Functional development in children with cerebral palsy in Uganda: population-based longitudinal cohort study

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ABBREVIATIONS

CFCS	Communication Function Classification System
GMFM-66	Gross Motor Function Measure-66 items
HIC	High-income countries
LMIC	Low- and middle-income countries
MACS	Manual Ability Classification System
PEDI-UG	Pediatric Evaluation of Disability Inventory, Ugandan version

AIM To follow the functional development of a population-based cohort of children with cerebral palsy (CP) in rural Uganda and compare their development with the developmental trajectories of children from high-income countries (HIC).

METHOD Eighty-one children (33 females, 48 males) aged 2 to 17 years (mean 8y 6mo, SD 4y 6mo) with CP were initially assessed in 2015 and then 4 years later using the 66-item Gross Motor Function Measure (GMFM-66), Pediatric Evaluation of Disability Inventory, Ugandan version (PEDI-UG), and functional classification systems. We calculated actual and reference scores (level of deviation from the developmental trajectories in HIC). A Wilcoxon signed-rank test was used for statistical analyses.

RESULTS Children and young people with CP in Uganda exhibited no differences in scores between the first and second assessments for the GMFM-66 and PEDI-UG mobility skills, whereas they exhibited increased PEDI-UG social function ($p<0.001$) and self-care skills scores ($p<0.001$). Reference scores were more negative at the second assessment than at the first for the GMFM-66 ($p=0.002$) and PEDI-UG mobility ($p=0.036$) but not for PEDI-UG self-care. The increased difference in reference scores over the 4 years was primarily driven by younger children (2–5y) and children with milder impairments.

INTERPRETATION The increased difference in reference scores between assessments suggests that children with CP in Uganda develop motor skills at a slower rate than peers in HIC. Limited access to health care and rehabilitation likely contributed to the lower scores and slower rate of development.

Cerebral palsy (CP) is one of the most common childhood-onset motor disorders and presents with a wide range of motor impairments. In its mildest form, children can perform most common activities of daily living independently, whereas children with the most severe impairments are unable to walk and require assistance in all activities of daily living.[1] Children with CP develop most of their functional skills at an early age; however, depending on the severity of their impairments, development follows different trajectories. In children with milder impairments, developmental trajectories closely follow those of typically developing children, while trajectories are much flatter in children with severe forms of CP.[2,3]

Over the last couple of decades, several assessment tools and functional classification systems have been created that can be used to predict the long-term development of children and young people with CP. This has been very helpful to both health care providers and caregivers since knowledge of children's functional prognosis can assist in planning interventions according to functional level.[2–4] Since most children and young people with CP worldwide live in low- and middle-income countries (LMIC),[5–7] it might be problematic that these assessment tools and classification systems were developed in high-income countries (HIC) where most children and young people have access to comprehensive rehabilitation programmes that are unavailable in LMIC. To fill the wide gap of tools measuring functioning and activity in children with disability living in sub-Saharan Africa, we previously translated and adapted the original Pediatric Evaluation of Disability Inventory (PEDI) developed in the USA to the culture and life conditions in Uganda (PEDI-UG) and showed that the PEDI-UG has good-to-excellent psychometric properties to measure the functional performance of typically developing children aged 6 months to 7 years 6 months in Uganda.[8,9] In a recent study of PEDI-UG in children and young people with CP in Uganda aged 2 to 22 years, we found a high validity (Amer et al., unpublished material).

When we used the PEDI-UG in our recent population-based study describing functional limitations among children and young people with CP in Uganda, we noted that these children and young people had not reached the same level of mobility and self-care skills as children and young people with CP in Sweden, suggesting that Ugandan children and young people with CP have a different developmental trajectory.[10] However, the previous study was cross-sectional and a longitudinal study is necessary to evaluate development over time more definitively.

The aims of this study were to follow the functional development over 4 years in children and young people with CP in Uganda with different levels of gross motor functioning and age groups to compare their development with children and young people with CP in HIC. Based on our previous observations, we hypothesized that children and young people in Uganda would exhibit both a lower attainment of functional skills and a slower rate of development than children and young people in HIC, which suggests that current classification systems are less accurate in predicting development in this group of Ugandan children.

What this paper adds

- Children with cerebral palsy in Uganda developed self-care skills and social function but not gross motor function and mobility.
- Functional classification level changed in more than one-third of children over the 4 years from first to second assessment.
- Children in Uganda attained lower motor skills scores and had a slower rate of development than children in high-income countries (HIC).
- The youngest and least impaired children deviated most from the developmental trajectories of children in HIC.

METHOD

Study design

This longitudinal study evaluated a population-based cohort of children and young people with CP in Uganda over a 4-year period in the Iganga Mayuge Health and Demographic Surveillance Site (IMHDSS), which covers 65 villages in a mainly rural area in eastern Uganda. Children and young people with CP were identified through a three-stage screening procedure in 2015 which included (1) door-to-door screening of all individuals aged 2 to 17 years living in the area (31 756), (2) follow-up by a trained CP team, (3) clinical neurological assessment and a triangulation of all villages in the IMHDSS using key village informants, as previously described.[5] The study was approved by the Higher Degrees Research and Ethics Committee of the School of Public Health, College of Health Sciences, Makerere University and the Uganda National Council for Science and Technology (protocols 280, 631, HS1787, and HS2608 respectively). All caregivers provided written informed consent in both 2015 and 2019; assent was obtained from study participants whenever possible.

