QUALITY OF LIFE ASSOCIATIONS WITH CARIES EXPERIENCE AND

BEHAVIORAL CHALLENGES IN THE DENTAL SETTING AMONG

CHILDREN WITH OROFACIAL CLEFTS

A Thesis

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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. Parent-reported 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 parent-reported 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.

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INTRODUCTION

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

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 wellbeing[7] and quality of life.[7, 9] Previous studies have

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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 (PedsQL), 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 HRQOL 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 in-

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office 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

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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 self-

reported group was Spanish for 48% (N = 11) and English for 52% (N = 12). Ninety-one 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 parent-reported 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 self-reported 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 self-reporting 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 self-reported 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 self-reported 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

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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.

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Table 1. Sociodemographic and Clinical Characteristics of Study Groups

	Parent-Reported Group (N = 79)	Self-Reported Group (N = 23)
	N *(%)*	N *(%)*
Child Gender		
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 *(1)*	0 *(0)*
Other	1 *(1)*	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 2. Summary of Spearman's Rank Test Correlations Results

	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

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

Figure 1. PedsQL Psychosocial Summary score distribution for parent-reported group (N = 79)
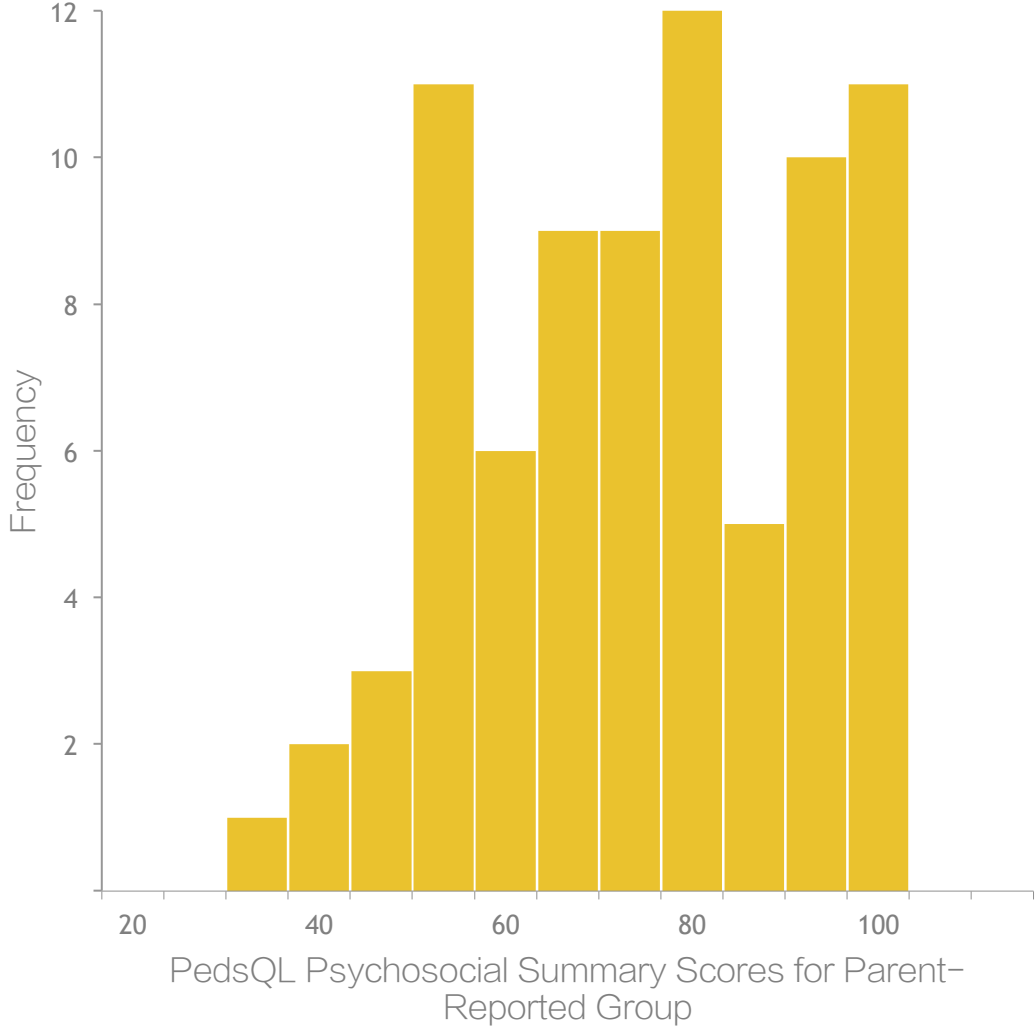
Figure 2. PedsQL Psychosocial Summary score distribution for self-reported group (N = 23)
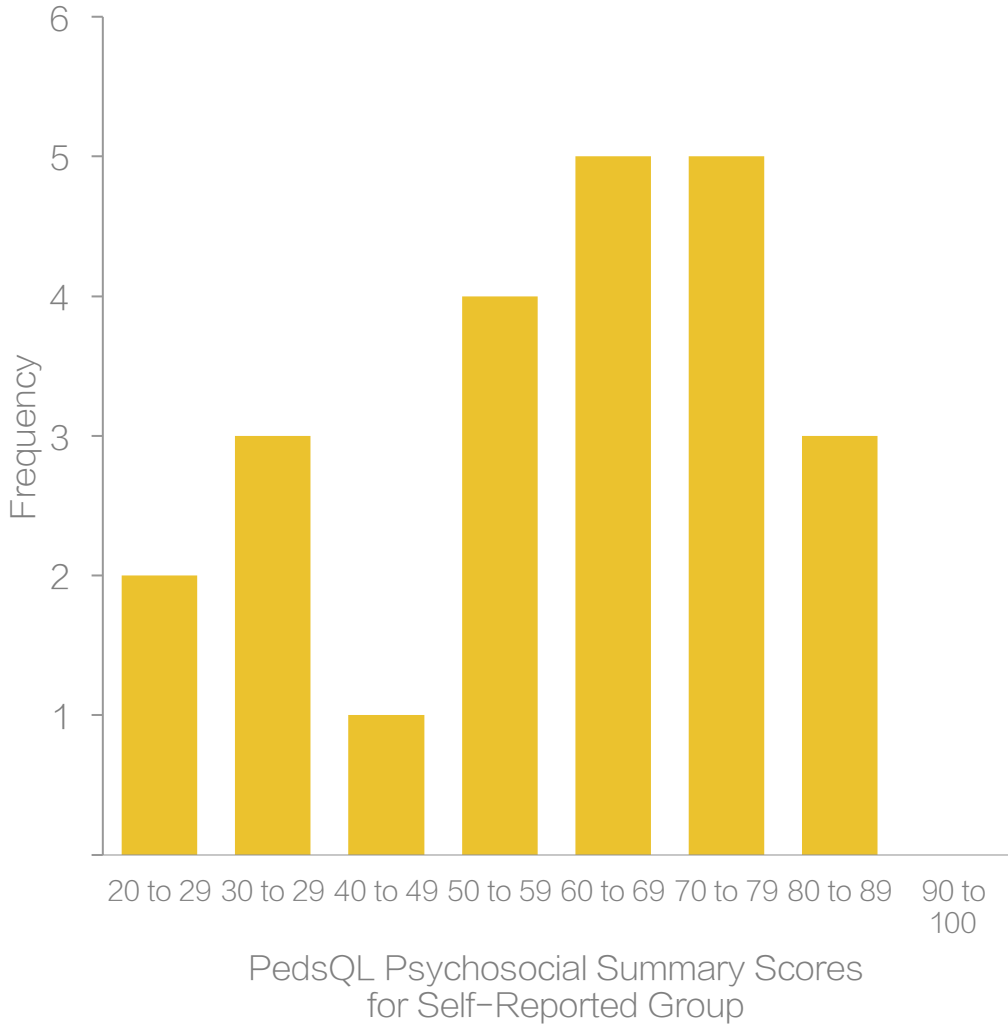
Figure 3. Surgical history distribution for parent-reported group (N = 79)
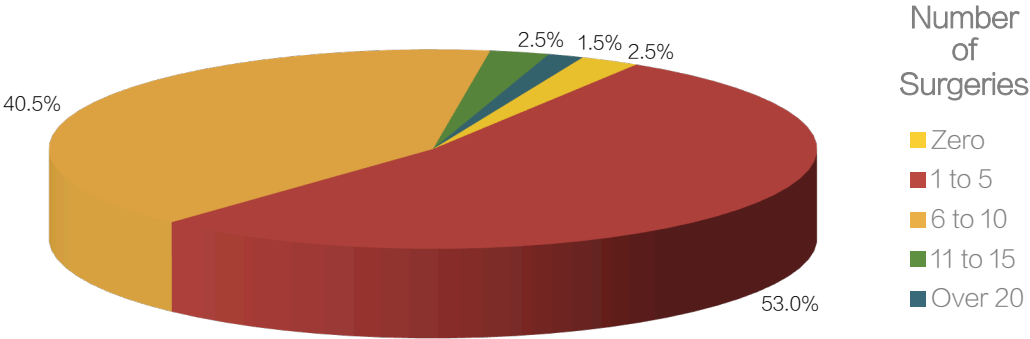
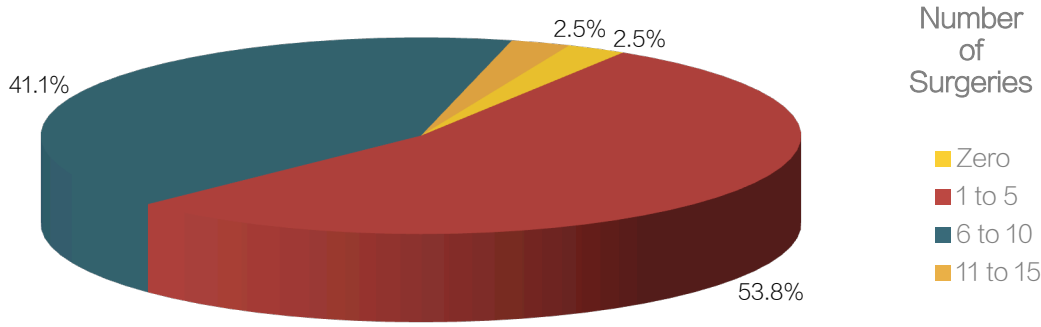Figure 4. Surgical history distribution for self-reported group (N = 23)

