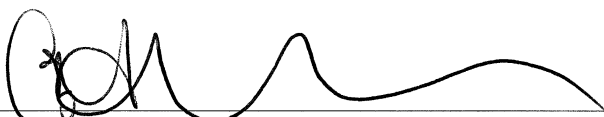
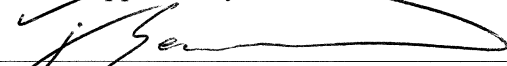


FACIAL ASYMMETRY AND SOCIAL FUNCTIONING
IN CHILDREN AND ADOLESCENTS WITH CLEFT LIP AND/OR PALATE

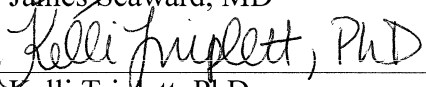
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DEDICATION

I would like to thank the members of my Graduate Committee for their time and participation in this project. Extended thanks to my thesis advisor and chair, Dr. Celia Heppner, for her guidance throughout the process. I would also like to thank my siblings, who painted the solar systems on the backs of my hands so that I could always find my way back home.

FACIAL ASYMMETRY AND SOCIAL FUNCTIONING
IN CHILDREN AND ADOLESCENTS WITH CLEFT LIP AND/OR PALATE

by

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THESIS

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Abstract

BACKGROUND: While orofacial clefts may affect the facial appearance of children and adolescents with this condition, research has yet to examine the impact of facial difference on social functioning in this population. Characteristics of facial appearance, such as symmetry, are important in social interaction. Given that individuals with CL/P often present with a degree of facial asymmetry, their social experience may differ from that of the general population. This study aimed to examine the relationship between facial asymmetry and social functioning in children and adolescents with CL/P.

PARTICIPANTS: Participants included children and adolescents seen in a multidisciplinary team clinic at a large plastic and craniofacial surgery center. Data was obtained from children and adolescents with a cleft lip and/or palate (CL/P) diagnosis, between the ages of 8 and 18 years of age. Participants with other complex medical or genetic diagnoses were excluded from this study. Participants were separated into groups based on cleft diagnosis (bilateral CL/P, unilateral CL/P, cleft lip only, and cleft palate only).

METHOD: Data was collected via retrospective chart review and included demographic information, medical and surgical history, and responses to self-report questionnaires measuring quality of life (PedsQL). Three-dimensional images of each patient were also taken as part of routine care at each clinic visit. This study utilized measurements obtained from the three-dimensional images, as well as scores on the social functioning scale from the PedsQL.

RESULTS: The current study found three-dimensional stereophotogrammetric analysis for facial asymmetry to have high interobserver reliability in the CL/P population. Overall, the current study found that there were no significant differences between diagnosis groups in regard to

facial asymmetry scores and reported social functioning. Furthermore, the current study found no significant correlation between reported social functioning and facial asymmetry scores.

DISCUSSION: The results suggest that three-dimensional image analysis is a useful and reliable tool for objectively evaluating facial asymmetry in youth with CL/P. The results also suggest that social functioning of youth with CL/P is not significantly associated with facial asymmetry.

Future studies should focus on evaluating other factors that may determine social functioning.

Keywords: cleft lip and/or palate, craniofacial anomalies, children and adolescents, social functioning, facial asymmetry, facial appearance, facial attractiveness, three-dimensional imaging.

TABLE OF CONTENTS

CHAPTER ONE: INTRODUCTION	10
CHAPTER TWO: REVIEW OF THE LITERATURE	12
Overview of CL/P	12
Medical Background	12
Associated Health Implications	14
Associated Psychosocial Implications	15
Social Functioning	16
Differences in Facial Appearance and Social Functioning	18
Social Functioning for Youth with CL/P	20
Symmetry	22
Symmetry as a Determinant of Attractiveness	22
Facial Symmetry and Social Functioning	25
Symmetry and CL/P	27
Three-Dimensional Imaging	29
Current Study	32
CHAPTER THREE: METHOD	34
Participants	34
Measures	34
Procedure	37
CHAPTER FOUR: RESULTS	38
Preliminary Analyses	38
Demographics	38

Descriptive Statistics	38
Results of Hypothesis Testing	39
CHAPTER FIVE: DISCUSSION	42
Limitations	46
Directions for Future Research	48
REFERENCES	52

LIST OF TABLES

TABLE 1	66
TABLE 2	67
TABLE 3	68
TABLE 4	69
TABLE 5	70
TABLE 6	71
TABLE 7	72

LIST OF FIGURES

FIGURE 1	73
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LIST OF APPENDICES

APPENDIX A	74
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CHAPTER ONE

Introduction

Facial appearance is one of the primary sources of information received during social interactions. Research has demonstrated that from this visual input, people make automatic personality inferences and social judgments, which may shape the behavior of the observer towards the perceived individual (Todorov, 2008; Todorov, Said, Engell, & Oosterhof, 2008). According to the ecological theory of social perception, facial appearance may convey messages about a person's attributes, particularly in the social context (Zebrowitz & Collins, 1997). Moreover, evolutionary theory suggests that humans have a preference for attractive facial traits, as facial attractiveness serves as a marker for both phenotypic and genetic quality (Penton-Voak & Perrett, 2000; Rhodes, 2006). One particular facial trait, symmetry, has been proposed as a major determinant for facial attractiveness (Rhodes, 2006). In several studies, individuals with greater facial symmetry have been rated as possessing social desirable traits (Fink, Neave, Manning, & Grammer, 2005; Pound, Penton-Voak, & Brown, 2007). Conversely, individuals with less facial symmetry are rated more negatively on parameters of social functioning, such as trustworthiness, honesty, employability, and popularity (Rankin & Borah, 2003; Shackelford & Larsen, 1997).

Negative social judgments due to facial differences may particularly affect school-age children and adolescents, as this is a developmental period in which self-awareness is strongly affected by peer approval, and concern for appearance and identity becomes more prevalent (Edwards et al., 2005; Strauss et al., 2007). Recent research indicates that children and adolescents with CL/P tend to experience more difficulties with social functioning when compared to non-affected peers (Berger & Dalton, 2011; Bradbury, 2012; Kapp-Simon et al.,

1992; Snyder & Pope, 2010). Given the impact of facial appearance on social experience, it may be that visible facial differences in youth with CL/P play a role in this relationship. The present study examined the relationship between facial asymmetry and reported social functioning for children and adolescents with cleft lip and/or palate (CL/P). Given that three-dimensional stereophotogrammetric analysis was used to assess facial asymmetry, the study also examined interobserver reliability of this method for measuring facial differences in the CL/P population.

CHAPTER TWO

Review of the Literature

Overview of CL/P

Medical Background

Cleft lip is one of the most common congenital anomalies, with an incidence of 1 in 940 live births (Parker et al., 2010). Cleft lip may present in isolation (CL) or combined with a cleft palate (CP); these diagnoses are collectively referred to as cleft lip and/or palate (CL/P).

Incidence of CL/P varies by race, with the highest incidence rates among individuals of American Indian and Asian descent and lowest among individuals of African descent (Robin et al., 2006). Cleft lip diagnosis is also twice as common in males as in females, whilst cleft palate diagnosis is more common in females (Robin et al., 2006). The condition affects lip and/or mouth formation in utero, resulting in significant difference in facial appearance as compared to non-affected individuals.

Youth with CL/P may receive multiple surgical interventions and continuing, multidisciplinary care. Timing and treatment vary by patient depending on type and severity of the cleft (Robin et al., 2006). Treatment may begin as early as one week to three months of age for infants with CL or CL/P with nasoalveolar molding (NAM), a presurgical therapy designed to reduce the size of the oronasal opening before surgical repair (CHOP, 2015). Surgical cleft lip repair or cheiloplasty is commonly performed at age 10 to 12 weeks, in order to close the lip and realign the muscle of the upper lip to provide normal lip function (Robin et al., 2006). During this time, simultaneous procedures, such as a primary rhinoplasty and myringotomy tube placement, may also be performed (Robin et al., 2006). Cleft palate is typically repaired between 6 and 12 months of age, as this procedure requires sufficient growth of facial bones (Robin et al.,

2006). The cleft palate repair or palatoplasty aims to repair the roof of the mouth and realign the palatal muscle so as to improve speech, eating, and swallowing (Robin et al., 2006). For patients with CL/P, orthodontic treatment often occurs in two phases, with the initial phase occurring around school age, when patients have mixed dentition, and the second phase in early adolescence when permanent teeth have fully erupted. Thus, between ages five and nine, approximately one-quarter of patients with CL/P will undergo further procedures with palatal expansion, and subsequent bone grafting if needed (CHOP, 2015). These surgeries involve aligning segments of and placing bone along the alveolus or gum to create space for permanent teeth or to prepare for orthodontic treatment and/or dental implants. During this age range, alveolar fistula, or remaining abnormal opening in the palate, is also repaired (Robin et al., 2006). During phase two, patients may receive orthodontic treatment, given that permanent teeth commonly erupt in abnormal positions (CHOP, 2015). Teeth are leveled, aligned, or replaced throughout the different phases of treatment. In some cases, a third phase is needed later in adolescence in conjunction with orthognatic or jaw surgery. Additional surgeries, such as secondary rhinoplasty or secondary palatal or lip procedures may also occur during adolescence (CHOP, 2015). These additional surgeries are directed at enhancing outcomes or addressing complications, such as residual functional or aesthetic limitations (Robin et al., 2006). Surgical outcome is impacted by many factors including timing of the repair and severity and type of cleft (Uwusu, Liu, Sidman, & Scott, 2013). When planning the overall surgical treatment process, surgeons concentrate on functionality, such as speech production, dental health, and facial growth, as well as aesthetics as their primary goals (Robin et al., 2006).

