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## Long-Term Effect of Multiple Surgeries on Psychosocial Function in Teenage Cleft Lip and Palate Patients

Hi'ilani M. K. Potemra, BS<sup>1</sup>, Johnny Lin, PhD<sup>2</sup>, Anthony A. Bertrand, MD, MBA<sup>1</sup>, Fransia S. De Leon, BA<sup>1</sup>, Jake A. Alford, MD<sup>1</sup>, Allison C. Hu, BA<sup>1</sup>, Libby Wilson, MD<sup>3</sup>, Justine C. Lee, MD, PhD<sup>1</sup>

<sup>1</sup>Division of Plastic and Reconstructive Surgery, University of California Los Angeles David Geffen School of Medicine, Los Angeles, CA, USA

<sup>2</sup>Institute for Digital Research and Education, Department of Statistics, University of California Los Angeles, CA, USA

<sup>3</sup>Cleft Palate Program, Orthopaedic Institute for Children, Los Angeles, CA, USA

### Abstract

**Background:** Cleft lip and palate (CLP) patients undergo a significant number of interventions during their childhood and adolescence. While the intention of such interventions is to improve psychosocial functioning, there exists a paucity of data on the psychosocial outcomes of the burden of care on cleft children. In this work, the long-term effects of quantity and timing of childhood surgeries on teenagers with CLP were evaluated.

**Methods:** CLP patients (age 14–17 years, n=55) and an age-matched unaffected cohort (n=14) prospectively enrolled from two institutions were administered the anger, anxiety, and depressive symptoms instruments from the Pediatric Patient Reported Outcomes Measurement Information System (PROMIS). Total number of surgeries and surgeries stratified by age groups (0–7, 8–10, 11–13, and 14–17 years) were evaluated in relationship to PROMIS scores. Descriptive statistics, independent t-tests, Pearson correlations, and multiple linear regression analyses were conducted.

**Results:** No differences in overall psychosocial functioning were found between the CLP and comparison groups. Total quantity of childhood surgeries did not correlate to psychosocial functioning of CLP teenagers. However, multiple linear regression analyses demonstrated that increased number of surgeries during 8–10 year-old age range predicted increased anxiety and depressive symptoms in CLP teenagers ( $\beta=0.38$ ,  $p=0.009$  and  $\beta=0.29$ ,  $p=0.03$ ; respectively).

**Conclusions:** It was previously reported by our group that the 8–10 year-old age range is an at-risk period for psychosocial distress in children with craniofacial anomalies. Our current work demonstrates that increased number of surgeries during this time frame may result in long-term consequences in anxiety and depression in CLP patients.

## Introduction

Treatment of patients born with cleft lip and palate (CLP) is focused on improving quality of life via reconstructing appearance, hearing, feeding, speech, dentition, and, ultimately, psychosocial functioning. While addressing mental health is an important component of multidisciplinary CLP care, unlike functional outcomes, quantitative long-term monitoring of psychosocial outcomes using validated, patient-reported outcome instruments has become more common only recently (1–3).

The current literature on psychosocial functioning in children with CLP compared to the general population has produced some contradictory results. While some reports have indicated significant differences in CLP patients compared to their unaffected peers, others have suggested no global differences could be found (1, 4–11). These discrepancies may be resultant of a multitude of factors. Whereas Kapp-Simon reported that adolescents with CLP did not have significant differences in global self-concept compared to their peers, she also reported that CLP patients between 5–9 years of age may have difficulties in self-concept (5, 6). Similarly, our group and others have reported that, overall, no significant differences are found between patients with craniofacial anomalies and comparison groups (4, 7). However, children with craniofacial differences between the ages of 8–10 years reported higher levels of anxiety, depression, and lower peer relationships compared to older children (1, 4). Recently, our group determined that children with craniofacial anomalies from recent immigrant households self-report worse psychosocial functioning compared to other craniofacial patients while such differences (12). The combination of these results suggests that specific subsets of children with craniofacial anomalies are likely to be at higher risk for psychosocial distress compared to others. Thus, one avenue for improvement in craniofacial care is the identification of such at-risk subsets and delineation of solutions which may or may not involve additional interventions.

