



Published in final edited form as:

Child Care Health Dev. 2016 January ; 42(1): 42–50. doi:10.1111/cch.12276.

Psychosocial Wellbeing of Parents of Children with Oral Clefts

Nichole Nidey, BA,

Research Specialist, University of Iowa, Department of Pediatrics, 500 Newton Road, 2182 ML, Iowa City, Iowa 52242, Phone: 319-353-4365, Fax: 319-335-6848

Lina M. Moreno Uribe, DDS, PhD.,

Assistant Professor, Orthodontics-Dows Institute, 401 DSB, University of Iowa, (319) 335-8912

Mary M. Marazita, Ph.D., and

Director, Center for Craniofacial and Dental Genetics, Professor and Vice Chair, Department of Oral Biology, School of Dental Medicine, Professor, Department of Human Genetics, Graduate School of Public Health, Professor, Clinical and Translational Science, Professor, Department of Psychiatry, School of Medicine, University of Pittsburgh, Suite 500 Bridgeside Point, 100 Technology Dr., Pittsburgh, PA 15219, ph: 412-648-8380, FAX: 412-648-8779

George L. Wehby, MPH, PhD.*

Associate Professor, University of Iowa, Research Associate, National Bureau of Economic Research, Departments of Health Management and Policy, Economics, and Preventive & Community Dentistry, and Public Policy Center, University of Iowa, 145 N. Riverside Dr., 100 College of Public Health Bldg., Room N248, Iowa City, Iowa 52242-2007

Nichole Nidey: nichole-nidey@uiowa.edu; Lina M. Moreno Uribe: lina-moreno@uiowa.edu; Mary M. Marazita: marazita@pitt.edu

Abstract

Background—Parents of children with oral clefts may be impacted psychosocially in several ways, but empirical evidence remains relatively sparse. The aim of this study was to identify predictors of psychosocial wellbeing of parents of affected children.

Methods—The study included a total sample of 287 parents (171 mothers, 116 fathers) of children with oral clefts. Parents completed validated psychosocial instruments to measure social avoidance and distress, fear of negative evaluation scale, self-esteem and interpersonal support. Regression analysis was used to evaluate how selected child, parent, and household characteristics relate to psychosocial outcomes focusing on child's cleft type, sex, and age, differences between mothers and fathers, marital status, and household income.

Results—Fathers had higher self-esteem than mothers ($p=0.01$) and lower concern of being negatively judged by others ($p<0.0001$) but also had lower perception of having someone to talk to about their problems ($p=0.01$). High household income was associated with greater self-esteem and perception of social support (<0.05). Parents of male affected children had greater perception of social support than parents of female affected children ($p=0.04$). No significant differences in parental psychosocial status measures were found by cleft type. Similarly, there is little evidence

*Corresponding Author: George L. Wehby, MPH, PhD., Phone: 319-384-3814, Fax: 319-384-4371, george-wehby@uiowa.edu.

of changes with child age, except for an increase in parental distress and decline in self-esteem during mid adolescence (age 15–17 years).

Conclusions—The results indicate that mothers and fathers of children with oral clefts may differ in their psychosocial adjustment and that mothers may overall experience more psychosocial problems than fathers. Also, parents from less wealthy households may be at greater risk. Parental psychosocial status should be considered in holistic family-based treatment approaches to reduce burden on affected families and improve their wellbeing.

Keywords

oral clefts; cleft lip; cleft palate; psychosocial status; depression; parent health; parental wellbeing

Introduction

Oral clefts including cleft lip with or without cleft palate or cleft palate alone are one of the most common congenital anomalies that affect 1 per 1250 births in the United States, or close to 7000 births annually (Parker et al., 2010). About 50% of cases with cleft palate only and 70% of cases with cleft lip with/without palate case are isolated occurring without any additional structural or cognitive abnormalities (Mossey et al., 2009). Genetic as well as environmental factors particularly maternal smoking and use of folic acid supplements influence the risk of having a child with an oral cleft (Wehby et al., 2011; Wehby & Murray, 2010).

Oral clefts bring a wide spectrum of ramifications for the health and quality of life of affected individuals throughout their lifespan (Wehby and Cassell, 2010). Early in life, affected individuals are at an increased risk of experiencing difficulties related to feeding (Nackashi, 2002) in addition to the need for cleft repair surgeries, typically done by 12 months of age to close the lip and 18 months to close the palate (Genisca et al., 2009). Later in childhood, problems with speech, hearing, dental, psychological, and learning issues may emerge, requiring multiple healthcare interventions and possibly creating a financial burden for affected families. The healthcare costs for children aged 0–10 years with oral clefts exceed those of unaffected children by about 8 times (Boulet et al., 2009). This is in part driven by a greater use of hospital services, which is more than doubled during ages 0–19 years compared to unaffected children (Wehby, Pedersen, et al., 2012). During adulthood, greater use of hospital care and a higher mortality risk have also been reported (Christensen et al., 2004; Wehby, Pedersen, et al., 2012).

