The Experience of Parents and Adolescents with the Medical Care for the Condition of Cleft Lip and Palate: A Phenomenological Study.

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THE EXPERIENCE OF PARENTS AND ADOLESCENTS WITH THE MEDICAL CARE FOR THE CONDITION OF CLEFT LIP AND PALATE: A PHENOMENOLOGICAL STUDY.

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ABSTRACT


The purpose of this qualitative study was to explore the experiences of adolescents born with cleft lip and/or palate and parents of adolescents with the condition. Individuals born with this condition experience extensive medical treatment from birth to late adolescence with impacts on both individuals and families. This study utilized a phenomenological design to explore and describe participants’ experiences of the impact of this condition, its medical treatment, as well as coping mechanisms utilized in adjusting to the condition. Criterion sampling was utilized to recruit adolescents between the ages of 12 and 18 and parents of this age group, all of whom were impacted by this condition. Twelve participants consisting of both individual parents and adolescents as well as parent/adolescent dyads were recruited and interviewed. Interviews were audio-recorded, transcribed, and analyzed according to phenomenological methods. Several themes emerged from participants’ descriptions of living with the condition of cleft lip and/or palate and undergoing treatment for this condition. Previous research noted the medical treatment for the condition is extensive and the findings of the present study noted its impact on identity, adjustment patterns, and emotional experiences of both adolescents and parents. The medical care was viewed with gratitude by most participants despite identified needs for additional communication.
and support from medical professionals. Parents and adolescents identified coping skills including social support, involvement in community or non-profit groups, and changing perspectives. Adolescents identified aspects of a process of self-acceptance within the context of social interactions and their medical treatment process. Parents identified their role as advocates for their children in social settings as a coping mechanism for social adjustment. The role of support groups in assisting parents and families to cope was key. Theoretical, research, and clinical implications for counseling psychologists and other mental health professionals were discussed. Future researchers are encouraged to consider family systems and feminist and social justice theoretical paradigms. Recruitment difficulties were discussed including the needs for more inclusion of fathers and diverse populations. The results of this research pointed to the need for an increased role for counseling psychologists and other mental health professionals in addressing the needs of children with cleft conditions and their parents. Incorporation of mental health as a discipline within cleft care medical teams would address many of the needs discussed by participants and facilitate successful adaptation and coping. Mental health professionals could provide psycho-education about the treatment process, expectations, and successful coping; screen for at-risk individuals; and provide tailored interventions for individuals, groups, or families. The need for social advocacy is also great among individuals with cleft conditions; mental health providers could work toward decreasing the stigma and increasing understanding of this population within medical, family, or school systems.

Keywords: Cleft lip and/or palate, adolescence, families, resilience, identity, social acceptance, decision making, phenomenology
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CHAPTER I

INTRODUCTION

We all need to realize just how unnatural the process is to a patient, especially a child, and think about what this might mean to a child psychologically. If the goal is to help a child be as normal as possible, where does the bizarre experience of all the treatment and surgeries fit into this? We have to stop concluding that it’s all nothing more than a necessary ‘evil’. These necessary evils are counterproductive to the full range of possibilities that really exist for people like me. (Eiserman, 2001)

In this phenomenological study, I attempted to explore and describe the experiences of adolescents who had the condition of cleft lip and palate (CL/P) as well as parents of adolescents. In particular, I examined the different perspectives of parents’ and adolescents’ emotional experiences related to medical interventions for the condition including interactions with medical teams, decision-making processes, and individual and familial coping mechanisms. By interviewing adolescents and parents, I sought to develop both structural and textural descriptions of these experiences with the ultimate goal of arriving at the invariant essence of this experience (Moustakas, 1994).

Participants of this study included a purposefully selected sample of seven parents (male and/or female) and five adolescents with CL/P, resulting in 12 participants with the final number dependent upon reaching saturation.

This study's findings addressed some of the deficits noted in the literature on cleft lip and palate care such as the emotional impact of cleft care treatment on parents and children individually, the impact on the parent/child relationship, and individual and
familial coping skills used to successfully navigate this condition. As such, it might provide useful information for medical and mental health professionals who work with cleft affected families.

In this chapter, I begin with a discussion of the background and context for the present study. I then present the rationale, statement of purpose, research questions, research approach, and limitations of the current study. I conclude this chapter with an overview of the condition of CL/P and the medical treatment it requires.

**Background and Context**

Cleft lip and palate is a common birth defect that affects 1 in 700 children born each year in the United States (Cleft Palate Foundation, 2013). This condition causes multiple aesthetic and functional challenges and requires interdisciplinary medical care from infancy to late adolescence. Children with this condition typically undergo multiple reconstructive surgeries of their mouth, jaw, and nose. Standard protocols on the number, order, and type of surgical interventions do not exist (Grollemund et al, 2012). They also undergo speech therapy as well as orthodontic and otolaryngological treatments. These interventions aim to restore not only appearance but also the ability to successfully eat and speak and hear optimally. Despite lengthy medical intervention, children and adults with this condition might face deficits in these areas of functioning as well as having visible facial scars. While there is consensus this condition puts the individual and the family at risk for less than optimal psychosocial functioning, the exact nature of this impact is a matter of considerable debate.

Research on the impact of CL/P on parents, children, and adolescents yielded varied and inconsistent results. A body of research made claims to the negative impact of
this condition on both parents and children in terms of psychopathology and/or psychosocial adjustment. Strauss and Broder (1991) noted studies that reflected the medical model consistently focused on the prevalence of psychopathology in patients. One estimate stated 30-40% of children with cleft conditions experienced difficulties with anxiety or depression, behavioral problems, learning problems, or social development (Endriga & Kapp-Simon, 1999). Reduced social acceptance and reduced achievement in school were also noted for children who were affected (Broder, 1997). Researchers also found difficulties with attachment, dependency, and separation anxiety (Pope & Ward, 1997). In parents, studies documented increased stress related to care of a child with CL/P, attachment difficulties, as well as the presence of depression and anxiety in mothers (Montirosso et al, 2011; Speltz, Armsden, & Clarren, 1990).

Yet other research contradicted these claims. For example, several authors found psychopathology was rare among individuals with CL/Ps although psychosocial adjustment problems were more common (Speltz & Richman, 1997). Other research noted children with CL/P and adolescents had neither significant emotional nor behavioral problems (Broder, 1997; Endriga & Kapp-Simon, 1999; Roberts & Shute, 2012). In addition, qualitative studies found positive aspects of the condition and factors contributing to successful adjustment and resilience (Nelson, Kirk, & Caress, 2013).

One writer noted research has failed to consistently show this population sustained negative psychological and psychosocial impacts (Mouradian, Edwards, Topolski, Rumsey, & Patrick, 2006). Another study found “a definitive answer” regarding these individuals’ personality and adjustment problems “cannot seem to be found in the literature” (Canady, 1995, p.122). It is clear the challenges some individuals
with CL/P face are significant while others with the same condition manage it with more success. What is not as clear is the factors that contribute to each.

Research on both parents and individuals with CL/Ps on the psychological aspects of the condition of CL/P examined psychosocial functioning, behavior, social experiences, quality of life, and satisfaction with treatment and appearance (Hall, Gibson, James, & Rodd, 2013). Most of this research utilized validated assessment tools and quantitative methods to assess particular domains of functioning with little focus on the individual as a whole or how these complex factors contributed to overall functioning (Berger & Dalton, 2009).

It was noted the use of quantitative measures alone contributed to deficit-focused data that did not capture the full experience of those affected by CL/P (Canady, 1995; Eiserman, 2001; Nelson et al., 2013). The specificity of focus in these studies necessarily excluded contextual factors and the presence of potentially positive experiences (Eiserman, 2001). Additionally, review studies revealed inconsistencies in variables studied and instruments used to assess individuals limited the generalizability of the CL/P literature (Hunt, Burden, Hepper, & Johnston, 2005). Hunt et al. (2005) also pointed out that while there is a large body of research on this topic in general, very few studies exist or have been replicated on specific constructs or topics within the broader domain of psychosocial functioning. The great variability in treatment teams and outcomes for CL/P also made comparisons between patients of different medical teams complicated—a factor not thoroughly taken into account in most studies (Strauss & Broder, 1993). A significant limitation of the research thus far was the absence of longitudinal studies that
could illuminate the severity and duration of psychosocial problems and the processes involved in the long-term adaptation to CL/P.

A handful of qualitative studies have generated findings that stand in contrast to the deficit-focused quantitative literature (Eiserman, 2001; Nelson et al., 2013; Sharif, Callery, & Tierney, 2013). Qualitative research has been used extensively to elicit patient values and meanings in the healthcare field (Nelson, Glenny, Kirk, & Caress, 2012). One author advises that research with the CL/P population should be focused on meaningful outcomes for affected individuals rather than objective measures (Nelson, 2009). Additionally, the contradictory nature of studies that explore discrete variables might be further explained by qualitative studies that include individual differences and contextual factors (Popay, Rogers, & Williams, 1998).

Bennett and Stanton (1994) noted the discrepancies in the literature reflected both methodological challenges as well as a lack of theoretical specificity regarding the etiology and development of psychological and emotional problems in individuals with CL/Ps. Without understanding the nature of the difficulties, implementing or advocating for successful coping mechanisms is challenging. The nature of the psychological burden on individuals and families is often not even discussed; it is merely assumed and then measured in terms of behaviors or psychosocial functioning. However, if researchers are to assist providers who work with CL/P patients and families, an understanding of the complex processes and varying individual factors relating to successful adjustment is needed. Mouradian et al. (2006) noted research on the psychosocial well-being of children and families that prioritizes “the individual’s subjective experience” (p.151) is critical.
Research on other chronic health conditions consistently found some individuals were more negatively impacted than others, suggesting research on individual factors contributing to successful coping is needed (Frank et al., 1998). Research on chronic health conditions generally and CL/P literature specifically found the severity of the condition was not correlated with psychological adjustment (Lansdown, Lloyd, & Hunter, 1991). Discrepancies between patients and physicians regarding the perceived success of and satisfaction with treatment and studies that found early negative social experiences played a role in later treatment satisfaction both pointed to the idea that assessing satisfaction with this condition required more than simply objective measures. The complexity and subjective nature of treatment were also highlighted by another study that found no correlation between the amount of interventions completed and outcome satisfaction for patients (Semb, Brattstrom, Molsted, Prahl-Anderson, & Shaw, 2005).

These findings led several authors to advocate for an increase in patient-centered care, particularly at the later stages of treatment for CL/P when surgical interventions are mainly elective rather than necessary for functioning (Canady, 1995; Noar, 1991). Because medical interventions impact both aesthetic appearance and functionality, the perceptions and emotional needs of patients are thought to be critical (Canady, 1995). Qualitative studies reported patients and parents also desired to be more involved in medical decisions and to have the emotional impact of medical treatment considered in addition to the medical outcome (Eiserman, 2001).

Mouradian et al. (2006) wrote,

Craniofacial surgeries are often undertaken with the goal of altering facial appearance to improve the individual’s well-being or psychosocial functioning. However, few studies with systematically collected outcome data are available to demonstrate that these interventions actually improve well-being. (p. 150)
While more research is needed on the outcomes of both surgical and non-surgical treatments for CL/P, what is also needed is a better understanding of the subjective experience of individuals and families going through these processes (Mouradian et al., 2006). However, currently few studies exist documenting the views of children or adolescents with CL/P (Sharif et al., 2013). A review of 64 studies found only 21 included patients’ perspectives (Hunt et al., 2005).

In a series of papers in 2000, several researchers noted the need for a change from a deficit-focused to a strengths-focused resiliency model in researching the cleft affected population (Broder, Slade, Caine, & Reisine, 2000; Mouradian, 2001; Strauss, 2001). Both concepts of individual resilience factors and family resilience factors are useful in understanding the CL/P population (Strauss, 2001). Mouradian (2001) noted such a shift “to promote health and well-being seems particularly appropriate in the craniofacial field, given the chronic and unremitting nature of most congenital ... craniofacial conditions” (p.255).

Mouradian (2001) described health promotion in terms of the World Health Organization’s (cited in Mouradian, 2001) definition of health that consisted of not only limiting disease but increasing physical, mental, and social well-being. She noted through focusing on the subjective experiences of individuals, researchers could find sources of resilience as well as stress. Strauss (2001) wrote for societal perceptions of cleft affected people to change, researchers and practitioners working with these populations need to listen to the stories of their experiences and ask questions that focus on coping.
While several studies suggested parenting practices were likely to influence the coping of children with CL/P, “little is known about parenting adaptations that may assist children … to overcome medical and social challenges” (Klein, Pope, & Tan, 2010). The experiences of parenting older children including parenting practices and the quality of the parent/child relationship in this population have yet to be examined. Studies documented discrepancies in parents’ and children’s experiences of CL/P treatment and pointed to the broad influence of parents on children’s understanding of their early condition and treatment through relying on parents’ memories. Yet these studies did not discuss the quality of this information and its potential impact. Research commonly suggested parents should assist children in coping with potentially negative social encounters and managing the stress of medical interventions but few studies within this population explored how this should be done (Sharif et al., 2013).

**Rationale**

Decades of research demonstrated CL/P predisposes individuals and their families to potentially adverse circumstances and numerous challenges. The chronic, yet sporadic medical treatment required for this condition begins in infancy and lasts until late adolescence, causing significant disruptions and making demands upon the resources of both patients and families. Parents with a child who has a chronic health condition face disruptions in their lives due to psychological distress relating to the diagnosis, treatment interventions, and the need to balance the care-taker role with that of a nurturing parent. Children with chronic health conditions similarly face a sequence of obstacles such as extensive hospitalization and treatment, missed school and social opportunities, and stigmatizing experiences. While the impact on children and parents has been studied
separately, research has not examined “the interaction between parent’s and children’s functioning within the family system” (Berger & Dalton, 2011, p. 82).

Chronic health literature pointed to the importance of the quality of the parent/child relationship in children’s adaptations to the stressors involved in long-term medical care and living with a medical condition (Case-Smith, 2004). Resilience literature also noted among other factors such as personality traits and the intensity of the adverse experience, the availability of support is critical in recovering from trauma or other difficult events (Cicchetti, 2010). Research indicated both family and child variables are more clearly associated with adjustment to disease than the disease variables (Lavigne & Faier-Routman, 1992). In addition, Roberts and Shute (2012) noted different coping strategies are used for different stressors and research should therefore aim to examine specific stressors rather than the entire disease process. More research is needed on factors that are condition-specific because the adjustment process to a particular condition varies greatly and is critical to successful adaptation (Broder, 2001).

Qualitative research is best suited for examining interactions and subjective experiences of complex processes. Many authors noted accurate psychological assessment of CL/P patients and their families is critical to promoting well-being and resilience (Broder, 2001; Canady, 1995). Yet, perspectives of children and adolescents and an understanding of family dynamics and relationships are missing from this assessment. This study explored some of these needs and addressed the gap in the literature of the phenomenological experience of adolescents and their parents who experienced the medical care for the condition of CL/P.
Statement of Purpose and Research Questions

The purpose of this qualitative study was to (a) explore and describe how both parents and adolescent children experienced living with the condition of cleft lip and palate, (b) specifically how the medical interventions impacted their lives both individually and in relationship to one another, and (c) what elements assisted them in successfully coping with this condition. Experiences with the medical procedures, decision-making process, interactions with providers, as well as how parents and adolescents coped with stressors individually and as a family unit were all focal points. Through interviews with the participants, a description of the essential elements of this experience was developed. The results of this study provided information on the experiences of undergoing medical treatments for CL/P for both adolescent patients and their parents including descriptions of the processes in detail and contextual factors. The results of this research might inform medical professionals who are members of cleft lip and palate teams, mental health professionals, or others who work with parents of children with CL/P. The study addressed the following research questions:

Q1   What is the essence of the experience of parents and adolescents with the process of CL/P treatment?

Q2   How do parents perceive that their children are impacted by CL/P treatment?

Q3   How do adolescents experience parental support in relationship to their medical treatment?

Q4   What are the coping strategies that parents and adolescents think that they have developed in response to CL/P and its treatment?
Research Approach

After receiving approval from my committee and the university's Institutional Research Board to conduct research (see Appendix A), interviews were conducted with 12 participants. Participants included adolescents who have CP/CL/P and parents of adolescents who have CP/CL/P.

Data were collected in the form of semi-structured interviews and my observational notes relating to the interviews. Semi-structured interviews followed a general topical guideline (see Appendices B and C) but also allowed for the exploration of relevant topics and concerns as they arose for participants (Merriam, 1998). Participants were interviewed individually.

Data were analyzed according to the phenomenological methods of Moustakas (1994). Data analysis began with my attempt to bracket both personal and empirical assumptions and biases about this topic to perceive and reflect upon the experiences of the participants. The first step in analysis, termed horizontalization, involved a broad reading of the data intended to generate a list of significant statements. The next stage of analysis involved grouping these statements into clusters of meaning or themes. These themes were then organized into what Moustakas referred to as the structural and textural descriptions of the phenomenon. Lastly, these two descriptions were combined into the final description of the essence of parents’ and adolescents’ experiences related to the medical process for children with cleft lip and palate.

I utilized several practices to augment the trustworthiness of the study including the use of an expert reviewer, member and peer checks, triangulation of data, the use of an audit trail, and the intentional use of bracketing to increase researcher reflexivity. The
findings are reported in a rhetorical style with direct incorporation of the participants’ quotations in an effort to provide the reader with an in-depth description of the participants’ experiences, thus ensuring transferability of the study.

**Delimitations**

Participants were adult parents of children with CL/P and adolescents with the condition of CL/P:

- Parents might be married, single, or divorced, and have a child (biological or adopted) who has CL/P.
- Parents must have cared for a child with CL/P during their surgical and other medical interventions.
- Adolescents were 12-years-old or over and had completed at least the initial surgeries (and possibly several revisions) for their condition of CL/P.
- All participants were willing to be interviewed.

**Summary**

In this chapter, an overview of the research on the psychosocial impact of CL/P for both individuals diagnosed with the condition and on parents was presented. Inconsistencies in the literature regarding the nature of this impact and the psychological outcomes were noted as were the needs for studying positive coping strategies and resilience factors in this population. Gaps in the literature were identified including the experiences of children and adolescents with CL/P, understanding how this condition impacted parent’s ability to parent their children, and what parenting factors contributed to successful adaptation and coping in children. The present study addressed the gap in
the literature and explored the phenomenological experience of parents and adolescents with the medical care for the condition of cleft lip and palate.
CHAPTER II

LITERATURE REVIEW

We can measure the size of the scar, but is the child better off? We need a much broader view of health to answer that question. (Mouradian et al., 2006)

The psychological, social, and functional impacts of cleft lip and palate (CL/P) for patients and parents have been the subject of much research. Parenting a child with a disability involves multiple complications for parents’ psychosocial functioning. In addition, research on children, adolescents, and adult patients with cleft conditions showed the potential for problems with school success, interpersonal relationships, and psychological stress.

While research has specified some areas of difficulty parents and children face, many areas have yet to be researched. The experiences of parents and children at different stages of development, individual and familial coping strategies for successful adaptation, and the impact on the family system and on fathers were some of the most frequently noted. In addition, while only a handful of qualitative studies have been conducted on this topic, they offered findings in contrast to much of the quantitative work. Most of the literature on children and adolescents who are cleft affected offered comparisons to healthy norm groups that outlined the presence of vulnerabilities but demonstrated little about how they experienced their life with the cleft condition.
Most existing research has focused on understanding outcomes through the use of standardized assessment measures either in terms of symptomology, coping, and adjustment factors or satisfaction with treatment. Fewer studies focused on understanding the complex subjective process related to living with this condition and its treatment process. This research has been quantitative in nature and has focused on identifying deficits and negative reactions (Eiserman, 2001). Many authors noted the use of quantitative research methods might not adequately address the complexity of the lived situation of parents and adolescents who are cleft affected (Eiserman, 2001; Masnari, Schiestl, Weibel, Wuttke & Landolt, 2013; Nelson, O’Leary & Wineman, 2009; Semb et al., 2005). The use of standardized measures to assess the specifics of this population is likely to miss important information unique to the condition (Eiserman, 2001; Hunt et al., 2005). In particular, those who are thriving with this condition might be missed (Eiserman, 2001). And it is precisely by studying those who are thriving that the information regarding factors that enhance resilience can be discovered.

Research has focused on the reactions and experiences of mothers with very little attention on the experiences of fathers or the family unit as a whole. Cross sectional research covering the period of birth through toddler-hood is the most well-researched area with much less attention on later stages of the child's life and how the parental reactions differed through time. Only recently have researchers begun to explore the experience of parents in depth through the use of qualitative methods. This work is “clinically informative” and broadens the portrait of parenting a child with CL/P through parental perceptions of their child's care, its impact on them, and healthy adaptations to this unique parenting experience (Collett & Speltz, 2007, p.139). While a few studies
compared parent and child perspectives of outcomes, psychosocial functioning, and coping strategies, almost none discussed the nature of the parent and child/adolescent relationship and how it impacted functioning.

This literature review begins with an overview of the condition of cleft lip and palate and its treatment as an introduction to the research on psychological functioning. Next, the literature on the psychosocial impact of CL/P is examined. Literature on children and adolescents is reviewed first, followed by research on parents, and finally research that includes parents and children together. Research topics included the psychosocial impact of the condition, coping and adjustment, quality of life, satisfaction with treatment and outcomes, and psychological interventions. To better understand the etiology of the psychosocial impacts on children and adults, several other relevant research areas were reviewed. Findings from the literature on chronic illness and disability conditions were reviewed as a comparison to the cleft lip/palate literature. Research studies on physical attractiveness, stigma, and resilience were also included.

The Condition of Cleft Lip and Palate

A cleft of the lip and/or palate (CL/P) is the fourth most common birth defect, affecting 1 in every 700 babies born in the United States (Bos & Prahl, 2011; Boston Children’s Hospital, 2013). It is also the most common craniofacial anomaly (American Cleft Palate-Craniofacial Association, 2013). Craniofacial anomalies (CFAs) are congenital conditions affecting the growth of the head and facial bones. Craniofacial anomalies impact feeding, growth, dental development, hearing, respiration, and speech (American Cleft Palate-Craniofacial Association, 2013). A birth defect is a condition or anomaly present at birth that causes physical or mental disabilities (Cleft Palate
Foundation, 2013). Cleft lip and palate can lead to disabilities relating to speech, hearing, and cognition (Cleft Palate Foundation, 2013).

In non-children with CL/P during typical neonatal development, separate areas of the face develop individually and then fuse together. In children born with a cleft, the tissue and bones of the upper mouth, nose, and lip fail to fuse, leaving a separation in either the lip, palate, or both (Cleft Palate Foundation, 2013). Clefts can occur solely in the lip or palate or they can occur simultaneously. About half (46%) of affected infants have cleft lip and palate, 32% have isolated clefts of the palate, and 21% have isolated clefts of the lip (Kasten et al., 2008). Clefts of the lip leave a gap in the upper lip that might only impact the lip itself (an incomplete cleft) or might extend up into the nostril and cause a flattening of the nose (a complete cleft). Unilateral clefts affect only one side of the mouth and palate and bi-lateral affects both (Boston Children’s Hospital, 2013). Clefts of the palate impact the roof of mouth and might cause an opening in the back of the mouth (soft palate) or they might cause a complete separation of the roof of the mouth including the hard palate nearer to the gums (McCorkell, McCarron, Blair, & Coates, 2012). Clefts of the palate can be either unilateral or bilateral (Kasten et al., 2008).

The cause of oral-facial clefts is unknown and is likely a combination of genetic and environmental factors (American Cleft Palate-Craniofacial Association, 2009). Maternal use of cigarettes, alcohol, illegal drugs, as well as some prescription medications during pregnancy increases the risk of oral-facial clefts (Cleft Palate Foundation, 2013). Incidence also increases with a family history of clefts. Cleft affected adults have an increased risk of passing on the condition to their children (O'Hanlon, Camic, & Shearer, 2012). The chance of having a second child with a cleft is
about 4% and when two siblings have the defect, the chance rises to 10% for the third child (Suslak & Desposito, 1988). Clefts occur more commonly in boys than in girls; are more frequent among Asians, Latinos and Native Americans; and are least common among African Americans (Bos & Prahl, 2011; Boston Children’s Hospital, 2013). The incidence rates for these groups are 3.6 out of 1,000 for Native Americans, 2 out of 1,000 for Asians, 1.5 out of 1,000 among Indians and Caucasians, and .3 out of 1,000 among African Americans (Mulliken, 2004). While oral-facial clefts can co-occur with other medical conditions or syndromes, about 70% of clefts are isolated birth defects (Centers for Disease Control and Prevention [CDC], 2013). Between 300 and 400 syndromes are associated with cleft lip and palate (Kasten et al., 2008). Prevalence rates for associated malformations or syndromes are imprecise (Kasten et al., 2008).

Clefts cause aesthetic problems as well as functional problems. Surgical repairs are required to repair the clefts of the lip and palate--not only to repair the visible deformities but also to assist the child in eating, breathing, speaking, and hearing (Lockhart, 2003). Palatal separation leads to an inability to create the suction needed to nurse or feed successfully with normal bottles, leading cleft children to require specialized bottles. Lactation consultants as well as occupational and speech therapists can assist parents in learning to feed infants following delivery. Due to feeding difficulties, close monitoring after release from the hospital is needed to ensure these infants continue to thrive. Clefts can also cause complications with chewing and breathing (Kasten et al., 2008).

Cleft children and adults are at risk for speech and language problems due to anatomical and structural differences of the oral region (Kasten et al., 2008). Chronic ear
infections due to a condition called otitis media, or inflammation of the middle ear, can lead to hearing problems or loss. Individuals might require ventilation tubes in the ears throughout their lifespan. Clefts of the palate lead to difficulties with speech including problems with articulation, phonation, and resonance (Kasten et al., 2008). Articulation disorders are most common and relate to the amount of air pressure, which can result in weak consonants and nasal emission that distorts speech. Disordered phonation relates to the larynx and involves compensatory mechanisms such as the overuse of glottal stops and decreased volume as an effort to reduce nasality. The development of vocal nodules can result, which can lead the voice to sound weak or hoarse. Resonance disorders concern the oral, nasal, and pharyngeal cavities. Problems with the velum (soft palate) and the walls of the larynx are termed velopharyngeal dysfunction. In CL/P children, this can result in several resonance problems such as hypernasality, hyponasality, and nasal air emission. The first of these affects the quality of vocal sounds while the latter results in an audible leaking of air through the nose. Velopharyngeal dysfunction sometimes requires additional surgery to correct these areas of the mouth and throat (Kasten et al., 2008).

Dental deformities such as missing or misaligned teeth are also common with most children requiring extensive orthodontic treatment including bone graft surgeries that typically occur around age seven. In addition, cleft children often require further orthognathic surgery in adolescence to realign the skeletal misalignment of the jaws as the upper jaw tends to be disproportionately smaller than the lower jaw.

Children and adults with cleft lip and palate are considered to be disabled when the condition leads to deficits in speech, hearing, or cognitive disabilities (Cleft Palate
Foundation, 2013). Under the Individuals with Disabilities Education Act (IDEA), children from birth to 18 years of age are eligible for public services relating to their condition (Cleft Palate Foundation, 2013). After evaluation by a team of specialists, families are given an individualized family service plan education plan (IFSP) to outline goals relating to special needs such as hearing or speech therapy (Cleft Palate Foundation, 2013). At age three, children receive an individualized education plan (IEP) that accompanies them throughout the school years.

Diagnosis of a cleft lip/palate occurs either in utero through the routine use of second trimester ultrasounds or at birth (Grollemund et al., 2012). In rare cases, clefts of the palate alone are not diagnosed until after an infant has been diagnosed with failure to thrive due to feeding difficulties. Parental reactions to the diagnosis of a cleft have been examined in several studies and appear to be similar regardless of the timing of the diagnosis (Nusbaum et al., 2008). This is discussed in further detail in the section about parental reactions in this chapter.

Treatment for CL/P is an extensive process that begins in early infancy and might continue into early adulthood. Surgical repairs and the related concerns about feeding, hearing, speech and orthodontic health are addressed at various stages of a child's life. The American Cleft Palate-Craniofacial Association (2009) recommends treatment for craniofacial anomalies such as cleft lip and palate be conducted by interdisciplinary teams of specialists who see a sufficient number of patients to maintain expertise. The interdisciplinary team might include the following professionals:

- anesthesiology
- audiology
- diagnostic medical imaging/radiology
- genetic counseling
- neurology
- neurosurgery
- nursing
- ophthalmology
- oral and maxillofacial surgery
- orthodontics
- otolaryngology
- pediatrics
- pediatric dentistry
- physical anthropology
- plastic surgery
- prosthodontics
- psychiatry
- psychology
social work, and speech-language pathology. (American Cleft Palate-Craniofacial Association, 2009, p. 9)

Referral to a craniofacial team is recommended within the first weeks of an infant's life (Kasten et al., 2008). The aim of such teams is to provide not only expert but highly coordinated and efficient care to children with craniofacial conditions. Surgical treatment conducted by specialized teams has been shown to have better outcomes for speech and facial growth, resulting in fewer revisions (Mulliken, 2004). Although many patients are treated by such interdisciplinary teams, many are also treated by independent providers.

In the United States, there is no standardized treatment protocol for surgical repairs and the timing, number, and types of surgical interventions vary greatly depending on the treatment team's experience (Grollemund et al., 2012). The treatment protocol involves the timing, type, and number of surgeries for both primary and secondary (revision) surgeries. Clinical guidelines are based upon expert opinion and case reports in the absence of randomized clinical control trials (American Cleft Palate-Craniofacial Association, 2013).

Craniofacial teams are responsible for assessing ongoing medical complications and for providing information and support to families. Regular team visits monitor growth; ear, nose, and throat; speech; and developmental issues. Initial surgeries for the closure of the cleft lip and palate typically occur sometime in the first year of the child's life. At times, the use of pre-surgical appliances or moldings assists with structural alignment. Additional revisions might occur throughout childhood and into adolescence. In addition, orthodontic treatments begin at around age five and continue through to age 18 (Kasten et al., 2008).
The Impact of Cleft Lip and Palate on Children and Adolescents

Psychosocial Impact

One of the most consistent statements in the CL/P literature is the inconsistency of the findings related to CL/P’s impact on children and adolescents (Collett & Speltz, 2007; Damiano et al., 2007; Hunt et al., 2005; Mouradian et al., 2006). A meta-analysis of this literature concluded the variations in study design and outcome measures made a true comparison impossible (Hunt et al., 2005). Hunt et al. (2005) further noted, “For every study reporting psychosocial problems among those with CL/P, there are others which refute this finding” (p. 282). These authors noted the lack of contemporaneous controls, lack of longitudinal data, variation in study design, and outcome measures made generalizing the data impossible. The lack of population-based samples was also noted as a concern (Collett & Speltz, 2007). Older studies found generally more negative outcomes than more recent ones; whether this was due to improvements in surgeries over time or research design was unclear (Mouradian et al., 2006).

Despite these concerns, most researchers stated while children and adolescents with CL/P were not particularly at risk for psychopathology, they were more vulnerable to psychosocial adjustment problems (Berger & Dalton, 2011). They were also at risk for cognitive deficits and behavioral problems, both of which were implicated in their social adjustment concerns (Broder & Strauss, 1989; Richmen, 1978; Tobiasen, 1987). Understanding why some individuals struggled with the challenges of CL/P while others coped well sat at the core of psychological research in this field (Stock, Feragen, & Rumsey, 2016). In an effort to understand the nature of these social adjustment concerns, psychological research has focused on several areas such as self-esteem and self-concept,
social behavior, satisfaction with appearance, adjustment and coping skills, and quality of
life (Benson, Gross, Messer, Kellum, & Passmore, 1991; Knapke, Bender, Prows,
Schultz, & Saal, 2010; Millard & Richman, 2001; Noar, 1991; Pope & Ward, 1997;

The vast majority of these studies utilized parent or adult retrospective reports
with few studies focusing on children's reports and experiences (Hall et al., 2013; Hunt et
al., 2005). Hunt et al. (2005) noted research found discrepancies between the reports of
parents and children and, therefore, advocated research that reported children’s subjective
experiences. Standardized assessments and quantitative methods are most common
although some observational and qualitative studies exist. Standardized assessments
might detect whether stress levels or psychological functioning were clinically elevated
or not but many authors noted they might fail to detect considerable stress that was not
clinically significant or to understand these variables as they related to this specific
condition (Collett & Speltz, 2007). Although many authors noted the need for
understanding the subjective experiences of this population, very few qualitative studies
exist with child or adolescent participants.

**Cognitive Functioning**

The cognitive functioning of infants and young children with CL/P have been
examined through parent/child attachment and motor or cognitive development. Because
attachment style and cognition have been studied together, both are included in this
section. A review study of cognitive functioning in children with CL/P noted research
has been inconsistent largely due to the variety of assessments used to measure cognitive
functioning, the use of differing types of control groups, and small sample sizes (Roberts,
Mathias, & Wheaton, 2012). Roberts et al. (2012) also noted current data were insufficient to analyze the effect of age on cognitive development.

One longitudinal study examined attachment styles of CL/P at three months and 12 months (Speltz, Endriga, Fisher, & Mason, 1997). This study found that overall, CL/P did not have a negative impact on attachment. However, they noted while mothers were initially less responsive to their babies, this improved over the first year of life, which was hypothesized to be the result of improved appearance due to completed surgeries.

Another study specifically examined whether the timing of the lip repair was significantly associated with the quality of the mother’s attachment and infant development (Murray et al., 2008). This study examined attachment style as well as cognitive and socio-emotional development of infants repaired at birth versus those repaired at three to four months. Measurements were taken at 2, 6, and 12 months. Attachment style and socio-emotional functioning were not significantly different between the two groups at 6 and 12 months but the later repair group showed lower scores on cognitive functioning. The mothers of infants with later repairs were also reported to be less interactive and their infants were reported to look at them less often. The authors hypothesized cognitive functioning later in childhood related to early mother/child interactions, which were influenced by the mother’s ability to accept her child’s facial difference (Murray et al., 2008).

A study by Collett and Speltz (2007) found the CL/P had a protective effect, fostering “increased rates of secure attachment” (p. 139); children with the most severe disfigurements had the highest rates of attachment. Findings from qualitative studies challenged the idea that the infant’s appearance hindered attachment, noting instead the
change in appearance post lip repair was emotionally challenging and often accompanied by feelings of loss and grief (Johansson & Ringsberg, 2004; Nelson, Kirk, Caress, & Glenny, 2012; Stock et al., 2016).

Studies that examined the mental and motor development of infants with CL/P found these children lagged behind norm groups and was more notable in children with only a cleft palate (CPO; Endriga, Speltz, Maris, & Jones, 1998). This was the case for both parent report measures and clinician administered tests. However, they noted CL/P infants were still found to be in the average range compared to test norms, whereas CPOs were slightly lower. In a study that followed children from three months to seven years, children with CL/P were found to be functioning on the same level as controls in cognitive and motor development by age five (Collett & Speltz, 2006).

Despite comparable intelligence to test norms, children and adolescents with CL/P frequently have learning disorders and low school achievement (Broder, Richman, & Matheson, 1998; Kapp-Simon, 1995). In some children, delays in speech and language have been linked to longer term problems with expressive language difficulty, auditory memory deficits, and reading disorders (Richman, McCoy, Conrad, & Nopoulos, 2012). Children with CL/P scored lower than their peers in basic reading skills, phonological memory, and reading fluency (Collett, Stott-Miller, Kapp-Simon, Cunningham, & Speltz, 2010). A study of neurological functioning found CL/P children performed worse on verbal skill measures and verbal memory but not on perceptual and non-verbal measures on the Wechsler Intelligence Scale for Children, yet there were no significant differences in executive functioning in the CL/P group (Conrad, Richman, Nopoulos, & Dailey, 2009). While most learning difficulties in children with CL/P improved through
adolescence and adulthood, speech and language difficulties persisted longer in those who had an isolated cleft of the palate than in those with either cleft lip and palate or isolated cleft lip (Richman et al., 2012).

One study (Knight, Cassell, Meyer, & Strauss, 2015) that utilized a population based sample size through looking at birth registries found children born with cleft conditions ages 5-12 were more likely to receive lower grades in school and had more frequent and lengthy absences from school than their comparison controls. This study found far fewer children with cleft conditions repeated grades than previous studies—a result the authors posited might be related to improvements in health care and intervention services or to differences between study design (clinical versus population based samples). Knight et al. (2015) noted while the study indicated “children with OFC may be more likely to have poorer academic outcomes than their peers without a major birth defect…the magnitude of this association is uncertain because of the small sample size (p. 266). Additionally, the authors noted the percentage of children with poor academic outcome was small and suggested “most children with OFC are successful in school” (p. 266). The authors noted similar studies on older children were needed.

The reasons for cognitive deficits and learning difficulties are not well established. Biologically-based neuronal differences might contribute to these differences (Richman et al., 2012). Recent research noted CL/P individuals have abnormal brain structures that might contribute to their cognitive functioning. Children with CL/P have smaller brain volumes, particularly in the frontal lobes and subcortical nuclei (Nopoulos, Langebehn, Canady, Magnotta, & Richman, 2007). Adult males with
CL/P were found to have normal brain volumes but enlarged frontal and parietal lobes and smaller temporal and occipital lobes (Nopoulos et al., 2001).

Strauss and Broder (1993) noted many factors could influence a child’s ability to perform cognitive tasks such as chronic surgeries, hearing loss, or specific learning disabilities, which were not considered in many study designs. In addition, psychological and environmental aspects were likely to play a role. One study found lower expectations of parents and teachers might be associated with lower rates of achievement (Richman, 1978). This same study found teachers’ ratings of academic ability were dependent upon the severity of the child’s facial disfigurement. Social inhibition might also lead to poorer school achievement (Richman et al., 2012).

The studies reviewed focused primarily on detecting the presence or absence of cognitive difficulties rather than ascertaining its cause. While the current study did not examine cognitive functioning, it is discussed briefly here because of the role it might play in the subjective experience by individuals and families, particularly its emotional impact and the impact of stigma.

**Behavioral functioning.** Behavioral research primarily utilizes parent or teacher reports, although a few observational studies exist. Richman et al. (2012) noted both children and adolescents with cleft lip and palate are at increased risk for externalizing behaviors such as oppositional behavior, non-compliance, and hyperactivity as well as internalizing behaviors such as anxiety, depression, and social inhibition. These behaviors are frequently attributed to frustration at poor speech ability and low satisfaction with facial appearance (Hunt et al., 2005; Richman, Clark, & Brown, 1985). Parent and teacher ratings of internalizing and externalizing behaviors are significantly
higher for these children than for those of non-affected children (Slifer et al., 2003). Yet another study found externalizing problems were less common in a CFA sample than in the normal population (Pope & Ward, 1997). Yet another study found internalizing behaviors were more common in school aged children with CL/P than externalizing behaviors (Murray et al., 2010). Differing results might be due to studying different age groups of children. For example, one study found the presence of internalizing and externalizing behaviors was not noted after the age of seven (Collett & Speltz, 2006).

**Social and emotional functioning.** Numerous studies supported the idea that children with CL/P have difficulties with social and emotional adjustments. Social interaction difficulties remain challenging for these individuals across their lifespans (Snyder & Pope, 2010; Tobiasen, Perkins, Weaver, & Hiebert, 1992). Several studies reported the negative social impact of cleft conditions on adults and stated adults born with cleft conditions were likely to be “educationally disadvantaged” (Stock et al., 2016, p. 551). One study noted adults were less likely to marry, marry later when they did so, and less frequently held positions of responsibility in the workplace (Hutchinson, Wellman, Noe, & Kahn, 2011). However, Stock et al. (2016) found adults in their study reported both successful long-term relationships and high educational and vocational attainment. Again, such discrepant findings call for further understanding of the nature of children’s social difficulties critical to fostering later successful development.

Multiple potential factors influence poor social adjustment in children born with cleft conditions (Pope, Klein, & Bergman, 2016). For example, research highlighted differences in facial appearance, self-acceptance, stigma, social behavior, academic difficulties, and the impact of ongoing medical care on ongoing social relationships as
factors associated with poor adjustment (Pope et al., 2016). Pope et al. (2016) noted these risk factors varied for each individual as did the strengths and resources present to cope with any difficulties.

The impact on self-perception of negative peer interactions due to facial disfigurement and/or speech difficulties is thought to be one contributor to the social difficulties in CL/P individuals. Early development delays due to hearing loss, problems with speech development, and cognitive deficits might influence early social and language skill development (Mouradian et al., 2006). However, findings were inconsistent. One study based on self-report measures of parents of children with CL/P found the most significant predictor of poor psychosocial functioning was being teased for having a CL/P (Hunt, Burden, Hepper, Stevenson, & Johnston, 2007). Self-concept in preschool and school-aged children was lower than for non-children with CL/P in one study (Broder et al., 1998) and higher than non-affected children in another study (Leonard, Brust, Abrahams & Sielaff, 1991). Parents reported their children with CL/P were unhappy with at least some aspect of their facial appearance but more so if they had been teased at school or had visible scars (Murray et al., 2010). However, another study found adolescents were more satisfied with their appearance than parents thought they were (Hunt et al., 2007).

Other research cited parenting practices, either restrictive and negative or permissive and over protective, as causal factors (Rubin & Wilkinson, 1995). In both cases, children with CL/P were considered to be at risk for being more dependent and less autonomous as young children, which negatively impacted their later social development. One study found preschoolers were less autonomous than their peers and displayed more
dependency (Speltz, Galbreath, & Greenberg, 1995). Studies have both endorsed and negated the fact that preschool and school aged children with clefts are more socially inhibited than peers (Richman & Millard, 1997). Children with CL/P have also been observed to be less assertive and less responsive to peers (Slifer et al., 2004).

One study hypothesized later social difficulties were due to negative parent-child interactions during the preschool age (Hutchinson et al., 2011). The authors predicted parents of children with CL/P would display negative parenting such as criticism and controlling behaviors and less positive reinforcement. However, their results did not confirm this hypothesis and found instead that parents of children with CL/P were more supportive and encouraging and their children were more autonomous than those in the control group (Hutchinson et al., 2011).

A study examining social anxiety and withdrawal in seven-year-olds with CL/P found they spent more time alone, had frequent negative peer experiences, and were less likely to engage in group play (Murray et al., 2010). Murray et al. (2010) utilized interviews of parents and teachers as well as observations of children at free play and semi-structured play and assessed children’s social relationships and symptoms of anxiety and depression. The study concluded maternal attachment, the parenting relationship, and the presence and significance of facial disfigurement and speech difficulties influenced psychosocial development.

