Developmental Trajectories of Mobility and Self-Care Capabilities in Young Children with Cerebral Palsy

Marjolijn Ketelaar, PhD1,2, Jan Willem Gorter, MD, PhD2,3, Paul Westers, PhD4, Steven Hanna, PhD3,5, and Marjolein Verhoef, MD, PhD1,2

Objective To describe development of mobility and self-care capabilities in young children (aged 1-4 years) with cerebral palsy, and to examine whether the development of mobility and self-care capabilities differs by cerebral palsy severity in terms of 5 distinct Gross Motor Function Classification System (GMFCS) levels.

Study design This prospective longitudinal cohort study included 100 children with cerebral palsy (aged 1.5 or 2.5 years at baseline) and their parents. Mobility and self-care capabilities were assessed by the Pediatric Evaluation of Disability Inventory during yearly assessments from inclusion up to age 4.5 years. Longitudinal data for 92 children were available for analysis. Repeated-measures analyses with random coefficient analysis were performed using linear mixed models.

Results Despite large variations among individuals in the development of mobility and self-care capabilities in young children with cerebral palsy, distinct developmental trajectories were found for children in different GMFCS levels. The estimated change per month differed significantly by GMFCS level for both outcomes.

Conclusions This longitudinal study provides an evidence base for prognosis in daily mobility and self-care skills in young children with cerebral palsy. The developmental trajectories for GMFCS levels can be helpful in communication between professionals and also in discussions of expectations and goal setting with families regarding mobility and self-care in the daily life of young children with cerebral palsy in neonatal follow-up and pediatric practice. (J Pediatr 2014;165:436-443).

With a prevalence varying from 2.0 to 3.6 cases per 1000 live births,1-3 cerebral palsy is the most common physical disability of childhood. Cerebral palsy is not a single, homogeneous health condition or disorder, but rather encompasses a wide variety of permanent disorders of development of movement and posture, attributed to nonprogressive disturbances occurring in the developing fetal or infant brain.4 The motor disorder is often accompanied by disturbances in sensation, perception, cognition, communication, and behavior; epilepsy; and secondary musculoskeletal problems.4,5 Thus, cerebral palsy can affect the development of children's everyday function, with a wide range of effects. In neonatal follow-up clinic or pediatric practice, parents often ask what their child’s future will hold, including the prognosis for daily activities, such as walking and self-care.7

The Gross Motor Function Classification System (GMFCS) has become the international standard for describing subgroups of children with cerebral palsy across the severity spectrum.6,8 It provides a 5-level classification of severity, ranging from walking without limitations (level I) to being transported in a wheelchair (level V).7 Classification with the GMFCS after age 2 years is reliable and valid and can be used to predict later gross motor functioning.9,10 The GMFCS facilitates communication between health professionals and families about developmental implications of cerebral palsy.6,11

Five distinct motor development curves based on the GMFCS have been described, identifying clinically relevant and statistically significant differences in the rates and limits of gross motor development in children and adolescents with cerebral palsy.12,13 In these curves, the Gross Motor Function Measure, 66-item version14 served as the outcome measure, used to assess gross motor capacity in a standardized environment (ie, therapy setting). A child’s capacity reflects what he or she can do in a standardized environment, and capability represents what he or she can do at home, at school, and in the community.15 For prognosis, it is important to create capability trajectories for mobility and self-care to enhance the Gross Motor Function Measure, 66-item version motor curves.

Little is known about the development of mobility and self-care capabilities of children with cerebral palsy.16,17 In both studies, the GMFCS was found to be a

<table>
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<tr>
<th>GMFCS</th>
<th>Gross Motor Function Classification System</th>
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<tr>
<td>PED</td>
<td>Pediatric Evaluation of Disability Inventory</td>
</tr>
<tr>
<td>PED-CAT</td>
<td>Pediatric Evaluation of Disability Inventory, computer adaptive testing</td>
</tr>
<tr>
<td>PED-FSS</td>
<td>Pediatric Evaluation of Disability Inventory, Functional Skills Scale</td>
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significant predictor of both mobility and self-care capabilities. To be able to inform parents and professionals early about a child’s prognosis and facilitate communication in neonatal follow-up and pediatric clinical practice, more knowledge is needed regarding early development of children with cerebral palsy. The present study examined the development of mobility and self-care capabilities in young children (aged 1-4 years) with cerebral palsy, and evaluated whether the development of mobility and self-care capabilities differs for distinct GMFCS levels.

