PATIENT-CENTERED OUTCOMES RESEARCH INSTITUTE

FINAL RESEARCH REPORT

Understanding the Development of Children with Cerebral Palsy and How Therapy May Affect Patient-Centered Outcomes

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ABSTRACT

Background: Knowledge of developmental trajectories of body structure/function and participation in family/recreation and self-care activities in children with cerebral palsy (CP) would help rehabilitation professionals and parents engage in joint decision-making on prognosis, goals, and services.

Objectives: (1) To create developmental trajectories of body structure/function and participation for children with CP and (2) to explore relationships between services and outcomes. We hypothesized the following: (1) The development of children with CP varies based on their functional ability classification, with lower as compared with higher functional ability leading to smaller improvements in their development; (2) younger children with CP would improve at a greater rate than older children; (3) children with CP who developed at the highest level would have received family-centered services with more focus on task-specific practice and environmental modifications, and parents would report their child's needs were met to a greater extent as compared with those developing at the lower levels.

Methods: We undertook a prospective longitudinal cohort design with 708 children with CP, 1.5 to 11.9 years old, and their parents recruited from 45 sites, clinics, or practices in the United States and Canada. Trained therapists assessed children 2 to 5 times over 2 years using valid, reliable, standardized measures of balance, spinal alignment/range of motion, strength, and endurance. A smaller substudy of children with CP in 2 sites tracked walking and physical activity. Parents completed questionnaires on family demographics and on their children's endurance, health, participation in family/recreation activities, ability to perform self-care activities, and rehabilitation services. Physical and occupational therapists and parents collaboratively classified children on the Gross Motor Function Classification System (GMFCS). We analyzed body structure/function and participation data using linear and nonlinear mixed-effects modeling to create developmental trajectories for GMFCS levels. We explored relationships between service amount, focus, family-centeredness, and the extent to which children's needs were met by services and outcomes of balance, endurance, and participation by (1) adding services to the longitudinal curves and evaluating model fits, and (2) using multinomial models to determine relationships to percentile categories of progressing "more than" (>90th percentile) and "less than" (<10th percentile) to the reference of "as expected" (20th to 80th percentile).

Results: Developmental trajectories overall indicate that children with lower functional ability had smaller improvements and improved more during younger ages. Four measures varied from this pattern: (1) impact of health conditions, which basically remained constant; (2) spinal alignment/range of motion limitations, which increased linearly; (3) strength, which increased and decreased linearly dependent on GMFCS level; and (4) the substudy measures of walking and physical activity, most of which showed decreases over time. Exploratory analyses of services to outcomes revealed several positive relationships to participation outcome percentile categories for the extent to which services were meeting children's needs; family-centeredness; and a focus on structured play/recreation activities, health, and well-being.

Amount of services showed relationships to children's functional ability level, with more services to children with lower functional ability who showed smaller changes on the outcomes.

Conclusions: Developmental trajectories for children with CP by GMFCS levels should help rehabilitation professionals and families discuss prognosis and collaborate on service planning. Services that are family-centered; engage in joint decision-making; consider the needs of the child; and focus on structured play/recreational activities, health, and well-being may enhance interventions for children with CP and their families.

Limitations: Generalization of the developmental trajectories results to children with diagnoses other than CP or for children from countries/cultures other than the United States or Canada should be applied cautiously due to differences in diagnoses' developmental patterns, parental beliefs, environments, and how health care services are provided. The design of the study was not well suited to the evaluation of services effects, and services were parent-reported estimates; therefore, generalization of the service results by policymakers should be done cautiously.

BACKGROUND

We designed the On Track study to provide rehabilitation professionals and families of children with cerebral palsy (CP) with evidence-based information about children's development that enables shared decision-making related to services in order to achieve meaningful outcomes. Cerebral palsy is the most prevalent childhood-onset neuromuscular condition, and more than 90% of all individuals with CP live well into adulthood.[1-4] Although the underlying pathophysiology of CP is nonprogressive, the clinical manifestations are variable and change with age. In previous research, a decline in performance, defined as changes in gross motor function in daily life, has been noted as early as the teen years and has been documented in adulthood.[5-8] Beginning when their children are young, families need evidence to guide decisions about effective and cost-efficient services and supports that build capacity and prepare children and youth for life as adults. Empirical research demonstrates that most parents want information about current services and advice to plan for the future.[9] This need is greater for parents of children with more significant motor limitations.[9] Responding to this need is a key focus of family-centered care,[10] which is considered best practice in pediatric rehabilitation.

Decisions on amount of physical therapy (PT) and occupational therapy (OT) for children with CP are often based on convention.[11] Bailes et al[12] recommended a frequency of 1 to 2 therapy sessions per week, or every other week for children who demonstrate continuous progress toward goals. This recommendation corresponds to the number of sessions of PT and OT that most children were receiving during our previous US/Canadian study, Move & PLAY.[11] The Move & PLAY study finding that there were no differences in the amount of PT and OT sessions received by children in different regions in the United States further supports the perspective that decisions are often based on convention. Models of service delivery, financial resources for publicly funded services, availability of therapists, physical space for intervention, and private health insurance plan benefits are factors that likely contribute to conventions for amount of services. The small percentage of children receiving more than 12 sessions per

month of PT or OT indicates that intensive therapy (4-5 times per week), as defined in research,[13-18] is not common in practice.

Our findings on the number of therapy sessions children received in the previous 12 months most likely reflect the financial cost and the family time commitment associated with a high intensity of therapy, coupled with research evidence that the effect of additional therapy is currently not fully substantiated.[19] Parent and professional advocacy are also likely to influence decisions on amount of therapy. In the previous Move & PLAY study, 32% of children with CP in the United States received PT and 27% received OT in both an education and a clinic setting; approximately 33% received both OT and PT.[11] This implies that many parents or professionals, or both, did not think that a single provider was meeting child and family needs. Coordination of services, both within and between settings, is therefore important for children receiving PT and OT.

We designed the On Track study to fill gaps in fundamental knowledge by creating developmental trajectories of performance of children's self-care abilities in daily life, an important priority for families with children with CP,[20] and 1 aspect of participation of children—the frequency of participation in family and community recreation.[21]

The gap addresses what has been described as a "pressing need" to "increase our understanding of the complexities of CP," which is required for families to be able to understand their child's development and to make appropriate choices about services in collaboration with service providers.[2] Furthermore, changes over time in postural control (a defining feature of CP), secondary impairments (muscle strength, spinal alignment/range of motion, and endurance for activity), and the impact of cooccurring health conditions have not been quantified. Recent reports of the high prevalence of cooccurring health conditions[2,22] suggest that this should also be a focus of monitoring so that families can be better informed about prognoses and expectations. Creation of developmental trajectories would enable families of children with CP and rehabilitation providers to (1) anticipate a child's future strengths and needs (prognosis); and (2) proactively and collaboratively plan efficient services

and supports to optimize a child's health, function, education, and social participation, and to mitigate secondary impairment risk.

The aims of our research are consistent with the consensus of an international workshop titled Adults With Cerebral Palsy.[23] Workshop participants advocated for research that "improves understanding of the natural history of musculoskeletal and neurological impairments across the lifespan in persons with CP."[23] Pragmatically, it is difficult to study the natural history of a childhood condition; therefore, On Track was a study of the clinical course—ie, documenting but not controlling the rehabilitation services received by participants. Others have advocated for the use of the International Classification of Functioning, Disability, and Health (ICF)[24] and have emphasized the need to pay particular attention to pain, mobility issues, and comorbidities[25,26]—all of which we included in this study. We also concur with the perspective that "research is needed in which . . . CP is categorized by a standard typology [using] Gross Motor Function Classification System (GMFCS) functional levels, with independent, longitudinal assessments of standardized outcome measures from childhood to adulthood."[25] We add that measures of determinants of outcomes and classification of manual ability and communication function ought to be considered, particularly for understanding developmental trajectories of activity and participation domains of the ICF.

PARTICIPATION OF PATIENTS AND OTHER STAKEHOLDERS IN THE DESIGN AND CONDUCT OF RESEARCH AND DISSEMINATION OR FINDINGS

Before our PCORI application, we received funding from the Canadian Institutes of Health Research (CIHR) to create percentile curves based on 2 data points, 1 year apart. Funding from PCORI allowed us to increase the sample size and to extend data collection for some participants to 5 data points; this enabled the creation of longitudinal developmental trajectories. We also were able to increase the number of parent research team members (our stakeholders) from 2 to 7. A limitation of our project is that stakeholders were not involved in determining the aims and methods. Our stakeholders are 7 parents of children with CP. Their children varied in age from 7 to 20 years at the start of the study and varied in motor, cognitive, and communication abilities. Investigators communicated with service agencies to identify parents who might be interested in being part of a research team and were comfortable sharing their thoughts and experiences. Consequently, parent members of our team most likely represented families who actively advocate for their children. Parents were selected to represent Ontario, Canada, and the 4 regions of the United States where the study was conducted: Seattle, Washington; Atlanta, Georgia; Oklahoma City; Oklahoma; and Philadelphia, Pennsylvania. The study investigator in each region identified parents and invited them to participate. We invited 1 parent each from the Seattle, Atlanta, and Oklahoma City metropolitan areas and 2 parents from the Philadelphia metropolitan area. Additionally, we invited 2 parents who had served as consultants for our previous Move & PLAY study. One resided in Pittsburgh, Pennsylvania, and 1 in Toronto, Ontario, Canada. In addition to providing regional representation, 7 parent members proved to be a good number for small group discussions and, combined with the academic researchers, this was a manageable number for team interaction.

All parents were mothers. Five families were White and 2 were African American. The children of 5 of the parent team members were older than the participants in our study. This was invaluable, as parents not only had seen their children through to adulthood but also

informed the research team of the importance of future planning, beginning early in childhood. This has important implications for sharing the developmental trajectories created in this study with families.

Parents engaged in conversations about their roles and contributions throughout the study. Monthly teleconferences were the primary method of engagement. An email including an agenda and other meeting materials was sent before each meeting. Parents were asked to submit agenda items, and meetings began with a request for additional items. Between meetings, parents sometimes worked individually or in groups to complete tasks. Several parents expressed that initially they did not know what to expect and had limited knowledge of the research process. Parents shared that the encouragement they received to share ideas, their perception that investigators valued their input, and tangible evidence of their contributions were key to their engagement. Investigators were sensitive to ensuring that requests did not exceed parents' comfort levels and time commitments; this proved to be a learning process. We became aware of parents' busy schedules and that participating in monthly meetings was a big commitment for some. We were fortunate that all members of the team respected one another and differences in opinion; consequently, we did not experience problems with our group process.

Activities included writing the "Parent to Parent" column in the On-Track Family Newsletter (distributed twice a year to study participants), preparing an Exit Survey of open-ended questions, and analyzing survey responses. Three parents presented at 2 national conferences and at a regional family day sponsored by the CanChild Centre for Childhood Disability Research. The theme for the presentations was "Bringing the Family's Voice to Research." Parents shared their desire to develop educational media for families. Two videos, *Checking Up and Checking In: Partnering With Families of Children With Cerebral Palsy* and *Creating the Future: Engaging Children With Cerebral Palsy in the Circle of Care*, were produced and are available on the CanChild website.[27] The videos feature 3 of the participating families.

A limitation of our study was that parents did not join the research team until after we finalized the research questions and study design. Additionally, we did not include any fathers;

including fathers is recommended for future projects. Parents advised on issues that emerged during the study (eg, guidance for assessors about what to do when parents of children with considerable limitations express frustration that their child is unable to perform items on standardized measures). Parents are currently engaged in dissemination of study results, including appropriate and potentially effective formats for families of children with CP. We envision their continuing contributions to interpretation of results and discussions to facilitate adoption of research evidence into practice. Study parents have had a 2-part article accepted for publication in eParent.com about participating in research, and they attended the final meeting of the research team in June 2017 related to dissemination efforts.

METHODS

Design and Aims

We used a prospective cohort design. This design offers an alternative to randomized controlled designs in providing an evidence base for realistic goal setting and intervention planning for heterogeneous groups of individuals. Comprehensive rehabilitation outcomes research using the prospective cohort design is useful for a less-uniform group of people (such as individuals with CP), when interventions are multidimensional and individualized, and when there are significant personal and environmental influences on outcomes.[28]

The On Track study was an international (Canada and United States), multisite study of children with CP, aged 1.5 to 11.9 years, that had 2 funding sources. A grant funded in 2012 by CIHR involved testing children twice over a 1-year period (at study onset and 12 months later) using the study measures with the aim to create percentile graphs of development. Funding in 2013 by PCORI involved testing children 3 additional times (at approximately 6, 18, and 24 months post–study onset) over a 2-year period.

The PCORI-funded aims are primarily prognostic. The first aim was to assess average developmental change and variability by creating longitudinal trajectories for balance (a primary impairment); spinal alignment/range of motion limitations, strength, and endurance (secondary impairments); impact of health conditions; participation in family/recreational activities; and performance of self-care activities by estimating the average pattern of change over time for children who were grouped by their functional ability levels. We expected that the children with CP would vary in their development based on their functional ability classification, with lower as compared with higher functional ability leading to smaller improvements in their development. We also hypothesized that younger children with CP would improve at a greater rate than older children with CP.

The second aim was to describe the relationship between the amount, focus, and family-centeredness of therapy services and outcomes in impairment and participation based on (1) trajectories of longitudinal development and (2) percentile categories. We hypothesized

that children who developed at the highest level would have family-centered services with more focus on task-specific practice and environmental modifications, and that parents of those children would report their child's needs were met to a greater extent as compared with those developing at the lower levels.

An additional substudy involved taking direct walking and physical activity measurements from a subset of children at 2 sites in the United States. By creating longitudinal developmental trajectories for walking and physical activity based on GMFCS functional ability levels, we aimed to compare average change over time in walking and physical activity (amount of steps per day, intensity of steps, amount and intensity of physical activity counts) between GMFCS levels.

Research Team Members

The research team for the On Track study consisted of physical therapist, physician, and biostatistician investigators from 2 universities in Canada and 4 universities in the United States; 2 project coordinators (1 in Canada and 1 in the United States); regional coordinators at each data collection site; 7 parents of children with CP; and 90 physical and occupational therapist assessors across the United States and Canada. Investigators, study coordinators, regional coordinators, and parent researchers (as available) participated in monthly team meetings about the study via Skype. Discussions in these meetings included resolving study recruitment, assessment, and budget issues, and reviewing data as they became available. During each team meeting, members reviewed recruitment tables to monitor progress and to identify if targeted recruitment was needed. For any participant for whom study eligibility was in question, team members reviewed questions and, with input from the physiatrist team member, documented eligibility decisions. Additionally, as therapist assessors had questions regarding scoring of assessment items, the team used a tracking table to document scoring decisions for use as a reference guide and for sharing with assessors. In addition, the team developed and updated a plan for study dissemination at professional conferences, and in the professional and lay literature. The team posted all dissemination products on the study website.

The therapist assessors assisted in the recruitment process; however, their primary role was to collect data for the study. Their training consisted of a standardized, full-day regional training workshop to learn about the study and the measures and equipment used for data collection. Information relating to safety, privacy, confidentiality, other ethical issues, and administrative procedures was also provided and discussed during training workshops. Standardized written materials were collated in resource binders for the assessors, supplemented by training CDs and PowerPoint presentations. Therapist assessors were provided with equipment kits containing the supplies needed to conduct assessments. After training on the measures, each assessor independently viewed and scored criterion test videos of assessments of children with CP. These results were compared with investigators' "gold standard" consensus scores. Each assessor was required to demonstrate at least 80% agreement with the investigators' consensus scores in order to be approved for starting data collection. Throughout the study, the assessors received a semiannual newsletter highlighting tips on conducting and scoring the assessments. Yearly telephone conferences invited assessors to share updates, to work together to address questions, and to share strategies and solutions in order to be proactive about how to respond in situations of uncertainty.

IRB/Ethics

IRB review and approvals for the study were maintained at each university, as well as at some clinical sites. Clinical sites without an independent review board accepted the local university IRB approval. Per IRB requirements, all research team members and US therapist assessors completed the Collaborative Institutional Training Initiative training associated with human subjects research. All parents, as well as children of appropriate age, signed approved consent and assent forms, according to each site's IRB approvals, before the start of data collection. Table 1 lists details of the approvals by data collection site.

Parent/Child Participants

Study participants were children with CP aged 18 months to 11 years old at the time of recruitment, and their families. Children were eligible for this study if they had a diagnosis of CP

or were suspected to have a diagnosis of CP (ie, if they exhibited delayed motor development, muscle stiffness, and difficulties with balance and moving).

For children who did not have an initial diagnosis of CP, their diagnosis was continually revisited by the therapist assessors throughout the study so that the final data set for analysis represented children with CP. If the therapist assessor at any study visit questioned that the child had a diagnosis of CP, then our study physiatrist reviewed the child's case in detail for determination of final study eligibility. We selected 18 months as the minimum age because (1) a diagnosis of CP is more certain at 18 months than at 12 months (see, for example, Nelson and Ellenberg[29]), (2) starting that young ensured that we had data from the earliest possible time for the developmental trajectories, and (3) it was still possible to administer the measures that we had selected to children of that age. Because reliability of the GMFCS is greater after age 2 years,[30,31] we confirmed the GMFCS level at the end of the first year for children younger than 2 years of age at study onset.

Table 1. IRBs Approving the Study

Region	IRB
Seattle, Washington	University of Washington Human Subjects Division
Philadelphia, Pennsylvania	Drexel University Human Research Protection Program
Oklahoma City, Oklahoma	University of Oklahoma Health Sciences Center Institutional Review Board
Atlanta, Georgia	Mercer University Institutional Review Board for Research Involving Human Subjects
London, Ontario, Canada	Western University Health Science Research Ethics Board
London, Ontario, Canada	Thames Valley Children's Centre Research Advisory Committee
Hamilton, Ontario, Canada	Hamilton Integrated Research Ethics Board, McMaster University
St. John's, Newfoundland, Canada	Health Research Ethics Authority
Winnipeg, Manitoba, Canada	University of Manitoba, Bannatyne Campus Health Research Ethics Board
Regina, Alberta, Canada	Regina Qu'Appelle Health Region Research Ethics Board
Victoria, British Columbia, Canada	Vancouver Island Health Authority Health Research Ethics Board

We selected 11 years as the oldest age at recruitment because it enabled data to be collected through late elementary school age. Parent questionnaires were available in English, French, and Spanish. Translations were completed by bilingual therapists, then back-translated and reviewed by the study investigators and translators to finalize the translations. Families who could not read or communicate in one of these languages were not eligible to participate in this study.

Recruitment

We recruited from 19 sites, clinics, and/or practices within the United States and 24 sites, clinics, and/or practices within Canada, spanning both countries from east to west coasts and in the United States from north to south borders. Table 2 displays details of the geographical locations for participants. Convenience sample recruitment was conducted by regional coordinators and managed centrally by the project coordinator for each country. The

first wave of recruitment was asking families who participated in the Move & PLAY study[28,32] and who had previously consented to have us approach them for future studies (n = 275). Of these families, 87 agreed to participate in the On Track study. In order to recruit more children, we used various methods, proposed by the investigators and approved by the parent researchers, that we previously found to be successful[33] and that were approved by the site IRBs. Recruitment methods included referrals from health care professionals; identification of "champions" at recruitment sites to promote the study; placement of informational flyers at sites where therapy is received; and screening of medical records from specific hospitals or programs by site staff, according to our inclusion criteria. The regional coordinators reviewed and verified inclusion criteria when they contacted the families to provide information regarding their assigned therapist assessor. Inclusion criteria were also revisited and confirmed at the first and each successive therapist assessment visit.

We did not track information about families who did not meet the inclusion criteria before the point of consent. However, we have documented information about eligible families who consented verbally or in writing but then later were not enrolled; the primary reason was that they did not answer our attempts to contact them after initially indicating interest in the study. Also, some families declined to participate due to already having too many commitments; they felt they did not have adequate time for the study.

Table 2. Recruitment and Assessment Sites and Regions

Sites in United States	
Pacific Northwest • Children's Therapy Center • Good Samaritan Children's Therapy Unit MultiCare Pediatric Therapy Services • Seattle Children's Hospital) Shriners Hospital for Children, Portland • South King Early Intervention Program UW • Medicine/Valley Medical Center Waypoint Pediatric Therapies	**Philadelphia** • Atlantic County Special Services Children's Specialized Hospital Cindy Miles & Associates Good Shepherd Rehabilitation Network HMS School • Kennedy Krieger Institute Private therapists Voorhees Pediatrics • Weisman Children's Hospital and Rehabilitation Center
Oklahoma • Early Intervention Heart Springs School Private therapists	**Georgia** • Private therapists
Sites in Canada	
Ontario	

• Hamilton, Ontario
  ○ McMaster Children's Hospital Developmental
  ○ Pediatrics and Rehabilitation Program
• Kingston, Ontario
  ○ Religious Hospitallers of Saint Joseph of the Hotel
  ○ Dieu Kingston
• London, Ontario
  ○ Thames Valley Children's Centre
• North Bay, Ontario
  ○ One Kids Place Children's Treatment Centre
• Ottawa, Ontario
  ○ Ottawa Children's Treatment Centre
	• Peterborough, Ontario
	  ○ Five Counties Children's Center
	• Simcoe York, Ontario
	  ○ Children's Treatment Network
	• St. Catharines, Ontario
	  ○ Niagara Children's Centre
	• Timmins, Ontario
	  ○ Cochrane Temiskaming Children's Treatment Centre
	• Toronto, Ontario
	  ○ Holland Bloorview Kids Rehabilitation Hospital
	• Windsor, Ontario
	  ○ John McGivney Children's Centre

Other Provinces	

• Halifax, Nova Scotia
  ○ IWK Health Centre
• Prince Albert, Saskatchewan
  ○ Victoria Hospital Therapies Department
• Regina, Saskatchewan
  ○ Wascana Rehabilitation Center
	• St. John's, Newfoundland
	  ○ Janeway Children's Health and Rehabilitation Centre
	• Winnipeg, Manitoba
	  ○ Children's Hospital of Winnipeg Provincial Outreach Therapy for Children St. Boniface Hospital
	  ○ Winnipeg Rehabilitation Center for Children

British Columbia	
• Surrey, British Columbia Centre for Child Development • Victoria, British Columbia Queen Alexandra Center for Children's Health	• Vancouver, British Columbia *B.C.'s Centre for Ability* *Sunny Hill Center (part of BC Children's Hospital)*

Once the children and families signed consent forms and were entered into the study, we collected further health information at the baseline and follow-up visits. If families indicated other diagnoses at any of the visits, or if the therapist assessors felt that there was some question about a continued pattern of CP, then our team physician reviewed the case information to determine whether the child was eligible to continue in the study (Figure 1).

The greatest obstacles for recruitment were administrative issues beyond our control: time lag in setting up contracts with actual therapy sites and approving payments for recruitment efforts, competing needs of therapists at participating sites, facilitation of multiple IRB applications, and therapist workloads in general.

