Hand use development in children with unilateral cerebral palsy

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ABBREVIATIONS

AHA	Assisting Hand Assessment					
СРОР	Cerebral Palsy Follow-up					
	Program					
MACS	Manual Ability Classification					
	System					
PDM	Peak and decline model					
SLM	Stable limit model					

AIM To describe the development of hand use during bimanual activities among children with unilateral cerebral palsy (CP).

METHOD A cohort of 166 children (79 females, 87 males; age range 18mo–13y, mean [SD] age at first assessment 37.6mo [20.5mo]) with unilateral CP, registered in the Norwegian CP Follow-up Program with two or more Assisting Hand Assessments (AHAs), were included in this longitudinal study comprising 524 AHAs. Developmental limits and rates were estimated by non-linear mixed effects models and compared between a stable limit model (SLM) and a peak and decline model. Development was described according to Manual Ability Classification System (MACS) levels and AHA performance at 18 months of age (AHA-18). **RESULTS** Children in MACS level I, or in the high AHA-18 group, reached highest limits and had the most rapid development (p<0.001). The developmental trajectories were different between MACS levels I, II, and III and between the high, moderate, and low AHA-18 groups. Seventy-five per cent of the children reached 90% of their estimated limit at 5 years 10 months or earlier. The SLM showed the best model fit (Akaike information criterion: 4008.99).

INTERPRETATION Most children approached a steady performance limit before 6 years of age. Although children in MACS levels I and II reached 90% of the expected limit at 3 and 4 years respectively, the corresponding age was 8 years for children in MACS level III. The better model fit for the SLM indicates that children with unilateral CP maintain their attained limit of hand use to at least the age of 13 years.

Independent performance of everyday activities is strongly related to hand function and manual ability.¹ The onset of a lesion to the immature brain may lead to the life-long condition of cerebral palsy (CP), and a common characteristic in children with CP is a limited ability to handle objects bimanually.²

Studies that describe the development of hand function among children with CP are limited, despite the desire for such information as expressed by parents who want to know the prospects for their child's development.³ For therapists, knowledge of factors that may influence the development of hand function is important for their communication with parents and for planning individualized and goal-directed rehabilitation services.⁴

For children classified with spastic unilateral CP the challenges of hand function relate specifically to using the most affected hand together with the dominant hand during bimanual activities.⁵ In a previous study, we were able to show improvements in the spontaneous use of the assisting hand over time, as measured with the Assisting Hand Assessment (AHA), among a cohort of children with unilateral CP aged 18 months to 6 years.⁶ Our results

supported the main findings from two earlier Swedish studies that spontaneous hand use improved between 18 months and 13 years of age, with the most rapid development occurring during the first 3 years of life.^{7,8} The results indicated that a plateau of performance was reached by 8 years of age, depending on the child's functional level and early performance on the AHA.⁶⁻⁸ Despite the young age range and the limited number of children classified in Manual Ability Classification System (MACS) level III, our population-based study provided more evidence to support the convenience-based Swedish studies.⁶⁻⁸ Nevertheless. there is still limited knowledge of hand use development in children with unilateral CP throughout the childhood years. Moreover, a Belgian 5-year follow-up study that included a convenience sample of children and adolescents with unilateral CP aged from 5 to 20 years, reported a general decrease in AHA scores after 9 years of age.⁹ Knowledge of the period of most rapid development, developmental trajectories over time, and potential developmental decline, has important clinical implications and needs to be further investigated. To produce generalizable information, efforts should be made to include population-

1462 DOI: 10.1111/dmcn.14957 © 2021 The Authors. Developmental Medicine & Child Neurology published by John Wiley & Sons Ltd on behalf of Mac Keith Press This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made. based cohorts of children within a wide age range and with representative distributions of functional levels.

