Unimanual and bimanual dexterity in unilateral cerebral palsy: psychometric properties of the Tyneside Pegboard Test and association with sensorimotor impairments

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Abbreviations

uCP	unilateral Cerebral Palsy
TPT	Tyneside Pegboard Test
MM	Mirror Movements
UNI_{large}	Unimanual TPT task with large pegs
UNI_{medium}	Unimanual TPT task with medium pegs
UNI_{small}	Unimanual TPT task with small pegs
BI_{MI-LI}	Bimanual TPT task more-impaired to less-impaired hand
BI_{LI-MI}	Bimanual TPT task less-impaired to more-impaired hand
JTHFT	Jebsen-Taylor Hand Function Test
AHA	Assisting Hand Assessment
CHEQ	Children's Hand-use Experience Questionnaire
MAS	Modified Ashworth Scale

ABSTRACT

AIM We explored the psychometric properties of the recently developed Tyneside Pegboard Test (TPT) for unimanual and bimanual dexterity in children with unilateral cerebral palsy (uCP) and investigated the impact of sensorimotor impairments on manual dexterity.

METHOD In this cross-sectional study, the TPT was assessed in 49 children with uCP (mean age 9y 8mo, SD 1y 11mo, 30 males, 23 right uCP). All participants additionally underwent a standardized upper limb evaluation at body function and activity level. We investigated (1) known-group, concurrent and construct validity and (2) impact of sensorimotor impairments including spasticity, grip force, stereognosis and mirror movements using ANCOVA, Spearman's rank correlation (r) and multiple linear regression (R^2), respectively.

RESULTS TPT outcomes significantly differed according to the Manual Ability Classification System ($p<0.001$, know-group validity). Relationships were found between the unimanual TPT tasks and the Jebsen-Taylor hand function test (r=0.86-0.88, concurrent validity). Bimanual TPT tasks were negatively correlated with the Assisting Hand Assessment, ABILHAND-kids and Children's Hand-use Experience Questionnaire (r=-0.38-(-0.78), construct validity). Stereognosis was the main determinant influencing all tasks ($p<0.001$, R^2=37%-50%). Unimanual dexterity was additionally determined by grip strength ($p<0.05$, R^2=8%-9%) and mirror movements in the more-impaired hand ($p<0.05$, R^2=4%-8%). Bimanual dexterity was also explained by mirror movements in the more-impaired hand ($p<0.01$, R2=10%-16%) and spasticity ($p=0.04$, R^2=5%).

INTERPRETATION The TPT is a valid test to measure unimanual and bimanual dexterity in uCP. The results further underline the importance of somatosensory impairments in children with uCP.

What this paper adds:

- The TPT is valid to measure unimanual and bimanual dexterity in uCP.
- Children with a poorer manual ability show worse unimanual and bimanual dexterity.
- Stereognosis is the main predictor of both unimanual and bimanual dexterity.
- Stronger mirror movements in the more-impaired hand result in worse bimanual dexterity.

Keywords: Cerebral palsy, upper limb, dexterity, sensorimotor impairments, manual ability

Shortened title: Unimanual and bimanual dexterity in uCP

Children with unilateral cerebral palsy (uCP) experience sensorimotor impairments[1], which are often more prominent in the upper limb compared to the lower limb.[2] Such sensorimotor impairments may compromise the development of manual dexterity[3], the ability to perform fast coordinated movements[4], which is crucial for performing everyday activities.

Recently, a quantitative tool was developed specifically for children with CP, to assess unimanual and bimanual dexterity, namely the Tyneside Pegboard Test (TPT).[5] The TPT is able to detect differences in unimanual and bimanual dexterity between typically developing children and children with uCP.[5] However, whether the TPT can also discriminate between children with uCP with different levels of manual ability, or known-group validity, is not yet investigated. Also concurrent validity and construct validity has not yet been fully examined.[5] Hence, further investigation of the psychometric properties of this test is needed.

Furthermore, an in-depth investigation of the influence of sensorimotor impairments on unimanual and bimanual dexterity is warranted as the development of dexterity depends on the sensorimotor experiences in early life[3], which are limited in children with uCP. Thus far, it has been shown that unimanual dexterity is related to grip strength[6],stereognosis[6,7] and spasticity[8]. Other studies have shown that mirror movements (MM) may impair how children with uCP use their more-impaired hand during the performance of bimanual tasks.[9] Finally, exteroception has shown to be a determining factor of treatment outcomes of unimanual dexterity after constrained-induced movement therapy.[10] However, it is not yet studied to what extent all these sensorimotor impairments affect both unimanual and bimanual dexterity. Hence, investigating the combined impact of these sensorimotor impairments on unimanual and bimanual dexterity will deepen our insights into the factors underlying manual dexterity. Such insights will aid to individualize treatment planning in terms of focussing on specific influencing factors.

We aimed to examine psychometric properties of the TPT establishing known-group validity, concurrent validity and construct validity and to investigate to what extent unimanual and bimanual dexterity are influenced by motor (spasticity, grip strength and MM) and somatosensory (exteroception and stereognosis) impairments. We hypothesized that unimanual and bimanual dexterity would be mostly influenced by grip strength and stereognosis, but bimanual dexterity would also be determined by the presence of MM.

