



Research article

Factors for changes in self-care and mobility capabilities in young children with cerebral palsy involved in regular outpatient rehabilitation care



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HIGHLIGHTS

- Capabilities of children with CP improve modestly over time in outpatient rehabilitation.
- Children with more severe CP reach maximum mobility and self-care levels at an earlier age.
- After this maintaining capabilities is more realistic than improvement.
- Important prognostic factors are GMFCS level and intellectual impairment.
- Routine monitoring can aid goal setting and expectation management in communication with families.

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ABSTRACT

Background: Assessing prognosis of self-care and mobility capabilities in children with cerebral palsy (CP) is important for goal setting, treatment guidance and meaningful professional-caregiver conversations.

Aims: Identifying factors associated with changes in self-care and mobility capabilities in regular outpatient multidisciplinary paediatric CP rehabilitation care.

Methods and procedures: Routinely monitored longitudinal data, assessed with the Paediatric Evaluation of Disability Inventory (PEDI-Functional-Skills-Scale, FSS 0–100) was retrospectively analysed. We determined contributions of age, gross-motor function, bimanual-arm function, intellectual function, education type, epilepsy, visual function, and psychiatric comorbidity to self-care and mobility capability changes (linear-mixed-models).

Outcomes and results: For 90 children (53 boys), in all Gross-Motor-Function-Classification-System (GMFCS) levels, 272 PEDI's were completed. Mean PEDI–FSS-scores at first measurement (median age: 3,2 years) for self-care and mobility were 46.3 and 42.4, and mean final FSS-scores respectively were 55.1 and 53.1 (median age: 6,5 years). Self-care capability change was significantly associated with age (2.81, $p < 0.001$), GMFCS levels III–V (-9.12 to -46.66, $p < 0.01$), and intellectual impairment (-6.39, $p < 0.01$). Mobility capability change was significantly associated with age (3.25, $p < 0.001$) and GMFCS levels II–V (-6.58 to -47.12, $p < 0.01$).

Conclusions and implications: Most important prognostic factor for self-care and mobility capabilities is GMFCS level, plus intellectual impairment for self-care. Maximum capability levels are reached at different ages, which is important for individual goal setting and managing expectations.

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1. Introduction

Cerebral palsy (CP) is a common physical disability in children (Hutton, 2006; Meihuizen-de Regt et al., 2015). It is a permanent neurological disorder caused by non-progressive brain injury or malformation in the developing brain (Meihuizen-de Regt et al., 2015; Rosenbaum et al., 2007). CP is characterized by large heterogeneity and primarily affects posture and movement, but sensory, perceptive, cognitive, communicative and/or behavioural disturbances or epilepsy also frequently occur (Meihuizen-de Regt et al., 2015; Rosenbaum et al., 2007). Children with CP experience limitations when performing daily activities from a very young age, for which their families often seek help in rehabilitation settings.

In rehabilitation care, the primary interest is obtaining the highest possible level of participation in daily life activities. To achieve this, multidisciplinary teams, led by rehabilitation physicians, are engaged in increasing or maintaining children's independence in performing daily life activities. Because functional skills are an important prerequisite for daily task performance and participation, self-care and mobility capabilities are considered key constructs in the development of young children with CP (Chiarello et al., 2011; Haley et al., 2010).

Several papers have been published that describe courses over time of outcomes relevant to CP throughout childhood, adolescence and beyond (Hanna et al., 2009; Smits et al., 2013; Tan et al., 2014; van Gorp, van Wely et al., 2019; Vos et al., 2013; Vos et al., 2014). Such knowledge is essential for making prognoses, planning interventions, monitoring progress, and guideline development. Recently, severity-grouped developmental trajectories for mobility and self-care capabilities as measured with the Paediatric Evaluation of Disability Inventory (PEDI) became available from a large international cohort (Smits et al., 2019). In the younger CP age group, the PEDI is commonly used for discriminative and evaluative purposes. In Dutch CP rehabilitation care the PEDI is often routinely measured (Custers et al., 2002; Haley et al., 1992; Wassenberg-Severijnen and Custers, 2005; Wassenberg-Severijnen et al., 2003).