Associated Health Implications

Children with CL/P tend to experience a range of medical issues and complications that extend beyond surgical repair of the congenital anomaly (Robin et al., 2006). A major concern that arises early in CL/P care is feeding and nutrition. Infants with CL/P are at risk for feeding difficulties due to interruption of the infant's ability to form a seal and maintain strong suction on the nipple (Robin et al., 2006). Speech problems can also be seen in patients with CL/P and may manifest as speech delay, articulation difficulties, language deficits, and/or hypernasal speech due to velopharyngeal insufficiency (VPI; Hartzell & Kilpatrick, 2014; Robin et al., 2006). VPI, a problem in the muscles and tissue behind the soft palate that control the flow of air between the nose and mouth, presents as a long-term complication that occurs in 10-20% of CL/P cases and can contribute to significant speech intelligibility difficulties (Robin et al., 2006). Infants and children with CL/P may also experience issues with hearing. Otitis media with middle ear effusion, an inflammatory disease of the middle ear, is common in patients with CL/P and results from eustachian tube dysfunction (Robin et al., 2006). These chronic middle ear complications can cause fluid to remain in the middle ear for weeks or months at a time; consequently, children with CL/P are at a higher risk of developing conductive hearing loss (Skuladottir et al., 2015).

Patients with CL/P also may experience dental and oral and maxillofacial sequelae, and as a result tend to have poorer dental health than children in the general population (McDonagh, Pinson, & Shaw, 2000). Due to maxillary growth disturbance, children with CL/P may present with dental malocclusion, cross bites, impacted teeth, missing teeth, midface deficiency, and sleep apnea (Robin et al., 2006). Children with CL/P also present with a higher incidence of dental caries (McDonagh et al., 2000). Approximately 25 to 30% of patients with repaired CL/P

require orthognatic surgery to correct jaw malformation and establish proper dental occlusion (Hartzell & Kilpatrick, 2014; Robin et al., 2006).

Because of the numerous medical issues and potential complications that accompany CL/P, patients with CL/P require multidisciplinary care and management that extends through adolescence. These multidisciplinary teams go beyond plastic surgeons and dental specialists, as they include pediatricians, audiologists, pediatric otolaryngologists, speech pathologists, occupational/feeding therapists, geneticists, dietitians, and clinical psychologists (Hartzell & Killpatrick, 2014; Robin et al., 2006). This care is commonly delivered in a multidisciplinary cleft clinic where patients are seen by all specialties routinely (Robin et al., 2006).

Associated Psychosocial Implications

Researchers have also investigated psychosocial outcomes of youth with CL/P, including academic performance, behavioral and social outcomes, and emotional difficulties. Specific issues with learning and academic functioning in children with CL/P have been identified (Collet, Stott-Miller, Kapp-Simon, Cunningham, & Speltz 2010; Conrad, McCoy, DeVolder, Richman, & Nopoulos, 2014; Wehby et al., 2014). When compared to their non-affected classmates, children with CL/P appear to underperform across multiple academic areas and grade levels (Wehby et al., 2014). Moreover, rates of learning disabilities in this population have been estimated to be 30% to 40%, compared to the estimated 10% to 20% in the general population (Broder, Richman, & Matheson, 1998). Research indicates that children with CL/P demonstrate poorer reading skills than children without CL/P, and one study further reported that word reading deficits were correlated with impairments in auditory memory (Conrad et al., 2014). Other studies have attempted to find connections between these academic deficits and a child's

psychological functioning. Richman, McCoy, Conrad, and Nopoulos (2012) documented an association between behavioral inhibition in children with CL/P and poor academic performance.

Theoretical frameworks have suggested that children and adolescents with craniofacial anomalies, such as CL/P, may encounter difficulties in psychosocial adjustment; empirical evidence seems to support this idea (Berger & Dalton, 2011; Kapp-Simon et al., 1992; Snyder & Pope, 2010). Specifically, research has suggested that youth with CL/P experience higher internalizing (i.e. depression, anxiety, and social inhibition) and externalizing symptoms (i.e. aggression, opposition, and noncompliance) than their same-aged peers (Richman et al., 2012). Additional psychological concerns that have been reported among youth with CL/P include depressive symptoms, higher rates of teasing, and dissatisfaction with appearance and speech (Hunt, Burden, Hepper, Stevenson, & Johnston, 2006). Studies examining youth with CL/P have identified adjustment concerns (Berger & Dalton, 2011; Feragen, Stock, & Kvaalem, 2015; Kapp-Simon et al., 1992; Snyder and Pope, 2010). Specific findings with regard to social functioning for youth with CL/P will be discussed in greater depth in the following sections.

Social Functioning

A growing body of social psychology research indicates that people make automatic and rapid personality inferences after minimal exposure to an individual's physical appearance (Todorov et al., 2008; Valla, Ceci, & Williams, 2011). An exposure time of 50-100 milliseconds to unfamiliar faces is sufficient for humans to make trait inferences (Bar, Neta, & Linz, 2006; Rule, Ambady, & Adams, 2009; Willis & Todorov, 2006). It is argued that because facial information is readily and continuously available to the perceiver during social interaction, facial appearance affects overall and subsequent evaluation of the perceived individual (Ambady, Bernieri, & Richeson, 2000; Kleck & Rubenstein, 1975). Additionally, information processing

and retrieval of visual characteristics, such as physical appearance, is presumed to be less complex than attitudinal or personality information, and therefore easier to use when evaluating others (Macrae & Bodenhausen, 2000).

Inferences about a person's social traits based on their face can influence how a perceiver thinks and behaves toward the individual, as well as how well the perceiver remembers the face (Rule, Slepian, & Ambady, 2012; Todorov, 2008). The ecological theory of social perception proposes that facial appearance may convey a person's attributes, particularly in the social context (Zebrowitz & Collins, 1997). A study that investigated memory found that faces perceived as untrustworthy are remembered better than faces perceived as trustworthy (Rule et al., 2012). Following the rationale of the ecological theory of social perception, because untrustworthy faces might signal dangers that should be avoided, untrustworthy faces are more noticeable to perceivers and can therefore be better remembered (Rule et al., 2012). Based on facial appearance, a perceiver may also make inferences about certain social dimensions, such as the perceived individual's character and social traits (Todorov et al., 2008). These inferences have been found to predict important social outcomes including electoral success, occupational outcome, and criminal sentencing decisions (Ballew & Todorov, 2007; Blair, Judd, & Chaplean, 2004; Todorov, Mandisodza, Goren, & Hall, 2005). For example, facial dominance predicted the final career rank of graduates from a military academy, as well as their ranks in later career (Mazur, Mazur, & Keating, 1984; Mueller & Mazur, 1996). Facial dominance was observed by judges who were instructed to rate images based on a given definition for 'a dominant person'. Similarly, inferences about competence, dominance, and maturity based on the facial appearance of CEOs have been found to positively predict company profits (Rule & Ambady, 2008).

Because of the consistency and automaticity of these appearance-based biases, one may speculate that facial appearance could play a role in social outcomes and overall experience.

Negative social experiences due to facial differences may particularly affect a child's development as they begin to build their self-concept and social identity. Middle childhood (8-11 years old) and adolescence (12 to 18 years old) are developmental stages in which self-awareness is strongly affected by peer approval, and concern for appearance and identity increases (Edwards et al., 2005; Strauss et al., 2007). By early school age, children make judgments about physical attractiveness in a very similar fashion to adults by showing preferences for globally attractive faces (Boothroyd, Meins, Vukovic, & Burt, 2014). Moreover, the specific influence of symmetry on attractiveness judgment typically emerges after five years of age and matures after nine years of age (Boothroyd et al., 2014; Vingilis-Jaremko & Maurer, 2013). Given the increasing importance of peer approval during this age range, youth with facial differences may experience a higher degree of teasing and bullying related to their appearance (Lovegrove & Rumsey, 2005; Masnari et al., 2012). Negative social interactions during childhood and adolescence may lead to more negative self-perceptions, which may affect a child's overall well-being (Masnari et al., 2012).

Differences in Facial Appearance and Social Functioning

Social and adjustment difficulties experienced by youth with CL/P may be related to their visible facial differences. Facial differences may affect an individual's overall psychological functioning, as they may contribute to challenges with self-image, self-esteem, and emotional development (Richman et al., 2012). For instance, children with craniofacial diagnoses, including CL/P, who endorse dissatisfaction with facial appearance, have been found to be more likely to report behavioral inhibition and negative self-appraisal (Kapp-Simon et al., 1992).

