Measuring changes of manual ability with ABILHAND-Kids following intensive training for children with unilateral cerebral palsy

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ABBREVIATIONS

AHA	Assisting Hand Assessment
CHEQ	Children's Hand-use Experience Questionnaire
CIMT	Constraint-induced movement therapy
HABIT	Hand-arm Bimanual Intensive Therapy
HABIT-ILE	Hand and Arm Bimanual Intensive Therapy Including the Lower Extremities
JTTHF	Jebsen-Taylor Test of Hand Function
MACS	Manual Ability Classification System
PEDI	Pediatric Evaluation of Disability Inventory

AIM ABILHAND-Kids is a parent-reported questionnaire measuring manual ability in children with cerebral palsy (CP). Its psychometric properties have been established, with the exception of responsiveness, which is examined here.

METHOD In this cohort study, 98 children (46 males, 52 females; range 6–19y, mean 11y, standard deviation [SD] 3.3y) with unilateral CP underwent three assessments of upper extremity function: at baseline (T1); after 80 to 90 hours of intensive training (T2); and at follow-up (T3). The responsiveness was analyzed using global, group (based on age and on Manual Ability Classification System [MACS] level), and individual approaches during two time periods (T1–T2 and T2–T3). Effect size was used to quantify magnitude of changes.

RESULTS The global approach showed significant improvements between T1 and T2 (p<0.001) but not between T2 and T3 (p=0.222). In the group analyses, effect size and SRM demonstrated large changes in younger children (6–12y, n=52, mean change=1.06 logit, effect size >0.8) and small changes in the older children (13–19y, n=46, mean change=0.71 logit, effect size >0.4). Children in MACS level II demonstrated larger changes than children in MACS level I or III.

INTERPRETATION The ABILHAND-Kids exhibited responsiveness in detecting changes after intensive training. Therefore, this scale is potentially useful in assessing the functional status of children with unilateral CP in clinical trials.

Cerebral palsy (CP) is the most common motor disability in childhood, frequently leading to limitations both in gross motor function and manual abilities.[1] To measure these limitations, an evidence-based approach requires the development of relevant and reliable assessment tools able to detect clinically significant changes over time. Measuring changes in bimanual ability remains a challenge in children with CP because there are only a few tools that are both CP-specific and demonstrate good psychometric qualities. Notably, these tools include the Assisting Hand Assessment (AHA),[2] the Children's Hand-use Experience Questionnaire (CHEQ),[3] and the ABILHAND-Kids.[4] Among these, the AHA is the only measure whose responsiveness has been studied.[5]

The ABILHAND-Kids questionnaire is a Rasch-built tool measuring the manual ability of children with all types of CP. It includes 21 specific items and has been developed in a process where the construct validity, test–retest reliability, linearity, and unidimensionality were verified (Appendix S1, online supporting information).[4] Parent-report questionnaires measuring functional abilities of children have been recognized as important in the assessment of clinical changes, especially because children do present a more dichotomous perception of their own abilities, potentially allowing measurement of capacity but not daily performance.[4,6] While validity and reliability have been demonstrated for ABILHAND-Kids[4] (Appendix S1), its responsiveness (i.e. its ability to detect changes over time[7]) remains unknown.

Over the past decade, intensive interventions developed to improve the manual ability of the children with unilateral spastic cerebral palsy (USCP) have demonstrated positive outcomes, providing potential to study the ABILHAND-Kids responsiveness in detecting changes. Constraint-induced movement therapy (CIMT) – where the use of the more affected upper extremity is induced by restraining the less affected side and providing repetitive and intensive training – was the first efficacious intensive treatment.[8] Based on the need of combined use of both hands to increase functional independence in everyday life, intensive bimanual training (e.g. Hand-arm Bimanual Intensive Therapy '[HABIT]') was subsequently introduced.[9] The efficacy of bimanual training has been recently demonstrated, with comparisons of CIMT and bimanual training showing positive and largely similar effects.[10] Recently, Hand and Arm Bimanual Intensive Therapy Including the Lower Extremities (HABIT-ILE) was introduced, where bimanual training is targeted concomitantly with engagement of postural and lower extremity control.[11] Its efficacy has been demonstrated for both upper extremity and lower extremity, as well as in social participation in children with USCP.[12] These interventions are all based on motor learning principles (practice specificity, types of practice, feedback), and apply principles of neuroplasticity (practice-induced brain changes arising from repetition, increasing movement complexity, motivation, and reward).[13]