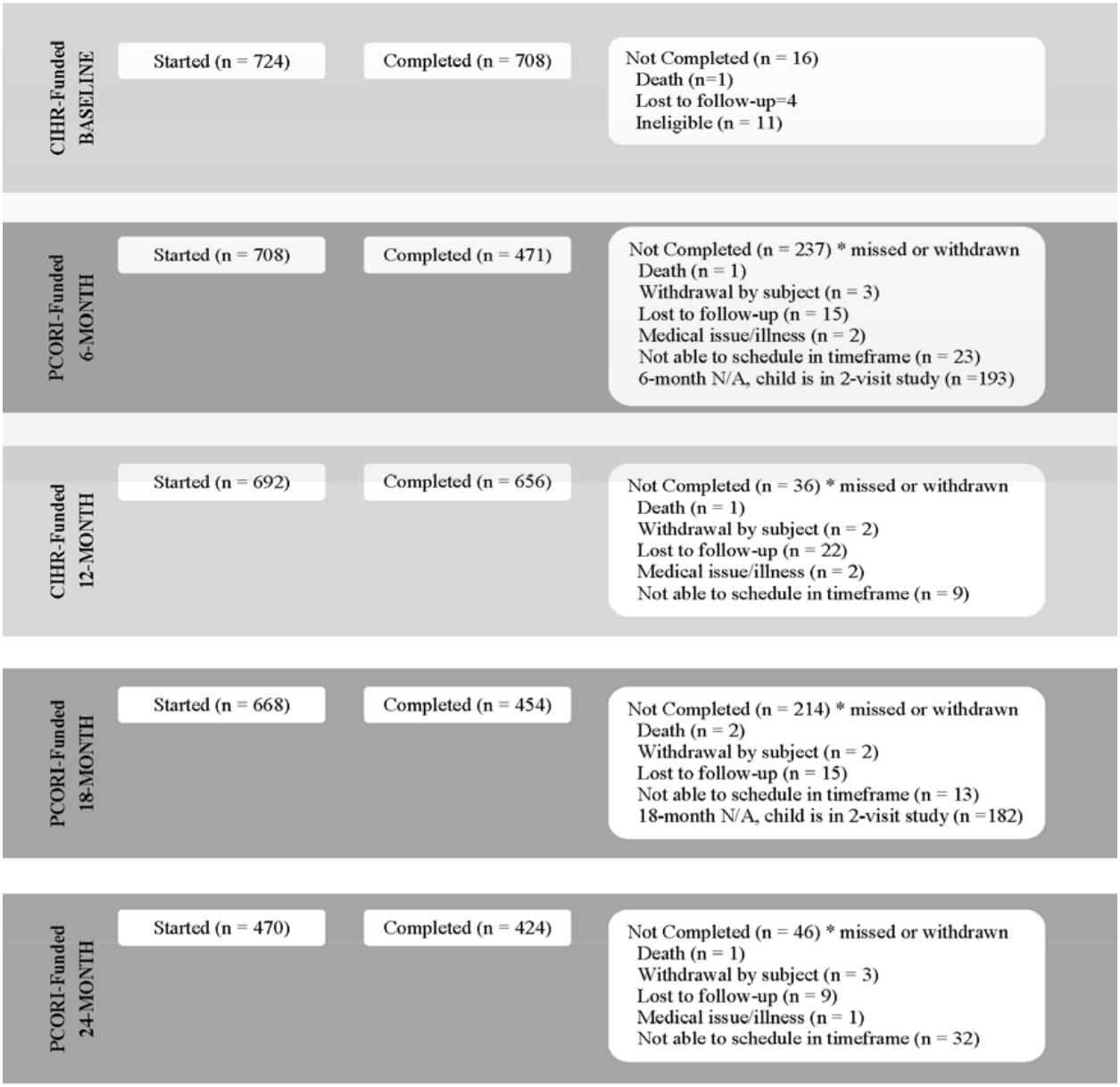
Retention

Once enrolled, all families were routinely contacted by the regional coordinators to let them know which assessor would be working with them. The assessor then contacted the families to schedule the assessment times. Assessors were to call and leave a message with families a maximum of 3 times. Use of email, if provided by the families, also assisted with more efficient communication. For families that did not respond via email, the regional coordinator mailed them a letter indicating that the assessor had been unable to reach them by phone or email; that they should contact the regional coordinator if they wanted to continue with the study; and that if they did not contact the study team, we would not make any further attempts to contact them. As the data were being checked and entered into the database for active families in the study, the assessors or regional coordinators contacted families as needed to obtain information about missing items on the parent questionnaires.

We enhanced retention through several methods. We disseminated a semiannual Family Newsletter (available to view on our study website under “Newsletters”).[27] Each newsletter shared a wide variety of information about the study as well as tips and perspectives from the parent researchers regarding raising a child with CP. We also offered tokens of appreciation to families by giving them a study magnet at the beginning of the study, a $20 gift card for the child at each study visit, remuneration for parking and travel to attend the visits as

applicable, and a feedback form summarizing assessment scores after each visit. Children received a “Junior Scientist” certificate after their final study visit.

Figure 1. Participant Flow Diagram



Abbreviations: CIHR, Canadian Institutes of Health Research; N/A, not applicable.

Parents were encouraged to share the feedback forms with their child's therapist(s) to facilitate discussions about how child, family, and service factors interact; planning and evaluating interventions; and supports to enhance the child's motor development and participation in daily activities.

Sample Size

We aimed to recruit a total sample size of 600 children (130 in each GMFCS level and approximate 18-month age bins). Because we experienced a 10% attrition rate in the Move & PLAY study, in which each child participated over a 1-year period, we modeled the same attrition rate; therefore, we planned to recruit 660 children.

We estimated the On Track study sample in terms of the requirements for the estimation of LMS centiles (CIHR grant aim), which are more demanding in terms of sample size than the longitudinal trajectories. We used calculations provided from Crawford and Garthwaite,[34] assuming estimation of 7 age bands (ages are not actually binned in the analysis). This sample size is also sufficient for the mixed-effects analyses of change trajectories and prediction, which are generally less demanding in terms of the number of children. We extended the longitudinal follow-up assessment number from 2, as funded in the CIHR grant, to 5 occasions, as funded in the PCORI contract, which compares favorably to change trajectories for gross motor function estimated with considerable precision by Rosenbaum et al.[35] For example, key parameters of the nonlinear model in Rosenbaum et al[35] included the predicted limit of gross motor function and the age by which 90% of this limit is achieved, estimated in GMFCS Level III from an average of 4.1 assessments each of 122 children with 95% CIs of ±3% and ±15%, respectively. We expected similar precision for our nonlinear models and considerably better precision for outcomes for which linear models could be used.

Measures

Demographics

We collected demographic information about children and families by having parents complete a study-specific demographic questionnaire. We adapted race and ethnicity questions

from the 2010 US Census and Statistics Canada's 2011 Census of Population. We did not collect information on the type of motor disorder because of known difficulties with reliability of this classification system[36]; however, we did have the assessors note the limb distribution of the CP. Table 3 includes details on the data collected.

Functional Classifications

We used 3 functional classification systems to describe the children according to their gross motor, manual ability, and communication functions: the GMFCS,[37] the Manual Abilities Classification System (MACS),[38] and the Communication Function Classification System (CFCS).[39] Table 3 includes details on the 3 classification systems.

Outcomes

The outcomes for this study have been identified as important by families of children with CP.[32,40,41] We chose measures by consensus among the investigators based on our previous findings within the Move & PLAY study about determinants (child, family, rehabilitation, and community services) of gross motor function, self-care, and participation in family and recreational activities in children with CP aged 1.5 to 5 years,[32,40] and based on measure reliability, feasibility to administer, ease of scoring and interpretation, use as an evaluative tool, and acceptability for parent and child participants.[40] Constructs measured included the following: (1) impairments and associated health conditions, specifically balance, spinal alignment/range of motion, muscle strength, endurance for activity, walking endurance (distance), impact of health conditions, and, for a subgroup from 2 US sites (Seattle and Atlanta), daily walking (intensity, amount) and physical activity (intensity, amount); (2) frequency of participation in family/recreational activities and performance of self-care activities in daily life (degree of independence); and (3) rehabilitation services. Table 3 includes a short description of each measure and the variable analyzed within the data analysis. Table 4 contains a summary of the psychometric properties of the measures used, as well as details of the data collected, and who collected data at various study visits.

Table 3. Description of Measurements Used Within the On Track Study

Measure (acronym)	Construct measured	Test/measure description
Demographics		
Demographic questionnaire	Demographics	The parent-completed questionnaire included child age at entry into the study, child gender, child race and ethnicity, parent age, parent respondent's relationship to child, parent highest education level achieved, parent race and ethnicity, number of children and adults living in the home, and total household income.
Limb distribution of CP diagram	Limb distribution of CP	Assessors circled on a diagram the configuration of the CP on the children's limbs, classifying as mono-, di-, tri-, or quadriplegic.
Functional classifications		
Gross Motor Function Classification System (GMFCS)[37]	Gross motor function	The GMFCS is a classification system based on functional body movement ability. GMFCS levels vary from I to V, with a level closest to I reflecting higher function. The following are the general descriptions of a child at 6-12 y of age: I = walks without limitations; II = walks with limitations; III = walks using a hand-held mobility device; IV = self-mobility with limitations (may use powered mobility); and V = transported in manual wheelchair. Descriptors for the 5 levels vary by age of the child.
Manual Ability Classification System (MACS)[38]	Manual ability function	The MACS is a classification system based on functional hand movement ability. MACS levels vary from I to V, with a level closest to I reflecting higher function. The following are the general descriptions for each level: I = handles objects easily and effectively; II = handles most objects with somewhat reduced quality and/or speed; III = handles objects with difficulty, needs help to prepare and/or modify activities; IV = handles a limited selection of easily managed objects; and V = does not handle objects and has severely limited ability to perform even simple actions.
Communication Function Classification System (CFCS)[39]	Communication function	The CFCS is a classification system based on functional communication ability. CFCS levels vary from I to V, with a level closest to I reflecting higher function. The following are the general descriptions for each level: I = effective sender and/or receiver with familiar and/or unfamiliar partners; II = effective but slower-paced sender and/or receiver with familiar and/or unfamiliar partners; III = effective sender and/or receiver with familiar partners; IV = inconsistent sender and/or receiver with familiar partners; and V = seldom effective sender and/or receiver with familiar partners.

Measure (acronym)	Construct measured	Test/measure description
Impairments and Associated Health Conditions		
Early Child Assessment of Balance (ECAB)[46,68]	Balance	The ECAB addresses postural control and balance across the developmental sequence. Part I has 7 items (1, 4, 5, 6, 7 scored bilaterally): (1) lateral head righting, (2) head righting in extension, (3) head righting in flexion, (4) rotation in the trunk, (5) equilibrium reactions in sitting, (6) protective extension to the side, and (7) protective extension backward. The items are scored on a scale of 0 = no response to 3 = complete and consistent response. Part II has 6 items: (1) sitting with back unsupported but feet supported, (2) moving from sitting to standing, (3) standing unsupported with eyes closed, (4) standing unsupported with feet together, (5) turning 360° in standing unsupported, and (6) placing alternate foot on the step while standing unsupported. Items are scored on a variable scale, which is weighted due to the increased difficulty of the items for a summed total score between 0 and 100. A higher score represents better balance.
Spinal Alignment and Range of Motion Measure (SAROMM)[69]	Joint range of motion	The SAROMM has 2 subscales. The Spinal Alignment Subscale contains 4 items and the Range of Motion and Extensibility Subscale has 22 items. Each item is scored on a 5-point Likert scale, with 0 = normal alignment and range, with or without active correction; 1 = normal alignment and range with passive correction; and 2, 3, and 4 = fixed contractures that are mild, moderate, or severe based on specified cut points, and illustrated by photographs in the training manual. We used the SAROMM average score across all items for analysis. High scores indicate greater deficits in range of motion than low scores.
Functional Strength Assessment (FSA)[70]	Muscle strength	We measured muscle strength using the FSA to examine neck and trunk flexors and extensors and hip extensors, knee extensors, and shoulder flexors bilaterally from a functional perspective. Each item is evaluated on a 5-point ordinal scale from 1 (no initiation of movement against gravity) to 5 (full available range against gravity and some or strong resistance). We used the FSA average score for analysis. Higher scores indicate greater muscle strength.

Measure (acronym)	Construct measured	Test/measure description
Early Activity Scale for Endurance (EASE)[71]	Endurance fitness	We measured the construct of endurance for activity from the perspective of level of energy using 4 items from the original parent-rated EASE. Items rated include the following: (1) my child's physical activity level is similar to other children his or her age; (2) my child has a high physical energy level and rarely needs to take rests when moving himself or herself around during daily activities and play time; (3) my child does enough activity so that he or she is breathing quickly or gets flushing in his or her face at least 1 time each day; and (4) my child spends a lot of his or her play or free time doing activities that require lots of physical energy. Items are scored from 1 (never) to 5 (always). We used the EASE average score for analysis. A higher score represents greater endurance for activity.
Six-minute Walk Test (6MWT)[72]	Endurance fitness	We obtained another estimate of endurance using the 6MWT for children at GMFCS Levels I, II, and III, once they were older than 3 y of age. This is a simple, submaximal clinical exercise test in which the distance walked under controlled conditions in 6 min is measured. For young children, assessors hold the child's hand; for older children, instructions provided by Maher and colleagues[73] are used to motivate the children to walk for 6 min. We used the distance walked in feet (measured using a survey-measuring wheel) for analysis.
Child Health Conditions Questionnaire (CHCQ)[22]	Impact of health problems	We used the CHCQ to measure the extent to which health conditions influence children's activities, based on the new definition of CP[74] and body functions contained in the ICF.[24] Health conditions include problems with seeing, hearing, learning, communicating, controlling emotions, seizures, the mouth, the teeth and gums, digestion, growth, sleeping, repeated infections, breathing, the skin, the heart, and pain. Parents respond "yes" or "no" to each health problem listed and, if the child has a problem, parents judge the impact of the problem on the child's daily life using an 8-point Likert scale (from 1 = not at all to 7 = to a very great extent). We imputed an impact of 0 if the child did not have the problem. We conducted the analysis using the average impact of the health problems on daily life. A higher score represents a greater health impact.

Measure (acronym)	Construct measured	Test/measure description
Participation		
Child Engagement in Daily Life (CEDL)[75,76]	Part 1: Participation in family/recreation Part 2: Participation in self-care	The CEDL version 2 is a 40-item parent-completed questionnaire. Part 1 captures participation of the child in family/community and family/recreational activities. Part 1 is scored on 2 Likert scales: (1) how often a child participates (1 = rarely/never to 4 = very often), and (2) the degree of enjoyment (1 = not at all to 5 = a great deal). Part 2 measures self-care, defined as the degree to which the child participates in daily feeding, dressing, bathing, and toileting. Part 2 is scored on a 5-point Likert scale, from 1 = does not do the activity to 5 = does the activity independently most of the time." A Rasch analysis has converted scores into 0 to -100 scaled scores for both parts. A higher score represents a higher degree of 'family/recreation' (Part 1) and self-care (Part 2) participation.
Services		
Services Questionnaire[11]	Services provided	Using a parent-completed study questionnaire, we collected data on the rehabilitation services provided, and major medical/surgical interventions in the 6-mo period preceding each data collection point. These data included the following: information on medical service visits and procedures; amount of PT, OT, and ST services (coded into categories of number of sessions/y = 0-1, 2-30, 31-52, 53-155, or 156 or more); focus of therapy rated on a Likert scale, with 1 = not at all to 5 = to a very great extent in the categories of primary and secondary impairments, activity, environmental adaptations, self-care activities, structured play and recreation activities, self-awareness and motivation, and health and well-being; family-centeredness of therapy (rated on the same Likert scale as for focus); number of community programs; coordination of care (rated on a Likert scale for how well providers have worked together to provide care, with 1 = not at all to 5 = excellent); and parents' perceptions of the extent to which their child's needs were being met (5-point Likert scale with 1 = not at all to 5 = completely) . For data analysis, we selected the following service variables based on previous services analyses from the Move & PLAY study[47,52,53]: (1) amount of PT, OT, and ST coded into categories of number of sessions/y = 0 -1, 2-30, 31-52, 53-155, or 156 or more; (2) average focus score for OT/PT/ST services across the 8 focus categories listed above; (3) average family-centeredness score across all items; (4) average parents' perception of the extent that their child's needs were being met.

Measure (acronym)	Construct measured	Test/measure description
Physical activity substudy		
StepWatch[77]	Walking activity	For a subgroup of participants who resided in 2 study sites (Seattle and Atlanta), were ambulatory (GMFCS Levels I-III), and were at least 3 y, we measured walking activity performance in the context of daily life using a StepWatch monitor (Modus Health). The StepWatch is a small (70 × 50 × 20 mm; 38 g), waterproof, self-contained device. Participants wore the StepWatch on their left ankle (inside a knit cuff) each day for 7 days. Specific variables collected were the average daily step counts and percentage time walking in low-, moderate-, and high-stride rates. We used the average single leg strides/day and the average strides/d faster than 30/min for analysis.[78]
ActiGraph[79]	Physical activity	For a subgroup of participants who resided in 2 study sites (Seattle and Atlanta), we measured physical activity within the context of daily life with a 3-dimensional accelerometer, ActiGraph wGT3X (ActiGraph, LLC). Participants wore the ActiGraph on their dominant wrist for a 7-d sample. We converted the wrist-mounted ActiGraph activity counts by axis to waist-worn raw activity counts[80] for calculation of average physical activity counts/min and the minutes of moderate to vigorous physical activity, which we used for analysis.

Abbreviations: CP, cerebral palsy; ICF, International Classification of Functioning, Disability, and Health; OT, occupational therapy; PT, physical therapy; ST, speech therapy.

Table 4. Psychometric Properties of the Measures Used

Measure	Psychometric properties	Timing
Completed by parent respondent and assessor		
Gross Motor Function Classification System (GMFCS)[31,37]	Content validity: Confirmed via nominal group technique and Delphi survey Interrater reliability: κ = 0.75 for children older than 2 y	At baseline, 12-mo, and 24-mo visits
Manual Ability[38] Classification System	Content validity: Via consensus Interrater reliability: Between therapists: ICC = 0.97 (95% CI, 0.96-0.98); between therapists and parents: ICC = 0.96 (95% CI, 0.89-0.98)	
Communication Function[39] Classification System	Content validity: Confirmed via Delphi process Preliminary reliability: Interrater reliability, weighted κ = 0.67; test-retest reliability, weighted κ = 0.84	
Completed by assessor		
Early Clinical Assessment of Balance (ECAB)[46,50,68]	Content validity (n = 410)[68]: Established through expertise on research team Internal consistency: Cronbach α = .92 Construct validity: Known groups study: ECAB scores differed significantly among all GMFCS levels ($P < .001$); correlation with GMFM = 0.97 ($P < .001$); children aged <31 mo had significantly lower ECAB scores than children aged 31-42 mo or 43-60 mo ($P < .01$) Factor loading[70]: ECAB loaded most highly onto the Move & PLAY construct of "primary impairment" with a loading of 0.95 reliability[50] (n = 28 children with CP, aged 2-7 y) Interrater reliability: ICC = 0.989 (95% CI, 0.976-0.995) Test-retest reliability (same raters): ICC = 0.987 (95% CI, 0.971-0.994) Test-retest reliability (different raters): ICC = 0.986 (95% CI, 0.971-0.994); SEM = 3.6; MDC95 = [missing data]	At each visit

Measure	Psychometric properties	Timing
Spinal Alignment and Range of Motion Measure (SAROMM)[69]	Content validity: Via consultation with experienced pediatric physical therapists through focus groups; administration details, testing protocol and scoring criteria refined through a Delphi process Internal consistency: Cronbach α = .95 (Move & PLAY, unpublished) Construct validity: Age and GMFCS level contributed significantly to SAROMM score (r^2 = 0.44); known groups validity: scores differentiate children at all GMFCS levels, except II and III (P < .006)[70] Factor Loading[70]: The SAROMM loaded second most highly onto the Move & PLAY construct of secondary impairment with a loading of 0.74 Interrater reliability (n = 25; 5 in each GMFCS level): ICC = 0.89 (95% CI, 0.76-0.95) Test–retest reliability: ICC = 0.93 (95% CI, 0.86-0.97); SEM = 3; MDC95 = 9	At each visit
Functional Strength Assessment (FSA)[70]	Construct validity: Supported by similarity to standard methods of manual muscle testing in children Internal consistency: Tested in Move & PLAY (n = 429) (unpublished); Cronbach α = .93 Construct validity (n = 429)[70]: Known groups validity: significant difference among all GMFCS levels (P < .001), except for Levels II and III Factor loading: FSA loaded most highly onto the Move & PLAY construct of secondary impairment with a loading of 0.95 Interrater reliability: Tested in Move & PLAY (n = 28 children with CP); ICC = 0.996 (95% CI, [missing data]	At each visit
Six-minute Walk Test (6MWT)[72,73]	Concurrent validity: With VO_2 max = 0.44 (P < .001) (typical children 12-16 y)[73,81,82] Test-retest reliability: ICC = 0.94 (95% CI, 0.89-0.96) (typical children aged 12-16 y)[73]	At each visit

Measure	Psychometric properties	Timing
Completed by parent respondent		
Early Activity Scale for Endurance (EASE)[71]	11-item version[71] Construct validity: Known groups validity = significant differences among children developing typically and children with CP in 5 levels of GMFCS ($P < .001$); post hoc tests NS for Levels II and III (n = 520) Internal consistency: Cronbach α = .93 Convergent validity: Spearman's correlation with 6MWT = 0.57 ($P = .001$) (n = 14 children with CP and 14 children developing typically) Test–retest reliability: ICC = 0.95 (95% CI, 0.90-0.98) (n = 32 children with CP); SEM = 2.9; MDC95 = 8.0 4-item version (tested in Move & PLAY [n = 429], unpublished) Good model fit: CFA – short version; χ^2 = 2.8 NS; CFI = 0.998, TLI = 0.993; RMSE = 0.03 Internal consistency: Cronbach α = .83 Factor Loading[70]: The EASE loaded significantly onto the Move & PLAY construct of secondary impairment with a loading of 0.66 Test-retest reliability: ICC = 0.75 (95% CI, 0.54-0.87) Convergent validity (On Track, unpublished data; n = 376): GMFCS Levels I to III, Pearson correlation of EASE to 6MWT = 0.30 ($P < .001$) Construct validity: Significant differences between GMFCS Levels I to III, Level I > II > III ($P < .03$), between age groups, 1.5-3 y > 6-9 y, and 9-12 y ($P = .006$; $P = .001$) and 3-6 y, > 9-12 years old ($P = .006$), between sex, boys > girls ($P = .02$)	At each visit

Measure	Psychometric properties	Timing
Child Health Conditions Questionnaire[22]	Content validity: Developed from the international definition of CP[74] using the ICF[24] Construct validity: Known groups validity: significant differences in both number and impact of health conditions among children developing typically and children in GMFCS groups (I, II, and III, and IV and V) *P* < .001 (n = 537), post hoc testing: all groups significantly different from each other for number (*P* < .01); for impact, all groups significantly different from each other (*P* < .001) except for GMFCS Levels I and II and III Test-retest reliability: For number of conditions: ICC = 0.80, 95% CI, 0.63-0.90 (n = 32); for average impact: ICC = 0.85, 95% CI, 0.72-0.93 (n = 32); for average impact: ICC = 0.85, 95% CI, =0.72- [missing data] Construct validity (n = 429 in Move & PLAY and 110 children developing typically): Internal consistency: Cronbach α participation = .86 (frequency), .91 (enjoyment); self-care = .90; known groups validity: frequency in and enjoyment of participation in recreation and self-care varied by age and GMFCS level (ie, children developing typically, GMFCS I, GMFCS II and III, GMFCS IV and V) (*P* < .001), there was an age by motor ability interaction for self-care, with the youngest children performing less than the 2 older age groups (*P* < .001). All motor ability groups performed significantly differently (*P* < .001). Rasch analysis: Participation performed well; self-care has been improved by adding items of intermediate difficulty for use in the On Track study Test–retest reliability (n = 33): Participation frequency: ICC = 0.70 (95% CI, 0.47-0.84); participation enjoyment: ICC = 0.70 (95% CI, 0.47-0.84); self-care: ICC = 0.96 (95% CI, 0.91-0.98) Analysis as an evaluative measure (n = 387)84:	At each visit
Services Questionnaire[11]	Content validity: Via experienced clinician review Test-retest reliability: Amount of therapy visits ICC = 0.92; focus of therapy services ICC = 0.55 to 0.95; family-centeredness ICC = 0.86; No. of recreation and leisure programs ICC = 0.95; coordination of services ICC = 0.88; perception that services meeting needs ICC = 0.61	At each visit

Measure	Psychometric properties	Timing
Physical activity substudy measures completed by assessor		
StepWatch[77,78,85,86]	Construct validity: StepWatch: A review of pedometers and accelerometers[86] reported that StepWatch is the most accurate pedometer ever designed for walking and is capable of capturing actual strides taken to within ±3% for speeds from 1-5 mph.[85] Calibration stride count to manual count: Typically developing youth and children with CP ranged 97.7% to 101.4%[87]; superior accuracy for stride counts as compared with waist-mounted pedometers during treadmill walking in lean and obese youth aged 10-12 y[88]; accuracy and precision of the StepWatch was documented for treadmill walking speeds up to 4 mph (ICC = 0.995).[89] Test-retest reliability: Stride curves from 5-d sample: *P* = .38-.95[87]; StepWatch to manual count treadmill walking test-retest: ICC = 0.99558, ICC for *X*, *Y*, and *Z* axes >70.9	Physical activity substudy maximum subsample of n = 79 at any visit
ActiGraph wGT3X[79]	Construct validity: Feasible[90] and valid if worn for 7 d[79,91,92]; good validity compared with indirect calorimetry (*r* = 0.82-0.89) across studies, with differing definitions of count cut points for metabolic equivalent levels[80,93,94]; when wearing ActiGraph on the hip, Evenson cut points provide valid estimates of time spent in MVPA in populations of ambulatory children with CP[95]; using hip-mounted ActiGraphs, MVPA was greater in ambulatory youth with CP compared with youth who were nonambulatory.[90] In a similar study with adults with cerebral palsy, wearing ActiGraphs worn on the wrist, authors found different activity counts for nonambulatory and ambulatory adults.[96] Instrument reliability: ICCs = 0.83-0.98; wrist-worn placement of the ActiGraph in typically developing children had good interdevice reliability (r = 0.72) and validity against indirect calorimetry (*r* = 0.8; *P* < .01).	Physical activity substudy maximum subsample of n = 79 at any visit

Abbreviations: 6MWT, 6-minute Walk Test; CFA, confirmatory factor analysis; CFI, comparative fit index; EASE, Early Activity Scale for Endurance; ECAB, Early Clinical Assessment of Balance; FSA, Functional Strength Assessment; GMFM, Gross Motor Function Measure; ICC, intraclass correlation coefficient; max, maximum; MDC95, minimal detectable change (at the 95% CI); MVPA, moderate to vigorous physical activity; NS, nonsignificant; RMSE, root mean square error of approximation; SAROMM, Spinal Alignment and Range of Motion Measure; TLI, Tucker Lewis Index; χ^2, chi-square analysis.

