06 | < 0.01 |
| Female relative to Male | -0.12 | 0.15 | 0.44 |
| Age × Group interaction | 0.14 | 0.09 | 0.12 |
| *Grip Strength, kg* | | | |
| Intercept** | 4.72 | 0.72 | < 0.001 |
| TD relative to USCP | 6.79 | 0.90 | < 0.001 |
| One year increase in age | 0.67 | 0.23 | < 0.05 |
| Female relative to Male | -0.21 | 0.54 | 0.7 |
| Age × Group interaction | 1.24 | 0.31 | < 0.001 |

*p-value calculated using Wald Stastic; **model was centered on age variable (intercept represents mean at age 7 years) with USCP group set as the reference group

*kg = kilogram TD = typically developing; USCP = Unilateral Spastic Cerebral Palsy;*

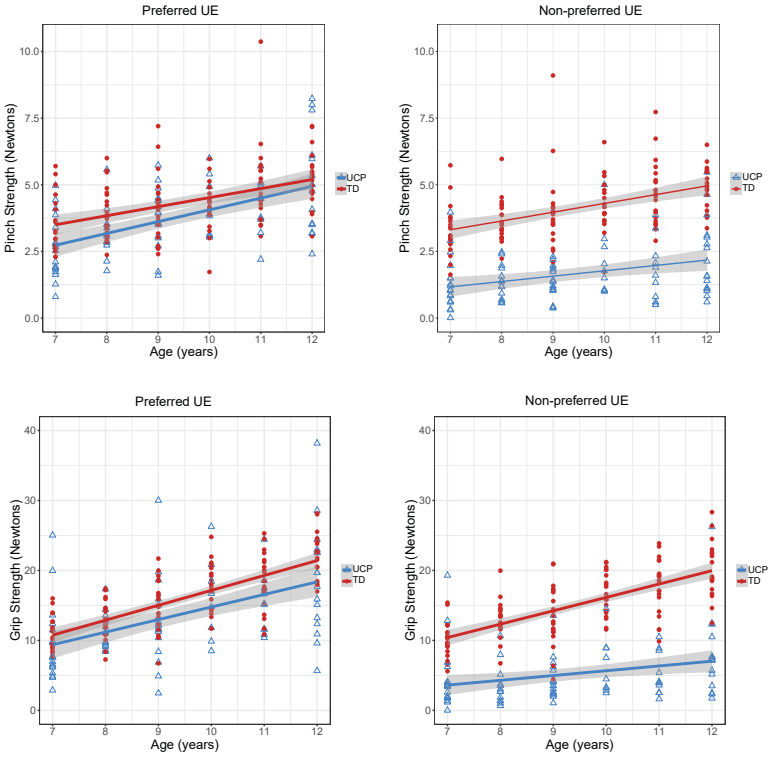


**Figure 2.** Age by group interaction

# Discussion

The aim of this study was to investigate whether the UE muscle strength between children with USCP and the children with TD differs for both the PUE and the non-preferred UE.

## Muscle strength in the preferred UE

Overall, children with USCP were consistently weaker than children with TD in their preferred UE, except for grip strength. These findings are almost consistent with the findings in adults with stroke. In adults with stroke impairments in strength of the total UE were found.[19,23]

Muscle weakness of the preferred UE of children with USCP seems to go beyond impairments in the hand. It is striking that the muscle strength of the elbow flexion and extension in the younger age groups is higher in children with USCP than in children with TD. In the older age groups, this difference between groups is reversed. A possible explanation could be the intensive (bimanual) training in children with USCP at the younger age and more disuse of the preferred UE at the older age. More research to explain this result is needed.

Rich et al.[12] and Tomhave et al.[14] assessed only differences in grip and pinch strength, so the results of these two strength measurements can be compared.

The results in our study regarding grip strength are comparable to the result of Tomhave et al.,[14] but different to that of Rich et al.[12] In the study of Rich et al., older children (8–18y, mean 14,1y, sd 2.4y) were included.[12] As figure 2 shows, the differences in grip strength becomes larger in the older age groups. This could explain why the differences in grip strength between the groups is probably not yet clear in our population.

Our findings regarding the difference in pinch strength contradict the study of Tomhave et al. (similar mean value compared to norm value).[14] Although all studies use the average of three measurements to determine the muscle strength, some differences in methodology exist that may explain these discrepancies.

A different measurement instrument was used to measure grip strength: the digitized Biometric E-Link evaluation system (Biometrics Ltd, Gwent, UK) in our study versus a Jamar hydraulic hand dynamometer (Patterson Medical, Warrenville, IL, USA) in the other studies.[12,14] The Biometric E-Link system has (digitized) incremental steps of 0.1 kg, whereas the Jamar hydraulic dynamometer has (visual) incremental steps of 2 kg/5 pounds. Therefore, small differences in muscle strength are more likely to be picked up using our E-Link system.

In our study and in the study of Rich et al.[36] children with TD were used as controls, but Tomhave et al.[14] compared the hand strength of children with USCP with previously published norms. These norms are based on 199 Brazilian children recruited within the same area and divided into ten age/gender groups.[37] About 37% of the American children were overweight or obese, whereas in Brazil this percentage is about 16–20%.[38,39] Within The Netherlands this percentage is about 12%.[40] As increasing weight status is associated with improved grip strength, [38] it is unclear whether the norm population sufficiently resembled the total population of children within the USA and The Netherlands.

Regarding our results, in most measurements the group of children with USCP showed a larger range of muscle strength compared to children with TD. These results cannot be compared to the other studies because this information is not available. With this larger range in muscle strength it is expected that specific characteristics related to children with USCP, such as MACS level and/or location of the lesion, may have an impact on muscle weakness. However, due to the small subgroups (e.g. there are only 7–9 children with MACS level III), a comparison of muscle strength values for different MACS levels was not possible between children with TD and children with USCP.

Because there was no information on the overall activity and participation levels of the children with USCP, there is a chance that reduced overall activity and participation levels might have affected the hand function of the non-affected UE.

Unfortunately, MRIs or neurophysiological data for the children with USCP were not available, so we could not examine whether the muscle weakness in children with USCP is related to a specific brain damage location. In addition, it is not known how much of the deficit might be due to bilateral involvement of the brain.

## Muscle strength in the non-preferred upper extremity

In the non-preferred UE, for all measures the children with USCP produced statistically significantly lower muscle strength values compared to children with TD. These differences in muscle strength are in accord with other studies.[4,5] Our study confirms the hypothesis that children with USCP can generate less muscle strength with the non-preferred side compared to children with TD. However, it is remarkable that the percentage difference in muscle strength is less in the proximal UE muscle groups compared to the distal UE muscle groups. A possible explanation is that the severity of hand function is closely related to the integrity and organization of direct corticospinal projections to the hand muscles and that these largely control distal movement/force.[41] A second explanation might be that because most children with USCP only use the non-preferred UE to support the preferred UE, the proximal muscle groups may be used more compared to the distal (fine motor) muscle groups.

## Limitations

We used a cross-sectional study design to compare differences in muscle strength between both groups. However, it should be noted that we did not study changes in muscle strength within each individual child. To do so a longitudinal study design is more appropriate, but such studies are logistically challenging to execute.

Because the age of our study population ranges from 7 to 12 years, these results cannot be extrapolated to other age groups.

The children with USCP are American and Dutch whereas the children with TD were exclusively Dutch children. Important patient characteristics, such as weight, height and age, did not differ significantly between the two groups, therefore the possible influence of country of residence is likely to be minimal. However, because most participants were Dutch, it is unknown if the differences in muscle strength are generalizable to populations with a lower or higher percentage of overweight or obese children.

We tried to have all therapists testing both the children with USCP and children with TD, but unfortunately this was not always possible for practical reasons. Therefore, personal measurement errors could have influenced the results. However, the measurement therapists also participated in a reliability study and showing excellent reliability,[32] indicating that they were likely sufficiently trained and consistent.

Although we already included more participants than most other studies on this topic, more participants are needed to be able to better differentiate which variables differs most between groups. Therefore, the results must be interpreted with some caution. A more global collaboration is needed to produce studies with a larger sample size.

## Conclusion

In children with USCP, muscle weakness in both upper extremities occurs. When uni- or bimanual ability limitations are present, investigation of muscle strength in the non-affected UE should be part of the assessment. Future research should focus on whether particular characteristics related to children with USCP can explain these differences in muscle strength and whether and where muscle weakness is present in the UE in adolescents with USCP.

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## 'Conflict of Interest: none declared'.

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# Chapter 6

# Can we measure changes in upper extremity strength in all children with unilateral spastic cerebral palsy? A perspective

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## Abstract

**Purpose:** The objective of this perspective paper is to provide a better insight into how useable the hand-held dynamometer (HHD) and the E-link (hand-dynamometer measuring grip- and pinch strength) are to detect changes in upper extremity (UE) muscle strength in children with unilateral spastic cerebral palsy (USCP).

**Method:** By comparing the ratio of muscle strength values/SDC values (12–520% for the affected UE, 9–204% for the non-affected UE) of the children with USCP, with published results of UE strength training (10–77%), we examined whether it is possible for children with USCP to show a gain in UE muscle strength that is at least equal to the SDC. An expert-based minimally important change was also determined (15% for the affected UE, 20% for the non-affected UE) and compared to the ratio of muscle strength values/SDC values.

**Results:** In clinical practice, it is possible for most children with USCP to measure clinically important changes in muscle strength in the affected UE. Only in the strongest children (and some measurements), the clinically important changes can be considered "real" changes.

**Conclusions:** Due to the high SDC values, only in children with USCP having a high baseline level of muscle strength, the clinically important changes can be considered "real" changes. Great caution in interpretation of the change score is recommended. For the majority of children with USCP the important changes cannot be distinguished from measurement error.

# Introduction

Muscle weakness of the affected upper extremity (UE) is one of the characteristics of a child with unilateral spastic cerebral palsy (USCP).[1–3] This muscle weakness may lead to limitations in the performance of daily activities. For example, grip strength is an important predictor for use of the affected arm in bimanual performance in children with USCP.[4,5] Although it was long assumed that muscle strength loss only occurred in the affected UE, two recent studies stated that the less affected UE also reported less muscle strength compared to typically developing children.[6,7] Also, reduced performance of the less affected UE compared to typically developing children, as measured with the Jebsen-Taylor Test of Hand Function, a computerized version of the Peg Moving Task, the Box and Blocks Test and the Tyneside Pegboard Test, has been reported.[7–10] Therefore, it seems important that when uni- or bimanual ability limitations are present, the muscle strength of both the affected and less affected UE are assessed.

Various methods exist to measure UE muscle strength in children with USCP. With cross-sectional measurements it is possible to investigate whether muscle weakness is present. Longitudinal measurements can be used to assess whether changes in muscle strength are the result of UE muscle strength training or due to natural development of the child.

Ideally, the clinimetric properties of the measurement instrument have been well researched and found to be good to excellent. The Consensus-Based Standard for the Selection of Health Measurement Instruments (COSMIN) taxonomy distinguishes three quality domains within clinimetrics: reliability, validity and responsiveness. Each domain contains one or more clinimetric properties.[11] To be sure that the difference between two measurements for the same person is not due to measurement error, the smallest detectable change (SDC: reliability domain) is considered the most important clinimetric property. If the change in score due to an intervention or to the child's development is equal to or more than the SDC, it can be concluded that there is a real (statistically significant) change of the measured variable, as there is less than 5% probability that the difference between scores is due to measurement error. In order to know whether a change score is also clinically important, the minimally important change (MIC: responsiveness domain) is considered the most important value. The MIC is the smallest change score in the construct to be measured that patients, clinicians or relevant others perceive as important.[11]

When the MIC is higher than or equal to the SDC, the measurement error is sufficiently small to detect the MIC. However, when the MIC is lower than the SDC, values lying between the MIC and SDC are considered important but they cannot be distinguished from measurement error.[11] (see Figure 1)

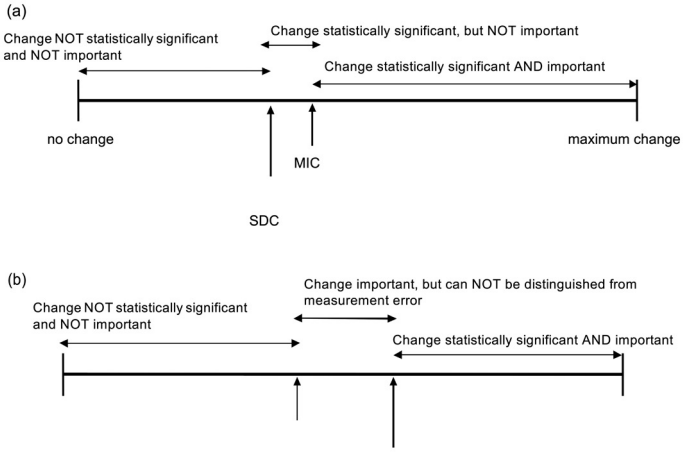


**Figure 1.** Minimal Important Change (MIC) versus Smallest Detectable Change (SDC).
*(a) Interpretation of change when MIC is larger than SDC. (b)Interpretation of change when MIC is smaller than SDC. Reprinted from Journal of Clinical Epidemiology, Vol 62, issue 10, Caroline B. Terwee, Leo D. Roorda, Dirk L. Knol, Michiel R. De Boer, Henrica C.W. De Vet, Linking measurement error to minimal important change of patient-reported outcomes, 2009, with permission from Elsevier.*

For most instruments used to measure UE muscle strength in daily practice the clinimetric properties have been examined only partly.[12] In a recent study in children with USCP, the reliability and the SDC of two frequently used measurement instruments – the Microfet2 hand-held dynamometer (HHD: Hoggan Scientific LLC, Salt Lake City, UT, USA) and the E-link grip/pinch dynamometer (Biometrics Ltd, Gwent, UK) – were studied.[13] Isometric arm strength was measured with the HHD and isometric grip and pinch strength with the E-link. It was concluded that both instruments can be used to measure UE muscle strength cross-sectionally. However, it was not clear whether both instruments can be used longitudinally because no information was available on whether children with USCP can achieve a gain in muscle strength above the SDC value. Therefore, caution is advised in the interpretation of changes in UE muscle strength. Additional research on the effects of UE strengthening in children with USCP is recommended before drawing firm conclusions about the usability of these instruments in clinical practice. Furthermore, because information regarding the MIC is lacking, it is not clear whether the HHD and the E-link are able to detect clinically important changes in muscle strength.

