EFFECTS OF SURGICAL INTERVENTION ON PARENT REPORTED QUALITY OF LIFE IN CHILDREN AND ADOLESCENTS WITH CRANIOFACIAL CONDITIONS

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DEDICATION

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Abstract

BACKGROUND: This study compared parent-reported quality of life pre- and post- surgery in children with craniofacial conditions. Many psychosocial difficulties associated with craniofacial conditions have been identified in the literature; however, research has not yet looked specifically at parent reports of patients' quality of life and psychosocial functioning before and after surgery.

OBJECTIVE: The aim of this study was to examine changes in parent-reported quality of life over time in relation to surgical intervention in children and adolescents with craniofacial conditions.

DESIGN: A retrospective chart review was conducted of patients seen in a multidisciplinary craniofacial team clinic. Data were examined for two time points: initial visit and follow-up visit to team clinic. Participants included in the chart review were patients seen twice by psychology in team clinic between March 2011 and August 2014, with PedsQL™ ratings from parents at both time points. Patients ages 0 to 23 years were eligible for inclusion in the sample. Data collected from patient charts included demographic information, medical and surgical history, and scores from the PedsQL™ ^{TM} 4.0 Generic Core Scales.

RESULTS: Parenting stress reported at follow-up had a significant relationship with parent reported quality of life reported at follow-up for this sample; however, surgery was not a significant predictor of outcome quality of life.

DISCUSSION: Current parenting stress and initial reports of quality of life may be important variables for clinicians to consider when working with the craniofacial population. Surgery should be investigated further in terms of its impact on quality of life in relation to other psychosocial variables.

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LIST OF ABBREVIATIONS

CLP- Cleft Lip and/or Palate

QoL- Quality of Life

OHRQoL- Oral Health-Related Quality of Life

ABG- Palate Alveolar Bone Graft

PedsQL™ - Pediatric Quality of Life Inventory

CHAPTER ONE

Introduction

While quality of life measures have been an area of focus in the craniofacial literature for several years now, studies on quality of life after surgical intervention in this population have only recently begun to emerge. One of the most common craniofacial conditions is cleft lip and/or cleft palate (CLP). Isolated cleft lip occurs at a rate of about 1 in 1,574 births, with cleft lip and cleft palate co-occurring in about 1 in 940 births (CDC, 2013). Among birth defects in the United States, isolated, or non-syndromic clefts, are the most common. Isolated clefts do not accompany another birth defect or known genetic syndrome (CDC, 2013). Asian and Native American populations have the highest reported birth prevalence rates of clefts (Dixon, Marazita, Beaty, & Murray, 2011).

In addition to isolated CLP, many other complex craniofacial diagnoses exist with co-occurring complications that may affect children's functioning. Children with orofacial clefts and other complex craniofacial conditions may experience a wide range of concurrent problems, including developmental delays, feeding concerns, dental and hearing problems, and psychosocial issues during childhood and adolescence (Kapp-Simon, 1995; Strauss & Broder, 1988). Because of the broad spectrum of these conditions, the severity of impact on functioning varies widely (Hunt, Burden, Hepper, & Johnston, 2005). Among the entire craniofacial population, 30% experience psychosocial or developmental problems associated with their condition (Schutte & Murray, 1999). Most commonly, the psychosocial concerns associated with these diagnoses include bullying, anxiety, and self-image problems (Kapp-Simon, 2006).

CHAPTER TWO

Review of the Literature

Medical Background

Cleft lip and palate (CLP) are the most common birth defects in children (CDC, 2013). These diagnoses are caused by many variables, including genetic and environmental factors (CDC, 2013). Common environmental factors involved in the development of craniofacial conditions include nutritional deficiencies, teratogens, maternal smoking or alcohol consumption, or pesticides (Dixon, Marazita, Beaty, & Murray, 2011; Little, Cardy, & Munger, 2002). CLP develops as a result of problems with fusion of tissue during the 4^{th}-7^{th} week of pregnancy (CDC, 2013). Around 3,000 infants in the United States are born with isolated cleft palate and approximately 4,437 babies are born with a cleft lip with or without a cleft palate each year (CDC, 2013). Approximately 70% of all cleft cases are non-syndromic, with the other 30% attributed to chromosomal anomalies or Mendelian disorders (Jones, 1988; Schutte & Murray, 1999). Non-syndromic conditions are characterized by the absence of additional developmental or physical problems.

It is common for children with CLP to experience a number of medical complications related to their diagnosis including feeding difficulties, with or without failure to thrive; chronic ear infections; need for repeated operations to correct functional and aesthetic concerns regarding their cleft; dental concerns; and speech difficulties (Kapp-Simon, 2006). Infants with an orofacial cleft often experience feeding difficulties due to difficulty maintaining adequate suction on a nipple or bottle (Clarren, Anderson, & Wolf, 1987; Ward et al., 2013). Children with CLP may experience dental or orthodontic concerns, such as missing, crooked, or small teeth (Ward et

al., 2013). Children with CLP are also at a higher risk for developing hearing and language problems related to their diagnosis (Kapp-Simon, 2006). Along with medical risks, children with CLP may have associated functional limitations related to their diagnosis. One of the most common functional limitations associated with CLP is speech and language impairment. This limitation can be due to missing or misplaced teeth, constricted maxillary arch form, skeletal discrepancies, oronasal fistulas, and velopharyngeal incompetence (Gorlin, Cohen, & Hennekam, 2001). Children with CLP are also at an increased risk for ear infections when compared to the general population. Research has shown that almost 90% of children with CLP develop some type of ear disease caused by buildup of fluid in the inner ear (Doyle, Cantekin & Bluestone, 1980; Schonweiler, Schonweiler & Schmelzeisen, 1994). Along with structurally-based speech difficulties, children with CLP may also experience language delays related to chronic ear infections and resulting hearing impairment in infancy (Sheahan, Miller, Earley, Sheahan, & Blayney, 2004). Children with speech and language delays may experience delayed acquisition of early babbling skills, difficulties with speech intelligibility, and poor communication skills (Persson, Elander, Lohmander-Agerskov, & Soderpalm, 2002). Continued problems with speech may result in ongoing speech therapy and additional surgery to address velopharyngeal dysfunction (Persson et al., 2002). A diagnosis of CLP is also associated with a higher risk of developmental problems such as learning disorders and poor academic functioning (Broder, Richman, & Matheson, 1998). Strauss and Broder (1993) found that individuals with CLP are 30-40% more likely than peers without a CLP to experience a learning disorder and are at a slight risk for intellectual disability (6%).

Associated Craniofacial Conditions.

