RESPONSIVENESS OF THE FUNCTIONAL MOBILITY SCALE FOR CHILDREN WITH CEREBRAL PALSY

SEREBRAL PALSİLİ ÇOCUKLARDA FONKSİYONEL MOBİLİTE SKALASININ YANIT BAŞARISI

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ABSTRACT

Objective: This study examined the ability of the Functional Mobility Scale (FMS) to detect minimal clinically important differences in mobility status in children with cerebral palsy (CP) having interventions.
Materials and methods: This prospective longitudinal study of instrument validation was conducted in a tertiary care paediatric hospital. A population-based sample of 84 children with CP aged 2-16 years (mean age 8.8 years), GMFCS levels I-IV recruited from CP clinics. Children had orthopaedic surgery for deformity correction and gait deviations, including single event multilevel surgery (SEMLS) or spasticity management with botulinum neurotoxin A (BoNT-A). Responsiveness of the FMS was examined using change scores and correlation with an external criterion, the Functional Assessment Questionnaire (FAQ) at four time points following the interventions. Comparison of the SEMLS and botulinum groups was used to examine the ability of the FMS to detect both change and stability in mobility status.
Results: Correlation of changes score between the FMS and FAQ showed six significant correlations, however the correlations were low overall (Spearman rho 0.01-0.36). More change on the FMS was seen in children who had SEMLS compared to those who had botulinum toxin injections.
Conclusion: The FMS was able to detect minimal clinically significant change in mobility in children with CP who had orthopaedic surgery and spasticity management.
Key words: Outcomes assessment, cerebral palsy, responsiveness, rehabilitation

ÖZET

Amaç: Bu çalışma tedavi gören serebral palsi (SP) li çocuklarda mobilite durumunda klinik olarak önemli en düşük değişimi tespit etmede Fonksiyonel Mobilite Skalasının (FMS) becerisini araştırdı.
Materyal-metod: Bu çalışma bir üçüncü basamak pediyatri hastanesinde yapılan propektif longitudinal validasyon çalışmasıdır. Çalışmaya SP kliniklerinden, populasyon tabanlı, SP tanısı almış yaş ortalamaları 2-16 yıl (ortalama 8.8 yıl), GMFCS düzeyi I-IV olan 84 çocuk alındı. Çocukların deformite ve yürüme anomalisi düzeltimi amaçlı tek aşamalı çok seviyeli cerrahi (SEMLS) veya botulinum neurotoxin A (BoNT-A) ile spastisite tedavisi öyküsü vardı. FMS nin yanıt başarısı değişim skorları ile ve tedavi sonrası dört noktada, dış bir kriter olarak seçilen Fonksiyonel Değerlendirme Sorgulaması (FAQ) arasındaki korelasyonun araştırılması ile değerlendirildi. SEMLS and botulinum gruplarının karşılaştırılması ile FMS'nin mobilite durumundaki stabilite ve değişimi yakalayabilmesi değerlendirildi.
Bulgular: FMS ve FAQ değişim skorları arasındaki korelasyon altı farklı korealsyon gösterdi ancak korelasyonlar genel olarak düşüktü (Spearman rho 0.01-0.36). SEMLS olan çocuklarda botulinum grubuna göre FMS'de daha fazla değişim gözlendi.
Sonuç: FMS ortopedik cerrhai ve spastisite tedavisi gören çocuklarda mobilitede minimum klinik anlamlı değişimi yakalayabilmektedir.
Anahtar kelimeler: Sonuç değerlendirimi, serebral palsi, yanıt başarısı, rehabilitasyon.

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INTRODUCTION

This study examined the responsiveness to change of the Functional Mobility Scale (FMS) in children with cerebral palsy (CP) following orthopaedic surgery and botulinum toxin injections. The FMS is a six level clinician administered self-report ordinal scale that rates mobility within the different environmental settings of the home, school and community based on the assistance children require (1) (Figure 1). It is an evaluative measure designed to measure the magnitude of longitudinal change in individuals or groups over time and to quantify treatment benefits (2). A fundamental property of evaluative instruments is proven responsiveness to change to ensure they can accurately detect change in function (2-8)

There is a lack of consensus in the literature regarding the definition of responsiveness (9, 10). For the purposes of this article responsiveness is defined as the ability of a measure to detect minimally clinical important differences in function when change is believed to have occurred (4). Internal responsiveness describes the ability of a measure to assess change over a pre-specified time frame, and external responsiveness reflects the extent to which changes in a measure over time relate to corresponding changes in a reference measure of health status (9). This study focuses on external responsiveness because it has a broader application by concerning the measure itself, rather than the treatment under investigation (9). An important aspect of assessing responsiveness is determining what constitutes a minimally clinical important difference (MCID) (4). The MCID is not a fixed property of the instrument and depends on each particular question being investigated. It requires a judgement, which is often subjective, as to what constitutes clinically important change (10). The MCID for the FMS has not previously been investigated.