Having a child with an oral cleft may affect the psychosocial wellbeing of parents in several ways. In addition to the parents' concern about the health and quality of life experiences of their affected children, parents may become financially burdened by the intensive healthcare needs and out-of-pocket expenditures as well as their time costs in seeking healthcare services (such as being away from work). Previous studies have reported that mothers of a child with a cleft experience a multitude of emotions including shock, guilt and grief after the birth of their child (Bradbury & Hewison, 1994). Many mothers experience concern about feeding their child (Chuacharoen et al., 2009), sensitivity towards reactions from others (Johansson, 2004), and making decisions regarding treatment and interventions for

their child (Nelson, Caress et al., 2012). These experiences may extend from the time when parents first know about their child's diagnosis (whether during pregnancy or at delivery) through childhood. The psychosocial wellbeing of parents may be further impacted by the psychological problems that children with oral clefts may be at greater risk for particularly separation anxiety disorder and inattention/hyperactivity (Tyler et al., 2013; Wehby, Tyler, et al., 2012) as well as academic achievement problems compared to unaffected children (Wehby et al., 2014). Finally, parents may be concerned about the risk of having another affected child and may modify their fertility behaviors subsequent to the birth of an affected child (Wehby, Nyarko, & Murray, 2014), which could further impact their psychosocial status.

To the best of our knowledge, only a handful of published empirical studies (summarized below) have directly evaluated the psychosocial status of parents of children with clefts. Most of these studies have focused on comparing outcomes of parents of affected children to those of unaffected ones. Less has been done however on examining factors that associate with psychosocial status of parents of affected children to identify parents at greatest risk of psychosocial problems in this population. The majority of studies had small samples (less than 150 parents of affected children) and included a limited number of psychosocial measures. Furthermore, the majority did not include data on fathers. The studies varied substantially in their sample sizes and their findings are generally mixed. The broader literature suggests that parents may experience emotional strain but that appears to fade once the affected child reaches preschool age (Nelson, Glenny et al., 2012), although little work has directly compared parental outcomes by child's age. Also, most of the research has excluded paternal outcomes (Nelson, Glenny et al., 2012).

A small study of 47 parents of children with oral clefts reported an increased parental stress during infancy and toddlerhood (Pope, Tillman, & Snyder, 2005). In contrast, Collett et al (2011) showed no significant differences in psychosocial status between 93 parents of children with oral clefts and 124 parents of unaffected children. Baker et al (2009) measured how families cope and levels of support affect adjustment to having a child with an oral cleft. The study found that families with high levels of social support had less distress and greater adjustment in a sample of 103 parents of children with clefts. That study also reported differences in parental stress by child's age, extent of medical need, and cleft type (better adjustment among parents of children with cleft lip with palate). Crerand et al (2014) evaluated 1200 children with clefts and their parents to examine family functioning utilizing the Family Environment Scale (FES) which measures the environmental and social characteristics of families. That study focused on the relationship domain of the FES which evaluates how helpful family members are to one another, family cohesion and support (Moos & Moos, 1994) They reported that families scored in normative range of this instrument but found differences in family functioning by race and ethnicity (African American and Hispanic families reported less expressiveness and cohesion than non-Hispanic Caucasian families), treatment needs (families who had a child in need of a surgery for a functional versus an aesthetic outcome reported to be closer), and type of insurance coverage (better outcomes among privately insured families compared to publicly insured ones). One recent study explored the father's perspective in parenting a child with clefts and reported that they have similar experiences to mothers (Stock & Rumsey, 2015)

In this study, we evaluate the psychosocial wellbeing of parents of affected children with clefts on several domains and measures of psychosocial status and focus on examining differences by child's cleft type, sex, and age, mothers versus fathers, marital status, and household income. We study a large sample of parents and evaluate a wide range of psychosocial domains. In doing so, our study augments the current knowledge of the psychosocial experiences of parents of children with oral clefts.

Methods

Study Sample

The study included a multi-site sample of parents of affected children enrolled between 2003 and 2011. Parents of children age 0–17 years with isolated oral clefts and no history of genetic conditions were eligible for the study. Since having multiple affected children may modify the impact on parental psychosocial status and given that parents of multiple affected children were only a small group in this study and could not be analyzed separately (27 parents), these parents were excluded from the analysis. Parents were identified and recruited at three craniofacial clinics: University of Iowa Craniofacial Clinic, St Louis Children's Hospital, and Cleft Craniofacial Center at Children's Hospital of Pittsburgh. In addition, parents of a population-wide sample of children with isolated oral clefts from the state of Iowa identified from the Iowa Registry of Congenital and Inherited Disorders (IRCID) were also invited to participate. Subjects were consented in person following IRB approved protocols and completed study procedures either at the study site where they were recruited or at their home during visits by study staff. A total of 287 parents including 171 mothers and 116 fathers were included in this study. The exact number of observations varied slightly between the regressions due to differences in missing data between the psychosocial outcomes and ranged from 248 to 274 (some individuals who had missing data on one instrument had complete data on another).

