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Original Research

Impact of the COVID-19 Pandemic on Health-related Quality of Life in Children with Early Onset Scoliosis

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Abstract

Background: Throughout the COVID-19 pandemic, decreases in health-related quality of life (HRQoL) have been observed in adults and children, with isolation, economic disruption, school closures, and health-related anxiety likely contributing. In this study, we evaluated the impact of COVID-19 on self-reported HRQoL of EOS patients and their caregivers using the Early Onset Scoliosis Questionnaire-24 (EOSQ-24).

Methods: Patients with EOS and their caregivers enrolled in the Pediatric Spine Study Group (PSSG) registry with EOSQ scores from the year before the COVID-19 pandemic and the first year during COVID-19 were included. Two years of before-COVID-19 baseline EOSQ scores were recorded for each patient. We recorded patient medical demographics, scoliosis etiology, and comorbidities.

Results: 618 patients met inclusion criteria (255 male, 363 female). All EOSQ subscores increased significantly from pre-COVID to early-COVID ($p < 0.001$, $p < 0.05$, respectively), though the mean difference was well below the proposed EOSQ-24 MCID. There was no evidence of change in Combined HRQoL or impact- and satisfaction-related scores between early-COVID to late-COVID ($p > 0.37$). When stratified by etiology (40.3% idiopathic, 17.6% syndromic, 17.8% neuromuscular, 23.0% congenital), there was no evidence of decrease in the HRQoL combined score or other subscores in any subgroup between Pre-COVID and during COVID.

Conclusions: Overall, there was no evidence of negative impact on HRQoL by the COVID-19 pandemic for children with EOS or their caregivers. In the future, protective factors contributing to the resilience for this population may be explored further.

Level of Evidence: III

Key Concepts

- Decreases in health-related quality of life have been observed among adults and children during the COVID-19 pandemic.
- The Early Onset Scoliosis Questionnaire allows for health-related quality of life measurement of patients with Early Onset Scoliosis and their caregivers.
- Among EOS patients in a national registry who filled out EOSQ-24 questionnaires before and during the COVID-19 pandemic, we found no evidence of decrease in HRQoL between pre-COVID, early COVID, and late COVID timepoints.

Introduction

EOS is a progressive, chronic condition often accompanied by a number of comorbidities and pulmonary complications requiring longitudinal treatment with the potential for repeated procedures. Therefore, monitoring health-related quality of life (HRQoL) and burden of care is vital for patients with Early Onset Scoliosis (EOS) and their families.[1-3] The COVID-19 pandemic has increased feelings of isolation and health-related anxiety among the general population and in subpopulations with chronic health conditions. Decreases in HRQoL have been observed in adults, adolescents, and young children.[4-6] School-age children have been greatly impacted by school closures, limited social contact, and suspension of extracurricular activities. In children whose internet usage increased during the COVID-19 pandemic, decreases in self-reported physical well-being, emotional well-being, self-esteem, family subscores, school-related subscores, and overall quality of life have been observed.[7] For parents and caregivers, personal and professional changes are compounded by disruptions in children's routines due to the pandemic, potentially contributing to physical, emotional, and economic stress. These changes may be particularly salient for individuals with pre-existing health conditions and their families.

The Early Onset Scoliosis Questionnaire-24 (EOSQ-24) measures HRQoL for both patients with Early Onset Scoliosis (EOS) and their caregivers and presents an opportunity to comprehensively evaluate HRQoL in this population.[3] This instrument has been utilized to measure and compare HRQoL and burden of care as outcomes of different treatments as well as its associations with various patient characteristics, such as EOS etiology.[8-12] Etiologies of EOS are divided conventionally into idiopathic, congenital, syndromic, and neuromuscular. Congenital scoliosis includes structural causes of curves such as hemivertebrae, fused ribs, VATER, etc. (vertebral defects, imperforate anus, tracheoesophageal fistula, and radial and renal dysplasia). Neuromuscular scoliosis is associated with spinal muscular atrophy, cerebral palsy, and other muscle tone disorders. Finally, syndromic scoliosis commonly includes Ehlers-Danlos and other connective tissue disorders, Prader-Willi syndrome, etc.[13] Comorbidities and patient characteristics associated with more complex etiologies, such as neuromuscular EOS, have demonstrated significantly lower HRQoL values in prior studies,[14] while idiopathic EOS has

demonstrated comparatively better results.[2] The purpose of this study was to compare HRQoL of EOS patients and their caregivers using the EOSQ-24 in the 2 years prior to and 1 year after the beginning of the COVID-19 pandemic, with stratification by etiology of EOS. These findings may allow identification of patient populations at greater risk of HRQoL changes with interruptions in daily life or in-person healthcare. We analyzed a single group of patients with HRQoL measures at multiple times, including before and during the pandemic. Due to increased psychosocial stress and closure of schools and activities, we hypothesized that HRQoL would decrease after March 2020 compared to before March 2020.

