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Resiliency and socioemotional functioning in youth receiving surgery for orofacial anomalies

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Abstract – *Objectives:* Restorative interventions for cleft lip and palate involve annual evaluations, adjunct treatment, and multiple surgeries. The purpose of this study was to investigate the longitudinal impact of cleft surgery on psychosocial functioning among youth with cleft. *Methods:* Data were derived from a 5-year, multicenter, prospective longitudinal study of children with cleft (N = 1196). Children completed psychological inventories for self-concept, anxiety, depression, mastery, and relatedness. Multilevel mixed-effects models were used to analyze the effects of craniofacial surgery for cleft on psychosocial outcomes over time. *Results:* There were 1196 participants at baseline, of whom 258 (27.5%) received a surgical intervention prior to their 1st follow-up visit. Approximately 78% of participants had cleft lip and palate, and 22% had cleft palate only. Surgery receipt was significantly associated with lower relatedness (β = −1.48, 95% CI = −2.91, −0.05) and mastery (β = −1.32, 95% CI = −2.49, −0.15) scores, although overall scores appeared to increase over time. Surgery was not related to anxiety (β = −0.15, 95% CI = −1.08, 0.79), depression (β = 0.18, 95% CI = −0.65, 1.01), and self-concept (β = −0.84, 95% CI = −1.83, 0.15). The treatment–time interaction was not significant. Significant differences in psychosocial functioning were found across sex, race/ethnicity, and age groups. *Conclusions:* Surgery may have negative short-term impacts on psychosocial functioning, although effects may diminish over time. Given the limited postsurgical follow-up period, long-term change in psychological well-being and the moderating effects of surgery may not be fully realized. Further follow-up of children with cleft through adulthood to explore developmental trajectories of psychosocial functioning in more detail is recommended.

Key words: orofacial anomalies; oral medicine; psychosocial aspects of oral health

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Restorative interventions for cleft lip and palate (CLP), the second most common birth defect in the United States, typically involve ongoing evaluations, adjunct treatment, and multiple surgeries[1]. Cleft is considered a chronic condition: treatment for cleft begins in infancy, often extending into adolescence and even young adulthood, with the bulk of revisionary surgeries occurring during school-aged years[2]. These surgical procedures include secondary palatal surgery to improve speech and functional well-being, alveolar bone graft surgery to improve functional well-being, such as tooth and bone development, and lip and nose revisions to improve facial appearance. Such changes are associated with social and emotional well-being. Yet, it is unclear what impact surgery has on psychosocial functioning of youth with cleft over time, particularly in relation to gender, race/ethnicity, age, payer status, and type of defect[3–6].

Despite research establishing the importance of psychological well-being in any comprehensive theoretical health model, questions remain regarding the effect of cleft and cleft treatment on psychosocial functioning in youth[7]. While numerous

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reports have suggested that children and adolescents with chronic conditions such as asthma, diabetes, and cleft have lower self-concept and are at risk for depression, others have found no evidence of higher levels of psychological distress or psychosocial adjustment difficulties[8–10]. Further, having a chronic condition such as CLP can foster greater resilience and coping[11]. One recent study found that young adults with CLP were happier and had higher levels of parental support than age-matched patients with dental-facial deformities other than CLP[12]. However, the extant literature on cleft and psychosocial functioning has been occasionally limited due to small sample sizes, inconsistent age groups, or lack a consistent conceptual framework[13].

Understanding psychological change requires examining not only negative states (depression, anxiety) but positive psychosocial indicators (resilience, sense of coherence, coping) as well, as 'a high rating of negative influence is not necessarily indicative of an absence of positive effect and vice versa'[14]. To our knowledge, no study has specifically explored the impact of treatment for cleft lip and palate on positive and negative psychosocial adjustment over time. The purpose of this study was to analyze the longitudinal effects of surgery for cleft lip and/or palate on resiliency and socioemotional functioning among school-aged youth. Multilevel mixed-effects models were used to analyze psychological data for self-concept, mastery, and resilience (as positive outcomes) and depression and anxiety (as negative outcomes). Primary hypotheses included (i) receipt of surgery does not significantly affect anxiety and depression for youth with cleft; (ii) those who receive cleft surgery will have higher self-concept, mastery, and resilience scores following receipt of surgery; and (iii) total prior surgical procedures received are negatively associated with depression and anxiety. We also analyzed change over time between males and females to explore gender as a possible protective factor in psychosocial functioning related to cleft and surgery for cleft[5].

