QUALITY OF LIFE ASSOCIATIONS WITH CARIES EXPERIENCE AND BEHAVIORAL CHALLENGES IN THE DENTAL SETTING AMONG CHILDREN WITH OROFACIAL CLEFTS

A Thesis

by

ANGELA KAY COOK

Submitted to the Office of Graduate and Professional Studies of Texas A&M University in partial fulfillment of the requirements for the degree of

MASTER OF SCIENCE

Chair of Committee,	Carolyn A. Kerins	
Committee Members,	Celia Heppner	
	Kavitha Viswanathan	
	Rosemarie Zartman	
	Alton G. McWhorter	
Head of Department,	Larry L. Bellinger	

May 2015

Major Subject: Oral Biology

Copyright 2015 Angela Kay Cook

ABSTRACT

Children with orofacial clefts experience many challenges beyond facial differences including risks for psychosocial and behavioral problems. As a result, evaluation for negative impacts on health-related quality of life (HRQOL) is highly important. Because the cleft condition manifests orally, more research is needed with regard to the dental impact on HRQOL. It is currently unknown how significantly HRQOL correlates with dental status or behavior of these children. The objective of this study was to evaluate HRQOL and surgical history of children with orofacial clefts to see if these measures correlate with caries experience and/or behavior in the dental chair.

Patient data was obtained with IRB-approval. The study population included patients with a non-syndromic orofacial cleft diagnosis between the ages of 4 and 10. Parent-reported data was obtained for 79 patients. Self-reported data was obtained for 23 patients. PedsQL Psychosocial Summary scores, PedsQL Total scores, number of surgeries and demographics were collected from the psychology department craniofacial clinic database. Decayed, missing, filled teeth (dmft) scores and Frankl scores during dental exams were collected from dental records. Median time between encounters with the psychology providers and the dental clinic for all patients was 42 days.

Spearman's Rank Correlation test identified significant correlations between parent-reported PedsQL Psychosocial Summary scores and dmft scores (p = 0.006) and PedsQL Total scores and dmft scores (p = 0.022), indicating that for the parent-reported group higher caries experience is significantly correlated with lower HRQOL. Parentreported PedsQL data were not correlated with Frankl scores. There is also no correlation between dmft or Frankl scores and the number of surgeries in the parentreported group.

For the self-reported group, significance was demonstrated between PedsQL Psychosocial Summary scores and Frankl scores (p = 0.002) and PedsQL Total scores and Frankl scores (p < 0.000), indicating that for the self-reported group lower HRQOL is significantly associated with poorer behavior in the dental chair. Self-reported PedsQL data were not correlated with dmft scores. There is also no correlation between dmft or Frankl scores and the number of surgeries in the self-reported group.

This study demonstrates that higher caries experience is associated with significantly lower HRQOL in children with orofacial clefts for the parent-reported group. Also, significantly lower HRQOL in children with orofacial clefts is associated with poorer behavior in the dental chair for the self-reported group. The number of surgeries a child with orofacial clefts has undergone is not associated with caries experience or behavior in the dental chair.

ACKNOWLEDGEMENTS

I am genuinely grateful for the guidance and mentorship given to me by my faculty, Drs. Carolyn A. Kerins, Kavitha Viswanathan, Rosemarie Zartman and Alton G. McWhorter through this research. I am thankful for the flexibility I was allowed to complete this task within my limited time as an active duty military resident. The assembly of this thesis was only possible because of the interdepartmental teamwork quarterbacked by Dr. Celia Heppner in the Psychiatry Department at Children's Medical Center. Your support, clarifications and editing were crucial to help me understand a different discipline.

Thank you.

TABLE OF CONTENTS

	Page	Э
ABSTRACT.		ii
ACKNOWLE	DGEMENTS i	ĪV
TABLE OF C	ONTENTS	v
LIST OF TAE	BLES	vi
LIST OF FIG	URES v	ii
CHAPTER I	INTRODUCTION	1
CHAPTER II	QUALITY OF LIFE ASSOCIATIONS WITH CARIES EXPERIENCE AND BEHAVIORAL CHALLENGES IN THE DENTAL SETTING IN CHILDREN WITH OROFACIAL CLEFTS	9
	Materials and Methods1Results1Discussion2	6
CHAPTER III	I CONCLUSIONS	:5
REFERENCE	2S	:6
APPENDIX	TABLES AND FIGURES 3	3

LIST OF TABLES

TABLE		Page	
1	Sociodemographic and Clinical Characteristics of Study Groups		
2	Summary of Spearman's Rank Test Correlations Results		

LIST OF FIGURES

FIGUF	RE P	Page
1	PedsQL Psychosocial Summary score distribution for parent-reported group	35
2	PedsQL Psychosocial Summary score distribution for self-reported group	36
3	Surgical history distribution for parent-reported group	37
4	Surgical history distribution for self-reported group	38
5	Caries distribution of parent-reported group	39
6	Caries distribution of self-reported group	40
7	Distribution of Frankl scores for parent-reported group	41
8	Distribution of Frankl scores for self-reported group	42

CHAPTER I

Orofacial clefts occur when tissues of the lip or palate fail to properly fuse during early embryonic development. The overall incidence of the condition is estimated to be approximately 1.7 per 1000 live births, which makes orofacial clefts among the most common craniofacial embryopathies worldwide.¹ Centers for Disease Control (CDC) estimates that every year in the United States 2,651 babies are born with a cleft palate and 4,437 babies are born with a cleft lip with or without a cleft palate.² With statistics like these, it is inevitable that pediatric dentists will encounter some, if not many, children with orofacial clefts while in practice. This literature review focuses on current collaborative treatment models employed by professionals treating children with orofacial clefts, the research that has been completed in this area and the need for further dental research within this unique population.

Although the etiology is unclear, cleft formation can be attributed to both exogenous factors (e.g. prenatal exposure to teratogenic agents) and endogenous factors (e.g. as part of Mendelian syndromes or as part of a phenotype resulting from chromosomal anomalies).³ For most patients, the diagnosis is made by clinical examination at birth; however, an increasing number of patients are being diagnosed via ultrasound *in utero*.⁴ The cleft lip or the cleft palate condition can manifest as an isolated occurrence or in combination. Cleft lip is associated with cleft palate in

1

approximately 68-86% of cases.⁵ Isolated cleft palate affects females most commonly whereas cleft lip with or without cleft palate predominantly occurs in males (60-80%).⁵ Unilateral cleft lip with or without cleft palate is two times more common than its bilateral counterpart and is more frequently seen affecting the left side.⁵

The treatment for orofacial clefts often includes multiple surgeries and medical interventions which generally begin within the first several months of life ⁶ and continue through adolescence depending on the cleft type and severity.⁵ These individuals are likely to suffer co-morbidities such as reduced fetal growth, feeding problems, or frequent ear infections, which can result in increased morbidity and mortality risks.^{6,7} The complex medical, surgical, ancillary and psychosocial interactions necessary in the rehabilitative process of these patients warrants a multidisciplinary team approach. The standard model for treating children with orofacial clefts is the organized cleft lip and palate team, which involves the collaborative efforts of multiple specialties and healthcare professionals.⁸ These teams may include professionals from plastic and reconstructive surgery, oral and maxillofacial surgery, pediatrics, audiology, speech pathology, social work, occupational therapy, psychiatry, pediatric dentistry and others. The advantage of the team structure is the ability to amalgamate the insights and skills of various specialties to coordinate complex, personalized services that meet the individual needs of the patients.