The aims of this study were to examine: (1) the responsiveness of the ABILHAND-Kids following intensive interventions; (2) whether the responsiveness of ABILHAND-Kids changes according to age or hand function level; and (3) whether ABILHAND-Kids changes correlate with changes in other bimanual assessment tools.

What this paper adds

- Good responsiveness of ABILHAND-Kids during intensive motor skill learning interventions was demonstrated.
- Larger mean ABILHAND-Kids changes in younger children than in adolescents were detected; larger mean ABILHAND-Kids changes in children with Manual Ability Classification System (MACS) II than in children with MACS I or III were highlighted.
- Higher responsiveness in ABILHAND-Kids than in other tools measuring manual skills (the self-care part of the Pediatric Evaluation of Disability Inventory and the Assisting Hand Assessment) was observed.

METHOD

This retrospective study includes 98 children (46 males, 52 females) with USCP who participated in three different cohort studies: Columbia University, NY (*n*=25);[14] Adelante Rehabilitation Center/Maastricht University (*n*=52);[15] and Université catholique de Louvain (*n*=21).[12] Ethical approval was obtained in the respective review boards of the universities. These studies were chosen because all children (age 6–19y, characteristics in Table I) were involved in a motor skill learning intensive rehabilitation camp; selected on common criteria (inclusion: ability to grasp light objects and lift the more affected arm 15cm above a table surface/typically developing school level/ability to follow instructions and complete testing; exclusion: uncontrolled seizures/orthopaedic surgery in the upper extremities within the previous 12mo or planned within the study period/visual problems likely to interfere with treatment/assessment); and assessed using the ABILHAND-Kids questionnaire.

Outcome measurements

Changes were systematically investigated during intervention period (T1, baseline, to T2, directly after intensive intervention) and at the follow-up (T3) where manual ability is expected to be stable. All children of this study were assessed with the parent-reported ABILHAND-Kids questionnaire and were classified using the Manual Ability Classification System (MACS).[16] In addition, those aged between 6 years and 12 years were assessed using the AHA[2] and the Pediatric Evaluation of Disability Inventory (PEDI, self-care subscale).[17] The adolescents (≥13y) were tested using the Jebsen–Taylor Test of Hand Function (JTTHF)[18] in addition to the ABILHAND-Kids (Appendix S1).

The ABILHAND-Kids questionnaire was completed by the parents.[4] Parents were instructed to rate their child's

Table I: Characteristics of the participants

Age	Mean (SD)	Median (range)
Mean age SD (y)	11.8 (3.3)	12 (6–19)
Children 6–12y (*n*=52)	9.1 (1.9)	9 (6–12)
Children 13–19y (*n*=46)	14.8 (1.4)	15 (13–19)
Hemiparetic side		
Right hemiparesis	54	
Left hemiparesis	44	
MACS		
Level I	26	
Level II	42	
Level III	29	
Level IV	1	
Intervention		
Modified CIMT	52	
HABIT	25	
HABIT-ILE	21	
MACS by age		
Children 6–12y		
MACS I	10	
MACS II	31	
MACS III–IV	11	
Children 13–19y		
MACS I	15	
MACS II	12	
MACS III	19	
MACS by intervention		
Modified CIMT		
MACS I	18	
MACS II	12	
MACS III	22	
HABIT		
MACS I	5	
MACS II	13	
MACS III-IV	7	
HABIT-ILE		
MACS I	3	
MACS II	17	
MACS III	1	

SD, standard deviation; MACS, Manual Ability Classification System; CIMT, constraint-induced movement therapy; HABIT, Hand-arm Bimanual Intensive Therapy; HABIT-ILE, Hand and Arm Bimanual Intensive Therapy Including the Lower Extremities.

perceived difficulty in performing each activity on a three-level response scale (impossible, difficult, easy). Parents were asked to take into account the performance of their child when performing the activity without technical or human assistance regardless of the limb(s) and the strategies used (Appendix S1).

