Oral Health-Related Quality of Life in Children with Orofacial Clefts

Thesis

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ABSTRACT

Orofacial clefts affect approximately 6,800 births in the United States each year.

Significant psychological and social burdens have been reported in relation to orofacial clefts. Oral Health Related Quality of Life (OHRQoL) is an extension of HRQoL and measures the impact of oral conditions on the child's physical and social functioning.

Objectives: To determine the impact of orofacial clefts on the OHRQoL of affected children and whether the OHRQoL of children with orofacial clefts differs among different age groups. Also, to assess whether the responses of children with orofacial clefts differ from the caregivers' perceptions of their child's OHRQoL. **Methods:** 75 patients from the Nationwide Children's Hospital Craniofacial Anomalies Clinic, as well as their caregivers, and 75 controls, met our inclusion criteria and completed the Child Oral Health Impact Profile (COHIP), a reliable and valid questionnaire designed to assess self-reported OHRQoL in children and teenagers. **Results:** Children with orofacial clefts had statistically significant lower quality of life scores than control subjects for overall OHRQoL and Functional Well-being. There was a statistically significant difference in the interaction of age group and Social-Emotional Well-being between children with orofacial clefts and control children. No statistically significant differences were found between the responses of children with orofacial clefts and their caregivers' reports for overall OHRQoL, nor for any of the subscales. **Conclusions:** Presence of an

orofacial cleft significantly decreases overall OHRQoL, Functional Well-being, and Social-Emotional Well-being in children and adolescents. The negative impact of orofacial clefts on Social-Emotional Well-being is greater in 15-18 year-olds than in younger age groups. Children with orofacial clefts and their parents were very similar in their evaluations of the child's OHRQoL.

Dedicated to my family

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CHAPTER 1

INTRODUCTION

Orofacial clefts, which include cleft lip with or without cleft palate and isolated cleft palate, affect approximately 6,800 births in the United States each year (Centers for Disease Control and Prevention, 2006). Orofacial clefts are associated with a variety of health complications, including feeding, speech, growth, and physical health problems such as recurrent middle ear infections. Children with orofacial clefts undergo various treatments, including surgical, medical, speech, dental, and other health interventions (Nackashi et al, 2002).

Significant psychological and social burdens have been reported in relation to orofacial clefts. Psychological adjustment is often challenging for children with orofacial clefts due to esthetic concerns, speech and hearing disabilities, and difficulty acquiring the social skills necessary for adjustment (Kapp-Simon, 1986). Children with orofacial clefts also have a higher risk of developmental problems, including cognitive performance (Jocelyn et al, 1996; Jelliffe-Pawlowski et al, 2003). Treatment of orofacial clefts extends throughout adolescence, during which psychological adjustment has been reported to be a problem as well (Kapp-Simon et al, 1992; Thomas et al, 1997).

Health Related Quality of Life (HRQoL) is used to assess the impact of disease severity on a child's physical, psychological, social and emotional well-being. Studies have found that children with visible facial differences have lower quality of life, and that their quality of life is similar to that of children with other chronic disorders (Topolski et al., 2005). Age-related differences among preadolescent children with oral clefts have been reported. Quality of life scores gradually improve for children with isolated cleft palate as they approach adolescence. However, quality of life scores for children with cleft lip decrease as they approach adolescence, when acceptance by their peers becomes more critical (Damiano et al, 2007).

Oral Health Related Quality of Life (OHRQoL) is an extension of HRQoL that more specifically measures the impact of oral disease on the child's physical and social functioning. Children and adolescents have been found to provide reliable information regarding their own OHRQoL when appropriate questionnaire techniques are used although few scales specifically designed for them have been developed (Jokovic et al, 2002; Jokovic et al, 2004; Filstrup et al, 2003). The Child Oral Health Impact Profile (COHIP), a questionnaire designed to assess self-reported OHRQoL in children and teenagers, was found to have excellent reliability and validity (Broder & Wilson-Genderson, 2007). Previous studies have also demonstrated the usefulness of obtaining caregiver reports of the child's QoL in addition to the report completed by the child (Wilson-Genderson et al, 2007).

The literature lacks reports of age-related differences in OHRQoL in adolescents with oral clefts. The purpose of this study is to identify and characterize age-related differences in OHRQoL in children and adolescents with orofacial clefts compared with

an unaffected control group. This study will also compare differences between ratings of children and adolescents with those of their caregivers.

COMPREHENSIVE LITERATURE REVIEW

Epidemiology

Orofacial clefts are the most common craniofacial birth defect (Peterka et al, 2000), occurring in approximately 1 in 700 live births (Mossey and Castillia, 2003). Prevalence at birth of cleft lip with or without cleft palate varies from 3.4 to 22.9 per 10,000 births among 57 registries internationally. Prevalence of isolated cleft palate varies even more, from 1.3 to 25.3 per 10,000 births (Croen et al., 1998). Prevalence of cleft lip with or without cleft palate is increased in China, Japan, and parts of Latin America and decreased in Israel, South Africa, and southern Europe (Croen et al., 1998). In Caucasian populations, cleft lip with or without cleft palate occurs more frequently in males than females by about 2:1 (Mossey and Little, 2002). Cleft palate alone appears more frequently in females than males (Mossey and Little, 2002). Orofacial clefts often occur in conjunction with other major congenital anomalies. This happens more frequently for individuals with cleft palate alone than for those with cleft lip with or without cleft palate (Mossey and Little, 2002). For cleft palate alone, 55% of cases are isolated, 18% are associated with other anomalies, and 27% are part of recognized syndromes (Calzolari et al., 2004). For cleft lip with or without cleft palate, 71% of cases are isolated and 29% are associated with other anomalies (Calzolari et al., 2007).

Developmental Pathogenesis

Orofacial clefts are usually caused by failure of the facial processes to fuse during embryogenesis (Johnson et al., 1990). By the 4th week of embryonic development, cranial neural crest cells have migrated to form the frontonasal prominence, the maxillary processes, and the mandibular processes, which surround the stomatodeum. By the end of the 4th week of embryogenesis, the lower portion of the frontonasal prominence develops into paired mesial and lateral nasal processes surrounding the nasal placodes. The upper lip and primary palate are formed when the medial and lateral nasal processes fuse with the maxillary processes on either side. The lateral nasal processes have accelerated cell division just before these processes are completed. Disruption of growth during this time can cause a failure of primary palate formation (Sperber, 2002).

During the 6th week of embryogenesis, development of the secondary palate begins as paired palatal shelves grow outward from the maxillary process. The palatal shelves initially grow vertically beside the developing tongue, but rise to a horizontal position above the tongue and fuse during the 7th week to form the secondary hard and soft palate. By the 10th week, the secondary palate has fused anteriorly with the primary palate and superiorly with the nasal septum (Sperber, 2002).

Orofacial clefts can occur as a result of teratogenic exposures, single gene disorders, or chromosomal abnormalities and are classified as syndromic when they occur with other congenital defects as one of over 400 described syndromes (Gorlin, 2001). In approximately 50% to 70% of cases there is no recognized pattern of malformation and the cause of the disorder is unknown (Gorlin, 2001). Such cases are classified as

nonsyndromic, and may either be isolated or nonisolated, depending on whether they occur with other congenital defects (Mitchell et al., 2002). Many syndromic clefts have simple Mendelian patterns of inheritance. Isolated clefts, however, are usually genetically complex traits (Lidral and Murray, 2004). Molecular studies have shown that mutations in genes such as *IRF6* (Kondo et al., 2002) and *MSX1* (van den Boogaard et al., 2000; Mossey and Little, 2002) can cause orofacial clefts. In addition to genetic factors, a number of other risk factors have been identified during early pregnancy when the lip and palate are developing. Teratogens such as anticonvulsant drugs and corticosteroids (Dravet et al., 1992; Abrishamchiam et al., 1994; Shaw et al., 1995; Park-Wyllie et al., 2000), as well as maternal smoking (Little et al., 2004; Honein et al., 2007), alcohol use (Romitti et al., 1999; Chevrier et al., 2005; Bille et al., 2007; Meyer et al., 2003; Romitti et al., 2007), and exposure to organic solvents and agricultural chemicals have all been associated with orofacial clefts (Shaw et al., 2003; Gordon and Shy, 1981; Garcia, 1998). Vitamin deficiencies (Wong et al., 1999; Munger et al., 2004; Kapels et al., 2004) and viral infections (Acs et al., 2005) can also increase the risk of orofacial clefts.

Psychosocial Factors

Numerous studies have demonstrated the negative impact that orofacial clefts can cause on the child's psychosocial development. Children with orofacial clefts have lower self-concept scores than children without clefts (Broder and Strauss, 1989). They are also more likely to have learning disabilities, grade retention, and low school achievement (Broder et al., 1998). Children with orofacial clefts are at risk for dyslexia

related to fluency and naming speed (Richman and Ryan, 2003) and 30-40% of them have early reading problems (Richman et al., 2005). Children with orofacial clefts report more problems with social well-being when compared to children with dental caries (Locker et al., 2005). Adolescents with orofacial clefts often demonstrate social inhibition (Kapp-Simon and McGuire, 1997). Low social competence in teens with clefts can lead to feelings of loneliness and social anxiety (Pope and Ward, 1997). Males with cleft lip and palate are more likely to have a midline brain anomaly and lower IQ than those without clefts (Nopoulos et al., 2001). Males with orofacial clefts are likely to have a smaller orbitofrontal cortex, which is associated with lower social functioning scores (Nopoulos et al., 2005).