Participants and measurements

In 2015, an expert team of physiotherapists and occupational therapists assessed 97 children and young people diagnosed with CP at the participants' home or nearby health facility.[10] A second assessment of each surviving child and young person was done 4 years later. Participants were assessed using the 66-item Gross Motor Function Measure (GMFM-66), which evaluates changes in gross motor function in children and young people with CP.[11,12] To measure mobility, self-care, and social function, caregivers were interviewed by a therapist using the PEDI-UG.

The level of functional impairment was assessed using the revised and expanded version of the Gross Motor Function Classification System (GMFCS), the Manual Ability Classification System (MACS), and the Communication Function Classification System (CFCS).[13–15] These systems use a 5-point ordinal scale from I (independent) to V (requires assistance in all activities).

Statistical analysis

GMFM-66 raw scores were converted to a 0 to 100 scaled score using the Gross Motor Ability Estimator software.[16] PEDI-UG raw scores for mobility, self-care, and social function were converted to 0 to 100 scaled scores using a conversion table developed from a study of typically developing children.[9] The median and interquartile range of the GMFM-66, PEDI-UG mobility, PEDI-UG self-care, and PEDI-UG social function scores were determined for the first and second assessments. The median and interquartile range were also determined for the 'score change', which was calculated as the difference in scores between the two assessment times (second minus first) for each participant.

To test the hypothesis that developmental rates differ between children and young people with CP in Uganda and children and young people with CP in HIC, we compared our data to developmental trajectories derived from studies in HIC. For the GMFM-66, we used tabulated reference centiles for children with CP developed from a large longitudinal study in Canada of children aged 2 to 12 years.[4,16,17] For the PEDI-UG mobility and self-care, we used modelled trajectories developed from large longitudinal studies of children and young people with CP aged 1 to 21 years in the Netherlands and Canada.[3] These reference centiles and modelled trajectories were developed to determine predicted development for children with CP based on age and GMFCS level. We determined the amount of deviation (either positive or negative) of the scores of each child and young person in our study from the 50th reference centile for GMFM-66 and the modelled developmental trajectories for the PEDI-UG mobility and self-care.[3,4,16,17] These 'reference scores' were calculated for both the first and second assessment using the GMFCS level at the first assessment and age at the respective assessments. The 'reference score change' was calculated as the difference in reference scores between the two assessment times (second minus first) for each participant. GMFM-66 reference centiles for 12-year-olds were used for participants older than 12 years.

To evaluate whether significant development occurred between the first and second assessment, we used a Wilcoxon signed-rank test to compare scores and reference scores. To analyse the effects of baseline motor function, we evaluated three subgroups based on GMFCS level at first assessment: GMFCS levels I and II; GMFCS level III; and GMFCS levels IV and V. To analyse the age effect, we evaluated three subgroups based on age at first assessment: 2 to 5 years; 6 to 11 years; and 12 to 18 years.

Stata v14.2 (StataCorp LLC, College Station, TX, USA) was used for all statistical analyses. Original *p*-values less than 0.05 were considered significant. A Bonferroni correction was applied to correct for multiple comparisons; adjusted alpha was set at 0.007 and is noted in the table legends.

RESULTS

Participants

Of the 97 children and young people with CP diagnosed in 2015,[5] 15 had died by the 4-year follow-up. Mean age at death was 10 years 2 months. Nine individuals who died were in GMFCS levels III to V.[18] Another child was unavailable for the 2015 assessment; thus, the number of study participants was 81 (33 females, 48 males) aged 2 to 17 years (mean 8y 6mo, SD 4y 6mo). The duration between first and second assessment ranged between 2 years 10 months and 4 years 7 months, with a mean of 4 years 1 month. Participant characteristics are presented in Table 1. More than half of participants were male (59%) and most families lived in rural areas (72%) as subsistence farmers (67%).

Table 1: Baseline characteristics of children with cerebral palsy and their main caregivers

Category	*n*=81 (%)
Age at first assessment	
2–5y	26 (32)
6–11y	30 (37)
12–18y	25 (31)
Sex	
Female	33 (41)
Male	48 (59)
Residence area	
Semi-urban	23 (28)
Rural	58 (72)
Main caregiver	
Mother	57 (70)
Grandmother	16 (20)
Father	4 (5)
Other	4 (5)
Main caregiver occupation	
Subsistence farmer	54 (67)
Petty trade	12 (15)
Other	9 (11)
Missing	6 (7)
GMFCS level at first assessment	
I	47 (58)
II	6 (7)
III	16 (20)
IV	5 (6)
V	7 (9)

All information presented in this table is from the first assessment in 2015, including age and Gross Motor Function Classification System (GMFCS) level at first assessment.

Functional classification levels

The GMFCS, MACS, and CFCS level ratings at the first and second assessment are shown in Table 2. Regarding the GMFCS, the classification level was unchanged in 57% of participants, increased (lower function) in 33%, and decreased in 10%. In four children, the GMFCS increased more than two levels. According to the caregivers, these children had been subjected to motorbike accident, fall from a veranda with injury to the hip and spine, typhoid fever, or no known adverse event. MACS level did not change in 60% of participants, increased in 20%, and decreased in 20%. CFCS level was unchanged in 55% of participants, decreased (better function) in 35%, and increased in 10%.