Several studies have investigated social perceptions of and reactions to facial differences in the CL/P population. Individuals with facial differences such as CL/P may experience negative social reactions (Bradbury, 2012). Reactions during social interchange may vary from intrusive staring to aggression, pity, or disgust (Rumsey & Harcourt, 2005). Children and adolescents with facial differences, including those with CL/P, were found to be at a higher risk of experiencing social stigmatization than same-age peers without a visible difference (Strauss et al., 2007). Reported stigma experiences included frequent expressions of pity, teasing, staring or startled reactions, and having frequent talks with others about how their face looks. Perhaps related to the social stigma of an appearance difference, increased rates of peer victimization may be present for youth with CL/P (Lorot-Marchand et al., 2015). Carroll and Shute (2005) also studied peer victimization in youth with CFA, including those with CL/P, by exploring craniofacial aggression. The authors defined craniofacial aggression as a group of characteristic behaviors identified in the craniofacial literature such as pointing, staring, standing further away than usual, impersonating, making jokes, and asking personal questions. The study found that severity of the facial difference, as measured by subjective rating, was associated with greater self-reported incidence and frequency of craniofacial aggression. A limitation of this study was that youth with more extreme facial differences were over-represented in the sample (Carroll & Shute, 2005). Hence, evaluation of the social impact of an appearance difference across a broader range of CL/P diagnoses is warranted.

Interpersonal reactions to facial differences can affect an individual's social functioning (Edwards, Topolski, Kapp-Simon, Aspinall, & Patrick, 2011). Behavioral conditioning may occur as a result of aversive experiences in social interactions, leading to behavioral inhibition, shyness, and social withdrawal (Edwards et al., 2011). Youth with CL/P in particular have been

found to exhibit higher social isolation and decreased participation in group-setting social interactions than non-affected youth (Murray et al., 2010). In another observational study that examined interactional patterns in adolescents, participants with facial differences took part in fewer social approaches than those without facial differences (Kapp-Simon & McGuire, 1997). Researchers have proposed that social withdrawal may act as a self-protective factor in this population to minimize peer rejection (Rubin, Coplan, & Bowker, 2009).

Social Functioning for Youth with CL/P

Various studies and system reviews have concluded that adults with cleft lip do not seem to be at increased risk for serious psychosocial adjustment or functioning problems, despite youth with cleft lip experiencing challenges in this area. (Endriga & Kapp-Simon, 1999; Hunt, Burden, Hepper, & Johnston, 2005; Pisula et al., 2014). Adults with cleft lip have been found to display less self-doubt and rumination over interpersonal relations than adolescents with the same diagnosis (Richman & Harper, 1980). Moreover, a recent qualitative study revealed that children and adolescents with cleft lip experience negative self-perception and social stigmatization, yet self-perception improves and perceived stigmatization declines as they reach adulthood (Alansari, Bedos, & Allison, 2014). Researchers attribute this improvement in psychosocial adjustment to the definitive treatment procedures that are undertaken at later stages, leading to significant improvement in appearance, as well as maturation of peer behavior (Alansari et al., 2014). As previously mentioned, childhood and adolescence are developmental stages in which humans build their self-concept and social identity. During this time period, individuals may have heightened sensitivity to peer stigmatization, as concern for appearance arises and youth increasingly seek peer approval (Edwards et al., 2005; Strauss et al., 2007). Given the increased importance of social experiences with peers during school age and

adolescence and the negative social outcomes associated with facial differences, this age group within the CL/P population may be at increased risk for social difficulties. These findings point out the importance of concentrating on children and adolescents when investigating the implications of facial attractiveness and symmetry on the social experience of individuals with CL/P.

Research has sought to identify predictors or factors that contribute to variability in adjustment for youth with CL/P. Although many factors have been proposed, cumulative research has identified a recurrent correlation between adjustment and social experience. One study within the craniofacial population, for which CL/P was the predominant diagnosis, found that the degree of social skills and overall quality of social behaviors reported by these youth were significant predictors of adjustment (Kapp-Simon et al., 1992). Similarly, a study by Berger and Dalton (2011) identified social experiences, defined as the frequency of social interactions and availability and helpfulness of social support, as the best predictor of psychosocial adjustment in adolescents with CL/P. Additionally, research suggests that adjustment problems for youth with CL/P are particularly related to anxiety regarding social acceptance and social judgment (Kapp-Simon et al., 1992). Therefore, social skills challenges and interpersonal difficulties may lead to poor adjustment outcomes in youth with CL/P.

Despite findings indicating that many youth with CL/P experience difficulties with social functioning and overall adjustment, some research suggests that many children with CL/P develop in an adaptive manner and do not experience significant psychosocial adjustment problems (Berger & Dalton, 2009). This discrepancy in findings warrants additional, in-depth research on the subject in order to further identify predictors of social outcomes and adjustment within this population.

Symmetry

Symmetry as a Determinant of Attractiveness

Evolutionary psychologists have hypothesized that attractive morphological characteristics are indicators of fitness, quality, mental and physical health, and reproductive value (Bashour, 2006). Facial attractiveness is also important in human mate preferences, as it predicts mating success (Rhodes, Simmons, & Peters, 2005). Additionally, meta-analytic research has established that perceptions of facial attractiveness in children influence impressions of social appeal, academic and developmental competence, psychosocial adjustment, and interpersonal competence (Langlois et al., 2000). Such findings suggest that physical attractiveness is seen in a positive light and can guide social perceptions.

Evolutionary theory suggests that preference for attractive facial traits arises from mate quality signaling, as facial attractiveness serves as a marker for both phenotypic and genetic quality (Rhodes, 2006; Penton-Voak & Perrett, 2000). In other words, facial attractiveness may reveal the quality of a person's observable physical traits, as well as the genes, which contribute to the expression of those traits. Theorists have proposed that there is a biological predisposition among humans to prefer attractive faces and argue that judgment of facial attractiveness is universal (Penton-Voak & Perrett, 2000). This idea is supported by the high cross-cultural agreement in facial attractiveness rating across multiple ethnicities, cultures, and geographical locations (Coetzee, Greeff, Stephen, & Perrett, 2014; Cunningham, Roberts, Barbee, Druen, & Wu, 1995). Extensive research has recognized four main determinants that influence facial attractiveness: averageness, sexual dimorphism, youthfulness, and symmetry (Bashour, 2006; Little, Jones, & DeBruine, 2011; Penton-Voak & Perrett, 2000; Rhodes, 2006; Saxton, DeBruine, Jones, Little, & Craig Roberts, 2011).

Averageness, or the prototypicality of a face, can be illustrated by averaging multiple individual faces together mathematically to create a physical configuration of an “average” face (Bashour, 2006). The evolutionary approach hypothesizes that a facial pattern close to the population average facial pattern signals genetic heterozygosity or diversity. It is argued that because genetic diversity increases an individual’s chance for survival by providing greater resistance to diseases and reducing the incidence of unfavorable inherited traits, averageness is a valued characteristic in mate choice and thereby conveys attractiveness (Lee et al., 2016; Thornhill & Gangestad, 1993).

Similarly, sexually dimorphic traits, or traits that are characteristically feminine and masculine, also influence perceptions of attractiveness. Adults seem to perceive extremes of secondary sexual characteristics as more attractive (Bashour, 2006). One study found that women ranked men with more masculine features, including thick eyebrows, small eyes, thin lips, and square jaws, as more attractive than those with more androgynous features (Keating, 1985). Correspondingly, men perceive feminine features, such as prominent cheekbones, large eyes, thick lips, and a small nose and chin, as attractive in women (Baudouin & Tiberghien, 2004).

Literature also supports youthfulness as another determinant of attractiveness; youthful faces are found to be more attractive than older looking faces (Henss, 1991; Mathes, Brennan, Haugen, & Rice, 1985). Relatedly, humans tend to display preference for neotenous features, those which approximate those of neonates, in infants and adults, in both genders (Bashour, 2006). Neotenous features include traits such as large eyes, small noses, and round cheeks. The evolutionary standpoint hypothesizes that preference for neoteny may occur because infants with more neotenous features attract more parental attention and care in order to survive and pass on

their genes (Bashour, 2006). As a consequence, adults with neonate features may evoke attraction and nurturance responses that were evolutionarily designed for infants (Bashour, 2006).

Symmetry is another proposed determinant of facial attractiveness. To examine this characteristic, researchers have manipulated the symmetry of individual faces in photographs and asked participants to assign attractiveness ratings to both original and manipulated faces (Perrett al., 1999; Rhodes, Proffitt, Grady, & Sumich, 1998). It was found that perceived attractiveness increased when symmetry was increased, and correspondingly decreased as symmetry was reduced. In other words, perfectly symmetric versions of faces were preferred to less symmetric versions of the same faces.

Bilateral symmetry of facial traits may reflect an overall high phenotypic and genetic quality. Phenotypic qualities may signal material benefits, such as increased parental care and protection from the environment (Ryan & Cummings, 2013). Similarly, genetic quality may signal the indirect benefit of passing on good genes to one's child (Ryan & Cummings, 2013). As a result, symmetry might be a desirable trait. Similar to averageness, symmetry of bilateral traits may signal genetic diversity (Bashour, 2006; Tomkins & Kotiaho, 2002). As previously stated, genetic diversity may signal ability to resist genetic and environmental disturbances via pathogen avoidance, heritable immunocompetence, and decreased vulnerability to genetic syndromes in offspring (Tomkins & Kotiaho, 2002; Tybur & Gangestad, 2011). Therefore, it is argued that symmetrical faces are preferred as they may have some evolutionary adaptive value (Bashour, 2006).

