HEALTH-RELATED QUALITY OF LIFE FOR CHILDREN WITH CLEFT LIP
AND/OR PALATE

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Abstract

A child’s health-related quality of life (HRQOL) is a multidimensional, subjective construct that is perceived as a result of the impact that a condition has on the child’s physical, emotional, social, and cognitive functioning (WHO, 1995). This study measured HRQOL for a culturally diverse sample of children with cleft lip and/or palate to include parent proxy reports and child self reports. A descriptive quantitative design was utilized. One hundred subjects participated in the study, including 62 parents and 38 children who completed the PedsQL instrument. Results revealed that parents of children with an oral cleft rated their children’s Physical Health higher than a normative sample of parents of healthy children which was an unexpected finding. Otherwise, parents of children with an oral cleft and the children themselves rated their Emotional Functioning, Social Functioning, School Functioning, overall Psychosocial Functioning, and Total HRQOL lower than healthy children and higher than children with a chronic condition. Parent-child concordance was quite low in all domains with the highest correlation being only .20 on the Emotional Functioning scale. Demographic variables, speech impairment, and hearing severity did not affect HRQOL scores. Wearing a hearing aid had a significant affect on School Functioning according to the parent’s perspective. Overall, the PedsQL was found to be an effective and quick measure of HRQOL for children with an oral cleft that could be utilized during the multidisciplinary cleft clinic to identify children who are at-risk for impaired HRQOL. Since parent-child agreement was low, children should be given every opportunity to rate their own HRQOL. Once individual concerns have been identified, health care providers can then implement care and treatment to maximize the child’s satisfaction with health and well-being.
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Chapter 1: Introduction

For over half of a century, nursing literature has focused on the care of children with oral clefts with the primary purpose of establishing optimal health-related quality of life (HRQOL). Post and Mason (1948) stated, “The nurse caring for a baby with a cleft lip knows that the infant has a better chance to approach his or her life in a normal manner as a result of the surgery and the careful nursing care which he has received (p.770).”

Despite tremendous advances in physical care and shifts in paradigm related to the psychosocial care of children who were born with oral clefts, achieving normalcy continues to be the overall goal. “The ultimate goal of cleft care is restoration of the patient as far as possible to a “normal” life, unhindered by handicap or disability” (Shaw, Semb, & Nelson, 2009, p. 2048).

This paper will provide an overview of cleft lip and palate, review normal growth and development in children, and discuss health-related quality of life in children overall as well as for children with cleft lip and/or palate. In addition, a conceptual model is presented that illustrates factors impacting health-related quality of life for children with cleft lip and/or palate. Finally, the paper will report the findings from this descriptive study focusing on health-related quality of life in children with cleft lip and/or palate.

Statement of the Problem

Orofacial clefts are one of the most common major birth defects (Spritz, 2001). Oral clefts are visible and stigmatizing defects which may affect a child’s physical and psychosocial development (Kapp-Simon & Gaither, 2009) and affect their overall health-related quality of life. For the child who was born with an oral cleft, a quantitative measurement of health-related quality of life may provide insight to nurses and other
health care professionals as to the child’s or parent’s perception of the child’s physical and psychosocial functioning.

Specific Aims and Research Questions

The overall goal of this study was to understand perceptions of health-related quality of life in children with cleft lip and/or palate. This goal was achieved through the exploration of four specific aims.

Specific Aim 1

To examine health-related quality of life of children with nonsyndromic oral clefts according to age category and compared to normative data of healthy children and children with a chronic condition as measured with the PedsQL instrument to include parent proxy reports for children ages 2 to 12 years and child self reports for children ages 5 to 12 years. The corresponding research questions for aim 1 are:

1. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life according to each age category.

2. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life according to each age category?

3. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of parents of healthy children?
4. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of healthy children?

5. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of parents of children with a chronic condition?

6. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of children with a chronic condition?

**Specific Aim 2**

To determine if there is parent-child agreement (concordance) between parent proxy reports and child self reports for 5 to 12 year old children with nonsyndromic cleft lip and/or palate when rating Physical Health, Psychosocial Health, and Total health-related quality of life as measured by the PedsQL. The corresponding research question for aim 2 is as follows:

7. Is there parent-child agreement between parent proxy reports and child self reports for 5 to 12 year old children with nonsyndromic cleft lip and/or palate when rating Physical Health, Psychosocial Health, and Total health-related quality of life?

**Specific Aim 3**

To examine if there are differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate that are attributed to demographic variables including gender, age,
and race of the children; and the geographic residence, educational level, income, marital status, and age of the parents? The corresponding research question for aim 3 is:

8. Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate that are attributed to demographic variables including gender, age, and race of the children; and the geographic residence, educational level, income, marital status, and age of the parents?

Specific Aim 4

To examine if there are differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate that are attributed to impaired speech or hearing. The corresponding research questions for aim 4 are:

9. Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate who do or do not have impaired speech?

10. Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate who do or do not have impaired hearing?

Significance of the Study

Healthy People 2020 focuses on improving the health of individuals, communities, and the Nation. The proposed study addresses two of the four overarching goals of Healthy People 2020 as follows: 1) “Promote quality of life, healthy development, and healthy behaviors across the life stages.” The measures of progress for this goal include
measurements of well-being/satisfaction; and measures of physical, mental, and social health-related quality of life, and 2) “Achieve health equity, eliminate disparities, and improve the health of all groups” (Healthy People 2020). The proposed study will generate new knowledge to enhance an understanding of health-related quality of life for culturally diverse children with cleft lip and/or palate.

Children with oral clefts usually have higher morbidity and mortality throughout life compared to their unaffected peers (Mossey, Little, Munger, Dixon, & Shaw, 2009). Further research is needed on health-related quality of life in children to understand health status and to identify the impact of illness and treatment in children (Matza, Swensen, Flood, Secnick, & Leitdy, 2004).

This study will increase an awareness of the importance of incorporating health-related quality of life measurement into the multidisciplinary cleft clinic. There are multiple reasons to incorporate health-related quality of life measurements for children in clinical practice as described by Varni, Burwinkle, and Lane (2005). First, if children have the opportunity to complete a health-related quality of life instrument, the patient-health care provider communication may be enhanced. Children may not have the verbal skills to articulate their symptoms or feelings verbally. When provided with a developmentally appropriate measure of health-related quality of life, an overall picture of both positive and negative aspects can be communicated, and the health care provider can intervene if problem areas are identified. Second, it is likely that parents of pediatric patients will report their satisfaction with care based on their perception of their child’s health-related quality of life secondary to treatment. Third, by providing a health-related quality of life measurement as a standard screening instrument, physical and
psychological health concerns for the child and parent may be identified that may otherwise be overlooked. Fourth, identifying health-related quality of life issues may influence clinical decision making for the health care provider. Lastly, incorporating health-related quality of life measurement into routine clinical practice may result in improved patient outcomes over time.

In addition to the importance of health-related quality of life measures in clinical practice, the subject matter of health-related quality of life for children who were born with an oral cleft continues to be a research topic of primary importance. The National Center on Birth Defects and Developmental Disabilities at the Centers for Disease Control and Prevention has identified the area of health-related quality of life for children and adolescents with cleft lip and palate to be a research priority. Gaps in the literature have been recognized, and the topic of health-related quality of life for children and adolescents with orofacial clefts was reported as being a priority with regard to public health importance, feasibility, and outcomes of interest (Yazdy, Honein, Rasmussen, & Frias, 2007).

The focus of this study is congruent with the areas of research emphasis for Healthy People 2020 and The National Center on Birth Defects and Developmental Disabilities by enhancing knowledge related to understanding the effect that cleft lip and/or palate has on a child’s health-related quality of life. This study will add to the body of nursing knowledge by increasing an understanding that the impact of being born with an oral cleft has on a child’s physical and psychosocial functioning. This study includes multiracial and multiethnic populations and generates new knowledge related to various physical and psychosocial variables with an Asian and Pacific Island population.
to further the goal of reducing health disparities. In addition, this information lays the groundwork for establishing interventions to improve the overall of health-related quality of life for children with cleft lip and/or palate as well as provides valuable information for future development of a condition-specific instrument to measure health-related quality of life for children who were born with an oral cleft.

To achieve the study aims and to answer the research questions, a descriptive quantitative design was conducted to measure health-related quality of life for children with cleft lip and/or palate to include parent proxy reports and child self reports. This study also describes how specific demographic variables, speech, and hearing impact health-related quality of life for these children. The conceptual model for this study has been emulated from Barnard’s Child Health Model (Barnard & Eyres, 1979) and the PedsQL (Varni, 1998).
Chapter 2: Literature Review

The literature review includes published papers that address health-related quality of life or quality of life for children with orofacial clefts as well as those addressing developmental considerations. The primary focus of this literature review includes health-related quality of life for children with oral clefts from 2 to 12 years of age. No studies could be found in the literature that addressed health-related quality of life for infants or children with oral clefts under 2 years of age from the parent’s perspective. Literature on health-related quality of life for adolescents and adults with cleft lip and/or palate was not addressed in this literature review. Literature that focused on children with facial differences other than oral clefts was not usually addressed unless relevant.

The paper trail for this literature review was completed according to the guidelines described by Garrard (2004). Key words and phrases that were used for the searches included the following: cleft lip, cleft palate, health-related quality of life, quality of life, children, etiology, incidence, embryology, growth and development, conceptual model, and theory.

The electronic bibliographic databases that were utilized and searched included PubMed, CINAHL, and PSYCHINFO. The review of literature covered a 62 year period from 1948 to 2011. A total of 107 journal articles were selected for the literature review. Several of the articles of interest were obtained from reviewing the reference lists from pertinent articles. Twenty-five text books were utilized which included books on oral clefts, child development, nursing theory, pediatrics, nursing research, literature reviews, and quality of life, and statistics.
Cleft Lip and Palate

Orofacial clefts can be classified as cleft lip, cleft palate, or cleft lip and palate. Although each of these congenital anomalies are different, epidemiologic studies frequently group them together since they are pathologically similar (Genisca, et al., 2009).

Embryology of Cleft Lip and Palate

On the 28th day postconception, the orofacial region can be identified in the embryo. From the 4th week after conception to the very early fetal period, cleft lip and palate occur from different developmental variations (Peterson-Falzone, Hardin-Jones, & Karnell, 2010). The development of the upper lip is complete by 7 weeks postconception, and the development of the palate is complete by 9 weeks postconception (Mitchell, 2009).

Upper lip formation occurs as a result of cell proliferation and tissue configuration. In order for the upper lip to form completely and in an intact manner, fusion must occur of the bilateral maxillary and two medial nasal prominences with the lateral nasal prominences excluded but wedged in between (Sperber & Sperber, 2009). Incomplete fusion of the embryonic structures that surround the primitive oral cavity result in cleft lip (McCormick, Mackey, & Wilson, 2007) which occurs from inadequate neural crest tissue migration to the lip area (Sperber & Sperber, 2009). The extent of the cleft can vary from a notch in the vermilion border to a deep, wide fissure extending to the nostril (McCormick, Mackey, & Wilson, 2007).

There are three embryological structures that compose an intact palate. These elements are derived from the median frontonasal prominence, the primary palate, and
the two lateral palatal shelves of the maxillary prominences. Cleft palate can result when fusion of the palatal shelves on either side fails to occur during embryologic development (Sperber & Sperber, 2009). The degree of cleft palate varies. The cleft may only occur in the soft palate or it may extend to the hard palate. Cleft palate may occur only in the midline of the posterior palate, or in more severe cases may extend to the nostril on one or both sides. Complete absence of the nasal septum may occur with wide central palatal clefts (McCormick, Mackey, & Wilson, 2007).

**Incidence of Cleft Lip and Palate**

Orofacial clefts of the lip and palate occur in approximately 1 per 700 births worldwide (Murray, 2002). Overall, the highest incidence of cleft lip and palate occurs among Asian or American Indian populations with a frequency of 1 per 500 births or higher. The Caucasian population has an intermediate incidence, and the African populations have the lowest incidence of approximately 1 per 2,500 (Murray, 2002).

Of the children who are born with cleft lip, approximately 50% also have cleft palate. Approximately 70% of children who have cleft lip or palate are nonsyndromic which refers to the child not having any other anomalies (Stanier & Moore, 2004).

The National Birth Defects Prevention Study (NBDPS) identified 3,344 infants with nonsyndromic oral clefts from 1997-2004. Of the 3,344 infants, 751 had cleft lip, 1,399 had cleft lip and palate, and 1,194 had cleft palate only with birth prevalence estimates at 0.3, 0.5, and 0.4/1,000 live births respectively (Genisca et al., 2009). For infants who had cleft lip and palate, approximately twice as many had unilateral versus bilateral involvement. For infants who had cleft lip, over 10 times as many had unilateral versus bilateral involvement. The majority of infants had left-sided involvement. Of the infants
who had cleft palate, approximately 25% had Pierre Robin sequence. The NBDPS was a multi-site, population-based, case-control study conducted with the purpose of identifying genetic and environmental risk factors for various birth defects. The NBDPS was the largest study in the United States for infants with oral clefts (Genisca et al., 2009).

Various factors were associated with nonsyndromic oral clefts in the NBDPS study. These factors included infant gender, maternal race and ethnicity, maternal age, and gestational age. With regard to infant gender, the incidence for cleft lip was higher among males, the incidence for cleft lip and palate was almost double for males than females, and the incidence for cleft palate was higher among females. The racial and ethnic categories only included Hispanic/non-Hispanic and black/white. The prevalence for all oral clefts was lower among the non-Hispanic blacks. The prevalence was lower for cleft lip and cleft palate for Hispanics.

For maternal age, the only result that was statistically significant was for mothers in the 30 to 34 year old group; whereby, the birth prevalence was lower for cleft lip and palate. With regard to gestational age, preterm birth at 20 to 36 weeks gestation was associated with cleft lip and palate and cleft palate (Genisca et al., 2009).

A global registry and database of craniofacial anomalies was created under the World Health Organization. The registry, known as the International Database on Craniofacial Anomalies (IDCFA), was created to assist with clinical and etiological research. The IDCFA includes 62 registries covering 2 million births per year as of March 2006. The IDCFA has listed Hawaii as the seventh highest area of increasing rates of oral clefts with a rate of 14.35 per 10,000 births (Mossey, 2007).
In the state of Hawaii, there were 681 infants/fetuses identified with oral clefts from 1986 to 2003 according to the Hawaii Birth Defects Program (HBDP). Oral clefts were categorized as isolated cleft palate or cleft lip with or without cleft palate. Of the 681 infants/fetuses, 249 had an isolated cleft palate, and 432 had cleft lip with or without cleft palate. The rate of oral clefts in Hawaii is 19.1 per 10,000 births (Merz & Forrester, 2004) which differs from the statistics provided by Mossey (2007); however, the HBDP was comprised of data to include all identified clefts, as opposed to live births only.

The HBDP statistics included the rate of oral clefts per 10,000 infants. With regard to infant gender, overall more males had oral clefts with a rate of 21.9 (n = 377) than females with a rate of 18.1 (n = 294). For maternal ethnicity, the highest incidence occurred among the Chinese group with a rate of 29.0 (n = 36), followed by Samoans with a rate of 22.2 (n = 21). The incidence for Japanese was 25.1 (n = 104), Filipino 20.6 (n = 128), and Hawaiian 19.9 (n = 168). The lowest incidence was for Caucasians at 15.7 (n = 129) and Blacks at 14.8 (n = 16). For maternal age in Hawaii, the highest incidence of oral clefts was for women ≥ 40 years of age with a rate of 35.8 (n = 28). The incidence of oral clefts was almost half for younger mothers. The lowest incidence was for women ≤ 19 years of age with a rate of 18.9 (n = 63). For gestational age, the highest rate of oral clefts occurred in the 31 – 33 week age group with a rate of 63.0 (n = 26), and the lowest incidence was for infants born at ≥ 41 weeks gestation with a rate of 13.5 (n = 63) (Merz & Forrester, 2004).

**Etiology of Cleft Lip and Palate**

Researchers continue to work toward the goal of identifying the causes of cleft lip and palate. There is currently no single cause that has been identified for oral clefts. Current
models continue to focus on predisposing factors which include genetic, environmental, or a combination of the two that may result in clefting (Peterson-Falzone, Hardin-Jones, Karnell, 2010). Oral clefts can be identified as nonsyndromic or syndromic types. Nonsyndromic oral clefts occur in individuals who do not have any other physical or developmental anomalies. Syndromic clefts can occur as a result of chromosomal syndromes, teratogens, or uncategorized syndromes (Murray, 2002). Over 300 syndromes are associated with oral clefts. Cleft palate is more frequently associated with syndromes than cleft lip with or without cleft palate (Arosarena, 2007). Nonsyndromic oral clefts may be etiologically and pathogenetically different than syndromic clefts (Genisca et al., 2009).

Various factors and exposures have been associated with the risk of orofacial clefts; however, further research is needed to ascertain those that are factual. Risk factors that may predispose the embryo to oral clefting include family history/genetics, maternal use of medication, maternal diseases, maternal characteristics and behaviors, nutrition, and exogenous exposures (Mitchell, 2009).

There is a strong genetic component involved in the etiology of oral clefts. In monozygotic twins, concordance rates are 40% to 60% which indicates a genetic etiology; however, since concordance is not 100% other causes must also be responsible for oral clefting (Murray, 2002).

The use of maternal medications such as retinoids, anticonvulsants, folate antagonists, benzodiazepines, and corticosteroids may increase the risk or oral clefting in the embryo. Certain maternal diseases such as diabetes mellitus, gestational diabetes, along with several other disease states may increase the risk oral clefts. Maternal characteristics and
behaviors including smoking, alcohol consumption, obesity, stress, and other additional maternal characteristics have been associated with the risk of oral clefts. Maternal nutritional factors including an intake of very high doses of Vitamin A may increase the risk of oral clefts, while the intake of folic acid may reduce the risk of clefting. Additional research is needed to confirm or dispel the various etiological factors related to cleft lip and palate (Mitchell, 2009).

Prevention of oral clefts includes the avoidance of smoking, alcohol, and anticonvulsants when possible based on the risk of withdrawal for the mother. Preliminary reports indicate that folic acid and vitamin B6 may help with the prevention of oral clefting. In addition, it is recommended that women should take prenatal vitamins preconceptually and during the pregnancy (Murray, 2002).

In conclusion, the embryological, incidence, and etiological factors for cleft lip and palate have been reviewed. Cleft lip and palate continues to be prevalent throughout the world. Variations of the defect may occur during the first trimester of embryological development. At this point in time, no definite specific etiology has been identified for cleft lip and/or palate, although various predisposing factors have been acknowledged in the literature. A synthesis of the literature is provided which will focus on health-related quality of life for children who were born with cleft lip and/or palate.

**Growth and Development of Children**

When evaluating health-related quality of life, physical and psychosocial aspects of the child’s functioning are considered. Physical functioning for a child may include the child’s gross motor skills, ability to participate in sports or exercise, ability to perform independent self-help skills such as taking a shower, routine activities of daily living such
as completing chores, the presence or absence of pain, and the child’s energy level. Psychosocial functioning aspects include emotional, social, and school functioning (Varni, 1998).

**Growth and Development of the Toddler**

**Overview of development.**

The toddler period represents the end of infancy as the child becomes more independent and an active participant in family life (Dixon & Stein, 2006). The toddler period is at times referred to as, “the terrible twos.” Children of this age are engaged in intense exploration and active learning. Temper tantrums, negativism, and obstinacy are common during this period (Wilson, 2007).

The psychosocial stage of the toddler according to Erikson (1963) is autonomy versus shame and doubt. This stage of development is characterized by the child’s sense of asserting control and autonomy. The child is determined to perform tasks independently and to be self-sufficient. The child may say “no” even when meaning “yes” as a way of asserting independence. When the child is encouraged to use newly acquired skills, the child becomes autonomous. If the child is made to feel inadequate when attempting new skills, is overly restricted, or harshly reprimanded, the child may become ashamed and experience a sense of doubt (Erikson, 1963; Muscari, 2005; Trawick-Smith, 2000).

The cognitive stage of development during the toddler period is the preoperational phase according to Piaget (1969). During this period, the child begins to form concepts, make simple classifications, engages in transductive reasoning, and exhibits egocentric thinking (Muscari, 2005). A toddler’s cognitive thinking is related to their perceptions of an event. Toddlers have the ability to problem solve based on their observations,
primarily what they see and hear directly as opposed to what they recall about objects and events (Wilson, 2007).

**Physical functioning.**

During the toddler period, children become more competent with locomotion. They usually have the ability to run fairly well with a wide stance, jump with both feet, and pick up objects without falling (Wilson, 2007). Children of this age may stumble frequently, especially when changing directions or stopping suddenly (McDevitt & Ormrod, 2002), and they are quite active (Dixon & Stein, 2006).

Toddlers exhibit increased independence from the mother or caregiver. By approximately 2.5 years of age, the child can usually put toys away after play (Hockenberry, 2004) and may be able to perform increased self-help skills such as putting on clothing, brushing their teeth, and washing and drying their hands (Denver II, 1990).

**Psychosocial functioning.**

Toddler-aged children may exhibit temper tantrums as a means of asserting independence. The toddler begins to develop awareness that their feelings and desires may be different from others. Children of this age have a desire to share their experiences with others and may pull people over to show them something of interest (Wilson, 2007). Children of this age typically engage in parallel play, whereby the child plays along side of others as opposed to with others. Toddlers may experience separation anxiety and stranger anxiety. Common fears of this age include loud noises, going to sleep, and large animals (Muscari, 2005). Toddlers may begin to experience life outside of the home by
beginning in a daycare or preschool where they are exposed to various differences related to structure and routines as well as additional adults and children.

**Growth and Development of the Preschool-Aged Child**

**Overview of development.**

The preschool period is marked by continued growth and maturity. Children of this age continue to become more independent in terms of personal and family life skills. During this time, children have a tendency to try to please their parents and conform to parental expectations as well as become less jealous of younger siblings (Hockenberry, 2004).

Erikson (1969) referred to the psychosocial stage of the preschooler as initiative versus guilt. During this stage, children play and work zealously and feel a sense of accomplishment and satisfaction from their activities. If children believe they have not behaved appropriately, they may experience a sense of guilt. These feelings of guilt may be accompanied by anxiety and fear (Erikson, 1963; Monroe, 2007).

The cognitive stage of development for the preschooler continues to be the preoperational phase which covers the span from 2 to 7 years of age. The preconceptual stage occurs from 2 to 4 years of age, and intuitive thought occurs from 4 to 7 years of age. During the preconceptual stage, the child is egocentric in thought and behavior, begins to understand time, and starts to have the ability to view concepts from another perspective. In the intuitive stage, children begin to become less egocentric, have increased social awareness, are more able to view other’s perspectives, and tolerate differences although they may not necessarily understand them (Piaget, 1969; Wilson, 2007) which may account for changes in their socializations skills during this period.
Physical functioning.

Preschool-aged children continue to develop more coordinated gross motor skills. They become more competent at climbing up and down stairs. Preschool-aged children are typically also able to leap, gallop, hop, and run in a more controlled manner (McDevitt & Ormrod, 2002).

Children of preschool age are usually able to perform self-help skills such as feeding, bathing, toileting, and dressing by the age of 4 years. They are also usually able to perform household chores such as simple cleaning and caring for their toys (McDevitt & Ormrod, 2002). Preschoolers also show significant strength and display tireless energy (Trawick-Smith, 2000).

Psychosocial functioning.

Preschool-age children know their own gender and the gender of others. They tend to be selfish and impatient at times, may experience mood swings, and display physical and verbal aggression. By age 4, the child usually becomes less rebellious and quarrelsome, and the child may display better manners. Play becomes associative and children enjoy imaginative play and begin to play games with simple rules (Monroe, 2007). Common fears for this age group include the dark, animals, imaginary creatures, masks, people with scars or deformities, and aggressive actions and threats (Dixon & Stein, 2006).

Children of this age frequently begin to attend preschool. Social competence refers to the child being liked by others as well as having the skills to effectively interact with others. As young as preschool-age, children may be “popular” and well-liked by others or “rejected” and considered to be an undesirable playmate which is based on various social and personal characteristics (Trawick-Smith, 2000).
Growth and Development of the School-Aged Child

Overview of development.

The middle years refers to children from 6 to 12 years of age. The school-aged child experiences significant changes in motor, social, and cognitive development. During this period, the child is strongly influenced by their school experience which has an impact on their development and relationships (Rodgers, 2007).

