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ORIGINAL ARTICLE

Oral clefts and behavioral health of young children

GL Wehby[1], MC Tyler[2], S Lindgren[3], P Romitti[4], J Robbins[5], P Damiano[6]

[1]*Department of Health Management and Policy, College of Public Health, University of Iowa, Iowa City, IA;* [2]*Public Policy Center, University of Iowa, Iowa City, IA;* [3]*Division of Pediatric Psychology, Department of Pediatrics, Carver College of Medicine, UI Children's Hospital, Center for Disabilities and Development, The University of Iowa, Iowa City, IA;* [4]*Department of Epidemiology, University of Iowa, Iowa City, IA;* [5]*Child Health Services Research, Department of Pediatrics, University of Arkansas for Medical Sciences, Arkansas Children's Hospital, Little Rock, AR;* [6]*Public Policy Center, Department of Preventative and Community Dentistry, University of Iowa, Iowa City, IA, USA*

OBJECTIVES: This study examined the behavioral health of young children with oral clefts, and effects of satisfaction with facial appearance, cleft team care, number of cleft-related surgeries, and socioeconomic status (SES).
SUBJECTS AND METHODS: The study included a population-based sample of 104 children aged 2–12 years with isolated oral clefts from the state of Iowa. Behavior was evaluated with the Child Behavior Checklist or the Pediatric Behavior Scale 30, depending on age, compared with normative samples.
RESULTS: Risks of behavioral problems were not significantly different from normative samples except for higher inattention/hyperactivity risks at age 6–12 years. Low satisfaction with facial appearance was associated with behavioral problems in all domains, except aggression. Team-care effects were not associated with behavioral problems. Number of cleft-related surgeries was associated with increased anxiety/depression and somatic symptom risks. Higher SES was associated with reduced inattention/hyperactivity, aggressive/oppositional behavior, and somatic symptoms.
CONCLUSIONS: Most children with oral clefts may have similar behavioral health outcomes to unaffected children, except for increased inattention/hyperactivity risks at older ages. However, low satisfaction with facial appearance, increased exposure to surgeries, and lower SES may significantly increase behavioral problems. Also, the findings emphasize the need to study the representation of behavioral health professionals on cleft teams and access to behavioral health care.
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Oral clefts are among the most common birth defects, occurring in about 1/1000 births with variation by geography and ancestry (Mossey *et al*, 2009). In the United States, more than 7000 babies were born with oral clefts per year between 2004 and 2006 (Parker *et al*, 2010). Oral clefts may impose a large burden on the physical health, psychosocial well-being, and quality of life of affected individuals (Wehby *et al*, 2006; Wehby and Cassell, 2010). Adverse effects begin early in life and can include reduced fetal growth (Wehby *et al*, 2011a), feeding problems, frequent ear infections, speech and hearing difficulties, and increased hospitalizations, healthcare expenditures, and costs (Nackashi *et al*, 2002; Cassell *et al*, 2008; Boulet *et al*, 2009; Weiss *et al*, 2009). A few studies on long-term outcomes suggest increased hospital admission and length of stay (Wehby *et al*, 2011b), mortality and suicide rates (Christensen *et al*, 2004), increased need for mental health services (Christensen and Mortensen, 2002), and increased risks of certain cancers (Bille *et al*, 2005). However, these studies have been limited to the Danish population, and long-term effects in more diverse populations are understudied.

The effects of oral clefts on the behavioral and psychosocial well-being of affected individuals have received considerable research attention (Hunt *et al*, 2005). However, study results have been somewhat inconsistent because of wide variation in measurement methods, use of small convenience and often clinic-based samples, and limited analytical models; thus, the need for further research in this area remains. Some studies have reported increased risk of mental health and psychosocial challenges from infancy throughout adulthood (Kapp-Simon *et al*, 1992; Kapp-Simon and McGuire, 1997; Kapp-Simon and Krueckeberg, 2000; Hunt *et al*, 2006; Brand *et al*, 2009), but other studies have not found elevated risks (Collett *et al*, 2011). Increased risks of behavioral/emotional or adjustment problems have been reported for children and adolescents with oral clefts compared with unaffected individuals (Slifer *et al*, 2006; Hunt *et al*, 2007) with speech and esthetic concerns identified as contributing factors (Richman, 1997; Hunt *et al*, 2005, 2006). Recent studies have reported adverse effects of oral clefts on neuropsychological outcomes among children and adolescents

Correspondence: George L. Wehby, MPH, PhD, Department of Health Management and Policy, College of Public Health, University of Iowa, E205 GH, 200 Hawkins Drive, Iowa City, IA 52242, USA. Tel: 319 384 5133, Fax: 319 384 5125, E-mail: george-wehby@uiowa.edu
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(Conrad *et al*, 2009) and differences in the brain structures of children with oral clefts compared with unaffected individuals (Nopoulos *et al*, 2005, 2007), some of which have been suggested to be related to social functioning. Increased rates of learning disabilities have also been reported in children with oral clefts (Richman *et al*, 1988; Broder *et al*, 1998).

While previous studies have provided valuable insights into potential effects of oral clefts on the behavioral health of affected children, some limitations need to be further addressed to better identify the psychosocial needs and develop interventions to improve the behavioral outcomes of at-risk children. One major shortcoming is the dearth of studies involving children younger than school age (< 6 years). Evaluating the behavioral outcomes of children with oral clefts at young ages is needed for earlier identification of and treatment for behavioral problems and improving the future health and well-being of affected individuals. Furthermore, there is a need to study the behavior of affected children using large and population-based rather than small clinic-based samples that are highly prone to bias to provide more definitive evidence on the relationship between oral clefts and behavior.