Although multidisciplinary treatment usually produces favorable functional and esthetic results, the orofacial cleft condition has been shown to impose a long-term burden on psychological wellbeing⁷ and quality of life.^{7,9} Previous studies have

2

reported increased risks for psychosocial challenges and mental health from infancy throughout adulthood.^{10, 11} Compared to their unaffected counterparts, children and adolescents with orofacial clefts have increased risks for behavioral and emotional problems.^{6, 10-13} Having to endure frequent bullying from peers,^{10, 11, 14} difficulties with speech,^{11, 15} and concerns with esthetics ^{6, 11} are all reasons that have been cited for these problems. There have also been reports of increased rates of learning disabilities, lower rates of school achievement,^{9, 14, 16-18} more depressive or anxious symptoms,^{9, 11, 18, 19} and lower self-esteem among these individuals.^{9, 13, 15, 18, 20} Wehby *et al.* (2012) found that an increased number of surgeries, lower socioeconomic status and lower satisfaction with facial appearance were predictors of behavioral problems for children with orofacial clefts.⁶ Other researchers have cited an increased number of surgeries as having a negative impact on the emotional development and physical wellbeing of this population.^{7, 9, 10} With regard to the general population, the psychology literature has found that psychosocial problems reported in childhood are predictive of analogous problems later in life;²¹ therefore, assessing behavioral outcomes of children with orofacial clefts at a young age is vital for early identification and treatment of psychosocial red flags.

The psychological care provided for children with orofacial clefts involves issues related to family adaptation, appearance, self-esteem, social interaction, emotional and behavioral adjustment and cognitive functioning.¹⁰ An umbrella construct that can capture such issues and numerically describe the general wellbeing of a patient is known as quality of life, or health-related quality of life (HRQOL). The term HRQOL is

defined as the impact of the disease and treatments on an individual's physical, psychological and social functioning.²² When investigating the HRQOL of a given population, researchers may choose a condition-specific instrument or a generic instrument. In many cases, established generic HRQOL instruments are advantageous over condition-specific measures as they often report normative data across a range of health conditions to facilitate comparisons across study populations.²³ Generic instruments are utilized for descriptive epidemiological research applications for children and adolescents. The two most widely utilized generic pediatric instruments are the Child Health Questionnaire (CHQ), a 98-item self-report measure ²⁴ and the Pediatric Quality of Life Inventory (PedsOL), a 23-item measure.²⁵ Due to the shorter format, PedsQL is often chosen in research. The PedsQL was designed to measure the core health dimensions defined by the World Health Organization (WHO) including physical, mental and social health as well as school functioning.²⁶ The American Cleft Palate Association's "Parameters for Care" advise that cleft teams assess social and emotional quality of life routinely during patient evaluations and treatment.²⁷ Since the PedsQL assesses for psychosocial quality of life, it is a useful tool for psychology providers on cleft teams.

The psychology providers of the Fogelson Plastic Surgery and Craniofacial Clinic team at Children's Medical Center (CMC) in Dallas, Texas are making efforts for early detection of psychosocial problems in children with orofacial clefts. Providers routinely obtain standard of care psychosocial evaluations starting from the initial visit with the team, which can be as early as birth, and continuing annually or biannually until 25 years

of age. If need be, these screenings serve as a referral point for further psychological evaluation and counseling. In addition, data from psychosocial screenings are used in an IRB-approved expedited study entitled, *Quality of Life in a Pediatric Craniofacial Population* (STU: 022012-032). In this study, PedsQL data from clinical evaluations, surgical history, medical treatment history and demographic variables are collected and recorded in a standardized datasheet.

One important health outcome associated with the HRQOL of children with orofacial clefts, which is currently lacking in exploration in the literature, is dental.²⁸ While it is well known that children with orofacial clefts have been shown to be at high risk for developing early childhood caries (ECC),²⁹⁻³³ currently, there is a dearth of information that evaluates dental status in relation to HRQOL. It may be possible that dental caries status could greatly impact HRQOL in this population. The term ECC is defined by the American Academy of Pediatric Dentistry (AAPD) as the, "presence of 1 or more decayed (noncavitated or cavitated lesions), missing (due to caries), or filled tooth surfaces in any primary tooth in a child 71 months of age or younger." ³⁴ It has been shown that 20-75% of children with orofacial clefts will develop ECC.²⁹ Due to ECC, children with orofacial clefts often have treatment needs that are extensive and complex. The contributing factors to dental caries in these patients are the presence of enamel defects,^{29, 35-37} the high *Streptococcus mutans* and *Lactobacilli* counts,^{29, 30, 38, 39} the use of infant orthopedic appliances,²⁹ deficiencies in oral hygiene practices,^{29-31, 33, 40} and slow oral food clearance.^{29, 30} Because of the high ECC risk, there is strong advocacy for frequent dental recalls and close management of these patients to offset the increased risks.

Past research has shown that the presence of ECC can lead to substantially negative effects on the HROOL of children within the general population.⁴¹⁻⁴⁸ Children with ECC may not verbally complain of tooth pain; however, a disruption of quality of life can manifest in different ways such as poor sleeping, eating and behavioral problems.^{43, 49} Sleeping patterns can be interrupted by dental pain which can then have an effect on glucocorticoid production and nighttime growth.⁴⁷ ECC can cause chewing discomfort and reduced total food intake. ECC is also associated with negative behaviors in children such as irritability,⁴⁷ aggression, lack of cooperation and difficulty playing well with other children.⁴³ Measuring dental pain indirectly by habits or behaviors can be as important as measuring dental pain directly. In a study by Low et al. (1999), questionnaire-based HRQOL surveys were completed by parents of children with ECC that were otherwise healthy before full-mouth dental rehabilitation treatment in the operating room and again 8 weeks after treatment. The dental treatment was shown to have a statistically significant effect in alleviating pain, reversing eating problems, improving sleep habits and improving child behavior.⁴³ While children with orofacial clefts already experience many threats to their overall HRQOL, it can be postulated that when coupled with the effects of ECC, these children could experience significantly poorer HRQOL.