Furthermore, symmetry preferences may also occur as a by-product of how brains process information (Rhodes, 2006). According to this argument, biases in the visual sensory

system influence an individual's perception and reaction to signals, with a preference for symmetrical input when recognizing and processing information (Enquist & Ghirlanda, 1998). Given the ease with which the visual system can recognize and process symmetrical stimuli, symmetry can be perceived as preferable and attractive (Jones et al., 2001; Little & Jones, 2006). Additionally, symmetric stimuli match the human visual system's own bilaterally symmetric framework, supporting the view that a perceptual bias to symmetry may result as a by-product of visual processing (Little & Jones, 2006). In other words, symmetry in the visual system's anatomy itself, consisting of a pair of eyes, two optic nerves, and the visual cortex on both cerebral hemispheres, may lead to a preference for symmetric stimuli.

Facial Symmetry and Social Functioning

The importance of attractiveness, and symmetry as its determinant of attractiveness, is particularly relevant in the context of an individual's social environment. In a foundational social psychology study, Dion, Berscheid, and Walster (1972) found that facial attractiveness elicits personality attributions, as individuals rated attractive people, with the attractiveness rating determined by a preliminary study, as possessing socially desirable traits and leading better lives than unattractive people. For instance, attractive people were perceived as being more competent spouses and being more successful occupationally. The attractiveness rating criteria were high-inter-rater agreement from the preliminary study and that the attractive and unattractive categories did not include extreme ends of attractiveness (Dion et al., 1972). Another foundational study in attractiveness and social perceptions found that for children, popularity with their classmates is associated with their physical attractiveness, as judged by adults (Dion & Berscheid, 1974). Similarly, Langlois and colleagues (2000) found that children and adults rated

as unattractive experienced more negative and fewer positive social interactions than those judged as attractive.

Multiple factors may be involved in determining the extent to which facial appearance affects social functioning. Extra-personal factors, such as cultural beliefs about the importance of appearance and tolerance in society or family, may be influential (Van Den Elzen et al., 2012). Intrapersonal components, such as severity of condition or dissatisfaction with appearance, appear to have a larger effect on social functioning. One study that explored social functioning in adults with orofacial clefts found that dissatisfaction with appearance, a subjective intrapersonal variable, was a predictor of poorer social functioning (Van Den Elzen et al., 2012). Berger and Dalton's (2011) study also supported this conclusion, as they found that self-reported satisfaction with appearance significantly predicted the social experience of young people with CL/P.

Facial symmetry may also impact social functioning. Symmetry appears to be a significant determinant of perception of facial attractiveness; thereby symmetry may have an effect on social desirability, as well as development and maintenance of interpersonal relationships (Bashour, 2006; Baudouin & Tiberghien, 2004). Various studies investigating the relationship between facial symmetry and psychosocial traits in late adolescents revealed that perceptions of universal personality factors from the five-factor model of personality were significantly related to facial symmetry (Fink et al., 2005; McCrae & Terracciano, 2005; Noor & Evans, 2003; Pound et al., 2007). The personality attributes measured included extraversion, openness, agreeableness, neuroticism, and conscientiousness (McCrae & Costa, 1987). These studies found a positive association between facial symmetry and traits traditionally viewed as pro-social, such as extraversion (Fink et al., 2005; Pound et al., 2007). Correspondingly, facial symmetry was negatively associated with less socially desirable traits such as disagreeableness,

neuroticism, and less conscientiousness (Noor & Evans, 2003). Other studies have investigated the relationship between facial symmetry and perceptions of social functioning. Findings revealed that individuals with greater degrees of facial asymmetry are more likely to be rated negatively on parameters of social functioning. For instance, they are more likely to be perceived as less trustworthy, less honest, less employable, less popular, and more impulsive (Rankin & Borah, 2003; Shackelford & Larsen, 1997). In comparison, faces high in symmetry tended to receive significantly higher ratings in sociability, liveliness, self-confidence, and genuineness (Fink, Neave, Manning, & Grammer, 2006; Shackelford & Larsen, 1997). The relationship found thus far between facial symmetry and the perception of pro-social characteristics may potentially shed light onto the role of facial symmetry in the context of the social experience of youth with CL/P.

The research summarized in this section suggests that facial symmetry may play a role in the social functioning of individuals with facial differences, as it influences perception of social functioning and prosocial personality traits. Nonetheless, the literature is still lacking a study that directly examines the relationship between facial symmetry and self-reported social functioning. Additionally, this relationship has not yet been examined in individuals with visible facial differences, such as those with CL/P.

Symmetry and CL/P

Patients with CL/P often present with a measurable and significant degree of facial asymmetry (Stauber et al., 2008). The nose and lip are the main facial features affected in CL/P, thus the contour of the upper lip and the nasal rim tend to be the most asymmetric structures (Bell et al., 2014). By comparing the nasolabial area to the upper area of the face, Bell and colleagues were able to confirm that asymmetry mostly concentrates on the nose and lips for

CL/P patients (Bell et al., 2014). Regions such as the area between nose and chin and the area between chin and cheek may exhibit asymmetry as well (Kuijpers et al., 2015). Additionally, whole face asymmetry has been found to be significantly higher in the CL/P population than in non-CL/P controls (Kuijpers et al., 2015; Meyer-Marcotty, Alpers, Gerdes, & Stellzig-Eisenhauer, 2010).

Unilateral orofacial clefts tend to be inherently asymmetric, as the condition phenotypically manifests one-sidedly (Kuijpers et al., 2015). Although bilateral clefts affect both sides of the mouth and/or nose, the cleft itself and scars after surgical repair may affect the two sides of the mouth unequally. Additionally, the presence of a cleft palate together with a cleft lip may contribute to asymmetry; one study found that asymmetry in patients with unilateral cleft lip and palate (UCLP) was higher than in patients with unilateral cleft lip only (UCL; Bell et al., 2014). Despite the impact that the presence of a cleft palate may have on asymmetry for patients with a cleft lip, morphological three-dimensional measurements of patients with cleft palate only (CP) appear to be more similar to those of non-affected peers, when compared to other CL/P diagnosis groups. (Bugaighis, Mattick, Tiddeman, & Hobson, 2014).

Although most orofacial clefts receive surgical intervention, reconstructive repairs may not be perfect. Russell, Kiddy, and Mercer (2014) and Hood, Bock, Hosey, Bowman, and Ayoub (2003) demonstrated that a significant degree of lip and nasal asymmetry remains in CL/P patients after primary repair. Similarly, Bell and colleagues (2014) found that asymmetry for the whole face is significantly higher in patients with unilateral CL/P than non-cleft groups, even after surgical repair.

Increased facial symmetry is perceived as an optimal surgical outcome, as a primary goal of cleft lip repair and revision procedures is to promote equal lip height bilaterally (Russell,

Patel, Skolnick, & Woo, 2015). Studies that assess for surgical treatment outcomes in the CL/P population use symmetry, nasolabial and entire-face, as an objective measure of treatment outcome, facial aesthetics, and surgical technique (Desmedt, Maal, Kuijpers, Bronkhorst, & Kuijpers-Jagtman, 2015; Hakim, Aschoff, Jacobsen, & Sieg, 2014; Sharma et al., 2012).

Three-Dimensional Imaging

In order to objectively diagnose and analyze the CL/P condition, it is important to measure facial form and structure (Mosmuller, Don Griot, Bijnen, & Niessen, 2013). Assessment of craniofacial anthropometry, or craniofacial measurement, has been used in the process of planning and evaluating treatment options, as well as to assess treatment outcomes (Othman, Ahmad, Asi, Ismail, & Rahman, 2014). Additionally, it may provide quantitative and objective measurements such as degree of asymmetry, level of residual scarring, and severity of condition (Bell et al., 2014). Various techniques of measuring facial morphology in the CL/P population have been reported, including direct clinical assessment, two-dimensional photographic evaluation by raters, two-dimensional photographic evaluation using measurements, and three-dimensional imaging (Mosmuller et al., 2013).

Assessment of morphology through direct visual examination may be used, but this technique is accompanied by several limitations. Although live observation may identify the most obvious facial disproportions, it often fails at finding more subtle details (Othman et al., 2014). Moreover, this assessment method may be time-consuming and requires the cooperation of patients. A more commonly used method has been two-dimensional rating, where raters score nasolabial appearance in photographs of patients with CL/P. This method has been studied using both medical professionals, like orthodontists or plastic surgeons, and laypeople as raters (Mosmuller et al., 2013). The five-point rating scale developed by Asher-McDade, Roberts,

Shaw, and Gallagher (1991) appears to be the most reliable for the assessment of two-dimensional photographs when compared to other scales (Mosmuller et al., 2013). Two-dimensional photographs have also been evaluated by performing linear measurements of facial landmarks, including frontal and lateral views. This type of assessment has shown high intraobserver and interobserver reliability, compared to the subjective rating system (Mosmuller et al., 2013). Quantitative assessment of facial morphology, particularly of asymmetry, may assist in overcoming the limitations of reliability and reproducibility inherent in subjective assessment (Al-Omari, Millett, & Ayoub, 2005).

Advancement in technological capacities in the last few years has allowed for the use of three-dimensional imaging in the assessment of craniofacial measurements of patients with CL/P. Three-dimensional imaging is identified as an effective assessment method because it provides a graphic representation of the image, allowing for a more accurate measurement of shape and anatomical features than that from a flat, two-dimensional image (Mosmuller et al., 2013). Studies using three-dimensional imaging systems have shown that intraobserver reliability in facial landmark placement and measurement is substantially higher than for two-dimensional photographic assessment, indicating that three-dimensional imaging provides a more reliable assessment of CL/P facial morphology (Mosmuller et al., 2013). Additionally, those studies that used multiple observers for landmark placement have shown high interobserver reliability (Schwenzer-Zimmerer et al., 2008; Van Loon et al., 2010). In fact, Mosmuller and colleagues concluded in their review of the literature that three-dimensional imaging is the most reliable method in the assessment of cleft-related anomalies.