Another limitation of the literature is the minimal identification of predictors of behavioral problems specifically for children with oral clefts. To our knowledge, there is no thorough evaluation of how socioeconomic status (SES), number of surgeries, and access to an organized cleft team[1] that can provide and coordinate the various types of needed specialty care are associated with the risk of behavioral problems among children with oral clefts. Identifying predictors of behavioral problems specifically among children with oral clefts is essential for effective screening of at-risk children as they may face specific risk factors, some of which are not as relevant to the general population. Mainly among these are the higher rates of low satisfaction with facial appearance, speech problems, and need for medical and surgical interventions, which are not only relevant for behavior on their own but may modify the effects of other factors such as SES on behavior. For example, even though higher SES may positively affect behavior in the general population (Roza *et al*, 2009), the greater need for medical and surgical interventions and higher rates of dissatisfaction with facial appearance and speech problems among children with oral clefts may intensify the effects of SES on behavior in the cleft population as children in high-SES households are more likely to obtain needed treatments for these problems. In addition, higher SES may directly compensate for some of the cleft-related risk factors for behavior such as satisfaction with facial appearance. On the other hand, children with oral clefts are at greater risk of being born in lower-SES households (Clark *et al*, 2003; Durning *et al*, 2007). Therefore, evaluating the impact of SES on behavioral health specifically for children with oral clefts is critical for understanding the role of social and economic factors in differences in behavioral health among affected children.

Similarly, the effect of number of surgical treatments on the behavioral health of children with oral clefts has not been adequately explored. Children with oral clefts typically undergo several cleft repair surgeries depending on the child's age, cleft type, and severity. These surgeries generally start within the first several months of life as is recommended (ACPA, 1993). Surgical treatments may have both positive and negative effects on the child's behavior, and the net effect is unknown. On the one hand, an increase in the number of surgical treatments can be very stressful to both children and parents and may have adverse effects on the child's emotional and psychological status (Kapp-Simon, 2004), although these effects have not been thoroughly investigated. On the other hand, obtaining needed surgeries may improve longer-term behavioral/emotional well-being through improving satisfaction with facial esthetics and speech performance. Therefore, the direction of the net effect of the number of surgeries on behavior at younger ages cannot be determined a priori and requires empirical evaluation. Assessing the effects of the number of surgical treatments on children's behavioral health is needed to identify and address any potential adverse effects on behavior when planning surgical treatments.

Providing care through organized cleft teams involving multiple specialties and health professionals has become the standard model for treating children with oral clefts (Strauss, 1999). However, there is a paucity of research on effects of team care on the risk of behavioral problems among children with oral clefts (Austin *et al*, 2010; Robbins *et al*, 2010). Such research is needed for evaluating the effectiveness of team care in improving the behavioral outcomes of children with oral clefts. Further, SES, number of cleft surgical treatments, and obtaining team care are likely to be related because of the effects of SES on access to health care and the potential effects of team care on treatment planning and quality. Therefore, it is important to simultaneously evaluate the effects of these factors on the behavioral outcomes of children with oral clefts.

This study addresses these limitations by evaluating the prevalence of being at risk of behavioral problems, using standardized instruments, in a population-based sample of children between 2 and 12 years of age with isolated oral clefts and by assessing the effects of satisfaction with facial appearance, team care, number of surgeries, and SES on the behavioral outcomes.

Methods

Study sample

A statewide population-based sample of living children born in Iowa between January 1, 1990, and December 31, 2000, with isolated oral clefts with no evidence of additional non-cleft structural birth defects, recognized etiology, or evidence of significant intellectual disability was identified by the Iowa Registry for Congenital and

[1]Cleft teams' can have several names (cleft and craniofacial team, cleft palate team, and others) and for purposes of this paper, they will be used synonymously throughout the paper.

Inherited Disorders (IRCID). Cases were reviewed by a clinical geneticist, and in most cases, physical examinations and family histories were obtained. The IRCID conducts active, population-based surveillance of pregnancy outcomes (elective terminations, stillbirths, and live births) diagnosed with a birth defect among Iowa residents. Identified cases were matched to State of Iowa death certificate data to determine vital status and remove deceased individuals. Data were collected via structured telephone interviews with the biological mothers by trained, professional research interviewers in the spring and summer of 2003 when children were between the ages of 2 and 12 years.

Iowa Registry for Congenital and Inherited Disorders employed an extensive search of local, Internet, and commercial databases to find current contact information for the study mothers. Mothers who currently did not have custody of the child were excluded from the study. IRCID mailed study packets to mothers including introductory letters and consent forms, which the mothers were requested to sign and mail back to the study. Once signed informed consent was received, the mother's phone number was released to the interviewers and a 20-min telephone interview was conducted with the mother, which included questions about the type and severity of the child's cleft, location and type of cleft care received thus far, access to general and cleft-related care, current health status, clinical outcomes (e.g. satisfaction with esthetics and speech), and social/family outcomes (e.g. school performance, parenting stress). Further details on sampling and data collection are described elsewhere (Damiano *et al*, 2007). All correspondence, consent forms and study instruments used were approved by the University of Iowa Institutional Review Board.