Because disruptions of HRQOL due to ECC can manifest as behavioral problems in children, another aspect of interest for pediatric dental providers is the potential for behavioral challenges that may occur when children with orofacial clefts present for inoffice dental treatment. These children have been shown to exhibit strong orally defensive behaviors,²⁹ especially in the oral cleft region(s), which can make accepting manipulation of the oral tissues and dental treatment extremely difficult. In the general pediatric population, early negative or painful medical experiences have been shown to create enduring memories which can lead to avoidance and negative attitudes toward future health care encounters.⁵⁰ These attitudes may be reflected in a child's HRQOL and may explain why children with orofacial clefts, who have had extensive histories of orally focused care, can be difficult to manage in the dental office. Given advanced knowledge of a child's current score on a HRQOL instrument, however, it may be possible to anticipate behavioral problems in children with orofacial clefts.

For reasons such as the high susceptibility of these children to dental caries and their difficulty coping with the demands of invasive dental treatment,^{9, 29} it is all the more important for dentists to be prepared to manage potential uncooperative behaviors and treat these patients successfully. A paucity of direct clinical research has been performed in the area of uncooperative behaviors exhibited by children with orofacial clefts in the dental setting; however, among pediatric dentistry residents and staff at the CMC Dental Clinic, it has been anecdotally noted that children with orofacial clefts tend to be some of the most behaviorally challenged dental patients. Wehby *et al.* (2012) found that children with orofacial clefts age 6 years and older exhibited elevated inattention/hyperactivity risks compared to the normative samples.⁶ A possible explanation for these behavioral problems proposed by this study is an increased number of surgeries.⁶ An increase in the number of surgeries has been shown to cause

significant stress in children and may adversely effect the child's emotional and psychological status.^{6, 10} Combining an understanding of a child's HRQOL and surgical past may prove useful when assessing which patients with orofacial clefts are at higher risk for negative behavioral outcomes in the dental chair.

The intent of this study was to evaluate the HRQOL and surgical history of children with orofacial clefts to see if correlations exist with caries experience and/or behavior in the dental chair. To date, no study has examined the HRQOL and surgical history of children with orofacial clefts and compared that information to a child's dental caries experience and behavior in the dental chair. The hypothesis is that children with orofacial clefts and lower HRQOL scores on the PedsQL will exhibit a greater number of dental caries and will display poorer behavior in the dental chair. Additionally, it is hypothesized that children who have had more surgeries will also have more dental caries and will display poorer behavior in the dental chair. If a correlation is found, information about HRQOL and/or the number of past surgeries may provide a better understanding of caries susceptibility and behavior in the dental chair. By providing pediatric dentists with meaningful predictors for caries susceptibility and behavioral challenges in children with orofacial clefts, preparations can be made for successful preventative strategies and positive dental encounters. With conscious improvements in dental healthcare practices, the ultimate goal is to enhance the dental health outcomes of affected children and lessen the overall burden of orofacial clefts at the individual, familial and societal levels.

CHAPTER II

QUALITY OF LIFE ASSOCIATIONS WITH CARIES EXPERIENCE AND BEHAVIORAL CHALLENGES IN THE DENTAL SETTING AMONG CHILDREN WITH OROFACIAL CLEFTS

Because cleft lip and palate is the second most common birth defect and the most common congenital craniofacial anomaly,²⁸ pediatric dentists are likely to encounter patients with orofacial clefts in clinical practice. Children with orofacial clefts experience a great number of challenges beyond differences in facial appearance. It is well understood that there are many important health outcomes associated with cleft care. As such, the process of rehabilitation for these patients requires the collaboration of a team of medical, surgical, ancillary and psychosocial specialists. The team care approach allows for the coordination and integration of complex services tailored to the individual patient's needs. Although team care usually yields favorable functional and esthetic results, it has been shown that the orofacial cleft condition imposes long-term burdens on the psychological health and quality of life of the affected individuals.

Compared to unaffected children, those with an orofacial cleft have increased risks for behavioral, emotional and adjustment problems.^{6, 10-13} Because psychosocial problems in childhood have been shown to be predictive of problems later in adulthood,²¹ it is important that children with orofacial clefts be evaluated routinely for possible negative impacts on psychological wellbeing. Early identification allows for

proactive treatment approaches to improve future health and quality of life. The psychological care provided for children with orofacial clefts includes issues related to family adaptation, appearance, self-esteem, emotional and behavioral adjustment, social interaction and cognitive functioning.¹⁰

The health-related quality of life (HRQOL) instrument is a common tool used to numerically illustrate a patient's wellbeing. HRQOL is defined as the impact of the disease and treatment on an individual's physical, psychological, and social functioning.²² A common, validated generic instrument for HRQOL is called the Pediatric Quality of Life Inventory (PedsQL).^{25, 26, 51-53} The design of the PedsQL focuses on the core health dimensions defined by the World Health Organization (WHO); namely, physical, mental, social health and school functioning.²⁶ Psychologists collect PedsQL data on children with orofacial clefts and use these data as referral points for further psychological evaluation and counseling.²³

As previously mentioned, the multifaceted nature of the orofacial cleft condition results in many health-related outcomes. It is important, however, to understand which aspects play the largest role in quality of life. Because the cleft lip and palate condition manifests orally, more research is needed specifically with regard to the oral health and dental impact on quality of life. It is known that children with orofacial clefts are at high risk for developing early childhood caries (ECC),²⁹⁻³³ which often creates dental treatment needs that are extensive and complex. The caries etiology in children with orofacial clefts is often attributed to enamel defects, ^{29, 35-37} high *Streptococcus mutans* and *Lactobacilli* counts, ^{29, 30, 38, 39} the use of oral obturator appliances during infancy,²⁹

poor oral hygiene practices, ^{29-31, 33, 40} and slow oral food clearance. ^{29, 30} Within the dental community, there is strong advocacy for close management of children with orofacial clefts, but it is currently unknown exactly how significantly the dental status of these children correlates with their quality of life. Within the general population, ECC has shown substantially negative effects on HRQOL.⁴¹⁻⁴⁸ Perhaps, those children with orofacial clefts who are unaffected by ECC will showcase a greater overall quality of life. Likewise, those who have undergone invasive dental procedures or have untreated ECC may have a poorer quality of life. Such findings might inspire parents and healthcare providers to take a more proactive role in the oral health of children with orofacial clefts.

One consequence of a reduced quality of life that has been shown in the literature is behavioral problems.⁴³ If the dental health of children with orofacial clefts negatively affects their quality of life, those negative health outcomes might also manifest in poor behavior in the dental chair. Research has also shown that behavioral problems might be the result of an increased number of surgeries.⁶ It may be advantageous to the treating pediatric dentist to use known information about a child's quality of life or number of past surgeries to help anticipate behavioral challenges in the dental setting.

The goal of this study is to evaluate the HRQOL and surgical history of children with orofacial clefts to see if these measures correlate with caries experience and/or behavior in the dental chair. There are no studies at this time that have specifically attempted to correlate dental status and behavior in the dental chair with quality of life or surgical history of children with orofacial clefts. If statistically significant correlations are found, this information may motivate parents and dental and healthcare providers to put greater emphasis on improving the dental health of children with orofacial clefts so that improvements will ultimately reflect in a greater quality of life.