One of the hallmarks of CLP patients is that they spend their childhood undergoing multiple reconstructive surgeries and non-surgical interventions. In a number of other pediatric conditions, the burden of medical care during childhood has been associated with detrimental psychosocial consequences (13–16). The first definitive evidence for an association between repeated and/or prolonged hospitalizations during childhood and psychological disturbances as teenagers was established in two population-based studies in the United Kingdom using parent or teacher questionnaires (17, 18). Following the advent of validated patient reported outcomes measures, the relationship between number of days of childhood hospitalization and psychosocial distress in adolescence and adulthood was reported in hematopoietic stem cell transplant recipients as well as congenital gastrointestinal malformations (14, 19). With respect to the quantity of surgical procedures, psychological consequences are less defined. Diseth and colleagues reported that both number of childhood hospital admissions and operations contributed to psychosocial distress as adolescents for patients with congenital gastrointestinal malformations (14, 20). Kain and colleagues reported that a subset of elementary school-aged children who underwent various elective ambulatory surgeries demonstrated persistent anxiety at 1 year follow up regardless of type of surgery (21). Two reports found that repetitive spinal lengthening in early onset scoliosis was associated with increased depressive and anxiety symptoms (13, 16). However,

the increasing understanding of an entity similar to post-traumatic stress disorder (PTSD), called pediatric medical traumatic stress (PMTS), suggests that a linkage between quantity of interventions and negative psychophysiological responses is present (22–25).

In this work, the relationships between number of surgeries, timing of surgeries, and long-term psychosocial functioning in teenagers with CLP was evaluated, with the hypothesis that increased number of surgeries may also result in mental health issues in CLP patients.

## Methods

### Patients

300 pediatric patients (ages 8–17) with craniofacial anomalies from the craniofacial clinics at the University of California, Los Angeles (UCLA) and the Cleft Palate Program at the Orthopaedic Institute for Children were consented and prospectively enrolled. Demographic information was collected via medical chart review and included age, sex, type of insurance, caregiver, caregiver English proficiency, diagnosis, and surgical history. A caregiver was defined as the person responsible for the patient's medical decision-making and included mothers, fathers, grandparents, other family members, and non-biological caregivers. To assess potential long-term psychosocial effects of multiple surgeries, patients 14–17 years of age were selected (n=118) and from this population only those with a CLP diagnosis were included in this study (n=55). Data regarding surgical history included number of surgeries and the child's age at each surgery. Cumulative number of surgeries between ages 0–17 was stratified into three age groups: 0–7 years, 8–10 years, 11–13 years, and 14–17 years (1, 4). Those who had a diagnosis other than CLP or had developmental delays that inhibited their ability to comprehend and complete the measures were excluded.

PROMIS measures were also gathered from various community events in Los Angeles County and from the pediatric primary care clinics at UCLA to create a comparison group of children (ages 8–17) with no medical diagnosis (n=50). For the purposes of this study, an age-matched comparison subgroup was selected (ages 14–17, n=14).

### Instruments

Patients completed the Patient-Reported Outcomes Measures Information System (PROMIS) pediatric self-report short forms (Version 2.0 Anger 5a, Anxiety 8a, Depressive Symptoms 8a) during multidisciplinary clinic visits (3, 26–29). Trained research associates administered PROMIS and each respondent completed the questionnaire in a private setting.

The PROMIS pediatric self-report measures constitute a multi-institutional collaborative effort to assess the quality of life across several generic health domains for youth ages 8 to 17. Initial calibration and validation of the pediatric anxiety, depression, peer relationships, and anger assessments were performed on 759 to 3,048 participants (depending upon the measure) in public schools, hospital-based outpatient clinics, and subspecialty pediatric clinics (26, 27, 30). PROMIS questions are rated on a 5-point Likert scale (e.g. never, almost never, sometimes, often, almost always) and are based on a 7-day recall period. The total raw score for each assessment was converted into a standardized transformed score (T-score) set to the United States population mean for a score of 50 and a standard deviation in 10

point increments (26). Higher T-scores represent more of the concept being measured (i.e. more anxiety, better peer relationships, etc.).