Changes in GMFM-66 and PEDI-UG

The GMFM-66, PEDI-UG mobility, PEDI-UG self-care, and PEDI-UG social function scores are shown in Table 3. GMFM-66 scores did not change between the first and second assessment in 77 children with two measures (p=0.792). When analysed according to age, these

Table 2: GMFCS, MACS, and CFCS levels for the two ratings

	GMFCS second rating					
GMFCS first rating	I	II	III	IV	V	Total
I	**29**	15	1	1	1	47
II	3	**1**	1	1	0	6
III	0	1	**11**	4	0	16
IV	0	0	0	**2**	3	5
V	0	0	0	4	**3**	7
Total	32	17	13	12	7	81
	MACS second rating					
MACS first rating	I	II	III	IV	V	Total
I	**13**	6	0	0	0	19
II	6	**23**	8	1	1	39
III	2	1	**4**	0	0	7
IV	0	0	5	**2**	0	7
V	0	0	1	1	**7**	9
Total	21	30	18	4	8	81
	CFCS second rating					
CFCS first rating	I	II	III	IV	V	Total
I	**20**	1	0	0	0	21
II	4	2	0	1	0	7
III	6	5	**8**	5	1	25
IV	0	0	4	**4**	0	8
V	0	0	1	8	**11**	20
Total	30	8	13	18	12	81

GMFCS, MACS, and CFCS for two ratings of 81 children with 4 years between ratings. Bold type indicates the number of children rated at the same level at both first and second rating. GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CFCS, Communication Function Classification System.

scores were unchanged in children younger than 12 years but decreased in children and young people aged 12 to 18 years (score change of −4.3). No significant changes in GMFM-66 scores were observed in any GMFCS level subgroup.

PEDI-UG mobility scores were unchanged between the first and second assessments in the 80 children assessed twice with the PEDI-UG (p=0.171) and in all age groups (Table 3). When analysed according to GMFCS level, PEDI-UG mobility scores increased in the GMFCS level III group (score change of +4.9) but were unchanged in the GMFCS levels I, II, IV, and V groups.

PEDI-UG self-care scores increased from the first to second assessment in the whole cohort (score change of +4.5, p<0.001; Table 3). These scores also increased in the younger age groups but did not change in the 12 to 18 years group. PEDI-UG self-care scores increased in the two lower GMFCS groups but were the same in the GMFCS levels IV and V group.

PEDI-UG social function scores increased between the first and second assessments in the whole cohort (score change of +10.0, p<0.001), in the two younger age groups, and in children with mild and moderate impairment (Table 3). It was unchanged for children and young people aged 12 to 18 years and children and young people with more severe impairment (GMFCS levels IV and V).

Comparison with developmental trajectories from HIC

Figure 1 shows the GMFM-66 scores at the first and second assessment of each participant and the developmental trajectory from HIC.[4,16,17] Reference scores and reference score changes are presented in Table 4. Most children and

Table 3: GMFM-66 and PEDI-UG mobility, self-care, and social function scores

Group	*n*	First assessment	Second assessment	Score change	*p*
GMFM-66					
All children	77[a]	60.1 (42.2–71.2)	57.9 (36.8–72.2)	0 (−5.0 to 4.1)	0.792
Age at first assessment					
2–5y	26	48.0 (32.3–66.3)	44.9 (18.9–69.2)	1.7 (−0.4 to 4.4)	0.124
6–11y	28	66.2 (46.0–76.6)	66.5 (46.1–76.9)	0.4 (−3.0 to 4.9)	0.531
12–18y	23	66.0 (41.4–78.3)	57.9 (31.8–73.1)	−4.3 (−19.7 to 2.7)	0.025
GMFCS level at first assessment					
I and II	49	68.9 (64.3–80.0)	70.4 (60.4–79.1)	0.8 (−11.8 to 4.5)	0.804
III	16	42.2 (36.8–44.5)	43.0 (35.5–45.1)	0.1 (−3.2 to 3.1)	0.756
IV and V	12	21.6 (13.5–28.0)	16.5 (14.2–21.0)	0 (−4.1 to 0)	0.268
PEDI-UG mobility					
All children	80[b]	67.0 (48.0–76.4)	68.3 (49.5–80.0)	1.2 (−3.5 to 10.6)	0.171
Age at first assessment					
2–5y	26	52.4 (34.8–64.7)	53.4 (30.5–72.7)	3.3 (−2.0 to 11.8)	0.162
6–11y	29	71.1 (57.6–76.4)	69.7 (61.5–85.1)	2.8 (−3.0 to 11.8)	0.202
12–18y	25	74.4 (64.7–81.4)	72.7 (43.6–78.7)	0 (−8.7 to 4.1)	0.607
GMFCS level at first assessment					
I and II	53	72.7 (67.0–78.7)	74.4 (68.3–85.1)	0 (−6.1 to 10.8)	0.378
III	15	48.0 (39.7–51.9)	51.9 (48.0–55.7)	4.9 (−1.0 to 11.3)	0.011
IV and V	12	27.7 (19.2–33.8)	23.5 (0–30.5)	0 (−15.3 to 5.9)	0.503
PEDI-UG self-care					
All children	80[b]	57.6 (42.2–77.8)	66.7 (43.8–81.5)	4.5 (−1.8 to 11.3)	<0.001
Age at first assessment					
2–5y	26	50.0 (33.9–56.0)	60.3 (35.1–67.1)	9.6 (−0.8 to 15.8)	0.006
6–11y	29	71.6 (55.3–79.5)	79.5 (59.9–87.2)	6.1 (0–10.7)	0.001
12–18y	25	69.7 (49.3–87.2)	64.8 (45.0–81.5)	0 (−5.7 to 4.8)	0.839
GMFCS level at first assessment					
I and II	53	71.6 (55.3–81.5)	79.5 (63.3–87.2)	3.2 (−3.4 to 10.7)	0.025
III	15	41.8 (35.1–54.0)	53.3 (40.9–71.6)	7.9 (4.8–14.9)	0.001
IV and V	12	28.6 (24.3–32.7)	27.9 (25.2–32.6)	2.7 (−5.7 to 5)	0.875
PEDI-UG social function					
All children	80	53.0 (37.2–68.5)	65.1 (43.4–85.1)	10.0 (0.4–16.7)	<0.001
Age at first assessment					
2–5y	26	47.0 (34.5–55.2)	60.8 (40.5–76.9)	13.8 (6.7–19.1)	0.001
6–11y	29	65.1 (49.2–75.3)	78.9 (53.0–91.1)	12.2 (5.9–17.0)	<0.001
12–18y	25	50.0 (36.7–71.2)	58.3 (39.6–73.8)	6.2 (−3.6 to 10.5)	0.106
GMFCS level at first assessment					
I and II	53	64.2 (47.7–75.3)	75.3 (53.0–91.1)	10.0 (4.4–18.1)	<0.001
III	15	43.0 (30.5–56.7)	59.9 (32.0–75.3)	15.3 (0–16.9)	0.006
IV and V	12	31.1 (19.8–39.4)	33.1 (14.8–43.3)	1.2 (−6.5 to 11.1)	0.610