Materials and Methods

Procedure

Approval for access to patient information was obtained via a modification to an ongoing IRB-approved expedited study entitled *Quality of Life in a Pediatric Craniofacial Population* (STU: 022012-032) granted by the Institutional Review Board of the University of Texas Southwestern Medical Center in Dallas, Texas with site-specific approval from Children's Medical Center. Consenting patients was not necessary due to the minimal risk of the study and because research procedures do not differ from the standard care of procedures. The patient population was filtered from the ongoing study datasheet to include patients between the ages of 4 and 10 years old with a non-syndromic orofacial cleft diagnosis. Patient demographics (age, gender, ethnicity, etc.), quantitative surgical history data and PedsQL scores were gathered from this datasheet. Inclusion criteria for this new study required that all study enrollees be patients of record at the Children's Medical Center Dental Clinic and have received either a new patient exam or a dental recall appointment within 6 months of the

psychology providers' evaluation. Data regarding behavior in the dental chair, in the form of Frankl Scores, were obtained from the new patient exams or dental recall appointments. Data concerning caries experience, in the form of dmft ("decayed-missing-filled teeth") scores, were collected by retrospective dental chart review.

Measures

The Pediatric Quality of Life (PedsQL) Inventory Version 4.0 is a generic measure, which can be used for evaluating quality of life outcomes in children with orofacial clefts. It consists of 23 questions on a five-point Likert scale from 'never' to 'almost always' and is intended to measure patient and parent perceptions of the patient's HRQOL. It has been shown to be valid and reliable.^{25, 26, 51-53} The PedsQL can be used for various pediatric health conditions, because rather than assessing specific health conditions, it evaluates the impact of health on function.^{25, 26, 51} Broder *et al.* (2014) assessed the PedsQL and found it to be sensitive to issues of clinical importance to children with orofacial clefts.²³ For children of all age ranges, PedsQL Inventory data is obtained via parent report forms. Developmentally appropriate test versions are available for ages 2-4, 5-7, 8-12 and 13-18. For children ages 8 and up, self-report forms are also available in two age-appropriate versions: 8-12 and 13-18. The target population for this study is age 4-10; therefore, most of the PedsQL scores were reported by parent proxy. However, when available, the self-reported data was compared to the parent-reported data and was analyzed as well. The validated Spanish language version ²⁵ of the PedsQL was used for the portion of our patients who are Spanish-speaking,

which accounted for 56% of the total patient population.

The data on the number of surgeries the child had undergone was obtained after the psychological clinical evaluation by retrospective medical chart review.

The Frankl Scale is a reliable and frequently used behavioral rating system in both clinical dentistry and dental research.⁵⁴ It divides observed behavior into four categories: 'definitely negative,' 'negative,' 'positive' and 'definitely positive,' which are assigned numbers on a scale from 1 to 4, respectively.^{54, 55} Frankl scores are routinely documented on treatment notes in most pediatric dental offices as a diagnostic aid for future visits.⁵⁴

The dmft/DMFT ("decayed, missing, filled teeth"/"Decayed, Missing, Filled Teeth") score is an index of dental caries in the primary and permanent dentitions developed by the World Health Organization (WHO) and is used in epidemiological surveys of dental status. It is a well established measure of caries experience and has been used for more than 70 years.⁵⁶ When applied to both dentitions, the dmf/DMF index equals the total number of teeth or surfaces that are decayed (d/D), missing (m/M), or filled (f/F) in an individual. Because of the difficultly in distinguishing between teeth that were not formed due to the presence of the cleft, teeth extracted due to caries and those that have exfoliated naturally, missing teeth was disregarded in this protocol.⁵⁷ Because most study participants are in an early or mixed dentition stage and none have a complete permanent dentition, for consistency the primary dentition was considered exclusively. In such case, caries experience will be stated as a "dmft score." Data Analysis

Excel (Excel 2011, Microsoft Corp., Redmond, Washington) and SPSS (SPSS 20.0, IBM Corp., Armonk, New York) were used to analyze data. The data analyzed included demographic information, PedsQL data, surgical history data, dmft scores and Frankl behavior scores. Prior to analysis, PedsQL scores were converted to Z-scores to minimize variability resulting from the different test forms administered to the defined age groups, as well as to control for variability in standard deviations for scores for the different age groups. Data analysis was accomplished with the Spearman's Rank Correlation test.

Results

The study sample included 79 parent-reported patient HRQOL (mean age = 7.55 years, s.d. = 2.05) and 23 self-reported patient HRQOL (mean age = 9.63 years, s.d. = 0.18) evaluated at the Fogelson Plastic Surgery and Craniofacial Clinic team and the dental clinic at Children's Medical Center (CMC) in Dallas, Texas. Demographic and clinical characteristics of the study samples are summarized on Table 1.

Of the patients reported by parent proxy, 89.8% had cleft lip and palate, 7.5% had cleft palate only, with the remaining 2.7% having cleft lip only. Sixty-five percent (N = 51) were male. Racial identification included the following: 78% Caucasian (N = 62), 6% Black (N = 5), 1% Asian (N = 1), 1% other (N = 1) and 14% (N = 10) not reported. Report for ethnicity found that 70% (N = 55) identified as Hispanic, the remaining identified as Not Hispanic. The primary language of the parent-reported group was Spanish for 56% (N = 44) and English for 44% (N = 35). Ninety percent of parent-reported patients were insured by government subsidies (N = 71), eight percent were private pay (N = 6) and two percent (N = 2) were uninsured.

Of the self-reported patients, 87% had cleft lip and palate, the 13% remainder had cleft palate only. Seventy percent (N = 16) were male. Racial identification included the following: 70% Caucasian (N = 16), 4% Black (N = 1), 4% other (N = 1) and 22% (N = 5) not reported. Report for ethnicity found that 61% (N = 14) identified as Hispanic, the remaining identified as Not Hispanic. The primary language of the selfreported group was Spanish for 48% (N = 11) and English for 52% (N = 12). Ninetyone percent of self-reported patients were insured by government subsidies (N = 21) and nine percent were private pay (N = 2).

The median number of days between encounters with the psychology providers and the dental clinic for all patients was 42.

The PedsQL subscale most relevant to the orofacial cleft population is the Psychosocial Summary, which is a composite score including the Emotional, Social and School Functioning scale scores. The PedsQL Psychosocial Summary score is distinct from the PedsQL Total Scale Score because it does not include the Physical Functioning scale score. Physical disabilities are not often associated with children with nonsyndromic orofacial clefts and, therefore, it is preferable to eliminate this scale score to allow for greater sensitivity to the more common psychosocial problems in this population. However, for comparison purposes, the PedsQL Total Scale Score, which includes Physical Functioning, will be reported also.

The PedsQL Psychosocial Summary scores were found to be normally distributed for both parent-reported and self-reported groups. The mean PedsQL Psychosocial score for the parent-reported group was 71.8 out of a total 100.0 with a standard deviation of 18.2 (Figure 1). The mean PedsQL Psychosocial score for the self-reported group was 71.1 out of a total of 100.0 with a standard deviation of 3.8 (Figure 2).