The aim of this study was to examine the ability of the FMS to detect minimal clinically important differences in mobility status, where change occurs, in children with CP having interventions. It was hypothesized that the FMS would be able to detect change and that it would show initial deterioration in mobility and subsequent improvement for children having major multilevel surgery and relative stability in mobility for children having injections of botulinum neurotoxin A (BoNT-A).

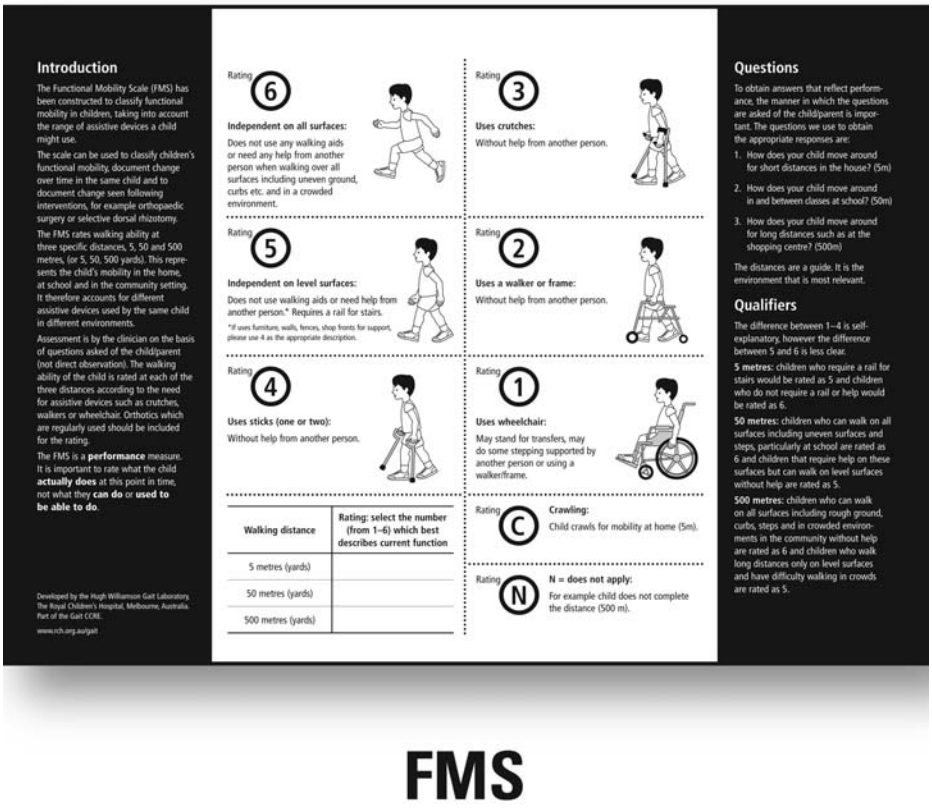

Figure 1. The Functional Mobility Scale

METHOD

This prospective longitudinal study used a consecutive sample of children. The study was approved by the institutional ethics committee of the centre where the study was performed as well as the associated university ethics research committee. Children were recruited consecutively from the orthopaedic CP clinics and gait laboratory of a tertiary care institution. All eligible children were invited to participate by the first author prior to their interventions. Written consent was obtained from each participating child's parent or legal guardian.

Included were children with CP aged 2-18 years, classified as Gross Motor Function Classification System (11) levels I-IV, who were scheduled to have orthopaedic surgery or spasticity management between January 2006 and May 2007. Excluded were children whose parents were not able to understand what was required of them due to cognitive or language problems.