The objective of this perspective paper is to provide a better insight into how useable the HHD and the E-link are to detect changes in UE muscle strength in children with USCP. Therefore, we examined whether it is achievable for children with USCP to show a gain in UE muscle strength that is at least equal to the SDC value. As determining the MIC is already a study in itself and there is no consensus on the best method to determine the MIC[11], we chose to ask a mix of clinicians with different professions, but all with extensive experience in muscle strength training in children with USCP, one simple question: "What percentage change in muscle strength is clinically important in your opinion", to determine an expert-based MIC. This expert-based MIC can provide insight into the extent to which the HHD and the E-link are able to measure clinically important changes in UE muscle strength. Finally, the expert-based MIC and SDC will be compared to get an impression of how to interpret changes in UE muscle strength in children with USCP.

## Materials and methods

Four steps were performed to gain better insight into the usability of the HHD and the E-link to measure changes in UE muscle strength for both the affected and less affected UE.

In the first step, the ratio of muscle strength values/SDC values was calculated. UE muscle strength data from the children and the SDC values of a recently published study by Dekkers et al.[13] were used. (See supplementary material for more information about for the standardized protocol with detailed descriptions of all procedures and measurements).

For the HHD-measurements, a total of 72 children (45 boys, 27 girls; mean age 9 years, 4 months, SD 1 year 9 months) with USCP participated. For the E-link measurements, a total of 86 children (53 boys, 33 girls; mean age 9 years, 3 months, SD 1 year 8 months) with USCP participated. See table 1 for details of the participants.

**Table 1.** Participants characteristics

| **Measurement** | **Characteristics** | |
|---|---|---|
| *Isometric Arm Strength measurements (HHD)* | Age group | Gender mix |
| n = 72. 45♂. 27♀.<br>Mean age 9.4 years. SD 1.9 years.<br>45 right side affected. 27 left side affected<br>MACS level; I: n = 23. II: n = 42. III: n = 7. | Age 7. n = 17 | 9♂. 8♀ |
| | Age 8. n = 12 | 5♂. 7♀ |
| | Age 9. n = 12 | 9♂. 3♀ |
| | Age 10. n = 8 | 5♂. 3♀ |
| | Age 11. n = 7 | 5♂. 2♀ |
| | Age 12. n = 16 | 12♂. 4♀ |
| *Isometric Grip and Pinch Strength (E-link system)* | Age group | Gender mix |
| n = 86. 53♂. 33♀.<br>Mean age 9.3 years. SD 1.8 years.<br>51 right side affected. 35 left side affected<br>MACS level; I: n = 29. II: n = 47. III: n = 10. | Age 7. n = 19 | 10♂. 9♀ |
| | Age 8. n = 14 | 6♂. 8♀ |
| | Age 9. n = 17 | 11♂. 6♀ |
| | Age 10. n = 9 | 5♂. 4♀ |
| | Age 11. n = 11 | 9♂. 2♀ |
| | Age 12. n = 16 | 12♂. 4♀ |

*HHD = hand-held dynamometer; ♂ = male; ♀ = female; SD = standard deviation; n = population size; MACS = Manual Ability Classification System*

Data from the following measurements were used for both the affected and less affected UE: isometric wrist extension with flexed and extended fingers, wrist flexion, elbow flexion and elbow extension (all measured with the HHD); and isometric grip and pinch strength (measured with the E-link). See table 2 for the more specific information about the mean, corresponding standard deviations and the minimum and maximum values of the measurements.

**Table 2.** Mean, standard deviation, minimum and maximum values of the measurements

| Movement | n | Mean | Standard Deviation | Minimum value | Maximum value |
|---|---|---|---|---|---|
| *Isometric Arm Strength measurements (HHD)* | | | | | |
| Wrist-extension AH | 72 | 25.9N | 18.3N | 5.0N | 93.1N |
| Wrist-extension (flexion fingers) AH | 72 | 29.7N | 18.5N | 9.6N | 98.7N |
| Wrist-flexion AH | 72 | 34.2N | 14.1N | 5.1N | 82.0N |
| Elbow-flexion AH | 72 | 70.8N | 28.4N | 22.7N | 144.0N |
| Elbow-extension AH | 72 | 63.6N | 23.0N | 27.0N | 125.4N |
| Wrist-extension NAH | 72 | 55.2N | 20.4N | 17.3N | 100.7N |
| Wrist-extension (flexion fingers) NAH | 72 | 60.1N | 25.4N | 18.3N | 139.8N |
| Wrist-flexion NAH | 72 | 63.1N | 25.1N | 21.7N | 147.3N |
| Elbow-flexion NAH | 72 | 102.0N | 37.8N | 34.0N | 217.7N |
| Elbow-extension NAH | 72 | 93.0N | 37.3N | 38.7N | 257.7N |
| *Isometric Grip and Pinch Strength (E-link system)* | | | | | |
| Grip strength AH | 86 | 5.5kg | 4.7kg | 0.7kg | 26.2kg |
| Pinch strength AH | 86 | 1.8kg | 1.4kg | 0.3kg | 8.6kg |
| Grip strength AH | 86 | 13.3kg | 6.6kg | 2.5kg | 38.2kg |
| Pinch strength AH | 86 | 3.8kg | 1.9kg | 0.8kg | 15.2kg |

*AH = affected hand; HHD = hand held dynamometer; kg = kilograms; n = population size; N = Newton; NAH = non-affected hand; SD = standard deviation;*



Based on the huge variability in the strength values for the children in this study (e.g. wrist extension range was 5–93.07 N; see Table 2), we identified, for each measurement separately and regardless of gender or age, the child with the lowest muscle strength, children with muscle strength values at the first, second and third quartiles and the child with the highest muscle strength.

Next, the percentage change (gain) in muscle strength needed to ensure that the change is similar to the corresponding SDC was calculated for these five children. For this, the ratio of muscle strength values to the corresponding SDC values was determined using the following equation: muscle strength values/SDC values = 1/(muscle strength/SDC) × 100.

The second step was to identify the reported effects of strength training studies of the UE in children with USCP using the HHD and the E-link. For each of these studies we calculated the percentage change in UE muscle strength. For quick clinical searches, Google Scholar returns twice as many relevant articles as PubMed,[14] therefore a Google Scholar search for the latest reviews and clinical trials on UE strength interventions was performed in January 2019. Papers were included if they met the following inclusion criteria: 1) children with USCP, 2) measuring UE muscle strength, and 3) describing an intervention or describing a (significant) change in muscle strength. Because we searched for the latest reviews and clinical trials, single case studies were excluded.

The following search string was used: "Cerebral palsy" AND Strengthening AND ("handheld dynamometer" OR "hand dynamometer" OR "e-link") AND (arm OR hand OR grip OR "upper limb" OR "upper extremity" OR "arm-strength" OR "hand-strength" OR "grip-strength"). The filter "publication date" was set at 5 years. Titles and abstracts were screened only by one author (KD) for relevance, as this was not meant to be a full systematic review.

After selection, the remaining papers were fully read and the percentage gains in UE muscle strength were derived from these papers. If in the paper the muscle strength change value was not displayed as a percentage, the percentage was calculated by dividing the change-value by the value before the training, and then multiply by hundred to get the change percentage.

Third, as determining the MIC is already a study in itself, we identified an expert-based MIC, for UE muscle strength changes in children with USCP, for the affected and the less affected UE separately. For patient-reported outcomes the MIC should be considered from the perspective of the patient, whereas for non-patient-reported outcomes a clinician's perspective of which change is minimally important could be more relevant.[11] As muscle strength is a non-patient-reported outcome, we chose the clinicians' perspective of which percentage change in muscle strength, as a result of an intervention, they consider to be clinically important.

Unfortunately, there is no consensus on the best method to determine the MIC.[11] Within our (Dutch) national network of clinicians working in the field of USCP, we invited clinicians to cooperate in determining the expert-based MIC. We determined an expert-based MIC by asking a total of 32 paediatric clinicians (physicians (n = 4) , physiotherapists (n = 11) and occupational therapists (n = 17)) with at least three years of extensive experience in UE strength training in children with USCP, working in five different Dutch rehabilitation centres, what percentage change in UE muscle strength they regard as clinically important.

All participants were contacted by email to answer the following question: What percentage change in UE muscle strength after a strength training programme is, according to your opinion, clinically important? Thereafter, we calculated mean values for the affected as well as for the less affected UE.

Finally, per measurement, we compared the expert-based MIC and SDC (ratio of muscle strength values/SDC values).

# Results

## Ratio of muscle strength values/SDC values

### *Affected UE*

Based on the SDC values per measurement, the child with the lowest muscle strength needs to gain from 117% (for elbow extension) up to 520% (for grip strength) in muscle strength to show a progression equal to the corresponding SDC value. The child with the highest muscle strength needs to gain from 12% (for pinch strength) up to 23% (for wrist flexion) in muscle strength to show a progression equal to the corresponding SDC value. See Table 3 for details and for values at the first, second and third quartiles.

### *Less affected UE*

The child with the lowest muscle strength must show a gain in muscle strength from 67% (for elbow extension) up to 204% (for grip strength) in order to reach the corresponding SDC value. The child with the highest muscle strength needs to gain from 9% (for pinch strength) up to 23% (for wrist extension) in muscle strength to reach the corresponding SDC value. See Table 3 for details and for values at the first, second and third quartiles.

**Table 3.** Ratio SDC-values/muscle strength values

| | SDC | Highest muscle strength value | Percent gain in muscle strength | 3rd quartile | Percent gain in muscle strength | 2nd quartile | Percent gain in muscle strength | 1st quartile | Percent gain in muscle strength | Lowest muscle strength value | Percent gain in muscle strength |
|---|---|---|---|---|---|---|---|---|---|---|---|
| Wrist-extension AH (Newton) | 13.79N | 93.07 | 15% | 35.67N | 39% | 22.33N | 62% | 16.73N | 82% | 5.00N | 276% |
| Wrist-extension (flexion fingers) AH | 17.51N | 98.73 | 18% | 36.25N | 48% | 25.08N | 70% | 17.58N | 100% | 9.57N | 183% |
| Wrist-flexion AH | 18.96N | 81.97 | 23% | 43.17N | 44% | 31.83N | 60% | 23.57N | 80% | 5.13N | 369% |
| Elbow-flexion AH | 30.45N | 144.00N | 21% | 87.00N | 35% | 67.67N | 45% | 52.42N | 58% | 22.67N | 134% |
| Elbow-extension AH | 31.49N | 125.40N | 25% | 79.53N | 40% | 56.33N | 56% | 48.18N | 65% | 27.00N | 117% |
| Grip strength AH | 3.47kg | 26.23kg | 13% | 7.40kg | 47% | 3.93kg | 88% | 2.52kg | 138% | 0.67kg | 520% |
| Pinch strength AH | 1.03kg | 8.57kg | 12% | 2.32kg | 44% | 1.40kg | 74% | 0.97kg | 107% | 0.30kg | 343% |
| | | | | | | | | | | | |
| Wrist-extension NAH | 23.22N | 100.67N | 23% | 69.83N | 33% | 56.00N | 41% | 39.07N | 59% | 17.33N | 113% |
| Wrist-extension (flexion fingers) NAH | 30.89N | 139.80N | 22% | 77.25N | 40% | 54.67N | 57% | 40.90N | 76% | 18.33N | 168% |
| Wrist-flexion NAH | 24.54N | 147.33N | 17% | 75.75N | 32% | 59.50N | 41% | 44.27N | 55% | 21.67N | 113% |
| Elbow-flexion NAH | 30.81N | 217.67N | 14% | 116.08N | 27% | 98.83N | 31% | 74.17N | 42% | 34.00N | 91% |
| Elbow-extension NAH | 25.93N | 257.67N | 10% | 104.86N | 25% | 89.00N | 29% | 69.42N | 37% | 38.67N | 67% |
| Grip strength NAH | 5.02kg | 38.17kg | 13% | 17.03kg | 29% | 11.75kg | 43% | 9.01kg | 56% | 2.47kg | 204% |
| Pinch strength NAH | 1.41kg | 15.17kg | 9% | 4.79kg | 29% | 3.53kg | 40% | 2.77kg | 51% | 0.80kg | 176% |

*AH = affected hand; kg = kilograms; N = Newton; NAH = non-affected hand; SDC = smallest detectable change; 3rd quartile: child a with muscle strength value at 75% of the data set; 2nd quartile: child a with muscle strength value at 50% of the data set; 1st quartile: child a with muscle strength value at 25% of the data set*

## Percentage change in muscle strength reported in studies

The Google Scholar search resulted in a total of 274 hits. After screening titles and abstracts, 245 studies were excluded from full text reading because they did not meet the inclusion criteria. After reading the remaining 29 full-text articles, another 24 articles were also excluded for not meeting the inclusion criteria; In fourteen papers UE muscle strength was not measured, in six papers no interventions were described (and no change reported) and in four papers, single case studies were described.

This left five papers; one of these was a narrative review and was excluded because the two relevant studies described were included in the other four papers.

In the four remaining articles a variety of therapeutic interventions, populations and results were described. In three articles an HHD was used to measure the progression in muscle strength and in the other article the E-link system was used to measure the grip and pinch strength: three articles reported an overall gain in muscle strength of the affected UE of 10–20%[15–17] and the other article reported an increase of 35–77%.[18] There were no results describing the effect of muscle strength training on the less affected UE. See Table 4 for a summary of the results.

