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The Final 24-Item Early Onset Scoliosis Questionnaires (EOSQ-24): Validity, Reliability and Responsiveness

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Abstract

Introduction—The goal of early-onset scoliosis (EOS) treatment is to improve health-related quality of life (HRQoL) for patients and to reduce the burden on their parents or caregivers. The purpose of this study is to develop and finalize the 24-item Early-Onset Scoliosis Questionnaire (EOSQ-24), and examine the validity, reliability, and responsiveness of the EOSQ-24 in measuring patients' HRQoL, the burden on their caregivers, and the burden on their caregiver's finances. The study also established age-matched normative values for the EOSQ-24.

Methods—The EOSQ-24 was administered to caregivers of male and female patients aged 0 to 18 years with EOS. Patients with EOS are diagnosed before 10 years of age. Criterion validity was investigated by measuring agreement between its scores and pulmonary function testing. Construct validity was established by comparing values across different etiology groups using the known-group method, and measuring internal consistency reliability. Content validity was confirmed by reviewing caregiver and health provider ratings for the relativity and clarity of the EOSQ-24

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questions. Test-retest reliability was examined through intraclass correlation coefficients. Responsiveness of the EOSQ-24 before and after surgical interventions was also investigated. Age-matched, healthy patients, without spinal deformity, were enrolled to establish normative EOSQ-24 values.

Results—The pulmonary function subdomain score in the EOSQ-24 was positively correlated with pulmonary function testing values, establishing criterion validity. The EOSQ-24 scores for neuromuscular patients were significantly decreased compared with idiopathic or congenital/structural patients, demonstrating known-group validity. Internal consistency reliability of patients' HRQoL was excellent (0.92), but Family Burden was questionable (0.64) indicating that Parental Burden and Financial Burden should be in separate domains. All 24 EOSQ items were rated as essential and clear, confirming content validity. All EOSQ-24 domains demonstrated good to excellent agreement (0.68 to 0.98) between test and retest scores. Neuromuscular patients improved their HRQoL postoperatively, whereas idiopathic patients worsened their HRQoL postoperatively, indicating the ability of the instrument to respond to different trajectories of HRQoL according to etiology in patients with EOS.

Discussion—The developed and finalized EOSQ-24 is a valid, reliable, and responsive instrument that is able to serve as a patient-reported outcome measure evaluating health status for patients with EOS and burden of their caregivers. This instrument will be able to serve as an outcome measure for future research including clinical trials evaluating the effectiveness of various treatments. In addition, the EOSQ-24 allows assessment of patients' HRQoL, and the burden on their caregivers relative to age-matched healthy peers.

Level of Evidence—Level II—diagnostic study with consecutive patients enrolled in national registries.

Keywords

early onset scoliosis; patient reported outcome; health related quality of life; burden of care; outcome measures

Children with early-onset scoliosis (EOS) and their caregivers face numerous health challenges associated with their progressive spinal deformity and compromised pulmonary function.^{1,2} Current surgical techniques have sought to address these complex health issues by halting the progression of scoliosis while allowing for spinal and thoracic growth.³ To this end, growing instrumentation with spine-based or rib-based distraction, notably the “growing rod” (GR) and “vertical expandable prosthetic titanium rib” (VEPTR) constructs, respectively, are utilized as an alternative to arthrodesis which has been shown to have negative effects in growing children.^{4–6}

Despite the promise of both techniques in supporting spinal growth and chest wall development,^{2,7,8} the comparative effectiveness of these surgical constructs is not well delineated.^{3,9} The risk of complications for both GR and VEPTR have been shown to be high^{3,9,10}; this is often attributed to the comorbid conditions in this population and the necessity for repetitive surgery with growing constructs. Complications are known to decrease the quality of life for patients, but in pediatric populations, additional mental and financial burdens are placed on the patients' caregivers as well.^{11–14} Matsumoto et al¹⁵

reported the negative psychosocial effects of repetitive surgeries on EOS patients and found these effects correlated with younger age. As such, it is important for EOS surgeons to have a comprehensive view of the patient and familial circumstances to anticipate and mitigate these deleterious psychosocial effects.

Implicit in the goals of EOS treatment, simultaneous scoliosis control and growth allowance, is the improvement of health-related quality of life (HRQoL) of patients and the reduction of caregiver burden. Previously, Vitale et al¹⁶ reported that limitations existed with generic HRQoL measures in reflecting changes before and after VEPTR surgery and emphasized a need for a disease-specific instrument. To this end, we launched a series of studies to develop and validate a patient-reported outcome instrument that measures caregiver evaluation of HRQoL of patients with EOS and the caregivers' burden.¹⁷ Items identified in the master list demonstrated good initial content and construct validity.