METHOD

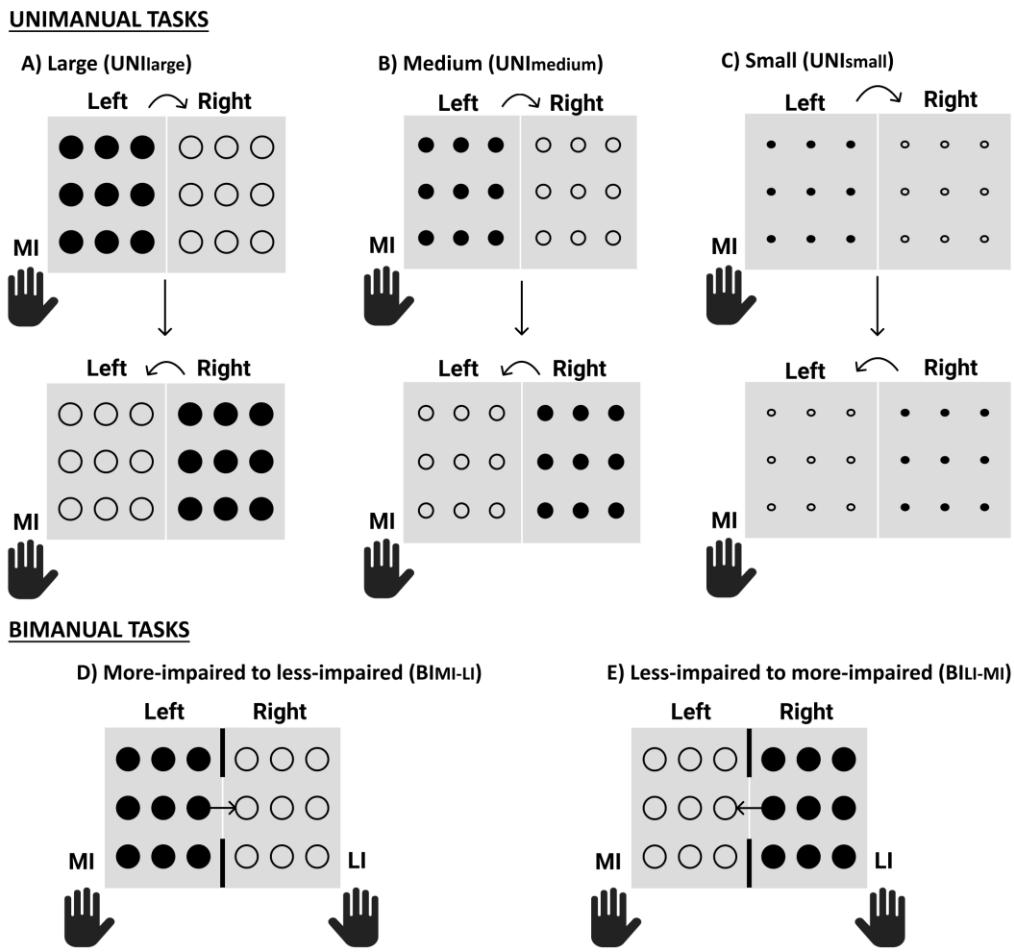
PARTICIPANTS

This cross-sectional study included children with uCP aged 6 to 15 years from the CP care program of the University Hospitals Leuven. For inclusion, children had to be (1) capable to comprehend the test instructions and cooperative to complete the tasks and (2) able to grasp and stabilize an object with the more-impaired hand (≥4 on the Modified House Functional Classification[11]). Children were excluded if they had received botulinum toxin to the upper limb in the 6 months prior to testing or had undergone upper limb surgery in the 2 years before testing. All parents gave written consent and children assented, in accordance with the Declaration of Helsinki. The study was approved by the Ethics Committee Research UZ/KU Leuven (S55555 and S56513).

ASSESSMENTS

We used the TPT (Newcastle University, Newcastle upon Tyne, United Kingdom) to measure unimanual and bimanual dexterity[5], where nine pegs were moved from one board to an adjacent board as quickly as possible (Figure 1). A lower score on the TPT presents a faster and better performance.[5] Normative data of 974 participants from 4-80 years is available. Moreover, moderate test-retest reliability (ICC between 0.74 to 0.91) and concurrent validity with the Purdue pegboard test (r=-0.61) was established in these healthy participants as well as construct validity in children with uCP (AHA, r=0.63-0.69; ABILHAND, r=0.62-0.65).[5] Unimanual tasks were performed first, in order of decreasing peg sizes (large; medium; small), using the less-impaired hand. A more detailed description of the 3 unimanual tasks (UNI_{large}, UNI_{medium} and UNI_{small}) can be found in figure 1 (A, B and C). If the child could not perform the task with a specific peg size, further testing with smaller peg sizes was not pursued to prevent frustration. Completion time was electronically collected and outputted via custom-written software.[5] In the bimanual condition, only large pegs were picked up one by one with one hand, passed through a hole in a Perspex screen to the other hand and placed in the adjacent board. For this study, completion time in both directions (Fig 1D from the more-impaired to the less-impaired hand (BI_{MI-LI}) and Fig 1E from the less-impaired to the more-impaired hand (BI_{LI-MI})) was used in the statistical analysis.

As with other dexterity tests[12], we implemented a maximum time of completion based on the collected data, for each child that was unable to perform a task or performed slower than this proposed threshold. For each of the five tasks, mean+2SD was calculated. The maximum time was set at 116.38s, 94.51s and 146.63s, respectively for UNI_{large}, UNI_{medium} and UNI_{small}. For BI_{MI-LI} the threshold was 75.48s and for BI_{LI-MI} 167.10s.



Known-group validity investigates a test's ability to differentiate between different groups of a specific characteristic[13] and will be assessed using the level of Manual Ability Classification System (MACS).[14] To assess concurrent validity, children performed the **Jebsen-Taylor hand function test** (JTHFT), measuring unimanual dexterity during six timed tasks*, where a lower score (shorter time) indicates better performance.*[12] Additionally, we included the Assisting Hand Assessment (AHA), the ABILHAND-Kids questionnaire and the Children's Hand-use Experience Questionnaire (CHEQ) for construct validity. The **AHA** evaluates how children with uCP spontaneously use their more-impaired hand during bimanual activities, resulting in 0–100 logit-based AHA units.[15] Next, parents completed the **ABILHAND-Kids questionnaire**, indicating if 21 predominantly bimanual daily activities are 'impossible', 'difficult', or 'easy', and converted to a logit score from -6 to 6.[16] The ***CHEQ*** is a 29 item online form (available at http://www.cheq.se/) assessing the child's experience when using the more-impaired hand in bimanual daily activities. Its 3 subscales measure (1) which hand is used (CHEQ-grip), (2) time needed (CHEQ-time) and (3) if the child feels 'bothered' by the bimanual activity (CHEQ-feeling). The raw score is transformed to a logit scoring from 0-100.[17] A higher score on these three measurements indicates better performance. [12,15–17] Psychometric properties were established for all these assessments. [12,15–17]

To investigate factors influencing dexterity, several impairments were assessed. **Spasticity** was measured with the Modified Ashworth Scale (MAS), providing a score of 0-4 for each muscle group (shoulder adductors and internal rotators, elbow flexors and pronators, wrist flexors, finger flexors and thumb adductors), resulting in a total score from 0 ('no spasticity) to 28 ('highest spasticity).[18] **Grip strength** was measured using the Jamar® dynamometer (Sammons Preston, Rolyan, Bolingbrook, IL, USA). The average of three maximum contractions was used for further analysis.[18] **Sensory function** included exteroception and stereognosis[1], during which the child's vision was occluded. ***Exteroception*** was determined with a reliable clinical test by lightly touching the index finger of the child three times. The child needed to indicate every time the touch was felt, and was graded 0 for absent, 1 for impaired (touch was not felt in one or more attempt(s)), or 2 for intact (all three attempts correct).[18] ***Stereognosis*** was assessed through tactile identification of six familiar objects.[18] A score ranging from 0 to 6 was given, according to the correct number of identified objects. In addition, **MM**, which are involuntary movements in one hand, that mirror voluntary movements in the other hand[19], were quantitatively assessed using the Windmill Task (Behavioural Science Institute, Nijmegen, the Netherlands)[20], which comprises a small windmill connected to an active and passive grip-force transducer. The child had to hold both transducers and repetitively squeeze in the active transducer to make the windmill turn. Mirroring activity were registered through the passive transducer. We calculated, per hand, MM-similarity (i.e. similarity between the two hand movements), and MM-intensity (i.e. strength of the mirroring activity). When the MM-similarity was ≥ 0.30, children were classified as having MM. [20] A detailed explanation of the assessment and MM quantification can be found elsewhere.[20]

