# Monitoring development of children with cerebral palsy: the On Track study. Protocol of a longitudinal study of development and services.

SARAH WESTCOTT MCCOY[1,2] | DOREEN BARTLETT[2,3] | MONICA SMERSH[1] | BARBARA GALUPPI[2] | STEVEN HANNA[4] | COLLABORATION GROUP: ON TRACK STUDY TEAM

**1** Department of Rehabilitation Medicine, University of Washington, Seattle, WA, United States; **2** CanChild Centre for Childhood Disability Research, McMaster University, Hamilton, ON, Canada; **3** School of Physical Therapy, Western University, London, ON Canada; **4** Department of Health Research Methods, Evidence, and Impact (HEI), McMaster University, Hamilton, ON, Canada.

Correspondence to Monica Smersh/Sarah Westcott McCoy at Department of Rehabilitation Medicine, University of Washington, Box 356490, 1959 NE Pacific St, Seattle, WA, 98195-6490, United States. Email: msmersh@uw.edu



***See Acknowledgements for additional members of the Collaboration Group.**

**BACKGROUND** The purpose of the On Track study is to determine how children with cerebral palsy (CP) progress in their physical development and participation in daily life. Study aims are to create longitudinal trajectories and percentile graphs for physical development and participation to help health care professionals and parents monitor development and track if children are progressing 'as expected,' 'more than expected,' or 'less than expected.' Services received will be explored in children within each developmental category.

**METHODS** On Track used a prospective cohort design, in which 708 children with CP were followed; 656 were assessed at least twice (baseline, 12-month) over 1 year and 424 were assessed up to 5 times (baseline, 6-, 12-, 18-, 24-months) over 2 years. Children, aged 1.5-11.9 years, and their families were from Canada and the United States. Children were assessed on standardized measures of body functions and structures, health conditions, activity, and participation. Trained physical and occupational therapists measured balance, range of motion, strength, endurance, and physical activity using valid and reliable tests. Parents completed questionnaires about their family demographics and about their children's endurance, health, participation in recreation and self-care, and health care services. Therapists and parents collaborated to classify children within five functional levels for gross motor, manual, and communication functions. Body function and participation data from all visits will be analyzed by linear and nonlinear mixed-effects modeling to create longitudinal trajectories by functional classification levels. Data from baseline, 12-month, and 24-month visits will be analyzed via quantile regression to construct cross-sectional reference percentile graphs for each measure by functional classification levels. Using separate multinomial models, service amount, focus, and family-centeredness, controlling for country, will be explored to understand how services relate to children's development.

**DISCUSSION** Developmental results including longitudinal trajectories and percentile ranks on children with CP by functional classification levels and exploration of services will assist health care professionals and families to monitor development and collaborate on service planning. These results will facilitate conversations to improve family-centered care in order to provide the most efficient and effective interventions for children with CP and their families.

Trial registration: Clinicaltrials.gov Identifier: NCT02391948
Keywords: Cerebral palsy, longitudinal development, percentiles, prognosis, rehabilitation services, parent researchers

## BACKGROUND

Cerebral palsy (CP) is the most prevalent childhood-onset neuromuscular condition, and over 90% of all individuals with CP live well into adulthood.[1-4] Although the underlying pathophysiology of CP is non-progressive, the clinical manifestations are variable and change with age. In previous research, a decline in performance 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 the majority of 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. Other contemporary frameworks such as a dynamic systems approach to motor development,[11] activity-focused motor interventions,[12] context therapy,[13] and participation-based therapy[14] have advocated for interventions for children with CP that focus on the child and family within their everyday environments. In the United States (US), family-centered services provided in natural environments is mandated for publicly-funded early intervention programs for infants and toddlers from birth to 3 years of age.[15]

Families have identified three fundamental outcome goals of rehabilitation for children with CP:[16,17] 1) optimize motor function, 2) prevent secondary impairments that impact life-long health, and 3) promote children's participation in their daily lives.[18,19] The On Track study addresses three patient-centered questions: 1) Given my child's personal characteristics, conditions, and preferences, what should I expect will happen to him/her?; 2) What can I do to improve the outcomes that are most important to my child?; and 3) How can clinicians and the care delivery systems they work in help me make the best decisions about my child's health and health care? The On Track study will provide service providers and families of children with CP with evidenced-based information about children's development that enables shared decision making related to rehabilitation services to achieve mutually important outcomes.

From previous research, it is suggested that current decisions on frequency and amount of physical therapy (PT) and occupational therapy (OT) for young children with CP are often based on convention.[20] Bailes et al.[21] recommended a frequency of 1 to 2 times a week or every-other-week for children who demonstrate continuous progress toward goals. This recommendation corresponds to the frequency of PT and OT that the majority of children were receiving during our previous North American study, Move & PLAY.[20] The Move & PLAY study finding that there were no differences in the amount of PT and OT received by children in different regions in the US further supports the perspective that decisions are often based on convention. Models of service delivery, financial resources for publicly funded services, and private health insurance plan benefits are factors that likely contribute to conventions for frequency and amount of services. The small percentage of

children receiving more than 12 sessions per month of PT or OT indicates that intensive therapy, as defined in research, is not common in practice. This most likely reflects 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.[22] Parent and professional advocacy are also likely to influence decisions on frequency and amount of therapy. In the previous Move & PLAY study, 32% of children in the US were receiving PT and 27% were receiving OT in both an education and clinic setting;[20] this implies that many parents, 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.

The On Track study will fill gaps in fundamental knowledge by creating developmental trajectories of participation in self-care abilities, an important priority for families with children with CP,[23] and participation in family and community recreation. The gap addresses what has been described as a "pressing need" to "increase our understanding of the complexities of CP,"[2] 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, range of joint motion, and endurance for activity), and impact of co-occurring health conditions have not been quantified. Recent reports of the high prevalence of co-occurring health conditions[2,24] suggest that this should also be a focus of monitoring so that families can be better informed of prognoses and expectations. Creation of longitudinal trajectories and percentile graphs would enable families of children with CP and health care providers to: 1) monitor a child's development (developmental surveillance), 2) anticipate a child's future strengths and needs (prognosis), and 3) 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 on *Adults with Cerebral Palsy*.[25] Workshop participants advocated for research that "improves understanding of the natural history of musculoskeletal and neurological impairments across the lifespan in persons with CP".[25] Pragmatically, it is difficult to study the natural history of a childhood condition; therefore, On Track is a study of the clinical course (i.e., documenting but not controlling the rehabilitation and medical services received by study participants). Others have advocated for the use of the International Classification of Functioning, Disability and Health (ICF)[26] and have emphasized the need to pay particular attention to pain, mobility issues, and comorbidities,[27,28] all of which were 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".[27] 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.

## METHODS

### Design and Aims

The On Track study is an international (Canada and US) multi-site prospective cohort design study of children with CP, age 1.5 to 11.9 years of age from all functional GMFCS levels. The study includes three aims and a smaller physical activity sub-study described below. The second aim was funded in 2012 by the Canadian Institutes of Health Research (CIHR) and involved testing children twice over a one-year period (at study onset and 12 months). The first, third, and sub-study aims were funded in 2013 by the US Patient-Centered Outcomes Research Institute (PCORI) and involved testing children three additional times (at approximately 6, 18, and 24 months post onset) over a two-year period. At the time of writing of this protocol paper, the data collection was complete and analyses were in progress.

The first study aim is to characterize average development and its variations, in balance (a primary impairment), range of motion limitations, strength, and endurance (secondary impairments), impact of health conditions, and participation in self-care and recreation/leisure activities. We will do this by creating longitudinal trajectories that estimate the average pattern of change over time and important individual variations in the pattern of change between children who are grouped by their functional ability levels. Functional ability levels are determined by one of the following: 1) Gross Motor Function Classification System (GMFCS),[29] 2) Manual Ability Classification System (MACS),[30] 3) Communication Function Classification System (CFCS).[31] The GMFCS, initially designed for service providers to classify usual gross motor performance of children with CP, was later validated for completion by parents. Both the MACS and the CFCS were developed for service providers to use in collaboration with parents. Establishment of longitudinal trajectories by functional ability levels will provide prognostic information to address the question: What will happen to my child? The following research questions will be answered: What are the unique developmental trajectories that describe change over time in balance, joint range of motion, muscle strength, physical activity, endurance for activity, impact of health conditions and participation in self-care and recreational/leisure activities, and how do these differ among children with CP between and within each of the five GMFCS levels?

The second study aim is to describe the changes in primary and secondary impairments, health conditions, and participation variables over a one-year period by functional ability levels. The data are being used to develop reference percentiles to assist families to determine if their children are progressing 'as expected,' 'more than expected,' or 'less than expected,' depending on their functional ability levels. Establishment of reference percentiles will: 1) provide easily understood and useful tools for families and service providers to discuss how well a child is doing in relation to other children with CP of similar functional ability levels; and 2) help families and service providers identify a child's strengths and areas for improvement, establish goals the child is capable of achieving, and plan for current and future needs. The following research question will be answered: At any given age, within GMFCS levels, what are the reference percentiles that best characterize relative standing in balance, joint range of motion, muscle strength, physical activity, endurance for activity, impact of health conditions, and participation in self-care and recreational/leisure activities?

The third study aim is to describe the relationship between the amount, focus, and family centeredness of therapy services and and percentile change in impairment and participation outcomes. Having this information should assist with collaborative decision making among family members and

service providers, to effectively and efficiently use rehabilitation services to meet families' goals. To date, this information does not exist; rather, many different types and intensities of services are recommended based primarily on convention, clinicians' past experiences, and education rather than on evidence of children's potentials to achieve goals. Knowledge of characteristics of services received will inform best practices and service delivery. The following research question will be answered: What characteristics of amount, focus, and family centeredness of physical, occupational, and speech therapy services, controlling for country, are associated with children with CP across all GMFCS levels who are progressing 'as expected' versus those who are progressing 'more than' or 'less than expected'?

An additional sub-study collected direct physical activity measurements from a sub-set of children at two sites in the US. These data will be correlated with the parents' endurance ratings of their children to provide additional validation for that measure and to create longitudinal trajectories of development for physical activity based on functional ability levels. The following research question will be answered: What are the unique developmental trajectories that describe change over time in walking and physical activity performance (number of steps/day, intensity of steps, number and intensity of physical activity counts), and how do these differ among children with CP between and within the GMFCS levels?