The purpose of this study was to develop and finalize the 24-item Early-Onset Scoliosis Questionnaire (EOSQ-24), and examine the validity, reliability, and responsiveness of the EOSQ-24. Criterion validity was investigated by measuring agreement between its scores and pulmonary function testing (PFT). In addition, content and construct validities of the finalized 24 items in the EOS patients were confirmed. The internal consistency reliability, test-retest reliability, and responsiveness of the EOSQ-24 were also investigated. In addition, we set out to establish age-specific normative scores of the EOSQ-24 as a reference of comparison to EOS caregiver responses. We hypothesized that the EOSQ-24 developed by qualitative and quantitative approaches emphasizing qualitative inputs from caregivers is a valid, reliable, and responsive instrument to measure the caregiver perspectives of HRQoL and burden of care of patients with EOS.

METHODS

The EOSQ-24 was administered to caregivers of male and female patients aged 0 to 18 years with EOS. Patients with EOS were diagnosed before 10 years of age. The mean age was chosen over the median age throughout this study as the ages were normally distributed. Only 1 caregiver of the patient was required to complete the EOSQ-24 for all tests as quality of life is a subjective measure. All treatments were by standard-of-care protocol of the sites and surgeons, and therefore were not for the purpose of this study.

Validity

Criterion validity of the EOSQ-24 was examined to evaluate the relationship between the EOSQ-24 and PFTs. Ten patients (mean age, 8.5 ± 2.6 y; range, 5.54 to 13.27 y) with EOS for whom the caregivers completed the EOSQ-24 and who completed PFTs within 12 days (mean, 3.5 ± 4.8 d; range, 0 to 12 d) at any stage of their treatments were identified from the Children's Spine Study Group (CSSG) and Growing Spine Study Group (GSSG) databases. Etiologies included idiopathic (N=3), neuromuscular (N=2), syndromic (N=3), and congenital/structural (N=2). Available PFT data included forced vital capacity (FVC), forced expiratory volume in 1 second (FEV₁), and the % predicted values of each. As FEV₁ and FVC were commonly available tests of pulmonary function in these patients, and because

they describe lung volume and elasticity well, correlations with these values and Pulmonary Function subdomain scores of the EOSQ-24 were examined using Pearson r .

Construct validity was examined by using the known-group method to compare scale scores across etiology groups using ANOVA and regression model using dummy coding to account for multiple comparisons.¹⁸ Ninety-five unique patients identified from the CSSG and GSSG databases completed the EOSQ-24. They were grouped based on their etiologies: idiopathic (N=29), neuromuscular (N=28), syndromic (N=18), and congenital/structural (N=15). For patients who underwent any form of treatment, a Child Satisfaction and Parent Satisfaction were measured. These patients included 8 idiopathic, 4 neuromuscular, 4 syndromic, and 6 congenital/structural patients. Internal consistency reliability was tested to address the extent that domains within the patient's HRQoL, Family Burden, and Satisfaction are related to each other. Cronbach α coefficients were calculated in which a coefficient of >0.70 are considered acceptable for aggregate data.¹⁹ The most commonly used sample size criteria for construct validity is at least 20 participants with a minimum of 10 additional participants for each variable. Therefore, it was determined that at least 90 participants would be needed for the 8 domains of the EOSQ-24. Post hoc power analyses demonstrated that all significance retained their power at 80%. Therefore, any significant differences seen as a result are due to actual large differences in the effect measures between groups and not due to the errors.

The previously developed master list of questions²⁰ was reviewed, and the clarity and relativity of the 24 items to be included in the EOSQ-24 were confirmed.

Reliability

Test-retest reliability was investigated to measure the reproducibility of the EOSQ-24. Fifteen caregivers of patients with EOS completed the EOSQ-24 twice within 1 month (7 to 29 d). Etiologies of scoliosis included idiopathic (N=6), neuromuscular (N=3), syndromic (N=2), and congenital/structural (N=4). Intraclass correlation coefficients (ICCs) were utilized to determine test-retest reliability and the designations of 0.40 (poor to fair agreement), 0.41 to 0.60 (moderate agreement), 0.61 to 0.80 (good agreement), and 0.81 to 1.00 (excellent agreement) were adapted to interpret the values.²¹ The recommendation for the sample size of a test-retest reliability testing is at least 10 participants and for ICCs to be at least 0.70 to 0.75 with the lower limits of the confidence intervals to be above 0.60.

