External validity of the Both Hands Assessment for evaluating bimanual performance in children with bilateral cerebral palsy

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AIM To investigate the external validity of the Both Hands Assessment (BoHA), a new test evaluating bimanual performance in children with bilateral cerebral palsy (CP), by analysing its relationship to established measurements of hand function and self-care skills.

METHOD In this cross-sectional study, we recruited children with bilateral CP and manual ability corresponding to Manual Ability Classification System (MACS) levels I to III attending three habilitation units in Norway. All participants completed the BoHA. Unimanual capacity was assessed using the Bimanual Fine Motor Function (BFMF) classification, the Box and Block Test (BBT), and the Melbourne Assessment 2 (MA2). Self-care skills were assessed with the Pediatric Evaluation of Disability Inventory (PEDI).

RESULTS Thirty-nine children (19 males, 20 females; mean age 8y 2mo, SD 2y 8mo; age range 2y 8mo–12y 6mo) were included. Spearman's correlation coefficient (ρ) suggested high correlation between the BoHA and MACS ($p=0.89$; 95% confidence interval [CI] 0.79–0.94), BFMF classification ($p=0.84$; 95% CI 0.71–0.91), and BBT for the non-dominant ($p=0.85$; 95% CI 0.68–0.95) and dominant hand ($p=0.72$; 95% CI 0.53–0.85). The Spearman's ρ between the BoHA and the MA2 subscales varied between 0.48 and 0.83, while the PEDI's ρ was 0.51 (95% CI 0.33–0.67).

INTERPRETATION The BoHA provides valid measures of hand use as suggested by its high correlation with other activity-based measures of hand function.

Children with bilateral cerebral palsy (CP) have varying degrees of manual impairments that may limit their ability to perform everyday activities successfully.[1,2] Extensive time and resources are used to improve hand function and independence in everyday life.[2] So far, evidence is scarce for interventions targeting hand function in children with bilateral CP; the use of appropriate outcome measures with sound measurement properties is needed for future research.[3]

One of the recommended outcome measures for evaluating hand function in children with bilateral CP is the newly developed Both Hands Assessment (BoHA).[4] The BoHA scale has been validated through Rasch analyses for children with bilateral CP classified in Manual Ability Classification System (MACS) levels I to III, aged between 18 months and 12 years.[5] The purpose of the BoHA is to measure and describe how effectively both hands are used in playful bimanual activities, as well as describing the degree of side difference between hands. The intent is to reflect the child's common and spontaneous hand use (performance), not the highest level of functioning (capacity),

as outlined in the conceptual framework of the International Classification of Functioning, Disability and Health (ICF).[6] A recent systematic review identified the BoHA as the only available observation-based instrument assessing bimanual performance in children with bilateral CP.[4] Very high interrater reliability has been reported for the BoHA, while test–retest reliability and responsiveness to change have not yet been established.[7]

In addition to scale measurement properties and reliability indices, it is important to investigate the external validity of the BoHA. External validity is an aspect of construct validity that describes how the BoHA results relate to results produced by instruments presumed to measure a similar construct (convergent validity) and those presumed to measure different constructs (divergent validity).[8]

The MACS and the Bimanual Fine Motor Function (BFMF) are used to describe hand function in children with CP in five different severity levels, with a lower score indicating higher ability.[9–11] Both classifications are related to the activity component of the ICF, providing

complementary information regarding manual performance (MACS) and fine motor capacity (BFMF).[11] Classifications describe hand function according to common characteristics but cannot be used to evaluate change. Thus, standardized tests providing more detailed information are required. Two frequently used tests are the Box and Block Test (BBT)[12] and the Melbourne Assessment 2 (MA2),[13] which measure unimanual motor abilities.[14,15] The BBT measures the speed related to fine hand use in the activity domain of the ICF, while the MA2 measures the qualitative aspects of hand function within the domains of body function and activity.[16,17]