Norway is a welfare state, where all citizens have access to the same healthcare services at low cost.¹⁰ All Norwegian children and adolescents with CP, born after 2006 (or 2002 for the south-eastern region) are invited to participate in the Cerebral Palsy Follow-up Program (CPOP), as part of standard care. National registry data from the CPOP thus provide an opportunity to conduct population-based studies and contribute to increasing the knowledge of the long-term development of motor functioning in children/ adolescents with CP. According to parents and occupational therapists in the CPOP, more than 60% of the young children perform training of hand skills as part of standard care.¹¹ The training usually occurs several times per week, both as individual sessions and integrated in everyday settings; children in MACS levels II and III are most likely to undergo the training.

The aim of this study was to describe the development of hand use during bimanual activities among a population of children with unilateral CP. More specifically, we wanted to: (1) explore whether development of hand use as measured with the AHA increases up to a stable limit of performance or follows a peak and decline curve, (2) describe developmental trajectories of hand use between children with unilateral CP classified in MACS levels I, II, and III, and (3) describe the developmental trajectories of hand use between children with unilateral CP who scored according to a high, medium, or low AHA category at 18 months of age (AHA-18).

METHOD

Design and procedure

The study had a prospective, longitudinal design and included national data from the CPOP, a surveillance programme that includes approximately 90% of the total population of children/adolescents with CP in Norway, born after 2006.^{12,13} In the CPOP, occupational therapists across the country monitor hand function in children with CP according to a standardized protocol.¹³ This study includes data that were previously published in a cohort study of younger children,⁶ but is expanded with more children and a wider age range.

Participants

All children with unilateral CP registered in the CPOP with results from two or more AHA assessments by 31st December 2018 were included in the analysis. The parents signed informed consent for their child's participation in CPOP, which also included permission to use the data in research. The included children had access to standard paediatric rehabilitation services similar to what was reported in a previous study.¹¹ The study was approved by the Data Protection Officer of Oslo University Hospital and the Regional Committee for Medical and Health Research Ethics of South-Eastern Norway (registration number 2019/30715).

What this paper adds

- Development of hand use between 18 months and 13 years follows a stable-limit pattern.
- Most children reach a steady limit on the Assisting Hand Assessment before 6 years of age.
- Manual Ability Classification System levels I, II, and III represent distinct developmental trajectories, level III having a slower rise.
- Early hand use is an important indicator of future development.

Classification and assessment

The MACS and Mini-MACS were used to classify the children's ability to handle objects in everyday activities. The MACS describes manual abilities according to five ordinal levels, where level I describes the ability to handle age-appropriate objects easily and successfully, while level V describes severely limited ability to perform simple actions.¹⁴ The Mini-MACS is an adaptation of the MACS for use with children younger than 4 years of age.¹⁵ The Norwegian translation of the Mini-MACS has been available since 2016, hence, the MACS was used for children under 4 years who were registered to the CPOP earlier than that. In cases where the MACS levels changed between assessments, the latest MACS level was used. In this paper the term MACS will be used for both versions.

The AHA is part of the standard CPOP assessment protocol and was used to evaluate changes in the use of the assisting hand over time. The AHA is a criterion-based, standardized test which measures how children with unilateral CP spontaneously use their affected hand during bimanual play.¹⁶ Occupational therapists across Norway who were certified to use the AHA completed and scored AHA assessments according to the 4.4 or 5.0 versions. The scores were converted to the comparable logit-based AHA 0 to 100 scale, with higher values indicating higher ability.¹⁷ A change of 5 or more AHA units (the smallest detectable difference) was considered a true change with 95% certainty.¹⁸

Statistical analysis

Descriptive analyses were conducted with IBM SPSS Statistics, version 25 (IBM Inc., Armonk, NY, USA) to describe the study population. To compare age at first assessment between MACS level I, II, and III, a one-way between-group analysis of variance was performed, since age was normally distributed. To compare the number of tests and time for monitoring between the MACS levels, Kruskal–Wallis tests were performed because the assumption of normality was not met.