Methods

Study Design and Settings

Upon the Institutional Review Board approval, a retrospective study using a multi-center international EOS registry was conducted. The data included patients from 27 institutions seen in the routine office visits from 2018 to 2021.

Study Patients

All patients with EOS, and their caregivers, who had office visits and received at least one Early Onset Scoliosis Questionnaire-24 (EOSQ-24) from April 2020 to February 2021 and at least one EOSQ-24 from March 2019 to February 2020 were included. Patients receiving any new intervention or change in treatment type (e.g., index spinal instrumentation, spinal fusion, initial implant or removal of a growth-friendly lengthening device, or initiation or completion of casting or bracing) from March 2019 to February 2021 were excluded. As a result, among the included patients, no changes in treatment type or index procedures were recorded during the Last Baseline, Pre-COVID, Early COVID, or Late COVID timepoints. This exclusion was made to limit the influence of changes in treatment, rather than the COVID-19 pandemic, on EOSQ-24 score.

Early Onset Scoliosis 24-Item Questionnaire (EOSQ-24)

All EOSQ-24 domain scores were used to measure HRQoL of patients and burden of care of their parents during the routine office visits. EOSQ-24 subscores consisted of General Health, Pain/Discomfort, Pulmonary Function, Transfer, Physical Function, Daily Living, Fatigue/Energy Level, and Emotion, which were averaged to create the "HRQoL Composite" score. Survey data also included Parental Impact, Financial Impact, Child Satisfaction, and Parent Satisfaction scores.

Baseline, Pre- and During-COVID Assessments

The EOSQ-24 scores were compared before and during COVID-19 to assess the impact of the COVID-19 pandemic on HRQoL of patients and burden of their caregivers. The Before-COVID assessment was defined as EOSQ-24 measures completed during visits from March 2019 to February 2020. This was then divided into two half-year timepoints: Last Baseline (March 2019 – August 2019) and Pre-COVID (September 2019 – February 2020). The During-COVID assessment was defined as EOSQ-24 measures administered during the COVID-19 pandemic from April 2020 to March 2021. The During-COVID was further categorized into two visit phases: Early COVID (earliest 4/2020 – 9/2020) and Late COVID (earliest 10/2020 – 3/2021), in order to divide the first year of the pandemic into 6-month sections. Early COVID and Late COVID scores were separated to evaluate for potential changes in HRQoL over the course of the pandemic. Due to the anticipated variation in the date of initial COVID-19 lockdown between patients in different communities and school systems, the month of March 2020 was treated as a transitional period and EOSQ-24 surveys from March 2020 were excluded.

Last Baseline, Pre-COVID, and During-COVID assessments were compared to differences in scores in HRQoL in the previous year (February 2018 to February 2019). Thus, in total, 2 years of baseline data before the COVID-19 pandemic were collected to establish the mean change in EOSQ-24 for individual patients over time. The baseline was further divided into another two phases at 6-month intervals (2/2018 – 8/2018, 9/2018 – 2/2019), making up the First and Second Baselines.

Other Variables of Interest

Patient age, sex, scoliosis etiology, ambulatory status, respiratory support, and body mass index (BMI) were recorded. Etiologies were categorized into idiopathic, syndromic, neuromuscular, and congenital.[13] Ambulatory status was categorized into ambulatory, limited-ambulation (walking with device, household ambulation, etc.), non-ambulatory, and pre-ambulatory. Respiratory support was categorized into no assistance, advanced volume-assure pressure (AVAP), bilevel positive airway pressure (BIPAP), continuous positive airway pressure (CPAP), nasal canula, tracheostomy, or other positive pressure vent. Treatment interventions at any of the above timepoints were recorded.

Statistical Analysis

Differences in demographics of patients with surveys at each time point were examined by t-tests or $\chi 2$ tests. EOSQ-24 scores were compared using Student's paired t-test with Welch's Correction. Statistical analysis was performed with Excel (Microsoft Inc, Redwood, WA) and RStudio Version 1.3.1093 (RStudio, Boston, MA).

Results

Study Patients

Overall, 618 patients were included (Figure 1). All patients received at least one EOSQ-24 survey during latest baseline and/or Pre-COVID timepoints (n = 423, n = 438, respectively). All patients also received at least one EOSQ-24 survey during either or both the Early COVID or Late COVID timepoints (n = 400, n = 366). Two hundred seventy-six patients received a survey at first baseline and 300 patients received a survey at second baseline.