Health-Related Quality of Life

The World Health Organization (1998, p.17) defines quality of life as an “individual’s perception of their position in life in the context of the culture and value system where they live, and in relation to their goals, expectations, standards, and concerns.” Health-related quality of life (HRQoL) is a patient-reported outcome that helps clinicians assess how patients are affected by their illness or condition by quantifying a child’s or parent’s perspective of the child’s functional and psychosocial well-being. Measuring HRQoL is becoming an increasingly important mechanism for evaluating different aspects of disease management. Children with a chronic disease and poor HRQoL (per parental report) had average monthly healthcare costs that were twelve times higher than children who had a chronic disease but better HRQoL (Seid et al., 2004). Hence, HRQoL may be used to identify patients that could benefit from

different treatment modalities or more aggressive preventative care. Increasing their HRQoL could potentially decrease their overall healthcare costs. HRQoL can also help clinicians make more informed treatment recommendations as they are able to better understand the patient's perspective and how the disease impacts their life (Ralstrom et al., 2009).

HRQoL in Children with Orofacial Clefts

Recent HRQoL studies have improved our understanding of how orofacial clefts affect children. Children with visible facial differences have lower quality of life scores compared to children with no facial differences, but when compared to children with chronic disorders, their scores are similar, and slightly better in the area of family relationships. (Topolski et al., 2005). Children with orofacial clefts showed few differences in HRQoL scores, oral symptoms, and emotional well-being when compared to children with dental caries (Locker et al., 2005). Health professionals who work with children with orofacial clefts report these children have a slightly lower HRQoL (Wehby et al., 2006).

Adults with cleft lip and palate had significantly lower HRQoL scores in the subscales social functioning and emotional role, but not in the subscales physical functioning, physical role, bodily pain, general health, vitality, and mental health (Sinko et al., 2004). Adults with cleft lip and palate who desired further treatment had significantly lower HRQoL scores compared to those who did not desire further treatment in the subscales physical role and vitality, and trends toward significant differences ($p <$

.10) in the subscales bodily pain, social functioning, and general mental health (Sinko et al., 2004).

One study (Damiano et al., 2007) used the Pediatric Quality of Life (PedsQL) survey and telephone interviews with parents to determine HRQoL in pre-adolescent (2-12 years old) children with nonsyndromic oral clefts. This study found that children with cleft lip and/or palate scored significantly lower than healthy children on the psychosocial health component scale. They found no difference on any of the PedsQL scales when comparing children according to type of cleft. Higher household income was associated with higher total quality of life scores, as well as with high physical and psychosocial scores. A higher number of household members, a higher health status rating, and the mother's being married were also associated with higher scores. The parent's perception of how happy the child was with his or her facial appearance was associated with higher total and psychosocial health scores. Quality of the child's speech was highly correlated with the total, physical, and psychosocial health rating of the child. After controlling for demographics, children with less severe speech problems had higher overall HRQoL as well as higher physical and psychosocial health domain scores. Children with cleft palate had significantly higher HRQoL than children with cleft lip and palate. Both categories of younger children (ages 2-4 and 5-7) had higher HRQoL than children who were 8-12 years old. The HRQoL was better among children with cleft lip and cleft lip and palate than among those with cleft palate until age 7, then declined for 8-12 year olds. Children with cleft palate had better HRQoL observed in the oldest age group. This suggests that the difference in outward appearance related to lip involvement has greater relative importance as the child gets closer to adolescence.

Another study (Warschausky et al., 2002) examined parents' perceptions of HRQoL in children ages 5 to 18 years with cleft and other craniofacial anomalies using the Child Health Questionnaire version PF28. In this study, parents reported that children with cleft lip and/or palate had physical and psychosocial scale scores that fell within normal limits. Parents had fewer concerns about global health status for the children with cleft lip and/or palate than for children with other craniofacial anomalies. There were no significant associations between either age or sex and physical or psychosocial health. Their findings suggest that there is a significant association between parentally perceived physical health and psychosocial adjustment in children with craniofacial anomalies.

A study using the KINDL, a German questionnaire for measuring HRQoL, found that 5-6 year-old children with orofacial clefts had scores that were lowest in the dimension of self-esteem. In all dimensions, the KINDL scores of the children were higher than those of the parents suggesting a superior HRQoL than the caregivers had estimated (Kramer et al., 2008).

Oral Health-Related Quality of Life

The impact of oral health on quality of life is usually referred to as oral health-related quality of life (OHRQoL). OHRQoL is recognized as a multidimensional concept, comprising both the presence or absence of oral disease as well as psychological aspects of oral health (Gift et al., 1997). Two major survey instruments have been developed to measure OHRQoL in children: the Child Perceptions Questionnaire (CPQ) and the Child Oral Health Impact Profile (COHIP).

The CPQ was developed to assess children's perception of the impact of oral disorders on physical and psychosocial functioning. The CPQ(8-10) and CPQ(11-14) were designed for children of different age groups according to cognitive ability and lifestyle. Different studies have shown mixed results regarding the CPQ's ability to detect OHRQoL differences between children with orofacial clefts and controls. In one study (Jokovic et al., 2002), the CPQ(11-14), designed for 11- to 14-year-old children, was found to be valid and reliable and able to discriminate OHRQoL differences between children with orofacial clefts versus those in the orthodontic and pediatric dentistry groups. In another study (Jokovic et al., 2004), the CPQ(8-10), designed for 8- to 10-year-olds, detected a slightly higher level of impact in the orofacial than in the pediatric dentistry group, but overall did not demonstrate discriminative validity. Similarly, Danish versions of CPQ(8-10) and CPQ(11-14) had limited ability to discriminate between children with cleft lip and palate and healthy children (Wogelius et al., 2009).

Child Oral Health Impact Profile

The Child Oral Health Impact Profile (COHIP, Appendix 1) is a 38 question survey designed to measure self-reported Oral Health Related Quality of Life (OHRQoL) in school age children ages 8-15 years. Its readability score has been assessed at a 3.5 grade reading level, making it suitable for children in third grade or higher. The COHIP contains 34 items to measure OHRQoL in five domains: Oral Health, Functional Well-Being, Social-Emotional Well-Being, School Environment and Self Image. It also contains 4 items to assess Treatment Expectations and Global Health of the child. The

questionnaire contains items to assess both positive and negative aspects of OHRQoL, following the World Health Organization concept that health is more than the absence of disease (Broder, 2007; Broder and Wilson-Genderson, 2007; Broder et al., 2007)

Reliability and validity of the COHIP were established by Broder et al. in a study of 523 children of diverse ethnicities recruited from pediatric dentistry clinics, orthodontic clinics, craniofacial clinics and elementary schools (Broder and Wilson-Genderson, 2007). It was found that the COHIP had excellent scale reliability and test-retest reliability (i.e., there was no significant change in COHIP scores over time.) The survey's discriminate validity was supported by the finding of significant differences in COHIP scores between the different clinical groups, with the craniofacial patients reporting lower OHRQoL than pediatric patients, orthodontic patients or non-clinical subjects. Within the orthodontic group, children with more severe orthodontic problems were also found to have lower COHIP scores. Hence, the COHIP was sensitive to differences between groups as well as to differences in disease severity for a single clinical condition (Broder and Wilson-Genderson, 2007). Convergent validity testing found that a significant positive relationship between Global Health and COHIP scores existed. The acceptable reliability and validity demonstrated preliminary support for the COHIP as a useful psychometric tool (Slade and Reisine, 2007). The COHIP has not been extensively used in large scale epidemiological studies or clinical trials to confirm its sensitivity, specificity or clinical utility (Broder, 2007; Broder and Wilson-Genderson, 2007).

Craniofacial Patients and COHIP

Broder and Wilson-Genderson (2007) compared COHIP scores among children recruited from pediatric, orthodontic, and craniofacial clinics. Their sample included 157 pediatric dentistry patients, 152 orthodontic patients, 110 patients with craniofacial anomalies, and 104 community-based participants. They found that the craniofacial group reported greater negative impact on their OHRQoL than either the general pediatric or the orthodontic patients. The craniofacial patients had significantly lower scores for Overall COHIP, as well as the subscales Functional well-being and School environment. The craniofacial group also had lower scores which approached significance for the subscale Social/emotional well-being.

Caregiver vs. Child Responses and COHIP

Recent studies have suggested that, when possible, both child and caregiver should be asked to provide ratings of OHRQoL in an effort to provide a more well-rounded depiction of the child's health care needs and quality of life issues (Wilson-Genderson et al., 2007). Although it is easy to argue that the child's opinion is the most valuable, there are certain factors which may impact the reliability and validity of a child's HRQoL responses. Some of these factors include dominance of short-term memory, strong influence of recent incidents, lack of a fully developed long-term perspective, language problems during interviews, and reading problems when completing a written questionnaire (Wilson-Genderson et al., 2007; Vogels et al., 1998; Eiser and Morse, 2001a).

Caregivers generally have low to modest agreement with the child's rating (Eiser and Morse, 2001b; Achenbach et al., 1987). The importance of facial appearance, time away from school, and symptomology may be over- or under-estimated by caregivers (Eiser and Morse, 2001b). Caregivers are also likely to have biases and expectations which can impact the QoL rating (Eiser and Morse, 2001b). When children have chronic or extreme health conditions, caregiver QoL assessments tend to report more negative impact than that reported by the child (Ennett et al., 1991; Garber et al., 1998).

Bos et al. (2010) used a Dutch version of COHIP to compare reports of OHRQoL by orthodontic patients (age 8-15) and their parents. They found only one significant though small difference on the subscale Oral Symptoms. Overall they found good agreement between parent and child reports and suggested that parents are suitable alternatives to their children in surveys measuring OHRQoL.

