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Measuring Physical Functioning in Children with Spinal Impairments with Computerized Adaptive Testing

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Abstract

Background—The purpose of this study was to assess the utility of measuring current physical functioning status of children with complex spinal impairments by applying computerized adaptive testing (CAT) methods. CAT uses a computer-interface to administer the most optimal items based on previous responses, reducing the number of items needed to obtain a scoring estimate.

Methods—This was a prospective study of 77 subjects (0.6 – 19.8 yrs) with spinal impairments who were seen during a routine clinic visit. Using a multidimensional version of the Pediatric Evaluation of Disability Inventory CAT program (PEDI-MCAT), we evaluated content range, accuracy and efficiency, known –groups validity, concurrent validity with the Pediatric Outcomes Data Collection Instrument (PODCI), and test-retest reliability in a sub-sample (n=16) within a two-week interval.

Results—We found the PEDI-MCAT to have sufficient item coverage in both self-care and mobility content for this sample, although a majority of the patients tended to score at the higher ends of both scales. Both the accuracy of PEDI-MCAT scores as compared to a fixed-format of the PEDI ($r = 0.98$ for both mobility and self-care) and test-retest reliability were very high (self-care: ICC (3,1)=0.98, mobility: ICC(3,1)=0.99). The PEDI-MCAT took an average of 2.9 minutes for the parents to complete. The PEDI-MCAT detected expected differences between patient groups, and scores on the PEDI-MCAT correlated in expected directions with scores from the PODCI domains.

Conclusion—Use of the PEDI-MCAT to assess the physical functioning status, as perceived by parents of children with complex spinal impairments, appears to be feasible and achieves accurate and efficient estimates of self-care and mobility function. Additional item development will be needed at the higher functioning end of the scale to avoid ceiling effects for older children.

Level of Evidence—This is a level II prospective study designed to establish the utility of computer adaptive testing as an evaluation method in a busy pediatric spine practice.

Keywords

computerized adaptive testing; assessment; outcomes; spine impairments

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Introduction

Children with spine impairments have a broad spectrum of physical abilities, ranging from typical development to significant limitations. For example, while the cause, degree and progression of idiopathic scoliosis may differ, those with late onset idiopathic scoliosis (after five years of age) typically present minor limitations in physical ability.¹ In fact, a diagnosis of late onset scoliosis is often brought about by recognizable asymmetry of the shoulders and hips while standing,² as opposed to any major limitations in physical functioning. In contrast, children with early onset idiopathic scoliosis,¹ congenital scoliosis, kyphosis and kyphoscoliosis,^{3,4} spinal dysraphism⁵ and neuromuscular scoliosis,⁶ such as cerebral palsy,⁷ have associated co-morbidities that often impose varying degrees of limitations in performing activities. Pediatric spine practices also manage children with recognized major limitations in physical capacity such as children with osteogenesis imperfecta, skeletal dysplasia and muscular dystrophy.

In the era of evidence and practice based medicine, there is a commitment among spine surgeons to carry out assessment protocols that evaluate outcomes of conservative and surgical treatment. Unfortunately, the instruments used to measure important outcomes in children with scoliosis and other spine disorders are only just evolving.⁸ For example, the Scoliosis Research Society 22-item form (SRS-22)⁹ originally designed for use with persons with Adolescent Idiopathic Scoliosis (AIS), evaluates physical activity performance, among other domains. It has been applied to the assessment of children and youth with poor results.^{10, 11} The Scoliosis Quality of Life Index (SQLI)¹² was developed based on the SRS-22 to evaluate outcomes in the younger adolescent population with AIS. Modifications included standardizing the time frame to a four-week period and replacing two questions. Like the SRS-22, the SQLI is a “disease-specific” instrument for the population with scoliosis and does not contain items that are useful for children with spine impairment due to reasons other than AIS. Other instruments have promise, but have not been developed for a broad patient age-range or set of clinical conditions, and have not been utilized widely in research or clinical care.^{13, 14} The Pediatric Orthopaedic Society of North America¹⁵ developed the Pediatric Outcomes Data Collection Instrument (PODCI) as a functional health outcomes instrument for children and adolescents with a focus on musculoskeletal health. While the instrument is used for outcomes research with populations with AIS, a significant number of questions in the physical function and sports as well as the transfers and mobility questions are geared toward ambulatory individuals and not relevant for most patients with neuromuscular scoliosis or patients who are non-ambulatory and thus, not used widespread in clinical practice.

