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Developmental Trajectories and Reference Percentiles for Range of Motion, Endurance, and Muscle Strength of Children With Cerebral Palsy.

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Title: Developmental trajectories and reference percentiles for range of motion, endurance, and muscle strength of children with cerebral palsy.

Short Title: Development Trajectories for the SAROMM, EASE, and FSA

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Abstract

Background: Children with cerebral palsy (CP) frequently present with secondary impairments in spinal alignment and extremity range of motion, endurance for activity, and muscle strength. Creation of developmental trajectories for these impairments will help guide clinical decision-making.

Objective: For children in each level of the Gross Motor Function Classification System (GMFCS) this study aimed to: 1). Create longitudinal developmental trajectories for range of motion (Spinal Alignment and Range of Motion Measures (SAROMM)), endurance (Early Activity Scale for Endurance (EASE)), and functional strength (Functional Strength Assessment (FSA)), and 2). Develop age-specific reference percentiles and amount of change typical over one-year for these outcomes.

Design: Longitudinal Cohort Design

Method: 708 children with CP across GMFCS levels, 18-months up to the 12th birthday and their families participated in 2-5 assessments every 6 months over 2 years. Trained therapists performed the SAROMM and FSA and parents completed the EASE questionnaire. For children in each GMFCS level, longitudinal trajectories using linear and nonlinear mixed-effects models from all visits and reference percentiles using quantile regression from the first, 12-month, and 24-month visits were created for each measure.

Results: Longitudinal trajectories and percentile graphs for SAROMM, FSA, and EASE are primarily linear, with different performance scores among GMFCS levels. Much variability in both longitudinal trajectories and percentiles exists within GMFCS levels.

Limitations: Limitations include a convenience sample, and varying numbers of participants assessed at each visit.

Conclusions: The longitudinal trajectories and percentile graphs have application for monitoring how children with CP are performing and changing over time compared with other children with CP. The resources presented allow therapists and families to collaboratively make decisions about intervention activities targeted to children's unique needs.

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Introduction

Children with cerebral palsy (CP) frequently present with primary impairments in muscle tone, postural stability, and motor coordination that are present at the time of diagnosis.[1,2] These primary impairments can lead to the development of secondary impairments of body function and structure as the children age.[1,3] Common secondary impairments in children with CP are decreased range of motion (ROM), endurance, and muscle strength, which are conceptualized as secondary impairments due to the known changes over time.[2,4] These secondary impairments have been identified in children as young as 18 months and across all functional motor ability levels.[3] Therapy for children with CP often focuses on the prevention or amelioration of these impairments using evidence-based interventions; therefore, knowledge of developmental trajectories for range of motion, endurance, and muscle strength is important.

Range of motion restrictions are common in children with CP.[5] In a population-based study (N=119; 6-19 year old),[6] 60% of the children with CP had ROM limitations in their lower and/or upper extremities. Research has noted an inverse relationship between children's functional motor ability and ROM restrictions.[3,5] Additionally, ROM was reported to decrease in children with CP between 2 and 14 years, with more pronounced restrictions for children with lower functional ability level.[7] Therefore, a means of monitoring change over time is important for determining intervention needs and strategies for prevention.

Compared to typically developing peers, children with CP are more sedentary and engage in fewer physical activities, particularly as they age.[8,9] In young children with CP (1.5-5 years old), better endurance was related to higher motor abilities.[10] Children with CP have demonstrated the ability to improve their physical activity and endurance following exercise and sports programs,[11,12] underscoring the importance of monitoring endurance for activity.

Children with CP have difficulty in force production contributing to impairments in muscle strength.[13,14] Young children with CP (1.5-5 years old) have demonstrated strength impairments that increased as functional mobility decreased.[3] Muscle strength can improve with strengthening exercises in children with CP[15,16] therefore, therapists are encouraged to monitor children's muscle strength and develop focused intervention programs.

Given the early development of secondary impairments in young children with CP, evidence-based resources for therapists and families to monitor changes in impairments over time is needed to support informed intervention planning. Longitudinal developmental trajectories provide information on a child's average prognosis for a particular impairment, and reference percentiles provide an understanding of a child's development relative to other children in the same GMFCS level as well as a system to interpret this change over time. Therefore, the purpose of this study was to create longitudinal developmental trajectories and reference percentiles for ROM, endurance and strength of children with CP grouped by Gross Motor Function Classification System (GMFCS) levels.[17]

Methods

This study was part of a multisite, prospective cohort study entitled 'Developmental Trajectories of Impairments, Associated Health Conditions, and Participation of Children with Cerebral Palsy' (short title: On Track Study)' which aimed to develop longitudinal developmental trajectories and reference percentiles for impairments, health conditions, and participation variables for children with CP.[18] Children were assessed across one to two years. Institutional Review Boards (IRBs) at all participating institutions and recruitment sites with IRBs reviewed and provided ethics approval. All parents or guardians provided informed consent and children, as appropriate and in compliance with the specific IRB, provided assent.

Participants

Participants were a convenience sample of 708 children with CP ages 18-months up to the 12th birthday, GMFCS levels I-V. Families agreed to be followed two times (baseline and 12 months later) or five times (baseline, 6, 12, 18, 24 months later). While 708 children with CP participated in the baseline testing, 656 completed at least 2 assessments (baseline and 12 months) and 424 participated in up to 5 assessment sessions. Complete details of the number of participants at each assessment session can be found in eFigure 1.[18] Children were recruited from six provinces across Canada, including British Columbia, Saskatchewan, Manitoba, Ontario, Nova Scotia, and Newfoundland, and four regions of the United States, including areas within and surrounding Georgia, Oklahoma, Pennsylvania, and Washington states. Participating children had a diagnosis of CP by a physician or demonstrated delay in gross motor development in addition to impairments consistent with a diagnosis of CP. Children were excluded if their parents were unable to speak and understand English, French or Spanish.