### Statistical Analyses

Descriptive statistics were used to evaluate demographic variables including age, sex, type of insurance, diagnosis, parental English proficiency, age at surgery, and total number of surgeries. Reliability of patient self-report PROMIS data was measured using Cronbach's alpha. Independent t-tests were used to compare PROMIS scores of patients with CLP to the comparison group. Pearson correlations were run to evaluate relationships between the different psychosocial outcome variables. Multiple linear regression analyses were conducted to determine the possible effect of sex, type of insurance, age at assessment, diagnostic severity, duration of follow-up, caregiver limited English proficiency, the total number of surgeries patients had stratified by age (number of surgeries between 0–7 years, 8–10 years, 11–13 years, and 14–17 years), and self-reported anger as a baseline psychosocial measure, on anxiety and depression. All data were analyzed using SPSS software Version 25 (Chicago, IL) with an alpha level of  $p < 0.05$ .

This study was approved by UCLA IRB #15–000979.

## Results

### Demographics

Of the 300 patients prospectively enrolled in the study, 55 CLP patients ages 14–17 (mean  $15.2 \pm 1.1$  years) met inclusion criteria (Table 1). The majority of patients had a unilateral cleft lip and palate ( $n=39$ , 70.9%), public insurance ( $n=40$ , 72.7%), and proficient English-speaking parents ( $n=39$ , 72.7%). The average total number of surgeries, including both cleft-related and non-cleft-related surgeries, between ages 0–17 was  $6.0 \pm 2.8$ . When stratified by age group, patients had an average of  $3.6 \pm 1.8$  surgeries between ages 0–7,  $0.9 \pm 0.9$  surgeries between ages 8–10,  $1.2 \pm 1.4$  surgeries between ages 11–13, and  $0.3 \pm 0.6$  between ages 14–17.

The most common surgeries between 0–7 years of age were cleft lip repairs ( $n=55$ ,  $6.0 \pm 5.7$  months old) and palatoplasties ( $n=50$ ,  $13.8 \pm 8.6$  months old) (Table 2). Five patients had cleft lip and alveolus (CLA) diagnosis and did not receive a palatoplasty. Alveolar bone grafts were the most common surgeries between ages 8–10 ( $n=26$ ,  $8.8 \pm 0.6$  years) and ages 11–13 ( $n=17$ ,  $11.3 \pm 0.5$  years). 37.5% of the surgeries between ages 14–17 were alveolar bone regrafts ( $n=6$ ,  $14.8 \pm 0.7$  years).

### Reliability and Pearson Correlations of PROMIS Measures

Both reliability of PROMIS measures and correlations between the measures were evaluated. Internal reliability of the pediatric self-reported anger, anxiety, and depressive symptoms measures had Cronbach's alpha levels of 0.8, 0.8, and 0.9, respectively. Pearson correlations demonstrated strong positive and significant correlation coefficients between scores of the measures: anger and anxiety ( $r=0.550$ ,  $p < 0.001$ ), anger and depression ( $r=0.675$ ,  $p < 0.001$ ), and anxiety and depression ( $r=0.615$ ,  $p < 0.001$ ).

### Psychosocial Functioning of Patients with CLP Compared to Unaffected Peers

To understand whether the CLP cohort had different psychosocial functioning compared to their unaffected peers, patients with CLP were age-matched to a comparison group with no medical diagnoses ( $n=14$ , mean  $15.9\pm 1.2$  years). Independent t-tests revealed no significant differences between overall PROMIS scores of the two groups for each of the measures (Table 3).

### Correlation Between Number of Surgeries and Psychosocial Functioning

To understand whether number of surgeries had any correlation to psychosocial functioning, Pearson correlations were performed between the scores and numbers of surgeries (Table 4). No significant correlations could be found for any of the measures in relationship to total number of surgeries. However, when number of surgeries were separated into age groups, both anxiety and depression demonstrated positive correlations to number of surgeries between 8–10 years of age ( $r=0.381$ ,  $p=0.002$  and  $r=0.248$ ,  $p=0.03$ ; respectively).