## Changes in score that are clinically important: expert-based MIC

According to the clinicians, a mean change of 15% (SD, 15%; range, 2–50%; median, 10%) was considered clinically important for the affected UE. For the less affected UE, a mean change of 20% (SD, 17%; range, 2–60%; median, 15%) was considered to be clinically important.

## Comparison of the expert-based MIC and SDC

With regard to the affected UE, in three measurements for the strongest child (wrist extension and grip and pinch strength) the ratio of muscle strength values/SDC values was lower or equal to the expert-based MIC value. For the other measurements, and in all measurements for the other children, this ratio exceeded the expert-based MIC value.

For the less affected UE, the ratio of muscle strength values/SDC values for the child with the greatest muscle strength was lower than the expert-based MIC in almost all measurements (except wrist extension with flexed and extended fingers). For all other measurements, and in all measurements for the other children, the ratio of muscle strength values/SDC values exceeded the expert-based MIC value. See Table 5 for the detailed results.

**Table 4.** Percent change in muscle strength that has been reported in studies

| Authors | Type of study | Population | Type of training | Measure-ment instrument | Joint/muscle | Pre test score | Post test score | Difference | Percentage changes in strength |
|---|---|---|---|---|---|---|---|---|---|
| Brauers et al. (2017) | Clinical trial | N=23. (12 boys and 11 girls) with a mean age of 14 y and 8 m (SD = 1 y 11 m). Hemiplegia was left-sided in 13 subjects and right-sided in 10 subjects. According to the MACS levels. 3 subjects (13%) were classified as level I. 7 subjects (30.4%) as level II. and 13 subjects (56.6%) as level III. | The children participated in a H-CIMT model organized in a therapeutic summer-camp. | E-Link H500 hand-kit (Biometrics; UK). | pinch strength AH | 2.88kg | 3.21kg | 0.33kg | 11.4% |
| Crowner et al. (2008) | Prospec-tive Clinical trial | N= 34. 19 boys. 15 girls. Mean age 7y 7 m (range 4-17y). 27 diplegic. 7 hemiplegic. GMFCS I=14. II=11. III=5. IV=4 | botulinum toxin type a | Baseline Hydraulic Hand-Held | Grip Strength all patients | 7.4N | 8.4N | 1N | 13.5% |
| | | | | Dynamometer (FEI. Irvington. NY) | Grip Strength low dosage | 9.2N | 10.9N | 1.7N | 18.5% |
| Megan Louise Auld & Leanne Marie Johnston (2014) | pre–post interven-tion study | N=10, 8-15y, 6♂, CP. GMFCS I= 6; II = 4; five diplegia; five hemiplegia) | Strength and balance training intervention were provided via a stat ion-based "gym group" held in a local community setting with low cost. portable equipment. Children attended one hour sessions once per week for eight weeks | Lafayette dynamometer (Lafayette Instrument Company. Lafayette. IN | elbow flexion AH | 20-Feb mmHg | 24-Mar mmHg | 4.1 mmHg | 20% |
| Vaz et al. (2008) | Clinical trail | 9 CP. 7-11 years. Spastic hemiplegia 1 MACS 1. 8 MACS 2 | Concentric training in the extended wrist range for flexors and extensors | Microfet-2 dynamometer (Hoggan | Wrist exten-sors AH. wrist extended 30° | 7.37N | 13.11N | 5.73N | 77.4% |
| | | | Manual resistance and added neuro muscular electro stimulation | Health Indus-tries. West Jordan. USA) | Wrist exten-sors AH. wrist at neutral | 19.23N | 25.97N | 6.74N | 35.0% |
| | | | | | Wrist flexors AH. wrist extended 30° | 31.02N | 42.01N | 10.98N | 34.7% |

*AH = affected hand; CP = Cerebral Palsy; HHD = hand held dynamometer; GMFCS; gross motor function classification system; H-CIMT = hybrid-constraint induced movement therapy; kg = kilograms; m = months; MACS = Manual Ability Classification; N = population size; SD = standard deviation; y = years, ♂ = male; ♀ = female.*

**Table 5.** comparison of the expert-based MIC-values and the SDC-values

| Movement | expert-based MIC | Max. muscle strength value; Ratio SDC-values/ muscle strength values | 3rd quartile muscle strength value; Ratio SDC-values/ muscle strength values | 2nd quartile muscle strength value; Ratio SDC-values/ muscle strength values | 1st quartile muscle strength value; Ratio SDC-values/ muscle strength values | Min. muscle strength; Ratio SDC-values/ muscle strength values |
|---|---|---|---|---|---|---|
| Wrist-extension AH | 15% | 15%* | 39% | 62% | 82% | 276% |
| Wrist-extension (flexion fingers) AH | 15% | 18% | 48% | 70% | 100% | 183% |
| Wrist-flexion AH | 15% | 23% | 44% | 60% | 80% | 369% |
| Elbow-flexion AH | 15% | 21% | 35% | 45% | 58% | 134% |
| Elbow-extension AH | 15% | 25% | 40% | 56% | 65% | 117% |
| Grip strength AH | 15% | 13%* | 47% | 88% | 138% | 520% |
| Pinch strength AH | 15% | 12%* | 44% | 74% | 107% | 343% |
| | | | | | | |
| Wrist-extension NAH | 20% | 23% | 33% | 41% | 59% | 113% |
| Wrist-extension (flexion fingers) NAH | 20% | 22% | 40% | 57% | 76% | 168% |
| Wrist-flexion NAH | 20% | 17%* | 32% | 41% | 55% | 113% |
| Elbow-flexion NAH | 20% | 14%* | 27% | 31% | 42% | 91% |
| Elbow-extension NAH | 20% | 10%* | 25% | 29% | 37% | 67% |
| Grip strength NAH | 20% | 13%* | 29% | 43% | 56% | 204% |
| Pinch strength NAH | 20% | 9%* | 29% | 40% | 51% | 176% |

*Red = minimal important change < smallest detectable change, green* = minimal important change > smallest detectable change.*

# Discussion

The objective of this perspective paper was to provide a better insight into how useable the HHD and the E-link are to detect changes in UE muscle strength over time in individual children with USCP.

## Usability of the measurement instruments: SDC

For the affected UE, three studies reported an average gain in muscle strength of 10–20%. This means that the difference in muscle strength, in three measurements for the strongest child, exceeded the SDC. It can, therefore, be concluded, that this is a real gain and not a gain due to measurement error. In all other measurements for this child and in all measurements in all the other children, there will be a higher probability that the measured gain in muscle strength is due to measurement error. Taking the results of the study by Vaz et al. into account (overall 35–77% gain in muscle strength after training), it could be possible that the child with muscle strength values at the third quartile is able to show a larger gain in muscle strength than the SDC. The child with muscle strength values at the second quartile had change values between 45% and 88%, therefore it can be concluded that for a large proportion of children with USCP a gain in muscle strength above the SDC of these measurements does not seem feasible. Hence, in many children with USCP there will be a high probability that measurement error is responsible for the change in muscle strength. Caution in interpreting the change score is thus recommended, especially in children with low levels of muscle strength. Because, the lower the muscle strength, the higher the probability of measurement error.

Unfortunately, the literature search revealed no information about a possible gain in muscle strength for the less affected UE. Although several studies identified some muscle weakness in the less affected UE [6,7], a comparison with the possible gain in muscle strength of the affected UE or the possible gain in muscle strength of typically developing children cannot be made. Considering the change values of the less affected UE (Table 3), the child with the lowest muscle strength needs a 67–204% gain in muscle strength to have a change value equal to the SDC value. It can, therefore, be concluded that there is a high chance for, at least several, children not to have a gain in muscle strength above measurement error; caution in interpreting the change score of muscle strength for the less affected UE is, therefore, recommended, especially in children with low levels of muscle strength. However, with current knowledge, it cannot be determined how many of the total sample children can meet the change value. More research on the muscle strength gain possibilities for the less affected UE is needed.

Also in other studies reporting on SDC values of muscle strength measurements in children with USCP, generally relatively high SDC values are found. Willemse et al. reported high SDC values in lower extremity strength measurements (>20.6% for knee flexors and >34.8% for ankle plantar flexors measured with the HHD) in children with cerebral palsy.[19] In this study, it was concluded that because previous strength training studies had reported lower extremity muscle strength increases of 11–74%, the HHD often will be insufficiently sensitive to detect individual strength gains.[19] De Groot et al. reported a 25–45% SDC value in isometric knee flexor and extensor muscle strength (measured with the Biodex system) for both the affected and less affected extremity.[20] Van Vulpen et al. reported a 40–128% SDC value for the standing heel-rise test in children aged 3–5 years and a 23–48% SDC

value in children aged 6–10 years for both the affected and less affected extremity.[21] However, they concluded that they found acceptable SDC values (9–30%) for the isometric calf muscle strength test (measured with the HHD) for both the affected and less affected extremity. It should be noticed, however, that isometric muscle strength for the plantar-/dorsiflexion muscles measured with a HHD underestimates the force capacity of children with CP during walking[22]

Taking the results of these other studies into account, for the majority of the children with USCP a change score (measured with the HHD) higher than the SDC value does not seem feasible.

## Usability of the measurement instruments: Clinically important changes

For the affected UE, the expert-based MIC as defined by the 32 clinicians in our study corresponds with the average gain in muscle strength after training reported in the literature. Based on the similarity of these results, it can be concluded that it seems possible for most children with USCP to achieve clinically important changes in muscle strength in the affected UE.

For the less affected UE, again we find that there is no information on possible gains in muscle strength available in the literature. As a result, we cannot firmly state that it is possible to measure clinically important changes in muscle strength for the less affected UE. However, because of the low value of the expert-based MIC, it is plausible that the HHD and E-link are able to measure clinically important changes in muscle strength for the less affected UE.

## Usability of the measurement instruments: SDC versus expert-based MIC

For the affected UE, only in three measurements (wrist extension and grip and pinch strength) for the strongest child, the ratio of muscle strength values/SDC values is sufficiently small to detect the expert-based MIC. For the other four children, the expert-based MIC values for all measurements are lower than the ratio of muscle strength values/SDC values. Therefore, it can be concluded that for the majority of the children with USCP it is possible to measure changes that are clinically important but they cannot be distinguished from measurement error.

The results for the less affected UE showed that for the child with the most muscle strength the ratio of muscle strength values/SDC values for five out of seven measurements is lower than the expert-based MIC. However, for the other four children the ratio of muscle strength values/ SDC values exceeds the expert-based MIC values. Thus, it can be concluded that in the majority of the children with USCP, changes in strength of the less affected UE, although important, cannot be distinguished from measurement error. Only in some children (and in some measurements) clinically important changes in muscle strength can be considered "real" changes (statistically significant).

## Usability of the measurement instruments: clinical use and implementation

To reduce the chance of measurement error, it is recommended to use a standardized protocol, to perform the measurements by the same assessor and taking the mean of multiple measurements. This would minimize the likelihood that the differences in muscle strength are caused by differences in initial posture, instrument placement (HHD) and handling (E-link).

It is important to note that as a child gets older, the length of the arms increases. Then, the outcome of the HHD (expressed as Nm (= Newton * lever arm)) will increase. Therefore, a better approach to measure changes in muscle strength over a long period of time (months or years) is to express strength in torque (Nm, including the length of the lever arm). However, the additional measurement to assess the length of the lever arm, could lead to additional measurement errors, and therefore to an increasing SDC-value.

## Limitations of this study

The dataset for this paper is for a population with an age range of 7–12 years and therefore the usability of these measurement instruments cannot be inferred for other age groups.

We only determined the ratio of muscle strength values/SDC values for the children with the lowest, highest and first/second/third quartile muscle strength values, therefore it is not possible to calculate exact values for the percentage of children in which these measurement instruments are useable. Because of the large sample size and distribution of important child characteristics, in our opinion the dataset contains a representative sample of the total population. However, small deviations in the distribution of muscle strength values and minimum/maximum muscle strength compared to the total population could have occurred.

The literature search was restricted to one academic search engine and with limited keywords, therefore relevant articles may have been missed. The goal of the literature search was to retrieve global information on the possible percentage gain in muscle strength.

Although the MIC is determined for several measurement instruments for children with cerebral palsy, none of them measure muscle strength. To be able to compare the SDC with the minimal important change, we had to determine a value which represents the minimal important change. Because this was not the goal of this study, we did not use a full Delphi to determine the minimal important change value. We chose to determine an expert-based MIC. For determining this expert-based MIC, a mix of clinicians from different professions were asked about their perspective on which percentage change in muscle strength is clinically important. Although only clinicians with extensive experience in UE training for children with USCP participated, the responses provided by the 32 therapists varied considerably (2–50%). A future study using a full Delphi design, i.e. an iterative, multistep process to reach consensus, and a more homogeneous population may reveal a MIC that deviates from ours.

Because of the high number of participants in this survey, we chose to use the mean of the answers instead of the median.[23] If we had taken the median, which is lower compared to the mean, this would have resulted in more children with USCP for whom it is possible to achieve clinically

important changes in muscle strength in the affected UE. However, this would not have affected our results significantly because for most children with USCP it is already possible to achieve clinically important changes in muscle strength.

We chose to determine the expert-based MIC by asking the clinicians, therefore clinically important change from the patient perspective is not clear. However, because the instruments measure muscle strength at the function level, it could be hard for the patient to estimate how much gain in muscle strength is needed to achieve goals at the activity level. A satisfaction assessment of the results of treatment by the child/parents could be added to obtain a better impression of whether the results of treatment are clinically important for the patient.

The current method to determine the SDC value uses measures of multiple children and gives the SDC as an absolute value. However, the SDC value is intended to interpret change scores in individual patients. As the muscle strength values of the participants vary greatly, the SDC value is harder to meet for a child with low values compared to a child with higher values. For example, if the SDC is 10 N, a child with a muscle strength value of 100 N has to gain 10% to meet the SDC value, whereas a child with a muscle strength of 10 N has to gain 100% to meet the SDC value. It is therefore arguable whether the current method of determining the SDC value is useable for this kind of outcome measure. The SDC value could be more useable/valuable if it were to be determined as a percentage. Alternatively, an SDC value determined for every child individually, by taking multiple measurements before starting the intervention, would be of great added value. More research about alternative methods to determine the SDC value in field-based (physiotherapeutic) tests is needed.