Methods

Data for analysis were derived from a 5-year, multicenter, prospective longitudinal study of youth with cleft. Youths and their caregivers participating in this study were followed at one of six major cleft treatment centers from the United States: New York University Langone Medical Center, Children's Hospital of Philadelphia, Lancaster Cleft Palate Center, Children's Healthcare of Atlanta, University of Illinois-Chicago, and University of North Carolina-Chapel Hill. The study was conducted from 2009 to 2015. Participants included any child having a cleft lip and palate or cleft palate only between 7.5 and 18.5 years of age who spoke English or Spanish. Children who were unable to read at a 2nd grade level or had a diagnosis of either an incomplete cleft lip without cleft of the alveolus, craniofacial syndrome, or other complex medical conditions were excluded from the study. Participants were assessed at baseline and observed over two or three subsequent follow-up visits. The average length of time observed in the study for participants was 414 days, and the length of time between follow-ups ranged from 6 months to 2 years. During the course of the study, some patients received a surgical intervention and some did not. Details of the study design, including study sample and surgical procedure descriptions, are available in a separate publication[15].

Measures

All participants in the study completed a brief demographic questionnaire including items for age, gender, surgery site, prior history of surgical intervention, academic grade of the participating child, ethnicity, and payer status (e.g., private insurance, Medicaid, etc.). In addition to demographic measures, participants were classified as having either a visible facial difference or a nonvisible functional difference, as well as by type of recommended surgical intervention. Functional surgery recommendations included those needing secondary palatal surgery, alveolar bone graft surgery, or fistula repair. Visible recommendations included lip/nose revision or orthognathic surgery.

Psychological inventories used in analysis included the Beck Youth Inventory for Emotional and Social Impairment (2nd edition)[16] and the Resilience Scales for Children and Adolescents[17]. Resilience is considered an important factor among individuals facing potential adversity, and emotional and social functioning are critical for future development. Three scales from the Beck Youth Inventory were considered in analysis: the Beck Depression Inventory for Youth, the Beck Anxiety Inventory for Youth, and the Beck Self-Concept Inventory for Youth. Resilience Scales used in anal-

ysis included mastery and relatedness. Each instrument is psychometrically validated and includes measures of internal consistency, validity, and test–retest reliability.

The Beck Youth Inventory consists of five separate scales assessing thoughts, emotions, and behaviors associated with psychological and social impairment in children and adolescents.[16] The Beck Depression Inventory is a 20-item assessment used to identify depressive symptoms following depression criteria of the Diagnostic and Statistical Manual of Mental Disorders, 4th Edition (DSM-IV), including negative thoughts about one's self, life, and future, as well as feelings of guilt, sadness, and sleep disturbances. Internal consistency scores were high in both children ($\alpha = 0.86$) and adolescents ($\alpha = 0.92$). The Beck Anxiety Inventory is a 20-item instrument used to assess worry over school performance, the future, and negative reactions of others, as well as fears surrounding loss of control and the physiological symptoms that manifest from anxious behavior. The Beck Self-Concept Inventory is a 20-item instrument designed to measure self-perceptions such as competence and positive self-worth. Test–retest reliability of the Self-Concept inventory was 0.81, with high internal consistency ($\alpha = 0.91$). The Mastery Scale is a 20-item questionnaire consisted of three content areas: optimism (positive attitudes about life), self-efficacy (sense that the individual can master his or her environment), and adaptability (ability to ask for help or problem solve). Internal consistency was high ($\alpha = 0.91$) with sufficient validity testing. Finally, the Relatedness Scale, a 20-item questionnaire, was used to measure trust, support, comfort, and tolerance. The Relatedness Scale had an overall coefficient alpha of .89. Both the Mastery and Relatedness Scales are included in the Resiliency Scales for Children and Adolescents[17].

Data and procedures

Summary scores for psychological indicators were calculated from specific individual item responses. Adjustment for missing data followed the established methods described in Beck and Resiliency protocols. First, participants with missing data on more than two items were not included in analysis. This resulted in missing summary data for 1.8% (Beck Self-Concept), 1.5% (Beck Anxiety), 1.8% (Beck Depression), 1.4% (Resilience Mastery), and 1.7% (Resilience Relatedness) of total observations. Second, at each observational period, participants with fewer than three items missing had missing data imputed using the raw mean score of the specified within-visit sample scores for each instrument.

Youth were classified as being in <12 or 12+ age groups. Race/ethnicity was categorized as white, Hispanic, black, Asian, and other. Payment type for surgical procedures was dichotomized as either private or nonprivate insurance. Extent of cleft (cleft lip or cleft lip/palate) and functional/visible surgery recommendations were determined using clinical records. Any participant with both a visible and functional surgical recommendation was classified as visible.