Thus, it seems apparent that a common instrument with a wide range of items, appropriate for the pediatric age range and breadth of abilities that typify all children with spine impairments – regardless of the type of spine impairment, needs to be developed and validated. We propose to evaluate for this population a modified physical functioning item bank and computerized adaptive test (CAT) based on the Pediatric Evaluation for Disability Inventory,¹⁶ but expanded for older children and children with severe physical disabilities.

Computerized adaptive testing (CAT) is a contemporary measurement technology that uses large item banks that are calibrated along unidimensional functional and quality of life content domains. CAT is currently being used in many outcome applications in both pediatric and adult health care services^{17–20} and it appears that it may be the assessment platform of choice in future health care assessment programs.^{21–26} Once an item bank, which can contain hundreds of calibrated items from multiple instruments, is created, then a CAT platform can be developed. The CAT employs a simple form of artificial intelligence that selects questions from the item bank that are directly tailored to an individual child, shortens or lengthens the test to achieve a desired precision, and scores each test on a standard metric so that results can

be compared across time points and across groups of children.^{25, 27} For example, for a child with paralytic spine impairments, a question about community ambulation may be unnecessary if the child is unable to walk. Similarly, a question about the ability to negotiate 3 to 5 steps is also pointless for a youth with adolescent idiopathic scoliosis who is currently walking up and down multiple flights of stairs. Administering items that are either too easy or too hard provides little information and wastes valuable time in a busy spine practice. As the child progresses and new test administrations are needed, new items are then selected that best reflects the child's ability at that time. We have previously demonstrated the potential usefulness of CAT programs in assessing physical functioning in children with developmental disorders²⁸ and rare lysosomal disorders such as Pompe disease.^{29, 30}

In this study, we used a multidimensional CAT model based on an extended version of the Mobility and Self-care Functional Skills of the Pediatric Evaluation of Disability Inventory (PEDI-MCAT) for the purpose of assessing physical functioning of children with complex spinal impairments. We use the model from the Pediatric Evaluation of Disability Inventory,¹⁶ by separating out self-care and mobility as two separate, but related concepts of physical functioning. Our goals were to use the CAT program in an ongoing, busy clinic to examine its feasibility, content range for the sample, accuracy, efficiency, known groups and concurrent validity, and test-retest reliability.

Methods and Materials

The study used a cross sectional research design. A convenience sample of legal guardians of 77 children between 6 months and 19 years of age (average age 9.9 years) with infantile idiopathic scoliosis (IS) (n=13), juvenile idiopathic scoliosis (JS) (n=13), adolescent idiopathic scoliosis (AIS) (n=21), congenital scoliosis (CS) (n=17), syndromic (SYN) (n=8), or kyphoscoliosis (KS) (n=5) being seen in routine office hours were included in this study. A majority of the sample was Caucasian (69%), however, it also included 7 subjects of African American and 2 subjects of Asian descent. See details of sample in Table 1. In addition to the PEDI-MCAT, parents were asked to complete a fixed-format version of the PEDI¹⁶ (55 items) and the Pediatric Outcomes Data Collection Instrument (PODCI)¹⁵ during the clinic visit. A small sub-sample (n=16) of subjects from the original 77 were asked to complete the PEDI-MCAT assessment on a second occasion, within 2-weeks of the first assessment. In most cases, the study staff traveled to the parents' home to conduct the second assessment.

The PEDI-MCAT is a multidimensional version of earlier work completed with children with Pompe disease, most of whom have serious physical deficits.^{31, 32} However, a number of children have relatively mild presentations, so there was a need to expand the original PEDI content to about 14-years of age to accommodate the children with more advanced levels of physical functioning. Because both self-care and mobility were highly correlated, we built a multidimensional model of physical functioning which included the two concepts into one item bank and one CAT program. This appears to have both advantages in efficiency, precision²⁰ and sensitivity to change over separate unidimensional models.^{33, 34} We have found in previous work that a CAT stop-rule of 15-items (per domain) appeared to generate accurate scores both at the group and individual level.²⁹ Items were administered iteratively based on the parents' responses, and mobility and self-care items were alternated. If a score for either content domain was estimated per internal standards of precision (standard errors), the CAT may stop before the prescribed 15-stop rule. Parents completed the self-report PEDI-MCAT on a tablet computer provided to them in the clinic waiting room prior to their clinic visit. A research nurse assigned to the project was available to the subjects during the administration process to answer any questions. Subject demographic information, impairment type were all recorded by the study nurse.