Examples of genetic diagnoses with a craniofacial component include Treacher-Collins syndrome, Crouzon Syndrome, and Wolf-Hirschhorn syndrome. Prevalence rates for these diagnoses range from 1 in 25,000 to 1 in 50,000 in live births (Genetics Home Reference, 2012; Gorlin, Cohen & Hennekam, 2001). Children with genetic diagnoses involving an orofacial cleft are at a greater risk of developing additional physiological problems, including hearing loss, vision problems, and cardiac anomalies (Heike & Hing, 2009). Children with these diagnoses typically undergo repeated surgeries to address a number of functional and aesthetic concerns. Commonly associated physical anomalies include mandibular hypoplasia, facial asymmetry, ear malformations, hearing loss, eye malformations, cardiac or renal anomalies, and cervical spine anomalies (Heike & Hing, 2009).

Treacher-Collins, Wolf-Hirschhorn, and Crouzon syndromes each require treatment for a wide variety of concerns associated with each diagnosis. Each of these syndromes typically involve CLP as part of the clinical presentation. Children with complex craniofacial conditions related to a genetic syndrome tend to have more widespread and significant medical problems related to their diagnosis than children with isolated, non-syndromic CLP (Bemmels et al., 2013; Pruzinksy, 1992). Children with these diagnoses require a multidisciplinary treatment approach from birth through adulthood because of the number and variety of problems associated with their diagnosis (Chang & Steinbacher, 2012; Genetics Home Reference, 2012; Turvey, Long & Hal, 1979). Research examining genetic syndromes that involve CLP continues to be an area of interest to help determine appropriate treatment options for patients with these syndromes.

Surgical Intervention

Surgical intervention is a key component of the management of craniofacial conditions, including the management of cleft lip and palate. A variety of surgeries aim to restore

functionality and form for patients with CLP. Factors which may impact surgical decision-making include the presence of other medical conditions, facial growth, and speech development (Kosowski, Weathers, Wolfswinkel, & Ridgway, 2012). Medical treatment of orofacial clefts usually occurs in two phases, with the first phase correcting the cleft and the second focusing on making the area more symmetric and aesthetically pleasing (Rohrich, Love, Byrd, & Johns, 2000). Current research has shown that early surgical intervention for CLP before the age of 5 years is beneficial for speech development but may inhibit facial growth (Kosowski et al., 2012).

After detection of CLP, either by ultrasound or newborn screening, individuals with these conditions routinely undergo closure of the cleft lip at 2 to 3 months of age via a surgical procedure also known as cheiloplasty (American Cleft Palate and Craniofacial Association [ACPA], 2007). The main goal of the lip closure is to promote satisfactory lip function and to improve the appearance and symmetry of the lip. It is also common for lip closure to improve the physical characteristics of the nose (Rohrich et al., 2000). At 6 to 18 months, a surgical procedure known as a palate closure, or palatoplasty, occurs. For children with cleft palate, this procedure assists them with developing normal speech and reducing eating and drinking complications. Children with an unrepaired cleft palate commonly have difficulty producing speech sounds due to air emission through the nose and often experience nasal regurgitation with certain foods and textures (ACPA, 2007). After palate closure, children are better able to articulate a range of sounds, thus improving their speech intelligibility (Murthy, Sendhilnathan, & Hussain, 2010). Palatal fistula, persistent velopharyngeal insufficiency, and sleep apnea are some of the most common complications after surgical repair of the cleft palate (Kosowski et al., 2012).

Along with initial surgical procedures, children with CLP may require additional surgeries and specific interventions, such as speech therapy, to increase speech development. With regard to speech development, children with a cleft palate may have difficulties resonating sounds and keeping air from escaping through the nose when speaking (ACPA, 2007). If problems with palate closure occur after a child's cleft palate is corrected surgically, they are at a higher risk of errors in their speech. Consequently, cleft palate may require secondary surgery to correct velopharyngeal insufficiency (VPI), a condition characterized by persistent hypernasal speech (ACPA, 2007). This condition tends to appear around 2 to 3 years of age, but can present later in childhood as well, and most frequently occurs in children with an existing cleft palate diagnosis. Surgeries associated with speech intervention in patients with CLP aim to assist with proper closure of the velopharyngeal sphincter to reduce air escape through the nose during speech. Technique and timing depend on the surgeon's preference and also depend on whether a child has airway problems or an extremely wide palate. There are a number of surgeries that may be used to correct this condition, such as posterior pharyngeal flap, pharyngoplasty, augmentation of the posterior pharyngeal wall, and speech prosthesis (ACPA, 2007).

Children with CLP may also require nasal reconstruction and lip revision prior to kindergarten. Surgeries during this age are intended to increase functionality of the lip and nose and to improve facial appearance during a time of increased peer interaction (Kosowski et al., 2012). School aged children may also undergo dental reconstruction or an alveolar cleft bone graft completed by an orthodontist (ACPA, 2007).

During adolescence, facial growth completes its final stages. During this time, patients with cleft palate may develop maxillary retrusion, a condition in which the upper jaw is posterior to its normal position (ACPA, 2007). This condition may require jaw surgery that aims to align

a child's dental arches. After jaw surgery, a final surgical procedure known as a septorhinoplasty may be needed to improve a child's breathing and nasal aesthetics. A septorhinoplasty improves the appearance of the nose and removes any internal obstructions that may be interfering with breathing (ACPA, 2007). Surgical intervention for CLP and other craniofacial conditions is usually complete after adolescence.

With regard to the interaction between surgery and psychosocial functioning, Millar and colleagues (2013) recently found that scarring and asymmetry related to surgery and facial differences are associated with lower self-esteem and anxiety. Adolescents with CLP have been found to perceive a main goal of surgery to be decreasing the stigma related to facial differences (Tiemens, Nicholas, & Forrest, 2013). Within the literature, findings are mixed regarding the differences in psychosocial outcomes for patients with visible and invisible conditions (Millar et al., 2013). Broder and Strauss (1989) found that individuals with surgically repaired cleft lip reported lower self-esteem, but they also noted that children with invisible craniofacial conditions (e.g., isolated cleft palate) also reported low self-esteem when compared to the general population. Thus, it is important for children with craniofacial conditions and their families to have realistic expectations and adequate social and coping skills in order to maximize psychosocial outcomes post-surgery (Turner et al., 1997).

Associated Psychosocial Factors with Cleft Lip and/or Palate

Along with the physical problems and surgical interventions associated with orofacial cleft diagnoses, individuals with CLP may experience a variety of psychosocial difficulties from infancy to adulthood. Research confirms that children and adults report facial disfigurements to be the least desirable type of disability (Pillavin et al., 1975; Richardson, Goodman, Hastorf, & Dornbusch, 1961). The literature in this area has also suggested that children who are at risk for

psychosocial concerns related to a craniofacial condition should receive intervention to promote their psychosocial well-being (Hunt, Burden, Hepper, Stevenson, & Johnston, 2005). In a study by Hunt et al. (2005), patients with CLP expressed concerns related to their facial appearance, specifically their nose, teeth, lips, and scars. Children and adolescents with CLP are also at an increased risk for experiencing problems with low self-esteem, anxiety, impairments in speech and language, feeding problems, self-image concerns, and bullying. Strauss and colleagues (1988) found that 60% of adolescents with CLP had some dissatisfaction with their facial appearance.