Another study examined psychosocial functioning through social competence, health-related quality of life, and parenting stress measures (Collett, Cloonan, Speltz, Anderka, & Werler, 2012). Participants included children (aged five to nine) and their mothers. This unique study recruited participants using population-based sampling rather
than clinical or convenience samples. Results showed both CL/P participants and control
groups scored within the average range on all measures and differences between the two
were small. Boys with CL/P had more behavioral problems than did peers while girl with
CL/P had fewer behavioral problems than their peers. Collett et al. (2012) concluded
population-based norms might reflect a more accurate picture of CL/P individuals than
clinical samples. Results also suggested school aged children showed resiliency despite
multiple stressors.

Another study examined self-reported social variables in 10- and 16-year-olds to
determine factors contributing to resilience (Feragen, Borge, & Rumsey, 2009). Factors
included the perception of being teased, satisfaction with appearance, and
depression/anxiety symptoms. For both boys and girls, the presence of a visible cleft
condition was not a factor in either satisfaction with appearance or perceived peer
harassment. Whereas the child’s satisfaction with appearance was critical in terms of
resilience, it was the subjective understanding of satisfaction that appeared to matter.

A Norwegian study compared adolescents with visible clefts, those with invisible
clefts, and a control group without CL/P on standardized measures of self-perception,
social acceptance, and emotional distress (Feragen, Kvalem, Rumsey, & Borge, 2010).
Feragen et al. (2010) examined whether visible clefts influenced perceptions of
friendships, social acceptance, and emotional distress, and whether self-perception would
mediate the associations among friendship, social acceptance, and distress. Adolescents
with a visible cleft reported better functioning on all variables in the study compared to
the comparison group including emotional distress--a finding the authors attributed to
resiliency. The study defined facial difference as a risk factor but the results suggested
self-perception of appearance is more important than objective measures of appearance, as self-perception mediated positive social experiences and emotional well-being (Feragen et al., 2010).

Others looked at the impact a disfigurement had as well as how this disfigurement influenced one’s ability to be socially competent. Pope and Ward (1997) focused on preadolescent peer social competence in a study based on adolescent and parent self-report. The authors hypothesized lower social competence would be associated with greater social anxiety, avoidance of social situations, and fewer friendships. They also hypothesized lower self-reported attractiveness and the presence of internalizing problems would play a negative role in social competence.

Results suggested children with less social competence rated themselves as more anxious and dissatisfied with relationships (Pope & Ward, 1997). Parents of these children also rated them as having internalizing problems and social withdrawal. Success in academics or athletics and satisfaction with appearance all related to good social competence. Dissatisfaction with appearance related to poor social competence. Less socially competent children also reported infrequent friendships. Pope and Ward (1997) suggested some children were at risk for social rejection due to lower social competence. This study was limited by its small sample size and the fact that it grouped various CFA conditions together, thus allowing distinctive factors to be overlooked.

Speltz et al. (1995) noted most individual with CL/Ps were similar to their non-affected peers in terms of social competence but a sub-group developed anxiety and social withdrawal. Pope and Ward (1997) noted that as a whole, individuals with CL/Ps were not significantly elevated on any measures of peer difficulties. They suggested
while children with CFA might not be prone to more difficulties, they might have different risk factors and pathways to social competence and social deficits in this group might differ from norms. While some studies suggested social inhibition was a concern for this population (Richman & Millard, 1997), others found the differences in areas of inhibition and social skills were not statistically significant from control groups (Hunt et al., 2005). One study that examined social inhibition, social skills, and adjustment found positive adjustment was correlated with social skills and a lack of inhibition more than it was to self-perception or self-worth (Kapp-Simon, Simon, & Kristovich, 1992). These authors note that the factors that contribute to the development of positive social skills and disinhibition may contribute to understanding how such resilience can be fostered.

One phenomenological study examined the perspectives of children aged 9-14 on their peer relationships and understanding of their medical condition (Pope et al., 2016). Children in this study were born with a range of cranio-facial conditions. Children reported being engaged in numerous social activities, having frequent contact with multiple peers, and feeling they participated in social activities as well as others their age (Pope et al., 2016). Friendships appeared to be long-lasting and the children reported high satisfaction of friendships. Only a minority reported difficulty sustaining friendships as a result of their condition or mistreatment by peers. The participants were knowledgeable about their conditions and held a balanced view of positive and negative aspects of the impact on their lives. None reported significant concerns for their future with regard to their medical condition.

The presence of teasing and bullying was a consistent finding for this population. A study of 17-year-olds from five European hospitals found teasing was the worst from
ages 8 to 11 and was followed by ages 12 to 15; very little teasing occurred in early childhood and later adolescence (Semb et al., 2005). However, more research is needed to understand the impact of bullying over time and which variables allowed some individuals to manage it while others did not. Studies demonstrating the impact of strategies to overcome the impact of bullying in this population are lacking (Pope et al., 2016).

**Adjustment and coping skills.** Adjustment refers to age appropriate behaviors viewed within the context of life-long development (Wallander & Thompson, 1995). Children and adolescents with chronic physical disorders have been noted to have an increased risk of adjustment difficulties (Lavigne & Faier-Routman, 1992). Factors that assisted in coping with chronic health problems and repetitive medical interventions were common subjects of research (Berger & Dalton, 2009). Research suggested children with chronic health conditions used the same coping skills other children used but different strategies were used for different stressors (Simis, Verhulst, & Koot, 2001). Further research is needed that examines specific disease processes.

Adjustment in children is influenced by many factors such as temperament, adverse experiences, and family and peer interactions (Kapp-Simon & McGuire, 1997). Self-perception might also play a role (Varni & Setoguchi, 1991). Individual factors hypothesized to contribute to adjustment in CL/P children included the visibility and severity of the disfigurement, self-perceived attractiveness, speech ability, coping styles, support from family and peers, family stress levels, and demographic factors (Broder & Strauss, 1991; Endriga, Jordan, & Speltz, 2003; Kapp-Simon, 1995; Pope & Ward, 1997). Some researchers noted psychosocial development and functioning is a complex process.
Therefore, in addition to measuring variables for which standardized measures are available, such as social skills or anxiety, other factors such as emotional well-being and self-confidence should also be used to “properly assess a child’s position in his or her environment” (Hutchinson et al., 2011, p. 498).

Understanding longitudinal adjustment within this population is critical because so many variables studied appeared to change through time. Speltz, Greenberg, and Endriga (1994) suggested CL/P impacts significant tasks within developmental periods--either with some individuals adapting and moving on while others face delays. However, Mouradian et al. (2006) noted while this model makes sense, studies on specific developmental tasks other than attachment have yet to be completed. Recent research suggested the adjustment process is a life-long one and there might be periods during which the stressor of the condition becomes present for individuals. Identifying factors that contribute to these periods of stress as well as successful ways to cope with them are critical (Stock, Feragen, & Rumsey, 2015).

One study examining the extent and nature of adjustment difficulties in adolescents utilized a cross sectional questionnaire design and compared CL/P children to published norm data on psychosocial measures (Berger & Dalton, 2009). Standardized measures were used to assess adjustment, coping strategies, satisfaction with appearance, and social experiences. Adolescent adjustment was within the normal range in this study; however, mothers reported elevated levels of distress in their children. Adolescents had higher levels of satisfaction with appearance than norm controls and most commonly reported coping by means of resignation, social support, and distraction. The most
helpful methods of coping included social support, cognitive restructuring, problem-solving, and emotional regulation.

Discrepancies between parents and children generally mirrored those found in chronic health literature. Berger and Dalton (2009) hypothesized this might be due to over-reporting of mothers who were more sensitized to their children’s stress levels. Adolescents might be underreporting problems as a way to maintain self-esteem or might actually be resilient in those areas. With only a 37% response rate, it might be those experiencing difficulties chose not to participate. Younger adolescents were more likely to utilize social support to cope than were older adolescents. Differences such as these led the authors to note the need for studies of coping at discrete ages as well as longitudinal research.

Berger and Dalton (2009) hypothesized adolescents had higher levels of satisfaction with appearance due to their specific adjustment process as CL/P patients. Their current satisfaction (post treatment) might relate to earlier levels of satisfaction or they might genuinely be more accepting and satisfied with their appearance compared to other adolescents of the same age who had not had their experiences. The results could also reflect overcompensating to maintain self-esteem as other research suggested individuals frequently were unsatisfied with their appearance.

In a review of 60 studies on coping in children and adolescents with chronic health problems, coping methods that were problem focused (problem solving, information gathering) and actively engaged (support seeking) led to better adjustment than did emotional problem solving (emotional expression, avoidance, denial; Compas, Connor-Smith, Saltzman, Thomsen, & Wadsworth, 2001). A cross sectional study by
Berger and Dalton (2011) found adolescents with CL/P reported important factors for their successful coping were positive social interaction with peers, satisfaction with their appearance and speech, and the use of problem-focused coping methods.

One recent study aimed to understand factors in successful psychological adjustment to the condition of CL/P from the perspective of adults (ages 22-77) born with the condition (Stock et al., 2016). The authors noted the individual variation in experience might itself be one reason for such conflicting results in the literature on this population. One final note of the authors was background factors such as age, gender, socio-economic background, ethnicity, and adoption require further research and might represent potential areas of increased risk for adjustment (Stock et al., 2016).

**Quality of life.** Several studies aimed to understand how the aesthetic and functional aspects of the cleft condition related to overall quality of life (Bos & Prahl, 2011; Damiano et al., 2007; Klassen et al., 2012; Munz, Edwards, & Inglehart, 2011; Topolski, Edwards, & Patrick, 2005). Quality of life refers to a “multidimensional and subjective construct” that measures an individual’s assessment of physical, social, and psychological functioning (Moreira et al., 2013, p. 1471).

Research suggested the quality of life of children with chronic conditions and their parents is negatively impacted (Moreira et al., 2013). As one author noted, the success of medical treatment consists not only of the absence of symptoms but the presence of factors that improve the quality of life, which are defined as physical, mental, and social well-being (Damiano et al., 2007). This approach attempts to shift “the focus onto the patient as a whole rather than merely the treatment site” (Munz et al., 2011, p. 71).
A literature review of quality of life (QOL) studies with this population found 26 studies from nine countries with sample sizes ranging from 23-661 (Klassen et al., 2012). Twenty-nine different questionnaires were used. The authors found the content of these measures varied widely, indicating “there are very different ideas about what constitutes the most important components of health” (Klassen et al., 2012, p. 55), and made cross comparisons very difficult. The results of these studies showed highly conflicting results on almost every variable studied. These studies examined areas such physical health, psychological health, behavior, satisfaction with appearance, psychological distress, cognitive health, and social health. Klassen et al. (2012) concluded a QOL scale specific to CL/P was needed and qualitative studies were a useful way to begin to define what variables are considered significant to health for this population.

One article examined QOL in 104 children through the use of a parent report scale designed to measure health-related QOL and distinguish between healthy children and those with chronic conditions (Damiano et al., 2007). In this study, QOL was associated with demographic factors such as higher socioeconomic status (SES), intact marriages, and a higher number of people per household. Overall, CL/P children scored significantly lower than published norms on psychosocial health. Children with less severe speech problems had higher QOL scores. The presence of visible deformity (CL/P, CL) related to overall QOL differently at different ages. Younger children (two to seven years of age) were less impacted by visible deformity than were older children (8 to 12).

A Dutch study examined health-related QOL in 122 participants (aged 8 to 15) and their parents (Bos & Prahl, 2011). Parents and children had significantly different
scores on QOL with children scoring higher than their parents. Cleft lip/palate patients older than 12 scored significantly lower on measures of emotional well-being than did younger participants. They hypothesized older individuals might be more aware of their condition and its impacts and have experienced more treatment interventions.

Another study compared adolescents ages 11 to 18 with a facial difference (FD), either CFA or burns to adolescents with mobility challenges (visible difference), adolescents with ADHD (non-visible challenge), and a control group. All groups with chronic conditions had a similar overall QOL lower than the control group. Groups with visible differences (FD) scored lower than the control group and non-visible difference group on peer relationships but higher on family relationships. Those with a visible difference also endorsed concerns about safety at school.

**Self-perception.** An assumption in the research indicates individuals with CL/Ps internalize negative treatment from others, which impacts their self-concept (Collett & Speltz, 2007). Self-concept, self-perception, and self-esteem are related ideas that are studied to better understand the psychosocial functioning of affected individuals. In a review article, Hunt et al. (2005) noted studies of self-esteem in cleft children and adults revealed discrepant results with some expressing lower self-esteem and others being unaffected. Children with visible clefts reported less satisfaction with their appearance than did those with non-visible clefts in one study (Broder, Smith, & Strauss, 1994). One study even found the self-esteem of children with CL/P was higher than a control group, a finding Broder et al. (1994) attributed to “skewed self-perception or a deliberate attempt to present a very positive image” (p. 282). A study in which children acknowledged having concerns about their appearance also found CL/P children reported having more
friends than others (Broder et al., 1994). Authors noted self-perception of appearance was only one aspect of the larger construct of self-perception (Collett & Speltz, 2007).

Relatively few studies examined self-perception as it related to body image (Crerand, Sarwer, Kazak, Clarke, & Rumsey, 2017). Crerand et al. (2017) noted body image is constructed of multiple constructs including investment in appearance, body image disturbance, or appearance related distress or impairment in functioning.” They hypothesized adolescents with cranio-facial conditions would report greater body image disturbance and lower satisfaction with appearance than non-affected adolescents and this poor body image would negatively impact their quality of life. In their study, adolescents between the ages of 14 and 18 who experienced a craniofacial condition were matched with appropriate comparisons (gender, age, and BMI). Both groups completed several measures of body image--both general and condition specific measures (facial appearance). Crerand et al. found the cranio-facial group did not differ from the comparison group in overall satisfaction with appearance and satisfaction with facial appearance. However, the cranio-facial group had more concerns about specific facial features, whereas the control group reported more concerns with weight or other bodily features (Crerand et al., 2017). Another finding was adolescents with cranio-facial conditions reported significantly less investment in their appearance, suggesting “as a group, appearance was less important to their sense of self-worth.” The authors noted this could relate to healthy adaptation to self-evaluation given their facial condition or a minimizing of the importance of appearance as a self-protective measure. Females in both groups were more invested in appearance and likely to report body image dissatisfaction (Crerand et al., 2017). Youth in this study who reported greater body
image dissatisfaction also reported lower quality of life, a finding that was consistent with the general body image literature (Crerand et al., 2017). Despite lower QOL scores, both groups reported an overall high quality of life. The authors concluded while past research hypothesized craniofacial populations were more at risk for negative impacts of body image satisfaction, this study found adolescents with craniofacial conditions did not experience significantly more problems compared with the general population (Crerand et al., 2017).

Several authors found the medical protocol for cleft individuals might differ from their desire and hopes for treatment (Mouradian et al., 2006; Semb et al., 2005). Alansari, Bedos, and Allison (2013) reported some adult patients compared their lengthy treatment process unfavorably when compared to treatment outcomes. Mouradian et al. (2006) noted no clear outcome studies that could recommend surgical treatment for CL/P as it now stands and questions remain whether treatment is advisable given the uncertainty of the results. Treatment is provided within the context of normalizing children to fit into societal norms and stigma is a powerful force in their lives. Yet, adaptation might require supportive treatments and therapies rather than only medical care (Mouradian et al., 2006). Given these considerations, there should be room in both research and medical care to consider patient values and opinions in order to provide more optimal outcomes and improve quality of life (Semb et al., 2005).

One phenomenological study examined the treatment experiences of adults (age 19 to 54) with CL/P through retrospectively examining their treatment experience (Alansari et al., 2013). Interestingly, this study found those who had a history of stigmatizing experiences early in life consequently had more negative self-perceptions
and treatment experiences. Stigmatizing experiences influenced self-perception, which was found to impact desires and expectations of treatment, perceived benefits/burdens of treatment, and satisfaction and decisions about future treatment. For individuals who experienced stigma, recollections of treatment experiences were mainly “difficult and burdensome” (Alansari et al., 2013, p. 2). They reported aspects of the treatment process contributed to a sense of being “defective or unworthy” including being demoralized in waiting rooms with only similarly affected people and experiencing shock at their post-surgical appearance (Alansari et al., 2013, p. 4). They reported having little importance or control during their treatment as professionals communicated only with their parents and focused exclusively on technical issues of treatment rather than personal ones.

Those individuals who reported less stigma in childhood reported more positive perceptions of their treatment experiences (Alansari et al., 2013). These participants reported positive interactions with doctors; time spent at hospitals and clinics was also time spent becoming closer to family members. Being around others with a similar condition was an experience that provided a sense of belonging and support as well as acceptance of their condition (Alansari et al., 2013).

The attitude toward the need for aesthetic surgeries differed according to stigmatizing experiences as well. Those who had not experienced stigma described a sense of acceptance for the condition and its impact, believing “God makes no mistakes” (Alansari et al., 2013, p. 6) and treatment was simply a way others might view them as normally as they saw themselves. Those who were stigmatized were dissatisfied and looked to surgery as a way to fix their differences.
Importantly, Alansari et al. (2013) found both stigmatizing and supportive interactions could take place both within the family and the treatment team. Still, not enough is known about how either of these could mediate potential stressful experiences and impact coping. This study suggested medical providers need to be aware of the psychological impact of this condition and treatment on an individual patient’s life. Alansari et al. noted the desire for optional revision surgeries might in fact be a “potentially covert pursuit of psychological outcomes through physical ones,” which could lead to disappointment with treatment outcomes (p. 7). This echoed other research that similarly found it common to experience high satisfaction with treatment with an equally strong desire for further treatment.

**Psychosocial Impact of Cleft Lip/Palate on Parents**

Research on parental experiences largely focused on the mothers of infants and of very young children; considerably less emphasis was placed on parenting older children and on the experiences of fathers (Nelson, Glenny et al., 2012). The relative absence of information on fathers mirrored research on chronic health conditions (Swallow, Macfayden, Santacroce, & Lambert, 2011). Considerable research has focused almost exclusively on the negative aspects of parenting a child with a cleft, specifically with identifying risk factors and deficits (Eiserman, 2001). This aforementioned body of research focused on the experience of caring for a child with a cleft from the perspective of sorrow and loss with little consideration of contextual factors and positive aspects (Baker, Owens, Stern, & Willmot, 2009; Eiserman, 2001). However, some recent qualitative studies have begun to “reveal a wider range of experience” including potential
rewards, complex emotional processes, and factors that enhance positive coping (Nelson, Glenny et al., 2011, p. 11).

The responses of parents to parenting a child with CL/P fell into two categories in the literature. The first looked at the psychological and emotional impact of parenting a child with CL/P including parenting stress levels, quality of life, and coping and adaptation. The second examined responses to medical care including satisfaction with care, treatment outcomes, and recommendations to providers. Although some anecdotal evidence existed, no studies specifically focused on the parent/child relationship in this population.

**Parental reactions to diagnosis.** Research on parental responses to diagnosis provided useful information about common parental reactions as well their perceptions and needs for medical support. The research included reactions to meeting the newborn, informing interviews after birth, as well as prenatal screenings. Both qualitative and quantitative studies in many countries documented a common parental reaction to the diagnosis of a cleft lip and palate involved feelings of shock and a sense of grief and loss (Byrnes, Berk, Cooper, & Marazita, 2003; Nelson, Glenny et al., 2012; Strauss, Sharp, Lorch, & Kachalia, 1995). Parents typically experience anger, guilt, and anxiety related to their child's future well-being (Bradbury & Hewison, 1994; Nelson, Glenny et al., 2012). In addition, parents consistently request information regarding the long-term implications of having a child with a cleft including treatment protocols, related health issues, and psychosocial concerns (Johansson & Ringsberg, 2004; Knapke et al., 2010; McCorkell et al., 2012). Parents request and are reassured by photos of other children with CL/P before and after surgery and by being given contact information for other
parents with children with CL/P (Kuttenberger, Ohmer, & Polska, 2010; McCorkell et al., 2012).

In contrast to most of the research, a recent qualitative study examining parents’ experiences with perceptions of support found both negative and positive reactions to the birth. While shock was still a common reaction, joy, acceptance, and a feeling of purpose in caring for a child with special needs were also noted responses (Johansson & Ringsberg, 2004).

Parents also had a need to understand the causes of the condition and often experienced a period of self-blame and guilt (Bradbury & Hewison, 1994; McCorkell et al., 2012). A survey of parents examining the type of causal attribution they made regarding the cause of the birth defect demonstrated the parents who self-blamed had significantly higher levels of anxiety and stress than parents who found no cause or attributed it to something external to themselves. Parents who self-blamed still did not experience clinical levels of anxiety or depression (Nelson et al., 2009).

Ideally, when a diagnosis is made, it is soon followed by an informational meeting with a medical professional. However, the character of these meetings in terms of their extent, nature, timing, and level of expertise varies greatly both internationally and nationally. Research demonstrated initial informational meetings with medical professionals had “long term effects on family's abilities to accept and adjust to a diagnosis” as well as on the parent/child relationship (Byrnes et al., 2003, p. 308). Studies consistently found parents who received accurate information and emotional support from specialists shortly after the birth of their child had better outcomes in terms
of immediate adjustment (Johansson & Ringsberg, 2004; Knapke et al., 2010; McCorkell et al., 2012; Nelson, Glenny et al., 2012).

Parents responded well to information that was accessible, individualized, and provided by informative and supportive professionals (American Cleft Palate-Craniofacial Association, 2013; Young, O’Riordan, Goldstein, & Robin, 2001). Immediate informational needs of parents included coaching on feeding, protocols for surgical treatment, surgical recovery, prognosis, and consultations with cleft care teams (Young et al., 2001). One study specifically examined the parents of cleft-affected children's perceptions of informational interviews at a large Midwestern hospital (Byrnes et al., 2003). Less than half of the parents felt the information conveyed was adequate. The expressed needs of the parents were for medical professionals to express caring, to make more effort to comfort them, to allow them to share their own feelings and to talk, to express their feelings, and to provide referrals to other parents with cleft children (Byrnes et al., 2003). Nelson et al. (2013) noted the emotional needs of parents have not been fully examined.

Parental adjustment at the time of diagnosis clearly varied greatly; some parents needed no adjustment time while others expressed varying levels of difficulty with the initial adjustment. Some evidence indicated parents who knew about the cleft condition prior to birth experienced fewer difficulties those who found out at birth (Bradbury & Hewison, 1994; Johansson & Ringsberg, 2004). While some participants reported their initial shock resolved quite quickly into acceptance, others reported anxiety, guilt, and shame about the child's condition for many years (Bradbury & Hewison, 1994). Maris, Endriga, Speltz, Jones, and DeKlyen (2000) found initial negative emotions in reaction to
the diagnosis subsided as the parents accepted the child and began the care-taking process.

**Parent’s satisfaction with medical care.** Studies consistently found general obstetrical teams lacked specific knowledge about cleft conditions, particularly feeding issues; as a result, insufficient support was provided to new parents of cleft children and compromised the infant's immediate care and well-being (Stock & Rumsey, 2015; Young et al., 2001). Without early expertise and coaching for parents in feeding cleft infants, many return to the hospital with unnecessary medical conditions such as failure to thrive and dehydration (McCorkell et al., 2012). Research suggested the quality of information and support received initially impacted later adjustment to cleft conditions (Stock & Rumsey, 2015). For example, parents reported unnecessary stress and frustration due to being given conflicting information regarding feeding techniques within the same hospital (Johansson & Ringsberg, 2004). Access to reliable information regarding the condition and its long-term care was also frequently absent in general maternity wards (American Cleft Palate-Craniofacial Association, 2013). Parents in one study reported they provided education to nurses who knew nothing about the condition (Johansson & Ringsberg, 2004). Similarly, a study in Switzerland found the lack of knowledge and insufficient care in maternity hospitals caused stress and anxiety to parents with new born children with CL/P (Kuttenberger et al., 2010).

Medical staff were also reported to struggle in providing emotional support to parents (Byrnes et al., 2003; Johansson & Ringsberg, 2004; McCorkell et al., 2012). Negative experiences such as being separated from their child at birth for a period of time without adequate explanation, witnessing doctors discussing their infant but not being
given any information or a diagnosis, and even being told a diagnosis without any further information were all examples in the literature of the lack of emotional support given to parents at the time of birth (Byrnes et al., 2003). Parents also reported subtler ways in which support was lacking such as reassurances from staff that everything would be okay, which emphasized to them a lack of acceptance of their children as they were at that time (Johansson & Ringsberg, 2004).

In contrast to these studies, one qualitative study in Ireland demonstrated it was possible to communicate relevant and appropriate information and support to parents at the time of the birth, noting this is a “basic but fundamental requirement in an often traumatic and emotional period” (McCorkell et al., 2012, p. 27). This unique study evaluated the services of a cleft liaison service providing in-home visits for parents of young infants. Parents valued the consistent, individualized emotional support and the continuity of care throughout the early treatment phase. In-home support provided an additional level of one-to-one support that was not rushed and could attend to the individual needs of families.

Parents’ involvement with medical decision-making. Only three studies examined parents’ involvement in treatment and the decision-making processes for treatment. A survey study of parents found although 90% of parents felt they understood a lot about treatment, 79% wanted more information, and 36% wanted to participate more in treatment decisions (Pannbacker & Scheurle, 1991). A grounded theory study examined beliefs and motivations that influenced parents’ decision-making (Nelson, Caress et al., 2012). Parents of children age 20 weeks to 21 years participated. Although the study intended to include parents who elected not to complete treatment for their
child, none could be located. Findings suggested parents’ decision-making and relationship with their child involved a “moral dimension” the authors termed “doing the right thing” for their child (Nelson, Caress et al., 2012, p. 799). Parents expressed a strong sense of responsibility to do what was best for their child, such as putting their child’s needs first, and following through with medical protocols that resulted in a “pro-treatment approach to decision making” (Nelson, Caress et al., 2012, p. 800). Parents in this study felt an obligation to accept any available cleft treatments with little sense of “explicit deliberation” (Nelson, Caress et al., 2012, p. 800). Only one parent refused a recommended treatment and expressed doubts about a treatment protocol. Parents instead expressed anxiety about not accepting treatment recommendations. They attempted to manage their own and their children’s complex emotions primarily through surgical treatment despite reports of experiencing a sense of conflict over “normalizing” the child they love. The expectation for treatment results were high, resulting in emotional upset and guilt if surgeries did not succeed as planned.

Parents experienced a heightened sense of trust in physicians because of their status as doctors even to the point of forgiving them for mistakes or failures in treatment. This allowed them to “maintain their vision of practitioners as…trustworthy, legitimize decisions to pursue their child’s treatments and continue with future treatments” (Nelson, Caress et al., 2012, p. 800). In part, this reliance on professionals reflected a sense of powerlessness in dealing with the condition. Parents also admitted that in leaving decision-making to professionals they then avoided the burden of emotional responsibility later on.
A mixed method pilot study also examined the perspectives of parents on their decision-making process (Eiserman, 2001). In this study, parents reported a desire to be fully informed and involved in every aspect of care in order to make good decisions for the long-term well-being of their child. Parents perceived in the medical perspective, performing procedures was considered to be a “necessary evil” not discussed or contemplated in any way (Eiserman, 2001, p. 267). Parents suggested that instead, concern should be for the overall well-being of the child and the family. Conflict over care resulted from the differing perceptions of doctors and parents toward the needs of the child and lack of communication about these differences. For example, parents felt doctors advocated for very early repairs because of their perceptions that a repaired child would be easier for parents to tolerate, whereas the parents already felt full acceptance of the child and felt having time to explore various options would have been more beneficial. Some parents also felt excluded or judged for disagreeing with decisions of doctors (Eiserman, 2001).

**Parents’ satisfaction with cleft care teams.** Research demonstrated overall parental satisfaction with the quality of services provided by cleft specialists or cleft care teams (Nelson, Glenny et al., 2012). Parents consistently expressed confidence in specialist teams because of their expertise and because of the continuity of care they provided (Canady, Means, Wayne, Thompson & Richman, 1997; Johansson & Ringsberg, 2004). However, Nelson, Caress et al.’s (2012) study pointed to the possibility that some of the high levels of satisfaction with care might reflect parents’ over-reliance on medical teams as a means of coping with feelings of powerlessness.
No internationally agreed upon outcome satisfaction measure exists for the aesthetic aspects of CL/P (Semb et al., 2005) as aesthetic outcomes are always subjective in nature. Both parents and patients perceived facial appearance based in part upon their psychological histories and experiences. In addition, difficulties in gathering data on satisfaction with outcomes of appearance included the potential for response bias and the likelihood satisfaction was influenced by the amount of behavioral commitment individuals had invested in the treatment process in terms of time, money, and emotional commitment (Semb et al., 2005).

For the purposes of understanding the complex adjustment process to CL/P for patients and parents, an understanding of the subjective aspects of satisfaction is more relevant than any objective measure. As discussed in the section on self-perception above, there was evidence to suggest satisfaction with appearance was related to self-concept, expectations, and psychological well-being.

One study assessed both parental and adolescent satisfaction with outcomes through a self-administered questionnaire that focused on team accessibility, the manner in which care was delivered, and treatment results and desires for further treatment (Semb et al., 2005). This study concluded patient/parent satisfaction had no association with the amount of treatment or success of treatment as judged objectively. Overall, patients and parents in this study had similarly high levels of satisfactions with outcomes despite 65% of patients and parents who wanted further treatment, particularly in revising lip and nose repairs. Response bias might have partly explained these results.

Pointing to the lack of research on parents’ satisfaction with care for older children and adolescents and for in depth understanding of parents’ views on delivery of
cleft care services, Nelson et al. (2013) conducted a grounded theory study regarding these concerns. Results indicated satisfaction with services rested not only the expertise of the practitioners but also on their ability to ease the medical process for their children including providing a continued long-term relationship and providing care and concern to frightened or confused children. Parents in this study expressed a need to understand the surgical process in more detail including risks, potential for failed surgeries, and revisions and postoperative care following surgery. Retrospectively, some parents felt they did not adequately understand procedures, terminology, or choices about surgical procedures.

The postoperative condition of their child was often a shock and cause for alarm as were feeding difficulties following surgeries. Importantly, Nelson et al. concluded that even when parents had experienced surgery with their child because each surgery is different, the need for in-depth information is always there. Across age ranges and types of treatments, the authors found parents expressed need for information, preparation, and support for themselves and their children (Nelson et al., 2013). Another qualitative study explored parents’ perspectives on the services of one cleft team and also found parents desired more preparation about post-operative care, feeding, and the length of the recovery period (Knapke et al., 2010).

The availability and expertise of cleft care teams varies from country to country. Mulliken (2004) noted Scandinavia has a long tradition of centralized cleft care in large hospitals. In the United Kingdom, the National Health Service mandated cleft treatment occurs in highly specialized centers that routinely see large numbers of patients. In the United States, no such regulations exist and parents can choose from a wide variety of cleft care providers. Geographical location, insurance coverage, and adequacy of referral
information influence these decisions. Specific research on the involvement of parents in decision-making related to the choice of a treatment team, or to the type of medical procedures, and how these impact outcomes is very limited.

**Parent’s emotional reaction to the condition of cleft lip and palate.** Until recently, most of the literature examining parents’ emotional reactions to their child's condition of cleft lip and palate consisted of quantitative studies that focused on identifying deficits and disorders (Eiserman, 2001; Nelson, Glenny et al., 2012). In part, this view developed from the almost complete focus on the early period of a child's life, which is characterized as a period of crisis for many individuals (Baker et al., 2009; Bradbury & Hewison, 1994). For example, several studies that noted elevated levels of maternal stress in mothers of CL/P children were carried out during the infancy period (Pelchat, Bisson, Bois, & Saucier, 2003).

Other research suggested by the preschool years, maternal stress levels were similar to control groups showing some level of adaptation to the condition occurred over time (Nelson, Glenny et al., 2012). Yet several authors noted sub-clinical stress levels still deserved the attention of researchers and clinicians (Collett & Speltz, 2007; Nelson, Glenny et al., 2012). Yet parents frequently requested psychological services be available, which indicated unmet emotional support needs (Johansson & Ringsberg, 2004; Kuttenberger et al., 2010). The nature of these needs past the early infancy stages has not been well established.

Studies on the parenting stress in parents of children with CL/P found mixed results. While a subgroup of parents appeared to experience symptoms of stress, depression, and feelings of low self-evaluations of competence (Dolger-Hafner, Bartsch,
Trimbach, Zobel, & Witt, 1997), they did not generally experience the same level of distress as did parents of children with other medical conditions (Pelchat et al., 2003). While general studies showed parenting stress related to poor outcomes in adjustment and functioning for children, no studies specifically linked parenting stress among this population to poor psychosocial adjustment in children with CL/P in part because of the lack of any longitudinal data (Pope, Tillman, & Snyder, 2005).

Parents reported being influenced by initial surgeries in the following ways: financial strain due to lost income for time taken off during treatment and its costs, difficulty planning the future, lack of family or free time, difficulty finding child care, strained family and social relationships, lack of certainty about how to parent a child with a special need, and disruption to sibling's lives with increased stress (Kramer, Baethge, Sinikovic, & Schliephake, 2007). Yet another study (Speltz et al. 1993) found mothers reported higher levels of emotional distress and marital strife than did controls.

Another study found five stressors typical in parents of children age 20: their child’s speech ability, future ability to marry and become independent, the nature and extent of medical visits, anxiety about witnessing painful treatment, and concern about the child’s appearance (Lei, Wang, Cheng, Chen, & Chin, 2010). Many of these were the same concerns noted by parents of newborns, indicating the life-long level of uncertainty and worry experienced by parents.

A recent grounded theory study also found the presence of conflicting emotions and tension to be a theme for parents across their child’s lifespan. Parents of children of varying ages were chosen for this study to represent different developmental stages in this population; 35 parents were interviewed and since 11 participants were fathers, this study
was more representative than most studies (Nelson, Caress et al., 2012). Results suggested parents experienced conflict caused by experiencing their child as normal while knowing others would not. A conflict around surgical treatment was also noted with parents expressing eagerness for their children to complete treatment as well as a strong desire to protect them from pain and distress.

Parents attempted to control their feelings to be strong for their child and coped with their anxiety through placing trust in and “surrendering themselves and their child” to their medical team (Nelson, Caress et al., 2012, p. 351). Parents also reported a sense of guilt for the multiple surgical repairs and both the physical and emotional pain this caused them. Another theme of this research was the sense of ongoing uncertainty faced by the parents due to the long-term nature of treatment and unclear prognosis (Nelson, Caress et al., 2012).

Parenting style and parent/child interactions are impacted by the level of stress parents experience (Krueckeberg, Kapp-Simon, & Ribordy, 1993). Higher levels of parenting stress have been associated with both restrictive and inattentive parenting practices. One study found mothers were over-controlling and discouraging of children’s autonomy (Pope & Ward, 1997). Another study found parents were more tolerant of behavioral problems than were controls (Tobiasen & Hiebert, 1984).

While researchers have theorized that restrictive practices result in the discouragement of autonomy in children, thereby negatively impacting their problem-solving capacities, several studies did not support this theory (Krueckeberg et al., 1993; Pope & Ward, 1997). Over-protectiveness in parents might be beneficial for the development of CL/P individuals (Hutchinson et al., 2011; Klein, Pope, Getahun, &
Thompson, 2006). Research from China suggested parents’ over-protectiveness served as a buffer to negative reactions from others and increased self-perception in CL/P adolescents (Hutchinson et al., 2011). Others noted parental anxiety and concern might not impact children’s social struggles but merely reflected parents’ own experiences (Pope & Ward, 1997). How parenting practices actually impact children and adolescents with CL/P is largely unknown.

The psychological health of mothers has been examined with mixed results. Murray et al. (2008) found significant levels of depression in mothers of children with CL/P although they did not find any impact on the mother/child relationship as a result. Montiroso et al. (2011) studied mother and child interactions in small groups through behavioral observations. Findings suggested cleft-affected infants were less communicative and engaged with their mothers and mothers appeared to be disengaged and depressed, which they attributed to the infant’s impaired facial and speech cues. Yet other research suggested while a subgroup of parents experienced symptoms of stress and depression, most did not generally experience the same level of distress as parents of children with other medical conditions (Collett & Speltz, 2007; Pelchat et al., 2003).

A German study examined mothers of children over one-year-old for prevalence rates of anxiety and depression by comparing them to a previous study of mothers whose children had isolated speech disorders and found no clinically significant levels of depression and anxiety in their sample (Weigl, Rudolph, Eysholdt, & Rosanowski, 2005). Research suggested the overall quality of life during the first two years of a cleft-affected child's life is less impacted than many other chronic conditions (Kramer et al., 2007).
One study compared parents of preschoolers with a CFA to those without measures of parenting stress, social support, and parenting style and on how these impacted the social skills of the children (Krueckeberg et al., 1993). This study utilized both standardized measurement tools and observation of children at play. Krueckeberg et al. (1993) hypothesized that parents of CFA children would have higher levels of stress and more restricted social support. The results did not demonstrate their hypothesis. Their finding contradicted others studies of younger children; the authors suggested this was due to the use of a non-disease specific measurement tool for parenting stress, which might not be adequate to detect CFA specific stressors. However, Krueckeberg et al. also noted the parents in their study were receiving regular emotional support from their medical team, which might also have made a positive difference.

While many studies focused on the impact on mothers and their response to parenting a child with cleft lip and palate, the perspective of fathers was almost entirely lacking from the literature (Klein et al., 2010; Nelson, Glenny et al., 2012). This followed a pattern in pediatric research in general (Stock & Rumsey, 2015). The social roles of fathers with regard to care-taking children's medical needs, the manner of showing emotion, and seeking health-related information might explain this in part (Swallow et al., 2011). Fathers reported feeling they had nothing to add in comparison to mothers, which might have impacted their lower response rates (Berger & Dalton, 2009). Research suggested paternal involvement in management of chronic illness was associated with more satisfactory parent/child relationships as well as increased treatment adherence in adolescents (Swallow et al., 2011). Further research on fathers’
involvement in medical management for their children is needed because of the increasing amount of time fathers spend with their children (Swallow et al., 2011).

A recent exploratory study examined fathers' perceptions of parenting a child with a craniofacial condition, four of whom had a CL/P (Klein et al., 2010). The small sample size and voluntary recruitment made generalizations beyond this study difficult; however, the results echoed many of the findings in the chronic illness literature and research on mothers. Topics for interviews included fathers’ feelings about their child’s future, ways in which they were involved in supporting social development, and their experience parenting a child with a CFA. Results indicated that overall, fathers had positive attitudes toward their children and utilized proactive parenting styles (Klein et al., 2010). All fathers noted many strengths in their children. Despite different conditions, the fathers’ concern for their child related to how other people’s perceptions might negatively impact their self-esteem, confidence, and opportunities in life. Fathers actively encouraged their children to include friends in their social activities (Klein et al., 2010).

A recent qualitative study in the United Kingdom examined the experiences of 15 fathers of children born with cleft conditions--both syndromatic and nonsyndromatic (ages 4 months to 24 years; Stock & Rumsey, 2015). Results of this study suggested fathers experienced several key concerns regarding their children and were not prepared by their medical team for addressing these concerns. Specifically, fathers reported concerns about the cause of cleft conditions and experienced both anxiety and guilt as a result. They also reported feeling unprepared for public reactions to their child’s condition and how to cope with these. They felt unprepared for their child’s appearance following surgery and how to address the topic of the condition, teasing, and acceptance
with their children. Fathers in this study reported the need for increased support and information from the medical providers specifically. Social support from other families was reported to be critical. While some fathers reported a lack of support specifically for fathers in the social networking arena, others felt the support their wives received in this area positively impacted them as well. Fathers in this study noted the importance of social support from other families facing the same treatment process, both in terms of providing and offering support to others.

**Parental coping and adaptation.** Parental adjustment, adaptation, and coping have been a frequent focus of study with little consensus on the overall impact of having a child with cleft lip and palate (O'Hanlon et al., 2012). Families must adapt to successfully accept a child with a healthcare condition and parental anxiety about a child's condition and well-being can become contagious and shape a child's view of him/herself (Kapp-Simon, 2006; Pope & Ward, 1997). Families play a large role in health-related behaviors and beliefs and can influence children’s compliance with treatment, perceptions of their condition, and health behaviors (Broder, 2001). While general studies showed parenting stress was related to poor outcomes in adjustment and functioning for children, no studies specifically linked parenting stress among this population to poor psychosocial adjustment in children with CL/P, in part because of the lack of any longitudinal data (Pope et al., 2005).

Researchers suggested it is critical to assess families for their coping strategies, communication, support, and cohesion; yet how parents cope has not been thoroughly studied (Broder, 2001). Eiserman (2001) wrote, “Little attention has been given to understanding the individuals who appear normal on the variables in which comparisons
are made between affected and non-affected samples” (p. 236). In studies where no clinically significant difference was found between cleft individuals and control groups, researchers cited faulty research methods or sample size for the lack of such findings. Eiserman indicated researchers might be mistaking “equating different with deficient” (p. 236) and the study of successful individuals might contribute to understanding more successful long-term adaptations.

Based on questionnaires and interviews with adults and parents, Eiserman (2001) explored “contributational” perspectives on facial differences (p. 237). The author noted precedents in the strengths model of disability, which took “into consideration how those affected by disability, might have very different but valued experiences attributed to disability” (Eiserman, 2001, p. 237). Eiserman’s study sampled parents of children from age 2 to 15 with a range of craniofacial conditions (eight CL/P), eight adults with CL/P, and two others with acquired facial disfigurement from disease and/or accident. Only positive outcomes were reported to produce an article on this underrepresented area in the research.

The parents in Eiserman’s (2001) study described making personal changes to best parent a child with CL/P or as a result of the experience itself. Several parents described overcoming initial negative responses such as fearing limited social interactions for their child or a limited future or goals. Others described experiencing personal growth related to stress such as increased compassion for others less fortunate or different from oneself, relinquishing a sense of personal control, actively seeking social support, and strengthening family relationships through increased reliance on one another, trust, and commitment. For example, one mother noted although she never felt
inclined to be “very understanding, patient and helpful” to those in need, these were exactly the qualities she hoped to have as a parent to a child with CL/P (Eiserman, 2001, p. 239). How these parents developed these coping strategies was not discussed.

Social support has been noted to be an important factor in coping with chronic illness. Benson et al. (1991) found social support was lacking among parents of children with CL/P; they hypothesized this was the result of real or perceived social stigma or the inability to sustain adequate social contacts due to the considerable time cleft care-taking requires. Yet two studies found parents reported adequate social support (Baker et al., 2009; O’Hanlon et al., 2012). Parents reported seeking the support of other families who were cleft-affected in a number of studies but this resource has not been closely examined.