STATISTICAL ANALYSIS

General characteristics of the participants were collected such as age, sex and side of hemiplegia and data quality was verified using outlier detection (value > 1.5 interquartile range). When an outlier was detected, clinical reasoning and statistical analyses without the outliers were performed to determine whether they should be removed or not. As the Shapiro-Wilk tests showed no normal distribution of the TPT parameters, non-parametric tests were used for construct and concurrent validity. For known-group validity and influencing factors, one-way analysis of covariance (ANCOVA) and multiple regression with backwards elimination, were used and normal distribution of the residuals was checked and confirmed for each fitted model.[21]

An ANCOVA was used to investigate the known-group validity of the TPT between MACS levels with age as covariates. If the interaction between age*MACS was not significant, the model with the main effects was retained. Bonferroni corrected post-hoc comparisons were computed to investigate differences between the three MACS levels, using a corrected p-value for multiple comparison (α=0.05). Effect sizes were calculated using partial η square (η_p^2) and interpreted as small (0.01-0.06), medium (0.06-0.14) and large (>0.14).[22] Effect sizes of post-hoc comparisons were calculated according to Cohen's d and interpreted as small (0.2-0.5), medium (0.5-0.8) and large (>0.8).[23] Spearman's rank correlation coefficients were used to assess construct and concurrent validity. Correlation coefficients were interpreted as no or little correlation (<0.3), low (0.3-0.5), moderate (0.5-0.7), high (0.7-0.9) and very high (>0.9).[24]

Finally, we investigated the influence of sensorimotor impairments on unimanual and bimanual dexterity using a multiple regression model with backward elimination. As only one independent variable should be included per 10 participants[25], simple linear regression analyses for the continuous variables (age, stereognosis, spasticity, grip strength and mirror movements) and univariate ANOVA for the categorical variable (exteroception) was used to reduce the number of independent variables. Variables with p-value>0.05 on all tasks were not included in multiple regression analysis. Multicollinearity between the independent variables was investigated using the variance inflation factor (VIF), of which a value above 10 indicates multicollinearity.[26] SPSS Statistics 26.0 (IBM, New York, USA) was used for all statistical analyses.

RESULTS

PARTICIPANTS

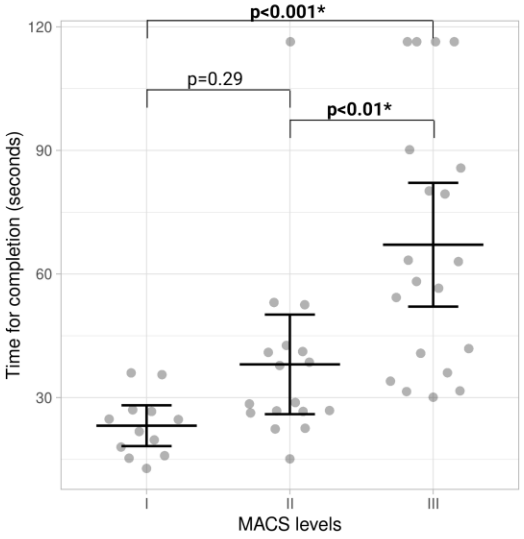
In total, 49 children with uCP (mean age 9y 8mo, SD 1y 11mo, 30 males, 23 right uCP) were included and classified according to their manual ability level (12 MACS I, 17 MACS II and 20 MACS III). Two children with MACS level II were an outlier (child 1 on all TPT tasks and child 2 on $BI_{LI\text{-}MI}$). These were the only two children with a MACS level II who received the implemented threshold due to inability or difficulty with performing the TPT. However, these children were still included in the analysis assuring that the whole spectrum of children with a MACS II classification were involved. According to MM-similarity, 59% children in our sample have MM (36% showed MM in both hands, 10% only in the more-impaired hand and 12% in the less-impaired hand). More information about the distribution of the MM characteristics, can be found in supplementary materials (SM 1). One child did not perform the bimanual tasks of the TPT due to technical problems, whereby only her unimanual TPT tasks were included. In case of missing data for the sensorimotor assessments, children were not included in the multiple linear regression analysis of those tasks, resulting in 43 children for the unimanual TPT tasks and 42 children for the bimanual TPT tasks (SM2). In total, 15 children received the implemented threshold, because they were unable to perform the task and/or because they performed the task in a slower pace than the threshold. A detailed overview regarding this implemented threshold according to the MACS levels is provided in Supplementary Materials (SM 2 and SM 3).

PSYCHOMETRIC PROPERTIES

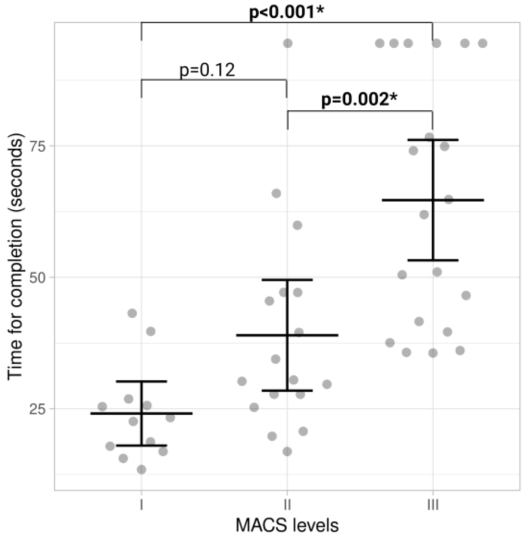
Known-group validity. The interaction between MACS level and age was not significant for any task (p=0.24-0.56) and left out from further analysis. A main effect of MACS levels was found with large effect sizes ($p<0.001$, $\eta_p^2>0.35$) during unimanual (UNI_{large}: $\eta_p^2=0.37$; UNI_{medium}: $\eta_p^2=0.43$; UNI_{small}: $\eta_p^2=0.57$, Fig. 2) and bimanual tasks ($BI_{MI\text{-}LI}$: $\eta_p^2=0.37$; $BI_{LI\text{-}MI}$: $\eta_p^2=0.35$). Post-hoc comparisons showed that children in MACS III performed significantly worse than children in MACS I ($p<0.001$) and MACS II ($p<0.05$) for all TPT tasks. No significant differences were found between MACS I and MACS II, except for the UNI_{small} task ($p=0.04$). An overview of the results is provided in Figure 2 and in the Supplementary Materials (SM 4).