Data Collection

The study began recruitment in April 2013 and completed recruitment in January 2015. At the first study visit, and approximately 12 and 24 months later, assessors collected data on GMFCS level,[37] the MACS,[38] and the CFCS,[39] as well as distribution of involvement. Both the parent and the assessor independently classified the child's function, then, in cases of disagreement, went through a structured process to reach a consensus about the child's classification levels.[42] We generated guidelines to reconcile disagreements for research purposes.[42]

We obtained information from parents either via paper booklets containing survey measures or, after the first visit, parents had the option to complete the survey measures online. We collected assessor data in paper booklets. We collected all data by the end of August 2016 and entered them into the online database (EmPOWER Health Research Inc). A second research staff member manually checked all data entries to ensure accuracy. We recorded data entry error rates; they are summarized by region in Table 5. These error rates were low and we corrected all errors.

We made various efforts, as described in detail in Table 6, to reduce missing data as much as possible. Across the 5 time points a total of 2713 assessments were completed. After the end of data collection, we assessed the amount of missing data and found it to be very small, ranging from 35 to 112 values for each measure. The number of children per assessment session with missing data on 6 of the 8 main measures was less than 2%. The other 2 measures, the Health Conditions questionnaire and the 6-minute walk test (6MWT), had missing data on children per assessment session ranging from 2.3% to 4.4%. We used mixed-effects random forests to impute missing data, instead of the originally planned multiple imputation method. Mixed-effects random forest imputation is relatively new, and treats children as clusters, differentiating person-level and sample-level data in the imputation process to produce more accurate child-level imputations. Details are described in Table 6.

Table 5. Data Entry Error Rates

	Assessor measures, all visits			Parent measures, all visits			Services questionnaire, all visits			Family demographics, baseline		
	(83 possible items)			(243 possible items)			(82 possible items)			(70 possible items)		
	No. of errors	No. of books	% error rate	No. of errors	No. of books	% error rate	No. of errors	No. of books	% error rate	No. of errors	No. of books	% error rate
Canada	113	1102	0.12	124	903	0.06	83	903	0.11	23	327	0.10
Philadelphia	47	293	0.19	78	285	0.11	24	285	0.10	2	71	0.04
Atlanta	28	522	0.06	49	471	0.04	17	471	0.04	0	112	0.00
Oklahoma	59	375	0.19	115	35	0.13	44	375	0.14	0	80	0.00
Seattle	8	434	0.02	26	356	0.03	8	256	0.03	0	99	0.00
All sites	**255**	**2726**	**0.12**	**392**	**2050**	**0.07**	**176**	**2290**	**0.08**	**25**	**689**	**0.03**

Table 6. Missing Data Plan

Efforts to reduce missing data during the study	
1	We reviewed our previous Move & PLAY documentation to identify specific items that were most often missing so we could address these with appropriate changes within the On Track study data measures. We alerted our assessors to the issues related to those items and offered proactive strategies to improve data collection via our Assessor Newsletter, assessor teleconferences, and specific communications as needed with assessors via regional coordinators and/or investigators.
2	Routine checks for missing data occurred at multiple levels, with assessors checking and making notes about missing data when collecting questionnaires from parents and regional coordinators and research assistants checking when entering data into the EmPOWER database. If missing data were detected, research personnel attempted to recover missing information from assessor and/or parents.
3	To track missing data carefully, we asked assessors and parents to provide brief notes about missing data within the comments boxes on the test forms. We also assigned descriptive missing data codes for all measures in our EmPOWER database.
4	Data from the parent and assessor forms were entered by a data entry assistant and then later checked by a different data entry assistant. Any errors were corrected and documented in an Excel tracking file and summarized in a detailed chart by site, over time. We reviewed data entry error rates 2 times/y to ensure that we were maintaining a high level of accuracy and to identify any specific measurement items that were frequently missing.
5	To minimize attrition, we worked diligently to keep participants engaged by providing individual feedback after every test session to families and by sending a parent newsletter from the team that was mailed out 2 times/y. We also tracked information about attrition via an attrition form within the EmPOWER database (ie, brief explanations when a participant was lost to follow-up before completing the study).
6	We discussed data queries as a standing agenda item within our monthly team meetings to determine the frequency of particular missing data, to determine if this appeared to be due to biased or unbiased reasons, and to make protocol decisions related to data collection and data entry. We tracked data queries cumulatively in a chart and communicated relevant information to regional coordinators and/or assessors as required.
Handling of missing data once all data were collected	
1	We did not calculate outcome scores if any item had missing data. Instead, we imputed scores on children who were assessed as follows.
2	We imputed missing outcomes scores using the mice package (multivariate imputation by chained equations) in R.[97] We imputed missing data only for those cases who attended an assessment. We did not impute data for children lost to follow-up.
3	Imputation order was according to the amount of missing data, with variables having the fewest missing cases imputed first.

4	For continuous variables, we used a MERF method, via a custom R function based on the code of Hajjem.[98] The MERF method incorporates random effects into a random forest model to improve the accuracy for clustered data. In our study, observations were clustered over time within child, so we used random effects to model within-child variability. The MERF algorithm is a fairly new development in imputation methods and is available only for continuous outcomes.
5	We imputed categorical outcomes with a conventional random forest model. In traditional multiple imputation methods, each imputed data set is analyzed and the results are combined according to established rules, and the variation in results across data sets is used to estimate the variability due to the imputation process.
6	We performed analyses on multiple imputations in our study since the MERF method is sampling based and because we observed so little between-imputation variance. We imputed 5 data sets and chose imputation 3 as the final data set for analysis.
7	If the amount of missing data is likely to affect the study results, sensitivity analyses are typically conducted to consider different assumptions about the causes of missing data and the effect on the results. We did not perform sensitivity analyses in our study because we imputed a relatively small number of values, ranging from 35 to 112 values out of 2713 values for each outcome. This amount of missing data was not enough to impact the distributional properties (mean and standard deviation) of our outcomes across the 5 imputed data sets.
8	We also reviewed the missing data codes with descriptors for missing data as a check for systematic bias in terms of cause.

Abbreviation: MERF, mixed-effects random forest.

We followed children for varying amounts of time, and we used all available information for every child. We excluded no children because of loss to follow-up. We imputed missing values only while a child was enrolled in the study and did not attempt to "forecast" future values. Primary methods for analysis for our study (mixed-effects modeling and quantile regression estimates of percentiles) inherently accommodate variations in assessment schedule, which effectively performs a required imputation at the time of analysis.

Data Analysis

Because there is great variability in children diagnosed with CP, we used the GMFCS[37] to group children for the longitudinal trajectories for balance, muscle strength, spinal alignment/range of motion, endurance for activity, health conditions, and physical activity; this will allow for appropriate prognostic use within clinical practice. We performed preliminary analysis of children's GMFCS, MACS, and CFCS levels from the first study visit to identify whether any combinations might be used to create the developmental curves for participation

in self-care and recreation activities; however, no consistent combinations of the 3 classifications systems emerged from the data.[43] In examining data, we found that performance in self-care and participation in family/recreational activities were better related to GMFCS than to the other 2 classifications, so longitudinal curves were produced using the GMFCS levels.

For Aim 1 and for the physical activity substudy aim, which were to assess average developmental change and variability by creating longitudinal trajectories for body function/structure and participation measures, we analyzed data from baseline and 6-, 12-, 18-, and 24-month visits by linear and nonlinear mixed-effects modeling to create longitudinal trajectories by GMFCS levels.

The youngest children in each level ranged from age 17 to 21 months and the oldest from 154 to 168 months; we chose to plot on the range of 24 to 144 months, for which the data were most dense. We used mixed-effects models to describe the relationship between age and each outcome. These models describe the nature of the relationship across the range of the data, while incorporating the dependency of multiple observations on a child. We used spider plots of the raw data to determine candidate models. Based on these plots, asymptotic nonlinear models were the model of choice for balance, endurance (Early Activity Scale for Endurance [EASE] and 6MWT), self-care performance, and participation in family/recreation for most ability levels. We fit linear models for range of motion, impact of health conditions, strides per day, and minutes of physical activity. We fit functional strength in children at GMFCS Levels I and II with asymptotic nonlinear models, and linear models were more appropriate for children at Levels III to V. We initially fit participation in family/recreation for children at GMFCS Level II with an asymptotic model, but this model showed essentially no change over time and, based on Akaike Information Criteria (AIC), a linear model was more suitable for this group. We created the longitudinal trajectories using the nlme package in R.[44,45] We used restricted maximum likelihood estimation in the final model fitting, although we used maximum likelihood estimation in the model selection phase to compare model AIC and to allow likelihood ratio tests (LRTs) to be performed. We used LRTs to determine if offset parameters improved the fit of asymptotic models. We evaluated final fitted models by examining residual

plots, by inspecting the model parameters for reasonableness, and by superimposing the fitted model onto raw data plots.

The goal of this aspect of the study was to model the change over time and the variability of children in different functional levels. Preliminary plotting of the outcomes by age for different genders, distribution, levels of communication ability (CFCS), and manual ability (MACS) did not suggest the need to include these variables in the model. We undertook analyses separately for GMFCS levels, instead of incorporating GMFCS level as a confounder covariate, because our work with the Gross Motor Function Measure (GMFM) indicated that the functional form of the relationship between outcome and age was influenced by GMFCS level. Modeling GMFCS as a confounder covariate would have enabled variations in the model parameters but not in the functional form. Nothing in the literature suggested that geographical factors would influence our outcomes, so we did not incorporate these into the longitudinal models.

Aim 2 was to describe the relationship between the amount, focus, and family-centeredness of therapy services and outcomes as trajectories of longitudinal change in impairment and participation. For this aim, we selected 13 service variables for analyses, including the categorized number of sessions of PT, OT, and ST; family-centeredness; and 8 focuses of service (primary impairments [relaxation of spasticity, balance, coordination], secondary impairments [strength, range of motion, endurance], activities to improve self-initiation, environmental [provision of assistive devices], self-care routines, structured play activities, self-awareness and motivation, and health and well-being); and parents' rating of the extent to which their child's needs were being met by services. The selection of these services encompasses the variables that we hypothesized would have positive relationships with the outcomes based on previous research, as well as representing the amount of service. We evaluated these services for the selected outcomes of balance, walking endurance, participation in family/recreation activities, and performance in self-care activities. We chose the outcome of balance as it has a strong correlation to gross motor development.[46] We selected walking endurance using the 6MWT data to represent functional mobility ability. We

selected participation in family/recreation activities and performance in self-care activities, as these outcomes represent the broadest and most important outcomes of effective therapy intervention.[47]

Country (ie, whether the children were from the United States or Canada) was the only confounder controlled for, which could be considered a limitation of the study. However, we expected country to have the greatest influence on the relationship between outcomes and services because of the differences in the health care systems between countries. We investigated the service variables individually to determine the effect of each on outcomes. There were 13 service variables of interest (8 related to focus, 3 related to intensity, and 2 related to perceptions of service). Had we decided to perform a multiple regression with all variables, we would have found the impact of each service variable, controlling for all other variables. Since the service variables measure related concepts, we thought a multiple regression approach would hinder our ability to find any relationships, since we expected the marginal contribution of each variable to be small. We did not investigate interactions between service variables because we had no a priori expectations of interactions, and the results of the univariate analysis did not suggest further work.

We used the nlme package in R to incorporate service variables into the longitudinal models[44]; for the multinomial regression, we used the nnet package.[48] We used service variables from the 12-month assessment because at this assessment families were asked about the services they had received in the preceding period. Furthermore, because we were investigating change from baseline to 12 months, we were interested in service provision in that same period.

We examined the relationship between the service variables and these outcomes using 2 different methods. First, we added the service variables individually to the GMFCS-specific longitudinal curves to determine if they had any impact on the outcomes. For nonlinear models, we added the service variable as a predictor of the model asymptote and rate of growth. For linear models, we added the service variable to the base model, in effect testing if the intercept (the value of the outcome at age 5) and the slope changed with respect to

services. We set the start value of the extra parameter to 0 in each case, and we made no attempt at adjusting start values to aid convergence. We assessed improvement in model fit with an LRT. We examined model fits for significance using $P < .05$. We calculated population predictions at 2, 5, and 12 years old for each level of service variable for those models in which the service variables improved model fit. For the Child Engagement in Daily Life (CEDL) participation in family/recreation and CEDL performance in self-care, we used the standard error of estimate (unpublished data) to create a confidence band around the prediction to determine if there was a meaningful difference between outcomes for those who received the least and most services.[49] For the Early Childhood Assessment of Balance (ECAB), we used the standard error of the mean to calculate 95% confidence bands.[50]

Second, we analyzed the selected service variables related to clinically significant change in percentile rank. We analyzed data on body structure/function and participation from the baseline and 12- and 24-month visits using quantile regression to develop reference centiles, which was the primary aim of the companion CIHR-funded study. We based the reference percentiles on data collected at annual intervals (maximum 3 per child) to minimize within-child dependency in the observations. We investigated associations between services and development using multinomial models. We classified development into 1 of 3 groups: developing "as expected," "more than expected," or "less than expected" based on the change in centile score over a 1-year period, from the baseline to 12-month visit, and controlled for each child's age and GMFCS level.

Those in the top 10% of change were the group developing more than expected, those in the middle 80% as expected, and those in the bottom 10% less than expected. We chose these cutoffs due to the variability of children's development and following a similar procedure as was done previously for the GMFM.[51] We used an LRT to determine if there was any relationship between the service variable and the child's progress classification (less than, more than, as expected), controlling for country. The LRT is a global test of predictor importance but does not indicate the direction of association. For those variables with a significant LRT, we examined multinomial regression coefficients. A multinomial regression is similar to a logistic

regression but with more than 2 groups of interest in the dependent variable. One category is treated as the reference category, and logistic regressions are modeled for each remaining category relative to the reference category. To facilitate interpretation, we restated the regression's coefficients as odds ratios and calculated the relative risks, comparing the group receiving the most services (ie, greater number of therapy sessions, focus to a greater extent, etc) to the group receiving the least services. We utilized service variables from the 12-month assessment in the analyses.

Potential Deviations From Methodology Standards

Deviations from PCORI Methodology Standards are noted and described in the accompanying Excel file.

RESULTS

Participants

Our recruitment and retention of participants are shown in the ClinicalTrials.gov (CTG) document, pages 5 to 9 (Participant Flow) and in Figure 1, yielding a final sample size of 708 for our longitudinal trajectories, except for the 6MWT, for which n = 456, as only children who were walking (GMFCS Levels I-III) and were at least 3 years old could participate. For the activity performance substudy utilizing measurement with the StepWatch (n = 50) and the ActiGraph(n = 79), the sample was smaller. For the analysis of services to the longitudinal data and percentile groups, we used all 708 participants' data. Our research includes a diverse sample for both the 2-assessment and 5-assessment studies that is comparable with population-based studies of children with CP.[22,40] The CTG document, pages 9 to 11 (Baseline Characteristics), contains child and parent baseline characteristics. The CTG document, pages 39 to 44, provides the number of participants per levels of the GMFCS and cross-tabulations of the number of participants within MACS and CFCS levels by the 5 GMFCS levels. We enrolled 34 children younger than 2 years of age and finalized their GMFCS at their 12-month assessment.

AIM 1: DEVELOPMENTAL TRAJECTORIES

We calculated 3 summary statistics for each combination of outcome and GMFCS level using linear or nonlinear mixed-effects modeling, as appropriate, to reflect the data for each measure: (1) the estimated value on the measurement scale at the age of 12 years for each GMFCS level to reflect differences based on functional ability; (2) a measure of rate of change, the Time-90 parameter (length of time required to achieve 90% of ability), to evaluate rate of development by functional ability (nonlinear models) and the slope parameter for linear models; and (3) the percent change in scores from 2 to 5 years old as compared with 5 to 12 years old, to determine if younger children with CP develop at a greater rate than older children with CP.

For detailed results, see Table 7 to view the descriptive data within the CTG tables and the longitudinal trajectories for each construct that includes an accompanying table with means (95% CI) at ages of 2, 5, and 12 years and the mean change (95% CI) in scores at 2 to 5 years and 5 to 12 years of age (Figures 2-13). A detailed summary by measure can also be found in Appendix A.

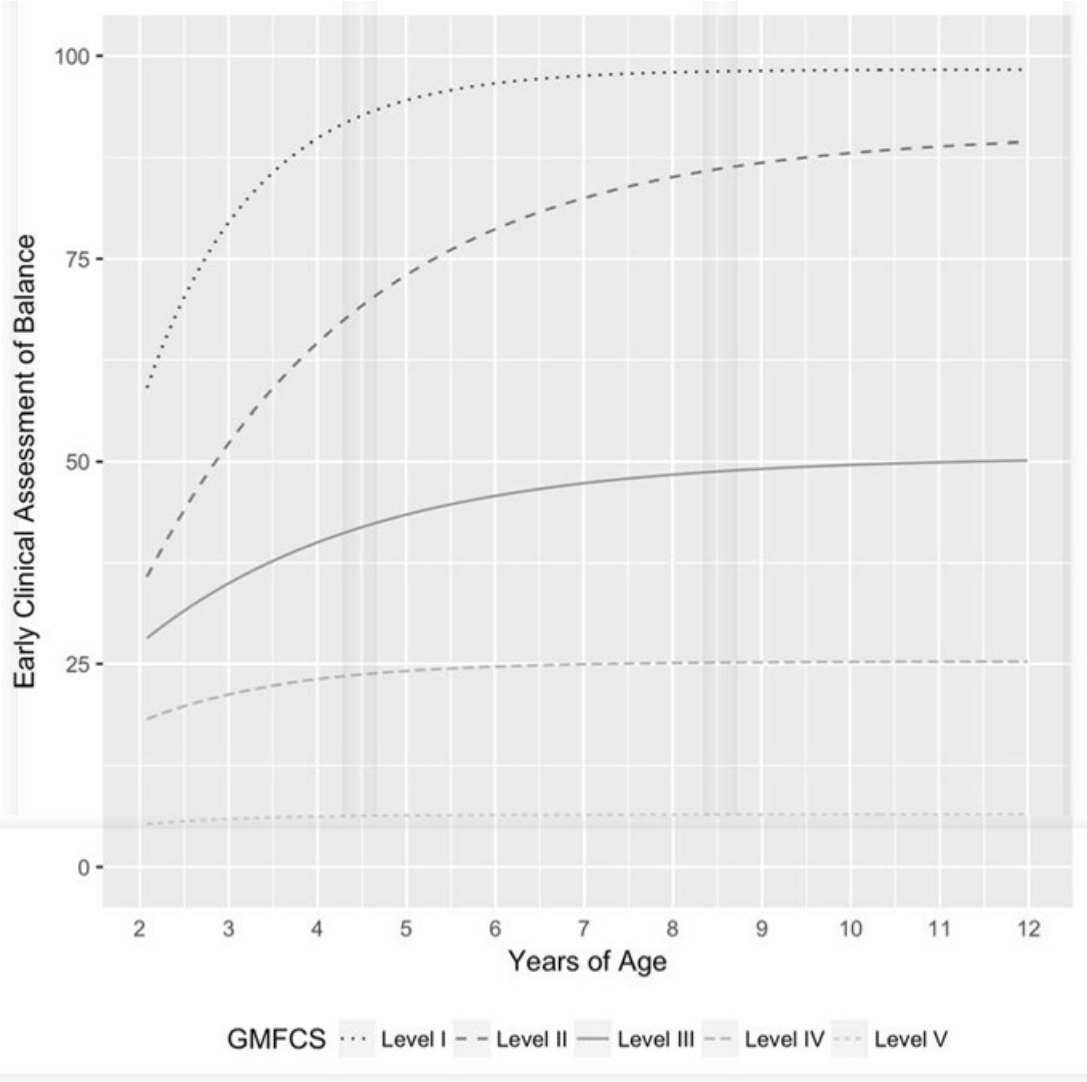
Table 8 shows summary results for all measures within each GMFCS level. Balance, strength for GMFCS Level I and II, endurance on both the measured test (6MWT) and on the parent-reported measure (EASE questionnaire), participation in family/recreation for all GMFCS levels except Level II, and performance in self-care activities all demonstrated nonlinear developmental trajectories. Spinal alignment/range of motion, strength for GMFCS Levels III to V, impact of health conditions, and direct physical activity measures demonstrated linear developmental trajectories. Overall, children with CP who have more functional ability (lower GMFCS levels) develop to a higher level for all measures. The time to reach 90% of development per construct varied and more often did not depend on the children's functional ability, suggesting that the rate of development is not fully dependent on the children's functional ability levels. From an age perspective, overall, children with CP develop more rapidly between 2 to 5 years of age as compared with 5 to 12 years, but this does not hold for the measures noted in which there was linear development.

Table 7. Location of Longitudinal Trajectory Data for Each Measure

	Descriptive data; estimated population value (95% CI) at age 12 y by GMFCS	Longitudinal curve; means (95% CI) at ages 2, 5, and 12 y mean change (95% CI) in ECAB scores at ages 2-5 y and 5-12 y
Balance (ECAB)	CTG document, pages 11-14	Figure 2
Range of motion (SAROMM)	CTG document, pages 14-17	Figure 3
Strength (FSA)	CTG document, pages 17-19	Figure 4
Endurance (6MWT)	CTG document, pages 19-22	Figure 5
Endurance (EASE)	CTG document, pages 19-22	Figure 6
Impact of health conditions (CHCQ)	CTG document, pages 25-28	Figure 7
Participation in family/recreation (CEDL Part I)	CTG document, pages 28-30	Figure 8
Performance in self-care (CEDL Part II)	CTG document, pages 30-31	Figure 9
Amount of walking (StepWatch, average single leg strides/d)	CTG document, pages 46-47	Figure 10
(StepWatch, average strides/d faster than 30/min)	CTG document, pages 44-46	Figure 11
Amount of activity (ActiGraph, average physical activity counts/min)	CTG document, pages 49-51	Figure 12
Intensity of activity (ActiGraph, minutes of moderate to vigorous physical min)	CTG document, pages 47-49	Figure 13

Abbreviations: 6MWT, 6-minute Walk Test; CEDL, Child Engagement in Daily Life; CHCQ, Child Health Conditions Questionnaire; EASE, Early Activity Scale for Endurance; ECAB, Early Clinical Assessment of Balance; FSA, Functional Strength Assessment; GMFCS, GMFCS, Gross Motor Function Classification System; ICC, intraclass correlation coefficient; MVPA, moderate to vigorous physical activity; SAROMM, Spinal Alignment and Range of Motion Measure; TLI, Tucker Lewis Index.