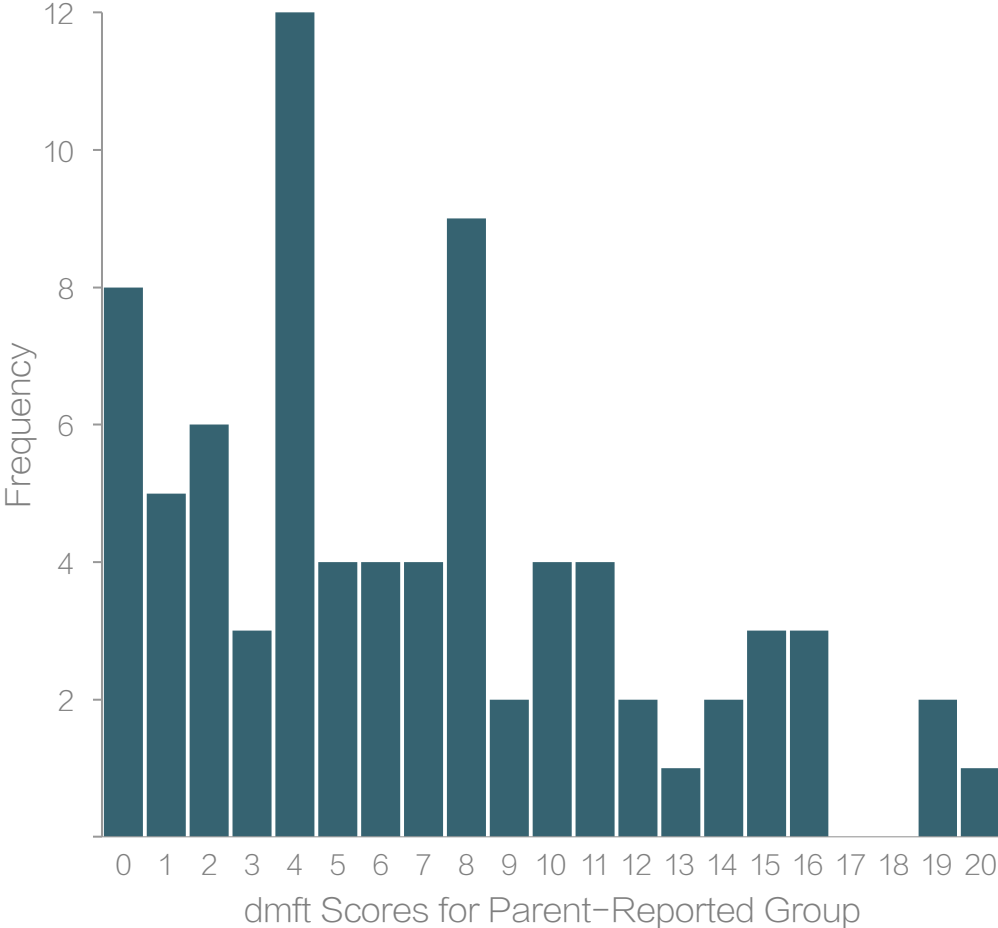Figure 5. Caries distribution of parent-reported group (N = 79)