Iowa Registry for Congenital and Inherited Disorders identified 455 children with isolated oral clefts who were born in Iowa between 1991 and 2000. Of these, 129 were unlocatable and the families of another 81 children could not be contacted by phone. Therefore, the study was able to locate and contact 245 mothers of eligible children. Of these, 181 consented to participate in the study and 64 refused. One hundred and fifty-one mothers actually completed an interview for participation rates of 62% of the locatable families and 83% of the consenting families. Non-response bias tests indicated that non-responders (i.e. consented but did not complete an interview or a written instrument) and non-participants (i.e. did not consent to participate) were similar to participants on relevant maternal and child characteristics such as the age of the mother and the child at the time of the interview, the gestational age of the child at birth (i.e. < 37 *vs* ≥ 37 weeks), and the child's cleft type. Following the telephone interviews, 104 mothers completed and returned the written behavior instruments to screen for child behavioral problems, for response rates of 69% of those participating, 57% of consenting, and 42% of locatable eligible subjects. Only results from the interviews with the 104 mothers who returned the behavior instruments are presented in these analyses.

Measures of behavioral/emotional problems

Standardized, validated written instruments were used to collect behavioral health outcome data. Mothers of the 59 children aged 6–12 completed the parent-report version of the Pediatric Behavior Scale-30 (PBS-30). A briefer version of the original 165-item PBS (Lindgren and Koeppl, 1987), the PBS-30 was developed for focused research and clinical applications and evaluates children's behavior based on 30 items in four broad areas: depression/anxiety (seven items), physical or somatic symptoms (five items), aggression/opposition (nine items), and inattention/hyperactivity (nine items) (McCarthy *et al*, 2002; Conrad *et al*, 2010). Reliability (based on internal consistency) coefficients are 0.80, 0.73, 0.83, and 0.87 for the PBS-30 Depression/Anxiety, Physical Health, Aggression/Opposition, and Hyperactivity/Inattention Scales, respectively. The seven items in the Depression/Anxiety Scale have been adopted to screen for internalizing problems as part of the Vanderbilt attention deficit hyperactivity disorder (ADHD) Parent Rating Scale (VADPRS) and Teacher Rating Scale (VADTRS) (Wolraich *et al*, 2003). The PBS-30 has been used in several studies of behavior of children with health problems including diabetes (McCarthy *et al*, 2002, 2003), preterm birth (Conrad *et al*, 2010), and ADHD (Wolraich *et al*, 2003).

Mothers of the 45 children aged 2–5 years completed the parent-report version of the Child Behavior Checklist for ages 1.5–5 (CBCL 1.5–5) (Achenbach *et al*, 1991; Achenbach and Rescorla, 2000), which assesses behavioral problems in younger children, including problems in the four areas addressed by the PBS-30: anxiety/depression, somatic symptoms, aggression, and attention problems. The CBCL 1.5–5 has good psychometric properties with a test–retest reliability of 0.85, inter-rater reliability of 0.65 (correlation within pairs of mothers and fathers), and higher scores being significantly related to higher risks of child referral to behavioral care (Rescorla, 2005). Further, there are no apparent age- or gender-related biases in the CBCL 1.5–5. The CBCL has been used in several studies of child behavior including children with oral clefts 5 years of age and older (Collett *et al*, 2011).

Both of these instruments have been standardized using normative samples that were selected to be generally representative of the population of children without major behavioral problems. The normative sample for the CBCL 1.5–5 was a multistate sample enrolled in 1999 and included 700 children (51.7% men) with diverse race/ethnicity (56% White; 21% African-American; 13% Hispanic; 10% other) and geographic distributions (40% were from the Midwest) (Rescorla, 2005). The PBS-30 norms were developed in 1991 based on a sample of 600 children (300 men; 300 women) from multiple communities in a single upper Midwestern state (Iowa). The normative sample was selected from urban, suburban, small town, and rural communities and was slightly more diverse (88% White; 2% African-American; 8% Hispanic; 2% other) than the general population in the state. Having a normative sample from the same geographic area and similar backgrounds as the children with oral clefts was ideal for making comparisons between the clinical and normative groups.

As mentioned below, we adjusted for the child's age as a continuous variable in the analysis to account for behavioral changes over age and the possibility that several of the model covariates also change with age. The instruments were self-administered by the mothers. Ninety-seven percent of the study mothers who completed the questionnaire had completed high school. The average number of years of maternal schooling in the sample was 14.7 (s.d. = 1.4) and more than 57% had education of at least 3 years post–high school, suggesting that the study mothers had adequate education to be able to complete these questionnaires on their own.

For each instrument, the raw scores for each domain were converted to standardized scores (T-scores). T-scores could range from 50 to 90, with 70 representing a score in the 98th percentile (top 2%) based on established norms for the instrument.[2] For this study, the clinical cutoff for each domain was defined as a T-score of 63, representing the 90th percentile of the instrument's normative sample.

We used the T-scores of the four behavioral domains described early as outcome measures. Furthermore, we used as alternative outcome measures four binary indicators for having a T-score of 63 or higher on the four behavioral domains. These risk indicators may represent more easily interpretable measures of the child's risk of behavioral problems than the continuous T-scores alone.

