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COVID-19 Pandemic Associated With Increased Self-reported Depressive Symptoms in Patients With Congenital Craniofacial Diagnoses

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Abstract

Objective: The current study investigated the influence of the coronavirus (COVID-19) pandemic on patients with congenital craniofacial diagnoses.

Methods: Patients (n = 66) with craniofacial diagnoses aged between 8 and 17 were prospectively evaluated with longitudinal psychosocial assessments using the anger, anxiety, depressive symptoms, and peer relationships instruments within the pediatric Patient-Reported Outcomes Measurement Information System (PROMIS). The COVID-19 cohort (n = 33) included patients with assessments within 2 years prior to the pandemic (t0) and during the pandemic (t1; March 2020 to March 2021). An age-matched comparison cohort (n = 33) with similar demographics and diagnoses included patients assessed twice over 3 years prior to the pandemic.

Results: All PROMIS measures were in the average range clinically for both groups across time points. However, the COVID-19 group reported a significant increase in depressive symptoms during the pandemic (t1) compared to pre-pandemic (t0) scores (48.2 ± 10.1 vs 44.3 ± 9.4, $P = .04$, $d = -0.37$), while the comparison group did not demonstrate any differences in psychosocial functioning between t0 and t1. For the COVID-19 cohort, only the pandemic timeframe ($r = 0.21$, $P = .03$) was significantly associated with increased depressive symptom scores, and no other sociodemographic or medical variables were associated with depressive symptoms.

Conclusions: Self-reported depressive symptoms increased during the COVID-19 pandemic in patients with congenital craniofacial diagnoses. Longitudinal studies are needed to elucidate whether such changes will be persistent or compound known variables associated with psychosocial functioning.

Keywords
COVID-19, craniofacial diagnoses, depressive symptoms

Introduction

Patients born with craniofacial diagnoses may face challenges including functional impairments, the need for extensive multidisciplinary medical care, possible social stressors, and impacts on self-image.[1–4] Such challenges can compound the effects of typical childhood stressors resulting in heightened vulnerability in the psychosocial well-being of patients with craniofacial diagnoses.[5–7] Previous studies have demonstrated varying differences between patients with craniofacial diagnoses and the general population, with some studies reporting greater behavioral problems and depressive symptoms whereas other studies found no differences.[1,2,4,6,8] Our group previously reported that the 8 to 10-year-old age range, a period of childhood that has been reported to correspond to increased bullying, is an at-risk period for immediate and long-term psychosocial distress in children with craniofacial diagnoses.[9–12] While the onset of the coronavirus (COVID-19) pandemic and its impact on the delivery of craniofacial care is starting

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to be studied,[13–16] the psychosocial consequences of social and medical changes during the pandemic for patients born with craniofacial differences are unknown.

Previous outbreaks, such as SARS, H1N1, and AIDS, have been associated with adverse childhood experiences, leading to acute stress disorders, post-traumatic stress, anxiety disorders, and depression.[17–21] Analogous effects are postulated to occur during the COVID-19 pandemic, as children have suddenly lost access to in-person social activities and have likely felt the stress of the pandemic on their parents or other family members.[22] In the context of patients with chronic medical conditions, an additional parameter exacerbated by the pandemic is the lack of access to medical care.[23,24] These changes as a result of the pandemic may have both positive and negative effects on patients with craniofacial diagnoses. Changes in typical social interactions during the pandemic may increase isolation, yet decrease negative peer interactions. Disruptions in medical care may increase anxiety due to the lack of progress in planned treatment, yet also provide a break from the numerous therapies and surgeries involved in the care pathway.

The craniofacial literature on COVID-19 has primarily focused on medical decision-making guidelines and virtual delivery of care with a paucity of studies examining the impact of the pandemic on psychosocial well-being.[13–16] In this study, we evaluated the effects of the COVID-19 pandemic by comparing the differences in psychosocial functioning before and during the pandemic in a single cohort, as well with a case–control using age-matched patients assessed prior to the pandemic across a similar timeframe.

Methods

Patients

Sixty-six patients with congenital craniofacial diagnoses completed each of the 4 pediatric Patient-Reported Outcomes Measurement Information System (PROMIS) measures from the multidisciplinary University of California, Los Angeles (UCLA) Craniofacial Clinic (Institutional Review Board protocol no. 15-000979). Thirty-three patients were included as part of the COVID-19 cohort, as defined by patients who had the battery of PROMIS instruments administered during the pandemic (March 2020 to March 2021; t1) as well as baseline scores within 2 years prior to the pandemic (March 2018 to February 2020; t0). The comparison group consisted of 33 age-matched patients with congenital craniofacial diagnoses who had the battery of PROMIS instruments administered twice over a similar timeframe before the pandemic (September 2015 to December 2019). The comparison group was matched to patients in the COVID-19 cohort by age and diagnoses; however, if diagnostic matches were unavailable, then patients were included based on similar age of assessment at both time points. The baseline score was designated (t0) and the subsequent score was designated (t1). Additional study variables including age at assessment, sex, health insurance type, ethnicity, parental English proficiency, type of diagnoses, presence of multiple diagnoses, and number of surgeries were collected.