Data analysis

The responsiveness was evaluated using global, group, and individual approaches.

The global approach was a statistical comparison using a repeated measures analysis of variance (RM ANOVA; T1 vs T2 vs T3). Newman–Keuls post-hoc comparisons with automatic correction for multiple comparisons were used.

In addition, the effect size was used to characterize the changes observed. The effect size consists of a ratio between the mean change in scores and the standard deviation of the change in scores,[19] and was interpreted using Cohen's benchmarks:[20] magnitude of the change was considered as small (0.2), moderate (0.5), or large (0.8).

Group approaches were also conducted to determine whether some subgroups would demonstrate greater responsiveness.

In an initial analysis, two groups were formed based on the child's age at the baseline assessment (6–12y, *n*=52; 13–19y, *n*=46), to separate adolescents from younger children, who according to the Kennard principle may encounter more neuroplasticity. Puberty was considered as starting in mean at 13 years, while early and late onset puberty is not uncommon in CP.[21] In a second analysis, three groups were formed based on the MACS level of the children scored at baseline (MACS I, *n*=26; MACS II, *n*=42; MACS III, *n*=29; MACS IV, *n*=1, excluded from this analysis). Effect size was used to determine magnitude of the changes in the three subgroups.

Finally, as it may be interesting to track changes for an individual (changes that have a meaning in groups could not be meaningful in individuals),[22] an individual approach was used, allowing us to evaluate changes in manual ability in each child using a *t*-value. The *t*-value is a self-relative score taking into account the score and its associated error for one person at two different time-points. It was calculated as:[23]

$$t = \frac{(\beta_{v2} - \beta_{v1})}{\sqrt{(\mathrm{SE}_{v2}^2 + \mathrm{SE}_{v1}^2)}}$$

where β_{v2} and β_{v1} represent the patient's manual ability at T2 and T1; SE_{v2} and SE_{v1} being their respective associated standard errors of measurement. This *t*-value indicates whether the change observed between two assessment times for a given child reflects more than the fluctuation of the measuring instrument.[23] Following individual calculation of the *t*-value (T1–T2 period), children were divided into five classes of clinical significance: clinically important improvement (t>1.96); improvement (0>t>1.96); no change (t=0); deterioration (–1.96<t<0); and clinically important deterioration (t<–1.96).

Finally, changes observed in the ABILHAND-kids were correlated (Pearson or Spearman correlation) with changes observed in other tools concomitantly used.

RESULTS

From the 98 children included, two dropped out between T2 and T3 as they did not appear for the follow-up assessment. These data were considered as missing values.

Global approach

Table IIa shows the results of the global approach for the ABILHAND-Kids scores. Using a one-way RM ANOVA, the global approach showed significant differences. The