The proportion of children in each GMFCS level is: GMFCS I – 32.1%; GMFCS II – 22.7%; GMFCS III – 11.2%; GMFCS IV – 18.2%; GMFCS V – 15.7%. Our distribution is comparable to data reported by Reid and colleagues.[19] Attrition was tracked across all study visits and is documented in eFigure 1.[18] Demographic information of the children and their families is included in eTable 1.[18]

Within this study of the clinical course of a large group of children with CP the amount and focus of the rehabilitation services that children received was not controlled; however, parents completed a services questionnaire at each assessment estimating the services received in the previous 6 or 12 months. On average, children received variable amounts of PT, OT and SLP services ranging from 0 to >156 sessions per year, with most children receiving 0-30 sessions per year. Detailed data on the amount and focus of services and analysis of the relationship of services to the children ‘s development are the focus of other reports.

Measures

Gross Motor Function Classification System (GMFCS)

The GMFCS is a five-point classification system used to describe gross motor function ability in children with CP. Distinctions between levels are made based on functional abilities, use of assistive technology, and quality of movement.[17] Content validity, construct validity, and inter-rater reliability have been previously supported.[17,20,21]

Spinal Alignment and Range of Motion Measure (SAROMM)

The SAROMM was developed to provide an estimate of overall *spinal alignment* and *range of motion and muscle extensibility* of children with CP.[22] The assessor observes the child’s sitting posture and alignment, scoring four *spinal alignment* items, using a 5-point ordinal score of 0 (“no alignment limitations with active correction”) to 4 (“Fixed” – limitation is structural, static, not reducible and severe).[22] The assessor then positions the child in supine for twenty lower extremity *range of motion and muscle extensibility* items and then positions the child into sitting for two upper extremity items. Scoring for the lower and upper extremity items uses a 5-point ordinal score of 0 (“normal” - no restrictions of ROM on passive testing and no postures typical of some children with CP observed) to 4 (“fixed” – limitation is structural, static, and irreducible and is severe).[23] The SAROMM has good inter-rater (ICC = 0.89, 95%CI = 0.76-0.95) and excellent test-retest (ICC = 0.93, 95%CI = 0.86-0.97) reliability with a standard error of measurement of 3.09 points.[22] Validity is supported by the contribution of GMFCS level and age to the SAROMM total score (r^2=0.44).[22] The mean of all item scores for each child was used for analysis.

Early Activity Scale for Endurance (EASE)

The EASE is a parent-completed assessment of the child’s perceived endurance for activity.[23] Parents rate four questions related to their child’s (1) physical activity related to peers, (2) physical energy level and need to take break, (3) frequency of breathing quickly and getting flushed during activity, and (4) frequency of daily activities requiring a lot of physical energy. Items are scored on a 5-point ordinal scale of (1 = Never; 5 = Always), with higher scores indicating greater endurance for activity. The EASE is moderately correlated (Spearman r = 0.41, *P* = 0.01) with the Six-Minute Walk test and has good test-retest reliability (ICC = 0.79, 95%CI = 0.62- 0.89).[3] The total score mean of all items was used for analysis.

Functional Strength Assessment (FSA)

The FSA provides an estimate of strength for major muscle groups for children as young as 18 months.[3] The assessor encourages the child to perform eight movements against gravity, providing resistance if possible. The muscle groups examined include the neck and trunk flexors and extensors, and bilateral hip extensors, knee extensors, and shoulder flexors. Therapy knowledge, skills, and creativity are used to encourage the movements and estimate the child's muscle strength. Multiple testing trials are allowed, based on judgment of the child's cooperation, to score the child's best performance for each muscle group. Items are scored on a 5-point ordinal scale ranging from 1 (only flicker of contraction or just initiates movement against gravity) to 5 (full available range against gravity and strong resistance).[3] The FSA has excellent inter-rater reliability (ICC = 0.996, 95%CI = 0.991- 0.998).[3] This method of muscle strength testing is similar to standard manual muscle testing of individual muscles supporting the FSA's construct validity. The mean of all the muscle group item scores was used for analysis.

The SAROMM, EASE, and FSA forms and training protocols (voice-over power point presentations for SAROMM and FSA) can be accessed through the CanChild website (http://www.canchild.ca), on the On Track study webpage.

Procedures

Portions of the overall procedures in this study have been reported elsewhere.[18] Specifics realted to these variables are described briefly. Prior to data collection, physical or occupational therapist assessors completed onsite training on the study and administration of measures. Therapists completed videotaped criterion tests of the SAROMM and FSA. Each therapist assessor obtained greater than or equal to 80% item agreement with the study investigators providing ‘gold standard’ responses.

Children were assessed in their home or clinic settings**.** The assessors completed the GMFCS via consensus with parents.[24] The GMFCS was independently completed by both the assessor and the parent, and then the child’s classification was discussed in attempt to reach consensus. Consensus was reached 97.8% of the time, and all disagreements were within one level.[24] The final classification used was the parent rating with specific rules applied to determine if the assessor classification should be used instead.[24] Therapists then completed the SAROMM and FSA, and parents completed the EASE.

Data Analysis

Longitudinal Developmental Trajectories

SAROMM and EASE

Using data from all 708 chidren (eFigure 1), both nonlinear and linear mixed effects models[25] were fit to the SAROMM and EASE longitudinal data, modelling score as a function of age; separately for each GMFCS level. Linear models were fit centering at five years of age (60 months) with random effects modelled for the intercept and slope parameters. Linear models are described by Model 1 in the e-supplement. Nonlinear models were used to model scores approaching a functional limit as age increases; random effects were modelled for the limit and rate of change parameters. Non-linear models are described in Model 2 of the e-supplement. Choice between models was dictated by a desire for parsimony and by model fit, as assessed with Akaike’s information criterion (AIC) on the maximum likelihood (ML) estimates. Although model selection was based on AIC of ML estimates the final models were fit using restricted maximum likelihood estimation (REML) to achieve better estimates of the random effects.[26] Models were fit using the nlme package in R.[25]

FSA

Inspection of the raw data indicated different trajectories based on GMFCS level. Scores of children in GMFCS Levels I and II increased and then plateaued while scores of children in levels III-V exhibited linear trends. Non-linear asymptotic models were fit to GMFCS levels I and II and simple age-centered linear models, as with the SAROMM and EASE, were fit to levels III-V. As with the SAROMM and EASE, random effects were fit to estimate the variability in trajectories among children.