Responsiveness

Sixty-five pediatric patients (mean age, 6.4 ± 2.9 y; range, 0 to 11.4 y) with a diagnosis of EOS and their caregivers were enrolled at 5 centers in this prospective study between 2008 and 2011. Patients with a coronal major curve >20 degrees (mean, 67.3 ± 17.0 degrees; range, 36 to 104 degrees) and scheduled to undergo a surgical treatment with GR (N=25), VEPTR (N=36), or Shilla (n=4) were included. These surgical patients represent a majority of EOS patients as many of them require a surgical treatment. Etiology of the curve included idiopathic (N=9, 14%), neuromuscular (N=23, 35%), syndromic (N=13, 20%), and congenital/structural (N=20, 31%).

Upon obtaining appropriate consent and HIPAA forms by the treating surgeons and/or research personnel at each participating site, patients and their caregivers were enrolled in the study. Caregivers were asked to complete the study questionnaire at 2 points: the immediate preoperative visit before implantation of a growing device, and at a postoperative visit before the first lengthening procedure. The exceptions to this data collection schedule were patients instrumented with self-distracting Shilla constructs. Caregivers of these patients completed the questionnaire before implantation and approximately 6 months postoperatively. Clinical data such as the magnitude of preoperative and postoperative coronal major curve measurements, implant type, instrumented levels, complications, comorbidities, use of assistive devices, and demographic information were also collected.

Normative Reference Data

Patients aged 0 to 18 years old were enrolled at a leading institution to establish normative data for the EOSQ-24. Otherwise healthy patients without a diagnosis of spinal deformity were eligible for enrollment. Patients were recruited using 2 methods. First, a retrospective chart review was conducted to identify patients receiving prior treatment for non-scoliosis-related issues. Accepted diagnoses included developmental dysplasia of the hip, fracture, femoral anteversion, tibial torsion, joint pain, gait abnormality, pes planus, and other orthopaedic evaluations that resulted in no diagnosis. Second, the treating physician's daily schedule was screened for patients receiving non-scoliosis-related care, using similar criteria as described above. Patients identified by the retrospective chart review were mailed a letter from their respective treating physician explaining the purpose of the study and inviting a caregiver to participate in collecting the normative data for this disease-specific instrument for EOS. Enclosed with each letter were a stamped return envelope, consent and HIPAA forms, and the EOSQ-24. Upon return of the completed set of forms, patients were enrolled and the questionnaire data answered by the caregivers were collected. For patients screened by the daily schedule, the treating physician obtained consent from these patients during the office visit. Appropriate consent and HIPAA clearance was collected before completing the questionnaire. A total of 150 patients were recruited. Caregivers of patients in the normative data study completed 1 questionnaire.

EOSQ-24

Questionnaire Items—The developed and examined EOSQ-24 consisted of 24 items pertaining to the patient's HRQoL, Family Burden, and Satisfaction. HRQoL includes 8 domains: General Health (2), Pain/Discomfort (2), Pulmonary Function (2), Transfer (1), Physical Function (3), Daily Living (2), Fatigue/Energy Level (2), and Emotion (2). Family Burden includes 2 domains: Parental Impact (5) and Financial Impact (1), and Satisfaction consists of Child Satisfaction and Parent Satisfaction (2).

Scoring Algorithm—Item values (1 to 5) were assigned to each response choice (poor to excellent), respectively. The algebraic means of the items within each domain were created and utilized in the equation below to transform raw scores into scale scores:

$$\left(\frac{(\text{Algebraic mean} - 1)}{4} \right) \times 100$$

These transformed scale scores should range between 0 and 100.

RESULTS

Validity

Criterion Validity—The EOSQ-24 domain scores of General Health, Pain/Discomfort, Pulmonary Function, Physical Function, Daily Living, Fatigue/Energy Level, and Emotion were positively correlated with % predicted values of FVC and FEV₁ (Table 1).

Known Groups Validity—The EOSQ-24 scores for all etiologies are shown in Table 2. The EOSQ-24 scores were significantly different in 6 of 8 children's HRQoL domains and Parental Burden among the etiologies. Neuromuscular patients had a significantly worsened General Health, Transfer, Physical Function, Daily Living, and Fatigue/Energy Level compared with idiopathic and congenital/structural patients. Neuromuscular patients also had a significantly increased Parental Burden compared with the other etiology groups. Syndromic patients had a significantly decreased General Health and Daily Living compared with idiopathic and congenital/structural patients. Although the sample size for Child and Parent Satisfaction scores was small, neuromuscular patients had the highest satisfaction scores after the treatments. Idiopathic patients had the worst satisfaction scores among all etiology groups.