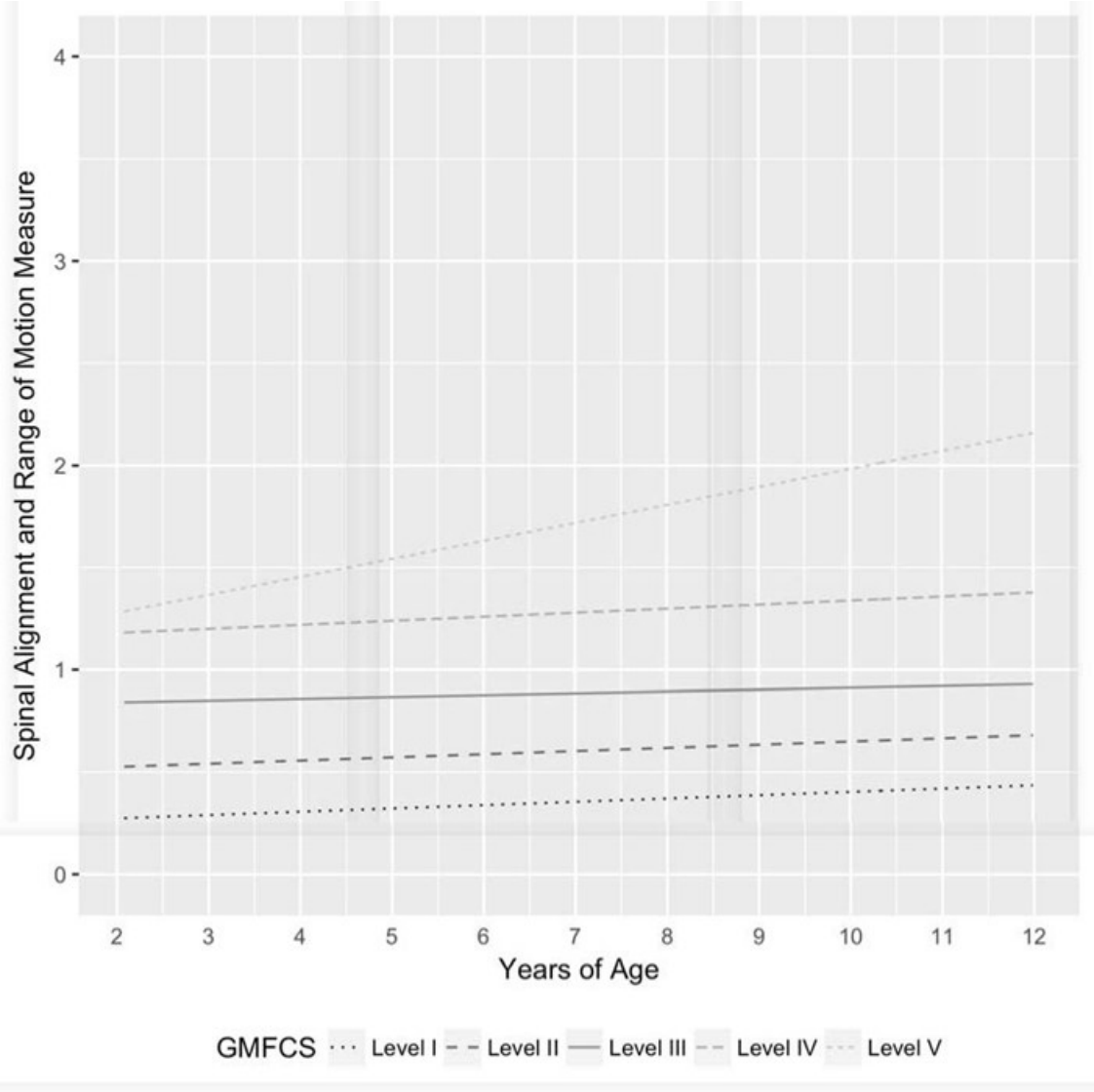
Figure 2. Longitudinal Curves for the Early Clinical Assessment of Balance



	GMFCS Level I Mean (95% CI)	GMFCS Level II Mean (95% CI)	GMFCS Level III Mean (95% CI)	GMFCS Level IV Mean (95% CI)	GMFCS Level V Mean (95% CI)
2 y	54.1 (27.4-71.6)	34.2 (27.9-39.6)	27.4 (24.5-30.5)	17.8 (16.0-19.7)	5.1 (3.8-6.2)
5 y	94.5 (93.4-95.4)	73.0 (71.3-74.7)	43.3 (40.8-45.9)	24.1 (22.6-25.6)	6.3 (5.5-7.1)
12 y	98.3 (97.8-98.6)	89.1 (86.6-91.1)	50.1 (46.5-53.6)	25.3 (23.7-27.0)	6.5 (5.7-7.4)
Change 2-5 y	40.4 (23.2-66.7)	38.8 (32.7-45.5)	15.9 (13.6-17.7)	6.2 (4.8-7.6)	1.2 (0.3-2.1)
Change 5-12 y	3.8 (2.9-4.9)	16.2 (12.5-19.1)	6.8 (4.0-9.9)	1.2 (0.5-2.2)	0.2 (0.0-0.8)

Abbreviation: GMFCS, Gross Motor Function Classification System.

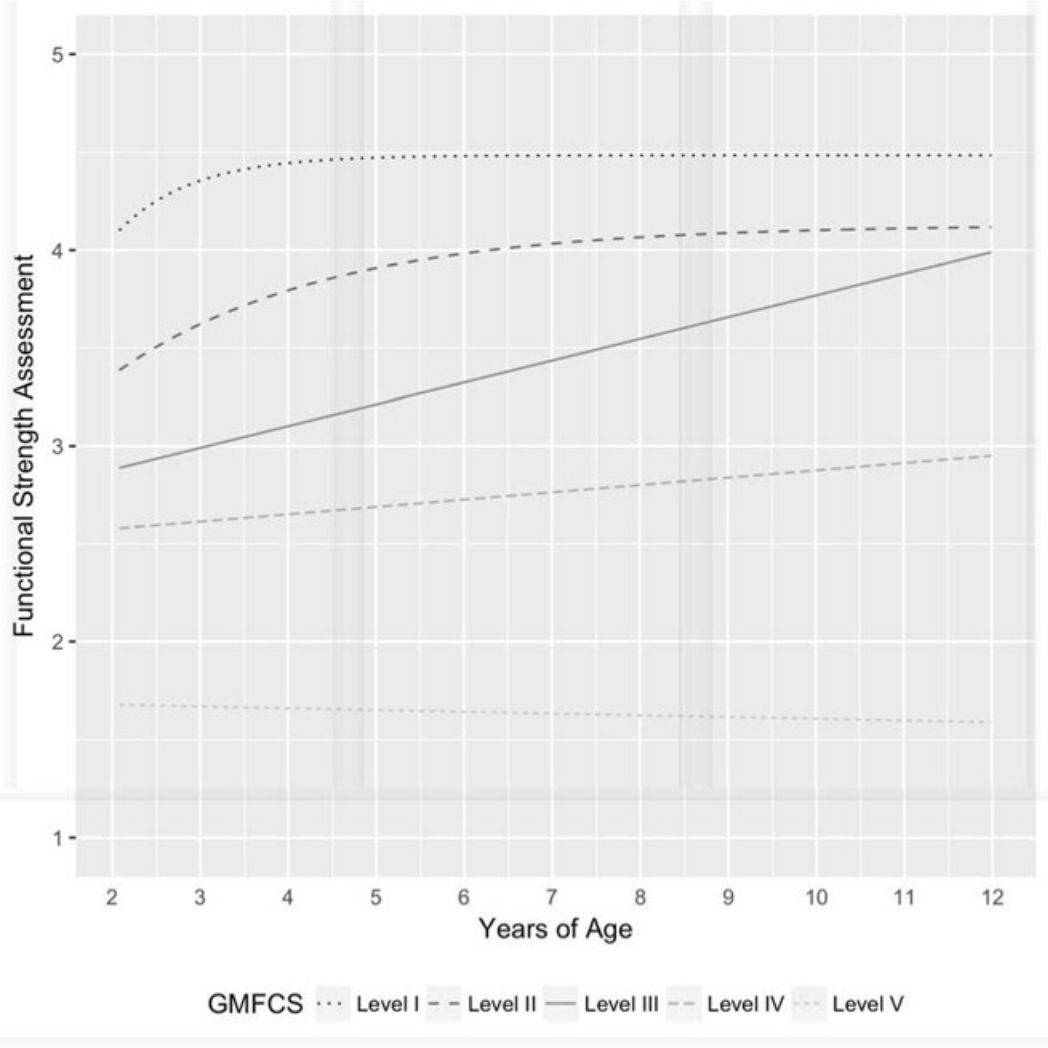
Figure 3. Longitudinal Curves for the Spinal Alignment and Range of Motion Measure



	GMFCS Level I Mean (95% CI)	GMFCS Level II Mean (95% CI)	GMFCS Level III Mean (95% CI)	GMFCS Level IV Mean (95% CI)	GMFCS Level V Mean (95% CI)
2 y	0.3 (0.2-0.3)	0.5 (0.4-0.6)	0.8 (0.7-1.0)	1.2 (1.1-1.3)	1.3 (1.1-1.4)
5 y	0.3 (0.3-0.3)	0.6 (0.5-0.6)	0.9 (0.8-0.9)	1.2 (1.2-1.3)	1.5 (1.4-1.6)
12 y	0.4 (0.4-0.5)	0.7 (0.6-0.8)	0.9 (0.8-1.1)	1.4 (1.2-1.5)	2.2 (2.0-2.3)
Change 2-5 y	0.0 (0.0-0.1)	0.0 (0.0-0.1)	0.0 (0.0-0.1)	0.1 (0.0-0.1)	0.3 (0.2-0.3)
Change 5-12 y	0.1 (0.1-0.2)	0.1 (0.0-0.2)	0.1 (–0.1 to 0.2)	0.1 (0.0-0.3)	0.6 (0.5-0.8)

Abbreviation: GMFCS, Gross Motor Function Classification System.

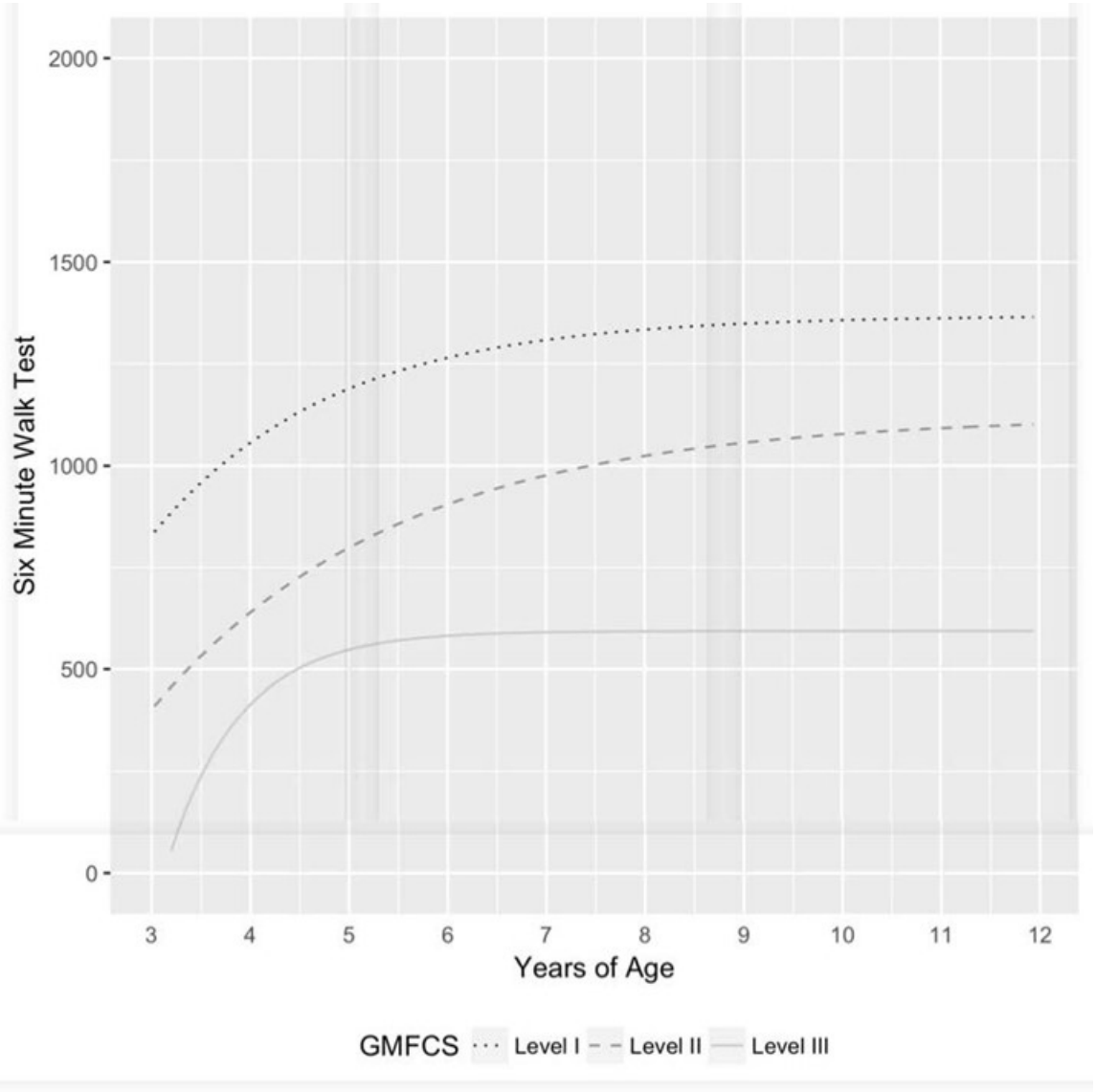
Figure 4. Longitudinal Curves for the Functional Strength Assessment



	GMFCS Level I Mean (95% CI)	GMFCS Level II Mean (95% CI)	GMFCS Level III Mean (95% CI)	GMFCS Level IV Mean (95% CI)	GMFCS Level V Mean (95% CI)
2 y	4.1 (3.9-4.2)	3.3 (2.9-3.5)	2.9 (2.7-3.1)	2.6 (2.4-2.7)	1.7 (1.5-1.8)
5 y	4.5 (4.4-4.5)	3.9 (3.8-4.0)	3.2 (3.1-3.3)	2.7 (2.6-2.8)	1.7 (1.6-1.8)
12 y	4.5 (4.4-4.5)	4.1 (4.0-4.2)	4.0 (3.7-4.3)	3.0 (2.7-3.2)	1.6 (1.4-1.8)
Change 2-5 y	0.4 (0.3-0.5)	0.6 (0.4-1.0)	0.3 (0.2-0.4)	0.1 (0.0-0.2)	0.0 (–0.1-0.1)
Change 5-12 y	0.0 (0.0-0.0)	0.2 (0.1-0.4)	0.8 (0.5-1.0)	0.3 (0.0-0.5)	–0.1 (–0.3 to 0.1)

Abbreviation: GMFCS, Gross Motor Function Classification System.

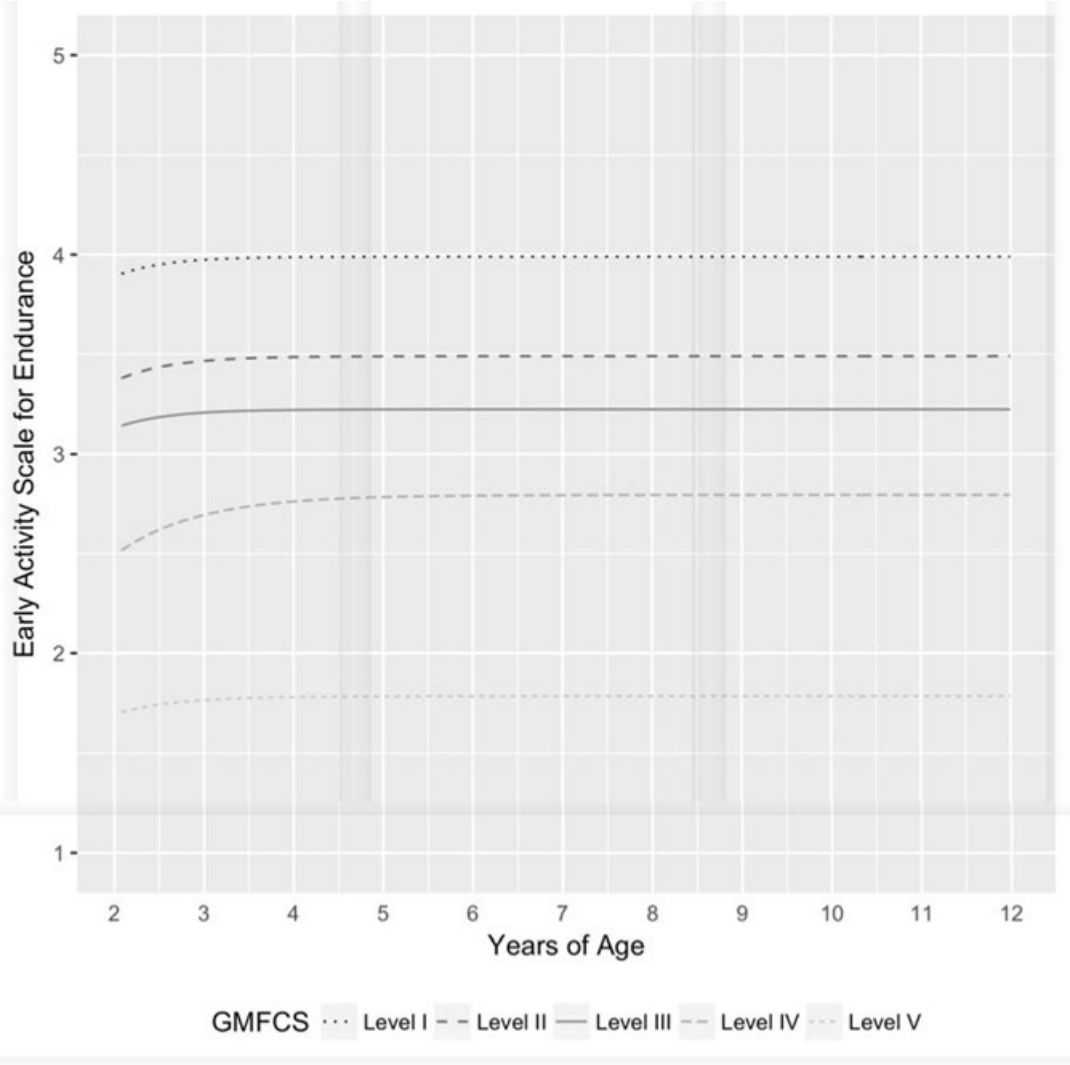
Figure 5. Longitudinal Curves for the 6-Minute Walk Test



	GMFCS Level I Mean (95% CI)	**GMFCS Level II Mean (95% CI)**	**GMFCS Level III Mean (95% CI)**
2 y	389 (77-581)	7 (0-208)	N/A
5 y	1184 (1142-1223)	793 (744-840)	540 (479-598)
12 y	1362 (1314-1411)	1096 (1029-1159)	593 (534-653)
Change 2-5 y	794 (574-1137)	786 (562-1132)	N/A
Change 5-12 y	179 (120-244)	303 (218-387)	53 (10-116)

Abbreviations: GMFCS, Gross Motor Function Classification System; N/A, not applicable.

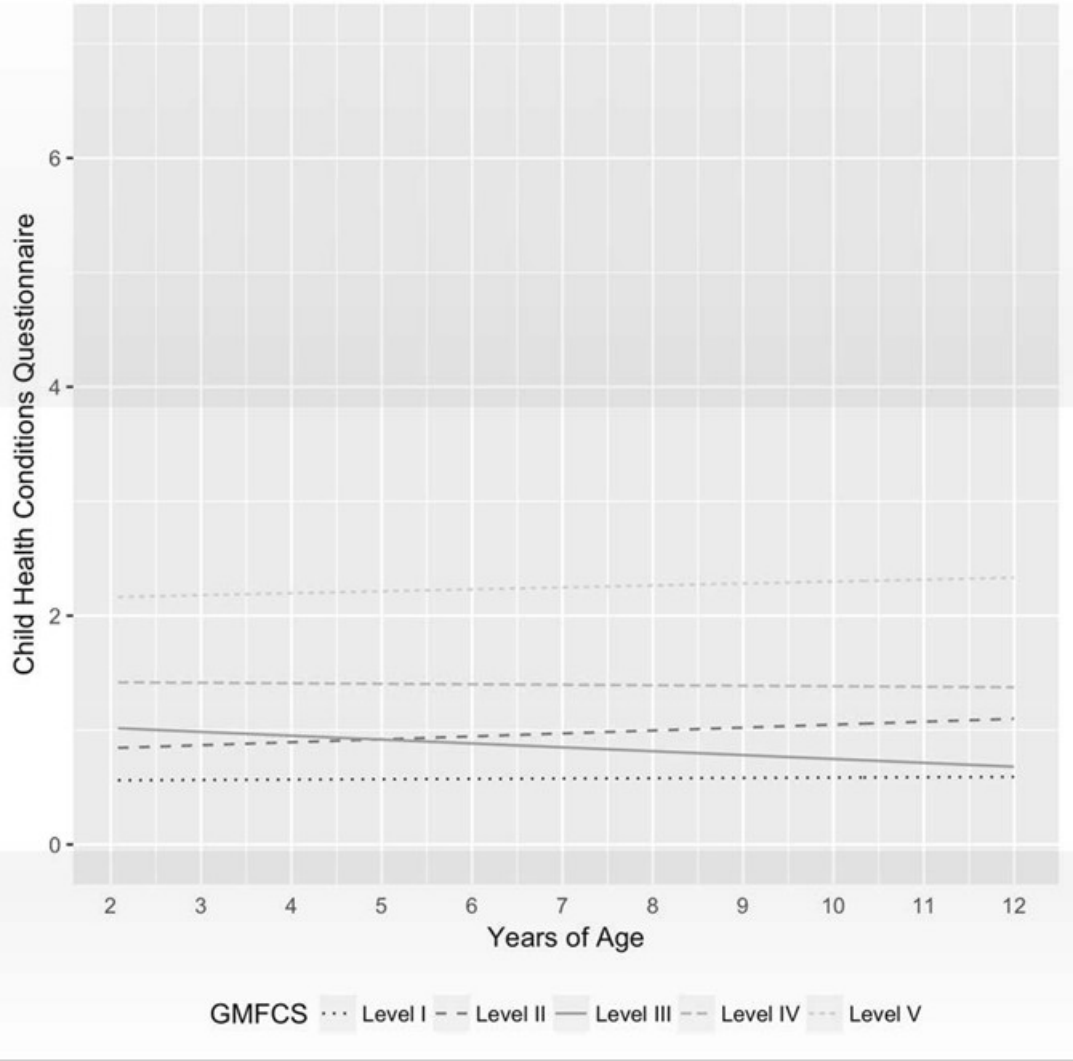
Figure 6. Longitudinal Curves for the Early Activity Scale for Endurance



	GMFCS Level I Mean (95% CI)	GMFCS Level II Mean (95% CI)	GMFCS Level III Mean (95% CI)	GMFCS Level IV Mean (95% CI)	GMFCS Level V Mean (95% CI)
2 y	3.9 (3.6-4.0)	3.3 (3.0-3.5)	3.0 (2.6-3.3)	2.5 (2.2-2.7)	1.6 (1.3-1.8)
5 y	4.0 (3.9-4.1)	3.5 (3.4-3.6)	3.2 (3.1-3.4)	2.8 (2.7-2.9)	1.8 (1.7-1.9)
12 y	4.0 (3.9-4.1)	3.5 (3.4-3.6)	3.2 (3.1-3.4)	2.8 (2.7-2.9)	1.8 (1.7-1.9)
Change 2-5 y	0.1 (0.0-0.3)	0.2 (0.0-0.5)	0.2 (0.0-0.6)	0.3 (0.1-0.6)	0.1 (0.0-0.4)
Change 5-12 y	0.0 (0.0-0.0)	0.0 (0.0-0.0)	0.0 (0.0-0.0)	0.0 (0.0-0.1)	0.0 (0.0-0.1)

Abbreviation: GMFCS, Gross Motor Function Classification System.

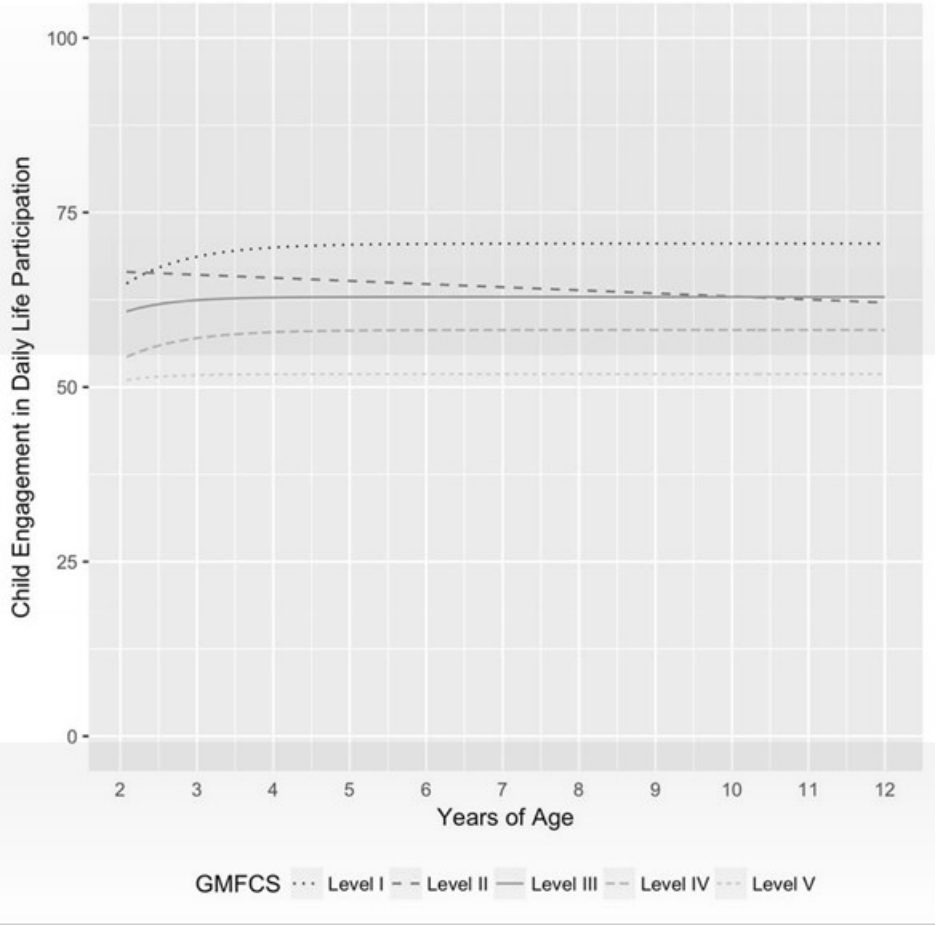
Figure 7. Longitudinal Curves for Child Health Conditions Questionnaire



	GMFCS Level I Mean (95% CI)	GMFCS Level II Mean (95% CI)	GMFCS Level III Mean (95% CI)	GMFCS Level IV Mean (95% CI)	GMFCS Level V Mean (95% CI)
2 y	0.6 (0.5-0.7)	0.8 (0.7-1.0)	1.0 (0.8-1.2)	1.4 (1.2-1.6)	2.2 (1.9-2.4)
5 y	0.6 (0.5-0.6)	0.9 (0.8-1.0)	0.9 (0.8-1.0)	1.4 (1.3-1.5)	2.2 (2.1-2.4)
12 y	0.6 (0.5-0.7)	1.1 (0.9-1.3)	0.7 (0.5-0.9)	1.4 (1.1-1.6)	2.3 (2.1-2.6)
Change 2-5 y	0.0 (0.0-0.1)	0.1 (0.0-0.2)	–0.1 (–0.2 to 0.0)	0.0 (–0.1 to 0.1)	0.1 (–0.1 to 0.2)
Change 5-12 y	0.0 (–0.1 to 0.2)	0.2 (0.0-0.4)	–0.2 (–0.5 to 0.0)	0.0 (–0.3 to 0.2)	0.1 (–0.1 to 0.4)

Abbreviation: GMFCS, Gross Motor Function Classification System.