Reference Percentiles

The reference percentiles describe the distribution of the SAROMM, EASE, and FSA scores at each age within each GMFCS level. To calculate reference percentiles, the authors used up to three observed scores, (first, 12-month, and 24 month visits) of each measure from each of the 708 children with no repeated measurements on a child within 9 months (eFigure 1). Each observation contributed to a cross-sectional (age-specific and GMFCS-specific) sectional reference percentile that was estimated using quantile regression (QR). The quantregGrowth package in R was used, which uses linear combinations of multiple basis functions to estimate smooth quantiles across the age continuum and constrains the percentiles to be non-crossing.[26] These reference percentiles describe the distribution of SAROMM, EASE, and FSA scores at each age and were computed for the five GMFCS levels.

Using these reference percentiles, which determine a child's percentile score based on their age and GMFCS level, we calculated percentile scores for all children with baseline and 12-month assessments. The amount of change in each child's percentile score over this 12-month period was calculated by subtracting the baseline centile score from the 12-month centiles score. The distribution of 12-month change scores was used to estimate bands that encompass 50% (range 25-75% change scores) and 80% (range 10-90% change score) of changes. These bands quantify the amount of change in percentiles that is typical in this clinical population. Following Hanna et al,[27] we recommend that children whose percentile changes are within the 80% limits can usually be described as 'developing as expected' for their age and GMFCS levels.

Results

Predicted average values of the SAROMM, EASE and FSA based on longitudinal models with bootstrapped confidence intervals are presented in eTable 2. Longitudinal developmental trajectories for the SAROMM, EASE and FSA by GMFCS level are shown in Figure 1. Accompanying model parameters for the SAROMM, EASE and FSA longitudinal developmental trajectories are in Tables 1-3. The longitudinal developmental trajectories provide average ability for a children with CP based on GMFCS level which will assist with prognosis.

Spinal alignment/range of motion (SAROMM) scores showed trends for average development in children to have increases in limitations at all functional ability levels, especially for children at GMFCS level V. The SAROMM predicted means values indicated very few overlaps of 95% CIs across GMFCS levels, indicating differences in ROM restrictions between functional levels except for young children in GMFCS levels IV and V. At younger ages, children in GMFCS levels IV and V present with similar ROM restrictions, which diverge as they get older, with children at GMFCS level V presenting with greater ROM restrictions.

Children with CP demonstrate a slight increase in endurance for activity (EASE) scores between ages 2 and 5 years, with endurance remaining relatively stable over the age of 5. In general, as children’s functional levels decrease, their average endurance for activities also decreases. However, there is some overlap noted in 95% CI in children in Levels II and III, and III and IV at the younger ages.

When examining strength (FSA scores), children in GMFCS levels I and II follow a similar average pattern to the EASE, with greater changes between 2 and 5 years than between 5 and 12 years. FSA in children in GMFCS levels III and IV follow a linear trend upward, meaning average scores for strength increase as children age, while FSA in children at GMFCS level V remains stable. Due to the continued increase in strength for children in GMFCS level III there is overlap noted in children at GMFCS levels II and III at younger ages.

The estimated reference curves for the SAROMM, EASE and FSA at each GMFCS level, plotted at the 3rd, 5th, 10th, 25th, 50th, 75th, 90th, 95th, and 97th percentiles are shown in Figure 2. Additional versions of these figures and the tabulated percentiles are available on the On Track study website: https://www.canchild.ca/en/research-in-practice/current-studies/on-track. Table 4 provides the mean and standard deviation of the change in percentile score over a one-year period by GMFCS level, along with the range of the central 50% and 80% of change scores.

Reference percentiles for the SAROMM, EASE and FSA show the variability that exists in the outcomes across the ages of 1.5-12 years in children with CP within all GMFCS levels. On the SAROMM, variability increases as children's functional abilities decrease (random effects residual SD = 0.12 [GMFCS I] - 0.27 [GMFCS V], Table 1). On the EASE, children at GMFCS level IV had the most variability (random effects residual SD = 0.53, Table 2), while on the FSA children at GMFCS levels I (random effects residual SD = 0.29, Table 3) and V (random effects residual SD = 0.24, Table 3) had less variability than children in GMFCS levels II-IV (random effects residual SD = 0.32-0.37, Table 3).

Discussion

Overall for the estimated outcomes of spinal alignment/ joint range of motion, endurance for activity, and strength as children age, there are small increases in average spinal alignment/ joint range of motion and endurance for activity limitations, and average strength. Unlike gross motor ability, which typically shows a more rapid rate of development in the early years,[20] only approximately 30% of the progression in spinal alignment/joint range of motion, endurance for activity, and strength (GMFCS levels III-V), occurred between 2-5 years as compared to between 5-12 years changes. This finding suggests that children across the ages of 1.5 to 12 years need monitoring related to these outcomes.

Variability in children's scores exists on all three secondary impairment measures, even when categorized by GMFCS. Due to this variability, therapists and families should use the longitudinal developmental trajectories to support prognosis in a broad context only. They can use the longitudinal developmental trajectories to discuss how a child with CP is developing in spinal alignment/joint range of motion, endurance for activity, and strength in relationship to the average performance of other children with CP with the same funcational ability.

To demonstrate the use of the longitudinal developmental trajectories, Joshua (pseudonym, GMFCS IV) was tested at age 4 years 0 months on the SAROMM, EASE, and FSA. At this visit, his SAROMM score was 0.8, his EASE score was 2.5, and his FSA score was 3.6. Using Figure 1, his SAROMM score is below the average trajectory for children at level IV, indicating less ROM restriction than his same aged peers, which is good but should be monitored, as ROM restrictions on average do increase slightly with age. His EASE score is just below the average trajectory for children at level IV, and over time, his endurance score is expected to remain stable. His FSA score is above the average trajectory for children at level IV, indicating strength is better than his same aged peers and should on average increase slightly over time.

The reference percentiles provide an understanding of a child's individual strengths and limitations related to the secondary impairments with specificity of how far above or below the 50^{th} percentile a child is performing. Using the reference percentiles at one time point, therapists can identify areas of strength, relative to peers in a similar manner to the use of the longitudinal trajectories, to develop a strengths-based intervention. Different from comparisons using the longitudinal curves, one can be more specific about how far from the 50^{th} percentile (middle of the average range) the child is functioning. Again, it is important to note that on the SAROMM, lower scores are indicative of fewer ROM restrictions, so lower percentile scores indicate less impairment. For the EASE and FAS, higher reference percentile scores indicate less impairment.