Internal Consistency Reliability—Patients' HRQoL (General Health, Pain/Discomfort, Pulmonary Function, Transfer, Physical Function, Daily Living Fatigue/Energy Level, and Emotion), as estimated by Cronbach α , was excellent (0.92). In Satisfaction (Child Satisfaction and Parent Satisfaction), Cronbach α was 0.87 indicating good consistency. Internal consistency reliability of Family Burden (Parental Burden and Financial Burden), however, was questionable (0.64), indicating that Parental Burden and Financial Burden should be separate domains.

Content Validity—All 24 EOSQ items were rated as essential and clear by caregivers and health care providers.

Reliability

Test-Retest Reliability—All ICCs for the EOSQ-24 domains demonstrated good to excellent agreement between the test and retest scores. The agreement results were excellent for General Health (0.84), Pain/Discomfort (0.85), Pulmonary Function (0.90), Transfer (0.84), Physical Function (0.97), Daily Living (0.98), Fatigue/Energy Level (0.92), Financial Burden (0.94), Child Satisfaction (0.93), and Parent Satisfaction (0.89). Emotion (0.68) and Parental Burden (0.80) yielded good agreement.

Responsiveness—Table 3 demonstrates significance and trends toward differences between preoperative and postoperative scores. In most of the domains during preoperative assessment, patients with idiopathic scoliosis had the highest scores among all etiology groups but the scores worsened in Physical Function, Emotion, and Financial Burden, and the scores for the rest of the domains stayed the same. Patients with neuromuscular scoliosis received lower scores postoperatively but significant improvements were seen in General Health, Fatigue, Pulmonary Function, Transfer, Emotion, and Parental Burden domains. The improvements in these domains were especially prominent among SMA patients as well as additional domains including Physical Function and Daily Living, and Transfer. Worsening was seen in the Financial Burden domain in the patients with SMA. Patients who had intraoperative complications experienced Pain to be worsened before the first lengthening. No consistent differences were noted in changes in scores between patients with GR and VEPTR.

Normative Data—A total of 141 caregivers of patients completed the EOSQ-24 (mean age, 6.9 ± 4.3 y) (Table 4). Four patients were excluded from the study for having significant comorbidities (3 asthma, 1 arthrogryposis). The distribution of diagnoses for normative patients completing the EOSQ-24 is shown in Table 5.

DISCUSSION

This study presents the final version of the EOSQ, newly named as the Early-Onset Scoliosis 24-Item Questionnaire (EOSQ-24). This developed EOSQ-24 is the first disease-specific instrument to measure HRQoL of patients with EOS, and parental and financial burden of their caregivers. This study demonstrates that the EOSQ-24 is a valid instrument to measure the HRQoL of patients with EOS, and parental and financial burden of their caregivers. In addition to content validity tested in the initial development,²⁰ criterion validity and construct validity were demonstrated in this study. This study also showed high test-retest reliability of the EOSQ-24. The EOSQ-24 is also sensitive to changes before and after surgical interventions, and intraoperative and postoperative complications.

In this study, criterion validity was demonstrated by high correlations between PFT values and HRQoL of patients with EOS. Patients with lower PFT values had lower EOSQ-24 scores for all HRQoL domains except for Transfer. Patients with EOS are known to have impaired respiratory function²² and this study indicates that patients who have the impairment do have low HRQoL, including the caregiver's perspective of the child's pulmonary function as well as general health, pain/discomfort, physical function, daily living, fatigue/energy level, and emotion. Therefore, targeting interventions to improve pulmonary function may in turn improve HRQoL of patients with EOS. Furthermore, for infants whose PFTs are difficult to obtain, it may be possible to use the Pulmonary Function domain in the EOSQ-24 to proximate pulmonary function of patients. Although this criterion validity testing is limited due to the small sample size, it provides preliminary information regarding the relationship with the clinical test and the caregiver's perspective of pulmonary function of these patients with EOS.