Data collection also involved administration of the PEDI expanded form for children with Pompe disease.²⁹ Since this version contains too many items for administration during a routine clinic visit, we chose 30 mobility and 25 self-care items to compare the accuracy of the scores between the PEDI fixed-form to the CAT. We also administered a parent-report version of the PODCI. The PODCI is used extensively in studies and clinical work for children with cerebral palsy and orthopedic conditions,³⁵ and serves in this study as a concurrent measure of physical functioning. All guardians provided written informed consent, as required by the Shriners Hospital for Children – Philadelphia and Boston University Medical Center institutional review committees.

We analyzed the content range of the PEDI-MCAT by examining the item locations of both self-care and mobility content as they corresponded to the children's physical functioning ability. For the analysis of known groups validity, we compared mean self-care and mobility scores between groups expected to perform differently in physical functioning status (AIS vs. Early Onset Scoliosis (CS&IS)). To compare the accuracy of the PEDI-MCAT scores to the PEDI fixed forms, we plotted the two scores and calculated simple Pearson correlations. We compared the PEDI-MCAT scores to the PODCI by calculating simple Pearson correlations between PEDI-MCAT and PODCI content domains. We concentrated on content domains that were somewhat similar to the PEDI domains of self-care and mobility. Finally, we used intraclass correlations (ICC (3,1))³⁶, to examine the test-retest reliability of the two administrations of the PEDI-MCAT separated by about a two-week interval. We established alpha rates at $<.05$.

Results

We examined the range of content covered by the PEDI-MCAT by plotting the scores of the 77 patients on the same metric as the items from both the mobility and self-care banks. The Mobility and Self-care graphs are depicted in Figure 1 and Figure 2, respectively. Since the PEDI-MCAT response scale is dichotomous, each tic mark reflects either the location of "unable" or "capable" for each item in the bank. Note that most of the person scores are at the more functional end of the scale (higher score = greater functional ability). We found no floor effects (person selected the lowest category for all the administered items) on the CAT. In contrast, the PEDI-MCAT has a 9.1 % ceiling effect (person selected the highest category for all the administered items) as opposed to the PEDI fixed for, which had a 28.6% ceiling effect.

As expected, the children with AIS had significantly higher functional scores than the children with early onset scoliosis, and these differences are discriminated easily by the PEDI-MCAT self-care (mean difference = 9.95; $t = 6.01$; $df = 1$; $p < .001$) and mobility (mean difference = 10.0; $t = 4.76$; $df = 1$; $p < .001$) scores. See Table 2.

As compared to the PEDI short form, the CAT scores were very accurate as shown in Figure 3 and Figure 4. Even though many of the scores were at the high end of the range, the PEDI-MCAT was able to accurately estimate person ability scores. The PEDI-MCAT was more efficient than the PEDI short form, as the average number of PEDI-MCAT items were 20 and the amount of time to complete the PEDI-MCAT was less than 3 minutes (2.87 min; range 1.2–6 min), while the average time to complete the short form (55 items) was nearly 5 minutes (4.9 min; range 2–14 min).

We also examined the correlations of PEDI-MCAT scores to the PODCI physical functioning domains as given in Table 3. The PEDI-MCAT self-care domain was moderately correlated with PODCI upper extremity function ($r = 0.68$). PEDI-MCAT is highly correlated with the PODCI domains of transfers and mobility ($r = 0.74$) and physical function and sports ($r = 0.84$).

We examined the stability of the PEDI-MCAT scores by recruiting a convenience sample of 16 parents who were willing to complete two PEDI-MCATs within an approximately two week period (average interval 10.5 days; SD= 3.6; range = 5–17 days.). Using intraclass correlations, scores on the two administrations of the PEDI-MCAT were remarkably consistent for both self-care (ICC (3,1)) = 0.98; and mobility (ICC (3,1)) = 0.99.

Discussion

Recent studies have highlighted the promise of CAT approaches in pediatric assessments, but most of the conclusions to date have come from CAT simulations^{20, 28, 29, 37, 38} rather than prospective use of the CAT in a busy clinic environment. There have been a few notable exceptions.^{28, 33} This study adds to the findings that CAT use in prospective studies can yield very accurate data. In comparison to earlier studies of the accuracy of using the PEDI in a CAT format,²⁸ the scores from the PEDI-MCAT were just as accurate in comparison to longer, fixed-length forms.

We found that the range of items included in the PEDI-MCAT did not ideally match the sample. This is not too surprising since the original PEDI¹⁶ was created for children under 7 years of age, and the PEDI-MCAT³⁹ was intended for children with very low skills. Although content was expanded to age 14 years, the major focus of item content was to eliminate floor effects. More work will need to be accomplished to add items to reduce ceiling effects for the older children and youth in clinical samples.