Psychosocial risks may occur and usually vary at different developmental stages for children with CLP. Regarding psychosocial difficulties in infancy, the literature has primarily focused on the family's experience, as well as stress related to medical treatment. Bradbury and Hewison (1994) found that families may experience stress related to overall treatment planning and may be overwhelmed by the amount of information they receive about their child's diagnosis. Upon learning their child has a cleft diagnosis, parents are often exposed to an abundance of new information from physicians, family, friends, and the internet. Along with initial stress related to CLP diagnosis, parents may experience difficulties cleaning and managing their child's surgical site, concerns about ongoing treatment, and fear of hospitalization (Drotan, Baskjewicz, Irvin, Kennell, & Klaus, 1975). Initial resiliency, flexibility, and strong emotional health of parents may strongly influence a child's adaptation throughout his/her life (Kapp-Simon, 1995). Along with parent resiliency, social support is an important resource for families of a child with CLP (Tiemens, Nicholas, & Forrest, 2013).

Once children with a craniofacial condition reach school age, they may have problems coping with the pressures of childhood and entering school (Kapp-Simon, 1986) as well as

adjusting to the school environment (Pope & Snyder, 2005). This period of development is very important; during this time children learn about social environments and making and maintaining relationships. Richman and colleagues (2012) found that psychosocial and behavioral difficulties experienced while entering the school environment may affect behavior, self-concept, and socio-emotional adjustment. Older school-age children with CLP may also have challenges academically (Kapp-Simon, 1986) and are at increased risk for learning disabilities (Broder, Richman, & Matheson, 1998). Additionally, they seem to have difficulties with peer interactions and relationships (Kapp-Simon, 1986), as well as mood and behavior concerns, including depression (Richman & Millard, 1997). Children with a craniofacial condition may also be at an increased risk for bullying because of the presence of facial differences and inhibition in social interactions (Hunt et al., 2005). Turner and colleagues (1997) found that 60% of children with a cleft reported being teased or bullied about their appearance and speech.

Adolescence and early adulthood marks a time of significant developmental transition for individuals in the general population, as well as for patients with craniofacial conditions (Broder, Richman & Matheson, 1998). Adolescents with CLP tend to exhibit more distress than peers without cleft when it comes to their appearance and how their peers view them (Richman & Millard, 1997). Turner and colleagues (1998) reported that a group of 15-20 year olds with CLP reported that self-confidence was very much affected by their facial difference. Subsequently, Hunt and colleagues (2005) found that adolescents with CLP may also be more depressed, less socially skilled, and more withdrawn than their peers. In context of this distress, adolescents with these diagnoses are at increased risk for not achieving in school or acting out (Richman & Millard, 1997). Specifically, increased levels of internalizing and externalizing behavior

problems have been found in adolescents with a cleft in comparison to adolescents without a cleft.

In summary, the literature has shown that many children and adolescents with a cleft encounter difficulties related to the social and emotional consequences of having a facial difference. Psychosocial concerns appear to often persist despite adequate social support and positive outcomes from surgical intervention.

Associated Psychosocial Factors with Other Craniofacial Conditions

Individuals with complex craniofacial conditions are also at an increased risk for experiencing psychological stress and poorer quality of life related to their facial difference (Pruzinsky, 1992). Complex craniofacial syndromes tend to involve not only greater degrees of facial disfigurement, but are also often associated with negative social and psychological outcomes (Bemmels et al., 2013). Because of possible co-occurring diagnoses, children with a more complex craniofacial condition are at a higher risk of developing psychiatric illnesses and have an increased incidence of suicide (Berger, 1973; Christensen & Mortensen, 2002). Individuals with a complex craniofacial condition also report social interactions to be difficult and tend to report higher rates of loneliness and social isolation (Bull & Rumsey, 1988). However, as is the case in patients with non-syndromic CLP, children with a complex craniofacial condition and high psychological resilience may be able to better withstand the difficulties associated with their diagnoses (Barden, 1990).

Parenting Stress

The transition to becoming a parent can be a stressful time in life (Menaghan, 1998). Stress is expected when beginning this new role, but is usually accompanied by positive emotions, such as joy and pleasure. Unexpected outcomes, such as a diagnosis of CLP, may

cause a family to experience additional stress and disruptions in the process of adapting to new parenthood (Despars et al., 2011). As it relates to CLP, high levels of parenting stress are not universal. For example, Pope and colleagues (2005) found that healthy parental adaptation and coping related to their child's diagnosis was linked to children's psychosocial functioning.

Along with individual stress, parents of children with CLP may experience emotional reactions such as confusion, denial, distress, and guilt related to their child's diagnosis (Bradbury & Hewison, 1994). During initial treatment, parents of children with CLP may have difficulty adjusting to their child's diagnosis and initiating strong attachment within the early parent-infant relationship under stressful circumstances (Despars et al., 2011). Research has indicated that parents may perceive their child with CLP as more vulnerable and needing more protection than a child without a cleft (Despars et al., 2011). Research examining early parent and child interactions and attachment has found that overprotective parenting may contribute to an anxious attachment style (Rholes, Simpson, & Blakely, 1995). Insecure or anxious attachment styles are commonly associated with negative outcomes including decreased resilience, guilt, complicated parental grief reactions, and marital stress (Bowlby, 1969). Because diagnosis of a cleft in a child can potentially be a stressful and frightening event for parents, it is important to address parents' well-being during treatment (Bradbury & Hewison, 1994).

Quality of Life

Quality of life (QoL) has been defined as an "individual's view of their cultural position in life and the values that they live by, their goals, expectations, concerns, and standards" (World Health Organization Quality of Life Group, 1994, p. 41). Quality of life has received increased attention in the craniofacial literature in recent years (Edwards et al., 2005; Patrick et al., 2007; Topolski, Edwards, & Patrick, 2005; Ward et al., 2013). In clinical work with the craniofacial

population, mental health clinicians are often focused on addressing quality of life issues, among many other common areas of concern (Patrick et al., 2007).

More recently, Oral Health Related Quality of Life (OHRQoL) has been researched as a measure of psychosocial adjustment and outcomes in the craniofacial population. Ward and colleagues (2013) created a questionnaire called the Child Oral Health Impact Profile that assesses oral health, functional well-being, social-emotional well-being, school environment, self-image, treatment expectancy, and global health. This questionnaire includes items that address positive and negative variables that affect children's overall quality of life. Using this questionnaire, Ward and colleagues (2013) found that children with orofacial clefts report more problems with functional and social-emotional well-being and overall quality of life than controls, with the greatest impact evident in 15- to18-year-olds. Research has also shown that multidisciplinary teams that prioritize evaluating and addressing quality of life are better able to assess impairments in functioning and overall burden of care caused by patients' medical conditions (Austin et al., 2010).

Use of Parent Proxy Reports.