The results of this study demonstrate that multiple domains in the EOSQ-24 respond to changes in HRQoL after growing instrumentation surgery, which further prove the usefulness of the instrument as an outcome measure for surgical interventions. Interestingly, this study revealed that, whereas patients with neuromuscular etiology improved their HRQoL after the surgery, patients with idiopathic etiology worsened their HRQoL postoperatively, which demonstrates the ability of the EOSQ-24 to differentiate the trajectory of 2 different populations. Although a goal of a treatment is to improve their ultimate HRQoL, the procedures on patients who are relatively healthy preoperatively, such as patients with an idiopathic etiology, are to prevent future progression of the spinal curvature and deterioration of their pulmonary function. Therefore, the result is consistent with our experience that idiopathic patients with high HRQoL preoperatively experience a decrease in HRQoL postoperatively due to surgical injury. An alternative explanation to the difference in trajectory is regression to the mean in both idiopathic and neuromuscular etiology groups. However, regression to the mean most likely happens when an instrument involves random guessing or luck when the instrument is measuring skills. The EOSQ-24 is measuring quality of life and not involving skills; therefore, regression to the mean is unlikely to happen.

During the development of the EOSQ-24, investigators raised 2 concerns. First, the item in the Physical Function domain, “How difficult has it been for your child to sit up on his/her own?,” was questioned. Their concern was that the difficulty of sitting up observed by caregivers was not reliable and caregivers could rate this as “easy” to sit up, while the positioning of the patients from a medical provider’s perspective is bad. Although this may be true, the patient-reported outcome measure is intended to assess the perspective of patient or their caregiver, which is different from a functional assessment made by a medical provider. Therefore, a caregiver’s subjective perception of their child’s health status is what we are aiming to measure through the EOSQ-24. Second, a concern about not including a phrase, “due to your child’s spinal deformity” at the end of each question item in the EOSQ-24 to accurately measure health status due to EOS was raised. However, we decided not to include the phrase because caregivers are not capable of identifying health status that is due to the child’s spinal deformity. For example, they may not realize the difficulty breathing is due to EOS.

This study included patients that are entered into 2 national registries and this instrument was tested on a representative group of participants with EOS who undergo surgical procedures. Currently, studies examining the responsiveness of the EOSQ-24 in patients undergoing casting treatment as well as studies evaluating the EOSQ-24 outside of the United States are underway. Further studies evaluating validity, reliability, and responsiveness of the EOSQ-24 beyond 6 months will be needed to confirm the applicability of the instrument to assess long-term follow-up. Although the EOSQ-24 was developed with participants that were representatives of the EOS population, future studies will be needed to evaluate the difference in countries, treatments, settings, or other variables. A new set of normative data for a corresponding group may also be needed.

The EOSQ-24 with proven validity, reliability, and responsiveness will be able to serve as a useful patient-reported outcome measure in studies evaluating interventions. In addition, the

developed EOSQ-24 allows us to assess the HRQoL of those patients, and the parental and financial burden of their caregivers at any time point of the treatment compared with the age-matched healthy children and their families.

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TABLE 1

Correlation Between EOSQ Domain Scores and Pulmonary Function Test

EOSQ Domain	Mean (95% CI)	
	Percent FVC Predicted	Percent FEV ₁ Predicted
General health	0.78 (0.17–1.09) *	0.78 (0.19–1.15) *
Pain/discomfort	0.58 (–0.10 to 0.85)	0.68 (0.12–0.92) *
Pulmonary function	0.62 (–0.23 to 3.53)	0.72 (0.24–3.38) *
Transfer	0.39 (–0.47 to 1.17)	0.36 (–0.61 to 1.37)
Physical function	0.74 (0.17–1.60) *	0.69 (0.06–1.67) *
Daily living	0.71 (0.25–2.30) *	0.69 (0.18–2.39) *
Fatigue/energy level	0.80 (0.36–1.53) **	0.78 (0.34–1.60) **
Emotion	0.79 (0.16–0.94) *	0.82 (0.23–0.99) **
Parental burden	0.55 (–0.21 to 1.00)	0.59 (–0.19 to 1.20)
Financial burden	0.14 (–0.90 to 1.22)	0.08 (–1.03 to 1.23)
Child satisfaction	0.43 (–0.78 to 1.90)	0.47 (–0.74 to 2.04)
Parental satisfaction	0.22 (–0.94 to 1.40)	0.28 (–0.92 to 1.53)

* $P < 0.05$.** $P < 0.01$.

CI indicates confidence interval; EOSQ, Early-Onset Scoliosis Questionnaire; FEV₁, forced expiratory volume in 1 second; FVC, forced vital capacity.