### Predictors of Psychosocial Functioning in Teenagers with CLP

To determine the effect of number of surgeries on psychosocial functioning, a multiple linear regression model was built using number of surgeries and other potential positive or negative predictors of psychosocial distress. Gender was included based on previous studies indicating discrepancies in psychosocial functioning between male and female children with CFA (31, 32). Unilateral versus bilateral CLP and CLA were included as a means of evaluating severity within the CLP diagnosis with the assumption that those with bilateral clefts had a more severe diagnosis than those with a unilateral cleft, and children with a CLA diagnosis are less severe than those with CLP. The presence of pharyngoplasty was included as an indicator of speech difficulties, which has been associated with increased psychosocial distress (33). Time between receipt of most recent surgery and PROMIS assessment was included to account for possible postoperative effects on psychosocial functioning. Considering the findings of previous works by our group, age at assessment and caregiver English proficiency were also included as possible predictors for the variance in psychosocial scores (1, 12). Insurance type was included as an indicator for socioeconomic status. As the self-reported anger PROMIS scores demonstrated no correlation with total number of surgeries or number of surgeries by age group yet correlated well to anxiety and depression scores, anger scores were used as an indicator of baseline psychosocial functioning (Table 5).

This model accounted for 54.5% of the variance in self-reported anxiety scores [ $F(14,40)=3.428$ ,  $p=0.001$ ] and 61.6% of the variance in depression scores [ $F(14,40)=4.591$ ,  $p<0.001$ ]. For both anxiety and depression, number of surgeries between ages 8–10 years ( $\beta=0.38$ ,  $p=0.009$  and  $\beta=0.29$ ,  $p=0.03$ , respectively) and self-reported anger ( $\beta=0.49$ ,  $p<0.001$  and  $\beta=0.61$ ,  $p<0.001$ , respectively) were found to be significant predictors.

### Discussion

This work evaluated whether the burden of multiple surgeries resulted in long-term psychosocial disturbances for teenagers with cleft lip and palate. Using the psychosocial

suite of PROMIS measures, no significant relationships were found between the total number of operations and psychosocial functioning among CLP teenagers. However, when number of surgeries was stratified by age groups, an increased number of surgeries between 8–10 years of age predicted higher self-reported anxiety and depression PROMIS scores as teenagers in multiple linear regression analyses. In combination with our previous work demonstrating the 8–10 year age range as an at-risk period for psychosocial distress (1), our current report suggests that increased interventions during this age range may translate to long-term negative psychosocial consequences.

Our report partially contrasts with the findings by Ruff and colleagues (34). In their work, a prospective, multi-institutional analysis of psychosocial outcomes in cleft children between 7.5–18.5 years of age was undertaken. While they found no association between surgery and negative psychosocial functioning, subgroup analysis by age was not performed, thereby potentially masking the impact of age (1, 15, 34). The rationale for stratifying surgeries by age groups in our current work is based on our previous report of age-related differences in psychosocial functioning in craniofacial children (1). In Volpicelli et al, children with craniofacial conditions between 8–10 years of age self-reported higher levels of anxiety, depression, and difficulties in peer relationships compared to their older cohorts, suggesting that this particular age range may be an at-risk period. Our current report now adds the understanding of the long-term consequences of increased surgeries during this at-risk period. Interestingly, increased surgeries at all other age groups demonstrated no association to psychosocial outcomes.

A number of limitations exist in this study. The effect of interventions on negative psychosocial outcomes is likely not limited to surgery or hospitalization alone (14, 20). As CLP patients are also actively undergoing orthodontic, dental, and potentially speech therapies at this age range, future work evaluating the additive effects of non-surgical care may be important, although quantifying such therapies are challenging. The relatively small size of our cohort limited the evaluation of the effects of complications or reoperations on psychosocial functioning. As receiving revision surgeries may potentially exacerbate PMTS symptoms, another avenue for investigation would be to assess the responses of CLP patients to repeat or revision surgeries. Similarly, our sample size may have not provided adequate power for finding significance among some of the non-significant variables. Thus, non-significant variables in this current work should not be interpreted as definitively insignificant. Rather, non-significant variables may require larger cohorts to determine its contribution or lack thereof. Another potential limitation is the use of PROMIS anger scores as a baseline measure during the study. Though the scores of the three measures correlated well, all measures were obtained simultaneously. Thus, anger scores may not represent a true baseline for psychosocial functioning. Future work using averaged scores collected longitudinally may improve on the estimation of the baseline psychosocial functioning. Lastly, while teenagers were specifically chosen to minimize the varied effects of adult stressors, inclusion of the adult population in larger multi-center studies may shed light on the ultimate consequences of multiple interventions.