Developmental trajectories with repeated AHA measures were estimated by non-linear mixed effects models and compared between a stable limit model (SLM) and a peak and decline model (PDM). The SLM has the shape of a negative exponential function and converges to an upper limit. It has two parameters corresponding to the limit and rate of change in AHA levels as a function of age (see Klevberg et al.⁶ for more details): AHA=limit×(1–exp(– rate×age)).¹⁵ For easier interpretability, the rate parameter was transformed to Age-90 (i.e. the age when the children reached 90% of their limit on the AHA).¹⁵ The PDM has

one extra parameter (AHA₀), which is the AHA score for any age_0 arbitrarily chosen within the range of ages observed in the data (here: 18mo). From the model, one can calculate the age at which the children reach their maximum performance (peak) before development starts to decline (Age-peak):¹⁹

$$\begin{array}{l} \mathrm{AHA} &= \left(-\mathrm{limit} \times \left(1 - \frac{\mathrm{age}}{\mathrm{age}_0}\right) + \frac{\mathrm{age}}{\mathrm{age}_0} \times \frac{\mathrm{AHA}_0 - \mathrm{limit}}{\mathrm{rate}^{\mathrm{age}_0}}\right) \\ &\times \mathrm{rate}^{\mathrm{age}} + \mathrm{limit} \end{array}$$

All model parameters were included as both fixed and random effects, except rate, which was not used as a random effect as its variation between participants was very small. Limits and rates (Age-90) of development were further described using the model (SLM or PDM), which proved the best fit to our data, as shown by the lower value of the Akaike information criterion statistics. In agreement with previous studies, developmental trajectories were described according to MACS levels I, II, and III, and according to a high (63–100), moderate (39–62), or low (0– 38) group for observed and estimated AHA units at 18 months of age.^{6–8} The third quartile was used to illustrate time-points for when the majority of the children approached their stable limits as defined by Age-90.

Sensitivity analysis of the mixed effects models was performed to explore the fit of the models when including children with two or more assessments versus including only children with three or more AHA assessments. The estimated values were almost identical in these two conditions and model fits were very similar (the within-group root mean square error was 4.30 and 4.32 respectively), indicating that the children with only two assessments are in line with the children with more assessments (i.e. there are no systematic differences). All children with two or more assessments were thus included in all further analyses. The mixed effects models were fitted with the statistical software R version 3.6.1 (R Foundation for Statistical Computing, Vienna, Austria), and, in particular, with the nlme version 3.1-141 package (R Foundation for Statistical Computing).

RESULTS

From the total population of 672 children who were registered in the CPOP and classified with unilateral CP, 166 children (25%) were registered with two AHAs (n=64) or more than two AHAs (n=102) and included in the study. Seventy-nine participants were female and 87 were male, with a mean (SD) age at first assessment of 37.6 (20.5) months (Table 1). The children were classified in MACS level I (n=28), level II (n=102), and level III (n=36) and assessed with the AHA on 524 occasions (median 3, range 2–7) for a mean (SD) period of 33.4 (24.2) months (Table 1). Comparison of participants and non-participants is shown in Table S1 (online supporting information).

The mean (SD) change of observed scores between the first and the last assessment was 9.2 (12.1) AHA units, with the largest improvement seen for children classified in MACS level III (mean 15.8 [12.4]) and the smallest (mean 4.9 [7.2]) for level I (Table 1). There were no significant differences between the MACS levels for age at first assessment (F=1.57, p=0.2) or number of tests (H=5.43, p=0.07), yet children classified in MACS level I were monitored over a shorter period than children classified in level II or III (H=9.05, p=0.01). In total, 102 participants improved their observed performance equal to or above the smallest detectable difference between the first and last AHA assessment (range 5–55 AHA units).

Table 2 shows the parameters of development as estimated with the SLM and the PDM, according to MACS levels I, II, and III. As indicated by the lower Akaike information criterion value for the global data, the SLM showed a better model fit (Akaike information criterion=4008.99) than the PDM (Akaike information criterion=6182.94), indicating that development follows an increasing curve up to a stable limit of performance before levelling off at various time-points. Also, when analyzing data for each MACS level separately, the SLM showed a better model fit.

As illustrated in Tables 2 and 3, the mean limit of performance for children classified in MACS level I was 81.4 AHA units, which was significantly higher than the limits for children classified in MACS level II (67.7 units) or level III (56.2 units). Whereas children classified in MACS level I reached their mean Age-90 at 35.5 months of age, this was achieved at 48.3 months and 92.1 months for children classified in MACS levels II and III respectively. We found significant differences between all MACS levels both for limit of performance and Age-90 (p<0.001).