Wilson-Genderson et al. (2007) also assessed child-caregiver concordance in children aged 8-15 and their parents using COHIP. Their sample included 141 pediatric dentistry patients, 135 orthodontic patients, and 100 craniofacial patients and their caregivers. They found low to modest rates of agreement between child and caregiver for the sample overall. Craniofacial patients tended to rate their OHRQoL higher than their caregivers' responses. Pediatric and orthodontic patients, on the other hand, were more likely to agree with their caregivers' responses or rate their OHRQoL lower.

Age-Related Differences in OHRQoL

Broder and Wilson-Genderson's study (2007) involved children of age 8-15. They found that the youngest children (age 8) had lower scores for the subscale Functional Well-being than some of the older children (ages 11 and 12), but cautioned that this finding may be due to the small number of 8-year-olds in the sample. They also found that the oldest children (age 15) had lower scores on functional well-being than some of the younger children (ages 11-13), and reasoned that older children had experienced untreated disease for longer than the younger children. These age-related differences were found in a post hoc examination of scores for all four groups (pediatric dentistry, orthodontic, craniofacial, and community) combined. This study did not discuss age-related differences within the craniofacial group alone. The literature lacks data on age-related differences in OHRQoL in adolescents with orofacial clefts.

As healthcare providers come to a greater understanding of how orofacial clefts impact children of different ages, they will be better able to provide treatment that meets the individual needs of those children. Evaluation of OHRQoL response agreement between children and their caregivers can also be helpful as healthcare providers seek methods of estimating the OHRQoL of patients who are either mentally or physically unable to complete a questionnaire.

OBJECTIVES

1. Assess the impact of oral health in the quality of life of children with cleft lip and/or palate;
2. Evaluate whether OHRQoL of children with orofacial clefts differs among different age groups;
3. Determine whether the parents' perceptions of OHRQoL differ from their children's responses.

NULL HYPOTHESES

- H₀1. Children with cleft lip and/or palate do not differ in oral health-related quality of life (OHRQoL) compared to healthy subjects.
- H₀2. There are no age-related differences in OHRQoL among patients with orofacial clefts.
- H₀3. Caregivers' perceptions of OHRQoL do not differ from their children's responses

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CHAPTER 2

MATERIALS AND METHODS

Study Approval

This study was approved by the Nationwide Children's Hospital Institutional Review Board (study ID #IRB09-00396). Funding for this study was provided by Delta Dental Foundation.

Selection of Subjects

To be eligible to enroll in this research project, patients had to meet the following inclusion and exclusion criteria:

Inclusion/Exclusion Criteria

i.) Cleft Group

1. Be between 8 and 18 years of age
2. Be diagnosed with a craniofacial anomaly
3. Be able to speak and read English
4. Not be pregnant
5. Not have a medically handicapping condition

6. Not be enrolled in a special class for mental disabilities
 7. Not have a mental disorder
- ii.) Control Group
1. Be between 8 and 18 years of age
 2. Have no significant medical history
 3. Be able to speak and read English
 4. Not be pregnant
 5. Not have a medically handicapping condition
 6. Not be enrolled in a special class for mental disabilities
 7. Not have a mental disorder

Data Collection

The sample size calculation indicated 75 subjects needed to be enrolled in both the study group and the control group. A convenience sample of 80 patients was recruited in the Nationwide Children's Hospital Craniofacial Anomalies Orthodontic Clinic (study group) and 32 patients were recruited in the Pediatric Primary Care Clinic (control group). Additionally, data from 57 patients recruited in the Adolescent Medicine Clinic during a previous study (Ralstrom, 2009) were also added to the control group. All subjects were recruited between March 1, 2009 and June 15, 2010. Parental consent and patient assent were obtained by an orthodontic or pediatric dental resident or research assistant after identifying patients who met eligibility criteria. After consenting to participate, subjects were assigned a number by which they were identified throughout the study in order to protect their confidentiality.

After assenting to participate, the child subjects completed the 38 question COHIP (Appendix 1). Caregivers of subjects in the cleft group also completed a 38 question COHIP (Appendix 2) re-worded for parents. Participants were given a gift card to compensate them for their time. As the survey has not been tested in foreign languages, only subjects who were able to read and speak English were eligible to participate. The following data was recorded for each subject and entered into a spreadsheet for statistical analysis:

1. Age
2. Gender
3. Race
4. Highest Education Level Attained in Household
5. Craniofacial Diagnosis
6. Orthodontic Treatment History
7. Medical Diagnosis
8. Current Medications
9. Responses to the survey

Scoring of COHIP

The COHIP (Appendix 1) consisted of 34 items forming 5 conceptually distinct subscales: *Oral Health*, *Functional Well-Being*, *Social/Emotional Well-Being*, *School Environment and Self-Image* (Broder and Wilson-Genderson, 2007). It also contains 2 items to measure *treatment expectancy* and 2 items to measure *global health*. Global health and treatment expectancy items are not used in computing the overall COHIP

score. The complete list of items is found in Appendix A. Below is a summary of the items and subscales (used with permission from Ralstrom, 2009).

- (1) ***Oral Health***: Items 1, 2, 3, 4, 5, 6, 7, 8, 9, and 10 measure specific oral symptoms that are not necessarily related to one another (e.g., pain, spots on teeth).
- (2) ***Functional Well-Being***: Items 11, 15, 20, 23, 25, and 27 relate to the child's ability to carry out specific everyday tasks or activities (e.g., speaking clearly, chewing).
- (3) ***Social-Emotional Well-being***: Items 12, 16, 17, 19, 22, 24, 28, and 29 pertain to peer interactions and mood states.
- (4) ***School-Environment***: Items 13, 18, 21, and 30 pertain to tasks associated with the school environment.
- (5) ***Self-image***: Items 14, 26, 31, 32, 33, and 34 address positive feelings about self.
- (6) ***Treatment Expectancy***: Items 35 and 36 measure expectations regarding the treatment process and outcomes.
- (7) ***Global Health***: Items 37 and 38 assess overall feelings about oral and systemic health.

The statements are formatted to elicit self-reports from the subject. Responses for the 7 positively-worded items are recorded as 'never' = 0, 'almost never' = 1, 'sometimes' = 2, 'fairly often' = 3, and 'almost all of the time' = 4. The positively scored items were:

- (1) *Item 14.* Been confident
- (2) *Item 26.* Felt you were attractive
- (3) *Item 31.* Have good teeth
- (4) *Item 32.* Believe you will have good teeth
- (5) *Item 33.* Believe you will have good health
- (6) *Item 34.* Feel good about yourself
- (7) *Item 35.* Will feel better when treatment is completed

Scoring of the remaining 31 negatively-worded items were reversed (0 = ‘almost all of the time’= 0, ‘fairly often’= 1, ‘sometimes’= 2, ‘almost never’= 3, ‘never’= 4).

Higher COHIP scores reflect more positive OHRQoL while lower scores reflect lower OHRQoL. Subscales scores are calculated by summing the responses of the items specific to the subscale (Broder and Wilson-Genderson, 2007). The overall OHRQoL score is computed by summing the subscales scores. Treatment expectation scores and the overall health response are not included in the overall COHIP scale since these items are relevant only when the COHIP is used as part of a treatment assessment. Scores range from 0-140 for the overall scale. If more than two-thirds of the items in a subscale are missing, the subscale and the overall score are set to missing. If fewer items are missing for a subscale, the average of available items used and the sum of the subscale is calculated. For this project, the questionnaire was transformed to a paper format that could be digitally scanned and processed using an HP Scanner and Cardiff TeleForm software. The responses were then checked manually for errors in scanning and/or data entry.

Statistical Analysis

Statistical analysis was performed using SAS 9.1.3. A significance level of $\alpha = .05$ was established. Descriptive statistics (frequency, mean, median, standard deviation) for the collected variables were estimated. The data from the COHIP survey was normally distributed so parametric tests were used for analysis.

The main outcomes of this cross-sectional study compared the OHRQoL measured by the COHIP between subjects with orofacial clefts and control subjects, between cleft subjects of different age groups, and between cleft subjects and their caregiver's responses. Overall OHRQoL and subscale scores were compared between groups using the Generalized Linear Model (GLM) Procedure, an analysis of variance (ANOVA). Responses between caregivers and children were compared using the Mixed Procedure, a repeated measures ANOVA. Least Squares Means analysis was used to compare scores of cleft and control subjects in different age groups. Age groups were selected according to other OHRQoL studies involving children with orofacial clefts (Jokovic et al., 2004; Locker et al., 2005).

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CHAPTER 3

MANUSCRIPT

Oral health-related quality of life in children with orofacial clefts

ABSTRACT

Objectives: To determine the impact of orofacial clefts on the oral health-related quality of life (OHRQoL) of affected children and whether the OHRQoL of children with orofacial clefts differs among different age groups. Also, to assess whether the responses of children with orofacial clefts differ from the caregivers' perceptions of their child's OHRQoL.

Design: Cross-sectional study.

Patients/Setting: 75 patients with cleft lip and/or cleft palate (mean age 13.0 y.o.) from the Nationwide Children's Hospital Craniofacial Anomalies Clinic, as well as their caregivers, and 75 control patients (mean age 13.9 y.o.).

Main Outcome Measure(s): Self-reported OHRQoL measured with the Child Oral Health Impact Profile (COHIP), a reliable and valid questionnaire designed for use with children and teenagers. Higher scores indicate better quality of life.

Results: Children with orofacial clefts had statistically significant lower quality of life scores than control subjects for overall OHRQoL and Functional Well-being. There was a statistically significant difference in the interaction of age group and Social-Emotional

Well-being between children with orofacial clefts and control children. No statistically significant differences were found between the responses of children with orofacial clefts and their caregivers' reports for overall OHRQoL, nor for any of the subscales.