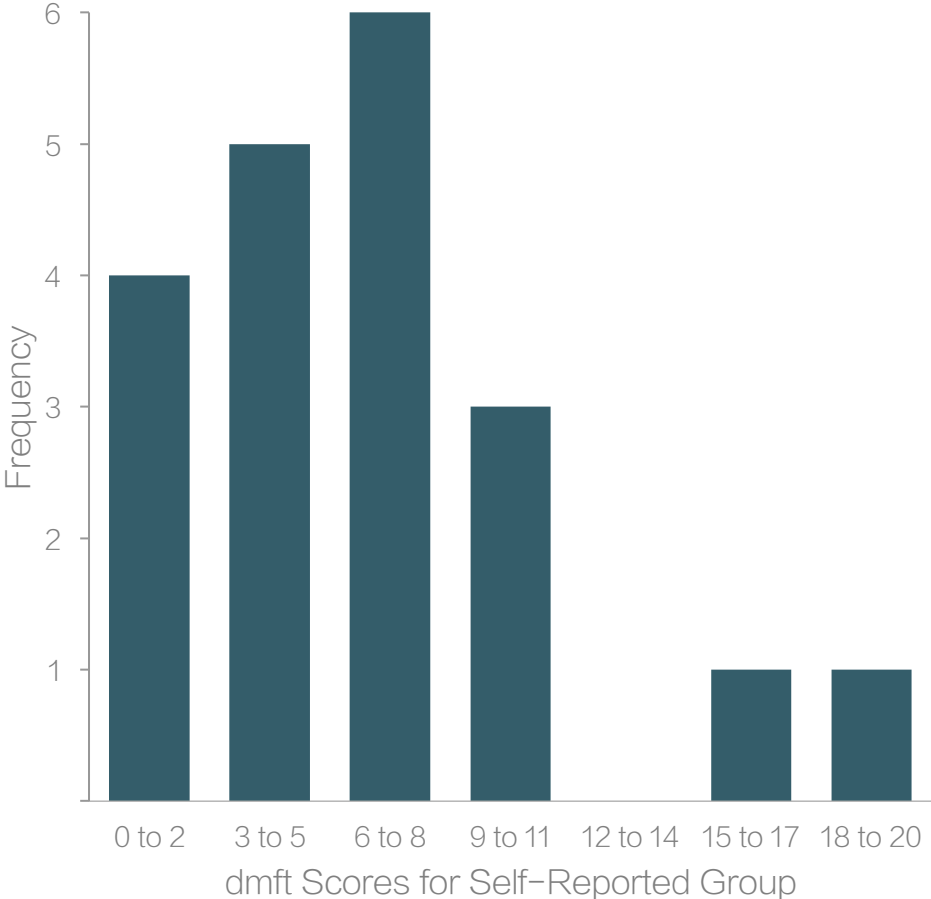Figure 6. Caries distribution for self-reported group (N = 23)

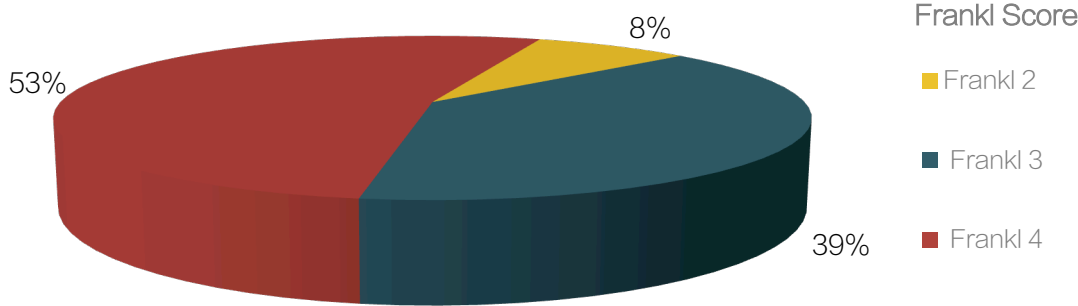Figure 7. Distribution of Frankl scores for parent-reported group (N = 79)

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