When comparing the limit of performance and Age-90 between children with a high-, moderate-, or low-level score

 Table 1: Age of the participants, period for monitoring, number of assessments, and observed change of performance on the AHA between the first and the last assessment for each MACS level

	n (%)	Age at first assessment, mean (SD) range, mo	Age at last assessment, mean (SD) range, mo	Time period, mean (SD) range, mo	Number of assessments, ^a median (range)	Change in AHA 0–100 units, mean (SD) range
All participants	166 (100)	37.6 (20.5) 18–126	71.2 (30.6) 21–159	33.4 (24.2) 3–107	3.0 (2–7)	9.2 (12.1) –18 to 55
MACS level I	28 (17)	43.8 (23.4) 18–95	66.2 (30.2) 24–133	22.4 (16.0) 4-64	2.0 (2-5)	4.9 (7.2) -14 to 23
MACS level II	102 (61)	37.1 (21.3) 18–126	73.1 (32.1) 21–159	35.6 (25.7) 3–107	3.0 (2-7)	8.1 (12.2) –18 to 55
MACS level III	36 (22)	33.9 (14.1) 19–93	69.8 (26.4) 30–155	36.0 (23.1) 9–85	3.0 (2–7)	15.8 (12.4) -12 to 45

^aNumber of children with two assessments were distributed within each Manual Ability Classification System (MACS) level as follows: level I *n*=15 (54%), level II *n*=36 (35%), level III *n*=13 (36%). AHA, Assisting Hand Assessment; SD, standard deviation.

	MACS			AHA-18 ^a				
	Level I	Level II	Level III	High	Moderate	Low	AIC	Residuals SD
SLM	ç	102	26	5	6	4	1000 00	11
AHA limit (95% CI)	20 81.4 (78.3–84.6)	67.7 (66.2–69.3)	56.2 (52.0–60.3)	12 83.8 (78.3–89.2)	71.7 (69.9–73.4)	/4 60.5 (57.7–63.2)	4000.00	
Age-90 (95% CI)	35.5 (32.2–38.7)	48.3 (44.2-52.4)	92.1 (78.6–105.6)	25.6 (22.0–29.2)	37.2 (35.4–39.0)	80.4 (72.8–87.3)		
Residuals SD	4.72	7.000	6.829	N/A	N/A	N/A		
AIC model fit	529.77	2398.30	903.88	N/A	N/A	N/A		
MO								
u	28	102	36	ო	76	87	6182.94	8.01
AHA peak (95% CI)	82.9 (79.7–86.0)	66.8 (64.8–68.7)	47.2 (43.7–50.7)	89.4 (78.5–100.4)	77.0 (75.3–78.8)	54.1 (51.9–56.3)		
Age peak (95% CI)	69.6 (68.4–70.9)	73.0 (71.8–74.2)	81.6 (78.9–84.3)	56.7 (54.3–59.1)	65.9 (64.8–66.9)	151.8 (70.9–232.6)		
Residuals SD	6.49	13.08	8.84	N/A	N/A	N/A		
AIC model fit	812.62	4207.48	1533.49	N/A	N/A	N/A		

used to estimate the model fit. ^aChildren grouped into a high (63–100), moderate (39–62), or low (0–38) group for observed and estimated AHA units at 18 months of age. Since the AHA-18 variable contains values estimated from the different statistical models, the *n* for the AHA-18 groups varies between the SLM and PDM. MACS, Manual Ability Classification System; AIC, pants are estimated to reach their peak of development according to the PDM; N/A, not applicable because the AHA-18 levels are based on estimated AHA-18 units and can thus not be Akaike information criterion; Cl, confidence interval; SD, standard deviation on the AHA at 18 months of age, significant differences were also found between all the three groups (Table 3).