Despite the fact that CL/P involves a complex treatment process that lasts through late adolescence with the potential for social, cognitive, and emotional adjustment difficulties (many of which are more pronounced after early childhood), research has not examined the experiences of parents and parent/child interactions past infancy. Parent adjustment, unique parenting challenges, and effective parenting styles within this population all need further exploration.

**Resilience**

Research on psychological resilience began as an effort to understand how some children facing adverse circumstances thrived while others did not (Masten & Powell, 2003). No single definition of resilience exists but the common goal of such research is to understand the process of adaptation to inform intervention and prevention strategies (Herrman et al., 2011; Masten, 1994). Understanding both factors that promote and
protect healthy development is critical (Masten, 2011). Masten (2001) concluded resilience is not a matter of extraordinary individuals but is a “common phenomenon arising from ordinary human adaptive processes (p. 234). Arbona and Coleman (2008) noted this focus on strengths rather than deficits is “consistent with counseling psychology’s traditional philosophical stance and professional emphasis (p.483).

Resilience research no longer focuses solely on children but has expanded into many populations and age groups. Initially, resilience was conceptualized as a matter of individual traits such as autonomy and self-esteem (Luthar, Cicchetti, & Becker, 2000). Later, researchers recognized external factors such as the family and environment are critical to the development of resilience. Still later, the focus shifted to understanding how these factors together contributed to resilience--defined as “a dynamic process encompassing positive adaptation within the context of significant adversity” (Luthar et al., 2000, p. 543). Individual traits such as intellectual functioning, self-esteem, and self-efficacy contribute to resilience but these factors are themselves often the result of family and/or environmental influences.

Researchers noted when studying resilience, three factors needed to be assessed: the presence of risk, positive developmental outcomes (often termed competence), and protective and vulnerability factors (Arbona & Coleman, 2008). The nature and level of risk is a matter of some controversy. Some studies concluded only adaptation following significant risk, such as trauma or chronic environmental adversity, could be defined as resilience (Masten & Coatsworth, 1998). Others broadened the concept of risk to include any significant challenge that required learning and growth (Robertson & Cooper, 2013). Others examined the accumulation of normative daily stressors as a process equivalent to
more significant risk factors (Patterson & McCubbin, 1983). Patterson (2002) noted practitioners interested in developing prevention and intervention strategies utilized the process orientation.

Because positive outcomes or competence are not viewed as a static set of variables but changes across development, the best way to measure it is in relation to the specific risk experienced (Arbona & Coleman, 2008). While resilience is not a uniform concept and is dependent upon the type of risk and context, some themes were noted in the literature regarding protective and vulnerability factors. Children and adolescents who experienced at least one positive, warm, and supportive relationship with an adult were more likely to overcome adversity while those who were mistreated or abused were less likely to do so (Luthar, 2006). Adults were also positively influenced by supportive relationships during times of adversity (Luthar, 2006).

The inoculation model of resilience posited stressors are not simply something to be overcome but can encourage the development of new capabilities and strengths (Zimmerman & Arunkumar, 1994). This stress-related growth could include both personal and social development (Folkman, 1997). Commonly noted characteristics of this included “feeling strengthened, more resourceful, more confident and developmentally advanced” (Simon, Murphy, & Smith, 2005, p. 427). A study of cleft affected families found parents experienced increased levels of community involvement, spirituality, and enhanced family relationships as a result of their child’s condition. Adults with CL/P experienced enhanced communication skills, commitment to social justice, and diverse social support as outcomes of their condition (Eiserman, 2001).
Research on chronic health conditions also found similar stress-related growth including increased empathy and family cohesion (Patterson & Leonard, 1994).

Several authors noted the complexity of the resilience process is challenging to research due to the disagreement about what outcomes are considered to be successful, how contextual factors could be sufficiently accounted for, and how factors influenced one other over time or across development (Arbona & Coleman, 2008; Masten, 2001; Ungar, 2003). Concepts of resilience are relative and dependent upon temporal as well as cultural contexts (Ungar, 2003). Ungar (2003) noted qualitative research is well suited to explore resilience factors because of its focus on variability, “complexity in people’s experiences and relationships, with one another embedded in social and political contexts”, and because it allows for authentic descriptions of marginalized people (p. 92).

**Family Resilience**

Nichols (2013) noted early family resilience research considered the family as a support in an individual’s resilience, whereas some viewed the family as “the unit” of resilience itself (p. 3). Family resilience expands the idea of resilience from the individual to the family system and examines “family relational processes wherein risk and protective mechanisms develop and result in some level of adaptation of the family system” (Patterson, 2002, p. 233). Family resilience is also defined as the ability to return to the level of functioning prior to the onset of the adverse event(s; Hawley & DeHaan, 1996). Inherent in studies of family resilience is the focus of identifying and fostering factors that contribute to it as well as reducing contextual risk factors (Hanson, 2001).
An understanding of family resilience builds upon research on stress and coping at the systemic level. Stressors are events that place demands on the family system (McCubbin & McCubbin, 1993). Such stressors either weaken or strengthen the family, depending on the family’s coping strategies. One model that examines family resilience is the family adjustment and adaptation response (FAAR) model. This approach looks at adaptation as an active process of balancing family demands with family capabilities and the interaction of family meanings (Patterson & McCubbin, 1983). Patterson (2002) noted this entire process including the nature of the demands, capabilities, and meanings is dependent upon the context in which a particular process occurs. Stressors can be daily strains and disruptions as well as larger demands. Capabilities include both resources and coping behaviors (Patterson & McCubbin, 1983). In this model, repeated levels of crisis result from a continual state of demands outweighing capabilities. Family meanings include a family’s appraisal of the stressor, the capabilities for coping with it, as well as family identity. Patterson argued a family’s appraisal of demands and their capabilities for coping are the underlying processes involved in the creation of resilience.

The length of the stressor, the life stage in which it occurs, and the types of internal and external support available all influence family resilience (McCubbin & McCubbin, 1988). McCubbin and McCubbin (1988) noted the following factors contribute to family resilience: marital communication, satisfaction with the quality of life, financial stability, family time and routines, and family traditions. Black and Lobo (2008) found a positive outlook, spirituality, cohesion, communication, and family time are critical to resilience. Family cohesiveness and flexibility are also critical to resilience (Patterson, 2002). Despite the differences among family’s structure, rituals, and
practices, an emphasis on relationships, meeting individual’s needs, and the family as source of comfort and security are commonly found factors (Parke, 2000).

A study of resilience in parents with children with disabilities found protective factors included the support of family and friends, a strong marital relationship, and “educational, therapeutic and psychological support for family members” (Heiman, 2002, p. 159). Positive parental feelings toward their marriage, child, and the ability to cope also contributed to resilience.

**Resilience in the Cleft Affected Population**

Several authors have suggested a strengths-based framework is needed in CL/P research (Mouradian, 2001; Strauss, 2001). Understanding both the risk factors and challenges as well as factors in positive coping and adjustment are critical in forming “the basis of a theory of craniofacial health” (Strauss, 2001, p. 226). Eiserman (2001) made the following recommendations for a strengths-based approach to research with this population. First, he stated this topic is best studied with the use of emergent designs that allow for a broad range of unexpected findings not predetermined by the researchers. He also noted that participants should be part of the research process to further increase the likelihood their experiences would be accurately represented. He also noted the use of comparative norm groups was something that should only be used with caution.

Three studies have examined resilience in the CL/P population. Feragen et al. (2009) examined psychosocial resilience among children with CL/P by examining adaptation in the areas of self-perception and self-esteem. Results suggested the variables associated with resilience included the presence of depression and anxiety, satisfaction with appearance, and the subjective experience of being teased. Children
who were more resilient reported fewer perceived negative interactions with other children. The authors noted children’s perceptions did not relate to the visibility of the cleft but they also did not control for speech quality, which also plays a role in children’s self-perceptions and social interactions. Feragen et al. noted children’s subjective experiences were critical and advocated more research to understand what contributes to these including the impact of parenting practices. Baker et al. (2009) examined resilience in parents of children with CL/P and found social support and approach rather than avoidance coping contributed to successful adaptation. Tiemens, Nicholas, and Forrest (2013) examined the resilience factors in adolescent girls with CL/P and found positive peer support contributed to positive self-perception and resilience.

**The Parent/Child Relationship**

Parenting a child with a cleft condition creates stress for the whole family. It is recognized that CL/P impacts emotional, financial, and social resources and might involve readjustment in the family system (Baker et al., 2009). Yet, few studies have focused on the impact of the family as a whole or have explored the nature of parent and child interactions in this population (Crerand et al., 2015; Hunt et al., 2005). Despite indications that the quality of parent/child interactions and parenting styles could influence children’ adjustment processes, medical adherence, and health-related outcomes, few studies have examined this dynamic with this population. And despite social development being a concern for CL/P children, socializing experiences within the family have not been studied in detail (Krueckeberg et al., 1993).

Anecdotal evidence points to parent and child interactions that influence the experience of living with this condition. Because the diagnosis and early interventions
for this condition happen in infancy, children’s understanding of their condition is significantly influenced by communications with their parents (Hall et al., 2013). Positive and negative parenting practices also influence children’s experiences and adaptations. Discrepancies in parent/child reports point at times to areas of misunderstanding and miscommunication. Key differences between parent and child perspectives have been noted in areas such as satisfaction with treatment outcomes, psychosocial concerns, and identified areas of stress (Berger & Dalton, 2011; Damiano et al., 2007; Noar, 1991).

Discrepancies in parent and child understanding and communication are commonly noted in chronic health literature and are critical for developing a complex understanding of patient needs (Damiano et al., 2007; Snyder, Bilboul, & Pope, 2005). While one study noted parents and adolescents with cranio-facial anomalies (CFAs) showed lower discrepancies than was commonly found and noted this might be a sign of particularly close parent/child relationships in this population, discrepancies are still to be found (Snyder et al., 2005).

One survey study found parents’ and children's perspectives matched with regard to overall satisfaction with treatment and appearance but parents were more concerned about the impact on social and educational functioning than were their children (Noar, 1991). Hunt et al. (2007) found adolescents were more satisfied with their appearance than parents thought they were while Berger and Dalton (2009) found children were more satisfied with their appearance than were their parents.

In a German qualitative study (Sharif et al., 2013), mothers reported their children were not being bullied, whereas their children stated they were. Mothers reported a belief
that talking about bullying would make things worse, whereas children reported it to be helpful. In this same study, children reported being most concerned with social issues, while mothers were concerned with their children’s physical condition. Another correlational study found that while both parents and adolescents reported satisfaction with outcomes of treatment, adolescents were less likely to recommend the same treatment than were parents (Munz et al., 2011).

One study compared parents of preschoolers with a CFA to those without on measures of parenting stress, social support, and parenting style and on how these impacted the social skills of the children (Krueckeberg et al., 1993). This study utilized both standardized measurement tools and observations of children at play. Krueckeberg et al. (1993) hypothesized the parents of CFA children would have higher levels of stress and more restricted social support. The results did not demonstrate this, thus contradicting others studies of younger children. The authors suggested this was due to the use of non-disease specific measurement tools for parenting stress that might not be adequate to detect CFA specific stressors. However, they also noted the parents in this study were receiving regular emotional support from their medical team, which might have also made a positive difference.

How such differing perspectives are experienced in the lives of these individuals and their families, and whether and how they influence the treatment process is unclear. Research did note the importance of open communication regarding the condition, its origin, and treatment are critical to good decision-making and adaptation (Hall et al., 2013). Increased adjustment difficulties in the absence of such communication were also evident (Whitehead, Tobiasen, & Hiebert, 1996).
In one study, adolescents who were in counseling reported the issue of their condition was not discussed adequately in their family (Chapados, 2000). Chapados’ (2000) case study on a 13-year-old with pre-surgical anxiety depicted how greatly parenting practices could influence a child’s adaptation. In this case, the child’s mother never discussed the condition with her daughter and lied to others about the origin of her scar. She kept details of treatment and surgery from her child until the day they were to occur. As a result, the child refused surgery; when referred to a psychologist, it was disclosed she felt her condition was a punishment from God and that she, therefore, did not deserve treatment. Both of these cases were from clinical examples that might reflect more challenges to adjustment than the general CL/P population.

One study that hypothesized children’s social adjustment difficulties were related to negative and controlling parenting practices leading to a lack of autonomy in children with CL/P found quite the opposite was the case (Gassling et al., 2013). This study compared parents of children with CL/P to parents of children with chronic migraines and children without any medical condition. The method of assessment consisted of laboratory observations of an achievement oriented task. Mothers of CL/P children reinforced their children more often and children showed more autonomous behavior toward both parents than in both control groups Gassling et al., 2013). Children with CL/P asked for help less often and interrupted parents less than both control groups. The authors concluded this condition was “associated with a more cautious, patient and encouraging parenting style which promotes greater initiative” in the child (Gassling et al., 2013, p. 956). They also hypothesized the increase in supportive behavior might be intended to compensate for the emotional problems faced by these children. A quality of
life study comparing CFA adolescents to a norm group found they had higher scores on the family relationships, and lower scores on peer relationships (Topolski et al., 2005), which might be the result of compensating for poor relationships with peer groups.

A recent cross-sectional study of 1,200 school aged children (7-18) and their parents examined levels of family functioning, specifically cohesion, expressiveness, and conflict among participants (Crerand et al., 2015). Expressiveness and cohesion were associated with psychological adjustment and quality of life in children with medical conditions (Crerand et al., 2015). Participants’ scores were within the normal range, suggesting “most families are functioning despite their child’s condition” (Crerand et al., 2015, p. 656). Factors that negatively influenced family functioning included imminent surgical interventions, low family support, and single parent families (Crerand et al., 2015). The authors noted longitudinal data and qualitative studies that included youths’ perceptions of the family were needed to further understand family functioning (Crerand et al., 2015).

Causes of Psychosocial Risks in Cleft Lip/Palate Individuals

The chronicity of medical care as well as speech and facial differences put children at risk for psychological difficulties. Studies pertaining to physical, attractiveness, and stigma as well as chronic health conditions and their impact on family functioning are beneficial in explaining these challenges; they are now discussed.

Physical Attractiveness and Stigma

Concern for physical appearance and the recognition it informs how others view us are an inherent part of our culture (Rumsey, Clark, & White, 2003). Social psychologists have documented the benefits of physical attractiveness and the challenges
of unattractiveness in our culture. Those perceived to be attractive are both treated better and thought to be more intelligent (Dion, Berscheid, & Walster, 1972). They are also thought to be more dominant, better adjusted, and more socially competent (Eagley, Ashmore, Makhijani, & Longo, 1991). They are also more likely to receive more positive social reactions such as peer acceptance and popularity (Langlois et al., 2000).

Some research suggested cleft conditions more closely fit the concept of stigma due to disfigurement than facial unattractiveness. Unattractiveness refers to simply not sufficiently meeting societal norms for beauty, whereas disfigurement refers to the presence of medically caused deficits to facial or bodily structure and appearance. The latter are often viewed with a sense of ambivalence based upon both avoidance and sympathy and a desire to help (Katz, 1981). Concepts of attractiveness are subjective but are based in cultural norms as are concepts of what constitutes disfigurement (Thompson & Kent, 2001). Thompson and Kent (2001) noted three types of disfigurement: congenital malformations such as CL/P, traumatic events, and disease processes.

There is evidence that individuals with facial disfigurement face challenges in forming relationships, face discrimination, and have negative self-perceptions (Jowett & Ryan, 1985; Rumsey et al., 2003). Studies found self-concept is closely linked to appearance-- both in the general population and in those with facial disfigurement (Rumsey et al., 2003). Across conditions and specifically with CL/P, facial differences might lead to social anxiety, social avoidance, lowered self-esteem, negative self-image, and depression (Jowett & Ryan, 1985; Richman et al., 2012; Rumsey et al., 2003; Turner, Thomas, Dowell, Rumsey, & Sandy, 1997).
One study found one-third to one-half of outpatients with disfiguring conditions had elevated levels of anxiety, depression, social anxiety, or social avoidance compared to published norms (Rumsey, Clarke, & Musa, 2002). Yet research on individuals with port-wine stains, burns, and disfiguring physical conditions all demonstrated that many individuals with disfigurements show only minimal impact in emotional disturbance, again pointing to the possibility that individual factors are critical in successful adjustment (Ben-Tobin & Walker, 1995; Bowden, Feller, Thorlen, Davidson, & James, 1980; Kalick, Goldwyn, & Noe, 1981). Other research suggested individuals with disfiguring conditions utilized adaptive factors. Such individuals utilized negative social experiences to bolster their self-esteem through attributing all negative reactions to being associated with their disfigurement rather than other personal characteristics (Crocker & Major, 1989).

Severity of disfigurement is not a good predictor of psychological distress (Bradbury, 1996). The visibility of disfigurements has also been examined with mixed results. Some studies reported visibility was the best predictor of psychological distress, while others found the opposite (Thompson & Kent, 2001). Researchers hypothesized that people with very visible disfigurements cannot avoid contact with others and are forced to build coping skills while others with more hidden disfigurements can practice avoidance. Other studies on the impact of disfigurement utilizing factors such as gender, age, and type of disability also found such inconsistent results that generalizations were difficult (Rumsey et al., 2003).

Factors contributing to resilience in the face of disfigurement have not been adequately researched. Social support and family acceptance are critical to successful
adaptation across many disfiguring conditions (Thompson & Kent, 2001). Positive early experiences and ongoing support also helped develop positive self-concept as well as positive attributions in social situations (Thompson & Kent, 2001). Parental reactions are likely internalized by the child but little is known about parent and child interactions specifically with disfigured populations.

Social adjustment in the CL/P population is frequently linked to research on disfigurement or unattractiveness. Being stigmatized by peers or teachers, either by being bullied or treated differently, negatively influences the self-development of CL/P children (Masnari et al., 2013). Such negative social interactions can lead to feelings of diminished control for disfigured individuals (Rumsey et al., 2003). It can also lead to preoccupation with social encounters as well as increased anxiety (Rumsey et al., 2003). In a study of children's perceptions of their psychosocial functioning, the experience of being teased for having this condition was a stronger predictor of poor functioning than that of simply having the condition itself (Hunt, Burden, Hepper, Stevenson, & Johnston, 2006). In another study, results showed that while parents assessed their children as having social adjustment concerns, the children reported a normal self-concept. These authors suggest that through social avoidance these children maintained their self-concept (Kapp-Simon et al., 1992).

Studies specific to children's perceptions of the appearance of CL/P found children attributed less favorable characteristics to photos of cleft children than non-affected children and they perceived them to be less likely choices as friends (Tobiasen, 1987). A recent study of children's perceptions of other children with other facial differences (not CL/P) similarly found these differences exerted a negative social
perception (Masnari et al., 2013). Testing perceptions of attractiveness based upon photos versus real encounters has been criticized as being inadequate because factors such as social skills, mannerisms, and vocal attractiveness are as important as facial features in perception of attractiveness. Thus, research that excludes social interactions is limited (Asch, 2006).

Importantly, self-perceived attractiveness is correlated more highly with positively experiences than with attractiveness itself (Asch, 2006). Research suggested individuals with a variety of disabilities reported better quality of life than outside observers would expect (Basnett, 2001). Problems they noted typically emphasized “how individuals and institutions respond to their impairments, not to unhappiness with their physical state” (Asch, 2006, p. 235).

Some challenged the idea that surgical treatments for facial differences are useful in protecting individuals from stigma. Instead, they noted Western cultures typically approach birth defects through “the desire to provide treatment and remediation, also known as medical activism” (Strauss, 2001, p. 228). This approach has been criticized by bioethicists and pediatricians as being too based in cultural values of physical attractiveness (Paren, 2006). Thus, the very surgery to remove the different and unacceptable appearance legitimized or was complicit with the idea that atypical appearances were reasons to avoid or mistrust people (Asch, 2006). Thus, on some level, surgical repairs for purely aesthetic problems supported “the practice of isolating, rejecting or denigrating people with atypical bodies” and failed to “promote the idea that disability and physical difference is a legitimate form of human variation” (Asch, 2006, p. 236). Despite reported needs to complete surgeries in individuals with CL/Ps to assist
with functional deficits, research suggested no clear link between surgical treatment and better overall quality of life (Mouradian et al., 2006).

**Chronic Health Conditions and Disabilities**

Parenting a child with extensive medical needs brings unique challenges to the parenting process. Similar to parenting a child with any chronic health condition, providing for the medical needs of children with CL/P impacts families financially, socially, and personally on many levels (Kramer et al., 2007). Kapp-Simon et al. (1992) noted that because cleft lip and palate involves the same stressors as other chronic illnesses including repeated medical visits, multiple hospitalizations, and differences in physical appearance, it is reasonable to compare CL/P to the broader literature on chronic illness. Thus, because CL/P is a disability, this literature is also discussed.

Parental reactions to and adjustment toward chronic illnesses follows a similar pattern despite the specificity of the medical condition (Cousino & Hazen, 2013). Challenges faced by families of children with chronic health needs are referred to as parental stress. Any demand that creates systemic change is a stressor; how it is experienced relates to the severity and nature of the stressor and the resources of the family (McCubbin & McCubbin, 1993). Parents with a child with a chronic health condition experience more stress than families without such a condition; although for some conditions this stress might occur sporadically, for others, the levels remain constant (Knafl et al., 1999). Despite difficulties, research also found positive family outcomes such as enhanced personal and spiritual growth, sense of purpose, and improved relationships (Hastings, Allen, McDermott, & Still, 2002; Stainton & Besser, 1998).
Some researchers defined family stress as the result of a stressful event, while others noted stress is a process of family change (Malia, 2007). Most theories note this includes an attempt by the family to maintain equilibrium while managing some event or situation (Malia, 2007). Hill’s (1949, 1958) ABC-X model of family is still the most cited model with its emphasis on the combination of a stressor, the family’s resources, and their definition or perception of the stressor. Subsequent models emphasized that the nature of the crisis is not as distinct as this because change and adaptation are always occurring (McCubbin et al., 1983; Walker, 1985). More recent models focused on strengths and resiliency (McCubbin & McCubbin, 1993; Patterson, 1988). Patterson’s (1988) family adjustment and adaptation response model (FAAR) used concepts of risk and protective factors, family resiliency, and adaptation. McCubbin, Thompson, and McCubbin’s (1996) typology model of family adjustment and adaptation included the following dimensions to understand adaptation to stress: (a) family hardiness and family coherence, (b) family flexibility and family bonding, (c) family time and routines, and (d) valuing family time and routines.

All of these models noted appraisals of illness were important for the adjustment process of caregivers. Across medical conditions, research supported the role cognitive processes played in parenting stress. Negative appraisals such as perceiving a child to be vulnerable and feeling insufficient at providing care for the child are associated with increased parenting stress. However, a review study noted specific research on coping mechanisms across conditions was lacking (Cousino & Hazen, 2013).

Conflicting emotions are common in parents who both wish for their children to receive medical interventions and want to protect them from pain and distress (Domurat
Dreger, 2006). Parents of children with health conditions or any noticeable “difference” often experience concern and anxiety for their children’s social acceptance, possible stigmatizing experiences, as well as internal conflict due to experiencing their child as normal while at the same time understanding they are not viewed this way by the majority of society (Ablon, 1990).

The effect of parenting a child with a disability varies across families and across time. Similar to research on CL/P, past research has focused more on negative aspects while recent research includes positive impacts as well (Case-Smith, 2004). Parents have decreased time for personal needs and have to adjust expectations and redefine what is normal (Kratz, Uding, Trahms, Villareale, & Kieckhefer, 2009). As with chronic health conditions, parents face stressors such as decreased time available for work and increased medical costs, increased time spent on medical and therapeutic activities both in and out of the home, and a decrease in social activities (Turnbull & Turnbull, 1997). Decreased socialization impacts not only parents but children as well who thus learn social skills in a more limited way. Parents frequently report that support networks become strained, resulting in isolation (Boland & Sims, 1996). They also noted the loss of relationships due to lack of understanding of the condition and its impact and shifting priorities within the marriages (Kratz et al., 2009). A parent’s self-identity and emotional well-being are also impacted (Turnbull & Turnbull, 1997). One way in which this occurs is the illness or disability status becomes part of a parent’s identity (Patterson & Blum, 1996).

Research suggested parents go through a period of initial adjustment after which they focus on minimizing the disruptiveness of the condition in the life of the child and other family members. This process is called normalization and consists of managing
both medical needs as well as prior family activities (Deatrick, Knafl, & Murphy-Moore, 1999). Normalization is usually equated with successful coping but for some families, it is an ongoing process and family life never returns to prior levels of functioning. Research suggested some families manage chronic conditions with minimal impact on family life. Understanding the factors that contribute to successful coping is critical.

Adults with chronic illness have a higher incidence of psychiatric co-morbidity than healthy norm populations. The American Psychological Association's (APA) Division of Health Psychology (cited in Broder, 2001) noted the following concerns when assessing and working with people with chronic conditions: psychological distress, cognitive function, psychosocial adjustment and personality, well-being, family cohesion and coping. These findings echoed the literature on CL/P, which also noted higher rates of adult morbidity for psychiatric disorders. Both of these findings highlight the need to better understand the development and adjustment of individuals with CL/P.

Disease-related parenting stress is unrelated to disease duration and severity (Streisand, Swift, Wickmark, Chen, & Holmes, 2005). However, in addition to experiencing generally elevated levels of parenting stress, parents of children with chronic illnesses also experience disease-specific parenting stress. These might be undetected in studies that utilize generalized measures of parenting stress (Cousino & Hazen, 2013). As there is no current measure for the CL/P population, qualitative studies could assist in laying the groundwork for the development of such measures (Mouradian et al., 2006).
**Psychological Interventions**

Broder (2001) noted psychological assessment in CFA populations is critical, is recommended by the Application and Practice in Health Psychology Series from the APA’s division of health psychology, and should be part of the protocol for medical teams. Cranio-facial teams frequently include mental health professionals, although the specifics of the services these professionals provide have not been documented in any detail. The range and frequency of these services are variable with only 50% of teams even screening for psychological concerns (Hood, Cradock, & Vander Wal, 2011). Bennett and Stanton (1994) suggested a low priority of services for publically funded healthcare, the difficulties in providing psychological services in the hospital setting due to the diversity of clients, and the lack of research on best practices for this population contributed to this trend; when this article was written, the authors could not find a controlled study on efficacious psychotherapy for this population. In 2007, Collett and Speltz noted that without well-designed psychosocial intervention studies, little is known about whether mental health professionals could help this population. A literature review of psychological interventions for this population found only two studies that fit their inclusion criteria, only one of which utilized a randomized control design. Norman et al. (2015) concluded that due to research design and small sample size, the reviewed studies “did not provide sufficient data to draw any firm conclusions regarding the best forms of psychosocial interventions for either adults or children affected by CL/P” (p. 310).

Bennett and Stanton (1994) noted inconsistencies in the literature about the emotional impact of having a cleft condition and the lack of theoretical understanding about how problems developed and could be changed made psychological intervention
difficult. These authors questioned whether general theoretical understandings of development were adequate for treating this specific population. They noted that in the absence of specific guidelines, clinicians must utilize general psychotherapy skills on an individualized basis (Bennett & Stanton, 1994).

Both qualitative and quantitative findings pointed to the need for evaluative and supportive services for parents as part of the treatment process (Eiserman, 2001; Nelson, Caress et al., 2012). Nelson, Caress et al. (2012) noted research is needed in developing therapies to support families throughout the treatment process and advocated routine family assessments be built into the treatment teams to screen for families that require further services. Several authors noted that parents could use assistance in understanding how to build their child’s self-confidence, social skills, and manage negative or stigmatizing experiences (Eiserman, 2001; Nelson. Caress et al., 2012). Stock and Rumsey (2015) noted that given the presence of reported needs from parents for ongoing support and the range of these needs, medical teams need to structure the availability of support in a step-wise fashion.

Mouradian et al. (2006) recommended individual supportive counseling, peer-group support, and education as potential interventions for individuals with clefts. They noted the adaptation process might be better served by focusing on emotional and psychological health rather than repeated surgical intervention. Two intervention programs have been designed for individuals with CFAs, both of which focus on social skill development being implemented in school settings. One designed by Kapp-Simon et al. (1992) includes workbooks, support groups, and class activities to help young people with CFAs become proactive in handling social situations. The other is called
Changing Faces (Partridge, 1990) and consists of in-school education for classes and teachers.

Individuals and families who are impacted by the condition of cleft lip and palate face multiple challenges. There is an assumption in the literature that because of these challenges the psychosocial functioning of individuals and families is negatively impacted (Collett & Speltz, 2007). Although studies have examined the nature of some of these challenges, the results offered more inconsistencies than certainties at this point in time. One of the gaps in the literature is an understanding of the processes underlying adaptation including factors in resilience. It was also noted that the subjective experiences of individuals with cleft conditions, particularly children and adolescents, have been largely ignored. The current study investigated these areas through examining the experiences of adolescents and parents.
CHAPTER III

METHODS AND PROCEDURES

Introduction

This study utilized qualitative research methods to explore the phenomenon of parents’ and adolescents’ experiences with the condition of cleft lip and palate. This study was guided by the following research questions:

Q1 What is the essence of the experience of parents and adolescents with the process of CL/P treatment?

Q2 How do parents perceive that their children are impacted by CL/P treatment?

Q3 How do adolescents experience parental support in relationship to their medical treatment?

Q4 What are the coping strategies that parents and adolescents think that they have developed in response to CL/P and its treatment?

Qualitative research is particularly useful in examining the meaning people make of their experiences and in examining processes (Morrow, 2005). Qualitative research is frequently used in healthcare research as a stand-alone method or as a complement to quantitative methods because of its ability to explore a patient's meanings and values (Nelson, 2009). Because this study aimed to understand the subjective experiences of individuals with a condition that could have complex psychological, social, and cognitive implications, qualitative research was a fitting method for this study. The current study aimed to inform families as well as both medical and mental health professionals
regarding the essence of participants’ experiences with the impact of the medical
treatment and its impact on their emotional lives. In conditions such as CL/P that have
both functional and aesthetic implications, the need for incorporating patients’
perspectives is critical to providing the best care (Canady, 1995). The subjective view
from those who live with this condition is still a relatively new perspective in this
research and has much to offer in broadening the understanding of this condition (Nelson,
2009). In this chapter, I discuss the purpose of the study, outline the research questions
and theoretical framework, as well as the methodology and research methods used. The
researcher's role and the methods utilized to ensure trustworthiness of the study are also
outlined.

Pilot Study

A pilot study for this research was conducted in the spring of 2013. The purpose
of this pilot study was to clarify the research questions and the interview protocol for the
larger study. Two adoptive parents were interviewed in a series of four interviews about
their experiences with the medical aspects of parenting children with CL/P. The parents
were interviewed individually in an initial session consisting of open ended questions and
again individually in follow-up interviews that focused on clarifying and extending the
information gained from the first interviews. The results of this study convinced me of
the need for further exploration of this topic. The parents described emotional difficulties
connected to the ongoing medical treatment of their children, the lack of preparation and
sense of isolation they faced in parenting children with this condition, and the
implications for their relationship and for extended family dynamics. In addition, this
couple described a strong sense of purpose and belief in their ability to provide a healing
home for children who were both adopted and cleft affected. This study was limited by
the fact that the parents interviewed adopted their children from overseas; therefore, their
experiences with the medical care and care-giving process were complicated by the
additional stressors related to the adoption process and the language barriers between
them and their children. Self-blame and guilt for the cleft condition were also not present
due to the fact that they and their children were not biologically related.

**Methods**

Qualitative research begins with an understanding of the researcher's theoretical
framework and is also influenced by worldviews and beliefs about the topic and field of
study. A theoretical framework might be defined as a set of beliefs that guide action
(Morrow, 2007). It consists of a philosophical standpoint that informs and guides the
research process (Creswell, 2007). This includes the researcher's ontological,
epistemological, axiological, and methodological assumptions (Morrow, 2007).

Ontology is concerned with the nature of reality: qualitative research views reality as
subjective and multiple (Creswell, 2007). Epistemology addresses how reality is known,
which includes the relationship between the researcher and participants (Morrow, 2007).
In qualitative research, the researcher tries to lessen the distance between him/herself and
his/her subject to minimize objective separateness (Creswell, 2007). Axiology refers to
the role of the researcher's values in the research process (Creswell, 2007). Qualitative
researchers acknowledge “the very nature of the data we gather and the analytic
processes in which we engage are grounded in subjectivity” (Morrow, 2005, p. 254). The
research methods are derived from the theoretical framework. Therefore, the framework
greatly influences the results and in turn determines how a particular study should be assessed in terms of rigor and trustworthiness (Havercamp & Young, 2007).

The theoretical framework that informed this study and guided interpretation of the data was constructivism-interpretivism (Schwandt, 1994). This framework posits “all knowledge, and therefore all meaningful reality...is contingent upon human practices, being constructed in and out of interaction between human beings and their world” (Crotty, 1998, p. 42). This is a relativist viewpoint in which multiple realities exist and are equally valid (Schwandt, 2001). Therefore, reality is not objectively discovered by observing researchers nor is it entirely dependent upon the subjective understanding of researchers. Instead, this framework posits that reality is thought to be constructed within each individual as he/she interacts with his/her social environment. Personal experiences, perceptions, and the social and historical context in which one lives can all influence the construction of personal meaning (Ponterotto, 2005). Therefore, the study of relational processes between people and the impact of contextual factors in shaping human experience are frequently topics of study within this framework (Creswell, 2007).

Constructivism-interpretivism also acknowledges the researcher's subjective experiences, knowledge, and values influence the research process and positions the researcher as a co-constructor of meaning (Morrow, 2005). The social relationship between the researcher and the participants is critical to the development of the research findings (Havercamp & Young, 2007). Indeed, constructivism-interpretivism is particularly characterized by the centrality of this relationship and the co-construction of findings based upon dialogue (Ponterotto, 2005). Schwandt (2001) noted that in this approach, meaning is thought to be hidden and arrived at only through deep reflection.
The transactional nature of the dialogue between researcher and participants is central to this type of reflection (Ponterotto, 2005). This collaborative process allows the researcher to broaden rather than abandon his/her understanding through interaction; the goal of such research is not concerned with “accuracy but with the emergence of a new, dialectical understanding of the phenomenon in question” (Havercamp & Young, 2007, p. 278). In this study, I attempted to create a dynamic research process in which the participants and I co-created a deeper understanding of their essential experience of the CL/P treatment journey.

**Research Design**

**Methodology**

Phenomenology holds that psychological reality--its meanings and subjective processes--can be faithfully discovered. Psychological realities need not be construed; they have essential features that can be intuited and described. (Wertz, 2005, p. 175)

In this study, I utilized phenomenological methodology to collect and analyze the data. Moustakas (1994) wrote, “The challenge facing the human sciences researcher is to describe things in themselves, to permit what is before one to enter consciousness and be understood in its meaning and essences in the light of intuition and self-reflection” (p. 27). Here he describes the goal of phenomenology, which is to accurately describe participants’ experiences with some phenomenon, and he acknowledges the role of the researcher's subjectivity in the process. Phenomenology is both a conceptual framework and a methodology that originated with the work of mathematician Edmund Husserl; it has come to influence many social science disciplines. Thus, it offers conceptual and practical guidelines for utilizing subjective processes to arrive at a description of participants’ experiences.
With its focus on the study of consciousness, Husserl's work (cited in Wertz, 2005) included several key concepts critical to later methodologies such as those of Moustakas. A central concept and methodology within Husserl's work was the use of *epoche*, which can be described as an “abstention from influences that could short-circuit or bias description” of the object of study (Wertz, 2005, p. 168). This is also referred to as bracketing of researcher assumptions (Wertz, 2005, p. 168). Husserl noted the necessity for two types of epoche. The first is an epoche of the natural sciences, which aims to bracket “natural scientific theories, explanations, hypotheses, and conceptualizations of the subject matter” (Wertz, 2005, p. 168). The second is described as an epoche of “the natural attitude,” which aims to suspend “naive belief in the existence of what presents itself” in order to reflect more deeply upon subjective meanings (Wertz, 2005, p. 168). This second type of epoche consists of being aware of one's own experiences, perceptions, and values while attempting to “empathically enter and reflect on the lived world of other persons in order to apprehend the meanings of the world as they are given to the first-person point of view” (Wertz, 2005, p. 168). In the current study, I utilized both types of epoche to generate findings that reflected the subjective meaning of participants. In designing and carrying out interviews as well as when analyzing data, it was important to bracket my own personal experiences as the mother of a child with CL/P as well as the theoretical knowledge I have gained from the necessary review of the literature.

The process through which this reflection occurred is defined further by Husserl and Moustaskas (cited in Wertz, 2005). Two analytic processes occurred that resulted in a written narrative describing the textural (what) and the structural (how) descriptions of
the phenomenon under study. In the first case, a process termed transcendental phenomenological reduction assisted in creating a textural description of the phenomenon (Moustakas, 1994). Moustakas (1994) wrote that this process consists of considering each experience “in its singularity, in and for itself” (p. 34). In this way, the phenomenon is described completely, containing every important element that contributes to its experience. In the current study, the first goal of analysis was to respectfully reflect the particulars of individual experiences without comparison to larger themes or patterns in the data.

In the second case, a method of analysis known as imaginative variation assisted in developing the structural description of the phenomenon (Moustakas, 1994). The goal of this practice was to grasp the psychological essence of whatever was being studied through trying to imaginatively vary whatever phenomenon or theme was being considered to distinguish what was essential to it and what was not (Wertz, 2005). The goal of imaginative variations is to arrive at a narrative--termed a structural description “presenting a picture of the conditions that precipitate an experience and connect with it” (Moustakas, 1994, p. 35). In the final analysis, multiple perspectives are eventually reduced to a universal essence typical to all participants (Creswell, Hanson, Plano, & Morales, 2007). This universal essence consists of what individuals experienced as well as how they experienced it (Creswell et al., 2007). Therefore, in this study, I attempted to gather and represent the experiences of participants as well as contextual factors influencing this experience.

Intentionality is another key concept, which refers to the fact that human consciousness is always directed toward some object or experience. Experiences are
thought to be conscious ones derived from the combination of our subjectivity and its relationship to something that has meaning (Wertz, 2005). Moustakas (1994) wrote that intentionality is the “internal experience of being conscious of something; thus, the act of consciousness and the object of consciousness are intentionally related” (p. 28). This is line with constructionism's understanding that meaning is made up of subjective perceptions of objective events. Giorgi (1985) noted that one of the tasks of phenomenological work is to explore the intentional relationship between people and situations. In the current study, I was informed by the concept of intentionality in recognizing that my experiences of the phenomenon and the participants were likely to differ based upon our conscious awareness and subjectivity.

While there are several approaches to phenomenology, I utilized the phenomenological method described by Moustakas (1994). The aim of phenomenological studies is to describe the meaning of a phenomenon experienced by several individuals (Moustakas, 1994). His approach is characterized by description and the use of epoche (Creswell et al., 2007). While Moustakas acknowledged complete self-awareness and the ability to set aside all of one's assumptions and prior knowledge is an impossible task, it was nonetheless a goal of this methodology to apprehend the direct experiences of others rather than provide interpretation. As a researcher, I strove to maintain a self-reflexive practice throughout the data collection and analysis phases to ensure the results honored and represented the experiences of the participants.

**Participants and Setting**

All participants were selected through purposeful sampling, which is best suited to qualitative research's goal of finding participants “whose experience most fully and
authentically manifests or makes accessible what the researcher is interested in” (Wertz, 2005, p. 171). Phenomenological research requires all participants have an in-depth experience with the phenomenon being studied. A participant group was selected based on the following criteria: parent participants must be either a parent or primary caretaker of a child (or children) with a non-syndromal cleft lip and palate, who are at least 12 years of age, and have completed at least the initial surgical repairs for the condition (i.e., initial repair of the cleft lip and palate). Due to the variation of medical protocols used by treatment teams as well as factors such as failed surgeries, adoption, etc., the number and type of treatments completed were not specified in this study. Because the perspective of fathers was lacking in the literature, fathers were recruited for this study. However, only one father agreed to interview. Adolescent participants were born with non-syndromal CL/P, were at least 12 years of age, and had completed at least the initial surgical repairs prior to the interview. In addition, all participants were fluent in the English language. Participants were not excluded based upon race, ethnicity, socio-economic status, age, gender, sexual orientation, or status as biological or adoptive parent.

To recruit participants, I contacted four cleft-lip and palate medical teams in the Rocky Mountain region and one cleft lip and palate medical team in the northeastern region of the United States to ask for permission to distribute information about the study to patients and their families. Only the hospital in the Northeast was willing to distribute flyers but no participants came forward from this recruitment effort. In addition, I emailed a speech therapist in Colorado who specializes in cleft care and a reading specialist who passed on recruitment information to one school district in Colorado. I also contacted another school district who posted information for one school district in
Colorado. Again, no participants were gained through these recruitment sources. I also recruited on the internet through a variety of cleft lip and palate support groups, blogposts and organizations, and through a study Facebook page. Finally, recruitment was completed through a snowballing process in which participants recommended other participants for the study.

Participants were recruited through these online sources. Contact people sent out recruitment information and then provided me with names, phone numbers, and email addresses of individuals who expressed interest in the study. I reached out to these individuals to answer questions and provide further information. I then sent written consent forms to those willing to participate by email.

Sample size in qualitative research depends upon the purposes and goals of the inquiry and is judged to be sufficient when the results extensively reflect the experiences of participants in depth (Morrow, 2007). Creswell (1998) recommended the number of individuals being interviewed in a phenomenological study be between 5 and 25. Guest, Bunce, and Johnson (2006) found that for studies with a high level of homogeneity, “a sample of six interviews may be sufficient” (p. 78). Yet, Wertz (2005) noted that in a phenomenological study, the number of participants cannot be determined beforehand but depends instead upon the researcher attempting to reach theoretical saturation and redundancy. Theoretical saturation refers to the point at which the themes of analysis fully account for all the data (Morrow, 2007). Redundancy refers to the point at which new data do not offer further findings. The goal was to continue recruitment until saturation was reached. However, one finding of this study was the target population was difficult to access. This is discussed further in the recommendations for future research.
The participants were four parent/adolescent dyads--three individual parents and one individual adolescent.