## Summary: Implications for physical therapists

In clinical practice, for most children with USCP it is possible to measure clinically important changes in muscle strength in the affected UE by means of the HHD and E-link, However, due to the high SDC values, only in those with a high baseline level of muscle strength, the clinically important changes can also be considered "real" changes, i.e. higher than measurement error. Hence, it is only possible to achieve a gain in muscle strength that is considered "real" for a small proportion of the children with USCP. Thus, great caution in the interpretation of the change score is recommended.

Regarding the less affected UE, only in some children (and in some measurements) the clinically important changes can also be considered "real" changes. For the majority of the children with USCP, the important changes cannot be distinguished from measurement error. Because of the lack of information on the muscle strength gain possibilities of the less affected UE, no firm conclusions can be drawn on whether it is possible to measure clinically important and/or statistically significant changes within one child.

To reduce the chance of measurement error, it is recommended to use a standardized protocol, to perform the measurements by the same assessor and to measure multiple times within one measurement moment, taking the mean value of the measurements. This would minimize the likelihood that the differences in muscle strength are caused by differences in initial posture, differences in placing of the measurement instrument (HHD) or differences in handling the measurement instrument (E-link).

A better approach to measure changes in muscle strength over a long period may be to include the lever arm (i.e. muscle strength measured as torque expressed in Nm). However, the additional measurement may lead to increased SDC-values.

As there is a high probability that the measured increase of strength is caused by measurement error, it is recommended that additional measurement instruments be used. To determine changes in (functional) muscle strength, functional strength measurement (FSM)[24] and the cup-and box task seem to be good alternatives.[25] As the goal of an intervention should be related to the activity in which the child wants to improve, it is also recommended to use instruments that measure changes in activity level, such as Goal Attainment Scaling (GAS),[26] ABILHAND-Kids,[27] Assisting Hand Assessment,[28] Melbourne Assessment-v2,[29] Quality of Upper Extremity Skills Test[30] and Shriners Hospital Upper Extremity Evaluation (SHUEE).[31] When multiple measurement instruments show gains between the measurements, there may be a higher probability that the intervention has a "real" positive result.

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## Declaration of interest statement:

The authors report no conflicts of interest.

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# Chapter 7

# General Discussion

Muscle weakness of the affected upper extremity (UE) is one of the main characteristics of a child with unilateral spastic cerebral palsy (USCP).[1–3] This muscle weakness appears to be one of the most important causes of impairment in performance of UE activities.[4–6] As a result, the primary goal of current therapy programmes is muscle strength training of the affected UE to facilitate performance during the numerous daily (uni- and bimanual) tasks.[7–9]

Measuring muscle strength is a common activity in daily practice for most clinicians working with children with USCP in order to determine whether muscle strength weakness is present (discriminative) or whether muscle strength training has been effective (evaluative).

To be able to measure changes in muscle strength, one needs to know the degree to which variations in the measurement results between repeated measurements occur. This so-called measurement error can arise from several sources: the measurement instrument itself, the examiner(s) performing the measurement, the patient undergoing the measurement and the circumstances under which the measurement is performed.[10]

Four out of five studies presented in this thesis focus on the clinimetric properties of various UE strength measuring instruments for children with USCP and on the interpretation of their outcomes, with the intention of clarifying how useful these measurement instruments actually are in clinical practice. The fifth study focuses on the muscle strength of the non-affected UE in children with USCP because it is not clear whether muscle weakness is a unilateral problem or whether the non-affected UE is also affected.

## Aim

The primary aim of our research was to determine the clinimetric properties of four measurement instruments used in clinical practice to measure UE muscle strength in children with USCP. In addition, the aim was to appraise their overall usability for discriminative and evaluative purposes in children with USCP. The final aim of our research was to determine whether muscle strength problems are also present in the 'non-affected UE' of children with USCP.

## Discussion outline

The outline of this thesis is divided into three main parts. First, the main findings will be discussed and contrasted to other findings reported in the international literature. Second, the 'lessons learned' about the methods used and the designs of our studies will be summarized and discussed. Third, there will be discussion on the current method of calculating the smallest detectable change (SDC) and whether this is suitable for muscle strength measurement instruments used in a (heterogeneous) rehabilitation population, as Chapters 3, 4 and 6 all show that the measurement instruments studied have a large SDC.

The discussion ends with two paragraphs on clinical implications and future research.

## 1. Interpretation of the outcomes of the studies

The overall conclusion from our systematic review (Chapter 2) was that research on UE strength measurements in children with CP needs to be improved, both in the number of subjects included and scientific quality. For example, we found that UE measurements with the hand-held dynamometer (HHD) were evaluated in two studies with a total of only 13 participants. In comparison, a systematic review from 2016 on the clinimetric properties of the HHD, used for measuring lower extremity muscle strength in children with CP, revealed seven studies with a total of 127 participants.[11] In contrast to the studies reported in our review, the primary aim of all studies in the review on the use of the HHD in the lower extremities was to determine reliability and validity.

After our systematic review, one other systematic review has been published in which UE muscle strength measurement instruments were researched in children with dyskinetic CP.[12] In this review only one paper was included in which a UE muscle strength measurement instrument was described (Biodex). However, the included paper did not report on clinimetric properties.[13] As far as we know, no other papers have been published in which the clinimetric properties of UE measurement instruments for children with USCP are described. Therefore, the conclusion of our systematic review, that only a few measurement instruments used for discriminative purposes have been researched (in studies of low methodological quality), still appears to be valid.

In Chapter 3, the reliability of the HDD and the Biometrics E-link system are described. Reviewing the literature up to May 2020, no other studies on the clinimetric properties of UE muscle strength measurement instruments for children with USCP have been published, making the HHD and E-link still the best researched measurement instruments for UE muscle strength in children with USCP.

Regarding the HHD and E-link, multiple measurement protocols are available. Although our protocol was developed by experts in paediatric rehabilitation, it would be useful to compare the results of our protocol with the results of other protocols. In this way, it is possible to determine which method, initial position of child and examiner and handling of the measurement instrument are most reliable. To our knowledge, our protocol is the only one that has been researched for its clinimetric properties in children with USCP, therefore we recommend our protocol for use in research and clinical practice.

In most manual activities of daily life (e.g. carrying or moving a heavy box) it is relevant not only to measure the muscle strength but also the ability to maintain and regulate that strength during the performance of a task. This ability is called functional strength and it needs to be measured during the performance of a specific task. Two types of functional muscle strength tests have been developed: the 'Cup task' for determining maximal functional unimanual UE strength and the 'Box task' for determining maximal functional bimanual UE strength. In both tests, a combination of functional grip and arm strength is measured by lifting the box or cup, which must be sustained for 5 s.

At the start of this PhD trajectory, besides the Cup and Box tasks (Chapter 4), no other UE measurement instruments or other types of UE functional strength measurement instruments were available. Since then, only two other UE functional muscle strength measurement instruments have been researched in children with USCP: 'functional strength measurement' (FSM)[14] and 'task-oriented arm–hand capacity' (TAAC) instrument.[15]

The FSM consists of eight items, of which four measure UE muscle strength: one measures anaerobic muscle endurance (lifting a box) and three measure muscle power (overarm throwing, underarm throwing, chest pass). In the anaerobic muscle endurance item (lifting a box), the number of repetitions in a 30-s time frame is assessed; in the muscle power items, the distance a sandbag is thrown is measured in centimetres. One study concluded that the FSM is feasible, reliable and valid to use in children with CP.[14] However, in contrast to the Cup and Box tasks, the UE items of the FSM are not specifically tailored to relevant therapy goals for children with USCP. For example, a goal for children with USCP related to lifting a box/object is mostly related to lifting (and holding) a heavy box/object instead of lifting it several times. The muscle power items of the FSM contain throwing items, which are less related to daily living activities.

The TAAC instrument is a computerized version of our Cup and Box tasks, where, by means of a sensor, the force is registered and shown on a display. The TAAC measures the peak force while the child lifts a cup or box and is holding it for 5 s. Among the activities of daily life, the lifting of objects is a relevant task and common therapy goal in children with CP (e.g. lifting a school bag). The TAAC showed good test–retest reliability for the (bimanual) Box tasks and for the Cup task for the non-affected hand for children (6–18 years) with USCP.[15] The results of the Cup task for the affected hand showed moderate test–retest reliability. An advantage of this digitalized version is that it can measure the force more precisely, as the minimum added weight, after the baseline weight was determined, was 100 g for the Cup task and 500 g for the Box task. Also, the chance of measurement error caused by a mistake in reading the computer display is lower than that of miscalculating the total amount of added weights by the tester. However, the costs of the TAAC are much higher compared to the Cup and Box tasks, therefore the TAAC is more applicable for research and the Cup and Box tasks for clinical practice.

As indicated above, it was not clear whether the muscle strength of the non-affected UE is affected. Alongside the expected weakness of the affected UE, we have hypothesized that the non-affected UE is also weaker compared to the norms of strength of the dominant hand of typically developing (TD) children. Several studies did report reduced performance in hand function of the non-affected UE compared to TD children.[16–19] However, it was not known whether this reduced performance is due to underlying muscle strength problems.

Only two studies have investigated the muscle strength of the non-affected hand in children with USCP, with opposite conclusions, therefore we compared the muscle strength of both hands to that in TD peers (Chapter 5).

Regarding the affected UE, our results support the studies that examined reduced muscle strength in the affected UE. [2,3,5] This muscle weakness of the affected UE appears to be one of the most important causes for the impairment in performance of UE activities.[4–6,25] Several proven effective therapy programmes focus on training the affected UE to facilitate its use in performing a multitude of daily (unimanual and bimanual) tasks: [7–9,20–24] programmes such as constraint-induced movement therapy (CIMT), bimanual intensive movement therapy (BIMT) and a combination of both (hybrid CIMT). All these interventions aim to improve the function and performance of the affected UE in unimanual (CIMT) or bimanual (BIMT and hybrid CIMT) activities.

With regard to the non-affected UE, the results of our study support the findings that muscle weakness in children with USCP is present also in the non-affected UE.

In addition to the fact that several magnetic resonance imaging (MRI) studies in children with supposed USCP have shown bilateral brain lesions[25–28] and that several studies reported reduced performance of the non-affected UE in children with USCP,[16–19] it is advisable not to speak of a purely unilateral disorder and to use the seemingly more appropriate term 'less-affected UE'.

The factor that causes muscle weakness of the less-affected arm/hand has not been researched yet but because of the often-present bilateral brain lesions it is most likely that the muscle weakness originates from reduced motor control. Furthermore, as children with USCP already use their less-affected UE more spontaneously in daily activities, muscle weakness as a result of disuse seems less likely.[29,30]

Whether the muscle weakness in the 'less-affected' UE leads to reduced performance has also not been researched. To perform UE activities, the strength of the affected hand in children with USCP and also in adults after unilateral stroke is an important component. [4–6,31,32] For example, in these populations it has been proved that strengthening interventions of the affected UE not only improve muscle strength but also the performance of activities.[33,34] Thus, it could be hypothesized that in children with USCP muscle weakness of the less-affected UE results in reduced performance, in which case bimanual activity training seems to be more valid than just providing unimanual training of the affected UE.

Based on the results of the reliability studies, we wrote a critical perspective paper (Chapter 6) on the usability of the measurement instruments for discriminative and evaluative purposes. In this paper it is concluded that for the majority of children with USCP there is a high probability (>5%, $p > 0.05$) that the change in muscle strength is due to a measurement error. As an aside, it should be mentioned that it is not yet known how much gain in muscle strength can be achieved through muscle strength training if the right training stimulus is given.

In addition to the perspective paper, it is interesting to get an impression of the probability that the change in muscle strength is due to measurement error. To do so, the formula by which the SDC values are calculated ($SDC = 1.96 \times \sqrt{2} \times SEM$, where SEM is the standard error of the mean) can be adapted. Within this formula, '1.96' represents the $Z$ score, which relates to a $p$ value of 0.05 (a 5% probability that the change is due to measurement error). With this knowledge, the formula can be adapted to $SDC = Z \times \sqrt{2} \times SEM$. By replacing the SDC value for the minimum important change (MIC) value, it is possible to calculate a new Z score: $MIC = Z \times \sqrt{2} \times SEM$. With the new $Z$ score, the corresponding $p$ value can be determined and the probability of a measurement error calculated.

When we use the SEMs displayed in Chapter 3 within the formula, the outcome shows that when a change equal to or higher than the MIC (15% gain in muscle strength for the affected UE and 20% for the less-affected UE) was measured, children with a muscle strength value at or below the 2nd quartile have at least an 18–74% chance that the change score is due to measurement error. This level of probability is much higher than the current standards and therefore, purely based on these results, the recommendation for clinicians would be that in evaluation studies one has to be aware that for the majority of children with USCP there is a high probability that the change score is caused by measurement error. In children with low muscle strength values, the probability that the measured (clinicallchange is due to measurement error can be up to 95%. These results indicate that the HHD and E-link instruments are not usable for measuring changes in muscle strength in individuals. As

this phenomenon occurs in the HHD and E-link but also in the Cup and Box tasks, one may wonder whether this is typical for these measurement instruments. We will further elaborate on this question later in the discussion, after this next section.

## 2. Lessons learned about the methods and designs used in our studies

In our systematic review on measurement instruments to determine UE muscle strength in children with USCP in clinical practice (Chapter 2), we used the COSMIN checklist to assess the methodological quality of the studies.