Erikson (1963) referred to the psychosocial phase of development for this age group as industry versus inferiority. During this period, the child has a desire to achieve and master tasks. During this time, the child strives to engage in tasks and activities which can be accomplished and completed. Since children of this age are often engaged in activities with other children, the child learns about rules, competition, and cooperation to achieve goals (Erikson, 1963; Muscari, 2005).

The cognitive stage of development for 7 to 11 year old children according to Piaget (1969) is known as concrete operations. During this stage, inductive reasoning, logical operations, and reversible concrete thought become the norm. Children are also able to understand the principle of conservation during this stage as well as classification. School-aged children transition from egocentric thought processes to objective thinking and begin to understand another person’s point of view. Children focus on the present physical reality as opposed to the future during this period (Piaget, 1969; Muscari, 2005).

Physical functioning.

The school-aged child continues to have steady increases in height and weight. During this period, their gross motor skills become refined and consolidated. The school-aged child may integrate their physical skills into play activities such as participation in
organized sports (McDevitt & Ormrod, 2002). School-aged children are typically very active and engage in constant activity. They have the ability to jump, chase, and skip (Rodgers, 2007). Pubescent changes may begin to appear for girls during the later school-age period (Hockenberry, 2004).

School-aged children exhibit increased independence regarding self-help skills. By age 6, children are usually able to take a bath without supervision. By ages 8 or 9, children may assume responsibility to participate and help with household chores such as dusting and sweeping (Rodgers, 2007).

**Psychosocial functioning.**

School-aged children typically experience a period of less intense emotions and are more even-tempered prior to entering into the adolescent period. Children of this age usually begin to identify with peers and become more perceptive regarding sex-role learning. Boys associate with boys, and girls tend to associate with girls, while each gender group pursues their own interests. Friendships and “best friends” are established with school-aged children, and clubs and peer groups take on significant meaning. Children become more aware of social norms and pressures of the peer group and may choose to modify their behavior to gain peer acceptance (Rodgers, 2007). Children this age typically enjoy family relationships, aim to please, and respect their parents. Body image and self-concept are more fully developed. Children evaluate their appearance and compare themselves with others. The head is the most noticeable and important part of the body to this age group. Children strive to be successful in school during this period (Rodgers, 2007). Common fears for this age group include school failure, personal failure, and bodily harm (Dixon & Stein, 2006).
The Child with a Chronic Condition

Definition of Chronic Condition

The Research Consortium on Children with Chronic Conditions was established in 1980 which has influenced research, clinical practice, and policy for children with special health care needs. Definition issues were later described that have become the basis for work in the field (Pless, Stein, & Walker, 2010). Stein and colleagues (1993) defined a chronic condition as one that would produce one or more of the following sequelae: limitation of functions appropriate for age and development, disfigurement, dependency on medication or special diet for normal functioning or control of condition, dependency on medical technology for functioning, need for more medical care or related services than usual for the child’s age, or special ongoing treatments at home or in school (Stein, Bauman, Westbrook, Coupey, & Ireys, 1993). Perrin and colleagues (1993) stated that although no single definition of chronic conditions fits all purposes, they recommended that a condition be considered chronic if it has lasted or is expected to last more than 3 months. The authors also stressed the importance of considering the impact that the condition has on the child, such as the level of impairment related to functioning.

Impact of a Chronic Condition on the Child

Children who have a chronic illness are at risk for difficulties which are related to physical as well as psychological stressors. Physical stress may include pain, not feeling well, or the need to endure unpleasant procedures. Psychological sequelae of a chronic illness may encompass feeling or being different, separations secondary to hospitalization, inability to meet developmental milestones similar to healthy peers, and real or imagined fears related to mortality (Northam, 1997).
Vessey and Rumsey (2004) described the impact of chronic conditions on child development. Factors of importance include characteristics of the condition such as the severity, visibility, prognosis, and iatrogenic insults (Patterson & Blum, 1996). The severity of the chronic condition as well as a prolonged disease state can alter a child’s development. Also, when a child has a chronic condition that is visible, the child may experience stigmatization which can affect the child’s psychosocial development. Physiological and psychological aspects of the child’s prognosis may also impede a child’s development. Iatrogenic effects which occur from various treatment regimes may affect the child’s developmental progress as well.

Other significant factors related to the impact of the chronic condition include characteristics of the child such as age, developmental level, and individualism. Toddler-aged children may experience regression associated with a chronic condition. The condition may pose limitations on the child in terms of the child not having the ability to achieve autonomy which may further hinder development at this age. Preschool-aged children may not have the energy to take initiative secondary to the chronic condition which is a developmental milestone of this age. The condition may also interfere with the child’s development of self-concept and body image. The school-aged child who is typically industrious may experience negative affects on development related to a chronic condition such as not having the opportunity to progress socially and achieve tasks that the child has set out to master (Vessey and Rumsey, 2004).

In summary, growth and development of the toddler, preschooler, and school-aged child encompasses the child’s physical, psychosocial, and cognitive realms. A child with a chronic condition may experience alterations in growth and development. How the
child’s condition impacts their life and level of functioning has overall implications related to the child’s overall health-related quality of life.

**Health-Related Quality of Life**

The terms quality of life and health-related quality of life are at times used interchangeably in the literature. The World Health Organization (WHO) is the primary authority for directing and coordinating health in the United Nations (WHO, 1995). The WHO defines quality of life as an, “individuals' perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” (WHO, 1995). Quality of life encompasses a broad concept which includes several facets including physical health, psychological state, level of independence, social relationships, and their relationships to features of their environment (WHO, 1993).

The WHO constitution defines health as, “a state of complete physical, mental and social well being and not merely the absence of disease” (WHO, 1995). The term health-related quality of life has become known as an appropriate term for, “… dimensions that represent the patient’s perception of the impact of an illness and its treatment…” (Varni, Limbers, & Burwinkle, 2007, p. 2).

Health-related quality of life for adults is different than health-related quality of life for children due to difference in developmental levels. The division of Mental Health of the World Health Organization (1993) suggested a list of facets which may be utilized to develop an instrument when measuring quality of life in children. The list includes the domains of family/social relationships, physical function, psychological aspects, physical
appearance, psychosocial relations to social and material environment, and the environment.

**Children, Chronic Conditions, and Health-Related Quality of Life**

Children who experience a chronic condition may perceive themselves to have a substandard health-related quality of life compared to their healthy peers due to the impact of the condition as well as the associated treatment modalities that the child must endure. Svavarsdottir and Orlygsdottir (2006) conducted a study with school-aged Icelandic children and found that children with chronic health conditions or illnesses reported that their health-related quality of life was significantly lower than their peers who did not have a chronic illness.

Although it may be expected that children with chronic illness would have a decreased quality of life as opposed to healthy children, one study conducted in Thailand concluded that chronic illness, acute illness, or severe illness did not have any significant impact on quality of life in a sample of school-aged children (Jirojanakul, Skevington, & Hudson, 2003). Since health-related quality of life is a multidimensional construct, various components impact a child’s perception.

Health-related quality of life may depend on the child’s perception of the impact of the condition as well as the actual condition itself. Health-related quality of life instruments are available to measure condition-specific aspects of disease and treatment relevant to various medical conditions. These instruments may be more sensitive in detecting changes and the impact of specific treatments (Matza, Swensen, Flood, Secnik, & Leidy, 2004).
Health-Related Quality of Life for Children with Cleft Lip and/or Palate

The focus of health-related quality of life on children with oral clefts encompasses the child’s physical and psychosocial well-being. The concept of psychosocial well-being may be subdivided into three domains which include emotional functioning, social functioning, and school functioning (Varni, Limbers, Burwinkle, & 2007).

In years past, care of the child who had a cleft lip or palate focused primarily on the physical defect. For almost two decades, health care providers have become cognizant of the fact that clefts are actually midline defects that affect not only the structures of the face but also affect the central nervous system. This body of knowledge has enhanced understanding related to the possible effects on the educational, social, and psychological aspects of development for these children (Peterson-Falzone, Hardin-Jones, & Karnell, 2010). In addition, children with isolated clefts of the lip and/or palate have abnormal brain structure, possibly related to abnormal brain development (Nopoulos, Langbehn, Canady, Magnotta, & Richman, 2007) which may impact the child’s health-related quality of life.

“The stigmata of cleft lip and palate can never be completely eliminated” (Turvey, Ruiz, & Tiwana, 2009, p. 838). “The projection and support of the lip are derived from the underlying skeletal structures. Without this skeletal support, regardless of how well the lip is closed, the stigmata of the cleft lip and palate remains,” (Turvey, Ruiz, & Tiwana, 2009, p. 838.) The facial differences and associated consequences that children experience as a result of being born with an oral cleft may affect the child’s health-related quality of life.
Four papers could be identified in the literature that measured health-related quality of life in children who were preschool through school-age with cleft lip and/or palate. Damiano and colleagues (2007) measured health-related quality of life in children 2 to 12 years of age with nonsyndromic oral clefts with the PedsQL from the mother’s perspective. The instrument measured physical functioning, emotional functioning, social functioning, and school functioning. The sample included 104 mothers. The researchers found that speech and aesthetic concerns were factors that affected the health-related quality of life of the children when demographic factors were controlled for. The child’s age and type of oral cleft affected health-related quality of life. Total health-related quality of life scores increased for children with cleft lip or cleft lip and palate from the 2 to 4 year old group to the 5 to 7 year old group. Total scores declined for children with cleft lip or cleft lip and palate for the 8 to 12 year old group which the authors contributed possibly to the developmental phase of physical appearance and social interaction becoming more important. For children with cleft palate, the reverse was found. Children in the 8 to 12 year old group had higher scores than the younger two groups which may have been related to improvement in speech resulting from speech therapy and surgery.

Sagheri, Ravens-Sieberer, Braumann, and Mackensen (2009) conducted an evaluation of health-related quality of life of 61 German children who were 4 to 7 years old with nonsyndromic cleft lip and/or palate from the parent’s perspective. The instrument utilized was the German KINDL=Kiddy HRQoL questionnaire designed for 4 to 7 year old children. The domains that were measured included physical well-being, emotional well-being, self-esteem, family life, friends, and school. Results revealed that the mean total scores were slightly lower for children with cleft lip and palate; however, no
significant differences were found between children with or without oral clefts. Interestingly, children with cleft lip and palate were found to have statistically higher values in the self-esteem category when compared to the normal population which the authors contributed possibly to increased parental and caregiver participation for children with oral clefts.

Kramer, Gruber, Fialka, Sinikovic, & Schliephake (2008) also measured health-related quality of life for German children with cleft lip and palate; however, health-related quality of life was evaluated by the child as well as by parent proxy report. The study also included a measure of family functioning. The sample included 147 children with an oral cleft who were 5 or 6 years of age as well as their families. The instrument utilized for the children was the KINDL developed for 4 to 8 year old children. The parents also completed the parent version of the instrument. Findings of this study differed in that the KINDL scores were lowest in the self-esteem dimension on both the child and parent reports. In all of the dimensions, children rated their health-related quality of life higher than their parents did.

In a similar study, Kramer, Gruber, Fialka, Sinikovic, Hahn, and Schliephake (2009) evaluated health-related quality of life in 132 German children who were 8 to 12 years of age utilizing the KINDL and Impact on Family Scale. The health-related quality of life scores were higher for children with oral clefts than the control group. Children with cleft lip and palate had lower scores on the family and friends component than children without cleft lip or cleft palate. Boys scored lower on health-related quality of life than girls. Children who had a cleft palate (with or without cleft lip) had a lower health-related quality of life than children who only had a cleft lip.
State of the Science: Health-Related Quality of Life for Children

with Cleft Lip and/or Palate

Theories, Conceptual Models, and Frameworks Guiding Health-Related Quality of Life Research

Theory is defined as a “creative and rigorous structuring of ideas that projects a tentative, purposeful, and systematic view of phenomena,” (Chinn & Kramer, 2004, p. 58). Conceptual models are abstractions that are arranged together because they share a common theme; whereas, a framework refers to the conceptual underpinnings of a research project (Polit & Beck, 2006). “Conceptual models of nursing inform thinking and give meaning and direction to nursing research,” (Fawcett & Gigliotti, 2001, p. 339). Conceptual models can provide researchers with a foundation for hypothesizing the causal process that leads to improved health-related quality of life (Lach et al., 2006). Various models have been reported in the literature to guide research related to health-related quality of life.

The theoretical framework utilized by Eiser, Cotter, Oades, Seamark, and Smith (1999) to develop a generic measurement for health-related quality of life for 6 to 11 year old children was based on the work of Calman’s (1984) self-discrepancy theory. This model “assumes that health-related quality of life is the result of discrepancies between an individual’s actual self and ideal self” (p. 87). This theory focuses on the individual’s ability to achieve daily goals, specifically related to the difference between one’s expectations and experiences of illness and in relation to the child’s developmental level.

Yeh (2002) utilized the Roy Adaptation Model (RAM) theory when studying health-related quality of life in pediatric patients with cancer. The RAM is based on the
proposition that individuals are biopsychosocial beings who are required to adapt to environmental stimuli (Roy & Andrews, 1991). Health-related quality of life was hypothesized as consisting of physical function, psychological function, peer/school function, treatment/disease symptoms, and cognition functions.

Lach and colleagues (2006) expanded on the work of Raeburn and Rootman (1996) to develop a theoretical model to describe health-related quality of life in youth with epilepsy. The researchers proposed that the child’s biomedical status/level of impairment related to the epilepsy and other co-morbidities. The researchers stated that child, family, and community variables also influenced health-related quality of life.

Another theoretical model utilized to enhance an understanding of the impact of illness on quality of life in children is the stress-coping model. This model was utilized to predict quality of life in children with asthma (Peeters, Bersma, & Koopman, 2008). The model was derived from the cognitive-appraisal model of Lazarus and Folkman (1984), whereby when a person is confronted with a stressor, the individual will evaluate the stressor and then determine one’s emotional or behavioral reaction. Maes, Levental, and de Ridder (1996) developed an extended model derived from the cognitive-appraisal model for coping with a chronic disease. Their extended model took into account other life events, disease treatment characteristics, disease-related events, and demographic characteristics. Peeters, Bersma, & Koopman found that disease characteristics, appraisal of the disease, coping, and quality of life are all significantly related to one another.

Wallender and Varni (1989) developed a Disability-Stress-Coping Model based on their prior research and the work of Pless and Pinkerton (1975). The authors utilized the model to study adjustment of children with chronic physical conditions. The variables
were conceptualized in a risk-and-resistance framework. The model includes disease/disability parameters, functional independence, and psychosocial stressors as being the major risk factors for causing adjustment problems in children with chronic physical conditions. The authors noted that children with similar risk factors may adjust to their condition in a variety of ways; therefore, the model includes resistance factors. Resistance factors include intrapersonal factors, social-ecological factors, and stress processing. These resistance factors influence the risk-adjustment relationship. In his later work on health-related quality of life in children, Varni (1998) utilized the theoretical framework that generic health-related quality of life is a multidimensional construct, consisting at the minimum of the physical, psychological (including emotional and cognitive), and social health dimensions delineated by the World Health Organization (J. Varni communicated by V. Martel, personal communication, August 22, 2010).

Kapp-Simon (2002) and Kapp-Simon and Gaither (2009) applied the Risk, Resistance, and Coping model to children with cleft lip and palate. Although application of the model was not to describe health-related quality of life, there is significant overlap in terms of conceptualizing components that affect health-related quality of life for children with cleft lip and palate. According to the authors, risk factors experienced by children with oral clefts include medical risks, functional risks, and psychosocial risks. Medical risks include feeding difficulty, chronic otitis media, multiple surgeries related to the cleft, dental concerns, velopharyngeal dysfunction, and possible conditions related to syndromes. Functional risks include speech and language impairment as well as developmental and cognitive problems. Psychosocial risks for parents of children with oral clefts include learning of the diagnosis and deciding on treatment options. Resilience
in the model refers to the “flexibility, buoyancy, spirit, hardiness, and toughness” of the child and family who are coping with the day-to-day challenges of living with an oral cleft (Kapp-Simon & Gaither, 2009, p. 1003). Adaptation of the family refers to the coping with the cleft which will influence the child’s adaptation. Parental recognition of their strengths, social support from extended family, cleft organizations, and financial resources are all factors that influence parental coping. Child factors that influence resilience include temperament and parenting skill.

Although no instrument exists to specifically measure health-related quality of life in children with oral clefts, one instrument was developed to measure quality of life of youth with facial differences (YQOL-FD). The authors utilized the WHO quality of life definition and the needs-based model to develop the instrument. “The needs based approach to development of QoL measures builds upon functional status measurement and views QoL as the net result of a person's evaluation of how much their needs have been met including their evaluation of functional status and interaction with the environment. It is based on Maslow's needs hierarchy and was the basis for the WHO-sponsored measure of QoL,” (YQOL-FD user manual and interpretation guide, 2008, p. 12).

For the purpose of this study, the Child Health Model (Barnard & Eyres, 1979) was utilized as the theoretical framework to describe the conceptual underpinnings of the study. This model was chosen based on the researchers’ experience in working and interacting with children and their families. This model in addition to the work of Varni (1998), Kapp-Simon (2002), and Kapp-Simon and Gaither (2009) was chosen to develop a conceptual model for further understanding health-related quality of life for children.
who were born with cleft lip and/or palate. Varni’s theoretical framework was chosen based on the researcher’s extensive work regarding health-related quality of life in children. The components of the conceptual model are based on the PedsQL 4.0 Generic Core Scales. Incorporation of the Risk, Resistance, and Coping model to children with cleft lip/or palate was utilized when developing the conceptual model due to the authors’ work and research regarding psychosocial aspects of children with cleft lip and palate (Kapp-Simon, 2002; Kapp-Simon & Gaither 2009). The work of additional theorists also influenced the proposed conceptual model which will be discussed.

Concept Analysis of Health-Related Quality of Life for Children with Cleft Lip and/or Palate

A concept analysis was conducted to enhance an understanding of the concept of health-related quality of life that provided richness for the proposed conceptual model. The summary of the attributes, antecedents, and consequences of health-related quality of life for children with cleft lip and/or palate is illustrated in Figure 1. Antecedents include the child with an oral cleft in addition to the child’s caregiver and the child’s environment. Attributes include the child’s physical, emotional, social, and cognitive/developmental factors. The consequences of health-related quality of life include the child’s and caregiver’s perception of satisfaction with health and perception of well-being on a continuum which ranges from optimal to sub-optimal health-related quality of life.
Figure 1. Concept Analysis of Health-Related Quality of Life for Children with Cleft Lip and/or Palate
**Child Health Assessment Model**

Barnard and colleagues engaged in the Nursing Child Assessment Project (NCAP) in the 1970’s to determine measures of health and caregiving environments for infants and children. Their work was based on developmental theory which emphasized the physical, emotional, social, and intellectual dimensions of child development (Sumner & Spietz, 1994).

The researchers’ focus on these particular domains of child health (physical, emotional, social, and intellectual) in the Nursing Child Assessment Project is congruent with the domains described in the Pediatric Quality of Life Inventory (PedsQL 4.0), an instrument developed by Varni (1998). The PedsQL 4.0 instrument’s items are categorized into four subscales which include physical functioning, emotional functioning, social functioning, and school functioning. The health-related quality of life for children with cleft lip and/or palate will be analyzed in the context of the Child Health Assessment Model developed by Barnard and Eyres (1979) and the PedsQL 4.0 instrument.

The framework of the Child Health Assessment Model (Barnard & Eyres, 1979) is depicted as three overlapping circles to include the caregiver, child, and environment as illustrated in Figure 2. Important personal characteristics of the child include physical appearance, temperament, feeding and sleeping patterns, and self-regulation. Characteristics of the caregiver include psychosocial assets, physical and mental health, life changes, expectations and concerns about the child, and the caregiver’s style and adaptation skills which are considered to be the most important characteristic. The environment for both the child and caregiver encompass social and financial resources.
including a supportive adult, adequate food and housing, a safe home, and community involvement. The most important part of the model is the intersection where the circles overlap, representing the interaction between child, caregiver, and environment. Each aspect of the model can facilitate or impede the interaction between the caregiver and child (Sumner and Spietz, 1994).

Figure 2. Child Health Assessment Model

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These factors will be applied to the health-related quality of life literature for children with cleft lip and/or palate. It is proposed that all of the factors emphasized in the Child Health Assessment Model (Barnard & Eyres, 1979) influence the child’s physical and/or
psychological well-being either directly or indirectly which ultimately affects their health-related quality of life. Caregiver and environmental factors will be reviewed; however, the primary emphasis of this paper will be on the child factors and how these factors affect the child’s health-related quality of life.

**Child Health Assessment Model Applied to Children with Cleft Lip and/or Palate**

Application of the Child Health Assessment Model (Barnard & Eyres, 1979) to children with cleft lip and/or palate is presented. The focus of this section will be on the child factors; however, caregiver factors and environmental factors are also discussed as well as an overview of the intersection of the primary components.

**Child factors impacting health-related quality of life.**

Multiple factors impact health-related quality of life for children. These factors will be discussed for the toddler, preschool, and school-aged child within the realm of the child’s physical functioning, psychosocial functioning, and cognitive/school functioning.

**Toddler and preschool-aged children with cleft lip and/or palate.**

Specific factors must be taken into account for young children with cleft lip and/or palate that may impact their health-related quality of life. Factors that may affect the toddler and/or preschool-aged child’s physical, psychosocial, and cognitive/school functioning are presented.

*Physical functioning.*

A child’s physical functioning may be affected by the type and severity of oral cleft as well as the presence of a genetic syndrome or other co-morbidities. Infants and children with cleft lip and/or palate have multiple health concerns which vary depending on the type of oral cleft and the child’s age. Health concerns may include (but are not limited to)
feeding, speech, hearing, dental and orthodontic abnormalities, and the need for surgical correction(s). A child who is born with a cleft lip typically does not require long term management following the cleft lip repair which occurs in early infancy; however, the child may experience alterations in psychosocial functioning due to the facial scarring secondary to the cleft lip repair. Children who were born with a cleft palate require more extensive long term management including speech therapy, surveillance for possible hearing loss, management of dental and orthodontic problems, and observation for potential psychosocial difficulty (McCormick, Mackey, & Wilson, 2007).

By the time the child with an oral cleft reaches 2 to 4 years of age, the child has usually undergone the initial surgical repair(s) for the cleft lip and/or palate (Chibbaro, Barzilai, and Breen, 2009), thus allowing the toddler-aged child to “be free of another surgical interruption in family life and child development” (p 376). However, for some toddlers with an oral cleft, a late palatal surgery or myringotomies may be required (Peterson-Falzone, Hardin-Jones, & Karnell, 2010). Revision of the lip may also occur in the preschool period with the goal of achieving a socially acceptable appearance by approximately 7 or 8 years of age at which time peer interactions may change from awareness and curiosity to teasing (Reinisch, Wai-Yee, & Urata, 2009).

Children who are born with a craniofacial anomaly may also have a congenital abnormality of the auditory structures and/or an increased incidence of otologic disease. In a study that followed children with cleft lip and palate and a control group of children from 1 to 5 years of age, the children with oral clefts experienced a significantly higher incidence of otitis media with effusion (74.7%) than children who did not have a cleft lip and palate (19.4%), and hearing loss was significantly more pronounced in the cleft
Children with cleft palate who are in need of an increased number of myringotomy tubes, middle ear surgery, and those with cholesteatomas are at increased risk for long term conductive hearing loss (Goudy, Lott, Canady, & Smith, 2006). The child with hearing loss may experience adverse effects on speech and language development which may impact educational, social, and psychological functioning. The child with an oral cleft should have audiological follow-up based on their history; however, even for children who have no history of hearing loss or ear disease, an audiologic examination or screening should take place at least yearly during the toddler and preschool years (ACPA, 2009).

Children in the toddler and preschool years are in the primary dentition stage. Dental and orthopedic management for the child with an oral cleft focuses on promotion of good oral hygiene due to the higher incidence of dental caries. Maxillary appliances may also be indicated for management of velopharyngeal dysfunction or to obdurate oronasal fistulae (Huebener, 2009).