In addition to knowledge of mobility and self-care capability courses for different CP-severity levels over time, it is important to control for factors that may influence capability courses (Smits et al., 2019). Identification of prognostic factors for changes in self-care and mobility capabilities in children with CP therefore is crucial to guide treatment. Several potentially important factors have been described in literature: age (Ketelaar et al., 2014), gross motor function level (Ketelaar et al., 2014; Palisano et al., 2000; Rosenbaum et al., 2002; Smits et al., 2011), bimanual arm function (Smits et al., 2011), selective motor control (Smits et al., 2011), visual function (Fedrizzi et al., 2000; Wu et al., 2004), and intellectual capacity (Beckung and Hagberg, 2002; Ostensjo et al., 2003; Palisano et al., 2000; Smits et al., 2011; Voorman et al., 2006). By analysing routinely monitored PEDI data from a heterogeneous group of young children with CP receiving regular prolonged multidisciplinary outpatient rehabilitation care, we aimed to identify factors associated with changes in self-care and mobility capabilities. We hypothesised that other factors than CP-severity motor function classifications would also be related to changes in capabilities.

2. Methods

2.1. Population and procedure

Longitudinal analyses were performed on retrospective health care data that was routinely collected for quality purposes in the paediatric outpatient setting of our rehabilitation centre (i.e. convenience cohort, not in a research setting). All children received regular multidisciplinary treatment over multiple years, involving rehabilitation physicians, psychologists, physiotherapists, occupational therapists, speech therapists and social workers. Treatment for very young children generally comprised two to three visits to 'therapeutic toddler groups' weekly. Children from 4-5 years of age generally received two treatment hours

weekly, either at the special school for disabled children linked to our centre or at the outpatient clinic in case of regular school type. Interventions were always specific to the severity of CP, age-appropriate, personalized, goal directed and 'state-of-the-art', according to best available evidence guidelines (Dutch Knowledge Institute of Medical Specialists, 2015).

We included children aged between six months and 12 years at their first available PEDI-NL assessment. Data was obtained from digital and hand-written patient records of CP-diagnosed children; codes G80-G83, International Classification of Diseases (ICD-10) (World Health Organization, 2016). Available records were searched for PEDI-NL scores (Dutch PEDI version): only records between April 2006 and January 2017 with at least two PEDI-NL scores were further screened to find data regarding potentially associated factors. Following standard procedures, parents/primary caregivers signed agreements at the start of rehabilitation, including approval to use anonymized data for research purposes. Children without signed agreement were excluded from analyses. The Medical Ethics Committee of Erasmus University Medical Centre Rotterdam approved the study (MEC-2017-016).

2.2. Outcome measures and factors

Primary outcomes were self-care and mobility capabilities as assessed with the Dutch version of the PEDI (PEDI-NL), Functional Skills Scale (FSS). The PEDI-NL is a standardized semi-structured interview assessment for systematic identification of the ability of daily functioning skills of developing children in the 0,5–7,5 year age range (Wassenberg-Severijnen and Custers, 2005; Wassenberg-Severijnen et al., 2003). For 73 self-care and 59 mobility skills, FSS capability scores ranging from 0-100 were created using a dichotomous scoring system per skill (Haley et al., 1992; Wassenberg-Severijnen and Custers, 2005). The PEDI-NL has a good validity, test-retest reliability and inter-rater reliability (Custers et al., 2002; Wassenberg-Severijnen and Custers, 2005). Interviews were conducted by a selected group of certified occupational- and physiotherapists. Adhering to our quality registration system, we aimed at annual PEDI-NL interviews.