Of the included patients, 41.3% were male. Mean age was 8.14 ± 3.74 years at latest baseline and 8.23 ± 3.84 years at Pre-COVID. Classified by etiology, 40.3% of patients were diagnosed as Idiopathic, 17.6% as Syndromic, 17.8% as Neuromuscular, and 23.0% as Congenital. Mean BMI in the year preceding the COVID-19 pandemic was 17.46 ± 4.09. A majority

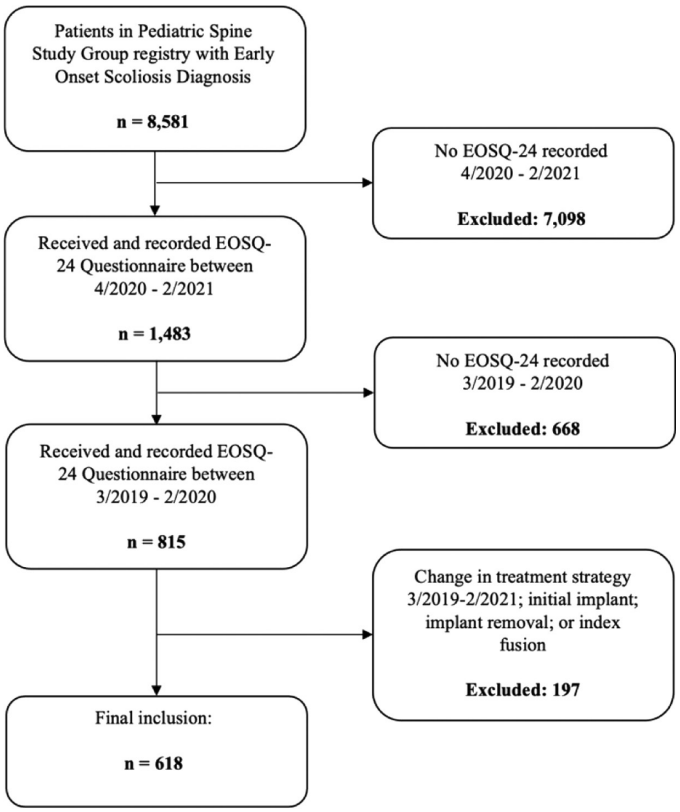

Figure 1. *Study patient inclusion and exclusion.*

of patients were ambulatory (66.7%) and required no respiratory assistance (75.7%) (Table 1).

Among the included patients, no changes in treatment type or index procedures were recorded during the Last Baseline, Pre-COVID, Early COVID, or Late COVID timepoints, as patients with interventions between March 2019 and February 2021 were excluded. Patients with new interventions during the first and second baseline periods were not excluded, as long as they had no new interventions from March 2019 to February 2021. During second baseline, 56 patients (18.7%) received new interventions, with no evidence of difference in HRQoL scores during second or third baseline (p = 0.77, p = 0.34), compared to patients with no interventions during second baseline. Forty-five patients (16.3%) received interventions during first baseline, with no evidence of difference in HRQoL at first baseline (p = 0.33) or second baseline (70.3 intervention cohort, 75.8 non-intervention cohort, p = 0.08).

Table 1. Characteristics of Included Patients

Sex	**Male, n (%)**	**255 (41.3)**
	Female, n (%)	**363 (58.7)**
Age at latest baseline, years ± stdev		8.14 ± 3.74
Age at Pre-COVID, years ± stdev		8.23 ± 3.84
Etiology	Idiopathic, n (%)	249 (40.3)
	Syndromic, n (%)	109 (17.6)
	Neuromuscular, n (%)	110 (17.8)
	Congenital, n (%)	142 (23.0)
	other, n (%)	8 (1.3)
BMI, mean ± stdev		17.46 ± 4.09
Ambulatory Status	Ambulatory, n (%)	412 (66.7)
	Limited Ambulation, n (%)	39 (6.3)
	Non-ambulatory, n (%)	76 (12.3)
	Pre-ambulatory, n (%)	11 (1.8)
	data not available, n (%)	80 (12.9)
Respiratory Support	No assistance, n (%)	468 (75.7)
	AVAP, n (%)	1 (0.2)
	BIPAP, n (%)	13 (2.1)
	CPAP, n (%)	6 (1.0)
	Nasal canula, n (%)	6 (1.0)
	Other positive pressure vent, n (%)	2 (0.3)
	Tracheostomy, n (%)	22 (3.6)
	data not available, n (%)	100 (16.2)

Pre- and During-COVID Health-Related Quality of Life

HRQoL Composite score was significantly increased in Early COVID versus Pre-COVID, despite a mean difference of only 3.1 points (75.6 ± 17.0 to 78.7 ± 16.0, $p < 0.001$). From Early COVID to Late COVID, there was no evidence of change in HRQoL Composite score (Table 2). In terms of subscores, General Health, Pulmonary Function, Fatigue/Energy Level, and Physical Function increased from Pre-COVID to either Early COVID or Late COVID ($p < 0.05$). From Pre-COVID to Early COVID, Parental Impact, Financial Impact, Child Satisfaction, and Parent Satisfaction all significantly increased (73.5 to 78.1, 78.2 to 83.0, 73.1 to 76.2, 74.2 to 77.3, respectively, $p < 0.05$), with no evidence of subsequent change from Early to Late COVID.