The significant differences presented in Table 3 for both the AHA limit and the Age-90 parameters between the three MACS levels and between the three AHA-18 levels were confirmed by the 95% confidence intervals (CIs) shown in Table 2. The graphical illustrations of the developmental trajectories (mean and 95% CI) for the total sample and for the MACS and AHA-18 subgroups show the considerable variation that was found within and between the groups (Fig. 1). The third quartile for the Age-90, according to MACS levels, was 3 years 7 months for level I, 5 years 1 month for level II, and 8 years 5 months for level III. For all the participants as a group, the median for Age-90 was 3 years 11 months and the third quartile was 5 years 10 months.

DISCUSSION

In our register-based sample of 166 children with unilateral CP, three quarters of the children had reached a steady limit of performance on the AHA by 6 years of age (as defined by the third quartile for Age-90). The most rapid development occurred before the age of 4 years for over half of the children, yet with large variability both between and within the groups. Children classified in lower MACS levels or with a better AHA performance at 18 months of age reached higher performance limits and approached their limits at younger ages compared with children in higher MACS levels and lower AHA-18 performance.

One aim of the study was to explore whether there was a decrease in development of hand use during bimanual activities after 9 years of age, as reported by Klingels et al.9 When comparing the SLM where the data are assumed to follow a steady curve until the limit of performance is reached,²⁰ with the PDM that assumes an initial increase followed by a performance decline,²¹ our results showed a better model fit for the SLM, both for the global data and when analyzing each MACS level separately. Consequently, our study agrees with previous studies that do not indicate a developmental decline.⁶⁻⁸ Three important aspects need to be considered regarding the contrasting findings. First, both the Belgian⁹ and the Swedish^{7,8} studies included convenience samples, whereas our Norwegian studies were population-based. However, this does not seem to explain the diverging developmental trajectories, as the Swedish and Norwegian results are very similar. On the other hand, Sweden and Norway monitor all children with CP through identical national surveillance programmes that aim to promote equal services for all children with CP.¹² Belgium does not have a similar programme, hence it could be that Sweden and Norway provide more equal services, and consequently their populations experience more similar development. Nevertheless, Belgium, Sweden, and Norway are welfare states where all citizens have equal access to healthcare services at a low cost, and the standard practice of interventions referred to in the Belgian study⁹ seems comparable to what

Table 3: Comparison between the mean estimated limits and rates of development on the AHA between children in MACS levels I to III, and between three groups of observed and estimated AHA units at 18 months of age

	Limit			Rate			Age-90		
	Difference	95% CI	p	Difference	95% CI	p	Difference	95% CI	p
Mini-MACS/MACS levels I–II Mini-MACS/MACS levels II–III AHA-18 high vs moderate ^a AHA-18 moderate vs low ^a	13.71 11.57 12.11 11.21	10.07–17.35 6.98–16.16 5.84–18.39 7.97–14.46	<0.001 <0.001 <0.001 <0.001	0.014 0.027 0.031 0.033	0.004-0.024 0.021-0.033 0.016-0.045 0.284-0.040	0.006 <0.001 <0.001 <0.001	-12.86 -43.79 -11.15 -41.59	-18.16 to -7.55 -58.31 to -29.27 -15.12 to -7.19 -49.39 to -33.80	<0.001 <0.001 <0.001 <0.001

^aChildren grouped into a high (63–100), moderate (39–62), or low (0–38) group for observed and estimated AHA units at 18 months of age. AHA, Assisting Hand Assessment; MACS, Manual Ability Classification System; Age-90, the age at which the participants are estimated to reach 90% of their performance limit; Cl, confidence interval.



Figure 1: Developmental trajectories of hand use illustrated according to three levels of the Manual Ability Classification System (MACS) and three levels of observed or estimated Assisting Hand Assessment (AHA) performance at 18 months of age (AHA-18). The solid lines represent the mean fitted curves for each group, surrounded by the 95% confidence intervals (CIs) illustrated by the shaded areas. The horizontal dotted lines represent the groupwise AHA limit values together with the 95% CI. The vertical dotted lines represent the groupwise Age-90 values and corresponding 95% CI.

we previously reported for children with CP in Norway.¹¹ Thus, it seems difficult to point out systematic differences that might explain the diverging developmental trajectories. Second, the statistical modelling of the developmental curves is important and should be reported in detail. In this study, we presented our results based on both the