Data are median (IQR) unless otherwise stated. Median scores and median score change in GMFM-66 and PEDI-UG mobility, self-care, and social function and results from the Wilcoxon signed-rank test. GMFM-66, 66-item Gross Motor Function Measure; GMFCS, Gross Motor Function Classification System; PEDI-UG, Pediatric Evaluation of Disability Inventory, Ugandan version; IQR, interquartile range. [a]The total number of children included in the GMFM-66 is 77 because four children were missing the 2015 measures. [b]The total number of children included in the PEDI-UG analysis is 80 because one child was missing the 2019 measure. When we adjust the significance level for multiple comparisons, the adjusted p-value is 0.007 for an original p-value of 0.05.

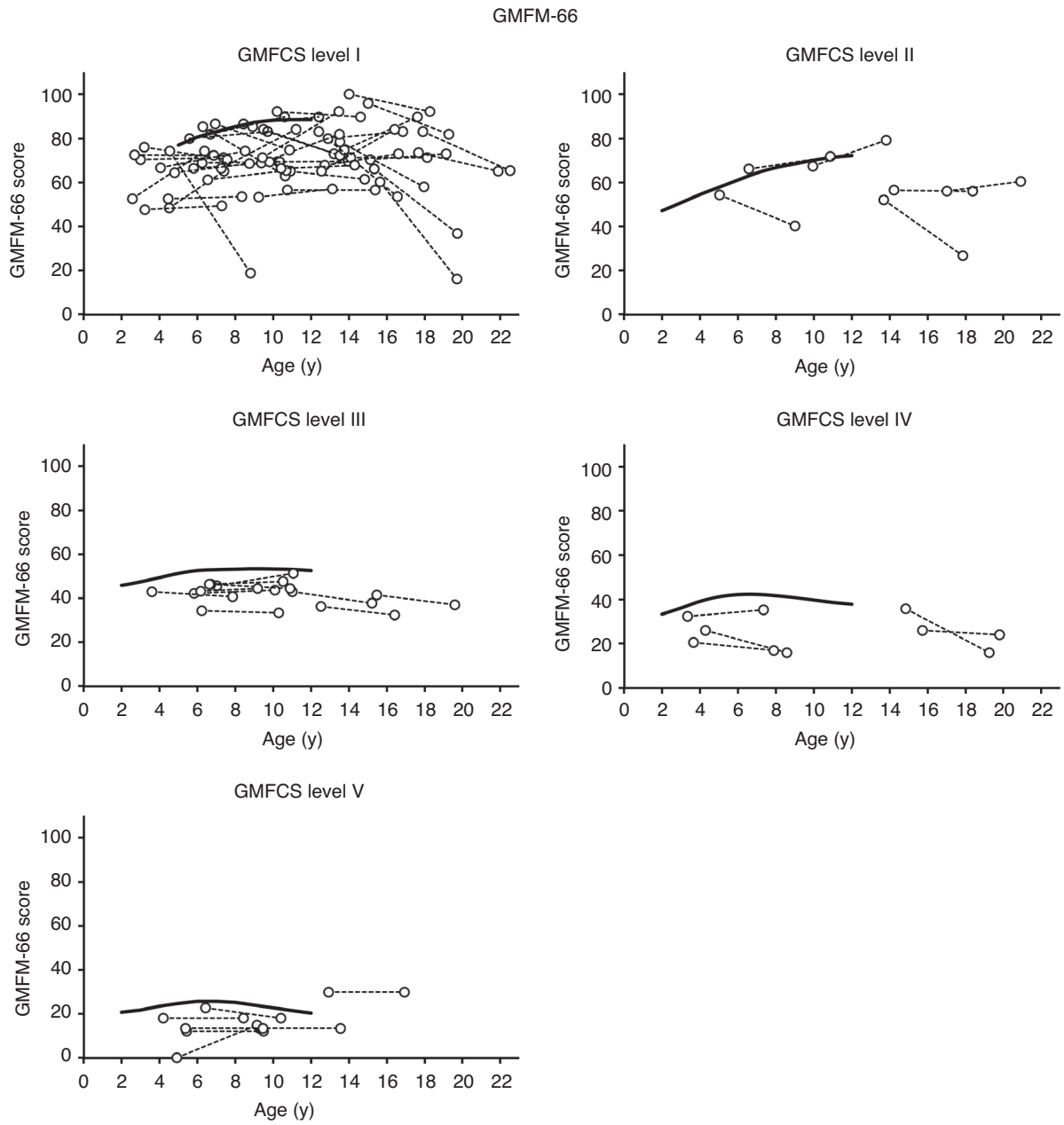

Figure 1: Scatter plots showing the 66-item Gross Motor Function Measure (GMFM-66) scores of 77 children and young people with cerebral palsy in Uganda (empty circles) in 2015 and 2019. The paired values for each participant are connected with a dashed line. Plots are shown for each Gross Motor Function Classification System (GMFCS) level at the first assessment. The solid line illustrates the 50th centile GMFM-66 score from a large population-based sample of children with cerebral palsy in Canada.[4,16,17]

young people in our Ugandan cohort scored below the 50th centile of the HIC reference at the first assessment, with 21% below −3 SDs. This proportion increased to 30% at the second assessment. In the whole cohort, median reference scores were negative at both the first and second assessment (−10.3 and −13.8 respectively). Negativity was greater at the second assessment (reference score change of −2.3, p=0.002), suggesting that children and young people in Uganda exhibited negative deviation from the developmental trajectory of individuals from HIC. When considering age subgroups, negative deviation was observed in the youngest (2–5y) and oldest (12–18y) groups but not in children aged 6 to 11 years. Negative deviation was observed in the GMFCS levels I and II group (reference score change of −5.7) but not in the GMFCS levels III, IV, and V groups.