Receipt of surgery was included as a time-varying predictor. Participants received surgery for facial differences (e.g., facial asymmetry or jaw discrepancies) in between observational periods throughout the duration of the study. Surgical visits and study observations were first temporally ordered in the dataset. Surgical visits were not included as observations in longitudinal analyses, but were used to indicate when a participant transitioned from the nonsurgical group to the surgical group, and were also used when calculating the total time passed from first surgery. Patients in the nonsurgical group only transitioned into the surgical group if they received an actual surgery prior to the final observational period. Those with scheduled surgical visits occurring after their last observational period were not classified as surgical recipients, as no postsurgery data were available.

To explore possible temporal delays in change in psychological outcomes associated with surgery, analysis included lagged predictors and multiple measures of time. First, a predictor for lag from first surgery received was calculated for each observational period indicating the total time in days that had passed since the date of first recorded surgery. Second, a lag of one observational period was used to reflect delayed response to receipt of surgery. For example, if a study participant received surgery between baseline and the first follow-up observation, then surgical status would reflect treatment at the second follow-up. Finally, a variable for the amount of time occurring between observed measurement occasions was included. Each lag was used in separate models in analysis.

Statistical analysis

Baseline descriptive statistics by visit were first calculated for cleft palate/cleft lip and palate status, surgical group (visible or functional), payer type,

gender, race/ethnicity, and age group, as well as means and standard deviations for each psychological outcome and the total number of prior surgeries received. Descriptive statistics were stratified by treatment group (surgery received/not received) for postbaseline visits.

Potential confounders were identified *a priori* as any covariate associated with both the receipt of surgery and psychological outcomes and were included in regression models. Bivariate associations between covariates and psychological indicators at baseline were assessed using t-tests and linear regression. Associations between covariates and surgical receipt at any time were assessed using chi-square tests and logistic regression.

Multilevel mixed-effects models were used to account for the nesting of observations within children and for children within treatment center. Intraclass correlation coefficients (ICCs) were calculated to estimate the proportion of variance explained at the child level, and within-patient unconditional models were used to estimate the average change over time. Primary predictors of interest included receipt of surgery (treatment), time, and their interaction. Adjusted models included additional covariates for gender, race/ethnicity, CPO/CLP status, age group, payer status, and prior surgical history. Random intercepts were included for individual and site-specific indicators. Following primary analyses, results were compared to models using lagged predictors for surgical receipt and observational period, and a stratified analysis comparing the rate of change between males and females was conducted. Sensitivity analyses included incorporating alternative measures of time, randomly varying slopes for receipt of surgery, and the use of generalized estimating equations. Statistical analysis was conducted using Stata v14.0[18] (StataCorp LP, College Station, TX, USA).

Results

There were 1196 participants in the study (Table 1), of whom 77.5% had cleft lip and palate and 22.5% had cleft palate only. At baseline, 191 (16%) participants presented with either a visible or visible/invisible surgical recommendation, 159 (13.3%) participants had an invisible surgical recommendation only, and 798 participants (66.7%) had no surgical recommendation. There were 580 participants (48.5%) under 12 years of age and 616 participants (51.5%) aged 12 years or older. For race/ethnicity, 731 participants were white (61.1%) followed by 189 Hispanic/Latino (15.8%), 126 Asian (10.5%), 99 black (8.3%), and 39 'other' race (3.3%). Finally, there were 671 males (56.1%) and 525 females (43.9%). Mean psychological scores at baseline were within normative levels as described in instrument manuals: self-concept = 45.2, SD = 9.9; anxiety = 12.1, SD = 9.6; depression = 7.9, SD = 8.1; mastery = 59.5, SD = 11.6; relatedness = 74.8, SD = 14.6[16,17].

There were 937 participants presenting at first follow-up. Between baseline and the first follow-up period, 258 participants (27.5%) received a surgical intervention (Table 2). Of the 715 participants presenting at second follow-up, 259 (36.2%) had received a surgical intervention. Only 52 participants returned for a third follow-up observational period, with 46 participants (88.5%) receiving a prior surgical intervention.