*Psychosocial functioning.*

The child’s temperament affects their response to stress and/or illness and influences the outcome. Approximately half of the child’s temperament is genetic and approximately half results from the environment and the child’s physical condition (Carey, 1998). Only one study could be found in the literature that addressed combined psychosocial variables with parent-infant feeding interactions which included infant temperament, maternal social support, and family socioeconomic status in association with growth of infants with oral clefts (Coy, Speltz, Jones, Hill, & Omnell, 2000). The authors reported that psychosocial variables do influence the early growth trajectory of
infants with oral clefts. In addition, Endriga and Speltz (1997) reported that difficult temperament (negative reactivity) was associated with low maternal involvement in infants with cleft lip and or palate.

One paper could be found in the literature that addressed self-regulation in children with oral clefts. Endriga, Jordan, and Speltz (2003) studied 83 children who were 5 years old and born with cleft lip and palate, isolated cleft palate, or no oral cleft. The children were videotaped during a disappointment situation that was conducted in a laboratory setting. Results indicated that children who had an oral cleft expressed significantly less disappointment than those in the comparison group. The authors concluded that the children with oral clefts may be more resilient due to their early physical and emotional challenges, or alternately that children with oral clefts may be overly controlled in response to a disappointing event.

During the toddler and preschool period, the child’s development of self as an individual separate from the immediate family becomes more pronounced (Dixon & Stein, 2006). Children 2 to 4 years of age may transition from spending most of his or her time at home to daycare or preschool. During this time, an important developmental task for children is to develop peer relationships (Hearst, 2007). As the child begins to spend more time outside of the home, children with oral clefts may develop concerns regarding the reaction of others who respond to their appearance and/or speech. Children who are born with an oral cleft may experience problems with psychological adjustment as early as 2 years of age (Cleft Palate Foundation, 2008). As the child transitions from toddlerhood to preschool age, the child’s central task is to establish a sense of identity (Dixon & Stein, 2006). The preschool-aged child begins to have an awareness of
desirable and undesirable appearances. At this time, children may begin to reflect the opinions of others regarding their appearance (Monroe, 2007). By age 4, children are most likely to notice differences between individuals (Dixon & Stein, 2006). Children who have facial differences such as a scar following a cleft lip repair may begin to be aware that they look different from other children. Visible differences in physical appearance can have a considerable psychological impact on children. Children with visible differences may experience challenges related to self-perception and social networks during early childhood (Rumsey & Harcourt, 2007). Other factors such as the appearance of the nose, improper tooth alignment, or hypernasal speech are other factors that may put the child with an oral cleft at risk for being teased by peers. During the preschool years, children gain an increased understanding of their cleft as a result of their increased sense of self-awareness and teasing from peers (Curtain & Boekelheide, 2004).

Speech and language difficulty may lead to psychosocial problems in the child with a cleft palate. If the child’s attempt at communication fails due to the words not being understood by others, the child’s frustration may become more problematic which may result in more frequent and intractable temper tantrums. Communication failures may lead to the child not attempting new words. Unintelligible speech may be difficult to correct. Speech evaluation must continue through the preschool years (Peterson-Falzone, 2009).

Surgical procedures may also have an impact on the child’s psychosocial adjustment. Toddlers and preschoolers who need to be hospitalized for surgical interventions may experience stress related to separation, loss of control, bodily injury, and pain. Toddlers may exhibit behavior reactions such as protest, physical aggression, verbal
uncooperativeness, regression, negativism, and temper tantrums. Preschoolers may also exhibit aggression and regression, despair, or detachment (Ireland, 2006). Preschoolers may believe that the hospitalization or surgery is a punishment for a prior misdeed (Stewart, Algren, & Arnold, 1994).

*Cognitive/school functioning.*

Language development and increased cognitive abilities support the interaction with others for the 2 year old child. There is a strong link between the child’s language and cognitive abilities (Dixon & Stein, 2006).

The results of one study indicated that 36 month old children with oral clefts demonstrated significantly lower performance in fine motor, gross motor, and expressive language when compared to 25 month old children who had oral clefts. The 36 month old children were considered to be at risk/delayed with regard to expressive language skills (Neiman & Savage, 1997). Another study revealed that 24 month old children with cleft lip and palate demonstrated significantly lower scores in cognition, comprehension, and expressive language when tested compared to matched controls (Jocelyn, Penko, & Rode, 1996).

**School-aged children with an oral cleft.**

*Physical functioning.*

Children, who are of school age with cleft lip only that has been repaired, typically do not have alterations in physical functioning, although they may undergo secondary surgical repair of the lip to restore normal appearance and function which is usually performed between the ages of 5 and 9. Children who require secondary repair of the nasal deformity due to the cleft lip will usually not have the repair until adolescence,
following completion of the postpubertal growth spurt (Byrd, El-Musa, & Yazdani, 2009).

School-aged children with cleft palate may undergo additional surgical procedures such as alveolar bone grafting and secondary palatal surgery. Children with cleft palate who undergo alveolar bone grafting will usually have the surgery between 6 and 12 years of age. The decision of when to repair the alveolar cleft is based on the child’s dental development and the recommendation of the orthodontist. The iliac crest is usually the site from which bone is harvested for the repair (Chibbaro, Barzilai, & Breen, 2009). In addition, school-aged children may undergo secondary palatal surgery for velopharyngeal dysfunction which may include a pharyngoplasty or pharyngeal flap. A tonsillectomy may also be indicated and performed at least 6 weeks prior to the secondary palatal surgery (Chibbaro, Barzilai, & Breen). Surgical or prosthetic closure of palatal fistulae may also be needed (American Cleft Palate-Craniofacial Association, 2009).

Children with cleft palate also require audiologic follow-up examinations throughout childhood to monitor hearing. Children with craniofacial anomalies are also in need of dental services for primary care and routine maintenance as a result of the medical condition (American Cleft Palate-Craniofacial Association, 2009).

*Psychosocial functioning.*

School-aged children may experience stress related to surgery and may react by becoming withdrawn or feel isolated despite their increased ability to express feelings. Developmentally appropriate preoperative preparation can reduce preoperative anxiety for children and their families (Ireland, 2006).
As children enter kindergarten and/or first grade, they are expected to achieve mastery of specific cognitive, motor, and social skills. Developmental tasks that children face as they enter school include the following: adapt to classroom learning, be able to tolerate prolonged separation from parents, integrate cognitive skills required to learn to read, form relationships with other children and adults, participate in group activities, and continue the formation of a sense of self (Dixon & Stein, 2006). As the young child’s self-concept continues to emerge, the child begins to be able to describe himself in terms of categorical identification. This type of identification includes basic descriptive features. Their description of self is concrete and often external (Damon & Hart, 1988; Dixon & Stein, 2006). As the child progresses into middle childhood, intellectual and social reasoning becomes more advanced. In addition, social expansion of activities with peers and social acceptance becomes a major developmental task (Dixon & Stein, 2006).

Children vary in terms of the impact of physical differences. Some children appear unaffected by their physical difference; however, some children may experience adverse effects on body image, self-esteem, quality of life, and difficulty with social encounters (Rumsey & Harcourt, 2007).

For children with cleft lip and/or palate, the overall goal is to “normalize the face with scars that are hardly visible” (Marcusson, Paulin, & Ostrup, 2002, p. 16). Children reported that a visible scar was the most concerning aspect of cleft lip and palate (Bernstein and Kapp, 1981). There is conflicting literature related to satisfaction of physical appearance for children with an oral cleft. While some studies report that children are satisfied with their facial appearance (Clifford, 1971; Bjornsson & Agustdottir, 1987; and Slifer et al., 2003), others reported dissatisfaction (Richman,
In a study that included 11 to 13 year old children with craniofacial anomalies, dissatisfaction with facial appearance was associated with peer relationship problems and low global self-esteem; however, it was not associated with other aspects of self-concept (Pope & Ward, 1997).

Although children with cleft lip and/or palate are confronted with increased risks which may affect psychological adjustment, most children have normal psychological adjustment (Kapp-Simon & Gaither, 2009). Hunt, Burden, Hepper, and Johnston (2005) conducted a systemic review which included 64 published articles that examined the psychosocial effects of cleft lip and palate. The authors found that the majority of children with cleft lip and palate do not experience major psychosocial problems although some problems may arise. Behavioral problems, satisfaction with facial appearance, depression, and anxiety have been reported. In one study that included 7 year old children with cleft lip (with or with cleft palate) and a control group, children who had clefts had higher rates of social problems as rated by teachers, higher anxious and withdrawn-depressed behavior, and difficulties in social relationships as opposed to their unaffected peers. The authors concluded that children with oral clefts are at risk for social and emotional problems during the school-aged years (Murray et al., 2010). In another study that included parent reports of the psychosocial functioning of children with cleft lip and/or palate, children with an oral cleft were more anxious, less happy with their appearance, less happy in general, had lower self-esteem, and had greater behavioral problems compared to their unaffected peers (Hunt et al., 2007).

A child’s resilience may also affect their health-related quality of life. Kapp-Simon and Gaither (2009) stated that child resilience is closely linked with child temperament
and parenting skill. One study explored psychosocial resilience in 10 year old children with oral clefts and found that resilience was associated with adequate emotional functioning, high satisfaction with appearance, and decreased frequency of teasing. Factors that did not affect resilience included visibility of the cleft, gender, and additional diagnoses (Feragen, Borge, & Rumsey, 2009).

**Cognitive/school/functioning.**

The success that a child achieves in school is related to various factors including the child’s intelligent quotient (IQ). For children with an oral cleft, school achievement or lack of success has been related to speech and hearing problems; expectations of peers, parents, and teachers; the child’s self-concept, learning disabilities, and the child’s social adjustment (Peterson-Falzone, Hardin-Jones, & Karnell, 2010). Controversy still exists regarding school performance for children with oral clefts. It is unknown if academic achievement is related more to biologic, cognitive, environmental, or social-emotional factors (Richman & Nopoulos, 2009). In an earlier study of 172 elementary school children with cleft lip and palate, approximately 35% of the sample had a moderate degree of reading disability, and 17% had a severe reading disability (Richman, Eliason, & Lindgren, 1988). In another study that included 84 children with an oral cleft, 46% had a learning disability, 47% had deficient educational progress, and 27% repeated a grade in school (Broder, Richman, & Matheson, 1998). In a recent longitudinal study which included children with oral clefts and controls, there were no significant group differences in language at ages 5 and 7 years, and the researchers concluded that their findings did not support the hypothesis that children with oral clefts score lower than
controls on neurocognitive and academic achievement measures (Collett, Leroux, & Speltz, 2010).

In addition to school functioning, the child’s understanding of the oral cleft may affect the child’s perception of health-related quality of life. A child’s understanding of their condition should also be determined. Yoos (1994) developed an expert/novice model as related to children’s understanding of their condition. The model proposed an alternate paradigm which can be applied to children’s concepts of illness as opposed to cognitive and developmental approaches. Yoos described an approach that elaborates on how children learn from experience. Yoos argued that what allows people to think at higher levels is related to changes in content as opposed to changes in cognitive structure. The model focuses on the notion that one can think at higher levels when they increase their knowledge base and experience within a particular domain.

No literature could be found on a child’s understanding of his or her congenital anomaly or specifically understanding of cleft lip or palate. Additionally, cleft palate is in somewhat of a different category than other conditions since it is neither an acute or chronic illness, yet it requires long term follow-up management. Based on previous research, it would be expected that children with cleft lip and/or palate would understand their condition in more detail with increased age. In addition, utilizing the paradigm proposed by Yoos (1994), one would expect children with cleft palate to become knowledgeable about his or her condition at a level higher than expected due to their multiple exposures and experiences with the multidisciplinary team. The child with cleft palate will typically require extensive follow-up care with appointments scheduled for speech, hearing, dental, and orthodontic care. In addition, the child with cleft palate may
become knowledgeable about the surgical experience due to multiple surgeries for repair of the cleft palate and associated complications.

In summary, for the child who was born with an oral cleft, multiple factors affect the young person’s health-related quality of life. These factors include those that impact the child’s physical functioning and multiple social, emotional, and cognitive factors that affect the child’s psychosocial functioning. Other factors that can affect a child’s health-related quality of life are caregiver factors, environmental factors, and the overlap of all the combined factors which will be briefly discussed.

**Caregiver factors impacting health-related quality of life.**

Caregiver factors may have a direct influence on a child’s health-related quality of life. There are several caregiver factors that affect the child’s health according to Barnard’s Child Health model (Barnard & Eyres, 1979). The most important factor is the caregiver’s style and adaptation skills. Other characteristics of the caregiver include psychosocial assets, physical and mental health, life changes, and expectations and concerns about the child (Sumner and Spietz, 1994).

For parents of children with cleft lip and/or palate, issues may include adaptation, cohesion, and emotional health prior to the birth of an infant as well as their acceptance of the infant’s oral cleft (Kapp-Simon & Gaither, 2009). The caregiver’s style and adaptation may be influenced by the parent’s experience in the prenatal and newborn period. Prenatal diagnosis and counseling may influence the way parents deal with their infant’s cleft. Prenatal diagnosis may help the family to adapt and prepare for feeding, family reactions, and sibling response (Peterson-Falzone, Hardin-Jones, & Karnell, 2010).
A mother’s perception of her newborn may also have significant implications regarding the child’s health-related quality of life. The mother-infant relationship is strongly dependent on the mother’s perception of her newborn (Broussard, 1979). Broussard noticed during clinical practice that the mother’s perception of her infant was determined by factors unrelated to the actual physical condition of the infant. Often times the physician and mother looked at the same infant, but saw the infant differently. Broussard postulated that certain genetic characteristics may be recognized soon after birth which may affect the mother’s perceptions regarding her child. The mother’s expectations of the child may become a self-fulfilling prophecy which may influence her interaction with the infant and may in turn influence the child’s behavior. The author found that mothers who did not have a positive perception of the infant as measured by the Neonatal Perception Inventories (NPI) had difficulty in responding to their infants needs.

For parents who have an infant with an oral cleft, the parent’s perception of the cleft may impact the child’s health and well-being. Parents who have an infant born with an oral cleft may experience a range of emotions and reactions. Parents may experience different perceptions related to the infant’s diagnosis (Beaumont, 2006). Parents may not perceive their child as having a disability when the child is born with an oral cleft since the condition can be corrected. Some parents can even experience distress following the cleft lip repair due the infant looking so different following the surgery (Nusbaum, Grubs, Losee, Weidman, Ford, & Marazita, 2008).

The caregiver’s locus of control regarding their child’s health care and the parent’s educational level may also impact the child’s health-related quality of life. Parents may
seek out information related to the child’s cleft. In one qualitative study, parents in the prenatal period planned for the infant’s needs by reading extensively about feeding, surgeries, and other challenges while another parent sought information through the Cleft Craniofacial Center. Another parent believed that if he had not known about the diagnosis prenatally, the baby would not have received optimal care. By knowing the infant’s diagnosis, he felt as though the knowledge resulted in being able to make better decisions regarding the infant’s care. Parents may be concerned if their infant is “normal” and what the long term implications may be for the child (Strauss, Sharp, Lorch, & Kachalia, 1995). In another qualitative study, parents of infants with an oral cleft indicated a desire to have more information and more of a discussion about the possibility of mental retardation (Nusbaum, Grubs, Losee, Weidman, Ford, & Marzita, 2008).

Stress and coping of parents who have a child with a chronic illness is dependent on the individual’s cognitive appraisal of stress according to the Family Resilience Model which addresses how the family responds and adapts to stress (Knafl & Santacroce, 2004). In one study that assessed the perception of the impact of a child’s chronic illness, factors that predicted maternal perceptions of the impact of the child’s condition included the need to watch for changes in the child’s condition, the presence of communication or speech problems, and the number of hospitalizations the previous year. Additionally, the mother’s perception of the impact of the child’s chronic illness had a direct effect on the mother’s mental health (Ireys & Silver, 1996).

Baker, Owens, Stern, & Willmot (2009) studied coping strategies and social support of parents who had a child with an oral cleft related to adjustment and psychological distress. The researchers found that negative outcomes such as family impact and
psychological distress were not high. The authors stressed that the perceived emotional support, practical support, and adaptive coping strategies is important to families who have children with an oral cleft, and stated that the information gained can be utilized to develop effective interventions to assist families. Stone and colleagues (2010) conducted focus groups with parents who had children with an oral cleft and found that parents identified the early need for support, credible information, and advice for daily living. Additionally, Strauss (2001) pointed out that past research on children with oral clefts focused on deficits and challenges which have led to an understanding of the physical and psychosocial challenges experienced by children. The author proposed launching a new model for craniofacial research around resilience, strengths, and success to maximize human potential.

**Environmental factors impacting health-related quality of life.**

The Child Health Assessment Model (Barnard & Eyres, 1979) includes environmental factors that play an important role in the child’s health. These factors include social and financial resources, a supportive adult, adequate food and housing, a safe home, and community involvement. Culture is an important factor that can affect an individual’s perception of health. In addition, “since quality of life is culturally constituted and patterned, it needs to be studied and understood from a transcultural nursing perspective in order to advance nursing as a discipline and profession” (Leninger, 1994, p. 22).

Basic material comfort such as adequate food and shelter affect a child’s health-related quality of life. In one study with a sample of 399 children, household food insecurity was significantly associated with total health-related quality of life and
physical functioning. Children who lived in households with food insecurity had poorer health-related quality of life (Casey et al., 2005).

Family dynamics can also affect a child’s health-related quality of life. Children who lived in two-parent original families had higher health-related quality of life than those who did not in one study of school-aged children (Spurrier, Sawyer, Clark, & Baghurst, 2003). When caring for chronically ill children at home, one of the coping strategies that was reported by parents as being the most helpful included utilizing family support (Ray & Ritchie, 1993).

Medical resources are a critical component of care and support for children who are born with an oral cleft. A multidisciplinary approach provides comprehensive care by essential specialists who are experts in the care and treatment of cleft lip and palate. Children and their families may benefit from longitudinal evaluation and treatment from specialties including audiologic care, cleft lip and palate surgery, craniofacial and maxillofacial surgery, dental care, genetic services, nursing care, otolaryngologic care, pediatric care, psychological and social services, speech-language services, and quality management (ACPA, 2009).

Perceived social support can affect family coping and is a factor influencing the resiliency of families who have a child with a chronic illness (Tak & McCubbin, 2002). Social support may also include organizations for families of a child with an oral cleft such as the Cleft Palate Foundation, AmeriFace, The Children’s Craniofacial Association, and Wide Smiles (Kapp-Simon & Gaither, 2009). In a study that included 103 parents of children or young adults with cleft lip and palate, high levels of social
support were predictive of less family impact, lower psychological distress, and more positive adjustment (Baker, Owens, Stern, & Willmot, 2009).

A family’s socioeconomic status can affect a child’s health-related quality of life. In a study that included 3,597 school-aged children, those who lived in families of higher income, had higher health-related quality of life. Children from lower socioeconomic backgrounds were found to have more negative experiences related to health and wellness (Spurrier, Sawyer, Clark, & Baghurst, 2003). In another study of pediatric patients with asthma, household income was most consistently associated with health-related quality of life for children and their parents (Erickson et al., 2002). Cope, Ungar, & Glazier (2008) also found that disparities exist in asthma control between children and families of different socioeconomic strata. Similar findings were reported regarding socioeconomic disparities as being important factors that determine health-related quality of life. In a study of children with a chronic condition, factors that had the greatest impact of poorer health-related quality of life included lower family income, lower parental education, and worse family functioning (Kulkarni, Cochrane, McNeely, & Shams, 2008).

Financial concerns are often problematic for families with a child who has an oral cleft due to the extensive treatment needed which may or may not be covered by the family’s insurance (Kapp-Simon & Gaither, 2009). Kramer, Baethge, Sinikovic, and Schliephake (2007) found that families whose children had isolated cleft lip experienced less financial and social impact than families whose children had cleft lip and palate or isolated cleft palate although problems in coping were increased.
School resources also influence a child’s health-related quality of life. School connectedness may be a factor that affects a child’s perception of their health-related quality of life since schools fundamentally contribute to a child’s intellectual, social, and emotional development (Mansour et al., 2003). School-aged children with oral clefts are at risk for socio-emotional difficulties, and interventions related to the transition to school may be required (Murray et al., 2010).

Cultural and spiritual beliefs may also directly or indirectly influence attitudes related to a child with a birth defect. Folklore regarding prenatal influences has existed since ancient times. Once such belief is if a pregnant woman was frightened by a rabbit, her child would be born with an oral cleft. Health conditions may also be viewed by individuals as a punishment from God or a test of strength (Hooke, 2007). Cultural beliefs vary geographically. For example in Zimbabwe, oral clefts are associated with witchcraft and ancestral spirits (Mzezewa & Muchemwa, 2010). In a study which took place in rural areas of India and Egypt, causation of oral clefts were thought to be related to nature, God’s will, God running out of skin, parental sins, punishment from God, solar eclipse, harmful ingestion, intrauterine contraception, witchcraft, and gazing at a camel. Various beliefs related to causation of the cleft may affect family and societal attitudes toward affected children (el-Shazly et al., 2010).

**Intersection of child, caregiver and environmental factors.**

The intersection of the Child Health Assessment Model is the most important part of the model. The dark area where the circles overlap represents the interactions between the child, caregiver, and environment. The area of the three-dimensional overlap focuses on how each component of the model affects the interaction of the other two (Barnard &
The Nursing Child Assessment Project team believed that the caregiver-child interaction would be an important predictor of a child's development. (Bee et al., 1982).

For a child with cleft lip and/or palate, the dynamic interaction between the child, caregiver, and environment is in a constant state of change as the factors in each domain change throughout the child and family's life. It is proposed that the interaction of the child, caregiver, and environment heavily influences the child's overall health-related quality of life. The child's perception as well as the caregiver's perception of each component affects the child's health-related quality of life.

A strong relationship between characteristics of early caregiver-child interactions and the development of later skills and qualities that the child exhibits has been demonstrated. Positive quality interactions in the child's early development tend to be strongly correlated with the child's subsequent intellectual and language skills. In addition, positive interactions between caregiver and child are linked with the child having more secure attachments (Barnard, Hammond, Booth, Mitchell, & Spiker, 1989; Barnard, Booth, Mitchell, & Telzrow, 1988; Barnard & Kelly, 1990). How the parent adjusts to the child's diagnosis may also have an impact on maternal-infant adaptation. Strong maternal attachment enhances healthy growth and development for a child. Maternal attachment can be defined as, "the unique, affectionate relationship that develops between an infant and persists over time" (Muller, 1994, p. 130). Maternal attachment begins during pregnancy and increases as the pregnancy progresses. Once the baby is born, the mother compares her "real" baby with the baby she fantasized about (Rubin, 1984). Some research has been conducted to assess if maternal attachment is altered when the infant is
born with a cleft lip due to the defect being highly visible and stigmatizing. This situation is a significant concern since difficulty in maternal interaction predicts lower IQ in children (Speltz et al., 2000; Wasserman & Allen, 1985). Overall, it appears as though maternal attachment is not affected due to the infant’s cleft lip (Slade, Emerson, & Freedlander, 1999; Coy, Speltz, & Jones 2002; Speltz, Endriga, Fisher, & Mason, 1997; Maris, Endriga, Speltz, Jones, & DeKlyen, 2000); In addition, a mother-infant pair may interact approximately 200 times a month while the mother feeds the infant at which time the relationship between the dyad is developed. If the feeding interaction does not go well, due to either problems with the mother or child, this may contribute to repeated failures in relationship building (Sumner & Spietz, 1994).

**Conceptual Model**

“A conceptual model is a set of relatively abstract and general concepts that address the phenomena of central interest to a discipline, the nonrelational propositions that broadly define those concepts, and the relational propositions that state relatively abstract and general linkages between two or more of the concepts,” (Fawcett & Gigliotti, 2001, p. 339, Fawcett). The abstractions, or concepts, are assembled in a conceptual model because of their relationship to a common theme (Polit & Beck, 2006).

A conceptual model for representing health-related quality of life for children with cleft lip and/or palate is illustrated in Figure 3. The model addresses the factors that have an impact on the child’s health-related quality of life and illustrates the conceptual links among the variables of interest. The primary components include caregiver factors, environmental factors, and child factors which have been included and modeled from Barnard’s Child Health Model (Barnard & Eyres, 1979) to emulate the theoretical
connections. The major components of the child factors are the physical, emotional, social, and cognitive domains which have been included and modeled from the PedsQL (Varni, 1998). The developmental component has been added to the cognitive domain since all aspects of the child’s factors must be viewed in terms of the child’s developmental level; however, the PedsQL focuses on school functioning.