Measures

Data were collected by trained study staff during interviews with the parents using similar instruments across all three sites. In addition to collecting data on demographic and socioeconomic characteristics, parents were asked to complete the following four self-administered instruments for measuring psychosocial status and social support: including the Social Avoidance and Distress (SAD) scale, Fear of Negative Evaluation (FNE) scale, Rosenberg Self Esteem (RSE), and Interpersonal Support Evaluation List (ISEL).

SAD—The Social Avoidance and Distress (SAD) scale was developed to evaluate the anxiety and distress an individual undergoes in social situations (D. Watson & Friend, 1969). This scale has been shown to be a valid measure of social anxiety in previous studies (internal consistency = .77, KR-20 = .94). The majority of the questions are related to how the respondent feels in various social situations. This scale is comprised of 28 true-false statements, half of which measure social avoidance while the other half measure social anxiety; therefore the measure can be analyzed aggregated or broken down into these two subscales. Total scores range from 0 to 28 where higher scores indicate more avoidance and distress (Leary, 1991).

FNE—The Fear of Negative Evaluation (FNE) scale was developed to evaluate the respondent's apprehension of being negatively judged by others and has high validity (internal consistency KR-20 = .92 & .94; (D. F. Watson, Ronald, 1969). The FNE scale is comprised of 30 items each answered on a five point Likert scale from not-at-all to extremely. Respondents choose the extent to which a statement for how much they worried about how others would evaluate them in social situations applied to them. Scores range from 30–150 with higher scores indicating increased apprehension of negative evaluation (Leary, 1991).

RSE—The Rosenberg Self Esteem (RSE) was utilized to evaluate subjects' feelings of self-acceptance and self-worth (Rosenberg, 1965). This scale has been widely used to measure self-esteem and has been shown to be a valid instrument (internal consistency 0.77 and reproducibility 0.90) (Rosenberg, 1965). The scale is made up of 10 items each answered on a four-point Likert scale from strongly disagree to strongly agree. Respondents are asked to state their agreement with statements related to how they feel about themselves. Scores can range between 10 and 40 with higher scores indicating higher self-esteem (Leary, 1991).

ISEL—The Interpersonal Support Evaluation List (ISEL) was developed to measure the respondent's perception of social support (Cohen, 1983) and has a consistency of 0.70 (Bates & Toro, 1999). The ISEL consists of 40 items each answered on a 4-point Likert scale on which respondents rank the applicability of statements to themselves from *Definitely False* to *Definitely True*. This instrument can be analyzed in its entirety or in four subscales (10 items each) that measure the respondent's perception of belonging, availability of tangible items, self-esteem scale, and appraisal. The appraisal subscale assesses the respondent's perception of having another person to discuss their feelings and problems as well as a person to obtain advice from. The tangible items subscale measures perception of having someone that can provide material assistance such as a loan or help fixing a car or appliance. The belonging scale measures the respondent's perception of availability of someone to do activities with and the self-esteem scale measures perception of self-worth.

Statistical Analysis

Since the psychosocial instruments provide continuous measures, we estimated regressions of these instrument scores, one at a time, over the explanatory variables of interest using ordinary least squares (OLS). Regressions were estimated for the total scores of each of the four instruments as well as the scores of six subscales (2 from the SAD and 4 from the ISEL). We examined the associations of these outcomes with several child and parent characteristics included simultaneously in the regression. Child's cleft type was represented by two indicators for cleft lip only and cleft palate only with cleft lip and palate as the reference group. Age of affected child was first captured in the main models by two indicators for pre-school age (<6 years) and adolescent years (13–17) with elementary school ages 6–12 as the reference category. We also explored narrower age groups including 0–2, 3–5, 6–9, 10–12, 13–14, and 15–17 years with 6–9 years as the reference category. Indicators for child's sex and for fathers versus mothers were also included. The models also included parental marital status and years of education as well as dummy variables (0/1) for household income categories. Finally, to account for potential geographic differences in

psychosocial status, we included dummy variables for the study site where parents were recruited to account for. We did not control for race since 98% of the sample were Caucasian.

Results

Sample Description

Tables 1 and 2 provide summary statistics for the explanatory and outcome variables, respectively. Sixty seven percent of the parents (n= 193) had a male affected child. The majority of affected children had cleft lip with palate (170) followed by cleft lip only (79), and cleft palate only (38). The majority of the parents were married (84%), and had incomes of 46K or higher (~7%); about half had incomes of 61K or higher. Parents had about 15 years of education on average.

Differences in Psychosocial Status

Tables 3, 4, 5, and 6 report the regression results for the SAD, RSE, ISEL, and FNE scales, respectively. We summarize below the main results for child and parental characteristics, household income, and study site.