In contrast to instruments measuring hand function, the Pediatric Evaluation of Disability Inventory (PEDI) is commonly used to measure children's capabilities and independence in self-care activities.[18] Although self-care skills are related to manual abilities, they are also dependent on other constructs, such as gross motor function and cognition.[1,19] Thus, self-care skills represent a broad construct, conceptualized in the ICF activities and participation domain.[18] The PEDI is recommended to assess self-care skills in children of elementary school age or younger and may therefore be used to explore the divergent validity of the BoHA.[18,20]

The aim of this study was to investigate the external validity of the BoHA by exploring its relationship to instruments assessing hand function (convergent validity) or self-care skills (divergent validity). We hypothesized that bimanual performance assessed with the BoHA would be highly correlated with instruments assessing various aspects of hand function but only moderately correlated with self-care skills.

METHOD
In this cross-sectional study using convenience sampling, we recruited children attending three paediatric habilitation units in Norway between 1st August 2012 and 28th February 2013. Children were eligible for participation if they (1) were diagnosed with bilateral CP of spastic, dyskinetic, or ataxic type; (2) had manual ability corresponding to MACS levels I to III; and (3) were aged between 30 months and 12 years to allow the use of additional test (s) of hand function besides the BoHA.[12,13] Parents provided written informed consent and the study was approved by the Regional Committee for Medical and Health Research Ethics in Mid-Norway (registration no. 2012/152). Children included in the current study were also included in the BoHA validation study.[5]

Assessments
Occupational therapists at three paediatric habilitation units in Norway (Trondheim, Oslo, and Tønsberg) administered the BoHA and the comparison instruments. In Trondheim, this was done by the first author (AKGE). Instruments were administered on the same day and in the same order for each participant. Breaks were provided between assessments when needed. Scoring of the MACS, BBT, and PEDI was completed by the occupational

What this paper adds
- The Both Hands Assessment (BoHA) is a valid test of hand function in children with bilateral cerebral palsy.
- The BoHA was highly correlated to activity-based measurements of hand function.
- Correlation between the BoHA and self-care skills measured with the Pediatric Evaluation of Disability Inventory was moderate.

therapists at each unit, while the BoHA, BFMF, and MA2 were scored by the first author from video recordings more than 14 days after the comparison tests had been scored. The MACS and BFMF were used to classify manual ability and fine motor function.[9,11] Furthermore, unimanual capacity was assessed with the BBT, which measures gross manual dexterity, and the MA2, which measures movement range, dexterity, accuracy, and fluency.[12,13,21] Self-care skills were assessed using the Norwegian version of the PEDI.[22] In addition, children's gross motor function was classified using the Gross Motor Function Classification System (GMFCS).

The BoHA is an observation-based assessment of bimanual performance that uses standardized, age-appropriate play activities. Spontaneous hand use is scored according to 11 unimanual and five bimanual items on a 4-point rating scale.[5] The unimanual items are scored for each hand separately and the sum scores are used to determine asymmetric or symmetric hand use (i.e. a difference of >20% or ≤20% between hands respectively). For the bimanual items, one common score is given for both hands and added to the unimanual sum scores, resulting in a possible raw score ranging from 27 to 108 points. The corresponding interval-level logit-based BoHA unit is reported on a 0 to 100 scale and was used in the statistical analyses.[5]

The MACS and Mini-MACS classify the ability to handle objects in relevant and age-appropriate daily activities in children aged 4 years or older or younger than 4 years respectively.[9,23] Both versions of the classification are valid and reliable; the classification describes five levels of manual performance, with lower levels indicating higher ability.[9,23] Children's MACS or Mini-MACS classification levels were determined by parent report.

The BFMF version 2 is a valid and reliable classification of fine motor capacity in children with CP aged 3 to 18 years and describes five levels, with lower levels indicating higher ability.[10,11] The classification level was determined by assessing the child's capacity to grasp, manipulate, and hold objects with each hand separately.