Conclusions: Presence of an orofacial cleft significantly decreases overall OHRQoL, Functional Well-being, and Social-Emotional Well-being in children and adolescents.

The negative impact of orofacial clefts on Social-Emotional Well-being is greater in 15-18 year-olds than in younger age groups. Children with orofacial clefts and their parents had very similar evaluations of the child's OHRQoL.

Key Words: children, adolescents, cleft lip and palate, oral health-related quality of life, COHIP

INTRODUCTION

Orofacial clefts occur in approximately 1 in 700 live births (Mossey and Castillia, 2003). In the United States, orofacial clefts affect 6,800 births each year (Centers for Disease Control and Prevention, 2006). Orofacial clefts can be subdivided into cleft lip with or without cleft palate, and isolated cleft palate because of the separate and distinct developmental origin of the lip and primary palate versus that of the secondary palate (Sperber, 2002). Orofacial clefts are associated with a variety of health complications, including feeding, speech, growth, and physical health problems such as recurrent middle ear infections. Children with orofacial clefts undergo multiple treatments, including surgical, medical, speech, dental, and other health interventions (Nackashi et al, 2002).

Functional difficulties associated with orofacial clefts include problems with speech, eating, and oral hygiene. Such difficulties are attributed to clinical findings such as missing and malpositioned teeth, constricted maxillary arch form, skeletal discrepancies, oronasal fistulas, and velopharyngeal incompetence (Gorlin et al., 2001).

Significant psychological and social burdens have been reported in relation to orofacial clefts. Psychological adjustment is often challenging for children with orofacial clefts, most likely due to esthetic concerns, speech and hearing disabilities, and difficulty acquiring the social skills necessary for adjustment (Kapp-Simon, 1986). Children with orofacial clefts also have a higher risk of developmental problems (Jocelyn et al, 1996; Jelliffe-Pawlowski et al, 2003). Treatment of orofacial clefts extends throughout adolescence, during which psychological adjustment has also been reported to be a problem (Kapp-Simon et al, 1992; Thomas et al, 1997).

Many studies have found problems with psychological well-being in children and adolescents with orofacial clefts. Such problems include lower self-concept scores (Broder and Strauss, 1989), learning disabilities, grade retention, and low school achievement (Broder et al., 1998). Children with orofacial clefts are also at risk for dyslexia (Richman and Ryan, 2003) and early reading problems (Richman et al., 2005). In addition, these children report more problems with social well-being compared to controls (Locker et al., 2005). Adolescents with orofacial clefts often demonstrate social inhibition (Kapp-Simon and McGuire, 1997). Low social competence in teens with orofacial clefts can lead to feelings of loneliness and social anxiety (Pope and Ward, 1997).

Health-related quality of life (HRQoL) is an outcome that helps health care professionals evaluate how disease, complications, and treatment affect the patient. Measures of HRQoL quantify a child's or parent's perception of the child's functional and psychosocial well-being and how they are affected by the child's illness and treatment. HRQoL measures can help clinicians to make better treatment recommendations as they come to understand the burden of disease from the patient's perspective (Ralstrom et al., 2009).

Recent HRQoL studies have increased our understanding of how orofacial clefts affect children. Topolski et al. (2005) found that children with visible facial differences have lower quality of life (QoL) scores compared to children with no chronic conditions. When compared to children with chronic disorders, their scores are similar, and slightly better in the area of family relationships. Damiano et al. (2007) found that children (2-12 years old) with cleft lip and/or palate scored significantly lower than healthy children on the psychosocial health component scale. Quality of the child's speech was highly correlated with the total, physical, and psychosocial health rating of the child. Other studies (Locker et al., 2005; Warschausky et al., 2002) have found orofacial clefts cause less of an impact on HRQoL in children.

Oral Health Related Quality of Life (OHRQoL) is an extension of HRQoL that specifically measures the impact of oral disease on the child's physical and social functioning. Children and adolescents provide reliable information regarding their own OHRQoL when appropriate questionnaire techniques are used although few scales specifically designed for them have been developed (Jokovic et al, 2002; Jokovic et al, 2004; Filstrup et al, 2003). The Child Oral Health Impact Profile (COHIP), a

questionnaire designed to assess self-reported OHRQoL in children and teenagers, was found to have excellent reliability and validity (Broder & Wilson-Genderson, 2007). Research has also demonstrated the usefulness of obtaining caregiver reports of the child's OHRQoL in addition to the report completed by the child (Wilson-Genderson et al, 2007).

Broder and Wilson-Genderson (2007) used COHIP to compare OHRQoL among children recruited from pediatric, orthodontic, and craniofacial clinics. They found that the craniofacial group reported greater negative impact on their OHRQoL than either the general pediatric or the orthodontic patients. The craniofacial patients had significantly lower scores for the overall COHIP, the Functional Well-being subscale, and the School Environment subscale.

There are few studies of age-related differences among children with orofacial clefts. Damiano et al. (2007) reported that QoL scores improve over time for children with isolated cleft palate as they approach adolescence. These children had no facial scars, although speech was most likely affected. These authors also reported that QoL scores for children with cleft lip decrease as they approach adolescence, when acceptance by their peers becomes more critical. However, the QoL survey used was not an OHRQoL survey and the study did not include adolescent children. Broder and Wilson-Genderson (2007) reported age-related differences in OHRQoL in their study of 8- to 15-year-olds. These differences were found in the combined scores for pediatric, orthodontic, craniofacial, and community patients. This study did not discuss age-related differences within the craniofacial group alone. There is a need for more studies of age-related differences in OHRQoL in children and adolescents with orofacial clefts.

The purpose of this study was to: 1. Assess the impact of oral health in the quality of life of children and adolescents with cleft lip and/or palate; 2. Evaluate whether OHRQoL of children and adolescents with orofacial clefts differs among different age groups; and 3. Determine whether the caregiver's perception of their child's OHRQoL differs from the child's perception.

MATERIALS AND METHODS

Participants / Subjects

To participate in the study, subjects had to be between 8 and 18 years of age and able to speak and read English. Exclusion criteria included pregnancy, having a medically handicapping condition, being enrolled in a special class for mental disabilities, or having a mental disorder. Additionally, participants of the cleft group had to be diagnosed with cleft lip and/or cleft palate. Participants of the control group were excluded if they had a significant medical history or craniofacial birth defect.

A sample size calculation, using 80% power and alpha of 0.05, estimated that 75 subjects for both the cleft group and the control group would be required to detect a difference of 13.30 points on the overall COHIP score. Differences of similar magnitude could be detected for the subgroup analyses of the COHIP. This calculation was based on results of a previous study using COHIP (Broder & Wilson-Genderson, 2007).

Eighty patients were recruited in the Nationwide Children's Hospital Craniofacial Anomalies Orthodontic Clinic (cleft group) and 32 patients were recruited in the Pediatric

Primary Care Clinic (control group). Additionally, data from 57 patients recruited in the Adolescent Medicine Clinic during a previous study (Ralstrom et al., 2009) were also added to the control group. Of the craniofacial subjects, one was excluded because he did not have an orofacial cleft, two were excluded because of mental disorders, one was excluded due to missing responses, and one was excluded due to age. Of the control subjects, eight were excluded due to mental disorders, one was excluded due to missing answers, and five were excluded due to age. A total of 75 cleft subjects and their parents, as well as 75 control subjects, met our inclusion and exclusion criteria.

Data Collection

The study was approved by the Institutional Review Board at Nationwide Children's Hospital. Caregivers consented to participate themselves and to have their child participate. Child subjects assented to participate and then completed the 38-item COHIP questionnaire. The accompanying caregiver of the subjects with orofacial clefts also completed a 38-item COHIP re-worded for caregivers. COHIP is designed to measure self-reported OHRQoL in children ages 8-15 years. Its readability score has been assessed at a 3.5 grade reading level, making it suitable for children in third grade or higher. The COHIP contains 34 items to measure OHRQoL in five domains: Oral Health, Functional Well-Being, Social-Emotional Well-Being, School Environment and Self Image. It also contains 4 items to assess Treatment Expectations and Global Health, which are not included in computing the overall COHIP score. The questionnaire contains items to assess both positive and negative aspects of OHRQoL, following the

World Health Organization concept that health is more than the absence of disease (Broder, 2007; Broder and Wilson-Genderson, 2007; Broder et al., 2007). The subscales are summarized in Table 3.1 (used with permission from Ralsrom, 2009).

COHIP Subscale	# of Items	Explanation
Oral Health	10	Measures specific oral symptoms that are not necessarily related to one another (e.g. pain, spots on teeth).
Functional Well-Being	6	Relates to the child's ability to carry out specific everyday tasks or activities (e.g. speaking clearly, chewing).
Social-Emotional Well-Being	8	Pertains to peer interactions and mood states
School Environment	4	Pertains to tasks associated with the school environment
Self Image	6	Addresses positive feelings about self.
Treatment Expectancy*	2	Measures expectations regarding the treatment process and outcomes.
Global Health*	2	Assesses overall feelings about oral and systemic health.

*Not included in calculation of Overall OHRQoL

Table 3.1 Summary of COHIP Subscales

Higher COHIP scores reflect more positive OHRQoL. Subscale scores are calculated by summing the responses of the items specific to the subscale and subscales are summed for the overall OHRQoL score (Broder and Wilson-Genderson, 2007). Treatment expectation scores and global health responses are not included in the overall COHIP scale since these items are relevant only when the COHIP is used as part of a treatment assessment. Scores can range from 0-140 for the overall COHIP scale. If more than two-thirds of the items in a subscale are missing, the subscale and the overall score are set to missing. If fewer items are missing for a subscale, the mean score of responses to scored items for the subscale is used. For this project, the questionnaire was

transformed to a paper format that could be digitally scanned and processed using an HP Scanner (Hewlett-Packard Company, Palo Alto, California) and Cardiff TeleForm software (New England Survey Systems, Brookline, Massachusetts). The responses were then checked manually for errors in scanning and/or data entry.