The focus of the present research is to study the child factors that impact health-related quality of life, specifically related to the child’s physical, emotional, social, and school functioning for those who were born with an oral cleft. The figure illustrates that the caregiver, environmental, and child factors overlap to represents that these entities are highly dependent on one another. The focal point of the model targets the child factors which have the greatest significance in terms of the individual child’s perception, functioning, and subjective view of one’s own health-related quality of life.
Figure 3. Conceptual Model: Factors Impacting HRQOL for Children with an Oral Cleft

Conceptual Model: Factors Impacting Health-Related Quality of Life for Children with Cleft Lip and/or Palate

ENVIRONMENTAL FACTORS
- Material comfort
- Family dynamics
- Medical resources
- Social resources
- Financial resources
- School resources
- Cultural/spiritual perceptions

CHILD FACTORS
- Physical
  - Type & severity of oral cleft
  - Associated genetic syndrome
  - Associated co-morbidities
  - Speech impairment/VPI
  - Hearing impairment
  - Dental/orthodontic impairment
  - Surgical interventions

- Emotional
  - Temperament
    - Self-regulation

- Social
  - Physical Appearance
    - Self-concept
    - Body esteem

- Cognitive/Developmental
  - Age
    - Developmental level
  - Learning disability
    - Child's understanding of oral cleft
Summary

Children who are born with cleft lip and/or palate are in need of comprehensive care throughout their lifespan. Nurses as well as the entire multidisciplinary team strive to provide ongoing physical and psychosocial support which may have a positive affect on the child’s overall health-related quality of life. Measurement of health-related quality of life from the child and parent provides insight as to the child’s and parent’s perception of the child’s physical and psychological well-being. This information can be utilized by health care professionals to provide individualized and holistic care which may enhance the child’s well-being. A conceptual model has been presented as a means of understanding the interrelated concepts of health-related quality of life for children with cleft lip and/or palate in a unified manner as well as to guide nursing research on the phenomena of interest within a conceptual frame of reference.
Chapter 3: Methodology

The overall purpose of this study was to measure health-related quality of life for children with cleft lip and/or palate to include parent proxy reports and child self reports. The information obtained was compared to healthy children and children with a chronic condition to determine how health-related quality of life was perceived for parents and children with oral clefts as compared to normative data. Data obtained was also utilized to determine how demographic variables, speech, and hearing impacted health-related quality of life for these children. This study was a part of a larger study that focused on psychosocial outcomes for children with cleft lip and/or palate.

Design

A descriptive quantitative design was used for this study. The design was non-experimental and cross-sectional.

Operational Definitions

Operational definitions for terms frequently used in this study are presented. For this study, quality of life for a child is defined as, “the child’s individual perception of their physical well-being, psychosocial well-being, and satisfaction with life in relation to their cognitive and developmental level which is influenced by caregiver and environmental factors.”

For the purposes of this paper, the health-related quality of life for a child is defined as, “the child’s individual perception of the impact that their medical condition, disease state, or congenital anomaly has on one’s physical well-being, psychosocial well-being, and satisfaction with life in relation to their cognitive and developmental level which is influenced by caregiver and environmental factors.” Hence, the health-related
quality of life for a child with cleft lip and/or palate is defined as, “the child’s individual perception of the impact that the oral cleft has on one’s physical well-being, psychosocial well-being, and satisfaction with life in relation to their cognitive and developmental level which is influenced by caregiver and environmental factors.” Although the child’s own perception is of utmost importance, the parent’s or caregiver’s perception of the child’s quality of life or health-related quality of life is also extremely significant.

Setting

Data were collected at Kapiolani Medical Center for Women and Children. The instruments were introduced to the parents and children during the Kapiolani Cleft and Craniofacial Center (KCCC) clinic visit.

Sample

The sample for this study was children who were born with cleft lip and/or palate, including children with nonsyndromic (no known genetic syndrome) oral clefts and their parents. A convenience sample of children from 2 to 12 years of age and their parents/caregivers, with a focus on the child’s mother when possible, were recruited. Other inclusion criteria included participants who could speak and write in English (when applicable for developmental level) and children of any race or ethnicity.

The Kapiolani Cleft and Craniofacial Center (KCCC) had been providing care to children and their families for 2-3 years at the time of data collection. Based on the number of visits at the KCCC clinic when implementation of the study began, it was estimated that approximately 50 families who had children between 2 and 12 years of age would be enrolled in this study in a one year time period. The data were collected from June 19, 2009 to July 28, 2010, and the data set was frozen for this analysis.
Measurements and Instruments

The Pediatric Quality of Life Inventory Version 4.0 (PedsQL)

Since there is not a condition-specific instrument to measure health-related quality of life for young children with cleft lip and/or palate, the Pediatric Quality of Life Inventory Version 4.0 (PedsQL) was chosen for this study as a generic tool to measure health-related quality of life in children. The PedsQL was developed over a 25 year period from a variety of data gathering methods such as patient and parent focus groups, individual interviews, item generation, cognitive interviewing, pre-testing, and field testing. Data was accumulated on over 35,000 children internationally who were healthy or had various chronic conditions (Varni & Limbers, 2009).

The PedsQL instrument has shown that children as young as 5 years of age can reliably and validly self report their health-related quality of life. The reliability and validity of the instrument has been demonstrated in an analysis of 8,591 children who were heterogeneouse with respect to race and ethnicity; this sample also included Asians and Pacific Islanders. The majority of the self report scales exceeded the minimum internal consistency reliability standard of .70 and the Total Scale Scores approached or exceeded .90 (Varni, Limbers, & Burwinkle, 2007).

In addition, the PedsQL demonstrated feasibility, reliability, and validity for parent proxy reports for children who were 2 to 16 years of age. The sample included parent proxy reports on 13,878 children from the PedsQL database (Varni, Limbers, & Burwinkle, 2007).
According to Drotar (2004), “The PedsQL appears to be the most promising measure for assessing quality of life in pediatric settings…” (p. 21). The Pediatric Quality of Life Inventory Version 4.0 (PedsQL) instrument is designed to measure the health-related quality of life in children from 5 to 18 years of age. The PedsQL includes child self report questionnaires for children 5 to 18 years of age and parent proxy questionnaires for parents who have children 2 to 18 years of age (Drotar, 2004). The parent proxy questionnaires are designed to complement, rather than substitute for the child’s self report (Varni, Limbers, & Burwinkle, 2007).

Several versions of the PedsQL instruments were used for this study. The parent and child reports included the following: Parent Report for Toddlers (ages 2-4) (see Appendix A), Young Child Report (ages 5-7) (see Appendix B), Parent Report for Young Children (ages 5-7) (see Appendix C), Child Report (ages 8-12) (see Appendix D), and Parent Report for Children (ages 8-12) (see Appendix E).

The PedsQL instrument consists of 23 items for children 5 to 18 years of age and 21 items for children 2 to 4 years of age rated on a Likert scale. The four domains of the instrument include Physical Functioning (8 items), Emotional Functioning (5 items), Social Functioning (5 items), and School Functioning (5 items for children 5 to 18 years of age and 3 items for children 2 to 4 years of age) (Scaling and Scoring of the Pediatric Quality of Life Inventory, 2008). The items included for the parent and child reports are almost identical. The participant instructions ask how much of a problem each of the items has been for the child in the past month (Varni, Limbers, & Burwinkle, 2007). For children 5 to 7 years of age, a 3 point scale is used. Selection responses are as follows:
0 = not at all, 2 = sometimes, and 4 = a lot. To aid the child, a visual analogue scale is included. The picture associated with the “not at all response” illustrates a smiling face, “sometimes” is associated with a face with a mouth that is a straight line, and “a lot” illustrates a frowning face. For children 8 years of age and older, a 5 point response scale is utilized. Selection responses include the following: 0 = never a problem, 1 = almost never a problem, 2 = sometimes a problem, 3 = often a problem, and 4 = a lot of a problem (Varni, 1998). The items are reverse scored and transformed to a 0 – 100 scale. Scoring for the PedsQL instrument is obtained by summing all items in a scale divided by the number of items that the participant answered. The Physical Health Summary score is obtained from the Physical Functioning domain score. The Psychosocial Health Summary Score is obtained from the sum of the Emotional, Social, and School Functioning scales divided by the number of items answered. Higher scores indicate higher health-related quality of life. A Total Score is obtained from all four domains by summing all items on the scale and dividing the total by the number of items answered (Mapi Research Trust, 2008).

**Demographic Data**

Demographic data were obtained from the Demographic Form (see Appendix F). Selected items from the form that were included for the study included gender, birth date, island of residence, and race.

**Data from Database Repository**

Selected questions from the Craniofacial Questionnaire (see Appendix G) were collected from the parents during the clinic visit. The data were later transferred and
obtained from the Kapiolani Cleft and Craniofacial Center repository and included parental information such as income, educational level, age, and marital status.

When available, genetic information was obtained during the screening process for the purpose of determining if the potential subject met the eligibility criteria. Prior to data analysis, information regarding if the child had a genetic syndrome (see Appendix H) and data related to the type of oral cleft (see Appendix I) was obtained from the repository.

Data related to speech (see Appendix J) and hearing (see Appendix K) were also obtained from the Kapiolani Cleft and Craniofacial Center database repository. Data were only included if information was available in the repository. No speech or hearing assessments were conducted for the purpose of this study.

Speech data included an assessment of articulation and speech impairment. The assessment was completed by a speech therapist from the oral cleft clinic who worked directly with the children who participated in the study. The articulation was entered into the repository and obtained from the repository for this study.

Articulation impression was obtained from subjective and objective measures and included number of errors on the Goldman Fristoe 2 Test of Articulation, standard score on the Goldman Fristoe 2 Test of Articulation (NCS Pearson, 2000), nasal emission of air, and miss-learning errors such as glottal stops and pharyngeal fricatives. Informal clinical impressions were also considered when ranking the child’s articulation impression. The child’s articulation was documented as one of the following: normal, mild impairment, moderate impairment, or severe impairment.
Data related to hearing included severity of hearing loss and information regarding the use of a hearing aid. Data related to severity of hearing loss was quantified in the database as follows: normal (0 – 20 dB), mild (21 – 40 dB), moderate (41-55 dB), moderate to severe (56 – 70), severe (71 – 90), and profound (91 – 100+). Information regarding use of a hearing aid was categorized as “yes” or “no.” All data related to hearing was described for the right and left ear in the repository, however was combined for the purpose of this study.

**Human Subject Protection and Confidentiality**

This study was approved by the Western Institutional Review Board (IRB) which is the Institutional Review Board utilized by Kapiolani Medical Center for Women and Children. Collaborative Institutional Training Initiative (CITI) was completed by the nurse researcher. Instruction related to protection of human subjects and research ethics was studied and discussed in multiple courses in the doctoral program at the University of Hawaii.

Potential risks for parents and children participating in the study included the possible inconvenience and/or discomfort associated with answering the questionnaire(s). The subjects were told that some of the questions may be considered personal and/or sensitive and that it was possible that they may feel uncomfortable when answering certain questions. Parents and children were told that they may choose not to answer any of the questions on the questionnaires by leaving the item(s) blank. It was possible that the child or parent could become anxious or bored while completing the questionnaires. If this happened, the participant was to be given a break, given the opportunity to discuss the
problem, or moved onto another activity; however, this did not occur during data collection.

Either Dr. Neal or Dr. Iwamoto, both Medical Directors for the Kapiolani Cleft and Craniofacial Center, was on site during clinic while data collection occurred. Either physician was to be notified immediately if an adverse event occurred. All adverse events were to be reported to the appropriate IRB as necessary. All serious adverse events were to be reported to the appropriate IRB within 24 hours of the event. A written report was to follow once the event was resolved. No adverse events occurred during the data collection for this study.

The plan for protecting participant identifiers from improper use and disclosure included providing a study identification number on the research questionnaires. Also, the research database was kept on a secure server at the Clinical Research Center and access to the research database was limited to authorized study personnel. Additional measures to ensure protection included keeping the server in a room with restricted access, having the computer and database password protected, and access to personal health information and direct identifiers was restricted to authorized study personnel. Only the principal investigator, co-investigators, data entry specialist, and information technology specialist had the ability to link the subject’s identity with the data.

**Data Collection Procedure**

The nurse researcher screened the parent and child to determine if the family met the eligibility criteria. The nurse researcher verbally reviewed the Informed Consent Form (see Appendix L) with the parent or legal guardian of the child and provided a written copy as well. For children 7 to 12 years of age, the nurse researcher reviewed the Assent
Form (also called the Information Sheet) and provided a written copy to the child (see Appendix M for children 7 to 10 years of age and Appendix N for children 11 to 12 years of age). Parents and children were given ample time to read the information and ask questions. Parents who consented to be in the study were given a copy of the signed Informed Consent Form. Children 7 years of age and older who agreed to participate in the study were given a copy of the age appropriate Assent Form. It should be noted that the Informed Consent Form and both Assent Forms were composed to include other studies in addition to the study for the doctoral dissertation.

If the parent and/or child agreed to participate in the study, compensation was provided for taking the time to be in the study. Children 5 years of age and under received a teddy bear. The teddy bear had “stitches” sewn on the bear to represent a cleft lip repair. For children older than 5 years of age, the child was given the choice to select a teddy bear or a $10 gift card to a book store.

Questionnaires were administered in a private examination room by the nurse researcher during the Kapiolani Cleft and Craniofacial Center at Kapiolani Medical Center for Women and Children. Directions were read to the parent and child verbatim as per the PedsQL directions printed on the instrument. For children 5 to 7 years of age, the directions from the PedsQL were followed to determine if the child understood the questionnaire. If the child appeared to understand, the questionnaire was administered. If the child did not seem to understand, the questionnaire was not administered to the child; however, the parent was still asked to complete the parent version. For children who had difficulty reading the questions, the nurse researcher assisted the child with reading, and when necessary, read the questions for the child. Parents and children were asked to
answer their own questionnaire without discussing the items with each other, and the nurse researcher stayed in the examination room to ensure that each parent and child answered their questionnaire independently of one another.

**Statistical Analysis**

The statistical analysis of data is presented by listing the specific aims with the corresponding research questions. The statistical test conducted to analyze each research question is included. Data was analyzed utilizing SPSS 19.0. For clarification, the score for the Physical Functioning domain is the same as the Physical Health Score. The Psychosocial Health Summary Score includes Emotional Functioning, Social Functioning, and School Functioning.

**Specific Aim 1**

To examine health-related quality of life of children with nonsyndromic oral clefts according to age category and compared to normative data of healthy children and children with a chronic condition as measured with the PedsQL instrument to include parent proxy reports for children ages 2 to 12 years and child self reports for children ages 5 to 12 years.

**Corresponding research questions for specific aim 1.**

1. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life according to each age category.

2. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life according to each age category?
3. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of parents of healthy children?

4. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of healthy children?

5. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of parents of children with a chronic condition?

6. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of children with a chronic condition?

**Data analysis for specific aim 1.**

Descriptive statistics were utilized to analyze the data for research questions 1 – 6. Means and standard deviations were calculated for the Physical Health Summary Score which included the Physical Functioning Score; the Psychosocial Health Summary Score which included the sum of the items over the number of items answered in the Emotional, Social, and School Functioning scales; and the Total Score which included the sum of items over the number of items answered on all of the scales (Mapi Research Trust, 2008). All of these scores were calculated for the parent proxy reports and the child self reports. The parent proxy reports were for 2 to 12 year old children and the child self reports were for 5 to 12 year old children.
Health-related quality of life scores for parents and children according to age category were analyzed. Minimum and maximum health-related quality of life scores were obtained in addition to the range of scores.

One-sample t tests were conducted to determine whether mean scores from the study sample differed significantly from normative data for each of the health-related quality of life scores for parents and children. The parent proxy report scores for parents of children with an oral cleft were compared with normative data of parent scores of those who had healthy children and parent scores of those who had a child with a chronic condition. Child self report scores for children with an oral cleft were compared with normative data of child self report scores of those who were healthy and child report scores of those who had a chronic condition. It should be noted that the term “healthy” was used for descriptive purposes that was documented in the literature, however was not meant to imply that children with an oral cleft are not healthy.

**Specific Aim 2**

To determine if there is parent-child agreement (concordance) between parent proxy reports and child self reports for 5 to 12 year old children with nonsyndromic cleft lip and/or palate when rating Physical Health, Psychosocial Health, and Total health-related quality of life as measured by the PedsQL.

**Corresponding research question for specific aim 2.**

7. Is there parent-child agreement between parent proxy reports and child self reports for 5 to 12 year old children with nonsyndromic cleft lip and/or palate when rating Physical Health, Psychosocial Health, and Total health-related quality of life?
Data analysis for specific aim 2.

Paired sample t tests were conducted and Pearson product moment correlation coefficients were obtained to measure the parent-child agreement to determine if parent’s perceptions were the same as their children’s perceptions with regard to the child’s health-related quality of life. The Physical Health, Psychosocial Health, and the Total Scores were compared between the parents and their 5 to 12 year old children.

Specific Aim 3

To examine if there are differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate that are attributed to demographic variables including gender, age, and race of the children; and the geographic residence, educational level, income, marital status, and age of the parents?

Corresponding research question for specific aim 3.

8. Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate that are attributed to demographic variables including gender, age, and race of the children; and the geographic residence, educational level, income, marital status, and age of the parents?

Data analysis for specific aim 3.

Analysis of variance (ANOVA) was conducted between the demographic variables and the Physical Health, Psychosocial Health, and Total Scores. The ANOVAs were conducted for parents and the children.
Specific Aim 4

To examine if there are differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate that are attributed to impaired speech or hearing.

Corresponding research questions for specific aim 4.

9. Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate who do or do not have impaired speech?

10. Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate who do or do not have impaired hearing?

Data analysis for specific aim 4.

For research question 9, the child’s articulation was documented in the oral cleft clinic database as one of the following: normal, mild impairment, moderate impairment, or severe impairment. For the data analysis, mild and moderate impairment were combined. Analysis of variance was conducted to determine if there were differences in Physical Health, Psychosocial Health, and Total health-related quality of life scores based on speech articulation from the parent’s and child’s perspectives.

For research question 10, the child’s severity of hearing loss was obtained from the oral cleft clinic database as one of the following: normal (hearing loss of 0-20 dB), mild (21-40 dB), moderate (41-55 dB), moderate to severe (56-70 dB), severe (71-90 dB), and profound (91-100+ dB). The hearing loss was documented separately for each ear. For the data analysis, hearing loss was regrouped into three categories which included normal
hearing (in both ears) (hearing loss of 0 to 20 dB), mild to moderate-severe hearing loss (in either ear) (21 to 70 dB of hearing loss), and severe to profound hearing loss (in either ear) (71 to 100+ dB of hearing loss). Analysis of variance was conducted to determine if there were differences in Physical Health, Psychosocial Health, and Total health-related quality of life scores based on severity of hearing loss from the parent’s and child’s perspectives as well as if wearing a hearing aid resulted in any differences of health-related quality of life scores.
Chapter 4: Results

The results of this study are presented beginning with a description of the sample. The demographic characteristics of the participants are presented next, followed by an overview of the results. The remaining four sections report the findings for the four specific aims and research questions.

Description of the Sample

The PedsQL was completed by a total of 68 families. Out of the 68 families, 63 were included in the final analysis. Five families were excluded from the final data analysis due to documentation in the oral cleft database repository of the children being diagnosed with a genetic syndrome which was not known at the time of data collection. One child had Kabuki syndrome, one child had caudal regression syndrome, two children were diagnosed with van der Woude syndrome, and one child had possible Noonan syndrome.

Of the 63 remaining families who were included in the final analysis, there were 100 subjects who completed the PedsQL. The 100 subjects included 62 parents and 38 children. There were 37 parent/child dyads, 25 parents (without children completing the survey), and one child (without a parent completing the survey) who participated in the study. The study sample was representative of the oral cleft clinic population. Approximately 90 to 95 % of those who were asked to participate in the study agreed to do so.

Demographic Characteristics of the Participants

Parents

Multiple demographic variables were obtained from the parents who participated in the study. Data obtained from the Craniofacial Questionnaire included selected questions
pertaining to the parents such as income, age, highest level of education, and marital status. Table 1 summarizes the characteristics of the parents and the children who participated in the study. Missing items are not included in the table.

Demographic data of the parents included island of residence, age, educational level, income, and marital status. Almost three-fourths of the families who participated in the study lived on Oahu (69.8%). The remaining (30.1%) families lived on the neighbor islands with half of them residing on the island of Hawaii. Regarding the parent’s highest level of education, the largest category for both mothers and fathers was completion of high school which accounted for 39.7% and 44.4% respectively. The question was not answered for 1 (1.6%) of the mothers and 5 (7.9%) of fathers. The majority of the parent’s combined income was reported to be at the higher and lower ends of the spectrum with 39.7% at greater than $50,000 and 38.1% at less than $25,000. Three (4.8%) did not report their income. The majority of parents were married (60.3%). For those who answered the question (90.5%), the mother’s ages at the time of the child’s birth varied greatly ranging from 15 years to 53 years (mean $= 27.6$ years). The two mothers who were 52 and 53 years of age when their child was born adopted their children.

**Children**

Data obtained from the Demographic Form which was utilized for the study included gender of the child, birth date of the child, island of residence, the child’s ethnicity/race, and race in which the child most identified with. There were slightly more male children (58.7%) than females (41.3%). Race/ethnicity was indicated by having the parent check off as many groups that applied to the child followed by a question asking which group
the child most identified with. The majority of the children were either Native Hawaiian or Japanese, both of which accounted for 19% each of the sample. Filipinos accounted for the next largest racial category (15.9%) followed by Caucasians (11.1%) and Chinese (7.9%). Eight percent of the children were Pacific Islanders (Samoan, Marshallese, and Micronesian). Of the remaining 15.9% of children, 9.5% were more than one of the Asian races and listed “Asian” as the race most identified with; and others were as follows: 3.2% Portuguese, 1.6% Mexican, and 1.6% Puerto Rican. The question was unanswered by 1.6%. Children ranged in age from 2 years 0 months to 12 years 7 months. The mean age of the children was 7 years 0 months. Table 1 summarizes the demographic characteristics of the parents and the children.
### Demographic Characteristics of the Parents and the Children

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Frequency</th>
<th>Percent</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Child’s Gender</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>26</td>
<td>41.3</td>
</tr>
<tr>
<td>Male</td>
<td>37</td>
<td>58.7</td>
</tr>
<tr>
<td><strong>Child’s Race</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>7</td>
<td>11.1</td>
</tr>
<tr>
<td>Chinese</td>
<td>5</td>
<td>7.9</td>
</tr>
<tr>
<td>Filipino</td>
<td>10</td>
<td>15.9</td>
</tr>
<tr>
<td>Japanese</td>
<td>12</td>
<td>19.0</td>
</tr>
<tr>
<td>Native Hawaiian</td>
<td>12</td>
<td>19.0</td>
</tr>
<tr>
<td>Pacific Islander</td>
<td>5</td>
<td>8.0</td>
</tr>
<tr>
<td>Other</td>
<td>10</td>
<td>15.9</td>
</tr>
<tr>
<td><strong>Family’s Residence</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hawaii (Big Island)</td>
<td>10</td>
<td>15.9</td>
</tr>
<tr>
<td>Kauai</td>
<td>4</td>
<td>6.3</td>
</tr>
<tr>
<td>Maui/Molokai</td>
<td>5</td>
<td>7.9</td>
</tr>
<tr>
<td>Oahu</td>
<td>44</td>
<td>69.8</td>
</tr>
<tr>
<td><strong>Mother’s Education Level</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Did not complete high school</td>
<td>8</td>
<td>12.7</td>
</tr>
<tr>
<td>High school</td>
<td>25</td>
<td>39.7</td>
</tr>
<tr>
<td>Vocational training or 2 year degree</td>
<td>13</td>
<td>20.7</td>
</tr>
<tr>
<td>4 year degree</td>
<td>10</td>
<td>15.9</td>
</tr>
<tr>
<td>More than a 4 year degree</td>
<td>6</td>
<td>9.5</td>
</tr>
<tr>
<td><strong>Father’s Education Level</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Did not complete high school</td>
<td>8</td>
<td>12.7</td>
</tr>
<tr>
<td>High school</td>
<td>28</td>
<td>44.4</td>
</tr>
<tr>
<td>Vocational training or 2 year degree</td>
<td>11</td>
<td>17.5</td>
</tr>
<tr>
<td>4 year degree</td>
<td>7</td>
<td>11.1</td>
</tr>
<tr>
<td>More than a 4 year degree</td>
<td>4</td>
<td>6.3</td>
</tr>
<tr>
<td><strong>Parent’s Income</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$0 - $25,000</td>
<td>24</td>
<td>38.1</td>
</tr>
<tr>
<td>$25,001 - $50,000</td>
<td>11</td>
<td>17.5</td>
</tr>
<tr>
<td>Over $50,000</td>
<td>25</td>
<td>39.7</td>
</tr>
<tr>
<td><strong>Parent’s Marital Status</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Married</td>
<td>38</td>
<td>60.3</td>
</tr>
<tr>
<td>Living with significant other</td>
<td>7</td>
<td>11.1</td>
</tr>
<tr>
<td>Separated</td>
<td>5</td>
<td>7.9</td>
</tr>
</tbody>
</table>
Overview of Results

An overview of the results are presented which includes the types of oral clefts of the children who participated in the study and versions of the PedsQL that were utilized. The method of how missing data was handled is also included.