The BBT assesses unilateral manual speed and dexterity and can be used from 3 years of age.[12,21] Studies have demonstrated evidence of test–retest reliability in typically developing children and children with CP, as well as responsiveness in children with CP.[12,24] The child is asked to move as many individual 2.5cm cubes as possible from one compartment to another within 60 seconds. After a 15-second trial period, the child first uses the dominant hand and then the non-dominant hand to move the cubes. The total number of cubes successfully moved with each hand is registered and was used in the analyses.

The MA2 measures the quality of unilateral upper-limb movements in children with CP between the ages of 30 months and 15 years.[13] The instrument consists of 14 functional tasks (e.g. pointing, grasping, releasing, manipulating) generating 30 item scores that are organized into four discrete subscales: dexterity; range of motion; accuracy; and fluency. The MA2 subscales are valid, reliable, and responsive in children with CP.[25,26] The total scores for each subscale are converted to percentage scores that were used in the analyses for the dominant and non-dominant hands.

The Norwegian version of the PEDI was used to assess self-care abilities through standardized interviews with the parents.[20,22] The PEDI is standardized for children aged from 6 months to 7 years 6 months; both norm- and criterion-referenced scores are provided. The criterion-referenced scaled scores give an interval measure between 0 and 100 that reflects functional ability regardless of age.[20] The PEDI has strong psychometric properties.[18] The criterion-referenced scaled scores for the self-care subscale were used in the analyses. Only data from children within the specified age range or with decreased self-care abilities (scaled scores below 100) were included.

Statistical analyses

All statistical analyses were performed using Stata v16.1 (StataCorp, College Station, TX, USA). Visual inspection of Q–Q plots and normality tests revealed that the variables for the PEDI self-care subscale and most of the MA2 subscales were skewed. Furthermore, the associations between the BoHA and MA2 subscales were curvilinear. For consistency, we chose to report the Spearman's rank correlation coefficient (ρ) for all comparisons. To test the equality of the two correlation coefficients between the BoHA and BBT for the dominant and non-dominant hands, we applied the Stata module cortesti. Confidence intervals (CIs) for the correlation coefficients were based on Fisher's transformation (the correlation values were interpreted as follows: very high, $p=0.9–1.0$; high, $p=0.7–0.89$; moderate, $p=0.5–0.69$; low, $p=0.26–0.49$; and very low, $p=0–0.25$).[27] A two-sided $p<0.01$ was deemed to indicate statistical significance.

Since the first author was not blinded to the test results for the children recruited in Trondheim, we performed sensitivity analyses to explore if the results persisted when the analyses were restricted to children recruited in Oslo and Tønsberg. In these analyses, the first author, who scored the BoHA, was blinded to the results of the MACS, BBT, and PEDI, while the local occupational therapists were unaware of the BoHA scores.

RESULTS

Thirty-nine children (Trondheim, $n=17$, Oslo, $n=14$, Tønsberg, $n=8$) were included in the study. The children (19 males, 20 females) had a mean age of 8 years 2 months, SD 2 years 8 months (age range 2y 8mo–12y 6mo). Thirty-four children were diagnosed with spastic bilateral CP, while four were diagnosed with the dyskinetic subtype and one with the ataxic subtype. Thirty-one children had symmetric hand use (Table 1). Twenty-eight children were classified in MACS levels I and II and 11 were classified in MACS level III. Fine motor (BMFM) and gross motor function (GMFCS) varied between levels I and IV with most children classified in levels I and II ($n=17$ and $n=28$ respectively) (Table 1). The MACS and BFMF levels were identical in 24 children.

The Spearman's ρ between the BoHA and classifications of hand function was 0.89 (95% CI 0.79–0.94, $p<0.001$) for the MACS and 0.84 (95% CI 0.71–0.91, $p<0.001$) for the BFMF (Table 2; Fig. S1, online supporting information).