Statistical Analysis

Statistical analysis was performed using SAS Version 9.1.3 (SAS Institute Inc., Cary, North Carolina). A significance level of $p < .05$ was established. Parametric statistics were used to analyze the data for comparison to other COHIP studies (Broder & Wilson-Genderson, 2007; Ralstrom, 2009).

Overall OHRQoL and subscale scores were compared between groups using the Generalized Linear Model (GLM) Procedure, an analysis of variance (ANOVA). Responses between caregivers and children were compared using the Mixed Procedure, a repeated measures ANOVA. The Least Squares Means analysis was used to compare the scores of cleft and control subjects in different age groups. Age groups were selected consistent with other OHRQoL studies involving children with orofacial clefts (Jokovic et al., 2004; Locker et al., 2005).

RESULTS

The demographic data are summarized in Table 2. The cleft group consisted of 48 (64%) males and 27 (36%) females. The control group had 29 (39%) males and 46 (61%) females. The majority of subjects in the cleft group were Caucasian (81%),

whereas the majority of the subjects in the control group were African American (71%). The cleft group had more subjects in the 11-14 age group 47%, whereas the majority of control subjects were in the 15-18 age group (53%).

	Cleft Group		Control Group	
	<i>n</i>	%	<i>n</i>	%
Gender				
Male	48	64%	29	39%
Female	27	36%	46	61%
Race/Ethnicity				
African Amer.	12	16%	53	71%
Asian	2	3%	1	1%
Caucasian	61	81%	19	25%
Hispanic	0	0%	2	3%
Middle Eastern	0	0%	0	0%
Other	0	0%	0	0%
Age Group				
8-10	22	29%	20	27%
11-14	35	47%	15	20%
15-18	18	24%	40	53%

Table 3.2 Demographic Characteristics

Table 3 summarizes the distribution of subjects according to cleft classification. The majority of cleft subjects had cleft lip and palate (28% bilateral and 33% unilateral). Less than one-fourth of the subjects had cleft lip only (4% bilateral and 19% unilateral). The remainder had either cleft palate (9%) or soft palate cleft (7%).

Cleft Classification	<i>n</i>	%
Bilateral Cleft Lip and Palate	21	28%
Unilateral Cleft Lip and Palate	25	33%
Bilateral Cleft Lip only	3	4%
Unilateral Cleft Lip only	14	19%
Cleft Palate only	7	9%
Soft Palate Cleft	5	7%

Table 3.3 Distribution of Subjects by Cleft Classification

Table 4 shows a comparison of overall and subscale scores for the cleft group and control group. There was a statistically significant difference between groups for the Overall COHIP ($p=0.012$), Functional Well-being ($p=0.001$), and Social-Emotional Well-being ($p=0.022$). The difference for School Environment approached significance ($p=0.070$).

COHIP Item	Cleft (n=75)		Control (n=75)		p-Value
	Mean	(STD)	Mean	(STD)	
Overall COHIP	95.6	(18.3)	108.6	(16.9)	0.012*
Oral Health Well-being	24.8	(5.9)	26.9	(6.5)	0.34
Functional Well-being	17.7	(4.7)	20.9	(3.6)	0.0011*
Social-Emotional Well-being	24.3	(6.7)	27.7	(6.1)	0.022*
School Environment	13.1	(3.0)	14.7	(2.0)	0.070
Self Image	15.7	(4.3)	18.4	(4.9)	0.17
Treatment Expectancy	5.1	(1.4)	5.0	(1.4)	0.19
Global Health	3.1	(0.8)	3.1	(0.9)	0.60
Global Health - Oral	2.8	(0.9)	2.9	(1.0)	0.53

* $p<0.05$

Table 3.4 Comparison of Overall and Subscale Scores for Cleft vs. Control Groups

A comparison of COHIP scores between age groups for the cleft subjects is shown in table 5. There were no statistically significant differences between age groups for overall OHRQoL, nor for any of the OHRQoL subscales. There was, however, a statistically significant difference in treatment expectancy, where the 15-18 age group had a slightly higher treatment expectancy than the younger age groups ($p=0.02$). There was also a statistically significant interaction between age group and subscale scores for Social-Emotional Well-being when comparing children with orofacial clefts to control children. This interaction is summarized in figure 1. No significant differences in Social-Emotional Well-being were found between cleft subjects in different age groups or between control subjects in different age groups. There were also no significant differences between cleft and control subjects in the 8-10 and 11-14 age groups. However, in the 15-18 age group, the cleft subjects had a significantly lower Social-Emotional Well-being than control subjects.

COHIP Item	age 8-10 (n=22)		age 11-14 (n=35)		age 15-18 (n=18)		p-Value
	Mean	(STD)	Mean	(STD)	Mean	(STD)	
Overall COHIP	93.3	(20.3)	98.6	(15.1)	92.7	(21.6)	0.71
Oral Health Well-being	25.5	(6.0)	25.2	(5.9)	23.5	(5.9)	0.58
Functional Well-being	15.9	(5.1)	18.7	(3.8)	18.0	(5.1)	0.23
Social-Emotional Well-being	24.0	(7.8)	25.3	(5.5)	22.6	(7.6)	0.92
School Environment	12.5	(4.1)	13.6	(1.6)	12.8	(3.4)	0.48
Self Image	15.6	(4.4)	15.8	(4.3)	15.8	(4.4)	0.86
Treatment Expectancy	5.0	(1.6)	4.9	(1.5)	5.5	(1.2)	0.02*
Global Health	3.3	(0.7)	3.1	(0.9)	3.0	(0.8)	0.85
Global Health - Oral	3.2	(0.8)	2.7	(0.9)	2.5	(0.9)	0.39

* $p<0.05$

Table 3.5 Comparison of Overall and Subscale COHIP scores between age groups for cleft subjects

*p<0.05

Figure 3.1 Social-Emotional Well-being Scores according to Age Group

Table 6 is a comparison of overall and subscale scores for the cleft subjects and their caregivers. We did not find any statistically significant differences between the responses of the children with orofacial clefts and their caregivers' reports for overall OHRQoL, nor for any of the subscales. Also, no statistically significant differences in COHIP scores were found according to cleft classification, race, or gender.

COHIP Item	Child (n=75)		Caregiver (n=75)		p-Value
	Mean	(STD)	Mean	(STD)	
Overall COHIP	95.6	(18.3)	90.2	(19.0)	0.36
Oral Health Well-being	24.8	(5.9)	23.9	(6.2)	0.38
Functional Well-being	17.7	(4.7)	16.1	(4.6)	0.56
Social-Emotional Well-being	24.3	(6.7)	21.5	(7.8)	0.49
School Environment	13.1	(3.0)	13.2	(2.7)	0.29
Self Image	15.7	(4.3)	15.5	(4.3)	0.20
Treatment Expectancy	5.1	(1.4)	5.3	(1.5)	0.98
Global Health	3.1	(0.8)	3.4	(0.8)	0.60
Global Health - Oral	2.8	(0.9)	2.6	(1.0)	0.11

Table 3.6 Comparison of Overall and Subscale Scores for Cleft Child vs. Caregiver Groups

DISCUSSION

The results of this study indicate that orofacial clefts have a significant impact on OHRQoL in children. Our results confirm that children with orofacial clefts had significantly lower OHRQoL than control children for the Overall COHIP score and for the Functional Well-being and Social-Emotional Well-being subscales.

These results are consistent with the findings of other studies. Broder and Wilson-Genderson (2007) reported that craniofacial patients had significantly lower Overall COHIP and Functional Well-being scores compared to orthodontic and pediatric patients. These investigators also reported that differences in Social-Emotional Well-being scores approached significance. Additionally, Broder and Wilson-Genderson reported that craniofacial patients had a significantly lower score for School Environment. Jokovic et al. (2002) reported lower OHRQoL in 11-14 year-old children with orofacial clefts using another OHRQoL questionnaire and Topolski et al. (2005)

reported children with visible facial differences have lower quality of life score compared with control children.

In contrast, some studies (Jokovic et al., 2004; Wogelius et al., 2009; Locker et al., 2005; Warschausky et al., 2002) have found orofacial clefts have less of an impact on HRQoL in children. These findings may be due to differences in sample size and/or study population, as well as the use of different questionnaires.

When evaluating which individual COHIP items demonstrated the greatest differences between cleft and control subjects, we found that for the Functional Well-being subscale cleft subjects were more likely to have difficulty eating, saying certain words, being understood when speaking, and keeping their teeth clean. These findings are consistent with the clinical manifestations of orofacial clefts. Individuals with orofacial clefts typically have malocclusion related to missing and rotated teeth, constricted maxillary arch form, and skeletal discrepancies, which impact mastication efficiency. Cleft palate is often associated with oronasal fistulas and/or velopharyngeal incompetence which result in hypernasality of speech. Difficulties with oral hygiene are consistent with rotated and mal-positioned teeth, which are often associated with orofacial clefts (Gorlin et al., 2001). Also, many of the subjects with orofacial clefts had orthodontic appliances, which can contribute to functional difficulties (Liu et al., 2011). For the Social-Emotional Well-being subscale, cleft subjects were more likely to be worried or anxious, experience bullying or teasing, feel like they looked different, be worried about what people think, and be upset or uncomfortable. These findings are supported by other studies (Hunt et al., 2006).