Completion of a subsequent assessment allows the therapists to determine how the impairments change over time within the individual child. Comparing the individual child's initial and subsequent reference percentiles allows a therapist to determine if the child's secondary impairments are progressing 'as expected,' 'more than expected,' or 'less than expected,' based on the child's initial score, age, and functional ability level. Change in individual percentiles between the 10-90% of the reference sample are labeled to be progressing "as expected," with change in percentiles from the initial score lower than the lowest 10% of the reference sample considered "less than expected," and change greater than highest 10% considered "more than expected."

Returning to Joshua, his initial SAROMM score (4 years 0 months) was 0.8, placing him at the 30-35th percentile, and at 5 years 0 months, his reassessment score was 0.7, placing him at the 20-25th percentile. Children of his age and GMFCS level change between -33 and +17 percentiles (Table 4) while remaining in the "as expected" (10-90% of reference sample) category. Joshua's change of -15 (35 minus 20) to -5 (30 minus 25) percentiles indicates that his ROM is progressing "as expected" even with the improvement (decrease) in his percentiles. Joshua's EASE first and follow-up scores of 3.6 and 3.0, respectively, translate into reference percentiles of 40% at the initial assessment and of 5% at the reassessment. This percentile drop of -35% represents a negative change that is greater than what the middle 80% of children at his age and GMFCS (-30 to +36; Table 4) demonstrate, signifying that his endurance is progressing "less than expected". Joshua's FSA score was initially 3.6 and then subsequently 3.0 one year later. His reference percentiles decreased from 90% to 65%, a difference of -25%. This percentile drop placed him just within the 10-90% of the reference sample range of percentile change for children at his age and GMFCS level (-27 to +36; Table 4), suggesting that he is still progressing "as expected," even with the decrease in percentile. These results suggest that collaborative intervention planning may emphasize activities to improve endurance, while focusing on strategies to maintain ROM and strength, potentially leveraging these areas of strength to support endurance activities.[28]

The example highlights the variability present in development in children with CP. Because of the variability, therapists and families can better interpret children's progress based on translation of raw test scores to percentiles according to the child's age and GMFCS level. Knowing that variability exists, large changes in reference percentiles are not unusual over 12 months. That said, decisions about services should always be supplemented with an analysis of the child's function and input from the child and family.

Limitations

This study sample is one of convenience; however, the proportion of children at each GMFCS level is comparable to international incidence data.[19] All participants did not complete all five assessments leading to a variation in the number of children included at each time point. Given that actual change in scores of the SAROMM, EASE, and FSA are relatively small and percentiles graphs reflect the large variability of scores for children with CP, interpretations from use of these data should be employed carefully with collaboration of therapists' and families' knowledge of the children. Finally, this was a study of clinical course, not natural history and the amount and focus of rehabilitation services that families sought and received within their communities was not controlled for within the development of the longitudinal trajectories and the percentile graphs.

Conclusion

Data on average development of estimated spinal alignment/joint range of motion, endurance for activity and strength by functional ability levels has been created for children with CP, age 1.5 to 12 years. Therapists can now compare performance on the secondary impairments of ROM, endurance for activity, and strength in children with CP to the average performance of same age peers with similar abilities. When used appropriately, the longitudinal developmental trajectories can assist with prognosis. Reference percentiles have also been created and can be used to establish status relative to similar children at specific GMFCS levels and ages and to monitor change in secondary impairments over time. Therapists and families can use the data to clearly identify the child's strengths and potential needs and proactively plan services. For children with CP, tracking secondary impairments and targeting interventions at opportune times may enhance the child's ability to participate in home, educational, and community activities.