The most widely used three-dimensional techniques in CL/P patients include computed tomography (CT), cone beam computed tomography (CBCT), magnetic resonance imaging

(MRI), laser surface scanning, and digital stereophotogrammetry (Kuijpers, Chiu, Nada, Carels, & Fudalej, 2014). For instance, pharyngeal space can be examined using MRI, CT, or CBCT (Kuijpers et al., 2014). Other craniofacial structures such as jaw relationship and the dental and alveolar arch can be evaluated with CBCT. Post-operative results of bone grafting and other surgical interventions are also commonly assessed with CT or CBCT (Kuijpers et al., 2014). In their recent review paper, Kuijpers and colleagues (2014) identified laser surface scanning and stereophotogrammetry as the best and most commonly used methods for quantitative analysis of facial soft tissues, based on high methodological quality (Kuijpers et al., 2014). The authors also argued that the low levels of measurement errors seen in both methods support their reliability for the quantitative measurement of asymmetry and changes in soft tissue after treatment (Kuijpers et al., 2014).

An increasing number of studies have been conducted utilizing three-dimensional digital stereophotogrammetry to evaluate orofacial clefts. Stereophotogrammetry is an imaging technique in which measurements are made from three-dimensional photographs (Tzou & Frey, 2011). Three-dimensional facial morphometry has been applied to numerous orofacial cleft groups, including unilateral cleft lip (UCL), unilateral cleft lip and palate (UCLP), unilateral cleft lip and alveolus (UCLA), unilateral cleft lip and palate and alveolus (UCLAP), bilateral cleft lip and palate (BCLP), and isolated cleft palate (ICP; Bugaighis et al., 2014; Schwenzer-Zimmerer et al., 2008). three-dimensional imaging has also become easier to use over time; for instance, acquisition time for images has been reduced to milliseconds in many cases, making it possible to capture usable images of infants and younger children (Mosmuller et al., 2013). Hence, because of its reliability, accuracy, and ease of use, three-dimensional digital

stereophotogrammetry may be a preferred three-dimensional imaging tool for the evaluation of facial characteristics in the CL/P population.

Current Study

Differences in facial appearance can influence a person's social experience (Van Den Elzen, 2012). For instance, variability in facial attractiveness can affect perceptions of pro-social personality traits and social functionality (Langlois et al., 2000). As a determinant of attractiveness, facial symmetry may thereby affect social desirability and relate to social experience (Bashour, 2006; Baudouin & Tiberghien, 2004). Given the facial asymmetry often perceivable in patients with CL/P (Stauber et al., 2008) and the association between facial asymmetry and social perceptions, it can be argued that children and adolescents with cleft lip, with or without cleft palate, may be at an increased risk for concerns with regard to social functioning. Existing literature seems to establish that youth with CL/P display different patterns of social interaction than non-affected youth (Kapp-Simon & McGuire, 1997), but the specific relationship between facial asymmetry and social functioning in this population has not been investigated.

The objective of the current study therefore was to examine the relationship between objectively measured facial asymmetry and social functioning in children and adolescents with CL/P. Additionally, the study aimed to evaluate the application of three-dimensional photogrammetric imaging in the CL/P population.

Aim 1: To quantify the facial asymmetry in children with CL/P using three-dimensional stereophotogrammetric analysis and evaluate the interobserver reliability of this assessment method in the CL/P population.

Hypothesis 1: Values generated for facial asymmetry through three-dimensional

stereophotogrammetric analysis by two different observers will be significantly correlated for images of patients with CL/P diagnoses.

Aim 2: To examine the relationship between facial asymmetry and social functioning in children and adolescents with CL/P, and to assess how this relationship varies by CL/P diagnosis.

Hypothesis 2a: It is hypothesized that facial asymmetry scores will differ for diagnosis group, such that unilateral groups will have higher asymmetry scores than bilateral groups, and cleft lip and palate (CLP) and cleft lip only (CL) groups will have higher asymmetry scores than cleft palate only (CP) group.

Hypothesis 2b: It is hypothesized that social functioning scores will differ for diagnosis groups, with cleft lip and palate (CLP) and cleft lip only (CL) groups having lower social functioning scores than cleft palate only (CP) group.

Hypothesis 2c: It is hypothesized that ratings of social functioning in children and adolescents with CL/P will be inversely proportional to their measured facial asymmetry. Specifically, it is predicted that children and adolescents who report lower scores for social functioning will have greater measured facial asymmetry.

CHAPTER THREE

Method

Participants

Participants were children and adolescents, between the ages of 8 and 18 years of age, with a diagnosis of cleft lip, cleft palate, or cleft lip and palate. Participants must have been seen in the multidisciplinary craniofacial team clinic within the Fogelson Plastic and Craniofacial Surgery Center at Children's Health/Children's Medical Center in Dallas, TX between March 2011 and March 2016 in order to have been included in the study. Additionally, participants must have been able to read and complete measures in either English or Spanish at the time of their clinic visit in order to be included in the sample, and must have had three-dimensional images taken as part of their team clinic visit. Children with previous facial trauma, facial scarring unrelated to orofacial cleft, or other facial anomalies were excluded from this study. Children with syndromic orofacial clefts were also excluded from this study. Children with significant cognitive deficits or learning disorders that prevent their ability to provide assent and complete study materials were excluded from this study.

Measures

Pediatric Quality of Life Inventory 4.0 Generic Core Scale (PedsQL) – Social Functioning

The Pediatric QL™ 4.0 Social Functioning subscale is a widely used 5-item self-report scale designed to assess social functioning in clinical pediatric populations (Varni et al., 2001). Items are rated on a 4-point Likert scale (1= *Never a problem*, 4= *Almost always a problem*), with the respondent rating the frequency of impairments in functioning over the past month, and an overall social functioning score is calculated using the average of the items within the social functioning subscale. Higher scores reflect more optimal social functioning. The PedsQL™ 4.0

has displayed strong construct validity and internal consistency reliability (Varni et al., 2001). Varni and colleagues agree that subscales of the PedsQL such as the social functioning subscale may be utilized for descriptive and exploratory analysis of specific domains of functioning. The quality of life inventory has been shown to correlate well with disease status and to differentiate clinical groups (Varni et al., 2001). It has also been extensively used in the CL/P population (Collett et al., 2012).

Three-Dimensional Imaging

Three-dimensional digital imaging is utilized for facial image acquisition and symmetry analysis, and has recently been used in the CL/P population (Kuijpers et al., 2014). Symmetry analysis using this technology has displayed strong intraobserver and interobserver reliability (Ayoub et al., 2003; Mosmuller et al., 2013).

The imaging system used in the current study is composed of 12 cameras positioned to obtain photographs from multiple angles of the craniofacial anatomy, all grouped into sets of three. Each set is composed of two black-and-white cameras and one color camera. All cameras captured images simultaneously, which were then merged together to compose a three-dimensional image. The process of capturing the images took an approximate of one second per patient. A three-dimensional image was constructed out of these different-angled images and digitized on the computer screen, which usually took one minute per patient.

For the current study, a perfectly symmetric model with left and right point correspondences was created from a three-dimensional scan and was used as a reference template, in order to calculate the asymmetry in each participant's three-dimensional data. The imaging software 3dMDvultus was used to manually place 29 landmarks on the template and each participant's three-dimensional image at recognizable and repeatable anatomic locations.

Two observers placed the landmarks on the images, and both observers followed the same standardized guidelines to placing landmarks. The observers were two graduate students in UT Southwestern Medical Center. For each of the participant images, registration was performed through rigid translation and rotation to match the template. Moreover, the template was scaled in width to appropriately match each three-dimensional image.

In order to allow point correspondence between the left and right sides of each participant, the template was deformed to each participant's three-dimensional surface scan. This was done by using thin-plate-splines with the landmarks described earlier, followed by closest-point deformation for detailed point-wise deformation.

Asymmetry was defined as the difference between two distances; the difference between corresponding points on contralateral sides of the midsagittal plane. This calculation was performed across every point on each data set. Finally, the overall nasolabial asymmetry (herein 'facial asymmetry') was calculated. The nasolabial area of interest was defined by the nasion point, a distinctly depressed area directly between the eyes and superior to the bridge of the nose, originating from the midsagittal plane and spreading out to the left and right oral commissures, or the corners of the mouth (Refer to Appendix A for landmark names and placement). Within this parameter, the areas examined for asymmetry are mainly the nose and lip. Given that the nose and lip are the main facial features affected and that the nasolabial area is the most asymmetrical facial area in individuals with CL/P, the current study focused on localized, rather than whole face asymmetry (Bell et al., 2014). This was also done so as to maximize variability in the sample.

Procedure

Data for this study was collected via retrospective chart review as part of a larger study examining psychosocial functioning and quality of life in patients with CL/P.

CHAPTER FOUR

Results

Preliminary Analyses

Preliminary analyses were conducted to screen the data for multivariate normal distribution, linearity, and outliers. Data was transformed in order to remove two extreme outliers for the facial asymmetry values. Data was linear and normally distributed.