TABLE 2

Known Group Validity: The EOSQ-24 Mean Scores Based on Etiology

	Mean (95% CI)				P
	Idiopathic (N = 29)	Congenital/Structural (N = 15)	Syndromic (N = 18)	Neuromuscular (N = 28)	
General health	80.6 ^a (74.3–86.9)	80.8 ^b (73.0–88.6)	62.5 ^c (51.2–73.8)	64.4 ^d (56.2–72.5)	a vs. b = 0.999 a vs. c = 0.010 a vs. d = 0.010 b vs. c = 0.034 b vs. d = 0.040 c vs. d = 0.988
Pain/fatigue	79.3 ^a (70.2–88.4)	76.7 ^b (62.6–90.7)	72.8 ^c (61.6–84.0)	70.8 ^d (63.0–78.7)	a vs. b = .983 a vs. c = 0.781 a vs. d = 0.502 b vs. c = 0.963 b vs. d = 0.854 c vs. d = 0.992
Pulmonary function	92.9 ^a (87.5–98.2)	92.5 ^b (86.2–98.8)	77.3 ^c (64.0–90.7)	78.8 ^d (67.7–89.9)	a vs. b = 0.999 a vs. c = 0.070 a vs. d = 0.066 b vs. c = 0.156 b vs. d = 0.171 c vs. d = 0.996
Transfer	89.3 ^a (79.6–99.0)	96.4 ^b (88.7–104.1)	85.3 ^c (74.1–96.5)	63.5 ^d (52.3–74.6)	a vs. b = 0.795 a vs. c = 0.947 a vs. d = < 0.001 b vs. c = 0.567 b vs. d = < 0.001 c vs. d = 0.021
Physical function	89.6 ^a (81.4–97.7)	89.9 ^b (77.9–101.8)	66.2 ^c (52.0–80.3)	47.6 ^d (29.0–66.2)	a vs. b = 0.999 a vs. c = 0.089 a vs. d = < 0.001

Mean (95% CI)					P
	Idiopathic (N = 29)	Congenital/Structural (N = 15)	Syndromic (N = 18)	Neuromuscular (N = 28)	
Daily living	80.8 ^a (71.3–90.3)	83.3 ^b (69.6–97.1)	47.7 ^c (27.3–68.0)	37.5 ^d (25.9–49.1)	b vs. c = 0.177 b vs. d = < 0.001 c vs. d = 0.252 a vs. b = 0.993 a vs. c = 0.003 a vs. d = < 0.001 b vs. c = 0.006 b vs. d = < 0.001
Fatigue/energy level	86.2 ^a (77.3–95.1)	86.7 ^b (76.4–97.0)	68.1 ^c (56.1–80.0)	55.8 ^d (45.1–66.5)	c vs. d = 0.684 a vs. b = 0.999 a vs. c = 0.068 a vs. d = < 0.001 b vs. c = 0.132 b vs. d = < 0.001
Emotion	82.4 ^a (74.1–90.7)	82.5 ^b (73.9–91.1)	78.5 ^c (70.6–86.4)	70.5 ^d (61.0–80.0)	c vs. d = 0.344 a vs. b = 0.999 a vs. c = 0.914 a vs. d = 0.142 b vs. c = 0.937 b vs. d = 0.256 c vs. d = 0.565 a vs. b = 0.619 a vs. c = 0.925 a vs. d = < 0.001 b vs. c = 0.364 b vs. d = < 0.001 c vs. d = 0.032
Parental burden	73.6 ^a (65.9–81.3)	81.1 ^b (73.5)	69.9 ^c (59.0–80.7)	53.0 ^d (44.9–61.1)	a vs. b = 0.527 a vs. c = 0.682 a vs. d = 0.592
Financial burden	74.1 ^a (64.8–83.5)	61.5 ^b (42.4–80.7)	64.7 ^c (48.9–80.5)	64.8 ^d (54.1–75.5)	

Mean (95% CI)					P
	Idiopathic (N = 29)	Congenital/Structural (N = 15)	Syndromic (N = 18)	Neuromuscular (N = 28)	
Child satisfaction	71.9 ^{*a} (51.2–92.6)	79.2 ^{*b} (44.3–114.0)	50.0 ^{*c} (17.5–82.5)	81.3 ^{*d} (43.2–119.3)	b vs. c = 0.990 b vs. d = 0.985 c vs. d = 0.999 a vs. b = 0.956 a vs. c = 0.551 a vs. d = 0.938 b vs. c = 0.355 b vs. d = 0.999
Parent satisfaction	65.6 ^{*a} (36.2–95.1)	87.5 ^{*b} (65.5–109.5)	68.8 ^{*c} (30.7–106.8)	87.5 ^{*d} (64.5–110.5)	c vs. d = 0.373 a vs. b = 0.460 a vs. c = 0.998 a vs. d = 0.563 b vs. c = 0.710 b vs. d = 0.999 c vs. d = 0.763

* N = 8 for idiopathic, N = 6 for congenital/structural, N = 4 for syndromic, and N = 4 for neuromuscular.