UNIMANUAL TASKS

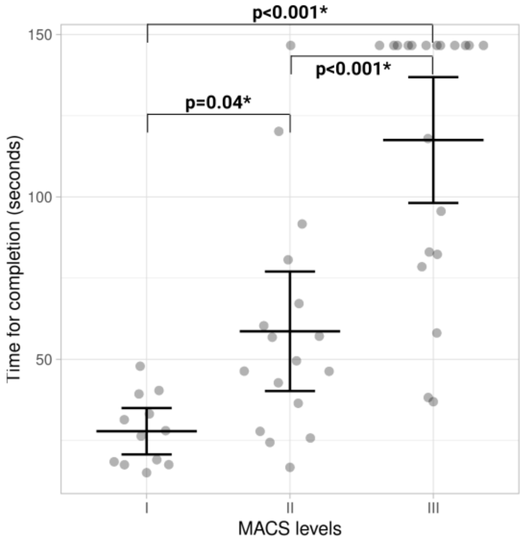
A) Large (UNI_{large})



B) Medium (UNI_{medium})

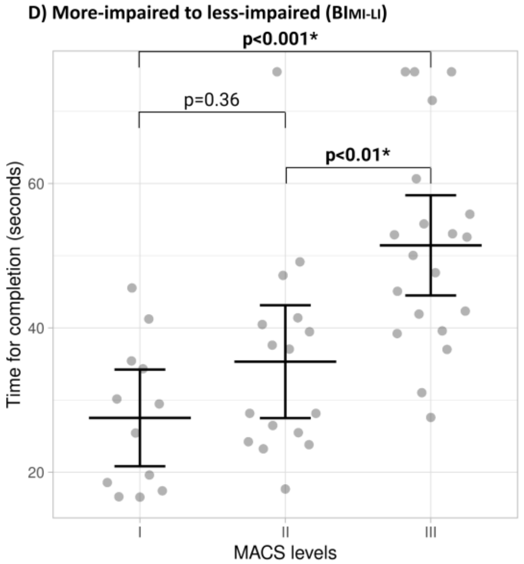


C) Small (UNI_{small})

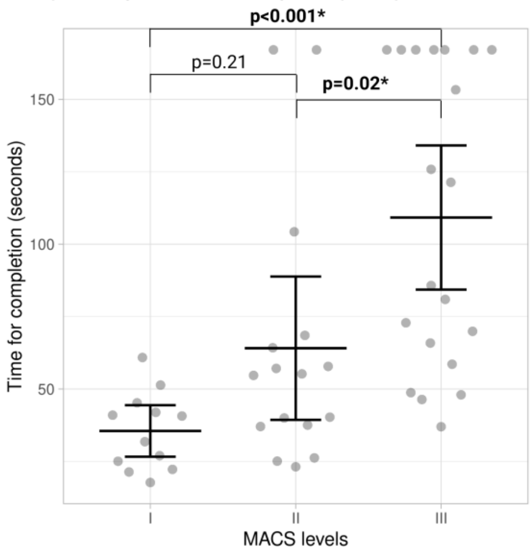


BIMANUAL TASKS

D) More-impaired to less-impaired (BI_{MI-LI})



E) Less-impaired to more-impaired (BI_{LI-MI})



Concurrent validity. High positive correlations were found between all three unimanual tasks and the JTHFT (UNI_{large}, r=0.88; UNI_{medium}, r=0.86; UNI_{small}, r=0.87; p<0.001).

Construct validity. A high negative correlation was found between the AHA and both bimanual tasks (BI_{MI-LI}: r=-0.78, p<0.001; BI_{LI-MI}, r=-0.76, p<0.001, Table 1). The ABILHAND-kids questionnaire correlated moderately with both bimanual tasks (BI_{MI-LI}: r=-0.64, p<0.001; BI_{LI-MI}: r=-0.68, p<0.001, Table 1), while for the CHEQ, mainly low negative correlations were found with both bimanual tasks (r=-0.38 – (-0.52), p<0.01, Table 1).

Table 1: Spearman's rank correlation coefficients (r) between the bimanual TPT tasks and AHA, ABILHAND and CHEQ

TPT	AHA r(p)	ABILHAND r(p)	CHEQ r(p)		
			Grip	Feeling	Timing
BI_{MI-LI}	-0.78 (<0.001*)	-0.64 (<0.001*)	-0.44 (0.002*)	-0.41 (0.005*)	-0.39 (0.007*)
BI_{LI-MI}	-0.76 (<0.001*)	-0.68 (<0.001*)	-0.38 (0.008*)	-0.48 (<0.001*)	-0.52 (<0.001*)

TPT = Tyneside Pegboard Test, BI_{MI-LI} = direction impaired to less-impaired hand, BI_{LI-MI} = direction less-impaired to impaired hand, AHA = Assisting Hand Assessment, CHEQ = Children's Hand-use Experience Questionnaire, r=r-value, p = p-value, * = statistically significant (p<0.05).

INFLUENCE OF SENSORIMOTOR IMPAIRMENTS ON UNIMANUAL AND BIMANUAL DEXTERITY

Based on simple linear regressions, the following variables were selected for the multiple regression analysis: stereognosis (p<0.001, R^2 =0.30-0.46), spasticity (p<0.05, R^2 =0.04-0.15), grip strength (p<0.001, R^2 =0.25-0.40) and MM-intensity in the more-impaired hand (p<0.05, R^2 =0.11-0.30). An overview of the individual relationships, simple linear regression analysis and univariate ANOVA between the TPT tasks and the factors can be found in the Supplementary Materials (SM6 and SM7). For all multiple regression analyses, VIF ranged from 1.08-1.46, indicating low multicollinearity between the independent variables. Correlation coefficients and scatter plots between the retained predictors can be found in the supplementary materials (SM 8).

In the multiple regression, stereognosis was the main factor explaining both unimanual and bimanual dexterity (p<0.001, R^2 =0.37-0.50). Unimanual dexterity was additionally determined by grip strength (UNI_{medium}, p <0.01, R^2 =0.09; UNI_{small}, p=0.03, R^2 = 0.09) and by MM-intensity in the more-impaired hand ($UNI_{large,}$ p=0.02, R^2 =0.08; UNI_{small}, p=0.04, R^2 =0.04). Bimanual dexterity was also determined by MM-intensity in the more-impaired hand (BI_{MI-LI}, p<0.01, R^2 =0.16; BI_{LI-MI}, p<0.01, R^2 =0.10) and by spasticity (BI_{MI-LI}, p=0.04, R^2 =0.05). A more detailed overview of the results is provided in Table 2.

Table 2: Overview of final models of the multiple regression analysis on unimanual and bimanual dexterity.

TPT tasks	R^2 final model	Retained predictors	p-value	Individual R^2	B (95%CI)	β
Unimanual dexterity						
UNI_{large}	0.46	Stereognosis	p<0.001	0.38	-7.19 (-10.43 – (-3.95))	1.60
		MM-intensity in the more-impaired hand	p=0.02	0.08	0.20 (0.03 – 0.67	0.08
UNI_{medium}	0.59	Stereognosis	p<0.001	0.50	-6.52 (-9.37 – (-3.68))	1.41
		Grip strength	p<0.01	0.09	-2.03 (-3.44 – (-0.61))	0.70
UNI_{small}	0.62	Stereognosis	p<0.001	0.49	-12.34 (-17.91 – (-6.77))	2.75
		Grip strength	P=0.03	0.09	-3.24 (-6.15 – (-0.34))	1.44
		MM-intensity in the more-impaired hand	p=0.04	0.04	0.29 (0.01 – 0.57)	0.14
Bimanual dexterity						
BI_{MI-LI}	0.61	Stereognosis	p<0.001	0.40	-3.55 (-5.30 – (-1.81))	-0.45
		MM-intensity in the more-impaired hand	p<0.01	0.16	0.14 (0.04 – 0.23)	0.34
		Spasticity	p=0.04	0.05	1.49 (0.11 – 2.87)	0.25
BI_{LI-MI}	0.47	Stereognosis	p<0.001	0.37	-12.27 (-18.30 – (-6.24))	2.98
		MM-intensity in the more-impaired hand	p<0.01	0.10	0.42 (0.11 – 0.74)	0.16

UNI_{large} = unimanual task with large pegs, UNI_{medium} = unimanual task with medium pegs, UNI_{small} = unimanual task with small pegs, BI_{MI-LI} = bimanual task from the more-impaired to the less-impaired hand, BI_{LI-MI} = bimanual task from the less-impaired to more-impaired hand, B = unstandardized coefficient, CI = confidence interval, β = standardized coefficients, R^2 final model = degree of variance of the TPT task that is explained by the retained predictors, individual R^2 = degree of variance of the TPT tasks that is explained by one specific retained predictor.