Individual PEDI mobility scores in relation to the developmental trajectories from HIC[3] are plotted in Figure 2

Table 4: GMFM-66 and PEDI-UG mobility and self-care reference scores

Group	*n*	First assessment	Second assessment	Reference score change	*p*
GMFM-66					
All children	77[a]	−10.3 (−17.5 to −3.4)	−13.8 (−20.8 to −6.9)	−2.3 (−11.5 to 1.4)	0.002
Age at first assessment					
2–5y	26	−9.4 (−13.5 to −3.4)	−11.8 (−19.2 to −9.3)	−5.9 (−12.1 to 1.3)	0.016
6–11y	28	−9.7 (−18.5 to −2.0)	−11.4 (−19.6 to −3.0)	−0.7 (−3.1 to 2.2)	0.494
12–18y	23	−14.8 (−18.0 to −5.4)	−16.3 (−23.4 to −6.5)	−4.3 (−19.7 to 2.7)	0.025
GMFCS level at first assessment					
I and II	49	−12.4 (−19.0 to −0.1)	−16.2 (−25.4 to −6.5)	−5.7 (−17.7 to 2.1)	0.003
III	16	−9.8 (−14.0 to −7.0)	−10.2 (−17.4 to −8.2)	−1.5 (−4.2 to 0.9)	0.379
IV and V	12	−11.0 (−13.4 to −3.8)	−9.8 (−17.8 to −6.9)	−1.5 (−5.0 to 1.5)	0.326
PEDI-UG mobility					
All children	80[b]	−13.2 (−19.2 to −4.6)	−15.7 (−24.4 to −6.8)	−1.9 (−12.6 to 4.3)	0.036
Age at first assessment					
2–5y	26	−6.0 (−10.8 to −0.6)	−15.4 (−23.5 to −5.9)	−9.8 (−17.0 to 0.5)	0.004
6–11y	29	−15.6 (−20.2 to −8.6)	−14.2 (−24.3 to −7.7)	−0.2 (−8.1 to 10.0)	0.991
12–18y	25	−16.5 (−22.4 to −13.8)	−19.2 (−25.2 to 8.6)	0 (−8.7 to 4.1)	0.589
GMFCS level at first assessment					
I and II	53	−15.0 (−19.2 to −10.2)	−17.0 (−25.0 to −8.6)	−2.0 (−12.8 to 4.7)	0.144
III	15	−10.0 (−20.0 to −5.3)	−13.4 (−20.7 to −10.8)	−2.3 (−9.1 to 4.2)	0.281
IV and V	12	−3.4 (−8.7 to 2.2)	−8.3 (−23.5 to 5.9)	−0.1 (−16.5 to 3.1)	0.289
PEDI-UG self-care					
All children	80[b]	−10.4 (−21.0 to −2.5)	−10.0 (−27.1 to −1.3)	−0.2 (−8.4 to 5.1)	0.300
Age at first assessment					
2–5y	26	−9.4 (−16.3 to −2.6)	−11.5 (−24.6 to −5.7)	−7.9 (−13.8 to 2.8)	0.059
6–11y	29	−10.3 (−18.1 to −2.4)	−4.3 (−17.2 to 1.2)	1.6 (−4.0 to 6.3)	0.496
12–18y	25	−14.0 (−35.2 to −2.6)	−15.5 (−29.8 to 8.2)	0 (−7.3 to 4.8)	0.657
GMFCS level at first assessment					
I and II	53	−7.4 (−17.3 to −1.6)	−10.3 (−25.1 to 0.7)	−1.5 (−9.3 to 3.4)	0.053
III	15	−21.7 (−35.2 to −12.2)	−17.5 (−33.9 to −1.4)	3.9 (−1.3 to 9.1)	0.078
IV and V	12	−8.6 (−13.6 to −1.4)	−7.1 (−17.9 to −2.8)	2.0 (−8.3 to 2.9)	0.530