### Methods

This study is part of the Pediatric Rehabilitation Research in the Netherlands Cerebral Palsy 0-5 study, a prospective longitudinal cohort study on the course and determinants of daily function in children aged 0 to 5 years with cerebral palsy. Data were collected at yearly assessments from inclusion at age 1.5 or 2.5 years up to age 4.5 years.

One hundred children and their parents were recruited at 5 university medical centers and 6 rehabilitation centers in the Netherlands. Parents were invited to participate in the study by the child’s pediatrician, child neurologist, or rehabilitation physician. Children aged 1 year, 6 months (±2 months, corrected age for prematurity if applicable) or 2 years, 6 months (±1 month) at study entry with a confirmed diagnosis of cerebral palsy were eligible. Children diagnosed with additional diseases and disorders affecting motor functioning and children whose parents lacked a basic knowledge of Dutch were excluded. All children participating in this study received regular care covered by the standard Dutch healthcare insurance, including rehabilitation services, such as physical and occupational therapy. The Medical Ethics Committee of University Medical Centre Utrecht and the Medical Ethics Committees of all participating centers approved the study, and all parents of participating children gave informed consent.

Mobility and self-care capabilities were assessed using the Pediatric Evaluation of Disability Inventory (PEDI), Functional Skills Scale (PEDI-FSS). The PEDI is a standardized instrument using parental reports in a structured interview. Trained research assistants administered the PEDI in a face-to-face interview with a parent (usually the mother).

The mobility and self-care domains of the PEDI-FSS consist of 65 and 74 items, respectively, with each item scored as positive (1) or negative (0). A score of 1 is given when the child is capable of accomplishing the activity. The mobility domain samples transfer and indoor and outdoor mobility skills. The self-care domain samples eating, grooming, dressing, bathing, and toileting skills. Mobility and self-care sum scores were transformed to interval-scaled scores (0-100), reflecting an increasing level of functioning.

Severity of cerebral palsy was classified with the GMFCS. The 5-level scale uses separate descriptions for different age groups and has shown to be stable over time. In this study, all participating children were classified at age 2.5 years using the GMFCS descriptions for age 2 up to age 4 (Table I; available at www.jpeds.com). For descriptive purposes, the child’s type of motor impairment (ie, spastic, dyskinetic, or ataxic cerebral palsy) and, within spastic cerebral palsy, limb distribution (ie, unilateral or bilateral) were classified according to the guidelines for the Surveillance of Cerebral Palsy in Europe.

Repeated-measures analysis with random coefficient analysis was performed using linear mixed models. This method considers the dependency of repeated measures within the same individual by allowing the regression coefficients for intercepts and slopes to differ among subjects. The number and timing of observations per person are free to vary. In effect, average trajectories are estimated while predicting how each child’s development differs from the average. First, the developmental trajectories of the outcome measures were modeled by age. The intercepts were treated as random effects to capture the variability in the data. Second, GMFCS was added to the model. The ratio likelihood test was used to compare models and to evaluate whether or not a random regression coefficient for age had to be considered in the models. Third, to test whether the development of mobility and self-care capabilities differed for distinct GMFCS levels, the interaction between age and GMFCS level was included. Finally, age was added to the model to test quadratic trajectories.

### Results

Out of 100 participants, 63 children were recruited at age 1 year, 6 months, and 37 were recruited at age 2 years, 6 months. Data from 8 children were excluded after baseline, including 3 children who were deemed ineligible because they did not fulfill the criteria for the diagnosis of cerebral palsy and 5 children who declined further participation. Thus, data for 92 children (Table II) were available for analysis. PEDI data for 52 children (56%) were available at age 1.5 years, for 86 children (92%) at age 2.5 years, for 86 children (92%) at age 3.5 years, and for 72 children (77%) at age 4.5 years. The main reason for dropout and missing data was the burden of the assessment on the child or family.