Besides potentially important factors already known from literature, three additional factors were added by clinical experts: 'epilepsy', 'type of education' and 'psychiatric comorbidity'. All factors were available from routinely collected data, except for 'selective motor control'. Thus, eight factors were studied:

- 'age' (in years and months) on each PEDI-NL measurement date;
- 'gross motor function level' as determined by the valid Gross Motor Function Classification System (GMFCS, valid Dutch version) (Gorter, van Tol, van Schie and Ketelaar, 2009), a five-level gross motor activity classification system, based on functional limitations, the need for assistive devices and quality of movement. GMFCS level I represents the highest (i.e. best) motor function level, level V the lowest (Gorter, Ketelaar, Rosenbaum, Helders and Palisano, 2009; Gorter, van Tol et al., 2009)
- 'bimanual arm function level' was scored using the Manual Ability Classification System (MACS, valid Dutch version) (van Meeteren et al., 2010), a five-level classification system for upper extremity use during daily activities, based on spontaneously undertaken ability to handle objects and the need for assistance or adaptations to perform bimanual skills in everyday life. MACS level I represents the highest level of bimanual performance, level V the lowest (Eliasson et al., 2006; van Meeteren et al., 2010);
- 'visual functioning' was evaluated as presence or absence of Cerebral Visual Impairment (CVI), diagnosed before or during the treatment period. CVI is a functional flaw in the retrochiasmatic part of the visual system (Dutton, 2013), including limited visual functioning not fully explained by improper eye functioning (i.e. eye movements, visual acuity and visual field) (Luyten, 2013);

- 'intellectual capacity' was evaluated as presence or absence of intellectual impairment, with impairment defined as intelligence quotient scores below 80 (IQ < 80 confirmed by official IQ test before the start or during treatment) (Wechsler, 2003);
- 'epilepsy' was evaluated as presence or absence of a confirmed diagnosis of epilepsy before the start or during treatment, with epilepsy defined as a neurological disorder marked by an increased susceptibility to recurrent unprovoked seizures (Stafstrom and Carmant, 2015);
- 'type of education' was distinguished as attending regular education or attending special education aged four years (please note this is the typical age at which Dutch children start primary school). Children who changed education type during treatment were assigned to the special education group;
- 'psychiatric comorbidity' was evaluated as presence or absence of secondary psychiatric disorders (i.e. ADHD, PDD-NOS and Autism/autistic behaviour) as diagnosed before the start or during treatment.

When possible, data on factors unavailable from treatment records were provided by clinicians to maximize statistical power. We chose to use their expertise instead of multiple imputation techniques to obtain most accurate estimates.

2.3. Statistical analyses

Several start-of-treatment characteristics of children with at least two PEDI-NL FSS-scores were first described. Subsequently, differences in age, gender, CP subtype and GMFCS level between the included and excluded children were tested.

After assumptions were checked, we used repeated-measures analysis using Linear Mixed Models (LMM) to generate models to identify factors for changes in self-care and mobility capabilities as determined by PEDI-NL FSS-scores. Full multivariate models with random intercepts, random slopes and a first-order autoregressive structure were chosen after comparing the robustness of different models using information criterion measures (AIC, BIC). These models allow adjustment for dependency of repeated measurements within the same child. To explore contributions of each factor together and to prevent overfitting, all factors were placed simultaneously. Multicollinearity was checked and if present, only one of the factors was retained in the model. The influence of possible outliers was identified by Cook's distance (Cook's D) and appropriate actions were taken when necessary (Cook and Weisberg, 1983; Field et al., 2012).

Because not all PEDI-NL measurements were performed exactly at one year intervals, all assessment dates were first rounded off in half years ($t = 0; 0.5; 1; 1.5$, up to 7.5) to create LMM time points. As this did not yield a balanced normal distribution, it was decided to round off all PEDI-NL assessment dates in whole years ($t = 0; 1; 2; 3$; up to 7) which resulted in a better fit. It therefore has to be noted that larger deviations from actual assessment dates may affect estimate precision of the factor 'age'. All statistical analyses were performed using R (version 3.2.4: R Foundation for Statistical Computing, Vienna, Austria).

3. Results

From a total of 416 children with signed parental agreements including research approval, there were 90 children with at least two PEDI interview measurements available (22%), resulting in a total of 272 PEDI-NL FSS scores. Thorough search of patient records, came to fourteen missing factor values for thirteen children ($14/720 = 2\%$); twelve missing for MACS levels, one missing for GMFCS level and for one child unclear intellectual capacity. These could be clarified by treating clinicians; the left column of Table 1 shows characteristics of our convenience cohort of 90 children. For comparison, some characteristics of the excluded group ($n = 326$) are also shown (right column, Table 1).