Data are median (IQR) unless otherwise stated. Median reference scores and median reference score change in GMFCS-66 and PEDI-UG (mobility, self-care, and social function) and results from the Wilcoxon signed-rank test. [a]The total number of children included in the GMFM-66 is 77 because four children were missing the 2015 measures. [b]The total number of children included in the PEDI-UG analysis is 80 because one child was missing the 2019 measure. When we adjust the significance level for multiple comparisons, the adjusted *p*-value is 0.007 for an original *p*-value of 0.05. GMFM-66, 66-item Gross Motor Function Measure; IQR, interquartile range; GMFCS, Gross Motor Function Classification System; PEDI-UG, Pediatric Evaluation of Disability Inventory, Ugandan version.

and reference scores are presented in Table 4. In the whole cohort, mean reference scores were negative at both the first and second assessment (−13.2 and −15.7 respectively). Scores were more negative at the second assessment compared with the first (reference score change of −1.9, p=0.036). This negative deviation from the HIC developmental trajectory was also found in the 2 to 5 years age group (reference score change of −9.8), while no significant deviations were found in the older age groups or in any GMFCS subgroup.

Individual PEDI-UG self-care scores are plotted in Figure 3 and reference scores are shown in Table 4. Reference scores in the whole cohort were negative at both the first and second assessment (−10.4 and −10.0 respectively). There was no significant difference in reference scores between the two assessments (reference score change of −0.2, p=0.300), indicating that the developmental trajectory of our cohort was like that of children and young people in HIC.

We could not calculate reference scores or the reference score change for PEDI-UG social function because the PEDI reference material[3] did not include any modelled trajectories for social function. Thus, no comparison could be made with developmental trajectories in HIC.

DISCUSSION

The main finding of this study was that children and young people with CP in Uganda attained lower scores in functional skills and had a slower developmental rate in gross motor function than children and young people in HIC. Younger children and those with milder impairments did not follow predicted developmental changes based on HIC data. This was a consequence of poor development of gross motor function and mobility. These children and young people had limited access to health care and rehabilitation services,[10] which may contribute to them failing to reach their full developmental potential.

The finding that children and young people with mild impairments (GMFCS levels I and II) exhibited no development in gross motor function or mobility during the 4-year follow-up was unexpected and differed from the results of studies from HIC, where children with milder impairments exhibited a steep developmental trajectory for gross motor function and mobility during the first 6 years of life.[2,3] When the development of children and young people in GMFCS levels I and II was compared to the steep developmental trajectory reported in HIC, we observed a negative deviation from the expected development for gross motor function. Similar negative deviation (although not