For both PEDI-FSS domains, mean scores increased with increasing age. Children with the most severe cerebral palsy had the lowest PEDI-FSS scores. Individual children showed wide variations in the development of motor capability and self-care capability (Figures 1 and 2; available at www.jpeds.com).

Repeated-measures analysis for both domains revealed a linear increase in all GMFCS levels between 1.5 and 4.5 years of age, with different baseline scores for the GMFCS levels. No significant random effect for age was found for either mobility or self-care, indicating no evidence of heterogeneity in the rate of change. Including fixed effects for GMFCS and the interaction between age and GMFCS in the models significantly improved the model fit (P < .001 for both models), indicating that children in different GMFCS level develop...
differently over time. Including age did not improve model fit for both models.

Inspection of residual plots for the total group and for each GMFCS level supported the linear models. Residuals were spread around 0, and no large differences over age and predicted value were found. Figures 3 and 4 describe the final models.

Given the significant interaction between age and GMFCS, insight into the differences among GMFCS levels for the effect of age is important (ie, to compare the slopes of the trajectories). For PEDI-FSS mobility, no significant differences in the slopes of GMFCS I, II, and III were noted. The slopes of GMFCS I, II, and III each differed from the slopes of GMFCS IV and V to a statistically significant degree (P < .001 for all). The slope of GMFCS V was significantly less steep than that of GMFCS IV (P < .01). The estimated monthly increase in PEDI-FSS mobility scores for children at GMFCS levels I, II, and III is much larger than that for children at GMFCS levels IV and V (Table III).

For PEDI-FSS self-care, in general lower GMFCS levels (ie, children with better gross motor function) were associated with steeper slopes compared with higher GMFCS levels; however, the slopes for consecutive GMFCS-levels did not differ from one another. A statistically significant group interaction was found for GMFCS levels IV and V compared with levels I and II (all P < .01). The slopes of GMFCS level III differed from the slopes of GMFCS level I (steeper; P < .01) and level V (less steep; P < .01). Slopes for children at GMFCS level I increased more than twice as much on average as those for children at GMFCS level V (Table III).

**Discussion**

This longitudinal study of a relatively large cohort of young children with cerebral palsy has revealed distinct developmental trajectories for mobility and self-care for children at different GMFCS levels. Development of both mobility and self-care was faster in children with lower GMFCS levels (indicating better gross motor function). For mobility, differences in the rate of development were found between GMFCS level I-III and GMFCS level IV/V. Mobility as defined in the PEDI includes more than just walking ability, including, for example, mobility in and around the house, distances, and independence regardless of the means of mobility. Even with the use of mobility devices (eg, wheelchair), development in daily mobility was much slower in children at GMFCS level IV/V compared with children at GMFCS level I-III. The estimated rate of development of mobility capability did not differ among children at GMFCS levels I, II, and III, with an average estimated increase of approximately 0.8 point per month. This translates to an estimated change of approximately 10 points per year on the PEDI scale of 0-100. The estimated increase in mobility capability of children with GMFCS level IV and V was much lower, 0.40 and 0.14 point per month, respectively.

For self-care, differences between subgroups based on GMFCS level were found for all subgroup comparisons, except for consecutive levels. In general, the differences in estimated rate of development were smaller than those in the mobility domain. The average increase in self-care score was 0.74 point (0.66-0.82 point) per month, or roughly 9 points per year, in children at GMFCS level I and 0.31 (0.17-0.44) point per month (3.7 points per year) children at GMFCS level V.

Our results add to the understanding of patterns of development of daily activities of young children with cerebral
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that children be reclassified at age 2 years or older.\textsuperscript{10} There-

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level determined when the child was 2 years, 6

tion on the development of children with cerebral palsy

these data and developmental trajectories provide informa-

palsy, a diagnosis characterized by its heterogeneous presen-

and prognosis. It is important to realize, however, that

that these data and developmental trajectories provide information on the development of children with cerebral palsy only from age 1 year, 6 months up to age 4 years, 6 months.