The interventions included in the study were; 1) bilateral single event multilevel surgery (SEMLS) for gait correction, 2) botulinum toxin injections into specific muscles for lower limb spasticity management and 3) other types of surgery including single level orthopaedic surgery for deformity correction (for example bony foot surgery or hamstrings surgery in isolation) and surgery for hip displacement (including varus derotation osteotomies and adductor longus lengthening with Phenol to the obturator nerve). These categories were chosen because they reflected the range of interventions routinely embarked on at the centre where the study took place.

SEMLS refers to the correction of all orthopaedic deformities in one session (12). It can be defined as at least two orthopaedic procedures at different anatomical sites in each limb i.e. a minimum of four procedures (13). The frequently used procedures are muscle-tendon lengthenings, tendon transfers, rotational osteotomies and bony stabilisation procedures (14). Injections of Botulinum toxin A are used to treat spasticity in selected muscle groups resulting in temporary chemodenervation of muscle (15). Injections are indicated primarily in younger children aged between 2 and 6 years (15, 16) to prevent the development of fixed contractures (12).

All categories of interventions were used in the correlations of overall change with an external criterion. In addition, the SEMLS and botulinum toxin group were isolated and analysed separately to examine change versus relative stability in mobility status. It was expected that there would be deterioration in mobility and subsequent improvement in the children having SEMLS based on a previous study that examined change over a 2 year period following SEMLS (17). Clinical experience suggests that the mobility status of children who have botulinum toxin does not change significantly and these children were expected to have stable mobility following the injections. The orthopaedic surgeon responsible for each child's management prescribed the surgery and performed the surgical operations or injected the botulinum toxin.

A sample size of 80 was aimed for based on a previous study of the FMS which found it was able to show change in 66 children with CP following SEMLS (17). This number would also allow for comparison between the group of children having SEMLS and those having botulinum toxin injections.

Pre-operatively and at each post-operative time point each child's GMFCS level was determined by the first author, an experienced physiotherapist. Other data collected for each child were; 1) an FMS rating administered by one of three experienced physiotherapists who were blind to the hypotheses of the study, and 2) the walking scale of the Gillette Functional Assessment Questionnaire (FAQ) (18) completed by the parents. The FAQ is a 10 level parent report ordinal scale that also assesses mobility and has been

Table-I
Participant characteristics.

Characteristic		Total group	SEMLS group	Botulinum toxin group
n		84	24	25
Age (y,m), mean (SD), range		8.8 (3.3), 2-16	10.3 (1.8), 7-14	6.6 (2.6), 2-12
Sex, n (%)	Male	59 (70%)	16 (67%)	15 (60%)
	Female	25 (30%)	8 (33%)	10 (40%)
GMFCS, n (%)	I	10 (12%)	1 (4%)	4 (16%)
	II	31 (37%)	11 (46%)	9 (36%)
	III	35 (42%)	11 (46%)	10 (40%)
	IV	8 (9%)	1 (4%)	2 (8%)

y.m: years:months, SD: standard deviation, SEMLS: single event multilevel surgery

shown to be reliable and valid (18). It differs from the FMS in that it has only one item and does not take into account the assistive devices the children require for mobility. The children were assessed at regular post-intervention time points; 3, 6, 9 and 12 months post SEMLS and 3, 6, 12 and 24 weeks post injections. At each post-intervention time point the child's GMFCS level, FMS rating and FAQ score were collected. The children did not have any additional surgery or injections before the last-follow-up.

Descriptive statistics for participant characteristics were calculated. The MCID was considered to be a change of one category up or down on the FMS based on consensus by four experienced clinicians. This decision was based on the literature that clinical experience with a measure is a valid method of determining the MCID where there is no "gold standard" for what represents a real change in clinical status (19). Each category within the FMS represents a different level of assistance required for mobility. A change of one level up or down therefore is a clinically meaningful level of improvement or deterioration. Measures of external responsiveness appropriate for categorical data were chosen to examine the responsiveness and the FAQ was used as an external criterion to assess change following the interventions.

Change scores (follow-up score minus initial score) for each post-intervention stage were used as a measure of responsiveness because FMS and FAQ data are ordinal (3). The percentage of children who either did not change or changed for the better (up) or worse (down) was calculated at each time point. This was done for the group as a whole as well as for the group who had SEMLS compared to the group who had botulinum toxin injections. This was to determine whether the FMS could show change as well as stability because a useful measure must reflect "no change" as well as it does "change" (9). Spearman rank correlation coefficients (20) were calculated for the correlation of change scores for each distance of the FMS with the FAQ over each time period based on groups of no change, improved or deteriorated.