Child Characteristics—There was little evidence of differences in parental psychosocial status by child's characteristics. No significant differences in any of the parental psychosocial measures were observed by child's cleft type (cleft lip only, cleft palate only, cleft lip with palate) or across the three age groups 0–5, 6–12, and 13–17 years examined in the main models. When examining the narrower age groups, none the differences were significant except for: 1-an increase in parental distress by about 2.8 on the SAD distress subscale, which was a large increase (about 70% relative to the sample mean reported in Table 2) but marginally significant (p=0.09); and 2- a decline in self-esteem on the ISEL subscale by 3.5 points (17% relative to sample mean; p=0.04), both among parents of adolescents aged 15–17 years compared to parents of children aged 6–9 years (additional detailed results available upon request).

No significant differences were observed by child's sex except for the ISEL scale. Parents of a male affected child had greater perception of social support than those of a female affected child (Table 5) by 4.33 units or by 4.6% compared to the sample average on the ISEL scale (p= 0.04).

Parental Demographic Characteristics—There were no significant differences between mothers and fathers on the SAD and the total ISEL scores. However, fathers had higher self-esteem than mothers (Table 4) by 1.57 units (6.6% of sample average) on the RSE scale (p=0.01) and less concern of being negatively judged by others (Table 6) by 12.71 points (15% of sample average) on the FNE scale (p=<0.0001). However, fathers also reported a lower perception of having someone to talk to about their problems than mothers by 1.7 points (7% of sample average) on the appraisal domain of ISEL (p=0.01). Interestingly, married parents also had lower self-esteem on the RSE. Although not

statistically significant, married parents tended to report worse outcomes on all psychosocial measures.

Household Income—The psychosocial status of parents was improved with higher household income on several domains and some of the differences were fairly large. For example, parents with an annual household income of \$105,000 or more had higher self-esteem on the RSE scale (Table 4) by 3.53 units ($p=0.02$) or by 15% relative to sample mean of the RSE score than those with a household income under \$16,000. They also had greater interpersonal support (ISEL) by 14 units or by 15% relative to sample mean ($p=0.01$; Table 5). Income gradients were also observed on the belonging, tangible items, and appraisal subscales of the ISEL scale (Table 5).

Two exceptions are worth noting however. Parents with an annual income of \$16,000–30,999 had lower self-esteem by 2.71 ($p=0.04$) units or by 13% relative to sample mean on the self-esteem subscale of the ISEL scale than those with an income under \$16,000 (Table 5). They also had lower self-esteem on the RSE scale although the difference was not statistically significant. Also, higher income was associated with an increase in fear of negative evaluation by others on the FNE scale but none of the differences were significant (Table 6). Interestingly, parental education was not significantly related to any of the psychosocial outcomes conditional on all the other covariates.

Study Site—No significant differences in outcomes were observed by study site except for two outcomes. Compared to parents enrolled in Iowa, parents enrolled in Pittsburgh reported less social avoidance by 1.14 units ($p=0.01$) or 40% relative to sample mean on the avoidance subscale of the SAD instrument (Table 3) and less fear of negative evaluation by 5.8 units ($p=0.04$) or by 7% relative to sample mean on the FNE instrument (Table 6). There were no significant differences between parents from Iowa and Saint Louis.

Discussion

Our study identifies important aspects of variation in the psychosocial status of parents of children with oral clefts. We find differences between fathers and mothers, with fathers showing greater self-esteem and reduced concern about negative judgement from others. At the same time, mothers report more having someone to talk to about their problems than fathers. Furthermore, parents with a high household income have greater self-esteem and perception of social support than those in poor households. Social support is reported to be higher among parents of male affected children compared to female affected children. Interestingly however, we find no differences in parental psychosocial outcomes by cleft type. Similarly, there is little evidence of changes with child age, except for some evidence of increased distress and reduced self-esteem among parents of children aged 15–17 years (compared to 6–9 years). While some of the differences such as by sex of the affected child may be considered small (~5% of sample mean), other differences such as in fear of negative evaluation between fathers and mothers and in self-esteem and social support by income as well as the increased distress and reduced self-esteem during mid adolescence are moderate to large (above 10%).

Understanding the psychosocial outcomes of parents of children with oral clefts can better prepare healthcare professionals to treat affected families. Our results support a family-based treatment approach as opposed to only focusing on the affected child. Besides the direct effect on parents, parental (especially maternal) wellbeing is an important predictor of a child's psychological outcomes (Despars et al., 2011) and the mental health of parents can impact child's health and educational achievement (Farahati, 2003; Frank, 2009; Propper, Rigg, & Burgess, 2007). Incorporating psychosocial assessments of parents can be readily done in settings that follow a team-based approach to treating children with oral clefts with psychosocial expertise as part of the team.