Other study measures

Cleft team care was measured by the mother's response to a yes/no question on whether the child is currently receiving care provided by an organized cleft team.[3] Number of cleft-related surgeries was mother's numeric response to a question on the number of cleft surgeries the child had undergone up until the time of the study.[4]

To measure SES, maternal education, total household income, and child's health insurance status/type (which are commonly used SES indicators) were aggregated into an SES index using principal component analysis (PCA) (Greene, 2003) with maximum likelihood estimated polychoric correlations between the index variables (Kolenikov and Angeles, 2004). The assumption for using PCA is that household SES explains most of the common variation in maternal education, household income, and the child's health insurance status. Given that these three indicators are highly correlated, an aggregate measure summarizing their variation is considered optimal to using separate variables in a multivariate model. PCA is commonly used to generate aggregate household wealth indices (Filmer and Pritchett, 2001). PCA has advantages over other methods such as those that arbitrarily assign equal or subjective weights to the individual variables. The scoring coefficients of the first principal component were used for generating the SES index. These are included in Table A1 in the Appendix. The first principal component explained 66.8% of the variation in the three index variables. The SES index is centered around 0 and ranges from −3 to 1.9. Higher values indicate higher SES.[5]

We also evaluated the relationships between the child's behavioral/emotional well-being and satisfaction with his/her own facial appearance and speech problems given the important role of these factors in influencing psychological adjustments and quality of life of affected children (Hunt *et al*, 2005; Damiano *et al*, 2007). Satisfaction with facial appearance was based on maternal report of how happy the child is with facial appearance on a four-category scale.[6] The majority (66.7%) of mothers indicated 'very happy'; about 25%, 7%, and 1% reported 'moderately happy', 'somewhat happy', and 'not at all happy'. Given the distribution of the responses, it is reasonable to compare the most optimal and common outcome of 'very happy' to the less common and optimal outcome of 'less than very happy'. Therefore, responses were dichotomized into an indicator of low satisfaction with facial appearance based on 'less than very happy' relative to high satisfaction based on 'very happy'. Combining 'moderately happy' with 'very happy' in one category may be suboptimal both theoretically and practically as only a few (seven) children in the study would serve as the reference group of unsatisfied with facial appearance.[7] The presence of speech problems was indicated by mother's response (yes or no) to a question of whether she or a health professional believed that the child needed speech therapy at any time during the past 12 months.[8] These questions about esthetics and speech outcomes have been used in previous oral cleft studies (Damiano *et al*, 2007).

Statistical analysis

We tested the significance in differences of proportions of T-scores at/above the 90th percentile between the

[2]For the CBCL 1.5–5, this was performed automatically by the instrument's official scoring program (Achenbach, 1999–2000).

[3]The question was: 'Do you feel your child is being cared for by an organized cleft care team? That is, an organized cleft care team made up of at least a surgeon, a dental professional and a speech professional'. Only 1 mother responded 'Don't know'.

[4]The question was: 'How many surgeries has your child had for his or her cleft thus far, not including placing tubes in his or her ears?'

[5]Table A1 shows how the various categories of the three variables forming the index (maternal education, total household income, and child's health insurance status/type) affect the index value.

[6]'Overall how happy would you say your child is with his or her facial appearance?' Response categories were 'very happy,' 'moderately happy,' 'somewhat happy,' or 'not at all'. This question was based on a clinical measure and developed in collaboration with expert clinicians at the University of Pittsburgh.

[7]As a sensitivity analysis, we repeated the regression models described below adjusting for an alternative dichotomous measure of satisfaction with appearance that combined 'very happy' and 'moderately happy' together in one category *vs* another category that combined 'somewhat happy' and 'not very happy'. The effects of team care, number of surgeries and SES on the behavioral outcomes were virtually unaffected with this change. Results are available from the authors upon request.

[8]The question was: 'Over the last 12 months, was there a time when you or a health professional thought your child needed speech therapy of any kind?'

study sample and the normative samples using a binomial test, separately for young (2–5 years) and older (6–12 years) children. As we describe below, the behavioral risk distributions were overall comparable between the two age groups, except for inattention/hyperactivity. Therefore, we combined the two age groups in additional analyses to increase sample size, but also conducted a separate analysis for inattention/hyperactivity for the older age group. We evaluated the bivariate relationships between the behavioral measures and cleft type, low satisfaction with facial appearance, and speech problems using chi-square tests of independence. We also estimated logistic regression models for the binary behavioral measures and ordinary least-squares regressions for the T-score measures of each of the four behavioral domains to evaluate simultaneously the effects of team care, number of cleft surgeries, and SES on the behavioral outcomes, adjusting for child age and cleft type, which are theoretically relevant for child behavior. Cleft type may have significant effects on behavior owing to differences in healthcare needs, with children who have both cleft lip and cleft palate generally requiring more healthcare interventions than those with either cleft alone. Child age (in years) was included because it is likely to be an important predictor of behavior and is also strongly correlated to number of surgeries. Child's age may also mediate the relationship between speech or facial appearance and behavioral health (Damiano *et al*, 2007).

Team care, number of surgeries, and SES may impact the study behavioral outcomes both directly as well as indirectly through their effects on satisfaction with facial appearance and speech performance. The bivariate analyses showed that satisfaction with facial appearance was related to behavioral outcomes, but perceived need for speech therapy was not. Therefore, in alternative models, we added low satisfaction with facial appearance as a covariate to evaluate how it mediates the effects of team care, number of surgeries, and SES on behavioral outcomes. We checked for and found no evidence of multicollinearity problems, with variance inflation factors of 1.5 or less in all regressions.

Results

Table 1 lists the study variables and their distributions. The average age of the children was 6.5 years. About 24% had cleft palate alone, 28% had cleft lip alone, and about 48% had cleft lip with palate. About one-third of the children were less than very satisfied with their facial appearance, and about 39% were reported to need speech therapy. About 78% of the children were reported to be cared for by an organized cleft team. The average number of surgeries was 2.2.