Table 1. Characteristics of the convenience cohort and excluded group.

		Convenience cohort (n = 90)	Excluded group (n = 326)	p-value
Median age at start rehabilitation*, (IQR, range)	Years;months	2;11 (2, 1–10)	5;3 (5, 0–16)	<0.001
Median age at first PEDI-NL measurement*, (IQR, range)	Years;months	3;2 (2, 1–11)		
Gender, n (%)	Male	53 (59)	196 (60)	1.00
	Female	37 (41)	130 (40)	
Subtype, n (%)	Spastic	67 (74)	290 (89)	<0.001
	Unilateral	14 (15)	153 (47)	
	Bilateral	53 (59)	137 (42)	
	Dyskinetic	18 (20)	26 (8)	
	Ataxic	5 (6)	10 (3)	
GMFCS, n (%)	Level I	15 (17)	173 (53)	<0.001
	Level II	22 (24)	72 (22)	
	Level III	18 (20)	16 (5)	
	Level IV	31 (35)	36 (11)	
	Level V	4 (4)	29 (9)	
MACS, n (%)	Level I	17 (19)		
	Level II	29 (32)		
	Level III	33 (37)		
	Level IV	7 (8)		
	Level V	4 (4)		
Intellectual impairment, n (%)	Yes	56 (62)		
	No	34 (38)		
Type of education, n (%)	Special	69 (77)		
	Regular	21 (23)		
Psychiatric comorbidity, n (%)	Yes	15 (17)		
	No	75 (83)		
Epilepsy, n (%)	Yes	25 (28)		
	No	65 (72)		
Visual impairment, n (%)	Yes	30 (33)		
	No	60 (67)		

IQR = interquartile range; GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System; Intellectual impairment = IQ < 80; Psychiatric comorbidity = the coexistence of a psychiatric diagnosis; Visual impairment = the presence of Cerebral Visual Impairment (CVI); p -value = $p < 0.05$ is statistically significant.

* Calendar age. These data are not a description of the developmental age of the children.

In our cohort, the PEDI-NL was, on average, completed three times per child during their rehabilitation periods (range 2–7); for 48% of the children three or more PEDI-NL FSS-scores were available. The mean of all first PEDI-NL FSS-scores for self-care was 46.3 (range 0–72.6) and for mobility this was 42.4 (range 13.3–87.6) at a median age of three years and two months (Figure 1a, and Table 1). At the final measurement points, the mean PEDI-NL FSS-score for self-care was 55.1 (range 2–83) and for mobility 53.1 (range 10.4–100), at a median age of 6 years and 5 months. In Figure 1, the course over time of mean self-care and mobility PEDI-NL FSS-scores of all 90 children is displayed (Figure 1a), as well as different courses over time for PEDI-NL FSS self-care and mobility scores per subgroup of children according to their GMFCS/MACS levels (Figure 1b-d).

For multivariable repeated-measures LMM analyses, one factor had to be removed from the full model due to collinearity between GMFCS and MACS levels. GMFCS level commonly is considered most relevant in discussions related to management of spastic CP (Love et al., 2010). However, since it can be argued that bimanual ability is conceptually