*Research Team members*

The research team for the On Track study consists of physical therapist, physician, and biostatistician investigators from two universities in Canada and four universities in the US, two project coordinators (one in Canada and one in the US), regional coordinators at each data collection site), seven parents of children with CP, and 90 physical and occupational therapist assessors across North America. The investigators conceived the study, obtained funding to undertake the study, and provided the overall guidance for the study. The project coordinators provided the very important guidance and infrastructure to implement the study, overseeing sample recruitment and retention, tracking study progress, monitoring data collection, entering and checking data into a central database (Empower), and managing budgets. The regional coordinators at each site worked directly with the therapist assessors to keep track of recruitment and data collection and, with site data entry assistants, to input data into the central database. The parent researchers provided consultation and collaboration to keep the study family-focused and acceptable to families of children with CP. 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, team 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, a tracking table was used by the team to document scoring decisions for use as a reference guide. In addition, we developed and updated a plan for study dissemination at professional conferences, and in the professional and lay literature; outputs were documented on the study website. The parent researchers were not always available to join the regular team meetings; however, the parent researchers met as a group with one or more study investigators monthly to provide input from the family perspective on study issues. Parent researchers also worked on dissemination for the website and within the lay literature related to topics associated with the study and family perceptions and needs related to rehabilitation.

The therapist assessors assisted in the recruitment process; however, their primary role was to collect data for the study. Training consisted of a standardized, full-day regional training workshop to learn about the study and the measures and equipment used for data collection, as well as additional information relating to safety, privacy, confidentiality, other ethical issues, and administrative procedures. 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. Each assessor was given feedback on any disagreements with the investigators' scores. Throughout the study, the assessors received a semi-annual newsletter highlighting tips on administration and scoring the assessments. Yearly phone conferences were also held inviting each assessor to participate 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*

Institutional Review Board (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 (CITI) training associated with human subjects research. All families and children of appropriate age signed approved consent and assent forms, according to each sites IRB approvals, prior to the start of data collection. Details of the approvals by data collection site are listed in Table 1.

**Table 1: Institutional Review Boards Approving the Study**

| Region | Institutional Review Board |
|---|---|
| Seattle, Washington, USA | University of Washington Human Subjects Division |
| Philadelphia, Pennsylvania, USA | Drexel University Human Research Protection Program |
| Baltimore, Maryland, USA | John Hopkins University Institutional Review Board |
| Oklahoma City, Oklahoma, USA | The University of Oklahoma Health Sciences Center Institutional Review Board |
| Atlanta, Georgia, USA | 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 |

**Parent/Child Participants**

The study participants were children with CP between the ages of 1.5 to 11.9 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, i.e., if they exhibited delayed motor development, muscle stiffness, and difficulties with balance and moving. Ongoing eligibility was maintained throughout the study so that the final dataset for analysis represented children with CP. Therapist assessors provided detailed information for consideration of eligibility of seventy-one unique cases either before or after recruitment to the study. A physiatrist reviewed and made recommendations to the team about any queries relating to study eligibility. As described in Figure 1, eleven cases were not included in the final sample for analysis due to not fulfilling the criteria for the diagnosis of CP. Eighteen months was selected 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),[32] 2) starting that young ensures that we have data from the earliest possible time for the developmental trajectories, and 3) it is still possible to administer the measures that we had selected to children of that age. Because reliability of the GMFCS is greater after 2 years,[33,34] the GMFCS level was confirmed at the 12-month visit for those children under 2 years of age at study onset. Eleven years was selected 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. Families who could not read or communicate in one of these languages were not eligible to participate in this study.

*Recruitment*

Our goal was to recruit a total sample size of 875 (175 in each GMFCS level) to be assessed twice (baseline, 12-month) over 1 year, and for 600 of the children (120 in each GMFCS level) to be assessed up to five times (baseline, 6-, 12-, 18-, 24-months) over 2 years. Our goal was to obtain a quota sample,[35] aiming for a relatively even distribution across the age and GMFCS spectrum. Within the CIHR-funded On Track study we planned to recruit 60% of the sample from sites in Canada and 40% from the US. For the PCORI-funded study, our goal was to increase the US sample size (n=660) to 60% to increase the generalizability of our results to the US and to ensure that we had representation from urban, suburban, and rural areas.

Recruiting was done by regional coordinators and was managed centrally by the project coordinator for each country. The first wave of recruitment was to ask families who participated in the Move & PLAY study and who had previously consented to have us approach them for future studies (n=275). Eighty-seven of these families agreed to participate in the On Track study. In order to recruit more children to meet our desired sample size, various methods were used that were approved by the site IRBs, such as advertisement through health professionals at sites where therapy is received and through patient lists from specific hospitals or programs where the organization staff screened their lists according to our inclusion criteria. We used the recruiting methods that we found to be most successful in the Move & PLAY study to approach new families.[36] We sent out letters of invitation to all eligible families identified in databases at several participating sites. We also identified "champions" at some recruitment sites who were enthusiastic about the study and who we could rely on to follow up with families to clarify aspects of the study, ascertain their interest, and obtain consent from those who were interested in taking part. At enrollment, families provided contact information, the child's GMFCS level, if known (using the parent report form for the appropriate age group), and date of birth. The US regional coordinators and Canadian site liaisons sent the signed consents to the project coordinators, if permitted through local ethics requirements; or alternatively, they were stored securely at the various regional facilities. The highest recruitment numbers came through the screening of patient lists. The greatest difficulty with recruitment was related to the time lag in setting up contracts with actual therapy sites, facilitation of multiple IRB applications, and approvals for payments for recruitment efforts**.**

*Retention*

Once enrolled, all families were contacted by the regional coordinators to introduce the therapist assessor who would be working with them. The therapist assessor then contacted the families to schedule the assessment times. Therapist assessors were instructed to leave a message no more than three times, if they did not speak with the family directly. Use of email, if provided by the families, also assisted with more efficient communication. If a family did not respond to assessor attempts to contact, then the regional coordinator sent a letter in the mail, indicating that the assessor had been unable to reach them successfully by phone and that if we did not hear from them we would not make any further attempts to contact. If families were to decide later on that they would like to continue with the study, they were asked to contact the regional coordinator.

Retention was enhanced through several methods. We disseminated a semi-annual family newsletter; the detailed Family Newsletters are available to view on our study website under Newsletters.[37] 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 throughout the study 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. 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 about supports to enhance the child's motor development and participation in daily activities.

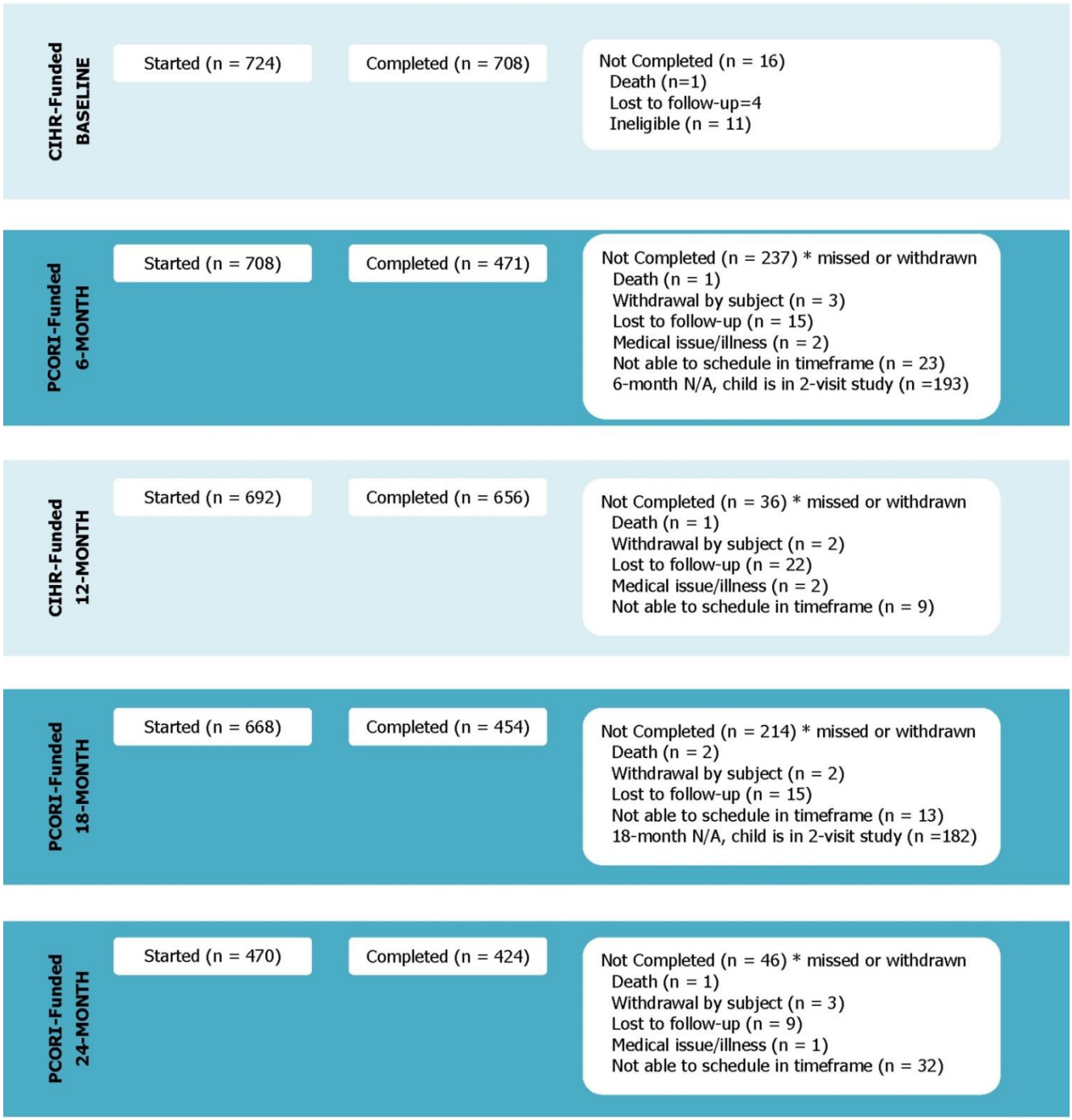
*Sample size*

We estimated the On Track study sample in terms of the requirements for the estimation of LMS percentiles by age and with GMFCS classification, using calculations from Crawford and Garthwaite[38] showing the adequacy of the width of the 95% CI for the 5th, 50th, and 95th percentiles. This sample size is sufficient also for the mixed-effects analyses of longitudinal trajectories and prediction, which are generally less demanding in terms of the number of children. The longitudinal follow-up was extended to five occasions which compares favourably to longitudinal trajectories for gross motor function estimated with considerable precision by Rosenbaum et al.[39] For example, key parameters of the nonlinear model in Rosenbaum et al. 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% CI of +/-3% and +/-15%, respectively. We expected similar precision for our nonlinear models and considerably better precision for outcomes where linear models can be used.