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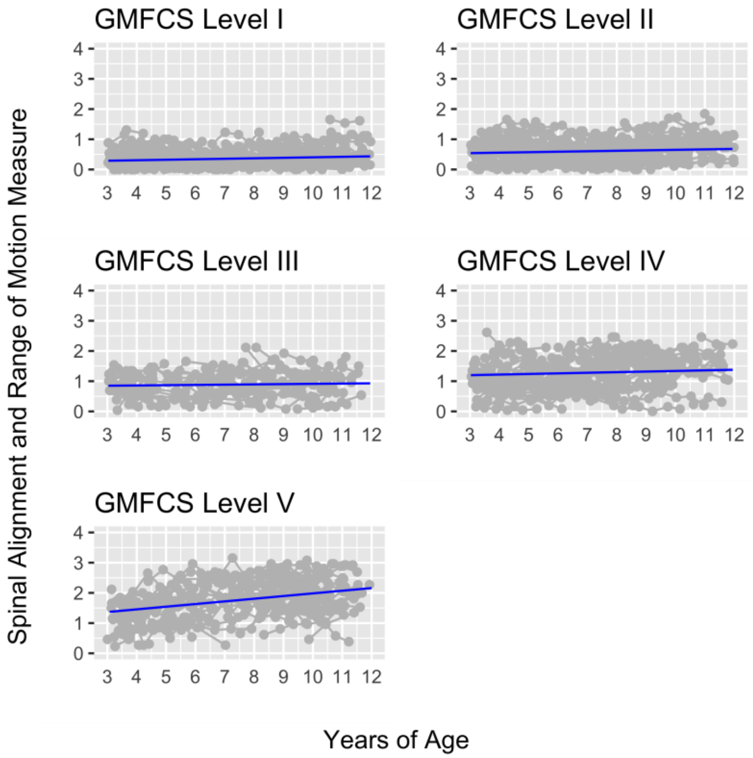
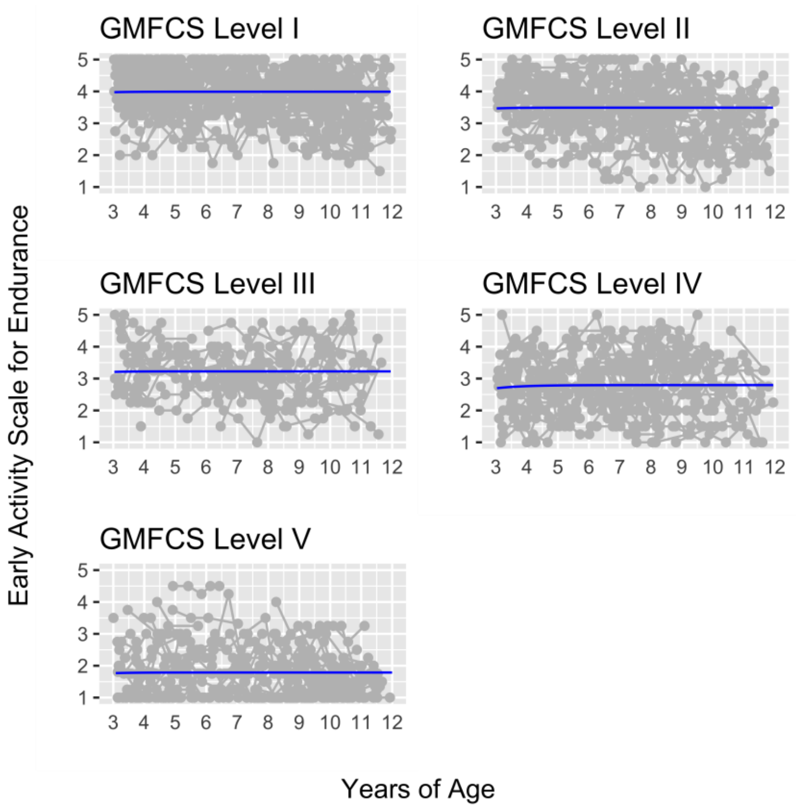
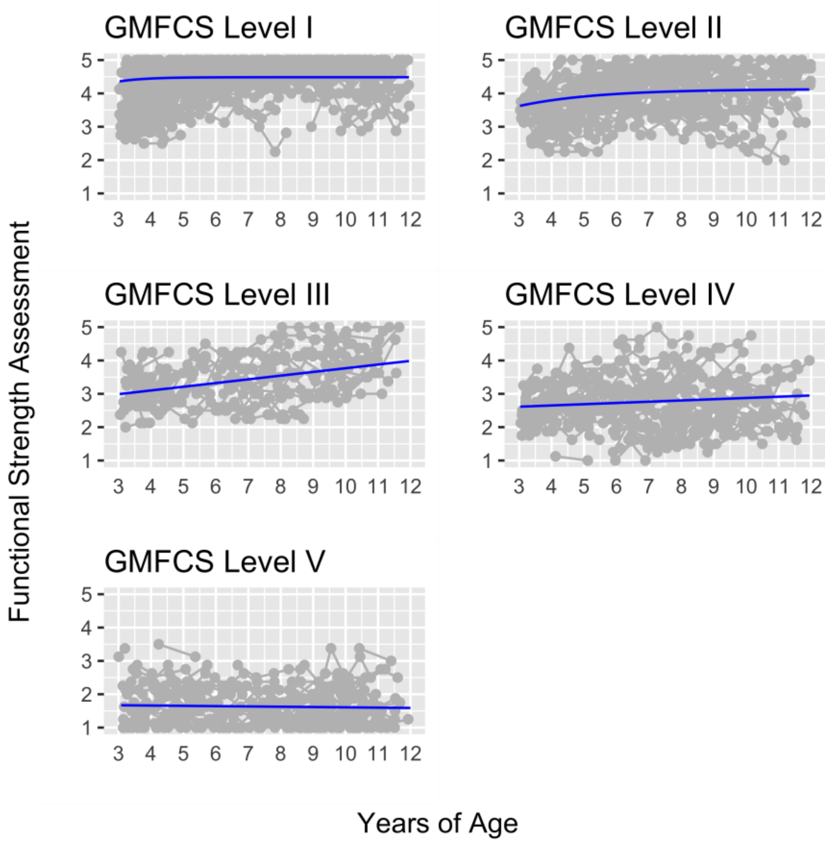
Figure 1. Longitudinal Developmental Trajectories for Range of Motion, Endurance, and Strength by GMFCS Level