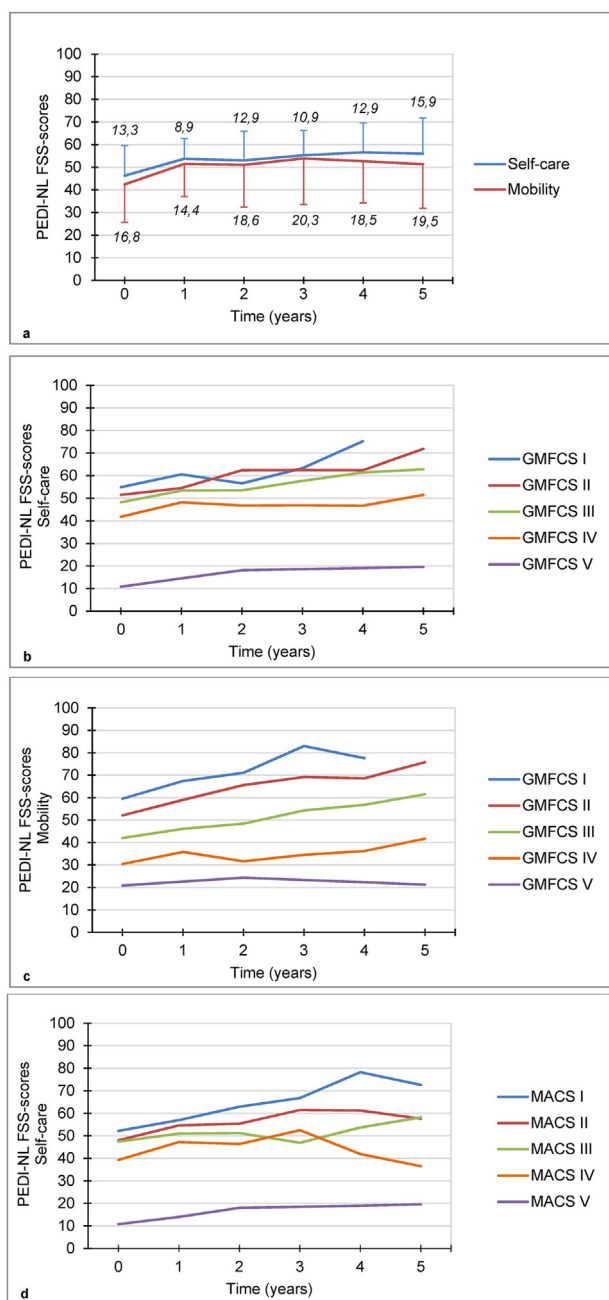


Figure 1. Course of mean PEDI-NL FSS-scores. Mean PEDI-NL FSS (Paediatric Evaluation of Disability Inventory, Functional Skills Scale) scores for each yearly taken measurement for all 90 children (including standard deviations for each time-point, for self-care upper part of bars are shown, for mobility lower part of bars are shown) (1a) per GMFCS level (Gross Motor Function Classification System), for the domains of self-care (1b) and mobility (1c), and per MACS level (Manual Ability Classification System) for self-care (1d). * Interpretation: time point 0 represents the mean PEDI-NL FSS-scores of the entire cohort at the first measurement (i.e. first PEDI assessment in the rehabilitation centre), time point 1 represents the mean PEDI-NL FSS-scores 1 year after the first measurement, et cetera. Please note that Time (years) on the X-axis does not represent age of the children. There is no data available from the GMFCS I group at time point 5. Time points 6 and 7 are not shown in the figures, due to insufficient measurements at these time points.

more related to self-care capabilities than gross motor function, and bimanual ability is generally considered as an important determinant of self-care performance (Palisano et al., 2020), we decided to create two separate models for changes in self-care capabilities. Assumptions for

LMM analyses were met and there were no outliers. Full models for self-care and mobility capabilities showed that at the time of the first PEDI-NL measurement the mean FSS-score of children following regular education with GMFCS/MACS level I without intellectual impairment, epilepsy, psychiatric comorbidity or CVI (representing the least affected CP children as a reference category) was 50.1/48.7 for self-care and 50.9 for mobility (see constant beta's, upper row Tables 2 and 3).

At the group level, changes in self-care and mobility capabilities were significantly associated with the factor 'age' ($p < 0.001$). Per year, the PEDI-NL FSS-score increased with a mean of 2.8/2.9 points for self-care and 3.3 points for mobility capabilities. With one exception for self-care (GMFCS/MACS II), the changes in self-care and mobility capabilities were also significantly associated with factors 'GMFCS level' or 'MACS level'. The higher GMFCS or MACS level (i.e. the worse the gross motor function or bimanual arm function was), the lower mean PEDI-NL FSS-scores (see Tables 2 and 3). Changes in self-care capabilities were significantly associated with the factor 'intellectual capacity' ($p < 0.01$ and $p = 0.02$ in the GMFCS and MACS as factor models, respectively): presence of intellectual impairment reduced the PEDI-NL FSS-score for self-care with 6.4/5.1 points. For the FSS mobility model, the reduction related to intellectual impairment of 3.4 points was not significant ($p = 0.09$). Changes in self-care capabilities were clearly negatively, but not significantly, associated with the factor 'psychiatric comorbidity' ($p = 0.06$ in the GMFCS as factor model), whereas the factor 'type of education' was positively but also not significantly associated with changes in mobility capabilities ($p = 0.08$).