Figure 8. Longitudinal Curves for the Child Engagement in Daily Life Participation

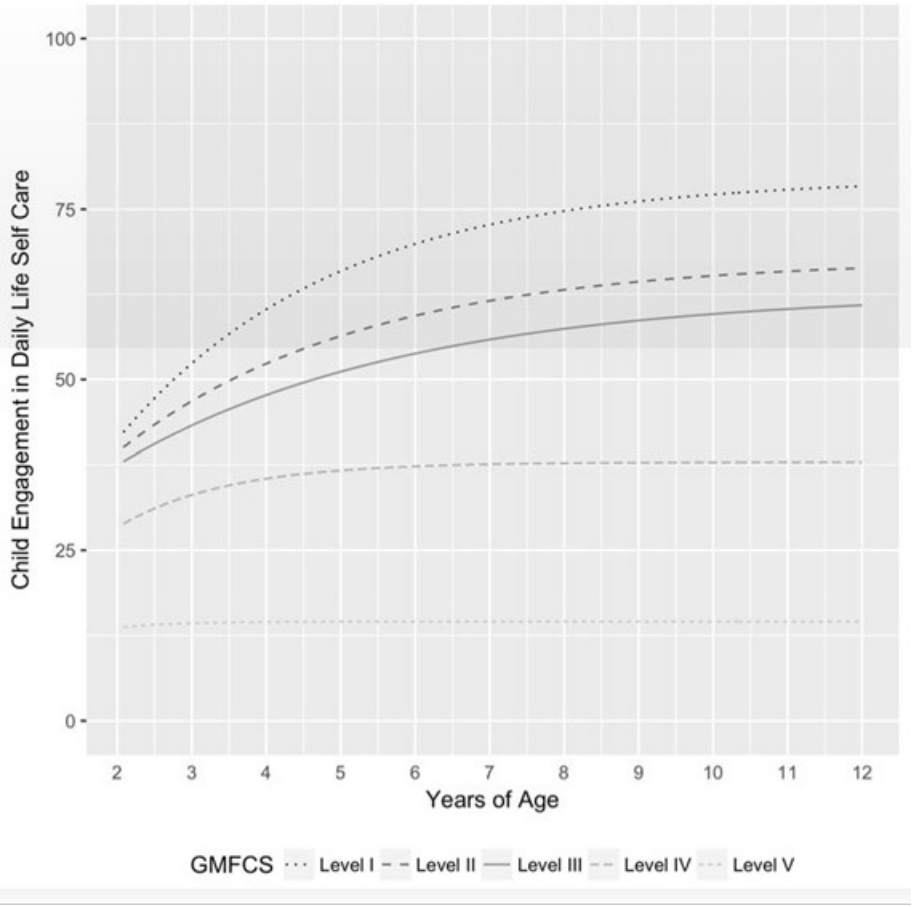


Participation in Family and Recreation (CEDL Part 1)

	GMFCS Level I Mean (95% CI)	**GMFCS Level II Mean (95% CI)**	**GMFCS Level III Mean (95% CI)**	**GMFCS Level IV Mean (95% CI)**	**GMFCS Level V Mean (95% CI)**
2 y	64.0 (60.7, 66.9)	66.5 (63.8, 69.2)	59.6 (54.0, 63.3)	53.4 (49.1, 56.7)	49.6 (43.6, 52.8)
5 y	70.3 (68.9, 71.8)	65.2 (63.5, 66.9)	62.8 (60.8, 64.9)	58.0 (56.6, 59.5)	51.8 (50.1, 53.6)
12 y	70.6 (69.1, 72.0)	62.1 (59.4, 65.0)	62.9 (60.9, 65.0)	58.2 (56.7, 59.7)	51.9 (50.2, 53.6)
Change 2-5 y	6.3 (3.5, 9.5)	–1.3 (–2.8, 0.1)	3.2 (0.3, 8.5)	4.6 (1.4, 8.8)	2.1 (0.0, 7.9)
Change 5-12 y	0.2 (0.0, 0.5)	–3.1 (–6.4, 0.3)	0.1 (0.0, 0.5)	0.2 (0.0, 0.6)	0.1 (0.0, 0.5)

Abbreviations: CEDL, Child Engagement in Daily Life; GMFCS, Gross Motor Function Classification System.

Figure 9. Longitudinal Curves for the Child Engagement in Daily Life Self-care

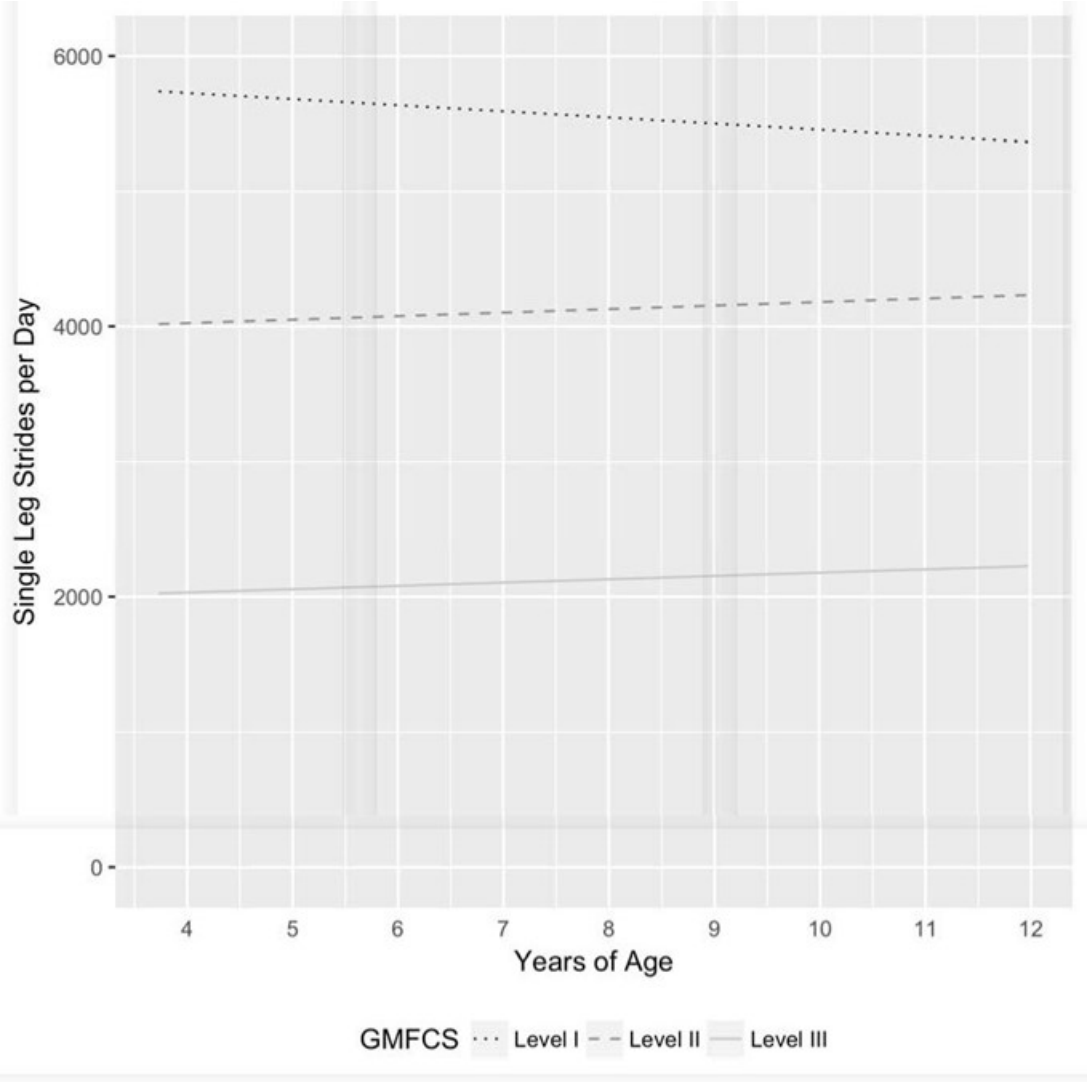


Participation in Self-care Activities (CEDL Part 2)

	GMFCS Level I Mean	**GMFCS Level II Mean**	**GMFCS Level III Mean**	**GMFCS Level IV Mean**	**GMFCS Level V Mean**
2 y	41.0	38.9	36.9	28.3	12.5
	GMFCS	GMFCS	GMFCS	GMFCS	GMFCS
	(38.8-42.9)	(35.4-41.7)	(33.6-39.7)	(25.3-31.4)	(7.3-15.7)
5 y	65.8	56.2	50.8	36.6	14.1
12 y	78.3	66.1	60.5	37.9	14.5
Change	24.8	17.3	13.9	8.3	1.6
2-5 y	(22.7-27.2)	(14.5-20.9)	(11.5-17.2)	(5.7-10.8)	(0.0-4.8)
Change	12.5	10.0	9.7	1.3	0.4
5-12 y	(10.0-14.8)	(7.2-12.7)	(6.5-12.8)	(0.4-2.6)	(0.0-2.6)

Abbreviations: CEDL, Child Engagement in Daily Life; GMFCS, Gross Motor Function Classification System.

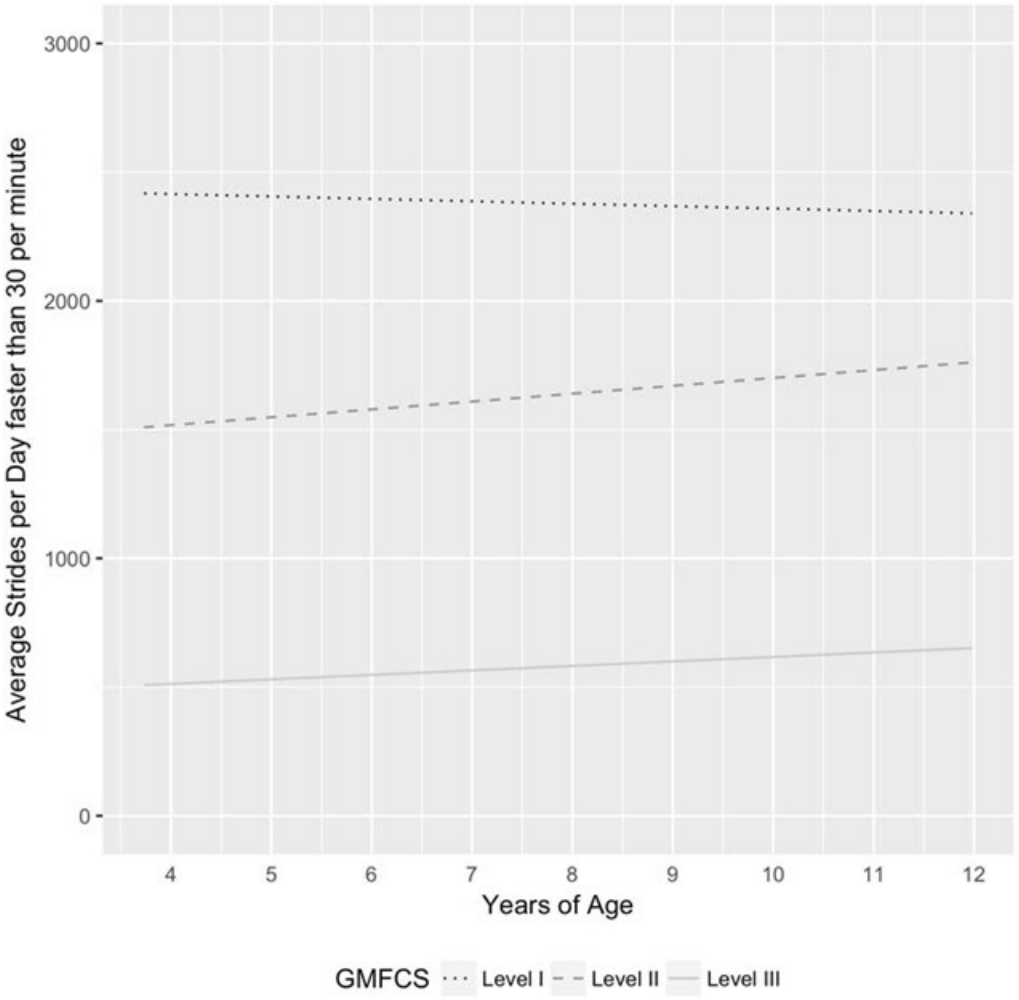
Figure 10. Longitudinal Curves for Average Single Leg Strides Per Day



	GMFCS Level I Mean (95% CI)	**GMFCS Level II Mean (95% CI)**	**GMFCS Level III Mean (95% CI)**
2 y	5685 (4849-6511)	4054 (3374-4723)	2062 (909-3198)
5 y	5552 (5089-6002)	4133 (3757-4500)	2142 (1197-3063)
12 y	5240 (3874-6670)	4319 (3258-5443)	2329 (189-4468)
Change 2-5 y	–133 (–718 to 486)	80 (–387 to 578)	80 (–703 to 900)
Change 5-12 y	–311 (–1676 to 1133)	186 (–904 to 1348)	187 (–1641 to 2099)

Abbreviation: GMFCS, Gross Motor Function Classification System.

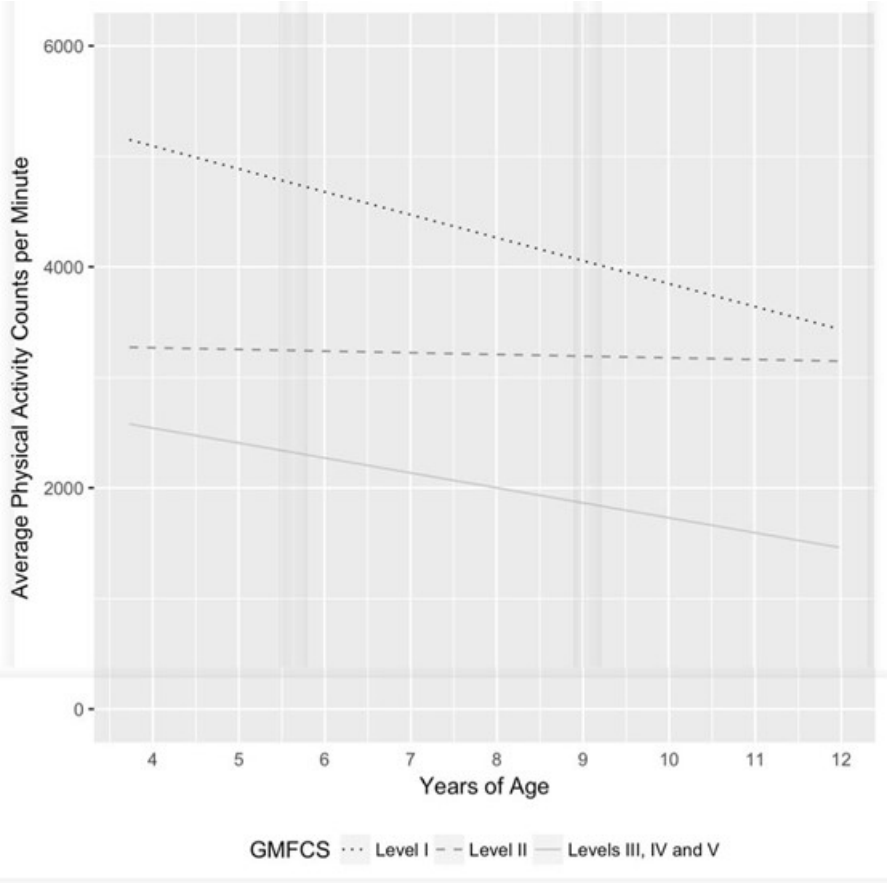
Figure 11. Longitudinal Curves for Average Strides Per Day Faster Than 30 Per Minute



	GMFCS Level I Mean (95% CI)	**GMFCS Level II Mean (95% CI)**	**GMFCS Level III Mean (95% CI)**
2 y	2408 (1890-2924)	1550 (1181-1915)	531 (145-913)
5 y	2381 (2098-2657)	1642 (1463-1816)	586 (314-851)
12 y	2319 (1460-3218)	1858 (1233-2530)	715 (0-1543)
Change	–27	93	55
2-5 y	(–396 to 363)	(–183 to 386)	(–267 to 386)
Change	–62	216	129
5-12 y	(–923 to 848)	(–426 to 902)	(–623 to 900)

Abbreviation: GMFCS, Gross Motor Function Classification System.

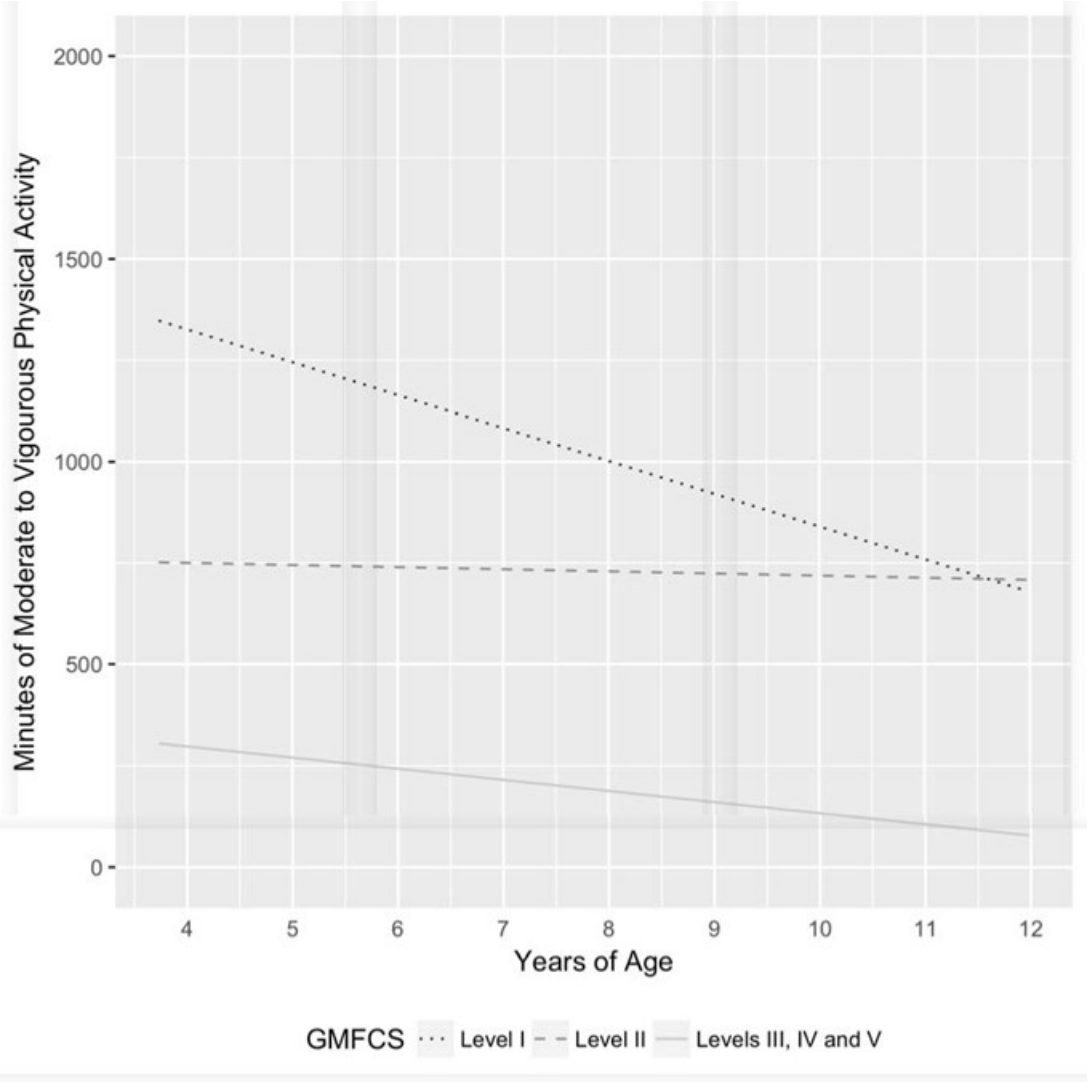
Figure 12. Longitudinal Curves for Average Physical Activity Counts Per Minute



	GMFCS Level I Mean (95% CI)	**GMFCS Level II Mean (95% CI)**	**GMFCS l III, IV, and V Mean (95% CI)**
2 y	4890 (4447-5327)	3259 (2791-3716)	2409 (2095-2718)
5 y	4268 (4058-4471)	3214 (2922-3499)	2004 (1789-2213)
12 y	2815 (2025-3650)	3109 (2502-3739)	1059 (471-1665)
Change 2-5 y	–623 (–957 to –267)	–45 (–315 to 240)	–405 (–631 to –164)
Change 5-12 y	–1453 (–2233 to –624)	–105 (–735 to 561)	–945 (–1472 to –382)

Abbreviation: GMFCS, Gross Motor Function Classification System.

Figure 13. Longitudinal Curves for Minutes of Moderate to Vigorous Physical Activity



	GMFCS Level I Mean (95% CI)	GMFCS Level II Mean (95% CI)	GMFCS Level III, IV, and V Mean (95% CI)
2 y	1246 (1048-1443)	747 (480-1005)	271 (198-343)
5 y	1004 (895-1109)	731 (577-882)	189 (145-231)
12 y	438 (110-781)	696 (311-1102)	N/A
Change 2-5 y	–242 (–383 to –93)	–15 (–187 to 165)	–82 (–140 to –20)
Change 5-12 y	–565 (–894 to –218)	–36 (–435 to 385)	N/A

Abbreviations: GMFCS, Gross Motor Function Classification System; N/A, not applicable.

Table 8. Developmental Trajectories Comparisons Among GMFCS Levels Related to Level of Improvement at 12 Years of Age, Time (Number of Months) to Attain 90% of Maximum Score, and Percentage of Improvement at 2-5 Years of Age Compared With 5-12 Years of Age for All Measures

Measure	Estimated population value (95% CI) at 12 y of age					No. of mo required to attain 90% of maximum score[a]					% of total mean score change occurring 2-5 y of age compared with 5-12 y				
GMFCS level	**I**	**II**	**III**	**IV**	**V**	**I**	**II**	**III**	**IV**	**V**	**I**	**II**	**III**	**IV**	**V**
ECAB (N = 708)	98.3 (97.8, 98.6)	89.1 (86.6, 91.1)	50.1 (46.5, 53.6)	25.3 (23.7, 27.0)	6.48 (5.66, 7.38)	35	71	71	45	34	91	71	70	84	86
SAROMM – limitation (N = 708)	0.44 (0.38, 0.49)	0.68 (0.60, 0.77)	0.93 (0.79, 1.07)	1.38 (1.22, 1.54)	2.16 (2.01, 2.31)	NP □	NP □	NP □	NP □	NP □	0	0	0	50	33
FSA (N = 708)	4.49 (4.43, 4.54)	4.10 (3.99, 4.20)	3.99 (3.75, 4.25)	2.95 (2.73, 3.19)	1.59 (1.44, 1.75)	23	67	NP □	NP □	NP □	100	75	27	25	0
6MWT (only GMFCS Levels I-III) (N = 408)	1362.3 (1313.6, 1410.7)	1096.33 (1028.80, 1158.62)	592.62 (533.79, 652.72)			50	69	20			82	72	99		
EASE (N = 708)	3.99 (3 91	3.49 (3 39	3.23 (3 08	2.80 (2 67	1.79 (1 67						100	100	100	100	100
CHCQ – impact (N = 708)	0.59 (0 47	1.10 (0 91	0.68 (0 48	1.38 (1 12	2.33 (2 08	NP □	NP □	NP □	NP □	NP □	NC	33	33	NC	50
CEDL – family/recreation (N	70.6 (69 1	62.1 (59 4	62.9 (60 9	58.2 (56 7	51.9 (50 2	23	NP □				97	30	97	96	95

Measure	Estimated population value (95% CI) at 12 y of age					No. of mo required to attain 90% of maximum score[a]					% of total mean score change occurring 2-5 y of age compared with 5-12 y				
GMFCS level	**I**	**II**	**III**	**IV**	**V**	**I**	**II**	**III**	**IV**	**V**	**I**	**II**	**III**	**IV**	**V**
CEDL – self-care (N = 708)	78.3 (76.0	66.1 (63.5	60.5 (57.3	37.9 (35.6	14.5 (12.4						66	63	59	86	80
SW –intensity (N = 46)	2319.0 (1459.9	1858.2 (1233.0	714.7 (–103.1			NP □	NP □	NP □			30	30	30		
SW – amount (N = 46)	5240.4 (3874.3	4318.8 (3258.1	2328.7 (189.4			NP □	NP □	NP □			30	30	30		
ActiGraph – intensity (GMFCS Levels II-V combined) (N = 72)	438.3 (110.5, 781.4)	695.6 (310.7, 1102.3)	–2.9 (–149.2, 147.1)			NP ↓	NP ↓	NP ↓			30		29		30
ActiGraph – amount (N = 72)	2815.0 (2024.7, 3649.7)	3109.0 (2501.9, 3739.0)	1058.7 (470.6, 1665.2)			NP ↓	NP ↓	NP ↓			30		30		30

Abbreviations: GMFCS, Gross Motor Function Classification System; NC, no change; NP, no plateau.