Our study offers several insights for improving treatment practices to address parental wellbeing. The results indicate that mothers and fathers of children with oral clefts differ in their psychosocial adjustment. While mothers may overall experience more psychosocial problems related to lower self-esteem and concern about judgement from others, fathers may have more internalizing problems. Furthermore, parents from poor households may be at greater risk of psychosocial problems. Evaluating the psychosocial status of parents with a focus on these domains while recognizing potential differences between fathers and mothers, and targeting parents of limited financial means may help to identify and remedy adjustment problems and improve family wellbeing. Addressing availability of social support especially among parents of female affected children may also be needed. The overall lack of differences by age indicates that parents should be screened as early as possible to identify and address any issues before they become more burdensome. However, the potential for increased distress and reduced self-esteem during child adolescence suggests that re-evaluating parental psychosocial status during this period maybe particularly useful. Finding no differences by cleft type indicates that it should not be a criterion for selecting parents for psychosocial assessments.

Our study has several strengths including being one of the largest studies to date in this area, the multiple psychosocial measures, and examining several child and parental characteristics in a multivariate analysis. However, it also has some caveats that should be addressed in future research. The study sample is essentially a convenience sample of individuals who were willing to participate in this study. It is possible that parents of children with greater health needs and those who seek more health services are more represented in the sample. We are unable to directly examine the extent of ascertainment bias with the available data. Furthermore, the sample included mostly Caucasian individuals (98%); focusing on minority groups in future studies is needed since the observed demographic and socioeconomic associations with parental outcomes may differ by race/ethnicity. While the nature of the sample may reduce the generalizability of our results, it is important to note that we focus on conditional relationships between the explanatory variables and the psychosocial outcomes rather than on the unconditional distribution of these outcomes, which may reduce the effect of ascertainment bias.

Our study focuses on parents but not unaffected siblings. Future studies of the psychosocial outcomes of unaffected siblings are needed in order understand how families as a unit are impacted by having a member with oral clefts and to help guide treatment and prevention approaches to improve the health and wellbeing of all family members. Finally, it is

important to identify the underlying pathways for the differences in psychosocial outcomes that we find in order to design specific and targeted interventions that can effectively prevent and reduce the psychosocial burden of parents of children with clefts and enhance their wellbeing.

Acknowledgments

The study was supported by the Centers for Disease Control and Prevention (CDC) grant 1R01DD000295. The contents of this work are sole responsibility of the authors and do not necessarily represent the official views of the CDC. None of the authors has any conflict of interest with this work.

References

- Baker SR, Owens J, Stern M, Willmot D. Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. *Cleft Palate Craniofac J*. 2009; 46(3):229–236.10.1597/08-075.1 [PubMed: 19642758]
- Bates DS, Toro PA. Developing measures to assess social support among homeless and poor people. *Journal of Community Psychology*. 1999; 27(2):137–156.
- Boulet SL, Grosse SD, Honein MA, Correa-Villasenor A. Children with orofacial clefts: health-care use and costs among a privately insured population. *Public health reports*. 2009; 124(3):447–453. [PubMed: 19445422]
- Bradbury ET, Hewison J. Early parental adjustment to visible congenital disfigurement. *Child Care Health Dev*. 1994; 20(4):251–266. [PubMed: 7955137]
- Christensen K, Juel K, Herskind AM, Murray JC. Long term follow up study of survival associated with cleft lip and palate at birth. *BMJ*. 2004; 328(7453):1405–1408. [PubMed: 15145797]
- Chuacharoen R, Ritthagol W, Hunsrisakhun J, Nilmanat K. Felt needs of parents who have a 0- to 3-month-old child with a cleft lip and palate. *The Cleft palate-craniofacial journal: official publication of the American Cleft Palate-Craniofacial Association*. 2009; 46(3):252–257. [PubMed: 19642744]
- Cohen S, Hoberman H. Positive events and social supports as buffers of life change stress. *Journal of Applied Social Psychology*. 1983; 13:99–125.
- Collett BR, Cloonan YK, Speltz ML, Anderka M. Psychosocial Functioning in Children with and without Orofacial Clefts and their Parents. *Cleft Palate Craniofac J*. 2011:397–405. [PubMed: 21214307]
- Crerand CE, Rosenberg J, Magee L, Stein MB, Wilson-Genderson M, Broder HL. Parent-Reported Family Functioning Among Children With Cleft Lip/Palate. *Cleft Palate Craniofac J*. 2014.10.1597/14-050
- Despars J, Peter C, Borghini A, Pierrehumbert B, Habersaat S, Muller-Nix C, Hohlfeld J. Impact of a Cleft Lip and/or Palate on Maternal Stress and Attachment Representations. *Cleft Palate Craniofac J*. 2011; 48(4):419–424. [PubMed: 20500071]
- Farahati FMDE, Wilcox-Gok V. The Effects of Parents' Psychiatric Disorders on Children's High School Dropout. *Economics of Education Review*. 2003; 22(2):167–178.
- Frank RGM, Ellen. The Effect of Maternal Depression and Substance Abuse on Child Human Capital Development. National Bureau of Economic Research. 2009
- Genisca AE, Frias JL, Broussard CS, Honein MA, Lammer EJ, Moore CA, National Birth Defects Prevention S. Orofacial clefts in the National Birth Defects Prevention Study, 1997–2004. *Am J Med Genet A*. 2009; 149A(6):1149–1158. [PubMed: 19441124]
- Johansson, BaR; Karin. Parents' Experiences of having a child with cleft lip and palate. *Journal of Advanced Nursing*. 2004; 47(2):165–173. [PubMed: 15196190]
- Leary, MR. Social Anxiety, Shyness and Related Constructs. In: Robinson, JP.; Shaver, PR.; Wrightsman, LS.; Andrews, FM., editors. *Measures of Personality and Social Psychological Attitudes*. Academic Press; 1991. p. 165-168.p. 177-179.
- Moos, RH.; Moos, BS. *Family environment scale manual*. Consulting Psychologists Press; 1994.