SLM and the PDM. Similar comparisons of results based on the two models have previously been presented for gross motor function development.²² Yet in the former studies of hand use development, this was only briefly addressed in the study by Nordstrand et al.⁷ Moreover, the Belgian study used a different statistical approach than the

Scandinavian studies, fitting linear mixed models and performing statistical tests between time-points to identify changes over time - separately for three groups of children as defined by their age at baseline. It is, therefore, unknown how a stable limit or a peak and decline curve based on non-linear mixed effects models would have fit the Belgian data. Third, and most importantly, the results illustrate the importance of following the participants longitudinally over a long period. Whereas the participants in the Belgian study were assessed between 5 and 20 years of age,⁹ the corresponding range was from 18 months to 8 years, and 18 months to 12 years, in the Swedish studies.^{7,8} In our previous Norwegian study, ages ranged from 18 months to 6 years,⁶ whereas children in the present study were assessed between 18 months and 13 years of age. We do not know how the skewed age distribution between the studies might have influenced the contrasting developmental curves. It is possible that the decline seen among the participants in Klingels el al.'s9 study reflects the lack of children in the youngest age range where the most rapid development occurs. Yet, it may also be that the failure to identify a decline in the Scandinavian studies reflects the lack of older children and adolescents.

Whether to expect hand use development to follow a steady-limit or a peak and decline trajectory is important from a clinical perspective. If early performance predicts rapid development during the first years of life, before development slows down up to a stable limit, it can be argued that early intervention is of the utmost importance. If a decrease in performance after 9 years of age is expected, however, it may be important to emphasize goal-directed interventions at older ages to maintain or further enhance performance. The development of alternate forms of the AHA, which enable measurements on the same scale from infancy through adulthood, provides the opportunity to study hand use among people with unilateral CP during the lifespan.²³ Studies that include participants within a large age range are difficult to complete, yet are urgently needed. Inclusion of performance-based assessments (like the AHA) in population-based surveillance programmes is therefore encouraged.

The finding of distinct, yet largely variable developmental trajectories between children classified in MACS levels I, II, and III has implications for clinical practice.^{6–8} Parents want to know the developmental prospects for their child,³ hence illustrations of the separate developmental curves may be useful for therapists and doctors for communication purposes. At the same time, the large variability within each MACS level (Fig. 1) should be acknowledged, and every child must be viewed as an individual with distinct needs and potential.

The results of this study confirmed the distinct developmental trajectories between children who were grouped according to a high, moderate, or low performance level on the AHA at 18 months of age.⁶ Following the rationale of Nordstrand et al.,⁷ the three AHA-18 groups are based on AHA units that represent active and efficient grasping by the assisting hand (high level), active yet sometimes not efficient grasping (moderate level), and passive or no grasping (low level). The finding that children in the high and moderate AHA-18 levels reached higher performance limits and had faster development than children in the low group is of significance for individualized goal-setting and planning of interventions. The three performance levels of the AHA-18 are more clinically specific than the MACS levels that represent the global manipulation of objects during everyday activity regardless of hand use. Since a large proportion of data included in this study was collected before the Mini-MACS became available, using the three levels of AHA-18 to predict development was considered relevant, and perhaps even more important from a clinical perspective than using MACS for prediction. The distinct trajectories between the three AHA-18 groups points to the importance of facilitating early grasping, which has also previously been suggested as an important clinical feature to predict hand use development in infants.²⁴ The role of early grasping ability for hand use development needs further investigation.

The developmental trajectories presented and discussed in this paper do not distinguish between children who have participated in specific interventions or who experience cognitive limitations. Short-term effectiveness of motor learning-based upper limb interventions is well documented,²⁵ vet the role of such interventions and the timing of these for long-term development remains unknown. A recent study of children's own perceptions of learning to perform bimanual activities illustrated how learning depends on the children's motivation, and how interventions must be embedded in their everyday life.²⁶ Although our study shows that the most rapid development of hand use occurs during preschool years, the great variability between and within each MACS level illustrates that development continues at least into early adolescence, particularly for those in MACS level III. Young people with CP have conveyed how interventions must be based on meaningful activities and provide opportunities to find strategies that enable them to perform in the way most suitable to them, something that evolves through time and practice.²⁷ This perspective is not captured in the AHA-based developmental trajectories and highlights the importance of describing development from various perspectives and with different tools.