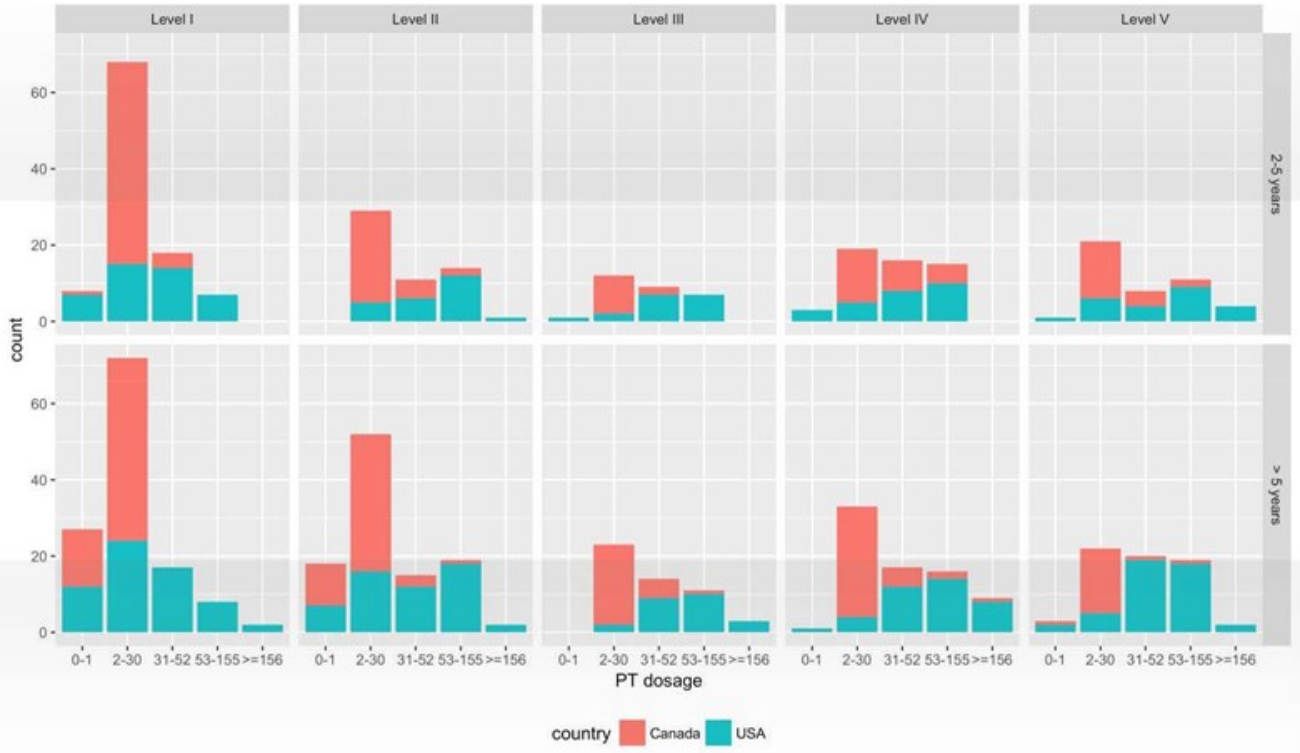
[a]Arrows = direction of the linear trajectory from 1.5 to 12 years of age.

AIM 2: SERVICES RELATIONSHIPS TO OUTCOMES

Services Description

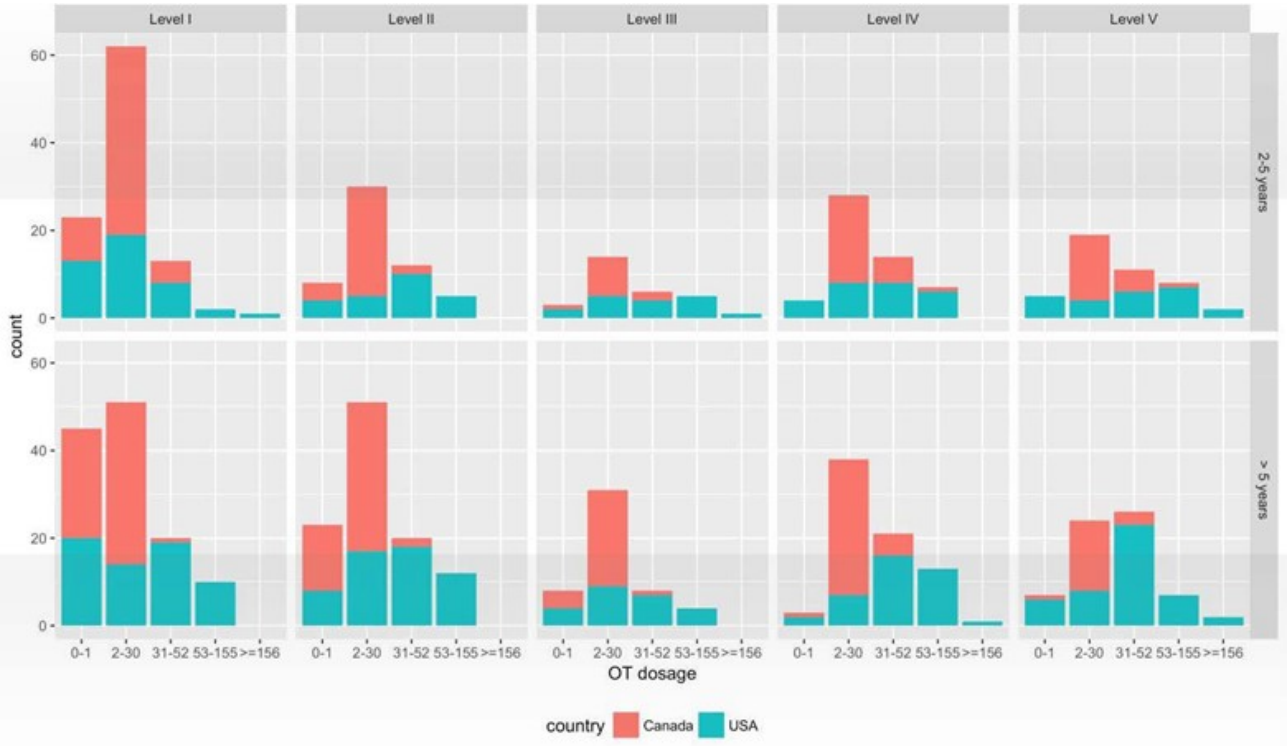
The number of service sessions per year reported by families, detailed by children's functional ability (GMFCS) and assessment session in which the services were reported across 24 months, is found in the CTG document, pages 31 to 38. The descriptive data reported in these tables reflect categorized service amounts as follows: 1 = 0 to 1 sessions, 2 = 2 to 30 sessions, 3 = 31 to 52 sessions, 4 = 53 to 155 sessions, 5 = ≥156 sessions per year. Figures 14 to 16 depict the amount of services for PT, OT, and ST, respectively, that parents reported for their children by country, age, and GMFCS level. Overall, the amount of service sessions per year for all 3 therapies provided to the children residing in Canada was lower (primarily 0-1 or 2-30 sessions per year) as compared with the United States, where the number of services was primarily 2 to 30 to 53 to 155 sessions per year. The amount of service sessions for 3 therapies increased as the children's functional limitations increased; however, ST services were provided at a lower level than PT and OT. The distribution of amount of service sessions for all 3 therapies based on age of the children (2-5 years of age as compared with >5-12 years of age) suggests that service amounts increase as children age, except for children with the highest functional ability (GMFCS Levels I and II), who received 0 to 1 sessions per year more often from >5 to 12 years of age.

Figure 14. Amount of PT Sessions Per Year by GMFCS Classification, Country, and Age (2-5 Years vs >5-12 Years)



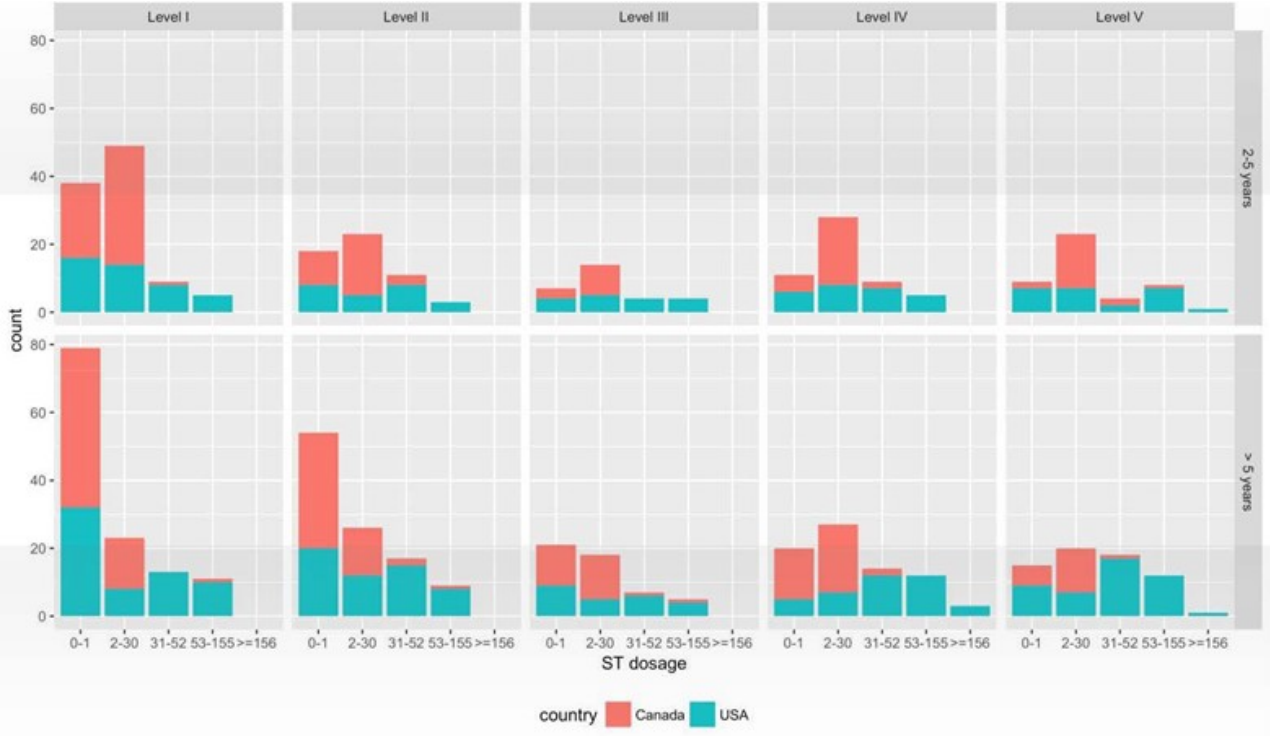
Abbreviations: GMFCS, Gross Motor Function Classification System; PT, physical therapy.

Figure 15. Amount of OT Sessions per Year by GMFCS Classification, Country, and Age (2-5 Years vs >5-12 Years)



Abbreviations: GMFCS, Gross Motor Function Classification System; OT, occupational therapy.

Figure 16. Amount of ST Sessions Per Year by GMFCS Classification, Country, and Age (2-5 Years vs >5-12 Years)



Abbreviations: GMFCS, Gross Motor Function Classification System; ST, speech therapy.

The focus of therapy was rated on a scale of 1 to 5, with 1 = not at all and 5 = to a very great extent. The lowest focus of therapy was reported to be on self-care activities, with ratings at approximately a "small extent" (mean, 1.8-2.9). The highest focus of therapy ratings, at approximately a "moderate to great extent," were for the secondary body structure/function domain (stretching tight muscles, strengthening muscles, and/or activities to increase fitness) (mean, 3.7-4.1); primary body structure/function domain (relaxation of spastic muscles, physically guiding movement of parts of the body [eg, head, neck, and trunk; lips and mouth; arms and legs], and/or balance activities in any position) (mean, 3.3-4.1); and a focus on self-initiation (activities to improve self-initiated abilities to transfer from one position to another, to move from one place to another, to use the hands in daily life activities, and/or to use the voice to make speech sounds or the hands to make gestures using sign language to communicate) (mean, 3.1-4.2). Parent ratings of the extent to which services were family-centered were similar across children's GMFCS levels and clustered around the "moderate

extent" level (mean, 3.2-3.5). Ratings of the extent to which parents believed their child's needs were being met were at a "moderate to great extent" for children at GMFCS Level I (mean, 3.7-3.8) as compared with approximately a "moderate extent" for children at GMFCS Level V (mean, 3.1-3.3).

Services Relationship to Outcomes

For the analyses of the relationships of the 13 selected service variables to the 4 selected outcomes generated by the GMFCS level-specific longitudinal trajectories, we tested 234 models; of those, 10 models were significantly ($P < .05$) improved. There was a statistically significant effect of the extent to which parents perceived their child's needs were being met by services for balance for children at GMFCS Level I (Table 9 and Figure 17). This effect was significant based only on 95% CIs at 2 years of age and showed that when parents indicated that their child's needs were met to a greater extent, outcomes were higher. There were no significant relationships of services to the 6MWT outcome. Analyses revealed 3 other effects of services for the outcome of participation in family/recreation for children at GMFCS Level II—a focus on health and well-being, self-care performance, and the extent to which parents perceived their child's needs were being met by services. But none of the service variables effected a meaningful difference in the CEDL scores as assessed with the 95% CIs (Table 10). For the outcome of performance of self-care activities for children at GMFCS Level I, 4 other service variables yielded an exploratory result—a focus on self-care activities, self-initiation, structured play and recreation activities, and the extent to which parents perceived their child's needs were being met by services. Again, none of the service variables effected a meaningful difference in the CEDL scores as assessed with the 95% CIs (Table 11). (Note that focus on "structured play and recreation activities" is included because of the *P* value for the effect of this therapy focus on the model limit but including it does not significantly improve model fit as assessed with the LRT.) PT, OT, and ST amounts of service were also significant for the outcome of participation in self-care activities for children at GMFCS Level I (Table 12). At each of the ages (2, 5, and 12 years), however, the 95% CIs for those receiving the least amount of services overlapped with the 95% CIs for those receiving the most services.

Table 9. Extent to Which Parents Perceived Their Child's Needs Were Being Met by Services Effect on Balance for Children at GMFCS Level I (Age Was Centered at 60 months [5 years]; Needs Met Scored on the Range of 1-5)

Early Clinical Assessment of Balance GMFCS Level I[a]

	Value	SE	*df*	*T* value	*P* value
Transformed limit[b]	4.12	1.46	643	2.81	.005
Needs met on transformed limit	0.07	0.41	643	0.16	.870
Log rate	–2.53	0.16	643	–15.44	.000
Needs met on log rate	–0.10	0.04	643	–2.23	.026
Offset	2.52	0.06	643	40.37	.000

Abbreviation: GMFCS, Gross Motor Function Classification System.
[a]Likelihood ratio test for adding needs met = 781.1 on 2 *df*; *P* value < .00001.
[b]Transformed limit = –ln(100/limit – 1).

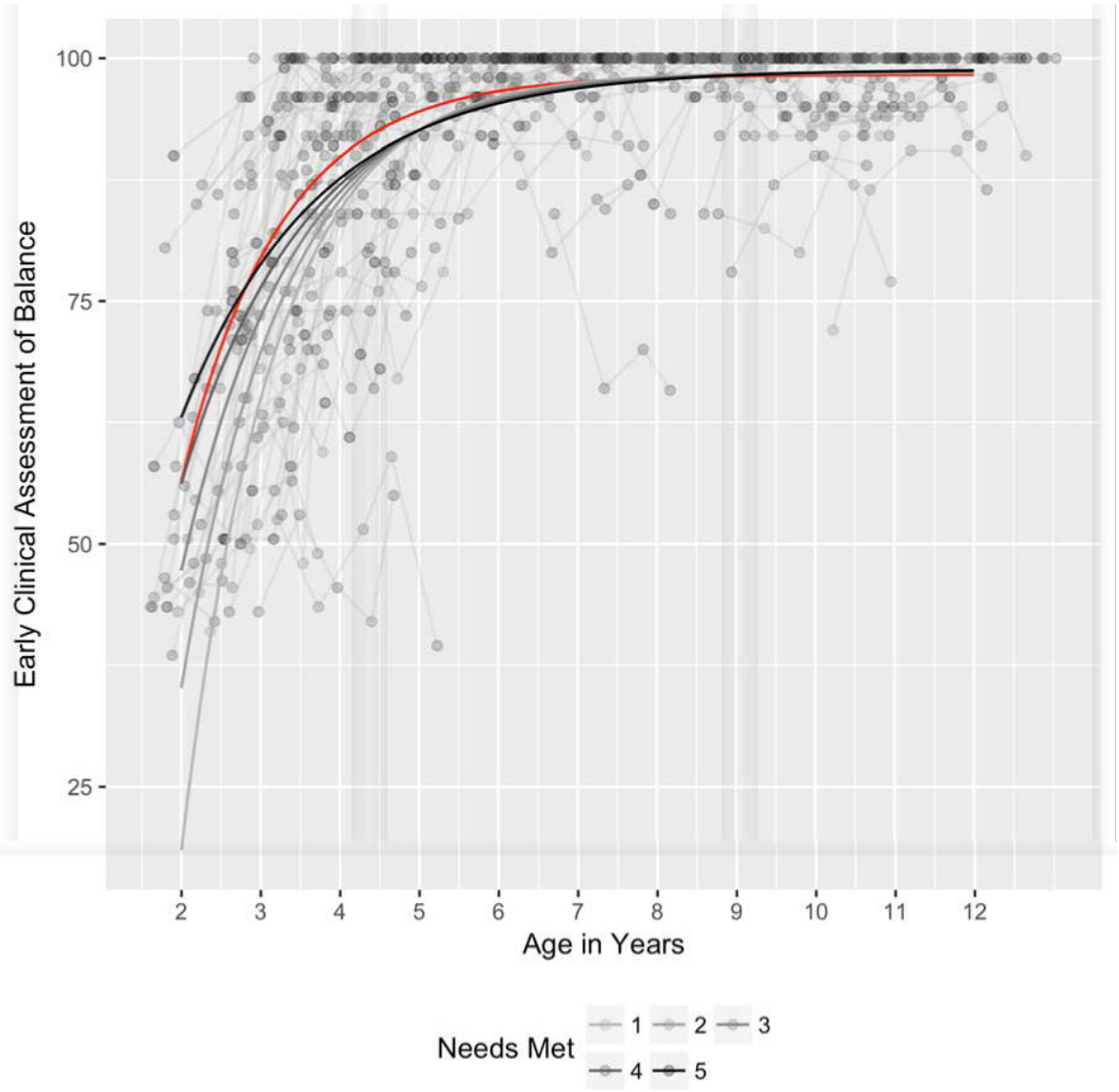
Population Predictions

Age, y	Not at all	To a small extent	To a moderate extent	To a great extent	To a very great extent
2	18.5	35.1	47.3	56.2	63.0
5	92.6	92.6	92.6	92.6	92.6
8		98.6	98.6	98.7	98.7

Age, y	Not at all, 95% CI	Completely, 95% CI
2[a]	(11.4-25.6)	(55.9-70.1)
5	(85.5-99.7)	(85.5-99.7)
12	(91.4-100)	(91.6-100)

[a]Confidence intervals do not overlap.

Figure 17. Growth Model Plots With Service Variables: Early Clinical Assessment of Balance GMFCS Level I



Abbreviation: GMFCS, Gross Motor Function Classification System.
Note: Red line = the predicted developmental trajectory.

Table 10. Focus on Health and Well-being, Self-care Activities, and the Extent to Which Parents Perceived Their Child's Needs Were Being Met by Services Effects on Participation in Family/Recreation Activities for Children at GMFCS Level II

Focus on Health[a]

	Value	SE	*df*	*T* value	*P* value
Intercept	63.20	1.40	447	45.26	.000
Focus on health	0.91	0.42	447	2.17	.031
Age, mo	–0.03	0.03	447	–0.90	.369
Focus on health: age, mo	0.00	0.01	447	–0.08	.939

Abbreviation: GMFCS, Gross Motor Function Classification System.
[a]Likelihood ratio test for adding focus on health = 8 on 2 *df*; *P* value = .01830. Age was centered at 60 months [5 years] and service variables scored with the lowest level at 0; intercept is the value of the outcome at 5 years of age for the lowest level of the service variables.

Population Predictions

Population predictions		To a small extent	To a moderate extent	To a great extent	To a very great extent
2	64.3	65.2	66.1	67.1	68.0
5	63.2	64.1	65.0	65.9	66.8
12	.7	61.6	62.4	63.3	64.1

95% Confidence Bounds

Age (Years)	Not at All	To a Very Great Extent
2	(56.5, 72.1)	(59.2, 76.8)
5	(55.6, 70.8)	(58.6, 75.0)
12	(53.4, 68.0)	(56.3, 71.9)

Focus on Self-care

	Value	SE	*df*	*T* value	*P* value
Intercept	63.28	1.16	447	54.46	.000
Focus on self-care	1.32	0.41	447	3.26	.001
Age (months)	–0.01	0.03	447	–0.32	.752
Focus on self-care: age (months)	–0.02	0.01	447	–2.00	.046

Note: Likelihood ratio test for adding focus on self-care = 10.5 on 2 *df*; *P* value = .00520. Population Predictions

Age, y	Not at all	To a small extent	To a moderate extent	To a great extent	To a very great extent
2	63.6	65.6	67.7	69.8	71.8
5	63.3	64.6	65.9	67.2	68.6
12	62.6	62.2	61.8	61.4	61.0

95% Confidence Bounds

Age, y		To a very great extent
2	(56.0-71.2)	(62.6-81.0)
5	(55.7-70.9)	(59.8-77.4)
12	(55.0-70.2)	(53.6,-68.4)

Extent to Which Parents Perceived Their Child's Needs Were Being Met by Services (Needs Met)[a]

	Value	SE	*df*	t-value	*P* value
Intercept	61.06	2.26	447	26.98	.000
Needs met	1.55	0.76	447	2.04	.042
Age, mo	–0.11	0.07	447	–1.55	.121
Needs met: age, mo	0.02	0.02	447	1.19	.236

[a]Likelihood ratio test for adding needs met = 13.4 on 2 *df*; *P* value = .00122.

Population Predictions

Age, y	Not at all	To a small extent	To a moderate extent	To a great extent	To a very great extent
2	64.2	64.9	65.7	66.5	67.2
5	61.1	62.6	64.2	65.7	67.3
12	53.8	57.2	60.5	63.9	67.3

95% Confidence Bounds

Age, y	Not at all	Completely
2	(56.4-72.0)	(59.0-75.4)
5	(53.7-68.5)	(59.1-75.5)
12	(46.9-60.7)	(59.1-75.5)

Likelihood ratio test results from multinomial models for each service variable of interest, controlling for country and setting the reference group as developing as expected, are expressed as ORs and relative risks and are detailed in Tables 13 to 15. The amount of PT, OT, and ST services did not significantly influence change in any of the outcomes. There were no significant relationships of services to the ECAB outcome. There were several exploratory results for the other 3 outcomes, as follows.

Six-minute Walk Test: Progress on the 6MWT was related to the degree of family-centered services, with a decreased likelihood of developing less than expected for those reporting more family-centeredness. For a unit increase OR = 0.57, 95% CI (0.38-0.88), corresponding to a relative risk of 0.16, children receiving the family-centeredness to the greatest extent are only 16% as likely to develop less than expected compared with those receiving the family-centeredness to the least extent.