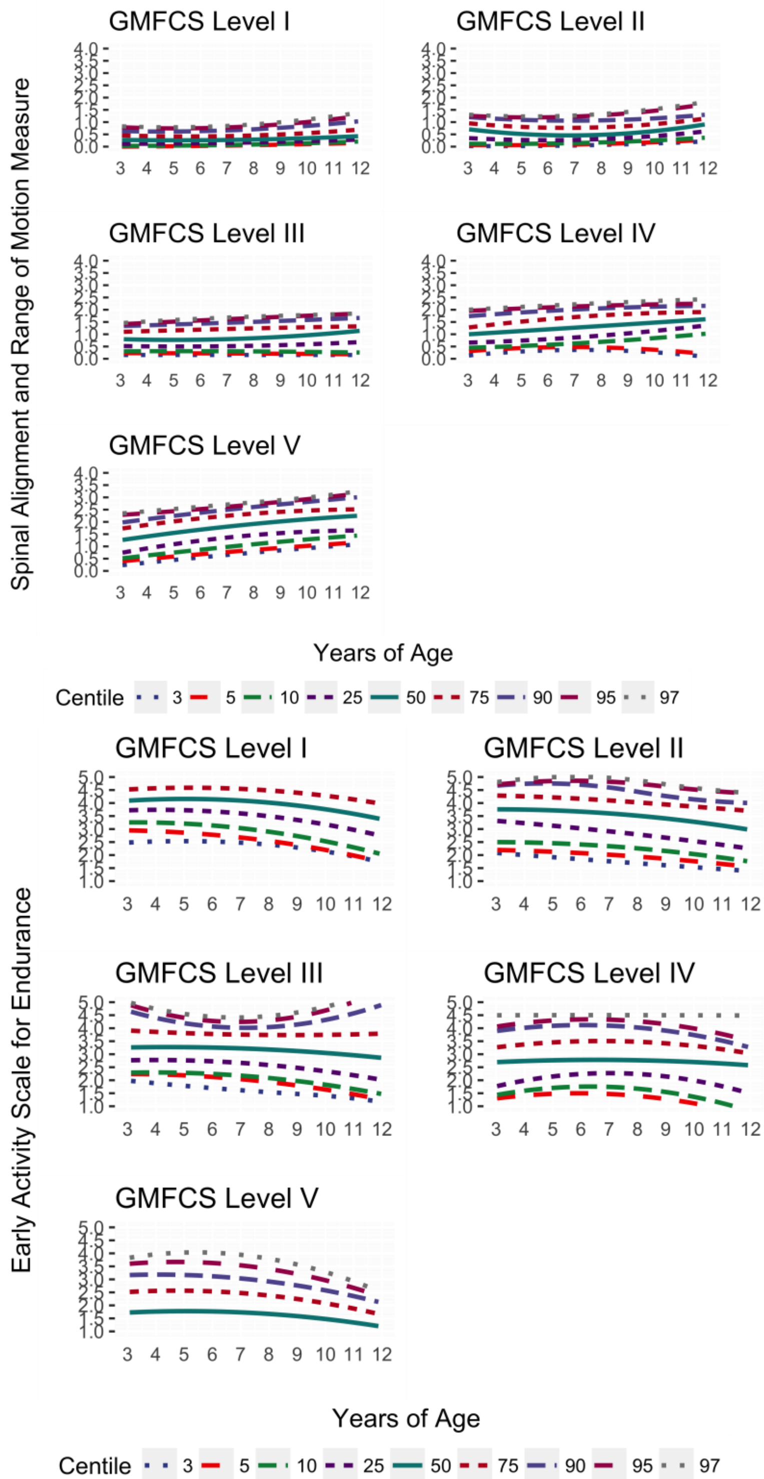


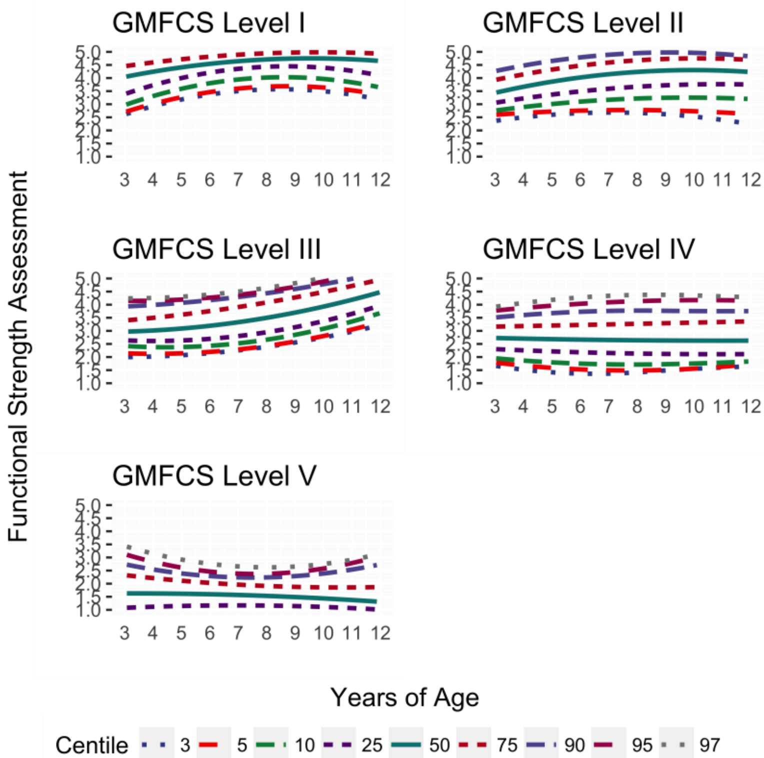
GMFCS Level I
GMFCS Level II
GMFCS Level III
GMFCS Level IV
GMFCS Level V
Early Activity Scale for Endurance
Years of Age



GMFCS Level I
GMFCS Level II
GMFCS Level III
GMFCS Level IV
GMFCS Level V
Functional Strength Assessment
Years of Age

Figure 2. Reference Percentiles for Range of Motion, Endurance, and Strength by GMFCS Level





GMFCS Level I
GMFCS Level II
GMFCS Level III
GMFCS Level IV
GMFCS Level V
Functional Strength Assessment
Years of Age
Centile 3 5 10 25 50 75 90 95 97

Table 1: Model parameters describing the linear change in SAROMM scores as a function of Age in months.

	Level I	**Level II**	**Level III**	**Level IV**	**Level V**
Fitted model	**Model 1**	**Model 1**	**Model 1**	**Model 1**	**Model 1**
Fixed Effects					
Intercept	0.32	0.57	0.87	1.24	1.54
(95% CI)	(0.29, 0.36)	(0.51, 0.64)	(0.78, 0.96)	(1.14, 1.34)	(1.43, 1.66)
Slope	0.001	0.001	0.001	0.002	0.007
(95% CI)	(0.001, 0.002)	(0.000, 0.003)	(-0.001, 0.003)	(-0.001, 0.004)	(0.005, 0.010)
Random Effects					
Residual SD	0.12	0.18	0.23	0.24	0.27
50% Ranges[a]					
Intercept	(0.18, 0.46)	(0.35, 0.79)	(0.64, 1.10)	(0.93, 1.55)	(1.20, 1.88)
Slope	(-0.001, 0.004)	(-0.002, 0.005)	(0.000, 0.002)	(-0.004, 0.007)	(0.004, 0.011)

[a] Expected range of the parameter for the central 50% of the population

Table 2: Model parameters describing the non-linear change in EASE scores as a function of Age in months. See Model 2 in the e-supplement for more details about the models.

	Level I	**Level II**	**Level III**	**Level IV**	**Level V**
Fitted model	**Model 2**	**Model 2**	**Model 2**	**Model 2**	**Model 2**
Fixed Effects					
Limit[a]	3.99	3.49	3.23	2.80	1.79
(95% CI)	(3.90, 4.08)	(3.36, 3.61)	(3.05, 3.39)	(2.64, 2.95)	(1.65, 1.93)
Age-90[b]	14.99	16.66	15.66	24.96	18.51
(95% CI)	(9.13, 24.62)	(8.59, 32.30)	(6.38, 38.43)	(15.04, 41.42)	(6.97, 49.10)
Random Effects					
Residual SD	0.43	0.41	0.46	0.53	0.47
50% Ranges[c]					
Limit	(3.51, 4.34)	(2.94, 3.94)	(2.72, 3.67)	(2.27, 3.30)	(1.37, 2.26)
Age-90	(14.99, 15.00)	(16.65, 16.67)	(15.66, 15.66)	(24.94, 24.99)	(18.45, 18.56)

[a] The asymptotoic limit as children age

[b] The time required (in months) to attain 90% of the limit.