Baseline Health-Related Quality of Life

No evidence of difference was found between HRQoL Composite scores at Baseline timepoints, with mean scores ranging from 75.1 to 76.4 out of 100 (Table 3). Financial Impact decreased from first baseline to second baseline, and Parental Impact increased from last baseline to Pre-COVID. No other evidence of difference

Table 2. Mean EOSQ-24 Subscores Before and After The COVID-19 Pandemic

	Pre-COVID	→	Early COVID	→	Late COVID
	n = 438	*p-value*	n = 400	*p-value*	n = 366
General Health	72.6 ± 19.3	**<0.001***	80.2 ± 18.1	0.173	80.3 ± 17.8
Pain/Discomfort	76.1 ± 20.4	0.584	75.5 ± 20.1	0.160	76.9 ± 20.4
Pulmonary Function	87.0 ± 18.9	**0.020***	88.2 ± 18.8	0.955	87.7 ± 18.4
Transfer	80.0 ± 24.4	0.389	82.8 ± 23.2	0.475	82.2 ± 25.4
Physical Function	77.1 ± 26.9	0.113	78.7 ± 27.2	**0.046***	79.0 ± 26.7
Daily Living	71.4 ± 29.2	**0.083**	73.1 ± 28.5	0.376	70.7 ± 29.3
Fatigue/Energy Level	75.6 ± 25.2	**0.045***	77.1 ± 25.2	0.999	76.8 ± 23.9
Emotion	77.2 ± 22.3	0.222	78.4 ± 21.6	0.409	76.4 ± 22.3
HRQoL Composite	75.6 ± 17.0	**<0.001***	78.7 ± 16.0	0.378	77.5 ± 16.3
Parental Impact	73.5 ± 23.5	**<0.001***	78.1 ± 21.9	0.754	74.3 ± 22.4
Financial Impact	78.2 ± 23.4	**0.001***	83.0 ± 21.6	0.671	80.9 ± 21.4
Child Satisfaction	73.1 ± 23.5	**0.023***	76.2 ± 21.8	0.377	74.2 ± 22.9
Parent Satisfaction	74.2 ± 23.2	**0.036***	77.3 ± 22.0	0.837	75.6 ± 22.4

was found between impact-related and satisfaction-related EOSQ-24 scores.

Categorical Analysis by Magnitude of Change Across Timepoints

While the minimally clinically important difference (MCID) for the EOSQ-24 has yet to be fully established, a threshold of ≥ 10 points has been suggested.[2] Using a threshold of ≥ 10 point change, both in the positive and negative directions, EOSQ-24 score changes are described categorically in Table 4. Between First, Second, and Third Baselines, 11.6% and 12.7% of patients reported increased EOSQ-24 scores by ≥ 10 points. From Third Baseline to Pre-COVID, Pre-COVID to Early COVID, and Early to Late COVID, 16.3%, 20.0%, and 12.8% of patients reported increased scores by ≥ 10 points.

Stratification by Scoliosis Etiology

HRQoL Composite score increased from Pre-COVID to Early COVID for Idiopathic, Neuromuscular, and Congenital etiologies (Table 5, Figure 2). The idiopathic cohort also showed increased Financial Impact scores at the same timepoints, and the congenital cohort showed increased Financial Impact and Parent Satisfaction scores ($p < 0.05$). The Neuromuscular cohort showed increases in all impact-related and satisfaction-related scores from Pre-COVID to Early COVID ($p < 0.05$). There was no evidence of difference in HRQoL Composite, impact-related, or satisfaction-related scores in the Syndromic cohort.

From Early COVID to Late COVID, there was no evidence of change in HRQoL scores in any etiology cohort except for a decrease in HRQoL Composite score in congenital patients (80.8 ± 17.4, 78.7 ± 15.0, $p = 0.05$).

Discussion

In this study, we analyzed EOSQ-24 surveys from a multicenter, cross-national registry to determine whether HRQoL changed over the course of the pandemic. We examined data from the first year of the COVID-19 pandemic and the preceding 2 years as a baseline.

Table 3. Mean EOSQ-24 scores with 2-year Baseline and 1 year During the COVID-19 Pandemic

	Baseline (2/2018 – 8/2018)	→	**Baseline (9/2018 – 2/2019)**	→	**Baseline (3/2019 – 8/2019)**	→	**Pre-COVID (9/2019 – 2/2020)**	→	**Early COVID (4/2020 – 9/2020)**	→	**Late COVID (10/2020 – 3/2021)**
	n = 276	*p-value*	n = 300	*p-value*	n = 432	*p-value*	n = 438	*p-value*	n = 400	*p-value*	n = 366
HRQoL Composite	76.4 ± 16.6	NS	75.1 ± 16.6	NS	76.1 ± 16.3	NS	75.6 ± 17.0	**<0.001***	78.7 ± 16.0	NS	77.5 ± 16.3
Parental Impact	74.0 ± 22.7	NS	72.3 ± 22.2	NS	73.3 ± 23.1	**0.026***	73.5 ± 23.5	**<0.001***	78.1 ± 21.9	NS	74.3 ± 22.4
Financial Impact	78.7 ± 23.8	**0.064**	77.8 ± 23.4	NS	79.3 ± 24.0	NS	78.2 ± 23.4	**0.001***	83.0 ± 21.6	NS	80.9 ± 21.4
Child Satisfaction	72.5 ± 24.5	NS	71.8 ± 23.8	NS	73.3 ± 23.6	NS	73.1 ± 23.5	**0.023***	76.2 ± 21.8	NS	74.2 ± 22.9
Parent Satisfaction	75.3 ± 23.6	NS	74.4 ± 22.9	NS	74.7 ± 23.4	NS	74.2 ± 23.2	**0.036***	77.3 ± 22.0	NS	75.6 ± 22.4