CEDL participation in family/recreation: The LRT testing indicated that family-centeredness influences change in CEDL participation in family/recreation (c^2 = 8.41; *P* = .015); children were more likely to develop better than expected when reporting increased family-

centered service, for a unit increase OR = 1.46, 95% CI (1.06-2.02). This corresponds to a relative risk of 3.9 for the highest family-centeredness score relative to the lowest; children reporting the highest level of family-centeredness are almost 4 times more likely to develop better than expected vs as expected with respect to participation than those reporting the lowest level of family-centeredness. The LRT indicated that the extent to which parents perceived their child's needs were being met by services influences change in CEDL participation in family/recreational activities (c^2 = 7.89; *P* = .019); children were more likely to develop "better than expected" than "as expected" when reporting an increase in the extent to which parents perceived their child's needs were being met by services. For a unit increase in needs met, OR = 1.48, 95% CI (1.07-2.03), corresponding to a relative risk of 4.14, parents reporting the highest degree of the extent to which they perceived their child's needs were being met by services were roughly 4 times more likely to develop better than expected than those reporting the lowest level of extent to which parents perceived their child's needs were being met by services. A focus on structured play and recreation activities was also associated with a greater likelihood of developing better than expected, OR = 1.03, 95% CI (1.07-1.58), corresponding to a relative risk of 2.55 so that children with the greatest focus on structured play and recreation activities were about 2.5 times more likely to develop better than expected than those with no focus on structured play and recreation activities. Finally, the focus on health was associated with a decreased likelihood of developing less than expected, OR = 0.81, 95% CI (0.67-0.99), corresponding to a relative risk of 0.61 so that children with the greatest focus on health are only 60% as likely to develop less than expected relative to those with no or very little focus on health.

CEDL self-care performance: Development in self-care was related to a focus on health, with an increased likelihood of developing better than expected for children with a greater focus on health, OR = 1.36, 95% CI (1.11-1.65), corresponding to a relative risk of 2.9 so that children receiving services with the greatest focus on health are almost 3 times as likely to develop better than expected than those with no or very little focus on health.

Table 11. Focus on Self-care Activities, Self-initiation, Structured Play and Recreation Activities, and the Extent to Which Parents Perceived Their Child's Needs Were Being Met by Services Effects on Participation in Self-care Activities for Children at GMFCS Level I

Child Engagement in Daily Life Self-care GMFCS Level I Focus on Self-care

	Value	SE	*df*	*T* value	*P* value
Limit	80.30	1.77	644	45.40	.000
Focus on self-care on limit	–0.62	0.24	644	–2.55	.011
Log rate	–3.54	0.11	644	–33.50	.000
Offset	–1.15	2.62	644	–0.44	.659

Abbreviation: GMFCS, Gross Motor Function Classification System.
[a]Likelihood ratio test for adding focus on self-care = 6.1 on 1 *df*; *P* value = .01371. Service variables were scored with the lowest level at 0; limit is the asymptotic limit of the measure for children in the lowest level of the service variable.

Population Predictions

Age, y	Not at all	To a small extent	To a moderate extent	To a great extent	To a very great extent
2	41.5	41.2	40.9	40.5	40.2
5	66.6	66.1	65.6	65.1	64.5
12	79.1	78.5	77.9	77.3	76.7

95% Confidence Bounds

Age, y	Not at all	To a very great extent
2	(36.8-46.2)	(35.3-45.1)
5	(61.1-72.1)	(59.4-69.6)
12	(69.7-88.5)	(68.3-85.1)

Focus on Self-initiation[a]

	Value	SE	*df*	*T* value	*P* value
Limit	80.78	1.86	644	43.52	.000
Focus on self-initiation on limit	–0.52	0.23	644	–2.25	.025

Log rate	–3.56	0.11	644	–33.01	.000
Offset	–1.77	2.74	644	–0.65	.517

[a]Likelihood ratio test for adding focus on self-initiation = 4.6 on 1 *df*; *P* value = .03211.

Population Predictions

Age, y	Not at all	To a small extent	To a moderate extent	To a great extent	To a very great extent
2	42.1	41.8	41.6	41.3	41.0
5	67.0	66.5	66.1	65.7	65.2
12	79.5	79.0	78.5	78.0	77.5

95% Confidence Bounds

Age, y	Not at all	To a very great extent
2	(37.4-46.8)	(36.3-45.7)
5	(61.5-72.5)	(59.9-70.5)
12	(70.1-88.9)	(69.1-85.9)

Focus on Structured Play and Recreation Activities (Focus on Structured Play)

	Value	SE	*df*	*T* value	*P* value
Limit	80.14	1.75	644	45.81	.000
Focus on structured play on limit	–0.49	0.23	644	–2.17	.030
Log rate	–3.53	0.11	644	–32.82	.000
Offset	–1.32	2.67	644	–0.49	.622

Note: Likelihood ratio test for adding focus on structured play = 2.6 on 1 *df*; *P* value = .10360. This was included because the effect of structure on limit had *P* < .05.

Population Predictions

Age, y	Not at all	To a small extent	To a moderate extent	To a great extent	To a very great extent
2	41.9	41.6	41.4	41.1	40.9
5	66.8	66.4	66.0	65.5	65.1
12	79.0	78.5	78.0	77.5	77.0

95% Confidence Bounds

Age, y	Not at all	To a very great extent
2	(37.2-46.6)	(36.0-45.8)
5	(61.3-72.3)	(59.6-70.6)
12	(69.6-88.4)	(67.6-86.4)

Extent Parents Perceived Their Children's Needs Were Being Met by Services (Needs Met)[a]

	Value	SE	*df*	*T* value	*P* value
Limit	76.57	2.04	644	37.61	.000
Needs met mean on limit	1.15	0.43	644	2.70	.007
Log rate	–3.56	0.11	644	–33.61	.000
Offset	–1.37	2.64	644	–0.52	.604

[a]Likelihood ratio test for adding needs met mean = 7.8 on 1 *df*; *P* value = .00519.

Population Predictions

Age, y	Not at all	To a small extent	To a moderate extent	To a great extent	Completely
2	39.5	40.1	40.6	41.2	41.8
5	63.3	64.2	65.2	66.1	67.1
12	75.4	76.5	77.6	78.8	79.9

95% Confidence Bounds

Age, y	Not at all	Completely
2	(34.6-44.4)	(36.9-46.7)
5	(58.4-68.2)	(61.6-72.6)
12	(68.0-82.8)	(70.5-89.3)

Table 12. Effects of PT, OT, and ST Amounts on Participation in Self-care Activities for Children at GMFCS Level I

Child Engagement in Daily Life Self-care GMFCS Level I OT Sessions per Year Categorized (OT Times)[a]

	Value	SE	*df*	*T* value	*P* value
Limit	82.46	1.84	643	44.85	.000
OT times on limit	–3.67	0.76	643	–4.85	.000
Log rate	–3.54	0.11	643	–33.16	.000
OT times on log rate	0.04	0.04	643	1.20	.230
Offset	–0.38	2.49	643	–0.15	.879

Abbreviation: GMFCS, Gross Motor Function Classification System; OT, occupational therapy; PT, physical therapy; ST, speech therapy.

[a]Likelihood ratio test for adding OT times = 40.4 on 2 *df*; *P* value < .00001. Service amounts were scored with the lowest level at 0; limit is the asymptotic limit of the measure for children receiving the least amount of therapy.

Population Predictions

Age, y	0-1	2-30	31-52	53-155	156 or more
2	41.7	41.1	40.4	39.5	38.6
5	68.1	66.1	64.0	61.7	59.4
12	81.2	77.8	74.3	70.8	67.3

95% Confidence Bounds

Age, y	0-1	156 or more
2	(36.8-46.6)	(33.5-43.7)
5	(62.4-73.8)	(54.9-63.9)
12	(71.8-90.6)	(61.8-72.8)

PT Sessions per Year Categorized (PT Times)[a]

	Value	SE	*df*	*T* value	*P* value
Limit	81.23	1.82	643	44.67	.000
PT times on limit	–2.27	0.72	643	–3.18	.002
Log rate	–3.52	0.11	643	–31.91	.000
PT times on log rate	0.01	0.03	643	0.26	.795

Offset	–0.93	2.63	643	–0.35	.724

[a]Likelihood ratio test for adding PT times = 20.4 on 2 *df*; *P* value = .00004.

Population Predictions

Age, y	0-1	2-30	31-52	53-155	156 or more
2	42.5	41.5	40.6	39.6	38.6
5	67.9	66.2	64.5	62.8	61.1
12	80.1	77.9	75.7	73.5	71.3

95% Confidence Bounds

Age, y	0-1	156 or more
2	(37.8-47.2)	(33.7-43.5)
5	(62.4-73.4)	(56.4-65.8)
12	(70.7-89.5)	(65.0-77.6)

ST Sessions per Year Categorized (ST Times)[a]

	Value	SE	*df*	*T* value	*P* value
Limit	79.94	1.74	643	45.84	.000
ST times on limit	–0.92	0.88	643	–1.04	.297
Log rate	–3.50	0.11	643	–32.20	.000
ST times on log rate	–0.05	0.03	643	–1.60	.110
Offset	–1.53	2.69	643	–0.57	.571

[a]Likelihood ratio test for adding ST times = 19.4 on 2 *df*; *P* value = .00006.

Population Predictions

Age, y	0-1	2-30	31-52	53-155	156 or more
2	42.8	40.8	38.9	37.0	35.2
5	67.4	65.3	63.3	61.2	59.1
12	78.9	77.8	76.6	75.3	74.0

95% Confidence Bounds

Age, y	0-1	156 or more
2	(38.1-47.5)	(29.9-40.5)
5	(61.9-72.9)	(54.6-63.6)
12	(69.5-88.3)	(67.1-80.9)

Table 13. Likelihood Ratio Tests for Removing Variables[a]

	Child Engagement in Daily Life participation			Child Engagement in Daily Life self-care			Early Clinical Assessment of Balance			Six-minute Walk Test		
	□□	*df*	*P* value	□□	*df*	*P* value	□□	*df*	*P* value	□□	*df*	*P* value
Family-centered	**8.41**	**2**	**.015**	4.00	2	.135	0.94	2	.624	**7.13**	**2**	**.028**
Needs met	**7.89**	**2**	**.019**	4.29	2	.117	3.12	2	.210	2.48	2	.289
Focus on stretching	1.24	2	.537	4.21	2	.122	4.04	2	.133	0.74	2	.691
Focus on self-initiation	3.20	2	.202	1.37	2	.505	2.65	2	.266	0.84	2	.657
Focus on health	**6.33**	**2**	**.042**	**10.37**	**2**	**.006**	0.77	2	.679	1.26	2	.531
Focus on motivation	4.58	2	.101	3.27	2	.195	4.50	2	.106	0.29	2	.866
Focus on relaxation	3.17	2	.205	3.58	2	.167	0.27	2	.876	2.74	2	.254
Focus on self-care	4.09	2	.130	0.23	2	.892	2.98	2	.225	3.37	2	.185
Focus on assistive devices	2.39	2	.303	0.72	2	.697	1.68	2	.433	0.99	2	.609
Focus on structured play	**10.28**	**2**	**.006**	0.78	2	.676	0.92	2	.630	0.61	2	.738
PT sessions/y	0.65	2	.723	0.90	2	.638	1.83	2	.400	0.17	2	.920
OT sessions/y	1.18	2	.554	0.45	2	.799	1.19	2	.551	1.97	2	.373
ST sessions/y	4.06	2	.131	0.11	2	.947	0.75	2	.687	3.43	2	.180

Abbreviations: OT, occupational therapy; PT, physical therapy; ST, speech therapy.

[a]Separate models were fit for each service variable, controlling for country. The bolded values correspond to the service variables with likelihood ratio test *P* values of < .05.

Table 14. Odds Ratios With 95% CIs[a]

Better than expected vs as expected	Child Engagement in Daily Life participation			Child Engagement in Daily Life self-care			Early Clinical Assessment of Balance			Six-minute Walk Test		
	OR	LB	UB	OR	LB	UB	OR	LB	UB	OR	LB	UB
Family-centered	1.46	1.06	2.02	1.37	1.00	1.88	1.06	0.78	1.44	0.83	0.54	1.26
Needs met	1.48	1.07	2.03	1.37	1.00	1.87	1.13	0.83	1.53	0.87	0.59	1.30
Focus on stretching	1.06	0.84	1.32	1.27	0.99	1.62	1.16	0.92	1.47	1.13	0.82	1.55
Focus on self-initiation	1.20	0.97	1.47	1.11	0.90	1.35	1.10	0.90	1.34	0.90	0.70	1.15
Focus on health	1.11	0.92	1.35	1.36	1.11	1.65	1.09	0.90	1.31	0.92	0.71	1.21
Focus on motivation	1.20	0.99	1.45	1.16	0.96	1.41	1.19	0.99	1.44	0.96	0.74	1.24
Focus on relaxation	1.16	0.94	1.44	1.21	0.98	1.51	1.00	0.82	1.22	0.88	0.68	1.14
Focus on self-care	1.22	1.01	1.47	1.00	0.82	1.22	0.97	0.79	1.18	0.90	0.68	1.19
Focus on assistive devices	1.15	0.96	1.38	1.08	0.90	1.29	1.02	0.86	1.22	0.89	0.70	1.13
Focus on structured play	1.30	1.07	1.58	1.09	0.90	1.31	1.10	0.91	1.32	0.97	0.76	1.24
PT sessions/y	0.93	0.72	1.21	0.90	0.70	1.15	1.19	0.92	1.53	1.05	0.74	1.48
OT sessions/y	0.98	0.74	1.28	0.93	0.72	1.21	1.15	0.89	1.50	0.95	0.65	1.38
ST sessions/y	1.25	0.97	1.60	1.03	0.81	1.31	1.10	0.87	1.39	0.86	0.60	1.25

Less than expected vs as expected	Child Engagement in Daily Life participation			Child Engagement in Daily Life self-care			Early Clinical Assessment of Balance			Six-minute Walk Test		
	OR	LB	UB	OR	LB	UB	OR	LB	UB	OR	LB	UB
Family centered	**0.81**	**0.60**	**1.09**	**1.07**	0.80	1.44	1.15	0.85	1.56	**0.57**	**0.38**	**0.88**
Needs met	**0.85**	**0.64**	**1.14**	**0.95**	0.71	1.26	0.81	0.60	1.07	0.75	0.51	1.09
Focus on stretching	0.90	0.74	1.11	1.09	0.88	1.36	1.21	0.96	1.52	0.96	0.72	1.27
Focus on self-initiation	0.97	0.81	1.17	1.07	0.89	1.29	1.15	0.94	1.41	0.94	0.73	1.20
Focus on health	**0.81**	**0.67**	**0.99**	**0.95**	**0.79**	**1.14**	1.02	0.85	1.23	0.87	0.67	1.14
Focus on motivation	0.93	0.77	1.11	0.94	0.79	1.12	1.12	0.93	1.35	0.94	0.73	1.21
Focus on relaxation	0.92	0.76	1.11	1.08	0.89	1.31	1.05	0.86	1.28	0.83	0.65	1.07
Focus on self-care	1.02	0.84	1.24	1.05	0.87	1.26	1.17	0.97	1.41	1.22	0.95	1.56
Focus on assistive devices	1.03	0.86	1.22	1.02	0.86	1.20	1.12	0.94	1.33	1.03	0.81	1.29
Focus on structured play	**0.87**	**0.72**	**1.04**	1.00	0.83	1.19	1.02	0.85	1.22	0.91	0.71	1.16
PT sessions/y	0.92	0.71	1.18	0.93	0.72	1.21	1.01	0.79	1.29	1.06	0.76	1.48
OT sessions/y	0.86	0.66	1.13	1.05	0.80	1.38	0.98	0.76	1.27	1.27	0.89	1.81
ST sessions/y	0.90	0.70	1.16	1.04	0.81	1.33	1.06	0.84	1.34	1.28	0.94	1.76

Abbreviations: LB, lower bound; OR, odds ratio; OT, occupational therapy; PT, physical therapy; ST, speech therapy; UB, upper bound.
[a]The bolded values correspond to the service variables with likelihood ratio test *P* values of <.05.

Table 15. Comparing the Relative Risk of Developing *Better Than Expected* vs *As Expected,* or Developing *Less Than Expected* vs *As Expected* for the Maximum of the Service Variable Score Relative to the Minimum of the Service Variable Score[a]

	Child Engagement in Daily Life participation		Child Engagement in Daily Life self-care		Early Clinical Assessment of Balance		Six-minute Walk Test	
	Better than expected	Less than expected	Better than expected	Less than expected	Better than expected	Less than expected	Better than expected	Less than expected
Family-centered	**3.95**	0.48	3.05	1.28	1.22	1.64	0.52	**0.16**
Needs met	**4.14**	0.57	3.05	0.82	1.54	0.48	0.63	0.39
Focus on stretching	1.21	0.70	2.33	1.38	1.71	1.96	1.53	0.87
Focus on self-initiation	1.91	0.90	1.43	1.28	1.39	1.64	0.70	0.81
Focus on health	1.46	**0.48**	**2.90**	0.83	1.34	1.07	0.76	0.63
Focus on motivation	1.91	0.77	1.70	0.80	1.84	1.49	0.86	0.82
Focus on relaxation	1.73	0.73	1.99	1.31	1.01	1.20	0.64	0.54
Focus on self-care	2.00	1.06	1.00	1.18	0.89	1.71	0.70	1.95
Focus on assistive devices	1.66	1.09	1.31	1.06	1.09	1.48	0.67	1.09
Focus on structured play	**2.55**	0.61	1.34	0.99	1.37	1.07	0.91	0.73
PT sessions/y	0.78	0.74	0.69	0.79	1.81	1.04	1.17	1.22
OT sessions/y	0.92	0.59	0.78	1.19	1.63	0.93	0.83	2.21
ST sessions/y	2.17	0.69	1.09	1.14	1.38	1.23	0.60	2.27

Abbreviations: OT, occupational therapy; PT, physical therapy; ST, speech therapy.

[a]The bolded values correspond to the service variables with likelihood ratio test *P* values of <.05.

DISCUSSION

Decisional Context

The On Track study results provide useful and important information to assist with answering 2 patient-centered questions: (1) How should I expect my child to develop? (2) How can clinicians and the care delivery systems they work in help me make the best decisions about my child's health and rehabilitation? To answer these questions, we used standard clinical measures to assess children with CP stratified by a standard functional classification system, the GMFCS. The use of standardized clinical measures could improve the consistency of physical therapist practice and communication between health care professionals. The use of a standardized classification tool to report on developmental progress decreases the variability of the diagnosis of CP, allowing for improved understanding of progress and focus of interventions.

The study results provide evidence-based data, not in existence before this study, that describe how children with CP progress in many aspects of their physical development and in participation in daily life from 1.5 to approximately 12 years of age. Creation of longitudinal trajectories by functional ability classification level enables families and health care providers to discuss a child's current development in comparison to others with similar disability in broad terms and to anticipate likely changes. The percentile graphs indicate how a child is developing relative to peers of the same ability level and of a similar age, permitting an understanding of a child's individual strengths and limitations. Information on the clinical course of children's physical development and participation assists therapists and parents in proactively and collaboratively (1) planning services around fundamental goals identified by families; (2) mitigating secondary impairment risk; and (3) optimizing a child's health, function, and participation.[32,40] Information on broad associations between aspects of therapy services and physical development and participation in daily life outcomes will help therapists and families determine the services that are most beneficial and meaningful for children and their family members, which should increase the efficiency and effectiveness of services.

Study Results in Context

Aim 1: Developmental Trajectories Determinants of Gross Motor Ability

Data on the longitudinal development of gross motor ability based on the GMFM have been available since 2002.[35] Use of this information within clinical practice has improved discussions about motor development prognosis between families with children with CP and health care providers; however, there has been a gap in knowledge of the longitudinal development of key determinants of children with CP's outcome of gross motor abilities.[47,52,53] From previous research, variables that were associated with gross motor abilities included primary impairments (spasticity, quality of movement, postural stability, and distribution of involvement)—with postural stability (balance) accounting for the highest variance—and secondary impairments (strength, joint range of motion, and endurance).[52] The impact of health conditions associated with the diagnosis of CP was also found to be a key determinant of self-care abilities.[53] Therefore, we created longitudinal trajectories for balance, spinal alignment/range of motion, strength, endurance, impact of health conditions, and physical activity variables.

Our purpose was to estimate the distribution of these measurements by characterizing the degree of variability. We used 2 methods (longitudinal models and percentiles), which we precisely chose to show this variability in a clinically meaningful way. The study was extremely successful. We expected that the children with CP would vary in their development based on their functional ability classification, with lower ability leading to smaller improvements in their development as compared with higher functional ability. Overall, we found this hypothesis to be supported. The longitudinal trajectories provide a guide for prognosis that should assist therapists and families in setting appropriate and achievable goals for improving determinants of body structure/function impairments related to gross motor development. We also expected that children with higher functional ability would progress over a longer period of time as compared with those with lower functional ability. This supposition was not supported overall, suggesting that children with more limited functional ability can continue to change on most of the determinants of gross motor ability during the ages of 1.5 to 12 years. Last, we

hypothesized and provided some support that younger as compared with older children with CP would improve at a greater rate, reinforcing that early services are very important. Details on qualifications of these general statements follow.

Overall, when we tracked our sample across 2 years, children with lower functional ability showed lower predicted scores for 12 years of age for almost all physical body structure/function variables tracked. There were 2 noticeable exceptions to this: (1) For the impact of health conditions, children at GMFCS Level III had a lower predicted health impact effect than those at Level II; and (2) for the amount and intensity of physical activity, children at GMFCS Level II demonstrated greater activity levels than those at GMFCS Level I. The reasons for these differences are not known and may just be a spurious finding within the data set.

Based on the GMFM motor growth curves for children with CP (1-13 years old), the age at which children were predicted to reach 90% of their motor development potential was shown to follow a sequence among children in the 5 GMFCS levels. We found longer development times in children with greater functional ability (GMFCS Level I = 5 years; II = 4.5 years; III = 3.75 years; IV = 3.5 years; V = 2.75 years).[35] Within this study, 6 of the physical body structure/function measures showed nonlinear developmental trajectories for some or all of the 5 GMFCS levels. However, no clear pattern of time to reach 90% of the predicted maximum for each outcome emerged based on functional ability levels. For these outcomes, the time to reach 90% of maximum was variable, ranging from 14 to 107 months, and was not associated consistently with the children's functional ability. This finding suggests that the rate at which children progress in development of the outcomes is not dependent on functional ability level and therefore supports continued work within therapy and home life toward improving outcomes.

We found linear developmental trajectories for spinal alignment/range of motion, impact of health conditions, muscle strength (for GMFCS Levels III-V), walking intensity/amount, and activity intensity/amount outcomes. Spinal alignment/range of motion showed trends for children to have increases in limitations at all functional ability levels, especially children with the lowest functional ability, indicating that current therapy activities

might be slowing increases in range of motion restrictions; however, the restrictions continued to increase between ages 1.5 to 12 years. The impact of health conditions essentially stayed the same among the functional ability levels, with the exception of children at GMFCS Level III showing a decreasing trend in the impact. This may be linked to the interventions provided, as described later in this report. Strength in the children with lower functional abilities (GMFCS Levels III and IV) increased markedly, while strength in children with the lowest functional ability (GMFCS Level V) decreased slightly. Even with these increases in strength based on isolated muscle group testing, walking for children with GMFCS classification levels of I to III and activity amounts and intensities in children with CP across all functional ability levels decreased, indicating the need for more focus on activity to prevent future health concerns and fitness.

The On Track longitudinal trajectories predicted that children would, on average, show a more rapid rate of development in the early years for all measures except spinal alignment/joint range of motion, impact of health conditions, strength (GMFCS Levels III-V), walking amount and intensity, and activity amount and intensity. In these outcomes, only approximately 30% of the development occurred between 2 and 5 years, as compared with between 5 and 12 years. This finding suggests that children across the ages of 1.5 to 12 years need services related to these outcomes.

Between 9 and 12 years of age, a drop in mean gross motor ability (GMFM scores) has been noted to occur in children with lower functional ability (GMFCS Levels III, IV, and V).[54] When children were tracked across more years (up to 21 years), a clinically significant drop in gross motor ability (GMFM) has been documented for children with lower functional ability (GMFCS Levels III, IV, and V).[54] Correlates of the drop were spinal alignment/range of motion, pain, and several anthropometric measurements (smaller circumference of midarm and midarm circumference/knee height ratio [a body mass index proxy]).[55] Exercise participation was not related but was noted to be low in the cohort followed, and declined across the study time (4 years). In the On Track data, much variability occurred within each of the determinant measures, but the overall trajectories of children 9-12 years old by functional ability level did not show a decline, except in certain measures for children at GMFCS Level V. Children at Level

V showed a slight decrease in strength, a slight increase in impact of health conditions (impact of pain was a health conditions item), and a marked increase in spinal alignment/joint range of motion limitations. Of note, there were also decreases in moderate to vigorous activity from 1.5 to 12 years of age in children within all functional ability levels. These developmental patterns may be a precursor to a future drop in gross motor function.