Table 2. Associations between changes in self-care and mobility capabilities and the independent factors (multivariable full model[#]).

	PEDI-NL FSS Self-care			PEDI-NL FSS Mobility		
	Beta	SE	p-value	Beta	SE	p-value
Constant	50.10	2.57	<0.001	50.94	2.66	<0.001
Age	2.81	0.29	<0.001	3.25	0.32	<0.001
GMFCS						
Level I	0 (ref)			0 (ref)		
Level II	-1.52	2.54	0.55	-6.58	2.55	0.01
Level III	-9.12	2.82	<0.01	-21.25	2.83	<0.001
Level IV	-14.49	2.71	<0.001	-34.60	2.71	<0.001
Level V	-46.66	4.58	<0.001	-47.32	4.61	<0.001
Intellectual impairment						
No	0 (ref)			0 (ref)		
Yes	-6.39	2.00	<0.01	-3.37	1.98	0.09
Type of education						
Regular	0 (ref)			0 (ref)		
Special	1.93	2.36	0.42	4.16	2.37	0.08
Psychiatric comorbidity						
No	0 (ref)			0 (ref)		
Yes	-4.71	2.43	0.06	-1.14	2.42	0.64
Epilepsy						
No	0 (ref)			0 (ref)		
Yes	-1.75	1.90	0.36	-0.69	1.89	0.72
Visual impairment						
No	0 (ref)			0 (ref)		
Yes	0.03	1.79	0.99	0.02	1.76	0.99

PEDI-NL FSS = Paediatric Evaluation of Disability Inventory, Functional Skills Scale; Beta = regression coefficient; SE = standard error; p-value = $p < 0.05$ is statistically significant; Age = in years; GMFCS = Gross Motor Function Classification System; Intellectual impairment = IQ < 80; Psychiatric comorbidity = the coexistence of a psychiatric diagnosis; Visual impairment = the presence of Cerebral Visual Impairment (CVI).

[#] To prevent effect modification no plausible interaction terms were included in the models.

Table 3. Associations between changes in self-care capabilities and the independent factors (multivariable full model[#]) – MACS instead of GMFCS.

	PEDI-NL FSS Self-care		
	Beta	SE	p-value
Constant	48.68	3.00	<0.001
Age	2.85	0.29	<0.001
MACS			
Level I	0 (ref)		
Level II	-2.55	2.77	0.36
Level III	-6.77	2.90	0.02
Level IV	-13.27	4.11	<0.01
Level V	-41.45	5.20	<0.001
Intellectual impairment			
No	0 (ref)		
Yes	-5.13	2.21	0.02
Type of education			
Regular	0 (ref)		
Special	-1.82	2.57	0.48
Psychiatric comorbidity			
No	0 (ref)		
Yes	-1.08	2.69	0.69
Epilepsy			
No	0 (ref)		
Yes	-1.08	2.22	0.63
Visual impairment			
No	0 (ref)		
Yes	-1.78	2.08	0.39

PEDI-NL FSS = Paediatric Evaluation of Disability Inventory, Functional Skills Scale; Beta = regression coefficient; SE = standard error; p-value = $p < 0.05$ is statistically significant; Age = in years; MACS = Manual Ability Classification System; Intellectual impairment = IQ < 80; Psychiatric comorbidity = the coexistence of a psychiatric diagnosis; Visual impairment = the presence of Cerebral Visual Impairment (CVI).

[#] To prevent effect modification no plausible interaction terms were included in the models.