Types of Oral Cleft

As illustrated in Table 2, over half of the children (65.1%) were born with cleft lip and palate. One quarter of the children had cleft palate only which comprised 25.4% of the subjects and the remainder of the children, (9.5%) were born with cleft lip only.

Table 2

<table>
<thead>
<tr>
<th>Category</th>
<th>Frequency</th>
<th>Percent</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft Lip Only</td>
<td>6</td>
<td>9.5</td>
</tr>
<tr>
<td>Cleft Palate Only</td>
<td>16</td>
<td>25.4</td>
</tr>
<tr>
<td>Cleft Lip and Palate</td>
<td>41</td>
<td>65.1</td>
</tr>
</tbody>
</table>

Versions of the PedsQL

Health-related quality of life was measured utilizing the PedsQL for 2 to 12 year old children with nonsyndromic oral clefts. The Physical Functioning score is also referred to as the Physical Health Score. The Psychosocial Health Score is the combined Emotional, Social, and School Functioning Scales. The Total Score is the combination of all of the scales. Higher scores indicate better health-related quality of life. Children from 5 to 12 years of age self reported their own health-related quality of life, and parents of children
from 2 to 12 years of age rated their perception of their child’s health-related quality of life.

Five versions of the PedsQL were utilized for the study which included three versions for parents and two versions for children. The parent reports included the following: Parent Report for Toddlers (ages 2-4), Parent Report for Young Children (ages 5-7), and Parent Report for Children (ages 8-12). The child versions included the Young Child Report (ages 5-7) and the Child Report (ages 8-12). Of the 63 families who completed the PedsQL, 23 (36.5%) had a child with cleft lip and/or palate in the 2 to 4 year old (toddler) age group, 12 (19.1%) had children in the 5 to 7 year old (young child) age group, and 28 (44.4%) were in the 8 to 12 year old (child) age group.

Missing Data

Excluding a few questions that were not completed by parents of 2 to 4 year old children which were not applicable such as questions regarding daycare or school, there were very few items missed or unanswered by both the children and their parents. The majority of questionnaires did not have any missing items, and the maximum number of missed items on any one questionnaire was two. Missed items were usually due to the subject inadvertently circling two numbers on one line and leaving the line above or below blank. The mean of the computed items in a scale replaced the missing data.

Results for Specific Aim 1, Including Research Questions 1-6

This section will describe the findings for the first six research questions that include: 1. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life according to each age category.
2. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life according to each age category?

3. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of parents of healthy children?

4. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of healthy children?

5. How do parents of 2 to 12 year old children with nonsyndromic cleft lip and/or palate rate their child’s Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of parents of children with a chronic condition?

6. How do 5 to 12 year old children with nonsyndromic cleft lip and/or palate rate their own Physical Health, Psychosocial Health, and Total health-related quality of life compared to a normative sample of children with a chronic condition.

**Description of Health-Related Quality of Life Scores for Parents and Children According to Age Category**

The highest mean quality of life scores as rated by the parents were in the Physical Functioning domain for all age groups, and the lowest scores were in the School Functioning domain for the 8 to 12 year old children. For the children’s self reports, the highest mean scores were in the Physical Functioning domain for the 8 to 12 year old children, and the lowest scores were in the School Functioning domain for the 5 to 7 year old children. Table 3 summarizes the mean health-related quality of life scores for parents.
and their children according to each domain that was measured with the PedsQL for each of the age categories.

Table 3

<table>
<thead>
<tr>
<th>Domain</th>
<th>Parent 2-4 N = 23</th>
<th>Parent 5-7 N = 12</th>
<th>Parent 8-12 N = 27</th>
<th>Child 5-7 N = 11</th>
<th>Child 8-12 N = 27</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical Functioning</td>
<td>88.32</td>
<td>83.60</td>
<td>88.20</td>
<td>77.27</td>
<td>82.53</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>77.39</td>
<td>82.22</td>
<td>71.30</td>
<td>75.76</td>
<td>74.63</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>81.30</td>
<td>71.67</td>
<td>75.93</td>
<td>72.73</td>
<td>81.67</td>
</tr>
<tr>
<td>School Functioning</td>
<td>76.47</td>
<td>70.00</td>
<td>63.47</td>
<td>71.14</td>
<td>76.25</td>
</tr>
<tr>
<td>Psychosocial Health</td>
<td>78.44</td>
<td>74.57</td>
<td>70.26</td>
<td>72.77</td>
<td>77.51</td>
</tr>
<tr>
<td>Health Summary Total</td>
<td>82.34</td>
<td>77.70</td>
<td>76.52</td>
<td>74.36</td>
<td>79.25</td>
</tr>
</tbody>
</table>

N = 17 for Parent School Functioning

Table 4 illustrates the ranges of health-related quality of life scores for the combined parent proxy reports and children self reports. The maximum score on all domains for parents and children was 100.00 (with the exception of the maximum Total Score for children being 97.83 due to imputing the mean of completed items in a scale for missing
data). The lowest minimum scores for the parent proxy reports were 25.00 for both Social Functioning and School Functioning. The lowest minimum score as reported by the children was 25.00 for Emotional Functioning.

Table 4

Minimum, Maximum, and Range of PedsQL Parent Proxy Scores for Their Children with Cleft Lip and/or Palate and the Children’s Self Report Scores

<table>
<thead>
<tr>
<th>Domain</th>
<th>Parent proxy report N = 62</th>
<th>Child self report N = 38</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Minimum</td>
<td>Maximum</td>
</tr>
<tr>
<td>Physical Functioning</td>
<td>43.75</td>
<td>100.00</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>40.00</td>
<td>100.00</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>25.00</td>
<td>100.00</td>
</tr>
<tr>
<td>School Functioning</td>
<td>25.00</td>
<td>100.00</td>
</tr>
<tr>
<td>Psychosocial Health Summary</td>
<td>48.33</td>
<td>100.00</td>
</tr>
<tr>
<td>Total Score</td>
<td>53.26</td>
<td>100.00</td>
</tr>
</tbody>
</table>

Note: 1) Scores that are not whole numbers are the result of imputing the mean of completed items in a scale for missing data, 2) N = 57 for parent proxy School Functioning Score.
Study Sample Data Compared to Normative Data of Healthy Children

Results are presented in this section that include the study sample of parent proxy reports of their children’s health-related quality of life compared to a normative sample of parent proxy reports for healthy children. In addition, the study sample of children’s self reports of their health-related quality of life compared to a normative sample of healthy children is also presented.

Parents of children with cleft lip and/or palate: PedsQL parent proxy reports of their children’s health-related quality of life compared to a normative sample of parent proxy reports for healthy children.

The normative sample used for comparison included over 5000 children who were heterogeneous with regard to race and ethnicity. With respect to gender, the sample included 52.1% boys and 47.9% girls. The sample was obtained from those who had enrolled in the state of California’s health-insurance program (Varni, Burwinkle, Seid, & Skarr, 2003). One-sample t tests were conducted to determine whether the means obtained from the study sample differed significantly from the normative data.

Interestingly, parents of children with cleft lip and/or palate rated their children’s Physical Functioning significantly higher (p = .05) than parents of healthy children from the normative sample. For all other health-related quality of life (HRQOL) scores, parents of children with an oral cleft rated their children’s scores to be significantly lower than healthy children as follows: Emotional Functioning (p = .01), Social Functioning (p = .01), School Functioning (p = .001), Psychosocial Health Summary score (p = .000), and overall Total Score (p < .05). Table 5 includes a summary of the results.
**Table 5**

*Comparison of PedsQL Parent Proxy Report Scores for Their Children with Cleft Lip and/or Palate (Study Population) and Normative Data of Parent Proxy Scores for Healthy Children*

<table>
<thead>
<tr>
<th>Domain</th>
<th>Children with cleft lip and/or palate (study population)</th>
<th>Healthy children</th>
<th>t score</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical Functioning</td>
<td>87.35 (12.87)</td>
<td>84.08 (19.70)</td>
<td>2.00</td>
<td>.05*</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>75.67 (16.48)</td>
<td>81.20 (16.40)</td>
<td>-2.64</td>
<td>.01**</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>77.10 (18.34)</td>
<td>83.05 (19.66)</td>
<td>-2.56</td>
<td>.01**</td>
</tr>
<tr>
<td>School Functioning</td>
<td>68.82 (21.07)</td>
<td>78.27 (19.64)</td>
<td>-3.36</td>
<td>.001***</td>
</tr>
<tr>
<td>Psychosocial Health Summary</td>
<td>74.13 (14.11)</td>
<td>81.24 (15.34)</td>
<td>-3.97</td>
<td>.000***</td>
</tr>
<tr>
<td>Total Score</td>
<td>78.91 (11.89)</td>
<td>82.29 (15.55)</td>
<td>-2.24</td>
<td>.03*</td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance. *P value = statistical significance at ≤ .05. **P = statistical significance at ≤ .01. *** P value = statistical significance at ≤ .001.

**Children with cleft lip and/or palate:** PedsQL self reports of their health-related quality of life compared to a normative sample of healthy children.

Children with cleft lip and/or palate rated their health-related quality of life lower on all dimensions compared to a normative sample of healthy children as shown.
in Table 6. The mean scores on Physical Functioning, Psychosocial Health Summary, and the overall Total Scores were significantly lower at the p < .05 level, and the School Functioning Scores were significantly lower at the p < .01 level for children with cleft lip and/or palate compared to a healthy normative sample of children. Although scores were lower on Emotional Functioning and Social Functioning for children with cleft lip and/or palate than healthy children, the differences were not statistically significant at the p < .05 level (p = .18 and p = .08., respectively).
Table 6

Comparison of PedsQL Self Report Scores for Children with Cleft Lip and/or Palate (Study Population) and Normative Data for Healthy Children

<table>
<thead>
<tr>
<th>Domain</th>
<th>Children with cleft lip and/or palate (study population)</th>
<th>Healthy children</th>
<th>t score</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical Functioning</td>
<td>81.01 (16.64)</td>
<td>87.77 (13.12)</td>
<td>-2.51</td>
<td>.02*</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>74.96 (19.19)</td>
<td>79.21 (18.02)</td>
<td>-1.37</td>
<td>.18</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>79.08 (20.16)</td>
<td>84.97 (16.71)</td>
<td>-1.80</td>
<td>.08</td>
</tr>
<tr>
<td>School Functioning</td>
<td>74.77 (14.69)</td>
<td>81.31 (16.09)</td>
<td>-2.75</td>
<td>.01**</td>
</tr>
<tr>
<td>Psychosocial Health Summary</td>
<td>76.14 (14.69)</td>
<td>81.83 (13.97)</td>
<td>-2.39</td>
<td>.02*</td>
</tr>
<tr>
<td>Total Score</td>
<td>77.83 (14.71)</td>
<td>83.91 (12.47)</td>
<td>-2.55</td>
<td>.02*</td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance. *P value = statistical significance at ≤ .05. **P = statistical significance at ≤ .01.

Study Sample Data Compared to Normative Data of Children with a Chronic Condition

Results are presented in this section that include the study sample of parent proxy reports of their children’s health-related quality of life compared to a normative sample of parent proxy reports for children with a chronic condition. In addition, the study
sample of children’s self reports of their health-related quality of life compared to a normative sample of children with a chronic condition is also presented.

Parents of children with cleft lip and/or palate: PedsQL parent proxy reports of their children’s health-related quality of life compared to a normative sample of parent proxy reports for children with a chronic health condition.

The normative sample of children with a chronic condition utilized for comparison consisted of over 500 children who had asthma, attention deficit hyperactivity disorder, depression, diabetes, or another chronic condition as reported by their parents (Varni, Burwinkle, Seid, & Skarr, 2003).

Parents of children with cleft lip or palate scored their children’s health-related quality of life higher in all domains than did parents of children who had a chronic health condition. The results are summarized in Table 7. Parents of children with an oral cleft rated their children’s Physical Functioning (p = .000), Emotional Functioning (p < .05), and overall Total Score (p = .000) significantly higher than parents of children with a chronic condition. Social Functioning, School Functioning, and Psychosocial Health Summary Scores were higher than the normative sample, although they were not statistically significant (p = .39, p = .26, and p = .09, respectively).
Table 7

*Comparison of PedsQL Parent Proxy Scores for Their Children with Cleft Lip and/or Palate (Study Population) and Normative Data of Parent Proxy Scores for Children with a Chronic Health Condition*

<table>
<thead>
<tr>
<th>Domain</th>
<th>Mean (SD)</th>
<th>Mean (SD)</th>
<th>t score</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical Functioning</td>
<td>87.35 (12.87)</td>
<td>76.99 (20.20)</td>
<td>6.34</td>
<td>.000***</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>75.67 (16.48)</td>
<td>71.08 (19.75)</td>
<td>2.19</td>
<td>.03*</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>77.10 (18.34)</td>
<td>75.06 (21.75)</td>
<td>.87</td>
<td>.39</td>
</tr>
<tr>
<td>School Functioning</td>
<td>68.82 (21.07)</td>
<td>65.58 (20.75)</td>
<td>1.15</td>
<td>.26</td>
</tr>
<tr>
<td>Psychosocial Health</td>
<td>74.13 (14.11)</td>
<td>71.04 (17.32)</td>
<td>1.72</td>
<td>.09</td>
</tr>
<tr>
<td>Total Score</td>
<td>78.91 (11.89)</td>
<td>73.14 (16.46)</td>
<td>3.82</td>
<td>.000***</td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance. *P value = statistical significance at ≤ .05. **P = statistical significance at ≤ .01. *** P value = statistical significance at ≤ .001.
Children with cleft lip and/or palate: PedsQL self reports of their health-related quality of life compared to a normative sample of children with a chronic condition.

When compared to children with a chronic condition, those with cleft lip and/or palate rated their health-related quality of life higher in all domains compared to the normative sample of children with a chronic condition. School Functioning Scores were significantly higher ($p = .01$), and the Psychosocial Health Summary Scores were significantly higher ($p = .05$) for children with an oral cleft than children with a chronic condition. Physical Functioning, Emotional Functioning, Social Functioning, and the overall Total Scores were higher for children with an oral cleft than those children with a chronic condition, although they were not statistically significant ($p = .57$, $p = .08$, $p = .41$, and $p = .13$, respectively). Results are illustrated in Table 8.
Table 8

Comparison of PedsQL Self Report Scores for Children with Cleft Lip and/or Palate (Study Population) and Normative Data for Children with a Chronic Health Condition

<table>
<thead>
<tr>
<th>Domain</th>
<th>Children with cleft lip and/or palate (study population)</th>
<th>Children with chronic condition</th>
<th>t score</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical Functioning</td>
<td>81.01 (16.64)</td>
<td>79.47 (17.07)</td>
<td>.57</td>
<td>.57</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>74.96 (19.19)</td>
<td>69.32 (21.36)</td>
<td>1.81</td>
<td>.08</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>79.08 (20.16)</td>
<td>76.36 (21.57)</td>
<td>.83</td>
<td>.41</td>
</tr>
<tr>
<td>School Functioning</td>
<td>74.77 (14.69)</td>
<td>68.27 (19.05)</td>
<td>2.73</td>
<td>.01**</td>
</tr>
<tr>
<td>Psychosocial Health</td>
<td>76.14 (14.69)</td>
<td>71.32 (17.13)</td>
<td>2.02</td>
<td>.05*</td>
</tr>
<tr>
<td>Summary</td>
<td>77.83 (14.71)</td>
<td>74.16 (15.38)</td>
<td>1.54</td>
<td>.13</td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance. *P value = statistical significance at ≤ .05. **P = statistical significance at ≤ .01.

Results for Specific Aim 2, Including Research Question 7

Research question 7 asks, “Is there parent-child agreement between parent proxy reports and child self reports for 5 to 12 year old children with nonsyndromic cleft lip and/or palate when rating Physical Health, Psychosocial Health, and Total health-related quality of life?” The findings are described below.
Children with Cleft Lip and/or Palate and Their Parents: Agreement between Parent Proxy Reports and Child Self Reports of Health-Related Quality of Life

Parent-child agreement was analyzed to determine if parent’s perceptions were the same as their children’s with regard to the child’s health-related quality of life. The data for the 37 parent-child dyads were analyzed to determine the parent-child agreement, or concordance, for the pairs.

Parents rated their children’s Physical Functioning higher than their children rated their own Physical Functioning. On all other domains, children with cleft lip and/or palate rated their health-related quality of life higher than their parents rated them. Children rated their Emotional Functioning, Social Functioning, and School Functioning higher than their parents did. Children rated their School Functioning significantly higher than their parents did (p < .05). The Psychosocial Health Summary Score and overall Total Scores were also rated higher by the children than their parents. Other than the School Functioning Score, the other health-related quality of life scores were not statistically significant. The results are summarized in Table 9 which includes the mean scores and standard deviations for parents and children as well as the paired sample t test scores, p values for the t test scores, the Pearson product moment correlation coefficients, and the p values for the correlations. It should be noted that the mean scores and standard deviations in Table 9 are different than those previously reported since only the paired parent/child dyads are reported (n = 37) as opposed to the entire sample.
Table 9

*Comparison of PedsQL Parent Proxy Scores for Their Children with Cleft Lip and/or Palate and Their Children's Self Report*

<table>
<thead>
<tr>
<th>Domain</th>
<th>Parent proxy report</th>
<th>Child report</th>
<th>t score</th>
<th>p value</th>
<th>r</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical Functioning</td>
<td>87.33 (13.76)</td>
<td>81.59 (16.47)</td>
<td>1.61</td>
<td>.12</td>
<td>-.03</td>
<td>.87</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>74.77 (17.87)</td>
<td>75.36 (19.29)</td>
<td>-.15</td>
<td>.88</td>
<td>.20</td>
<td>.24</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>74.86 (19.88)</td>
<td>79.19 (20.43)</td>
<td>-.98</td>
<td>.33</td>
<td>.12</td>
<td>.48</td>
</tr>
<tr>
<td>School Functioning</td>
<td>66.59 (19.50)</td>
<td>74.90 (14.87)</td>
<td>-2.19</td>
<td>.04*</td>
<td>.11</td>
<td>.50</td>
</tr>
<tr>
<td>Psychosocial Health Summary</td>
<td>72.08 (14.64)</td>
<td>76.35 (14.83)</td>
<td>-1.24</td>
<td>.22</td>
<td>-.02</td>
<td>.93</td>
</tr>
<tr>
<td>Total Score</td>
<td>77.40 (11.99)</td>
<td>78.17 (14.76)</td>
<td>-.24</td>
<td>.81</td>
<td>-.09</td>
<td>.61</td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance. *P value = statistical significance at ≤ .05. Effect sizes are as follows: small (.10), medium (.30), large (.50) (Cohen, 1987).

**Children with Cleft Lip and/or Palate and Their Parents: Agreement between**

**Parent Proxy Reports and Child Self Reports on Individual Items**

Paired sample t tests and correlations were conducted on individual items of the PedsQL. Only the significant findings will be presented. In the Physical Functioning domain, for the item “lifting something heavy,” the means differed significantly between parent and child (t = 3.11, p = .004). In the Emotional Functioning domain, correlations
with a medium effect size occurred between parent and child on the “feeling afraid or scared” ($r = .33, p = .05$) and the “trouble sleeping” items ($r = .34, p = .04$). In the Social Functioning domain, the correlation between parents and children related to “getting teased by other children” was close to a large effect size ($r = .47, p = .004$). The School Functioning domain showed significant differences between means on the item “paying attention in class” ($t = -3.27, p = .002$) and “doing the same school activities as peers” ($t = -2.02, p = .05$).

**Results for Specific Aim 3, Including Research Question 8**

Research Question 8 asks, “Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate that are attributed to demographic variables including gender, age, and race of the children; and the geographic residence, educational level, income, marital status, and age of the parents?” The results are described below.

Independent $t$ tests were conducted between gender and health-related quality of life scores which included Physical Health, Psychosocial Health, and Total scores for the parents and the children. For all other demographic variables, a one-way analysis of variance (ANOVA) was conducted between each demographic variable and the health-related quality of life scores including Physical Health, Psychosocial Health, and Total scores for the parents and the children. The demographic variables included the following: race child identified with the most, island of residence, mother’s highest level of education, father’s highest level of education, parent’s combined annual income,
parent’s marital status, mother’s age when child was born, and father’s age when child was born. Parent’s ages were categorized into ordinal variables.

Results revealed that there were no significant differences identified on any of the health-related quality of life scores from the parent’s or the children’s perspective based on any of the demographic variables. Regarding gender, there were no significant differences on the health-related quality of life scores related to gender from the parent’s or children’s perspective. Parent results are as follows: Physical Health Score $t(60) = -0.11, p = 1.91$; Psychosocial Summary Score $t(60) = 0.09, p = .93$; and Total Score $t(60) = 0.05, p = .96$. Child results are as follows: Physical Health Score $t(36) = 0.19, p = .85$; Psychosocial Summary Score $t(36) = -0.13, p = .90$; and Total Score $t(36) = -0.01, p = .99$. For the remainder of the findings, Tables 10 and 11 illustrate the ANOVA results including the F-ratios, degrees of freedom, and probability values for the parents and children respectively.
Table 10

*Relationship between Parent Proxy Reports of Health-Related Quality of Life Scores and Demographic Variables*

<table>
<thead>
<tr>
<th>Demographic Variable</th>
<th>Physical Health Score</th>
<th>F (df)</th>
<th>p</th>
<th>Psychosocial Summary Score</th>
<th>F (df)</th>
<th>p</th>
<th>Total Score</th>
<th>F (df)</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Child’s Age</td>
<td>0.50</td>
<td>2.17</td>
<td>0.12</td>
<td>0.53</td>
<td>0.23</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>(2, 59)</td>
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<td></td>
<td>(2, 59)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Child’s Race</td>
<td>0.72</td>
<td>0.58</td>
<td>0.83</td>
<td>0.61</td>
<td>0.81</td>
<td></td>
<td></td>
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<tr>
<td>(11, 49)</td>
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<td></td>
<td>(11, 49)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Island of Residence</td>
<td>0.32</td>
<td>2.00</td>
<td>0.11</td>
<td>1.39</td>
<td>0.25</td>
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<tr>
<td>(4, 57)</td>
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</tr>
<tr>
<td>Mother’s Education</td>
<td>0.80</td>
<td>0.58</td>
<td>0.72</td>
<td>0.69</td>
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<td>(5, 55)</td>
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<td></td>
<td></td>
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</tr>
<tr>
<td>Father’s Education</td>
<td>0.87</td>
<td>1.46</td>
<td>0.22</td>
<td>1.46</td>
<td>0.22</td>
<td></td>
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<td></td>
</tr>
<tr>
<td>Family Income</td>
<td>1.9</td>
<td>0.37</td>
<td>0.69</td>
<td>0.06</td>
<td>0.94</td>
<td></td>
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</tr>
<tr>
<td>Marital Status</td>
<td>0.52</td>
<td>0.27</td>
<td>0.85</td>
<td>0.12</td>
<td>0.95</td>
<td></td>
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<td>(3, 57)</td>
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<td></td>
<td>(3, 57)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mother’s Age</td>
<td>0.69</td>
<td>0.55</td>
<td>0.70</td>
<td>0.57</td>
<td>0.69</td>
<td></td>
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</tr>
<tr>
<td>Father’s Age</td>
<td>0.25</td>
<td>0.53</td>
<td>0.59</td>
<td>0.24</td>
<td>0.79</td>
<td></td>
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<td>(2, 38)</td>
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<td>(2, 38)</td>
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<td></td>
<td></td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance. df = degrees of freedom.
Table 11

Relationship between Child Self Reports of Health-Related Quality of Life Scores and Demographic Variables

<table>
<thead>
<tr>
<th>Demographic Variable</th>
<th>Physical Health Score</th>
<th>Psychosocial Summary Score</th>
<th>Total Score</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>F (df)</td>
<td>p</td>
<td>F (df)</td>
</tr>
<tr>
<td>Child’s Age</td>
<td>.77 (.36)</td>
<td>.39</td>
<td>.81 (.36)</td>
</tr>
<tr>
<td>Child’s Race</td>
<td>1.03 (.25)</td>
<td>.45</td>
<td>1.31 (.25)</td>
</tr>
<tr>
<td>Island of Residence</td>
<td>1.31 (.34)</td>
<td>.29</td>
<td>.22 (.34)</td>
</tr>
<tr>
<td>Mother’s Education</td>
<td>.99 (.32)</td>
<td>.44</td>
<td>.46 (.32)</td>
</tr>
<tr>
<td>Father’s Education</td>
<td>.56 (.28)</td>
<td>.73</td>
<td>1.08 (.28)</td>
</tr>
<tr>
<td>Family Income</td>
<td>1.76 (.33)</td>
<td>.19</td>
<td>.77 (.33)</td>
</tr>
<tr>
<td>Marital Status</td>
<td>1.82 (.34)</td>
<td>.16</td>
<td>1.06 (.34)</td>
</tr>
<tr>
<td>Mother’s Age</td>
<td>.71 (.32)</td>
<td>.55</td>
<td>.24 (.32)</td>
</tr>
<tr>
<td>Father’s Age</td>
<td>1.33 (.21)</td>
<td>.29</td>
<td>1.46 (.21)</td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance. df = degrees of freedom.
Results for Aim 4, Including Research Questions 9 and 10

Question 9 asks, “Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate who do or do not have impaired speech?” Question 10 asks, “Are there differences between Physical Health, Psychosocial Health, and Total health-related quality of life scores for 2 to 12 year old children with nonsyndromic cleft lip and/or palate who do or do not have impaired hearing?” The results are addressed for the fourth aim of this study.