The PEDI-MCAT was more efficient than the fixed, short form PEDI as evidenced by the 36% reduction of items and 58% of time required by the PEDI-MCAT versus the 55-item fixed length version of the PEDI. In this study, we set a rather high threshold for the stop-rule, primarily since we had no experience in testing physical functioning using a CAT in this sample. Efficiency could improve if fewer items on the PEDI-MCAT were allowed, but this would need to be balanced by the required precision of the scores and the efficiency needed in the clinic in order to collect data consistently.

Despite the need for short-term test-retest data in the field of CAT in prospective studies, we know of no reported data in either the adult or pediatric literature. Since CAT is administered in real time, and item selection is based on previous responses, the items used for CAT sessions could vary. Theoretically, score estimates can be obtained from any subset of items, and when enough items are administered, similar scores can be obtained even if different items are administered. Our excellent test-retest data suggest that within a short 2-week interval, in which memory of the answers to specific items is probably not a major factor,^{40, 41} that CAT scores are stable. These initial findings will be important to replicate in future work with larger samples.

Parents welcomed involvement in this study and did not appear hesitant about using the computer to answer questions. The majority (70%) of subjects preferred the PEDI-MCAT over the paper-based fixed-length PEDI form. The tablet-interface and CAT process allowed parents to easily complete a questionnaire while waiting for their physician.

This study demonstrated the utility and efficiency of CAT technology in a busy pediatric spine clinic. Not only were parents able to complete the computer interface associated with the PEDI-MCAT, the majority of caregivers preferred it over traditional surveys. The accuracy of the PEDI-CAT and discriminatory ability between known groups were demonstrated. The agreement between repeated PEDI-CAT measures was high. As anticipated, the PEDI-MCAT, if used in this population, will require the construction of additional mobility and self care items for higher functioning children with spine impairments.

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Figure 1. Plot of Self-care scores (left) and item locations (right)

Plot of Self Care Scores and Item Locations. Range of content covered by PEDI-MCAT of 77 subjects on self care item bank.

Figure 2. Plot of Mobility scores (left) and item locations (right)
 Plot of Self Care Scores and Item Locations. Range of content covered by PEDI-MCAT of 77 subjects on mobility item bank.

Figure 3. Accuracy of PEDI short form (25 items) and CAT (15 items) for Self-care Domain
Accuracy of the PEDI-CAT self-care when compared to fixed length traditional paper-pencil PEDI short form.

Mobility Domain

Figure 4. Accuracy of PEDI short form (30 items) and CAT (15 items) for Mobility Domain
Accuracy of the PEDI-CAT mobility when compared to fixed length traditional paper-pencil PEDI short form

Table 1

Sample Characteristics

Diagnostic Group	N (%)	Mean Age (SD)	Gender (N; Female %)	Race (N; Caucasian%)	Ethnicity (N; Hispanic%)
AIS	21(21.27%)	14.74(2.35)	17(80.95%)	20(95.24%)	1(4.76%)
CS	17(22.08%)	6.7(4.59)	8(47.06%)	13(76.47%)	0(0%)
IS	13(16.88%)	6.08(5.16)	10(76.92%)	11(84.62%)	0(0%)
JS	13(16.88%)	9.44(2.33)	8(61.54%)	12(92.31%)	1(7.69%)
SYN	8(10.39%)	8.05(3.83)	4(50%)	8(100%)	2(25%)
KS	5(6.49%)	15.04(4.80)	4(80%)	5(100%)	0(0%)
TOTALS	77	9.93(5.20)	51(66.23%)	69(89.61%)	4(5.19%)

Table 2

Known-groups Validity for PEDI-MCAT scores

group	AIS	Early Onset Scoliosis (CS & IS)	<i>t</i> -test
N	21	30	
Average Selfcare Score (SD)	63.43(1.63)	53.48(7.45)	6.01 (p<0.0001)
Average Mobility Score (SD)	63.56(5.52)	53.56(8.44)	4.76 (p<0.0001)

Table 3
Correlations between PEDI-MCAT and PODCI physical function domains

PODCI Domains	PEDI-MCAT Domains	
	<i>Selfcare (N)</i>	<i>Mobility (N)</i>
Upper extremity function	0.682 (72)	0.823 (72)
Transfers and mobility	0.400 (75)	0.739 (75)
Physical function and sports	0.665 (77)	0.839 (77)