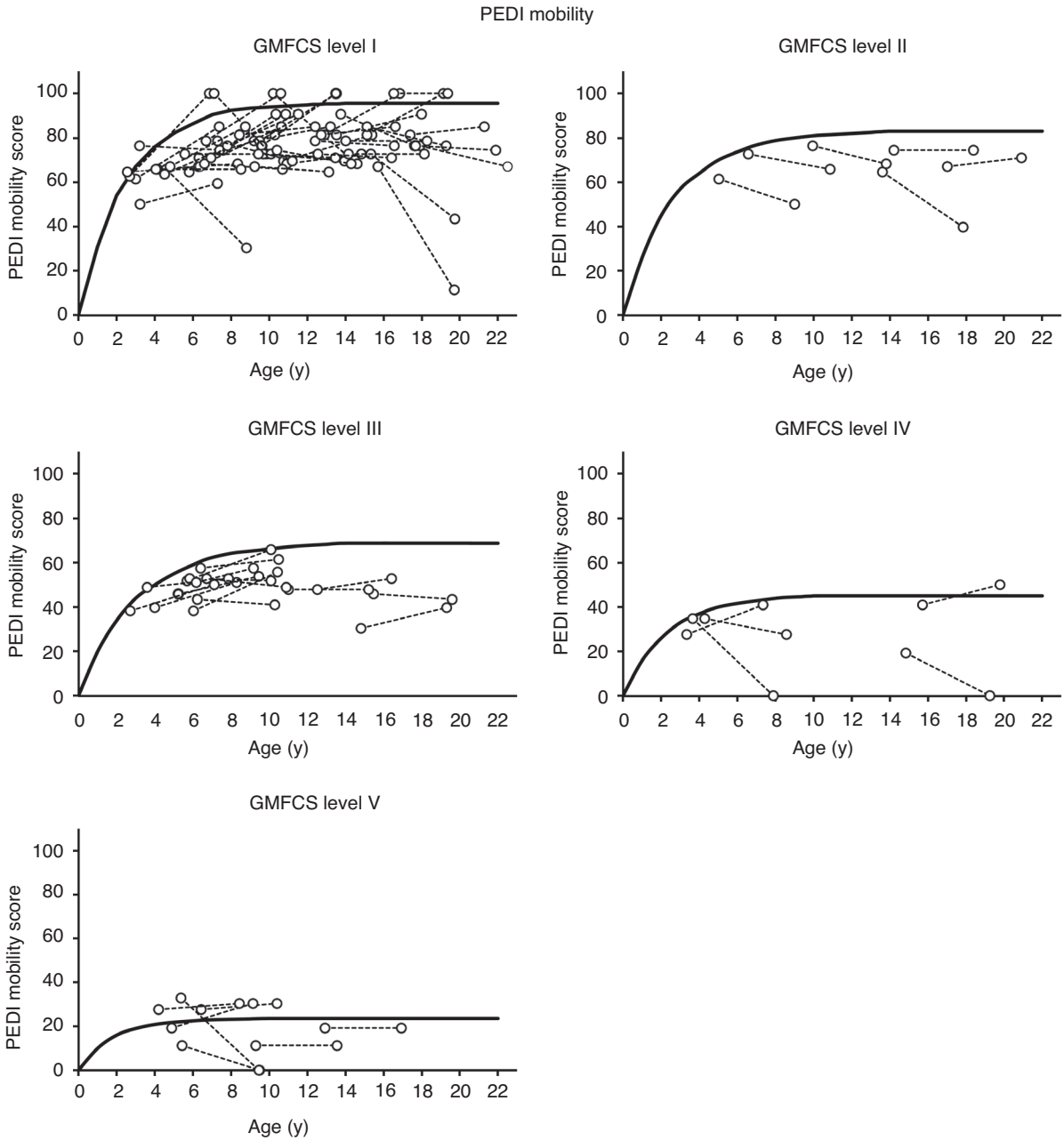

Figure 2: Scatter plots showing the Pediatric Evaluation of Disability Inventory, Ugandan version (PEDI-UG) mobility scores of 80 children and young people with cerebral palsy in Uganda (empty circles) in 2015 and 2019. The paired values for each participant are connected with a dashed line. Plots are shown for each Gross Motor Function Classification System (GMFCS) level at the first assessment. The solid line illustrates a modelled PEDI mobility trajectory developed from a large sample of children with cerebral palsy from Canada and the Netherlands.[3]

significant) from the HIC developmental trajectory for self-care skills was found in children and young people with mild impairments, despite their increased function over time, indicating that the developmental rate was lower than in children with CP from HIC. These negative deviations from developmental trajectories suggest that children and young people with milder forms of CP in Uganda do not achieve their full developmental potential. While previous studies of clinical cohorts of children with CP in Africa reported low proportions of children with milder motor impairments (23–30%),[19-21] the current and other population-based studies from Africa[22] suggest that 58% to 65% of children and young people are in GMFCS levels I and II. The difference in proportions likely reflects a recruitment bias of clinical cohort studies because children with milder impairments are probably not receiving health care to the same extent. Thus, children with CP who may benefit most from