Table II: Responsiveness analysis using a global and group approach

	T1 mean (SD)	T2 mean (SD)	T3 mean (SD)	RM ANOVA			Effect Size	
(a) Global approach				Main effect	T1 vs T2	T2 vs T3	T1 vs T2	T2 vs T3
ABILHAND-Kids (*n*=98)	2.93 (1.97)	3.95 (2.02)	4.12 (1.98)	*F*=31.22; *p*<0.001	*p*<0.001	*p*=0.222	0.596	0.139
(b) Group approach by age								
Younger (6–12y) (*n*=46)								
ABILHAND-Kids (logits)	2.13 (1.75)	3.19 (2.01)	3.34 (1.83)	*F*=29.89; *p*<0.001	*p*<0.001	*p*=0.263	0.916	0.158
AHA (% of logits – AHA units)	63 (12.9)	68 (13.7)	68 (13.2)	F=15.437; *p*<0.001	*p*<0.001	*p*=0.789	0.758	0.047
PEDI (functional care) (raw scores)	60(11.9)	66 (10.9)	67 (10.5)	*F*=33.755; *p*<0.001	*p*<0.001	*p*=0.093	0.877	0.262
Adolescents (13–19y) (*n*=52)								
ABILHAND-Kids (logits)	4.07 (1.56)	4.78 (1.67)	4.99 (1.75)	*F*=9.202; *p*<0.001	*p*<0.001	*p*=0.535	0.437	0.119
JTTHF more affected hand (sec)	201 (134)	190 (122)	187 (128)	*F*=0.586; *p*=0.558			−0.051	0.119
JTTHF less affected hand (sec)	42 (17)	46 (51)	51 (73)	*F*=0.331; *p*=0.719			0.071	0.049
(c) Group approach by MACS levels								
MACS I (*n*=26)								
ABILHAND-Kids (logits)	4.09 (1.63)	4.76 (1.64)	4.98 (1.68)	*F*=7.264; *p*=0.002	*p*=0.008	*p*=0.357	0.616	0.172
MACS II (*n*=42)								
ABILHAND-Kids (logits)	2.33 (1.70)	3.60 (1.86)	3.72 (1.68)	*F*=19.788; *p*<0.001	*p*<0.001	*p*=0.633	0.653	0.089
MACS II (*n*=42)								
ABILHAND-Kids (logits)	2.98 (2.02)	3.91 (2.20)	4.12 (2.24)	*F*=6.235; *p*=0.004	*p*=0.012	*p*=0.454	0.513	0.165

RM ANOVA, repeated measures analysis of variance; AHA, Assisting Hand Assessment; PEDI, Pediatric Evaluation of Disability Inventory; JTTHF, Jebsen–Taylor Test of Hand Function; MACS, Manual Ability Classification System; SD, standard deviation; T, testing session. For effect size and standardized response of the mean scores: ■ 0.2 small change; ■ 0.5 moderate change; ■ 0.8 large change.

post-hoc test highlighted significant differences between T1 and T2 scores (p<0.001) but not for T2 versus T3 (p=0.263). In addition, the effect size measured on the whole sample (n=98) presented moderate changes (Table II). The global approach could only be applied to the ABILHAND-Kids (only assessment completed by all children).

Group approach

Groups based on age. Based on the hypothesis that age may have an impact on the amount of improvement, the sample was divided in two groups: younger children (6–12y) and adolescents (13–19y). As shown in Table IIb, during the intervention period (T1–T2), the mean change in manual ability was higher in the younger (1.06 logits) than in the older group (0.71 logits). This was confirmed by the effect size where the changes detected were categorized as large for the younger group and small for the adolescent group.

Other tools used to measure manual ability during the intervention period, (i.e. the AHA and the self-care subtest of the PEDI for the youngest children and the JTTHF for the adolescents) are also shown in Table IIb. For the youngest children, the AHA demonstrated moderate changes, while the PEDI showed large changes.

In the adolescent group, no changes could be detected using the JTTHF in both hands. During the follow-up period, few changes were observed in both groups. Only the PEDI showed small changes as measured with the effect size in the youngest group (Table II).

Groups based on MACS level. Table IIc shows the responsiveness of the ABILHAND-Kids scale in groups based on the various MACS levels. Groups of MACS I and III presented a moderate change. Children with a MACS II presented a higher value than the two other groups, with changes that can be described as moderate.

Individual approach

Table III shows the percentage of children in the sample demonstrating changes in each t-value category. Following intervention, most of the children improved on the ABILHAND-Kids score (70%), with 19% showing an important improvement (t>1.96) and 51% presenting an improvement (1.96>t>0); 12% did not change; 15% tended to decline; and 3% presented an important decline.

Figure 1 shows the percentage of children with different t-values in each age group. A larger percentage of younger children did demonstrate ‘clinically important improvement’ (22%) and ‘improvement’ (59%), compared to the adolescent group (‘clinically important improvement’ 4%; ‘improvement’ 52%). The adolescents showed a larger percentage in the ‘no change’ category (20% vs 5%), but did not show larger percentages in the ‘significantly decreased group’ (2% in each age group).