All participants were interviewed by telephone due to geographical distance. While this method of interviewing did not allow for the same level of intimacy as face-to-face contact, research suggested telephone interviews also limit participant distress and are particularly useful in discussing sensitive topics (Mealer & Jones, 2014). Participants were informed their confidentiality could not be ensured with the use of technology in the same way as it would be in person.

**Role of the Researcher**

Creswell (2007) wrote that the researcher is the primary instrument of data collection, organization, and analysis. Therefore “qualitative researchers acknowledge both the existence of and benefits of subjectivity; yet they also attempt to explore or manage their biases through reflexivity” (Williams & Morrow, 2009, p. 579). In phenomenological work, the study begins with “a topic and question rooted in autobiographical meanings and values” (Moustakas, 1994, p. 103), which are then “set aside” or put in brackets (Moustakas, 1994, p. 85) in order to bring a fresh perspective to the topic of interest. In the current study, part of the reflective process included my own awareness of my interest in and relationship to this topic and how it might impact my role as a researcher.

My interest in this topic would likely not have arisen had it not been for my own experiences as a parent of a child born with a cleft lip and palate. My son's condition was diagnosed at a 20-week ultrasound; my own emotional and psychological reactions to this condition as well as extensive needs for medical services began at that time. In an effort
to find support, I sought out other parents through online groups and met many more during hospital stays and through referrals from my son's surgeon. In seeking out support, I also frequently sought information and understanding of my own experiences. The deficit-focused literature did not match my experiences of mothering or my observations of my child. Instead, I found the presence of a stigmatizing lens in some of the research that went unacknowledged. The emotional experiences beyond the early stages of diagnosis and feeding focused on depression and anxiety, whereas I experienced a more complex range of responses. With other parents of children with CL/P, I discussed both emotional experiences as well as practical details relating to care-taking pertinent to this condition. Through much of the early management of my child's condition, I sought information only as it related to my particular concerns. But at the same time as a trainee psychologist, I have, at times, watched my own emotional reactions with some sense of curiosity and have been drawn into the literature in the field with an eye toward recommendations for medical and mental health practice as well as personal interest.

In my reading of the relevant literature thus far, I have seen my experiences reflected often as well as finding perspectives very different from my own. However, I found few studies examined in sufficient depth the complex reactions and experiences of parents having a child with this condition. In talking with other parents, I also realized some similarities and some differences in our experiences, and I am aware that each family's experience is unique. I was intrigued by these different experiences and hoped to illuminate them further in this study. Because the “bracketing of presuppositions” is the hallmark of phenomenological research, it was important for me to maintain self-
awareness and utilize reflective practices throughout the study to, as Wertz (2005) wrote, maintain “an attitude of wonder that is highly empathic” and to describe the “meaning of the situation purely as it is given from the participant's experience” (p. 172)

**Data Collection**

Data collection for this study included transcribed interviews as well as researcher notes based upon observations during interviews. Participants were all interviewed individually by phone for 35 to 75 minutes. The interviews were semi-structured (see Appendices B and C for sample interview questions). Semi-structured interviews allowed me to respond to the emerging discussion and topics as they evolved (Merriam, 1998). Phenomenological interviews typically follow an “informal, interactive process and utilize open-ended comments and questions” (Moustakas, 1994, p.114). I audiotaped and transcribed the interviews verbatim.

**Procedures**

**Data Analysis**

Data were analyzed to answer the research questions by identifying themes relating to the emotional experiences of parents and adolescents with the medical process and the impact on parent and child relationships. Data analysis followed the phenomenological methods outlined by Moustakas (1994). The first step in analysis involved my attempt to use epoche and to bracket any prior personal or theoretical understanding of the topic in order to reflect fully on the data that had been collected. While Moustakas acknowledged it is an impossible goal to fully set aside subjective understanding, he encouraged the researcher's intentions to become more fully aware of thoughts and beliefs that might intrude upon perceptions of the data.
The second step in analysis involved a process termed horizontalization in which the transcripts were first read through in their entirety in an effort to recognize “every horizon or statement relevant to the topic and question as having equal value” (Moustakas, 1994, p.118). As I read the transcripts, I made notes of significant statements from each participant in turn. The third step involved organizing these statements into larger units of meaning or themes (Moustakas, 1994). In this step, I began to organize statements from the horizontalization process into themes for each participant. The fourth step involved reordering themes into a structural description of how the phenomenon was experienced by participants and a textural description of what was experienced in terms of the assignment of meaning by participants (Moustakas, 1994). At this stage when I had common themes, I utilized color coding to mark the transcripts according to each categorical theme. Lastly, these structural and textural descriptions were further combined into a common, typifying description known as the “essential invariant essence” of the experience (Creswell, 2007, p. 62). The final themes were organized after consultation with the peer reviewer who first came up with her own themes, which were then compared to mine.

**Ethical Considerations**

Because all qualitative research utilizes the subjective understanding of the researcher in its design, collection, and data analysis processes, special care must be taken to ensure that participants believe their viewpoints are respected and fully reflected in the final research results without any interpretive bias from the researcher. In this study, I attempted to be reflexive and aware of my biases throughout the research process. In constructing interview questions, I attempted to obtain the general descriptions of
experiences without leading the direction of responses based upon my values, experiences, or training. In each interview, I strove to approach the dialogue with curiosity, compassion, and interest, and to listen for the unique experiences of each participant. I attempted to check any biases that might have intruded upon the data without my awareness through dialogues with both participants, peer reviewers, and my research advisor.

Havercamp (2005) noted many ethical considerations in qualitative research are specific to its intensely relational nature. She pointed to one concern being “the asymmetrical power relationship (that) exists between researcher and participant, despite our best intentions to transform our participants into co-researchers” (Havercamp, 2005, p. 146). Because of this power differential, I considered it my responsibility to protect the interests of participants, both as a researcher and as a psychotherapist. In being mindful of this power differential and the ways it might impede the autonomy and well-being of the participants, I provided participants with information regarding the nature of the research, its goals, as well as my role as a researcher and the potential for risks and benefits to them. It was important to remember my role was that of a researcher and not a psychotherapist. Therefore, my goal was to gather information from participants rather than try to influence them in any way through the use of therapeutic procedures.

Another concern involved “a greater level of engagement offered the researcher is paralleled by a greater level of engagement for participants” (Havercamp, 2005, p. 147). The potential existed for dual relationships, emotional distress, and breaches in confidentiality to arise. No personal friends or acquaintances of mine were selected as participants. Due to the nature of the in-depth personal disclosures, participants were at
risk of vulnerability and emotional distress and it was my responsibility to protect them from harm. In this study, participants were informed of the potential for experiencing emotional distress due to the nature of the topic being discussed—both during the informed consent process as well as before each interview (see Appendix D for adult informed consent and Appendix E for adolescent assent form). Participants were provided with a list of mental health referrals should these be necessary and were free to withdraw from the study entirely or to request that any specific information be left out of the findings at any time. Participants were provided with the opportunity to review themes and the final written results to ensure they felt their views were being respected and accurately portrayed.

Participants were given pseudonyms and any identifying information was removed from the written results. Data were stored in a locked cabinet or locked electronic file and no one had access to this information other than my dissertation chair and peer reviewer.

**Rigor in Qualitative Research**

The manner in which qualitative research is judged to be rigorous and scholarly is different from the way in which quantitative research is judged. The term trustworthiness refers to the standards by which a study has been conducted and written and to its ability to accurately and convincingly describe the topic with sufficient depth. In this study, I was guided by Lincoln and Guba's (1985) categories of trustworthiness: credibility, transferability, dependability, and confirmability.

Credibility refers to whether the findings are believable when compared to the direct data as quoted in the study as well as to whether they are useful to and worthwhile
for the field of study (Lincoln & Guba, 1985; Schwandt, 2001). Researcher reflexivity is critical to ensuring credibility. Reflexivity refers to the practice of becoming self-aware of biases, experiences, and values that might influence the research process and making these explicit to the reader (Creswell, 2007). I kept a reflective journal throughout the research process to record my experiences, reactions, and awareness of biases that might influence the research. Intentional bracketing of a researcher's biases is critical to the phenomenological method and also ensures credibility through helping researchers stay aware of their own and participant's experiences (Williams & Morrow, 2009). Bracketing of my views and biases was practiced both prior to each interview and during data analysis.

Credibility was also supported by the use of expert, peer, and member checking. Dr. Basilia Softas-Nall, my research advisor, has expertise in qualitative research and methodology; a graduate of the counseling psychology program with training in qualitative methods was my peer advisor. These advisors were utilized throughout the analysis process to discuss concerns with the ongoing research as well as themes and analysis. Peer checking involved reading of transcripts, identifying significant information through horizontalization, coding of themes, and comparing this coding to my own code themes. Member checking involved making sure my interpretations honored the meaning as perceived by participants (Lincoln & Guba, 1985). Participant feedback was elicited to check for accuracy and faithfulness of the themes. Member checking also augments the researcher-participant relationship, increases trust and collaboration, which in turn increases trustworthiness (Williams & Morrow, 2009).
Triangulation was utilized to insure credibility of the present study. Triangulation refers to the use of multiple sources of data, methods, investigators, or theories to ensure adequate and sufficient data (Merriam, 1998). My goal was to increase triangulation through interviewing both fathers and mothers and through interviewing pairs of parents/adolescents to reveal different perspectives of the experience of CL/P. However, gaining interviews from different perspectives within the same family was not always possible due to recruitment difficulties.

Transferability refers to whether the data were presented in enough detail so the reader could generalize them to similar situations or research findings. Transferability is judged by the reader and dependent upon the use of direct quotations and the use of “thick and rich descriptions” in which the “writer describes in detail the participants or setting under study” (Creswell, 2007, p. 209). In this study, I conducted in-depth interviews with each participant with the goal of understanding in sufficient depth the experiences they disclosed. The use of extensive participant quotes and detailed contextual description was also utilized to increase transferability. Accuracy in the portrayal of participant's experiences was verified through member checks following transcription of interviews and during theme development.

Dependability refers to whether the research design and process are traceable and logical (Lincoln & Guba, 1985). Dependability can be augmented by providing a clearly articulated theoretical framework, methodology, and methods section as well as by documenting any changes that emerged in the research design and the decision-making process that led to these changes. An audit trail consists of a chronological list of all research procedures and methods used, thereby providing documentation of the entire
research design and process (Creswell, 2007). In this study, the audit trail consisted of a chronological and detailed list of all research activities as well as blank copies of documents used such as consent forms, interview questions, and any referrals given to participants. A reflexive journal provided further information on decisions made throughout the research process.

Confirmability refers to whether the written findings make sense to the reader when compared to the data that were collected. The use of an audit trail, member and peer checks, and thick and rich descriptions all increased confirmability (Creswell, 2007).

Summary

In this chapter, I outlined the methodology for the current study. I discussed the pilot study, the theoretical framework, methodology, and research methods used in the study. I also included the researcher's stance, ethical considerations, and a discussion of the trustworthiness of the study.
CHAPTER IV

RESULTS

Introduction

In this chapter, participants’ experiences of having the condition of cleft lip and/or palate and the medical treatment process for this condition are described and analyzed. The current study explored both the experiences of adolescents born with the condition of cleft lip and/or palate (CL/P) and parents of adolescents. Specifically, this study described participants’ experiences with the medical treatment for CL/P and its impact on their emotional lives. This study was guided by the theoretical framework of constructivism-interpretivism (Schwandt, 1994), which posits that reality is constructed within each individual through the interaction of personal experiences, perceptions, and social and historical contexts (Ponterotto, 2005). Through semi-structured interviews, this study explored the meaning the participants had created regarding their experiences with this condition and specifically how they coped and adapted to these experiences.

The methodology informing this study was phenomenology, which aims to describe the meaning of a phenomenon as experienced by several individuals (Moustakas, 1994). This chapter describes each participant’s experiences with the condition of CL/P. According to Moustakas’ (1994) phenomenological methods, the first goal of data analysis is to reflect the particulars of an individual's experiences without comparison to larger themes or patterns in the data. This chapter provides a textural-
structural description for each individual participant. Individual participants included four parent and adolescent pairs, three individual parents, and one individual adolescent.

In this study, participants’ experiences were gathered from in-depth interviews by telephone due to the geographical distance between the researcher and the participants. Direct quotes from participants are included throughout this chapter as part of the thick description and to increase trustworthiness.

The following questions were addressed and answered in the following section:

Q1 What is the essence of the experience of parents and adolescents with the process of CL/P treatment?

Q2 How do parents perceive that their children are impacted by CL/P treatment?

Q3 How do adolescents experience parental support in relationship to their medical treatment?

Q4 What are the coping strategies that parents and adolescents think that they have developed in response to CL/P and its treatment?

Four parent and adolescent dyads were interviewed for this study--two mothers, one father, and one adolescent—as well as interviewed individually. The participants came from different areas of the United States and all met the criteria for participation in this study: the adolescent was born with a cleft lip and/or palate, had completed the initial surgeries for this condition, and was between the ages of 12-18; and the parents were either biological or adoptive parents. All participants spoke English. The following section includes the description and the background information of each family (or individual) such as family structure, when the condition was diagnosed, the nature of the treatment team, and any significant experiences that occurred in connection to living with the cleft condition. All participants were given pseudonyms that were used in the
descriptions to tell the story of the family (or individual) and describe the phenomenon of living with and coping with the medical treatment for the condition of cleft lip and/or palate before introducing themes that emerged from all participants (see Table 1).

Table 1

*Family Participants*

<table>
<thead>
<tr>
<th>Family</th>
<th>Participant</th>
<th>Role</th>
<th>Age (of child)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Smith Family</td>
<td>Paula</td>
<td>Mom</td>
<td>15</td>
</tr>
<tr>
<td></td>
<td>Kristin</td>
<td>Daughter of Paula</td>
<td></td>
</tr>
<tr>
<td>Bennett Family</td>
<td>Lisa</td>
<td>Mom</td>
<td>14</td>
</tr>
<tr>
<td></td>
<td>Anne</td>
<td>Daughter of Lisa</td>
<td></td>
</tr>
<tr>
<td>Nelson Family</td>
<td>Francine</td>
<td>Mom</td>
<td>17</td>
</tr>
<tr>
<td></td>
<td>Jennifer</td>
<td>Daughter of Francine</td>
<td></td>
</tr>
<tr>
<td>Potter Family</td>
<td>Jan</td>
<td>Mom</td>
<td>18</td>
</tr>
<tr>
<td></td>
<td>Lily</td>
<td>Daughter</td>
<td></td>
</tr>
<tr>
<td>Grant Family</td>
<td>Jessie</td>
<td>Mom</td>
<td>(Daughter 18)</td>
</tr>
<tr>
<td>Johnson Family</td>
<td>Margaret</td>
<td>Daughter</td>
<td>15</td>
</tr>
<tr>
<td>Jacobs Family</td>
<td>Janet</td>
<td>Mom</td>
<td>(Son 16)</td>
</tr>
<tr>
<td>Baxter Family</td>
<td>Jeff</td>
<td>Dad</td>
<td>(Son 17)</td>
</tr>
</tbody>
</table>

*Family Participants*

**The Smith Family**

The Smith family lives in a small town in the Pacific Northwest of the United States within easy access of a major metropolitan area. The family consists of mom
Paula, husband Joe, and five children. Paula is Caucasian and Joe is Native American. Kristin, age 14, is the youngest of the children and the only child born with a cleft lip and palate. Kristin’s condition was diagnosed prenatally and was a relief according to Paula compared to the original diagnosis of Trisomy 18, which is a fatal condition: “Clefting runs in my husband’s family and there is a lot...for us the cleft was like, whatever.” Kristin’s paternal aunt as well as seven of her cousins were born with cleft conditions. Despite this family experience with the condition, Paula did not reach out to her in-laws for support or information, stating they never talked about it: “They pretend it’s not there.” Although Joe’s family provided assistance and support to the family and to Kristin, they did not visit her in the hospital or until she had completed her initial surgeries.

Paula had previous experience working with children with cleft conditions as a dental assistant; through this, she had “an idea of what to expect.” At Kristin’s birth, Paula said she and her husband were not bothered by the condition; rather, they were “kind of fascinated, ‘O look, that’s what it looks like before it’s been fixed.’” Caring for an infant with special needs, however, with four other children was challenging and exhausting for Paula.

The family initially utilized a medical team recommended by their insurance company but the family had a bad experience with them. Kristin’s surgeon did not believe in pain medication for infants so following a surgery, “my daughter was screaming post-operatively and they had to call in a pediatrician that was on call at the hospital to give her pain medication. It was horrid, utterly horrid.” The family then transferred her care to a local Shriner’s hospital where they have remained ever since.
Paula’s brother’s father-in-law was a Shriner and sponsored them to receive care in the hospital.

Paula is a stay-at-home mother and former dental assistant and her husband works in construction. Paula stated her husband is “super supportive” and she believes it is her role to be the “foundation of the family.” Paula has also found a support group online of other parents with children born with cleft lip and palate. Paula and Joe and Paula’s family try not to treat Kristin as if she’s special or different; they treat the cleft condition as a “non-issue.”

Kristin has had a total of eight surgeries so far, several ear tube placements, and extensive orthodontic work. Paula noted her surgeon assessed Kristin as one of the 10% of “super hard” cases of cleft lip and palate as several of her surgeries have failed and had to be revised.

The Bennett Family

The Bennett family lives in a metropolitan area in the Midwest of the United States. The family consists of Lisa, her biological daughter Anne--aged 14, Lisa’s husband Bill, his biological daughter, and the couple’s son. The family’s ethnicity is Caucasian. Anne’s biological father chose to leave Lisa and Anne when the cleft condition was diagnosed at birth. In every way, Anne’s birth challenged her mother’s expectations: “nothing went as planned.” The relationship ended suddenly although the pregnancy had been planned after four years of commitment. The condition was a shock as the couple had multiple ultrasounds, one a 3-D, and none detected it. Lisa had a scheduled induction but due to fetal distress, an emergency cesarean-section was performed. Shortly after the birth of her daughter, Lisa had to face raising her child alone
as well as an unpredicted medical condition. She expressed being in shock and having
difficulty comprehending what the delivery staff told her: “I think my whole life was a
whirlwind right then…I didn’t really process what was said to me.”

Anne’s condition was described by her mom: “on a scale of 1-10 severity wise,
she had like a 9. She had a bilateral lip and palate. It was very severe.” Lisa herself was
adopted as a toddler but knew her biological mother was also affected by a cleft
condition:

My biological mom had a cleft lip; she had just a unilateral lip and her palate was
not affected. She had four children and none of us were cleft affected. My older
brother has six kids and none of them had it, my older sister had three, none of
them were affected, and then all of a sudden boom out of nowhere I got the most
severe of the severe.

Lisa recalls being overwhelmed with concerns about how to parent and how to
manage being a single parent immediately after Anne’s birth. Of the time in the hospital,
she recalls:

Yeah, I remember being bombarded with medical information and because I
didn’t know ahead of time I was in complete shock. I didn’t know how to
function. I didn’t know how we were going to survive from minute to minute and
they were pouring information into me, and none of it registered. The only thing
that registered with me and I so want to go find this nurse even today, was when I
was rocking her in ICU, and a nurse came up to me and knelt down beside me and
out of her one side of her white coat pocket she pulled out a picture of a cleft
baby, who was a little a boy and out of the other pocket she had a picture when he
was seven or nine after he had surgeries and that spoke volumes to me. It just let
me know that things were going to be okay, and that’s the only thing that
registered with me in the hospital.

Lisa returned home to live with her adoptive parents who were very supportive
and helped tremendously in the early phase of Anne’s life. Lisa stated the challenges of
caring for and feeding an infant with a cleft condition were “overwhelming, like 45
minute feedings. It was a lot you know…it was very overwhelming for a first time young mother.”

Lisa could not recall any emotional support being offered from her daughter’s medical team; instead, she felt lonely and isolated in parenting a special needs child. She recalled a story from her daughter’s early infancy that highlighted this experience:

When Anne was brand new, her cleft was completely open because it was so bilateral and she had like the ball of tissues outside of her mouth that should have been gum tissue for her upper teeth. When you looked inside her mouth, her palate was gone, it looked hollow and empty all the way up to what I thought was the base of her eyeballs. It was so hollow and empty there was nothing. I just remember when she was brand new going to her plastic surgeon and I just wanted to know can she smell? When I sit and rock her, does she know what her mother smells like? Is she going to be able to smell flowers? Is she going to be able to smell things in the world? “In your opinion do you think that she can she smell?” His answer was “Well how should I know that.” That was a really big deal. I needed to know that she knew what I smelled like. It seems dumb, but it was a big deal. I was so angry and so hurt and so mad.

Because of her own perceived need for more support, she became involved in social advocacy for other parents of children born with cleft conditions. With other parents, she co-founded an organization that visits parents of cleft affected children in the hospital after birth, providing information and support--something she felt was lacking from her experience:

I remember I thought I don’t ever, ever want anybody to feel the way I did again, and so I wanted to get together with other parents and be resources for people in the hospital so that we could be that person who would be there to say it’s going to be ok. We made up pamphlets and before and after pictures and all our contact information and gave it to the hospital.

Lisa chose her daughter’s medical team through the advice of her father who is a primary care physician and her sister who is a surgical technologist. She felt it vital to be involved with a cranio-facial team and trusted medical decision-making to their expertise.
Despite feeling a lack of emotional support from her team, she described feelings of gratitude for their expertise and the help they provided her daughter.

Lisa described the changes that took place in terms of parenting a child at different stages with regard to treatment. When her daughter was an infant and unaware, she did not experience distress prior to surgery but Lisa felt “I had that anxiety build up.” Later, her daughter’s awareness and understanding caused her some anxiety, which in turn impacted Lisa.

Lisa and Bill met when her daughter Anne was three and he later adopted her. Both parents work for the police force. Lisa noted there have been challenges involved in co-parenting in a blended family and Anne’s history and condition played a role in this. Lisa and her daughter were very close, having spent the first three years of Anne’s life alone together. She stated, “We went through a lot together. I was a young mother and that was stressful and I was alone and that was stressful and we had to survive it and get through it together.” She is protective of her daughter in relation to her condition:

I was a young mother and that was stressful and I was alone and that was stressful and we had to survive it and get through it together. My husband coming into the picture when we started dating was difficult because I was like mama with bear claws out all the time –“Don’t tell me how to parent my child, because you haven’t been here- you’ve no idea what we’ve gone through, and you have no right to suggest anything to me- we’ve gone through a lot together, we’re tight and I know how to raise my kid” and so it’s been difficult.

Lisa noted her husband and Anne still have a strained relationship and she believes it causes her daughter significant stress.

Anne was diagnosed with Perthes disease at the age of four. Perthes disease is a rare orthopedic illness that can require several years of medical treatment. At the time, Lisa said it felt “traumatic and tragic. I thought here we survived all the cleft stuff and
surgeries and now she has a disease.” This condition increased Lisa’s concern that Anne would experience social alienation as in addition to her lip scar, she entered kindergarten in a wheelchair and leg braces. In Anne’s case, medical intervention was successful in resolving this condition and education and advocacy in the classroom helped Anne’s social acceptance.

Anne has also been diagnosed with attention deficit hyperactivity disorder (ADHD) and has an individualized education plan (IEP) in school for this condition. Lisa advocated for her daughter in the school district and even changed schools to receive adequate accommodations. Since receiving additional services, Anne’s school performance has improved. Anne also experiences some anxiety with regard to academic performance and depression in connection to a bullying incident. She is on anti-depressants prescribed by her family doctor.

Lisa and family are Christian and their church community is an important source of support. Lisa also stated that both her and her spouse’s families are supportive. At the time of the interview, Anne had had six surgeries.

The Grant Family

The Grant family lives in a small city in the southern United States. The family consists of mom Jessie, dad Steve, two biological children-- Cammy (18) and Ian (16), both of whom were born with cleft conditions, and an adopted child Dan (13), also born with a cleft condition. The family’s ethnicity is Caucasian. Cammy was born with a unilateral cleft lip and palate. Ian was born with a bilateral cleft lip and palate. Dan was born with a unilateral cleft lip only. The couple did not know of any prior family history of cleft conditions before the birth of their children. Cammy’s condition was not known
until birth. Although unexpected, Jessie said, “I don’t remember a time that we were upset about it. We’re kinda like okay this is it, let’s deal with it.” Ian’s condition was detected prenatally, which Jessie said was “the hardest part for me.” With a second child impacted by a cleft condition, she blamed herself: “How could I do this to another child?”

Their last child was adopted at the age of one month after he was abandoned by his mother shortly after birth. His mother had been incarcerated and was allowed to leave the hospital for the birth. While his cleft condition was less significant than his siblings, he has struggled in other ways: being diagnosed with Tourette’s syndrome, obsessive compulsive disorder (OCD), and ADHD.

Jessie is a former Latin teacher and her husband works in banking. Jessie has not worked since Ian’s birth due to caretaking needs of her children. This has been a financial strain on the family at times. The children completed the initial surgeries at a children’s hospital in a metropolitan community in the southern United States. The children’s hospital was located across the street from the hospital in which Jessie gave birth to her oldest and the cranio-facial team was on site in the hospital the day after Cammy’s birth. The family recently moved to a smaller city in a different state and Jessie has arranged for several providers to work in consultation with one another to provide care to her kids in the absence of a large cranio-facial team in the area.

Jessie stated she had no knowledge of cleft lip and palate conditions until her daughter was born. Jessie mentioned that information was less available prior to widespread use of the internet and national advocacy groups. The information was also less positive than it has become. Jessie also noted incredible changes in technology that positively impacted the treatment process for cleft conditions.
While public awareness has improved during her children’s lifetime, Jessie finds that many people are still unaware of it. Even insurance companies have been unaware of the necessity of procedures for her children and have questioned her choices. She and her family initially became involved with fundraisers for the cranio-facial team when Cammy was a young child; this experience led them to participate actively in other fund-raising and awareness raising events in their community. Jessie wrote a children’s book with her children about cleft lip and palate, which has sold copies worldwide. She initially wrote this at the request of Ian who wanted her to explain to his classmates in kindergarten about his condition.

Cammy has had six surgeries to date, Ian has had seven, and Dan has had two. Ian was placed in intensive care for 13 days after his birth because the staff did not read his parent’s birth plan and attempted to feed him with a non-cleft specialized bottle, which led to aspiration. During this time, she could not hold her infant, which was more upsetting to her than his condition. Ian also had to have hearing aids for a period of time in elementary school due to a complication of his cleft condition, which resolved itself with time. Jessie signed waivers with the hospital when her children were all infants and toddlers so they could have a bed rather than a crib, which enabled her to hold them after surgeries.

The Johnson Family

The Johnson family lives in a large metropolitan community in a Gulf Coast state. The family consists of mom Jennifer, dad Andrew, a step mom, and two daughters, one of whom is cleft affected. The family’s ethnicity is Caucasian. Margaret, the cleft affected daughter, was 15 at the time of the interview. All of her medical care has taken place at
the only children’s hospital with a cranio-facial team in her city. She was uncertain about
the number and extent of her surgeries thus far. Margaret lives with her mother and sister
and has frequent contact with her father and stepmother. Margaret’s parents have
encouraged her to make decisions about her treatment including asking the team
questions about her concerns and goals. Margaret and her older sister have founded a
nonprofit that raises money for the local cranio-facial team in order to provide resources
for families who travel from out of state to pay for travel and lodging expenses. Margaret
attends a local high school where she does very well academically and has many friends.

The Jacobs Family

The Jacobs family lives in a metropolitan area in the Midwest of the United
States. The family consists of mom Janet, dad George, and two sons--Frank (16) and
Stan (18). The family’s ethnicity is Caucasian. Frank was born with a bilateral cleft lip
and palate. There is no known family history of cleft conditions; Janet stated she felt
some guilt about his condition because she was using alcohol and cigarettes prior to
knowing she was pregnant. Frank receives medical care at a Shriner’s hospital in a
metropolitan city in a bordering state, requiring them to drive about four hours for
medical care and visits. Shriner’s hospitals provide all medical care free of charge.
Frank’s father’s family have been very involved in the Shriner’s association for
generations; Janet stated that because of their involvement and knowledge of the hospital,
they never even considered going anywhere else. She and her husband have also been
involved in fund raising activities for the hospital.

At 18-months-old, Frank’s throat swelled around a breathing tube put in place
during a surgery, which obstructed its removal. At this time, they also learned he had a
negative reaction to morphine. It took 24 hours for this situation to be managed, a time Janet called “the scariest nightmare God ever put on earth.” Because of this surgery, Frank lost considerable weight and had to send time in the intensive care unit. While no other emergency complications have occurred in his treatment course, he has not been a “typical case.” Frank has experienced many of the rarer side effects and had procedures not work as well as expected. Janet expressed concern over the overall quantity of invasive procedures he has had, mainly anesthesia and x-rays. Frank has struggled with sleep apnea, a fairly common side effect of cleft conditions, and has had surgical interventions for it since early elementary school. Janet has encouraged Frank to be involved in medical decision-making from a fairly young age, although it was made clear declining needed treatment was not an option.

Janet is a former preschool teacher who has worked part time or been a stay at home mom since Frank’s birth. Frank’s treatments and the travel distance to his medical team has made returning to work impossible. Janet reported Frank’s medical needs have had a negative impact on her older son. As a child, Janet stated Stan acted out in response to the time she necessarily spent both taking Frank to treatment and taking care of him. As an older child, Stan has been able to verbalize his belief that she loves Frank more than him and still has difficulty with the time involved in Frank’s care. Despite his jealousy, Stan is also very protective of his brother with other children.

Janet and her family have had the support of her family, her husband’s family, neighbors, and parents of Frank’s friends. Janet became heavily involved in one of the major cleft lip and palate advocacy groups and both provided and found support through this organization. She has also served as a parent-consultant to several cranio-facial
teams. She ceased to be as involved as Frank got older and the treatment needs decreased. He has had about 15 surgeries to date.

The Baxter Family

The Baxter family lives in a large metropolitan area in the Midwestern region of the United States. The family consists of mom Samantha and dad Jeff, who were recently separated, and a daughter and a son. The family’s ethnicity is Caucasian. Both children currently live with their dad. The couple’s son, Aidan, was born with a bilateral cleft lip and palate. He was diagnosed prenatally and the couple researched several options for surgical care prior to his birth. Samantha found it very distressing that he would not be able to nurse; the couple considered a clinic in another state that provided very early repairs, which might enable nursing to occur. They decided against this option due to the distance from the hospital during recoveries. Her inability to nurse Aidan was something her husband stated she “never fully got over.”

Jeff stated Aidan experienced complications and setbacks and was frequently required to repeat surgeries as a result. Jeff stated, “We had really bad luck with surgeries.” One example was the surgery Aidan had to improve his speech, which resulted in the creation of sleep apnea, a condition that has never been well managed for him. Jeff stated this constant need for repeated surgical interventions was exhausting for the whole family.

For the past four years, Aidan has experienced behavioral difficulties including opposition toward his parents, violence toward them, and depression and anxiety symptoms. His behavioral challenges have led to his refusal to continue care for his cleft condition including attending to orthodontic work, utilizing his C-pap machine for sleep
apnea, and completing a recommended jaw surgery. He has also dropped out of high school. Aidan had an IEP in school for speech originally and later for general educational needs. His aggressive behavior toward his mother was the reason she sought a place to stay outside of the home, which eventually led to a separation in the marriage. At this point in time, she has no contact with Aidan who refuses to engage with her. Aidan has seen many mental health professionals including individual therapists, family therapists, a psychiatrist, and an inpatient behavioral health team. He has been diagnosed with depression and anxiety disorders, ADHD, and pathological demand avoidance, which is an autism spectrum disorder recognized in Europe.

Jeff stated a belief that Aidan’s mental health problems are not related to his cleft lip and palate. However, he also acknowledged that Aidan’s personality and behavior changed considerably following being bullied in middle school. Jeff stated after Aidan was bullied, Aidan “lost his smile.” At the time this occurred, his parents were unaware of it and never knew the extent or exact nature of this experience. While Jeff and his wife have worked together to find mental health support and resources for Aidan until recently, at this point, he stated he was parenting on his own. He discussed making ongoing attempts to get Aidan the help he needs despite a considerable financial burden.

The Nelson Family

The Nelson family lives in a small city in the Midwestern region of the United States. The family consists of mom Francine, dad Joe, and four adopted children ages 18, 17, 15, and 13. The family’s ethnicity is Caucasian. The two middle children were both born with cleft conditions. The oldest is Jennifer and the middle child is John. John has also been diagnosed with autism. All four children were adopted internationally. When
Jennifer was adopted at 20 months, she had not had any treatment for her cleft lip and palate. When the Nelsons adopted their two middle children, it was extremely common to find children born with cleft conditions in their country of origin. Jennifer was lucky as she was loved and fairly well cared for in her orphanage including being fed in a manner in which she could manage to eat despite her unrepaired cleft palate. John was less lucky; he was physically restrained while formula was poured down his throat.

Francine believes this mistreatment was the cause of his autism. The children receive cleft care at a local teaching hospital and have had two different surgeons since beginning their treatment. The parents have left most of the decision-making to the cranio-facial team, although they tailor the timing of surgeries to fit the family’s schedule. Francine stated as parents they were naïve about how extensive the medical treatment for this condition would be. Francine has created her own support network of parents of children with cleft conditions, not having had a former support network. Francine currently is a stay at home mother and her husband works for the U.S. government. Both parents formerly worked in information technology.

**The Potter Family**

The Potters live in a rural part of the Pacific Northwest of the United States. The family consists of mom Jan, Dad Dave, four adult biological children from the couple’s previous marriages, and 21 internationally adopted children (seven are in the home currently). The parents’ ethnicity is Caucasian. The couple have raised children in three successive groups. Several of their adopted children were adopted in sibling groups--all were special needs and most were adopted as older children. Jan stated they “did not set out to adopt this many kids” but kept encountering more children who needed their help:
“It’s been one heck of a ride that’s for sure.” Several of the children they adopted would have died without urgent medical care and none of them would have experienced stability without long-term medical treatment. While it has been challenging, Jan says what keeps her going is the knowledge of how much they have changed these children’s lives for the better. Jan stated not all of the adoption process has been positive:

It hasn’t been easy and some of the kids have not been wonderful. Some have had a lot of social issues that we didn’t realize…so we learned along the way that sometimes love doesn’t heal everything. We had a lofty idea that love heals everything. No, it does not.

The family’s situation was written about in local newspapers; after this, an orthodontist contacted them and offered free services for their children. Not all responses have been so positive, however. When the family lived in a small town in the Midwest, neighbors and others in the town spread rumors that the family was receiving government pay for their adoptive children and the government had paid for their house. Jan stated this “hurt the most because people were looking for the negative…there must be some gain from it…it was upsetting.”

Lily (18) is the only one of their children who was born with a cleft lip and palate, a condition Jan says is much more “minor…compared to some of the stuff we’ve dealt with.” Lily’s ethnicity is Asian. Lily had had one surgery in her orphanage for her lip repair at the time they adopted her. She also had a club foot, a missing leg, and deformed fingers. Lily currently receives care at a Shriner’s hospital in a nearby metropolitan area and formerly was under care at a children’s hospital in the Midwest where the family used to live. Jan estimates Lily has had 10-12 surgeries related to the cleft lip and palate.

Jan has home schooled her adopted children from kindergarten through ninth grade because she believed that society and school have “changed a lot…there’s a lot of
yucky stuff going on.” Widespread drug use was one of her concerns. Jan’s goal is to give her children a “good foundation to be able to you know make it through life and not be sucked into whatever is going on and really be able to think it through better.” Because the children were homeschooled together, were all adopted, and all had special needs, they never felt “I’m adopted that’s awful.” Jan is a former interior designer who has been a stay-at-home mother. Dave works as an architect.

**Participant Themes**

In the following section, themes that emerged from the data are described. The themes are illustrated with details from the individuals’ lives as well as quotes from the participant interviews.

**Identity Development and Acceptance of Perceived Difference**

Four families described awareness of identity as a factor in living with a cleft condition. One mother felt her three children’s sense of identity was helped by the fact that as parents they focused on and encouraged individualism. Two of the adolescents described experiencing an impacted sense of identity due to the presence of a cleft condition and one described no concern about her identity due to a level of acceptance for her condition.

**Anne.** When reflecting on living with the condition of cleft lip and palate, Anne said, “I kinda like it because it makes me different from other people and I’m like not the same and things as other people.” For her, the condition made her unique: “There’s kids out there just like me. I have my own special group of other people like me.” This special group “can’t judge me cause they are the same as me, like we all have the same thing in common.” While her uniqueness made her “different” in the eyes of others, she
also expressed she was equal nonetheless. She said, “I might look different but I’m the same personality wise as other people. You just don’t understand I’m still the same as everybody else.” Clearly, she experienced this difference as a potential source of alienation or misunderstanding.

**Jessie.** In discussing parenting, Jessie said that perhaps she and her husband’s personalities have influenced their children’s level of self-acceptance, although not consciously: “We are who we are and that’s how the kids have been raised. You don’t have to be like everybody else, be the best you you can be.” They did not parent in this way specifically because their children all had this condition” “I don’t know if we were doing it differently, these are just our kids.” Of her oldest, she said, “She thrives on being not like anybody else” in terms of fashion, music choices etc. And her younger brother “follows in her footsteps.” She did worry when they were younger about social acceptance, which she now feels was “a waste of energy.”

**Lily and Jan.** Lily’s overall attitude toward her condition and treatment was very positive. She said, “It’s not a huge deal. I think people have it a lot worse.” She expressed less concern about living with the condition than having to endure the treatment process. Jan indicated this process itself has “made me think about myself differently...if I could have taken the surgeries out I could’ve been a little happier I guess. It kinda makes you insecure, you know there’s something not right and you’re working on it.” This sense of not being “right” felt like a burden to her as did the chronic invasiveness of the treatments. Having to explain the treatments to others also bothered her. She admitted that after one surgery, she told people at school she had gotten into a
fight when they asked her about the bandages rather than have to explain that she got a “nose job.”

**Extensive and Lengthy Medical Treatment (With Significant Impact)**

**Lack of certainty in medical process.** Every family described a treatment process full of changes and shifts due in part to development and growth of the children, in part to less than successful outcomes or side effects, and in part to changes in technology. Four out of eight families described a desire for their medical teams to communicate more clearly with regard to some aspects of the treatment process. Although praising some experiences within the long course of treatment, these families were unsatisfied with the communication of expectations for timing of interventions, recovery processes, and the overall treatment course. While all families expressed an understanding of the difficulty of providing specifics and certainty in what could be a fairly fluid treatment process, most felt this should at least be attempted. However, two families expressed a preference for vague and open-ended communication about the course of treatment due to the high potential for change and to lessen any unnecessary expectations of the patients.

**Paula.** While praising her daughter’s medical team in terms of expertise and ability, Paula stated that as a parent, she needed more information about the nature of the treatment process:

I would have liked to be a little bit more informed about the extent of what we were talking about as far as surgeries and stuff. I was left with the impression that have some surgeries and ear tubes and she’ll be fine…(I) kind of felt like I was not well informed or were (they) feeding bits at a time. I kind of would have like to have that life time warning that this is a 20-year ordeal, don’t get complacent after year two. I kind of felt cheated and maybe they think parents don’t want to hear it or maybe they can’t handle it…I felt like they had sugar coated it for me.
Jeff. Jeff stated he felt a need for a more thorough understanding of the reality of treatment outcomes:

I don’t know if there’s a better way to prepare parents that things don’t always go exactly the way they’re supposed to. We had really bad luck with the surgeries. The team was great but they would say there is a “very low percentage we have to do it again” and Aidan always experienced that. His surgeries were always successful but if there was a side effect we were going to get it… that was really tough to keep having to deal with those things.

Francine. Francine felt her daughter’s team communicated poorly regarding pain management options and treatment planning. Her daughter was “really anxious to have some of these repairs because it changes your appearance and your ability to talk.” Francine thought this would “improve her self-esteem.” They were initially told to wait a year; at the end of which, they were then told they had to wait for several physical changes to occur. More information “would have been nice…so she didn’t get her hopes up… she was really disappointed, really disappointed.” Francine saw her daughter mentally prepare for it: “You think okay here it comes, this tough surgery...you get prepared mentally and emotionally and then nothing. It makes her angry.” Francine also noted that poor communication about treatment expectations made her job as a parent more difficult since she liked to support the preparation for treatment. Instead, Francine tried to temper any excitement her daughter felt about upcoming treatments with reminders about the inherent uncertainty of the process. Jennifer was open about her negative feelings towards her medical team: “I mean as a kid I was pretty okay with them but as I got older I actually started to really hate them…I get mad that sometimes I think they’re postponing stuff on purpose.” She believed her surgery was being delayed because they had other patients who were first on the list.
**Lily and Jan.** Lily gave several examples of times she felt communication was not adequate from her team. As a child, she said she did not have a clear understanding of what was going to happen and why with regard to surgery. She also noted she had to have the same orthodontic procedures twice due to growth and did not understand the purpose of this. She said the “surgeries keep popping up, out of nowhere…like o yeah you have to have this- like the bone graft I just had I had no idea I had to have it. When I found out, I was really upset about it.” After one surgery, the doctor did not provide enough information about eating restrictions; as a result, she ate foods that caused excessive bleeding from her mouth and nose. On the other hand, Jan thought less specific information regarding the overall course of treatment was better in the long run. About the extent of the surgeries needed, Jan said, “I think honestly I’m glad I didn’t know, we still would have adopted her but I’m glad I didn’t know. I didn’t need to be burdened down with knowing there were going to be a million surgeries ahead for her.” She believed doctors were unable to prepare people accurately for what lay ahead because of the inherent uncertainty in the process and therefore, “They don’t want to burden the parent.” As an example, she related that her daughter’s first team made the final jaw surgery sound horrible; in fact, they used the word “gruesome.” Whereas the second team was much more positive; this difference was striking to her and she felt the first team created more anxiety than was needed about that intervention.

**Jessie.** Jessie also stated a preference for a less detailed understanding of the treatment regimen and viewed it as a sign of care. She stated, “It’s a shock for parents to know what lies ahead” and recommended that doctors “be gentle and vague because things change so quickly, especially now the treatments have changed.”
developments in technology have altered and improved the original treatment plan for her children.

**Financial impact.** Three of the families discussed experiencing financial burden as one impact of the condition of cleft lip and palate. The other five families did not specifically discuss any financial impact. Three of the eight families were receiving care at a Shriner’s hospital, which offered care free of charge, excluding orthodontic work in some cases. All the families possessed health insurance and one specifically mentioned an ongoing struggle in having procedures reimbursed by their insurance company. One family mentioned financial burden due to the unexpected cost of orthodontic work. Two families experienced the loss of one parent’s income due to the time involved in managing the treatment process. Multiple children with the same condition and traveling long distances for care increased the difficulty of maintaining employment. One parent mentioned the difficulty of financing mental health treatment for their adolescent.