Data for speech and hearing were obtained from the oral cleft database repository. No hearing or speech assessments were conducted for the purpose of this study. If more than one speech or hearing assessment was documented in the repository, data were utilized from the date closest to when the parent or child completed the PedsQL. If there was no speech or hearing data in the repository for the subjects, they were not included in the analysis.

Speech: Articulation Impression and Speech Impairment

Parent proxy report scores related to articulation impression and speech impairment.

There were 39 children who had an articulation impression reported in the oral cleft repository whose parents completed the PedQL. The child’s speech articulation was categorized according to speech impairment. Of the 39 children, 6 (15.4%) had normal speech, 21 (53.8%) had mild to moderate speech impairment, and 12 (30.8%) had severe speech impairment. For Physical Functioning, School Functioning, Psychosocial Health Summary, and Total Scores, mean scores were less for children who had speech
impairments versus children who had normal speech according to the parent proxy reports; however, scores varied on the Emotional and Social Functioning domains. None of the mean differences between the groups displayed statistical significance as illustrated in Table 12. Also, individual items did not reveal statistically significant differences.

Table 12

*Relationship between Parent Proxy Reports of Health-Related Quality of Life Scores for Their Children and the Child’s Speech Impairment Status*

<table>
<thead>
<tr>
<th>Domain</th>
<th>Normal speech Mean (SD)</th>
<th>Mild to moderate speech impairment Mean (SD)</th>
<th>Severe speech impairment Mean (SD)</th>
<th>F</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical Functioning</td>
<td>94.27 (5.73)</td>
<td>88.40 (12.58)</td>
<td>88.28 (14.05)</td>
<td>.58</td>
<td>.56</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>78.33 (21.13)</td>
<td>71.90 (16.54)</td>
<td>80.42 (17.77)</td>
<td>.98</td>
<td>.39</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>75.00 (21.45)</td>
<td>77.62 (16.93)</td>
<td>68.75 (22.17)</td>
<td>.81</td>
<td>.45</td>
</tr>
<tr>
<td>School Functioning</td>
<td>78.75 (20.16)</td>
<td>65.50 (24.89)</td>
<td>64.24 (14.48)</td>
<td>.71</td>
<td>.50</td>
</tr>
<tr>
<td>Psychosocial Health Summary</td>
<td>76.39 (18.00)</td>
<td>71.91 (14.04)</td>
<td>71.28 (13.21)</td>
<td>.28</td>
<td>.76</td>
</tr>
<tr>
<td>Total Score</td>
<td>83.27 (13.15)</td>
<td>77.90 (11.35)</td>
<td>77.26 (11.91)</td>
<td>.59</td>
<td>.56</td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance.
Child report scores related to articulation and speech impairment.

Articulation impression was documented in the oral cleft repository on 24 of the children who completed the PedsQL. Speech articulation was categorized according to speech impairment. Only 3 (12.5%) of the children had normal speech. Thirteen (54.2%) of the children had mild to moderate articulation impairment, and 8 (33.3%) had severe articulation impairment.

Mean health-related quality of life scores were lower in each domain for those with a speech impairment compared to those children with normal speech according to the child’s perspective, although they were not statistically significant. For the child’s individual item scores, the only item that approached statistical significance related to speech impairment was “getting teased by other children” ($F = 3.33$, $p = .056$). Results are summarized in Table 13.
Table 13

*Relationship between Child Self Reports of Health-Related Quality of Life Scores and Speech Impairment Status*

<table>
<thead>
<tr>
<th>Domain</th>
<th>Normal speech</th>
<th>Mild to moderate speech impairment</th>
<th>Severe speech impairment</th>
<th>F</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical Functioning</td>
<td>86.46 (16.04)</td>
<td>78.13 (19.26)</td>
<td>75.39 (19.15)</td>
<td>.37</td>
<td>.69</td>
</tr>
<tr>
<td>Emotional Functioning</td>
<td>81.67 (20.21)</td>
<td>69.23 (18.13)</td>
<td>74.79 (24.11)</td>
<td>.51</td>
<td>.61</td>
</tr>
<tr>
<td>Social Functioning</td>
<td>86.67 (15.28)</td>
<td>76.15 (20.02)</td>
<td>68.75 (25.18)</td>
<td>.80</td>
<td>.46</td>
</tr>
<tr>
<td>School Functioning</td>
<td>85.00 (5.00)</td>
<td>69.81 (13.48)</td>
<td>75.00 (17.29)</td>
<td>1.40</td>
<td>.27</td>
</tr>
<tr>
<td>Psychosocial Health Summary</td>
<td>84.45 (13.47)</td>
<td>71.67 (13.92)</td>
<td>72.36 (17.98)</td>
<td>.88</td>
<td>.43</td>
</tr>
<tr>
<td>Total Score</td>
<td>85.15 (14.35)</td>
<td>73.93 (14.82)</td>
<td>73.42 (18.18)</td>
<td>.67</td>
<td>.52</td>
</tr>
</tbody>
</table>

All p values are reported as 2-tailed significance.

**Hearing: Severity of Hearing Loss**

In the sample of 60 children who had hearing data documented in the repository and who had parent proxy reports completed, 23 (38.3%) had normal hearing, 33 (55%) had mild to moderate-severe hearing loss, and 4 (6.7%) had severe to profound hearing loss. Since there were only 4 children with severe to profound hearing loss, data were
regrouped into normal hearing, 23 (38%) or some degree of hearing loss, 37(62%). The hearing loss category ranged from mild to profound hearing loss. Results are presented for the parent proxy reports and the child self reports.

**Parent proxy report scores related to severity of hearing loss.**

Results revealed that there was no significant effect of severity of hearing loss on health-related quality of life scores including Physical Functioning ($F = .29, p = .59$), Emotional Functioning ($F = .72, p = .40$), Social Functioning ($F = .50, p = .48$), School Functioning ($F = .90, p = .35$), Psychosocial Health Summary ($F = .10, p = .75$), and the Total Score ($F = .01, p = .93$). When individual items were analyzed with ANOVAs, the only item that was significantly affected by severity of hearing loss according to the parent’s perspective was, “missing school/daycare to go to the doctor or hospital” ($F = 4.07, p = .05$).

**Child self report scores related to severity of hearing loss.**

For the child self reports, there were 36 children who had hearing data documented in the repository. Of the 36 children, 14 (38.9%) had normal hearing, and 22 (61.1%) had some degree of hearing loss.

Child self report scores for health-related quality of life were compared with severity of hearing loss. Results indicated that severity of hearing loss did not significantly affect health-related quality of life scores. The results of the ANOVAs are as follows: Physical Functioning ($F = .84, p = .37$), Emotional Functioning ($F = 1.66, p = .21$), Social Functioning ($F = .19, p = .67$), School Functioning ($F = .18, p = .68$), Psychosocial Summary Score ($F = .03, p = .86$), and Total Score ($F = .22, p = .65$). None of the individual items were statistically significant.
Wearing a Hearing Aid

Data were gathered from the oral cleft database repository to determine if the child wore a hearing aid or not. Data were categorized as “yes” or “no.” If the child wore a hearing aid in either ear or in both ears, the child was placed in the “yes” category. Children in the “no” category did not wear a hearing aid in either ear. Analysis of variance was conducted to determine if health-related quality of life scores differed significantly for children who did and did not wear a hearing aid. Out of the 60 children who had data hearing data documented in the repository, 53 (88.3%) did not wear a hearing aid and 7 (11.7%) did wear a hearing aid.

Parent proxy report scores related to wearing a hearing aid.

According to the parents, children who wore a hearing aid had lower health-related quality of life scores in each domain. A significant effect was found in School Functioning ($F = 4.05$, $p = .05$) between children who did and did not wear a hearing aid according to the parent’s perspective. Results revealed no significant effect of wearing a hearing aid on Physical Functioning ($F = .47$, $p = .50$), Emotional Functioning ($F = .06$, $p = .81$), or Social Functioning ($F = 2.73$, $p = .10$). The Psychosocial Summary Health Score ($F = 3.23$, $p = .08$) and Total Health Scores ($F = 2.87$, $p = .10$) were also not significantly different based on whether or not the child wore a hearing aid according to the parents. An individual item that was significantly affected by wearing a hearing aid was “forgetting things” according to the parents ($F = 7.10$, $p = .01$).
Child self report scores related to wearing a hearing aid.

Mean total scores were similar regardless of whether or not the child wore a hearing aid according to the child self reports, and no significant differences were found. Mean scores on Physical Functioning ($F = .50, p = .48$), Emotional Functioning ($F = 1.03, p = .32$), Social Functioning ($F = .10, p = .75$), Psychosocial Health Summary ($F = .17, p = .69$), and the Total Score ($F = .29, p = .59$) were actually higher for children who wore a hearing aid compared to those who did not. Mean School Functioning scores for children who wore a hearing aid were lower than their counterparts who did not wear a hearing aid, although the scores were not significantly different ($F = .30, p = .59$). Individual items did not differ significantly based on wearing a hearing aid.
Chapter 5: Discussion

Health-related quality of life encompasses physical and psychosocial factors including social, emotional, and cognitive domains (Varni, Seid, & Rode, 1999). As described in the concept analysis presented in this paper, health-related quality of life can be perceived on a continuum from optimal to sub-optimal which is dependent upon one’s perception of satisfaction with health and perception of well-being. This study was conducted to increase an understanding of health-related quality of life for children with cleft lip and/or palate from the parent’s perspective as well as the child’s perspective. Subjective measurement of parent’s and children’s perceptions of health-related quality of life can be useful in clinical practice as a screening tool to monitor and evaluate health, to obtain information needed to provide prophylactic intervention, to aid in clinical decision making, and as an end point in clinical trials (WHO, 1994).

This is the first study in the United States to the researcher’s knowledge to measure health-related quality of life for toddler, preschool, and school-aged children from the parent’s and children’s perspectives (excluding child perspective for toddlers) for those individuals with cleft lip and/or palate. It is also the first study known to include a culturally diverse population of children with an oral cleft that focuses on health-related quality of life. While literature exists that focuses on health-related quality of life for adolescents with an oral cleft, the literature is scant related to understanding the concept for toddler, preschooler, and school-aged children. The studies conducted in Germany (Kramer et al., 2008; Kramer et al., 2009; Sagheri, Ravens-Sieberer, Braumann, & von Mackensen, 2009) add to the body of knowledge in understanding health-related quality of life for children with oral clefts. In the United States, Damiano and colleagues (2007)
evaluated health-related quality of life of children with oral clefts from the mother’s perspective although the children’s perceptions were not measured. Since parent proxy reports of health-related quality of life were designed to complement rather than substitute for the child’s self report (Varni, Limbers, & Burwinkle, 2007), this study aimed to understand health-related quality of life for children with cleft lip and/or palate by including both the parent’s and the children’s perspectives.

The discussion section is presented for each of the specific aims with the purpose of drawing conclusions about the meaning of the findings and results (Polit & Beck, 2006). Interpretation of the results will focus on the practical meaning within the scope of the conceptual model that was developed for the study. The study limitations, contributions to the nursing literature, and implications for future research will also be discussed.

**Discussion for Specific Aim 1**

This study compared parent proxy and child self report scores of health-related quality of life for children with cleft lip and/or palate compared to normative data of healthy children and children with a chronic condition. “Generic health-related quality of life instruments enable comparison across pediatric populations and facilitate benchmarking with healthy population norms,” (Varni & Limbers, 2009, p. 845).

**Parent Proxy and Children’s Report Scores Compared to Healthy Children**

**Physical health.**

An unexpected finding of this study was that parents of children with an oral cleft rated their children’s health-related quality life as being significantly higher than parents of healthy children. Since children with oral clefts, especially those with cleft palate, may have physical conditions that could have a negative impact on health-related quality of
life, this finding was not anticipated. These findings may be related to three factors. First, the PedsQL 4.0 is a generic instrument; therefore, it does not measure specific physical conditions that may be problematic for children with an oral cleft. For example, the Physical Health section of the PedsQL measures items such as walking more than a block, running, and heavy lifting which children with musculoskeletal, respiratory, neurological, or other conditions may have difficulty with, however would not usually be expected to be a problem for children with cleft lip and/or palate. Since there is currently no condition-specific instrument to measure health-related quality of life for children with cleft lip and/or palate, the PedsQL is not as reflective of Physical Health as it may be for other childhood chronic conditions, although the Psychosocial Health domain is very applicable for children with oral clefts.

A second possible reason for parents in the study rating their children’s Physical Health higher than those who have healthy children may be due to resilience factors which lead to the parent focusing on the child’s unaffected body systems. As noted when conducting a focus group with parents who had children with an oral cleft, parents may center on the positive aspects of their child’s health. For example, one parent said, “We are so happy that she’s healthy, this (the cleft) is just cosmetic. Her heart and lungs are fine.” Parents may also focus on “it could have been worse,” again emphasizing the child’s positive aspects of health (Sinclair & Neal, 2011).

A third possible reason for the parents of children with an oral cleft rating their children’s Physical Health higher than those parents who have healthy children is that the literature has shown that parents are likely to rate their children’s health-related quality of life in relation to their satisfaction of care. As stated by Varni and colleagues, “…parents
of pediatric patients are likely to report satisfaction or dissatisfaction with medical care
based on their perception of their child’s health-related outcomes as a result of
treatment,” (Varni, Burwinkle, & Lane, 2005, p. 2). The PedsQL has been shown to be
related to quality of pediatric primary care (Seid, et al., 2001). Since the parents in the
current study rated their child’s health-related quality of life higher than even those
parents who had healthy children, it is reasonable to believe that their reason for doing so
is directly related to their satisfaction with care in the oral cleft clinic. Since the clinic had
only been established for 2 to 3 years at the time of data collection, parents may have
been very grateful to have the opportunity to experience comprehensive care from a
variety of specialists all in one visit as opposed to nonintegrated care which some of the
families in the study had prior to development of the cleft clinic. Overall satisfaction with
care in other cleft centers has been rated by parents as “excellent” when measured
(Knapke, Bender, Prows, Schultz, & Saal, 2010). In addition, since the size of the clinic
that the study population attended is small in comparison to many cleft clinics, the care is
personal and individualized which may also increase parent satisfaction. Since parent
satisfaction with pediatric care is also related to getting their questions answered
regarding care and treatment, having confidence in the physicians, and being satisfied
with the behavior of the physicians and nurses (Ammentorp, Mainz, & Sabroe, 2006), it
is reasonable to conclude that these factors may have also influenced parents in this study
to have rated their children’s Physical Health Score so highly.

Unlike their parents, children with cleft lip and/or palate rated their Physical Health
lower than their healthy peers. Although not necessarily true for children with cleft lip
only, these results would be expected based on the fact that children with cleft palate
have hearing, speech, and dental conditions that require longitudinal evaluation, treatment, follow-up, and intervention including surgical intervention (ACPA, 2009). As previously stated, the PedsQL did not specifically measure these particular health items.

**Psychosocial health: social, emotional, and cognitive functioning.**

Parents of children with cleft lip and/or palate in this study rated their children’s Emotional Functioning and Social Functioning lower than parents who rated their healthy children. Findings are congruent with Damiano and colleagues (2007) who also reported that mothers of children with cleft lip and/or palate rated their children’s Psychosocial Health significantly lower than norms of healthy children.

Children in this study also rated their Emotional Functioning and Social Functioning lower than healthy children. Since children with oral clefts are known to be at risk for social and emotional difficulty (Murray et al., 2010), the findings of this component of the study were expected. Any type of visible difference can have psychological sequela which may have a negative effect on a child’s body image, self-esteem, social interaction, and quality of life (Rumsey & Harcourt, 2007). These findings are consistent with other researchers who found that parents of children with cleft lip and/or palate reported that their children were more anxious, less happy with appearance, had lower self-esteem, had greater behavior problems, and were teased more often compared to those without an oral cleft (Hunt, Burden, Hepper, Stevenson, & Johnston, 2007). Preschool-aged children with oral clefts may be considered to be less friendly and less helpful when interacting with peers (Kapp-Simon, 1986; Krueckeberg, Kapp-Simon, & Ribondy, 1993) while school-aged children with oral clefts are at risk for teasing, bullying, and social exclusion (Kapp-Simon, 2004). These factors are likely to have contributed to lower health-related quality
of life scores on the Psychosocial Health Summary domain. In contrast, Kramer and colleagues (2009) found that German school-aged children with oral clefts had similar or higher health-related quality of life scores on the KINDL when matched with unaffected peers with the exception of the social dimension, whereby children with oral clefts had reduced scores. The researchers also conducted a health-related quality of life study with 5 to 6 years olds with oral clefts and found that the KINDL scores were lowest in the self-esteem dimension (Kramer et al., 2008). In a similar study conducted in Germany with 4 to 7 year old children, results revealed that the children with oral clefts had slightly lower total health-related quality of life scores than unaffected peers although these differences were not statistically significant (Sagheri, Ravens-Sieberer, Braumann, & von Mackensen, 2009).

In the School Functioning domain, parents and their children with oral clefts both rated the child’s health-related quality of life lower compared to normative data of healthy children. These findings were expected based on current research related to cognitive and school functioning in children with oral clefts. While most studies indicate that children with oral clefts have overall average IQs, there is some evidence that shows that the children have lower verbal IQs related to mild language deficits which may result in a high incidence of learning disabilities, specifically reading disabilities, memory deficits, and language disorders (Richman & Nopoulos, 2009). In a longitudinal study, researchers found that 7 year old children with oral clefts scored significantly lower than controls on tests of cognitive development such as IQ, language, and school achievements compared to children who did not have an oral cleft (Hentges et al., 2011).

From a social standpoint at school, Murray and colleagues (2010) found that children
with oral clefts had increased rates of social problems and social relationships based on teacher reports as well as direct observations and doll play.

**Parent Proxy and Children’s Report Scores Compared to Children with a Chronic Condition**

**Physical health.**

Parents of children with cleft lip and/or palate rated their children’s Physical Health higher in all domains than did parents of children with a chronic condition. The children in the study with cleft lip and/or palate also rated their Physical Health higher than did children who had a chronic condition. For children with cleft lip, the condition is not usually considered to be a “chronic condition” since after the surgical repair, physical limitations are resolved although secondary surgical intervention may occur between the ages of 5 to 8 or during early adolescence (Reinisch, Wai-Yee, & Urata, 2009). Children with cleft palate are considered to have a “chronic condition” since these children experience the need for ongoing longitudinal treatment and follow-up. Some children with other types of chronic conditions experience a more severe disability and resulting affect on physical functioning than do children with an oral cleft. For example, many children with other chronic conditions may require daily medication, respiratory therapy, routine catheterization, wheelchair for mobility, or gavage feedings. Others with a chronic condition may experience a life-threatening illness (Allen & Vessey, 2004). Children with cleft palate are considered to be “somewhere in the middle” in terms of having a chronic condition, although not as severe as many children who also have other types of chronic conditions which may account for their health-related quality of life scores when compared to healthy children and children with a chronic condition.
**Psychosocial health: social, emotional, and cognitive functioning.**

Parents of children with cleft lip and/or palate rated their children’s psychosocial health-related quality of life higher in all domains than did parents of children with a chronic condition which included Emotional Functioning, Social Functioning, School Functioning, overall Psychosocial Health, and Total Score. The children in the study with cleft lip and/or palate also rated their psychosocial health-related quality of life higher in all domains than did children who had a chronic condition.

As with physical functioning, children with an oral cleft may have challenges with social, emotional, and school functioning; however, these challenges may not be as pronounced as those children with more severe types of chronic conditions. The effect that the child’s condition has on their psychosocial health should take into account how the condition affects the child’s progress of achieving developmental milestones. While a chronic condition can interfere with progression of normal development, it also provides an opportunity for the child to master and enhance self-esteem (Dixon & Stein, 2006).

There are many factors that affect school functioning for children with a chronic condition. A child’s ability to attend school, manage the condition, sense of self, teacher and family expectations, and components of the school environment are all factors that can promote or hinder academic success. In addition, absenteeism, mobility issues, fatigue, medications, and treatments may interfere with school functioning (Allen & Vessey, 2004). While children with cleft palate may experience some challenges related to school functioning as previously mentioned, children with other types of chronic conditions may experience a higher degree of impact on school functioning which most
likely accounts for the middle range health-related quality of life scores when compared to healthy peers and those with a chronic illness.

**Discussion for Specific Aim 2**

As described in the conceptual model presented in this paper, important factors influencing the health-related quality of life for children with an oral cleft are the parent’s perception of the impact that the cleft has on the child’s life as well as the child’s perception of the impact that the cleft has on his or her life. This study compared the health-related quality of life scores between parents and their children to determine if there was concordance between the scores.

Parents rated their children’s physical functioning higher than the children rated their own physical functioning; however, the children’s scores were higher than their parents in all other domains, especially with regard to school functioning. These findings were similar to those reported by Kramer and colleagues, whereby the parents of German 5 to 6 year old (2008) and 8 to 12 year old children (2009) with cleft lip and/or palate rated their children’s health-related quality of life lower in all dimensions than the children did on the KINDL.

Children in this study demonstrated different views from their parents, with very small effect sizes in most of the domains. The highest parent-child agreement was in the Emotional Functioning domain which still only had an effect size of .20.

Upton and colleagues (2008) conducted a systematic review of the literature to evaluate parent-child agreement on health-related quality of life domains. The authors found that 17 out of the 19 papers reported Pearson or Spearman correlation coefficients or intraclass correlation coefficients. For studies that utilized the PedsQL, there was
moderate (.30-.50) or good (> .50) agreement for all subscales of the PedsQL in the majority of the studies, whereas four studies found poor agreement. The authors reported that 17 of the studies found differences between parent proxy mean scores and child self report mean scores although most of the differences were small and not statistically significant. Other researchers reported that differences in the levels of parent-child agreement previously reported may be an artifact of the statistical method used. The researchers found consistency between child and parent proxy reports to be low on the PedsQL with intra-class correlation coefficients ranging from .02 to .23. The child’s age, domains that are measured, and the parent’s own quality of life can also affect levels of agreement (Cremeens, Eiser, & Blades, 2006).