Patient and parent-proxy reports can be useful in evaluating how children with a craniofacial condition function psychosocially. The literature suggests that parent reports should be used when the child is unable to answer questionnaires related to health related quality of life (Varni, Limbers, & Burkwinkle, 2007). Examiners should be aware of potential reporting bias when interpreting parent report measures (Varni et al., 2007). For example, mothers may report lower quality of life for their child with a cleft compared to the child's own self report, possibly because mothers may experience a significant amount of stress related to their child's medical situation or may be strongly invested in their child's status as a patient with a medical condition

(Broder & Strauss, 1989). An additional concern with parent-proxy reports is that parents may not be aware of the amount of bullying their child experiences and may underreport some issues (Turner, 1997). However, children may also underreport problems on self-report measures to put forth a more positive representation of themselves or to preserve their self-esteem (Thompson & Kent, 2001). According to Thompson and Kent (2001), children with self-image concerns may be using strategies to maintain a sense of acceptability or protect against the judgment of others. Considering the aforementioned concerns regarding both parent reports and patient self-reports, reporting bias is likely present to some degree for both parent and self-reported information, and it is important to consider possible biases when interpreting any measure of a patient's quality of life.

Limitations of Current Research

Current research has identified many psychosocial difficulties associated with craniofacial conditions. Current research has also shown that overall quality of life in children can be greatly affected by a facial difference related to CLP (Patrick et al., 2007). Research has not yet looked specifically at quality of life before and after surgery. Another limitation in existing research is that some prior studies in this area utilized samples of only males or only females rather than samples that included both genders (Tiemens et al., 2013; Topolski et al., 2007). It is important to look at both genders to assess differences in how children cope with these conditions and to improve the generalizability of findings. Additionally, the validity of some prior studies may be questionable because of small sample sizes. For example, many studies examining psychosocial functioning in patients with craniofacial conditions used sample sizes of 7 to 10 (Tiemens et al., 2013; Topolski et al., 2007).

Along with issues in sampling and the generalizability of prior studies, inconsistencies in methodology exist for many studies addressing the craniofacial population. The current literature also suggests mixed findings regarding how children with craniofacial conditions cope with their diagnoses from childhood to adolescence. Some studies have found no difference between psychosocial functioning for craniofacial samples and control groups, while other studies have identified significant differences in functioning for specific psychosocial variables (Hunt et al., 2007). The inconsistency in findings related to psychosocial difficulties in the craniofacial population may be attributed to several factors, including the use of broad measures of psychosocial functioning versus symptom-specific measures. Additionally, some researchers have used a control group of non-cleft patients while others made comparisons between patients with different types of craniofacial conditions. As many patients with CLP demonstrate resilience with regard to coping and adjustment (Berger & Dalton, 2009), it may be most useful to identify factors that differentiate patients who experience psychosocial difficulties from those who do not. Research has not yet addressed variables which may account for significant differences in psychosocial functioning among patients with craniofacial conditions.

Current research has not addressed the effects of surgical intervention on quality of life and psychosocial functioning in the cleft population. Specifically, the relationship between surgical intervention and quality of life should be examined and factors which affect this relationship should be researched to help identify psychosocial risks and benefits associated with surgery, as well as potential interactions between surgery, other psychosocial variables, and quality of life for patients with CLP.

Current Study

Based on the extant literature, it appears that children with a craniofacial condition are at a higher risk of developing psychosocial problems than children in the general population. Surgical intervention is necessary for most children with CLP to address functional and aesthetic concerns. Children undergoing surgery and their families experience a variety of stressors over a long duration of time. Individuals with a good support system may cope effectively with stressors associated with their diagnosis (Baker, Owens, Stern, & Willmot, 2009). For others, self-image concerns may contribute to a variety of psychosocial problems (Kapp-Simon, 1986). This study will build on the current body of literature by addressing parent-reported quality of life in patients with CLP as it relates to surgical intervention.

Research has identified a number of variables that may contribute to overall quality of life in children with CLP. Parenting stress, diagnosis, and surgical intervention appear to be factors which may contribute to the variability of functioning in CLP patients. For example, Pope and colleagues (2005) found that parental adaptation was linked to healthier psychosocial functioning in children with CLP. In patients with CLP, surgical intervention is aimed to increase quality of life by decreasing functional limitations and/or improving aesthetic appearance (Millar et al., 2013). Considering that current literature has found a relationship between severity of diagnosis (e.g., scarring, asymmetry, visibility of facial difference) and psychosocial functioning (Thompson & Kent, 2001; Ward et al., 2013) it is likely that diagnosis also plays a role in the relationship between parenting stress, surgical intervention, and parent-reported quality of life. Further, given that different surgical procedures to address CLP yield different results in terms of visibility of outcomes, surgical visibility may also play a role in the relationship between surgery and outcome QoL. The goal of this study is to examine the relationship among

these variables in an effort to gain greater understanding of the impact of surgery on patients' quality of life.

Aim 1:

Identify variables contributing to changes in parent-reported quality of life over time in patients with craniofacial conditions.

Hypothesis 1:

Follow up parent reported scores on the PedsQL™ would be predicted by diagnosis and parenting stress, after controlling for initial parent reported PedsQL™ scores.

Aim 2:

Compare parent-reported quality of life for patients with craniofacial conditions who received surgical intervention to those who did not.

Hypothesis 2:

Patients who received surgical intervention between initial and follow-up visits will have higher parent-reported PedsQL™ scores at follow-up than those who did not have surgery, after controlling for baseline PedsQL™ scores and time between visits.

Aim 3:

Examine the role of surgical intervention in changes in parent-reported quality of life over time.

Hypothesis 3a:

Interim surgery will predict higher parent-reported quality of life scores, after controlling for parenting stress, diagnosis, and initial parent-reported PedsQL™ scores.

Hypothesis 3b:

Interim surgical procedures with more immediately visible results will be associated with higher parent-reported PedsQL™ scores at follow-up than procedures with less immediately visible results.

CHAPTER THREE

Method

The present study was conducted via a retrospective chart review of data collected for patients seen in the multidisciplinary craniofacial team clinic in a pediatric hospital in an urban area from March 2011 through August 2014. Each patient received screening and follow-up evaluations by a psychologist or supervised psychology trainee during the two consecutive team clinic visits. Parents also completed written parent-report measures (craniofacial questionnaire and PedsQL™) at two consecutive team clinic visits.

Participants

This study was approved by the Institutional Review Board at UT Southwestern Medical Center as an expedited study. This study utilized clinical data collected as part of standard of care, thus consent was waived. Participants included patients seen in the craniofacial team clinic at Children's Medical Center between March 2011 and August 2014. The sample included 139 participants. Caregivers were required to be able to read English or Spanish to complete the written questionnaires. Children under the age of 2 years with a parent who did not speak English were excluded, as a Spanish form of the PedsQL™ was not available at the time of this study for caregivers of patients ages 0 to 24 months. Additionally, in order to be included in this study, a parent must have completed the questionnaires utilized in this study at both initial and follow-up visits. Patients in the sample ranged in age from 0.10 years to 16.82 years.