Table 1 Distribution of the study outcome, explanatory, and descriptive variables

Variable	*Complete data sample*	*% or Mean (s.d.) [Range]*
Outcome measures[a]		
Depression/anxiety risk (yes *vs* no; %)	104	8.7
Inattention/hyperactivity risk (yes *vs* no; %)	104	13.5
Aggressive/oppositional risk (yes *vs* no; %)	104	12.5
Somatic symptom risk (yes *vs* no; %)	104	13.5
Depression/anxiety T-score	104	52.7 (5.1) [50–73]
Inattention/hyperactivity T-score	104	54.4 (7.2) [50–83]
Aggressive/oppositional T-score	104	53.5 (6.4) [50–79]
Somatic symptom T-score	104	54.3 (6.5) [50–74]
Explanatory variables		
Team-care use (yes *vs* no; %)	103	77.7
Number of cleft surgeries	102	2.2 (1.4) [1–7]
SES index[b]	96	−0.01 (1.2) [−3.0–1.9]
Child's age (years)	104	6.5 (3.1) [2–12]
Cleft palate only (yes *vs* no; %)	104	24.0%
Cleft lip only (yes *vs* no; %)	104	27.9%
Cleft lip with cleft palate (yes *vs* no; %)	104	48.1%
Child not very satisfied with own facial appearance (yes *vs* no; %)	93	33.3%
Child needs speech therapy (yes *vs* no; %)	104	38.5%
Additional descriptive variables		
Child's race reported as White (yes *vs* no; %)	104	95.2%
Maternal age (years)	104	35.4 (7.0)
Child is 2–5 years old (yes *vs* no; %)	104	56.7%
Child is 6–12 years old (yes *vs* no; %)	104	43.3%

SES, socioeconomic status.
Complete data sample represents the number of children with complete data for the variables.
[a]The binary behavior indicators (yes/no) are based on a T-score of 63 or higher (at/above 90th percentile for normative samples) within each domain.
[b]The SES index is based on the first component scoring coefficients from a principal component analysis of maternal education, total household income, and child's health insurance status/type. The index is estimated for cases with complete data on all these characteristics and on behavioral outcomes.

T-scores at/above the 90th percentile indicating elevated risks were most prevalent for somatic symptoms and inattention/hyperactivity at about 13.5%, followed by aggressive/oppositional behavior (12.5%) and depression/anxiety (8.7%). None of these rates were statistically different from the 10% prevalence in the normative samples. Table 2 reports the rates of elevated risks separately for ages 2–5 and 6–12 years. None of these rates was significantly different from the normative samples or between the two age groups, except for inattention/hyperactivity in the older age group, which was 20.3% (compared with 10% in the normative sample and 4.4% in the younger sample).

Table 3 reports the distribution of the behavioral outcomes by satisfaction with facial appearance, presence of speech problems, and cleft type. Children who were not very satisfied with their own facial appearance were at significantly higher risk of behavioral problems on all domains, except for aggression. Reported need for speech therapy was not significantly correlated with the behavioral outcomes, although insignificantly higher rates of aggression and depression risks were observed among children with reported need for speech therapy. Some differences were observed by cleft type, but these were generally not statistically significant, except for the inattention/hyperactivity rate in children age 6 years and older, which was higher among children with both cleft lip with palate.

Table 4 reports the adjusted odds ratios (OR) of the effects of team care, number of surgeries, SES, and other model covariates on the child's behavioral outcomes from the logistic regression that simultaneously included all these variables. Table 5 reports the adjusted effects of these variables on the T-scores of the four behavioral domains as estimated from ordinary least-squares regression. Two different models are presented: the first excludes satisfaction with facial appearance as a covariate while the second adjusts for this variable. Team care did not have any significant effects on the binary risk or T-score outcome measures. The number of cleft surgeries was associated with a twofold increase in the risk of depression/anxiety with each additional surgery. However, the surgery effect on depression/anxiety decreased and became statistically insignificant when adjusting for low satisfaction with facial appearance. A similar result was observed with the T-score outcome measure, with a 1.2-point increase per additional surgery in the model that excludes satisfaction with facial appearance.

Table 2 Distribution of behavioral outcomes by age group

Variable	*Age 2–5 years (*N = *45)*	*Age 6–12 years (*N = *59)*
Depression/anxiety risk	11.1	6.8
Inattention/hyperactivity risk	4.4	20.3**
Aggressive/oppositional risk	11.1	13.6
Somatic symptom risk	13.3	13.6

**Significantly different at $P < 0.05$ from 10% rate in normative sample.

Furthermore, the number of cleft surgeries was significantly associated with an increase in the somatic symptom T-score by 1.3 points per additional surgery, with the effect being virtually insensitive to adjusting for satisfaction with facial appearance. The effect on the somatic symptom binary risk indicator was only marginally significant when adjusting for satisfaction with facial appearance. The regression results for inattention/hyperactivity separately for the older age group were similar to those from the analysis combining all ages.[9]

Higher SES was significantly associated with a decrease in the risks of inattention/hyperactivity, aggressive/oppositional behavior, and somatic symptoms. A one-point increase in the SES index (about one standard deviation) was associated with a 0.2-fold decrease in the inattention/hyperactivity risk and a 0.5-fold decrease in the aggressive/oppositional behavior and somatic symptom risks. Higher SES was significantly associated with a decrease in the T-scores of all four behavioral domains, ranging from 1-point decrease for depression/anxiety to 2.3-point decrease for inattention/hyperactivity. The SES effects were overall insensitive to adjusting for dissatisfaction with facial appearance except for aggressive/oppositional behavior (which became insignificant).