Model results (Table 2) show that change in psychosocial functioning over time does not differ across treatment group (nonsignificant interaction for each outcome) and that receipt of surgery for a craniofacial abnormality is associated with lower relatedness ($\beta = -1.5$, 95% CI = −2.9, −0.1) and mastery ($\beta = -1.3$, 95% CI = −2.5, −0.2) scores (Table 3). Surgery was not significantly related to anxiety, depression, and self-concept at the 5% level. While time was not significant in the noninteraction model (Table 3), effects appear to increase over time for self-concept, relatedness, and mastery and decrease for anxiety and depression. Psychological scores did not significantly differ with respect to cleft type, visible/functional defect, or prior surgery history. Compared to females, males had significantly lower anxiety ($\beta = -1.9$, 95% CI = −2.8, −0.9), depression ($\beta = -1.2$, 95% CI = −2.1, −0.4), relatedness ($\beta = -1.8$, 95% CI = −3.3, −0.3), and mastery ($\beta = -1.4$, 95% CI = −2.6, −0.2) scores. Compared to Whites, Hispanic children had significantly higher anxiety ($\beta = 2.5$, 95% CI = 1.0, 4.0) and depression ($\beta = 1.8$, 95% CI = 0.5, 3.1) and significantly lower self-concept ($\beta = -2.8$, 95% CI = −4.4, −1.3), relatedness ($\beta = -5.5$, 95% CI = −7.8, −3.3), and mastery ($\beta = -4.1$, 95% CI = −5.9, −2.4), while blacks had lower relatedness ($\beta = -4.9$, 95% CI = −7.7, −2.1) and Asians had lower self-concept ($\beta = -2.9$, 95% CI = −4.6, −1.2), relatedness ($\beta = -3.0$, 95% CI = −5.4, −0.5), and mastery ($\beta = -2.9$, 95% CI = −4.8, −0.9) (Table 3). Finally, those participants aged 12 years or older had significantly lower anxiety ($\beta = -2.3$, 95% CI = −3.0, −1.5),

Table 1. Demographic characteristics over time by surgical group

	Baseline		First follow-up				Second follow-up				Third follow-up			
	Full sample		Surgery		No surgery		Surgery		No surgery		Surgery		No surgery	
Variables	*N*	%	*N*	%	*N*	%	*N*	%	*N*	%	*N*	%	*N*	%
Overall	1196	100	258	27.5	679	72.5	259	36.2	456	63.8	46	88.1	6	11.5
CPO/CLP														
CLP	927	77.5	235	91.1	518	76.3	237	91.5	343	75.2	43	93.5	6	100
CPO	269	22.5	23	8.9	161	23.7	22	8.5	113	24.8	3	6.5	0	0
Visibility														
Visible + Both	191	16.0	62	24.0	38	5.6	48	18.5	12	2.6	4	8.7	1	16.7
Invisible	159	13.3	58	22.5	44	6.5	55	21.2	21	4.6	4	8.7	0	0
Not Accepted	798	66.7	135	52.3	568	83.7	151	58.3	407	89.3	34	73.9	5	83.3
Missing	48	4.0	3	1.2	29	4.3	5	1.9	16	3.5	4	8.7	0	0
Insurance type														
Private	613	51.3	112	43.4	383	56.4	117	45.2	262	57.5	18	39.1	4	66.7
Nonprivate	531	44.4	128	49.6	267	39.3	127	49.0	161	35.3	25	54.4	2	33.3
Missing	52	4.3	18	7.0	29	4.3	15	5.8	33	7.2	3	6.5	0	0
Gender														
Female	525	43.9	112	43.4	300	44.2	115	44.4	204	44.7	20	43.4	3	50
Male	671	56.1	146	56.6	379	55.8	144	55.6	252	55.3	26	56.5	3	50
Race/ethnicity														
White	731	61.1	128	49.6	423	62.3	128	49.4	295	64.7	19	41.3	3	50
Hispanic/Latino	189	15.8	61	23.6	101	14.9	60	23.2	64	14.0	18	39.1	2	33.3
Black	99	8.3	18	7.0	59	8.7	18	7.0	35	7.7	2	4.4	1	16.7
Asian	126	10.5	36	14.0	72	10.6	40	15.4	44	9.7	4	8.7	0	0
Other	39	3.3	13	5.04	16	2.4	10	3.9	12	2.6	1	2.2	0	0
Age (Categorized)														
<12	580	48.5	122	47.3	312	46.0	128	49.4	203	44.5	11	23.9	2	33.3
12 and older	616	51.5	136	52.7	367	54.0	131	50.6	253	55.5	35	76.1	4	66.7
	X	SD	X	SD	X	SD	X	SD	X	SD	X	SD	X	SD
Beck Scores														
Self-concept	45.2	9.9	43.6*	11.1	45.9*	9.8	43.8*	10.6	46.1*	9.9	44.2	9.9	51	7.4
Anxiety	12.1	9.6	12.6	9.0	11.7	9.7	11.9	8.7	10.8	9.5	11.9	8.6	12.2	1.9
Depression	7.9	8.1	8.6	8.1	7.9	8.6	8.5	8.6	7.3	8.5	8.3	10.8	5.4	3.4
Resilience scores														
Mastery	59.5	11.6	58.0*	12.3	60.2*	11.6	58.1*	12.3	61.2*	12.0	58.9	12.7	65.7	14.4
Relatedness	74.8	14.6	72.6*	15.6	75.6*	14.7	73.4*	15.4	76.8*	14.3	74.7	13.1	77.8	19.8
Surgery history	4.0	3.1	5.7	3.5	4.9	2.8	5.6	3.2	4.9	2.8	5.4	2.6	4.7	2.6