**Jessie.** Prior to the birth of their second child, Jessie was a full-time teacher. The loss of Jessie’s income cut their family earnings by exactly half. She said, “That was impactful, very impactful financially.” The three children’s treatment needs as well as the loss of her income have been a “financial burden.” Yet Jessie said, “There are things that are more important than other things.” When her children were younger, they required more frequent surgery but as adolescents, they required frequent orthodontic work. They do not live near a cranio-facial team and she has had to find independent providers to complete their medical care. She said, “You should look at my calendar, setting up, finding care and getting three kids into orthodontists and dentists. It was a full-time job.”
Janet. Janet ran a daycare prior to Frank’s birth. After he was born, she has only worked occasionally due to Frank’s treatment needs and the three to four-hour drive between their home and his cranio-facial team. Janet said there was “no way I could” work with Frank’s treatment schedule.

Relationship with Medical Team

Positive relationship with medical providers. All eight families described overall positive relationships with their medical providers. One family had a significant negative experience that led to them to choose a new medical team. Three families experienced significant complications or negative interactions with their providers yet chose to stay with their medical teams due to their level of expertise. Two families experienced changes in staff within the same team and two changed teams due to relocating. Despite significant challenges in the treatment process including frequent complications, setbacks, and need for revision surgeries, families described a continued sense of trust in the medical provider’s expertise and appreciation and gratitude for their care, concern, and availability. Two families discussed a sense of closeness with their medical providers, likening them to “family.”

Kristin and Paula. Kristin described her experiences with her doctors and nurses as supportive, saying they were always kind, available, and helpful during surgical treatments. She described the yearly visits with her craniofacial team as informative as well as social, stating they were a time to “catch up with them and ask them questions if I need to.” When it came to guiding the treatment protocol for her daughter, Paula trusted the medical team. This trust was partly based on the professional’s availability, concern, and attentiveness to the family’s needs. Paula said, “They have been amazing about
consulting with us, making sure concerns that we have are taken care of.” She stated the surgeon is “available, easy to talk to.” Trust in the surgeon was based on recognition of his expertise as well as their shared experiences over many years. Paula affirmed, “If he says something I really do believe--you know he’s been there the whole time. If he tells us that she needs something I still research it but I trust it.”

Lisa. Lisa expressed a sense of trust in the craniofacial team due to the level of expertise:

Being part of a cleft team is super, super important. I love it that they all come together and say okay…this is how we are going to work together as a team for your child. I think I went along with what they said because they have so much expertise and they all work together to say we’re here and we’re here and work together to make an overall plan for your child.

Lisa’s relationship with her daughter’s first surgeon was challenged by his lack of empathy and his inability to answer questions, which concerned her as a parent. However, through time, she recognized his expertise lay in his surgical work; she gained acceptance of the fact that he could not understand her experience as a parent or guide her in this experience. Lisa expressed gratitude for the help he provided her daughter. When he retired, she felt his loss as personal: “It was like losing a member of our family because he had done so much to help my daughter.”

Janet. Janet described a professional but also a personal relationship with her son’s providers:

We took our first trip there when he was two-weeks old. The lady that’s the speech pathologist on the team she took a liking to him from the first time she saw him and she has pretty much taken him under her wing and gone way out of her way to try to make things better for him.

The speech pathologist has not only coordinated appointments for them on weekends but met the family at the appointment and then met them for lunch. Whenever Frank visits
the hospital, his surgeon tries to stop in briefly to see him. Several providers on their team have gone out of their way to be accommodating, supportive, and available. Janet said, “I can’t say enough about those people. If it wasn’t for them and the speech therapist… he would not be the child he is today. There’s no way he could have endured what he went through and become the type of person he is.”

**Jeff.** Jeff expressed gratitude for the experimental treatment his team provided that made a bone graft surgery unnecessary. Despite the fact that his son experienced many complications, side effects, and failed treatments, Jeff had “nothing but praise for the team… they’ve been wonderful.” He felt the team had worked well with Aidan’s reluctance to undergo treatment and provided clear information in an attempt to work with him. He was appreciative of their expertise as well as their efforts to work with Aidan and attempted to persuade him to comply with treatment.

**Francine.** Francine felt a close relationship with the head nurse on her daughter’s medical team, with whom she had the most frequent communication. She said this nurse had been a constant on the team and their “relationship is a good one.” She was available and listened to the family’s needs and preferences. For example, Francine stated a desire to only see certain providers at the yearly clinic meeting due to insurance coverage and personal preference and the nurse arranged this for them. Of her daughter’s surgeon, she said, “He’s good, he’s kind, he’s patient… he’s really good about telling me this is the surgery and this is what it’s going to look like.”

**Lily.** Lily spoke of her current team as being “one of the best. They know what they’re doing. They work a lot with kids and a lot with this problem specifically.” She
stated she felt the team had real expertise and much more experience with this condition than her former team.

**Need for increased emotional support from team.** Three families described a desire for the medical team to have more awareness of the emotional impact of the treatment process and/or condition and to provide some support for this. Only two families were offered formal emotional or psychological support by their medical team in the form of regular screenings or check-ins with mental health professionals at the yearly clinic visit. One parent stated she never felt the need to further utilize this support system; the other did for her daughter but not for herself.

**Lisa and Anne.** At various times throughout her daughter’s childhood, Lisa had felt the need for more support and understanding from the medical team. She called this support critical for a parent:

> Even if it has to be part of the cleft team, just having a parent and having a room where you can go and say I’ve been through this and you’re going to be okay. You need that emotional support. They all have their area of expertise…ENT’s, neurologists, whatever …whatever… but nobody is an expert in being a parent like somebody’s that’s parented a cleft child. I think that’s super, super important.

Anne expressed a desire for her medical team to understand her experience both physically and emotionally. She felt the medical providers did not understand the uniqueness of the condition and how it could impact treatment adherence and, in particular, pain. For example, because her mouth was proportionately smaller than other children her age, completing some orthodontic work was much more painful than it would be otherwise. Anne also said she thought the medical team should understand what it is like to live day to day with this condition--from being misunderstood or
mistreated by others, to facing constant uncertainty about treatment outcomes, to how
difficult and frustrating it is to eat with fistulas in her mouth:

There’s a lot of foods and things I can’t eat. I have like this hole in between my
nose and my mouth. If I eat like peas or things they’ll go up into my hole and it’s
disgusting. It’ll come like between my nose and mouth. And so the cleft team
can kinda help, like the bone graft surgery was supposed to help cover up one of
the holes. And it kinda did help.

If they knew what this experience was like, she said, “They can know what surgeries to
do or how to help me more.”

Jessie. Jessie said she would like the physicians who work with cleft affected
children to know that “these are our babies.” While overall her experiences have been
positive, she said, “This is not a flight plan.” Her recommendation for them was to be
gentle, stating, “It’s their job but it needs to include a bit of compassion.”

Jennifer. Jennifer felt emotional awareness and emotional support were lacking
in the interaction with her doctors. This feeling stemmed from childhood. Jennifer said,
“I mean when you have surgeries as a kid you can definitely get traumatized with
it….since it happened over and over I got pretty used to it and so did the medical staff.
They were obviously nice about it they would comfort me.” However, she felt they did
not realize the impact it had on her. Communication about each intervention and the
expectations for recovery were very poor. In her opinion, doctors should “maybe talk to
the kid about it- tell them exactly what’s going on …instead of getting something dumped
on them and not knowing what’s happening.” She also thought the teams should include
a “child psychiatrist who comes in to talk to the kids about their feelings…because a kid
can’t just bottle this up their whole life. They have their mom to talk to like ‘this hurts
and this hurts’ but not exactly like what they emotionally go through.” She still felt this struggle currently: “I don’t think it got any easier I just got used to it.”

**Coping Through Social Support**

Seven out of eight families discussed the importance of social support in coping with the medical treatment and the daily impact of the condition of cleft lip and palate. Three families discussed having practical support from family members such as medical recommendations, help with childcare, and recovery care. Two families mentioned the importance of emotional support and acceptance from family and friends. Four adolescents mentioned their parents provided caretaking and practical guidance and advice surrounding medical interventions. Four mentioned parents helping through with emotional concerns related to their condition. Two discussed the importance of the support of siblings or outside friends.

**Kristin and Paula.** Paula noted that having support of her spouse, her older children, her family, and her husband’s family was critical in managing the time-consuming medical treatments. Extended family members took time out of their schedules to help with childcare during surgery and recoveries. Her brother’s father-in-law sponsored Kristin’s care at Shriner’s Hospital. Kristin’s parents helped her before, during, and after treatment with their presence, caretaking, understanding, and through providing guidance and decision-making regarding the course of treatment. Kristin’s mother was the primary one involved in the day-to-day physical care and assistance following a surgery. In addition, her mom provided emotional support by normalizing and validating her experience: “She always (says) it’s okay to be scared and that it’s normal.”
Anne and Lisa. Lisa mentioned the role of supportive friends and family in coping. Her parents provided her with a home and emotional and practical support during Anne’s infancy. In particular, she mentioned that friends who had witnessed what Anne had been through and who communicated understanding and acceptance of her were the most important to Lisa: They have “really taken the time to know Anne for who she is, what a beautiful human being she is…so obviously they can look past the differences and see her for who she is.” Lisa also mentioned the invaluable support of a relative who offered her valuable insight and perspective:

She is so good for me, she is so good for me. She is so good for me because she always has such words of wisdom for me that really, really help me. Anne has a lot of gay friends--pretty much almost every friend she has. As a parent that worries you…she said, “You know they’re kinda living their own scary little world and situation and she’s living her different own scary little situation. They’re safe for each other they know they’re supposed to have relationships with girls and she knows she is supposed to have relationships with guys but in her mind no normal guy is going to accept her because she’s different. So this is a safe place for her to have a relationship with guys--safe for them, so they won’t judge each other and they are there for each other and its safe for all of them.”

Anne said that mostly her mom helped during surgery recovery periods through providing caretaking as well as reassurance. Anne felt the support of her friends at church played a large part in her ability to regain her confidence after being bullied. She reflected the people in her church were always available to talk, very supportive, and she found it easier to talk to them at times than to her family. These friends helped her to be able to “not care what they think if people make fun of me or whatever.” She learned that “you just have to stay strong inside and not let them bother you.”

Margaret. Despite some anxiety about surgery, Margaret noted the surgeries brought her family together. At her last surgery, her mother spent the night and the “whole family stays” throughout the day to support her. Margaret saw another side to her
sister, whom she said is “always really nice” but who becomes incredibly caring and compassionate during her surgeries and recoveries. She was grateful for the closeness these times brought to her and her family. Margaret attributed her mother’s support to helping her overcome self-consciousness about her appearance. During this time, she talked with her mother a lot and said her mom helped her put her struggles in perspective: “She didn’t only tell I shouldn’t feel that way, but she understood what I was saying and going through. I just remember my mom always making me feel super confident which helped a lot.” Her mom also encouraged her to be proactive about making changes in her appearance; she “always said there were options that I could do.”

**Janet.** In terms of family support, Janet said, “We had the best of the best situations. We had a kid with a cleft but we had everything in place to deal with it. My parents have always been there, there’s always aunts, grandparents, everybody.”

**Jennifer and Francine.** Jennifer’s mother helped her cope with medical treatments through emotional support and taking care of her physical needs. Jennifer stated she managed the last difficult recovery with “the support of my family and mostly my mother cause she was my caretaker…she would get up at night and make sure I was okay. She would obviously make food I was able to have because I couldn’t have most food.” When facing the unknown, fear, or pain, her mother said, “Don’t worry it will hurt but it helps you.” In discussing helping her children cope with treatments, Francine said, “We just parent the way we parent.” They also tried to spoil the recovering child with special treats, pajamas, etc. and to minimize as much time as possible spent in the hospital.
**Lily and Jan.** Lily was appreciative of her parent’s support. She said they were always there for her and “really supportive. They don’t say it’s a problem--just supportive.” Living in a family made up of internationally adopted, special needs children provided Lily with a supportive social group. It also normalized their many differences: “None of them feel Oh I’m adopted that’s awful because they know we’re crazy about adoption.” Jan provided the type of emotional support during treatment she felt Lily needed. She said she was not someone who liked to be babied, instead “I just let her go through it. I used to try and comfort her and hold her. No, she wants people to stay away. I honor that.” Rather than speaking about what she did for Lily specifically to support her in terms of her condition, Jan noted ways she has encouraged the environment to be one that encourages positive development more generally. Jan believed homeschooling was one critical way she provided emotional support to Lily. She said it provided a “good foundation to be able to you know make it through life and not be sucked into whatever is going on and really be able to think it through.”

**Jessie.** Jessie believed the fact that her children all had the same condition helped them cope. As younger children, this made it very normal to them: “It was so funny taking them for walks through the neighborhood and there’d be a baby and they’d look like ‘Oh mom where’s his cleft?’” And she was aware they could provide a level of understanding for each other she could not provide: “At this point in time right now where I don’t know what it’s like being a teenager with a cleft, they have each other.” Jessie described physically caring for her children after surgeries as well as letting them choose special foods, gifts, and distractions to help them through recoveries “so they’re part of it.”
Support Groups and Community Involvement

Six of eight families described becoming involved with support groups and/or advocacy groups of some kind. Families sought both support for themselves and offered advocacy and support to others. Of the two families that did not mention social support groups, one family stated their medical team had a support group that seldom met and the other felt that having been involved in other types of parenting support groups over the years, they no longer needed that type of support at that stage in their lives.

Lisa and Anne. Lisa sought out support and information from other parents of children with cleft conditions and found it to be critical because doctors “don’t know how to be parents to cleft kids.” The reassurance of families who were further along the treatment process was vital in providing hope and understanding. In this way, she learned details of upcoming treatments and what the recoveries were like; she shared this with her daughter to help create realistic expectations. Lisa co-created a parent network for her daughter’s hospital that provided information and support to new parents of children born with cleft lip and palate.

Jessie. Jessie said, “Getting involved in the hospital really, really helped shape our lives and maybe our personalities.” The advocacy work she and her family have done has helped them all gain a community, understanding, and a sense of purpose. As a toddler, Cammy was chosen as a poster child for a hospital fund raising mission, which led to the whole family becoming involved in fundraising and outreach for cleft conditions in their community. Jessie noted it “opened up opportunities and friendships and a community that we didn’t know.” She noted this type of charity work was “good for the soul.” Jessie found information and support groups “vital” and noted they had
become more positive and encouraging in their tone over the years. While their team initially provided social gatherings for children to meet one another, this ceased with the advent of social media and online support groups.

**Margaret.** Margaret noted her involvement in nonprofit work was one of the main means by which she gained perspectives that allowed her to adjust to her own condition. Margaret founded a non-profit at the age 13 with her sister who is two years older than her. The non-profit raises money for families traveling to her city for treatment for cleft conditions and pays for extraneous expenses such as hotels, gas money, etc. This work has helped Margaret “realize how fortunate I am that I’m able to afford all the surgeries and stuff and definitely puts a perspective on things and makes me realize that it’s not as serious as it could be.” She learned about running a non-profit and discussed her excitement at the real contributions she has been able to provide to families in need.

**Janet.** Janet sought out the support of other parents, which eventually led her to become very involved at a senior level with one of the national advocacy groups for cleft lip and palate. She felt the support of advocacy groups was critical for parents: “If it hadn’t been for that organization, I wouldn’t have handled things as I did.” She sought information about living day-to-day and parenting a child with this condition. Other parents were critical because doctors don’t know “what the day to day is...they don’t live it the second we walk out of the hospital.” She also believed the medical team had learned from her experiences and thought “every team should have some type of parental representation on their team.”
Francine. Francine has found support from other parents critical, which she found through online support groups. She also said she would like to have an organized support group as part of her daughter’s team.

Lily. Lisa said it would have been nice to have the support of other children going through similar experiences: it “could have helped me accept it more or understand it more or just know other kids like me…like I’m not the only one, why me?” Interestingly, Jan stated she felt that as a family, they had outgrown support groups for adoption and therefore she never looked into this type of support specific to Lily’s condition.

Paula. Paula also sought out the support of other parents of cleft affected children online. This mutual support led to friendship: “It goes beyond cleft support now but we also know that if we have something going on with our kids we can talk together and we all get it.”

Varied Emotional Impacts of Condition and Its Treatment

All eight families mentioned being emotionally impacted by the presence of cleft lip and palate and the treatment process for it. Parents discussed sadness, guilt, and anxiety with regard to their children’s medical interventions and the struggles they managed in living with the condition. Adolescents discussed anxiety related to medical outcomes and surgeries, anger and frustration at physical limitations, the length of the treatment process, and negative social interactions. Two adolescents experienced more significant emotional distress due to incidents of bullying. Three adolescents reported feelings of ambivalence toward their medical treatment and treatment providers. This ambivalence revolved around recognizing the need for and yet resenting treatment
interventions and about whether the outcomes were worth the painful process. None of
the parents mentioned any ambivalence toward the medical process.

**Kristin and Paula.** Kristin stated that prior to recent surgeries, she experienced
some anxiety: “I was a little scared but at the same time it wasn’t that bad. But I was still
a little scared.” Her main concern related to the uncertainty of the treatment outcome,
how surgery would impact her current appearance, and what it would mean for the
ongoing course of treatment. She said, “I’m um…scared…I’m really nervous…like after
like how it looks—after…and down the road like is everything going to be done.” Paula
also noted increased anticipatory anxiety in her daughter toward surgery as she grew
older: “Everything went great until her first bone graft when she was nine, old enough to
know what surgery was. So it was scary.” While Kristin experienced some social
anxiety, Paula did not attribute it to the cleft condition:

Socially she’s a little slow but I don’t think it necessary cleft related I think it’s
more anxiety related…in social situations she doesn’t deal with people all that
well most people think she’s quiet and shy…but in reality some of those social
situations are pretty scary for her.

Paula described struggling to set aside her own emotions in order to show strength
for her child around the time of surgeries:

I try really hard to not be really emotional when she’s really emotional. Because I
think it’s my job to show her that it’s okay for her to be scared. I don’t want her
to see that I’m scared… so I wait until she goes back and then I fall apart… I
want her to see that I’m strong and confident and that I’ll be fine if I’m a nervous
wreck along with her she would wonder why. I have to be strong so that she can
have somebody she can rely on.

The emotional impact of the medical interventions on Paula included having to
witness her child’s physical pain. Speaking about the immediate recovery period, she
said, “I see her at her worst and that’s really hard.” When asked about how she coped
with this, she said simply, “I feel horrible… I don’t like to think about it… you wish you could take their place but you can’t.” The emotional strain of the treatment process challenged the role Paula saw for herself as a mom. Her belief that being a mom meant being the strong one was challenged by parenting a child undergoing painful medical treatment:

I really was raised that mom is supposed to be the foundation of the family. It’s my job to hold everything together. It was a challenge when she was young... it was really hard for me to balance holding that together with not having a nervous breakdown with everything. It really was… I don’t get to break down or say I’m just so sick of this, that’s not my job and not letting anyone down.

**Anne and Lisa.** Lisa discussed her daughter’s different reactions to surgery at different ages. All of them were challenging and caused emotional reactions in parents witnessing this process. Surgery during adolescence was complicated by the increased awareness of her child, which in turn created new challenges for parents. Lisa said, “She’s 14 now and so that was really hard because she’s old enough now to know… that one was really hard on me too because it was hard on her because she’s old enough to understand.” Anne’s last surgery was only months before our interview. Anne recalled being frightened by “not knowing what was going to happen.” Her worry was so heightened she could not remember how or whether anyone tried to help her prepare for the surgery: “I was really worrying a lot. I don’t remember anything.” She also recalled experiencing pain during recovery that was far worse than she had expected: “I was in a lot of pain and it lasted about a month.” Anne experienced ambivalence about the treatment process. She felt resentment toward treatment and her providers but also recognized benefits from it. In discussing recent treatment experiences, Anne said, “As
much as I don’t want to admit it, I can tell things are getting better.” Another time, she said, “It was the most painful thing in the world but it helped bring the teeth close.”

**Jennifer and Francine.** Jennifer’s feelings toward her medical team are currently negative. In explaining this, she said, “I mean as a kid I was pretty okay with them but as I got older I actually started to really hate them…I get mad that sometimes I think they’re postponing stuff on purpose.” She could only imagine surgery was being delayed because they had other patients who were first on the list. The next surgery was one she really wanted because it would change her appearance greatly for the better and the delays were very upsetting for her. Jennifer described her last surgery as “like probably the worst year of my life.” The pain she experienced was far worse than she had been led to expect as well as the debilitation: “When I came home I was on the couch for weeks. I could barely walk and stuff.” Jennifer described ambivalence about treatment as an ongoing experience: “Some days I’d be excited for it…and other days I’d dread it.” Jennifer described her excitement for treatment in connection to her hope that “I was getting fixed. I had it in my mind that I would be able to eat right without pain or talk right.” Her sense of dread related to the physical pain involved. With her daughter’s last surgery, Francine was not given options for managing pain other than a pain pump. Her pain did not seem well managed and the recovery did not go well: “That was sort of a turning point for things going well for her because that thing was horrible…she was more fearful and worried about pain.” It was such a bad experience for the whole family that when her son faced the same surgery, he refused the pain pump and had a much better recovery. Francine said she was frustrated with this: “I was like what, there’s an option? I didn’t know there was an option?”
**Lily and Jan.** Lily’s surgeries were successful and proceeded as planned or even better than they were led to believe. Jan said she just “didn’t realize there were going to be so many surgeries.” Some of the success she attributed to Lily whom she described as incredible...she is a tough little cookie. The first 48 hours she’s really withdrawn seems kind of angry...and right before surgery too she’s like she doesn’t want to talk and I understand that. And she hardly ever takes pain medicine...she has a high threshold for pain.

However, Lily mentioned one bone graft surgery that was particularly difficult for her, saying the recovery took two weeks: “I couldn’t actually walk…there’s like three places you have to worry about with incisions. It was a lot of work. I couldn’t eat very much, it took a long time for me to actually eat anything.” Lily also described ambivalence toward the treatment process. Two of Lily’s cosmetic surgeries led to very little apparent change. While she had not wanted the surgeries to begin with and was somewhat anxious about having a drastic alteration in her appearance, she felt somewhat disappointed in having to undergo the preparation, worry, and treatment for such little result:

Honestly we’d been planning it for years I was always really scared--you’d look different you’d look quote unquote better…and it’s your face too. I go to public school…people were going to ask questions. But the result felt to me it was kinda a waste of time... I wouldn’t do it again.

**Janet.** Janet expressed feeling some guilt for Frank’s condition due to the uncertainty as to whether exposure to alcohol and nicotine led to the development of his cleft lip and palate. She also hated to witness him going through the pain of the medical treatment:

I guess you do it. You have to. There’s been tears shed and words flying around and there’s been moments when you want to beat anything close but you don’t do that. I get my frustrations out in the car by myself. There would be screaming sessions when nobody’s around to hear me.
But the emotional impact was never debilitating: “We dealt with it… it was trying at times but there were never any issues that kept us from being logical about it.” Janet stated if she had one regret it was not handling the impact of Frank’s treatment on his brother. As a child, he acted out; even now, he had “ill feeling” toward Frank and believed she loved Frank more because of the time she spent with him due to his treatment. Juggling the needs of these two young boys was difficult and although she felt she handled it well at the time, she knew he was still bothered by it. She also had concerns that repeated anesthesia and x-rays might have impacted his health or cognition. She acknowledged these worries were based on her “instinct” and she had no proof of anything being wrong.

**Jeff.** Jeff also described the challenge of witnessing his child suffer pain and anxiety in relation to surgery. But the “biggest impact on the family was my wife’s inability to nurse him. That really had a big effect on her … that was something my wife never fully got over. She tried to nurse and express milk and it just didn’t work.” Because of this, he and his wife considered an experimental surgical technique offered in another state. This involved very early surgical intervention in the hope this would allow him to nurse. They decided against it due to geographical distance and the recovery process. Although not all cleft lip and palate related, the impact on the family of Aidan’s emotional and behavioral problems has also been “pretty devastating.” Aidan’s behavioral problems and escalating violence put strain on both individuals and the family system. Jeff and his son were having regular physical fights; his wife felt afraid of her son and eventually chose to live apart from the family. All of this together led to marital separation. Aidan has no contact with his mother currently by his own choice. Jeff says he tiptoes around him and does not put demands on Aidan. Jeff stated he is “not dealing
very well” at the moment but is still hopeful about finding treatment options for him in the future. Aidan has dropped out of regular and online high school. His only social contact is online. Jeff is very “concerned that the way he is now that he can’t hold a job…” and he will never be independent. His concerns about medical compliance and long term outcomes of his cleft lip and palate are minimal by comparison.

**Margaret.** Margaret stated she wished the treatment course could be completed more quickly and described a strong desire for surgeries that would continue to improve her appearance and ability to bite.

**Management of the Condition Includes Ongoing Decision Making)**

Families chose a medical team either through familiarity with the team or hospital or simply by geographical proximity to their home. Only one family left a team due to dissatisfaction with care. One family researched team protocols and approaches prior to choosing a team and the rest followed the protocol of their chosen team after the fact. Only one family stated they had decided against a treatment intervention altogether, feeling it was unnecessary and would need to be repeated later in their child’s development.

**Kristin and Paula.** Paula described trusting her medical team to make medical decisions but still felt the need to be fully informed and to ask questions regarding interventions: “I never blindly do what they say, and I ask bizillions of questions and I research everything. I’m crazy that way. I don’t just go along with it. I’m really anal about researching it.” Planning for medical treatment involved not only information seeking, consultation with the team and her daughter, but also considered the schedules of the entire family. Paula and her family planned surgeries with the goal of minimal
disruption to the entire family system. Surgery impacted her husband’s work, which she stated was thankfully flexible enough to allow for such accommodations. Paula encouraged Kristin to be involved in decisions regarding her medical care. She asks Kristin to write down her concerns and questions for the medical team; “I encourage her to ask the questions herself. As she’s gotten older, I tried to get her to be more proactive with the doctor.” She also allows Kristin to make some medical decisions: “anything that was functional she has no choice…it the doctor said it was cosmetic, just cosmetic she could choose.” Kristin was comfortable with her parents and the team making medical decisions on her behalf. She trusted them and knew she could communicate her concerns to them.

Jessie. Choosing a team for her children was easy as there was a craniofacial team “already in place” across the street from the hospital where Jessie gave birth to Cammy. Making decisions about the course of treatment has been mainly left up to the team and has been “mapped out” for them, although with changes due to technological advances rather than to unexpected complications or alterations in the protocol. Although she mentioned several surgeries that needed to be repeated, she did not express any sense of frustration or concern about them.

Margaret. Margaret expressed gratitude that her parents had given her the option to make her own medical decisions. Of the three most recent surgeries she has experienced, Margaret stated they were all “definitely cosmetic” and her parents let her decide whether or not to have them. Margaret also instigated the idea of one of them to her mother, who directed her to her surgeon for his suggestions:

They are pretty lenient with asking me what I want to have done which I appreciate. I mean there are definite boundaries they wouldn’t let me do anything
I would regret. They give me advice because I know they know best and it’s hard
to decide when I want everything to change when I’m younger.

Janet. Janet said her family never considered another hospital besides Shriners
because her husband’s family has been involved with this organization for generations.
Since Frank’s birth, she and her husband have run charities at times to raise additional
funds for the hospital. Janet began to include Frank in treatment decisions as soon as he
was old enough to understand. She presented him with choices and felt it was important
to try to honor his choices. At age seven, he first chose to try a less invasive treatment for
his sleep apnea but after struggling with it, he requested a surgery. Janet said, “I wasn’t
going to tell him no. He was the one that had to sleep with that tube around him; he had
full right to make that decision.” She described the struggle involved in allowing him to
make choices for himself as he got older. Currently, Janet believes he should have a nose
revision surgery but “he thinks it’s not an issue.” She said, “It’s kind of an awkward
conversation” because it involves criticism of his appearance. She is reluctant to openly
criticize his appearance but has concerns that later in life he might regret not having this
surgery done. But she recognizes that “right now I think all of this stuff is a distraction
that he doesn’t want to talk about.” She tried to have the doctor intervene but this
backfired as the doctor referred it back to her dissatisfaction with his appearance. She
stated she has resolved to give him several options while not doing anything is not an
option.

Jeff. Jeff and his wife researched several teams and protocols when Aidan was a
baby with the hope that one might allow his wife to nurse him. Eventually, they chose to
remain with a hospital in their city to make any follow-up care needs easier. They were
also excited to be involved in an experimental treatment that eliminated the need for a
bone graft surgery. Since he was a young child, Jeff and his wife tried to involve Aidan in decision making where possible: “We always tried to give him the illusion of choice.” While the team was more direct, he and his wife attempted to explain the choices and discuss the benefits and potential consequences of these with him. Jeff recalled Aidan frequently saying, “I don’t want to do this surgery. I don’t want to do this surgery.’ And then I’d sit down and explain it to him and he’d give me this and look and go, ‘okay I’ll do it.’” Lately, however, Aidan has become more resistant to any type of treatment or care. Jeff is accepting the fact that at Aidan’s age he has the right to make decisions regarding his care and said, “He is making what I don’t think are good decisions, but there’s not much I can do about them.”

**Jennifer.** Jennifer’s parents made the decisions with the advice of the medical team. She is accepting of this, saying, “I mean it is what it is. They just know this is good for me and we’re going to do it.”

**Lily and Jan.** There were two surgeries Lily felt she did not want and would not have chosen to complete without the influence of her parents. “I feel like they think it’s necessary when I would have rejected a couple,” she said. “Mom pushed me into it. Gotta listen to mom.” However, about the nose surgery she also said that “friends did tease me for my nose, so I guess I was sorta into it, but I was just really unsure.” Because both of these surgeries did not have the results she had been led to believe they would have, she felt they were unnecessary. However, she was glad she did them because it was “just one less thing to think or worry about.” On decision-making, she had very definite ideas: “I feel like its right to let the kids choose mainly because it’s their own body…I was like mom it’s not you how about you go to school with a beat up swollen face see
how you feel. Don’t just tell them what’s right.” She stated parents cannot understand what the impact of these surgeries are physically and emotionally.

**Reframing and Changing Perspectives**

**Personal growth result of living with cleft lip/palate.** Two parents and one adolescent described the condition of cleft lip and palate or its treatment process as contributing to positive personal growth or development.

**Paula.** One way parenting a child with this condition has impacted Paula has been to increase her empathy for others. She described herself as trying to always be a tough person and having little understanding for “people that feel sorry for themselves or their situations.” But this experience made her realize some people might not be feeling sorry for themselves but for their children, and made her “more empathetic towards people who have children with issues.” The difficulties she has faced also made her aware that many people face much more difficult conditions and gave her a sense of perspective in the difficulties people overcome.

**Margaret.** Margaret called the condition a “blessing,” saying it has made her stronger and given her so much:

There’s so many things I should be grateful for. It might not be the easiest thing in the world but I founded this nonprofit out of it. It definitely helped me care less what other people say about me because I used to and it hurt my feelings a lot and then I realized it doesn’t matter and it’s just not that big a deal anymore.

**Jan.** Jan thought having this condition and the treatment process have only been positive for Lily in the long run: “I think it’s made her a stronger person and made her not sweat the little stuff.” Lily was also aware that the blessings of her situation outweighed any negatives; if she had not been adopted, her life would have been very difficult. For Jan, parenting a child with a cleft condition has been eye opening and personally
transforming: “Years ago, I would see kids with clefts and it would really turn me off.”

Yet when she met her daughter,

She looked so beautiful to me...and I thought how foolish of me to think I
couldn’t adopt a cleft lip child. I’ve really grown in this process...I have grown so
much as a person and what I think is beautiful. I was so limited in my thinking
before, looking so narrowly.

**Spirituality is impacted by the condition and is a coping skill.** Two parents
specifically mentioned spirituality impacting their perspective and providing coping
mechanisms for their experience.

*Lisa.* Lisa’s religious faith and community were a source of support for her. Her
faith also provided a different perspective for understanding Anne’s struggles with social
acceptance:

> We try to always just say “who you are is on the inside and how you treat people.
> God loves you exactly as you are. It doesn’t matter what people say about you. If
> they are mean, it’s probably because they have something going on so in their life
> and they’re trying to make you feel bad so they can have control.” We try to fill
> her mind with positive things.

*Jessie.* Jessie experienced depression and guilt after her second child was
diagnosed with a cleft lip and palate. She said this experience changed her sense of
spirituality. After Ian’s diagnosis, she said, “I was so mad at myself. I felt like I did
this.” An encounter with a psychic at this time had a strong impact on both her emotional
state and her spiritual beliefs. Jessie’s sister, prior to being informed of Ian’s diagnosis,
met a psychic who mentioned a “facial condition” and said there was another child in the
home with the same condition who had “called him to this family.” Jessie said, “It may
sound like hokey-pokey but it really calmed me...in my head it was okay.” Since this
experience, she has continued to identify ways in which there is a sense of purpose to her
children’s health conditions for both themselves and the entire family.
Acceptance and Adjustment Is a Complex Process

Four parents specifically discussed the importance of acceptance in their child’s adjustment process and things they did to try to help with this process. Acceptance of both the reality of the condition as well as the treatment needs were mentioned. Two adolescents discussed acceptance of the treatment process helping them cope with specific interventions that were difficult and two specifically mentioned the role of maturity in the adjustment process.

Anne and Lisa. Lisa has tried to encourage her daughter’s acceptance through openly discussing what she has been through:

Something that we did we had her photographed every six months and nine months, like every other parent does and we hung them in our home. She grew up seeing herself before she was repaired and all that because we loved her and we were proud of her. So she’s always seen herself before repair, and known she went through some things.

Anne said that to cope with each surgery, she tries to remember the overall goal: “one less surgery I have to do to be better.” She was able to have viewed even being bullied through a larger perspective when she said, it “helped me gain my confidence back so everything happens for a reason.” The process of regaining confidence took some effort on Anne’s part as well as support from others.

Kristin and Paula. Paula’s acceptance of the severity of her daughter’s cleft helped her understand and tolerate the difficulties that have occurred during Kristin’s course of treatment. As an infant, her daughter’s surgeon told her that “‘10% are easy, 80% are normal, and 10% are super hard. She’s going to be in the 10%.” Paula said, “Her cleft was super wide and the soft palate was almost bilateral…it was super, super wide. She’s just been a challenge.” Paula associated this with the fact that not every
surgery has gone as planned or had the outcome was hoped for, stating, “So nothing was easy with her.” Paula has tried to communicate acceptance to Kristin with regard to treatment:

We didn’t have a choice. We had to do it so that she’ll be functional throughout her whole life. So it’s not like we were just doing something for the heck of it…now she knows she’s having a big nasty surgery in a year even though she worries about it she knows that she doesn’t have a choice she’s pretty cooperative about it.

Kristin denied any significant impact of the condition on her day-to-day life, saying, “It doesn’t affect me. It doesn’t upset me or anything. I’m fine with it.” She mentioned minor interruptions in school attendance and missing extracurricular activities during recovery periods but stated the teachers were very supportive and helped her manage the missed time. Paula did not believe Kristin was greatly impacted by her condition on a daily basis:

I don’t think it’s really affected her. She’s never come to me and said “Oh my God, this really bothers me.” I think she views it as an inconvenience… We’ve never treated it as something odd or something special. It just is. That seems to be how she accepts it. She’s not odd, not special, it’s just part of her.

Paula denied any current concerns for her daughter’s future with regard to her condition other than the impact of surgeries, which she called, “Kind of a mom worry.”

**Janet.** Janet described her son’s cleft condition as “not a typical case.” Not only was his cleft bilateral, requiring more intervention than a unilateral cleft, but he experienced numerous unusual complications: “He just seems to be the kid that if it can happen it will.” Both for herself and her son, Janet recognized the need for flexibility and patience in coping with this condition: “You have to be able to take things as they come. It’s a snowflake, no two cases are the same. You have to be open minded and very
patient cause things don’t go according to plan.” For example, they have been waiting for two years for two teeth to come in and now have been told to wait another six months.

Lily and Jan. Lily is able to focus on the end goal during times of frustration with treatment: “I just tell myself I’m only here to get better. They’ll fix me…it’ll soon be over. Just bear with it.” While Lily was clear the overall impact of this condition was not as bad as many other health conditions, she also said there were times when she was younger that she felt like it was not going to be okay and that the process took a lot of patience: “It kinda takes until you become an adult.” Jan said Lily’s cleft lip and palate “is minor stuff compared to some other stuff we’ve dealt with.” This perspective helped both she and her daughter cope more successfully she thinks. Having so many children with special needs created a sense or normality in having frequent health interventions. Without an acceptance of what these conditions entailed, Jan warned: “I think you can almost make your child feel inadequate you have to be really careful. This is what’s wrong this is how we’re going to deal with it. Even if it is a big deal to us, we try not to make that a big deal to the child.” She has tried to teach Lily to have the attitude that “it is an inconvenience to have surgeries and it isn’t a deficit…so much of life is how you handle it.” Jan has no concerns about her daughter with regard to her condition, saying, “She has an outstanding personality.” She does well in school, socially, and has a positive outlook. While she once worried about the possibility of social difficulties, this has never happened. She said, “I think it has to do with the child’s personality...if they feel they are a victim...she doesn’t…they are going to bring that on.”
Maturity Eases Emotional Impact for Adolescents

Margaret. When discussing her self-consciousness about her appearance, Margaret said, “I just remember thinking I don’t know why I’m self-conscious about it. I’m proud that I’ve been able to overcome it… the surgeries have definitely helped too. But I think it’s just getting older and more mature and realizing that it doesn’t matter.”

Anne. Anne attributed her ability to cope with both the condition and mistreatment from others to maturity, gaining experience, and increased understanding of others:

   Going back to like kindergarten or second grade I wasn’t as mature. Like if you make fun of me, then I’m going to cry but if you make fun of me now it’s okay like I don’t really care. Like I don’t care what you think about me. Like I’m different, I know I’m different. I don’t really care what you have to say about me.

In general, she felt less bothered by others’ negativity as she grows older.

Communication and Honesty

All eight parents felt open and honest communication with their children about their condition and medical treatment helped them cope with difficulties and adjust more easily.

Paula. Paula witnessed what she felt was poor coping with cleft conditions in her husband’s family’s treatment of his sister: “The family just doesn’t really talk about it. They pretend it’s not there.” Paula stated open communication with her child about her condition and her care was critical: “We’re open about it. I’m just an open blunt down to earth type of mom. Here’s the facts and here’s how it’s going to be--and do you have any questions?” Validation and information increased her daughter’s acceptance and ability to cope with the emotions connected to surgery. Paula described listening and validating
her experience, saying, “We never blow her off.” Paula encouraged her daughter to be open with communication as well: “I might just say ‘So if we see the doctor, do you have any questions? Are you worried about anything?’”

**Lisa.** Lisa stated she has tried to prepare her daughter for treatment by being open with her: “If I learn something, I share it with her…by saying, this is what I know, I’ve been in cleft groups online and I’ve seen people go through bone graft surgeries and I kinda remember the things they’ve said. So just be really open and honest about the things that I had seen, that this is what we can expect.”

**Jessie.** Jessie stated open, honest communication was key to coping in her family: “We talk a lot.” In terms of treatments, she prepares her children by providing information and says, “It’s never really been a problem.” As her children grew older, they communicated openly with her as well: “Once they can verbalize, you know they don’t feel good. It makes it a little more challenging but it’s good because at least they can say it.” And she was open to their emotional experience: “I try to talk to them about it with them to see how they’re feeling… ‘I’m a little anxious’…‘of course you are, I’m anxious for you.’”

**Janet.** Regarding preparing for treatment, Janet’s belief was it was best to “always be up front and honest… whether they understand it or not they deserve to know what’s going on. They just don’t have these surprises.” Janet has tried to prepare her son for the degree of uncertainty in every procedure and even every team meeting: “I don’t care how upset they get, they need to know. Don’t sugarcoat anything. It’s their body and they’re the ones who have to go through it.”
Jeff. Open communication about treatment helped Aidan in the past to cope with and prepare for interventions: “We’ve always been very upfront with him about the different treatment options that he’s had. We always let him know--we never promise that it’s not going to hurt but we try to explain the benefits and things and in the past that’s worked.” Although Jeff still tries to communicate in this way, he said, “You can’t force him to do anything now that he’s older and bigger.”

Francine. Whether discussing medical treatment or social concerns, Francine tries to be open and direct in her communication with her kids. She offers problem solving about ways to manage social concerns, which she says her daughter sometimes accepts and sometimes does not. She tries to offer realistic assessment of what to expect of surgeries as well as to give them perspective and support: “Sometimes there’s going to be pain but we’re going to make choices that are always good for you and I tell them I’ll be right there with you.”

Jan. Jan said regarding decision-making and preparing for medical treatments, “We discuss it…she really listens to us. We talk frankly with her about the pros and cons.”

Social Acceptance Is a Concern for Parents

All but three of the parents interviewed described having significant concerns about social acceptance for their children during their early childhood. Only one parent expressed ongoing concern about negative judgment and lack of acceptance at this stage of development. Those parents who were not concerned described positive experiences with peers and their own children’s adjustment and outlook as the reasons for their changed perspective. One adolescent mentioned being aware that others noticed their
appearance and felt frustrated by this but attributed it to ignorance rather than ill will.

One adolescent described herself as still learning to cope with the negative judgements of others and one adolescent clearly struggled to feel accepted by her peers.

**Paula.** Paula said when Kristin started school, she worried about her being teased and bullied and made fun of but then she got to school and it was such a non-issue. But before she got to school, I worried a lot about it. Are they going to be mean, are they going to tease her, are they going to point at her? Never came to be.

**Anne and Lisa.** Immediately after Anne’s birth, Lisa felt acceptance for her daughter’s appearance but feared others would not: “I was proud of her, but I was torn about what to do…do I want people to come or not because I don’t want them to see her and be like ‘ew’ and not accept her. I couldn’t take that. I want them to love and accept her.”

Lisa acknowledged Anne struggled to feel accepted by others her age. She recently joined an online support group for teenagers born with cleft conditions. After chatting with a boy on this website, she made the comment to her mom that maybe they could get married, saying, “He couldn’t judge me because he’d be just like me”

Reflecting on this, Lisa stated she thought her daughter needed to have friends who could relate to her and with whom she could find acceptance and support:

“I know what’s it like to be a parent to a cleft child but I don’t know what it’s like to have a cleft. I don’t know what that feels like physically or emotionally...she needs another child that has a cleft that she can talk to—“I’m scared about this surgery” or “I’m being treated this way.” They need that from each other.