For methodological quality there are various types of checklists. Some can be used to check for the presence of items reflecting methodological quality, for example the STROBE (STrengthening the Reporting of OBservational studies in Epidemiology) checklist.[35] Others can be used to score the methodological quality of a study, for example the Cochrane Collaboration's tool for assessing risk of bias in randomized trials[36] and the Physiotherapy Evidence Database quality scale for randomized controlled trials (PEDro).[37]

Regarding assessment of the methodological quality of clinimetric properties, several standards and criteria have been proposed, as in the study of Lohr et al.[38] and the quality criteria proposed by Audigé et al.[39] However, these standards and criteria have not been transformed into applicable user-friendly checklists. Worldwide, there are only a few checklists by which the methodological quality of a clinimetric property study can be determined: examples are the Guidelines for Reporting Reliability and Agreement Studies (GRRAS) and the Quality Appraisal of Reliability Studies (QAREL) checklists.[40] However, only the COSMIN checklist includes all clinical properties and is consensus based.

According to the original COSMIN checklist (2011),[41] a study must include at least 50 participants to allow a good methodological quality score for population size. This is a very strict criterion that is not easy to achieve in a clinical study like ours. In 2018, the COSMIN group published a new checklist, the COSMIN Risk of Bias checklist,[42] replacing the original COSMIN checklist.[43] In this checklist it is recommended to take the aggregated sample size of the available studies into account when assessing the overall quality of evidence for a measurement property in a systematic review.[44] A total of 50–99 participants is needed to allocate the label of adequate quality and at least 100 participants for a very good methodological quality score.[44] But even if we pooled the sample sizes of all included studies in our review, no measurement instrument would receive a higher methodological quality score. If we totally omitted the quality criterion of population size, some measurement instruments would be given a slightly better methodological quality score. However, the primary goal of most studies was not to determine the clinimetric properties, therefore these studies did not report on several other important criteria and the conclusion that there are only limited studies of poor methodological quality still holds.

The original COSMIN and the later COSMIN Risk of Bias checklists were developed for assessing the quality of studies on the measurement properties of patient reported outcome measures (PROMs). Because clinician reported outcome measures (ClinROMs) and performance-based outcome measures (PerBOMs) are typically more complex, requiring strict protocols, specific equipment and the involvement of personnel, the COSMIN group is currently developing a new version of the checklist that can hopefully soon can be used on ClinROM and PerBOM studies. To reach consensus about the content

of this new checklist, the COSMIN group performed an international Delphi study among experts from various biomedical fields. A PerBOM checklist would have been more appropriate in our case. What influence the use of such a checklist would have had on the results presented in this thesis is hard to predict, as the checklist is still in the developmental stage. However, with the experience gained in our studies, we will now report our ideas on which items of the current COSMIN Risk of Bias checklist are useful in the new PerBOM checklist (related to reliability) and which new items are needed.

The current COSMIN Risk of Bias checklist contains four items on the statistical method: one item related to the patient (stable in the interim period), two items on the design (time interval appropriate, test conditions similar for the measurement) and one item named 'other important flaws'. These four items are also appropriate to use in the ClinROM and PerBOM checklists.

The main difference between a PROM and a ClinROM/PerBOM is the difference in obtaining the results, namely by the patient (PROM) versus the examiner/test (ClinROM/PerBOM). There is no item on how the results were obtained, which is a particular feature of PerBOMs. Therefore, we recommend adding an item about the examiner. One well-trained examiner may not properly reflect how the results of the measuring instrument are commonly obtained in clinical practice. Another difference between a PROM and a ClinROM/PerBOM is that some ClinROMs/PerBOMs have strict protocols in which the instruction of the patient is precisely described. When these protocols are not followed by the examiner, there is a higher chance that larger measurement errors occur. Related to measurement errors caused by deviations of the protocol/test instructions, the item 'Were the test conditions similar for the measurements? e.g. type of administration, environment, instructions' partly covers this topic. However, some measurements also require specific protocols in the use of the measurement instrument by the examiner. In such cases, an additional question, on whether the existing specific protocol of the measurement instrument has been properly used, should be taken into account.

In Chapters 3 and 4, the clinimetric properties of the HHD, E-link and Cup and Box tasks were described. To determine overall usability it is also necessary to look at the other clinimetric properties (validity and responsiveness for the HHD and E-link; responsiveness for the Cup and Box tasks).

The validity of the HHD has never been researched in the upper and lower extremities of children with USCP. Studies have involved other populations and often an isokinetic dynamometer (like the Biodex system) as the gold standard.[45–47] However, during isokinetic muscle strength testing the patient needs to be able to cooperate with the examiner and to perform a maximal concentric contraction of one muscle group. For many children with CP this is a very difficult task to perform due to co-contraction of antagonists or agonists and/or cognitive limitations.[48,49] Because of these specific characteristics of children with CP, we regard the isokinetic dynamometer not to be an appropriate comparator to serve as the gold standard. Therefore, research into the criterion validity of the HHD in children with CP using the isokinetic dynamometer for comparison is not appropriate. As the tester tries specifically to measure muscle strength on the function level of the ICF-CY (International Classification of Functioning, Disability and Health – Children and Youth Version) with the HHD, research into the construct validity is more appropriate.

Regarding the validity of the E-link, the study by Allen et al. indicates that both the Jamar and Biometrics dynamometers measure the same construct of grip strength and that the E-link is a valid tool for measuring maximum voluntary grip strength in healthy university students (aged 18–25 years).[50] Unfortunately, no studies are published on the validity of the E-link in children with USCP,

therefore it is not certain that the Jamar can be used to check the validity of the E-link in children with CP, as these children have specific characteristics, such as cognitive limitations, that could influence the validity. Further research on this topic is needed to gain more clarity.

No research has been done on the responsiveness of the HHD, E-link and Cup and Box tasks for UE measurements in children with USCP. Given the high measurement error, we have doubts about the responsiveness. Further research is therefore needed before firm conclusions can be drawn about the usability of these instruments for evaluative purposes.

Chapter 5 focused on the differences in muscle strength between children with USCP and TD peers. When examining differences between populations, it is important that both groups are representative samples of their population. The TD peers were recruited from different parts of the Netherlands to obtain a representative sample. Regarding the children with USCP, only those who have had therapy at a rehabilitation centre were included. However, it cannot be determined whether this had a positive or negative impact on the results. Children with USCP who have (multiple) problems in activities visit a rehabilitation centre, therefore the children could have already had a muscle strength training programme and it is unknown whether this has influenced our results.

In the comparison of muscle strength between the children with USCP and TD peers, we used the outcome of the HHD, which depends on the distance between the HHD placement and the joint. To avoid differences in HHD outcome due to differences in arm length, one should calculate the torque (torque = lever-arm ´ force). Because we did not have information on the arm length of the children, we were not able to calculate this torque. In our study, the length, weight and age did not differ at group level, therefore standardization of the HHD outcome by calculating the torque would not have led to an altered conclusion. Nevertheless, when evaluating muscle strength over a longer period (years), it is advisable to include the lever-arm in measurements because it is likely that the lever-arm of the child increases due to natural growth of the child between both measurements. The same applies when muscle strength norm values are developed.

In Chapter 6 we discussed the usability of the measurement instruments from a critical perspective, where the most important weaknesses/flaws of the study design and measurement instruments used can be found.

## 3. Determining the smallest detectable change

The current method to determine the SDC uses measures of multiple participants and results in an absolute value for the total group. This method is recommended by the members/experts of the COSMIN group and is well researched within the focus area (PROMs) of the COSMIN.

A measurement error can be caused by four sources: the measurement instrument itself, the examiner(s) performing the measurement, the patient undergoing the measurement and the circumstances under which the measurement is performed.[10] With the design of our studies, the use of a standardized protocol and training of skilled therapists, we tried to minimize the influence of these sources on the measurement error. Despite all these points of attention, relatively high SDC values were found in all the measuring instruments that we have researched. The question arises whether such high values are specific for these measurement instruments or related to the population (USCP).

Therefore, we have examined the SDC values for the HHD in the lower extremities in children with USCP (the E-link measures grip/pinch strength) and for the HHD and E-link in other populations. As we developed the Cup and Box tasks recently, no other information about these measurement instruments could be found.

To gain more insight into the size of the SDC value, we converted absolute to relative SDC values (in %) by dividing the SDC-value by the average group value.

— SDC values for the HHD in the lower extremities in children with CP:
Willemse et al. reported high relative SDC values in lower extremity strength measurements (>20.6% for knee flexors and >34.8% for ankle plantar flexors) in children with CP using the HHD.[51] In this study, it was concluded that because previous strength training studies had reported lower extremity muscle strength increases of 11–74%, the HHD often will be insufficiently sensitive to detect individual strength gains.[51]
Van Vulpen et al. reported a 40–128% SDC value for the standing heel-rise test in children aged 3–5 years and a 23–48% value in children aged 6–10 years for both the affected and less affected extremity.[52] However, they concluded that they found acceptable SDC values for the isometric hip abductors (11%), knee extensor (9%) and calf (23% for M. gastrocnemius and 30% for M. soleus) muscle strength tests for both the affected and less affected extremity. In that study, all the measurements were performed by the same experienced/well-trained examiner. These results were achieved by taking the mean values of two or three test occasions (separate days) and repetitions. By using fewer repetitions and moments, the SDC values increased up to 57%.
Summarizing, high SDC values are found also in the lower extremities of children with USCP.

— SDC values for the HHD and E-link in other populations:
For hand strength in TD children aged 4–12 years (performed with a Lode dynamometer, which is similar to the Jamar dynamometer), an SDC value of 23.2–27.0% of the mean maximum voluntary contraction was found.[53]
Also, high SDC values were found in healthy adults (20–30%),[54] in adults with shoulder pain and functional loss (15% and 28%),[55] in elderly with diabetes/chronic condition (28%, elbow flexion)[56] and in women with systemic lupus erythematosus (21%, grip; 21%, shoulder abduction; 18%, shoulder flexion).[57]
Summarizing, the HDD and E-link also have high relative SDC values in other populations.

Because high SDC values are found in both the lower extremities of children with CP and in other populations, we also examined the SDC values for other instruments that measure the same construct (muscle strength or endurance).

— The 'functional strength measurement' (FSM) in children with USCP also reveals high SDC values. The SDC values for the majority of the individual's items were larger than the median. For the total score, the SDC value for the younger population (4–6 years) is just below the median (2.33 vs. 3.0) but for the older group (7–10 years) is more than half the median (2.8 vs. 5).

For the muscle power sprint test, on comparing the SDC values and means a relative SDC value of 25–40% (mean power/peak power) was found.[58]

Two studies were published in which the SDC value of the Wingate anaerobic test in children with CP was reported. In one study, the SDC value was 16.3% compared to mean power and 28.9% for the peak power.[59] In the other study it was concluded that only large improvements (16–45%) can be detected when monitoring individual changes.[60]

Summarizing, other measurement instruments that measure muscle strength or endurance in children with CP also have high relative SDC values.

As the SDC value seems high irrespective of the measurement instrument used or population studied, one should question whether the current form of the SDC (as an absolute value valid for the total population) is appropriate for these types of measurement. Perhaps we should consider alternative ways to calculate the SDC, considering its intended use for interpreting change scores in individual patients.

Alternatives:

1) Calculate the SDC value as a percentage:

   For example, when measuring the height of a person multiple times within a short period using a measuring rod, it is likely that the measurement error is about the same in all measurements, independent of the height of the person. Changes in height are equally different/easy to measure, independent of the individual, therefore an SDC value as an absolute number seems logical. However, when fixed/absolute values are used for measuring changes in muscle strength, a 5 N gain in muscle strength could be easier to achieve for a person who has a muscle strength of 100 N compared to a person who has a muscle strength of 2 N. After all, the person with the muscle strength of 100 N needs to gain 5% muscle strength, whereas the person with the muscle strength of 2 N needs to gain 250% muscle strength to achieve an increase of 5 N. As the muscle strength values of the children with USCP vary greatly, an increase equal to the SDC value is harder to meet for a child with low baseline muscle strength values compared to a child with higher values.

   In this example it seems better to calculate the SDC value as a percentage so that it is equally difficult for every child to achieve an increase equal to the SDC value. This could be possible if the SEM was also calculated as a percentage, instead of an absolute value. However, until now, this has not been done.

2) Multiple measurements within one individual:

   Alternatively, an SDC value could be determined for every child individually by taking multiple measurements before starting the intervention. The difference between the highest and the lowest muscle strength value at baseline can then be used to determine the child-specific SDC value. In this way, child-specific SDC values are available that can be used for evaluative purposes.

3) Divide the SDC by the square root of *N*:

In the book *Measurement in Medicine*, it is indicated that for research with groups of patients the SDC value can be divided by the square root of the number of patients, *N*.[61] This implies that in clinical research smaller changes are needed at group level to detect changes beyond the measurement error. This formula would also mean that for a large population it is always possible to measure an effect above the measurement error at group level. With this information, all measuring instruments will be useful in research with larger groups of children with USCP. However, in personal consultation with the COSMIN steering group, it was advised not to use this method. One of the authors declared that, in their opinion, the SDC value is only related to changes in individuals and not to changes in groups.

Further research on this topic is definitely needed. Until there is more clarity about alternative methods for calculating the SDC value, it is recommended that additional strength measurement instruments are used. When multiple measurement instruments (preferably on different ICF levels) all show gains between the pre-/post-intervention measurements, there may be a higher probability that the intervention has a 'real' positive result.

# Clinical implications

## Who should be tested?

In children with USCP suspected of muscle weakness, the HHD, E-link and Cup and Box tasks are suitable in order to gain an impression of their muscle strength.

During muscle strength measurements, it is important that the child is able to fully cooperate. In the study by van Vulpen et al., lower reliability and higher SDC values were found in the age group 3–5 years compared to children aged 6–10 years.[52] Therefore, the chance of measurement error in younger children seems to be higher. It should also be kept in mind that strength training programmes are recommended no earlier than at the age of 7 years[62] so it is not possible to set a 'hard' minimum age limit for measuring muscle strength.