CPO/CLP = Cleft palate only/Cleft lip and palate. Visibility refers to surgical recommendations for visible facial difference or invisible (functional) difference or that the recommendation was not accepted by the patient.
*$P < 0.05$.
Hypothesis tests for surgical group comparisons of psychological outcomes only.

depression (β = −1.2, 95% CI = −1.9, −0.5), and self-concept (β = −1.5, 95% CI = −2.3, −0.6) scores compared to participants younger than 12 years old.

Change over time did not differ by gender (Table 4) for any of the included psychological outcomes, indicating comparable group effects (nonsignificant slope differences) over time. A treatment lag of one observational period was unrelated to psychological outcomes, and the effect of receiving surgery did not change as a function of lag time between observations (results not shown). GEE model results were comparable to those from mixed models.

Discussion

This study examined psychosocial adjustment among school-aged youth with cleft receiving secondary cleft-related surgery, using data from a multicenter prospective cohort study. We hypothe-

Table 2. Change in psychological functioning over time due to receipt of surgery, with treatment*time interaction

Variables	(1) Anxiety	(2) Depression	(3) Self-concept	(4) Relatedness	(5) Mastery
Surgery	−0.278	−0.0713	−0.680	−0.654	−0.233
	(−2.057, 1.501)	(−1.664, 1.521)	(−2.572, 1.213)	(−3.383, 2.074)	(−2.482, 2.016)
Visit	−0.167	−0.0865	0.0563	0.584*	0.457*
	(−0.561, 0.227)	(−0.438, 0.265)	(−0.361, 0.473)	(−0.0178, 1.186)	(−0.0381, 0.952)
Surgery*Visit	0.0901	0.171	−0.110	−0.555	−0.734
	(−0.935, 1.115)	(−0.747, 1.089)	(−1.198, 0.978)	(−2.124, 1.015)	(−2.031, 0.562)
Gender (Male)	−1.880***	−1.225***	−0.543	−1.843**	−1.419**
	(−2.847, −0.913)	(−2.076, −0.374)	(−1.580, 0.495)	(−3.343, −0.342)	(−2.615, −0.224)
Hispanic	2.475***	1.755***	−2.832***	−5.518***	−4.132***
	(0.998, 3.951)	(0.464, 3.046)	(−4.389, −1.276)	(−7.764, −3.272)	(−5.919, −2.346)
Black	1.418	0.144	1.306	−4.916***	−1.784
	(−0.381, 3.216)	(−1.429, 1.717)	(−0.611, 3.223)	(−7.703, −2.130)	(−3.999, 0.431)
Asian	0.394	1.083	−2.886***	−2.983**	−2.881***
	(−1.178, 1.966)	(−0.301, 2.466)	(−4.572, −1.199)	(−5.433, −0.533)	(−4.830, −0.933)
Other	1.387	2.498**	−1.333	−1.467	−1.184
	(−1.258, 4.033)	(0.167, 4.829)	(−4.183, 1.518)	(−5.574, 2.640)	(−4.460, 2.091)
CPO versus CLP	−0.885	−0.848	−0.127	0.492	−0.788
	(−2.166, 0.397)	(−1.970, 0.274)	(−1.490, 1.236)	(−1.481, 2.464)	(−2.359, 0.782)
Age 12+	−2.262***	−1.229***	−1.455***	−0.391	−0.380
	(−3.034, −1.490)	(−1.914, −0.544)	(−2.277, −0.633)	(−1.580, 0.797)	(−1.343, 0.583)
Invisible	−0.0697	0.353	−0.163	−0.404	−0.920
	(−1.411, 1.272)	(−0.846, 1.552)	(−1.598, 1.272)	(−2.470, 1.661)	(−2.608, 0.768)
Not Accepted	−0.697	−0.288	1.451***	0.639	0.760
	(−1.719, 0.324)	(−1.201, 0.625)	(0.358, 2.544)	(−0.931, 2.210)	(−0.526, 2.046)
Nonprivate Insurance	0.354	0.515	−1.056**	−1.595**	−0.984*
	(−0.538, 1.246)	(−0.273, 1.303)	(−2.001, −0.112)	(−2.960, −0.229)	(−2.086, 0.117)
Prior Surgery History	−0.0148	−0.0320	−0.0153	−0.0316	−0.0895
	(−0.192, 0.162)	(−0.186, 0.122)	(−0.200, 0.170)	(−0.300, 0.236)	(−0.302, 0.123)
Constant	14.18***	9.108***	46.65***	78.17***	62.42***
	(12.17, 16.19)	(7.460, 10.76)	(44.74, 48.55)	(75.41, 80.93)	(60.21, 64.62)
Observations	2585	2582	2577	2579	2587
Number of Groups	6	6	6	6	6