*Final sample*

Our recruitment and retention of participants is shown in Figure 1, yielding a final sample size of 708 for the three main study aims. Our research includes a diverse sample for both the two-assessment and five-assessment studies that is comparable with population-based studies of children with CP.[18,24] Table 2 contains child and parent demographics. Table 3 provides cross-tabulations of levels of the GMFCS, MACS, and CFCS by five age categories. We recruited from 19 sites, clinics and/or practices within the US and 24 sites, clinics and/or practices within Canada, which span the two countries from east to west coasts and in the US from north to south borders. Details of the geographical locations for participants are displayed in Table 4.

**Figure 1: On Track Study Participant Flow Diagram**




**Included in Longitudinal Curves Analysis (n = 708):** Using all available data points. Cases in analysis with 1 visit = 27, 2 visits = 198, 3 visits = 18, 4 visits = 89, 5 visits = 376.

**Included in Percentiles Analysis (n = 708):** Using Baseline, 12- and 24- Month data points with no repeated measurements on a child within an age group.
Cases in analysis with 1 visit = 42, 2 visits = 252, 3 visits = 414.

**Included in the Six-Minute Walk Test Longitudinal Curves Analysis (n=456):** Using all available data points.
Cases in analysis with 1 visit = 33, 2 visits = 136, 3 visits = 29, 4 visits = 71, and 5 visits = 187.

**Included in the Activity Performance Sub-Study Longitudinal Curves Analysis:**
Actigraph (n=79): Using all available data points. Cases in the analysis with 1 visit = 4, 2 visits = 6, 3 visits = 25, 4 visits = 25, 5 visits = 19.
StepWatch (n=50): Using all available data points. Cases in the analysis with 1 visit = 4, 2 visits = 4, 3 visits = 15, 4 visits = 18, 5 visits = 9.

**Table 2: Child and Parent Respondent Demographics**

| | | Participants | | |
|---|---|---|---|---|
| | | **Baseline Completed** n=708 (%) | **12-Month Completed n=656 (%)** | **24-Month Completed N=424 (%)** |
| **Child Age, years** | Mean (SD) | 6.0 (2.7) | 7.1 (2.7) | 7.9 (2.7) |
| | Minimum - Maximum | 1.5 – 11.9 | 2.4 – 13.0 | 3.1 – 14.0 |
| **Child Gender** | Male | 396 (56) | 369 (56) | 242 (57) |
| | Female | 312 (44) | 287 (44) | 182 (43) |
| **Child GMFCS Level** | I | 227 (32) | 217 (33) | 135 (32) |
| | II | 161 (23) | 147 (22) | 97 (23) |
| | III | 80 (11) | 73 (11) | 48 (11) |
| | IV | 129 (18) | 116 (18) | 75 (18) |
| | V | 111 (16) | 103 (16) | 69 (16) |
| **Child Distribution of Involvement*** | Monoplegia | 8 (1) | 8 (1) | 6 (1) |
| **Baseline (n = 707)** | Hemiplegia | 198 (28) | 184 (28) | 114 (27) |
| **12-Month (n = 655)** | Diplegia | 184 (26) | 172 (26) | 114 (27) |
| **24-Month (n = 424)** | Triplegia | 39 (6) | 38 (6) | 20 (5) |
| | Quadriplegia | 278 (39) | 253 (39) | 170 (40) |
| **Child race*** | American Indian/Alaska | 15 (2) | 11 (2) | 3 (1) |
| **Baseline (n = 699)** | Asian | 40 (6) | 37 (6) | 18 (4) |
| **12-Month (n = 649)** | Black/African American | 60 (8) | 56 (8) | 45 (11) |
| **24-Month (n = 419)** | White | 503 (72) | 472 (73) | 310 (74) |
| | Multi | 81 (12) | 73 (11) | 43 (10) |
| **Child ethnicity*** | Hispanic | 49 (7) | 43 (7) | 32 (8) |
| **Baseline (n = 703)** | Non-Hispanic | 654 (93) | 610 (93) | 390 (92) |
| **12-Month (n = 653)** | Aboriginal | 31 (4) | 26 (4) | 9 (2) |
| **24-Month (n = 422)** | Non-Aboriginal | 672 (96) | 627 (96) | 413 (98) |
| **Parent respondent race*** | American Indian/Alaska Native | 15 (2) | 12 (2) | 4 (1) |
| **Baseline (n = 698)** | Asian | 51 (7) | 45 (7) | 22 (5) |
| **12-Month (n = 648)** | Black/African American | 56 (8) | 52 (8) | 42 (10) |
| **24-Month (n = 419)** | White | 550 (79) | 517 (80) | 339 (81) |
| | Multi | 26 (4) | 22 (3) | 12 (3) |
| **Parent respondent ethnicity*** | Hispanic | 32 (5) | 30 (5) | 20 (5) |
| **Baseline (n = 701)** | Non-Hispanic | 669 (95) | 621 (95) | 400 (95) |
| **12-Month (n = 651)** | Aboriginal | 20 (3) | 16 (3) | 5 (1) |
| **24-Month (n = 420-421)** | Non-Aboriginal | 681 (97) | 635 (97) | 416 (99) |
| **Parent respondent age, years*** | Mean (SD) | 37.8 (7.9) | 37.9 (8.0) | 37.4 (7.1) |
| **Baseline (n=694)** | | | | |
| **12-Month (n = 644)** | | | | |
| **24-Month (n = 415)** | | | | |
| **Parent respondent relationship to child*** | Mother | 628 (89) | 578 (88) | 382 (90) |
| **Baseline (n = 704)** | Father | 51 (7) | 51 (8) | 26 (6) |
| **12-Month (n = 654)** | Other | 25 (4) | 25 (4) | 15 (4) |
| **24-Month (n = 423)** | | | | |
| **Parent respondent education*** | High School or less | 160 (23) | 147 (23) | 92 (22) |
| **Baseline (n = 700)** | Community College / Associate's Degree | 212 (30) | 196 (30) | 114 (27) |
| **12-Month (n = 650)** | | | | |
| **24-Month (n = 420)** | University | 328 (47) | 307 (47) | 214 (51) |
| **Family Income*** | ≥$75,000 | 306 (52) | 293 (53) | 190 (52) |
| **Baseline (n = 594)** | $60,000 - $74,999 | 78 (13) | 72 (13) | 43 (12) |
| **12-Month (n = 553)** | $45,000 - $59,999 | 50 (8) | 47 (8) | 34 (9) |
| **24-Month (n = 363)** | $30,000 - $44,999 | 58 (10) | 49 (9) | 35 (10) |
| **(CAD or USD)** | ≤$30,000 | 102 (17) | 92 (17) | 61 (17) |
| **Family Composition** | Adults (mean, SD) | 2.1 (0.7) | 2.1 (0.7) | 2.1 (0.7) |
| **Baseline (n= 667)** | Children (mean, SD) | 2.3 (1.1) | 2.3 (1.1) | 2.3 (1.1) |
| **12-Month (n = 620)** | | | | |
| **24-Month (n = 404)** | | | | |
| **Country** | Canada | 347 (49) | 330 (50) | 137 (32) |
| | United States | 361 (51) | 326 (50) | 287 (68) |

GMFCS= Gross Motor Function Classification System Level CAD = Canadian Dollars USD = United States Dollars SD = standard deviation

* report based on the available information

Notes: 'mother' includes mother, adoptive mother, foster mother, or custodial mother; 'father' includes father, adoptive father, or step father; 'other' includes grandparent, nursing supervisor, or aunt.

**Table 3: Distribution of Child Age by Classification Systems**

| Consensus **GMFCS** at baseline for Children over 2 years at enrolment or at 12 Month Visit for Children under 2 years at baseline | | Age at baseline | | | | | |
|---|---|---|---|---|---|---|---|
| | | 1.5 years up to 3rd birthday | 3 years up to 6th birthday | 6 years up to 9th birthday | 9 to 11 years | over 11 years | Total |
| | Level I | 51 | 73 | 64 | 39 | 0 | 227 |
| | Level II | 23 | 48 | 63 | 26 | 1 | 161 |
| | Level III | 17 | 20 | 31 | 12 | 0 | 80 |
| | Level IV | 21 | 45 | 50 | 13 | 0 | 129 |
| | Level V | 19 | 30 | 38 | 23 | 1 | 111 |
| | Total | 131 | 216 | 246 | 113 | 2 | 708 |

| Consensus **MACS** at baseline for Children over 2 years at enrolment or at 12 Month Visit for Children under 2 years at baseline | | Age at baseline | | | | | |
|---|---|---|---|---|---|---|---|
| | | 1.5 years up to 3rd birthday | 3 years up to 6th birthday | 6 years up to 9th birthday | 9 to 11 years | over 11 years | Total |
| | Level I | 30 | 46 | 47 | 21 | 0 | 144 |
| | Level II | 50 | 89 | 91 | 49 | 1 | 280 |
| | Level III | 20 | 31 | 47 | 9 | 0 | 107 |
| | Level IV | 25 | 38 | 36 | 17 | 1 | 117 |
| | Level V | 6 | 12 | 25 | 17 | 0 | 60 |
| | Total | 131 | 216 | 246 | 113 | 2 | 708 |

| Consensus **CFCS** at baseline for Children over 2 years at enrolment or at 12 Month Visit for Children under 2 years at baseline | | Age at baseline | | | | | |
|---|---|---|---|---|---|---|---|
| | | 1.5 years up to 3rd birthday | 3 years up to 6th birthday | 6 years up to 9th birthday | 9 to 11 years | over 11 years | Total |
| | Level I | 33 | 88 | 97 | 46 | 0 | 264 |
| | Level II | 18 | 39 | 45 | 23 | 1 | 126 |
| | Level III | 41 | 41 | 40 | 12 | 0 | 134 |
| | Level IV | 27 | 34 | 45 | 22 | 1 | 129 |
| | Level V | 12 | 14 | 19 | 10 | 0 | 55 |
| | Total | 131 | 216 | 246 | 113 | 2 | 708 |

GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System; CFCS = Communication Function Classification System.