[c] Expected range of the parameter for the central 50% of the population

Table 3: Model parameters describing the linear (Levels III-V) and non-linear (Levels I-II) change in FSA scores as a function of Age in months. See the e-supplement for more details about the models.

	Level I	**Level II**	**Level III**	**Level IV**	**Level V**
Fitted model	**Model 2**	**Model 3**	**Model 1**	**Model 1**	**Model 1**
Fixed Effects					
Intercept (score at age 5yrs)			3.21	2.69	1.65
(95% CI)			(3.08, 3.34)	(2.57, 2.81)	(1.53, 1.77)
Slope (change in score per month)			0.009	0.003	-0.001
(95% CI)			(0.006, 0.013)	(0.000, 0.006)	(-0.003, 0.002)
Limit[a]	4.49	4.13			
(95% CI)	(4.42, 4.55)	(3.96, 4.30)			
Age90[b]	23.4	66.63			
(95% CI)	(20.30, 26.98)	(34.69, 127.97)			
Offset[c]		-24.62			
(95% CI)		(-56.56, 7.33)			
Random Effects					
Residual SD	0.29	0.32	0.37	0.36	0.24
50% Ranges[d]					
Intercept			(2.91, 3.52)	(2.30, 3.07)	(1.31, 2.00)
Slope			(0.003, 0.015)	(-0.003, 0.010)	(-0.008, 0.006)
Limit	(4.12, 4.71)	(3.73, 4.53)			
Age-90		(66.60, 66.66)			
Offset		Not estimated			

[a] The asymptotoic limit as children age [b] The time required (in months) to attain 90% of the limit. [c] The offset parameter is a nuisance parameter required to achieve better model fit but without a useful interpretation. [d] Expected range of the parameter for the central 50% of the population

Table 4. Mean and standard deviation of change in percentile score over a one-year period by GMFCS level.

	GMFCS				
	Level I	Level II	Level III	Level IV	Level V
Spinal Alignment and Range of Motion Measure					
N	217	147	73	116	103
Mean Centile Change	-2	0	-4	-7	-3
SD Centile Change	23	23	23	20	21
Range 25-75% Change Scores	-13, +11	-16, +14	-12, +11	-16, +3	-14, +4
Range 10-90% Change Scores	-31, +24	-29, +28	-30, +20	-33, +17	-26, +21
Early Activity Scale for Endurance					
N	217	147	73	116	103
Mean Centile Change	5	6	8	1	3
SD Centile Change	23	21	22	27	24
Range 25-75% Change Scores	-6, +18	-4, +22	-4, +20	-12, +19	-10, +13
Range 10-90% Change Scores	-24, +36	-22, +30	-20, +36	-30, +36	-23, +39
Functional Strength Assessment					
N	217	147	73	116	103
Mean Centile Change	3	1	-1	3	4
SD Centile Change	23	22	24	24	18
Range 25-75% Change Scores	-7, +15	-10,+13	-18, +13	-10, +16	-7, +14
Range 10-90% Change Scores	-25, +30	-28, +25	-30, +29	-27, +36	-17, +27

eFigure 1: On Track Study Participant Flow Diagram (reprinted with permission)[18]

CIHR-Funded BASELINE

Started (n = 724)

Completed (n = 708)

Not Completed (n = 16)
- Death (n=1)
- Lost to follow-up=4
- Ineligible (n = 11)

PCORI-Funded 6-MONTH

Started (n = 708)

Completed (n = 471)

Not Completed (n = 237) * missed or withdrawn
- Death (n = 1)
- Withdrawal by subject (n = 3)
- Lost to follow-up (n = 15)
- Medical issue/illness (n = 2)
- Not able to schedule in timeframe (n = 23)
- 6-month N/A, child is in 2-visit study (n =193)

CIHR-Funded 12-MONTH

Started (n = 692)

Completed (n = 656)

Not Completed (n = 36) * missed or withdrawn
- Death (n = 1)
- Withdrawal by subject (n = 2)
- Lost to follow-up (n = 22)
- Medical issue/illness (n = 2)
- Not able to schedule in timeframe (n = 9)

PCORI-Funded 18-MONTH

Started (n = 668)

Completed (n = 454)

Not Completed (n = 214) * missed or withdrawn
- Death (n = 2)
- Withdrawal by subject (n = 2)
- Lost to follow-up (n = 15)
- Not able to schedule in timeframe (n = 13)
- 18-month N/A, child is in 2-visit study (n =182)

PCORI-Funded 24-MONTH

Started (n = 470)

Completed (n = 424)

Not Completed (n = 46) * missed or withdrawn
- Death (n = 1)
- Withdrawal by subject (n = 3)
- Lost to follow-up (n = 9)
- Medical issue/illness (n = 1)
- Not able to schedule in timeframe (n = 32)

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.

eTable 1. Child and Parent Demographics (repinted with permission)[18]

		Participants		
		Baseline Completed n=708 (%)	**12-Month Completed n=656 (%)**	**24-Month Completed N=424 (%)**
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 Native	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)
	White	550 (79)	517 (80)	339 (81)
24-Month (n = 419)	Multi	26 (4)	22 (3)	12 (3)
Parent respondent ethnicity*	Hispanic	32 (5)	30 (5)	20 (5)
	Non-Hispanic	669 (95)	621 (95)	400 (95)