For cleft and craniofacial diagnoses, the majority of the literature has been focused on specific surgeries with, primarily, reports of postoperative improvement in psychosocial



functioning (35–38). Our current study and work by Ruff and colleagues (34) demonstrated that overall increased number of surgeries had no association to psychosocial functioning in CLP patients. While these conclusions may be considered positively in that CLP patients have not been negatively impacted by more surgery, the negative aspect is that they have not demonstrated any benefit by having more surgery either. These data suggest that a thoughtful evaluation of intervention, both surgical and non-surgical, in a prospective manner should be considered among the field as it remains unclear if the comprehensive, well-intentioned care provided by first world multi-disciplinary cleft and craniofacial teams may actually become a burden for some children with craniofacial anomalies. Immediately, our current work indicates that at least one practice change can be implemented. The potentially negative consequences of having more operations during the at-risk 8–10 year age range in cleft patients suggest that efforts to limit surgeries by combining procedures, either intradisciplinary or interdisciplinary, or modification in timing of surgeries, such as earlier bone grafting, should be considered to the extent possible based on the timing of dental eruption.

## Conclusions

Increased number of surgeries between ages 8–10 predicts an increase in long-term anxiety and depressive symptoms in cleft lip and palate patients. The significant association between multiple surgeries and psychosocial functioning suggests a need to develop strategies for modifying timing or consolidating procedures during that age range.

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**Table 1.**

## Patient Characteristics

	<b>Patients with CLP, Age 14–17 n=55</b>
Age, mean (SD)	15.2 (1.1)
Male, n (%)	29 (52.7)
Diagnosis, n (%)	
Unilateral CLP	35 (63.6)
Bilateral CLP	15 (27.3)
Unilateral Cleft Lip Alveolus	4 (7.3)
Bilateral Cleft Lip Alveolus	1 (1.8)
Total Number of Surgeries, mean (SD)	6.0 (2.8)
Number of Surgeries 0–7 years	3.6 (1.8)
Number of Surgeries 8–10 years	0.9 (0.9)
Number of Surgeries 11–13 years	1.2 (1.4)
Number of Surgeries 14–17 years	0.3 (0.6)
Time Between Surgery and PROMIS, n (%)	
<1 Year	9 (16.4)
1 Year	46 (83.6)
Amount of Time in Years, mean (SD)	3.1 (2.3)
Insurance Status, n (%)	
Public Aid	40 (72.7)
HMO	4 (7.3)
PPO	11 (20.0)
Caregiver, n (%)	
Mother	35 (63.6)
Father	15 (27.3)
Grandparent	4 (7.3)
Alternative Caregiver	1 (1.8)
English Proficient Caregiver, n (%)	39 (70.9)