Table 1: Demographic characteristics of the study participants

Characteristics	Participants n (%)	BoHA units Mean (SD)	Age (y:mo) Mean (range)
Sex			
Male	19 (48.7)	66.6 (14.9)	8:3 (3:3–12:4)
Female	20 (51.3)	67.7 (16.5)	8:2 (2:8–12:6)
CP subtype			
Spastic bilateral	34 (87.2)	69.2 (15.2)	8:1 (2:8–12:6)
Dyskinetic	4 (10.3)	50.3 (10.0)	8:9 (6:6–12:4)
Ataxic	1 (2.5)	67.0	8:9
MACS/MINI-MACS level			
I	16 (41.0)	81.6 (10.7)	8:4 (2:8–12:6)
II	12 (30.8)	63.2 (7.7)	8:1 (5:6–11:2)
III	11 (28.2)	50.6 (5.6)	8:0 (3:9–12:4)
BFMF level			
I	14 (35.9)	81.7 (12.0)	9:0 (3:3–12:6)
II	15 (38.4)	64.7 (9.2)	8:2 (2:8–12:4)
III	6 (15.4)	53.8 (3.0)	7:9 (4:3–11:2)
IV	4 (10.3)	45.5 (5.7)	6:3 (3:9–8:10)
GMFCS level			
I	11 (28.2)	79.6 (14.0)	8:6 (2:8–12:6)
II	17 (43.6)	67.8 (13.5)	8:1 (3:3–11:9)
III	7 (17.9)	55.6 (6.4)	8:0 (3:9–12:0)
IV	4 (10.3)	51.0 (11.0)	8:0 (5:1–12:4)
Hand dominance			
Right	31 (79.5)	66.7 (15.4)	8:2 (2:8–12:4)
Left	5 (12.8)	65.0 (20.7)	10:1 (8:10–12:6)
Mixed	3 (7.7)	75.3 (6.8)	5:6 (4:1–7:2)
BoHA			
Total sample	39 (100)	67.2 (15.5)	8:3 (2:8–12:6)
Symmetric hand use	31 (79.5)	71.5 (14.3)	8:3 (2:8–12:6)
Asymmetric hand use	8 (20.5)	50.5 (5.9)	8:0 (3:9–12:0)
BBT			
Dominant hand	34 (89.7)	68.4 (15.6)	8:9 (4:1–12:6)
Non-dominant hand	33 (87.2)	68.2 (15.8)	9:0 (2:5–12:6)
MA2			
Dominant hand	37 (94.9)	67.2 (15.9)	8:4 (3:3–12:6)
Non-dominant hand	36 (92.3)	67.0 (16.1)	8:6 (3:3–12:6)
PEDI self-care			
Total sample[a]	36 (92.3)	65.4 (14.9)	7:11 (2:8–12:4)
Younger than 7y 6mo	18 (46.2)	65.9 (14.0)	5:8 (2:8–7:4)
Older than 7y 6mo[a]	18 (46.2)	68.2 (17.0)	10:1 (8:8–12:4)

[a]Within the standardized age range. BoHA, Both Hands Assessment; CP, cerebral palsy; MACS, Manual Ability Classification System; BFMF, Bimanual Fine Motor Function; GMFCS, Gross Motor Function Classification System; BBT, Box and Block Test; MA2, Melbourne Assessment 2; PEDI, Pediatric Evaluation of Disability Inventory.

Some children did not complete the BBT (dominant hand, $n=5$; non-dominant hand, $n=6$) and MA2 (dominant hand, $n=2$; non-dominant hand, $n=3$). These children dropped out because they had difficulties complying with the task instructions and one child was below 3 years of age and thus too young for the BBT (Table 2). The results indicate somewhat higher correlations between the BoHA and speed and dexterity (BBT) for the non-dominant ($p=0.85$; 95% CI 0.68–0.95, $p<0.001$) compared with the dominant hand ($p=0.72$; 95% CI 0.53–0.85, $p<0.001$) but the correlations did not differ significantly ($p=0.33$) (Table 2; Fig. S1). The correlation coefficients between the BoHA and MA2 subscales varied from $p=0.48$ to 0.58 ($p<0.003$) for the dominant hand and $p=0.76$ to 0.83 ($p<0.001$) for the non-dominant hand (Table 2). Less variability was evident for the dominant hand with several children reaching the ceiling for the MA2 subscales of range of motion ($n=13$), fluency ($n=13$), accuracy ($n=11$), and dexterity ($n=3$) (Fig. 1). Fluency of the non-dominant hand was the MA2 subscale with the highest correlation with the BoHA ($p=0.83$, 95% CI 0.69–0.91, $p<0.001$).