Our study did not find any significant differences in OHRQoL among cleft patients in different age groups. However, there was a significant difference in the interaction between age group and Social-Emotional Well-being when comparing children with orofacial clefts and control children. Orofacial clefts had a greater impact on Social-Emotional Well-being in 15-18 yr-olds than they did in 8-10 or 11-14 yr-olds, possibly because concerns about appearance and the opinions of others become more important as children with orofacial clefts approach the age of serious relationships with the opposite sex. Interestingly, we also found that cleft subjects in the 15-18 age group had a statistically significant higher treatment expectancy score than the cleft subjects in younger age groups, meaning that they were more hopeful about how they will feel after treatment and less nervous about receiving treatment.

Only a limited number of studies have looked at age-related differences in OHRQoL in children and adolescents with clefts. Although Broder and Wilson-Genderson's study (2007) found age-related differences when combining subjects from the craniofacial, orthodontic, pediatric, and control groups, differences within the craniofacial group alone were not reported. Results from a study by Damiano et al. (2007) demonstrated age-related differences in HRQoL in pre-adolescent children with orofacial clefts. The results of their study indicate that the difference in external appearance related to lip involvement has greater relative importance as the child gets closer to adolescence.

When comparing the responses of children with orofacial clefts to the reports of their caregivers, we found no significant differences for overall OHRQoL nor any of the subscales. Generally, caregivers have low to modest agreement with their child's QoL ratings (Eiser and Morse, 2001; Achenbach et al., 1987). Caregiver/Child agreement in other studies using the COHIP survey varies. Bos et al. (2010) found good agreement overall between reports of orthodontic patients (age 8-15) and their parents using the Dutch version of COHIP. On the other hand, Wilson-Genderson et al. (2007) found low to modest rates of caregiver agreement for their sample of craniofacial, orthodontic, and pediatric children. They found that craniofacial patients tended to rate their OHRQoL higher than their caregivers' responses, whereas pediatric and orthodontic patients were more likely to agree with their caregivers' responses or rate their OHRQoL lower.

Study Limitations

One of the limitations of this study is the difference in demographics between the study and control groups. There were significant differences between the cleft and control groups for age, gender, and race. The cleft group had significantly more males and fewer females than the control group. This is not surprising, considering the majority of subjects in the cleft group had cleft lip with or without cleft palate, which in Caucasian populations occurs more frequently in males than females by about 2:1 (Mossey and Little, 2002). However, there were no significant differences between responses for male and female subjects, so this difference should not affect the results.

Additionally, the control group had fewer Caucasians and more African Americans and Hispanics than the cleft group. This difference is explained by orofacial clefts being more prevalent in Caucasians than in African-Americans (Croen et al., 1998). Also, the control subjects were recruited from primary care clinics which draw many of their patients from urban, predominantly African-American neighborhoods near the hospital. The study subjects were recruited from the Craniofacial Anomalies Clinic which draws patients from a larger geographical area, with patients traveling up to 3 hours to attend the clinic. Although there were differences in racial composition of the cleft and control groups, there were no significant differences in OHRQoL between races, so the racial differences should not affect the results.

The cleft group had more subjects in the 11-14 age group (47%), whereas the majority of control subjects were in the 15-18 age group (53%). The reason for this difference is that a larger percentage of the control group came from the Adolescent Medicine Clinic than the Pediatric Primary Care Clinic, therefore the control subjects tended to be older. The majority of subjects from the Craniofacial Anomalies Clinic were in active orthodontic treatment and therefore more likely to be in the 11-14 age group, when orthodontic treatment is most prevalent. There were no significant differences among subjects in different age groups for the overall OHRQoL nor for any of the subscales except for Social-Emotional Well-being, for which there was a significant interaction between age group and COHIP score between the cleft and control groups.

We analyzed our data using parametric statistics so that our results could be compared with the statistical analysis in previous COHIP studies (Broder & Wilson-

Genderson, 2007; Ralstrom, 2009). However, we have concerns about the use of parametric statistics for several reasons. First, the COHIP questionnaire uses a Likert-like scale which could be argued to be ordered-categorical and not interval-level because respondents may not perceive response levels as equidistant. Second, the extremes at each end of the scale are not equal in magnitude. The possible responses for the first 30 items are ‘Never,’ ‘Almost Never,’ ‘Sometimes,’ ‘Fairly Often,’ and ‘Almost all the time.’ The opposite of ‘Never’ should be ‘All the time’ instead of ‘Almost all the time.’ This could be problematic because scoring is reversed for negatively-worded items but not for positively-worded questions, which could result in skewing of the data toward one end of the scale depending on whether the question was negatively or positively worded. Third, using Bonferoni corrected tests for normality we found many deviations from a normal distribution in our data.

To address these concerns, future studies may consider replacing the Likert-like scale with a visual analog scale. For the first 30 items, words of equal magnitude should anchor the scale (i.e. ‘Never’ and ‘Always’). Another possibility would be to leave the questionnaire as it is currently worded and alter the scoring and data analysis. Responses for items 1-30 could be assigned values between 0-100% based upon respondent perception (e.g. ‘Never’ might be assigned 0%, ‘Almost Never’ 10%, ‘Sometimes’ 40%, ‘Fairly Often’ 70%, ‘Almost all the time’ 90%) rather than assigning equidistant values for the responses.

CONCLUSIONS

1. Presence of an orofacial cleft significantly decreases overall OHRQoL, Functional Well-being, and Social-Emotional Well-being in children and adolescents.
2. The negative impact of orofacial clefts on Social-Emotional Well-being is greater in 15-18 year-olds than in younger age groups.
3. Children with orofacial clefts and their caregivers had very similar evaluations of the child's OHRQoL.

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CHAPTER 4

CONCLUSIONS

1. Presence of an orofacial cleft significantly decreases overall OHRQoL, Functional Well-being, and Social-Emotional Well-being in children and adolescents.
2. The negative impact of orofacial clefts on Social-Emotional Well-being is greater in 15-18 year-olds than in younger age groups.
3. Children with orofacial clefts and their caregivers had very similar evaluations of the child's OHRQoL.

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APPENDIX A

COHIP Questionnaire - Child

HELLO!! Thank you for helping us with our study. We are doing a study to better understand how children feel about their teeth and themselves.

DIRECTIONS: Please read carefully each statement and choose the answer that best describes you in the **PAST 3 MONTHS** regarding your teeth, mouth, or face. There are no right or wrong answers. We want to know how you really feel.

Example: In the past 3 months, how often have you felt shy because of your teeth, mouth, or face?

If you have felt shy because of your teeth, mouth, or face then choose the appropriate response. If you felt shy for other reasons, choose "Never."

<u>Never</u>	<u>Almost Never</u>	<u>Sometimes</u>	<u>Fairly Often</u>	<u>Almost all the time</u>
☐	☐	☐	☐	☐

Some things to keep in mind:

- * Answer the questions as honestly as you can.
- * Don't talk to anybody about the questions when answering them.
- * Before you answer, ask yourself:
 - "Does this happen to me because of my teeth, mouth, and face?"
- * Choose the answer that best describes you in the past 3 months.

	Never	Almost Never	Sometimes	Fairly Often	Almost all the time
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In the past 3 months, how often have you?

1. Had <u>pain</u> in your teeth/toothache.	☐	☐	☐	☐	☐
2. Been <u>breathing through your mouth</u> or <u>snoring</u> .	☐	☐	☐	☐	☐
3. Had <u>discolored teeth</u> or <u>spots</u> on your teeth.	☐	☐	☐	☐	☐
4. Had <u>crooked teeth</u> or <u>spaces</u> between your teeth.	☐	☐	☐	☐	☐
5. Had <u>sores</u> or <u>sore spots</u> in or around your mouth.	☐	☐	☐	☐	☐
6. Had <u>bad breath</u> .	☐	☐	☐	☐	☐
7. Had <u>bleeding gums</u> .	☐	☐	☐	☐	☐
8. Had <u>food sticking in or between</u> your teeth.	☐	☐	☐	☐	☐
9. Had <u>pain or sensitivity</u> in your teeth with <u>hot or cold things</u> .	☐	☐	☐	☐	☐

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	Never	Almost Never	Sometimes	Fairly Often	Almost all the time
In the past 3 months, how often have you?					
10. Had <u>dry mouth or lips</u> .	☐	☐	☐	☐	☐
11. Had <u>trouble biting off or chewing</u> food such as apple, carrot or firm meat.	☐	☐	☐	☐	☐
12. Been <u>unhappy or sad</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
13. <u>Missed school</u> for any reason because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
14. Been <u>confident</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
15. Had <u>difficulty eating foods</u> you would like to eat because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
16. Felt <u>worried or anxious</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
17. Felt <u>shy or withdrawn</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
18. Had <u>difficulty paying attention</u> in school because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
19. <u>Avoided smiling or laughing</u> with other children because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
20. Had <u>trouble sleeping</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
21. <u>Not wanted to speak/read our loud</u> in class because of your teeth, mouth, or face.	☐	☐	☐	☐	☐

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	Never	Almost Never	Sometimes	Fairly Often	Almost all the time
In the past 3 months, how often have you?					
22. <u>Been teased, bullied, or called names</u> by other children because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
23. <u>Had difficulty saying certain words</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
24. Felt that you <u>look different</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
25. Had people had <u>difficulty understanding</u> what you were saying because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
26. Felt that you were <u>attractive (good looking)</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
27. Had <u>difficulty</u> keeping your <u>teeth clean</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐
28. Been <u>worried about what other people think</u> about your teeth, mouth, or face.	☐	☐	☐	☐	☐
29. Been <u>upset or uncomfortable</u> with being asked questions about your teeth, mouth, or face.	☐	☐	☐	☐	☐
30. <u>Not wanted to go to school</u> because of your teeth, mouth, or face.	☐	☐	☐	☐	☐

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The following items are about your teeth, mouth, face, or general health. Please read each carefully and choose the answer that best describes you. There are no right or wrong answers. We want to know how you feel.