**Table 2.**

## Surgical History

	Age 0–7	Age 8–10	Age 11–13	Age 14–17
<b># Patients, n (%)</b>	55 (100)	32 (58.2)	32 (58.2)	13 (23.6)
<b>Surgery, n (%)</b>				
Cleft Lip Repair	55 (100)	--	--	--
Cleft Lip Revision	4 (7.3)	3 (5.45)	12 (21.8)	--
Palatoplasty	50 (90.9)	--	--	--
Palatoplasty Revision	6 (10.9)	1 (1.8)	--	--
Intermediate Cleft Tip Rhinoplasty or Revision	16 (29.1)	--	8 (14.5)	--
Septorhinoplasty	--	--	--	3 (5.45)
Alveolar Bone Graft	4 (7.3)	16 (32.7)	17 (30.9)	2 (3.6)
Alveolar Bone Regraft	--	5 (9.1)	6 (10.9)	6 (10.9)
Pharyngoplasty	11 (20.0)	5 (9.1)	6 (10.9)	2 (3.6)
Pharyngoplasty Revision	--	4 (7.2)	--	--
Alveolar Bone Graft + Pharyngoplasty	4 (5.45)	10 (18.2)	--	--
Tympanoplasty or Revision	11 (20)	--	2 (3.6)	--
Myringotomy	14 (25.45)	1 (1.8)	1 (1.8)	--
Lip Adhesion	1 (1.8)	--	--	--
Circumcision	2 (3.6)	--	--	--
Circumcision Revision	1 (1.8)	--	--	--
Closure of Oronasal Fistula	--	3 (5.45)	2 (3.6)	--
Le Fort I	--	--	1 (1.8)	2 (3.6)
Distractor Removal	--	--	--	1 (1.8)
Ophthalmologic Surgery	--	--	2 (3.6)	--
Dental Procedure or Placement of Orthodontic Device Under Anesthesia	8 (14.5)	1 (1.8)	4 (7.2)	--
None	--	23 (41.8)	23 (41.8)	42 (76.4)
<b>Total # of Surgeries</b>	187	49	61	16

**Table 3.**

Differences in Psychosocial Functioning Between Comparison Group and Patients with CLP

<b>PROMIS Measure</b>	<b>Comparison Group, n=14 Mean (SD)</b>	<b>Cleft Lip and Palate, n=55 Mean (SD)</b>	<b>p-value</b>
<b>Anxiety</b>	51.8 (14.2)	47.3 (9.4)	NS
<b>Depression</b>	49.4 (10.5)	47.2 (10.4)	NS
<b>Anger</b>	49.4 (8.2)	46.2 (10.0)	NS

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**Table 4.**

Pearson Correlations Between Number of Surgeries and PROMIS Scores

	Anxiety, r (p value)	Depression, r (p value)	Anger, r (p value)
<b>Total Number of Surgeries</b>	0.0023 (NS)	0.046 (NS)	0.048 (NS)
<b>Total Number of Surgeries by Age Group</b>			
0–7 years	–0.043 (NS)	–0.013 (NS)	–0.048 (NS)
8–10 years	0.381 (0.002)	0.248 (0.03)	0.083 (NS)
11–13 years	–0.161 (NS)	–0.053 (NS)	0.094 (NS)
14–17 years	0.006 (NS)	–0.015 (NS)	0.023 (NS)

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**Table 5.**

Linear Regression with Possible Predictors of Anxiety and Depressive Symptoms

	Anxiety: $R^2=0.545$ ; $F(14,40)=3.428$ ; $p=0.001$ ;			Depression: $R^2=0.616$ ; $F(14,40)=4.591$ ; $p<0.001$ ;		
Predictors	$\beta$	t-test	p-value	$\beta$	t-test	p-value
Gender	-0.08	-0.54	NS	-0.14	-1.02	NS
Diagnostic Severity:						
Bilaterality	-0.09	-0.56	NS	-0.17	-1.13	NS
Cleft Lip Alveolus	-0.21	-1.60	NS	-0.03	-0.22	NS
Pharyngoplasty	-0.04	-0.24	NS	-0.26	-1.91	NS
Age at Assessment	0.10	0.79	NS	0.14	1.17	NS
Parental English Proficiency	0.19	1.38	NS	0.05	0.37	NS
Insurance:						
Public Aid (Reference)	--	--	--	--	--	--
HMO	0.10	0.70	NS	0.05	0.37	NS
PPO	0.18	1.45	NS	0.13	1.11	NS
Self-Reported Anger Scores	0.49	3.81	<0.001	0.61	5.16	<0.001
Time Between Surgery and PROMIS	-0.13	-0.72	NS	0.08	0.52	NS
Number of Surgeries:						
0-7 years	-0.14	-1.08	NS	0.04	0.36	NS
8-10 years	0.38	2.76	0.009	0.29	2.31	0.03
11-13 years	-0.18	-1.07	NS	0.02	0.10	NS
14-17 years	0.12	0.65	NS	-0.004	-0.03	NS