Upton and colleagues (2008) pointed out that the PedsQL is comprised of items that focus primarily on what a child can do (18 out of 23) as opposed to how the child feels; therefore, it is possible that good agreement occurs when the nature of the item is observable. Several studies have shown that high correlations occur in observable areas such as physical aspects and a low correlation occurs in areas that are not directly observable such as emotional domains (Eiser & Morse, 2001). With the exception of the Physical Health Score, the children in the study felt that they did better in all other dimensions than what their parents perceived. Some researchers focus on health-related quality of life in terms of self-discrepancy theory and the individual’s ability to achieve daily goals (Eiser et al., 1999). During the school-age years, the child’s psychosocial development is heavily focused on being industrious, becoming more independent, developing increased socialization skills, competing, and comparing ones self with others (Hockenberry, 2004) which may account for perceived satisfaction of well-being.
resulting in higher health-related quality of life scores from the child’s perspective.
Furthermore, parents of school-aged children often lack first hand experience regarding
their child’s social interactions and school experiences (Ravens-Sieberer, et al., 2006);
therefore, they may not have an accurate account of how the child interacts with others in
the school environment. Parents may compare their child’s psychosocial functioning with
siblings or other children who do not have an oral cleft and perceive their child’s
psychosocial health as being sub-optimal compared to others. The parent’s expectations
and concerns about the child may also influence the parent’s perception related to the
child’s health related quality of life.

Another factor that must be taken into consideration is the method of data collection
utilized for obtaining parent proxy and child self reports. For questionnaires that are
obtained through mail surveys, the possibility exists that the parent and child discussed
items on the questionnaire which could result in answers that are more similar. In
addition, even if the questionnaire was administered in a clinic, and the parent and child
were left unattended, it is possible that the parents and children could discuss or compare
answers. For example, in the study utilized for comparative normative data for this study
(Varni, Burwinkle, Seid, & Skarr (2003), the procedure for data collection was a mail
survey. Parents and children were instructed to complete the PedsQL separately. For 5 to
7 year old children, parents were instructed to complete their survey first before assisting
children with their questionnaire. Although one would expect that parents followed the
directions provided, there is no guarantee that the procedure was followed as instructed.
It is also possible that the parent influenced the child’s answers. In this study, the nurse
investigator stayed in the room with the parent and child to discourage any communication about specific items on the questionnaire.

Although parent proxy reports are beneficial to utilize if the child is too young or incapacitated to self-report, the negative drawback from parent proxy reports is that they may not accurately describe the child's perception of how the child is functioning or feeling (Connonlly & Johnson, 1999). This study confirms that for children with cleft lip and/or palate, the children should have the opportunity to rate their own health-related quality of life. Although self-report from the children is advocated, parent proxy reports are valued in providing additional, complementary information (Varni, Limbers, & Burwinkle, 2007).

**Discussion for Specific Aim 3**

None of the demographic variables had an effect on health-related quality of life scores for children or parents in this study. These findings were not consistent with those reported by Damiano and colleagues (2007) who reported that higher income and the mother's marital status (being married) were associated with higher quality of life scores for children with an oral cleft. The researcher's utilized a more incremental income scale than the one utilized for this study which may have contributed to the difference in findings. Kramer et al. (2009) found for school-aged children that boys had lower quality of life scores than girls with orofacial clefts, although no significant differences were found regarding gender for this study. Kramer et al. (2008) also found no differences with regard to gender for preschool-aged children with oral clefts.

Hutchinson & colleagues (2010) conducted a meta-analysis of the psychosocial effects of cleft lip and palate with regard to cultural variations. The study focused on
adolescents and adults. The authors found that individuals with an oral cleft had a higher chance of experiencing lower psychosocial adjustment compared to unaffected peers regardless of culture. No studies could be found that analyzed the cultural influences that affect health-related quality of life for children with oral clefts.

Since Hawaii is unique with regard to the state being comprised of islands, it was not known whether or not health-related quality of life would be perceived as lower for those living in rural neighbor islands versus those on Oahu where the multidisciplinary cleft clinic is more readily accessible. Since results indicated that island of residence did not affect health-related quality of life scores, it is reasonable to assume that the distance was not perceived by parents to negatively affect their children’s health-related quality of life. This is most likely due to the diligent care of the Department of Health nurse, social worker, and oral cleft clinic nurse manager with regard to planning and arranging clinic schedules and flights to accommodate those families who live on neighbor islands.

No other literature could be found that compared health-related quality of life for children with an oral cleft with demographic variables such as parental age, parental education, or geographic residence. These caregivers factors and environmental factors may also affect health-related quality of life for children with oral clefts.

**Discussion for Specific Aim 4**

**Speech**

There were no significant differences found in this study related to the impact of speech impairment on health-related quality of life scores on the Physical Health, Psychosocial Health, or Total Scores for the parent proxy reports or the children’s reports. Advances in management during the past decades have led to better outcomes. It
is likely that children with an oral cleft will achieve normal or near normal speech
(Peterson-Falzone, Hardin-Jones, & Karmell, 2010) which may have explained the
findings of no significant differences in health-related quality of life scores for the study
related to speech. Even for those who had speech impairments, it is possible that with
ongoing speech therapy, if improvements were noted, the parent’s and children’s
perceptions were positive. Damiano and colleagues (2007) reported different results,
whereby according to the mother’s perspective, the child’s speech was highly correlated
with Physical, Psychosocial, and Total health-related quality of life scores.

Speech was a factor with regard to teasing for children from the child’s perspective in
this study. Other studies have also reported that children with an oral cleft experience
teasing which was associated with greater psychosocial problems (Hunt, et al., 2006;
Hunt et al., 2007).

**Hearing**

Severity of hearing loss did not significantly affect Physical, Psychosocial, or Total
health-related quality of life scores from the parent’s or child’s perspective. The only
item that was significantly affected by hearing loss was “missing school/daycare to go the
doctor or hospital” according to the parents. This finding may be due to the increased
incidence of otitis media (OM) and middle ear effusion in children with cleft palate,
although the incidence of OM decreases with age. In addition, children with cleft palate
require routine monitoring for hearing at every visit with their cleft team (Sabo & Probst,
2009) which may also necessitate missing school, although necessary since longstanding
OM may result in hearing and speech impairment (Hubbard, Paradise, McWilliams,
Elster, & Taylor, 1985).
For children who wore hearing aids, parents reported their child’s health-related quality of life scores lower in each domain with a significant effect on school functioning. “Forgetting things” was also significantly affected by wearing a hearing aid according to the parents. The children’s ratings were the opposite of their parents, whereby those who wore a hearing aid actually had higher health-related quality of life scores compared to those who did not wear a hearing aid. This finding may be related to specialized care provided in schools for children who have special needs. Children who wore a hearing aid did have lower mean scores than those who did not in the School Functioning domain, although not significant. Hearing aids provide amplification for children with cleft palate who have permanent sensorineural or conductive hearing loss. Children with hearing aids need frequent visits to monitor hearing to ensure adequate amplification (Sabo & Probst, 2009) which may also contribute to missing school and lower school functioning, although vigilant monitoring of hearing is essential to maximize speech and language outcomes (Sabo & Probst, 2009).

Limitations

This study had several limitations which must be considered in the context of the findings. First, The PedsQL which was utilized to measure health-related quality of life is a generic instrument. It would have been preferable to measure health-related quality of life with a generic and condition specific instrument if one were available (Patrick & Deyo, 1989; Sprangers, Cull, Bjordal, Groenvold, & Aaronson, 1993; Varni, Seid, & Rode 1999). Condition specific measurements provide enhanced measurement sensitivity in areas of health that are particular to the condition of interest, therefore enabling the researcher or practitioner to gain a more comprehensive evaluation of the patients.
health-related quality of life (Varni & Limbers, 2009). The PedsQL generic instrument does not measure physical functioning in areas that are problematic for children with oral clefts such as dental, speech, and hearing items.

Secondly, this study had a small convenience sample. Although the incidence of cleft lip and palate is high in the state of Hawaii, the oral cleft clinic has a relatively small number of children who attend the clinic in comparison to some other major multidisciplinary clinics. After approximately one year of data collection, almost all children who were eligible to participate in the study were recruited. Although the sample was representative of those attending clinic (approximately 90 to 95% of those who were asked to participate in the study agreed to do so), having a small convenience sample limits the ability to generalize the study findings to other populations of children with cleft lip and/or palate.

This study combined children with cleft lip only, children with cleft lip and cleft palate, and those with cleft palate only. There may be subtle or significant differences in health-related quality of life among children with these three conditions, and this study did not address this issue. A larger sample would have provided the opportunity to compare findings across children with these three conditions.

Another limitation was that due to the small sample size, some variables were regrouped to increase the number. For example, for speech articulation, mild and moderate speech impairment was grouped together. Hearing loss was regrouped to include mild and moderate hearing loss and severe and profound hearing loss. Results may have shown different outcomes if the sample was larger to evaluate the items without regrouping. Also, overall articulation impairment was determined by subjective
and objective measures. It is possible that two different speech therapists could have had different impressions of the child’s speech.

**Contributions to the Nursing Literature**

Preliminary work for this study that focused on concept development of health-related quality of life for children with cleft lip and/or palate adds to nursing science by providing a clear understanding of the concept. The analysis resulted in a clear, succinct definition of health-related quality of life that can accurately reflect its use in the literature and be utilized for clinical research, theory construction, and instrument development for children with an oral cleft.

This study contributes to nursing overall, by describing health-related quality of life for children with cleft lip and palate, and by providing insight as to the parent’s perspective and the child’s perspective of how the oral cleft impacts the child’s physical and psychosocial functioning. One of the overarching goals of Healthy People 2020 is to attain high quality of life; therefore, nurses and health care professional must be cognizant of “what that means” to parents of children who have an oral cleft as well as the children themselves. By measuring health-related quality of life, nurses and health care professionals can gain information from the parents and children that are unique to the individual in order to provide guidance and interventions that can ultimately improve health outcomes.

The results of this study identified how parents of children with an oral cleft and the children themselves rated their quality of life compared to healthy children and children with a chronic illness which sheds light on overall perceptions of satisfaction with health and well-being for this population of children. Since parental perceptions and the
children’s perceptions were found to be significantly lower than healthy children with regard to physical, psychosocial, and total functioning, this study confirms that nurses and health care professional need to focus on and provide interventions to assist parents and children in these areas. While the multidisciplinary clinics are geared toward providing optimal medical and dental care, families would benefit by having psychosocial and school needs addressed to improve health-related quality of life for children with oral clefts.

This study has also revealed that the children should be given every opportunity to rate their own health-related quality of life since parent-child agreement was low for parents of children who have an oral cleft and the children themselves. This information is clinically important to enable nurses and other health-care professionals to seek information and allow disclosure of parents and children to express their concerns and needs in order to gain assistance where needed to improve the child’s overall health-related quality of life.

Results of this study also contribute to nursing science by providing preliminary data that can be utilized to establish normative condition-specific means of health-related quality of life scores that can be utilized to determine at risk-cut off scores to identify impaired health-related quality of life for children with an oral cleft. Although more research is needed in this area, this study has provided a basis to expand on in addition to findings that were reported by Damiano and colleagues (2007). By measuring health-related quality of life with the PedsQL in the multidisciplinary cleft clinic, parents and children are given the opportunity to identify, quantify, and disclose concerns. This information is of utmost importance in that it can identify at-risk status for impaired
health-related quality of life, enabling the health-care provider to intervene in the appropriate area/domain identified or result in referring the child for psychological services if warranted.

**Future Research**

This study provided information which serves as a foundation for understanding health-related quality of life for children with cleft lip and/or palate in the United States with a culturally diverse sample. Further research is needed in this area to include a larger sample size.

Conducting focus groups with parents and children to further understand the impact that the oral cleft has on the child’s physical and psychosocial functioning would add richness to data obtained from quantitative research. Qualitative studies and focus groups that center on the concept of health-related quality of life, particularly related to the day-to-day life experiences of the child with cleft lip and/or palate would increase awareness of the impact that the oral cleft has on a child’s life with regard to challenges, treatment, and positive influences gained from the experience. Focus groups conducted with parents related to their child’s health-related quality of life as well as focus groups conducted with school-aged children would likely yield excellent data to better understand the meaning of the influence of an oral cleft on health-related quality of life.

Research with specific cultural/racial groups would also be beneficial as a means of understanding how cultural beliefs affect health-related quality of life for children with cleft lip and/or palate. For example, a traditional view of the concept of health for Native Hawaiians is related to maintaining health through unity, balance, and harmony (lokahi), which in turn provides a connection with God, people, and the land (Palakiko, 2005).
Since no specific instrument exists to measure health-related quality of life for children with an oral cleft, research geared toward instrument development would be beneficial. Qualitative research could be valuable in developing a condition-specific instrument to measure health-related quality of life for children with cleft lip and/or palate.

As described by Hutchinson and colleagues (2010), even though numerous studies have described the psychosocial effects of cleft lip and/or palate, some craniofacial teams do not address the importance of psychological intervention. This research can serve as a baseline for establishing normative population data with regard to the dimensions of Physical Health, Psychosocial Health, and Total scores on the PedsQL for children with cleft lip and/or palate. More research is needed in this area to establish normative data with this population in order to identify at-risk status for impaired health-related quality of life. Scores that are approximately one standard deviation below the population mean have been proposed by researchers to use as a reference for cut-off point scores for at-risk status (Varni, Burwinkle, & Lane, 2005; Matza, et al., 2004).

Conclusion

A child’s health-related quality of life is a multidimensional, subjective construct that is perceived in terms of the impact that a condition has on the child’s physical, emotional, social, and cognitive functioning (Varni, 1998). The health-related quality of life for a child with cleft lip and/or palate can be defined as “the child’s individual perception of the impact that the oral cleft has on one’s physical well-being, psychosocial well-being, and satisfaction with life in relation to their cognitive and developmental level which is influenced by caregiver and environmental factors.” Although the child’s own perception
is of utmost importance, the parental or caregiver’s perception of the child’s quality of life or health-related quality of life is also paramount.

This study was conducted to increase an understanding of health-related quality of life for children with cleft lip and palate from the parent’s and child’s perspective within the conceptual framework of the Child Health Assessment Model (Barnard & Eyres, 1979) with an emphasis on the child’s physical, emotional, social, and cognitive functioning (Varni, 1998). The data yielded information which can be utilized in clinical practice and research.

Overall, with the exception of parents of children with an oral cleft rating their children’s Physical Functioning higher than parents of healthy children, parents of children with an oral cleft and the children themselves rated their Physical Functioning, Emotional Functioning, Social Functioning, School Functioning, overall Psychosocial Functioning, and Total health-related quality of life lower than healthy children and higher than children with a chronic condition. In other words, children with cleft lip and/or palate fall between healthy children and children with a chronic condition in terms of health-related quality of life. Despite the middle-range classification, attention must be provided to those who have Total Scores that fall one standard deviation below the population mean. It is imperative to intervene with these families to provide physical, psychological, and school support to improve health-related quality of life.

Parent-proxy reports and child self reports were similar in terms of means on most of the health-related quality of life scales although parent-child concordance was quite low in all domains with the highest correlation being only .20 on the Emotional Functioning scale. All other domains had an even smaller effect size. When parent proxy and child
self report scales are to be utilized in clinical practice, the health care provider must take into account the purpose of measuring health-related quality of life. If the purpose is to obtain results from the parent’s perspective, then parent-child agreement is not important; however, if the purpose is to take the place of the child’s own report, in other words to “speak for the child,” then one would want parent and child report agreement to be parallel (Upton, Lawford, & Eiser, 2008). Based on the findings of this study where the parent-child concordance was poor, the results suggest that for children with cleft lip and/or palate, the children should be given every opportunity possible to rate their own health-related quality of life.

While parents of children with an oral cleft and children themselves rate their health-related quality of life to be middle-range compared to healthy peers and children with a chronic condition, the goal of health care is always to strive to improve quality of life as much as possible. By understanding each family’s perception of quality of life, caregivers can aim to individualize care with regard to specific concerns identified on the health-related quality of life measure in addition to following overarching parameters for evaluation and treatment of patients with cleft lip and/or palate as established by the American Cleft Palate-Craniofacial Association. “A major hypothesized benefit to incorporating health-related quality of life measurement into routine clinical practice is the potential for identifying symptoms and problems that may result in improved patient outcomes over time,” (Varni, Burwinkle, & Lane, 2005, p. 2). Once individual concerns have been identified, health care providers then can implement care and treatment to maximize the child’s satisfaction with health and well-being.
APPENDICIES

APPENDIX A: PedsQL – PARENT REPORT FOR TODDLERS (ages 2-4)

C:\Documents and Settings\Sanc

APPENDIX B: PedsQL – YOUNG CHILD REPORT (ages 5-7)

C:\Documents and Settings\Sanc

APPENDIX C: PedsQL – PARENT REPORT FOR YOUNG CHILDREN (ages 5-7)

C:\Documents and Settings\Sanc

APPENDIX D: PedsQL – CHILD REPORT (ages 8-12)

C:\Documents and Settings\Sanc

APPENDIX E: PedsQL – PARENT REPORT FOR CHILDREN (ages 8-12)

C:\Documents and Settings\Sanc

APPENDIX F: DEMOGRAPHIC FORM

C:\Documents and Settings\Sanc

APPENDIX G: QUESTIONS FROM CRANIOFACIAL QUESTIONNAIRE

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APPENDIX H: SYNDROME DATA FROM REPOSITORY

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APPENDIX I: SURGICAL DATA FROM REPOSITORY

C:\Documents and Settings\Sanx

APPENDIX J: SPEECH DATA FROM REPOSITORY

C:\Documents and Settings\Sanx

APPENDIX K: HEARING DATA FROM REPOSITORY

C:\Documents and Settings\Sanx

APPENDIX L: INFORMED CONSENT FORM

C:\Documents and Settings\Sanx

APPENDIX M: ASSENT FORM (7 TO 10 YEARS OLD)

C:\Documents and Settings\Sanx

APPENDIX N: ASSENT FORM (11 TO 17 YEARS OLD)

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APPENDICES A - E

The following instruments are not included due to copyright laws:

Appendix A: PedsQL – Parent Report for Toddlers (ages 2-4)

Appendix B: PedsQL – Young Child Report (ages 5-7)

Appendix C: PedsQL – Parent Report for Young Children (ages 5-7)

Appendix D: PedsQL – Children Report (ages 8-12)

Appendix E: PedsQL – Parent Report for Children (ages 8-12)
APPENDIX F: DEMOGRAPHIC FORM
The Impact of a Multidisciplinary Approach to Growth, Development, Hearing and Speech, and General Quality of Life in Children born with Cleft Lip and/or Palate in the State of Hawaii

Principal Investigator Charles Neal, M D

PLEASE PROVIDE THE FOLLOWING INFORMATION (AS COMPLETE AS POSSIBLE)

Person Completing This Form ____________________________ Your relationship to child ________________

Child’s Name ________________________________________ Sex □ male □ female

Mailing Address ________________________________________ Birth date ________________________________

Street _____________ Apt # ______________________________

City _____________ State _____________ Zip Code ________________ Home Phone # ____________________________

What island do you live on? □ Oahu □ Big Island □ Maui □ Kauai □ Molokai □ Lanai

Parent’s Name ________________________________________ Other Phone # ____________________________

Child’s School ________________________________________ Child’s Grade in School □ □

What is the child participant’s ethnicity? (check one box)

□ Hispanic or Latino □ Not Hispanic or Latino

What is the child participant’s race? (check all that apply)

□ African American
□ Alaskan Native
□ American Indian
□ Asian (please specify)
    □ Chinese
    □ East Indian
    □ Filipino
    □ Other Asian __________________________
□ Caucasian
□ Native Hawaiian
□ Pacific Islander (please specify)
    □ Fijian
    □ Marshallese
    □ Other ethnicity/race __________________________

□ Hawaiian
□ Micronesian
□ Tongan
□ Tahitian
□ Samoan
□ Vietnamese
□ Thai
□ Korean
□ Japanese
□ Malaysian
□ Laotian
□ East Indian
□ Filipino
□ Other Asian __________________________

Which ethnicity does the child most identify with? (please choose only one) __________________________
APPENDIX G: QUESTIONS FROM CRANIOFACIAL QUESTIONNAIRE
Selected Questions from Craniofacial Questionnaire

What is the parents' combined annual (for 1 year) income before taxes?
$0 - $25,000
$25,001 - $50,000
Over $50,000

What is the FATHER’S highest level of education?
Did not complete high school
Completed high school
Completed vocational training
Completed 2 year degree
Completed 4 year degree
More than a 4 year degree

What is the MOTHER’S highest level of education?
Did not complete high school
Completed high school
Completed vocational training
Completed 2 year degree
Completed 4 year degree
More than a 4 year degree

Age of MOTHER when child was born
Age of FATHER when child was born

Which of the following best describes the parent's marital status?
Married
Living with a significant other
Separated
Divorced
Never married
APPENDIX H: SYNDROME DATA FROM REPOSITORY
Syndrome Data from Repository

<table>
<thead>
<tr>
<th>Data Field</th>
<th>Possible Responses</th>
</tr>
</thead>
<tbody>
<tr>
<td>Does patient have a syndrome</td>
<td>yes, no</td>
</tr>
<tr>
<td>Syndrome name</td>
<td>name of type of syndrome</td>
</tr>
</tbody>
</table>
APPENDIX I: SURGICAL DATA FROM REPOSITORY
<table>
<thead>
<tr>
<th><strong>Data Field</strong></th>
<th><strong>Possible Responses</strong></th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft lip</td>
<td>yes, no</td>
</tr>
<tr>
<td>Cleft lip left</td>
<td>complete, incomplete, none</td>
</tr>
<tr>
<td>Cleft lip right</td>
<td>complete, incomplete, none</td>
</tr>
<tr>
<td>Cleft palate</td>
<td>yes, no</td>
</tr>
<tr>
<td>CP type</td>
<td>complete, incomplete, none</td>
</tr>
<tr>
<td>Alveolar cleft</td>
<td>yes, no</td>
</tr>
</tbody>
</table>
APPENDIX J: SPEECH DATA FROM REPOSITORY
Speech Data from Repository

<table>
<thead>
<tr>
<th>Data Field</th>
<th>Possible Responses</th>
</tr>
</thead>
<tbody>
<tr>
<td>Articulation Assessment</td>
<td></td>
</tr>
<tr>
<td>Standard Score on GFTA-2</td>
<td>numerical value</td>
</tr>
<tr>
<td>Number of errors on GFTA-2</td>
<td>numerical value</td>
</tr>
<tr>
<td>Voice/Vocal Resonance Assessment</td>
<td></td>
</tr>
<tr>
<td>Vocal resonance</td>
<td>normal, slight hypernasality, hypernasality, hyponasality, mixed, cul-de-sac</td>
</tr>
<tr>
<td>Nasal emission of air</td>
<td>yes, no</td>
</tr>
<tr>
<td>Velopharyngeal insufficiency indicated</td>
<td>yes, no, suspected, undertermined</td>
</tr>
<tr>
<td>Articulation Impression</td>
<td>normal, mild impairment, moderate impairment, severe impairment</td>
</tr>
</tbody>
</table>

GFTA-2 is the Goldman Fristoe 2 Test of Articulation

Mixed vocal resonance refers to hypernasality and hyponasality

Articulation data was obtained from the repository on the date closest to when the parent and/or child completed the PedsQL.
APPENDIX K: HEARING DATA FROM REPOSITORY
Hearing Data from Repository

<table>
<thead>
<tr>
<th>Data Field</th>
<th>Possible Responses</th>
</tr>
</thead>
<tbody>
<tr>
<td>Severity of hearing loss left</td>
<td>normal, mild, moderate, moderate to severe, severe, profound</td>
</tr>
<tr>
<td>Severity of hearing loss right</td>
<td>normal, mild, moderate, moderate to severe, severe, profound</td>
</tr>
<tr>
<td>Hearing aid left</td>
<td>yes, no</td>
</tr>
<tr>
<td>Hearing aid right</td>
<td>yes, no</td>
</tr>
</tbody>
</table>

Type of hearing loss was categorized according to the guidelines utilized at KMCWC as follows:

- Normal: 0 – 20 dB
- Mild: 21-40 dB
- Moderate: 41-55 dB
- Moderately severe: 56-70 dB
- Severe: 71-90 dB
- Profound: 91-100+ dB

Hearing data was obtained from the repository on the date closest to when the parent and/or child completed the PedsQL.
RESEARCH SUBJECT INFORMATION AND CONSENT FORM

TITLE: The Impact of a Multidisciplinary Approach on Growth, Development, Hearing and Speech, and General Quality of Life in Children Born with Cleft Lip and/or Palate in the State of Hawaii

PROTOCOL NO.: 2008-01-Neal-OCC
WIRB® Protocol #20090567

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United States

STUDY-RELATED PHONE NUMBERS: Charles R. Neal Jr., M.D., Ph.D.
808-983-6000 (hospital operator – pager)

Sandra Sinclair, R.N., M.S.N.
808-983-6247

This consent form may contain words that you do not understand. Please ask the study doctor or the study staff to explain any words or information that you do not clearly understand. You may take home an unsigned copy of this consent form to think about or discuss with family or friends before making your decision.