	Strongly Disagree	Somewhat Disagree	Don't Agree or Disagree	Somewhat Agree	Strongly Agree
31. I have <u>good</u> teeth.	☐	☐	☐	☐	☐
32. When I am <u>older</u> , I believe (think) I will have <u>good teeth</u> .	☐	☐	☐	☐	☐
33. When I am <u>older</u> , I believe (think) I will have <u>good health</u> .	☐	☐	☐	☐	☐
34. I <u>feel good about myself</u> .	☐	☐	☐	☐	☐
35. I will <u>feel better about myself</u> when <u>treatment</u> for my teeth, mouth, or face <u>is completed</u> .	☐	☐	☐	☐	☐
36. I am <u>nervous (anxious)</u> about the <u>treatment that I need</u> for my teeth, mouth, or face.	☐	☐	☐	☐	☐

	Poor	Fair	Average	Good	Excellent
37. Overall I feel my health is	☐	☐	☐	☐	☐
38. Overall I feel my oral health is	☐	☐	☐	☐	☐

Thank you for your time!!!

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APPENDIX B

COHIP Questionnaire - Caregiver

Oral Health Survey-Parent

Thank you for helping us with our study. We are doing a study to better understand how children feel about their teeth and themselves.

DIRECTIONS: Please read carefully each statement and choose the answer that best describes your child in the **PAST 3 MONTHS** regarding his/her teeth, mouth, or face. There are no right or wrong answers. We want to know **how you really feel**.

Example: In the past 3 months, how often has your child felt shy because of his / her teeth, mouth, or face?

If your child has felt shy because of his / her teeth, mouth, or face then choose the appropriate response. If your child has felt shy for other reasons not related to his / her teeth, mouth, or face choose "Never."

<u>Never</u>	<u>Almost Never</u>	<u>Sometimes</u>	<u>Fairly Often</u>	<u>Almost all the time</u>
☐	☐	☐	☐	☐

	Never	Almost Never	Sometimes	Fairly Often	Almost all the time
In the past 3 months, how often has your child?					
1. Had <u>pain</u> in his / her teeth/toothache.	☐	☐	☐	☐	☐
2. Been <u>breathing through his / her mouth</u> or <u>snoring</u> .	☐	☐	☐	☐	☐
3. Had <u>discolored teeth</u> or <u>spots</u> on his / her teeth.	☐	☐	☐	☐	☐
4. Had <u>crooked teeth</u> or <u>spaces</u> between his / her teeth.	☐	☐	☐	☐	☐
5. Had <u>sores</u> or <u>sore spots</u> in or around his / her mouth.	☐	☐	☐	☐	☐
6. Had <u>bad breath</u> .	☐	☐	☐	☐	☐
7. Had <u>bleeding gums</u> .	☐	☐	☐	☐	☐
8. Had <u>food sticking in or between</u> his / her teeth.	☐	☐	☐	☐	☐
9. Had <u>pain or sensitivity</u> in his / her teeth with <u>hot or cold things</u> .	☐	☐	☐	☐	☐
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	Never	Almost Never	Sometimes	Fairly Often	Almost all the time
In the past 3 months, how often has your child?					
10. Had <u>dry mouth or lips</u> .	☐	☐	☐	☐	☐
11. Had <u>trouble biting off or chewing</u> foods such as apple, carrot or firm meat.	☐	☐	☐	☐	☐
12. Been <u>unhappy or sad</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
13. Missed school for any reason because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
14. Been <u>confident</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
15. Had <u>difficulty eating foods</u> he / she would like to eat because of his/her teeth, mouth, or face.	☐	☐	☐	☐	☐
16. Felt <u>worried or anxious</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
17. Felt <u>shy or withdrawn</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
18. Had <u>difficulty paying attention</u> in school because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
19. <u>Avoided smiling or laughing</u> with other children because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
20. Had <u>trouble sleeping</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
21. <u>Not wanted to speak/read out loud</u> in class because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐

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	Never	Almost Never	Sometimes	Fairly Often	Almost all the time
In the past 3 months, how often has your child?					
22. <u>Been teased, bullied, or called names</u> by other children because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
23. <u>Had difficulty saying certain words</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
24. Felt that he / she <u>looks different</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
25. <u>Had people have difficulty understanding</u> what he / she was saying.	☐	☐	☐	☐	☐
26. Felt that he / she was <u>attractive (good looking)</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
27. <u>Had difficulty keeping his / her teeth clean.</u>	☐	☐	☐	☐	☐
28. <u>Been worried about what other people think</u> about his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
29. <u>Been upset or uncomfortable with being asked questions</u> about his / her teeth, mouth, or face.	☐	☐	☐	☐	☐
30. <u>Not wanted to go to school</u> because of his / her teeth, mouth, or face.	☐	☐	☐	☐	☐

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	Strongly Disagree	Somewhat Disagree	Don't Agree or Disagree	Somewhat Agree	Strongly Agree
31. He / She has <u>good</u> teeth.	☐	☐	☐	☐	☐
32. When he / she is <u>older</u> , he / she believes (thinks) that he / she <u>will</u> have good teeth.	☐	☐	☐	☐	☐
33. When he / she is <u>older</u> , he / she believes (thinks) that he / she <u>will</u> have good health.	☐	☐	☐	☐	☐
34. He / She <u>feels good about him / herself</u> .	☐	☐	☐	☐	☐
35. He / She will <u>feel better about himself / herself</u> when <u>treatment</u> for his / her teeth, mouth, or face <u>is completed</u> .	☐	☐	☐	☐	☐
36. He / She is <u>nervous (anxious)</u> about <u>the treatment</u> that he / she needs for his / her teeth, mouth, or face.	☐	☐	☐	☐	☐

	Poor	Fair	Average	Good	Excellent
37. Overall I feel his / her health is	☐	☐	☐	☐	☐
38. Overall I feel his / her oral health is	☐	☐	☐	☐	☐

Thank you for your time!!!

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APPENDIX C

Demographic Information Form

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APPENDIX D

Additional Tables

COHIP Item	Domain	Never		Almost Never		Sometimes		Fairly Often		Almost all the time	
		n	%	n	%	n	%	n	%	n	%
Pain in teeth	Oral Health	24	32%	24	32%	22	29%	3	4%	0	0%
Mouth breathing/snoring	Oral Health	20	27%	11	15%	20	27%	11	15%	13	17%
Discolored teeth	Oral Health	35	47%	15	20%	19	25%	3	4%	3	4%
Crooked teeth/spaces	Oral Health	10	13%	12	16%	20	27%	3	4%	27	36%
Sores in mouth	Oral Health	31	41%	22	29%	12	16%	6	8%	2	3%
Bad breath	Oral Health	20	27%	18	24%	26	35%	3	4%	8	11%
Bleeding gums	Oral Health	30	40%	14	19%	21	28%	8	11%	1	1%
Food stuck in teeth	Oral Health	7	9%	14	19%	33	44%	13	17%	6	8%
Pain/Sensitivity	Oral Health	26	35%	11	15%	19	25%	14	19%	5	7%
Dry mouth/lips	Oral Health	6	8%	16	21%	35	47%	8	11%	10	13%
Trouble Biting/Chewing	Functional	31	41%	18	24%	15	20%	7	9%	4	5%
Been unhappy	Social-Emo.	39	52%	12	16%	15	20%	4	5%	4	5%
Missed school	School	32	43%	5	7%	24	32%	10	13%	2	3%
Been confident	Self-image	22	29%	10	13%	20	27%	9	12%	14	19%
Difficulty eating	Functional	32	43%	18	24%	19	25%	4	5%	2	3%
Worried/anxious	Social-Emo.	33	44%	19	25%	15	20%	4	5%	4	5%
Shy/withdrawn	Social-Emo.	36	48%	13	17%	18	24%	4	5%	4	5%
Difficulty paying attention	School	55	73%	10	13%	7	9%	1	1%	2	3%
Avoided smiling/laughing	Social-Emo.	43	57%	16	21%	10	13%	1	1%	5	7%
Trouble sleeping	Functional	54	72%	12	16%	7	9%	2	3%	0	0%
Not wanted to speak	School	48	64%	12	16%	8	11%	3	4%	4	5%
Been teased/bullied	Social-Emo.	54	72%	12	16%	3	4%	3	4%	3	4%
Had difficulty saying words	Functional	22	29%	16	21%	25	33%	6	8%	5	7%
Looked different	Social-Emo.	25	33%	20	27%	21	28%	5	7%	4	5%
Felt difficult to understand	Functional	25	33%	13	17%	28	37%	3	4%	6	8%
Felt Attractive/good looking	Self-image	29	39%	14	19%	24	32%	1	1%	7	9%
Difficulty keeping teeth clean	Functional	35	47%	17	23%	17	23%	4	5%	2	3%
Worried what people think	Social-Emo.	32	43%	17	23%	17	23%	3	4%	6	8%
Been upset/uncomfortable	Social-Emo.	37	49%	12	16%	16	21%	5	7%	5	7%
Not wanted to go to school	School	57	76%	8	11%	5	7%	0	0%	5	7%

Table D.1 Cleft Group Response Frequencies (Items 1-30)