4. Discussion

By analysing routinely collected PEDI-NL data, we aimed at identifying factors associated with changes in self-care and mobility capabilities in young children with CP involved in prolonged multidisciplinary outpatient rehabilitation care. At the group level, self-care and mobility capabilities improved modestly over time with large variation at each time point confirming the heterogeneity known to CP. The ambulatory subgroup (GMFCS levels I-III) showed gradually improved capabilities over time, whereas children with more severe CP (GMFCS levels IV-V) showed marginal changes yearly. For the latter, maximum self-care and mobility capabilities levels were reached at an earlier age. After limits are reached in more severe CP, maintaining capabilities over time thus appears more realistic than improvement.

The three multivariable models showed that changes in self-care capabilities were significantly associated with older age, better gross motor function/bimanual ability and higher intellectual capacity, and that changes in mobility capabilities were significantly associated with older age and better gross motor function. The hypothesis that changes in capabilities over time would differ across GMFCS and MACS CP-severity levels was confirmed. Besides, intellectual impairment also appeared an important prognostic factor, especially regarding development of self-care capabilities. This finding is relevant in communication with families because, together with worse motor function levels, intellectual disability has been shown to limit participation in domestic life and/or interpersonal relationships later in life (van Gorp, Roebroek, et al., 2019).

Our findings were largely in line with previous studies on (changes in) mobility and self-care capabilities in CP (Beckung and Hagberg, 2002; Ketelaar et al., 2014; Morris et al., 2006; Ohrvall et al., 2010; Ostensjo et al., 2003; Palisano et al., 2020; Phipps and Roberts, 2012; Smits et al., 2019), despite differences regarding the age range studied, composition of cohorts, instruments used to assess self-care and mobility capabilities, intensity and content of interventions received, treatment settings, and/or statistical methods applied. Similar to mobility and self-care trajectories described by Smits and colleagues (Smits et al., 2019), we found distinct courses for different GMFCS and MACS levels, with lower maximum capability limits that were reached sooner in more severe CP. There also was extensive variation regarding courses over time at the (sub)group level(s) in both self-care and mobility capabilities, which is inherent to CP (Damiano, 2014; Shevell, 2018). The relatively smaller variation at the 1-year time point (Figure 1a) may reflect the rapid 'pre-plateau' development of mobility and self-care capabilities in early childhood as described recently (Smits et al., 2019); the steepest parts of Smits' developmental trajectories, before performance limits are reached, generally occur before the age of three. Divergence of self-care and mobility capability courses for GMFCS and MACS levels IV and V (Figure 1b-d) may be explained by some kind of lower capability limit required for performing self-care activities in comparison to mobility activities.

Except for visual function, all factors identified from scientific literature were significantly associated with changes in self-care and mobility capabilities, whereas none of the additional factors brought in by experts appeared significant. The absence of a significant associations for the factor 'visual function' may be related to a higher incidence of visual impairments in children with more severe CP (Hou et al., 2010; Jeon et al., 2019): post-hoc analysis indeed showed a significantly higher occurrence of CVI in GMFCS levels IV-V compared to levels I-III ($p < 0.05$). The finding that children at special schools (where therapy is generally integrated during school hours) showed more favourable changes in PEDI-NL mobility scores than children at regular schools might be related to higher treatment intensity (Storvold, Jahnsen, Evensen and Bratberg, 2018; Storvold, Jahnsen, Evensen, Romild and Bratberg, 2018). Overall, presence of one (or more) impairment(s) and/or comorbidity appears to be associated with less favourable changes in self-care and mobility capabilities, and severity of CP is a primary factor for self-care and mobility capability courses. Since limits are reached earlier in more severe CP, maintaining capabilities should be treatment goal rather than improving by the time maximum levels are reached.