Unfortunately, we were not able to research whether specific child characteristics related to USCP, such as the degree of spasticity or the Manual Ability Classification System (MACS) level, are related to the muscle strength of a child with USCP. As muscle weakness appears to be associated with impairment in the performance of UE activities.[4–6] it is possible that children with USCP with less arm/hand abilities have lower muscle strength compared to those with more arm/hand abilities. There are several classification methods to make a distinction in arm/hand abilities. In the Dutch CP guidelines, only the MACS is recommended for classification of the UE possibilities. Worldwide, the Bimanual Fine Motor Function (BFMF) classification is often used, which classifies fine motor function according to the child's best ability (capacity) to grasp, hold and manipulate objects for each hand separately.

Because the MACS classifies bimanual possibilities, less can be said about the unimanual possibilities. Furthermore, because the muscle strength of the less-affected UE can also be affected, deviations in the less-affected UE can have a major impact on the MACS level. As a result, it is not self-evident that the MACS level is appropriate to use for stratification.

## Who should do the testing?

Our results showed that in most measurements the intraclass correlation coefficient (ICC) of the test-retest reliability is a fraction higher compared to the interrater reliability. However, differences in the ICC values for test-retest and interrater reliability are usually negligible and the 95% confidence intervals are largely overlapping. This is opposite to our expectations, as in interrater reliability measurements (where multiple testers are involved) there seems to be a higher chance for measurement error. After all, differences between measurements as a result of differences in interpretation and execution of the measurement protocol are expected to be much higher when different raters are involved. This particular finding is propably due to the use of a standardized protocol and well-trained examiners, and can be regarded as an important indication for clinical practice that the use of both well-trained tester(s) and a clear testing protocol can reduce measurement error.

Around a single measurement there is always measurement error (SEM) but by taking several measurements the range between which the real value is located becomes narrower. To reduce the chance of measurement error, it is recommended that a standardized protocol be used and that the measurements are performed on different short successive moments (separate days) and measured multiple times within one measurement moment, taking the mean value of the measurements. This will minimize the likelihood that the differences in muscle strength are caused by differences in initial posture, specific influential child characteristics (e.g. fatigue), differences in placing of the measurement instrument (the HHD) or differences in handling the measurement instrument (the E-link).

## Clinical implications of the measurement instruments for discriminative purposes

As the reduced performance of the affected UE in children with USCP could be caused by several impairments in body functions, such as disturbances in both the passive and active range of motion, muscle tone, sensibility and muscle strength, task analyses and/or multiple tests need to be carried out to determine which factor has the largest contribution to the activity impairments.[4–6]

In most activities, not only is maximum muscle strength important but other components such as muscle strength generation, regulation and timing of the strength are also important in the execution of activities. If there is a suspicion that muscle strength is a limiting factor in activities, then all the different components have to be examined. Therefore, it is recommended that various strength measurement instruments are used, covering different strength components and different ICF levels.

To determine if the maximal UE muscle strength is reduced in children with USCP, both the HHD and E-link are excellent measurement instruments. However, to gather information about muscle strength in a functional context (activity level of the ICF-CY), the Cup and Box tasks can be used.

As norm values for all these instruments are not yet available, it cannot be determined whether a child has 'normal' (functional) muscle strength. Overall, the less-affected UE in children with USCP is weaker compared to the norm of TD peers.[63] A comparison of the affected UE with the less-affected UE, instead of with normative data, is therefore less appropriate to identify deficits in the less-affected UE: the critical clinical gaze of a professional will always be needed in the interpretation of measure-

ment results. The development of norm values will be of great added value in interpreting the results in discriminative measurements. Furthermore, the lever-arm of the child could be included within the norm values, as the length of the lever-arm influences the outcome of the HHD and Cup and Box tasks.

### *Core set*

Combining the results of different measurements of body functions (passive and active range of motion, muscle tone, sensibility) and constructs of muscle strength (grip and pinch: the E-link; arm: the HHD; functional strength: the Cup and Box tasks), but also tests to map muscle strength generation, regulation and timing of the strength, leads to a complete picture of the UE. With this knowledge, a more specific therapy programme can be tailored to the specific impairments of the individual child and, of course, to the goal of the child.

## Clinical implications for evaluative purposes

Caution in the interpretation of changes in UE muscle strength measurements is needed. Based on our results, it is only possible to achieve a gain in muscle strength that is considered 'real' for the strongest children with USCP. By using the core set of instruments that measure muscle strength on multiple ICF levels, a little more certainty about changes in muscle strength as a result of training can be obtained. In addition, it is advisable to involve the child and the parents in formulating the expected progress of the therapy, by letting them determine the minimal change (on activity/participation level) that is needed to be clinically relevant (MIC). Also, specific measurements (e.g. Goal Attainment Scaling)[64] related to the goals (on activity/participation level of the ICF-CY) are needed for evaluative purposes.

When evaluating muscle strength over a longer period (years), it is advisable to include the lever-arm in the measurements. With this method the torque can be measured. This prevents measured differences in muscle strength between measurements that may not be due to changes in muscle strength but arise because the arm has grown longer.

# Future research

1. More research on the statistical alternatives for calculating the SDC value for field-based tests is needed. As displayed earlier in this discussion, it seems that in many field-based tests excellent ICC values are found but also high relative SDC values. Due to the high SDC values, most instruments seem unusable for evaluative purposes.
2. It must be examined if, and by how much, the reduced maximum muscle strength of the less-affected UE is the cause of the reduced uni- and bimanual performances in children with USCP compared to TD children.
3. More research on what causes the muscle weakness of the less-affected UE is needed. In addition, research on the possibilities and results of training the less-affected UE is needed to determine the best type of intervention.

4. It must be examined if a core set of UE (muscle strength) measurement instruments can be achieved. With the use of this core set, it should become clearer whether and where muscle weakness occurs and what the consequences are for the performance of activities. In evaluative measurements, with this core set it is easier to register changes in muscle strength. When multiple measurement instruments show gains between measurements, there may be a higher probability that the intervention has a 'real' positive result.
5. More research about the norm values of the measurement instruments is needed. The development of norm values will be of great added value for the clinician in interpreting the results. These norm values must be based on TD children and ideally should include the lever-arm.
6. Our research only focuses on children with USCP in the age group 7–12 years. It would be very interesting to find out if our results also apply to those who are younger than 7 years old and to those aged 13–18 years.

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# Valorisation Addendum

In this dissertation, valorisation is considered as "the process of creating value from knowledge, by making knowledge suitable and/or available for social (and/or economic) use and (by making it suitable) for translation into competing products, services, processes and new activities" (definition taken from the report of the National Valorisation Committee, Waardevol: Indicatoren voor Valorisatie [Valuable: Indicators for valorisation] (2011) The Hague: Rathenau Institute, p. 8).

In this valorisation-addendum the following topics will be addressed:

1. social relevance of research results;
2. target groups to whom research results are of interest;
3. products/activities in which research results can be applied and formalized;
4. the extent to which research results can be called innovative;
5. how the valorisation plan will be implemented.

## 1. Social relevance of research results

Worldwide, CP is the most common motor disorder in childhood, with a prevalence in Europe of about 1.8–2.1 per 1,000 live births. Within the Dutch pediatric rehabilitation, CP is the largest group (>32%) receiving interdisciplinary pediatric rehabilitation treatments, resulting in high healthcare costs. Healthcare costs in Dutch pediatric rehabilitation in 2018 amounted to approximately 146 million euros.

Approximately 30% of all children with CP are diagnosed with unilateral spastic cerebral palsy (USCP). Muscle weakness of the affected upper extremity (UE) is one of the main characteristics of a child with USCP. Measuring muscle strength is a common activity in daily practice for most clinicians working with children with USCP in order to determine whether muscle strength weakness is present (discriminative) or whether muscle strength training has been effective (evaluative). In order to be able to measure changes in muscle strength, one needs to know the extent to which variations in measurement results occur between repeated measurements. This so-called measurement error may have several causes: the measurement instrument itself, the examiner(s) performing the measurement, the patient undergoing the measurement and the circumstances under which the measurement is performed.

This dissertation gives more insight into the (im)possibilities of measuring UE muscle strength in children with USCP in clinical practice.

Regarding the less-affected UE, only two studies had investigated muscle strength of the less-affected hand in children with USCP, with opposite conclusions. As it was not clear whether muscle weakness is a unilateral problem or whether both UEs have muscle weakness, we also researched the muscle strength of both UEs in children with USCP. This dissertation provides new insights into UE muscle strength in children with USCP (both affected and less-affected).

## 2. Target groups to whom research results are of interest

The results presented in this thesis are of interest to clinicians and researchers working with children with USCP. Our results can be used to 1) further optimize diagnostics and therapy (including the less-affected UE) and 2) to improve the use and interpretation of strength measurements for evaluative purposes.

Moreover, information about the Standard Error of the Measurement (SEM), Smallest Detectable Change (SDC), and the introduction of two new functional UE muscle strength measurement are valuable to the clinicians.

As the SDC values seem highly irrespective of the measurement instrument used or population studied, we advise that alternative ways to calculate the SDC should be considered. Therefore, the discussion of the dissertation is also interesting for statisticians and/or clinical epidemiologists who are interested in the smallest detectable change of measurement instruments.

To a lesser extent, policy makers can use our results to determine which interventions can be included during the development or update of clinical guidelines. Finally, this indirectly concerns health insurers because they can decide which (effective) muscle strength training intervention is reimbursed. and which is not.

## 3. Products/activities in which research results will be applied and formalised

Our measurement protocol is freely available and a large part of the measurement protocol has already been published as an appendix of the paper presented in chapter 4. If needed, we can train therapists in the (right) use and interpretation of measurement instruments.

We have also disseminated our results by presentations and mini symposiums at national and international conferences. After finishing this PhD, the dissemination of the results at national and international conferences will continue and a one-day symposium will be organised by Revant, Rehabilitation Centre Breda and Adelante Kenniscentrum in Hoensbroek, in collaboration with the Department of Rehabilitation Medicine (Care and Public Health Research Institute (CAPHRI)) of Maastricht University. The symposium is intended for clinicians and researchers.

As far as the cup and box tasks are concerned, we have chosen to use commonly available objects, so that they can easily be used by every clinician.

Recently, a computerised version of the cup and box tasks has been developed (activity Daily Life test and training Device-ADL-TTD), which can be used in research but also for training UE muscle strength. The ADL-TTD is also the first measuring instrument in which muscle strength during multiple (long lasting) dynamic functional tasks can be measured by a computer. Clinimetric properties of the

ADL-TTD are currently being researched. If the product is usable in clinical practice and clinimetric properties are acceptable, the product will become commercially available (under the license of Umaco b.v./ Procare b.v.)

## 4. The extent to which research results can be called innovative

Our study was the first study in which clinimetric properties of UE measurement instruments for children with USCP were researched in a study of good methodological quality as suggested by the COSMIN.

Although a few functional UE muscle strength measurement instruments are available, the Cup and Box tasks are the first functional muscle strength measurements in which the ability to maintain the strength in a sustained contraction can be measured.

Another innovation is that we showed that in children with USCP, muscle weakness is present in both UEs. As a result, measurement of both hands separately must be done in the diagnostic phase, and a bimanual strength measurement must be added. Besides, in case of muscle weakness of the less-affected UE, bimanual activity training seems to be more valuable than just providing unimanual training of the affected UE.

To our knowledge, we are the first research group to criticise the calculation of the smallest detectable change in upper extremity muscle strength measurements in children with USCP. Alternative methods should be considered.

## 5. How the valorisation plan will be implemented

As our research did not contain any intervention or product development, less can be said about the implementation of the valorisation plan. Information on how the knowledge/results are disseminated has already been mentioned in paragraph 3 of this addendum. Additionally, within our research network, the results have been disseminated with the message that:

- the measurement instruments studied are useful for clinical practice,
- measure the muscle strength in both arms
- measure different constructs of muscle strength, such as peak and endurance strength and certainly functional strength
- have the measurements performed by trained therapists
- measure multiple times, as described in our standardized measurement protocol, and
- use the measurement instruments we have researched.

What we can and will implement, is the use of the measurement instruments in regular rehabilitation and in highly intensive existing UE therapy camps for children with USCP. The findings regarding the muscle weakness of the less-affected UE will be communicated to the project leaders of the UE training camps and we will discuss with them what possibilities there are to pay more attention to the less affected UE. The "Handen in elkaar" platform and the CP-Net organisation will be invited to spread the knowledge in the Netherlands.

Regarding the calculation of the SDC, presented in chapter 6, we made an appeal that future research should focus on alternative ways to calculate the SDC. In the near future we will contact the members of the COSMIN team to discuss our results and recommendations regarding this topic to facilitate this research.

# Summary

"CP describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, behavior, by epilepsy, and by secondary musculoskeletal problems." By far the largest group (>85%) consists of children with spastic CP. This group can be divided into bilateral and unilateral spastic CP. Overall, almost 30% of all children with CP are diagnosed with unilateral spastic cerebral palsy (USCP). USCP is characterized by motor impairments lateralized to one side of the body, resulting in an "affected" and a "non-affected" body side.

Children with USCP are typically more limited in manual ability than in gross motor function. They usually have difficulty with grasping, reaching, releasing, and manipulating objects with the affected upper extremity (UE), limiting their ability to execute activities of daily living and restricting their independence and participation. About 60% of the children between the age of 4- and 16-years experiences problems with their arm-hand function during daily activities predominantly caused by impairments in muscle function.

Muscle weakness appears to be one of the most important causes of impairment in performance of UE activities.