Table 4. Per Patient Change in EOSQ-24 Score Grouped by ≥ 10-point Difference

	Baseline (2/2018 – 8/2018)	→	**Baseline (9/2018 – 2/2019)**	→	**Baseline (3/2019 – 8/2019)**	→	**Pre-COVID (9/2019 – 2/2020)**	→	**Early COVID (4/2020 – 9/2020)**	→	**Late COVID (10/2020 – 3/2021)**
Increase ≥ 10 points n (%)		20 (11.6)		30 (12.7)		41 (16.3)		56 (20.0)		19 (12.8)	
Δ < 10 points n (%)		132 (76.3)		163 (68.8)		179 (71.3)		199 (71.1)		115 (77.7)	
Decrease ≥ 10 points n (%)		21 (12.1)		43 (18.1)		31 (12.4)		25 (8.9)		14 (9.5)	

Table 5. Mean EOSQ-24 Score by Timepoint, Stratified by EOS Etiology

	Baseline (2/2018 – 8/2018)	→	**Baseline (9/2018 – 2/2019)**	→	**Baseline (3/2019 – 8/2019)**	→	**Pre-COVID (9/2019 – 2/2020)**	→	**Early COVID (4/2020 – 9/2020)**	→	**Late COVID (10/2020 – 3/2021)**
Idiopathic (n = 249)	115	*p-value*	135	*p-value*	180	*p-value*	180	*p-value*	161	*p-value*	141
HRQoL Composite	82.2 ± 13.9	NS	80.2 ± 14.3	NS	82.6 ± 13.3	NS	83.6 ± 13.9	**0.041***	84.6 ± 13.2	NS	84.7 ± 13.6
Parental Impact	81.3 ± 18.2	NS	78.5 ± 19.6	NS	81.4 ± 17.4	**0.021***	84.3 ± 17.8	**0.073**	86.0 ± 16.2	NS	83.5 ± 17.1
Financial Impact	81.5 ± 22.4	NS	80.3 ± 21.9	NS	81.6 ± 23.3	NS	82.2 ± 21.0	**0.034***	83.8 ± 21.3	NS	83.3 ± 19.9
Child Satisfaction	79.7 ± 21.5	NS	78.6 ± 20.6	NS	80.1 ± 21.3	NS	81.0 ± 22.4	NS	82.9 ± 20.1	NS	82.4 ± 20.6
Parent Satisfaction	82.6 ± 19.2	NS	80.6 ± 19.8	NS	81.4 ± 21.8	**0.056**	83.7 ± 20.1	NS	83.1 ± 20.3	NS	83.8 ± 19.6
Syndromic (n = 109)	55		52		81		73		77		64
HRQoL Composite	70.2 ± 16.1	NS	69.3 ± 17.3	NS	70.9 ± 13.5	NS	68.9 ± 13.9	NS	72.3 ± 13.3	NS	73.4 ± 15.0
Parental Impact	66.6 ± 22.8	NS	65.7 ± 22.1	NS	65.7 ± 22.9	NS	64.3 ± 20.9	NS	69.7 ± 22.2	NS	68.7 ± 22.9
Financial Impact	74.5 ± 23.8	NS	77.0 ± 23.4	NS	76.9 ± 23.9	NS	75.4 ± 24.8	NS	79.3 ± 21.1	NS	81.3 ± 22.0
Child Satisfaction	65.7 ± 22.4	NS	64.8 ± 24.4	NS	66.6 ± 21.3	NS	62.9 ± 21.8	NS	66.0 ± 18.7	NS	69.0 ± 22.9
Parent Satisfaction	67.1 ± 23.7	NS	64.7 ± 23.0	NS	65.6 ± 23.3	NS	62.3 ± 23.2	NS	67.0 ± 21.0	NS	69.8 ± 22.2
Neuromuscular (n = 110)	53		51		75		80		63		73
HRQoL Composite	67.7 ± 17.5	NS	66.4 ± 18.0	**0.083**	65.8 ± 16.4	NS	63.3 ± 16.3	**<0.001***	68.9 ± 15.9	NS	66.2 ± 16.2
Parental Impact	63.2 ± 24.1	NS	60.0 ± 24.8	NS	60.0 ± 24.0	NS	55.2 ± 23.4	**0.001***	64.5 ± 24.5	NS	57.6 ± 23.0
Financial Impact	73.5 ± 26.2	**0.019***	72.1 ± 26.3	NS	73.6 ± 26.4	NS	73.7 ± 23.4	**0.017***	81.3 ± 24.6	NS	78.7 ± 20.5
Child Satisfaction	60.8 ± 27.5	NS	60.1 ± 26.9	NS	61.9 ± 24.1	NS	60.2 ± 20.1	**0.039***	65.2 ± 20.0	NS	60.8 ± 21.6
Parent Satisfaction	64.6 ± 26.2	NS	65.3 ± 24.9	NS	64.8 ± 23.0	NS	59.3 ± 20.8	**0.039***	65.9 ± 20.0	NS	61.0 ± 21.8
Congenital (n = 142)	52		60		92		100		94		84
HRQoL Composite	79.0 ± 16.5	NS	76.4 ± 15.1	NS	76.6 ± 17.8	NS	76.8 ± 16.5	**0.019***	80.8 ± 17.4	**0.050***	78.7 ± 15.0
Parental Impact	76.2 ± 23.8	NS	75.7 ± 20.0	NS	75.1 ± 25.6	NS	75.4 ± 23.2	NS	80.4 ± 21.8	NS	77.8 ± 20.2
Financial Impact	82.5 ± 23.3	NS	77.5 ± 25.7	NS	81.5 ± 23.2	NS	77.9 ± 25.4	**0.027***	86.0 ± 20.6	NS	79.5 ± 23.1
Child Satisfaction	75.5 ± 24.5	NS	72.0 ± 22.8	NS	74.2 ± 24.6	NS	75.0 ± 22.7	NS	80.4 ± 22.1	NS	75.0 ± 22.0
Parent Satisfaction	77.9 ± 24.1	NS	77.5 ± 22.4	NS	76.9 ± 22.3	NS	76.3 ± 21.0	**0.018***	83.2 ± 21.0	NS	77.7 ± 20.9