Measures

The Pediatric Quality of Life Inventory Core Scales (PedsQL™) were used to measure health-related quality of life in children with a craniofacial condition. The PedsQL™ includes parent proxy report questionnaires. The parent proxy reports were given to caregivers of all

patients included in the sample. Three composite scores are calculated from the PedsQL™ parent proxy questionnaire: Physical functioning, Psychosocial functioning, and a Total score which combines the Physical and Psychosocial composite scores. For each of the items, parents use a 5 point Likert scale to report how frequently their child experienced specific problems within the past month. Items on the PedsQL™ focus on health, activities, feelings, school, attention/concentration, and social interactions. For the present study, the parent-reported Psychosocial score was used, as social and emotional functioning were the primary domains of interest for this study, more so than physical functioning.

Parents were also asked to fill out a 5-item questionnaire ("craniofacial questionnaire;" see Appendix A) designed by a psychologist in the clinic to evaluate domains identified in the craniofacial literature as being important factors for psychosocial outcomes. Items on the parent craniofacial questionnaire were related to children's abilities, parental characteristics, and social support. On the craniofacial questionnaire, the item "I feel overwhelmed by my child's needs" was used to measure the level of stress a parent may have experienced related to their child's diagnosis and ongoing treatment. Items were scored on a 1 to 5 scale. Two of the items on the questionnaire, including the parenting stress question, were reverse scored so that higher scores on all items were associated with better functioning, and lower scores represented poorer functioning (reverse-scored items were "I feel overwhelmed by my child's needs" and "My child shows repetitive or unusual behaviors").

Procedure

As part of standard clinical care, parents were asked to fill out the PedsQL™ and the craniofacial questionnaire at each clinic visit. Medical chart reviews also were conducted to obtain information about medical and surgical history, diagnoses, and treatment. Scores from the

PedsQL™ and craniofacial questionnaire, as well as information from patients' medical chart reviews, were entered into a clinical database.

Due to the wide age range included in the study sample and the different forms of the PedsQL™ administered for different patient ages, parent-reported PedsQL™ scores were converted to z-scores. This conversion was done to be able to compare parent-reported PedsQL™ scores across different forms of the measure for the various age groups included in the study. Z-scores were calculated separately for each age group covered by a different form of the PedsQL™ (i.e., 0 to 12 months, 13 to 24 months, 2 to 4 years, 5 to 7 years, 8 to 12 years, and 13 to 18 years) and were also calculated separately for data from each time point (initial and follow up visits).

For patients who underwent craniofacial surgery between visits, surgery type was categorized as either producing immediately visible results or producing less visible results. Surgeries included in the "immediately visible" category included cheiloplasty, rhinoplasty, lip repair, lip revision, and orthognathic surgery. Procedures included in the "less visible" category included palate alveolar bone graft (ABG), palatoplasty, pharyngeal flap, and palatal fistula closure. Patients who underwent more than one type of surgery between initial and follow-up visits were included in the "immediately visible" category if they underwent any of the procedures listed in that category, even if they also received a less visible surgery.

Analyses

Various statistical analyses were conducted to examine the relationships between variables of interest for the present study; and these analyses are described below.

Aim 1:

Identify variables contributing to changes in parent-reported quality of life over time in patients with craniofacial conditions

Hypothesis 1:

Follow up parent reported scores on the PedsQL™ were expected to be predicted by diagnosis and parenting stress, after controlling for initial parent-reported quality of life.

Analyses 1:

Multiple regression analyses were used to determine whether diagnosis, parenting stress at initial visit, and parenting stress at follow-up predicted outcome quality of life and if so, the amount of variance each independent variable contributed to the equation.

Aim 2:

Compare parent-reported quality of life for patients with craniofacial conditions who received surgical intervention to those who did not.

Hypothesis 2:

Patients who received surgical intervention between initial and follow-up visits were expected to report higher will have higher parent-reported PedsQL™ scores at follow-up than those who did not have surgery, after controlling for baseline PedsQL™ scores and time between visits.

Analyses 2:

An ANCOVA was conducted to determine group differences for parent-reported PedsQL™ scores between the groups who did and did not receive surgery, with baseline PedsQL™ scores and time between visits as covariates.

Aim 3:

Examine the role of surgical intervention in changes in parent-reported quality of life over time.

Hypothesis 3a:

Interim surgery was expected to predict higher parent-reported quality of life scores after controlling for parenting stress, diagnosis, and initial parent-reported PedsQL™ scores.

Analyses 3a:

Hierarchical regression analysis was used to determine whether surgery predicted outcome parent-reported quality of life, after controlling for parenting stress, diagnosis, and baseline PedsQL™ scores at initial visit.

Analyses 3b:

Hierarchical regression analysis was used to determine whether surgical category predicted outcome parent-reported quality of life after controlling for parenting stress, diagnosis, and baseline PedsQL™ scores at initial visit.

CHAPTER FOUR

Results

Sample

One hundred thirty-nine participants were included in the present study. Of the 139 patients, 71 were males, with over half of the sample (61.2%) identifying English as their primary language. The average age of the sample was 7.14 years (SD = 4.84), with a range of 0.10 to 16.82 years (see Table 1 for a full summary of demographic information and Table 2 for average PedsQL™ scores for each age group.)

Statistical Analyses

A one-tailed two-sample independent means t-test, equal variances assumed, revealed that significant differences were not present in demographic variables (i.e., language, race/ethnicity, gender, and insurance payor status) for the surgery vs. no surgery groups. A one-way analysis of variance for PedsQL™ scores and age range was not significant, $F(1, 139) = 1.69$, $p = .14$. $\alpha = .05$, suggesting that PedsQL™ scores did not significantly differ across the age groups included in the sample. However, one-way analysis of variance did reveal a statistically significant difference in surgical category (visible vs. less visible) by age group, $F(1, 139) = 6.97$, $p = <.001$. $\alpha = .05$. However, post-hoc analyses using the Bonferroni adjusted alpha level of .003 (.05/15) per test were not significant.

To evaluate differences between the sample and normative groups on the PedsQL™ (Varni, Seid, & Kurtin, 2001), t-tests were used to compare parent-reported PedsQL™ scores at initial and follow–up visits to previous research on healthy versus chronically ill populations (Varni, Limbers, & Burkwinkle, 2007). Scores for the study sample were significantly lower than the normative healthy sample at both the initial and follow-up visits respectively, $t(854) =$

10.32, p = .001; $t(854) = 9.24$, p = <.001. When compared to the chronically ill normative group, significant differences were not found for initial or follow-up scores respectively, $t(798)$= .87, p = .39; $t(798)$= .14, p = .88.