Instruments

Four psychosocial pediatric PROMIS instruments (Short Form version 2.0 Depressive Symptoms 8a, Peer Relationships 8a, Anxiety 8a, and Anger 5a) were prospectively administered to all patients on an annual basis as part of our longitudinal, prospective study on psychosocial outcomes in patients with craniofacial diagnoses.[25–31] PROMIS measures were developed as a multi-institutional collaborative effort to measure health via validated, quantitative patient-reported outcomes measures. Initial calibration and validation of the pediatric anger, anxiety, peer relationships, and depressive symptoms assessments were performed on 759 to 3048 participants in public schools, hospital-based outpatient clinics, and subspecialty pediatric clinics.[28–30] PROMIS measures are widely used in pediatric care, including with craniofacial populations.[8,9,24] Questions are rated on a 5-point Likert scale and based on a 7-day recall period. Raw scores for each assessment were converted to standardized T-scores based on national norms with a mean score of 50 and a SD of 10. Higher scores on the anger, anxiety, and depressive symptoms measures represent more distress, while a higher score on the peer relationships measure indicates higher social functioning. Trained research associates administered the PROMIS instruments using paper and pencil during in-person clinic visits pre-pandemic and verbally through virtual clinic meetings and phone calls during the pandemic.

Statistical Analysis

Descriptive statistics were conducted to summarize patient characteristics. Two-tailed paired samples *t*-tests were then used to compare PROMIS scores over time between t0 and t1 in each cohort. PROMIS scores were also compared between groups cross-sectionally at both t0 and t1. Effect sizes were calculated using Cohen's *d*. To determine whether a relationship between COVID-19 and psychosocial scores exists, Pearson correlations were performed using all PROMIS scores assessed at t0 and t1 in the COVID-19 cohort. Variables analyzed included PROMIS scores and time of assessment (during vs before COVID-19). Other sociodemographic and clinical variables analyzed included age, primary diagnosis, presence of multiple diagnoses, health insurance type, sex, ethnicity, parental limited English proficiency, and number of surgeries between t0 and t1.[10] All data were analyzed using IBM SPSS Version 26 (IBM Corp.) with an alpha level of $P < .05$.

Results

Patient Characteristics

Participants aged 8 to 17 years in the COVID-19 cohort (mean age 13.2 ± 2.5 years) and comparison cohort (mean age 13.3 ±

2.6 years) demonstrated similar ages at baseline, as well as similar distribution of sex, insurance type, and presence of multiple diagnoses. Among the COVID-19 cohort, 48.5% of patients were diagnosed with cleft lip and/or palate compared to 66.7% of the comparison cohort. The second most common diagnosis was craniofacial microsomia, which composed 30.3% of the COVID-19 cohort and 24.2% of the comparison cohort. Other craniofacial diagnoses included mandibular micrognathia, congenital vascular malformation of the face, velopharyngeal incompetence, cherubism, and malocclusion. In the COVID-19 cohort, 3 patients were additionally diagnosed with syndromes, including Treacher Collins syndrome, cleft palate with Nager syndrome, and cleft palate with Turner syndrome. In the comparison cohort, 2 patients were diagnosed with cleft palate and Stickler's syndrome, 1 patient had cleft palate and cerebrocostomandibular syndrome, and 1 patient had craniofacial microsomia and Goldenhar syndrome. None of the sociodemographic or medical variables were significantly different between the 2 groups (Table 1).

Table 1. Patient Characteristics of the Coronavirus (COVID-19) Cohort and the Age-Matched Comparison Cohort.

	COVID-19 cohort	Comparison cohort	*P value*
n	33	33	
Age, mean ± SD (years)			
t0	13.2 ± 2.5	13.3 ± 2.6	NS
t1	14.6 ± 2.6	14.5 ± 2.5	NS
Female, n%	18 (54.5)	17 (51.5)	NS
Diagnosis, n%			NS
Cleft lip and/or palate	16 (48.5)	22 (66.7)	
Craniofacial microsomia or Goldenhar syndrome	10 (30.3)	8 (24.2)	
Other	7 (21.2)	3 (9.1)	
Presence of other diagnoses, n%			NS
One diagnosis	21 (63.6)	17 (51.5)	
Multiple diagnoses	12 (36.4)	16 (48.5)	
Insurance, n%			NS
Public aid	25 (75.8)	24 (72.7)	
Private	8 (24.2)	9 (27.3)	
Ethnicity, n%			NS
Hispanic	25 (75.8)	19 (57.6)	
Not Hispanic	8 (24.2)	14 (42.4)	
Parental English proficiency, n%			NS
Limited English proficiency	15 (45.5)	11 (33.3)	
English proficient	18 (54.5)	22 (66.7)	
Number of surgeries between t0 and t1, mean ± SD	0.19 ± 0.48	0.42 ± 1.09	NS