Three children older than 7 years 6 months reached the ceiling for the PEDI self-care skills and were therefore excluded from the analyses. The correlation between the BoHA and PEDI self-care skills was $p=0.51$ (95% CI 0.33–0.67, $p=0.002$) for the remaining 36 children (Table 2; Fig. 2). For children younger than 7 years 6 months ($n=18$), the correlation was $p=0.34$ (95% CI 0.13–0.59, $p=0.168$) and not statistically significant. In the older age group ($n=18$), the correlation was $p=0.64$ (95% CI 0.41–0.87, $p=0.004$).

In the sensitivity analyses including only children recruited in Oslo and Tønsberg ($n=22$), the main results were essentially unchanged (Appendix S1, online supporting information).

DISCUSSION

The results of this study correspond well with our initial hypotheses, suggesting high external validity of the BoHA for assessing hand function in children with bilateral CP and classified in MACS levels I to III. Overall, correlations were higher between the BoHA and activity-based instruments assessing hand function (convergent validity), compared with the correlation between the BoHA and self-care skills (divergent validity). This corroborates that the BoHA measures important aspects of hand function.

High correlations between the BoHA and classifications of hand function imply that the BoHA can distinguish between different severity levels of hand function and that children in lower classification levels of the MACS and BFMF have more efficient bimanual performance. In previously published results from our research group, Rasch analyses indicated that the BoHA can distinguish between six and seven different ability levels.[5] However, the responsiveness of the BoHA must be investigated to determine the evaluative validity of the test.

Our results also showed high correlations between the BoHA and BBT, supporting the relevance of considering speed and dexterity when evaluating bimanual performance in children with bilateral CP. During the development of the BoHA, slowness of movement during bimanual play activities was observed in several children with bilateral CP.[5] Therefore, the item 'speed of movement' was developed and slowness was added as an indicator of ineffective hand use for several BoHA items (i.e. grasps, releases, moves fingers, manipulates).[5] Furthermore, high variability in BBT scores was found for both the non-dominant and dominant hands (range 1–62 and 14–64 respectively; Fig. S2, online supporting information). This shows that several children with bilateral CP may also have severe functional limitations in the dominant hand and underscores the importance of assessing both hands in bimanual tasks for these children.

The BoHA items measure hand function mainly within the ICF activity domain but they also measure concepts in the body function domain, such as arm control and coordination and finger movements.[4,5] Thus, we expected a high correlation between BoHA and MA2 subscales. This was confirmed for the non-dominant hand, while correlation was only low to moderate for the dominant hand. The lower correlation for the dominant hand is probably due to less variability in MA2 scores, with several children reaching the ceiling level. This suggests that having adequate movement quality of the dominant hand is not sufficient for effective bimanual performance. Instead, bimanual performance may depend more on the degree of limitation in movement quality of the non-dominant hand.

Table 2: Spearman's ρ with 95% CI for correlations between the BoHA and comparison instruments

Comparison measure	Participants (n)	BoHA units	95% CI
Convergent validity			
MACS	39	0.89[a]	0.79–0.94
BFMF	39	0.84[a]	0.71–0.91
BBT			
Dominant hand	34	0.72[a]	0.53–0.85
Non-dominant hand	33	0.85[a]	0.68–0.95
MA2 dominant hand			
Range of motion	37	0.56[a]	0.29–0.75
Accuracy	37	0.48[b]	0.18–0.70
Dexterity	37	0.58[a]	0.32–0.76
Fluency	37	0.49[b]	0.20–0.70
MA2 non-dominant hand			
Range of motion	36	0.77[a]	0.59–0.88
Accuracy	36	0.79[a]	0.62–0.89
Dexterity	36	0.76[a]	0.58–0.87
Fluency	36	0.83[a]	0.69–0.91
Divergent validity			
PEDI self-care			
Total sample[c]	36	0.51[a]	0.33–0.67
Younger than 7y 6mo[c]	18	0.34	0.13–0.59
Older than 7y 6mo	18	0.64[a]	0.36–0.83