95% Confidence Intervals in parentheses. ***$P < 0.01$, **$P < 0.05$, *$P < 0.1$. CPO/CLP=Cleft palate only/Cleft lip and palate. Visibility refers to surgical recommendations for visible facial difference or invisible (functional) difference or that the recommendation was not accepted by the patient. The reference category for facial difference include those with Visible and both visible/invisible differences.

sized that surgery would improve positive psychosocial functioning and show no change in negative outcomes. Findings indicated no significant changes in anxiety, depression, and self-concept scores due to receipt of surgery, time, and the time–treatment interaction, but that relatedness and mastery scores were negatively associated with surgery. We found no evidence of an association between the number of prior surgeries received and depression and anxiety. These findings are intriguing, as other research has found that surgery improves psychological adjustment for individuals with chronic conditions such as obesity and epilepsy and increases the risk of psychological maladjustment in adolescents with other chronic conditions[19–21].

Although surgery is negatively associated with psychosocial adjustment in the short term, questions remain regarding long-term effects and the mediating role of surgery over the life course. A majority of youth in this study either did not receive a surgical intervention or had limited follow-up observations after receipt of surgery, both of which may mask differences in change over time. The lack of change among such psychological parameters within a one to two year period of time may be due to treatment expectations or other developmental issues related to body image. Both average change over time and group-specific change may require longer periods of follow-up or the use of more flexible trajectories (such as fractional polynomials) to more fully understand psychological development.

Overall psychological scores within each treatment group were high for positive outcomes and low for negative outcomes. This finding supports the existing literature indicating that youth treated

Table 3. Change in psychological functioning over time due to receipt of surgery, no interaction

Variables	(1) Anxiety	(2) Depression	(3) Self-concept	(4) Relatedness	(5) Mastery
Surgery	−0.145	0.181	−0.842*	−1.476**	−1.320**
	(−1.075, 0.785)	(−0.650, 1.012)	(−1.832, 0.147)	(−2.905, −0.0466)	(−2.491, −0.149)
Visit	−0.155	−0.0648	0.0423	0.514*	0.364
	(−0.527, 0.216)	(−0.397, 0.267)	(−0.351, 0.436)	(−0.0544, 1.081)	(−0.103, 0.832)
Gender = 1, Male	−1.880***	−1.225***	−0.542	−1.841**	−1.418**
	(−2.848, −0.913)	(−2.076, −0.375)	(−1.580, 0.495)	(−3.342, −0.341)	(−2.613, −0.223)
Hispanic	2.475***	1.757***	−2.833***	−5.521***	−4.136***
	(0.999, 3.952)	(0.466, 3.047)	(−4.390, −1.277)	(−7.767, −3.275)	(−5.922, −2.349)
Black	1.418	0.144	1.306	−4.918***	−1.786
	(−0.380, 3.216)	(−1.429, 1.717)	(−0.612, 3.223)	(−7.704, −2.132)	(−4.002, 0.429)
Asian	0.394	1.082	−2.885***	−2.981**	−2.879***
	(−1.178, 1.966)	(−0.302, 2.466)	(−4.572, −1.199)	(−5.431, −0.532)	(−4.828, −0.931)
Other	1.386	2.495**	−1.331	−1.457	−1.170
	(−1.260, 4.031)	(0.164, 4.826)	(−4.181, 1.520)	(−5.563, 2.649)	(−4.446, 2.105)
CPO versus CLP	−0.884	−0.848	−0.127	0.492	−0.788
	(−2.166, 0.397)	(−1.970, 0.274)	(−1.490, 1.236)	(−1.481, 2.464)	(−2.359, 0.782)
Age 12+	−2.261***	−1.227***	−1.456***	−0.399	−0.389
	(−3.033, −1.489)	(−1.912, −0.542)	(−2.278, −0.634)	(−1.588, 0.790)	(−1.352, 0.574)
Invisible	−0.0683	0.356	−0.164	−0.412	−0.930
	(−1.410, 1.273)	(−0.843, 1.555)	(−1.599, 1.270)	(−2.477, 1.654)	(−2.619, 0.758)
Not Accepted	−0.695	−0.284	1.449***	0.626	0.741
	(−1.717, 0.326)	(−1.197, 0.629)	(0.356, 2.542)	(−0.944, 2.196)	(−0.545, 2.027)
Nonprivate Insurance	0.355	0.517	−1.057**	−1.600**	−0.993*
	(−0.537, 1.247)	(−0.271, 1.305)	(−2.002, −0.112)	(−2.966, −0.235)	(−2.094, 0.109)
Prior Surgery History	−0.0147	−0.0319	−0.0153	−0.0318	−0.0900
	(−0.192, 0.162)	(−0.186, 0.122)	(−0.200, 0.170)	(−0.300, 0.236)	(−0.303, 0.123)
Constant	14.17***	9.088***	46.66***	78.24***	62.50***
	(12.16, 16.18)	(7.443, 10.73)	(44.76, 48.56)	(75.48, 80.99)	(60.31, 64.70)
Observations	2585	2582	2577	2579	2587
Number of Groups	6	6	6	6	6