Comparison of Pre-Pandemic and Pandemic Psychosocial Functioning

PROMIS scores at t0 and t1 were all in the average range clinically when compared to national norms. To understand changes in psychosocial functioning over time, PROMIS scores between t0 and t1 in the COVID-19 cohort were compared. No significant differences were found among anger, anxiety, and peer relationships scores (Table 2), albeit anxiety scores trended down during the pandemic. However, depressive symptoms were significantly increased by nearly half a SD (44.3 ± 9.4 vs 48.2 ± 10.1, $P = .04$, $d = -0.37$). The proportion of patients who were 1 SD above the national mean (ie, >60) for depressive symptoms was 12.1% at both t0 and t1.

Comparison of Changes in Psychosocial Functioning of Patients Affected by the Pandemic Versus an Age-Matched Cohort of Patients Prior to the Pandemic

As changes in psychosocial PROMIS scores may be age-related differences, we compared the changes found in the COVID-19 cohort to an age-matched comparison group of patients. As with the COVID-19 cohort, all mean scores for the comparison cohort at both time points were in the average range. The change in scores of the comparison group over time from t0 to t1 were compared to determine typical age-related changes in psychosocial outcomes for patients with craniofacial diagnoses (Table 2). Unlike the COVID-19 cohort, no significant differences in any of the scores were found between the 2 time points in the comparison cohort. In particular, there was only a fractional change in depressive symptom scores (46.4 ± 10.9 vs 46.2 ± 10.0), suggesting that the change in depressive symptoms observed in the COVID-19 cohort may be associated with their experiences during the pandemic. In addition, t0 and t1 PROMIS scores for depressive symptoms, peer relationships, anxiety, and anger were compared between the two cohorts and found to demonstrate no statistically significant differences.

Correlations With Increased Depressive Symptoms in Children With Craniofacial Diagnoses During the COVID-19 Pandemic

Sociodemographic and clinical factors were evaluated within the COVID-19 cohort using Pearson correlations to determine whether any other variables may be potentially associated with increased depressive symptoms scores (Table 3). Both t0 and t1 depressive symptom scores for the COVID-19 cohort were correlated with the other PROMIS measures (anger, anxiety, and peer relationships), timing of PROMIS assessment before or during COVID-19, age, cleft lip and/or palate diagnosis (vs all other diagnoses), presence of multiple diagnoses, private insurance (vs public insurance), sex, ethnicity, parental limited English proficiency, and number of surgeries between

Table 2. Psychosocial Patient-Reported Outcomes Measurement Information System (PROMIS) Scores of the Coronavirus (COVID-19) Cohort and the Comparison Cohort Over Time.

PROMIS measure	t0	t1	Cohen's *d*	*P value*
COVID-19 cohort				
Depressive symptoms	44.3 ± 9.4	48.2 ± 10.1	−0.37	.04
Peer relationships	49.5 ± 11.0	48.3 ± 10.4	0.10	NS
Anxiety	48.0 ± 9.7	45.8 ± 8.8	0.24	NS
Anger	45.1 ± 11.0	45.0 ± 10.3	0.01	NS
Comparison cohort				
Depressive symptoms	46.4 ± 10.9	46.2 ± 10.0	0.02	NS
Peer relationships	47.8 ± 8.8	46.6 ± 7.2	0.16	NS
Anxiety	48.6 ± 9.3	46.7 ± 8.3	−0.05	NS
Anger	45.1 ± 8.4	45.6 ± 8.9	0.20	NS

Table 3. Pearson Correlations for Increased Depressive Symptoms Patient-Reported Outcomes Measurement Information System (PROMIS) Scores Within the Coronavirus (COVID-19) Cohort.

	Depressive symptoms	
Variables	*r*	*P*
Anger PROMIS score	0.61	<.001
Anxiety PROMIS score	0.61	<.001
Peer relationships PROMIS score	−0.60	<.001
PROMIS assessment during COVID-19[a]	0.21	.03
Cleft lip and/or palate (vs all other diagnoses)[a]	0.00	NS
Multiple diagnoses[a]	−0.10	NS
Private insurance[a]	−0.15	NS
Female[a]	0.15	NS
Age at PROMIS assessment	−0.08	NS
Hispanic[a]	−0.18	NS
Parental limited English proficiency[a]	0.12	NS
Number of surgeries between t0 and t1	−0.10	NS

Note: [a]Dichotomous variable.

t0 and t1. The other 3 PROMIS measures demonstrated significant moderate to strong correlations to depressive symptoms scores, either positively (anger and anxiety) or negatively (peer relationships). Among all other variables, only timing of the assessment during the COVID-19 pandemic versus pre-pandemic ($r = 0.21$, $P = .03$) was significantly associated with increased depressive symptoms.