DISCUSSION

This study established psychometric properties of the TPT, namely known-group, concurrent and construct validity in children with uCP. Furthermore, stereognosis was found to be the main factor explaining unimanual and bimanual dexterity, followed by grip strength for unimanual dexterity and MM-intensity in the more-impaired hand for bimanual dexterity.

First, we investigated psychometric properties of the TPT. Our results showed that the TPT can discriminate between manual ability levels in children with uCP, establishing known-group validity. The TPT tasks discern MACS III from other levels very well. Between MACS I and MACS II, a significant difference was only found in the unimanual task with small pegs. During the analysis, two participants with MACS level II were identified as outliers. These two participants had more difficulties with performing the tasks, resulting in the implementation of the threshold. When removing these outliers from the analysis, only the results of the UNI_{small} between MACS levels I and II changed to a non-significant result. This is not unexpected, since the two children with the most impaired performance in the MACS II group were left out of the analysis. However, not including these children may potentially bias our results as we would not include the whole spectrum of children with a MACS II classification compromising the generalizability of our results. Subsequently, we opted to report the results of the whole group. Still, an overview of the results without these outliers is provided in the supplementary materials (SM5). Second, we established concurrent validity of the unimanual TPT tasks by showing agreement with the commonly-used JTHFT. Nevertheless, the TPT has the advantage to measure both unimanual and bimanual dexterity electronically and that it is easily adapted using the different peg sizes. Moreover, norm values are available.[5] Finally, we found that children with a lower score on bimanual assessments performed slower on the TPT, establishing construct validity. Slightly higher correlations were found between the TPT and the AHA and ABILHAND-kids compared to the findings of Basu et al. 2018.[5] This might be explained by the fact that in this study also children with a more severely impaired hand function could still be included due to the implementation of the thresholds. Next to this, the TPT correlated higher with the AHA, compared to the CHEQ, indicating that the CHEQ in particular measures a different aspect of bimanual performance. The CHEQ evaluates the perceived abilities of the more-impaired hand during bimanual daily life activities.[27] In contrast, the AHA focusses more on the observed spontaneous use during bimanual play, and also specifically evaluates grasping abilities during task performance (e.g. grasps, grip stability, readjust grasp…)[15], which may explain the higher correlation with the TPT.

Second, results from the univariate linear regression were in line with our hypothesis and showed moderate to high relation between stereognosis and grip strength with all TPT tasks. Multiple regression analysis identified stereognosis as the main predictor of both unimanual and bimanual dexterity. This is in line with literature showing that stereognosis is highly correlated with the unimanual JTHFT.[6] Due to an impaired stereognosis, the ability to make a mental representation of the object during the TPT task might be affected, which has an influence on the anticipatory control to adapt the correct grip force with timed accuracy.[8] Hence, due to an impaired stereognosis, the child may have to rely more on visual feedback, which could slow down the sensorimotor feedback[28], resulting in a slower performance in the TPT tasks.Also grip strength has been shown to be an important predictor of unimanual dexterity.[6] In this study, grip strength was identified as an additional explanatory factor of the more difficult unimanual tasks with medium and small pegs. Also, the univariate correlations between grip strength and unimanual dexterity were high. These results underline the importance of both grip strength and somatosensory function for unimanual dexterity. Stronger MM in the more-impaired hand determined a small part of the variance in the *unimanual tasks* with large and small pegs. This is an unexpected result, as MM are suggested to mostly have an effect on bimanual tasks[9]. A possible explanation is the presence of ipsilateral corticospinal tract projections from the dominant hemisphere to the more-impaired hand in children with MM.[29] It has been shown that children with ipsilateral and bilateral corticospinal tract projections have indeed worse unimanual dexterity, compared to children with a contralateral corticospinal tract.[30] Another explanation could be that children with a weak hand function use the MM in the more-impaired hand, as support to perform the unimanual tasks.[31] Furthermore, stronger MM in the more-impaired hand were found to be the second largest determinant of the *bimanual TPT tasks.* Surprisingly, MM-intensity in the less-impaired hand and MM-similarity in both hands showed no to low correlations with the bimanual TPT tasks. Zielinski et al. (2016) found the same trend for bimanual performance, where higher correlations were found with MM-intensity compared to MM-similarity in the more-impaired hand.[31] Different neuropathological mechanisms of MM could possibly explain the different outcomes between both hands.[32] Both the presence of ipsilateral corticospinal tract projections as well as the lack of interhemispheric inhibition have been put forward to explain the occurrence of MM in children with uCP.[32] Future research is warranted to unravel the complex relationship between MM and its underlying neuropathological mechanisms and how this affects bimanual dexterity in children with uCP. Lastly, spasticity had a minor significant contribution for the bimanual task performed from the more-impaired to the less-impaired hand. Spasticity is most often present in the distal muscle groups of the upper limb such as the wrist flexors and forearm pronators.[1] Hence, increased spasticity in these muscle groups may compromise a good orientation of the peg through the hole, impeding the peg transfer from the more-impaired towards the less-impaired side.

Some limitations of this study also need to be addressed. First, some clinical factors were measured with an ordinal score and qualitative scale, like exteroception and spasticity. Nevertheless, reliability of both measurements has been shown previously.[18] Second, in this study we specifically aimed to investigate the impact of sensorimotor impairments. However, our results show that the variability in manual dexterity cannot be fully explained by these sensorimotor impairments alone. Other factors, such as vision, cognition and motor planning may further influence the performance of these tasks. More research with a larger sample size is needed to elucidate which factors fully explain the variability of manual dexterity. Third, due to missing data in the sensorimotor impairments, not all children could be included in the final multiple regression analysis. Nevertheless, as the pattern of missing data is at random, the analysis remains unbiased.[33] Lastly, we implemented a maximum time in case children with a more severely impaired hand function (MACS III) struggled to complete the most difficult tasks. As no maximum time was determined before the assessment, we decided to set a threshold based on the mean+2SD, resulting in thresholding the data of children who performed the task slower than that cut-off or who were not able to perform that task. As a result, also children with a more severely impaired hand function could still be included, since not being able to perform the tasks also provides information on the manual dexterity level of these children. Hence, as in accordance with the JTHFT, we propose the implementation of a threshold value for future administration with the TPT, limiting frustration in children with a more impaired hand function and prevent empty data. Nevertheless, due to this implementation, 12 of the 20 children with MACS level III received this threshold resulting in a low number of children with unchanged data (8 children) for the task UNI_{small} compared to the other MACS levels (MACS level I: 12 children, MACS level II: 16 children). Hence, current study results need to be validated in a new study sample including the implementation of our proposed threshold. Based on our data and the data of Basu et al. 2018[5], we propose 120 seconds as a threshold for UNI_{large}, UNI_{medium} and BI_{MI-LI} for future testing, and 150 seconds for UNI_{small} and BI_{LI-MI} as these tasks are perceived as more difficult in children with uCP.