Child age had no significant associations with either the binary risk or T-score outcome measures. Cleft lip with palate was negatively associated with somatic symptom risk and with T-scores when adjusting for satisfaction with facial appearance. Finally, dissatisfaction with facial appearance was associated with an increase in depression/anxiety risk and T-scores and with an increase in aggressive/oppositional behavior T-scores.

Discussion

The study is one of the first to evaluate the risks of behavioral/emotional problems in children with oral clefts and include preschool-age children and to assess the effects of team care, number of surgeries, and SES on these risks in a population-based sample. While rates of T-scores at/above 90th percentiles indicating elevated risk were slightly higher for inattention/hyperactivity, aggressive/oppositional behavior, and somatic symptoms than the expected 10% based on normative samples, the differences were not statistically significant in the combined sample. The only exception was that elevated inattention/hyperactivity risks were twice as common for children age 6 years and older compared with the normative sample and about three times as common for the children in this age group with both cleft lip and palate. These findings suggest that most young children with oral clefts have similar behavioral health outcomes compared with unaffected children, but that older children may be at elevated risks of specific behavioral problems such as inattention/hyperactivity.

[9]Detailed results are available from the authors.

Table 3 Distribution of behavioral outcomes by satisfaction with facial appearance, need for speech therapy, and cleft type

	% of children at behavioral risk						
	Not very happy with facial appearance		Need speech therapy		Cleft type		
	Combining age groups						
Outcome	Yes (N = 31)	No (N = 62)	Yes (N = 40)	No (64)	Cleft lip only (N = 29)	Cleft palate only (N = 25)	Cleft lip with palate (N = 50)
% of children at behavioral risk							
Depression/anxiety risk	22.6***	3.3	12.5	6.3	6.9	8.0	10.0
Inattention/hyperactivity risk	22.6**	8.1	15.0	12.5	10.3	8.0	18.0
Aggressive/oppositional risk	16.1	6.5	17.5	9.38	13.8	12.0	16.0
Somatic symptom risk	22.6**	8.1	12.5	14.1	20.7	12.0	10.0
T-score Mean (s.d.)							
Depression/anxiety T-score	55.5*** (7.2)	51.3 (3.1)	53.1 (5.0)	52.4 (5.1)	51.9 (4.2)	52.4 (5.5)	53.2 (5.3)
Inattention/hyperactivity T-score	57.0 (7.5)	52.8 (5.1)	54.4 (6.9)	54.2 (6.2)	55.1 (7.5)	53.9 (6.4)	54.0 (6.0)
Aggressive/oppositional T-score	55.7 (8.8)	53.5 (6.2)	55.6 (7.8)	53.7 (6.7)	53.8 (5.4)	53.9 (7.0)	55.1 (8.2)
Somatic symptom T-score	55.6*** (8.3)	51.7 (4.0)	54.3 (6.9)	53.1 (6.1)	53.0 (5.6)	52.3 (5.3)	54.4 (7.3)
	Age 6–12 years						
	Yes (N = 25)	No (N = 33)	Yes (N = 40)	No (64)	Cleft lip only (N = 20)	Cleft palate only (N = 16)	Cleft lip with palate (N = 23)
Inattention/hyperactivity risk (%)	28.0	15.2	17.0	33.3	10.0	12.5	34.8*

The association between each of the binary risk indicators for behavioral problems and each of speech therapy need, satisfaction with facial appearance, and cleft type was assessed using a chi-square of independence. The associations with the T-scores were evaluated using ordinary least-squares regression separately for each of speech therapy need, satisfaction with facial appearance, and cleft type. The analysis for association with speech therapy need and with cleft type included 104 children. The analysis for association with satisfaction with facial appearance included 93 children (11 children had unreported data on this question).
** and ***$P < 0.05$ and < 0.01, respectively.

This suggests that extensive screening of all children with oral clefts for behavioral problems may be unnecessary given that the risks are low and that it may be burdensome to families and children. On the other hand, targeted screening focusing on inattention/hyperactivity (particularly for older children and those with both cleft lip with palate) and children from lower-SES households, who are less satisfied with their facial appearance, and who are undergoing or have undergone multiple surgeries may be cost-effective and more productive.

The study found no evidence that increasing team-care utilization has significant reductions in the risk of behavioral problems among children with oral clefts. Cleft team care is commonly expected to cover all the health needs of affected children including behavioral health. However, the results suggest limited effects of team care in addressing the behavioral health needs of children with oral clefts in the study population, despite the fact that the study had reasonable power to detect moderate effects of team care on behavior.[10] It is possible that parents who are concerned about behavioral issues are more likely to receive team care, which might result in underestimation of the team-care effects. Nonetheless, the study results highlight the importance of studying the current behavioral health professionals' representation on cleft teams and access to and effectiveness of behavioral care available through cleft teams to identify gaps and improve the provision and availability of behavioral care to affected children as needed.

The study provides some evidence that an increase in the number of surgeries may be associated with increased risk of behavioral or adjustment problems, particularly in the areas of depression/anxiety and somatic symptoms. The effect on anxiety/depression risk but not on somatic symptoms was attenuated by controlling for the child's dissatisfaction with his/her facial appearance, which was in fact a strong predictor for the risk anxiety/depression. This suggests that other factors besides satisfaction with facial appearance are mediating the effects on somatic symptoms. Therefore, it is important to consider these effects when planning surgical treatments for the child. Of course, the study does not assess the net effects of surgical treatments on the child's well-being, but rather highlights the importance of identifying why risks of behavioral problems are higher with an increase in cleft surgeries and finding ways to reduce these risks.