Figure 2 shows the percentage of children with different t-values in each MACS group. A larger percentage of children with a MACS II demonstrated ‘clinically important improvement’ (30%), compared to the two other levels (‘clinically important improvement’ 4% and 14% in MACS I and III respectively). A higher percentage of children in the MACS I and III groups was observed in the ‘improvement’ (58% and 54% for MACS I and III respectively, vs 47% in MACS II) and ‘no change’ categories (27% and 14% respectively, vs 2% in MACS II). MACS I and III did not have many participants in the ‘clinically important deterioration group’ (0% for MACS I and III, vs 5% in MACS II).

Table III: Responsiveness analysis/individual approach (t-values)

		T1–T2 (%)	T2–T3 (%)
t>1.96	Clinically important improvement	19	7
1.96>t>0	Improvement	51	29
0	No change	12	31
0>t>–1.96	Deterioration	15	32
t<–1.96	Clinically important deterioration	3	1

T, testing session; t, t-value.

Correlations

To assess further the age effect that was highlighted by the group approach, individual changes in t-values during intervention (T1–T2) were correlated with the age of the children. A small but significant correlation was observed (n=98, r=–0.215, p=0.030) showing that changes in t-values decrease as age increases.

Correlations were also computed between changes in the ABILHAND-Kids scores and changes in other manual skills tools during intensive intervention (T1–T2).

In the younger group, while a moderate correlation was found between changes in the PEDI and in the ABILHAND-Kids (Spearman, r=0.430, p=0.003), no correlation was found between AHA-changes and ABILHAND-Kids changes (Pearson, r=–0.104, p=0.493).

In the older group no significant correlations were seen between the changes in the ABILHAND-Kids scores and the changes in the JTTHF performance in either hand (r=–0.025, p=0.860, and r=–0.046, p=0.751, for the more affected and less affected hand respectively).

DISCUSSION

This study investigated the responsiveness of the ABILHAND-Kids in the context of intensive interventions for children with USCP. Responsiveness of the ABILHAND-Kids was demonstrated by all approaches; however, the magnitude of changes was larger in younger children and in children with a MACS level II.

The question of the age at which children are most responsive to intensive treatments is critical. Previously, similar changes in the JTTHF have been observed for children aged 4 to 8 years and 9 to 13 years following CIMT.[24] In the present study, a difference in responsiveness was observed between the two age groups (i.e. large changes in the younger children [6–12y] and small changes in the adolescents [13–19y]). One possible explanation,

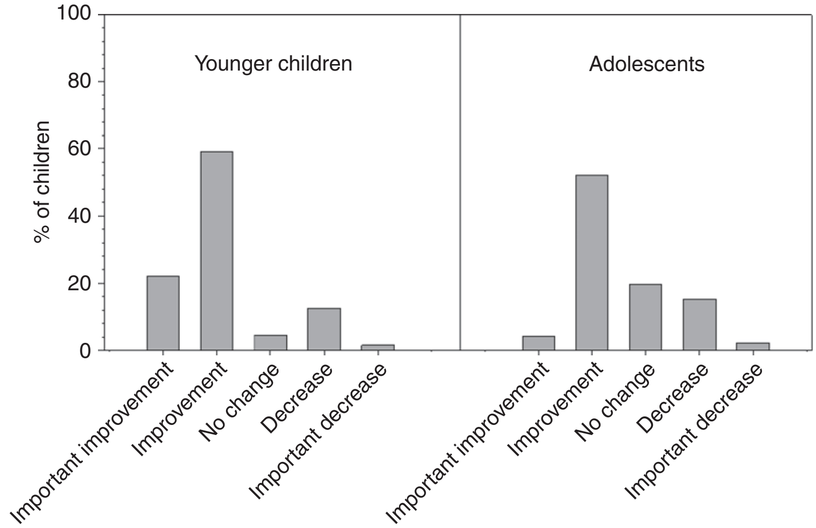

Figure 1: The percentage of children in each *t*-value category, based on their individual ABILHAND-Kids change following intervention. Five classes are represented for each age group regarding their *t*-value significance: clinically important improvement (t>1.96); improvement (0>t>1.96); no change (t=0); deterioration (–1.96<t<0); and clinically important deterioration (t<–1.96). Percentages are represented here according to the two age groups: younger (6–12y) and adolescents (13–19y).