- Mossey PA, Little J, Munger RG, Dixon MJ, Shaw WC. Cleft lip and palate. *Lancet*. 2009; 374(9703): 1773–1785.10.1016/S0140-6736(09)60695-4 [PubMed: 19747722]
- Nackashi, JA.; Dedlow, ER.; Wood-Dixon, V. Health care for children with cleft lip and palate: comprehensive services and infant feeding. In: Wyszynski, DF., editor. *Cleft lip and palate: from origin to treatment*. Oxford University Press USA; 2002. p. 303-316.
- Nelson P, Glenny AM, Kirk S, Caress AL. Parents' experiences of caring for a child with a cleft lip and/or palate: a review of the literature. *Child: care, health and development*. 2012; 38(1):6–20.
- Nelson PA, Caress AL, Glenny AM, Kirk SA. 'Doing the "right" thing': how parents experience and manage decision-making for children's 'normalising' surgeries. *Social science & medicine*. 2012; 74(5):796–804. [PubMed: 22305806]
- Parker SE, Mai CT, Canfield MA, Rickard R, Wang Y, Meyer RE, National Birth Defects Prevention N. Updated National Birth Prevalence estimates for selected birth defects in the United States, 2004–2006. *Birth Defects Res A Clin Mol Teratol*. 2010; 88(12):1008–1016. [PubMed: 20878909]
- Pope AW, Tillman K, Snyder HT. Parenting stress in infancy and psychosocial adjustment in toddlerhood: a longitudinal study of children with craniofacial anomalies. *Cleft Palate Craniofac J*. 2005; 42(5):556–559. [PubMed: 16149839]
- Propper C, Rigg J, Burgess S. Child health: evidence on the roles of family income and maternal mental health from a UK birth cohort. *Health Econ*. 2007; 16(11):1245–1269. [PubMed: 17311355]
- Rosenberg, M. *Society and the Adolescent Self-image*. Princeton University Press; 1965.
- Stock NM, Rumsey N. Parenting a child with a cleft: the father's perspective. *Cleft Palate Craniofac J*. 2015; 52(1):31–43.10.1597/13-035 [PubMed: 24295441]
- Tyler MC, Wehby GL, Robbins JM, Damiano PC. Separation Anxiety in Children Ages 4 Through 9 with Oral Clefts. *Cleft Palate Craniofac J*. 2013:520–527. [PubMed: 23451755]
- Watson D, Friend R. Measurement of social-evaluative anxiety. *Journal of consulting and clinical psychology*. 1969; 33(4):448–457. [PubMed: 5810590]
- Watson D, Ronald F. Measurement of Social-Evaluative Anxiety. *Journal of Consulting and Clinical Psychology*. 1969; 33(4):448–457. [PubMed: 5810590]
- Wehby G, Collet B, Barron S, Romitti P, Ansley T, Speltz M. Academic Achievement of Children and Adolescents with Oral Clefts. *Pediatrics*. 2014 In Press.
- Wehby G, Jugessur A, Murray JC, Moreno L, Wilcox A, Lie RT. Genes as Instruments for Studying Risk Behavior Effects: An Application to Maternal Smoking and Orofacial Clefts. *Health Serv Outcomes Res Methodol*. 2011; 11(1–2):54–78. [PubMed: 22102793]
- Wehby GL, Murray JC. Folic acid and orofacial clefts: a review of the evidence. *Oral Dis*. 2010; 16(1):11–19. [PubMed: 20331806]
- Wehby GL, Nyarko KA, Murray JC. Oral cleft recurrence risk and subsequent maternal fertility preferences and behavior in Brazil. *Birth Defects Res A Clin Mol Teratol*. 2014; 100(1):48–56. [PubMed: 24382743]
- Wehby GL, Pedersen DA, Murray JC, Christensen K. The effects of oral clefts on hospital use throughout the lifespan. *BMC Health Serv Res*. 2012; 12:58–75. [PubMed: 22405490]
- Wehby GL, Tyler MC, Lindgren S, Romitti P, Robbins J, Damiano P. Oral clefts and behavioral health of young children. *Oral Dis*. 2012; 18(1):74–84. [PubMed: 21883709]