As a result of this conversation, Lisa was looking for ways to increase Anne’s contact with others with cleft conditions and considered taking her to a conference in a neighboring state. Even as far back as kindergarten, Anne remembers being treated
differently and people not understanding her. “It was the hardest thing,” she said. People have made fun of her, said she was different, and questioned her about her experiences. The judgement feels ongoing: “When I go places like when I go out to eat there’s always that one person who stares at me but you can’t let that bother you because they don’t understand.”

Janet. Janet does not have concerns for Frank’s social acceptance: “In the big picture…he’s a normal kid. Nothings different other than the obvious appearance of a lip scar… he plays sports, he has an active social life, he has a girlfriend, he’s a normal 16-year-old kid.” He also does well in school. She is aware of minor social incidents but says both of her sons handle them on their own. She attributes part of his adjustment to having a longstanding and positive peer group: “Some of them have been together since preschool. They’ve been very protective of him and accepting of him. Their parents have made sure they’re protective and accepting.”

Jennifer and Francine. Jennifer wearily discussed her frustration with reactions to her appearance from other kids at school. She believes their reactions are based on ignorance rather than ill will but the process is still trying for her: “A lot of kids at my school have asked me about it for sure…it’s really hard explaining…sometimes I explain it and sometimes I don’t sometimes cause I just don’t feel like explaining it.” Francine is aware that socially Francine’s condition can cause her frustration. Sometimes children ask “stupid” questions, which makes her angry, and “affects her attitude. It makes her self-conscious.” Francine is concerned about her self-esteem although she has never spoken about this to her mother: “She seems fine but I know she’s kind of grumpy and angry sometimes.” Jennifer is aware of behavior that points toward low self-worth: “She
seems under confident in like saying her reported or speaking out loud or speaking up for herself.” Francine attributes her behavior as much to the repetitive treatments she has undergone as to her appearance: “She’s had so many more experiences than the normal child it effects how she experiences herself.”

**Margaret.** While Margaret says she is still impacted by some insecurity, she is “able to deal with it better” the older she becomes. She compares herself now to when she was in elementary school and says, “It made me upset at the time…I realize they didn’t mean it to be mean. I’m definitely more confident now. I used to be self-conscious about it. I’ve learned to deal with it.”

**Social Advocacy Seen as Key by Parents**

Four of the six parents described taking a proactive approach to educating and advocating for their children among their peers, starting in elementary school. The other parents did not specifically mention doing any specific advocacy on behalf of their children.

**Lisa.** Lisa feared teasing and bullying or negative judgment for her daughter from the time she was an infant. She made a decision be proactive with education and advocacy in her daughter’s school beginning in kindergarten:

The very first day I went in and I wanted to talk to the class. I just said you know you’re going to notice several different things about Anne and I wanted to talk to you about those differences…I said “you might notice that she has a scar on her lip, how many of you have scars” and every hand shot up. So it made it not different and scary, because everybody could relate to having a scar.

Lisa explained the treatments Anne had been through and how brave she had been to the children. Lisa felt the children who understood and accepted Anne from an early age continued to support her in later years. While she remained in the same school
system, this early acceptance and understanding contributed to her social success. When Anne was bullied, Lisa advocated for her within the school system and also took steps to have her seen professionally by a primary care doctor who prescribed anti-depressants.

**Paula and Kristin.** Paula taught Kristin how to advocate for herself and educate others about her condition from a young age. She attributed this skill to her daughter’s successful social adjustment: “If you teach your child to educate their peers at a young age, then it becomes a non-issue as they get older.” Kristin’s first presentation to her class regarding her condition and treatments took place when she was in the first grade. Kristin was also prepared to answer awkward questions with direct, factual information in an assertive manner. Although she has occasionally experienced negativity from peers, her ability to advocate for herself, “she lets it rip,” contributed to her ongoing ability to cope. Kristin’s parents were also proactive in educating peers with the goal of increased support and understanding: “We sent pictures from the hospital to the classroom so the teacher could show the class this is what Kristin is looking like today so when she comes back you can’t run up to her and huge her or poke her in the face. You have to be careful.” She said children who had already experienced this once were more likely to speak up for Kristin during the next surgery and educate children less familiar with her condition.

**Jessie.** Ian had hearing aids when he began kindergarten and Jessie read a book to the class about his condition. Ian later requested she come in and read a book about his cleft after it came to their awareness that he was upset by people asking him about it all the time. When she told Ian she could not find one, he asked her to write one: “So we
sat down together and wrote a book.” The whole family wrote the book together, which Jessie described as a “work of love.” It was later published and sells internationally.

**Janet.** Janet was very involved in both of her children’s schools and coaching their sports teams. She also taught for a while in the same preschool as Frank but not the same classroom. This allowed her to intervene if needed and to meet other parents and develop relationships. Janet felt protective of Frank and concerned about his acceptance: “I think I needed to because I didn’t trust people.” Some of her concern related to his speech:

> When Frank was in preschool, he couldn’t be understood very well so I had to translate for people at times…not so much of a necessity but to make sure that…I didn’t want him to have to struggle and I didn’t want people to struggle to try to understand him either.

Janet gradually lessened her involvement as she witnessed Frank’s independence and growing relationships with others outside of the family.

**Bullying due to facial appearance and speech ability.** Two families out of eight reported their children were bullied because of their appearance and/or speech.

**Anne and Lisa.** When Anne was in seventh grade, she switched middle schools to receive academic accommodations that had been denied at her school. In this new school, she was bullied by another girl. Lisa stated that being the newcomer probably contributed to being bullied: “It might be when you’re outsider coming in you’re easy prey.” Despite Lisa’s efforts to intervene in the school, the bullying continued and they returned to Anne’s original school. Anne does not know the extent or nature of the bullying: “To this day, she won’t tell me what she said or what she did.” But the impact was significant and immediate: “It completely changed her. I don’t know…what was done to her at school but she never said anything to me about it…she really is fighting
anxiety and depression.” Anne stated she felt she was working through this experience with the help of friends in her church.

Jeff. Jeff said Aidan was bullied in middle school because of his appearance and speech. It had a profound impact on his son and his behavior:

When Aidan was in elementary school, he was the happiest little kid you’ve ever seen, was the light of the party, he would walk into a room and he would light up everybody knew his name and everybody loved him at school and then somewhere in middle school that changed. I think part of it had to do with he was getting bullied because of his appearance and his speech and things went downhill from there.

The family never knew the extent of the bullying and Aidan still would not talk about it: “I say he lost his smile. Up to that point he always had a smile on his face and after that he very seldom had a smile on his face.”

**Summary**

In this chapter, the themes provided by the participants were thoroughly described to present how the condition of cleft lip and palate and the medical treatment process for it impacted adolescents and parents of the adolescents and how they coped with those impacts. The data were collected during individual interviews in which the participants described their experiences. The uniqueness of individuals and families was presented both in the themes and in the contextual and background descriptions for each participant.
CHAPTER V

CONCLUSION AND DISCUSSION

In the previous chapter, I described the themes of each parent and adolescent participant. The thematic findings of this study were obtained following individual interviews. An analysis of the data included reviewing transcriptions, thematic coding, and member and expert checks. Twelve individual participants were interviewed including four parent/adolescent dyads, one individual adolescent, and three individual parents. All participants provided his/her unique perspective on the impact of living with the condition of CL/P, undergoing treatment for it, and mechanisms utilized for coping with both the condition and its treatment. In this chapter, the research questions and the purpose of this study are reviewed. A summary of the findings in the context of previous research is presented as well as limitations of the study and implications for future research.

**Purpose of the Study**

The condition of CL/P commonly impacts facial appearance and can impact speech and hearing as well as cognitive functioning. These differences in functional and aesthetic abilities place individuals with this condition at risk for psychosocial adjustment difficulties. Many studies have found a percentage of individuals with CL/P are at risk for difficulties such as social acceptance, behavioral problems, and/or emotional distress (Hunt et al., 2005). Individuals born with CL/P often undergo medical treatment for the
condition from infancy through late adolescence if not throughout their lifespan. The burden of medical care impacts not only the individual child but the entire family system (Baker et al., 2009). The treatment process places demands of time, energy and financial burden on many families (Kramer et al., 2007). Healthy family functioning positively impacts children’s abilities to adapt to the stress associated with chronic health concerns (Crerand et al., 2015). Therefore, research on families and coping styles within this population was warranted.

Research on risk factors and the adjustment process to the condition of CL/P has found highly inconsistent results due in part to the inconsistency of research designs and measures utilized and to the multifactorial nature of the adjustment process itself (Rumsey & Stock, 2013). Research on resilience provides a framework through which to understand the adjustment process. The general literature on resilience noted individuals often simultaneously experienced risk in some areas and resilience in others (Luthar, 2006). The concept of family resilience examined dynamic processes between family members as sources of strength in addition to individual traits (Luthar et al., 2000). Identifying the risk areas within this population remains of critical importance to provide the support needed (Feragen, Stock, & Kvalem, 2015; Stock et al., 2016).

The concept of resilience is a relative one dependent upon culture and context (Unger, 2003). Because of this, several authors have argued that qualitative research with its focus on complexity, contextual factors, and emergent designs is best suited to explore this area (Eiserman, 2001; Unger, 2003). Many authors noted the subjective experience of individuals who live with the condition is often lacking (Feragen et al., 2015; Nelson et al., 2013). In addition, the voices and experiences of children and adolescents have
been largely absent from the research on this population (Hunt et al., 2005). This study aimed to understand the experiences of both parents and adolescents with the impact of the condition as well as its treatment and to understand how both individual and family coping strategies contributed to successful adjustment. Patient perspectives are the foundation of patient centered care; therefore, it is my hope the results of this study might support medical providers who work with this population (Canady, 1995). This study utilized a qualitative design and phenomenological theoretical framework. It was guided by the following research questions.

Q1  What is the essence of the experience of parents and adolescents with the process of CL/P treatment?

Q2  How do parents perceive that their children are impacted by CL/P treatment?

Q3  How do adolescents experience parental support in relationship to their medical treatment?

Q4  What are the coping strategies that parents and adolescents think that they have developed in response to CL/P and its treatment?

**Discussion of the Findings**

In the previous chapter, I provided detailed description of the participants’ themes. In this chapter, I discuss the themes in terms of the research questions and integrate them within the context of previous literature.

Any discussion of the impact of the condition of CL/P must include the medical treatment for it, both because it is so consuming and because the outcomes directly and considerably impact quality of life in every foreseeable way. In this study, both adolescent and adult participants acknowledged the treatment process for CL/P was extensive and time consuming. The medical treatment for cleft lip and palate begins in
infancy, or even prior to that for parents, and ends in early adulthood. One recent study noted this journey does not actually end with late adolescence although coordinated medical support often does (Stock et al., 2015). Parents in this study reported impacts on the entire family system and none felt prepared for the quantity and intensity of treatment required. Adolescents reported chronic medical care being a normal experience for them and having strong emotional reactions to it.

The medical treatment as experienced by patients and their families was characterized by uncertainty, both about the practicalities of the timing and nature of interventions and what their impact would be physically, emotionally, and functionally. In the United States, surgical treatment for CL/P is highly varied with no standardized protocol for treatment, making the results also highly variable (Grollemund et al., 2012). In the current study, some participants experienced anxiety, frustration, and anger in response to this uncertainty; others found more acceptance and tolerance for the process. Why some individuals coped with this uncertainty better than others is critical to understand as is what could be done by institutions serving these individuals to mitigate the level of uncertainty experienced.

All of the adolescents in this study had undergone revision surgeries for primary repairs. Participants reported between 6 and 15 surgeries, whereas the number of primary repairs including repair of the lip, palate, and bone graft is typically five or less. Surgical repairs are frequently unsuccessful either in terms of the goal for aesthetic outcome or functionality. While the number of revision surgeries has not been researched extensively, a growing number of authors have noted the burden of repeated medical intervention might not warrant the outcomes for patients (Sitzman, Coyne, & Britto,
Systematic reviews of revision rates found such variations: “it is not possible to estimate the burden of care” for patients (Sitzman et al., 2016, p. 92). One study noted the average number of surgeries per individual in one hospital between the ages of birth and 21 was eight (McIntyre et al., 2016). Sitzman et al. (2016) noted the burden of care (number and intensity of surgeries) should be warranted by the final aesthetic outcome; yet research showed revisions were often costly in terms of time, finances, and decreased satisfaction (Trotman et al., 2007). Based on the findings of the current study, it would seem the need for revision surgery was not the exception but the norm.

Despite the surgical interventions, several participants still experienced ongoing health problems including breathing difficulties, sleep apnea, and bite concerns. Some but not all participants mentioned a desire for further aesthetic surgery to improve appearance. The literature also noted that despite surgical repairs, individuals with cleft conditions often continued to experience functional difficulties with eating, talking, and breathing (Kasten et al., 2008). All participants had experienced or were undergoing significant orthodontic work; this was similar to the literature that stated orthodontic work began at age five and continued through late adolescence (Kasten et al., 2008). While none of the participants reported continued speech problems, all of them had undergone speech therapy in their past, which was common to this population (Kasten et al., 2008).

Factors involved in variations of revision surgery included quality of initial repair, preference for revision surgery by the surgeon, and differences in the treatment protocol (Semb et al., 2005). Socio-economic status, health access, and insurance availability might have also influenced whether families were offered revision surgeries (Cassell,
Studies have shown the incidence of surgical revision could be reduced by the improved quality of primary repairs, yet few surgeons employed published techniques guaranteeing improvement (Mulliken & Martinez-Pérez, 1999; Sitzman, Girotto, & Marcus, 2008; Sitzman et al., 2016). While adjustment to the realities of the need for medical care and the potential for functional complications and some degree of physical difference are realistic goals for this population, adjustment to substandard surgical interventions should not be.

While the level of uncertainty in both expectations and outcomes was clearly communicated well by some providers, this was not consistently the case. Many participants reported successful and adequate communication with their providers while several felt the need for further communication. A great deal of this communication had to do with expectations. For example, four parents expressed a desire for better communication regarding the course of treatment, treatment outcomes, and postoperative recovery. One parent stated she felt “cheated” by the lack of information regarding the real extent of the medical process. One parent specifically mentioned the lack of clear information made parenting a challenge as she could not properly inform her child about the future. Another parent, although reporting successful surgeries, mentioned many complications and revisions were needed and expressed that more information regarding the possibility of set-backs would have been helpful. The goal for these parents was to better prepare their children and reduce their suffering. Because each new intervention was unique, parents experienced a continuous need for information (Nelson et al., 2013). They also needed realistic prognoses regarding success in both functionality and aesthetic outcomes.
Parents who felt the inherent uncertainty or difficulty of the process had been adequately explained to them had more tolerance for setbacks and variations to the protocol. Therefore, two parents actually reported preferring or being satisfied with less specific information because they felt it better reflected the inherent uncertainty of the treatment process. They considered this less burdensome to the family. Two parents who acknowledged what they considered more than the average number of complications or revision surgeries also stated they had been warned about this likelihood early on and, therefore, did not express dissatisfaction with information provided. Shared communication and a feeling of being involved in the medical process has been linked to positive coping for parents (Nelson, Caress et al., 2012). Eiserman (2001) found parents desired to be fully informed about the medical process in order to make better decisions for their children. Yet several studies found parents felt relatively uninvolved in the treatment process for this condition (Jeffrey & Boorman, 2001; Turner et al., 1997).

In addition to the need for increased information, three families also reported the need for increased emotional support from the cranio-facial team. While studies have shown parents consistently requested psychological support from their medical teams and medical and psychological organizations in several countries have made recommendations that mental health providers be a core part of the cranio-facial treatment team, such support is still relatively rare (Colbert, Green, Brennan, & Mercer, 2015; Johansson & Ringsberg, 2004; Kuttenberger et al., 2010). Only two families were offered regular mental health check-ins during their yearly team visits. Two sought mental health treatments outside of their medical team. Other parents did not specifically express a need for emotional support from the team but described having such needs met
through support groups. Most parents did not identify mental health services as an answer to that need nor did they consider mental health needs to be within the scope of their cranio-facial teams.

Parents felt anxiety and wanted reassurance about the implications of treatment and the condition on their children's quality of life. Parental coping involved providing assurance their children would be well cared for. One parent expressed a desire for reassurance the family would be okay, that it was possible to cope with the process, and the child would find acceptance and a normal life. Another parent mentioned the desire to understand and be reassured about aspects of her child’s experience given her physiological differences. Another parent expressed a desire that the medical team be gentle and compassionate with her children. Research noted parents’ satisfaction with care in part hinged on how much medical providers eased the process for their children (Nelson et al., 2013).

Parents in this study were aware of their adolescents’ physical pain and anxiety in relation to their treatment experiences. Despite this, some adolescents experienced a sense of isolation with these very emotional experiences. Some stated their parents just could not understand the extent of their emotions and most felt the physicians were unaware of the emotional impact of the treatment process. They expressed a belief that treatment could be better informed through an understanding of their pain, fear, and uncertainty. One expressed her parents minimized the impact of surgeries and recoveries. And three adolescents specifically stated needs for either peer support or psychological support in addressing their feelings of isolation and uncertainty.
Despite the challenges and stated needs of these participants, every parent and most adolescents in this study expressed positive relationships and overall satisfaction with treatment. Research demonstrated parents’ satisfaction with their cranio-facial specialists was overwhelmingly positive (Nelson, Glenny et al., 2012). Only one family changed teams due to dissatisfaction with care. Three families experienced unexpected medical outcomes complications with post-operative care due to poorly managed pain yet remained trusting in the expertise of their medical teams. Every family interviewed mentioned having to repeat a surgery and often believed this was exceptional to their experience and their child’s condition. Theorists have speculated reasons for such positive appraisal of medical providers. A heightened sense of trust in the medical providers might allow parents to forgive or overlook bad experiences and a reliance on surgical interventions could serve as a means of coping with anxiety and helplessness (Nelson, Caress et al., 2012, 2013).

Families in this study stated their trust in providers was due to their expertise, care and concern, their availability, and the length of the relationship. The duration and quality of these relationships led parents to describe their providers as extended family. Two adolescents also reflected feeling a closeness and warmth toward their providers. One adolescent was very matter of fact about her relationship with her doctors; nonetheless, she stated she had great trust in their expertise despite experiencing frustration with many aspects of her care.

Research has shown providers’ expertise and continuity of care contributed to a sense of trust (Canady et al., 1997). The dependency on providers parents feel has been linked to the idea of obligatory trust (Carnevale, 2004). This trust is natural given the
needs and level of expertise on cranio-facial teams; however, this trust should not be blind but ideally formed through adequate communication that allows for realistic assessment of expertise.

While decision-making plays a large role in the management of this condition given the variability of treatment options, survey results indicated parents of children with CL/P felt relatively uninvolved in decision-making (Pannbacker & Scheuerle, 1993). All families chose their medical teams either through recommendation of family or medical doctors or for close proximity. Only one family drove a distance to their medical team and only one family chose a team based on an understanding of an aspect of the treatment protocol. All families utilized large craniofacial teams except one. This family utilized a large team until a move required them to find independent providers. One family left a team due to dissatisfaction with pain management and two switched teams due to geographical moves. Research on parental medical decision-making in general found decision-making was highly dependent on context and difficult to predict (Knopf, Hornung, Slap, DeVellis, & Britto, 2008).

All parents but one felt comfortable in allowing their medical team to make decisions and other than two who questioned recovery management, none expressed disappointment with any aspect of the treatment protocol or its outcomes. Only one parent declined a surgery in elementary school and opted to wait for this treatment until adolescence to avoid duplicating the procedure. Other parents were frustrated by setbacks, changes in timing, or types of surgeries but viewed this as a communication problem rather than one of planning or protocol.
Previous research suggested parents’ reliance on their medical teams’ decision-making might relieve them of the burden of this emotional responsibility; parents might be overly reliant on following treatment advice in order to feel they are responsibly parenting their child (Nelson, Caress et al., 2012). While parents might wish to do their utmost medically and therefore rely heavily on medical treatment, the fact that families reported a lack of awareness of many aspects of treatment until they were informed by other parents points to a problematic lack of communication with medical providers.

In adolescence and even before, many of the surgeries for CL/P are elective and done for aesthetic reasons rather than for critical life functions. These surgeries involve a different type of decision-making; they are influenced by emotional and cultural factors—to help the child achieve a more “normal” appearance (Daniel, Kent, Binney, & Pagdin, 2005). Research has shown parents typically do not challenge societal norms when it comes to appearance in terms of acceptance of treatment protocols (Nelson, Carress et al., 2012; Sanders, Carter, & Goodacre, 2007). Yet numerous studies have found parents do challenge this notion of normality internally; they fully accept their infants prior to repairs and they grieve the loss of their natural appearance (Nelson, Glenny et al., 2012; Stock & Rumsey, 2015). The result can be an internal struggle between acceptance of one’s child for who they are and recognition that societal expectations require significant surgical changes (Nelson, Caress et al., 2012).

Two adolescents expressed significant hope for future surgeries and were impatient to be completed with the protocol. Both of these stated they wanted to be better and both had experienced some stigmatizing experiences. Research has linked stigmatizing experiences to a desire for further treatment (Alansari et al, 2013). Two
families had or were requesting further revision surgeries outside of the planned protocol, both with the aim of improving aesthetic appearance. Those who had planned revision surgeries in their near future were anxious and hopeful these would create further change and discussed recognizing a need to be “fixed.” The practice of “normalization” is complex as it is fueled by societal expectations of attractiveness and might not be the most efficient route to adaptation and adjustment. Yet adolescents in the present study described their hope of being “fixed” as enabling them to endure medical interventions. The only adolescent who had completed the surgical protocol expressed the outcomes were not worth the pain and inconvenience of the medical process. She expressed more frustration with the constant assumption that something was wrong with her than with the burdens of the condition itself. A recent article found that following an aesthetic surgery, most individuals experienced a period of stress followed by a return to normal levels of stress; in general, participants were happy with the outcomes (Albers, Reichelt, Nolst-Trenité, & Menger, 2016). However, another recent study of adults who underwent additional aesthetic surgeries noted several felt regret (Stock et al., 2015). Clearly there are individual variations in these reactions as well as surgical variations causing differing outcomes. What would help patients make more informed decisions regarding these potentials is more informed communication and patient centered care.

While parents expressed the desire to allow their children to make decisions, the degree to which this autonomy was actually given varied among participants. One adolescent was extremely appreciative of her parent’s support in making her own decisions. One mother described a strong desire for her son to undergo an elective surgery she felt would improve his appearance. She had hoped his doctor would discuss
treatment options for him but he had not. Research found tension about these topics hindered open communication between parents and their children’s medical providers (Silverman, 1983). In one dyad, the parent stated her daughter could make decisions, while her daughter felt her parents made choices without her input. One parent recognized he did not have the power to influence his son’s decisions at his age. Most adolescents stated acceptance that their parents and the team made the decisions for them. Only one adolescent mentioned frustration with her parents persuading her to accept treatment she did not really want or feel she needed.

Several adolescents described a complex sense of identity with regard to their medical condition. Their identity was strongly influenced by their facial appearance and the opinion others had of their facial differences. They described knowing others perceived them to be different and had adjusted to this fact. This adjustment evolved through a process involving understanding that others’ perceptions were based on a lack of understanding and growing self-acceptance. Part of their ability to accept themselves hinged on a sense of the relative lack of importance in outward appearance. Crerand et al. (2017) found adolescents with CL/P were less invested in the importance of appearance than others their age. Parents in the current study who had encouraged self-acceptance and perceived these efforts to be helpful also noted their children exhibited a strong sense of individuality.

Identity was connected to the progress of medical treatment as well as social interactions. None of the participants expressed difficulty or unhappiness with regard to this sense of identity currently, although they recognized periods when this was more difficult due to social interactions, age, or stigmatizing experiences. Those who had
planned revision surgeries in their near future were anxious and hopeful these would create further change, whereas the one adolescent who had completed the surgical protocol expressed the outcomes were not worth the pain and inconvenience of the medical process. She expressed more frustration with the constant assumption that something was wrong with her than with the burdens of the condition itself. Some noted societal expectations and stigma played a large role in treatment protocols and others’ perceptions of them (Mouradian et al., 2006).

Studies have repeatedly demonstrated that adolescents with CL/P fare well on measures of appearance and self-acceptance (Pope et al., 2016). Authors concluded the reason for this was either the presence of resilience and successful adjustment or the product of overcompensation. One author posited it was only through avoidance of socializing that adolescents maintained a positive self-concept (Kapp-Simon et al., 1992). Others have considered this adaptation a matter of minimizing and self-protection (Crerand et al., 2017). The concept of self-presentation posits cognitive and behavioral strategies assist individuals in maintaining a positive self-concept in the face of lack of social acceptance (Thompson & Kent, 2001). It is also well known that patient satisfaction with appearance and outcomes has no association with the amount of treatment or any objective measure of aesthetic normalization (Semb et al., 2005). This points to some internal process that is as important as are outward appearances in the process of developing a healthy self-concept and identity, although this process is not necessarily one involving denial or minimization. More specific understanding is needed on the development of self-acceptance with regard to the course of treatment for CL/P.
The emotional impact of the treatment process for CL/P has been well documented in numerous studies (Collett & Speltz, 2007; Hunt et al., 2005). Parents in this study experienced guilt, sadness, and anxiety with regard to their children’s condition and medical treatments. They discussed difficulty balancing their emotional reactions with the role of being a parent and caretaker. They also discussed some anxiety about their children’s futures, acceptance, and self-esteem. Two families whose children were impacted by mental health concerns experienced anxiety and frustration about their children’s level of psychosocial stress. It is noteworthy that four of the adopted children in this study were abandoned by their biological parents due to their condition. While not participants in the study, the emotional reactions of these parents were part of the story and a potential for many parents who face the challenges inherent in this condition.

Literature found the type of concerns parents had for their children with CL/P was relatively stable over time (Lei et al., 2010). In this study, the anxiety and sadness parents felt watching their children undergo painful treatment remained stable over time. All parents expressed a degree of anxiety with regard to their child’s medical interventions or overall functioning. One parent expressed increased anxiety as the child matured given their increased understanding of the procedures. Even two parents who acknowledged their children coped well with surgeries stated they still felt anxiety as parents. In contrast, parents’ concerns about social acceptance abated over time unless their children were experiencing bullying or mistreatment.

Adolescents’ emotional reactions included fear of the outcomes and of pain, frustration at the process and at poor communication, and gratitude about positive changes. Difficult social interactions caused a range of emotions—from frustration and
annoyance, to insecurity or self-consciousness, and sadness. Two adolescents reported anxiety prior to treatment both with regard to the procedure and recovery and to the uncertainty about the outcome of treatment. Pain and debilitation were worse than expected by two adolescents. Two adolescents experienced anxiety and impatience to be finished with surgeries to have better outcomes both aesthetically and functionally.

It has often been noted that parents and adolescents’ perspectives on coping and emotional impact differ from one another. Authors suggested parents might be overly concerned about their adolescents or adolescents might be self-protectively minimizing their reactions. In this study, there was some agreement between parents and adolescents as well as some discrepancies. Parents were aware of pain and anxiety when their children were vocal about this. For example, one mother who perceived her child to have a high pain tolerance not reported by her daughter also reported her daughter’s response to surgery was to isolate and stop communicating. Another mother stated more concern about her daughter’s anxiety and depression than she herself noted; again, this adolescent stated a preference to speak with friends about her emotions than to her parents.

One emotion not clearly understood or perceived by parents was their adolescent’s ambivalence toward treatment. None of the parents expressed any ambivalence with regard to treatment protocols or outcomes. It was noted the parents rarely questioned treatment recommendations as a means of coping and might have overly relied on medical intervention (Nelson, Caress et al., 2012). Two adolescents experienced complex emotions in response to their medical treatment and providers. One expressed anger and resentment about poor communication and pain management but also acknowledged the surgical interventions resulted in improvements in her appearance
and functionality. Another experienced both wanting a surgery that would improve her appearance but also feared drastic change. When the surgery did not have a significant impact, she then felt it had not been worth the effort. Alansari et al. (2013) found some patients with CL/P at the end of the treatment negatively compared the process to the outcomes. Both of these adolescents appeared to struggle with reconciling the outcomes with the significant costs of undergoing treatment. The latter one stated the sense that something was always needing to be fixed was a burden to her. In contrast, none of the parents expressed ambivalence with regard to treatment protocols or outcomes, perhaps due to over-reliance on medical intervention as a means of coping (Nelson, Caress et al., 2012).

Resilience can be understood as the process through which individuals or families cope well with adversity. Understanding factors and strategies of coping for individuals and families with CL/P is critical and can help inform intervention and prevention strategies. Several factors were identified by participants as assisting them in not only adjusting but at times thriving with the condition of CL/P.

Both parents and adolescents noted the importance of social support in coping with the treatment burden and their emotional reactions to it. Baker et al. (2009) found social support was one of the main coping skills mentioned by parents of children with CL/P. Research on resilience also noted the role social support plays in helping children adjust to risk factors (Luthar, 2006). Practical support in managing caretaking needs came from extended family as well as older siblings. Emotional support came from friends who offered acceptance of their children’s condition as well as guidance or perspectives in coping with challenges related to it. Coping required developing
understanding and acceptance of their child's situation, which was often facilitated by the advice of others. Authors noted changes in personal beliefs often accompanied parents’ efforts to be the best parent and support they could be for their child (Eiserman, 2001).

Adolescents described support in managing expectations and understanding of treatment interventions, physical recovery, and emotional responses to chronic medical care and social difficulties. Four adolescents noted their parents offered guidance and emotional support as well as caretaking throughout the treatment process. Parents validated their experiences, which increased self-acceptance. Social support and family acceptance are critical in positive adjustment to disfiguring conditions (Thompson & Kent, 2001). Parents also offered problem solving regarding concerns presented by their children such as social problems and treatment options. They provided perspective by helping their children manage concerns with low self-esteem and self-confidence and managing medical treatment. One adolescent stated a preference for receiving emotional support from friends in dealing with bullying and self-esteem. Family relationships were enhanced through the treatment process for one adolescent. Research on cleft conditions and chronic illness in general found improved relationships could be both an outcome of this process and served as a protective factor for adolescents with cleft conditions (Eiserman, 2001; Hastings et al., 2002; Hutchinson et al., 2011).

Parents reported methods of providing support that matched their adolescent’s perceptions of this support. They provided post-operative care-taking, tried to be informed about treatment and encouraged self-advocacy, offered choices surrounding medical care when possible, maximized normality through the use of familiar routines
during stressful periods, provided acceptance and support, and advocated for a positive environment at home and school.

A majority of families noted the central importance of involvement in community support or advocacy groups. The needs for emotional support, validation, and information in this population are noteworthy. Two parents formed their own peer support groups. One parent created a parent support network for parents of newborn infants and two parents joined large community advocacy groups involved in support, education, and fund raising. Another served as a parent consultant for craniofacial teams. And one adolescent founded a nonprofit organization for families. In addition to emotional support and information, other benefits included generating community, giving back, and increased family cohesion. The support and advice of other parents was reassuring and filled gaps left by the medical community (McCorkell et al., 2012). Increased empathy and understanding leads to a desire to give back and is common with individuals with chronic health conditions (Stock & Rumsey, 2015).

Interestingly, parents had not sought out similar support groups specifically for cleft conditions for their adolescents, although they advocated for them with their peers at school. Two parents mentioned having siblings with cleft conditions or other medical conditions, which helped their children feel less isolated and alienated. One adolescent stated a current desire for more peer interaction and support, which her mother was actively seeking for her. Another adolescent stated she would have liked to have been part of a support group, feeling it would have helped her feel less alone and provided hope going through the medical process.
All but three of the parents had significant concerns about their children’s social acceptance from the time their children were very young. This was cited as a major concern for this population given the known role physical attractiveness plays in societal values and acceptance (Nelson, Glenny et al., 2012). Whether this concern is warranted, however, is unclear based upon the results of multiple studies examining the nature of social adjustment in this population. While lack of social competence has been associated with reduced friendships and more rejection among adolescents with CL/P, other studies found no significant difference in social competency in this population compared to norm groups (Collett et al., 2012; Pope & Ward, 1997). Self-acceptance in terms of satisfaction with appearance is not related to objective understandings of attractiveness (Feragen et al., 2009). Rather, it involves a complex interplay of factors including self-acceptance, stigma, social competence, and appearance (Pope et al., 2016). Stock et al. (2015) noted that earlier research pointed toward greater difficulty in educational achievement, career development, and intimate adult relationships in connection to poor social adjustment. Yet their research found adults with CL/P reported successful work and relationship adjustment including committed relationships and parenting (Stock et al., 2015).

Despite early concerns, only one parent expressed ongoing concern about their child’s adjustment based upon the adolescent’s stated fear of rejection. The remainder noted positive and plentiful peer interactions. One parent denied any negative social experiences. One mentioned minor comments that were handled by the child and his brother and did not cause any negative impact. Two parents mentioned peers who asked questions and made occasional comments, which caused frustration and irritation. For
these adolescents, the theory that self-acceptance comes at the expense of social interaction seemed inadequate. The adolescents in this study who expressed positive self-acceptance also expressed realistic assessments of their differences in appearance and how these were likely to be perceived. They did not appear to be either minimizing or discounting that such interactions were a likely response in our culture.

Interestingly, two parents who reported their adolescents having social anxiety did not associate this with the CL/P. The children of these two parents corroborated these impacts; both stated that as they grew older, they were less negatively impacted by these types of comments because they could better understand their origin lay in ignorance rather than ill will. One parent stated their child had withdrawn from face-to-face social interaction although this adolescent likely had comorbid mental health concerns in addition to negative experiences with social peers due to the CL/P.

Two adolescents experienced bullying. For both, the bullying occurred in middle school, which was consistent with a study on this population that found the most common period for bullying was early adolescence (Semb et al., 2005). Both families thought the bullying was related to their children’s appearance and possibly speech but did not know the exact nature due to their children’s refusal to discuss the events. One parent intervened rapidly, involved the school, and sought mental health support for her child in the community. The other family was less aware of the situation until sometime after it had occurred but sought mental health support years later. Both parents noted a significant and almost immediate impact of the bullying on their children’s mood, self-confidence, and behavior. Bullying is a significant predictor of poor self-concept (Hunt et al., 2006; Murray et al., 2010). Bullying can lead to social anxiety, social avoidance,
and negative self-development (Masnari et al., 2013; Rumsey et al., 2003). The adolescent with more significant and comorbid mental health concerns still struggles with behavioral problems, mood swings, and social interaction. While research supports that adolescents with CL/P are at higher risk for both externalizing behaviors and internalizing behaviors including non-compliance, oppositional behavior, and depression and anxiety, the etiology of these problems remains unclear (Richman et al., 2012). If social rejection plays a role in this, then why do some adolescents manage to navigate this and others do not? One adolescent reacted to bullying with a drastic decline in behavior and mood; it was unclear whether this was solely the result of bullying or whether a comorbid condition contributed. The other adolescent reported she felt she had worked through the impact and was even grateful for lessons learned through it.

Four families described taking an active approach in relieving their anxiety regarding their child’s acceptance among peers. Three parents presented information to their children’s elementary school classes about the condition of CL/P and their own child’s particular experiences including updates about surgical interventions taking place during school. One parent taught her daughter to present the information as well as to answer questions or comments assertively. Another parent supported her child’s social development by being highly involved in both his school and sports activities. Research also demonstrated that parents of children with CL/P took steps to encourage active involvement with peers (Klein et al., 2010). The same parent stated she felt the need to be involved due to not trusting others’ reactions to her son. Some evidence indicated over-protectiveness might have served as a buffer for negative reactions in this population (Hutchinson et al., 2011). All of the parents in this study felt their advocacy
had positively influenced their children’s success within their peer group. The parents’ perceptions of their child’s successful adjustment aligned with research that pointed toward positive peer support contributing to positive self-perception and resilience among adolescents with CL/P (Tiemens et al., 2013). Crerand et al. (2017) found adolescents with CL/P were less invested in the importance of appearance than others their age. Several parents actively worked to foster and encourage self-acceptance in their children. While the adolescents described had some sense of self-acceptance for their appearance, they also described this relative sense of acceptance was impacted by surgical outcomes, recovery periods, and social interactions. Self-acceptance appeared to be a chronic and periodic stressor in this population. In addition to the stressors of medical treatment and changes in appearance, stigma might play a role in the struggle for self-acceptance.

The development of new strengths and abilities as a result of stressful situations is a well-known phenomenon (Zimmerman & Arunkumar, 1994). Two parents and two adolescents described such personal growth with regard to the treatment process and condition of CL/P. One parent described having increased empathy for others and an increased perspective on the different types of struggles people face. Another parent described gaining acceptance of facial differences and changing her perception of beauty. She and her daughter both mentioned positive perspectives gained as a result of these experiences. One adolescent described her own experiences and her work with a non-profit as benefiting her perspective and growth. She viewed her condition as a blessing because it allowed her to do this work, through which she gained such perspective.

Two parents discussed spirituality as a coping mechanism. One mother’s religious faith provided a perspective for her to understand and assist her daughter in
understanding the condition and in coping with it. Their religious community also provided a source of support in particular for her daughter, who stated this community helped her heal after being bullied. Another parent stated that experiences with regard to her children’s CL/P had altered her spirituality, specifically in understanding a larger sense of purpose in her children’s being born with this condition. Previously, parents had noted that increased spirituality, acceptance, commitment to social justice, and increased empathy were outcomes of the CL/P experience (Eiserman, 2001).

Adults with CL/P noted acceptance and perspective were key coping mechanisms (Berger & Dalton, 2009). The current study suggested parents played a key role in the development of these coping mechanisms. Four parents specifically mentioned encouraging acceptance and perspective in children in order to foster adjustment. Parents assisted children in accepting the facts of their condition and they educated them about ongoing treatment. Others described helping their children accept the inevitability of the treatment process and felt this acceptance led to more compliance with treatment and successful recoveries. Minimizing negative reactions to the condition and reframing it was an inconvenience that also served to accelerate adjustment. One adolescent even expressed acceptance of being bullied, stating everything happens for a reason and bullying had started a process through which she had improved her self-confidence. In looking back on her life, one adolescent noted there were times when it was more difficult to remain hopeful that things would turn out well. Continued parental support and perspective were important aspects of successful adjustment.

Two adolescents stated maturity was a factor in being better able to accept their condition and cope with social struggles. With maturity, they felt they had gained
experience, a better understanding of the negative reactions of others, and were less concerned with appearance. Parents perceived their children coped more successfully as they grew older due to confidence and long-standing, positive peer groups. Some evidence indicated a more settled social environment could decrease social difficulties in this population (Turner et al., 1997).

One parent whose child was bullied perceived that growing older was more difficult as adolescents became more concerned with appearance. Adolescence has been cited as a risk factor for social alienation in this population. One study that found lower quality of life for older children posited the reason for this was increased self-awareness of the condition (Bos & Prahl, 2011). However, this hypothesis directly contradicted what two adolescents in this study stated about improvements due to increased self-awareness.

All eight parents felt open and honest communication was critical to helping their children cope with both the condition and its medical treatment. All the parents talked about the importance of providing accurate information, listening to their child’s concerns and questions, and validating their experiences. Hall et al. (2013) noted children’s understanding of this condition is significantly impacted by parents. All parents felt communication helped their children accept treatment recommendations and make better decisions regarding treatment. Open communication has been deemed critical to decision-making and adaptation (Whitehead et al., 1996). Two parents noted communication from adolescents about their experiences was difficult to hear in comparison to earlier stages of development. Yet they also noted the importance of sharing experiences.
Resilience has been noted to be not a single factor or characteristic but a dynamic process of adapting to stressful situations or events (Luthar et al., 2000). Living with and adapting to a chronic condition such as CL/P, the treatment for which occurs over a 20-year period, certainly entails a process. This condition entails the chronic, periodic stressors of medical interventions and the changes these bring as well as more sustained stresses of living with functional and aesthetic challenges. In this study, as in resilience research generally, critical protective factors included support and educational and psychological resources. Most of the participants, both parents and adolescents, described a path of developing resilience and successful adaptation. The parents reported ongoing, intermittent stressors with regard to their children's medical treatment and any problems that arose in relation to the condition but did not report being chronically overwhelmed by these problems or unable to function as a result of them. Adolescents reported success in school, social relationships and a relative degree of acceptance to their condition and treatment. Parents and adolescents reported coping through social support, changing perspectives and personal growth, spirituality, and social advocacy. While those participants who reported successful adaptation managed to find the types of support and understanding they needed, their emotional and social needs could be met in a more systematic and accessible manner if they were a structured part of the medical team.

Integrated care could serve as a model for improving many of the stated needs of participants in this study. The term integration refers both to the integrated location of medical and mental health care and to the fact that medical and mental health treatment are integrated into one treatment plan (Blount, 2003). Research on integrated care
demonstrated it fosters communication between mental health providers and medical providers, allowing for increased understanding for both (Blount, 2003). The role of mental health providers in this setting is not only to provide assessment and therapeutic services but to educate and advocate for patient’s mental health needs and facilitate physicians’ understanding of their experiences (Blount, 2003). Medical providers are more likely to introduce mental health concerns to patients, “knowing that if they discover a situation that seems beyond their expertise, there is someone down the hall who could be involved within a reasonable period of time” (Blount, 2003, p. 8).

Outcomes research demonstrated integrated care improved access, increased patient satisfaction with medical care treatment adherence, and improved clinical outcomes (Blount, 2003).

Parents and adolescents experienced inadequate support needs both during periods of medical intervention and routinely. Both could be identified and addressed during yearly cranio-facial visits. Having a mental health provider tracking the development and progress of patients from infancy would increase the likelihood of detecting any mental health or educational concerns and would insure consistent and informed mental health care.

Communication between patients and cranio-facial specialists is critical because the condition of CL/P involves aesthetic outcomes that involve societal and subjective factors. This aspect of the decision-making process is one studies show is seldom questioned by patients or even discussed with medical providers (Nelson et al., 2013). Yet, as in the current research, patients mentioned dissatisfaction with treatment with regard to expectations after the fact. The role of cultural expectations for facial
appearance or within those expectations the presence of stigma is something that should be acknowledged and discussed by medical providers from the beginning of the treatment course. A recent study evaluated responses to individuals with cleft lip and palate and found that despite surgical advances and increased outreach and advocacy for this population, individuals still responded with stigmatizing attitudes (Pausch et al., 2016). Despite more social advocacy for this population, stigmatizing attitudes remain a part of our culture.