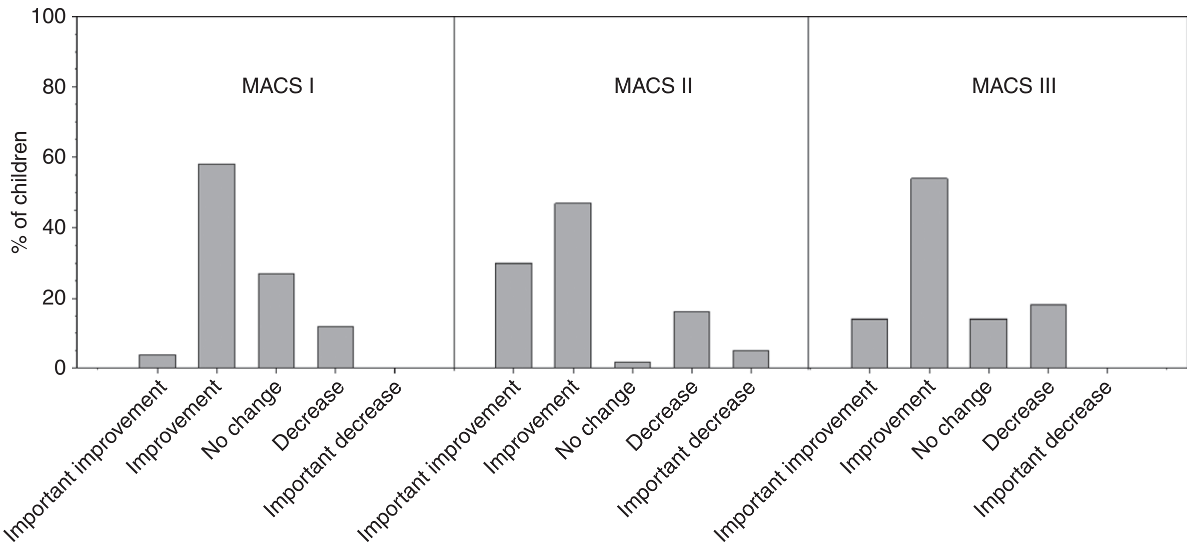

Figure 2: The percentage of children in each *t*-value category, based on their individual ABILHAND-Kids change following intervention. Five classes are represented for each Manual Ability Classification System (MACS) level group regarding their *t*-value significance: clinically important improvement (t>1.96); improvement (0>t>1.96); no change (t=0); deterioration (–1.96<t<0): and clinically important deterioration (t-value<–1.96). Percentages are represented here according to the three MACS levels (I, II, and III).

consistent with the general idea of the 'Kennard Principle',[25] might be that neuroplasticity is lower in adolescents and that less manual ability improvement can be obtained in this group over a 2 to 3 weeks' intervention in comparison to younger children. While it has previously been suggested that older children may encounter reduced changes because of less plasticity of the brain, notably during CIMT,[26,27] opposite findings were proposed by Sakzewski et al.,[28] suggesting that best responders were older children. However, the sample of children from the latter study had a mean age of around 10 years (with a confidence interval at 95% reaching less than 12y). This means that 'older' children of this study match our 'younger group'. So it seems congruent that adolescents might respond less to intensive interventions. Alternatively, while it is currently used in adolescents because of its unique psychometric qualities (valid, reliable, linear, and unidimensional measure of manual ability), the ABILHAND-Kids has been calibrated in children from 6 to 15 years. Therefore the tool might be less sensitive for children from 15 years. Another possible explanation is that older children develop habits based on assistance provided by caregivers or compensations, which are more difficult to change than in younger children.