We performed repeated-measures LMM analyses using both random intercepts and slopes to identify factors for changes in self-care and mobility capabilities in one cohort of young children with CP, whereas Smits and colleagues used non-linear mixed-effects modelling to estimate limits, rates, and variations of development in a composite cohort (Smits et al., 2019). We would like to point out these differences, because our graphs should consequently be interpreted differently as we put calendar age in the model as a factor for changes in capabilities over time. Regarding interpretation of findings, it should also be noted that in our cohort, all children received standard rehabilitation care and none were involved in clinical trials. In research settings, it is more likely that (part of) the study population received intensive/intensified treatment for some period. Analysis of routinely monitored data during prolonged outpatient rehabilitation care in one cohort from one rehabilitation centre provides complementary evidence about self-care and mobility capability courses in young children with CP. Such clinical practice reference data is valuable for decisions on services for children growing up with CP (van Trijffel et al., 2019).

The generic PEDI is a valid and reliable outcome measure, but also known to be limited by ceiling effects when assessing better-functioning children (Damiano et al., 2005), and its responsiveness has also been discussed (James et al., 2014; Vos-Vromans et al., 2005). Therefore, it is possible that children in our cohort actually showed more self-care

and/or mobility progress than reflected in their PEDI-NL FSS-scores. The fact that the PEDI-NL was scored less frequently in better functioning and in older children in our centre was probably related to the age limit for evaluative purposes (i.e. developmental ages 0,5–7,5 years). Assessing these children's capabilities is probably often only done for discriminative and diagnostic purposes (Haley et al., 2010).

Due to the nature of our study, there are some limitations that have to be discussed. An important limitation is that both GMFCS and MACS classification levels were not allowed to vary in the LMM analyses, whereas changes of levels over time are not uncommon in a proportion of the CP population (Gorter, Ketelaar, et al., 2009; Nylén and Grooten, 2021; Palisano et al., 2018). Another limitation is that for better functioning children, the time consuming PEDI-NL assessments were frequently omitted because of clinical judgements that 'functioning went well'. Similar clinical reasoning led to consciously omitting 'frustrating/confronting' interviews for GMFCS level IV-V children. Other reasons for absence of PEDI-NL FSS-scores were referral to other healthcare settings, missing annual appointments, too little time, and failing procedures to secure annual PEDI-NL interviews. It is therefore important to take potential selection bias into account when interpreting our findings. Since only 22% of the screened patient records had two or more PEDI-NL FSS-scores available, our convenience cohort likely is more severely affected than the general CP population. And it is probably also not representative for the CP population generally seen in rehabilitation care. Another limitation of our work is the rather small cohort size with negative effect on statistical power. Nevertheless, we found clear differences between different CP-severity levels, which adds to the existing evidence because we controlled for several factors potentially important for changes in self-care and mobility capabilities. For future studies, analysing factors related to personal or environmental context, natural development, motor selectivity, the content, intensity and effect sizes of interventions, and the interplay between these factors is critically important.

In summary, our models developed to identify factors for changes in self-care and mobility capabilities in young children with CP within a multidisciplinary outpatient rehabilitation setting, showed that self-care and mobility capabilities improve modestly over time at the group level. Most important prognostic factors for self-care and mobility capability development as measured with the PEDI-NL are motor function levels and intellectual impairment. Maximum capability limits are reached at different ages for different CP-severity levels: children with more severe CP reach their maximum levels of self-care and mobility capabilities at an earlier age, and after this maintaining capabilities is more realistic than improvement. Goal setting should be adapted accordingly. Analysis of routinely monitored patient data can support meaningful conversations with families about realistic goals and expectations and allows more informed decision making in the continuous clinical challenge to maximize independence of children with CP.

Declarations

Author contribution statement

Marleen J. de Leeuw: Conceived and designed the experiments; Performed the experiments; Analyzed and interpreted the data; Contributed reagents, materials, analysis tools or data; Wrote the paper.

Fabienne C. Schasfoort, Bea Spek, Tessa Westendorp and Robert F. Pangalila: Conceived and designed the experiments; Analyzed and interpreted the data; Wrote the paper.

Inez van der Ham and Stella Verschure: Conceived and designed the experiments; Contributed reagents, materials, analysis tools or data; Wrote the paper.

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Data availability statement

Data will be made available on request.

Declaration of interests statement

The authors declare no conflict of interest.

Additional information

No additional information is available for this paper.

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