Medical teams must make sure they do not inadvertently support this stigma. When adaptation, acceptance, and adjustment concerns are framed within societal conceptualizations of “normality,” there is a risk for unacknowledged bias in their approach to this population. This could influence both research and medical practice. The theory of disability paradox could inform this bias. This theory is when individuals with disabilities often report an excellent quality of life--one that is informed by a realistic assessment of lower functioning, social isolation, and discrimination, whereas external observers expect them to report a low quality of life (Albrecht & Devlieger, 1999). This paradox points to the importance of researchers understanding individuals’ subjective experiences of their disability within the context of their social relationships and for its own unique perspective. Richman and Millard (1997) noted perhaps the pathway to social adjustment is simply different for this group of individuals rather than distorted.

Medical ethicists have advised medical teams to create awareness of the role of stigma and to support families in considering the implications of surgical change in light of societal expectations and the patient’s identity (Aspinall, 2006). Research on stigma
noted any social interaction has the potential to be either stigmatizing or supportive including interactions within the family and with medical professionals (Alansari et al, 2013). Physicians should not make the assumption that patients are dissatisfied with their appearance but rather discuss openly the variety of options for improving quality of life. Rather than advising more medical treatments, supportive therapies might be more beneficial and less detrimental to the adjustment process (Mouradian et al., 2006).

Several of the parents in this study described considerable distress and overwhelming even temporary thoughts of self-harm at the time of diagnosis of the cleft condition. While several infants in this study were adopted, none of the adoptive parents knew for certain the reason they were given up for adoption. While not a finding of this study, it is possible some parents chose adoption in response to the emotional reaction to the condition of their children. Having mental health providers available at the time of diagnosis is also critical given the outcomes for these children are mainly positive. Specific information and support would assist parents in better managing their distress.

**Limitations**

Qualitative research acknowledges the role of subjectivity throughout the research process and has as its goal the accurate and faithful representation of the subjective experience of a participant or group of participants (Creswell, 2007). This study, like all qualitative studies, did not aim to present generalizable results; thus, the findings of this study are not necessarily applicable to all individuals within the study population. In addition, the constructivist-interpretivist posits all individuals create personal meaning through their experiences and interactions with their environment (Schwandt, 1994). Therefore, while participants might share some experiences in relation to the condition of
cleft lip and palate, their perceptions and understanding might also differ according to their unique circumstances.

I designed the interview questions through my own perspective of the research topic, which might not have addressed the particular perceptions and experiences of the participants. While the interviews were open ended and attempted to elicit participants’ unique perspectives, it is possible the participants did not share aspects of their experiences due to the structure of my interview questions.

The fact that I am the mother of a child who was born with a cleft lip and palate might have impacted the study in several ways. It might have been a strength as my personal experience created a sense of comfort for participants to share their experiences with me. Despite my attempt to bracket my own experiences, my familiarity with the subject might have influenced my understanding of participants’ reflections.

Despite efforts to include a diverse sample of parents and adolescents, my participant group differed on several measures from the more general population--only one father agreed to participate and all of the adolescents who participated were female despite the fact that CL/P is more likely to impact boys than girls. The reason why boys were unwilling to participate was unclear. Two parents offered their views of why their sons were unwilling to participate. One mother stated her son was less focused on undergoing treatment or discussing any aspect of his condition due to being less preoccupied with the condition at this stage of life and more interested in socializing and other activities. The other parent, a father, described his son as being non-compliant with most aspects of his life due to mental health concerns. Despite the fact that the condition occurs more frequently in Asian and Native American individuals, 10 of the 12
participants were Caucasian. All families were financially stable and had insurance or the ability to receive adequate medical treatment and all but one reported having support from extended family. Most of the families interviewed consisted of two heterosexual parents--most often the biological parents of the adolescent with CL/P. Two families had adopted children and one family was a blended family. More diversity on all of these measures would likely produce more varied results.

Given that a large proportion of the participants were involved in community support or outreach and they reported a desire to give back and help others in the CL/P community, this might have impacted their desire to participate in the study in the first place. Therefore, the experiences of these individuals and families might have overly reflected the positive experiences of social support and advocacy than was warranted in the more general CL/P population.

A final limitation of this study is the interviews all took place over the telephone due to geographical distances between the participants and myself. Research suggested strengths in telephone interviews although there were few empirically based comparisons between face-to-face and telephone interviews (Irvine, Drew, & Sainsbury, 2013). Advantages of telephone interviews include increased ease with discussing sensitive topics and greater accessibility and cost effectiveness (Chapple, 1999). It might be more difficult to develop rapport over the telephone without the sharing of food and drink, small talk, and social cuing offered through body language (Opdenakker, 2006). However, a recent study pointed to several challenges in telephone interviews that might have impacted data collection. Irvine et al. (2013) noted participants make more frequent checks regarding their comprehension of questions as well as the adequacy of the
responses they offer and researchers tend to offer fewer verbal acknowledgements than in face-to-face interviews. Their findings also noted a tendency for participants to provide less detail, resulting in shorter interviews (Irvine et al., 2013).

**Implications**

**Theoretical Implications**

I chose the constructivism-interpretivism as a theoretical orientation for this study. With its focus on relative truth and personal meaning developed from individuals’ experiences, relationships, and cultural context, it seemed suited to inform a study on phenomenological experiences. Creswell (2007) noted this theory is commonly utilized in research that explores relational processes. Understanding the lived experiences of participants with a unique medical condition that requires extensive medical intervention and which impacts individual’s social acceptance necessarily involves consideration of social context. Findings of this study informed relational processes within families, between families and their medical providers, and social support systems. Because of this, a family systems orientation would also be beneficial in future research with this population.

In future research, a feminist research approach might be a useful addition to the study of this population. As Creswell (1998) noted, feminist researchers view gender as a basic principal that shapes individuals’ experiences. Feminist research is often concerned with power imbalances and social justice reform (Creswell, 1998). Recent findings suggested girls with CL/P were more negatively impacted by societal expectations regarding physical appearance than were boys (Crerand et al., 2017). The authors noted this finding mirrored body image research in the general population (Crerand et al.,
Because there is more emphasis culturally on the value of physical appearance for females, studying this population through this theoretical lens would be of great value.

Findings from this study revealed both parents and individuals with CL/P might experience stigma due to facial and speech differences linked to the condition. While healing from the impacts of being stigmatized could be an individual or family process appropriate for psychotherapy, impacting the conditions under which stigma continues to exert force in our culture requires larger efforts to inform and change societal norms.

Counseling psychologists who are committed to social justice recognize the need for a social justice orientation in all aspects of their work--from research to clinical work and efforts to influencing policy to bring about social change (Vera & Speight, 2003). Future research that includes a social justice theoretical orientation might be beneficial both in better understanding this population as well as in soliciting societal transformation.

**Research Implications**

Gaining access to this population was a significant challenge. Recruitment for this study lasted over a year, causing data collection to take place in several successive waves followed by gaps in which recruitment continued. Both schools and hospitals either would not allow access due to privacy constraints or would allow research only with the involvement of one of their own researcher team. Online support groups for parents and children affected by CL/P yielded many interested parents of young children and infants but far fewer parents of adolescents. This might be due to the fact that by the period of adolescence, most families and children have already formed their support networks and are not present on the internet groups in the same way as are parents of young children. Finally when parents did respond, often their adolescent children did not
wish to participate. Recruitment strategies for this study produced a sample of mainly mothers and daughters with only 1 of 12 participants being a male (father). Finding other means to access and recruit this population is critical, in particular increasing strategies to target adolescents and male participants.

Utilizing a qualitative design for this study helped to explore contextual and relational factors that contributed to an understanding of the risk factors and adjustment process for this population. Previous research noted the difficulty in studying the complexity, variability, and subjective nature of resilience (Ungar, 2008). Qualitative research is best suited to examine resilience factors because of its emphasis on unnamed processes, contextual factors, descriptive content, and giving voice to marginalized populations (Ungar, 2003).

Clinical Implications

Several implications emerged from this study for both medical and mental health providers who work with this population including specific concerns parents and adolescents had with regard to the impact of the treatment process, the impact of the condition on daily life, and the types of coping skills developed in response to these events. Based on the results of this study, there is a significant need for mental health providers to bring their training and services to the aid of this population. Counseling psychologists and other mental health professionals could serve a vital function in providing targeted information and support that would contribute to a successful adjustment process for individuals and families dealing with CL/P. Firstly, most participants either stated the desire for the involvement of mental health professionals or discussed emotional needs well suited to the training and skills of the mental health
profession. Secondly, based on this study and previous research, a percentage of individuals impacted by CL/P experienced symptoms of mental health disorders, indicating the need for a mental health professional to provide regular screenings and appropriate referrals for families who were more at risk. The results indicated counseling psychologists and mental health professionals should be aware that concerns about identity could significantly impact adolescents with CL/P. Successful adjustment might be aided by supportive therapies that assist adolescents in finding balance in terms of their overall identity.

The long and often uncertain medical process for CL/P has significant impacts on both parents, patients, and families. Both parents and adolescents in this study requested additional information regarding most aspects of the treatment process, emotional support and reassurance, treatment course, and post-surgical recovery. It is outside of the scope of practice for medical providers to provide information about emotional and psychological responses to medical interventions; however, this is a role counseling psychologists and other mental health professionals are well equipped to fill. In this era of integrated care, psychologists often play a vital role in assisting patients in a medical setting with understanding and adapting to the demands of medical treatment. Psychologists working with this population could provide psycho-education on the CL/P process and common responses as well as provide brief therapy and screenings for those families that might be more at risk.

Social support plays a large role in successfully coping with CL/P and its treatment process. Counseling psychologists and other mental health professionals should screen families for the availability of adequate social support, provide psycho-
education on its benefits, and provide referrals to appropriate support or therapeutic groups and/or family therapy.

The emotional impact of the condition and its treatment is significant and varied. The uncertainty of the treatment process and outcomes and the inherent risks in surgical interventions create anxiety in both parents and patients. It is clear parents would benefit greatly from learning to regulate their anxiety; this would in turn make them more available to support their children in their treatment. Counseling psychologists and other mental health professionals could assist in both normalizing and learning ways to regulate this anxiety. Sadness, grief, and anger are also emotions participants discussed as a response to treatment and for which mental health professionals could provide support and validation. Adolescents in this study experienced ambivalence about treatment and a sense of isolation despite positive family support. Group therapy with other adolescents and family therapy could both reduce alienation and provide a context for positive shared experiences.

The potential for educational difficulties and cognitive disabilities in this population also makes the role of psychologist on a medical team critical. Psychologists could provide routine assessments of cognitive functioning and provide appropriate interventions or referrals as needed. Parents expressed significant concern about their children’s peer acceptance. Mental health professionals could assist with helping parents navigate this anxiety as well as providing social skills training or therapy for social concerns for adolescents.

Counseling psychologists and other mental health professionals should be aware this is a developmental stage for families in which they and their adolescents renegotiate
autonomy and decision-making. Parents described struggling to allow their children to make decisions for themselves regarding treatment options and found it especially hard to witness what they considered to be poor decisions. Because decision-making regarding elective surgeries is a complex process involving personal and societal factors, values, and emotions rather than medical necessity, differences of opinion between family members are to be expected (Nelson, Caress et al., 2012). Counseling psychologists and other mental health professionals could assist families in navigating the complexity of these decisions as well as providing psycho-education regarding satisfaction with treatment, outcomes, and the factors in adjustment.

Based on the results of this study, counseling psychologists and other mental health professionals should be aware that individuals and families impacted by CL/P often grow from this experience in terms of increased empathy for others, desire for prosocial activity, and increased family cohesion. Participants described ways in which they reframed their experiences in a positive light and gained in perspective, wisdom, and spirituality. Therefore, the use of a strengths-based perspective with this population is likely to be critical.

The overall results of this study indicated most adolescent participants achieved successful adaptation including school success, social integration, and self-acceptance. While counseling psychologists should be aware most adolescents with CL/P show signs of resiliency, some individuals require more support. Three adolescents in this study experienced symptoms of mental health disorders and two of these had been bullied. Two parents felt the CL/P condition was independent of their child’s mental health concerns or only partly related and one attributed it solely to bullying and self-
acceptance. Understanding the etiology of these struggles is something psychologists could do both in clinical practice and in future research.

Two adolescents stated maturity allowed them to realize the relative importance of physical appearance and gave them perspective on others’ reactions to them. Growing older also led to increased ability to regulate their emotions, causing less significant reactions to difficult situations.

The potential for stigma to influence this population is very great. Both adolescents and parents expressed experiences when they felt stigmatized because of the condition of CL/P or had constant concerns about its potential impact. While all families in this study were supportive of their adolescents, it is possible this was not always the case. In addition, the subtle presence of stigma was noted in the preference for surgical treatment over other forms of adaptation such as supportive psychotherapy (Nelson Caress et al., 2012). Counseling psychologists and other mental health professionals could advocate for this population in medical settings, school settings, and assist parents in advocating for their children. United efforts between counseling psychologists and medical teams could provide support to patients and their families in an effort to de-stigmatize this condition.

**Future Research Directions**

Further research is needed to understand more about those individuals and families who are at risk and those who thrive with the condition of CL/P. There is clear evidence that while most individuals with this condition thrive, a portion of them experience adjustment concerns and/or mental health problems. Research is needed to understand the characteristics of individuals who struggle as well as their context,
environment, and family factors that might contribute to their struggles. Early psychological research illustrated the presence of risk in this population. Qualitative studies have illuminated the complexity of the process of adjustment and the need to consider the dynamics and contextual factors inherent in the treatment process (Nelson, Glenny et al., 2012). Qualitative research also revealed resilience factors and the critical importance of representing the unique voice of individuals with the CL/P condition (Eiserman, 2001). Continued research is also needed to highlight the critical aspects of successful adjustment. Further defining the unique experience with grounded theory research would provide more specificity and inform interventions with this population.

Most of these studies explored the experiences of individuals who volunteered to share their experiences and who had access to the researchers. The experience of individuals of diverse ethnic and cultural backgrounds and those of lower socio-economic class are not often represented. The voices of adolescents are still needed and efforts to include parent perspectives in this research would help illuminate the adjustment process. Longitudinal research has been lacking in the literature and is greatly needed to understand how this process develops through time and what if any risk factors contribute at various stages of development. Finally, research on psychological intervention is only in its infancy as is its integration into cranio-facial team care. Research is needed on specific treatment approaches that could assist adolescents with their unique concerns such as self-acceptance, social competence, and the impacts of bullying.

**Conclusion**

The condition of CL/P is one that requires long-term medical care and has the potential to impact both long-term functionality and appearance as well as psycho-social
adjustment. As a chronic condition requiring surgical treatment from infancy through late adolescence, CL/P also impacts family functioning. This study aimed to address a gap in the literature on parents’ and adolescents’ perspectives of successfully coping with the condition of CL/P. Research noted the need for a subjective understanding of the resilience factors within this population in order to better understand successful adaptation (Mouradian, 2001). This study utilized a phenomenological methodology to examine the impact of CL/P on adolescents and the coping skills utilized by both parents and adolescents. The findings reflected the participants’ experiences of living with a long, uncertain medical condition and its treatment process. The findings also described a lengthy adjustment process with many identified risk and resilience factors. Identification of this adjustment process and risk factors within it provided useful information to mental health providers who might work with this population.
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APPENDIX A

INSTITUTIONAL REVIEW BOARD APPROVALS
DATE: December 19, 2014

TO: Sarah Breske, MA
FROM: University of Northern Colorado (UNCO) IRB

PROJECT TITLE: [663152-5] The experiences of parents and children with the medical care for the condition of cleft lip and palate; a phenomenological study.

SUBMISSION TYPE: Amendment/Modification

ACTION: APPROVED
APPROVAL DATE: December 19, 2014
EXPIRATION DATE: December 19, 2015
REVIEW TYPE: Expedited Review

Thank you for your submission of Amendment/Modification materials for this project. The University of Northern Colorado (UNCO) IRB has APPROVED your submission. All research must be conducted in accordance with this approved submission.

This submission has received Expedited Review based on applicable federal regulations.

Please remember that informed consent is a process beginning with a description of the project and insurance of participant understanding. Informed consent must continue throughout the project via a dialogue between the researcher and research participant. Federal regulations require that each participant receives a copy of the consent document.

Please note that any revision to previously approved materials must be approved by this committee prior to initiation. Please use the appropriate revision forms for this procedure.

All UNANTICIPATED PROBLEMS involving risks to subjects or others and SERIOUS and UNEXPECTED adverse events must be reported promptly to this office.

All NON-COMPLIANCE issues or COMPLAINTS regarding this project must be reported promptly to this office.

Based on the risks, this project requires continuing review by this committee on an annual basis. Please use the appropriate forms for this procedure. Your documentation for continuing review must be received with sufficient time for review and continued approval before the expiration date of December 19, 2015.

Please note that all research records must be retained for a minimum of three years after the completion of the project.

If you have any questions, please contact Sherry May at 970-351-1910 or Sherry.May@unco.edu. Please include your project title and reference number in all correspondence with this committee.

Hello Sarah,
Thank you for these modifications. I apologize for the process being this tedious for you. We had a miscommunication that Dr. Roehrs had further feedback, but this was not sent to you because she did approve your application.

I would like to commend you for your perseverance and will now approve your study and hopefully you can begin shortly.

Thank you for your attention and good luck with your research.

Sincerely,

Nancy White, PhD, IRB Co-Chair

This letter has been electronically signed in accordance with all applicable regulations, and a copy is retained within University of Northern Colorado (UNC) IRB's records.
DATE: September 11, 2015

TO: Sarah Breeseke, MA
FROM: University of Northern Colorado (UNCO) IRB

PROJECT TITLE: [653152-6] The experiences of parents and children with the medical care for the condition of cleft lip and palate: a phenomenological study.

SUBMISSION TYPE: Amendment/Modification

ACTION: APPROVED
APPROVAL DATE: September 9, 2015
EXPIRATION DATE: December 19, 2015
REVIEW TYPE: Expedited Review

Thank you for your submission of Amendment/Modification materials for this project. The University of Northern Colorado (UNCO) IRB has APPROVED your submission. All research must be conducted in accordance with this approved submission.

This submission has received Expedited Review based on applicable federal regulations.

Please remember that informed consent is a process beginning with a description of the project and insurance of participant understanding. Informed consent must continue throughout the project via a dialogue between the researcher and research participant. Federal regulations require that each participant receives a copy of the consent document.

Please note that any revision to previously approved materials must be approved by this committee prior to initiation. Please use the appropriate revision forms for this procedure.

All UNANTICIPATED PROBLEMS involving risks to subjects or others and SERIOUS and UNEXPECTED adverse events must be reported promptly to this office.

All NON-COMPLIANCE issues or COMPLAINTS regarding this project must be reported promptly to this office.

Based on the risks, this project requires continuing review by this committee on an annual basis. Please use the appropriate forms for this procedure. Your documentation for continuing review must be received with sufficient time for review and continued approval before the expiration date of December 19, 2015.

Please note that all research records must be retained for a minimum of three years after the completion of the project.

If you have any questions, please contact Sherry May at 970-351-1910 or Sherry.May@unco.edu. Please include your project title and reference number in all correspondence with this committee.
Sarah,

Sorry this took so long.

So glad to see your research continuing!

Maria

This letter has been electronically signed in accordance with all applicable regulations, and a copy is retained within University of Northern Colorado (UNCO) IRB's records.
DATE: November 30, 2016

TO: Sarah Breske, MA

FROM: University of Northern Colorado (UNCO) IRB

PROJECT TITLE: [663152-8] The experiences of parents and children with the medical care for the condition of cleft lip and palate: a phenomenological study.

SUBMISSION TYPE: Amendment/Modification

ACTION: APPROVED

APPROVAL DATE: November 29, 2015

EXPIRATION DATE: December 19, 2015

REVIEW TYPE: Expedited Review

Thank you for your submission of Amendment/Modification materials for this project. The University of Northern Colorado (UNCO) IRB has APPROVED your submission. All research must be conducted in accordance with this approved submission.

This submission has received Expedited Review based on applicable federal regulations.

Please remember that informed consent is a process beginning with a description of the project and insurance of participant understanding. Informed consent must continue throughout the project via a dialogue between the researcher and research participant. Federal regulations require that each participant receives a copy of the consent document.

Please note that any revision to previously approved materials must be approved by this committee prior to initiation. Please use the appropriate revision forms for this procedure.

All UNANTICIPATED PROBLEMS involving risks to subjects or others and SERIOUS and UNEXPECTED adverse events must be reported promptly to this office.

All NON-COMPLIANCE issues or COMPLAINTS regarding this project must be reported promptly to this office.

Based on the risks, this project requires continuing review by this committee on an annual basis. Please use the appropriate forms for this procedure. Your documentation for continuing review must be received with sufficient time for review and continued approval before the expiration date of December 19, 2015.

Please note that all research records must be retained for a minimum of three years after the completion of the project.

If you have any questions, please contact Sherry May at 970-351-1910 or Sherry.May@unco.edu. Please include your project title and reference number in all correspondence with this committee.
This letter has been electronically signed in accordance with all applicable regulations, and a copy is retained within University of Northern Colorado (UNCO) IRB's records.
DATE: January 2, 2016

TO: Sarah Breske, MA
FROM: University of Northern Colorado (UNCO) IRB

PROJECT TITLE: [663152-9] The experiences of parents and children with the medical care for the condition of cleft lip and palate: a phenomenological study.

SUBMISSION TYPE: Continuing Review/Progress Report

ACTION: APPROVED
APPROVAL DATE: January 2, 2016
EXPIRATION DATE: December 19, 2016
REVIEW TYPE: Expedited Review

Thank you for your submission of Continuing Review/Progress Report materials for this project. The University of Northern Colorado (UNCO) IRB has APPROVED your submission. All research must be conducted in accordance with this approved submission.

This submission has received Expedited Review based on applicable federal regulations.

Please remember that informed consent is a process beginning with a description of the project and insurance of participant understanding. Informed consent must continue throughout the project via a dialogue between the researcher and research participant. Federal regulations require that each participant receive a copy of the consent document.

Please note that any revision to previously approved materials must be approved by this committee prior to initiation. Please use the appropriate revision forms for this procedure.

All UNANTICIPATED PROBLEMS involving risks to subjects or others and SERIOUS and UNEXPECTED adverse events must be reported promptly to this office.

All NON-COMPLIANCE issues or COMPLAINTS regarding this project must be reported promptly to this office.

Based on the risks, this project requires continuing review by this committee on an annual basis. Please use the appropriate forms for this procedure. Your documentation for continuing review must be received with sufficient time for review and continued approval before the expiration date of December 19, 2016.

Please note that all research records must be retained for a minimum of three years after the completion of the project.

If you have any questions, please contact Sherry May at 970-351-1910 or Sherry.May@unco.edu. Please include your project title and reference number in all correspondence with this committee.
This letter has been electronically signed in accordance with all applicable regulations, and a copy is retained within University of Northern Colorado (UNCO) IRB's records.
Institutional Review Board

DATE: October 3, 2016

TO: Sarah Breske, MA
FROM: University of Northern Colorado (UNCO) IRB

PROJECT TITLE: [663152-10] The experiences of parents and children with the medical care for the condition of cleft lip and palate: a phenomenological study.

SUBMISSION TYPE: Continuing Review/Progress Report

ACTION: APPROVED

APPROVAL DATE: October 3, 2016
EXPIRATION DATE: October 3, 2017
REVIEW TYPE: Expedited Review

Thank you for your submission of Continuing Review/Progress Report materials for this project. The University of Northern Colorado (UNCO) IRB has APPROVED your submission. All research must be conducted in accordance with this approved submission.

This submission has received Expedited Review based on applicable federal regulations.

Please remember that informed consent is a process beginning with a description of the project and insurance of participant understanding. Informed consent must continue throughout the project via a dialogue between the researcher and research participant. Federal regulations require that each participant receives a copy of the consent document.

Please note that any revision to previously approved materials must be approved by this committee prior to initiation. Please use the appropriate revision forms for this procedure.

All UNANTICIPATED PROBLEMS involving risks to subjects or others and SERIOUS and UNEXPECTED adverse events must be reported promptly to this office.

All NON-COMPLIANCE issues or COMPLAINTS regarding this project must be reported promptly to this office.

Based on the risks, this project requires continuing review by this committee on an annual basis. Please use the appropriate forms for this procedure. Your documentation for continuing review must be received with sufficient time for review and continued approval before the expiration date of October 3, 2017.

Please note that all research records must be retained for a minimum of three years after the completion of the project.

If you have any questions, please contact Sherry May at 970-351-1910 or Sherry.May@unco.edu. Please include your project title and reference number in all correspondence with this committee.
Sarah -

Your continuation application is approved. Best wishes with your continued work on this project.

Sincerely,

Dr. Megan Stelliño, UNC IRB Co-Chair

This letter has been electronically signed in accordance with all applicable regulations, and a copy is retained within University of Northern Colorado (UNCO) IRB's records.
APPENDIX B

INTERVIEW GUIDE—PARENTS
Describe your experiences with your child’s medical care.

- When did you find out about your child’s diagnosis? How did you respond?
- Describe the process of choosing a medical team for your child?
- Describe your role in making decisions for your child’s care?
- What treatment has your child undergone thus far?
- What were your experiences with this team? With surgery and recovery?
- What were the positive and negative aspects of their medical care?
- Describe how these experiences affected you?
- How did you cope with these experiences?
- How do you make sense of these experiences?
- How do you think these experiences continue to impact you, your family and your child?
- Describe how this treatment impacts your parenting?
- How do you try to help your child with aspects of this treatment process?
- Is there anything else you would like to add?
APPENDIX C

INTERVIEW GUIDE--ADOLESCENTS
Describe your experience with the medical treatment for CL/P:

- What treatments have you undergone for CL/P?
- What do you remember about these treatments?
- What have you been told about treatments you don’t remember?
- What has it been like to undergo treatment?
- Describe your experience with your medical team?
- Describe your experience with surgery and recovery?
- How do these treatments impact you today?
- Describe how your parents try to help you with this process?
- Describe your involvement in making decisions about treatment?
- How do you make sense of these experiences?
- How do you cope with these experiences?
- Is there anything else you would like to add?
APPENDIX D

CONSENT FORM (ADULT)
CONSENT FORM FOR HUMAN PARTICIPANTS IN RESEARCH
UNIVERSITY OF NORTHERN COLORADO

Project Title: The Experience of Parents and Adolescents with the Medical Care for the Condition of Cleft-lip and Palate. A Phenomenological study

Researcher: Sarah Breseeke, M.A. Department of Counseling Psychology
Phone: (573) 639-0112 Email: bres6545@bears.unco.edu
Research Advisor: Basilia Softas-Nall, Ph.D. Department of Counseling Psychology.
Phone: (970) 351-1640 Email: Basilia.softas-nall@unco.edu

Purpose and Description: The purpose of this study is to explore the experiences of parents and adolescents with the medical treatment for cleft lip and palate, and in particular the impact on parent and child relationships, and factors involved in coping with the condition. This study aims to produce a description of the essential experiences of parents and adolescents with their medical treatment journey.

Participation in this study is voluntary and you may choose to end participation at any time. If you agree to participate in this study, you will be interviewed two times for a total time of 1-3 hours. The interviews will take place in person if possible, and by telephone if geographical distance prohibits in person interviews. Parents and children will decide if the children would like to be interviewed alone or with a parent present. The interviews may take place in your home, or at some other location that is convenient, comfortable and confidential. The interviews will be recorded and the recordings will later be typed verbatim by the researcher. Types of questions that may be asked include 1) Describe your experience with the surgical care you/ or your child received? 2) What was the impact of this experience on you/ or your family? 3) Describe how this experience is impacting you today. You will also be sent the researchers analysis of the interviews in order to provide feedback regarding how well I captured your experience, and to provide suggestions or comments. The entire research process is expected to be completed by May of 2015. Discussing in depth your child's medical treatment, and the impact on yourself and your family may be upsetting, and you are free to end the interviews or change their direction at any point in time. It may also be beneficial to discuss your experiences in depth.

The researcher will make every attempt to maintain the confidentiality of the participants in this study. You will be asked to select a pseudonym during your interview, unless you wish to use your real name. You will only be referred to by the name chosen during data collection and report writing. The doctoral student who will serve as a research advisor will not be aware of your real name, or any identifying information, again, unless you specifically request that your real name be used. Requests to eliminate any of the information shared in the interview process from the final written analysis will be honored by the researcher. Participant information such as signed consent forms, and transcripts of interviews will be kept on a password protected computer, and destroyed three years after the completion of the study.

There are no foreseeable risks involved in participation in this study. However, the participant may experience emotional discomfort due to the sensitive nature of the topic. It may also be beneficial to you focus on your experiences in depth. If you wish I will provide a list of counseling referrals. While this study may not benefit participants directly, it will help us learn more about the nature of the medical treatment process for cleft children from the perspective of parents. This information will be useful to other parents, medical personnel and mental health professionals who work with cleft-affected families.
Participation is voluntary. You may decide not to participate in this study and if you begin participation you may still decide to stop and withdraw at any time. Your decision will be respected and will not result in loss of benefits to which you are otherwise entitled. Having read the above and having had an opportunity to ask any questions, please sign below if you would like to participate in this research. A copy of this form will be given to you to retain for future reference. If you have any concerns about your selection or treatment as a research participant, please contact the Office of Sponsored Programs, Kepner Hall, University of Northern Colorado Greeley, CO 80639; 970-351-2161.

________________________________________________________
Participant Signature

________________________________________________________
Researcher Signature
APPENDIX E

ASSENT FORM (ADOLESCENTS)
Hi,

My name is Sarah Breseke and I am a student at the University of Northern Colorado. I am doing research about what the treatment process for cleft lip and palate is like for adolescents and their parents. I would like to talk to you about your experiences. If you wish you can talk to me about this.

If you talk to me I will ask you about the types of medical treatments you have had, those you remember and those your parents have told you about, and what these were like for you. I will ask you about how you and your family make decisions about your treatment, and what your relationship is like with your doctors. We will also talk about how your parents try to help you with these experiences. If there is anything you do not wish to talk about you do not have to. I will record the interview but I won’t use your name or any identifying details about you in the written report.

I will interview you either in person or on the phone. In person interviews will take place in a location that feels comfortable, confidential and convenient for you, like your home or a library. I will interview you twice, for a total of between 1-3 hours.

Talking to me is unlikely to have any risks for you, and may be beneficial. Your parents have given permission for you to talk to me, but you don’t have to. You can also stop anytime you want to if you change your mind. You can choose if you would like to have your parents present or not during the interview.

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APPENDIX F

MANUSCRIPT FOR PUBLICATION
THE EXPERIENCE OF PARENTS AND ADOLESCENTS WITH
THE MEDICAL CARE FOR THE CONDITION OF CLEFT
LIP AND PALATE: A PHENOMENOLOGICAL STUDY

Introduction

Cleft lip and palate (CL/P) is a common birth defect that affects 1 in 700 children born each year in the United States (Cleft Palate Foundation, 2013). This condition causes multiple aesthetic and functional challenges and requires interdisciplinary medical care from infancy to late adolescence. Children with this condition typically undergo multiple reconstructive surgeries of their mouth, jaw, and nose. Standard protocols on the number, order, and type of surgical interventions do not exist, causing uncertainty in treatment expectations (Grollemund et al., 2012). Patients also undergo speech therapy as well as orthodontic and otolaryngological treatments. Despite lengthy medical interventions, children and adults with this condition might face deficits in these areas of functioning as well as having visible facial scars. While there is consensus that this condition puts the individual and the family at risk for less than optimal psychosocial functioning, the exact nature of this impact is a matter of considerable debate.

Research on the impact of CL/P on parents, children, and adolescents has yielded varied and inconsistent results. A body of research makes claims to the negative impact of this condition on both parents and children in terms of psychopathology and/or psychosocial adjustment. Strauss and Broder (1991) noted studies that reflect the medical model have focused on the prevalence of psychopathology in patients. Qualitative studies have focused on factors pertinent to successful adjustment and resilience (Nelson, Kirk, & Caress, 2013), generating findings away from the focus on
deficits (Eiserman, 2001; Nelson et al., 2013; Sharif, Callery, & Tierney, 2013). Challenges some individuals with CL/P face are significant, while others with the same condition manage more successfully. Factors that contribute to this success are not clear.

Research has examined psychosocial functioning, behavior, social experiences, quality of life, and satisfaction with treatment and appearance (Hall, Gibson, James, & Rodd, 2013). Most of this research utilized validated assessment tools and quantitative methods to assess particular domains of functioning with little focus on the individual as a whole or how these complex factors contributed to overall functioning (Berger & Dalton, 2009). The specificity of focus in these studies necessarily excluded contextual factors and the presence of potentially positive experiences (Eiserman, 2001). Inconsistencies in study variables and assessment instruments limited the generalizability of the CL/P literature (Hunt, Burden, Hepper, & Johnston, 2005). Thus, while there is a large body of research on this topic in general, very few studies exist or have been replicated on specific constructs or topics within the broader domain of psychosocial functioning (Hunt et al., 2005). The great variability in treatment teams and outcomes for CL/P also complicates comparisons between patients of different medical teams, a factor not thoroughly taken into account in most studies (Strauss & Broder, 1993).

The discrepancies in the literature reflect methodological challenges as well as a lack of theoretical specificity regarding the etiology and development of psychological and emotional problems in individuals with CL/Ps (Bennett & Stanton, 1994). Without understanding the nature of the difficulties, implementing or advocating for successful coping mechanisms is challenging. If researchers are to assist providers, an
understanding of the complex processes and varying individual factors relating to successful adjustment is needed.

Adjustment to a chronic health condition and its treatment is complex and varied. Chronic illness research found some individuals are more negatively impacted than others, suggesting research on individual factors contributing to successful coping is needed (Frank et al., 1998). Research on chronic health conditions generally and the CL/P literature specifically found the severity of the condition was not correlated with psychological adjustment (Lansdown, Lloyd, & Hunter, 1991). One study found no correlation between the amount of interventions completed and outcome satisfaction for patients with CL/P (Semb, Brattstrom, Molsted, Prahl-Andersen, & Shaw, 2005).

Qualitative research has been used extensively to elicit patient values and meanings in the healthcare field (Nelson, Glenny, Kirk, & Caress, 2011). Nelson (2009) advised that research with the CL/P population should be focused on meaningful outcomes rather than objective measures.

These findings have led to a call for an increase in patient-centered care, particularly at the later stages of treatment for CL/P when the surgeries are elective rather than functional (Canady, 1995; Noar, 1991). Because these surgeries impact both aesthetic appearance and functionality, the perceptions and emotional needs of patients are thought to be critical (Canady, 1995).

More research is needed on the outcomes of both surgical and non-surgical treatments for CL/P and on the experiences of individuals and families going through these processes (Mouradian, Edwards, Topolski, Rumsey, & Patrick, 2006). A review of 64 studies found only 21 included patients’ perspectives (Hunt et al., 2005). Strauss
(2001) wrote that in order for societal perceptions of cleft affected people to change, researchers and practitioners need to listen to their experiences and ask questions that focus on coping. Currently, few studies document the views of children or adolescents with CL/P (Sharif et al., 2013).

While several studies suggested parenting practices are likely to influence the coping of children with CL/P, “little is known about parenting adaptations that may assist children…to overcome medical and social challenges” (Klein, Pope, & Tan, 2010, p. 300). The experiences of parenting older children, parenting practices, and the quality of the parent/child relationship have yet to be examined. Research commonly suggested parents should assist children in coping with potentially negative social encounters and managing the stress of medical interventions but few studies within this population explored how this should be done (Sharif et al., 2013).

The purpose of this qualitative study was to (a) explore and describe how both parents and adolescent children experienced living with the condition of CL/P, (b) specifically how the medical interventions impacted their lives both individually and in relationship to one another, and (c) what elements assist them in successfully coping with this condition. Experiences with the medical procedures, decision-making processes, interactions with providers, and how parents and adolescents coped with stressors individually and as a family unit were all focal points.

Method

The theoretical framework that informed this study and guided data interpretation was constructivism-interpretivism (Schwandt, 1994). This framework posits “all knowledge, and therefore all meaningful reality...is contingent upon human practices,
being constructed in and out of interaction between human beings and their world” (Crotty, 1998, p. 42). This is a relativist viewpoint in which multiple realities exist that are equally valid (Schwandt, 2001). Therefore, reality is not objectively discovered by observing researchers nor is it entirely dependent upon the subjective understanding of researchers. Instead, reality is constructed within each individual as he/she interacts with his/her social environment. Personal experiences, perceptions, and social and historical contexts can all influence the construction of personal meaning (Ponterotto, 2005). The study of relational processes between people and the impact of contextual factors in shaping human experience are frequently topics of study within this framework (Creswell, 2007).

Constructivism-interpretivism also acknowledges the researcher's subjective experiences, knowledge, and values influence the research process and position the researcher as a co-constructor of meaning (Morrow, 2005). The social relationship between the researcher and the participants is critical to the development of research findings (Havercamp & Young, 2007). A collaborative process allows the researcher to broaden rather than abandon his/her understanding through interaction; the goal of such research is not concerned with “accuracy” but with “the emergence of a new, dialectical understanding of the phenomenon in question” (Havercamp & Young, 2007, p. 278).

In this study, I utilized phenomenological methodology to collect and analyze the data. Moustakas (1994) wrote the aim of phenomenological studies is to describe the meaning of a phenomenon as experienced by several individuals. His approach emphasized description and the use of epoche or the intentional setting aside of any prior
experience, values, or knowledge on the researcher’s part (Creswell, Hansen, Plano & Morales, 2007).

**Procedure**

All participants were selected through purposeful sampling. Purposeful sampling is best suited to qualitative research's goal of finding participants “whose experience most fully and authentically manifests or makes accessible what the researcher is interested in” (Wertz, 2005, p. 171). A participant group was selected based on the following criteria: parent participants must be either a parent or primary caretaker of a child (or children) with a non-syndromal cleft lip and palate, who are at least 12 years of age, and have completed at least the initial surgical repairs for the condition (i.e., initial repair of the cleft lip and palate).

Participants were recruited through online sources such as cleft support groups and a Facebook page. Sample size in qualitative research depends upon the purposes and goals of the inquiry and is judged to be sufficient when the results extensively reflect the experiences of participants in depth (Morrow, 2007). Wertz (2005) noted that in a phenomenological study, the number of participants cannot be determined beforehand but depends instead upon the researcher attempting to reach theoretical saturation and redundancy. Saturation refers to the point when the themes of analysis fully account for all the data (Morrow, 2007). Redundancy refers to the point at which new data do not offer new insights. The goal was to continue recruitment until saturation was reached; however, one finding of this study was the target population was difficult to access.

Four parent and adolescent dyads were interviewed for this study as well as three individual parents and one individual adolescent. The participants came from different
areas of the United States. Ten of the 12 participants were Caucasian, one was Asian, and one was bi-racial. All families reported financial stability and had insurance or the ability to receive adequate medical treatment. Most of the families interviewed consisted of two heterosexual parents, most often the biological parents of the adolescent with CL/P. Two families had adopted children and one family was a blended family.

All participants were interviewed by telephone. While this method of interviewing did not allow for the same level of intimacy as face-to-face contact, research suggested telephone interviews also limit participant distress and are, therefore, useful particularly in discussing sensitive topics (Mealer & Jones, 2014). Participants were informed their confidentiality could not be ensured with the use of technology in the same way as it would be in person.

**Data Collection**

Data collection for this study included transcribed interviews and researcher notes based on observations during interviews. All participants were interviewed individually by phone for between 35 and 75 minutes. The interviews were semi-structured to allow the researcher to respond to the emerging discussion and topics as they evolved (Merriam, 1998). Phenomenological interviews typically follow an “informal, interactive process and utilize open-ended comments and questions” (Moustakas, 1994, p. 114). I audiotaped and transcribed the interviews verbatim. Participants were assigned pseudonyms to protect confidentiality.

**Analysis**

Data analysis followed the phenomenological methods outlined by Moustakas (1994). The first step in analysis was the researcher's attempt to use epoche and to
bracket any prior personal or theoretical understanding of the topic in order to reflect fully on the data that had been collected. The second step in analysis involved a process termed horizontalization in which the transcripts were first read through in their entirety in an effort to recognize that “every horizon or statement relevant to the topic and question as having equal value” (Moustakas, 1994, p. 118). The third step involved organizing these statements into larger units of meaning or themes (Moustakas, 1994). The fourth step involved reordering themes into a structural description of how the phenomenon was experienced by participants and a textural description of what was experienced in terms of the assignment of meaning by participants (Moustakas, 1994). Lastly, these structural and textural descriptions were further combined into a common, typifying description known as the “essential invariant essence” of the experience (Creswell, 2007, p. 62).

The manner in which qualitative research is judged to be rigorous and scholarly is different from the way quantitative research is judged. The term trustworthiness refers to the standards by which a study has been conducted and written and to its ability to accurately and convincingly describe the topic with sufficient depth. In this study, I was guided by Lincoln and Guba's (1985) categories of trustworthiness: credibility, transferability, dependability, and confirmability.

Credibility refers to whether the findings are believable when compared to the direct data as quoted in the study and whether they are worthwhile for the field of study (Lincoln & Guba, 1985; Schwandt, 2001). Researcher reflexivity is critical to ensuring credibility. Intentional bracketing of researcher’s biases is critical to the phenomenological method and also ensures credibility (Williams & Morrow, 2009).
Credibility was also supported by the use of expert, peer, and member checking. Advisors were utilized throughout the analysis process to discuss concerns with the ongoing research as well as themes and analysis. Member check involved making sure my interpretations honored the meaning as perceived by participants (Lincoln & Guba, 1985). The use of triangulation through interviewing both mothers and fathers and pairs of adolescents to reveal different perspectives also increased credibility. However, gaining interviews from different perspectives within the same family was not always possible due to recruitment difficulties. Transferability refers to whether the data are presented in enough detail so the reader can generalize them to other similar situations or research findings. Transferability is judged by the reader and is dependent upon the use of direct quotations and the use of “thick and rich descriptions” in which the “writer describes in detail the participants or setting under study” (Creswell, 2007, p. 209). Extensive use of participant quotes and detailed contextual description are also utilized to increase transferability. Dependability refers to whether the research design and process are traceable and logical (Lincoln & Guba, 1985). Dependability can be augmented by providing a clearly articulated theoretical framework, methodology, and methods section as well as by the use of an audit trail. Confirmability refers to whether the written findings make sense to the reader when compared to the data that were collected. The use of an audit trail, member, and peer checks and thick and rich descriptions all increase confirmability (Creswell, 2007).