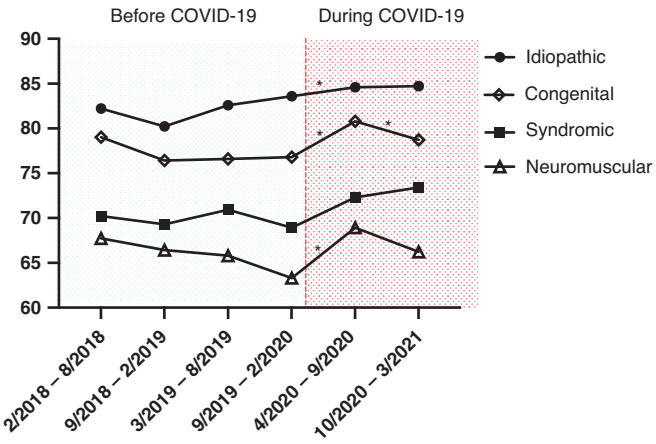

Figure 2. *Mean HRQoL Composite Score by Etiology.*

Despite disruptions to daily living for children during COVID-19, there was no overall evidence of decrease in HRQoL during the pandemic among EOS patients. When stratified by etiology, increases in HRQoL were found in idiopathic, neuromuscular, and congenital cohorts from before COVID-19 to immediately after. No evidence of difference was found in the syndromic cohort. Over the course of the first year of COVID-19, no evidence of decrease in HRQoL was found in idiopathic, syndromic, and neuromuscular cohorts. We found decrease only in the HRQoL Composite score in the congenital cohort over the course of the pandemic, with no evidence of difference in impact- and satisfaction-related scores.

While statistically significant increases in HRQoL were found both in the overall population and when stratified by etiology in all cohorts except syndromic patients, none of the mean differences from before onset of the pandemic to immediately after exceeded 10 points out of 100. As a result, increased scores may not reach a clinically important difference, which has been suggested to be ≥ 10 points.[2] Further studies to more fully establish the minimally clinically importance difference for the EOSQ-24 are ongoing. However, the lack of evidence of decrease in HRQoL during the COVID-19 pandemic is in itself an important finding.

The results from this study differ from several other child/adolescent population groups that have demonstrated decreased HRQoL scores during the pandemic.[5,15] One study by Riiser et al. of 2,205 adolescents between 16 and 19 years old in Norway found quarantine isolation to be associated with a significant decrease in HRQoL, although increased health literacy was associated with an increased HRQoL.[16] There are notable differences between EOS patients and the patient population in the study by Riiser et al., with EOS patients in this study being much younger (with an average age around 8 years old) and likely having on average more frequent interactions with healthcare systems. Another study by Adibelli et al. of children ages 7-13 found higher HRQoL scores during the pandemic among younger children (≤ 10 years), which corresponds better to the EOS population in our study, although overall decreases in HRQoL were still noted.[7] Increased involvement in healthcare due to chronic conditions for EOS patients and their caregivers may correspond loosely to the increased health literacy mentioned in Riiser et al., which was found to mitigate decreases in HRQoL during the pandemic.