Bolded *P*-values indicate significant values.

CI indicates confidence interval; EOSQ-24, 24-item Early-Onset Scoliosis Questionnaire.

TABLE 3

Responsiveness: The EOSQ-24 Domain Scores Significantly Different Between Preoperative and Postoperative Assessments

Cohort	Domain	Mean (95% CI)				P (Pre vs. Post)	P (Norm vs. Pre)	P (Norm vs. Post)
		Preoperative	Postoperative	Norm				
Neuromuscular	General health	61 (53–72)	71 (62–79)	87 (85–89)	0.204	< 0.001	< 0.001	< 0.001
	Fatigue	53 (41–65)	68 (54–78)	92 (91–93)	0.083	< 0.001	< 0.001	< 0.001
	Pulmonary function	77 (64–90)	87 (73–101)	98 (97–99)	0.131	0.003	0.110	0.110
	Transfer	59 (48–73)	69 (55–82)	98 (98–99)	0.367	< 0.001	< 0.001	< 0.001
	Emotion	67 (57–78)	80 (69–91)	94 (93–95)	0.046	< 0.001	0.010	0.010
Spinal muscular atrophy	Parental burden	49 (41–59)	59 (50–68)	92 (91–93)	0.020	< 0.001	< 0.001	< 0.001
	General health	56 (34–79)	75 (66–84)	87 (82–92)	0.256	0.013	0.007	0.007
	Pulmonary function	61 (31–91)	80 (48–113)	98 (98–99)	0.093	0.022	0.222	0.222
	Transfer	34 (15–54)	61 (28–93)	99 (98–100)	0.045	< 0.001	0.028	0.028
	Physical function	33 (–19 to 86)	13 (–4 to 29)	98 (96–99)	0.482	0.023	< 0.001	< 0.001
	Daily living	28 (6–50)	18 (3–33)	88 (82–94)	0.476	< 0.001	< 0.001	< 0.001
	Fatigue	36 (19–53)	52 (35–69)	93 (90–95)	0.078	< 0.001	0.001	0.001
	Emotion	50 (37–63)	63 (41–84)	94 (92–97)	0.286	< 0.001	0.011	0.011
	Parental burden	42 (30–54)	51 (33–70)	94 (92–97)	0.428	< 0.001	0.002	0.002
	Financial burden	78 (57–99)	64 (42–87)	98 (96–99)	0.407	0.065	0.010	0.010
Idiopathic	Physical function	94 (84–105)	84 (70–98)	98 (96–99)	0.048	0.417	0.096	0.096
	Emotion	83 (72–95)	61 (48–75)	95 (93–97)	0.006	0.048	< 0.001	< 0.001
	Financial burden	46 (22–71)	36 (3–68)	98 (96–100)	0.141	0.003	0.004	0.004
With intraoperative complication		76 (66–79)	59 (40–78)	92 (86–97)	0.092	0.165	0.013	0.013

Bolded *P*-values indicate significant values.

CI indicates confidence interval; EOSQ-24, 24-item Early-Onset Scoliosis Questionnaire.