Participation

Participation in activities of daily life has been identified by families of children with CP to be a fundamental goal of rehabilitation services. Health care professionals, including physical therapists, have agreed and supported an ultimate goal of optimal participation outcomes for children with CP. Data on the longitudinal development of participation in family/recreation activities and performance in self-care activities of children with CP were missing in the literature; therefore, we created longitudinal trajectories for these outcomes. For participation in family/recreation, mean scores at 12 years of age became lower as functional ability decreased (higher GMFCS levels). Children with CP in all functional ability levels, except Level II, in general showed a rapid increase in participation to reach 90% of their predicted maximum (14-23 months); therefore, most of the progress in development occurred in the children between 2 to 5 years of age. Children at Level II, from age 2 years to 12 years, showed a linear downward trend in participation ability. This may be related to these children's functional ability (not as functional as children at GMFCS Level I but not showing an obvious need for environmental accommodations [use of walkers, etc.] as for children at GMFCS Level III) and a lack of community programs geared at their functional ability level. These results suggest the need for greater intervention focus to improve participation, the potential need for more community programs tailored to the children's functional ability levels, and better family-centered services, as noted later in this report.

For performance in self-care activities, mean scores at 12 years of age also became lower as functional ability decreased. Children at GMFCS Levels I to III appeared to make continual improvements in self-care abilities from 1.5 to 12 years of age, with approximately 60% of development occurring between 1.5 to 5 years. Children at Levels IV and V showed a

very steep early development, reaching 90% of their predicted maximum within 20 to 40 months. These data are informative for families and therapists in guiding when to move from a focus on changing motor abilities to provision of appropriate supports, environmental modifications, and training in self-determination.

Aim 2: Services Relationship to Outcomes

Decision-making regarding the most effective and efficient amount, focus, and delivery mode of therapy services is complex. Decisions must rely on the effectiveness of the intervention and on the availability of therapists and families, training of therapists, therapist's and children's environments, and health care and insurance policies.[11,56] Even if the overriding issues of time, training, and policies are ignored, evidence regarding the amount of services to provide is conflicting. Studies of specific goal-focused interventions, such as use of constraint-induced movement therapy (CIMT) or bimanual intensive intervention, confirm that a higher intensity of services over a short period of time leads to a better specific outcome of improved use of an arm/hand in children with hemiplegic CP.[57,58] In more general studies of physical therapy intervention designed to improve overall outcomes of gross motor and participation abilities, while the results favor greater intensity, studies do not consistently show that greater numbers of sessions with therapists in the longer term lead to better outcomes.[14,19,59] Our previous research examining the relationship of amount of rehabilitation services in combination with attributes of the child and family to the outcomes of gross motor function, self-care, and participation in family/recreation activities revealed only that children who participated in more community activities had better abilities.[47,52,53] This relationship may simply reflect that children with higher functional abilities participate in more community programs, which in turn may be due to the availability of appropriate programs. Further analysis of the amount of PT, OT, and ST within the Move & PLAY study revealed that amounts were primarily related to the functional ability of the children; ie, the lower the functional ability of the children, the greater the amount of therapy services provided. It should be noted, however, that the Move & PLAY study was not designed to specifically draw cause–effect

conclusions. In summary, determination of the amount of therapy services provided depends on many factors and has not been fully resolved based on previous research.

Within the On Track study, we examined the relationships of amount of therapy sessions to the children's longitudinal outcomes. We collected our services information from parents of children with CP across 2 years but did not control the services, so we report relationships within the clinical course of children with CP at the present time. Regarding amount of services, we again found that more service sessions are provided to children with lower functional ability, and given that these children showed smaller improvements than those with greater functional ability, the service amount did not relate to greater improvement in the outcomes we measured. We do not know what the relationship of the amount of service would be to individualized goal attainment or for specific family/child outcomes. We also do not know if the amount of service was decreased, if development of the outcomes we measured would also decrease. Changes in movement ability are thought to relate to the amount of time individuals are able to practice throughout daily life, which does not have to relate directly to the number of sessions with a therapist. Many activities designed to be performed outside of therapy sessions are recommended by therapists. This aspect of intensity is difficult to capture and was not measured within our study.

A systematic review of effective intervention foci for children with CP has shown that infants with CP respond best to interventions that include task-specific practice of child-initiated movements and include environmental modifications and parent education.[60] Another systematic review from a very broad perspective, a "helicopter view," reported that to improve motor activities, function, and self-care, the most effective interventions were CIMT, goal-directed training, bimanual training, home programs, and context-focused therapy.[61] A meta-analysis of intervention foci for children at GMFCS Level I and for those with developmental coordination disorder suggests a small positive effect of traditional and task-specific training.[62] Most recently, a meta-analysis of the effect of therapy and behavioral change interventions on habitual physical activity in children with CP revealed a modest but clinically insignificant intervention effect.[63] Researchers noted that interventions within the studies evaluated were

overall not goal directed or participation focused, and practice of skill activities did not improve participation in recreation activities.[63] Previous research has suggested that progress in 1 ICF domain, for instance "body structure/function," does not necessarily predict change in other domains, for instance, "activity"[64] or "activity to participation."[56,63]

Within the On Track study, while the service associations (amount, focus, family-centeredness) with the longitudinal trajectories were unimpressive, relationships of service variables to the percentile graph classifications (developing more than [top 10 %] and less than [lower 10%] to the reference of as expected [middle 80%]) yielded interesting information. Family-centeredness and parent perception of their child's needs being met were associated with developing more than expected in the participation and endurance outcomes. A focus on health and well-being and on structured play/recreation activities also showed interesting relationships of significance to the participation in our family/recreation outcome. Despite a general report by parents of a strong focus on primary and secondary body structure/function interventions during therapy, these focus variables did not predict the balance or endurance outcomes.

Blending the On Track results with information from systematic reviews and meta-analyses of therapy intervention effectiveness, the following are suggestions about service delivery for children with CP. Parents' rating of the extent to which they perceive services are meeting their child's needs as a predictor of outcomes is consistent with our earlier Move & PLAY study[47,52,53]; it also resonates with comments from our parent researchers in that parents have a great understanding of their children and have knowledge about what their children need. This and our findings regarding family-centeredness suggest that therapists should discuss and collaborate with families as service decisions are made and should value the families' thoughts and desires. While this idea has already received support in previous literature, our results relate this practice to the outcome of participation in family/recreation and to the performance of self-care activities. Our findings also suggest that therapists in general do provide services that are perceived by families to be family-centered to a large extent; however, providing services with the greatest focus on family-centeredness does make

a difference in outcomes. The provision of rehabilitation services should focus on both the health and well-being of the child as a protective issue, to prevent less than expected development, and on structured practice of play and recreational activities, to promote more than expected participation development.

In summary, based on our clinical tracking of children with CP, the amount of therapy received was related to the country in which they received the services; their functional ability level; and, to a certain extent, their age. We found no significant relationships for service amount with the children's ability to improve in physical impairments or participation. Greater family-centeredness of services and higher parent perception that their child's needs were being met were associated with better performance on the 6MWT, reflecting endurance, participation in family/recreation activities, and performance of self-care activities. The relationship of services' focus to the outcomes of participation in family/recreation and performance of self-care activities also showed clinically meaningful associations, with a focus on health and well-being and a focus on structured practice of play and recreational activities. Our hypotheses that children with the most optimal change would have services with more focus on practice of specific tasks, more family-centeredness, and more parent reports that their child's needs were being met to a great extent by their rehabilitation services, were supported.

Implementation of Study Results

There is great potential for implementing the results of Aim 1, development of longitudinal trajectories (in addition to knowledge of percentiles and the system to interpret change over time), into practice to assist with discussing prognosis and determining the focus of plans of care. Findings from analyzing the relationship of services provided to children's outcomes (Aim 2) underscore the importance of emphasizing aspects of holistic care, such as family-centeredness and ensuring that children's needs are met, as well as putting more consideration on the child's overall health and well-being and participation.

The research team's goal is to develop a manual of the study results for therapists and to offer teaching symposiums at national and local conferences to explain how to implement

the information in practice. Our team presented a half-day, preconference workshop associated with the American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) in September 2017, presented similar information at the Ontario Association for Children's Rehabilitation Services Annual conference in November 2017, and will present at the Combined Sections Meeting of the American Physical Therapy Association in February 2018.

We have planned to hold (and archive) a series of webinars for the Pediatric Division of the Canadian Physiotherapy Association, which can also be introduced within the United States via the American Physical Therapy Association's Learning Center in fall 2017/spring 2018. We will make a therapy "how-to" manual, including the longitudinal trajectories, available online for download. One team member has also led several students in qualitative research knowledge translation projects related to the On Track study. One project was working with parents and youth to determine how they would best like to receive the assessment information to help with check-ups and check-ins. The second project used a deliberative dialogue approach with a wide variety of stakeholders to explicate what is required at a systems level (ie, the Ontario Association of Children's Rehabilitation Centres) to implement the findings. The results of these studies will guide the production of appropriate dissemination materials for families and health care decision makers. These post–PCORI award activities are funded by the CIHR grant.

We also have a dissemination plan that includes many papers that will be submitted to various professional journals for review for potential publication. Journals will include those related to physical therapy, rehabilitation medicine, and parent magazines. For all manuscripts submitted for potential publication, our parent researchers will be polled to determine if they have the time and desire to participate. The parent researchers will be authors, dependent on their desire to review and participate in the publications. We will consider the PCORI data sharing plan, NIH options for potential data sharing, and the developing CP registry through the AACPDM.

Generalizability

The developmental trajectories, reference percentiles, and services data collected in this study are generalizable to other children with CP in the United States and Canada, as our sample is reflective of population numbers within GMFCS levels, our participants came from a wide geographical region, and our participants represent a wide range of socioeconomic statuses. Populations of children with other disorders and children from other countries/cultures, where services may not be as available or where child-rearing methods vary, would need to be studied, as they may demonstrate different results.

Subpopulation Considerations

Not applicable.

Study Limitations

Limitation of Research Design

We did not follow a single cohort from 1 year to 11 years of age; rather, we tracked children from 1.5 to 10 years of age at the study onset over a course of 2 years. We created developmental trajectories across ages (1.5-12 years) by collecting data from children of a wide range of ages instead of collecting repeated measures over 10 years.

Although this specific research design can be considered acceptable[28] (if not optimal) given limited time and resources, interpretations and applications should be made with caution. Confounding by indication can be a problem when trying to draw cause–effect inferences from dose-response relationships; therefore, these interpretations should not be made. We used an observational design and did not attempt to control intervention for the children. The study did not include a comparison group without CP; we believe that growth patterns are different for children with and without CP and were interested in tracking children with CP. We planned to recruit children to match the population numbers by the GMFCS but ended up recruiting all children and families who were willing to participate. In the end, our sample demographics were similar to the reported population of children with CP.

Limitations of Outcomes

For participation in family and recreational activities, we used only frequency as an outcome for the 2 aims. The construction of participation also includes involvement of participation. Although the Child Engagement in Daily Life measure also allows reporting of enjoyment of participation, one aspect of involvement, we did not use these data in the analysis, as enjoyment is subjective, is an individualized experience, and may not be appropriate for creation of longitudinal trajectories or percentile ranks. However, future research needs to consider and examine other aspects of involvement in participation in order to monitor participation in a more comprehensive manner. Our measurement of self-care activities on the CEDL is a performance-level assessment and is different from how often a child participates in and enjoys activities.

Limitations to the Developmental Trajectories Aim Include the Following:

(1) Even though we have collected data on 708 children with CP, the variability of the children on all measures was large; therefore, the longitudinal trajectories need to be used as a prognostic guide but not to evaluate an individual child's progress over time. (2) We tracked a smaller number of children for the measures of walking endurance and for actual activity measurements; therefore, these results are more preliminary for children with CP in the United States. (3) The balance measure demonstrated a ceiling effect for the children with CP at GMFCS Level I; therefore, the developmental trajectories are limited for this construct for children at this functional level. (4) It is important to consider that within this study we report development of children receiving medical services; this should be considered when interpreting the longitudinal trajectories. (5) We considered no confounding factors and possible interactions in data analyses. We did not take into account potential effects of multiple sites.

Limitations in the Relationship of Services Received to Outcomes Experienced Include the Following:

(1) We collected services data from parents rather than directly from therapists; therefore, data should be considered estimates from the parents' perspectives and may not fully reflect how therapists would report their services.[65] (2) Services were not manipulated within the study and neither were discrete periods "on" or "off" service known, so conclusions about the comparative effectiveness of service are not warranted. (3) Services findings relating to the longitudinal trajectories analyses were marginally statistically significant and also related specifically to a GMFCS level, so generalization should be done with caution. (4) Services findings could have been confounded by indication, as noted earlier (ie, children with the greatest functional limitations often receive the most services and yet experience the smallest change in the constructs measured).

Future Research

Future research needs related to the developmental trajectories aim include the following:

(1) extension of the tracking through young adulthood to better understand the declines in gross motor and participation outcomes and (2) extension of tracking to include nutritional and anthropometric data to better understand changes during adolescence.

Future research needs related to the relationship of services received to outcomes aim include the following:

(1) prospective study of the quantity of generic physical therapy services for large samples of children grouped within GMFCS levels; (2) prospective comparisons of therapy services that conform to the guidelines for best practice vs generic physical therapy service; (3) practice-based evidence studies[66,67] on a large sample recording service goals, amount, and focus of service provided, and relating that to standardized goal outcomes across a specified time; (4) study of the linkage of services with the engagement of the child and family in the services and the child's practice within the "real world" to standardized outcomes; (5) controlled studies of the effectiveness of the family-centered services and a focus on health and

activity within therapy services; and (6) controlled studies testing the value of the use of the trajectories to inform patient care.

CONCLUSIONS

We tracked a large sample of US and Canadian children with CP (N = 708), aged 1.5 to 11.9 years, across 2 years for their development of physical body structure/function, participation in family/recreation activities, and performance in self-care activities. We created longitudinal trajectories and reference percentiles for children within each GMFCS level through nonlinear or linear modeling, as appropriate, to represent the clinical course of development of each variable. These developmental trajectories supplement previous longitudinal curves developed for gross motor activities using the GMFM. From examination of the longitudinal trajectories, we observed several specific issues. Walking for children with GMFCS classification levels of I to III and activity amounts and intensities in children with CP across all functional ability levels decreased, indicating the need for more focus on activity to prevent future health concerns and fitness. The time to reach 90% of maximum scores on the outcomes was not associated consistently with the children's functional ability. This finding suggests that the rate at which children progress in development of the outcomes is not dependent on functional ability level and therefore supports continued work within therapy and home life toward improving on the outcomes. Therapists should consider using the longitudinal trajectories to inform collaborative discussions with families regarding children's prognosis and service needs. Percentile graphs can be used to evaluate individual children over time in all informed shared decision-making relative to service planning.

We evaluated the relationship of the amount, focus, family-centeredness, and extent to which parents perceived their child's needs were being met by services to 4 outcomes: balance, walking endurance, participation in family/recreation activities, and performance in self-care activities. Relationships of services to outcomes based on longitudinal trajectories were few and very modest. Amount of services was related to the children's GMFCS levels, their country, and their age, but more sessions of therapy did not relate to better outcomes. Children with lower functional abilities received more therapy sessions but did not show higher outcomes, which likely was confounded by indication, as noted earlier. Relationships of services to outcomes based on categorical percentile groups showed clinically meaningful positive

relationships for a focus on health and well-being and a focus on structured play and recreation activities to participation outcomes. The family-centeredness of therapy and the extent to which parents perceived their child's needs were being met by services also showed meaningful positive relationships with participation outcomes. These types of services delivery and therapy foci have generally been supported by previous systematic reviews and meta-analyses of intervention effectiveness for children with CP. Therapists and families should consider these holistic concepts as they make service decisions for children with CP.

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APPENDIX

Appendix A. Details on Longitudinal Trajectories by Outcomes Measured

Impairments and Associated Health Conditions Balance (ECAB)

The longitudinal curves for the ECAB total score were nonlinear for each GMFCS level. The estimated population value (95% CI) at age 12 years was the largest for children at GMFCS Level I and decreased for each GMFCS level, with no overlap of 95% CIs indicating that development is different by functional ability classification. The Time-90 parameter increased from Level V (34 months) to Level III (71 months), then maintained for Level II (71 months) but was very short for Level I (35 months). This lends some support to indicate that children with lower functional ability reach their maximum ability sooner than those with greater functional ability. The mismatch with children at Level I and II may have occurred as there was a ceiling effect for the ECAB measure which started in individual children as early as 3-5 years age; therefore, we do not know what their highest balance ability was. The mean changes in scores from age 2-5 years represented a larger percent change (70-91%) as compared to age 5-12 years, indicating that younger children with CP develop balance ability at a greater rate than older children with CP.

Spinal Alignment/Range of Motion (SAROMM)

The longitudinal trajectories for the SAROMM average score were linear for each GMFCS level. The estimated population value (95% CI) at 12 years of age was the smallest for children at GMFCS Level I and increased (representing more limitations) for each GMFCS level, with no overlap of 95% CIs indicating that development is different by functional ability

classification. Due to the continual increase in the mean SAROMM scores, we could not calculate a Time-90 parameter. There was no difference in percent change in scores at 2-5 years of age as compared with 5-12 years of age for children at GMFCS Levels I to III and minimal differences for children at GMFCS Levels IV (50%) and V (33%), indicating that, in general, younger children do

not develop spinal alignment and range of motion restrictions at a greater rate than older children.

Muscle Strength (Functional Strength Assessment)

The longitudinal trajectories for strength were nonlinear for GMFCS Levels I and II and linear for Levels III to V. The estimated population value (95% CI) at 12 years of age was the largest in children at GMFCS Level I and reduced through the levels, with the smallest value for GMFCS Level V. There was no overlap in the 95% CI between levels, except for children at GMFCS Levels II and III, generally supporting that development is different by functional ability classification. The Time-90 parameter increased from Level II (23 months) to Level I (67 months) and could not be calculated for Levels III to V. This lends some support to the contention that children with lower functional ability (Level II) reach their maximum sooner than those with greater functional ability (Level I). The mean percent change in scores from 2-5 years of age was large for children at GMFCS Levels I and II (75%-100%) but small for children at Levels III to V (0%-27%), as compared with 5-12 years of age. This indicates that younger children with greater functional ability develop strength at a greater rate than older children, but those with lower functional ability do not develop strength at a greater rate than older children.

Endurance (6MWT)

The longitudinal curves for endurance (6MWT) were nonlinear for children with CP who were ambulatory (GMFCS Levels I-III). The estimated population value (95% CI) at 12 years of
a ge was highest at GMFCS Level I and decreased for each GMFCS level, with no overlap of 95% CIs indicating that development is different by functional ability classification. The Time-90 parameter increased from Level III (20 months) to Level II (69 months) then dropped for Level I (50 months). This lends some support to the hypothesis that children with lower functional ability reach their maximum score sooner than those with greater functional ability. The percent change in mean scores from 2-5 years of age represented a larger percent change (72%-

99%) as compared with 5-12 years of age, indicating that younger children develop endurance for walking at a greater rate than older children.

Endurance (EASE)

The longitudinal curves for endurance for activity were nonlinear for each GMFCS Level. The estimated population value (95% CI) at 12 years of age was the largest for children at GMFCS Level I and decreased for each GMFCS level, with no overlap of 95% CIs indicating that development is different by functional ability classification. The Time-90 parameter varied from 15 to 25 months and was not related to the children's GMFCS level. The percent change in
mean scores occurred 100% within 2-5 years of age as compared with 5-12 years of age, indicating that younger children develop endurance for activity at a greater rate than older children.

Impact of Health Conditions (Child Health Conditions Questionnaire)

The longitudinal curves for the mean impact of health conditions were linear for each GMFCS level. The estimated population value (95% CI) at 12 years of age was lowest for children at GMFCS Level I and increased across the GMFCS levels; however, there was an overlap in 95% CIs between Level I and III and between Level II and IV, indicating some support that development is different by functional ability classification. Due to the linear nature of development, we could not calculate a Time-90 parameter. Children at Level I and IV essentially remain the same through the age range. Children at Level II and V continue to increase slightly, and those at Level III decrease slightly through 12 years of age. The percent change in mean scores from 2-5 years of age represented a smaller percent change (33%-50%) as compared with 5-12 years of age, indicating that younger children with CP do not increase the impact of health conditions at a greater rate than older children with CP.

Participation

Child Engagement in Family/Recreation Activities (CEDL Part 1)

The estimated population value (95% CI) at age 12 years does gradually decrease from GMFCS Level I to V; however, there is a large overlap of the 95% CI between children at Levels II and III and a very small overlap between Levels II and IV, indicating some support that development is different by functional ability classification. The Time-90 parameter varied from 14 to 23 months and was not related to the children's functional ability (GMFCS level). Children at GMFCS Level II showed a slight decrease in development over time; this does not support that the rate of development is associated with functional ability. The percent change in mean scores occurring at 2-5 years of age represented a

larger percent change (95%-97%) as compared with 5-12 years of age in all GMFCS levels, except for Level II, for which the mean score change occurring between 2 and 5 years of age was 30%. The mean change score for Level II, however, represents a smaller decrement in score from 2-5 years of age as compared with 5- 12 years of age. This supports that younger children develop participation in family/recreation activities at a greater rate than older children.

Child Performance in Self-Care Activities (CEDL Part 2)

The estimated population value (95% CI) at 12 years of age was the largest for children at GMFCS Level I and decreased for each GMFCS level, with no overlap of 95% CIs, indicating that development is different by functional ability classification. The Time-90 parameter varied from 20 to 107 months with children who had the most functional ability (GMFCS Levels I-III), developing their maximum across a mean of 80 to 107 months and those with less functional ability meeting their Time-90 parameter more quickly (20-40 months). This supports that functional ability is associated with the development of children's performance in self-care activities. The percent change in mean scores occurring at 2-5 years of age represented a larger percent change (59%-90%) as compared with 5-12 years of age in all GMFCS levels. This supports that younger children develop performance in self-care at a greater rate than older children.

Physical Activity Substudy *Amount of Walking (StepWatch)*

The estimated population value (95% CI) at 12 years of age decreases from GMFCS Level I to III, but there are overlaps in the 95% CIs between consecutive levels, indicating some support that development is different by functional ability classification. Due to the linear nature of development, we could not calculate a Time-90 parameter. Scores for

children at Level I decrease slightly and children at Level II and III increase slightly from 1.5 to 12 years of age. The percent change in mean scores occurring at 2-5 years of age represented a small percent change (30%) as compared with 5-12 years of age in all GMFCS levels; however, for Level I this represented a smaller decrease at 2-5 years as compared with 5-12 years. This does not support that younger children develop their amount of walking at a greater rate than older children.

Intensity of Walking (StepWatch)

The estimated population value (95% CI) at age 12 years decreases from GMFCS Level I to III, but there are overlaps in the 95% CIs between consecutive levels, indicating some support that development is different by functional ability classification. Due to the linear nature of development, we could not calculate a Time-90 parameter. Scores for children at Level I show a gradual decrease from 1.5 to 12 years of age. For children at Level II and III, they increase slightly through 12 years of age.

The percent change in mean scores occurring at 2-5 years of age represented a small percent change (30%) as compared with 5-12 years of age in all GMFCS levels; however, for Level I this represented a smaller decrease at 2-5 years of age as compared with 5-12 years of age. This does not support that younger children develop their intensity of walking at a greater rate than older children.

Amount of Activity (ActiGraph)

The estimated population value (95% CI) at 12 years of age was highest for children at GMFCS Level II, but the 95% CIs overlapped between Levels I and II. Children at Level III to V (combined due to small sample size) had the smallest values, and their CI did not overlap with Levels I or II. This

indicates some support that development is different by functional ability

classification. Due to the linear nature of development, we could not calculate a Time-90 parameter. Children in all GMFCS levels showed a decrease in activity counts per minute from
1.5 to 12 years of age. The percent change in mean scores occurring at 2-5 years of age represented a small percent change (30%) as compared with 5-12 years of age in all GMFCS levels. This does not support that younger children develop their amount of activity at a greater rate than older children.

Intensity of Activity (ActiGraph)

The estimated population value (95% CI) at 12 years of age was highest for children at GMFCS Level II, but the 95% CIs overlapped between Levels I and II. Children at Level III to V (combined due to small sample size) had the smallest values and their CI did not overlap with Levels I or II. This indicates some support that development is different by functional ability classification. Due to the linear nature of development, we could not calculate a Time-90 parameter. Children in all GMFCS levels showed a decrease in activity counts per minute from
1.5 to 12 years of age. The percent change in mean scores occurring at 2-5 years of age represented a small percent change (30%) as compared with 5-12 years of age in all GMFCS levels. This does not support that younger children develop their intensity of activity at a greater rate than older children.

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https://www.pcori.org/research-results/2013/understanding-development-children-cerebral-palsy-and-how-therapy-may-affect