Nevertheless, our study suggests that both sensory (i.e. stereognosis) and motor (i.e. grip strength) function are important factors for manual dexterity, corresponding to the suggestion that these are both key ingredients for upper limb intervention in children with uCP. [34] Whilst motor-based training forms the typical approach to improve functionality, somatosensory function should also be taken into account. A recent study in adult stroke survivors already showed beneficial results on unimanual capacity after intensive somatosensory discrimination training during which texture discrimination, proprioception and stereognosis were trained.[35] The effect of an integrated sensorimotor training program on unimanual and bimanual dexterity in children with uCP still warrants further investigation.

CONCLUSION

This study established known-group, construct, and concurrent validity of the TPT assessment in children with uCP. The main determinant of both unimanual and bimanual dexterity was stereognosis. Unimanual dexterity was additionally determined by grip strength and MM-intensity in the more-impaired hand, and bimanual dexterity by MM-intensity in the more-impaired hand and spasticity. For future purposes, we recommend to use a threshold in order to minimize frustration and prevent empty data.

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FIGURES AND TABLES LEGENDS

Figure 1: Overview of unimanual and bimanual TPT tasks.

Figure 2: Single data points of different unimanual and bimanual TPT tasks across the three MACS levels.

Table 1: Spearman's rank correlations (r) between the bimanual TPT tasks and AHA, ABILHAND and CHEQ.

Table 2: Overview of final models of the multiple regression analysis on unimanual and bimanual dexterity.

SUPPORTING INFORMATION

The following supplementary materials may be found online:

SM 1: Distribution of the MM characteristics (MM-similarity and MM-strength) in both hands

SM 2: Missing and descriptive statistics (mean and standard deviation (mean(SD)) of the outcome measures for TPT tasks.

SM 3: Overview of number of children who received the identified threshold for each TPT task as a function of the MACS levels

SM 4: Descriptive statistics (mean and standard deviation (mean (SD)) and the one-way ANCOVA results of the main effect of MACS levels on TPT outcome, with age as a covariate**.**

SM 5: Descriptive statistics (mean and standard deviation (mean (SD)) and the one-way ANCOVA results of the main effect of MACS levels on TPT outcome, with age as a covariate, without outliers (child 1 for all TPT tasks and child 2 for the $BI_{LI\text{-}MI}$).

SM 6: Pearson's correlation coefficients(r) between TPT tasks and nine clinical factors.

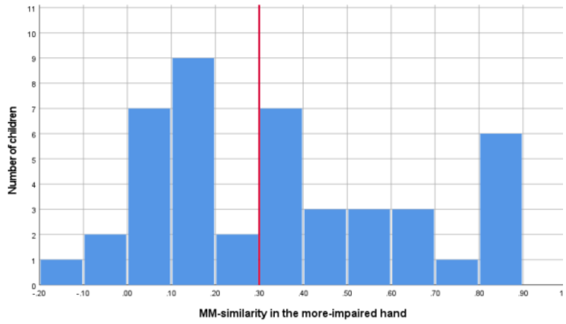
SM 7: Simple linear regression analysis (R^2(p-value)) between TPT tasks and eight clinical factors and univariate ANOVA (R^2(p-value)) between TPT tasks and exteroception.

SM 8: Pearson's correlation coefficients (r) with scatter plots between retained predictors of the multiple regression analysis

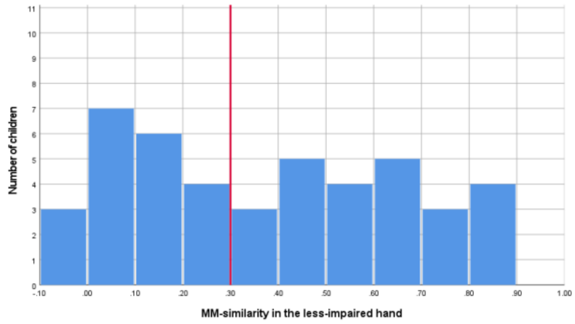
Supplementary materials

SM 1: Distribution of the MM characteristics (MM-similarity and MM-strength) in both hands

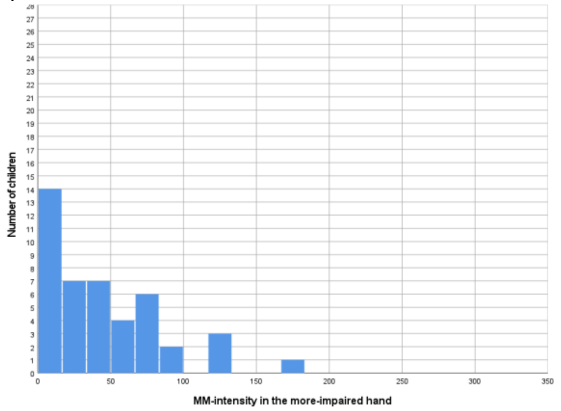
A)



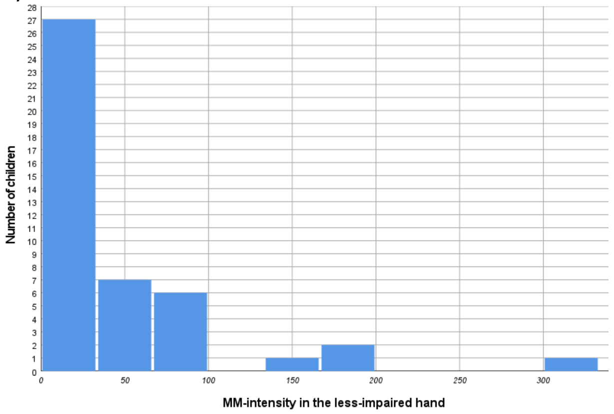
B)



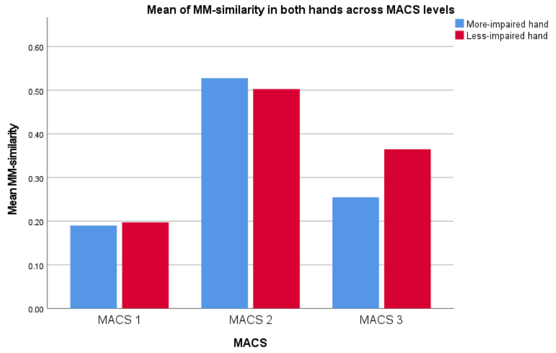
C)



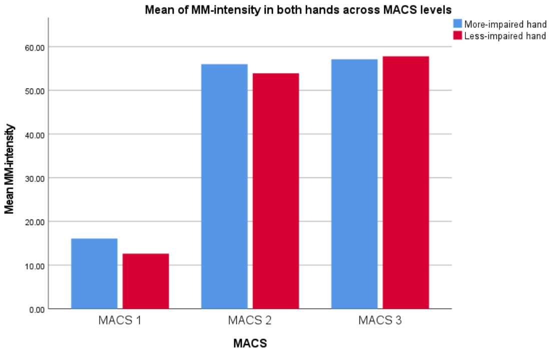
D)



E)



F)



Overview of distribution of MM-similarity (A,B) and MM-intensity (C,D) across whole population and across the different MACS levels (E,F). According to Zielinski et al. (2016), children were classified with MM when showing a MM-similarity ≥ 0.30[30] (red line in A and B). MM = mirror movements, MACS = Manual ability classification system.