Results

Themes that emerged from the findings included identity development and acceptance of perceived difference, extensive and lengthy medical treatment (with
significant impact), relationship with the medical team, coping through social support, varied emotional impact of condition and treatment, management of the condition includes ongoing decision making, reframing and changing perspectives, acceptance and adjustment as a complex process, communication and honesty, and social acceptance is a concern for parents. Sub-themes are also discussed.

**Identity Development and Acceptance of Perceived Difference**

Several adolescents described a complex sense of identity in relation to their medical condition. This identity was described as the result of a long internal process of defining their self-concept in combination with their perception of their place in their cultural context. They described knowing others perceived them to be different and had accepted this. They also described knowing they were not fundamentally different from other people and had come to this realization through a process involving understanding that others’ perceptions were based on a lack of understanding. Anne stated, “I might look different but I’m the same personality wise as other people. You just don’t understand I’m still the same as everybody else.”

Adolescents were able to accept themselves though realizing the relative lack of importance in outward appearance. Crerand, Sarwer, Kazak, Clarke, and Rumsey (2017) found adolescents with CL/P were less invested in the importance of appearance than others their age. Parents in the current study who had encouraged self-acceptance and perceived these efforts to be helpful also noted their children exhibited a strong sense of individuality. Jessie stated, “You don’t have to be like everybody else, be the best you you can be…she thrives on being not like anybody else…and her brother follows in her footsteps.”
Identity is connected to the progress of medical treatment as well as social interactions. None of the participants expressed current difficulty with their sense of identity although they recognized periods when this was more difficult due to age or stigmatizing experiences. Those who had planned revision surgeries in the near future were anxious and hopeful these would create further change, whereas the one adolescent who had completed the surgical protocol felt the outcomes did not warrant the process. Lily said, “[It] made me think about myself differently...if I could have taken the surgeries out I could’ve been a little happier I guess. It kinda makes you insecure, you know there’s something not right and you’re working on it.”

Studies have shown that adolescents with CL/P fare well on measures of appearance and self-acceptance (Pope Klein, & Bergman, 2016); the reasons are usually thought to be either the presence of resilience and successful adjustment or the product of overcompensation. Kapp-Simon, Simon, and Kristovich (1991) posited it was only through avoidance of socializing that adolescents maintained a positive self-concept. Others considered this adaptation to consist of minimizing and self-protection (Crerand et al., 2017). The concept of self-presentation posits that cognitive and behavioral strategies assist individuals in maintaining a positive self-concept in the face of lack of social acceptance (Thompson & Kent, 2001). It is known that patient satisfaction with appearance and outcomes has no association with the amount of treatment or any objective measure of success (Semb et al., 2005). This points to an internal process that is as important as are outward appearances in the process of developing healthy self-concept and identity.
When adaptation, acceptance, and adjustment concerns are framed within societal conceptualizations of “normality,” there is a risk for unacknowledged bias in the approach to understanding the experience of this population. The theory of disability paradox could inform this bias. This phenomenon finds individuals with disabilities often report an excellent quality of life--one that is informed by realistic assessment of lower functioning, social isolation, and discrimination; whereas external observers expect them to report a low quality of life (Albrecht & Devlieger, 1999). This paradox points to the importance of researchers exploring individuals’ subjective experiences of their disability within the context of their social relationships and for its own unique perspective.

Richman and Millard (1997) noted perhaps the pathway to social adjustment is simply different for this group of individuals rather than distorted. Medical ethicists have advised medical teams to create awareness of the role of stigma and to support families in considering the implications of surgical change in light of societal expectations and the patient’s identity (Aspinall, 2006). Rather than advising more medical treatments, supportive therapies might be more beneficial and less detrimental to the adjustment process (Mouradian et al., 2006).

**Extensive and Lengthy Medical Treatment (with Significant Impact)**

Both adolescent and adult participants acknowledged the treatment process for CL/P is extensive and time consuming. Parents reported impacts on the entire family system and none felt prepared for the quantity and intensity of treatment required. All the adolescents in this study had undergone revision surgeries for primary repairs. One parent and one adolescent specifically mentioned the burden of having to repeat surgeries. All participants had experienced or were undergoing significant orthodontic
work and several experienced ongoing difficulties with hearing, taste, breathing, and eating. The literature also noted that despite surgical repairs, individuals with cleft conditions often continued to experience functional difficulties with eating and breathing (Kasten et al., 2008). While none of the participants reported continued speech problems, all of them had undergone speech therapy in the past, which is common to this population (Kasten et al., 2008).

**Lack of certainty in the medical process.** Four parents expressed a desire for better communication from the medical team regarding the course of treatment, treatment outcomes, and postoperative recovery expectations. Paula said:

> I would have liked to be a little bit more informed about the extent of what we were talking about as far as surgeries and stuff. I was left with the impression that have some surgeries and ear tubes and she’ll be fine…(I) kind of felt like I was not well informed. I kind of would have like to have that life time warning that this is a 20-year ordeal, don’t get complacent after year two. I kind of felt cheated and maybe they think parents don’t want to hear it or maybe they can’t handle it…I felt like they had sugar coated it for me.

One parent specifically mentioned the lack of clear information made parenting a challenge as she could not properly inform her child about timing and expectations for treatments. Another parent who experienced several complications and revisions stated more information regarding the possibility of set-backs would have been helpful.

> I don’t know if there’s a better way to prepare parents that things don’t always go exactly the way they’re supposed to. We had really bad luck with the surgeries. The team was great but they would say there is a “very low percentage we have to
do it again’ and Aidan always experienced that. His surgeries were always successful but if there was a side effect we were going to get it… that was really tough to keep having to deal with those things. (Jeff)

The goal of these parents was to better prepare their children, thereby reducing their suffering. Because each intervention was unique, parents experienced a continuous need for information (Nelson et al., 2013).

Two adolescents reported the need for better communication regarding treatment. Francine stated, “I mean as a kid I was pretty okay with them but as I got older I actually started to really hate them…I get mad that sometimes I think they’re postponing stuff on purpose.” Lily stated, “Surgeries keep popping up, out of nowhere…like o yeah you have to have this- like the bone graft I just had I had no idea I had to have it. When I found out, I was really upset about it.”

In the United States, there is no standardized protocol for treatment; rather, treatment depends upon each provider’s experience, often informed by case studies (Grollemund et al., 2012). Thus, these perspectives reflected a very real condition of cleft care at this time. There is also no consensus on the acceptable amount of revision surgeries (Sitzman, Coyne, & Britto, 2016). Reviews of revision rates found such variations that “it is not possible to estimate the burden of care” for patients (Sitzman et al., 2016, p. 92). McIntyre et al. (2016) noted the average number of surgeries per child in one hospital between birth and the age of 21 was eight. Sitzman et al. (2016) noted the number and intensity of surgeries should be warranted by the final aesthetic outcome, yet Trotman et al. (2007) showed revisions were often costly in terms of time and finances.
Factors involved in variation of revision surgery included quality of initial repair, preference for revision surgery by the surgeon, and treatment protocol differences (Semb et al., 2005). Socio-economic status, health access, and insurance availability might also influence whether families are offered revision surgeries (Cassell, Daniels, & Meyer, 2009). The incidence of surgical revision could be reduced by the improved quality of primary repairs but few surgeons have employed published techniques that guarantee this (Mulliken & Martinez-Perez, 1999; Sitzman, Girotto, & Marcus, 2008; Sitzman et al., 2016).

By contrast, two parents reported preferring or being satisfied with less specific information, which they felt better reflected the inherent uncertainty of the treatment process. They considered this less burdensome to the family. These parents also did not perceive any significant disappointments or set-backs in the treatment process for their children despite acknowledging several revision surgeries. Jessie stated, “It’s a shock for parents to know what lies ahead.” Doctors need “be gentle and vague because things change so quickly, especially now the treatments have changed.”

Two parents who were warned about the likelihood of revisions or complications early on did not express dissatisfaction. Shared communication and a feeling of being involved in the medical process has been linked to positive coping for parents (Nelson, Caress, Glenny, & Kirk, 2012). Eiserman (2001) found parents desired to be fully informed about the medical process in order to make better decisions for their children. Several studies found parents felt relatively uninvolved in the treatment making process for this condition (Jeffrey & Boorman, 2001; Turner, Thomas, Dowell, Rumsey, & Sandy, 1997).
Financial impact. Three families experienced financial burden as a result of the medical treatment. One family struggled with insurance covering care and two families experienced loss of income as one parent stopped working to meet caretaking needs. Both families discussed making this choice to meet their expectations for parenting, which they felt was more important than income. One had to travel several hours away to another state for medical care and one had multiple children with CL/P and no extended family near enough to assist with childcare. One family struggled to finance their child’s significant mental health needs. Burdens on parent’s time, careers, and financial stability were noted in the literature (Locker, Jokovic, & Tompson, 2005).

Relationship with the Medical Team

Need for increased emotional support from the medical team. Three families expressed a desire for more emotional support from their team. Anne said, “You need that emotional support. They all have their area of expertise…ENT’s, neurologists, whatever …whatever… but nobody is an expert in being a parent like somebody’s that’s parented a cleft child. I think that’s super, super important.” Studies have shown some parents consistently requested psychological support from their medical teams (Johansson & Ringsberg, 2004; Kuttenberger, Ohmer, & Polska, 2010). In several countries, medical and psychological organizations have made recommendations that mental health providers be a core part of the cranio-facial treatment teams (Colbert, Green, Brennan, & Mercer, 2015).

Parents wanted to understand the implications of both the treatment and the condition on their child’s quality of life. Reassurance that the family would be okay, that it was possible to cope with the process, and the child would find acceptance and a
normal life were common concerns. One parent mentioned the desire to understand and be reassured about aspects of her child’s experience given her physiological differences; another desired that the medical team be gentle and compassionate. Jessie stated, “These are our babies…this is not a flight plan…it’s their job but it needs to include a bit of compassion.” Nelson et al. (2013) noted parents’ satisfaction with care in part hinged on how much medical providers eased the process for their children.

Parents who did not specifically express the need for emotional support from their teams nonetheless sought this type of support elsewhere and discussed its critical value (see Support Groups section). Only two families were offered regular mental health check-ins as part of their cranio-facial team; two sought mental health treatment outside of their medical team. Interestingly, most parents considered mental health needs to be outside of the scope of the cranio-facial team.

Several adolescents expressed a sense of isolation. They felt treatment could be better informed through an understanding of their pain, fear, and uncertainty. One expressed her parents did not always appreciate her experience of surgery and recovery. Three stated needs for either peer or psychological support in addressing these feelings of isolation and uncertainty. Jennifer stated,

I don’t know if they have this or not but I think they should have like a child psychiatrist who comes in to talk to the kids about their feelings...because a kid can’t just bottle this up their whole life. They have their mom to talk to like ‘this hurts and this hurts’ but not exactly like what they emotionally go through.

**Positive perception of the medical team.** Research demonstrated parents’ satisfaction with their cranio-facial specialists was overwhelmingly positive (Nelson et
In this study, every family expressed positive relationships with providers and overall satisfaction with treatment. Only one family switched teams due to dissatisfaction with care. Three families experienced unexpected medical complications with post-operative care due to poorly managed pain yet remained trusting in the expertise of their medical teams. Every family interviewed mentioned having to repeat a surgery and often believed this was exceptional to their experience. Nelson et al. (2012) found the heightened sense of trust in the medical providers allowed parents to forgive or overlook bad experiences. Reliance on surgical intervention was viewed as a coping skill for parents who experienced anxiety and helplessness (Nelson et al., 2013). Families trusted their providers due to their expertise, care and concern, availability, and the length of the relationship.

[They have] been amazing about consulting with us, making sure concerns that we have are taken care of… If he says something I really do believe- you know he’s been there the whole time. If he tells us that she needs something I still research it but I trust it. I love it that they all come together and say okay…this is how we are going to work together as a team for your child. I think I went along with what they said because they have so much expertise and they all work together to say we’re here and we’re here and work together to make an overall plan for your child. (Lisa)

I can’t say enough about those people. If it wasn’t for them and the speech therapist… he would not be the child he is today. There’s no way he could have endured what he went through and become the type of person he is. (Janet)
Other research has shown a provider’s expertise and continuity of care contributes to a sense of trust (Canaday, Means, Wayne, Thompson, & Richman, 1997). The dependency parents feel on providers has been linked to the idea of obligatory trust (Carnevale, 2004). The duration and quality of these relationships led parents to describe their providers as extended family. Jessie said, “[It has been] very supportive, very positive. It’s like a family…when you’ve seen people for 17 years, they’re family.” Two adolescents also reflected feeling a closeness and warmth toward their providers. One adolescent was very matter of fact about her relationship with her doctors, stating she had great trust in their expertise despite experiencing frustration with many aspects of her care.

**Coping Through Social Support**

Seven out of eight families discussed the critical need for social support in coping with the condition of CL/P and its treatment. Baker, Owens, Stern, & Willmot (2009) found social support was one of the main coping skills mentioned by parents of children with CL/P. Research on resilience also noted the role social support played in helping children adjust to risk factors (Luthar, 2006). Practical support came from extended family as well as older siblings. Parents valued supportive friends who provided acceptance of their children’s condition as well as guidance or advice in coping with the emotional challenges related to it. Anne said, “I mean people in my life have really taken the time to know Anne for who she is, what a beautiful human being she is…so obviously they can look past the differences and see her for who she is.” Two parents felt having siblings with medical conditions helped their children with CL/P feel less isolated and
alienated. Jessie stated, “At this point in time right now where I don’t know what it’s like being a teenager with a cleft, they have each other.”

Adolescents discussed the support of parents in offering guidance and emotional support as well as caretaking throughout the process. Research on disfigurement showed social support and family acceptance are critical in positive adjustment (Thompson & Kent, 2001). Parents normalized and validated their experience and offered problem solving regarding social problems and medical care. They provided perspective as well, which helped their children accept and adjust to problems such as low self-esteem and managing medical treatment. Margaret said, “She didn’t only tell me I shouldn’t feel that way, but she understood what I was saying and going through. I just remember my mom always making me feel super confident which helped a lot.”

One adolescent stated a preference for receiving emotional support from friends in dealing with bullying and self-esteem. One discussed an enhanced relationship with her sibling as a result of her treatment and their joined community outreach work. This participant also discussed the enhancement of her whole family’s connection as a result of the medical journey. Research on cleft conditions and chronic illness found increased willingness to seek social support and improved relationships were a common outcome (Eiserman, 2001; Hastings, Allen, McDermot, & Still, 2002).

All of the families in this study reported acceptance and validation of their children. It is important to remember this might not always be the case. Research on stigma noted any social interaction has the potential to be either stigmatizing or supportive including interactions within the family and with medical professionals (Alansari, Bedos, & Allison, 2013).
Six families noted the importance of involvement in community support or advocacy groups. Two parents formed their own peer support groups. Janet said, “If it hadn’t been for that organization I wouldn’t have handled things as I did. [The doctors don’t know] what the day to day is...they don’t live it the second we walk out of the hospital.”

Research has shown parents frequently seek the support and advice of other parents and are reassured by this advice (McCorkell, McCarron, Blair, & Coates, 2012). One parent created a parent support network for parents of newborn infants and two parents joined large community advocacy groups involved in support, education, and fund raising. Another served as a parent consultant for craniofacial teams. And one adolescent founded a nonprofit organization for families impacted by CL/P. The benefits noted from this involvement included generating community, giving back, increased family cohesion, and gaining support, information, and perspective. Other literature found a desire to give back was a common reaction to this and other chronic health conditions (Stock & Rumsey, 2015). Jessie said, “It opened up opportunities and friendships and a community that we didn’t know. [It is] good for the soul.” And Margaret said, “It made me realize how fortunate I am that I’m able to afford all the surgeries and stuff and definitely puts a perspective on things and makes me realize that it’s not as serious as it could be.”

Interestingly, while parents sought the support of other parents impacted by CL/P, they did not seek similar support for their children but did advocate social inclusion in other ways. One adolescent stated a current desire for more peer interaction and support, which her mother was actively seeking out for her. Another stated she would have liked
to have been part of a support group to feel less alone and to have had more hope going through the process.

**Varied Emotional Impact of the Condition and Its Treatment**

The emotional impact of the treatment process for CL/P was well documented in numerous studies (Collett & Speltz, 2007; Hunt et al., 2005). All families interviewed expressed a variety of emotional responses to their experience. Janet said,

There’s been tears shed and words flying around and there’s been moments when you want to beat anything close but you don’t do that. I get my frustrations out in the car by myself. There would be screaming sessions when nobody’s around to hear me.

Parents noted reactions of guilt, sadness, and anxiety in response to their children’s medical treatments. Paula said, “I feel horrible… I don’t like to think about it… you wish you could take their place but you can’t.” During these times, three parents discussed difficulty balancing their emotions with the role of being a parent. Paula continued,

I try really hard to not be really emotional when she’s really emotional. Because I think it’s my job to show her that it’s okay for her to be scared. I don’t want her to see that I’m scared… so I wait until she goes back and then I fall apart… I want her to see that I’m strong and confident and that I’ll be fine if I’m a nervous wreck along with her she would wonder why. I have to be strong so that she can have somebody she can rely on.

Families also discussed some anxiety about their children’s futures, acceptance, and self-esteem. Two families whose children were impacted by mental health concerns
experienced anxiety and frustration about their children’s emotional stress. While most parents acknowledged their experience eased with the maturity of the children, they still expressed some degree of anxiety with regard to their child’s medical interventions or overall functioning. One parent expressed increased anxiety as their child matured given their increased understanding of the procedures. Even two parents who acknowledged their children coped well with surgeries stated they still felt anxiety prior to surgery.

Literature found the type of concerns parents had for their children with CL/P was relatively stable over time (Lei, Wang, Cheng, Chen, & Chin, 2010).

One parent noted her daughter’s acceptance and compliance aided her emotional stability. Another noted her daughter had a high pain tolerance, although this did not match the daughter’s report of her experience. Two adolescents reported anxiety prior to treatment both in relation to the procedure and recovery and to the uncertainty about the outcome of treatment. Kristin said prior to surgery, “I’m really nervous…like after like how it looks –after… and down the road like is everything going to be done.” Anne stated that she worried: “I don’t remember anything before surgery cause I was really worrying a lot. I don’t remember anything.” Pain and debilitation was worse than expected by two adolescents. Two adolescents experienced anxiety and impatience to be finished with surgeries in order to have better outcomes both aesthetically and functionally.

Two adolescents experienced complex emotions in response to their medical treatment and providers. One expressed anger and resentment about poor communication and pain management but also acknowledged the surgical interventions resulted in improvements in her appearance and functionality. Anne said, “As much as I don’t want
to admit it, I can tell things are getting better. It was the most painful thing in the world but it helped bring the teeth close.” Another experienced both wanting a surgery that would improve her appearance, yet also fearing drastic change. When the surgery did not have a significant impact, she then felt it had not been worth the effort. She said, “Honestly we’d been planning it for years I was always really scared--you’d look different you’d look quote unquote better…and it’s your face too. I go to public school …people were going to ask questions”

Alansari et al. (2013) found some patients with CL/P at the end of the treatment negatively compared the process to the outcomes. Two adolescents appeared to struggle with reconciling the outcomes with the significant costs of undergoing treatment. Yet, none of the parents expressed ambivalence with regard to treatment protocols or outcomes, perhaps due to over-reliance on medical interventions as a means of coping (Nelson et al., 2012).

**Management of the Condition Includes Ongoing Decision-Making**

Research on parental medical decision-making in general found decision-making is highly dependent on context and difficult to predict (Knopf, Hornung, Slap, DeVellis, & Britto, 2008). Decision-making includes the choice of a medical team and the types and timing of interventions. All families chose their medical teams either through recommendation of family or medical doctors or for close proximity. Only one family drove a distance to their medical team and only one family chose a team based on an understanding of an aspect of their treatment protocol. All families utilized large craniofacial teams except one family forced to find independent providers following a
move. One family left a team due to dissatisfaction with pain management and two families switched teams due to geographical moves.

Decision-making for some participants involved planning around family and school schedules. One parent mentioned that while trusting the team, she still felt the need to be fully informed and ask questions. Paula said, “I never blindly do what they say, and I ask bizillions of questions and I research everything. I’m crazy that way. I don’t just go along with it. I’m really anal about researching it.” Other parents were frustrated by setbacks, changes in timing, or types of surgeries, but viewed this as a communication problem rather than one of planning or protocol.

All parents but one felt comfortable in allowing their medical team to make decisions and other than two who questioned recovery management, none expressed disappointment with any aspect of the treatment protocol or its outcomes. For example, one parent stated the protocol had been laid out for her and despite many alterations to this map, she remained comfortable with the team’s decision-making. Survey results indicated parents of children with CL/P felt relatively uninvolved in decision-making (Pannbacker & Scheuerle, 1993). Only one parent declined a surgery and opted to wait for this treatment until adolescence to avoid duplicating the procedure. Nelson et al. (2012) suggested parents’ reliance on medical team’s decision-making might relieve them of the burden of this emotional responsibility as parents might be overly reliant on following treatment advice to feel they are responsibly parenting their child.

In adolescence and even before, many of the surgeries for CL/P are elective and done for aesthetic reasons rather than for critical life functions. These surgeries involve a different type of decision-making as they are influenced by emotional and cultural
factors; the goal is to help the child achieve a more “normal” appearance (Daniel, Kent, Binney, & Pagdin, 2005). Research has shown parents do not typically challenge societal norms when it comes to appearance in terms of acceptance of treatment protocols (Nelson et al., 2012). Numerous studies found parents do challenge this notion of normality internally; however, they fully accept their infants prior to repairs and grieve the loss of their natural appearance (Nelson et al., 2012; Stock & Rumsey, 2015). For parents, there is an internal struggle between acceptance of their child for who they are and recognizing that societal expectations require surgeries to alter them (Nelson et al., 2012).

Two adolescents expressed impatience to be completed with the protocol. Both of these stated they wanted to be better and both had experienced some stigmatizing experiences. Research has linked stigmatizing experiences to a desire for further treatment (Alansari et al, 2013). One parent and one adolescent had or were requesting further revision surgeries outside of the planned protocol, both with the aim of improving aesthetic appearance. Margaret greatly appreciated her parents’ support in making her own decisions:

They are pretty lenient with asking me what I want to have done which I appreciate. I mean there are definite boundaries they wouldn’t let me do anything I would regret. They give me advice because I know they know best and it’s hard to decide when I want everything to change when I’m younger.

While several parents discussed wanting to give their children autonomy with decision-making, the amount given varied. Janet described a strong desire for her son to undergo an elective surgery she felt would improve his appearance but he was not
interested: “Right now I think all of this stuff is a distraction that he doesn’t want to talk about.” She had hoped his doctor would discuss treatment options for him but he had not. Silverman (1983) found tension about these topics hindered open communication between parents and their children’s medical providers. One dyad expressed a discrepancy in decision-making with the parent stating her daughter could make decisions while her daughter stated her parents made the decisions. Lily stated,

I feel like they think it’s necessary when I would have rejected a couple. I feel like its right to let the kids choose mainly because it’s their own body…I was like mom it’s not you how about you go to school with a beat up swollen face see how you feel. Don’t just tell them what’s right.

While parents expressed the desire to allow their children to make decisions, they also struggled with witnessing what they felt were poor choices. Another parent stated recognition that he did not have the power to influence his son’s decisions at his age.

Most adolescents whose parents were responsible for decision-making felt comfortable with this. Jennifer said, “I mean it is what it is. They just know this is good for me and we’re going to do it.”

Reframing and Changing Perspectives

Personal growth as a result of living with cleft lip/palate. The development of new strengths and abilities as a result of stressful situations is a well-known phenomenon (Zimmerman & Arunkumar, 1994). Two parents and two adolescents described such personal growth pertinent to their treatment process and condition. Paula described having increased empathy for others and an increased perspective on the different types of struggles people face:
You know for me I think it has made me a little more empathetic towards people who have children with issues…I’m a really tough person, I try to be, and so I don’t understand people that feel sorry for themselves or their situations. But this kind of let me see a little more into okay they have a kid who needed something. They are sorry for what their child has to go through. Made me more empathetic.

Jan described gaining acceptance of facial differences and changing her perception of beauty:

Years ago, I would see kids with clefts and it would really turn me off…she looked so beautiful to me…and I thought how foolish of me to think I couldn’t adopt a cleft lip child. I’ve really grown in this process…I have grown so much as a person and what I think is beautiful. I was so limited in my thinking before, looking so narrowly.

She and her daughter both said her daughter’s perspective had been altered by her experiences. One adolescent described her own experiences and her work with a non-profit as benefiting her perspective and growth. She viewed her condition as a blessing because it allowed her to do this work through which she gained such perspective.

Margaret said,

There’s so many things I should be grateful for. It might not be the easiest thing in the world but I founded this nonprofit out of it. It definitely helped me care less what other people say about me because I used to and it hurt my feelings a lot and then I realized it doesn’t matter and it’s just not that big a deal anymore.

**Spirituality is impacted by the condition and is a coping skill.** Two parents discussed spirituality as a coping mechanism. Lisa’s religious faith provided a
perspective for her to understand and assist her daughter in understanding the condition and in coping with it:

We try to always just say “who you are is on the inside and how you treat people God loves you exactly as you are. It doesn’t matter what people say about you. If they are mean it’s probably because they have something going on so in their life and they’re trying to make you feel bad so they can have control.” We try to fill her mind with positive things.

Their religious community also provided a source of support in particular to her daughter who stated this community helped her heal after being bullied. Jessie stated that experiences in relation to her children’s CL/P had altered her spirituality, specifically in understanding a larger sense of purpose in her children having been born with this condition:

My sister has this lady who talks to spirit guides. So take this just as a story but it made me okay…this woman…said to my sister “Your sister…is pregnant…its healthy except it has a facial something…there’s another child in the home with the same condition and she called him.

Previously, parents had noted increased spirituality, acceptance, commitment to social justice, and increased empathy were outcomes of the CL/P experience (Eiserman, 2001).

Acceptance and Adjustment as a Complex Process

Four parents specifically mentioned encouraging acceptance and perspective in their children fostered adjustment. Others described helping their children accept the inevitability of the treatment process and felt this acceptance led to more compliance and successful recoveries. Janet said, “You have to be able to take things as they come. It’s a
snowflake, no two cases are the same. You have to be open minded and very patient cause things don’t go according to plan.” Paula added,

We didn’t have a choice. We had to do it so that she’ll be functional throughout her whole life. So it’s not like we were just doing something for the heck of it…now she knows she’s having a big nasty surgery in a year even though she worries about it she knows that she doesn’t have a choice she’s pretty cooperative about it.

Two parents tried to minimize negative reactions through reframing. Paula said, “We’ve never treated it as something odd or something special. It just is. That seems to be how she accepts it. She’s not odd not special it’s just part of her.” Jan stated, “I think you can almost make your child feel inadequate you have to be really careful. This is what’s wrong this is how we’re going to deal with it. Even if it is a big deal to us we try not to make that a big deal to the child.” Two adolescents discussed how acceptance of the need for treatment helped cope with each individual procedure; one even felt that being bullied happened for a reason. Adults with CL/P noted acceptance and perspective as key coping mechanisms (Berger & Dalton, 2009).

Three adolescents cited maturity as a factor in being better able to accept their condition and cope with social struggles. Maturity helped them gain a better understanding of others and be less concerned with appearance. Parents perceived their children coped better as they grew older due to confidence and positive peer groups.

Going back to like kindergarten or second grade, I wasn’t as mature. Like if you make fun of me then I’m going to cry but if you make fun of me now it’s okay like I don’t really care. Like I don’t care what you think about me. Like I’m
different, I know I’m different. I don’t really care what you have to say about me.

(Anne)

Margaret stated, “I just remember thinking I don’t know why I’m self-conscious about it. I’m proud that I’ve been able to overcome it… the surgeries have definitely helped too. But I think it’s just getting older and more mature and realizing that it doesn’t matter.”

There is some evidence a more settled social environment can decrease social difficulties (Turner et al., 1997). One parent whose child was bullied perceived growing older was more difficult as adolescents became more concerned with appearance. While this has been cited as a risk factor, a study found lower quality of life for older children increased self-awareness as the reason for this (Bos & Prahl, 2011). However, this hypothesis contradicted what two adolescents in this study stated about self and other understanding creating ease of acceptance.

**Communication and Honesty**

All eight parents felt open and honest communication was critical to helping their children cope with both the condition and its medical treatment. Janet said,

 Always be up front and honest…whether they understand it or not they deserve to know what’s going on. They just don’t have these surprises…I don’t care how upset they get they need to know. Don’t sugar coat anything. It’s their body and they’re the ones who have to go through it.

They discussed the importance of providing accurate information, listening to their child’s concerns and questions, and validating their experiences. Lisa stated,

 If I learn something I share it with her…by saying, this is what I know, I’ve been in cleft groups online and I’ve seen people go through bone graft surgeries and I
kinda remember the things they’ve said. So just be really open and honest about the things that I had seen, that this is what we can expect.

Hall et al. (2013) noted children’s understanding of this condition is significantly impacted by parents. All parents felt communication helped their children accept treatment recommendations and make better decisions regarding treatment.

Open communication has been deemed critical to decision-making and adaptation (Whitehead, Tobiasen, & Hiebert, 1996). One parent witnessed the ongoing impact of poor communication on her sister-in-law, an adult born with CL/P. She felt this had led to poorer treatment outcomes, more stigmatizing within the family, and was determined to provide a different experience for her child. Two parents noted while hearing about the children’s perceptions of treatment as they became older, more verbal, and self-aware was difficult as a parent, it also allowed for more communication and sharing of these experiences.

**Social Acceptance Is a Concern for Parents**

All but three of the parents had significant concerns about their child’s social acceptance from the time their children were very young. At birth, Anne described, “I was proud of her, but I was torn about what to do…do I want people to come or not because I don’t want them to see her and be like ‘ew’ and not accept her. I couldn’t take that. I want them to love and accept her.” This was cited as a major concern for this population, given the role physical attractiveness plays in societal values and acceptance (Nelson et al., 2011). Factors involved in social adjustment likely include self-acceptance, stigma, social competence, and appearance (Pope et al., 2016). Self-
acceptance is not related to objective understandings of attractiveness but is linked to resilience (Feragen, Borge, & Rumsey, 2009).

Stock and Rumsey (2015) noted earlier research pointed toward greater difficulty in educational achievement, career development, and intimate adult relationships in connection with poor social adjustment. Yet their own research found adults reported successful work and relationship adjustment including committed relationships and parenting (Stock & Rumsey, 2015). While lack of social competence has been associated with reduced friendships and more rejection among adolescents with CL/P, other studies found no significant difference in social competency when compared to norm groups (Collett, Cloonan, Speltz, Anderka, & Werler, 2012; Pope & Ward, 1997).

Despite early concerns, only two parents expressed ongoing concerns about their children’s adjustment based on comments the child made or evident social avoidance. One parent noted his child had withdrawn from social interaction although this adolescent likely had comorbid mental health concerns in addition to negative experiences with social peers due to the CL/P. Both parents reported their children had social anxiety and some social avoidance but did not associate this with the CL/P. One parent denied any negative social experiences. One mentioned minor comments that were handled by the child and his brother and did not cause any negative impact. These parents mentioned peers asked questions and made occasional comments that caused frustration and irritation; one mother thought perhaps this impacted her daughter’s self-confidence.

Berger and Dalton (2009) also found the majority of social problems named in their study took the form of teasing and questioning. The children of these two parents corroborated these impacts and both stated as they grew older, they were less negatively impacted by
these types of comments because they could better understand their origin lay in ignorance rather than ill will. The parents pointed toward positive social experiences, long-term friendships, and positive self-concept of their children as reasons they did not feel the same level of concern.

**Bullying due to facial appearance and speech.** Surprisingly, only two families reported their children were bullied as it was thought to be quite common in this population (Semb et al., 2005). For both children, the bullying occurred in middle school, which was consistent with a study on this population that found the most common period for bullying was early adolescence (Semb et al., 2005). Both families thought the bullying was related to their children’s appearance and possibly speech but did not know the exact nature due to their children’s refusal to discuss the events. One family intervened quite rapidly, involved the school, and sought mental health support for their child in the community. The other parent was less aware of the situation until sometime after it had occurred but had been seeking mental health support for several years in part with regard to these events. Both parents noted a significant and almost immediate impact of the bullying on their children’s mood, self-confidence, and behavior. Lisa said, “To this day she won’t tell me what she said or what she did…It completely changed her. I don’t know…what was done to her at school but she never said anything to me about it…she really is fighting anxiety and depression.” Jeff stated,

> When Aidan was in elementary school, he was the happiest little kid you’ve ever seen, was the light of the party, he would walk into a room and he would light up everybody knew his name and everybody loved him at school and then somewhere in middle school that changed. I think part of it had to do with he was
getting bullied because of his appearance and his speech and things went downhill from there...I say he lost his smile. Up to that point, he always had a smile on his face and after that he very seldom had a smile on his face.

Bullying is a significant predictor of poor self-concept (Hunt, Burden, Hepper, Stevenson, & Johnston, 2006; Murray et al., 2010). Bullying can lead to social anxiety, social avoidance, and negative self-development (Masnari, Schiestl, Weibel, Wuttke, & Landolt, 2013; Rumsey, Clarke, & White, 2003). The adolescent with more significant and comorbid mental health concerns struggled with behavioral problems, mood swings, and social interaction. Richman and Millard (2012) noted adolescents with CL/P are at higher risk for both externalizing behaviors and internalizing behaviors including non-compliance, oppositional behavior, depression, and anxiety. While the parent of this adolescent noted bullying led to drastic changes in his son, he was less certain that bullying was the only factor. Interestingly, the other adolescent reported she felt she had worked through the impact and was even grateful for lessons learned through it, while her mother reported significant concerns about its continued impact.

**Social advocacy seen as key by parents.** Four families described taking an active approach to relieve their anxiety regarding their child’s acceptance among peers. Three parents presented information to their children’s elementary school classes about the condition of CL/P and their own child’s particular experiences including updates about surgical interventions taking place during school. Lisa said,

The very first day I went in and I wanted to talk to the class. I just said you know you’re going to notice several different things about Anne and I wanted to talk to you about those differences...I said “you might notice that she has a scar on her
lip, how many of you have scars” and every hand shot up. So it made it not different and scary, because everybody could relate to having a scar.

Paula taught her daughter to present the information as well as to answer questions or comments assertively: “If you teach your child to educate their peers at a young age, then it becomes a non-issue as they get older.” Another parent supported her child’s social development by being highly involved in both his school and sports activities:

When Frank was in preschool, he couldn’t be understood very well so I had to translate for people at times…not so much of a necessity but to make sure that…I didn’t want him to have to struggle and I didn’t want people to struggle to try to understand him either.

Research also demonstrated that parents of children with CL/P take steps to encourage active involvement with peers (Klein et al., 2010). One parent stated she felt the need to be involved due to not trusting others’ reactions to her son. Some evidence indicates over protectiveness may serve as a buffer for negative reactions in this population (Hutchinson, Wellman, Noe, & Kahn, 2011). All of these parents felt their advocacy had positively influenced their children’s success within their peer group. The parents’ perceptions of their children’s successful adjustment aligned with research that pointed toward positive peer support contributing to positive self-perception and resilience among adolescents with CL/P (Tiemens, Nichols, & Forrest, 2013).

**Recommendations for Mental Health Professionals**

Several implications emerged from this study for both medical and mental health providers who work with this population. Based on the results of this study, counseling
psychologists and other mental health professionals could serve a vital function in providing targeted information and support that would contribute to a successful adjustment process for individuals and families dealing with CL/P. In discussing coping with medical treatment, many participants either stated the desire for the involvement of mental health professionals or discussed emotional needs well suited to the training of mental health professions. A portion of participants experienced symptoms of mental health disorders, indicating the need for a mental health professional to provide regular screenings and referrals for families more at risk.

Based on the findings of this research, counseling psychologists and other mental health professionals should be aware that identity concerns are critical for adolescents with CL/P. Successful adjustment might be aided by supportive therapies that aim to assist adolescents to find balance in terms of their overall identity. Both parents and adolescents requested additional information about treatment. In this era of integrated care, psychologists often play a vital role in assisting patients in a medical setting with understanding and adapting to the demands of medical treatment. Psychologists working with this population could provide psycho-education on the CL/P process, common responses, as well as offer therapy and screenings for those families more at risk. Social support is key for successful coping. Psychologists could screen for adequate support and provide referrals where indicated. Emotional responses to treatment are common including anxiety, guilt, sadness, and depression. Psychologists could provide emotion regulation skills and psychotherapy to assist in coping. Isolation and ambivalence about treatment occur in adolescents despite family support. Group therapy could reduce alienation and provide a context for positive shared experiences. Parents expressed
significant concern about their children’s peer acceptance. Mental health professionals could assist with helping parents navigate this anxiety as well as providing social skills training or therapy for such social concerns. Maturity aids coping for adolescents through providing perspective and improving emotional regulation.

This is a developmental stage for families in which they and their adolescents renegotiate autonomy and decision-making. Parents struggle to allow their children to make decision for themselves regarding treatment options and find it especially hard to witness what they consider to be poor decisions. Because decision-making regarding elective surgeries is a complex process involving personal and societal factors, values, and emotions rather than medical necessity, differences of opinion between family members are to be expected (Nelson et al., 2012). Counseling psychologists and other mental health professionals could assist families in navigating these decisions as well as provide psycho-education regarding satisfaction with treatment, outcomes, and the factors in adjustment.

Individuals and families impacted by CL/P often grow from this experience in terms of increased empathy for others, desire for prosocial activity, and increased family cohesion. Participants described ways in which they reframed their experiences in a positive light and gained in perspective, wisdom, and spirituality. Therefore, the use of a strengths-based perspective with this population is likely to be critical.

While counseling psychologists should be aware most adolescents with CL/P show signs of resiliency, some individuals require more support. Three adolescents in this study experienced symptoms of mental health disorders. Psychologists could provide screening, psychotherapy, and determine the role of the CL/P in such difficulties.
Both adolescents and parents expressed experiences in which they felt stigmatized because of the condition of CL/P. The subtle presence of stigma was noted in the preference for surgical treatment over other forms of adaptation such as supportive psychotherapy (Nelson et al., 2012). Counseling psychologists and other mental health professionals could advocate for this population in medical settings, school settings, and assist parents in advocating for their children. United efforts between counseling psychologists and medical teams could provide support to patients and their families in an effort to de-stigmatize this condition.

**Discussion and Conclusion**

Many studies found a percentage of individuals with CL/P were at risk for difficulties such as social acceptance, behavioral problems, and/or emotional distress (Hunt et al., 2005). Individuals born with CL/P often undergo medical treatment for the condition from infancy through late adolescence if not throughout their lifespan. The burden of medical care impacts not only the individual child but the entire family system (Baker et al., 2009). Research on risk factors and the adjustment process to the condition of CL/P found highly inconsistent results—in part due to the inconsistency of the research designs and measures utilized and in part due to the multifactorial nature of the adjustment process itself (Rumsey & Stock, 2013). Resilience literature noted individuals often simultaneously experienced risk in some areas and resilience in others (Luthar, 2006). The concept of resilience is a relative one dependent upon culture and context (Ungar, 2003). Because of this, several authors have argued that qualitative research with its focus on complexity, contextual factors, and emergent designs is best suited to explore this area (Eiserman, 2001; Ungar, 2003). The goal of this study was to
understand the experiences of both parents and adolescents with the impact of the condition as well as its treatment and to understand how both individual and family coping strategies contributed to successful adjustment. Patient perspectives are the foundation of patient-centered care; therefore, it is my hope the results might support medical providers who work with this population (Canady, 1995).

Qualitative research does not aim to be generalizable; rather, it provides an accurate and faithful representation of the subjective experience of a participant or group of participants (Creswell, 2007). Interview questions were designed from the perspective of the researcher and might not have addressed particular perceptions and experiences of the participants. While the interviews were open-ended and attempted to elicit participants’ unique perspectives, it was possible they did not share aspects of their experiences due to the structure of the interview questions.

I am the mother of a child who was born with CL/P. Despite my attempt to bracket my own experience, my familiarity with the subject might have influenced my understanding of participants’ reflections. My experience might have also created a sense of comfort for participants to share their experiences with me.

Despite efforts to include a diverse sample of parents and adolescents, males and individuals of varied ethnic and socio-economic backgrounds were not well represented in this study. Given a large proportion of the participants are involved in community support or outreach and reported a desire to help others in the CL/P community, this might have impacted their desire to participate in the study. The experiences of these individuals and families might have overly reflected the positive experiences of social support and advocacy than is warranted in the more general CL/P population.
A limitation of this study was the interviews all took place over the telephone due to geographical distances between the participants and myself. Research suggested strengths in telephone interviews, although there were few empirically based comparisons between face-to-face and telephone interviews (Irvine, Drew, & Sainsbury, 2013). Advantages of telephone interviews included increased ease with discussing sensitive topics and greater accessibility and cost effectiveness (Chapple, 1999). It might be more difficult to develop rapport over the telephone without the sharing of food and drink, small talk, and social cuing offered through body language (Opdenakker, 2006). However, a recent study pointed to several challenges in telephone interviews that might have impacted data collection. Irvine et al. (2013) noted participants make more frequent checks regarding their comprehension of questions as well as the adequacy of the response they offer and researchers tend to offer fewer verbal acknowledgements than in face to face interviews. Their findings also noted a tendency for participants to provide less detail, resulting in shorter interviews (Irvine et al., 2013).

Further research is needed to understand more about those individuals and families at risk and those who thrive with the condition of CL/P. Qualitative studies have illuminated the complexity of the process of adjustment and the need to consider the dynamics and contextual factors inherent in the treatment process. Continued research is needed to highlight the unique pathways to successful adjustment. Most of these studies explored the experiences of individuals who volunteered to share their experiences and had access to researchers. The experiences of individuals of diverse ethnic and cultural backgrounds and those of lower socio-economic class are not often represented. Finally,
research on psychological intervention is only in its infancy as is its integration into cranio-facial team care. As Lily stated,

I feel like the whole thing has affected my life. It’s made me think about myself differently like what would have happened if I didn’t do or I did do that. I honestly think I don’t have any choice about any of the surgeries that was just that --but if I could have taken the surgeries out I could’ve been a little happier I guess. It kinda makes you a little insecure you know there’s something not right and you’re working on it. It’ll have to be something I just live with my whole life of course.
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