95% Confidence Intervals in parentheses. ***$P < 0.01$, **$P < 0.05$, *$P < 0.1$. CPO/CLP = Cleft palate only/Cleft lip and palate. Visibility refers to surgical recommendations for visible facial difference or invisible (functional) difference or that the recommendation was not accepted by the patient. The reference category for facial difference include those with Visible and both visible/invisible differences.

by cleft teams appear to have normal psychological functioning[5,10,22,23]. It is possible that psychological status is formalized in the early school-age years and maintains initial trajectories despite short-term changes associated with surgery. As a result, the included measures of psychosocial adjustment may not be sensitive enough to detect changes over time, and further study using other measures of psychological adjustment or analysis of clinically meaningful change is warranted. Research exploring alternative quality-of-life outcomes (e.g., SF-36, Youth Quality of Life: Facial Differences, Cleft Audit Protocol for Speech) may yield further knowledge of the impact of surgery for craniofacial abnormalities. For example, other unpublished findings from this cohort indicate that surgery is associated with positive change in oral health-related quality of life, and research has shown that oral health-related quality of life is lower in youth with cleft who are recommended for surgery than those who have no such treatment needs[3]. Finally, the use of mixed methods that incorporate qualitative data may provide meaningful insight for future study[24].

Although there were significant gender differences in psychological outcomes, with females having higher levels of anxiety, mastery, and relatedness than males, the rate of change in each group was not significant. This finding is in partial contrast to recent research showing gender to be the main influencing factor in psychological adjustment in 16-year-old adolescents with cleft, as well as an important moderator of the effect of chronic illness on depressive symptoms among adolescents[5,25]. Additionally, the observed differences across race/ethnic groups indicate that disparities in psychosocial well-being are plausible. Previous research shows that children with special health needs tend to face more barriers than children without special health needs[26]. Such differences

Table 4. Change in psychological functioning over time, including gender*visit interaction