SM 2: Missing and descriptive statistics (mean and standard deviation (mean(SD)) of the outcome measures for TPT tasks

Assessment	Number of participants	Missing data	Minimum	Maximum	Mean (SD)	Median (IQR)
TPT tasks						
UNI_{large}	49	0	12.80	116.38*	46.27 (30.19)	35.99 (26.45–57.37)
UNI_{medium}	49	0	13.44	94.51*	45.82 (25.66)	39.51 (25.52–63.37)
UNI_{small}	49	0	15.08	146.63*	75.12 (49.76)	57.10 (32.29–146.63)
$BI_{MI\text{-}LI}$	48	1	16.55	75.48*	40.09 (16.60)	39.34 (26.75–49.82)
$BI_{LI\text{-}MI}$	48	1	17.71	167.10*	75.74 (52.06)	56.16 (38.15–117.09)
Assessments for psychometric properties						
ABILHAND-kids	48	1	-0.50	3.90	1.64 (1.18)	1.57 (0.85–2.63)
AHA	49	0	21	94	58.84 (16.12)	NA
CHEQ – Timing	47	2	2.59	82.62	40.69 (15.84)	NA
CHEQ – Feeling	47	2	10.20	94.79	47.63 (18.16)	46.65 (38.13–55)
CHEQ – Grip	47	2	0.00	76.16	45.50 (15.15)	46.03 (35.23–55.55)
JFHFT	49	0	40.50	720	237.11 (177.60)	171.98 (86.48–347.87)
Clinical assessments						
Exteroception	49	0	0	2	1.86 (0.41)	N (%)
<u>Absent</u>						1 (2)
<u>Impaired</u>						5 (10.2)
<u>Intact</u>						43 (87.8)
Stereognosis	48	1	0	6	3.83 (1.96)	4 (2.25-5.75)
Spasticity	49	0	0	15	5.62 (2.74)	5.5 (4.50-7.25)
Grip Strength	49	0	1	15	5.71 (4.00)	4.33 (2.67-7.25)
MM-intensity						
in the more-impaired hand	44	5	0	168.86	46.45 (38.81)	38.67 (14.48-72.90)

in the less-impaired hand	44	5	0	301.65	45.09 (62.10)	21.49 (6.05-53.97)
MM-similarity						
in the more-impaired hand	44	5	-0.14	0.87	0.34 (0.29)	0.34 (0.11-0.57)
in the less-impaired hand	44	5	-0.01	0.87	0.37 (0.28)	0.35 (0.11-0.61)

TPT = Tyneside pegboard test, UNI_{large} = unimanual task with large pegs, UNI_{medium} = unimanual task with medium pegs, UNI_{small} = unimanual task with small pegs, BI_{MI-LI} = bimanual task from the more-impaired to the less-impaired hand, BI_{LI-MI} = bimanual task from the less-impaired to more-impaired hand, AHA = Assisting Hand Assessment, JTHFT = Jebsen-Taylor hand function test, MM = mirror movements, *= implemented threshold of mean+2SD for tasks of the TPT, IQR = interquartile range, NA = not applicable as these variables were normally distributed.

SM 3: Overview of number of children who received the identified threshold for each TPT task as a function of the MACS levels

TPT tasks	MACS level	Children unable to perform	Children with a slower pace	Total children (percentage of total participants for each task)
Unimanual TPT				
UNI_{large}	MACS 1	0	0	5 (10%)
	MACS 2	1	0	
	MACS 3	1	3	
UNI_{medium}	MACS 1	0	0	7 (15%)
	MACS 2	1	0	
	MACS 3	3	3	
UNI_{small}	MACS 1	0	0	13 (26%)
	MACS 2	1	0	
	MACS 3	11	1	
Bimanual TPT				
BI_{MI-LI}	MACS 1	0	0	4 (8%)
	MACS 2	0	1	
	MACS 3	2	1	
BI_{LI-MI}	MACS 1	0	0	9 (19%)
	MACS 2	1	1	
	MACS 3	6	1	

TPT = Tyneside Pegboard Test, MACS = Manual Ability Classification System, UNI_{large} = unimanual task with large pegs, UNI_{medium} = unimanual task with medium pegs, UNI_{small} = unimanual task with small pegs, BI_{MI-LI} = bimanual task from the more-impaired to the less-impaired hand, BI_{LI-MI} = bimanual task from the less-impaired to more-impaired hand

SM 4: Descriptive statistics (mean and standard deviation (mean (SD)) and the one-way ANCOVA results of the main effect of MACS levels on TPT outcome, with age as a covariate.

	MACS I	*MACS II*	*MACS III*	***p*** **(η_p^2) (MACS)**	***Group differences***		
					I vs II (d)	***I vs III (d)***	***II vs III (d)***
Unimanual TPT tasks							
L	23.18 (7.46)	38.06 (22.81)	67.11 (31.28)	p<0.001* (0.37)	p=0.29 (0.88)	p<0.001* (1.93)	p<0.01* (1.06)
M	24.10 (9.19)	38.98 (19.87)	64.68 (23.84)	p<0.001* (0.43)	p=0.12 (0.96)	p<0.001* (2.25)	p=0.002* (1.17)
S	27.85 (10.76)	58.62 (34.71)	117.51 (40.34)	p<0.001* (0.57)	p=0.04* (1.20)	p<0.001* (3.04)	p<0.001* (1.56)
Bimanual TPT tasks							
MI-LI	27.53 (10.09)	35.32 (14.20)	51.44 (14.46)	p<0.001* (0.37)	p=0.36 (0.65)	p<0.001* (1.93)	p<0.01* (1.12)
LI-MI	35.51 (13.40)	64.08 (44.97)	109.20 (51.84)	p<0.001* (0.35)	p=0.21 (0.86)	p<0.001* (1.95)	p=0.02* (0.93)

TPT = Tyneside Pegboard Test, L = large pegs, M = medium pegs, S = small pegs, MI-LI = direction more-impaired to less-impaired hand, LI-MI = direction less-impaired to more-impaired hand, MACS = Manual Ability Classification System, SD = standard deviation, p = p-value, η_p^2 = partial eta squared, d = effect size. *comparison is significant at 0.05 level (2-tailed).

SM 5: Descriptive statistics (mean and standard deviation (mean (SD)) and the one-way ANCOVA results of the main effect of MACS levels on TPT outcome, with age as a covariate, without outliers (child 1 for all TPT tasks and child 2 for the $BI_{LI\text{-}MI}$).