Moreover, the analyses in the present study were based on GMFCS level determined when the child was 2 years, 6 months of age. GMFCS classification of children aged <2 years is known to be less precise, and thus it is recommended that children be reclassified at age 2 years or older.\textsuperscript{10} Therefore, our findings for different GMFCS levels are most useful for children aged ≥2 years. For younger children, the graphs can be best interpreted for GMFCS levels I-III combined versus GMFCS levels IV and V combined, with no specific reference to the estimated changes. A previous study found a 96% probability that young children initially classified at GMFCS level I, II, or III would be subsequently classified at the same level at age ≥2 years.\textsuperscript{10}

In interpreting the results of the present study, some limitations should be considered. First, sample sizes were relatively small in each GMFCS subgroup, especially GMFCS level II (n = 12) and level V (n = 9), which might have limited the fit of the models. However, inspection of residuals by GMFCS subgroup confirmed the final models for each subgroup, with residuals spread around 0 and no large differences over age and predicted value.

Second, our study group included only 2 children with the dyskinetic subtype of cerebral palsy, and thus no specific conclusions for this subtype can be drawn from our data. In most previous studies, the large majority of children had predominantly spastic cerebral palsy,\textsuperscript{12,13} reflecting the typical distribution found in population-based studies.\textsuperscript{23,24} Third, we lack detailed information on the frequency of services provided to the participants in this study. In general, in The Netherlands, children aged <4 years with cerebral palsy receive physical and occupational therapy 1-3 times per week. This is important with regard to the generalizability of the results to other countries and healthcare systems. Fourth, the trajectories are specific to the PEDI. A new version of the Pediatric Evaluation of Disability Inventory, computer adaptive testing (PEDI-CAT), was published recently.\textsuperscript{25} The PEDI-CAT differs from the PEDI in the scope of the constructs, age group (0-20 years), method of administration, and number of items. We foresee use of the PEDI and PEDI-CAT as complementary measures in the future, with the PEDI being very informative in young children with cerebral palsy because of the many relevant items for this group.

One of the most important questions for parents is whether their child will become independent in self-care and mobility activities.\textsuperscript{6} Independence relates to performance, what a child actually does in his or her daily environment.\textsuperscript{15} Capacities and capabilities of a child are important prerequisites for the child’s daily functioning, the actual performance of activities. In earlier studies, the GMFCS proved very helpful in understanding distinct gross motor development curves, that is, curves on capacity in gross motor functions such as sitting, standing, and walking as measured in a standardized environment, typically a therapy setting. In the present study, the user-friendly GMFCS also proved helpful in creating and understanding developmental trajectories for capabilities in real life activities, that is, activities in the child’s own daily environment, including indoor and outdoor mobility, various transfers, eating, dressing, and toileting. It is hoped that these trajectories of mobility and self-care activities can provide healthcare professionals with helpful information when discussing prognosis issues beyond basic gross motor functions with the family. Knowledge of the rate of development (Figures 3 and 4 and Table III) would be useful for characterizing the estimated changes that could be expected in the next year, as well as the variation among children within a particular level. For prognosis in individual children, it is important to realize that this information holds some uncertainties, as reflected in the wide CIs as well as the degree of individual variation in trajectories. Of note, we found that not all GMFCS levels have the same slope of development.

![Graph](image.png)

Figure 4. Predicted average development of PEDI-FSS self-care by GMFCS level.