Baseline (n = 701) 12-Month (n = 651) 24-Month (n = 420-421)	Aboriginal Non-Aboriginal	20 (3) 681 (97)	16 (3) 635 (97)	5 (1) 416 (99)
Parent respondent age, years* Baseline (n=694) 12-Month (n = 644) 24-Month (n = 415)	Mean (SD)	37.8 (7.9)	37.9 (8.0)	37.4 (7.1)
Parent respondent relationship to child* Baseline (n = 704) 12-Month (n = 654) 24-Month (n = 423)	Mother Father Other	628 (89) 51 (7) 25 (4)	578 (88) 51 (8) 25 (4)	382 (90) 26 (6) 15 (4)
Parent respondent education* Baseline (n = 700) 12-Month (n = 650) 24-Month (n = 420)	High School or less Community College / Associate's Degree University	160 (23) 212 (30) 328 (47)	147 (23) 196 (30) 307 (47)	92 (22) 114 (27) 214 (51)
Family Income* Baseline (n = 594) 12-Month (n = 553) 24-Month (n = 363) (CAD or USD)	≥$75,000 $60,000 - $74,999 $45,000 - $59,999 $30,000 - $44,999 ≤$30,000	306 (52) 78 (13) 50 (8) 58 (10) 102 (17)	293 (53) 72 (13) 47 (8) 49 (9) 92 (17)	190 (52) 43 (12) 34 (9) 35 (10) 61 (17)
Family Composition Baseline (n= 667) 12-Month (n = 620) 24-Month (n =	Adults (mean, SD) Children (mean, SD)	2.1 (0.7) 2.3 (1.1)	2.1 (0.7) 2.3 (1.1)	2.1 (0.7) 2.3 (1.1)

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.

eTable 2. : Predicted average values of the SAROMM, EASE and FSA based on longitudinal models with bootstrapped confidence intervals

GMFCS					
	Level I	Level II	Level III	Level IV	Level V
Spinal Alignment and Range of Motion					
Predicted Values Mean (95% CI)					
2 years	0.3 (0.2, 0.3)	0.5 (0.4, 0.6)	0.8 (0.7, 1.0)	1.2 (1.1, 1.3)	1.3 (1.1, 1.4)
5 years	0.3 (0.3, 0.3)	0.6 (0.5, 0.6)	0.9 (0.8, 0.9)	1.2 (1.2, 1.3)	1.5 (1.4, 1.6)
12 years	0.4 (0.4, 0.5)	0.7 (0.6, 0.8)	0.9 (0.8, 1.1)	1.4 (1.2, 1.5)	2.2 (2.0, 2.3)
change 2 to 5 years	0.0 (0.0, 0.1)	0.0 (0.0, 0.1)	0.0 (0.0, 0.1)	0.1 (0.0, 0.1)	0.3 (0.2, 0.3)
change 5 to 12 years	0.1 (0.1, 0.2)	0.1 (0.0, 0.2)	0.1 (-0.1, 0.2)	0.1 (0.0, 0.3)	0.6 (0.5, 0.8)
Early Activity Scale for Endurance					
Predicted Values Mean (95% CI)					
2 years	3.9 (3.6, 4.0)	3.3 (3.0, 3.5)	3.0 (2.6, 3.3)	2.5 (2.2, 2.7)	1.6 (1.3, 1.8)
5 years	4.0 (3.9, 4.1)	3.5 (3.4, 3.6)	3.2 (3.1, 3.4)	2.8 (2.7, 2.9)	1.8 (1.7, 1.9)
12 years	4.0 (3.9, 4.1)	3.5 (3.4, 3.6)	3.2 (3.1, 3.4)	2.8 (2.7, 2.9)	1.8 (1.7, 1.9)
change 2 to 5 years	0.1 (0.0, 0.3)	0.2 (0.0, 0.5)	0.2 (0.0, 0.6)	0.3 (0.1, 0.6)	0.1 (0.0, 0.4)
change 5 to 12 years	0.0 (0.0, 0.0)	0.0 (0.0, 0.0)	0.0 (0.0, 0.0)	0.0 (0.0, 0.1)	0.0 (0.0, 0.1)
Functional Strength Assessment					
Predicted Values Mean (95% CI)					
2 years	4.1 (3.9, 4.2)	3.3 (2.9, 3.5)	2.9 (2.7, 3.1)	2.6 (2.4, 2.7)	1.7 (1.5, 1.8)
5 years	4.5 (4.4, 4.5)	3.9 (3.8, 4.0)	3.2 (3.1, 3.3)	2.7 (2.6, 2.8)	1.7 (1.6, 1.8)
12 years	4.5 (4.4, 4.5)	4.1 (4.0, 4.2)	4.0 (3.7, 4.3)	3.0 (2.7, 3.2)	1.6 (1.4, 1.8)
change 2 to 5 years	0.4 (0.3, 0.5)	0.6 (0.4, 1.0)	0.3 (0.2, 0.4)	0.1 (0.0, 0.2)	0.0 (-0.1, 0.1)
change 5 to 12 years	0.0 (0.0, 0.0)	0.2 (0.1, 0.4)	0.8 (0.5, 1.0)	0.3 (0.0, 0.5)	-0.1 (-0.3, 0.1)

eModel 1

$$y = \alpha + \beta(age\text{-}60)$$

The linear models has parameters α, the intercept and the estimated value of the outcome at age 5 (60 months) and β, the slope or rate of change of the outcome as the child ages, expressed in change per month of age.

eModel 2

$$y = \frac{y_{max}}{1 + e^{-L}}\left(1 - e^{-\lambda \cdot age}\right)$$

This asymptotic regression model has parameters L and λ. The function is constrained to stay below y_{max}, the maximum value of the outcome (y_{max} is 5 for the EASE and 4 for the SAROMM). The L and λ parameters are re-parameterized as *Limit* and Age_{90}, respectively, using the formulas below.

eModel 3

$$y = \frac{100}{1 + e^{-L}}\left(1 - e^{-\lambda(age\text{-}Offset)}\right)$$

As with Model 2, the two parameters are L and λ and are re-parameterized in the results as Limit and Age90. In this model the offset parameter is a nuisance parameter included to improve model fit, but with no useful interpretation.

Re-parameterizations

$Limit = \frac{100}{1+e^{-L}}$ is the asymptote of the ECAM score as age approaches infinity

$Age90 = \frac{-\log(1-0.9)}{e^{\lambda}}$ is the length of time required for 90% of the Limit to be achieved.

50% Ranges

50% Ranges represent the expected range of the parameter for the central 50% of the population and were calculated as fixed effect ± $z_{0.25}$ x random effect.