The data were stored and organised using the EpiData for Windows program (http://www.epidata.dk/.). All analyses were performed using Stata [StatCorp 2005 Stata statistical software Release 9.0. College Station; TX. Stata Press].

RESULTS

Ninety six children were recruited for the study. Nine children dropped out of the study in the initial period due to parents not returning the FAQ in the mail. Another three children recruited did not have their surgery on the expected date within the recruitment period. This left a total sample of 84 children. The children had a mean age of 8.8 years (range 2-16 years). There were 59 males and 25 females. Table 1 summarises the participant characteristics.

Twenty four children (29%) had bilateral SEMLS, 25 (30%) had botulinum toxin injections and 35 (41%) had other types of surgery. The mean age of the children who had SEMLS was higher than that for those who had botulinum toxin injections.

The correlations between change scores on the FMS and FAQ using Spearman rank correlation coefficients are presented in Table 2. The correlations were generally low with only six comparisons statistically significant.

The change scores are shown in Table 3. The percentages of children who showed no change, changed up one or more level (change for the better) and changed down one or more level (changed for the worse) based on the MCID of one category at each post-intervention time point compared to baseline are presented. This is for the group as a whole and for the SEMLS and botulinum toxin groups.

For the group as a whole there were more than 50% of children who did not change at each time point on the FMS compared to baseline. There was more change for the FAQ. At the first post-intervention time point more children showed deterioration than improvement but the levels were relatively even at the second time point. At the third and fourth time points more children improved than deteriorated compared to baseline.

A pattern emerged when comparing children who had SEMLS with those who had botulinum toxin injections. A greater number of children showed no change on the FMS and FAQ in the toxin group at each time point. Children who had SEMLS showed change, particularly at the first time point with more than 50% deteriorated on all three FMS distances. The differences in change in these two groups can be clearly seen in the bar graphs in Figures 2-4. The graphs illustrate greater change in the SEMLS group compared to the relative stability of FMS in the botulinum toxin group for each FMS distance.

DISCUSSION

The results of this study provide some evidence of the responsiveness to change of the FMS. A higher percentage of children who had SEMLS changed compared to those who had botulinum toxin injections, thus suggesting the FMS is able to detect both change and relative stability in the mobility status of children with CP.

Table-II

Correlation of change scores between the FMS and FAQ.

Change period	FMS distance	Rho	p value
Change from baseline to first post intervention point	5	0.28	0.0088*
	50	0.36	0.0007*
	500	0.23	0.04*
Change from baseline to second post intervention point	5	0.17	0.1282
	50	0.18	0.1075
	500	0.06	0.6016
Change from baseline to third post intervention point	5	0.01	0.5815
	50	0.26	0.03*
	500	0.04	0.7391
Change from baseline to fourth post intervention point	5	0.05	0.7279
	50	0.29	0.05*
	500	0.22	0.14

* denotes statistically significant result

The differences in change observed were expected based on previous research examining changes in mobility following SEMLS (17) and indications for botulinum toxin. One of the aims of the botulinum toxin injections is to prevent the development of fixed contractures (12) and defer the need for more extensive deformity correction until the child is at an optimal age where the most benefits are obtained from surgical interventions (14). Thus, children who have botulinum toxin injections tend to be younger than those having SEMLS as seen in the differences in the participant characteristics of each group in this study. Because botulinum toxin injections result in temporary chemodenervation of targeted muscles to improve muscle length (15), it is not expected that mobility status will change significantly. SEMLS on the other hand is more invasive and aims to correct musculoskeletal deformities contributing to gait deviations (21). It is expected that following SEMLS mobility will deteriorate in the initial post-operative phase and then improve as the children become stronger and the gait pattern improves throughout the rehabilitation period.

Responsiveness in this study was examined using the FAQ as an external criterion. The correlations of change scores on the FMS with change scores on the FAQ were lower than expected. This possibly reflects the differences between the two measures. The walking scale of the FAQ is a one item 10 level categorical scale

Table-III

Percentage of children (%) changed from baseline categories for the FMS and FAQ at post-intervention time points.