Table III. Final model: Estimated changes in mobility and self-care capability per month of age by GMFCS level

<table>
<thead>
<tr>
<th>GMFCS level</th>
<th>Mobility capability</th>
<th>Self-care capability</th>
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<tbody>
<tr>
<td></td>
<td>Estimated change (SE)</td>
<td>95% CI</td>
</tr>
<tr>
<td>I</td>
<td>0.87 (0.05)</td>
<td>0.77 to 0.98</td>
</tr>
<tr>
<td>II</td>
<td>0.84 (0.07)</td>
<td>0.70 to 0.98</td>
</tr>
<tr>
<td>III</td>
<td>0.82 (0.06)</td>
<td>0.71 to 0.93</td>
</tr>
<tr>
<td>IV</td>
<td>0.40 (0.06)</td>
<td>0.28 to 0.51</td>
</tr>
<tr>
<td>V</td>
<td>0.14 (0.09)</td>
<td>-0.04 to 0.31</td>
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The information provided by this study can be used by physicians to discuss different scenarios with parents. For example, expectations for development in mobility and self-care are quite different for a 2-year-old child at GMFCS level III than for a 2-year-old child at level IV, although neither may be able to stand and walk.

For parents, it is important to feel supported in their search for information. Clinicians can now use evidence-based knowledge to communicate prognoses with families. The presented developmental trajectories may support realistic goal setting and treatment planning. There are many choices to be made in terms of therapy goals, supportive devices, and adaptations at home. Given the information in the PEDI manual on items and on the hierarchy of difficulty of the items, in combination with the expected changes for the individual child, realistic goals can be set in terms of daily activities. However, it must be kept in mind that the PEDI includes many, but not all, aspects of self-care and mobility skills, and thus might not reflect all of the goals and changes important for young children with cerebral palsy. Moreover, capabilities are important prerequisites for a child’s functioning, but actual performance is also affected by environmental factors, such as the amount of assistance provided by caregivers, and personal factors, such as the child’s level of motivation. Insight into the development of performance is important, but also challenging, because of the various factors that play a role in performance, especially in young children, in whom parents play a crucial role in providing opportunities to perform activities independently.

Finally, the development and use of outcome measures that assess functional daily activities reflect an important paradigm shift in recent childhood disability research and pediatric clinical practice. Researchers and clinicians are shifting from thinking in terms of neurologic status and motor milestones to thinking in terms of the consequences of cerebral palsy on daily functioning. This is what matters most to children with cerebral palsy and their parents.

We thank the children and their parents who participated in this study, as well as the research assistants who administered the measures. We also express our gratitude to the university medical centers and rehabilitation centers that participated.

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Figure 1. Development of mobility capability of individual children.

Figure 2. Development of self-care capability of individual children.
Table I.  GMFCS levels for children between the second and fourth birthdays

<table>
<thead>
<tr>
<th>Level</th>
<th>Description</th>
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<tbody>
<tr>
<td>Level I</td>
<td>Children floor sit with both hands free to manipulate objects. Movements in and out of floor sitting and standing are performed without adult assistance. Children walk as the preferred method of mobility without the need for any assistive mobility device.</td>
</tr>
<tr>
<td>Level II</td>
<td>Children floor sit but may have difficulty with balance when both hands are free to manipulate objects. Movements in and out of sitting are performed without adult assistance. Children pull to stand on a stable surface. Children crawl on hands and knees with a reciprocal pattern, cruise holding onto furniture and walk using an assistive mobility device as preferred methods of mobility.</td>
</tr>
<tr>
<td>Level III</td>
<td>Children maintain floor sitting often by “W-sitting” (sitting between flexed and internally rotated hips and knees) and may require adult assistance to assume sitting. Children creep on their stomach or crawl on hands and knees (often without reciprocal leg movements) as their primary methods of self-mobility. Children may pull to stand on a stable surface and cruise short distances. Children may walk short distances indoors using a hand-held mobility device (walker) and adult assistance for steering and turning.</td>
</tr>
<tr>
<td>Level IV</td>
<td>Children floor sit when placed, but are unable to maintain alignment and balance without use of their hands for support. Children frequently require adaptive equipment for sitting and standing. Self-mobility for short distances (within a room) is achieved through rolling, creeping on stomach, or crawling on hands and knees without reciprocal leg movement.</td>
</tr>
<tr>
<td>Level V</td>
<td>Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At level V, children have no means of independent movement and are transported. Some children achieve self-mobility using a powered wheelchair with extensive adaptations.</td>
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