Despite being registry-based, only 25% of the children with unilateral CP registered in the CPOP had repeated AHA assessments, which enabled inclusion in this study. Comparison of the included versus non-included children shows a skewed distribution of MACS levels, with a larger proportion of children classified in MACS level I among the non-included group (Table S1). Although the AHA is part of the standard CPOP protocol, many therapists find it time-consuming and do not complete the assessments in their regular follow-up of children in the highest functional level. The developmental curves, particularly for MACS level I, should be interpreted with this in mind. The number of participants with only two assessments is also a limitation to this study, particularly with respect to the group of children classified in MACS level I. Nevertheless, statistical testing of the mixed effects models supported the inclusion of children with a minimum of two assessments, and even showed the best fit for MACS level I (see Table S2, online supporting information). Although the study is populationbased, generalization of the results to countries with a different healthcare system should be taken with caution. The developmental trajectories reported in this study are estimated for children with access to evidence-based interventions as part of the Norwegian public healthcare system, such as botulinum neurotoxin A injections, motor-learningbased therapy, and continuous monitoring over time.

CONCLUSION

Distinct developmental trajectories of hand use were identified between children with unilateral CP classified in MACS levels I, II, and III and between children grouped according to a high, moderate, or low performance level on the AHA at 18 months. Children at higher functional levels and with better early performance had a faster speed of development and reached higher developmental limits. The results indicate that children with unilateral CP maintain their attained level of hand use at least until the age of 13 years. Studies over longer time periods are encouraged to further investigate the developmental trajectories in older children and adolescents.

DATA AVAILABILITY STATEMENT

Data available on request due to privacy/ethical restrictions.

SUPPORTING INFORMATION

The following additional material may be found online:

 Table S1. Distribution of MACS levels and sex among study

 participants and non-participants, recruited from the Norwegian

 CPOP

Table S2. Developmental parameters for participants with a minimum of three AHAs according to MACS levels and levels of performance on the AHA at 18 months of age

REFERENCES

- Burgess A, Boyd RN, Ziviani J, Ware RS, Sakzewski L. Self-care and manual ability in preschool children with cerebral palsy: a longitudinal study. *Dev Med Child Neu*rol 2019; 61: 570–78.
- Klevberg GL, Ostensjo S, Krumlinde-Sundholm L, Elkjaer S, Jahnsen RB. Hand function in a population-based sample of young children with unilateral or bilateral cerebral palsy. *Phys Occup Ther Pediatr* 2017; 37: 528–40.
- Rosenbaum P. Cerebral palsy: what parents and doctors want to know. BMJ 2003; 326: 970–4.
- Novak I. Evidence-based diagnosis, health care, and rehabilitation for children with cerebral palsy. *J Child Neurol* 2014; 29: 1141–56.
- Gordon AM, Bleyenheuft Y, Steenbergen B. Pathophysiology of impaired hand function in children with unilateral cerebral palsy. *Dev Med Child Neurol* 2013; 55(Suppl 4): 32–7.
- Klevberg GL, Elvrum A-KG, Zucknick M, et al. Development of bimanual performance in young children with cerebral palsy. *Dev Med Child Neurol* 2018; 60: 490–7.
- Nordstrand L, Eliasson AC, Holmefur M. Longitudinal development of hand function in children with unilateral spastic cerebral palsy aged 18 months to 12 years. *Dev Med Child Neurol* 2016; 58: 1042–8.
- Holmefur M, Krumlinde-Sundholm L, Bergström J, Eliasson AC. Longitudinal development of hand function in children with unilateral cerebral palsy. *Dev Med Child Neurol* 2010; 52: 352–7.
- Klingels K, Meyer S, Mailleux L, et al. Time course of upper limb function in children with unilateral cerebral palsy: a five-year follow-up study. *Neural Plast* 2018; 2018: 2831342.
- Ringard A, Sagan A, Sperre Saunes I, Lindahl AK. Norway: health system review. *Health Syst Transit* 2013; 15: 1–162.