COHIP Item	Domain	Strongly Disagree		Somewhat Disagree		Don't Agree or Disagree		Agree Somewhat		Strongly Agree	
		n	%	n	%	n	%	n	%	n	%
Have good teeth	Self-image	8	11%	14	19%	12	16%	23	31%	18	24%
Will have good teeth	Self-image	2	3%	4	5%	3	4%	18	24%	48	64%
Will have good health	Self-image	0	0%	2	3%	2	3%	23	31%	48	64%
Feel good about myself	Self-image	1	1%	4	5%	6	8%	19	25%	45	60%
Will feel better about self when treatment is complete	Tx expect.	2	3%	2	3%	10	13%	19	25%	42	56%
Am nervous about treatment	Tx expect.	14	19%	10	13%	13	17%	23	31%	15	20%

Table D.2 Cleft Group Response Frequencies (Items 31-36)

COHIP Item	Domain	Poor		Fair		Average		Good		Excellent	
		n	%	n	%	n	%	n	%	n	%
Overall Health	Global Health	0	0%	2	3%	13	17%	33	44%	27	36%
Overall Oral Health	Global Health Oral	0	0%	7	9%	17	23%	34	45%	17	23%

Table D.3 Cleft Group Response Frequencies for Global Health(Items 37-38)

COHIP Item	Domain	Never		Almost Never		Sometimes		Fairly Often		Almost all the time	
		n	%	n	%	n	%	n	%	n	%
Pain in teeth	Oral Health	20	27%	20	27%	23	31%	5	7%	3	4%
Mouth breathing/snoring	Oral Health	26	35%	6	8%	22	29%	9	12%	12	16%
Discolored teeth	Oral Health	44	59%	16	21%	8	11%	6	8%	0	0%
Crooked teeth/spaces	Oral Health	34	45%	13	17%	10	13%	8	11%	9	12%
Sores in mouth	Oral Health	45	60%	10	13%	18	24%	1	1%	1	1%
Bad breath	Oral Health	21	28%	25	33%	23	31%	4	5%	2	3%
Bleeding gums	Oral Health	37	49%	11	15%	16	21%	8	11%	3	4%
Food stuck in teeth	Oral Health	13	17%	11	15%	39	52%	8	11%	3	4%
Pain/Sensitivity	Oral Health	22	29%	7	9%	28	37%	9	12%	9	12%
Dry mouth/lips	Oral Health	10	13%	14	19%	34	45%	11	15%	4	5%
Trouble Biting/Chewing	Functional	38	51%	20	27%	14	19%	2	3%	1	1%
Been unhappy	Social-Emo.	43	57%	12	16%	13	17%	3	4%	3	4%
Missed school	School	54	72%	10	13%	9	12%	1	1%	1	1%
Been confident	Self-image	8	11%	6	8%	18	24%	13	17%	29	39%
Difficulty eating	Functional	50	67%	9	12%	10	13%	1	1%	4	5%
Worried/anxious	Social-Emo.	53	71%	11	15%	7	9%	1	1%	3	4%
Shy/withdrawn	Social-Emo.	48	64%	15	20%	9	12%	2	3%	1	1%
Difficulty paying attention	School	61	81%	6	8%	8	11%	0	0%	0	0%
Avoided smiling/laughing	Social-Emo.	50	67%	15	20%	8	11%	2	3%	0	0%
Trouble sleeping	Functional	63	84%	6	8%	5	7%	1	1%	0	0%
Not wanted to speak	School	61	81%	8	11%	3	4%	1	1%	2	3%
Been teased/bullied	Social-Emo.	63	84%	1	1%	7	9%	3	4%	1	1%
Had difficulty saying words	Functional	53	71%	11	15%	7	9%	2	3%	2	3%
Looked different	Social-Emo.	58	77%	7	9%	6	8%	1	1%	3	4%
Felt difficult to understand	Functional	60	80%	6	8%	5	7%	2	3%	2	3%
Felt Attractive/good looking	Self-image	13	17%	3	4%	14	19%	8	11%	36	48%
Difficulty keeping teeth clean	Functional	55	73%	9	12%	11	15%	0	0%	0	0%
Worried what people think	Social-Emo.	50	67%	12	16%	8	11%	3	4%	2	3%
Been upset/uncomfortable	Social-Emo.	55	73%	9	12%	6	8%	2	3%	3	4%
Not wanted to go to school	School	65	87%	3	4%	6	8%	0	0%	1	1%

Table D.4 Control Group Response Frequencies (Items 1-30)

COHIP Item	Domain	Strongly Disagree		Somewhat Disagree		Don't Agree or Disagree		Agree Somewhat		Strongly Agree	
		n	%	n	%	n	%	n	%	n	%
Have good teeth	Self-image	2	3%	8	11%	9	12%	27	36%	29	39%
Will have good teeth	Self-image	2	3%	5	7%	6	8%	21	28%	41	55%
Will have good health	Self-image	1	1%	3	4%	4	5%	24	32%	42	56%
Feel good about myself	Self-image	0	0%	3	4%	4	5%	15	20%	52	69%
Will feel better about self when treatment is complete	Tx expect.	13	17%	5	7%	16	21%	8	11%	31	41%
Am nervous about treatment	Tx expect.	31	41%	6	8%	12	16%	17	23%	7	9%

Table D.5 Control Group Response Frequencies (Items 31-36)

COHIP Item	Domain	Poor		Fair		Average		Good		Excellent	
		n	%	n	%	n	%	n	%	n	%
Overall Health	Global Health	0	0%	3	4%	15	20%	26	35%	31	41%
Overall Oral Health	Global Health Oral	3	4%	4	5%	17	23%	28	37%	23	31%

Table D.6 Control Group Response Frequencies for Global Health (Items 37-38)

COHIP Item	BCLP		UCLP		BCL		UCL		CP		SPC		p-Value
	Mean	(STD)	Mean	(STD)	Mean	(STD)	Mean	(STD)	Mean	(STD)	Mean	(STD)	
Overall COHIP	96.5	(18.3)	93.6	(16.9)	98.3	(5.5)	92.6	(23.8)	96.0	(15.7)	108.7	(18.2)	0.59
Oral Health Well-being	25.0	(5.9)	23.9	(6.1)	27.3	(5.9)	25.4	(7.5)	23.9	(5.2)	26.4	(3.0)	0.74
Functional Well-being	18.0	(3.8)	17.1	(4.8)	19.0	(2.6)	17.1	(6.2)	17.6	(4.0)	20.6	(4.4)	0.24
Social-Emotional Well-being	24.5	(6.7)	24.0	(6.0)	23.7	(4.7)	22.4	(9.1)	25.3	(6.6)	28.8	(4.1)	0.60
School Environment	13.2	(2.4)	13.2	(2.8)	12.7	(0.6)	12.4	(4.6)	13.7	(1.8)	13.6	(2.6)	0.90
Self Image	15.9	(4.8)	15.2	(4.6)	15.7	(3.8)	15.3	(3.3)	15.6	(3.1)	19.2	(5.1)	0.87
Treatment Expectancy	5.1	(1.6)	5.1	(1.4)	4.7	(0.6)	5.1	(1.6)	5.4	(1.6)	4.6	(0.9)	0.38
Global Health	3.0	(0.9)	3.0	(0.8)	3.3	(0.6)	3.4	(0.7)	3.6	(0.5)	3.4	(0.5)	0.63
Global Health - Oral	2.9	(0.9)	2.8	(1.0)	3.3	(0.6)	2.5	(0.9)	2.7	(0.5)	3.4	(0.9)	0.66

Table D.7 Comparison of Overall and Subscale Scores according to Cleft Classification

COHIP Item	African Amer.		Asian		Caucasian		Hispanic		p-Value
	Mean	(STD)	Mean	(STD)	Mean	(STD)	Mean	(STD)	
Overall COHIP	106.4	(19.5)	101.0	(9.0)	98.1	(17.6)	124.0	(5.7)	0.95
Oral Health Well-being	27.1	(6.6)	27.0	(3.6)	24.7	(6.0)	30.5	(2.1)	0.48
Functional Well-being	20.0	(4.6)	20.0	(4.0)	18.7	(4.3)	23.0	(1.4)	0.80
Social-Emotional Well-being	26.5	(7.2)	24.3	(5.0)	25.4	(6.2)	31.5	(0.7)	0.61
School Environment	14.3	(2.6)	13.7	(2.1)	13.5	(2.6)	15.0	(1.4)	0.99
Self Image	18.4	(4.6)	16.0	(3.6)	15.9	(4.7)	24.0	(0.0)	0.45
Treatment Expectancy	5.2	(1.5)	5.3	(0.6)	5.0	(1.4)	3.5	(0.7)	0.21
Global Health	3.1	(0.9)	3.3	(0.6)	3.1	(0.8)	3.5	(0.7)	0.99
Global Health - Oral	2.9	(1.1)	3.3	(0.6)	2.8	(0.9)	3.0	(1.4)	0.88

Table D.8 Comparison of Overall and Subscale Scores among Different Races

COHIP Item	Male		Female		p-Value
	Mean	(STD)	Mean	(STD)	
Overall COHIP	100.3	(17.5)	104.1	(19.9)	0.84
Oral Health Well-being	25.8	(5.8)	25.9	(6.7)	0.81
Functional Well-being	18.9	(4.3)	19.8	(4.6)	0.90
Social-Emotional Well-being	25.6	(6.4)	26.3	(6.9)	0.88
School Environment	13.5	(2.5)	14.2	(2.7)	0.64
Self Image	16.4	(4.5)	17.8	(5.0)	0.76
Treatment Expectancy	4.9	(1.5)	5.2	(1.4)	0.59
Global Health	3.1	(0.8)	3.1	(0.9)	0.28
Global Health - Oral	2.8	(0.9)	2.8	(1.0)	0.20

Table D.9 Comparison of Overall and Subscale Scores between Genders