The PedsQL Total Scale scores were also found to be normally distributed for both parent-reported and self-reported groups. The mean PedsQL Total Scale score for the parent-reported group was 73.7 out of a total of 100.0 with a standard deviation of 18.0. The mean PedsQL Total Scale score for the self-reported group was 77.4 out of a total of 100.0 with a standard deviation of 3.5.

Due to difficulties ascertaining an exact surgical history, patients were categorized into groups to minimize error in surgical history reporting. For the parentreported group, patients with zero surgeries and 11 to 15 surgeries accounted for 2.5% each (N = 2 for both), 1 to 5 surgeries accounted for 53% (N = 42), 6 to 10 surgeries accounted for 40.5% (N = 32), and over 20 surgeries accounted for 1.5% (N = 1) (Figure 3). For the self-report group, patients with zero surgeries and 11 to 15 surgeries accounted for 4% each (N = 1 for both), 1 to 5 surgeries accounted for 52% (N = 12), and 6 to 10 surgeries accounted for 40% (N = 9) (Figure 4).

Caries experience in both parent-reported and self-reported groups were not found to be normally distributed. The median dmft score for the parent-reported group was 6 with an interquartile range of 7 (Figure 5). The median dmft score for the selfreported group was 4 with an interquartile range of 6 (Figure 6).

Frankl Score data during new patient exams or dental recall appointments for the parent-reported group resulted in a Frankl score = 2 in 8% of the patients (N = 6), Frankl score = 3 in 39% of the patients (N = 31) and Frankl score = 4 in 53% of the patients (N = 42) (Figure 7). The self-reported group displayed a Frankl score = 3 in 40% (N = 9) of the patients and Frankl score = 4 in 60% of the patients (N = 14) (Figure 8).

Table 2 summarizes all correlation results for parent-reported and self-reported groups. Using Spearman's Rank Correlation Test in the parent-reported group, PedsQL

Psychosocial Summary scores were significantly correlated with dmft scores (p = 0.006), indicating that a higher HRQOL is significantly correlated with a lower caries experience. No correlation (p = 0.693) was found for Frankl scores, indicating that for parent-reported scores, HRQOL is not correlated with behavior in the dental chair. For comparison, PedsQL Total Scale scores were also correlated significantly with caries experience (p = 0.022), but not with behavior in the dental chair (p = 0.489).

In the parent-reported group, Spearman's Rank Correlation Test was also used to determine a correlation between the number of surgeries patients have undergone and caries experience and behavior in the dental chair. Between number of surgeries and caries experience, there was only a slight absence of statistical significance (p = 0.052); whereas number of surgeries and behavior in the dental chair yielded no correlation (p = 0.489).

For patients with self-reported PedsQL scores, there was shown to be no correlation between PedsQL Psychosocial Summary scores and PedsQL Total Scale scores with dmft scores (p = 0.649 and 0.833, respectively). Unlike parent-reported scores, however, self-reported PedsQL scores in both subscales yielded significant correlations with Frankl scores; p = 0.002 for Psychosocial Summary score and p < 0.000 for PedsQL Total score. No correlation between number of surgeries and caries experience/Frankl scores was found (p = 0.265 and 0.459, respectively).

Discussion

This cross-sectional study examined the HRQOL and surgical histories of children with orofacial clefts to determine if significant correlations exist among those measures and caries experience and/or behavior in the dental chair.

The most remarkable finding in this study is that within the observed population of children with orofacial clefts, those with few or no dental caries exhibit a quality of life that is significantly greater than those with many dental caries. This study is the first of its kind to demonstrate that increased dental caries is associated with a significantly negative effect on the overall HRQOL of children with orofacial clefts. This finding is reflected in both the PedsQL Psychosocial Summary and the PedsQL Total Scale score for the parent-reported sample group (p = 0.006 and 0.022, respectively). Because the orofacial cleft condition manifests in the oral cavity, a possible explanation for this finding is that any other disruption in the mouth leading to added discomfort and further invasive treatment could produce strong, negative psychological impacts. This finding is consistent with two other studies (in non-cleft patients) that demonstrated worse quality of life in healthy children with dental caries. One study found that cavitated caries lesions on anterior and posterior teeth in healthy preschool children caused a negative impact on quality of life.⁴¹ The other study determined that dental caries was associated with poorer quality of life in 5 to 6 year old children as reported by children and their

parents.⁴² No other study has demonstrated a significant inverse correlation between quality of life and caries experience in children with orofacial clefts.

While a correlation between HRQOL and caries experience was demonstrated for the parent-reported group, it was not duplicated in the self-reported group. An explanation for this difference is that sample size of the self-reported group was small (N = 23, versus N = 79 for the parent-reported group). Given a larger sample of selfreporting patients, it may have been possible to demonstrate this important finding categorically.

The connection between HRQOL and caries experience are important for pediatric dentists who treat children with orofacial clefts. By focusing on improving the dental health of children with orofacial clefts with frequent dental recall appointments and conscientious preventative strategies, it may be possible to greatly improve their overall quality of life. Such an effect may be long lasting, as shown by studies that report experiences in childhood carrying over into adulthood.²¹ This finding should inspire pediatric dentists to take a proactive role in the multidisciplinary care of these children.

This study also sought to correlate HRQOL with behavior in the dental chair. Anecdotally, it has been found that children with orofacial clefts tend to be some of the most behaviorally challenged patients at the CMC Dental Clinic. It was hypothesized that children with poorer PedsQL scores would exhibit worse behavior in the dental chair during new patient exams and dental recall appointments. This hypothesis was true for the self-reported group, but not for the parent-reported group. This discrepancy may be a reflection of parent reporting bias either by over-reporting psychosocial difficulties when they may be age-appropriate or by under-reporting difficulties that their child did describe in their self-report forms.

Another possible explanation could be that because the parent-reported group contains a wider age range, it may not be possible to isolate the negative behaviors that are developmentally "age-appropriate" from the negative behaviors that are a reflection of a poor quality of life. Because the self-reported group includes only children from ages 8 to 10 years old (which are the oldest children included in this study) it is possible that poor behavior in the dental chair cannot be defended as "age-appropriate." If an 8 to 10 year old child is displaying poor behavior in the dental chair, it is possible that it is a manifestation of a severe disruption in quality of life and not immaturity. To test this explanation, 8 to 10 years olds were separated from the parent-reported group and their PedsQL scores were compared with behavior scores. Interestingly, a correlation was found with PedsQL Total scores and behavior in the dental chair (p = 0.050), but there was no correlation with PedsQL Psychosocial Summary scores and behavior (p = 0.087). This study finding shows that parent-reported PedsQL Total scores may serve as a predictor for behavior in the dental chair, but only for children ages 8 to 10 years old.