Measuring muscle strength is a common activity in daily practice for most clinicians working with children with USCP in order to determine whether muscle strength weakness is present (discriminative) or whether muscle strength training has been effective (evaluative). In order to make inferences about muscle strength, either in clinical practice or in research, strength has to be measured with an instrument that has sound clinimetric properties. To know the extent to which the strength measurement instrument is suitable for discriminative and evaluative purposes in clinical practice, the clinimetric properties "reliability" and "responsiveness" of the instrument need to be known. Reliability indicates the degree to which the measurement is free from measurement error. With an Intraclass Correlation Coefficient (ICC) value it is possible to determine whether the measurement instrument is suitable for discriminative purposes. The related Standard Error of Measurement (SEM) is a measure of how far apart the outcomes of repeated measurements are; it is the standard deviation around a single measurement.

Responsiveness indicates the ability of a measurement instrument to detect changes over time in the construct being measured. For the interpretation of a change in score, the smallest detectable change (SDC) and the minimally important change (MIC) are important. With the SDC, it can be determined whether the difference between two (evaluative) measurements can be distinguished from

a measurement error. In order to know whether a change score is also clinically important, the MIC is considered the most important value. The MIC is the smallest change score in the construct to be measured that patients, clinicians or relevant others perceive as important.

The quality of each clinimetric property can be rated as positive, negative or indeterminate, according to internationally accepted criteria. In addition to the value of the clinimetric property, it is important that the research of the determination of the clinimetric property is conducted in a study of (at least) good methodological quality.

Four out of five studies presented in this thesis focus on the clinimetric properties of various UE strength measuring instruments for children with USCP and on the interpretation of their outcomes, with the intention of clarifying how useful these measurement instruments actually are in clinical practice. The fifth study focuses on the muscle strength of the non-affected UE in children with USCP because it is not clear whether muscle weakness is a unilateral problem or whether the 'non-affected' UE is also affected.

The first step was to obtain a detailed overview of the existing UE muscle strength tests for children with USCP. Therefore we performed a systematic review in which the reported clinimetric properties were derived from the studies and also the methodological quality of the studies was determined. A data-synthesis of both aspects was performed to determine which measurement instruments are useable in clinical practice. The systematic review is presented in Chapter two. In the few (a total of seven) studies studying a total of six measurement instruments for measuring upper extremity strength, only test-retest, intrarater, and interrater reliability were investigated in a select group of age ranges, Manual Ability Classification System (MACS) levels, and Gross Motor Function Classification System (GMFCS) levels. No conclusions can be made regarding the responsiveness, the SDC or SEM. Furthermore, it is not clear whether all measurement instruments specifically measure muscle strength, as validity has not been investigated. Although the assessed clinimetric properties for most measurement instruments were rated positive, the methodological quality of the studies was mostly poor. As a result, the found clinimetric properties have hardly any scientific value. Three measurement instruments can be used in clinical practice. First; to measure grip strength, it is recommended to use the Jamar dynamometer. Second; for measuring other upper extremity muscle groups, it is recommended to use the Hand Held Dynamometry (HHD). Third; manual muscle testing can be used when measuring the "total upper extremity" or wrist strength in children with CP who have very limited muscle strength ((≤ grade 3).

Based on these results, only two instruments (HHD and Jamar) were identified as potentially useful UE muscle strength measurement instruments. Next, we performed a study to determine test-retest and interrater reliability of these measurement instruments in children with USCP, in a study designed using the guidelines of the COSMIN consortium. The results are presented in Chapter three. Instead of the Jamar dynamometer, we used a digitalized version (E-link) of this measurement instrument.

Our results showed that almost all arm/hand strength measurements (performed with the HHD and E-link) have excellent test–retest reliability and excellent interrater reliability in children with USCP, aged 7–12 years. Only the interrater reliability of the elbow flexion of the affected UE was classified as "good", meaning there was more variability in the performance of this measurement. We concluded that the HDD and E-link system are usable measurement instruments in cross-sectional measurements of UE muscle strength in children with USCP. Because there is no clear information available

on how much improvement a child with USCP can achieve after a strength-training program, it is not clear if both instruments are usable for measuring changes in UE muscle strength within one person, especially if a child with USCP has low muscle strength.

In most daily life manual activities, e.g. during carrying/moving a heavy box, not only a certain amount of muscle strength is required, but also the ability to maintain/regulate that strength for a certain time. This ability is called functional strength and needs to be measured during the performance of the specific task. As there were no existing measurement instruments available which measure UE strength in the context of functional activities, two specific functional muscle strength tests were developed. These tests measure unimanual and bimanual sustained contractions, i.e., the "Cup-Task" for determining maximal functional unimanual UE strength and the "Box-Task" for determining maximal functional bimanual UE strength. In both tests, a combination of functional grip and arm strength is measured by lifting the Cup or Box, which must be sustained for five seconds.

We determined the reliability and validity of these two new functional muscle strength measurement instruments in children with USCP, also following the guidelines of the COSMIN consortium. The results are presented in Chapter four. We concluded that the Cup-Task and Box-Task are reliable and valid measurement instruments for measuring functional hand and upper extremity muscle strength in children with USCP who can perform such tasks. However, most of the children with USCP and MACS level III will not be able to perform the Cup-Task with the affected UE. Due to the lack of information about the possibilities in gaining functional muscle strength, the same conclusion as with the HHD and E-link was drawn regarding their evaluative use, namely that it is not clear whether both instruments are usable for measuring changes in UE muscle strength within one person, especially if a child with USCP has low muscle strength.

As only two studies compared the non-affected UE muscle strength of children with USCP and children with typically development (TD), and these studies had opposite conclusions as to whether muscle weakness only occurs in the affected UE, we performed a study to compare the isometric muscle strength (measured with the HHD and E-link) of the affected and non-affected UE of children with USCP to the isometric muscle strength of children with TD. The results are presented in Chapter five.

In the affected UE (non-preferred UE), for all measures the children with USCP produced statistically significantly lower muscle strength values compared to children with TD. Our study confirms the hypothesis that children with USCP can generate less muscle strength with the non-preferred side compared to children with TD. However, it is remarkable that the percentage difference in muscle strength is less in the proximal UE muscle groups compared to the distal UE muscle groups. A possible explanation is that the severity of hand function is closely related to the integrity and organization of direct corticospinal projections to the hand muscles. A second explanation might be that because most children with USCP only use the non-preferred UE to support the preferred UE, the proximal muscle groups may be used more compared to the distal (fine motor) muscle groups.

Muscle weakness of the non-affected UE (preferred UE) of children with USCP seems to go beyond impairments in the hand. It was striking that the muscle strength of the elbow flexion and extension in the younger age groups is higher in children with USCP than in children with TD. In the older age groups, this difference between groups is reversed. A possible explanation could be the intensive (bimanual) training that (most) of the participating children with USCP received at the younger age. More research to explain this result is needed.

An important difference between the study we have performed and the other studies that researched the muscle strength of the non-affected UE, is that we used a different measurement instrument to measure grip strength and small differences in muscle strength are more likely to be picked up by the E-Link system.

An important recommendation based on our results is that when uni- or bimanual ability limitations are present, investigation of muscle strength in the non-affected UE should be part of the assessment.

In the last study, we present a critical perspective on how to interpret changes in UE muscle in children with USCP strength, measured with the HHD and E-link, taking an expert-based MIC and the measurement error of the measurement instruments into account. The critical perspective is presented in Chapter six.

We concluded that in clinical practice, for most children with USCP it is possible to measure clinically important changes in muscle strength in the affected UE by means of the HHD and E-link, However, due to the high SDC values, only in those with a high baseline level of muscle strength, the clinically important changes can also be considered "real" changes, i.e. higher than measurement error. Hence, it is only possible to achieve a gain in muscle strength that is considered "real" for a small proportion of the children with USCP. Great caution in the interpretation of the change score is recommended.

Regarding the less affected UE, only in some children (and in some measurements) the clinically important changes can also be considered "real" changes. For the majority of the children with USCP, the important changes cannot be distinguished from measurement error. Because of the lack of information on the muscle strength gain possibilities of the less affected UE, no firm conclusions can be drawn on whether it is possible to measure clinically important and/or statistically significant changes within one child.

To reduce the chance of measurement error, we recommend to use a standardized protocol, to perform the measurements by the same assessor and to measure multiple times within one measurement moment, taking the mean value of the measurements. This minimize the likelihood that the differences in muscle strength are caused by differences in initial posture, differences in placing of the measurement instrument (HHD) or differences in handling the measurement instrument (E-link).

Chapter seven contains the general discussion, in which the main results are evaluated. Also, the methodological considerations are discussed and implications for clinical practice and future research are presented.

As the HHD, E-link, Cup-and box task all have a large SDC-value, we also discussed whether the SDC value, calculated/determined according to the current method, is suitable for muscle strength measurement instruments used in a (heterogeneous) rehabilitation population. Therefore, we made an overview of the SDC values of the HHD and E-link in other populations, and of the SDC values of other measurement instruments which measure muscle strength in children with USCP. The results show that the SDC value seems high irrespective of the measurement instrument used or population studied. Alternative ways to calculate the SDC should be considered, considering its intended use for interpreting change scores in individual patients. Suggested alternatives are: calculate the SDC value as a percentage; or take multiple measurements within one individual and use the difference between the highest and the lowest muscle strength value to determine the child-specific SDC value.

Further research on this topic is definitely needed. Until there is more clarity about alternative methods for calculating the SDC value, it is recommended that additional strength measurement instruments are used. When multiple measurement instruments (preferably on different ICF levels) all show gains between the pre-/post-intervention measurements, there may be a higher probability that the intervention has a 'real' positive result.

# Nederlandse samenvatting (Dutch summary)

"Cerebrale Parese (CP) beschrijft een groep van permanente stoornissen in de ontwikkeling van beweging en houding, waardoor beperking in activiteiten optreedt, die wordt toegeschreven aan niet-progressieve stoornissen, die zich hebben voorgedaan in de zich ontwikkelende foetale- of zuigelingenhersenen. De motorische stoornissen van CP gaan vaak gepaard met stoornissen van gevoel, waarneming, cognitie, communicatie, gedrag, door epilepsie en secundaire spier- en skeletproblemen."
Veruit de grootste groep (>85%) bestaat uit kinderen met spastische CP. Deze groep kan worden onderverdeeld in bilaterale (tweezijdige) en unilaterale (eenzijdige) spastische CP. Gemiddeld genomen is bijna 30% van alle kinderen met CP gediagnosticeerd met unilaterale spastische cerebrale parese (USCP). USCP wordt gekenmerkt door motorische stoornissen aan een zijde van het lichaam, wat resulteert in een "aangedane" en een "niet-aangedane" lichaamszijde.

Kinderen met USCP zijn meestal meer beperkt in fijn motorische arm/handvaardigheden dan in grof-motorische activiteiten. Ze hebben meestal moeite met het grijpen, reiken, het vasthouden en manipuleren van objecten met de "aangedane" bovenste extremiteit (BE). Hierdoor wordt hun vermogen om activiteiten van het dagelijks leven uit te voeren beperkt en hun onafhankelijkheid en participatie belemmerd. Ongeveer 60% van de kinderen tussen 4 en 16 jaar ondervindt problemen met hun arm/handfunctie tijdens dagelijkse activiteiten, voornamelijk veroorzaakt door beperkingen in de spierfunctie. Spierzwakte lijkt een van de belangrijkste oorzaken van de beperkingen in de uitvoering van BE-activiteiten.

Het meten van spierkracht is een veel voorkomende activiteit in de dagelijkse praktijk voor de meeste clinici die werken met kinderen met USCP. Enerzijds wordt spierkracht gemeten om te bepalen of spierzwakte aanwezig is (discriminerend), anderzijds om te onderzoeken of een interventie effectief is geweest (evaluatief). Om conclusies te kunnen trekken over spierkracht, hetzij in de klinische praktijk of in onderzoek, moet kracht worden gemeten met een instrument dat goede klinimetrische eigenschappen heeft. Om te weten in hoeverre het krachtmeetinstrument geschikt is voor discriminerende en evaluatieve doeleinden in de klinische praktijk, moeten de klinimetrische eigenschappen "betrouwbaarheid" en "responsiviteit" van het instrument bekend zijn. Betrouwbaarheid geeft aan in welke mate de meting vrij is van meetfouten. Met een Intraclass Correlatiecoëfficiënt (ICC) is het mogelijk om te bepalen of het meetinstrument geschikt is voor discriminerende doeleinden. De bijbehorende standaardmeetfout (SEM) is een maat voor hoe ver de resultaten van herhaalde metingen zijn; het is de standaarddeviatie rond een enkele meting.

Responsiviteit geeft het vermogen van een meetinstrument aan om veranderingen in de tijd te detecteren. Voor de interpretatie van een verandering in score is de kleinste detecteerbare verandering (smallest detectable change; SDC) en de minimaal belangrijke verandering (minimal important change; MIC) belangrijk. Met de SDC kan worden bepaald of het verschil tussen twee (evaluatieve) metingen kan worden onderscheiden van een meetfout. Om te weten of een veranderingsscore ook klinisch belangrijk is, wordt de MIC beschouwd als de belangrijkste waarde. De MIC is de kleinste veranderingsscore die patiënten, clinici of relevante anderen als belangrijk ervaren.

Aan de hand van internationaal aanvaarde criteria voor kwaliteitsbeoordeling kan de kwaliteit van elke klinimetrische eigenschap worden beoordeeld als positief, negatief of onbekend. Naast de kwaliteit van de klinimetrische eigenschap is het belangrijk dat het onderzoek naar de bepaling van de klinimetrische eigenschap wordt uitgevoerd in een studie van (ten minste) goede methodologische kwaliteit.

Vier van de vijf studies die in dit proefschrift worden gepresenteerd, richten zich op de klinimetrische eigenschappen van verschillende BE-spierkrachtmeetinstrumenten voor kinderen met USCP en op de interpretatie van hun uitkomsten, met de bedoeling te verduidelijken hoe bruikbaar deze meetinstrumenten eigenlijk zijn in de klinische praktijk. De vijfde studie richt zich op de spierkracht van de “niet-aangedane” BE bij kinderen met USCP, omdat het niet duidelijk is of spierzwakte een eenzijdig probleem is of dat spierzwakte ook in de 'niet-aangedane' BE aanwezig is.