Variables	(1) Anxiety	(2) Depression	(3) Self-concept	(4) Relatedness	(5) Mastery
Surgery	−0.150	0.194	−0.842*	−1.482**	−1.341**
	(−1.080, 0.781)	(−0.636, 1.025)	(−1.832, 0.147)	(−2.912, −0.0532)	(−2.512, −0.171)
Gender (Male)	−1.990***	−0.881*	−0.546	−2.019**	−1.885***
	(−3.073, −0.908)	(−1.837, 0.0753)	(−1.705, 0.612)	(−3.694, −0.343)	(−3.230, −0.540)
Visit	−0.235	0.184	0.0395	0.385	0.0301
	(−0.746, 0.277)	(−0.274, 0.642)	(−0.502, 0.581)	(−0.397, 1.168)	(−0.612, 0.672)
Gender*Visit	0.144	−0.449	0.00513	0.232	0.607
	(−0.492, 0.779)	(−1.018, 0.120)	(−0.668, 0.678)	(−0.741, 1.204)	(−0.194, 1.408)
Hispanic	2.479***	1.745***	−2.833***	−5.515***	−4.121***
	(1.002, 3.956)	(0.453, 3.036)	(−4.390, −1.276)	(−7.761, −3.269)	(−5.908, −2.334)
Black	1.420	0.136	1.306	−4.916***	−1.779
	(−0.378, 3.218)	(−1.438, 1.709)	(−0.612, 3.223)	(−7.703, −2.130)	(−3.995, 0.437)
Asian	0.399	1.065	−2.885***	−2.973**	−2.859***
	(−1.173, 1.971)	(−0.319, 2.450)	(−4.572, −1.198)	(−5.423, −0.523)	(−4.808, −0.909)
Other	1.393	2.474**	−1.330	−1.444	−1.139
	(−1.252, 4.039)	(0.142, 4.805)	(−4.181, 1.521)	(−5.551, 2.663)	(−4.415, 2.138)
CPO versus CLP	−0.884	−0.851	−0.127	0.494	−0.785
	(−2.165, 0.397)	(−1.973, 0.271)	(−1.490, 1.236)	(−1.479, 2.466)	(−2.357, 0.786)
Age 12+	−2.260***	−1.229***	−1.456***	−0.398	−0.387
	(−3.032, −1.489)	(−1.914, −0.545)	(−2.278, −0.634)	(−1.587, 0.791)	(−1.350, 0.576)
Invisible	−0.0764	0.386	−0.165	−0.425	−0.963
	(−1.418, 1.265)	(−0.812, 1.585)	(−1.600, 1.271)	(−2.491, 1.641)	(−2.651, 0.725)
Not Accepted	−0.703	−0.259	1.449***	0.614	0.714
	(−1.725, 0.319)	(−1.172, 0.653)	(0.355, 2.542)	(−0.956, 2.185)	(−0.571, 2.000)
Nonprivate Insurance	0.355	0.514	−1.057**	−1.600**	−0.987*
	(−0.537, 1.247)	(−0.274, 1.302)	(−2.002, −0.112)	(−2.965, −0.234)	(−2.089, 0.114)
Prior Surgery History	−0.0147	−0.0323	−0.0153	−0.0316	−0.0893
	(−0.192, 0.162)	(−0.186, 0.122)	(−0.200, 0.170)	(−0.300, 0.236)	(−0.302, 0.124)
Constant	14.24***	8.883***	46.66***	78.34***	62.78***
	(12.21, 16.27)	(7.216, 10.55)	(44.74, 48.59)	(75.55, 81.13)	(60.55, 65.01)
Observations	2585	2582	2577	2579	2587
Number of Groups	6	6	6	6	6

95% Confidence Intervals in parentheses. ***$P < 0.01$, **$P < 0.05$, *$P < 0.1$. CPO/CLP=Cleft palate only/Cleft lip and palate. Visibility refers to surgical recommendations for visible facial difference or invisible (functional) difference or that the recommendation was not accepted by the patient. The reference category for facial difference include those with Visible and both visible/invisible differences.

can manifest through disproportionate access to care and have potential public health implications for treating youth with cleft and other chronic conditions[7,27–29].

While these findings contribute to the existing cleft literature, some study limitations should be noted. First, the effects of surgery may be related to child, caregiver, surgeon, and speech pathologist perceptions of facial and speech differences, respectively. Further research should investigate the interrelationships among psychological functioning and patient/professional ratings of extent of difference. Second, approximately one-third of the participants either refused or postponed surgery or received another surgical recommendation prior to completing the study. Similarly, a majority of individuals receiving surgery did not complete the study protocol or present for all follow-up visits, in part associated with the additional interventions throughout the study period. Thus, there are a number of unanswered questions related to surgery for cleft and psychological adjustment over time that cannot yet be fully resolved. As a result, participant attrition may bias results. The use of mixed models allowed analysis to draw from all available data, although future studies incorporating multiple imputation for multilevel data may be useful. Based on active surgery histories among participants, it is clear that additional follow-up into young adulthood is needed to fully measure psychological sequelae[30–32].

In conclusion, surgical intervention for cleft lip and palate was significantly related with mastery and relatedness in a prospective study of children with cleft but appeared to have no significant effect on anxiety, depression, and self-concept. The num-

ber of prior surgeries was also unrelated to psychosocial functioning. Further study of long-term change, as well as effects on other quality-of-life outcomes, may reveal additional impacts of surgery for cleft.

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