Historically, HRQoL studies in EOS populations have shown lower scores associated with non-ambulatory status, complications, and neuromuscular etiology.[14] Idiopathic EOS has been associated with relatively better EOSQ-24 scores.[2] This corresponds well with the results of etiology stratification in our study, which found lowest EOSQ-24 scores at all time points in the neuromuscular cohort and highest scores in the idiopathic cohort. This prior research has demonstrated lower EOSQ-24 scores in syndromic or neuromuscular etiologies even prior to treatment.[17] This may reflect the increased care effort and demands of daily living for many patients in the neuromuscular cohort, such as those with spinal muscular atrophy, muscular dystrophy, or cerebral palsy. Further research may be warranted on the demands of daily living during the COVID-19 pandemic for both patients and caregivers between the etiologies to identify protective factors for idiopathic and syndromic versus congenital and neuromuscular diagnoses.

In a previous survey of parents and caregivers of children with EOS, major factors affecting quality of life included

emotional "rollercoaster rides" and feelings of self-blame and depression, lack of resources and information from healthcare professionals, lack of community among other patients, financial grief, and exhaustion from travel to and from healthcare centers.[1] Established support systems among family and community have also been associated with improvements in HRQoL in a 7-11 year old chronic illness population, especially in children with low physical self-esteem.[18] Parents of children with cerebral palsy have noted that initial isolation improves over time with repeated clinic visits.[19] The impact of COVID-19 on these factors is unclear, as feelings of lack of community and resources may be increased by the pandemic but travel exhaustion may have actually improved due to rollout of telehealth during the pandemic. It is possible that early pressure to establish greater support systems well before the pandemic, as expressed by parents of children with EOS, may have had some protective effect during the isolation of the COVID-19 pandemic.

Because inclusion criteria for this study necessitated EOSQ-24 administration during the pandemic, patients who continued to attend either telehealth or in-person appointments would be more likely to be included. This is a limitation of the study, as it could bias the population toward better-resourced patients or caregivers who maintained greater involvement in their child's healthcare, and this population may be more likely to report higher HRQoL scores. In terms of the statistical analysis and data availability, not every patient took an EOSQ at every timepoint, so it was not feasible to track the HRQoL scores for a single patient across all timepoints. For comparisons between timepoints, paired t-tests were used, which excluded patients who took only one of the two surveys being compared. A third consideration for interpretation of the results of this study is the nature of the EOSQ-24 survey, which is filled out by the parent or caregiver of each patient. This means that, especially for younger patients, parent perceptions of the child's quality of life are measured in place of the patient's own self report. For instance, greater parent health-related anxiety could potentially correspond to decreased emotion scores even without increased anxiety in the child with EOS. The level of direct involvement in survey response that each patient has often depends on their age, as older patients are more likely to answer questions themselves. Furthermore, it is possible that the EOSQ-24 as an instrument was unable to detect differences in domains specifically affected by the COVID-19 pandemic, whether through gaps in the areas assessed by the questionnaire or through inadequate resolution of the 5-point scoring system for each survey question. Finally, in order to reduce confounding of HRQoL by new interventions, patients with new interventions (e.g., index spinal instrumentation) during March 2019 to February 2021 were excluded. As a result, further studies are warranted to investigate the effect of the pandemic on HRQoL for patients who had less stable treatment courses. Despite the limitations, there are several notable strengths of this study. We included over 600 patients using a multicenter registry, evaluating data from a span of 3 years to capture temporal changes in HRQoL of individual patients. This allowed us to stratify by scoliosis etiology, with still over 100 patients in the smallest etiology subgroup. The EOSQ-24 questionnaire also enabled the analysis of twelve distinct aspects of HRQoL.

In sum, the results of this study suggest some element of resilience in the EOS population, which may have buffered the impact of the COVID-19 pandemic on these patients. Additional explorations of the individual roles of increased telehealth availability, remote schooling, health-related media, and well-established patient support networks are warranted. On the other hand, it is possible that changes were not detected with the current instruments utilized for evaluating HRQoL. Thus, the study also highlights the need for a comprehensive investigation of EOS patient and caregiver experiences relating to COVID-19 to identify aspects of HRQoL that may have in fact changed during this time. These could then be incorporated into future evaluations of HRQoL in this population. These inquiries may provide a better framework for assessing and promoting the HRQoL of children with chronic health conditions and their families, perhaps in preparation for future widespread interruptions in healthcare. Finally,

while few changes in HRQoL during the pandemic were found overall, we did find a significant decrease in scores among patients with congenital scoliosis, as well as a decrease in mean score without statistical significance in the neuromuscular cohort, throughout the first year of the pandemic. These findings suggest that vulnerability to decreased HRQoL varied between etiologies, and additional support may be warranted for neuromuscular and congenital EOS patients.