De eerste stap was het verkrijgen van een gedetailleerd overzicht van de bestaande meetinstrumenten voor het meten van de spierkracht in de BE bij kinderen met USCP. Daarom hebben we een systematische review uitgevoerd waarbij de gerapporteerde klinimetrische eigenschappen werden afgeleid van de onderzoeken. Tevens werd ook de methodologische kwaliteit van de studies bepaald. Een datasynthese van beide aspecten werd uitgevoerd om te bepalen welke meetinstrumenten bruikbaar zijn in de klinische praktijk. De systematische review wordt gepresenteerd in hoofdstuk twee.

In een beperkt aantal (in totaal zeven) studies werden in totaal zes meetinstrumenten voor het meten van de BE-spierkracht bestudeerd. In deze studies werden alleen test-hertest betrouwbaarheid, intrabeoordelaars- en interbeoordelaarsbetrouwbaarheid onderzocht. De populaties die in de studies werden beschreven betroffen ook nog eens een selecte groep wat betreft leeftijd, MACS-niveaus (Manual Ability Classification System) enGross Motor Function Classification System (GMFCS) niveaus. Er konden geen conclusies worden getrokken over de responsiviteit, de SDC of SEM. Bovendien was het niet duidelijk of alle meetinstrumenten specifiek de spierkracht meten, omdat de validiteit niet is onderzocht. Hoewel de klinimetrische eigenschappen voor de meeste meetinstrumenten positief werden beoordeeld, was de methodologische kwaliteit van de studies meestal slecht. Als gevolg hiervan hebben de gevonden klinimetrische eigenschappen nauwelijks wetenschappelijke waarde. In de klinische praktijk kunnen drie meetinstrumenten worden gebruikt. Ten eerste, de Jamar dynamometer om de grijpkracht te meten. Ten tweede, de Hand Held Dynamometer (HHD) voor het meten van andere spiergroepen in de BE. Ten derde, handmatige spiertesten voor het meten van de "totale bovenste extremiteit" of polssterkte bij kinderen met CP die een zeer beperkte spierkracht hebben (≤ graad 3).

Op basis van deze resultaten werden slechts twee instrumenten (HHD en Jamar) geïdentificeerd als potentieel nuttige BE-spierkrachtmeetinstrumenten. Vervolgens hebben we een studie uitgevoerd om de test-hertest betrouwbaarheid en interbeoordelaarsbetrouwbaarheid van deze meetinstrumenten bij kinderen met USCP te bepalen, in een studie van goede methodologische kwaliteit (volgens de richtlijnen van het COSMIN-consortium). De resultaten worden gepresenteerd in hoofdstuk drie. In plaats van de Jamar dynamometer gebruikten we een gedigitaliseerde versie (E-link) van dit meetinstrument.

Onze resultaten toonden aan dat bijna alle arm/hand spierkracht metingen (uitgevoerd met de HHD en E-link) een uitstekende test-hertest betrouwbaarheid en uitstekende interbeoordelaarsbetrouwbaarheid hebben bij kinderen met USCP, leeftijd 7–12 jaar. Alleen de interbeoordelaarsbetrouwbaarheid van de elleboog-flexie van de aangedane BE werd geclassificeerd als "goed", wat betekent dat er meer variabiliteit is tussen metingen. We concludeerden dat het HDD- en E-linksysteem bruikbare meetinstrumenten zijn voor discriminerende BE-spierkracht metingen bij kinderen met USCP. Omdat er geen duidelijke informatie beschikbaar is over hoeveel verbetering een kind met USCP kan bereiken na een krachttrainingsprogramma, is het niet duidelijk of beide instrumenten bruikbaar zijn voor het meten van veranderingen in BE-spierkracht binnen één persoon, vooral als een kind met USCP een lage spierkracht heeft.

In de meeste dagelijkse activiteiten, bijvoorbeeld tijdens het dragen of verplaatsen van een zware doos, is niet alleen een bepaalde hoeveelheid spierkracht vereist, maar ook de mogelijkheid om die kracht gedurende een bepaalde tijd te behouden/te reguleren. Dit vermogen wordt functionele spierkracht genoemd en moet worden gemeten tijdens de uitvoering van de specifieke taak. Aangezien geen bestaande meetinstrumenten beschikbaar waren die de BE-spierkracht in het kader van functionele activiteiten meten, werden twee specifieke functionele spierkracht testen ontwikkeld. Deze tests meten unimanuele en bimanuele functionele spierkracht, d.w.z. de "maatbeker-taak" voor het bepalen van maximale functionele unimanual BE-spierkrachten de "krat-taak" voor het bepalen van maximale functionele bimanuale BE-spierkracht. In beide tests wordt een combinatie van functionele hand- en armspierkracht gemeten door de maatbeker of krat op te tillen, en deze gedurende vijf seconden in een bepaalde positie te houden.

We hebben de betrouwbaarheid en validiteit van deze twee nieuwe functionele spierkrachttesten bepaald bij kinderen met USCP, volgens de richtlijnen van het COSMIN-consortium. De resultaten worden gepresenteerd in hoofdstuk vier. We concludeerden dat de maatbeker-taak en krat-taak betrouwbare en valide meetinstrumenten zijn voor het meten van functionele hand- en armspierkracht bij kinderen met USCP die dergelijke taken kunnen uitvoeren. Echter, de meeste van de kinderen met USCP en MACS niveau-III zullen niet in staat zijn om de maatbeker-taak uit te voeren met de aangedane BE. Vanwege het gebrek aan informatie over mogelijke resultaten van functionele spierkracht training, werd dezelfde conclusie getrokken als bij de HHD en E-link met betrekking tot hun evaluatief gebruik, namelijk dat het niet duidelijk is of beide instrumenten bruikbaar zijn voor het meten van veranderingen in BE (functionele) spierkracht binnen één persoon, vooral als een kind met USCP een lage spierkracht heeft.

Aangezien slechts twee studies de niet-aangedane BE-spierkracht van kinderen met USCP en typisch ontwikkelende (TD) kinderen hebben vergeleken, en deze studies tegengestelde conclusies hadden over de vraag of spierzwakte alleen optreedt in de aangedane BE, voerden we een studie uit om de isometrische spierkracht (gemeten met de HHD en E-link) van de aangedane en niet-aangedane BE van kinderen met USCP te vergelijken met de isometrische spierkracht van kinderen met TD. De resultaten worden gepresenteerd in hoofdstuk vijf.

Voor wat betreft de aangedane BE hadden kinderen met USCP in alle spiergroepen statistisch aanzienlijk minder spierkracht in vergelijking met niet-voorkeurs BE van kinderen met TD. Onze studie bevestigt de hypothese dat kinderen met USCP minder spierkracht kunnen genereren met de aangedane BE in vergelijking met kinderen met TD. Het is echter opmerkelijk dat het procentuele verschil in spierkracht minder is in de proximale BE-spiergroepen in vergelijking met de distale BE-spiergroepen. Een mogelijke verklaring is dat de ernst van de handfunctie nauw verwant is aan de integriteit en organisatie van directe corticospinale projecties van de handspieren. Een tweede verklaring zou kunnen zijn dat, omdat de meeste kinderen met USCP alleen gebruik maken van de aangedane BE ter ondersteuning van de niet- aangedane BE, de proximale spiergroepen meer worden gebruikt in vergelijking met de distale (fijne motorische) spiergroepen.

Ook in de niet-aangedane BE van kinderen met USCP is spierzwakte aanwezig en deze lijkt in de hele niet-aangedane BE voor te komen (in vergelijking met de voorkeurs BE van kinderen met TD). Het is dus beter om te spreken van een "minder-aangedane" BE. Het was opvallend dat kinderen met USCP in de jongere leeftijdsgroepen sterker zijn in de elleboogflexie en -extensie in vergelijking met kinderen met TD. In de oudere leeftijdsgroepen is dit verschil tussen groepen omgekeerd. Een mogelijke verklaring zou de intensieve (bimanuele) training kunnen zijn die (de meeste) van de deelnemende kinderen met USCP op jongere leeftijd kregen. Om dit resultaat te verklaren, is meer onderzoek nodig.

Een belangrijk verschil tussen de studie die wij hebben uitgevoerd en de andere studies die de spierkracht van de "minder-aangedane" BE hebben onderzocht, is dat we een ander meetinstrument gebruikten om grijpkracht te meten en kleine verschillen eerder zullen worden opgepikt door het door ons gebruikte E-Link-systeem.

Een belangrijke aanbeveling op basis van onze resultaten is dat wanneer uni- of bimanuele beperkingen aanwezig zijn, moet het in kaart brengen van de "minder-aangedane" BE-spierkracht deel uitmaken van het onderzoek.

In de laatste studie presenteren we een andere kijk op het interpreteren van veranderingen in BE-spierkracht bij kinderen met USCP, gemeten met de HHD en E-link, rekening houdend met een MIC en de meetfout van de meetinstrumenten. Dit perspectief wordt gepresenteerd in hoofdstuk zes.

We concludeerden dat in de klinische praktijk, bij de meeste kinderen met USCP het mogelijk is om klinisch belangrijke veranderingen in spierkracht in de aangedane BE te meten door middel van de HHD en E-link. Echter, als gevolg van de hoge SDC-waarden, kunnen alleen bij kinderen met veel spierkracht de klinisch belangrijke veranderingen ook worden beschouwd als "echte" veranderingen, d.w.z. niet veroorzaakt door meetfouten. Vandaar dat een 'echte' winst in spierkracht alleen bereikbaar is voor een klein deel van de kinderen met USCP. Grote voorzichtigheid bij de interpretatie van een veranderingsscore is noodzakelijk.

Met betrekking tot de "minder-aangedane" BE, kunnen alleen bij sommige kinderen (en in sommige metingen) de klinisch belangrijke veranderingen ook worden beschouwd als "echte" veranderingen. Voor de meerderheid van de kinderen met USCP kunnen de belangrijke veranderingen niet van metingsfout worden onderscheiden. Door het gebrek aan informatie over de mogelijkheden van spierkrachtwinst van de "minder-aangedane" BE als gevolg van training, kunnen geen harde conclusies worden getrokken met betrekking tot de vraag of het mogelijk is om klinisch belangrijke en/of statistisch significante veranderingen te meten binnen een kind.

Om de kans op meetfouten te verkleinen, raden we aan om ons gestandaardiseerde meetprotocol te gebruiken, de metingen door dezelfde geschoolde beoordelaar uit te laten voeren en meerdere keren binnen één meetmoment te meten, waarbij de gemiddelde waarde van de metingen wordt gemeten. Dit minimaliseert de kans dat de verschillen in spierkracht worden veroorzaakt door verschillen in uitgangshouding, verschillen in plaatsing van het meetinstrument (HHD) of verschillen in de hantering van het meetinstrument (E-link).

Hoofdstuk zeven bevat de algemene discussie, waarin de belangrijkste resultaten worden geëvalueerd. Ook worden de methodologische overwegingen besproken en implicaties voor de klinische praktijk en toekomstig onderzoek gepresenteerd.

Aangezien de HHD, E-link, maatbeker-taak en krat-taak allemaal een grote SDC-waarde hebben, hebben we ook bediscussieerd of de SDC-waarde, berekend/bepaald volgens de huidige methode, geschikt is voor spierkracht-meetinstrumenten gebruikt in een (heterogene) revalidatie populatie. Daarom hebben we een overzicht gemaakt van de SDC-waarden van de HHD en E-link in andere populaties, en van de SDC-waarden van andere meetinstrumenten die spierkracht meten bij kinderen met USCP. De resultaten tonen aan dat de SDC-waarde hoog is, ongeacht het gebruikte meetinstrument of de onderzochte populatie. Alternatieve manieren om de SDC-waarde te berekenen moeten worden overwogen. Voorgestelde alternatieven zijn: bereken de SDC-waarde als percentage; of neem meerdere metingen binnen één persoon en gebruik het verschil tussen de hoogste en de laagste spierkrachtwaarde om de kindspecifieke SDC-waarde te bepalen.

Verder onderzoek over dit onderwerp is zeker nodig. Totdat er meer duidelijkheid is over alternatieve methoden voor het berekenen van de SDC-waarde, wordt aanbevolen om extra spierkracht-meetinstrumenten te gebruiken. Wanneer meerdere meetinstrumenten (bij voorkeur op verschillende ICF-CY niveaus) allemaal winst laten zien tussen de pre-/post-interventiemetingen, is de kans groter dat de interventie een "reëel" positief resultaat heeft.

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# About the author

Koen Dekkers was born on November 6th, 1978 in Roosendaal, the Netherlands. He attended Senior general secondary education (HAVO) at Gertrudis Lyceum, Roosendaal, where he obtained his diploma in 1997. From 1997 to 2001 he studied physiotherapy at Hogeschool Brabant (nowadays Avans) in Breda.

After obtaining his physiotherapy diploma he started working as a physiotherapist at Revant rehabilitation centre in Breda at the pediatric rehabilitation department. In 2003 he started the "Pediatric Physical Therapy" training at Avans+, Breda, after which he graduated as the first male Master Pediatric Physiotherapist in 2007. After graduation he remained involved as a (part-time) teacher.

During his career at Revant, he was involved in multiple external research programmes. In 2011 he got the opportunity to start his own (external/part-time) PhD trajectory at the Maastricht University Rehabilitation Medicine department, in addition to his job at Revant. He was supervised by Prof. dr. Rob Smeets, Dr. Yvonne Janssen-Potten and Dr. Eugene Rameckers. During (and before) the PhD trajectory Koen Dekkers followed multiple courses, like "quantitave data-analysis" (Open University; 2006/2007) "introduction in epidemiology" (Maastricht University; 2013), Clinimetrics Assessing Measurement Properties of Health Measurement Instruments (EpidM/VuMC 2015), Regression techniques (EpidM/VuMC 2016) and the BROK (NFU, 2017) in order to obtain more knowledge about scientific research.