TABLE 4

Normative Data Breakdown by Age and Domains

Mean (95% CI)										
Age (y)	General Health	Pain/Discomfort	Pulmonary Function	Transfer	Physical Function	Daily Living	Fatigue/Energy Level	Emotion	Parental Burden	Financial Burden
0	80 (71-89)	92.5 (84-101)	98.75 (96-102)	97.5 (92-103)	92.5 (84-101)	86.1111 (72-100)	86.1111 (70-102)	98.4375 (95-102)	77.5 (60-95)	92.5 (80-105)
1	81.25 (75-88)	86.4583 (75-98)	94.7917 (83-106)	95.8333 (87-105)	92.3611 (82-103)	90.6250 (82-100)	91.6667 (83-100)	94.3182 (86-102)	87.5 (81-94)	95.8333 (90-102)
2	76.9231 (69-84)	82.6923 (73-92)	97.1154 (94-100)	100 (100-100)	100 (100-100)	79.8077 (71-88)	91.3462 (86-96)	94.2308 (89-99)	85.7692 (78-94)	96.1538 (90-100)
3	80.25 (72-89)	94.6429 (86-100)	100 (100-100)	98.2143 (95-100)	100 (100-100)	96.4286 (93-99)	99.1071 (97-100)	98.2143 (95-100)	95 (88-98)	100 (100-100)
4	80.25 (76-86)	86.4583 (79-96)	97.9167 (95-100)	100 (100-100)	99.3056 (98-100)	89.5833 (81-97)	97.9167 (96-100)	98.9583 (97-100)	95 90-99)	100 (100-100)
5	90.6667 (89-96)	94.7917 (86-100)	98.9583 (97-100)	97.9167 (94-100)	95.8333 (88-100)	87.5 (75-97)	93.75 (89-98)	93.75 (85-100)	93.3333 (85-99)	97.9167 (94-100)
6	84.9099 (78-91)	90.9091 (84-98)	98.8636 (97-100)	100 (100-100)	96.9697 (91-100)	77.2727 (61-91)	92.0455 (82-99)	93.1818 (86-100)	89.5455 (79-98)	95.4545 (86-100)
7	90 (85-95)	93.75 (88-100)	97.5 (92-100)	97.5 (93-100)	98.3333 (95-100)	92.5 (81-100)	93.75 (85-100)	95 (85-100)	93 (85-99)	100 (100-100)
8	87.5 (77-97)	87.5 (77-96)	97.9167 (94-100)	97.9167 (94-100)	97.9167 (94-100)	97.9167 (95-100)	90.6250 (79-100)	95.8333 (88-100)	94.5833 (85-100)	97.9167 (94-100)
9	82.5 (89-96)	85 (74-94)	97.5 (94-100)	100 (100-100)	99.1667 (98-100)	96.25 (91-100)	97.5 (94-100)	92.5 (81-100)	95.5 (88-100)	97.5 (93-100)
10	87.5 (79-93)	75 (58-89)	97.2222 (93-100)	91.6667 (83-100)	92.5926 (80-100)	88.8889 (75-99)	93.0556 (83-100)	91.6667 (85-99)	90 (83-97)	97.2222 (92-100)
11	85.143 (80-91)	76.7857 (64-89)	94.6429 (87-100)	100 (100-100)	100 (100-100)	100 (100-100)	76.7857 (61-89)	96.4286 (93-100)	97.8571 (96-100)	100 (100-100)
12	80.5556 (69-92)	72.2222 (56-89)	97.2222 (93-100)	94.4444 (78-100)	99.0741 (96-100)	100 (100-100)	88.8889 (81-96)	93.0556 (72-100)	92.2222 (73-100)	91.6667 (78-100)
13	90 (80-100)	85 (65-100)	95 (85-100)	100 (100-100)	100 (100-100)	92.5 (78-100)	82.5 (73-93)	92.5 (78-100)	98 (96-100)	95 (85-100)
14	91.6667 (88-100)	100 (100-100)	95.8333 (88-100)	100 (100-100)	100 (100-100)	100 (100-100)	95.8333 (88-100)	100 (100-100)	100 (100-100)	100 (100-100)
15	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)	75 (75-75)	75 (75-75)	100 (100-100)	100 (100-100)	100 (100-100)
16	82.5 (79-100)	91.6667 (75-100)	95.8333 (88-100)	100 (100-100)	100 (100-100)	100 (100-100)	75 (67-84)	87.5 (75-100)	96.6667 (93-100)	100 (100-100)
17	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)	100 (100-100)
18	87.50 (87.5-87.5)	37.50 (37.5-37.5)	100 (100-100)	25.00 (25-25)	83.33 (83.33-83.33)	37.50 (37.5-37.5)	62.50 (62.5-62.5)	62.50 (62.5-62.5)	60 (60-60)	75 (75-75)

CI indicates confidence interval.

TABLE 5

The Diagnoses Distribution for Normative Subjects Completing EOSQ

Diagnosis	Patients [n (%)]
Developmental dysplasia of hip	23 (16)
Fracture	57 (40)
Femoral anteversion	12 (9)
Tibial torsion	13 (9)
Joint pain	8 (6)
Gait abnormality	18 (13)
Pes planus	7 (5)
Other	3 (2)
Total	141 (100)

EOSQ indicates Early-Onset Scoliosis Questionnaire.