[a]$p<0.001$. [b]$p<0.005$ [c]Excluding children who reached the ceiling for PEDI. CI, confidence interval; BoHA, Both Hands Assessment; MACS, Manual Ability Classification System; BFMF, Bimanual Fine Motor Function; BBT, Box and Blocks Test; MA2, Melbourne Assessment 2; PEDI, Pediatric Evaluation of Disability Inventory.
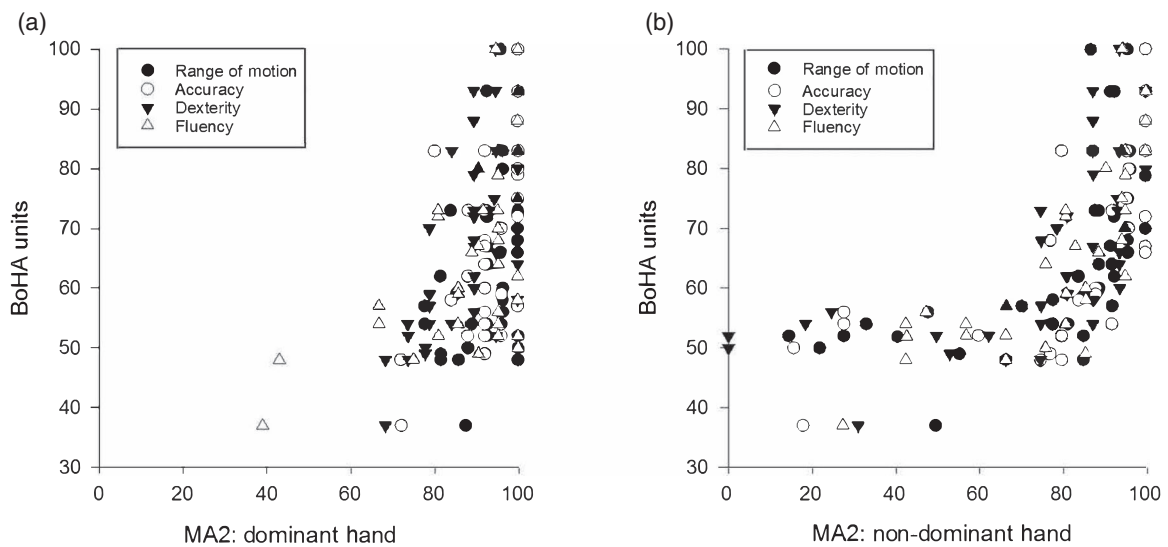
Figure 1: Scatter plot showing the correlation between the Both Hands Assessment (BoHA) and Melbourne Assessment 2 (MA2) for the dominant (a) and the non-dominant hands (b) for the subscales range of motion, accuracy, dexterity, and fluency.
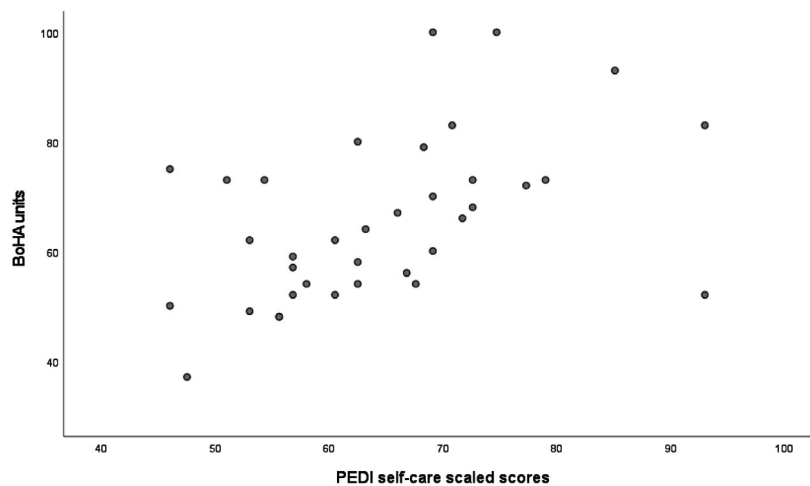
Figure 2: Scatter plot showing the correlation between the Both Hands Assessment (BoHA) units and Pediatric Evaluation of Disability Inventory (PEDI) self-care scaled scores for the 36 children included in the analysis.