Demographics

Table 1 lists demographic information for the study sample. The mean age for participants was 13.20(2.88). The sample consisted of 39.5% females and 60.5% males. Of the participants, 49.4% were Hispanic, 49.4% were Non-Hispanic, and 1.2% were of unknown ethnicity. Regarding race of patients in the sample, 2.5% were Asian, 4.9% were African American/Black, 86.4% were Caucasian/White, and 6.2% identified their race as “Other.” The sample was predominantly English-speaking, with most participants living in households with English as the preferred language (66.7%). The second most common language preference represented in the sample was Spanish (32.1%), followed by other languages (1.2%). Regarding insurance, 75.3% of participants had government-subsidized insurance, whereas 24.7% had private insurance. Cleft diagnostic groups represented in the sample included bilateral CL/P (24.7%), unilateral CL/P (59.3%), cleft lip only (9.9%), and cleft palate only (6.2%).

Descriptive Statistics

Table 2 lists descriptive statistics for the variables of interest. When comparing social functioning by diagnostic categories, participants with bilateral CL/P reported the highest social functioning, with an overall mean PedsQL social functioning score of 86.75(19.75). Participants with cleft palate only reported a markedly lower overall mean PedsQL social functioning score,

70.00(21.51). All diagnostic categories had variable social functioning scores, with scores ranging from 25 to 100. According to published norms for the PedsQL, a score of 66.61 or lower represents at-risk status for impaired social functioning (Varni, Burwinkle, Seid, & Skarr, 2003). In the current study, 24.36% of participants reported social functioning scores below the cut-off point score indicating at-risk status. For the 81 participants included, measured facial asymmetry scores ranged from 0.45mm to 2.05mm. The overall mean measured facial asymmetry was 0.89(0.38) for the bilateral CL/P group and 1.02(0.38) for the unilateral CL/P group. The cleft lip only group displayed an overall mean of measured facial asymmetry of 0.84(0.20), compared to 1.11(0.50) displayed by the cleft palate only group.

Results of Hypothesis Testing

The first aim of this study was to examine interobserver reliability for the method utilized to analyze three-dimensional images and generate values for facial asymmetry. It was hypothesized that values generated for facial asymmetry through three-dimensional stereophotogrammetric analysis by two different observers would be significantly correlated for images of patients with CL/P diagnoses. A series of tests, collectively known as Bland-Altman method, were computed to test interobserver agreement for facial asymmetry measurements between two raters. In the initial step of this process, a one-sample t-test was conducted to compare the difference between symmetry values. This test revealed no significant difference in symmetry values between observers; $t(62) = -0.33, p = 0.74$. A Bland Altman plot was constructed to visually examine the global agreement between the two measurements. Data for mean facial asymmetry was mostly concentrated between 0.5mm and 1.5mm of total asymmetry. No extreme outliers were observed. The data appeared to be distributed evenly above and below the mean difference. Linear regression analysis was also conducted to evaluate for the presence

of proportional bias, or a linear trend of the differences between raters' measurements. The model was not significant, suggesting that differences between measurements of the two observers were proportional; ($F(1,61) = 0.53, p = 0.47, R^2 = 0.01$). The results from these analyses suggest that there is a strong level of agreement between observers, and that the degree of agreement remains consistent through the range of measurements.

The second aim of this study was to examine the relationship between facial asymmetry and social functioning in children and adolescents with CL/P, and to assess how this relationship varies by CL/P diagnosis. Facial asymmetry scores were hypothesized to differ for diagnosis group; specifically, it was predicted that patients with a diagnosis of unilateral CL/P would have higher asymmetry scores than patients with bilateral CL/P. Additionally, it was hypothesized that CLP and CL groups would have higher asymmetry scores than the CP group. An independent samples *t* test revealed no significant differences between unilateral and bilateral diagnosis groups with regard to facial asymmetry scores, $t(74) = 1.17, p = 0.24$. Similarly, a one-way ANOVA revealed no significant differences between the three general diagnosis groups (CL, CLP, and CP) with regard to facial asymmetry scores, $F(2, 78) = 0.84, p = 0.44$.

It was also hypothesized that social functioning scores would differ across diagnostic group, with lower social functioning scores reported by the cleft lip and palate (CLP) and cleft lip only (CL) groups than the cleft palate only (CP) group. A one-way analysis of variance revealed no significant differences between the groups (CL, CLP, and CP) with regard to social functioning scores, $F(2, 78) = 1.20, p = 0.31$. In order to examine the relationship between diagnostic category and social functioning while controlling for age, an analysis of covariance was conducted. Initial analyses found that the relationship between social functioning and age significantly differed across groups, indicating that the ANCOVA model, controlling for age,

was not appropriate (See Figure 1 for more information). To determine whether there were any gender differences for the relationship between diagnostic category and social functioning, an analysis of covariance controlling for gender was also completed. Results indicated that after controlling for gender, significant differences still were not present for social functioning across CL/P diagnosis group, $F(2, 78) = 1.33, p = 0.27$. Overall, no significant differences in social functioning were found between the diagnostic groups, after controlling for gender.

Finally, social functioning was hypothesized to be correlated with facial asymmetry in children and adolescents with CL/P, such that ratings of social functioning would be inversely proportional to facial asymmetry scores. Pearson correlation analysis found no significant correlation between the social functioning scores and facial asymmetry scores, $r(79) = -0.07, p = 0.55$.

CHAPTER FIVE

Discussion

Although previous studies have found that children and adolescents with craniofacial anomalies experience greater difficulties with social functioning than unaffected peers, no studies to date have examined the relationship between objectively measured differences in facial appearance on social functioning, specifically in children and adolescents with CL/P (Berger & Dalton, 2011; Kapp-Simon et al., 1992; Snyder & Pope, 2010). The purpose of the present study was to examine the relationship between facial differences, as defined by facial asymmetry, and social functioning in youth with CL/P. Additionally, this study evaluated the interobserver reliability of the three-dimensional stereophotogrammetric method in quantifying facial asymmetry in the CL/P population.

The first aim of this study was to examine interobserver reliability for the method utilized to analyze three-dimensional images and generate values for facial asymmetry. It was predicted that values generated for facial asymmetry by two observers would be significantly correlated. This hypothesis was supported, as results indicated that there were no significant differences between the facial asymmetry values computed by two different observers. This finding suggests that there is high interobserver reliability in the three-dimensional stereophotogrammetric method in quantifying facial asymmetry in individuals with CL/P. Other recent studies that used three-dimensional stereophotogrammetric assessment in the CL/P population also found this method to have high interobserver reliability (Schwenzer-Zimmerer et al., 2008; Van Loon et al., 2010). There is considerable research that supports three-dimensional imaging as a reliable method in the assessment and quantification of cleft-related facial differences (Mosmuller et al., 2013). The current study expanded upon these previous findings, as it displayed the applicability

and reproducibility of three-dimensional image analysis as an objective assessment of facial asymmetry in a substantially large sample as compared to what previous studies have used. The potential clinical application of quantitative analysis of facial asymmetry via three-dimensional imaging could greatly contribute to surgical intervention planning and postoperative monitoring of facial morphology in youth with CL/P.

The second aim of this study was to examine the relationship between facial asymmetry and social functioning in children and adolescents with CL/P, and to assess how this relationship varies by CL/P diagnosis. It was hypothesized that facial asymmetry scores would differ for various CL/P diagnoses. This hypothesis was not supported. Instead, the current study found that facial asymmetry did not significantly differ based on diagnosis group. Previous research has shown that degree of facial asymmetry differs by diagnostic category, when comparing isolated CL to isolated CP and CL/P cases, as well as when comparing unilateral and bilateral cleft presentations (Bell et al., 2014; Bugaighis et al., 2014; Kuijpers et al., 2015). However, findings of the current study are not consistent with previous findings. A factor that may have played a role in the lack of significant findings for facial asymmetry differences among diagnosis groups may be the unequal diagnosis group sizes. The CL (N=5) and CP (N=8) groups were small, compared to the CL/P group (N=68). Additionally, although patients with unilateral clefts may be perceived as having inherently greater facial asymmetry than patients with bilateral clefts, given the one-sided phenotypic manifestation of the diagnosis, surgical corrections may diminish such dissimilarity in asymmetry. As symmetry is the primary goal of cleft lip repairs, it may be that this objective is being attained to the extent that patients with varying degrees of congenital facial asymmetry display relatively similar outcomes. Interestingly, the presentation of different cleft diagnoses in the sample was not consistent with the incidence rates seen in the general

population. Unilateral cleft diagnoses are more common than bilateral clefts in the general population with a ratio of 4:1; however, in this study the ratio observed was markedly lower, with a ratio of 2.4:1 (Allam & Stone, 2014). This inconsistency may be because a function of the setting in which participants presented for treatment. Given that data was collected within a multidisciplinary team clinic where patients are typically seen for surgical planning and addressing other medical needs related to the cleft diagnosis, it is possible that patients seen in this setting were more likely to have specific concerns that needed to be addressed, such as those imposed by a more complex cleft diagnosis.