Because of the low demands of new patient exams and recalls, perhaps a better dental encounter to illustrate behavior and correlate with PedsQL scores would be an operative dentistry appointment with dental restorative treatment or a tooth extraction. Due to the variability of these types of appointments, however, a standardized operative dentistry encounter was unable to be defined and studied. Perhaps future prospective research in the area of behavior of children with orofacial clefts in the dental setting could focus on operative dentistry appointments.

Overall, the findings of this study do not uphold the anecdotal notion that children with orofacial clefts display worse behavior in the dental chair. Further research in the area of randomized, controlled clinical trials would be required to make such a claim.

The last component of the hypothesis sought to determine if an increased number of surgeries resulted in a greater dental caries experience and worse behavior in the dental chair. In the parent-reported group, there was a slight absence of significance between dental caries experience and number of surgeries (p = 0.052). For the selfreported group, no correlation was found between dental caries experience and number of surgeries (p = 0.265). Given a greater sample size in both the parent-reported and self-reported groups, it may have been possible to demonstrate a correlation between dental caries experience and number of surgeries. In both the parent-reported and selfreported groups, no correlation was found between Frankl Scores and number of surgeries. With these results, no specific comment can be made with regard to a greater number of surgeries placing a child with an orofacial cleft at greater risk for dental caries or for demonstrating worse behavior in the dental chair.

On a practical level, this study helps further the dental practitioner's understanding of the risk factors for the psychological wellbeing in children with orofacial clefts. Dental caries has been shown to cause a significant impact on the quality of life of these patients. It is, therefore, incumbent on the pediatric dentist to utilize preventative strategies such as optimized home care, more frequent professional visits with regimented topical fluoride application, dietary counseling and anticipatory guidance to help minimize dental caries risks in this population. It is also recommended that pediatric dental providers work in close coordination with psychology providers and other team care members to proactively manage the complex treatment needs of these patients.

The limitations of this study included the retrospective nature of the data collection method, the limited sample size of the self-reported groups and the concentration on a single location.

The American Cleft Palate Association's Parameters for Care advises cleft teams around the US to collect ongoing quality of life data for cleft team patients. An interesting research endeavor would be to see if dental caries experience is equally affecting the quality of life of orofacial cleft patients in different geographic locations. Such findings may increase national advocacy for the pediatric dentist to assume a more central role in the long-term care of these patients. This may lead to further and more comprehensive investigations into strategies to best approach the oral and dental health of children with orofacial clefts with a primary goal of helping these children realize their greatest potential quality of life.

CHAPTER III

CONCLUSIONS

- 1. Higher dental caries experience is associated with significantly lower HRQOL in children with orofacial clefts.
- 2. For children with orofacial clefts between the ages of 8 and 10, significantly lower HRQOL is associated with poorer behavior in the dental chair.
- 3. The number of surgeries a child with orofacial clefts has undergone was not associated with dental caries experience or behavior in the dental chair.

REFERENCES

- Mossey PA, Little J, Munger RG, Dixon MJ, Shaw WC. Cleft lip and palate. Lancet 2009;374(9703):1773-85.
- Facts about cleft lip and cleft palate. Centers for Disease Control and Prevention. Accessed July 15 2013. http://www.cdc.gov/ncbddd/birthdefects/cleftlip.html.
- Marazita ML, Mooney MP. Current concepts in the embryology and genetics of cleft lip and cleft palate. Clin Plast Surg 2004;31(2):125-40.
- Jones MC. Prenatal diagnosis of cleft lip and palate: detection rates, accuracy of ultrasonography, associated anomalies, and strategies for counseling. Cleft Palate Craniofac J 2002;39(2):169-73.
- Arosarena OA. Cleft lip and palate. Otolaryngol Clin North Am 2007;40(1):27-60.
- Wehby GL, Tyler MC, Lindgren S, et al. Oral clefts and behavioral health of young children. Oral Dis 2012;18(1):74-84.
- Wehby GL, Cassell CH. The impact of orofacial clefts on quality of life and healthcare use and costs. Oral Dis 2010;16(1):3-10.
- 8. Strauss RP. The organization and delivery of craniofacial health services: the state of the art. Cleft Palate Craniofac J 1999;36(3):189-95.
- Vogels WE, Aartman IH, Veerkamp JS. Dental fear in children with a cleft lip and/or cleft palate. Cleft Palate Craniofac J 2011;48(6):736-40.

- Kapp-Simon KA. Psychological issues in cleft lip and palate. Clin Plast Surg 2004;31(2):347-52.
- Hunt O, Burden D, Hepper P, Stevenson M, Johnston C. Self-reports of psychosocial functioning among children and young adults with cleft lip and palate. Cleft Palate Craniofac J 2006;43(5):598-605.
- Slifer KJ, Pulbrook V, Amari A, et al. Social acceptance and facial behavior in children with oral clefts. Cleft Palate Craniofac J 2006;43(2):226-36.
- Tobiasen JM, Hiebert JM. Clefting and psychosocial adjustment. Influence of facial aesthetics. Clin Plast Surg 1993;20(4):623-31.
- Richman LC. Behavior and achievement of cleft palate children. Cleft Palate J 1976;13:4-10.
- Kramer FJ, Gruber R, Fialka F, Sinikovic B, Schliephake H. Quality of life and family functioning in children with nonsyndromic orofacial clefts at preschool ages. J Craniofac Surg 2008;19(3):580-7.
- Broder HL, Richman LC, Matheson PB. Learning disability, school achievement, and grade retention among children with cleft: a two-center study. Cleft Palate Craniofac J 1998;35(2):127-31.
- Strauss RP, Broder H. Directions and issues in psychosocial research and methods as applied to cleft lip and palate and craniofacial anomalies. Cleft Palate Craniofac J 1991;28(2):150-6.
- Broder HL. Using psychological assessment and therapeutic strategies to enhance well-being. Cleft Palate Craniofac J 2001;38(3):248-54.

- Pope AW, Ward J. Factors associated with peer social competence in preadolescents with craniofacial anomalies. J Pediatr Psychol 1997;22(4):455-69.
- Pope AW, Ward J. Self-perceived facial appearance and psychosocial adjustment in preadolescents with craniofacial anomalies. Cleft Palate Craniofac J 1997;34(5):396-401.
- Hofstra MB, Van Der Ende J, Verhulst FC. Adolescents' self-reported problems as predictors of psychopathology in adulthood: 10-year follow-up study. Br J Psychiatry 2001;179:203-9.
- 22. Varni JW, Seid M, Rode CA. The PedsQL: measurement model for the pediatric quality of life inventory. Med Care 1999;37(2):126-39.
- 23. Broder HL, Norman RG, Sischo L, Wilson-Genderson M. Evaluation of the similarities and differences in response patterns to the Pediatric Quality of Life Inventory and the Child Oral Health Impact Scores among youth with cleft. Qual Life Res 2014;23(1):339-47.
- 24. Landgraf JM, Maunsell E, Speechley KN, et al. Canadian-French, German and UK versions of the Child Health Questionnaire: methodology and preliminary item scaling results. Qual Life Res 1998;7(5):433-45.
- 25. Varni JW, Seid M, Kurtin PS. PedsQL 4.0: reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. Med Care 2001;39(8):800-12.