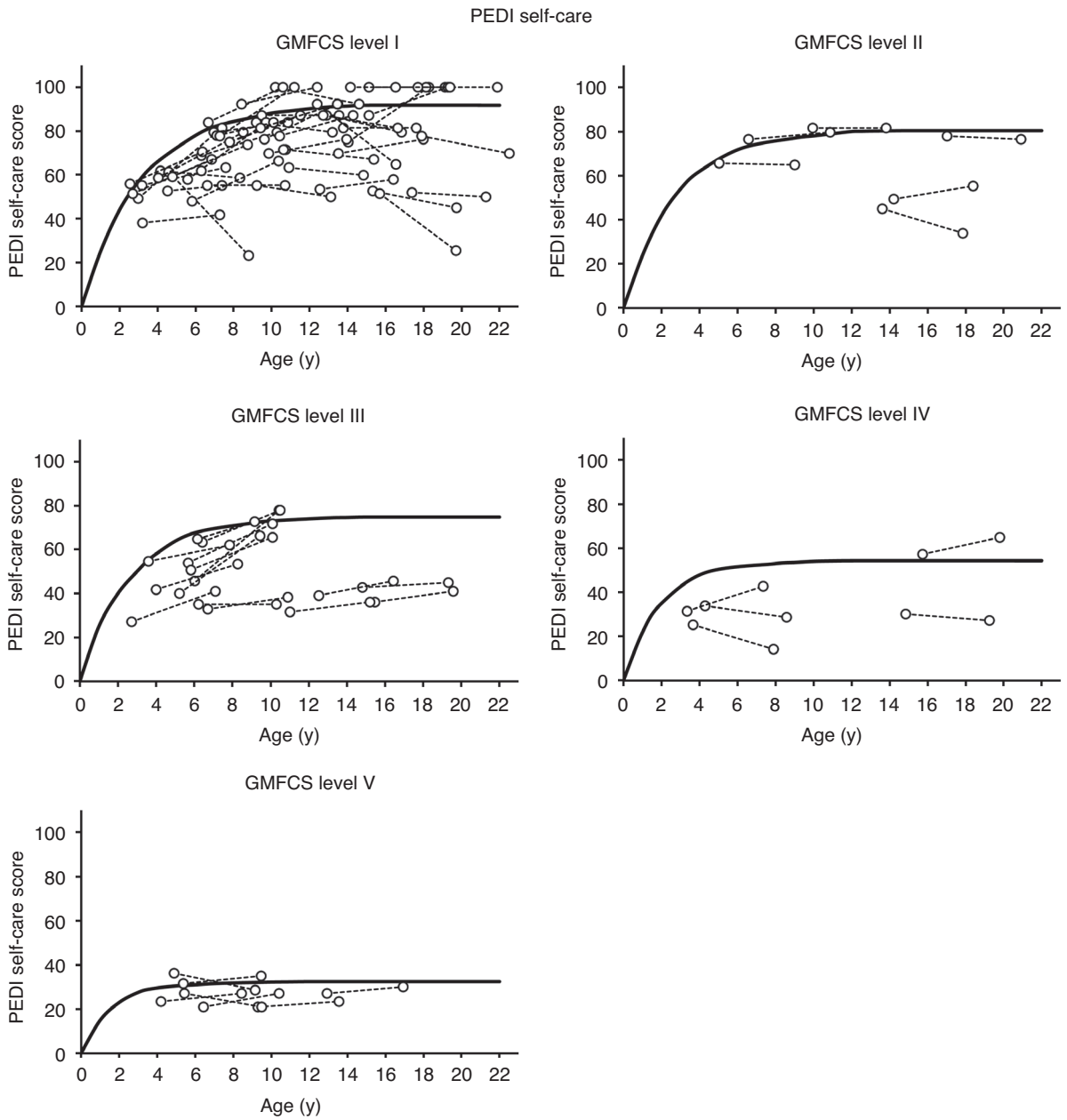

Figure 3: Scatter plots showing the Pediatric Evaluation of Disability Inventory, Ugandan version (PEDI-UG) self-care scores of 80 children and young people with cerebral palsy in Uganda (empty circles) in 2015 and 2019. The paired values for each participant are connected with a dashed line. Plots are shown for each Gross Motor Function Classification System (GMFCS) level at the first assessment. The solid line illustrates a modelled PEDI mobility trajectory developed from a large sample of children with cerebral palsy from Canada and the Netherlands.[3]

early intervention may fail to be identified by health services.

Our results suggest that the largest deviation from HIC developmental trajectories of all assessed functions occurred in the youngest children (2–5y) and that the mechanisms underlying this deviation changed with age. While there was no or little development in younger children who lagged behind predicted trajectories, gross motor development deteriorated in children and young people aged 12 to 18 years. This might seem paradoxical but likely reflects the steep predicted development during the youngest years, which plateaus as children age. Recent studies from HIC indicate a similar decline of gross motor function in young people and young adults with CP in GMFCS levels III to V.[23] Since we used reference values for 12-year-old children for this older group, this group may not have deviated from the developmental patterns of young people from HIC. Nevertheless, the finding that young children lagged the most behind predicted development suggests that this age group would benefit most from interventions.

More than one-third of children and young people in our study exhibited changes in GMFCS, MACS, and CFCS classification levels over the 4-years. This contrasts with the results of studies from HIC, which reported stability of MACS[24,25] and GMFCS[25,26] over time, with most children remaining at the same level. This stability has been a critical attribute of the classification systems, enabling health professionals and families to predict functional development, plan interventions, and allow stakeholders to plan community needs.[24,26] The instability in our Ugandan cohort suggests that classification systems developed in HIC cannot be used to predict development in LMIC unless the systems are adapted to the specific population and its developmental trajectories.

When speculating about the causes underlying the lower score attainment and slower development in our cohort, findings from this study may provide some of the best evidence regarding the effectiveness of comprehensive rehabilitation and medical interventions for young children with CP. Although the lower scores and slower development may reflect biological and pathological differences between children with CP in sub-Saharan Africa and HIC – as partly supported by different patterns of aetiology described earlier for this cohort with a high proportion of children with postneonatal CP and a small proportion of children being born preterm[5] – these differences are unlikely to be major factors because we used international diagnostic and classification systems and compared children of the same age and with the same functional impairment level. A more plausible reason is that the children have different opportunities for health interventions and are living under different socioeconomic conditions. In HIC, from an early age children have access to medical treatments and interventions aiming to stimulate development and improve motor and self-care skills.[2] Conversely, children with CP in LMIC have limited access to health and rehabilitation services and are vulnerable to medical conditions that may restrict their development, including malnutrition, malaria, and untreated epileptic seizures.[10,18]

Children and young people in our cohort had very poor access to adequate medical treatment, rehabilitation services, assistive devices, and education.[10] Unfortunately, this situation is repeated in many low-resource settings with shortages of health care and rehabilitation services for children with disabilities.[27,28] Additionally, the families of children with disabilities in Africa face many barriers when attempting to access care. These include poverty, stigma and negative attitudes, lack of knowledge about the disability, and poor availability of qualified health care professionals and health care services.[10,29,30]

Study strengths and limitations

To our knowledge, this is the first longitudinal study from a low-income country describing functional development of a population-based cohort of children and young people with CP. The use of international diagnostic, classification, and assessment methods enabled comparisons with data from other countries; these comparisons provided novel results and demonstrated significant differences. Since there is no validated method for comparing functional development in CP between populations, we utilized trajectories originally created to describe the development of children and young people in HIC. Another strength of this study was our use of the same therapist team at both assessments, which provided high reliability.

The relatively small number of children limited our ability to analyse data for different functional levels and age groups. Since we performed multiple tests, this will influence the statistical significance and increase the possibility that p-values were lower than the nominal 5% just by chance. To avoid type I errors, we performed Bonferroni corrections and have indicated the adjusted alpha levels in the table legends; this could increase the risk of type II errors, although only few of the comparisons lost significance. Since this study was performed in a mainly rural setting in Uganda, our results may not be transferrable to more urban settings in Uganda or other LMIC.

CONCLUSION

This study suggests that children and young people with CP in Uganda do not reach their full developmental potential. Their poorer functional development is likely at least partly attributed to a lack of services providing medical treatment and other early interventions. Relatively slower development was most apparent in younger children with mild impairments, suggesting that these children would probably benefit most from interventions. This is important since children with milder impairments are most of the children with CP in sub-Saharan Africa according to population-based studies; clinical cohort studies suggest they are not seeking health care. Our results underscore the urgent need for universal coverage and access to early screening and intervention services for children and young people with developmental delay and disability in LMIC to promote achievement of full functional potential.

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DATA AVAILABILITY STATEMENT

The data that underlie the results reported in this article are described at the Swedish National Data Service. Data are made available upon request after ensuring compliance with relevant legislation. DOI: https://doi.org/10.5878/txww-hg72.

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