**Table 4: Recruitment and Assessment Sites and Regions**

| **SITES IN UNITED STATES** | **SITES IN CANADA** |
|---|---|
| *Pacific Northwest* | *Halifax, NS* |
| Children's Therapy Center (CTC) | IWK Health Centre |
| Good Samaritan Children's Therapy Unit (CTU) | *Hamilton ON* |
| MultiCare Pediatric Therapy Services | McMaster Children's Hospital Developmental Pediatrics and Rehabilitation Program |
| Seattle Children's Hospital (SCH) | *Kingston ON* |
| Shriners Hospital for Children, Portland | Religious Hospitallers of Saint Joseph of the Hotel Dieu Kingston |
| South King Early Intervention Program (SKIP) | *London ON* |
| UW Medicine/Valley Medical Center | Thames Valley Children's Centre |
| Waypoint Pediatric Therapies | *Mississauga ON* |
| | ErinoakKids Centre for Treatment and Development |
| *Philadelphia* | *North Bay ON* |
| Atlantic County Special Services | One Kids Place Children's Treatment Centre |
| Children's Specialized Hospital | *Ottawa ON* |
| Cindy Miles & Associates | Ottawa Children's Treatment Centre |
| Good Shepherd Rehabilitation Network | *Peterborough, ON* |
| HMS School | Five Counties Children's Center |
| Kennedy Krieger Institute | *Prince Albert, SK* |
| Private Therapists | *Victoria Hospital Therapies Department* |
| Voorhees Pediatrics | *Regina, SK* |
| Weisman Children's Hospital and Rehabilitation Center | Wascana Rehabilitation Center |
| | *Simcoe York ON* |
| *Oklahoma* | Children's Treatment Network |
| Early Intervention | *St. John's NL* |
| Heart Springs School | Janeway Children's Health and Rehabilitation Centre |
| Private Therapists | *St. Catharines ON* |
| | Niagara Children's Centre |
| *Georgia* | *Surrey BC* |
| Private Therapists | The Centre for Child Development |
| | *Timmins ON* |
| | Cochrane Temiskaming Children's Treatment Centre |
| | *Toronto, ON* |
| | Holland Bloorview Kids Rehabilitation Hospital |
| | *Vancouver, BC* |
| | B.C.'s Centre for Ability |
| | Sunny Hill Center (part of BC Children's Hospital) |
| | *Victoria BC* |
| | Queen Alexandra Center for Children's Health |
| | *Windsor ON* |
| | John McGivney Children's Centre |
| | *Winnipeg, MN* |
| | Children's Hospital of Winnipeg |
| | Provincial Outreach Therapy for Children |
| | St Boniface Hospital |
| | Winnipeg Rehabilitation Center for Children |

## Measures

*Demographic information*

We collected demographic information about children and families including: child age at entry to the study, child gender, child race and ethnicity, child age, parent respondent's relationship to child, parent highest education level achieved, and parent race and ethnicity. Race and ethnicity questions were adapted from the 2010 United States Census and the 2011 Statistics Canada Census. We also collected information about the number of children and adults living in the home and total household income. We did not collect information on the type of motor disorder because of known difficulties with reliability of this classification system,[40] but we did collect information on the limb distribution of CP.

The outcomes for this study have been identified by families of children with CP.[17-19] Measures were chosen by consensus among the academic researchers, based on their previous findings about determinants (child, family, rehabilitation, and community services) of gross motor function, self-care, and participation in community and recreational activities in children with CP 1.5-5 years of age,[18,19] 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.[18] A short description of each measure follows.

*Functional Classifications*

Gross Motor Function Classification System (GMFCS)[29]

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 general descriptions of a child at 6 to 12 years of age are: 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 five levels vary by age of the child.

Manual Ability Classification System (MACS)[30]

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 general descriptions for each level are: 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)[31]

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 general descriptions for each level are: I: Effective sender/receiver with familiar/unfamiliar partners, II: Effective but slower-paced sender and/or receiver with familiar/unfamiliar partners, III: Effective sender and receiver with familiar partners, IV: Inconsistent sender and/or receiver with familiar partners, and V: Seldom effective sender and receiver with familiar partners.

*Impairments and Associated Health Conditions*

Balance, a primary impairment of children with CP, was measured using the Early Clinical Assessment of Balance (ECAB).[41] The ECAB was developed in the Move & PLAY study to provide a measure appropriate for use with young children with CP across the spectrum of GMFCS ability levels. This new measure comprises seven items from the Automatic Reactions section of the Movement Assessment of Infants (MAI)[42] and six items from the Pediatric Balance Scale (PBS),[43] both of which have been shown to have adequate inter-rater reliability. It can be administered and scored in about 10 to 15 minutes, depending on the child's functional ability level. Items from the MAI include lateral head righting (left and right), head righting in extension, head righting in flexion, rotation in the trunk (left and right), equilibrium reactions in sitting (left and right), protective extension to the side (left and right), and protective extension backwards (left and right). Reponses to these are rated as: 0 (no response), 1 (partial response), 2 (incomplete or inconsistent response), or 3 (complete and consistent response), using variable descriptors for each item. Items from the PBS include six newly numbered 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 degrees, and 6) placing alternate foot on the step while standing unsupported. Each of these items is scored from 0 (cannot do) to 4 (fully completes). Items from the MAI and PBS are summed, after reweighting the PBS items 1 and 2 by 1.5, items 3 and 4 by 2.5, and items 5 and 6 by 4, to account for increased difficulty of execution. The ECAB total sum score will be used for analysis.

Range of Motion, a secondary impairment of children with CP, was measured using the Spinal Alignment and Range of Motion Measure (SAROMM).[44] 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 meaning normal alignment and range, with or without active correction, 1 meaning normal alignment and range with passive correction, and 2, 3, and 4 indicating fixed contractures that are "mild," "moderate," or "severe" based on specified cut points, and illustrated by photographs in the training manual. The SAROMM takes approximately 15 minutes to complete. The Spinal Alignment Subscale score is obtained by summing the four items; the Range of Motion and Extensibility Subscale score is obtained by summing the 22 items. A total SAROMM score is obtained by summing the two subscale scores. Items "not tested due to pain" will be treated as missing. The SAROMM average score will be used for analysis.

Muscle strength, another secondary impairment of children with CP, was measured using the Functional Strength Assessment (FSA) [45] 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). In training, we used suggestions by Gajdosik[46] to enhance our ability to obtain accurate strength assessments. The FSA takes 10 minutes to complete. As with the SAROMM, we sum the item scores to obtain an overall estimate, with low scores indicating greater deficits in muscle strength than high scores. The FSA average score will be used for analysis.

Endurance fitness, our final secondary impairment of children with CP, is typically defined as the time a person can persevere before exhaustion limits exercise involving rhythmic motions of large muscle groups. We measured the construct of endurance for activity from the perspective of level of energy using a newly-constructed, parent-rated, Early Activity Scale for

Endurance (EASE).[47] In the Move & PLAY study, we determined that just four items capture this construct well reducing the response burden, now taking only 5 minutes to complete. Items include: 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 one 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. These items are scored from 1 (Never) to 5 (Always). As with the previous two secondary impairments, we will use the EASE average score for analysis.

We obtained a second estimate of endurance using the 6-Minute Walk Test (6MWT) for children in GMFCS levels I, II, and III, once they were older than 3 years of age. This is a simple, submaximal clinical exercise test in which the distance walked under controlled conditions in 6 minutes is measured.[48] For young children, assessors hold the child's hand; for older ones, instructions provided by Maher and colleagues[49] were used to motivate the children to walk for 6 minutes. Speed is to be self-paced. Location of walking was variable; we tried to ensure that the terrain was level and flat. Distance walked was measured using a survey-measuring wheel. The 6MWT distance walked in feet will be used for analysis.

Impact of health problems was measured using the Child Health Conditions questionnaire.[24] We developed this tool to measure the extent to which health conditions influence children's activities, based on the new definition of CP[50] and body functions contained in the ICF.[26] Health conditions include problems with seeing, hearing, learning, communicating, controlling emotions, seizures, the mouth, teeth and gums, digestion, growth, sleeping, repeated infections, breathing, the skin, heart, and pain. Parents respond "yes" or "no" to each health problem listed and, if the child has a problem, are asked to 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); note an impact of 0 is imputed if the child does not have the problem. This measure is completed in 5 minutes. Analysis will be conducted based on the average impact of the health problems on daily life.

*Participation*

Participation was measured using two domains of the Child Engagement in Daily Life Measure (CEDL),[51,52] which is a 29-item questionnaire developed by the research team. Part one of the CEDL captures interaction with others and play; specifically, participation of the child in family/community life and leisure/recreational activities. This domain is scored on 4-point Likert scale for how often a child participates ("Very often" to "Never or Almost Never"), and on a 5-point scale for the degree of enjoyment ("A great deal" to "Not at all). For this study, data on frequency of participation were collected. Part two of the CEDL measures self-care, defined as the degree that the child participates in his or her daily self-care activities (feeding, dressing, bathing, and toileting). The 5-point Likert ratings for daily self-care activities (from "Does the activity independently most of the time" to "Does not do activity") distinguish the need for physical assistance of an adult and, for children who do not require adult assistance, whether the child is able to perform the activity consistently. The CEDL measure is completed in 10 minutes. The Rasched CEDL Participation and Self-Care scores will be used for analysis.

*Services*

Services Received.[20] Because the On Track study investigates *clinical course,* we collected data on the services provided, as well as on major medical and surgical interventions in the 6-month period preceding each data collection point. These data can be summarized across the study period as: number of primary care visits; number of medical service visits; number of medical and surgical procedures; amount (# of sessions) of physical, occupational, and speech and language therapy services; focus of therapy (in the categories of primary impairments, 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; number of community programs; coordination of care; and parents' perceptions that their children's needs were being met. This measure takes 10 minutes to complete.