- Varni JW, Seid M, Knight TS, Uzark K, Szer IS. The PedsQL 4.0 Generic Core Scales: sensitivity, responsiveness, and impact on clinical decision-making. J Behav Med 2002;25(2):175-93.
- 27. American Cleft Palate-Craniofacial Association. Parameters for evaluation and treatment of patients with cleft lip/palate or other craniofacial anomalies. March, 1993. Cleft Palate Craniofac J 1993;30 Suppl:S1-16.
- Klassen AF, Tsangaris E, Forrest CR, et al. Quality of life of children treated for cleft lip and/or palate: a systematic review. J Plast Reconstr Aesthet Surg 2012;65(5):547-57.
- 29. Karp JM. Dental surgery under general anesthesia for preschool patients with orofacial clefts. Pediatr Dent 2009;31(5):426-31.
- 30. Ahluwalia M, Brailsford SR, Tarelli E, et al. Dental caries, oral hygiene, and oral clearance in children with craniofacial disorders. J Dent Res 2004;83(2):175-9.
- Bokhout B, Hofman FX, van Limbeek J, Kramer GJ, Prahl-Andersen B.
 Increased caries prevalence in 2.5-year-old children with cleft lip and/or palate.
 Eur J Oral Sci 1996;104(5-6):518-22.
- 32. Bian Z, Du M, Bedi R, et al. Caries experience and oral health behavior in Chinese children with cleft lip and/or palate. Pediatr Dent 2001;23(5):431-4.
- Paul T, Brandt RS. Oral and dental health status of children with cleft lip and/or palate. Cleft Palate Craniofac J 1998;35(4):329-32.
- American Academy of Pediatric Dentistry (AAPD). Definition of Early Childhood Caries (ECC). Pediatr Dent 2008;30:15.

- Ruiz LA, Maya RR, D'Alpino PH, Atta MT, da Rocha Svizero N. Prevalence of enamel defects in permanent teeth of patients with complete cleft lip and palate. Cleft Palate Craniofac J 2013;50(4):394-9.
- 36. Maciel SP, Costa B, Gomide MR. Difference in the prevalence of enamel alterations affecting central incisors of children with complete unilateral cleft lip and palate. Cleft Palate Craniofac J 2005;42(4):392-5.
- Malanczuk T, Opitz C, Retzlaff R. Structural changes of dental enamel in both dentitions of cleft lip and palate patients. J Orofac Orthop 1999;60(4):259-68.
- de Soet JJ, Bokhout B, Buijs JF, et al. Transmission of mutans streptococci between mothers and children with cleft lip and/or palate. Cleft Palate Craniofac J 1998;35(5):460-4.
- Bokhout B, van Loveren C, Hofman FX, et al. Prevalence of Streptococcus mutans and lactobacilli in 18-month-old children with cleft lip and/or palate. Cleft Palate Craniofac J 1996;33(5):424-8.
- 40. Dahllof G, Ussisoo-Joandi R, Ideberg M, Modeer T. Caries, gingivitis, and dental abnormalities in preschool children with cleft lip and/or palate. Cleft Palate J 1989;26(3):233-7; discussion 37-8.
- Gomes MC, Pinto-Sarmento TC, Costa EM, et al. Impact of oral health conditions on the quality of life of preschool children and their families: a crosssectional study. Health Qual Life Outcomes 2014;12(1):55.

- 42. Abanto J, Tsakos G, Paiva SM, et al. Impact of dental caries and trauma on quality of life among 5- to 6-year-old children: perceptions of parents and children. Community Dent Oral Epidemiol. 2014;42:385-94.
- Low W, Tan S, Schwartz S. The effect of severe caries on the quality of life in young children. Pediatr Dent 1999;21(6):325-6.
- 44. Acs G, Pretzer S, Foley M, Ng MW. Perceived outcomes and parental satisfaction following dental rehabilitation under general anesthesia. Pediatr Dent 2001;23(5):419-23.
- 45. Cunnion DT, Spiro A, 3rd, Jones JA, et al. Pediatric oral health-related quality of life improvement after treatment of early childhood caries: a prospective multisite study. J Dent Child (Chic) 2010;77(1):4-11.
- 46. Filstrup SL, Briskie D, da Fonseca M, et al. Early childhood caries and quality of life: child and parent perspectives. Pediatr Dent 2003;25(5):431-40.
- 47. Sheiham A. Dental caries affects body weight, growth and quality of life in preschool children. Br Dent J 2006;201(10):625-6.
- Casamassimo PS, Thikkurissy S, Edelstein BL, Maiorini E. Beyond the dmft: the human and economic cost of early childhood caries. J Am Dent Assoc 2009;140(6):650-7.
- Finucane D. Rationale for restoration of carious primary teeth: a review. J Ir Dent Assoc 2012;58(1):31-42.

- Pate J BR, Cohen L, Smith A. Childhood Medical Experience and Temperament as Predictors of Adult Functioning in Medical Situations. Children's Health Care 1996;24(4):281-98.
- 51. Stein RE, Bauman LJ, Westbrook LE, Coupey SM, Ireys HT. Framework for identifying children who have chronic conditions: the case for a new definition. J Pediatr 1993;122(3):342-7.
- 52. Chan KS, Mangione-Smith R, Burwinkle TM, Rosen M, Varni JW. The PedsQL: reliability and validity of the short-form generic core scales and Asthma Module. Med Care 2005;43(3):256-65.
- 53. Varni JW, Limbers CA. The PedsQL 4.0 Generic Core Scales Young Adult Version: feasibility, reliability and validity in a university student population. J Health Psychol 2009;14(4):611-22.
- AAPD. Guideline on behavior guidance for the pediatric dental patient. Pediatr Dent 2008;30(7 Suppl):125-33.
- 55. Aminabadi NA, Adhami ZE, Oskouei SG, Najafpour E, Jamali Z. Emotional intelligence subscales: are they correlated with child anxiety and behavior in the dental setting? J Clin Pediatr Dent 2013;38(1):61-6.
- 56. Larmas M. Has dental caries prevalence some connection with caries index values in adults? Caries Res 2010;44(1):81-4.
- Cappelli DP MC. Prevention in Clinical Oral Health Care. Philadelphia, Pa: Mosby Elsevier; 2007.