Multiple regression was used to examine the first hypothesis, which anticipated that initial reports of parenting stress and child's diagnosis would predict follow-up parent-reported PedsQL™ scores after controlling for initial parent-reported PedsQL™ scores. Initial PedsQL™ scores were entered as the control variable, parenting stress and diagnosis category were entered as the independent variables, and follow-up PedsQL™ score was entered as the dependent variable. The overall model was significant. In this model, initial PedsQL™ scores accounted for approximately 24% of the variance in PedsQL™ scores at follow-up, R^2 = .24, $F(1, 138)$ = 12.30, p < .001. However, neither parenting stress at initial visit, β = .07, t =.87, p =.39, nor diagnosis, β = -.02, t = -.21, p =.84, were significant predictors of follow-up PedsQL™ scores within this model.

Further regression analyses were conducted to examine the relationship between follow-up PedsQL™ scores and parenting stress reported at follow-up, after controlling for initial PedsQL™ scores. Initial PedsQL™ scores were entered as the control variable, parenting stress at follow-up was entered as the independent variable, and follow-up PedsQL™ scores were entered as the dependent variable. The overall model was significant. Within the model, follow-up parenting stress accounted for a significant amount of variance in follow-up PedsQL™ scores at the specified 0.05 alpha level, β =.19, t = 2.62, p <.01 (see Table 2 for summary of results from regression analysis of PedsQL™ scores and parenting stress at follow-up).

For the second hypothesis, an analysis of covariance was used to compare follow-up parent-reported PedsQL™ scores for patients who underwent surgery between visits and those

who did not. No difference was found between those who had surgery and those who did not, $F(1, 135) = .08$ $p = .77$. Initial scores on the PedsQL™ and days between surgery and follow-up visit were entered as covariates to control for initial quality of life reports and time from surgery to follow-up visit. The mean PedsQL™ z-score at follow-up for the group that had surgery between visits was .06, while the mean z-score at follow-up for the group that did not have surgery was -.01.

To examine hypothesis 3a, a hierarchical multiple regression analysis was conducted to identify whether interim surgery predicted follow-up parent-reported PedsQL™ scores, after controlling for initial PedsQL™ scores and parenting stress at follow-up. No significant differences were found. As diagnosis was not found to be significantly associated with PedsQL™ scores at follow up in previous analyses, it was excluded from this model. In step one of the regression analysis, initial PedsQL™ scores and follow-up parenting stress accounted for a significant amount of variance in follow-up PedsQL™ scores, $R^2 = .24$, $F(2, 136) = 21.76$, $p < .001$. In step two, the variable surgery was added to the model. Interim surgery was not a significant predictor of PedsQL™ scores at follow-up within the model itself, $\beta = .03$, $t = .33$, $p = .74$.

With regard to hypothesis 3b, an *a priori* power analysis revealed that a sample size of 40 patients who underwent surgery between initial and follow-up visits would be needed to sufficiently power this analysis. This analysis showed no significant difference between the visible and less visible surgery categories. As only 35 patients in the sample underwent interim surgery, this analysis is underpowered; however, as the relationship between type of surgery and outcome quality of life has yet to be researched in the craniofacial population, results are presented here as exploratory data. A hierarchical regression analysis was conducted for patients

receiving interim surgery to examine the relationship between surgical category (visible results or less visible results) and outcome parent-reported PedsQL™ scores. In step one of the regression analysis, initial PedsQL™ scores accounted for a significant amount of variance in follow-up PedsQL™ scores, $R^2 = .28$, F (1, 33) = 12.55, p < .001. In step two, the variable surgical category was added to the model. Surgical category was not a significant predictor of PedsQL™ scores at follow-up within this model, $R^2 = .26$, $F(2, 34) = 6.99$, $p = .262$.

CHAPTER FIVE

Discussion

The present study aimed to contribute to the literature on psychosocial concerns in children with a cleft lip and/or palate. More specifically, this study endeavored to determine the relative impact of surgery on parents' rating of children's psychosocial functioning on the Pediatric Quality of Life Inventory. A main hypothesis for this study was that parent reported child quality of life would be higher for patients who received surgical intervention than those who did not. It was also hypothesized that diagnosis and parenting stress would influence parent reported child quality of life. Lastly, the present study hypothesized that surgical visibility would influence parent reported quality of life.

An initial aim of the present study was to identify variables contributing to changes in parent-reported quality of life over time in patients with craniofacial conditions. It was hypothesized that follow-up parent report scores on the PedsQL™ would be predicted by diagnosis and parenting stress. The results for diagnosis and for baseline parenting stress did not support this hypothesis. These findings suggest that diagnostic category (i.e., cleft lip only, cleft palate only, and cleft lip/palate combined) and parenting stress reported at baseline did not play a significant role in outcome parent reports of child quality of life. However, parenting stress reported at follow-up did have a significant relationship with outcome parent reported quality of life for this sample, suggesting that current parenting stress may be more meaningful in parents' perception of children's quality of life than parenting stress more longitudinally. This finding elaborates on previous research by Pope et al. (2005) that found that for parents of a child with a craniofacial diagnosis, healthy coping and lower rates of stress related to their child's diagnosis were linked to more positive child psychosocial functioning. Within the context of the present

study, parenting stress as more of a "state" variable appears to be related to parent perceptions of their child's psychosocial functioning more so than historical reports of stress.

The second aim of the present study was to compare changes in parent-reported quality of life for patients with craniofacial conditions who received surgical intervention to those who did not. It was predicted that patients who received surgical intervention between initial and follow-up visits would have higher parent-reported PedsQL™ scores at follow-up than those who did not have surgery, after controlling for baseline parent-reported PedsQL™ scores. There was not a significant difference in outcome parent-reported quality of life between craniofacial patients who had interim surgery and those who did not. These findings suggest that parent reports of quality of life after a craniofacial surgery seem to be similar to parent reports of quality of life for children who did not undergo surgery. This finding was unexpected, as the goal of many craniofacial surgeries, especially later in childhood and adolescence, is to reduce stigma around having a facial difference (Tiemans, Nicholas, & Forrest, 2013), which could be expected to improve quality of life. Previous research on quality of life among children and adolescents with a cleft has found that quality of life ratings tend to be lower for these patients than for controls, especially in late adolescence (Ward et al., 2013). Given that the mean age for the sample in the present study was 7.14 years, more young children and school-age children were present within the sample than older adolescents. Specifically, only 15 participants in the sample were between the ages of 15 and 18 years, which is the age range associated with lowest quality of life for this population (Ward et al., 2013). It is possible that for the younger patients in this sample, parent-reported quality of life scores were not yet low enough to improve significantly after surgery, and the sample did not include enough older adolescents who underwent surgery to demonstrate a significant difference from the group who did not have surgery.