*Physical Activity sub-study measures*

The following measurements were collected on a subgroup of participants within the Seattle, WA and Atlanta, GA study sites in order to acquire more specific measurement of activity amounts and levels of exertion.

Walking Activity Measurement: StepWatch[53]

For participants who were ambulatory, walking activity performance was measured in the context of daily life using a StepWatch monitor (Modus Health, Washington, DC), which is a small (70 x 50 x 20 mm; 38 g), waterproof, self-contained device. StepWatch data were collected within the physical activity sub-study on a subset of 50 participants in GMFCS Level I, II or II. Participants wore the StepWatch on their left ankle (inside a knit cuff) each day for seven days. Specific variables are the average daily step counts and percent time walking in low-, moderate-, and high-stride rates based on pediatric values for the seven-day sample. Of the commercially available monitors today, the StepWatch device has one of the highest levels of accuracy for detecting the movement of stepping across walking speeds.[54,55] The average single leg strides per day and the average strides per day faster than 30 per minute will be used for analysis.[56] The StepWatch takes 10 to 15 minutes to calibrate each time it is worn.

Physical Activity Measurement: ActiGraph[57]

ActiGraph data were collected within the physical activity sub-study on a subset of 79 participants. For all participants, physical activity was measured within the context of daily life with a 3-dimensional accelerometer (ActiGraph wGT3X) (ActiGraph LLC, Pensacola, FL). Participants wore the ActiGraph on their dominant wrist for a seven-day sample. The wrist mounted ActiGraph activity counts by axis were converted to waist worn raw activity counts[58] for calculation of average physical activity counts per minute and the minutes of moderate to vigorous physical activity will be used for analysis. The ActiGraph takes 5 minutes to calibrate each time it is worn.

1-minute and 6-minute walk test (1MWT, 6MWT)[49,59,60]

During the extra visits to the participants' home for the activity performance sub-study, both the 6MWT, explained above, and the 1MWT were completed. The 1MWT utilizes the same methods as for the 6MWT; however, the distance walked in 1 minute is also recorded. For exploratory purposes, 1MWT data were collected within the physical activity sub-study on a subset of children in GMFCS level I, II, or III, with a maximum of 40

participants at any time point. 1MWT distance walked in feet will be used for analysis. This measure takes 10 minutes with instruction and set up.

1-minute to 6-minute push test (1MPT, 6MPT)[61]

For exploratory purposes, 1MPT and 6MPT data were collected within the physical activity sub-study on a subset of children in GMFCS level II, III, or IV, with a maximum of 5 participants at any time point. The 1MPT to 6MPT are submaximal, clinical exercise tests in which the total distance propelled in a manual wheelchair in meters in 1minute and 6 minutes, under controlled conditions, are measured. In this study, for children who use a manual wheelchair for mobility, the 1MPT/6MPT were conducted indoors or outdoors on a large, flat, hard terrain. A survey measuring wheel was used to calculate the total distance wheeled and a stopwatch was used to keep track of the allocated time. Standardized directions were used to encourage the child to wheel as far as possible. The distance pushed in feet will be used for analysis. This measure takes 10 minutes including instruction and set-up.

1-stroke push test (1SPT)[61]

For exploratory purposes, 1SPT data were collected within the physical activity sub-study on a subset of children in GMFCS level III or IV, with a maximum of 4 participants at any time point. The 1SPT is a clinical exercise test in which the distance rolled in a manual wheelchair, under controlled conditions, with one push using both hands if possible, is measured. In this study a subsample of children who use a manual wheelchair for mobility were tested on this measure. A survey measuring wheel was used to calculate the total distance wheeled. Standardized directions were used to encourage the child to wheel as far as possible. The distance rolled in feet will be used for analysis. This measure takes 5 minutes including instruction and set-up.

Environment Section of the Participation and Environment Measure - Children and Youth (PEM-CY)[62]

PEM-CY environment data were collected within the physical activity sub-study on a subset of 79 participants. The PEM-CY environment section is a caregiver completed, 45-item questionnaire about the facilitators and barriers that might impact the child's participation in the home, school, and community environments. Twenty-five items include ratings on things that help or make it harder for the child to participate in activities in each environment (4-point scale: "not an issue," "usually helps," "sometimes helps/sometimes makes harder," "usually makes harder"). Twenty items include ratings of the availability of supports for the child's participation in each environment (4-point scale: "not needed," "usually, yes," "sometimes yes/sometimes no," "usually no"). Caregivers can also write in what family members do that help the child participate in each setting. A percentage score is given for each setting. The higher the percentage the more support the environment provides for the child's participation within the setting. The score used for analysis will be the percent total of parent perception that the home/community/school environment is supportive for participation in that setting. This measure takes 10 minutes to complete.

Table 5 contains a summary of the psychometric properties of the measures used, as well as details of who collected data at various study visits.

**Table 5: Psychometric Properties of the Measures Used**

| MEASURE | PSYCHOMETRIC PROPERTIES | TIMING |
|---|---|---|
| **Completed by Parent Respondent and Assessor** | | |
| **Gross Motor Function Classification System (GMFCS)[29,34]** | Content validity: confirmed via nominal group technique and Delphi survey.<br><br>Inter-rater reliability: Kappa = 0.75 for children older than 2 years | At Baseline, 12 Month, and 24 Month Visits |
| **Manual Ability Classification System (MACS)[30]** | Content validity: via consensus;<br><br>Inter-rater 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 Classification System (CFCS)[31]** | Content validity: confirmed via Delphi process;<br><br>Preliminary reliability: Inter-rater reliability, Weighted Kappa = 0.67; Test–retest reliability, Weighted Kappa = 0.84 | |
| **Completed by Assessor** | | |
| **Early Clinical Assessment of Balance[41]** | Content validity: (n = 410)[41] established through expertise on research team; Internal consistency: Cronbach's alpha = 0.92; Construct validity: known groups study: ECAB scores differed significantly among all GMFCS levels ($p < 0.001$); correlation with GMFM = 0.97 ($p < 0.001$); Children aged less than 31 months had significantly lower ECAB scores than children aged 31-42 or 43-60 months ($p < 0.01$); Factor Loading:[45] ECAB loaded most highly onto the Move & PLAY construct of "primary impairment" with a loading of 0.95<br><br>Reliability;[63] (n = 28 children with CP, aged 2-7 years); Inter-rater 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 = 10 | At Each Visit |
| **Spinal Alignment and Range of Motion Measure[44]** | 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's alpha = 0.95 (Move & PLAY, unpublished); Construct validity: age and GMFCS level contributed significantly to SAROMM score (r2 = 0.44); Known groups validity: scores differentiate children at all GMFCS levels, except II and III ($P < 0.006$);[45] Factor Loading:[45] the SAROMM loaded second most highly onto the Move & PLAY construct of 'secondary impairment' with a loading of 0.74<br><br>Inter-rater reliability: (n = 25; 5 in each GMFCS level) – ICC = 0.89 (95% CI = 0.76 | At Each Visit |

| | | |
|---|---|---|
| | – 0.95); test-retest reliability – ICC = 0.93 (95% CI = 0.86 – 0.97); SEM = 3; MDC95 = 9 | |
| **Functional Strength Assessment (FSA)[45]** | Construct validity: supported by similarity to standard methods of manual muscle testing in children; Internal consistency: Tested in Move & PLAY (n = 429) (unpublished), Cronbach's alpha = 0.93; Construct validity: (n = 429);[45] Known groups validity: significant difference among all GMFCS levels ($P < 0.001$), except for levels II & III; Factor Loading: FSA loaded most highly onto the Move & PLAY construct of 'secondary impairment' with a loading of 0.95.<br><br>Inter-rater reliability: Tested in Move & PLAY (n = 28 children with CP), ICC = 0.996 (95% CI = 0.991 – 0.998); Test-retest reliability: ICC = 0.97 (95% CI 0.95-0.99) | At Each Visit |
| **Six-minute Walk Test (6MWT)[48,49]** | Concurrent validity: with VO2 max = 0.44 ($P < .001$) (typical children 12-16 years);[49,64,65]<br><br>Test-retest reliability: ICC = 0.94 (95% CI 0.89-0.96) (typical children aged 12-16 years)[49] Test-retest reliability: ICC = 0.98 (children with CP aged 11- 17 years)[61] | At Each Visit |
| **Completed by Parent Respondent** | | |
| **Early Activity Scale for Endurance[47]** | **11-item version[47]** Construct validity: Known groups validity – significant differences among children developing typically and children with CP in 5 levels of GMFCS ($P < 0.001$); post hoc tests NS for levels II and III (n = 520); Internal consistency: Cronbach's alpha = 0.93; Convergent validity: Spearman's correlation with 6MWT = 0.57 ($P = 0.001$) (n = 14 children with CP and 14 children developing typically)<br>Test-retest reliability: ICC = 0.95 (95% CI = 0.90-0.98) (n = 32 children with CP); SEM = 2.9; MDC95 = 8.0<br><br>**4-item version**: (tested in Move & PLAY (n = 429), unpublished)<br>Good model fit: CFA – short version, X2 = 2.8 NS, CFI = 0.998, TLI = 0.993, RMSE = 0.03; Internal consistency: Cronbach's alpha = 0.83; Factor Loading:[45] the EASE loaded significantly onto the Move & PLAY construct of 'secondary impairment' with a loading of 0.66<br>Test-retest reliability: ICC = 0.75 (95% CI 0.54-0.87)<br>Convergent validity: (On Track, unpublished data), (n=376): GMFCS levels I-III, Pearson correlation of EASE to 6MWT = 0.30 (p<0.001); Construct validity: Significant differences between GMFCS levels I-III, Level I>II>III (p<0.03), between age groups, 1.5-3 years-olds > 6-9 and 9-12 year-olds (p=0.006, p=0.001) and 3-6 year-olds > 9-12 year-olds (p=0.006), between sex, boys > girls (p=0.02) | At Each Visit |
| **Child Health Conditions Questionnaire[24]** | Content validity: Developed from the international definition of CP[50] using the ICF;[26]<br>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&III, and IV&V) $P < 0.001$ (n = 537), post hoc testing: all groups significantly different from each other for number ($P < 0.01$); for impact, all groups significantly different from each other ($P < 0.001$) except for GMFCS Levels I and II&III<br><br>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) | At Each Visit |
| **Child Engagement in Daily Life Measure[51,52]** | Construct validity: (n = 429 in Move & PLAY and 110 children developing typically); Internal consistency: Cronbach's alpha<br>Participation = 0.86 (frequency), 0.91 (enjoyment), Self-care = 0.90; Known groups validity: frequency in and enjoyment of participation in recreation and self-care varied by age and GMFCS level (i.e. children developing typically, GMFCS I, GMFCS II & III, GMFCS IV & V) ($P < 0.001$), there was an age by motor ability interaction for self-care, with the youngest children performing less than the 2 older age groups ($P < 0.001$) in GMFCS levels I-III only. All motor ability groups performed significantly differently ($P < 0.001$).<br>Rasch analysis: Participation performed well; self-care has been improved by adding items of intermediate difficulty for use in the On Track study<br><br>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)<br><br>Analysis as an evaluative measure:[66] (n = 387):<br>Sensitivity to change over the period of 1 year: participation and self-care had significantly higher scores at the end of 1 year for children in GMFCS Levels I and II&III ($P < 0.01$); effect sizes for participation, were 0.22, 0.34 and 0.13 for children in GMFCS Levels I vs II and III vs IV & V, respectively; for self-care were 0.56, 0.58, and 0.08 for children in Levels I, II and III, and IV and V, respectively.<br><br>Psychometric properties of the new 29-item version (expanded and revised to be appropriate for children up to 12 years of age) are being re-calculated within the On Track Study. | At Each Visit |
| **Services Questionnaire[20]** | Content validity: via experienced clinician review,<br><br>Test-retest reliability: Amount of therapy visits ICC = 0.92; Focus of therapy services ICC = 0.55 – 0.95; Family Centeredness ICC = 0.86; Number 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 |
| **Physical Activity Sub-study Measures Completed by Assessor** | | |
| **StepWatch[53-56]** | Construct validity: StepWatch: A review of pedometers and accelerometers[55] reported that StepWatch is the most accurate pedometer ever designed for walking | Physical Activity Sub-Study maximum subsample of n=79 |