- Klevberg GL, Ostensjo S, Elkjaer S, Kjeken I, Jahnsen RB. Hand function in young children with cerebral palsy: current practice and parent-reported benefits. *Phys Occup Ther Pediatr* 2017; 37: 222–37.
- Hollung SJ, Vik T, Wiik R, Bakken IJ, Andersen GL. Completeness and correctness of cerebral palsy diagnoses in two health registers: implications for estimating prevalence. *Dev Med Child Neurol* 2017; 59: 402–6.
- Alriksson-Schmidt AI, Arner M, Westbom L, et al. A combined surveillance program and quality register improves management of childhood disability. *Disabil Rebabil* 2017; 39: 830–6.
- Eliasson A-C, Krumlinde-Sundholm L, Rösblad B, et al. The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Dev Med Child Neu*rol 2006; 48: 549–54.
- Eliasson AC, Ullenhag A, Wahlstrom U, Krumlinde-Sundholm L. Mini-MACS: development of the Manual Ability Classification System for children younger than 4 years of age with signs of cerebral palsy. *Dev Med Child Neurol* 2017; 59: 72–8.
- Krumlinde-Sundholm L, Eliasson A-C. Development of the Assisting Hand Assessment: a Rasch-built measure intended for children with unilateral upper limb impairments. Scand J Occup Ther 2003; 10: 16–26.
- Holmefur MM, Krumlinde-Sundholm L. Psychometric properties of a revised version of the Assisting Hand Assessment (Kids-AHA 5.0). *Dev Med Child Neurol* 2016; 58: 618–24.
- Krumlinde-Sundholm L. Reporting outcomes of the Assisting Hand Assessment: what scale should be used? Dev Med Child Neurol 2012; 54: 807–8.
- Ratkowsky D, Giles D. Handbook of Nonlinear Regression Models. New York: Marcel Dekker, 1990.

- Rosenbaum PL, Walter SD, Hanna SE, et al. Prognosis for gross motor function in cerebral palsy: creation of motor development curves. *JAMA* 2002; 288: 1357–63.
- Hanna SE, Rosenbaum PL, Bartlett DJ, et al. Stability and decline in gross motor function among children and youth with cerebral palsy aged 2 to 21 years. *Dev Med Child Neurol* 2009; **51**: 295–302.
- Smits D-W, Gorter JW, Hanna SE, et al. Longitudinal development of gross motor function among Dutch children and young adults with cerebral palsy: an investigation of motor growth curves. *Dev Med Child Neural* 2013; 55: 378–84.
- Louwers A, Beelen A, Holmefur M, Krumlinde-Sundholm L. Development of the Assisting Hand Assessment for adolescents (Ad-AHA) and validation of the AHA from 18 months to 18 years. *Dev Med Child Neurol* 2016; 58: 1303–9.
- Sakzewski L, Sicola E, Verhage CH, Sgandurra G, Eliasson AC. Development of hand function during the first year of life in children with unilateral cerebral palsy. *Dev Med Child Neurol* 2019; 61: 563–69.
- Novak I, Morgan C, Fahey M, et al. State of the evidence traffic lights 2019: systematic review of interventions for preventing and treating children with cerebral palsy. *Curr Neural Neurosci Rep* 2020; 20: 3.
- 26. Lidman G, Himmelmann K, Peny-Dahlstrand M. Managing to learn bimanual activities – experiences from children and adolescents with cerebral palsy – a qualitative analysis. *Disabil Rebabil* 2020; 1–9. https:// doi.org/10.1080/09638288.2020.1768305. Online ahead of print.
- Bergqvist L, Öhrvall A-M, Himmelmann K, Peny-Dahlstrand M. When I do, I become someone: experiences of occupational performance in young adults with cerebral palsy. *Disabil Rebabil* 2019; 41: 341–7.