	MACS I	*MACS II*	*MACS III*	***p*** **(η_p^2) (MACS)**	***Group differences***		
					I vs II (d)	***I vs III (d)***	***II vs III (d)***
Unimanual TPT tasks							
L	23.18 (7.46)	33.17 (10.97)	67.11 (31.28)	p<0.001* (0.46)	p=0.46 (1.06)	p<0.001* (1.93)	p<0.001* (1.45)
M	24.10 (9.19)	35.50 (14.23)	64.68 (23.84)	p<0.001* (0.50)	p=0.16 (0.95)	p<0.001* (2.25)	p=0.001* (1.49)
S	27.85 (10.76)	53.12 (27.14)	117.51 (40.34)	p<0.001* (0.62)	p=0.08 (1.22)	p<0.001* (3.04)	p<0.001* (1.87)
Bimanual TPT tasks							
MI-LI	27.53 (10.09)	32.64 (9.66)	51.44 (14.46)	p<0.001* (0.45)	p=0.36 (0.52)	p<0.001* (1.93)	p<0.001* (1.53)
LI-MI	35.51 (13.40)	49.36 (21.61)	109.20 (51.84)	p<0.001* (0.47)	p=0.21 (0.77)	p<0.001* (1.95)	p=0.001* (1.51)

TPT = Tyneside Pegboard Test, L = large pegs, M = medium pegs, S = small pegs, MI-LI = direction more-impaired to less-impaired hand, LI-MI = direction less-impaired to more-impaired hand, MACS = Manual Ability Classification System, SD = standard deviation, p = p-value, η_p^2 = partial eta squared, d = effect size. *comparison is significant at 0.05 level (2-tailed).

SM 6: Pearson's correlation coefficients (r) between TPT tasks and nine clinical factors.

TPT tasks	Age	Stereo-gnosis	Extero-ception	Spasticity	Grip Strength	MM-similarity		MM-intensity	
						MI hand	LI hand	MI hand	LI hand
Unimanual TPT									
UNI_{large}	-0.21	-.55	-0.16	.22	-.50	0.20	0.13	0.35	0.11
UNI_{medium}	-0.24	-.66	-0.25	.32	-.61	0.13	0.13	0.33	0.24
UNI_{small}	-0.15	-.67	-.19	.36	-.63	0.08	0.09	0.43	0.22
Bimanual TPT									
BI_{MI-LI}	-0.06	-.58	-0.11	.39	-.57	0.10	0.18	0.55	0.27
BI_{LI-MI}	-0.23	-.55	-0.09	0.20	-.56	0.23	0.22	0.43	0.21

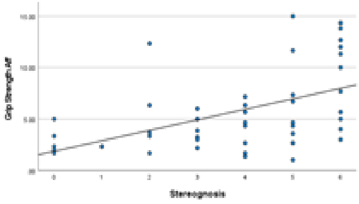
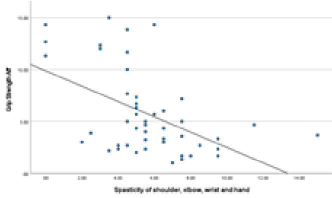
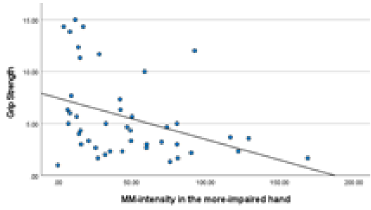
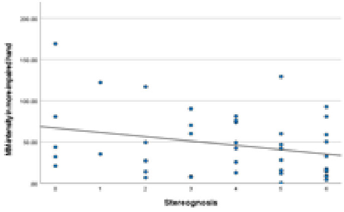
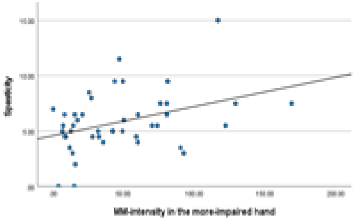
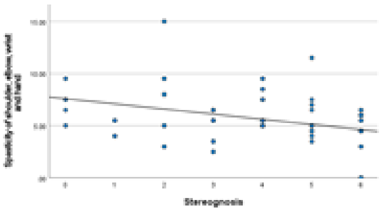
TPT = Tyneside Pegboard Test, MI = more-impaired hand, LI = less-impaired hand, UNI_{large} = unimanual task with large pegs, UNI_{medium} = unimanual task with medium pegs, UNI_{small} = unimanual task with small pegs, BI_{MI-LI} = bimanual task from the more-impaired to the less-impaired hand, BI_{LI-MI} = bimanual task from the less-impaired to more-impaired hand, MM = mirror movements.

SM 7: Simple linear regression analysis (R^2(p-value)) between TPT tasks and eight clinical factors and univariate ANOVA (R^2(p-value)) between TPT tasks and exteroception

TPT tasks	Age	Stereo-gnosis¥	Spasticity¥	Grip Strength¥	MM-similarity		MM-intensity¥		Extero-ception¤
					MI hand	LI hand	MI hand¥	LI hand	
Unimanual TPT									
Large	0.04 (0.15)	0.30 (<0.001*)	0.05 (0.13)	0.25 (<0.001*)	0.04 (0.19)	0.02 (0.40)	0.13 (0.02*)	0.01 (0.50)	0.06 (0.25)
Medium	0.06 (0.10)	0.44 (<0.001*)	0.10 (0.03*)	0.37 (<0.001*)	0.02 (0.39)	0.02 (0.4)	0.11 (0.03*)	0.06 (0.12)	0.07 (0.21)
Small	0.02 (0.30)	0.46 (<0.001*)	0.13 (0.01*)	0.4 (<0.001*)	0.01 (0.60)	0.01 (0.55)	0.19 (<0.01*)	0.05 (0.15)	0.09 (0.11)
Bimanual TPT									
MI-LI	0.003 (0.71)	0.34 (<0.001*)	0.15 (<0.01*)	0.32 (<0.001*)	0.01 (0.54)	0.03 (0.24)	0.3 (<0.001*)	0.07 (0.08)	0.03 (0.50)
LI-MI	0.05 (0.12)	0.31 (<0.001*)	0.04 (0.17)	0.31 (<0.001*)	0.05 (0.14)	0.05 (0.16)	0.19 (<0.01*)	0.05 (0.17)	0.03 (0.50)

TPT = Tyneside Pegboard Test, MI = more-impaired hand, LI = less-impaired hand , MI-LI = bimanual task from the more-impaired to the less-impaired hand, LI-MI = bimanual task from the less-impaired to more-impaired hand, MM = mirror movements, ¥ = included clinical factors, ¤ = univariate ANOVA, *comparison is significant at 0.05 level (2-tailed).

SM 8: Pearson's correlation coefficients (r) with scatter plots between retained predictors of the multiple regression analysis

	Stereognosis	Spasticity	MM-intensity in the MI hand
Grip strength	 r = 0.50	 r = -0.51	 r = -0.38
MM-intensity in the MI hand	 r = -0.27	 r = 0.38	
Spasticity	 r = -0.36		

r = correlation coefficient, MM = mirror movements, MI = more-impaired.