The moderate correlation between the BoHA and PEDI is reasonable since these two instruments aim to assess different constructs, namely hand function and self-care skills. Still, our results suggest that effective bimanual performance may affect the ability to achieve independence in self-care. This is in line with a recent study showing that bimanual performance and cognitive abilities were the strongest determinants of self-care skills among children with bilateral CP aged between 8 years and 12 years.[19] No similar results exist for younger children. In the former study, the newly developed computer adaptive test version of the PEDI, validated from birth up to 20 years, was used.[19] A Norwegian version of the computer adaptive test version of the PEDI was not available when the current study was performed. Thus, to confirm our results, future studies should investigate the correlation between the BoHA and self-care skills using the computer adaptive test version of the PEDI and include children from 18 months to 12 years of age.

Study limitations
This study involved a convenience sample of relatively few children with bilateral CP, which could lead to selection bias or chance findings. The latter is unlikely as indicated by the very low *p*-values and CIs. However, some selection bias is possible, as suggested by a lower proportion of

children with asymmetric hand function (21%) in the current study compared with a population-based study from Norway (36%).[2] Thus, including a larger proportion of children with asymmetric hand use may have given other results and requires further study. Furthermore, the very small number of children with the dyskinetic and ataxic CP subtypes precludes reasonable subgroup analyses.

Compared with unpublished population-based data from the Cerebral Palsy Registry of Norway, a lower proportion of children in our study were classified in MACS level I (41%) and a higher proportion were classified in MACS level III (28%) than children registered in the Cerebral Palsy Registry of Norway (48% and 21% respectively). This may also indicate some selection bias. However, for the purpose of the present study, that is, investigating the external validity of the BoHA, the more even distribution of MACS levels may be an advantage. Furthermore, the distribution of MACS levels in our study may be more representative for a clinical sample since children classified in MACS level III usually have more follow-up from health care services than children classified in MACS level I.

Lack of complete blinding of the first author is a potential limitation. However, the BoHA was scored from video recordings more than 2 weeks after the other test scores were completed; for the 22 children where the first author was blinded, the results were essentially unchanged. Therefore, it is unlikely that lack of complete blinding explains the main results.

CONCLUSION

Our results suggest good external validity of the BoHA for assessing hand function in children with bilateral CP and classified in MACS levels I to III. As hypothesized, a high correlation was found between the BoHA and activity-based instruments assessing hand function, while a more moderate correlation was evident between the BoHA and self-care skills. Thus, our results provide further evidence to recommend using the BoHA as a measure of bimanual performance in children with bilateral CP. However, additional research is needed to assess the responsiveness of the BoHA.

DATA AVAILABILITY STATEMENT
Data available on request from the authors.

SUPPORTING INFORMATION
The following additional material may be found online:

Figure S1 Box plots showing the medians of the Both Hands Assessment units in each Manual Ability Classification System and Bimanual Fine Motor Function level.

Figure S2 Scatter plot showing the correlation between the Both Hands Assessment and Box and Block Test for the dominant and non-dominant hands.

Appendix S1: Sensitivity analyses for the 22 children recruited in Tønsberg and Oslo.

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