APPENDIX

TABLES AND FIGURES

	Parent-Reported Group (N = 79)	Self-Reported Group $(N = 23)$	
	N (%)	<u>N (%)</u>	
Child Gender	1 (70)	11(70)	
Female	28 (35)	7 (30)	
Male	51 (65)	16 (70)	
Ethnicity			
Hispanic	55 (70)	14 (61)	
Not Hispanic	24 (30)	9 (39)	
Race			
Caucasian	62 (78)	16 (70)	
Black	5 (6)	1 (4)	
Asian	1 <i>(Î)</i>	0 (0)	
Other	1 <i>(Î)</i>	1 (4)	
Not Reported	10 (14)	5 (22)	
Language			
English	35 (44)	12 (52)	
Spanish	44 (56)	11 (48)	
Insurance Type			
Govt. Subsidies	71 (90)	21 (91)	
Private Pay	6 (8)	2 (9)	
None	2 (2)	0 (0)	
Diagnosis			
Cleft lip & palate	71 (89.8)	20 (87)	
Cleft lip only	2 (2.7)	0 (0)	
Cleft palate only	6 (7.5)	3 (13)	

Table 1. Sociodemographic and Clinical Characteristics of Study Groups

	Parent-Reported Data			Self-Reported Data		
	PedsQL Psychosocial Summary Score	PedsQL Total Score	Number of Surgeries	PedsQL Psychosocial Summary Score	PedsQL Total Score	Number of Surgeries
dmft Score	0.006*	0.022*	0.052	0.649	0.833	0.265
Frankl Score	0.639	0.881	0.489	0.002*	< 0.000*	0.459

Table 2. Summary of Spearman's Rank Test Correlations Results

* denotes correlations that are statistically significant at p < 0.05.

Figure 1. PedsQL Psychosocial Summary score distribution for parent-reported group (N = 79)

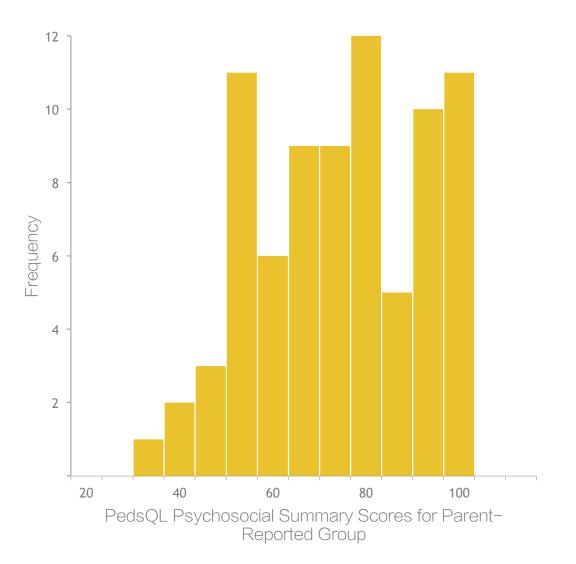


Figure 2. PedsQL Psychosocial Summary score distribution for self-reported group (N = 23)

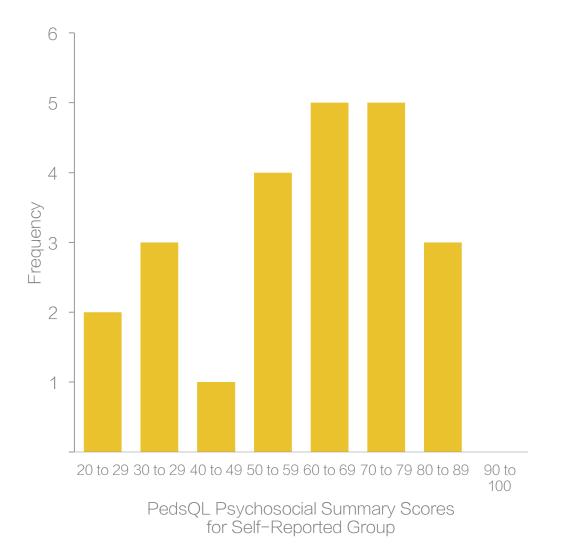


Figure 3. Surgical history distribution for parent-reported group (N = 79)

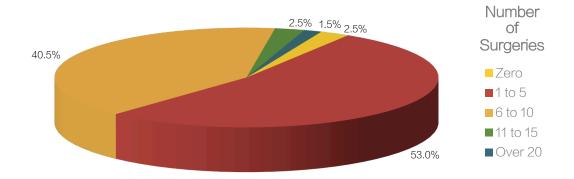


Figure 4. Surgical history distribution for self-reported group (N = 23)

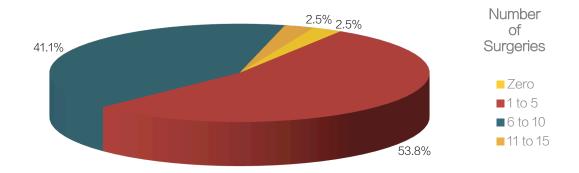


Figure 5. Caries distribution of parent-reported group (N = 79)

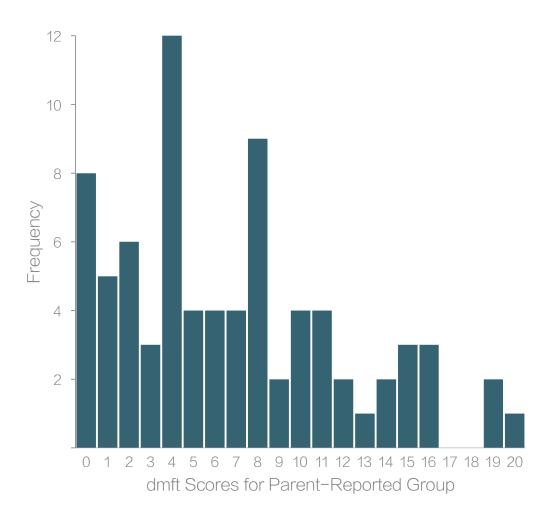


Figure 6. Caries distribution for self-reported group (N = 23)

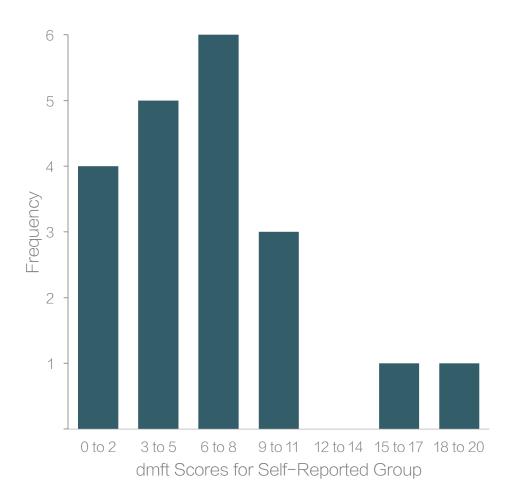


Figure 7. Distribution of Frankl scores for parent-reported group (N = 79)

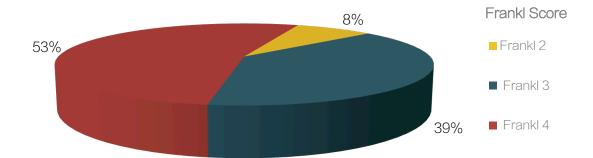


Figure 8. Distribution of Frankl scores for self-reported group (N = 23)