Within the second aim, it was also hypothesized that social functioning scores would differ across diagnostic groups. Results also did not support this hypothesis, as social functioning scores did not significantly differ across diagnostic groups. Although the literature has not directly investigated the variance of social functioning among different types of CL/P diagnoses, previous studies found an association between negative social experiences and presence of visible facial differences (Bradbury, 2012; Kapp-Simon & McGuire, 1997; Kapp-Simon et al., 1992; Murray et al., 2010; Strauss et al., 2007). In the current study, diagnosis was conceptualized as a variable representing different degrees of visibility of facial difference, such that the externally visible diagnoses (CL and CL/P) represented a higher degree of facial difference than CP only. Given that results from the current study were not consistent with previous research findings on social experiences and facial differences, it is possible that the use of diagnosis as a proxy for severity of facial difference may have contributed to the lack of significant findings. Alternatively, findings of the present study may suggest that patients with facial differences in general, regardless of visibility or severity, report comparable social functioning. The fact that patients with an observable facial difference (those in the CL only

group) and patients with speech-related problems (those in the CP only group) report similar social functioning to patients with both problems (those in the CL/P group) may denote that having a combination of both problems is not related to differences in social functioning. In other words, the presence of both diagnosis-specific problems may not be associated with poorer social functioning, when compared to the presence of only one diagnosis-specific problem. It should also be noted that the average social functioning score for the overall sample, 82.90, was comparable to the average for the general population, 84.04 (Varni et al., 2003). It is possible that there is no significant variation in social functioning among CL/P diagnosis groups because there are no significant concerns in the population overall, regardless of diagnosis.

The final hypothesis proposed that facial asymmetry scores would be correlated with social functioning scores, such that youth with high facial asymmetry would report lower social functioning. Results indicated that there was no significant correlation between reported social functioning and facial asymmetry for children and adolescents with CL/P in the current sample. This finding may imply that facial asymmetry alone may not be a significant factor in influencing social functioning. As previously noted, there is considerable evidence that there is an association between negative social experiences and differences in facial appearance (Bradbury, 2012; Kapp-Simon & McGuire, 1997; Kapp-Simon et al., 1992; Murray et al., 2010; Strauss et al., 2007). Following the theory that facial symmetry is a strong determinant of facial attractiveness, the current study used facial asymmetry as a measurement of overall differences in facial appearance (Bashour, 2006; Little et al., 2011; Penton-Voak & Perrett, 2000; Rhodes, 2006; Saxton et al., 2011). Since findings reveal that facial asymmetry may not be significantly associated with social functioning, perhaps overall facial appearance may be more accurately represented by another measure, such as degree of residual scarring. In fact, Millar et al. (2013)

found an association between poor psychological adjustment and visibility of postoperative cleft scarring, but no significant associations between psychological adjustment and facial asymmetry. Alternatively, it may be possible that differences in facial appearance alone do not have a significant correlation with the social functioning of youth with cleft lip. Other relevant factors, such as speech intelligibility, may have a more significant impact on social functioning than facial difference in this population.

Limitations

A number of limitations in this study merit discussion, as they may have influenced results and generalizability. Although the overall sample size was large in comparison to other studies utilizing three-dimensional imaging in this population, the sizes of diagnostic category groups were disproportionate. The sample consisted of 48 participants with unilateral CL/P, 20 with bilateral CL/P, 8 with cleft lip only, and 5 with cleft palate only. It is possible that the uneven group sizes skewed the results when comparing the group means to examine differences between the diagnosis groups. The particularly small CL group size could have potentially prevented the findings from being extrapolated. Additionally, overall sample size limited the grouping of diagnoses; consequently, subtle differences may have been present within categorized groups. For instance, the CL/P and CL groups could have been further subcategorized based on complete or incomplete presentations. Moreover, the results of the study may have limited generalizability to other demographic groups, as the sample was predominantly Caucasian/White (86.4%) and it had a high percentage of participants with government-subsidized insurance (75.3%).

Furthermore, examination of the distribution of social functioning scores among the three diagnosis groups found that the cleft palate only group had the lowest mean social functioning

score. This finding was surprising, as isolated cleft palate is the only cleft condition examined in this study that is not characterized by an altered external facial appearance. As previously noted, it is possible that social functioning may be influenced by other factors, such as speech abnormalities. Given that speech problems are common in individuals with CL/P, it would have been pertinent to control for speech-related concerns (Hartzell & Kilpatrick, 2014; Robin et al., 2006). The lack of a uniform measure with which to control for speech impairments is a limitation of the present study, as such impairments could potentially contribute to negative social experiences. For instance, a child with articulation difficulty may experience aversive social cues and peer stigmatization when reading aloud in front of peers.

The use of a single subscale on the PedsQL as a measure of social functioning may have been a limitation to the current study, as it is possible that it was not a comprehensive measure of the variable of interest. It should also be noted that social functioning data collected in this study is self-reported. Although the social functioning subscale on the PedsQL has strong reliability and validity, response bias is still possible and may have influenced patients' scores (Varni et al., 2001). The high degree of face validity in the measure allowed the participants to understand what was being assessed and thus, their response style may have been impacted. Certain variables may have influenced the way in which participants responded to the items. For example, because the patients attended to the items as they waited for providers during their clinic visit, the presence of multiple potential interruptions and distractions may have led to careless or indifferent responding. Being in the same examination or waiting room as their parents while completing the measure may have also affected patients' responses; for instance, some patients may have responded to items in a way that gave an unrealistically positive or negative impression of their functioning to parents. As participants were instructed by the

measure to base their responses on experiences that occurred over the past one month, the data gathered provides a relatively small snapshot of social functioning over time, which may be easily affected by life events, and thus could limit the comprehensiveness of patients' reported functioning. Additionally, this self-report measure requires the respondent to have an intrinsic understanding and enough insight so as to comprehend what their social functioning is like.

Directions for Future Research

Research focusing on social functioning in the pediatric CL/P population has had limited emphasis on objectively measured facial differences and appearance-related concerns. Contributions to this area of research would greatly advance the understanding of this population and add to the provision of care, both medical and psychosocial. The findings of this study suggest that facial asymmetry does not have a significant correlation with social functioning; however, it may be beneficial to assess for relationships between social functioning and other measurements of facial difference. For instance, future studies may attempt to examine the relationship between social functioning and visibility of residual scarring, perhaps by using similar three-dimensional imaging methodology to detect luminance ratio and redness of scarring. Future research investigating facial appearance and social functioning in youth with CL/P should also aim to recruit a larger sample of participants, in an effort to better assess for a wide range of facial differences. Specifically, a larger sample size may provide better homogeneity in group distribution, as well as allow for the examination of additional diagnostic categories (e.g. complete vs. incomplete clefts) that the current study was not able to account for. Additionally, future studies may wish to recruit a sample size with greater demographic diversity in order to increase generalizability.

The literature has supported the idea that social functioning follows a developmental trend in the CL/P population, with negative social experiences and social difficulties being more evident at a early primary school age (5 to 7 years old; Border & Strauss, 1989; Endriga & Kapp-Simon, 1999; Speltz, Morton, Goodell, & Clarren, 1993). Future studies may consider expanding the age range to include participants younger than eight years old, so as to better understand the developmental trend of social functioning in the CL/P population and assess the relationship between social functioning and appearance differences during other phases of medical treatment of a cleft. Secondly, future research may move towards evaluating these variables longitudinally to determine whether there are any changes in the relationship between an individual's facial difference and social functioning over the course of their childhood and adolescence. Studies of this nature would inform the timing and specificity of optimal psychosocial intervention addressing appearance-related concerns. In addition, longitudinal studies would allow further investigation of the impact of changes in facial appearance on patients' psychosocial functioning over time. For example, by collecting pre- and postoperative imaging data, studies may be able to track how repair of the physical presentation of the condition may be associated with changes in social functioning.

Furthermore, future studies should be directed at including measures of speech intelligibility and studying its relevant components in the specific context of social functioning in the CL/P population. A significant limitation for this study was the lack of a validated measure to control for the relationship between speech problems and social functioning. As this was a retrospective study that was conducted using an existing database, introducing additional measures was beyond the scope of the study. Future studies that examine social functioning and facial appearance in youth with CL/P may wish to incorporate a quantitative assessment of

speech abnormalities that can be easily administered to all participants. In this manner, studies may be able to account for several speech dimensions such as articulation deficits, dysfluency, language deficits, and hypernasal speech, which may also affect an individual's social experience. Inclusion of a speech-specific measure may allow researchers to identify and compare the impact that facial appearance and speech problems may have on an individual's social functioning.

In future studies examining the social functioning of youth with CL/P, a measure dedicated specifically to social functioning that can better account for variables such as social skills, social exclusion, and social support may be beneficial as it could provide more detailed information on the individual's overall social functioning. In addition to investigating social behaviors and interpersonal interactions, this measure may examine more intrapersonal dimensions related to the individual's social functioning, such as awareness of their social experience. Using collateral reports of social functioning, such as parental reports, may also be of value, as it would augment the scope of the data. Given the lack of findings regarding social difficulties in this study's sample, as well as the fact that this is inconsistent with previous literature on patients with facial differences, use of a broader measure and/or multi-informant method to assess social functioning may be needed in subsequent studies. Future research may also identify factors that protect youth with CL/P from social functioning problems in order to better understand factors that may be associated with better outcomes in this population. This additional data may help develop interventions geared to enhancing social skills and improving the overall social experience of patients with CL/P.

Regarding the use of three-dimensional stereophotogrammetry, future research should continue to assess its reliability as a tool for examining facial morphology in the CL/P

population. Additionally, future studies should further investigate the feasibility and appropriateness of this new technique for clinical use, as it may have significant clinical applications. For instance, it could potentially be utilized for surgical planning, surgical individualization, and monitoring of improvements in facial morphology over time.