| | | |
|---|---|---|
| | and is capable of capturing actual strides taken to within +/- 3% for speeds from 1-5 mph;[54] Calibration stride count to manual count: Typically developing youth and children with CP ranged 97.7 to 101.4%.;[67] superior accuracy for stride counts as compared with waist-mounted pedometers during treadmill walking in lean and obese youth ages 10–12 years;[68] accuracy and precision of the StepWatch was documented for treadmill walking speeds up to 4 mph (ICC = 0.995).[69]<br><br>Test-retest reliability: stride curves from 5-day sample: *P* = 0.38 to 0.95;[67] StepWatch to manual count treadmill walking test-retest: ICC= 0.99558 ICC for X, Y, and Z axes > 70.9 | at any visit |
| **ActiGraph wGT3X[57]** | Construct validity: Feasible[70] and valid if worn for 7 days;[57,71,72] good validity compared with indirect calorimetry (r=.82 to .89) across studies, with differing definitions of count cut points for metabolic equivalent levels;[58,73,74] When wearing Actigraph on the hip, Evenson cut points provide valid estimates of time spent in MVPA in populations of ambulatory children with CP.[75] Using hip-mounted ActiGraphs, MVPA was greater in ambulatory youth with CP compared with youth who were nonambulatory.[70] In a similar study with adults with cerebral palsy, wearing ActiGraphs worn on the wrist, authors found different activity counts for non-ambulatory and ambulatory adults.[76]<br><br>Instrument reliability: ICCs =.83 to.98; Wrist-worn placement of the ActiGraph in typically developing children had good inter-device reliability (r = 0.72) and validity against indirect calorimetry (r = 0.8, p < 0.01). | Physical Activity Sub-Study maximum subsample of n=79 at any visit |
| **One-minute/Six-minute Walk Test (1MWT/6MWT)** | 6MWT described above.<br>1MWT: Convergent validity: with GMFM, r=0.92, p < 0.001 (children with CP)[59]<br><br>Test-retest reliability: ICC = 0.94 (children with CP)[77] | Physical Activity Sub-Study maximum subsample of n=79 at any visit |
| **One-minute/Six-minute Push Test (6MPT)** | Convergent validity: 6MPT to 1SPT (r=0.73; p<0.001), and 6MPT to heart rate during the 6MPT (r=0.29, p=0.014).<br><br>Test-retest reliability: ICC = 0.97 for 6MPT (children with CP)[61] | Physical Activity Sub-Study maximum subsample of n=79 at any visit |
| **One-Stroke Push Test (1SPT)** | Convergent validity: 6MPT to 1SPT (r=0.73; p<0.001<br><br>Test-retest reliability: ICC = 0.97 for 1SPT (children with CP)[61] | Physical Activity Sub-Study maximum subsample of n=79 at any visit |
| **Physical Activity Sub-study Measures Completed by Parent Respondent** | | |
| **(PEM-CY) Environment Section[62]** | Test-retest reliability: Environmental Section – "Supportiveness" 0.76 for the home, 0.87 for school, 0.96 for community. | Physical Activity Sub-Study maximum subsample of n=79 at any visit |

## Data Collection

*Study Timeline*

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,[29] the MACS,[30] and the CFCS,[31] as well as distribution of involvement. Both the parent and the assessor collected these data individually then went through a structured process to come to consensus about the child's final classification level which took approximately 15 minutes. Parents were asked to complete the three classification systems independently, prior to the assessor visit.[78] During the visit, parents and therapists discussed the classifications and the therapist documented: 1) immediate agreement with the parent, 2) consensus with the parent after discussion, or 3) disagreement with the parent. We generated guidelines to reconcile disagreements for research purposes.[78]

Information from parents was obtained via either paper booklets containing survey measures or, after the first visit, parents had the option to complete the survey measures online. Assessor data were collected on paper booklets. Completed paper booklets were sent by courier to the regional coordinators (US) or to the PI (Canada) where the data were entered into the online database (EmPOWER Health Research Inc.). All data were collected by the end of August 2016. All data entry was manually checked by a second research staff member to ensure accuracy. Data entry error rates were recorded and are summarized by region in Table 6. As can be seen, these error rates were low and all errors were corrected.

**Table 6: Data Entry Error Rates**

| | **Assessor Measures All Visits** (83 possible items) | | | **Parent Measures All Visits** (243 possible items) | | | **Services Questionnaire All Visits** (82 possible items) | | | **Family Demographics Baseline** (70 possible items) | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| **Site** | # errors | # books | % error rate | # errors | # books | % error rate | # errors | # books | % error rate | # errors | # 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** |

*Missing Data*

Various efforts were made to reduce missing data as much as possible during the study:

1) We had very little missing data, using most of the same measures within the Move & PLAY study.[18] 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 around those items and offered pro-active 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 assessors 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 assistant or research staff member. Any errors were corrected and documented in an Excel tracking file and summarized in a detailed chart by site, over time. Data entry error rates were reviewed two times/year 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 mailing a Family Newsletter from the team two times/year. We also tracked information about attrition via an attrition form within the EmPOWER database, i.e., 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 it appeared to be due to biased or unbiased reasons, and to make protocol decisions related to data collection and data entry. Data queries were tracked cumulatively in a chart and relevant information was communicated to regional coordinators and/or assessors as required.

*Missing Data Plan*

Once data were collected, we finalized how missing data were handled within the full dataset. Outcome scores were not calculated if any item had missing data. Missing outcomes scores were imputed using the mice package (Multivariate imputation by chained equations) in R.[79] Missing data were imputed only for those cases who attended an assessment. Data were not imputed for children lost to follow-up. Imputation order was according to the amount of missing data, with variables having the fewest missing cases imputed first. For continuous variables a mixed-effects random forest (MERF) method was used, via a custom R function based on the code of Hajjem.[80] 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 random effects were used to model within-child variability. The MERF algorithm is a fairly new development in imputation methods and is available only for continuous outcomes. Categorical outcomes were imputed 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 datasets is used to estimate the variability due to the imputation process. Analyses on multiple imputations were not done in our study since the MERF method is sampling-based and because so little between-imputation variance was observed. We imputed five datasets and chose imputation three as the final dataset for analysis. 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. Sensitivity analyses were not done in our study because a relatively small number of values were imputed; ranging from 35 to112 values out of 2, 713 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 five imputed datasets. The missing data codes with descriptors for missing data were also reviewed as a check for systematic bias in terms of cause.

*Data Analysis*

Previously, we demonstrated that when children with CP are considered a homogeneous group there is no relationship between age and gross motor development.[81] When children with CP are grouped by classification level, however, distinct patterns of development emerge that enable comparison of a child's development to children of the same age and classification level.[39,82] The GMFCS[29] will be used to group children for the trajectories and graphs for balance, muscle strength, joint range of motion, physical activity, endurance for activity, and health conditions. Preliminary analysis of children's GMFCS, MACS, and CFCS levels from the first study visit was done to identify whether there were combinations that might be used to create the developmental trajectories for participation in self-care and recreation activities; however, no consistent combinations of the three classifications systems emerged from the data.[83] In examining data we found that participation in self-care and recreational/leisure activities were better related to GMFCS than the other two classifications, so percentiles and trajectories will be produced from the GMFCS.

Body function and participation data from baseline, 6-, 12-, 18-, and 24-month visits will be analyzed by linear and nonlinear mixed-effects modeling to create longitudinal trajectories by functional classification levels. Data on body function and participation data from the baseline, 12-month and 24-month visits will be analyzed via quantile regression to construct cross-sectional reference percentiles to create percentile graphs for each measure by functional classification levels. The distribution of change in reference centiles will be estimated by describing the range of changes over 12 months observed for the middle 50% and 80% of children. These ranges of typical change can be used to

classify children into three development groups: those who change 'as expected,' 'more than expected,' and 'less than expected.' Differences in services profiles for these groups will be explored to understand how amount of physical, occupational, and speech therapy services, service focus, family-centeredness, and extent services met the children's needs, relate to children's development.

Data from all available time points will be used in the development of the longitudinal trajectories. For the centile curves only data from the baseline, 12-month, and 24-month visits will be used to ensure no repeated measurements on a child within an age group. Age groups will be in three-month bands in the tabulated centile tables. Once centiles are tabulated, the change in centile score from baseline to 12 months will be used to categorize children into performance profiles ('as expected,' 'more than expected,' or 'less than expected') and we will investigate the relationship between services and performance profiles.