Table 1

Descriptive Statistics for the Explanatory Variables

Variable	N [%] or Mean (SD)
Cleft Type	
Cleft Lip Only	77 [28.10]
Cleft Palate Only	38 [13.87]
Cleft Lip with Palate*	159 [58.03]
Child's Age	
<6 years	129 [47.08]
6–12 years*	120 [43.80]
13–17 years	25 [9.12]
Gender of Affected Child	
Male	182 [66.42]
Female*	92 [33.58]
Parent	
Father	112 [40.88]
Mother*	162 [59.12]
Marital Status	
Married	230 [83.94]
Unmarried*	44 [16.06]
Parent's Years of Education	14.87 (2.74)
Household income	
<\$16,000*	17 [6.20]
16,000–30,999	27 [9.85]
31,000–45,999	55 [20.07]
46,000–60,999	31 [11.31]
61,000–75,999	38 [13.87]
76,000–90,999	26 [9.49]
91,000–105,000	36 [13.14]
105,000 and Up	44 [16.06]
Enrollment Site	
Iowa*	145 [52.92]
Pittsburgh	116 [42.34]
Saint Louis	13 [4.74]

Notes: Sample counts and frequencies, N [%], are shown for categorical variables. Mean (SD) is shown for years for education. The descriptive statistics are calculated for the subsample of 274 individuals (out of a total of 287 participants) with complete data on all explanatory variables and on the RSE instrument, which had the lowest rate of missing data.

Table 2

Descriptive Statistics for the Psychosocial Outcomes

Outcome	N	Mean	SD
SAD	248	6.30	6.27
Distress	272	3.95	4.08
Avoidance	251	2.77	2.98
RSE	274	23.86	4.78
ISEL	266	93.72	16.90
Belonging	274	23.25	5.19
Self-esteem	269	21.12	4.22
Tangible Items	273	25.34	4.58
Appraisal	274	24.15	5.10
FNE	264	84.69	20.59

Notes: All individuals have complete data on the explanatory variables in Table 1.

Author Manuscript

Author Manuscript

Author Manuscript

Author Manuscript

Table 3

Regression Results for the Parental SAD Scale and Distress and Avoidance Subscales

Explanatory Variables	SAD Full Scale	Distress	Avoidance
Cleft Type			
Cleft Lip Only	-0.12(0.95)	-0.04(0.583)	-0.12(0.45)
Cleft Palate Only	-1.55(1.22)	-0.65(0.76)	-0.73(0.57)
Child's Age			
Age <6 years	0.59(0.89)	0.36(0.55)	0.42(0.42)
Age 13–17	2.30(1.62)	1.33(0.96)	1.07(0.75)
Male vs Female Affected Child			
Father vs Mother	0.82(0.83)	-0.22(0.51)	0.60(0.39)
Married vs Unmarried			
Parent's Years of Education	-0.06(0.16)	-0.07(0.10)	-0.03(0.08)
Household income			
\$16,000–30,999	0.74(2.29)	0.67(1.31)	0.58(1.07)
\$31,000–45,999	-2.59(2.11)	-1.58(1.27)	-0.89(0.98)
\$46,000–60,999	-2.37(2.18)	-1.55(1.32)	-0.88(1.02)
\$61,000–75,999	-1.87(2.21)	-1.21(1.34)	-0.67(1.03)
\$76,000–90,999	-4.55(2.38)*	-2.70(1.45)	-1.67(1.12)
\$91,000–105,000	-2.33(2.23)	-1.40(1.35)	-1.12(1.04)
\$105,000 and Up	-3.47(2.21)	-2.23(1.36)	-1.54(1.03)
Enrollment Site			
Pittsburgh	-1.39(0.93)	0.02(0.57)	-1.14(0.43)***
Saint Louis	-0.49(1.95)	0.10(1.21)	-0.41(0.92)

Note: The table reports regression coefficients and standard errors in parentheses;

* $p < 0.1$,

** $p < 0.05$,

*** $p < 0.01$; reference categories for the explanatory variables are as shown in Table 1. An increase in the scores (positive coefficients) indicates a decline in psychosocial wellbeing.

Table 4

Regression Results for the Parental RSE Scale

Explanatory Variables	RSE Scale
Cleft Type	
Cleft Lip Only	0.56(0.66)
Cleft Palate Only	0.47(0.84)
Child's Age	
Age <6 years	-0.50(0.62)
Age 13-17	-0.73(1.06)
Male vs Female Affected Child	0.08(0.60)
Father vs Mother	1.57(0.58)***
Married vs Unmarried	-2.31(0.92)**
Parent's Years of Education	0.15(0.11)
Household income	
\$16,000-30,999	-1.91(1.48)
\$31,000-45,999	0.16(1.44)
\$46,000-60,999	2.34(1.49)
\$61,000-75,999	0.93(1.51)
\$76,000-90,999	2.02(1.64)
\$91,000-105,000	1.93(1.52)
\$105,000 and Up	3.53(1.53)**
Enrollment Site	
Pittsburgh	0.52(0.64)
Saint Louis	0.90(1.36)

Note: The table reports regression coefficients and standard errors in parentheses;

* $p < 0.1$,

** $p < 0.05$,

*** $p < 0.01$; reference categories for explanatory variables are as shown in Table 1. An increase in the scores (positive coefficients) indicates an improvement in psychosocial wellbeing.