The final aim of the study was to examine the role of surgical intervention in changes in parent-reported quality of life over time. The initial hypothesis for this aim anticipated that interim surgery would predict higher parent-reported PedsQL™ scores after controlling for parenting stress, diagnosis, and initial parent reports of quality of life. The results did not support this hypothesis. Consistent with previous research, (Kapp-Simon, 1986; Richman & Millard, 1997; Turner et al., 1997), this finding suggests that surgery alone may not significantly contribute to changes in parents' perception of their child's quality of life for children and adolescents with craniofacial conditions. Parents' expectations of surgery may have affected their perception of their child's quality of life. Because there was a large range of variability in surgeries, age, and time between visits, parents of children included in the study may not have reported considerable differences in their child's quality of life. It is also possible that parents' expectations may have changed between initial and follow-up visits, which may have affected how they reported their child's quality of life at each visit. It was expected that surgery would be associated with higher parent ratings of child quality of life, given that craniofacial surgery tends to be aimed at both functional and aesthetic improvements (Rohrich et al., 2000). However, prior studies also found that children with craniofacial conditions tend to experience psychosocial difficulties such as low self-esteem, even after their facial difference has been surgically repaired (Broder & Strauss, 1989; Millar et al., 2013). The results for this hypothesis appear to support the idea that children and adolescents with a craniofacial condition may not experience significant, direct, observable improvements in quality of life as a result of surgery alone.

The last hypothesis for this study predicted that interim surgical procedures with more immediately visible results would be associated with higher parent-reported PedsQL™ scores at follow-up than procedures with less immediately visible results. Results indicated that surgical

category was not a significant predictor of outcome parent-reported PedsQL™ scores. Although this analysis was underpowered and thus potential for interpretation of these findings is limited, exploratory findings suggest that the visibility of surgical results may not be a significant factor in parents' perceptions of children's quality of life after a craniofacial surgery. This finding was surprising, given that prior research in the craniofacial population has found that visibility of facial differences is associated with negative psychosocial outcomes. Specifically, one study found that scarring and asymmetry related to surgery was linked to lower self-esteem and anxiety (Millar et al., 2013). Accordingly, it would be expected that patients undergoing a surgery that visibly alters a facial difference might experience a difference in quality of life after surgery. Results for this hypothesis may have been influenced by the way in which surgical procedures were categorized in this study. Varying degrees of risk and need for post-surgical care and follow-up are associated with different surgical procedures, and there may have been a significant amount of variability in these factors within the two surgical categories used in this study. Additionally, some patients underwent multiple surgical procedures at one time. Although it was expected that visible surgeries would have a more pronounced effect on parent-reported quality of life, it is possible that the less visible procedures had an unexpected relationship with parent-reported quality of life. Parents also may have perceived changes in quality of life after surgery differently for children and adolescents in different age groups. Further, it is possible that parents may have perceived any surgery, whether visible or less visible, as having similar effects on quality of life (e.g., parents may have perceived all surgeries as an equally important step towards completing their child's treatment plan).

Limitations

Several limitations should be considered in the interpretation of the results of this study. First, the small sample size of this study limited the number of individuals with surgery between initial and follow up visits. Subsequently, there was a large range of variability in time between visits and in time after surgery. As we were unable to collect data at the same time after surgery for each individual included in the study, we were not able to control for time between surgery and follow-up. There also was not a way that individuals could be assigned to surgery versus no surgery category, meaning that the study could not control for differences related to not having surgery. Additionally, we were not able to gather follow-up data at regular intervals after surgery. As a result, we are not able to identify differences in quality of life at different time intervals after surgery, which may be an important factor to consider.

The study sample also spanned a large age range, which may have impacted the results. Because of this large age range, parents may have reported different concerns due to various developmentally normative events in their child's life. Developmental expectations for the age range included in this study vary dramatically, and domains measured to determine quality of life are different across age groups. Children and adolescents included in the study likely had different experiences, emotional reactions, and coping strategies related to surgery and ongoing treatment as a result of developmental differences. Consequently, the study aimed to standardize quality of life scores for age groups by converting to z-scores. However, potential age range-related issues may still have been present.

There is a large variety of treatment options offered for individuals across the age range included in the sample. Initial treatment options for this population begin with increasing functionality of the nose and mouth (e.g. palatoplasty, lip revision). As children move into

adolescence and early adulthood, surgical options tend to emphasize aesthetics more so than function. Surgical procedures with more immediately visible results tend to cluster in infancy (e.g., lip repair) and later adolescence (e.g., rhinoplasty), whereas procedures with less immediately visible results tend to occur more frequently in middle childhood and school age (e.g., ABG). Naturally occurring age differences for surgical category may have introduced confounding variables into the analyses for this hypothesis. Additionally, this study did not include a quantitative measure for determining asymmetry related to a child's diagnosis and surgical intervention at initial and follow-up visits. By not having a measure to determine degree of asymmetry and degree of repair after surgery, this study was unable to measure differences related to overall severity of a child's CLP and effects of surgery.

Because of the relatively small number of patients who received surgery between initial and follow-up visits, we were not able to consider patients who received surgery specifically targeting speech intelligibility (e.g., pharyngeal flap) as a separate group. These patients were included with the "less visible" surgery group. Although speech surgery is not associated with immediate, visible results, patients undergoing speech surgery may still experience an observable improvement in speech intelligibility, which may significantly contribute to improved quality of life after surgery. Future studies should attempt to evaluate this group separately.

Additionally, the use of both parent and child reported data would have been beneficial to minimize reporting bias. Many other studies looking at psychosocial outcomes in the craniofacial population have utilized child self-report data (e.g., Millar et al., 2013). Findings in the present study, which do not align with previous research findings, may have been influenced by the sole use of parent-reported data alone. Developmental differences may also have been a limitation related to using the parent report form only. Research has found that teens tend to rely more on

friends than parents for social support (Helsen, Vollebergh, & Meeus, 2000). Consequently, parent report of adolescents' quality of life may be less accurate than parent report of quality of life for younger patients who may be more likely to express concerns and share detailed information about their psychosocial functioning with a parent.

An additional potential limitation of this study is that the geographic area where the study was held has a larger portion of Spanish-speaking families than most areas in the United States. Thus, the results of the study may not be generalizable to other geographic areas due to the differences in language and cultural factors, such as parents' perception of their child's treatment plan, prognosis, and trust in the medical system.

Patients included in the sample for this study were receiving treatment at one center at a pediatric hospital. The current craniofacial team has been at this center since early 2011; thus, patients in this sample older than 3 years of age likely received at least some of their care from a different craniofacial provider or team. It is possible that receiving some care and surgical intervention elsewhere could have affected quality of life reports for these patients as a result of differing expectations for treatment and previous experiences, which were not accounted for in this study.

Additionally, individuals in this study may have experienced some type of significant or life-changing event between initial and follow-up visits, such as entering school for the first time, experiencing abuse or significant changes in the family structure. Consequently, parent reports of child quality of life at follow-up may have been affected by these events and not surgery or other variables of interest.

Directions for Future Research

Based on the results and limitations of the present study, several recommendations can be made regarding future research in this area. First, it would be helpful to include a measure of life stressors to capture events that may occur between visits. By controlling for these variables, future studies may be able to gather more accurate information regarding a child's quality of life in relation to surgery or others areas of interest. A prospectively-designed study could evaluate quality of life at the same point in time for each patient post-surgically, which may help minimize interference from time between surgery and follow-up assessment as an extraneous variable. Additionally, a larger sample size is needed in future studies examining the relationship between different types of surgery and children's quality of life.