Discussion

In this study, we evaluated the effects of the COVID-19 pandemic on the self-reported psychosocial well-being of patients with congenital craniofacial diagnoses. As part of our long-term effort prospectively evaluating psychosocial outcomes using a battery of PROMIS instruments, we compared the scores of 33 patients who were assessed during COVID-19 with their scores within 2 years prior to the pandemic and found that self-reported depressive symptoms scores were higher during COVID-19. While the depressive symptom scores significantly increased, it is important to note that scores remained in the average range clinically and do not reflect levels of clinical distress. The anxiety, anger, and peer relationship scores were also all within the average range across time points and cohorts. To understand whether the increase in depressive symptom scores was due to age-related changes independent of the pandemic, scores were compared with an age-matched cohort of 33 patients with assessments across a similar timeframe of about 3 years. Unlike the COVID-19 cohort, no significant differences in PROMIS scores for any of the measures were seen over the timeframe analyzed. Similarly, only the pandemic timeframe was significantly associated with increased depressive symptom scores in the COVID-19 cohort when Pearson correlations were conducted for sociodemographic and medical variables.

One of the known effects of the COVID-19 pandemic on the child and adolescent population is the increase in depressive symptoms and anxiety.[32] Longitudinal studies on pediatric mental health have reported significant increases in depressive symptoms and anxiety during COVID-19 in the context of government restrictions and the transition to online learning.[33–35] A recent meta-analysis pooling data from 29 global studies with over 80,000 patients estimated that the prevalence of depressive symptoms and anxiety was 25% and 21%, respectively, during the first year of the pandemic.[31] In contrast to reports of increased pediatric anxiety and depressive disorders, the current study showed only increases in depressive symptoms during COVID-19 for patients with congenital craniofacial diagnoses. There are several interpretations as well as limitations of this finding. First, this longitudinal elevation of depressive symptoms during the pandemic corroborates previous longitudinal studies conducted in the general population, suggesting the influence of shared experiences between patients with and without craniofacial diagnoses. In particular, studies have documented a marked increase in loneliness, which is associated with increased depressive symptoms, potentially stemming from social and physical distancing

measures.[36,37] The transition to virtual learning and limited social interactions during the pandemic may have also contributed to the worsening of depressive symptoms.[34,38] However, it is also possible that the experiences of children with craniofacial diagnoses may have been impacted by COVID-19 in a different way. For example, the lack of typical school interactions could also contribute to the lack of increased anxiety over negative peer interactions or bullying. In addition, patients assessed during COVID-19 demonstrated a trend toward fewer surgical interventions during a similar period of time than the comparison group which could have contributed to lowering anxiety.[39]

The current study is limited by the number of patients included as our criteria required not only that we have assessed the patients during a period when patient care was generally limited, but that we also had relatively recent assessments as baseline comparisons. Hence, results may be secondary to the limited sample size. In addition, other variables may have played a role in shaping psychosocial outcomes, but the limited sample size precluded our ability to account for other factors. Unlike pre-pandemic assessments that were administered using pencil and paper, assessments during the pandemic were performed verbally either via phone or virtual meetings. While we cannot conclude what difference, if any, could exist in the responses between the different modalities, other investigators who have compared telephone interviews versus self-survey via hardcopy in the clinic found a high correlation between the survey methodologies.[40] In addition, this study did not include a cohort without craniofacial diagnoses as our ongoing longitudinal work encompasses only patients with craniofacial diagnoses. A cross-sectional study conducted by our research group before the pandemic found no differences between the PROMIS scores of patients without craniofacial diagnoses and those with craniofacial diagnoses.[41]

The association of increased depressive symptoms with the COVID-19 pandemic in patients with craniofacial diagnoses suggests that team providers should complete psychosocial screening and facilitate access to intervention for at-risk patients. As the impact of the pandemic continues to shift over time, ongoing reassessments can help determine whether the increased depressive symptoms identified in this study are transient or long-term. Further research may clarify whether risk factors related to successive surges in the pandemic, such as multiple instances of activity and school restrictions, treatment disruption, family financial stressors, COVID-19 illness, and death in families, result in additive effects on psychosocial functioning.

Conclusion

While in the average range clinically, self-reported depressive symptoms increased in patients with congenital craniofacial diagnoses during the first year of the COVID-19 pandemic from March 2020 to March 2021. Ongoing psychosocial surveillance with access to mental health resources is recommended during and in the aftermath of the pandemic.

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Declaration of Conflicting Interests

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