Table 5
 Regression Results for the Parental ISEL Scale and the Belonging, Self-Esteem, Tangible Items and Appraisal Subscales

Explanatory Variables	ISEL- Full Scale	Belonging	Self Esteem	Tangible Items	Appraisal
<i>Cleft Type</i>					
Cleft Lip Only	-0.19(2.35)	-0.26(0.72)	-0.16(0.60)	0.02(0.64)	0.03(0.70)
Cleft Palate Only	-2.62(3.01)	-0.28(0.93)	-1.27(0.76)*	0.01(0.82)	-1.11(0.90)
<i>Child's Age</i>					
Age <6 years	0.75(2.24)	-0.46(0.68)	-0.26(0.56)	0.28(0.60)	0.84(0.66)
Age 13-17	-2.39(3.82)	-1.44(1.16)	-0.06(0.97)	-1.02(1.03)	0.36(1.13)
Male vs Female Affected Child	4.33(2.13)**	1.04(0.65)	1.05(0.54)	0.63(0.58)	1.75(0.63)***
<i>Father vs Mother</i>					
	-3.14(2.09)	-0.76(0.63)	0.39(0.53)	-0.68(0.56)	-1.70(0.62)***
<i>Married vs Unmarried</i>					
	-2.27(3.31)	-1.22(1.00)	-1.17(0.84)	-0.23(0.89)	-0.16(0.98)
<i>Parent's Years of Education</i>					
	0.22(0.42)	-0.06(0.13)	0.10(0.11)	0.06(0.11)	0.11(0.12)
<i>Household income</i>					
\$16,000-30,999	-6.91(5.22)	-1.42(1.61)	-2.71(1.32)**	-0.67(1.42)	-1.73(1.55)
\$31,000-45,999	6.00(5.04)	2.98(1.55)	0.13(1.28)	1.29(1.37)	1.69(1.50)
\$46,000-60,999	6.26(5.34)	2.08(1.61)	0.31(1.36)	3.19(1.43)**	0.82(1.56)
\$61,000-75,999	6.14(5.35)	3.33(1.63)**	-0.33(1.35)	1.50(1.46)	1.69(1.59)
\$76,000-90,999	10.10(5.82)*	4.27(1.77)**	1.13(1.48)	2.59(1.58)	2.68(1.73)
\$91,000-105,000	8.53(5.41)	3.99(1.65)**	1.20(1.37)	2.54(1.47)*	1.58(1.61)
\$105,000 and Up	14.00(5.43)**	5.09(1.66)***	1.62(1.38)	3.82(1.47)**	3.49(1.61)**
<i>Enrollment Site</i>					
Pittsburgh	-0.75(2.29)	0.01(0.70)	0.12(0.58)	-0.09(0.62)	-0.56(0.68)
Saint Louis	0.42(5.23)	0.34(1.49)	0.70(1.33)	1.47(1.33)	-0.31(1.45)

Note: The table reports regression coefficients and standard errors in parentheses;

* p<0.1,
 ** p<0.05,
 *** p<0.01.

reference categories for explanatory variables are as shown in Table 1. An increase in the scores (positive coefficients) indicates an improvement in psychosocial wellbeing.

**
*
p<=0.05,

Author Manuscript

Author Manuscript

Author Manuscript

Author Manuscript

Table 6

Regression Results for the Parental FNE Scale

Explanatory Variables	FNE Scale
Cleft Type	
Cleft Lip Only	-0.95(2.86)
Cleft Palate Only	1.06(3.80)
Child's Age	
Age <6 years	4.68(2.72)*
Age 13-17	4.95(4.59)
Male vs Female Affected Child	0.59(2.60)
Father vs Mother	-12.71(2.53)***
Married vs Unmarried	7.39(4.06)*
Parent's Years of Education	-0.43(0.51)
Household income	
\$16,000-30,999	9.38(6.62)
\$31,000-45,999	2.48(6.40)
\$46,000-60,999	4.44(6.73)
\$61,000-75,999	10.64(6.77)
\$76,000-90,999	10.27(7.32)
\$91,000-105,000	7.37(6.84)
\$105,000 and Up	1.77(6.90)
Enrollment Site	
Pittsburgh Enrollment Site	-5.80(2.79)**
Saint Louis Enrollment Site	-2.14(6.09)

Note: The table reports regression coefficients and standard errors in parentheses;

* p<0.1,

** p<0.05,

*** p<0.01; reference categories for explanatory variables are as shown in Table 1. An increase in the scores (positive coefficients) indicates a decline in psychosocial wellbeing.