To our knowledge, this is one of the first studies to assess and find large socioeconomic disparities in risks of behavioral problems among children with oral clefts. It is well-known that higher SES may attenuate early life developmental deficits, while low SES may intensify their impacts (Feinstein, 2003). The positive SES effects on health are not unique to the cleft population and

[10]For an outcome rate of 10%, a sample of 93 observations and 5% type 1 error, the study had about 77% and 95% power to detect ORs of 0.8 and 0.75, respectively.

Table 4 Adjusted odds ratios for effects of study explanatory variables on the binary indicators for risk of behavioral problems

	Depression/anxiety		*Inattention/hyperactivity*		*Aggressive/oppositional*		*Somatic symptoms*	
	Model 1	*Model 2*	*Model 1*	*Model 2*	*Model 1*	*Model 2*	*Model 1*	*Model 2*
Team care	1.8 [0.2,17.7]	3.9 [0.3,53.3]	0.9 [0.1,11.2]	1.2 [0.1,16.6]	0.4 [0.1,2.2]	0.9 [0.1,6.0]	0.8 [0.2,3.9]	2.8 [0.4,19.5]
Number of cleft surgeries	2.1** [1.0,4.5]	1.7 [0.7,4.2]	1.7 [0.8,3.6]	1.6 [0.7,3.4]	0.9 [0.5,1.7]	0.8 [0.4,1.6]	1.6 [0.9,3.1]	1.9* [0.9,4.1]
SES index	0.9 [0.4,1.6]	0.8 [0.3,1.9]	0.2*** [0.1,0.5]	0.2*** [0.1,0.6]	0.5** [0.3,0.9]	0.6 [0.3,1.1]	0.6* [0.4,1.0]	0.5** [0.3,0.9]
Child's age	0.8 [0.6,1.1]	0.7** [0.5,1.0]	1.4 [0.9,2.0]	1.4 [0.9,2.2]	1.1 [0.8,1.4]	1.1 [0.8,1.5]	1.0 [0.8,1.2]	0.9 [0.7,1.2]
Cleft lip with palate[a]	0.2 [0.0,2.5]	0.1 [0.0,2.6]	2.4 [0.2,32.5]	1.7 [0.1,27.2]	4.4 [0.6,33.7]	3.4 [0.4,32.0]	0.2* [0.0,1.4]	0.04** [0.002,0.5]
Dissatisfied with facial appearance		17.5** [1.4,212.9]		2.4 [0.3,22.2]		2.3 [0.4,14.1]		3.2 [0.5,22.1]
Sample size[b]	93	84	93	84	93	84	93	84

SES, socioeconomic status.
The table lists the adjusted odds ratios for the explanatory variables included simultaneously in the logistic regression. Model 1 excludes satisfaction with facial appearance while Model 2 adjusts for this variable. For each categorical variable, the reference categories are as listed in Table 1.
[a]We combined cleft lip alone and cleft palate alone in the reference category for cleft lip with cleft palate as we did not observe significant behavioral differences between the two cleft types and because children who had cleft lip with cleft palate had about two surgeries more on average than children with cleft lip or cleft palate alone (mean number of surgery of 3.2 *vs* 1.3). Also, including two indicators of cleft type in the regression resulted in some stability problems in the regressions for some outcomes because of the small number of children with cleft lip alone or cleft palate alone.
[b]This is the sample with complete data on the outcome and *all* variables included simultaneously in the regression. The 95% confidence intervals for the odds ratios are in brackets.
*, ** and ***$P < 0.1$, < 0.05, and < 0.01, respectively.

Table 5 Adjusted effects of study explanatory variables on the T-scores of the behavioral outcomes

	Depression/anxiety		*Inattention/hyperactivity*		*Aggressive/oppositional*		*Somatic symptoms*	
	Model 1	*Model 2*	*Model 1*	*Model 2*	*Model 1*	*Model 2*	*Model 1*	*Model 2*
Team care	−1.17 (1.24)	−0.80 (1.33)	0.09 (1.67)	0.11 (1.84)	−1.28 (1.56)	0.38 (1.64)	−2.53 (1.52)	−0.87 (1.60)
Number of cleft surgeries	1.21** (0.53)	0.75 (0.58)	1.00 (0.72)	0.92 (0.80)	−0.09 (0.67)	−0.53 (0.71)	1.34** (0.65)	1.41** (0.70)
SES index	−1.14*** (0.43)	−0.99** (0.48)	−2.33*** (0.58)	−2.30*** (0.66)	−1.38** (0.54)	−1.01* (0.59)	−1.68*** (0.53)	−1.76*** (0.57)
Child's age	−0.12 (0.18)	−0.26 (0.21)	0.08 (0.25)	0.13 (0.29)	0.24 (0.23)	0.19 (0.25)	−0.04 (0.23)	−0.17 (0.25)
Cleft lip with palate[a]	−1.33 (1.50)	−0.90 (1.66)	−0.60 (2.03)	−0.71 (2.30)	2.74 (1.90)	2.32 (2.04)	−3.08* (1.85)	−4.60** (1.99)
Dissatisfied with facial appearance		3.23** (1.37)		0.31 (1.89)		3.46** (1.68)		2.60 (1.64)
Intercept	52.16*** (1.59)	52.61*** (1.70)	51.32*** (2.16)	51.05*** (2.35)	51.43*** (2.01)	50.35*** (2.09)	54.72*** (1.96)	53.92*** (2.04)
Sample size[b]	93	84	93	84	93	84	93	84