The current study examined the relationship between social functioning and facial differences, as measured by facial asymmetry, in children and adolescents with CL/P. Findings seem to indicate that facial asymmetry is not correlated with self-reported social functioning. Type of physical presentation of the condition, based on cleft diagnosis group, did not appear to be significantly related to social functioning, either. These findings may be interpreted as a lack of association between facial differences and social functioning in the CL/P population; however, these findings may also indicate that further investigation is warranted and the use of other measures for facial difference, such as degree of scarring or overall difference from a norm, may help in this exploration. Regardless, the finding that a number of self-reported social functioning scores represented a clinically important difference from the reported social functioning of a healthy population is meaningful (Varni et al., 2003). Findings of the current study should be used to direct future research in exploration of potential factors that may be related to social functioning in children and adolescents with CL/P.

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Table 1

Demographic Statistics for Participants (N=81)

Demographics	N (%)	<i>M (SD)</i>	Range
Gender			
Female	32 (39.5)		
Male	49 (60.5)		
Ethnicity			
Hispanic	40 (49.4)		
Non-Hispanic	40 (49.4)		
Unknown	1 (1.2)		
Race			
Asian	2 (2.5)		
African	4 (4.9)		
American/Black			
Caucasian/White	70 (86.4)		
Other	5 (6.2)		
Language			
English	54 (66.7)		
Spanish	26 (32.1)		
Other	1 (1.2)		
Insurance			
Government	61 (75.3)		
subsidized			
Private	20 (24.7)		
Diagnostic Category			
Bilateral CL/P	20 (24.7)		
Unilateral CL/P	48 (59.3)		
Cleft Lip Only	8 (9.9)		
Cleft Palate Only	5 (6.2)		
Age		13.20 (2.88)	8-18

Table 2

Descriptive Statistics

Variable	Categories			
	Bilateral CL/P (n=20)	Unilateral CL/P (n=48)	Cleft Lip Only (n=8)	Cleft Palate Only (n=5)
Social Functioning				
Mean	86.75	82.92	81.25	70.00
SD	19.75	19.29	22.80	21.51
Range	30-100	25-100	50-100	50-100
Facial Symmetry				
Mean	0.89	1.02	0.84	1.11
SD	0.38	0.38	0.20	0.50
Range	0.45-1.77	0.54-2.05	0.48-1.14	0.82-2.00

Note. Social Functioning scores on a scale from 0 to 100, with high scores indicating better social functioning.

Facial Symmetry values = Symmetry of nasolabial area in mm.

Table 3

Results of One-sample t-test and Descriptive Statistics for Mean Difference Between Observers

Outcome	M	SD	n	Comparison Value	95% CI for Mean Difference	t	df
Mean difference	-0.01	0.21	63	0	-0.06, 0.04	-0.33	62

Table 4

One-Way Analysis of Variance of Symmetry by Diagnostic Group

Source	<i>df</i>	<i>SS</i>	<i>MS</i>	<i>F</i>	<i>p</i>
Between Groups	2	0.24	0.12	0.84	0.44
Within Groups	78	11.15	0.14		
Total	80	11.38			

Table 5

Independent Samples t-test for Symmetry by Cleft Laterality

	Type of Clefting						95% CI for Mean Difference	t	df
	Unilateral			Bilateral					
	M	SD	n	M	SD	n			
Symmetry	1.00	0.37	55	0.88	0.38	21	-0.08, 0.30	1.17	74

Table 6

One-Way Analysis of Variance of Social Functioning by Diagnostic Group

Source	<i>df</i>	<i>SS</i>	<i>MS</i>	<i>F</i>	<i>p</i>
Between Groups	2	942.84	471.42	1.20	0.31
Within Groups	78	30600.37	392.31		
Total	80	31543.21			

Note. Controlled for gender using ANCOVA.

Table 7

Correlations Between Symmetry and Social Functioning

	Symmetry	Social Functioning
Symmetry	1	-0.07
Social Functioning	-0.07	1

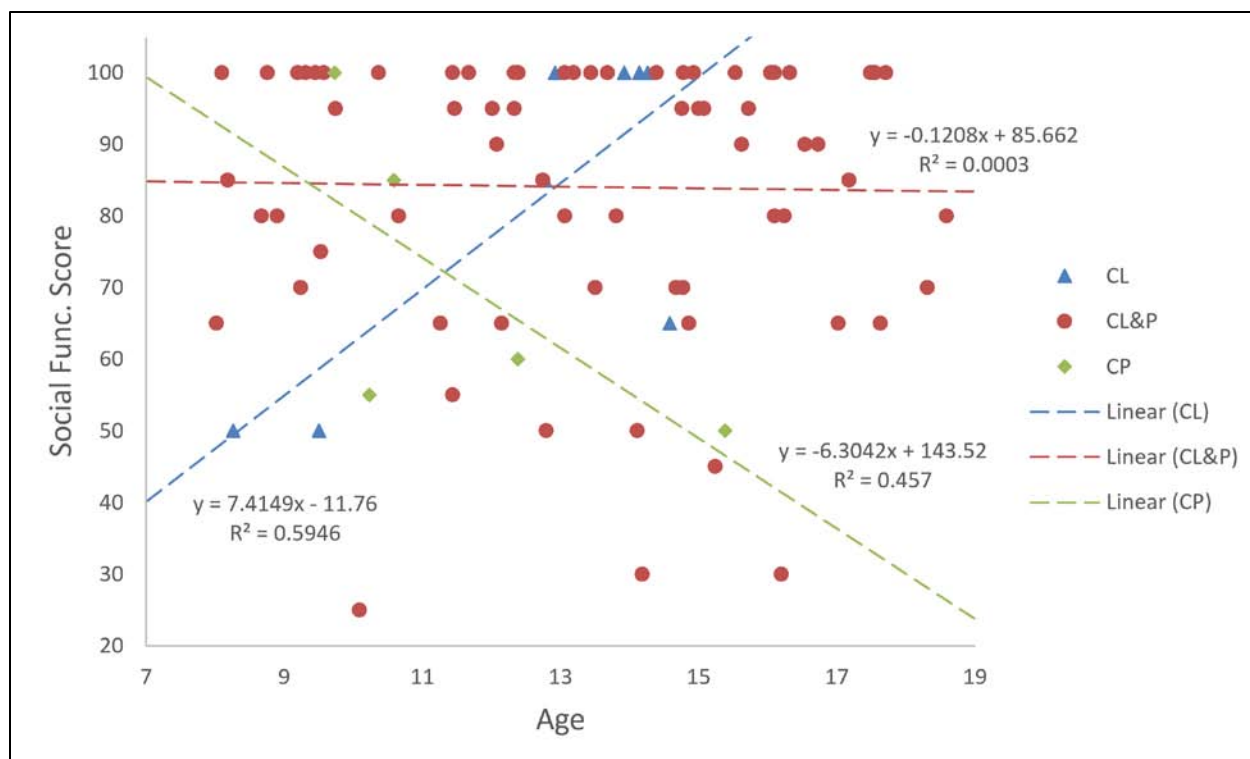


Figure 1. Social Functioning and Age by Diagnosis Group.

Appendix A

Landmark Placement Guidelines

<i>Label</i>	<i>Name</i>
REAR	Right ear, junction of tragus/helical root
RS	Right scalp at superior helical rim
RLOB	Right lobule
REYE	Right eye midlobe
RME	Right mideyebrow
RLC	Right lateral canthus
RMC	Right medical canthus
LEYE	Left eye midlobe
LME	Left mideyebrow
LLC	Left lateral canthus
LMC	Left medical canthus
RA	Right ALA most lateral rim
Subnasale	Subnasale
LA	Left ALA most lateral rim
ROC	Right oral commissure
ULCB	Upper lip cupid bow
LOC	Left oral commissure
LI	Labiale inferius
NA	Nasion
NT	Nasal tip most anterior projection
Chin	Chin midline most anterior projection
LEAR	Left ear, junction of tragus/helical root
LSC	Left scalp at superior helical rim
LLOB	Left lobule
Inion	Inion
BOH	Back of head, most posterior projection
TOH	Top of head, most superior projection
ACR	Alare crest right
ACL	Alare crest left

BIOGRAPHICAL SKETCH

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EDUCATION/TRAINING

INSTITUTION AND LOCATION	DEGREE	YEAR	FIELD OF STUDY
University of Houston – Honors College	B.S.	2014	Biology
The University of Texas Southwestern School of Health Professions	M.S.	2016	Clinical Rehabilitation Counseling

Positions and Employment

2011-2012	Research Assistant at University of Houston – Abramson Center for the Future of Health
2015-2016	Research Assistant at Children’s Health – Fogelson Plastic and Craniofacial Surgery Center
2016	Testing Specialist at UNT Health Science Center – Geriatrics Memory Disorders Clinic

Clinical Experience

2015-2016	Psychology Intern, Consult Liaison Psychiatry Service at Parkland Memorial Hospital
2015-2016	Rehabilitation Counseling Intern, University Rehabilitation Services
2016	Rehabilitation Counseling Intern, Physical Medicine & Rehabilitation Units at Zale Lipshy University Hospital

Presentations and Publications

2012	<i>Educational Campaign for Prevention of Buruli Ulcer in Rural West Africa</i> , Poster presented at the University of Houston Undergraduate Research Day
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Professional Memberships

2016	American Psychological Association
2016	International Association of Rehabilitation Professionals
2016	National Rehabilitation Association
2012	Alpha Epsilon Delta