## DISCUSSION

The On Track study will determine how children with CP progress in many aspects of their physical development and participation in daily life from 18 months to approximately 12 years of age. The information collected from this study will help therapists and parents monitor if a child is developing 'as expected' in his or her physical development and participation. Then, the results of this study can be used by health care professionals in collaboration with families to provide the services that are most beneficial and meaningful for each child and their family members.

Results of this study will lead to improved efficiency in services for children with CP as the focus and intensity for individual children will be informed by research on the characteristics of services received by children making more or less optimal progress. A survey of pediatric physical and occupational therapists in Quebec indicated that frequency and duration of therapy services varied by profession, characteristics of children, and practice setting.[84] Availability, accessibility, cost, and service setting are additional factors likely to impact decisions regarding PT and OT services.[85,86] Parents of children receiving rehabilitation programs incorporating group and community services expressed a desire for more individual services but reported quality was not influenced by the type and intensity of service.[87] Health care delivery will be improved by knowing the extent of the need for services (amount) and the area that those services should focus on.

We anticipate that our study findings, combined with those of the Move & PLAY study will provide evidence to support episodic services based on child and family readiness to work towards a goal that the child is capable of achieving. Similarly, for children whose trajectory is 'less than expected' we anticipate services will address contributing factors. This evidence-based focus should contribute to improving motor function, preventing the development of secondary impairments, and enhancing health, activity, and participation in self-care and recreation.

Members of our Parent Research Team have been directly involved in all aspects of the study, and most importantly, they have created knowledge translation products for families of children with CP. The products will provide highly clinically relevant reference values that are user-friendly to those providing services to children with CP and their families. With this information, families of children with CP will be able to better answer the questions: "What can I do to improve the outcomes that are most important to my child and family?"; and "How can clinicians and the care delivery systems work in helping me make the best decisions about my child's health and healthcare?"

## ACKNOWLEDGEMENTS

This work was supported through the Canadian Institutes of Health Research (MOP # 119276, 2012–2017) and the Patient Centered Outcomes Research Institute (PCORI) Award (# 5321, 2013–2016).

The funders have had no involvement in study design, data collection, data analysis, manuscript preparation, and/or publication decisions.

The statements presented in this work are solely the responsibility of the authors and do not necessarily represent the views of either the Canadian Institutes of Health Research or the Patient-Centered Outcomes Research Institute (PCORI), including its Board of Governors or Methodology Committee.

***Collaboration Group:***
***On Track Study Team:***

*Academic Researchers*
Doreen Bartlett, Canada
Sarah Westcott McCoy, United States
Lisa Chiarello, United States
Robert J. Palisano, United States
Lynn M. Jeffries, United States
Alyssa Fiss, United States
Jan Willem Gorter, Canada
Steven Hanna, Canada
Kristie Bjornson, United States
Cheryl Kerfeld, United States
Lisa Avery, Canada

*Study Coordinators*
Barb Galuppi, Canada
Monica Smersh, United States

*Parent Researchers*
Lisa Diller, United States
Paula Drew, United States
Nancy Ford, United States
Marquitha K. Gilbert, United States
tina hjorngaard, Canada
Kimberly Rayfield, United States
Barbara Sieck Taylor, United States

*Regional Coordinators*
Jennifer Miller, United States
Amanda Porter, United States
Tracy Stoner, United States
Erin English Wentz, United States
Allison Yocum, United States

We thank the participating children and families, whose continued involvement made this study possible. We also acknowledge the important contributions of the regional coordinators as well as the 90 therapists across North America who assessed children during the course of the study.

**REFERENCES**


1. Statistics Canada . Births and birth rate, by province and territory. Available at: http://www40.statcan.ca/101/cst01/demo04a-eng.htm. Accessed July 2, 2010.
2. Kirby RS, Wingate MS, Van Naarden Braun K, et al. Prevalence and functioning of children with cerebral palsy in four areas of the United States in 2006: A report from the Autism and Developmental Disabilities Monitoring Group. *Res Dev Disabil.* 2011;32:462-469.
3. Stanley F Blair E, Alberman E. *Cerebral Palsies: Epidemiology and Causal Pathways.* Mac Keith Press; 2000.
4. Strauss D, Shavelle R, Reynolds R, Rosenbloom L, Day S. Survival in cerebral palsy in the last 20 years: Signs of improvement? *Dev Med Child Neurol.* 2007;49:86-92.
5. Andersson C, Mattsson E. Adults with cerebral palsy: A survey describing problems, needs, and resources, with special emphasis on locomotion. *Dev Med Child Neurol.* 2001;43:76-82.
6. Ando N, Ueda S. Functional deterioration in adults with cerebral palsy. *Clin Rehabil.* 2000;14:300-306.
7. Bottos M, Feliciangeli A, Sciuto L, Gericke C, Vianello A. Functional status of adults with cerebral palsy and implications for treatment of children. *Dev Med Child Neurol.* 2001;43:516-528.
8. Murphy KP, Molnar GE, Lankasky K. Medical and functional status of adults with cerebral palsy. *Dev Med Child Neurol.* 1995;37:1075-1084.
9. Palisano RJ, Almarsi N, Chiarello LA, et al. Family needs of parents of children and youth with cerebral palsy. *Child Care Health Dev.* 2010;36:85-92.
10. King S Teplicky R, King G, Rosenbaum P. Family-centered service for children with cerebral palsy and their families: A review of the literature. *Semin Pediatr Neurol.* 2004;11:78-86.
11. Darrah J, Bartlett D. Dynamic systems theory and management of children with cerebral palsy: Unresolved issues. *Infants Young Children.* 1995;8:52-59.
12. Valvano J. Activity-focused motor interventions for children with neurological conditions. *Phys Occup Ther Pediatr.* 2004;24:79-107.
13. Darrah J, Law MC, Pollock N, et al. Context therapy: A new intervention approach for children with cerebral palsy. *Dev Med Child Neurol.* 2011;53:615-620.
14. Palisano RJ, Chiarello LA, King GA, et al. Participation-based therapy for children with physical disabilities. *Disabil Rehabil.* 2012;34:1041-1052.
15. National Dissemination Center for Children with Disabilities. Individuals with Disabilities Education Act, Part C, Early Intervention for Babies and Toddlers. Available at: http://nichcy.org/laws/idea/partc. Accessed 11/28/2012.
16. Øien I FB, Østensjo S. Goal-setting in paediatric rehabilitation: Perceptions of parents and professionals. *Child Care Health Dev.* 2009;36:558-565.
17. Vargus-Adams JN, Martin LK. Domains of importance for parents, medical professionals and youth with cerebral palsy considering treatment outcomes. *Child Care Health Dev.* 2011;37:276-281.
18. Bartlett DJ, Chiarello L, McCoy SW, et al. The measurement model of the Move & PLAY study: The protocol of a study using comprehensive outcomes research in rehabilitation. *Phys Ther.* 2010;90:1660-1672.
19. Chiarello LA Palisano R, Bartlett DJ, McCoy SW. A multivariate model of determinants of changes in motor abilities and engagement in self-care and play of young children with cerebral palsy. *Phys Occup Ther Pediatr.* 2011;31:150-168.
20. Palisano RJ, Begnoche DM, Chiarello LA, et al. Amount and focus of physical therapy and occupational therapy for young children with cerebral palsy. *Phys Occup Ther Pediatr.* 2012;32:368-382.
21. Bailes AF, Reder R, Burch C. Development of guidelines for determining frequency of therapy services in a pediatric medical setting. *Pediatr Phys Ther.* 2008;20:194-198.
22. Arpino C, Vescio MF, De Luca A, Curatolo P. Efficacy of intensive versus nonintensive physiotherapy in children with cerebral palsy: A meta-analysis. *Int J Rehabil Res.* 2010;33:165-171.
23. Chiarello LA, Palisano RJ, Maggs JM, et al. Family priorities for activity and participation of children and youth with cerebral palsy. *Phys Ther.* 2010;90:1254-1264.
24. Wong C, Bartlett DJ, Chiarello LA, Chang HJ, Stoskopf B. Comparison of the prevalence and impact of health problems of preschool children with or without cerebral palsy. *Child Care Health Dev.* 2012;38:128-138.
25. Tosi LL Maher N, Moore DW, Goldstein M, Aisen ML. Adults with cerebral palsy: A workshop to define the challenges of treating and preventing secondary musculoskeletal and neuromuscular complications in this rapidly growing population. *Dev Med Child Neurol.* 2009;51(suppl 4):2-11.
26. World Health Organization (2001). International Classification of Functioning, Disability, and Health. Geneva, Switzerland.
27. Haak P, Lenski M, Hidecker MJ, Li M, Paneth N. Cerebral palsy and aging. *Dev Med Child Neurol.* 2009;51(suppl 4):16-23.
28. Kembhavi G, Darrah J, Payne K, Plesuk D. Adults with a diagnosis of cerebral palsy: A mapping review of long-term outcomes. *Dev Med Child Neurol.* 2011;53:610-614.
29. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. *Dev Med Child Neurol.* 2008;50:744-750.
30. Eliasson AC Krumlinde-Sundholm L, Rösblad B, et al. The Manual Ability Classification System (MACS) for childen with cerebral palsy. Scale development and evidence of validity and reliability. *Dev Med Child Neurol.* 2006;48:549-554.
31. Hidecker MJC, Paneth N, Rosenbaum PL, et al. Developing and validating the Communication Function Classification System (CFCS) for individuals with cerebral palsy. *Dev Med Child Neurol.* 2012;54:737-742.
32. Nelson KB, Ellenberg JH. Children who "outgrew' cerebral palsy. *Pediatrics.* 1982;69:529-536.
33. Gorter JW, Ketelaar M, Rosenbaum P, Helders PJM, Palisano R. Use of the GMFCS in infants with CP: The need for reclassification at age 2 years or older. *Dev Med Child Neurol.* 2008;51:46-52.
34. Palisano R, Rosenbaum P, Walter S, et al. Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Dev Med Child Neurol.* 1997;39:214-223.
35. Portney LG Watkins MP. *Foundations of Clinical Research. Applications to Practice.* 2nd ed. Upper Saddle River, NJ: Prentice Hall Health; 2000.
36. LaForme Fiss AC, Westcott McCoy S, Bartlett DJ, et al. Sharing of lessons learned from multi-site research. *Pediatr Phys Ther.* 2010;22:408-416.
37. On Track Study website. Available at: https://www.canchild.ca/en/research-in-practice/current-studies/on-track.
38. Crawford JR, Garthwaite PH. On the "optimal" size for normative samples in neuropsychology: Capturing the uncertainty when normative data are used to quantify the standing of a neuropsychological test score. *Child Neuropsychol.* 2008;14:99-117.
39. Rosenbaum PL, Walter SD, Hanna SE, et al. Prognosis for gross motor function in cerebral palsy: creation of motor development curves. *JAMA.* 2002;288:1357-1363.
40. Gorter JW, Rosenbaum PL, Hanna S, et al. Limb distribution, type of motor disorder and functional classification of cerebral palsy: How do they relate? *Dev Med Child Neurol.* 2004;46:461-467.
41. McCoy SW, Bartlett DJ, Yocum A, et al. Development and validity of the Early Clinical Assessment of Balance for young children with cerebral palsy. *Dev Neurorehabil.* 2014;17:375-383.
42. Chandler LS, Andrew M, Swanson MW. Movement Assessment of Infants: A Manual. Rolling Bay, WA: P.O Box 46311980.
43. Franjoine MR, Gunther J, Taylor MJ. Pediatric Balance Scale: A modified version of the Berg Balance Scale for the school-aged child with mild to moderate motor impairment. *Pediatr Phys Ther.* 2003;15:114-128.
44. Bartlett DJ, Purdie B. Testing of the Spinal Alignment and Range of Motion Measure: A discriminative measure of posture and flexibility for children with cerebral palsy. *Dev Med Child Neurol.* 2005;47:739-743.
45. Jeffries L, Fiss A, McCoy SW, Bartlett DJ. Description of primary and secondary impairments in young children with