SES, socioeconomic status.
The table lists the adjusted effects the explanatory variables included simultaneously in the ordinary least-squares regression. Model 1 excludes satisfaction with facial appearance while Model 2 adjusts for this variable. For each categorical variable, the reference categories are as listed in Table 1.
[a]We combined cleft lip alone and cleft palate alone in the reference category for cleft lip with cleft palate as we did not observe significant behavioral differences between the two cleft types and because children who had cleft lip with cleft palate had about two surgeries more on average than children with cleft lip or cleft palate alone (mean number of surgery of 3.2 *vs* 1.3). Also, including two indicators of cleft type in the regression resulted in some stability problems in the regressions of some outcomes because of the small number of children with cleft lip alone or cleft palate alone.
[b]This is the sample with complete data on the outcome and *all* variables included simultaneously in the regression.
*, ** and ***$P < 0.1$, < 0.05, and < 0.01, respectively.

have been shown to be relevant for child health in the general population (Currie, 2009). Nonetheless, the large socioeconomic gradients in behavior highlight significant socioeconomic disparities in the behavioral health of children with oral clefts and suggest that children in less affluent households may be at significantly higher risks of behavioral problems. Therefore, additional attention to these children may be needed when providing behavioral care. The consistent and large associations of SES with all four behavioral domains indicate that household SES is one of the most relevant factors influencing child behavioral health. Further studies are needed to evaluate the access of children with oral clefts to behavioral treatments and how this varies by SES to assess the need for policies to improve access to this care.

While the study makes several contributions to this area, some caveats need to be considered when interpreting the results. First, the findings may be less generalizable to more racially diverse populations, given that about 95% of the sample's children were White. However, there is no information a priori that the effects of SES, number of surgeries, and access to team care on behavioral health vary significantly by race. Future studies with large sample sizes from diverse populations that allow stratification by race are needed to address this question. Second, while the participants in the study were similar to non-participants on maternal and child age and child's gestational age at birth and cleft type, it is possible that the participants may not be representative of the population of children with oral clefts in Iowa on certain unobserved clinical characteristics that may also relate to behavior. While there is no evidence that children with lower risks of behavioral problems were more likely to participate in the study, such sample-selection problem, if present, would bias the estimated rates of behavioral problems downward, making the study sample appear more similar to the normative sample. Third, even though we adjusted for cleft type and satisfaction with facial appearance, it is possible that other unmeasured confounders (such as cleft severity, developmental delay or parental concerns about child's behavior, and the resulting demand for more cleft team care) may be positively related to both increased cleft-team-care use and higher behavioral risks. These factors could result in underestimation of cleft-team-care effectiveness. However, given that the sample includes only isolated cleft cases and that we adjusted for cleft type, it is unlikely that this is a major bias. Future studies that can identify the causal effects of team care using designs such as instrumental variable analysis (with instruments such as distance to the nearest cleft team) are needed to evaluate the extent of such biases.

Finally, we were unable to include a matched group of children without oral clefts to compare the effects of SES on behavioral health between affected and unaffected children. However, we were able to compare the behavioral outcomes in the oral cleft sample to those in the normative samples used for standardizing the behavioral instruments. The normative samples are thought to be well representative of the general population of children without major mental/behavioral health complications. However, our cleft sample may differ somewhat from these normative samples on factors such as race, geographic location, and SES. These differences are more likely to affect comparisons with the CBCL normative data because the sample for the PBS-30 is based on children from the same geographic area as the cleft sample, and characteristics are similar for the percentage of mothers completing high school (Cleft sample = 97%; PBS-30 sample = 95%) and minority representation (Cleft = 5%; PBS-30 = 12%). Given the increase in behavior problems associated with lower SES, higher socioeconomic levels in the cleft sample, if present, could suggest that the percentage of children at elevated risk of behavioral problems may be biased downward. However, this is unlikely to bias the estimated effects of team care, number of surgeries, and SES on behavioral health, although it may increase the variance of the estimated effects and thus reduce statistical significance. Nonetheless, future studies that include unaffected controls from the same population as the group with oral clefts are important to validate the appropriateness of comparisons with normative data from standardized instruments.

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Disclosure

The authors have no financial interests or any conflict of interest in this work.

Author Contributions

All authors have approved this submission to the journal and take full responsibility for this work. Dr. Wehby designed and conducted the analysis and wrote the manuscript. Drs. Damiano, Romitti and Robbins designed the study and contributed to writing the manuscript. Mrs. Tyler contributed to the statistical analysis and to writing the manuscript. Dr. Lindgren contributed to the definition and interpretation of the behavioral outcomes and to writing the manuscript.

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Appendix

Table A1 First principal component scoring coefficients of the SES Index

	Scoring coefficients
Household income	0.61
Mother's completed years of education	
12	−1.23
13	−0.61
14	−0.23
15	0.02
16	0.41
17	0.99
Child insurance status/type	
Private insurance	0.17
Medicaid/CHIP	−0.81
Other	−1.30
Total variance explained by 1st principal component (%)	66.8

SES, socioeconomic status.
The table shows the SES index scoring coefficients from the first principal component. Positive and negative coefficients indicate increases and decreases in SES, respectively. The index is estimated for cases with complete data on the index and outcome variables.