Time	1st time point			2nd time point			3rd time point			4th time point		
Change	**0**	**v**	**^**	**0**	**v**	**^**	**0**	**v**	**^**	**0**	**v**	**^**
Total (n=84)												
FMS 5	60	33	7	74	14	12	73	8	19	65	15	20
FMS 50	57	38	5	67	17	16	60	13	27	63	13	24
FMS 500	67	27	6	70	12	18	71	9	20	85	6	9
FAQ	40	47	13	49	24	27	45	19	36	30	22	48
SEMLS (n=24)												
FMS 5	29	62	9	54	34	12	50	21	29	52	24	24
FMS 50	37	59	4	58	25	17	59	12	29	52	10	38
FMS 500	42	50	8	54	26	20	59	16	25	81	14	5
FAQ	25	67	8	52	26	22	44	26	30	24	23	53
BOTOX (n=25)												
FMS 5	88	4	8	92	4	4	88	0	12	74	9	17
FMS 50	84	12	4	80	4	16	71	8	21	74	13	13
FMS 500	96	4	0	100	0	0	96	0	4	92	0	8
FAQ	64	20	16	64	12	24	50	13	37	39	17	44

0; no change, ↓; changed down by at least 1 category (worse), ↑; changed up by at least one category (better)
SEMLS: single event multilevel surgery group, BOTOX; Botulinum toxin group

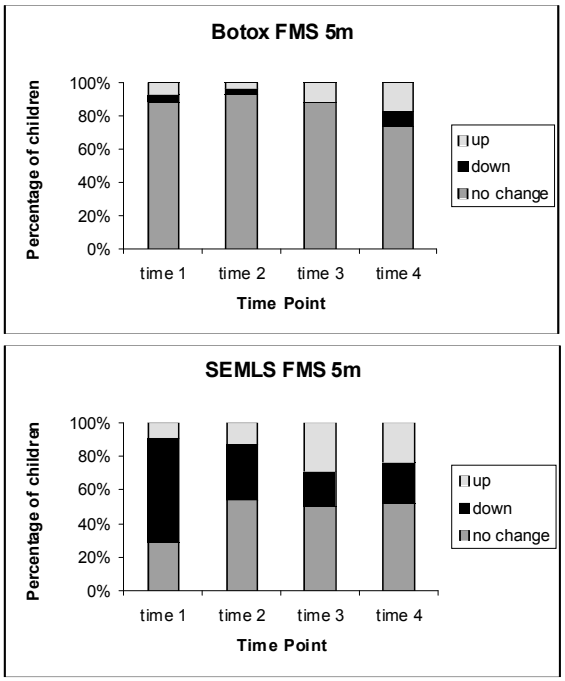

Figure 2. Change in FMS 5m score of botulinum group compared to SEMLS group

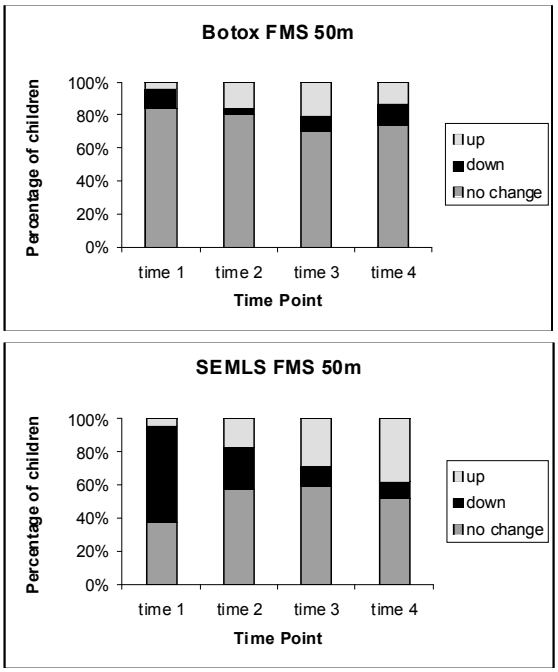

Figure 3. Change in FMS 50m for botulinum group compared to SEMLS group

where one category is chosen by the parent to reflect the child's usual walking ability. The FMS is a six level categorical scale using clinician interpretation of self or parent report and has three different items reporting three different levels of mobility according to environmental setting. Although they are both measuring mobility, the methods of reporting differ. Correlations of change scores were calculated for all three FMS items with the FAQ. It is interesting to note that there were higher correlations of the FAQ with the FMS 50 meter distance, suggesting that of the three FMS distances the 50 meter distance is most closely related to the FAQ. The higher number of levels to select from on the FAQ may have resulted in a higher variability of categories chosen, with a smaller chance of selecting the same category each time.