Future studies of quality of life in the craniofacial population should incorporate data from multiple informants, such as parent, child, and teacher, which would aid in accuracy and generalizability of findings. Additionally, use of a craniofacial-specific quality of life or psychosocial measure might capture psychosocial concerns that are most relevant to children with craniofacial conditions and their parents (e.g., bullying, self-image, and speech intelligibility) and might yield different results than a more general, broad measure of quality of life such as the PedsQL™. In addition, future studies should consider using a full parenting stress questionnaire to look at a variety of domains related to stress a parent may feel regarding their child's functioning. Furthermore, studies in this area should examine the effects of different types of surgery on quality of life to determine specific differences in immediately visible and invisible surgery types. For example, future studies should consider evaluating differences in quality of life after initial cleft lip repair, speech surgery, rhinoplasty and other aesthetic surgeries in later adolescence, and ABG and other surgeries without immediate visible

or speech results. Furthermore, future studies should aim to include a quantitative of asymmetry related to a child's diagnosis and results from surgery. Lastly, researchers should examine the effects of surgery on quality of life within specific age groups to determine whether differential effects are observed for different developmental stages.

Conclusion

Even though most hypotheses for this study were not supported, these findings do offer contributions to the existing body of literature within the craniofacial community. In this study, surgery proved to not be a significant a predictor of outcome ratings of quality of life as anticipated. Rather, a number of factors likely contribute to psychosocial outcomes for this population, including developmental concerns, language delays and communication problems, and school and family functioning (Kapp-Simon, 1995). These variables should be further investigated in terms of possible interaction effects with surgery on quality of life. Additionally, this study suggests that current parenting stress may be a variable of interest to clinicians working with families of patients with craniofacial conditions.

As expected, initial parent reports of quality of life also appear to be a factor that may be of importance to clinicians working with this population. Given that initial parent-reported PedsQL™ scores were consistently a significant predictor of parent-reported PedsQL™ scores at follow-up, it may be beneficial for clinicians to routinely screen for quality of life as an indicator of likely quality of life at subsequent visits. Additionally, it would be beneficial for both clinicians and researchers to attempt to identify variables which may maintain low scores on measures of quality of life across time.

Parents of children with a cleft may experience a number of stressors related to their child's diagnosis, and may develop different views of their child's quality of life depending on

external factors and treatment stages. Although the overall results of this study were not significant, the effects of surgery on parent-reported quality of life within this craniofacial population should continue to be examined in order to better understand the relationship between these important variables. There is a need for further research addressing psychosocial concerns and surgical intervention in the craniofacial population.

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Table 1
Demographic Information (N=139)

Demographics	*n (%)*
Gender	
Female	66 (47.5%)
Male	71 (51.1%)
Age	
0-12 Months	20 (14.4%)
13-24 Months	7 (5.0%)
2-4 Years	26 (18.7%)
5-7 Years	29 (20.9%)
8-12 Years	38 (27.3%)
13-18 Years	18 (12.9%)
Race	
Asian	6 (4.7%)
White/Caucasian	105 (82.0%)
Black/African American	8 (6.3%)
Other	9 (7.0%)
Ethnicity	
Hispanic	82 (59.0%)
Non-Hispanic	56 (40.3%)
Insurance Type	
Government Subsidized	112 (80.6%)
Private	22 (15.8%)
None	5 (3.6%)
Language	
English	85 (61.2%)
Spanish	54 (38.8%)
Diagnosis	
Cleft Lip/Palate	107 (77%)
Cleft Lip	6 (4.3%)
Cleft Palate	26 (18.7%)
Surgical Category	
Visible	7 (5.0%)
Less Visible	27 (19.4%)

Table 2

Mean PedsQL™ raw scores by age group

Age	Initial Visit	Follow-Up
0-12 Months	83.11	80.98
13-24 Months	73.01	74.25
2- 4 Years	77.79	81.19
5-7 Years	73.70	70.89
8-12 Years	67.13	68.58
13-18 Years	70.29	73.55

Table 3
Regression Analysis for Hypothesis 1

Variable	B	SE B	β
Initial PedsQL™ Scores	.441	.077	.476**
Parenting Stress at follow-up visit	.125	.048	.198*

* $p < .01$; ** $p < .001$

Appendix A

Parent Craniofacial Questionnaire

Ages 13-24 Months

Please place an "X" in the box that describes how you feel about the statement. For example, if you agree with the statement, "I sometimes worry about my child," you would complete the question in this way:

	Strongly Disagree	Disagree	Neither Agree nor Disagree	Agree	Strongly Agree	Unsure
I sometimes worry about my child.				X		

Please answer the following items the same way:

	Strongly Disagree	Disagree	Neither Agree nor Disagree	Agree	Strongly Agree	Unsure
I have people I can count on when I need help.						
When I have a problem, I like to face it head on and figure out ways to solve it.						
I feel overwhelmed with my child's needs.						
I have the strategies I need for parenting my child.						
My child shows repetitive or unusual behaviors.						
I enjoy playing with my baby or toddler						

Please describe the best things about your child:

Please describe what concerns you about your child:

Wetherington, 2011

Ages 8-12 Years

Please place an "X" in the box that describes how you feel about the statement. For example, if you agree with the statement, "I sometimes worry about my child," you would complete the question in this way:

	Strongly Disagree	Disagree	Neither Agree nor Disagree	Agree	Strongly Agree	Unsure
I sometimes worry about my child.				X		

Please answer the following items the same way:

	Strongly Disagree	Disagree	Neither Agree nor Disagree	Agree	Strongly Agree	Unsure
I have people I can count on when I need help.						
When I have a problem, I like to face it head on and figure out ways to solve it.						
I feel overwhelmed with my child's needs.						
I have the strategies I need for parenting my child.						
My child shows repetitive or unusual behaviors.						
My child reads well.						

Please describe the best things about your child:

Please describe what concerns you about your child:

BIOGRAPHICAL SKETCH

Lauren Perrin
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EDUCATION/TRAINING

INSTITUTION AND LOCATION	DEGREE	YEAR(s)	FIELD OF STUDY
Texas A&M University	B.A.	2010	Psychology
The University of Texas Southwestern School of Health Professionals	M.R.C.	2014	Rehabilitation Counseling

Positions and Employment

2009-2014: Owens Ear Center
Allergy Consultant
2013-Present: Coaching for Academic Success
Executive Functioning Homework Coach
2014-Present: Matthew Housson & Associates

Clinical Experience

2009 Texas A&M University, Educational Psychology Department: Project Achieve
2010 Texas A&M University, Educational Psychology Department: Project ABC
2013 Matthew Housson & Associates: Graduate student intern
2014 Zale-Lipshy Psychiatric Unit: Graduate student intern
2014 University Rehabilitation Services (PSAT): Graduate student intern

Professional Memberships

2013 International Association of Rehabilitation Counselors