Additional Links

- The Pediatric Spine Foundation: The Pediatric Spine Foundation is a community dedicated to improving the quality of care and the outcome of treatment for patients and families dealing with chest wall and spine deformities.
- The Final 24-Item Early Onset Scoliosis Questionnaires (EOSQ-24): Validity, Reliability and Responsiveness, Matsumoto et al. 2018, *Journal of Pediatric Orthopaedics*.
- POSNA COVID-19 Updates Blog: Sharing experience, perspectives and information about the current crisis brought on by the COVID pandemic.

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References

1. Lauder B, Sinclair PM, Maguire J. Mothers' experience of caring for a child with early onset scoliosis: a qualitative descriptive study. *J Clin Nurs*. 2018;27(7-8):e1549-e1560.
2. Campbell M, Matsumoto H, St. Hilaire T, et al. Burden of care in families of patients with early onset scoliosis. *J Pediatr Orthop B*. 2020;29(6):567-571. https://journals.lww.com/jpo-b/Fulltext/2020/11000/Burden_of_care_in_families_of_patients_with_early.8.aspx.
3. Matsumoto H, Williams B, Park HY, et al. The final 24-item early onset scoliosis questionnaires (EOSQ-24): validity, reliability and responsiveness. *J Pediatr Orthop*. 2018;38(3):144-151.
4. Ping W, Zheng J, Niu X, et al. Evaluation of health-related quality of life using EQ-5D in China during the COVID-19 pandemic. *PLoS One*. 2020;15(6):e0234850.
5. Nobari H, Fashi M, Eskandari A, et al. Effect of COVID-19 on health-related quality of life in adolescents and children: a systematic review. *Int J Environ Res Public Health*. 2021;18(9):4563.
6. O'Dwyer MC, Meixner K, Albiac LC, et al. Health-related quality of life for people with acute and chronic illnesses during the COVID-19 pandemic. *J Am Board Fam Med*. 2021;34(3):509-521.
7. Adıbelli D, Sümen A. The effect of the coronavirus (COVID-19) pandemic on health-related quality of life in children. *Child Youth Serv Rev*. 2020;119:105595.
8. Matsumoto H, Skaggs DL, Akbarnia BA, et al. Comparing health-related quality of life and burden of care between early-onset scoliosis patients treated with magnetically controlled growing rods and traditional growing rods: a multicenter study. *Spine Deform*. 2021;9(1):239-245.
9. Matsumoto H, Marciano G, Redding G, et al. Association between health-related quality of life outcomes and pulmonary function testing. *Spine Deform*. 2021;9(1):99-104.
10. Matsumoto H, Auran E, Fields MW, et al. Serial casting for early onset scoliosis and its effects on health-related quality of life during and after discontinuation of treatment. *Spine Deform*. 2020;8(6):1361-1367.
11. Roye BD, Simhon ME, Matsumoto H, et al. Bigger is better: larger thoracic height is associated with increased health related quality of life at skeletal maturity. *Spine Deform*. 2020;8(4):771-779.
12. Roye BD, Fano AN, Matsumoto H, et al. The impact of unplanned return to the operating room on health-related quality of life at the end of growth-friendly surgical treatment for early-onset scoliosis. *J Pediatr Orthop*. 2022;42(1):17-22.
13. Williams BA, Matsumoto H, McCalla DJ, et al. Development and initial validation of the Classification of Early-Onset Scoliosis (C-EOS). *JBJS*. 2014;96(16):1359-1367.
14. Hell AK, Braunschweig L, Behrend J, et al. Health-related quality of life in early-onset-scoliosis patients treated with growth-friendly implants is influenced by etiology, complication rate and ambulatory ability. *BMC Musculoskelet Disord*. 2019;20(1):588.
15. Ravens-Sieberer U, Kaman A, Otto C, et al. Mental health and quality of life in children and adolescents during the COVID-19 pandemic-results of the copsy study. *Dtsch Arztebl Int*. 2020;117(48):828-829.
16. Riiser K, Helseth S, Haraldstad K, et al. Adolescents' health literacy, health protective measures, and health-related quality of life during the Covid-19 pandemic. *PLoS One*. 2020;15(8):e0238161.
17. Ramo B, McClung A, Jo CH, et al. Validation of the early onset scoliosis questionnaire (EOSQ) as applied to the classification of early onset scoliosis (C-EOS) etiology designation before scoliosis treatment. *Spine Deform*. 2019;7:1013.
18. Chernoff RG, Ireys HT, DeVet KA, et al. A Randomized, controlled trial of a community-based support program for families of children with chronic illness: pediatric outcomes. *Arch Pediatr Adolesc Med*. 2002;156(6):533-539.
19. Coffey JS. Parenting a child with chronic illness: a metasynthesis. *Pediatr Nurs*. 2006;32(1):51-9.