Considering the differences between the scales a different external criterion might have produced higher correlations. A major difference between the scales is that children can improve or deteriorate in mobility as measured by the FMS and remain in the same category on the FAQ. This is because the FAQ does not consider the assistive devices children use. This implies that although both scales measure mobility, their underlying constructs are different. There is, however, no other scale or measure that can be considered a "gold standard" for mobility to use as an alternative external criterion. A weakness of relying on an external standard to assess responsiveness is that a new outcome measure may be designed specifically because it reflects a different aspect than currently available measures (9). The FMS was developed because no other measures available considered the assistive

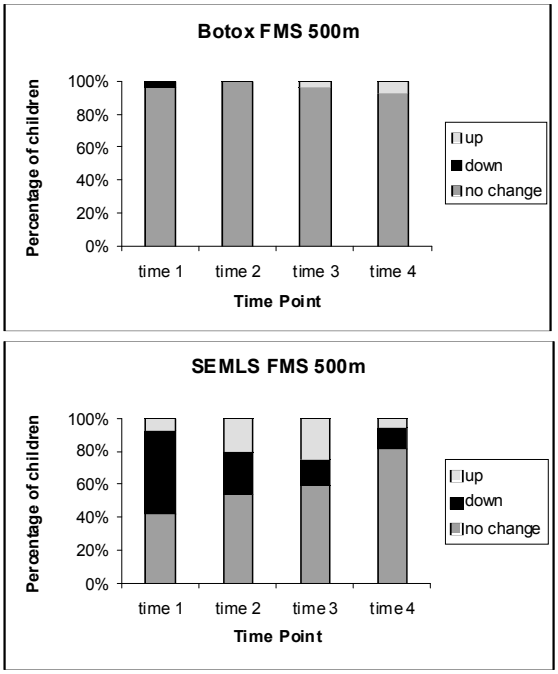

Figure 4. Change in FMS 500m in botulinum toxin group compared to SEMLS group

devices children use in different environmental settings. It therefore measures different aspects of mobility than the FAQ. Other methods of assessing responsiveness that do not require the use of an external criterion, such as effect size and standardized response means, are not appropriate for categorical data. These methods tend to assess the magnitude of treatment effect rather than the ability of the tool to detect clinically important differences and do not provide information about the quality of the instrument to serve its purpose (10).

Another option for a criterion measure in the absence of a gold standard that has been suggested is using parent or clinician judgment (3). Parental opinions can be subjective and potentially introduce a bias in reporting. It is possible for an instrument which includes some subjective components to detect differences between interventions and thus suggest responsiveness without being a valid measure (5). These subjective methods of change were not chosen for this study because of the lack of evidence for reliability and validity of such methods. Determining the best methods for assessing responsiveness of evaluative tools remains an ongoing process.

There are some limitations of this study. The sample included more children classified as GMFCS levels II and III as this reflects the demographic of children who are appropriate for spasticity management and orthopaedic surgery for gait correction. The children were recruited from one centre only. Further investigations of the responsiveness of the FMS with children from a range of centers with a more even spread of GMFCS levels is appropriate. It is also important to demonstrate the FMS is able to detect changes in mobility following other interventions such as physiotherapy programs and other forms of spasticity management as well as changes that occur that are not related to interventions as children grow and develop over time.

The FMS is measured on a categorical scale and this limits the range of statistical methods that can be used to analyze the data. A different criterion measure such as the mobility domain of the Pediatric Evaluation of Disability Inventory (PEDI) (22) or the Activities Scale for Kids (ASK) (23) may have been more suitable. Receiver operating characteristic (ROC) curves could have been used for data analysis (9). They were not used due to the lack of a completely stable group to compare change against. Clinical experience suggests that children having botulinum toxin injections show less change,, however there is no expectation that this group would be completely stable.

In conclusion, the FMS was able to detect changes in mobility in children with CP. The FMS was able to detect more change in children who had SEMLS compared to those who had botulinum toxin injections.

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