cerebral palsy. *Pediatr Phys Ther.* 2016;28:7-14.
46. Gajdosik CG. Ability of very young children to produce reliable isometric force measurements. *Pediatr Phys Ther.* 2005;17:251-257.
47. Westcott McCoy S, Yocum A, Bartlett DJ, et al. Development of the Early Activity Scale for Endurance (EASE) for children with cerebral palsy. *Pediatr Phys Ther.* 2012;24:232-240.
48. Li AM, Yin J, Yu CC, et al. The six-minute walk test in healthy children: reliability and validity. *Eur Respir J.* 2005;25:1057-1060.
49. Maher CA, Williams MT, Olds TS. The six-minute walk test for children with cerebral palsy. *Int J Rehabil Res.* 2008;31:185-188.
50. Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol Suppl.* 2007;49(suppl109):8-14.
51. Chiarello L, Chang H, and Move & PLAY. Child engagement in daily life of young children with cerebral palsy. *Dev Med Child Neurol.* 2009;51(suppl 5):72-73.
52. Chiarello LA, Palisano RJ, McCoy SW, et al. Child Engagement in Daily Life:A measure of participation for young children with cerebral palsy. *Disabil Rehabil.* 2014;36:1804-1816.
53. StepWatch 2015; Available from: https://modushealth.com/ Accessed Nov 17, 2015.
54. Bassett DR, John D. Use of pedometers and accelerometers in clinical populations: Validity and reliability issues. *Phys Ther Rev.* 2010;15:135-142.
55. Karabulut M, Crouter SE, Bassett DR, Jr. Comparison of two waist-mounted and two ankle-mounted electronic pedometers. *Eur J Appl Physiol.* 2005;95:335-343.
56. Bjornson KF, Zhou , C, Stevenson, R.D, Christakis, D. Relation of stride activity and participation in mobility-based life habits among children with cerebral palsy. *Arch Phys Med Rehabil.* 2014;95:360-368.
57. Actigraph Products. Available from: http://www.theactigraph.com. Accessed 2/2012.
58. Evenson KR, Catellier DJ, Gill K, Ondrak KS, McMurray RG. Calibration of two objective measures of physical activity for children. *J Sports Sci.* 2008;26:1557-1565.
59. McDowell BC, Kerr C, Parkes J, Cosgrove A. Validity of a 1 minute walk test for children with cerebral palsy. *Dev Med Child Neurol.* 2005;47:744-748.
60. MacKay A. One Minute Walk Test in cerebral palsy. Personal communication; 2008.
61. Verschuren O, Ketelaar M, De Groot J, Vila Nova F, Takken T. Reproducibility of two functional field exercise tests for children with cerebral palsy who self-propel a manual wheelchair. *Dev Med Child Neurol.* 2013;55:185-190.
62. Coster WJ, Bedell G, Law M, et al. Psychometric evaluation of the Participation and Environment Measure for Children and Youth (PEM-CY).*Dev Med Child Neurol.* 2011;53:1030-1037.
63. Randall KE, Bartlett DJ, McCoy SW. Measuring postural stability in young children with cerebral palsy: a comparison of 2 instruments. *Pediatr Phys Ther.* 2014;26:332-337.
64. Fitzgerald D, Hickey C, Delahunt E, Walsh M, O'Brien T. Six-Minute Walk Test in children with spastic cerebral palsy and children developing typically. *Pediatr Phys Ther.* 2016;28:192-199.
65. Nsenga Leunkeu A, Shephard RJ, Ahmaidi S. Six-minute walk test in children with cerebral palsy gross motor function classification system levels I and II: Reproducibility, validity, and training effects. *Arch Phys Med Rehabil.* 2012;93:2333-2339.
66. Palisano R, Chiarello L, McCoy SW, Bartlett D, An M. Use of the Child Engagement in Daily Life and Ease of Caregiving for children to measure change in young children with cerebral palsy. *Phys Occup Ther Pediatr.* 2015;35:280-295.
67. Bjornson KF, Song K, Zhou C, et al. Walking stride rate patterns in children and youth. *Pediatr Phys Ther.* 2011;23:354-363.
68. Mitre N, Lanningham-Foster L, Foster R, Levine JA. Pedometer accuracy for children: Can we recommend them for our obese population? *Pediatr.* 2009;123:e127-131.
69. Bjornson K, Song K, Lisle J, et al. Measurement of walking activity throughout childhood: Influence of leg length. *Pediatr Exerc Sci.* 2010;22:581-595.
70. Gorter JW, Noorduyn SG, Obeid J, Timmons BW. Accelerometry: A feasible method to quantify physical activity in ambulatory and nonambulatory adolescents with cerebral palsy. *Int J Pediatr.* 2012;2012:329284.
71. Trost SG. State of the Art Reviews: Measurement of physical activity in children and adolescents. *Am J Lifestyle Med.* 2007;1:299-314.
72. Trost SG, McIver KL, Pate RR. Conducting accelerometer-based activity assessments in field-based research. *Med Sci Sports Exerc.* 2005;37(suppl 11):S531-543.
73. Puyau MR, Adolph AL, Vohra FA, Zakeri I, Butte NF. Prediction of activity energy expenditure using accelerometers in children. *Med Sci Sports Exerc.* 2004;36:1625-1631.
74. Wen LM, van der Ploeg HP, Kite J, Cashmore A, Rissel C. A validation study of assessing physical activity and sedentary behavior in children aged 3 to 5 years. *Pediatr Exerc Sci.* 2010;22:408-420.
75. Clanchy KM, Tweedy SM, Boyd RN, Trost SG. Validity of accelerometry in ambulatory children and adolescents with cerebral palsy. *Eur J Appl Physiol.* 2011;111:2951-2959.
76. Claridge EA, McPhee PG, Timmons BW, et al. Quantification of physical activity and sedentary time in adults with cerebral palsy. *Med Sci Sports Exerc.* 2015;47:1719-1726.
77. McDowell BC, Humphreys L, Kerr C, Stevenson M. Test-retest reliability of a 1-min walk test in children with bilateral spastic cerebral palsy (BSCP). *Gait Posture.* 2009;29:267-269.
78. Bartlett DJ, Galuppi B, Palisano RJ, McCoy SW. Consensus classifications of gross motor, manual ability, and communication function classification systems between therapists and parents of children with cerebral palsy. *Dev Med Child Neurol.* 2016;58:98-99.
79. van Buuren S, Groorhuis-Oudshoorn K. Multivariate imputation by chained equations in R. *J Stat Software.* 2011;45:1-67.
80. Hajjem A, Bellavance F, Larocque D. Mixed-effects random forest for clustered data. *J Stat Comput Sim.* 2012;84:1313-1328.
81. Palisano RJ, Hanna SE, Rosenbaum PL, et al. Validation of a model of gross motor function for children with cerebral palsy. *Phys Ther.* 2000;80:974-985.
82. Hanna SE, Rosenbaum PL, Bartlett DJ, et al. Stability and decline in gross motor function among children and youth with cerebral palsy aged 2 to 21 years. *Dev Med Child Neurol.* 2009;51:295-302.
83. Bartlett D, Dyszuk E, Galuppi B, Gorter JW. Inter-relationships of functional status in children with cerebral palsy: A descriptive study and an extension. *Pediatr Phys Ther.* 2018:30:1-16.
84. Mazer B, Feldman D, Majnemer A, Gosselin J, Kehayia E. Rehabilitation services for children: Therapists' perceptions. *Pediatr Rehabil.* 2006;9:340-350.
85. Majnemer A, Shevell MI, Rosenbaum P, Abrahamowicz M. Early rehabilitation service utilization patterns in young children with developmental delays. *Child Care Health Dev.* 2002;28:29-37.
86. Weindling AM, Cunningham CC, Glenn SM, Edwards RT, Reeves DJ. Additional therapy for young children with spastic cerebral palsy: A randomised controlled trial. *Health Technol Assess.* 2007;11:iii-iv, ix-x, 1-71.
87. Camden C, Swaine B, Tetreault S, Brodeur MM. Reorganizing pediatric rehabilitation services to improve accessibility: Do we sacrifice quality? *BMC Health Serv Res.* 2010;10:227.

To cite this article: Sarah Westcott McCoy, Doreen Bartlett, Monica Smersh, Barbara Galuppi & Steven Hanna. Collaboration Group: On Track Study Team (March 2018). Monitoring development of children with cerebral palsy: the On Track study. Protocol of a longitudinal study of development and services. Available at: https://www.canchild.ca/en/resources/294-monitoring-development-of-children-with-cerebral-palsy-the-on-track-study-protocol-of-a-longitudinal-study-of-development-and-services.