In younger children, where a high responsiveness was found, the ABILHAND-Kids provided higher responsiveness than tools concomitantly used. This shows the potential importance of using this short questionnaire in addition to the AHA, currently considered as the criterion standard for measuring manual function in children with USCP.[29] The interest in using both instruments is further highlighted by the absence of correlation between changes in the ABILHAND-Kids and changes in the AHA. Thus, these tools do not measure the same aspect of manual function. The parent-reported ABILHAND-Kids provides information on actual everyday manual ability (i.e. autonomy of all children with CP in manual activities),[4] and the AHA provides specific information on the quality of bimanual manipulation and the effectiveness of the child to use their more impaired hand in activities requiring skilled bimanual abilities. The results obtained in this study emphasize the need to use both measures to capture changes in manual function in children with USCP.[2,5] In the future, it would be interesting to test together the changes in the ABILHAND-Kids[4] and another questionnaire, the CHEQ,[3] which has a different measurement goal than ABILHAND-Kids. Indeed, it is likely that these two scales do not capture the same variable, as the CHEQ specifically evaluates the experience of children (i.e. grasp efficacy, time taken to perform the activity, and degree of feeling bothered) with decreased function in one hand in using the affected hand in activities where usually two hands are needed. Furthermore, the CHEQ is scored by children and/or parents, while the ABILHAND-Kids is scored by parents only. As it is known that children can assess their own ability accurately but dichotomously ('I can do' or 'I cannot'),[4] the CHEQ is more likely to capture a capacity measure, while the ABILHAND-Kids is more likely to be graded regarding everyday life performance. Comparing these two well-designed tools for capturing upper extremity activity would be needed in the future to assess whether they measure similar or different, and potentially complementary, variables.

An absence of correlation was also observed in older children between changes in the ABILHAND-Kids and in the JTTHF. This highlights the different focus of the tests. JTTHF is more focused on dexterity per se, without actual context of everyday life (simulation of different tasks). Dexterity has been demonstrated in a large sample as related – but not strongly – to manual ability in children with CP.[30] In addition, while ABILHAND-Kids provided responsiveness even in that group, our results suggest that specific tools should be calibrated for adolescents.

When groups were considered regarding the MACS level, children in MACS II systematically exhibited higher indices than those in MACS I and III. It has generally been suggested that children with a higher deficit (higher MACS level) are likely to be best responders compared to children with mild (typically MACS level I) deficit.[28] However, the fact that children in a low MACS level (III is usually the lowest level for a child with USCP) are equally trainable as children with MACS I is of interest: while a bit less responsive than children with MACS II, these children may benefit from intensive interventions. Moreover, both in MACS I and III, when taking into account their individual evolution, no children exhibited an 'importantly decreased' score after the intensive interventions. Most of them (>70%) improved following the camp intervention.

The individual approach, using *t*-value, also showed that the percentage of children demonstrating improvements in ABILHAND-Kids is higher in younger children than in adolescents. It also demonstrated that while the phenomenon concerns a small percentage of children, some of them may not respond to intensive interventions. Future studies should focus on the characteristics of these non-responding children.

Limitations of the study

The retrospective aspect of this study may have introduced some bias because the timing of the follow-up assessment differed slightly between the studies and the number of children involved in each study was different. While these limitations impair the possibility of comparing these different motor-skill learning interventions, they are not likely to decrease the responsiveness of a tool used to measure their effect on manual ability. A prospective randomized design (HABIT, HABIT-ILE, CIMT) would have allowed, in addition, a comparison of the interventions.

This study included only children with UCP while the ABILHAND-Kids scale has been designed for a group of children with all subtypes of CP. Therefore the responsiveness in other subgroups of CP cannot be extrapolated from these results. We also drive the attention of the users to the importance of asking the same parent/caregiver to answer the questionnaire at different time-points to assess changes, in order to avoid bias. In studies with a long-term follow-up, it might be useful to check with parents if the environment remained stable over time (i.e. having a new lunch box might induce differences in the opening system).

CONCLUSION

The ABILHAND-Kids questionnaire exhibited responsiveness in detecting changes after intensive training in children with USCP. Therefore, this scale is useful in assessing the functional status of children with USCP in such clinical trials. Differences of responsiveness have been observed, notably regarding age of the children, which suggests the need to build new tools dedicated for use in adolescents.

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SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Measuring changes of manual ability with ABILHAND-Kids following intensive training for children with unilateral cerebral palsy.

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