INTRODUCTION

You and your child are being asked to be in this research study because your child has a cleft lip or a cleft palate. This is a study for children from birth to age 18 who are born with cleft lip and/or palate and their parent. Your child will be one of about 160 subjects in this study. As a parent/legal guardian, you will also be a subject in this study.
Before you decide if you and/or your child will be in this study, read this consent form to find out about the purpose, how it may help, any risks involved, and your and your child’s rights as research subjects. This process is called informed consent. The consent form will be discussed with you. If you decide that your and your child will be in the study, you will be asked to sign this consent form. A signed copy will be given to you to keep for your reference.

Before you learn about the study, it is important that you know the following:

- Taking part in this study is of your own free will
- You and your child may decide not to take part in the study or stop being in the study at any time. If you decide that you don’t want to be part of this study, you and your child will still receive medical care and any benefits that you are entitled to

PURPOSE

The three purposes and parts to this study are:

1. The information collected in this study is to be used for future research in a research database (an information bank called a repository) on children with cleft lip and/or palate. The information collected will be used to look at the physical and psychological health of children who were born with cleft lip and/or palate. The study doctor wants to learn more about:
   - The effects of having a craniofacial clinic where a child can receive care from several doctors and specialists on one day compared to having the parent organize several separate appointments,
   - The growth and feeding patterns of children during the first 24 months of life,
   - The hearing and language development of children.

2. The quality of life assessments and questionnaires to learn more about:
   - the child’s health-related quality of life (well-being), self-concept (how the child feels about him or her self),
   - how the parent and child interact,
   - the mother’s awareness and beliefs about her child, and
   - the child’s temperament (manner of behaving and reacting to situations).

   Some information from these assessments will also be banked in the repository for future research.

3. The focus groups will provide more information about the parent’s experiences related to caring for a child with cleft lip/palate. This information will be stored in the repository. You or your child may chose not to answer any of the questions on the questionnaires or during the focus group.
DURATION

If you consent to be in the study and to permit your child to be in the study, you and your child may be enrolled in the study for as long as your child receives care at the Oral Cleft Clinic. The number of clinic visits you attend each year will depend on the age of your child and the recommendations of the specialists at the clinic. The study will possibly continue from the time your child starts being seen at the Oral Cleft Clinic until your child is 18 years.

PROCEDURES

Storage of information in the research database
The information obtained from you and your child during the Oral Cleft Clinic visits will be kept in the research database for future research. The database will include any information collected during visits such as medical evaluations, physical exams, assessments from specialists, results from tests, and some information collected from the research questionnaires and scales (described below).

Research Questionnaires and Assessments
The study doctor would like to learn more about children and families who come to the clinic. Depending on your child’s age, you and your child will be asked to fill out the questionnaires and be part of a one-time focus group. See the Study Procedures Table described below. Your child can still be in the repository component of the study even if you or your child decide not to participate in some or all of the research questionnaires, assessments or the focus group.

The table of study procedures below includes specific information about the questionnaires, scales, and focus group. Most the research procedures will be completed during your scheduled visits to the Oral Cleft Clinic. If you/your child have another scheduled visit at KMCWC, the study visit could take place on that day in the Clinical Research Center at KMCWC if you prefer. Some of the assessments may be done during a home visit since it would be a more natural environment for you and your child. Please see Appendix I at the end of this consent form for more information about the questionnaires, scales, and focus groups.

<table>
<thead>
<tr>
<th>Age of child</th>
<th>Assessment Tool</th>
<th>Child</th>
<th>Parent</th>
<th>Time Required</th>
<th>Description</th>
<th>How Often</th>
</tr>
</thead>
<tbody>
<tr>
<td>First study visit</td>
<td>Demographic form</td>
<td>X</td>
<td></td>
<td>3-5 minutes</td>
<td>Questionnaire</td>
<td>Once</td>
</tr>
<tr>
<td>First study visit</td>
<td>Craniofacial questionnaire</td>
<td>X</td>
<td></td>
<td>5-10 minutes</td>
<td>Questionnaire</td>
<td>Once</td>
</tr>
<tr>
<td>Birth and 1-4 months</td>
<td>NPI (Neonatal Perception Inventories)</td>
<td>X</td>
<td></td>
<td>5 minutes</td>
<td>Questionnaire</td>
<td>Twice</td>
</tr>
<tr>
<td>Birth and 1-4 months</td>
<td>Your Infant questionnaire</td>
<td>X</td>
<td></td>
<td>5 minutes</td>
<td>Questionnaire</td>
<td>Twice</td>
</tr>
<tr>
<td>1-4 months and 6-9 months</td>
<td>MAI (Maternal Attachment Inventory)</td>
<td>X</td>
<td></td>
<td>5-10 minutes</td>
<td>Questionnaire</td>
<td>Twice</td>
</tr>
<tr>
<td>Age Range</td>
<td>Assessment/Scale Description</td>
<td>Administration Time</td>
<td>Description</td>
<td>Frequency</td>
<td></td>
<td></td>
</tr>
<tr>
<td>-----------------</td>
<td>------------------------------------------------------------------------------------------------</td>
<td>---------------------</td>
<td>---------------------------------------------------------------------------------------------------</td>
<td>-----------</td>
<td></td>
<td></td>
</tr>
<tr>
<td>1-4 months and 6-9 months</td>
<td>NCAFS (Nursing Child Assessment Feeding Scale)</td>
<td>X 15 to 60 minutes</td>
<td>Observation of parent and child during feeding time Twice the first year of life, once from age 1-3, 3-7, and 8-12 years</td>
<td>Twice</td>
<td></td>
<td></td>
</tr>
<tr>
<td>1 month – 12 years</td>
<td>CTS (Carey Temperament Scale)</td>
<td>X 20 minutes</td>
<td>Questionnaire</td>
<td>Yearly</td>
<td></td>
<td></td>
</tr>
<tr>
<td>2 years – 18 years</td>
<td>PedsQL (Pediatric Quality of Life Questionnaire)</td>
<td>X &gt; or = 5 yrs X &gt; or = 2 yrs 5 minutes</td>
<td>Questionnaire</td>
<td>Yearly</td>
<td></td>
<td></td>
</tr>
<tr>
<td>2-3 yrs</td>
<td>NCATS (Nursing Child Assessment Teaching Scale)</td>
<td>X X 5-10 minutes</td>
<td>Observation of parent and child during teaching situation</td>
<td>Once or Twice</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3 yrs – 6 yrs</td>
<td>Joseph Picture Self-Concept Scale</td>
<td>X 5-10 minutes</td>
<td>Picture book and talking</td>
<td>Yearly</td>
<td></td>
<td></td>
</tr>
<tr>
<td>7 yrs – 18 yrs</td>
<td>Piers-Harris Children’s Self-Concept Scale</td>
<td>X 10-15 minutes</td>
<td>Questionnaire</td>
<td>Yearly</td>
<td></td>
<td></td>
</tr>
<tr>
<td>11 yrs – 18 yrs</td>
<td>YQOL-FD (Youth Quality of Life – Facial Differences)</td>
<td>X X 15 minutes</td>
<td>Questionnaire</td>
<td>Yearly</td>
<td></td>
<td></td>
</tr>
<tr>
<td>All ages</td>
<td>Focus Group</td>
<td>X 1 hour</td>
<td>Audio taped focus group for parents to express experiences</td>
<td>Once</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**KNOWN RISKS OR DISCOMFORTS**

The study may involve the following risks and/or discomforts. There also may be risks and discomforts that are not yet known.

Some of the questions are personal or sensitive. You or your child may feel uncomfortable when answering and may chose not to answer any of the questions on the questionnaires or during the focus group. You or your child may become anxious or bored during the testing. If this happens, you or your child will be given a break, given the opportunity to discuss the problem, or moved onto another activity. You may have a loss of privacy if you are breastfeeding and being observed by the nurse for the feeding assessment. The nurse will try to decrease this loss of privacy by having the feeding time in a private area such as an exam room if it is being done at KMCWC instead of during a home visit.
BENEFITS

You and your child may benefit by the results the questionnaires which the research nurse may discuss with you. Also, if you participate in the assessments for breast feeding or the child teaching assessments (NCATS), the study nurse will provide you with tips that may help you and your child interact. If you participate in the assessment about your child’s temperament (CTS), the study nurse will provide you with information about your child’s behavioral style. There may be no direct benefits to you or your child for participating in this study. Your and your child’s participation may help us learn more about the physical and psychosocial health of children who are born with cleft lip and/or palate. This may help health care professionals determine better ways to assist these children and their families in the future.

COSTS

There will be no charge for the research tests done for this study.

PAYMENT FOR PARTICIPATION

For participating in the first study visit, children 5 years and under will receive a teddy bear. If your child has a cleft lip, the teddy bear will have “stitches” sewn on the bear to represent a cleft lip repair. If your child has a cleft palate, the bear will not have “stitches.” For participating in the first study visit, if your child is older than 5 years of age, you may choose a teddy bear or a $10 gift card to a book store for your child. Parents who participate in the focus group will be paid $20.

ALTERNATIVES

You and your child may choose not to participate in this study and may obtain care for your child at the Oral Cleft Clinic in the usual manner.

NEW FINDINGS

You will be told about any new information that might change your decision for you and your child to be in this study. You may be asked to sign a revised consent form if this occurs.

CONFIDENTIALITY

Every effort will be made to maintain the confidentiality of your and your child’s study records. All information about you and your child will remain strictly confidential to the extent required by law. Results from the questionnaires may be shared with the members of the Oral Cleft Clinic multidisciplinary team in order to best manage and care for your child. Results from this study may be published or shared with other interested parties such as other physicians, research institutions, and/or federal authorities, but you and your child will not be personally identified. In the process of evaluating the study, your and your child’s records may be examined by and the Western Institutional Review Board® (WIRB®). Records from this study will be kept in a secured area. You and your child will be assigned a study number which will be used on all study documents.
USE AND DISCLOSURE OF YOUR HEALTH INFORMATION

By signing this consent form you are authorizing the use and disclosure of individually identifiable information. Your and your child’s information will only be used/disclosed as described in this consent form and as permitted by state and federal laws.

This consent covers all information about you and your child that is used or collected for this study. It includes identifiable information and health information, including existing or new information that will be obtained for this study. For example your and your child’s name, birth date, identification numbers, codes, and information gathered about you and your child during the study visits. The study-related information includes information that is described in the Procedures section of this consent form.

Your authorization to use your and your child’s identifiable health information will not expire even if you terminate your and your child’s participation in this study or you and your child are removed from this study by the study doctor. However, you may withdraw your authorization to use your and your child’s identifiable information at anytime by writing to the study doctor named at the top of this consent form. If you decide to withdraw your authorization, your and your child’s identifiable health information collected or created for this study shall not be used or disclosed by the study doctor after the date of receipt of the written notice except to the extent that the law allows us to continue using your and your child’s information. The study doctor is not required to destroy or retrieve any of your and your child’s health information that was created, used or disclosed for this study before receiving your written notice. Audiotapes from the focus group will be destroyed 3 years following the completion of the study.

By signing this consent form you authorize the following parties to use and/or disclose your and your child’s identifiable health information collected or created for this study:

- Charles R. Neal Jr., M.D., Ph.D.; Lynn Iwamoto, M.D., Sandra Sinclair, R.N., M.S.N., Mari Uehara, M.D., and their research staff for the purposes of conducting this research study
- University of Hawaii Clinical Research Center personnel
- Medical records department at Kapiolani Medical Center for Women and Children

The individuals named above may disclose your and your child’s medical records, this consent form, and the information about you and your child created by this study to:

- Members of the Oral Cleft Clinic multidisciplinary team for the purpose of medical management and evaluation of care
- Other research investigators who are conducting studies related to cleft lip and/or palate who have obtained approval to obtain data from the repository
- Federal, state and local agencies having oversight over this research, such as the Office for Human Research Protections in the U.S. Department of Health and Human Services, and the National Institutes of Health
- University of Hawaii Clinical Research Center Data and Safety Monitoring Board
- The Hawaii Pacific Health (HPH) Officials, and the HPH Research Institute
• The Western Institutional Review Board® (WIRB®) for purposes of overseeing the research study and making sure that your ethical rights are being protected
• University of Hawaii dissertation committee members for purposes of overseeing the doctoral dissertation component of the study

Your and your child’s medical records may contain information about AIDS or HIV infection, venereal disease, treatment for alcohol and/or drug abuse, or mental health or psychiatric services. By signing this consent form, you authorize access to and use of this information if it is in the records used by members of the research team.

Some of the persons or groups that receive your study information may not be required to comply with federal privacy regulations, and your information may lose its federal privacy protection if those persons or groups disclose it. Information that could identify you or your child will be removed before it is used by another investigator so that your and your child’s identity will not be disclosed.

COMPENSATION FOR INJURY

There will be minimal risk involved in participating in this research study. There are no plans to compensate you or your child or provide medical treatment in the event that you or your child are injured as a result of participating in this study.

SOURCE OF FUNDING

Funding for this research study will be provided by Cleft Palate Foundation.

VOLUNTARY PARTICIPATION OR WITHDRAWAL

Your and your child’s participation in this study is voluntary. You and your child may decide not to participate, or you may leave the study at any time. Your decision will not result in any penalty or loss of benefits to which you and your child are entitled. If you and your child withdraw from the study, we will continue to use information that has already been collected but no new information about you and your child will be collected unless the information concerns an adverse event (a bad effect) related to the study. If such an adverse event occurs, your and your child’s entire medical records may need to be reviewed.

The study doctor or the sponsor has the right to withdraw you and your child from the study at any time without your consent for any of the following reasons:

• for reasons related solely to you (for example, not following study-related directions from the study doctor; or a serious adverse reaction);
• because the entire study has been terminated;
• you do not later consent to any future changes that may be made in the study plan;
• or for any other reason.
You have the right to refuse to sign this consent form. This would prevent those listed above from having access to or using any of your and your child’s health information for the study. Refusal to sign this consent form means that you and your child cannot be in this study.

QUESTIONS

If you have any questions, concerns or complaints about this study, or if at any time you feel you or your child has had a research-related injury you may call Dr. Charles R. Neal Jr. at 808-983-6000 (hospital operator – pager), or Sandra Sinclair, the nurse coordinator at 808-983-6247.

If you have any questions about your rights as a research subject, or if you have questions, concerns or complaints about the research, you may call:

Western Institutional Review Board® (WIRB®)
3535 Seventh Avenue, SW
Olympia, Washington 98502
Telephone: 1-800-562-4789 or 360-252-2500
E-mail: Help@wirb.com.

WIRB is a group of people who perform independent review of research.

WIRB will not be able to answer some study-specific questions, such as questions about appointment times. However, you may contact WIRB if the research staff cannot be reached or if you wish to talk to someone other than the research staff.

Do not sign this consent form unless you have had a chance to ask questions and have received satisfactory answers to all of your questions.

If you agree to be in this study, a signed copy of this consent form will be given to you.

VOLUNTARY CONSENT:

By signing this consent form, you are agreeing to storage of information collected as part of standard care at the Oral Cleft Clinic to be maintained in the research database and used to study children with oral clefts and their families.

Your child can still be in the study even if you decide not to participate.

I give consent for my child to participate in research questionnaires and assessments

☐ I agree   ☐ I do not agree   Parent’s initials ___________ Date ___________

I consent to participate in research questionnaires and assessments.

☐ I agree   ☐ I do not agree   Parent’s initials ___________ Date ___________
I consent to participate in the focus group

☐ I agree ☐ I do not agree Parent’s initials ___________ Date ___________

I have read the information provided above (or it has been read to me). All my questions about the study and my participation in it have been answered. I voluntarily agree to participate and have my child participate in this study.

I authorize the use and disclosure of my and my child’s health information to the parties listed in the authorization section of this consent for the purposes described above.

By signing this consent form, I have not given up any of my or my child’s legal rights.

**Consent and Assent Instructions:**

**Consent:** For subjects under 18, consent is provided by the parent or guardian

**Assent:** Is not required for subjects 6 years and younger

*Verbal assent is required for subjects ages 7 through 10 years using the Assent section below and the Information Sheet for Children.*

*Verbal assent is required for subjects ages 11 through 17 years using the Assent section below and the Information Sheet for Adolescents.*

________________________________________
Child’s name (print)

________________________________________
Parent/legal guardian’s name (print)

Parent/legal guardian’s signature Date

________________________________________
If signed by someone other than the subject, describe your legal authority to act on behalf of the Research Participant

________________________________________
Signature and title of person conducting informed consent discussion and identification of role in the study Date
ASSENT SECTION:

Statement of person conducting assent discussion:

1. I have explained all aspects of the research to the subject to the best of his or her ability to understand.
2. I have answered all the questions of the subject relating to this research.
3. The subject agrees to be in the research.
4. I believe the subject's decision to enroll is voluntary.
5. The study doctor and study staff agree to respect the subject's physical or emotional dissent at any time during this research when that dissent pertains to anything being done solely for the purpose of this research.

Signature of Person Conducting Assent Discussion

Statement of Parent or Guardian:

My child appears to understand the research to the best of his or her ability and has agreed to participate.

Signature of Parent or Guardian

----------------------------------------------- Use the following only if applicable --------------------------------------------

If this consent form is read to the subject because the subject is unable to read the form, an impartial witness not affiliated with the research or investigator must be present for the consent and sign the following statement:

I confirm that the information in the consent form and any other written information was accurately explained to, and apparently understood by, the subject. The subject freely consented to be in the research study.

Signature of Impartial Witness

Note: This signature block cannot be used for translations into another language. A translated consent form is necessary for enrolling subjects who do not speak English.
Appendix 1: Description of Assessment Tools and Focus Group

Demographic Form
The demographic form includes basic information such as name, address, birth date, and race. The questions can be answered in about 3-5 minutes. We will ask you to complete this form at your first study visit.

Craniofacial Questionnaire
The craniofacial questionnaire includes information about the child and parent such as feeding practices, number of surgeries, and parental age at your child’s birth. The questions can be answered in about 5-10 minutes. We will ask you to complete this form at your first or second study visit.

Neonatal Perception Inventory (NPI)
The NPI is a questionnaire that looks at what the mother expects and feels about her infant’s behaviors. Each question is answered from considering the behavior of the average baby and the behavior of your own baby. The questions are answered on a scale ranging from “none” to “a great deal”. The questionnaire asks about your infant’s crying, spitting, feeding, bowel movements, sleeping, and behaviors. The questions can be answered in about 5 minutes. If possible, we will ask you to complete this questionnaire within a few days of your baby’s birth while you are still in the hospital and then again about 1 to 4 months after your baby’s birth.

Your Infant Questionnaire
The Your Infant questionnaire looks at how you interpret your baby’s behaviors and how you feel about your baby. The questions can be answered in about 5 minutes. If possible, we will ask you to complete this questionnaire within a few days of your baby’s birth while you are still in the hospital and then again about 1 to 4 months following your baby’s birth.

Maternal Attachment Inventories (MAI)
The MAI is a questionnaire that looks at the mother’s affection and bonding with her baby. The questions are answered on a scale ranging from “almost always” to “almost never.” The questions can be answered in about 5-10 minutes. We will ask you to complete the questionnaire when your baby is about 1-4 months of age and again when your baby is about 6-9 months of age.

Carey Temperament Scale (CTS)
The CTS is a questionnaire that looks at your infant’s or child’s temperament. Temperament is the way your child behaves or responds to situations such as activity, routine, mood, and sensitivity to sounds and events. The questions can be answered in about 20 minutes. We will ask you to complete the questionnaire when your child is: 1-4 months of age, 4-12 months of age, 1-3 years of age, 3-7 years of age, and 8-12 years of age.

The Nursing Child Assessment Feeding Scale (NCAFS)
For the NCAFS, the nurse observes the behaviors between the you and your child during a feeding situation and fills out a form. The feeding situation can be during breast-feeding, bottle feeding, or feeding the child with eating utensils. The observation would last for the length of time needed to feed your child (about 15 to 60 minutes). We will ask you to participate when your child is about 1-4 months old and again when your child is about 6-12 months old.
The Pediatric Quality of Life Questionnaire (PedsQL)
The PedsQL has 23 questions. It looks at four areas of your child’s life and is a way to measure your child’s well-being. The four sections are physical functioning, emotional functioning, social functioning, and school performance. The questions can be answered in about 5 minutes. When your child reaches 2 years of age, we will ask you to complete the questionnaire each year. When your child reaches age 5, he or she will also be asked to complete the questionnaire each year. If your child is unable to read or answer the questions, the research nurse will help your child.

The Nursing Child Assessment Teaching Scale (NCATS)
For the NCATS, the nurse observes the behavior between you and your child during a teaching situation and fills out a form. You will choose a teaching activity that would be suitable for your child’s age (for example, how to complete a jigsaw puzzle). The observation would occur as long as the parent wishes to teach the child (usually about 5-10 minutes). We will ask you to participate in the NCATS once or twice, when your child is about 2-3 years of age.

The Joseph’s Picture Self-Concept Scale
Your child will be asked to draw his or her face in a drawing that outlines a child’s head and shoulders. Next, your child will be shown pairs of pictures in everyday situations. Your child will be asked to choose between a picture that shows a positive event (for example, a picture that shows a child being praised) and another that shows a negative event (for example, a picture that shows a child being scolded). Your child will be asked to say or point to the situation that happens to him/her more often. The questions can be answered in about 5-10 minutes. When your child is 3 years old, we will ask your child to complete the scale on a yearly basis through 6 years of age.

The Piers-Harris Children’s Self-Concept Scale
The Piers-Harris Children’s Self-Concept Scale has 60 questions that can be answered as either “yes” or “no.” It asks about physical appearance, anxiety, school status, happiness and satisfaction, and popularity. The questions can be answered in about 10-15 minutes. When your child is 7 years old, we will ask your child to complete the questionnaire each year.

The Youth Quality of Life - Facial Differences (YQOL-FD)
The YQOL-FD has 48 questions. It was created for children with head and face conditions and asks about feelings such as shame or feeling different. The questions can be answered in about 15 minutes. When your child is 11 years old, we will ask you and your child to complete the questionnaire each year.

Focus Group
Parents may be in a focus group to discuss their experiences caring for a child who was born with cleft lip and/or palate. The focus group will be audio taped and notes will be taken during the session. The audiotapes will be transcribed without names and will be destroyed 3 years following completion of the study. The focus group will last about one hour.
APPENDIX M: ASSENT FORM (7 TO 10 YEARS OLD)
ASSENT INFORMATION SHEET
(Ages 7 to 10 years old)

TITLE: The Impact of a Multidisciplinary Approach on Growth, Development, Hearing and Speech, and General Quality of Life in Children born with Cleft Lip and/or Palate in the State of Hawaii

PROTOCOL NO.: 2008-01-Neal-OCC
WIRB® Protocol #20090567

STUDY-RELATED PHONE NUMBERS:
Charles R. Neal Jr., M.D., Ph.D.
808-983-6000 (hospital operator – pager)

Sandra Sinclair, R.N., M.S.N.
808-983-6247

You are being asked to be in a study because you have a cleft lip and/or palate. We want to learn more about how you feel about different things that happen as you grow up.

If you agree to be in this study, you will come to the clinic to see your doctors. You will answer some questions. Sometimes you will draw your face. You do not have to answer any questions or do any tests if you don’t want to. Just tell your Mom or Dad or the doctor.

This study will last until you are 18 years old. You can stop anytime you want to. But, you will still have to see your doctors.

Benefits

This study will help us learn more about kids with cleft lip and palate. We hope that what we learn can help other kids and their families in the future.

What you will get

You will receive a teddy bear or a $10 gift card to a book store for taking the time to be in the study. Some of our bears have sewn on “stitches” to look like a cleft lip repair.

Important things to know

It’s Your Choice - You don’t have to be in this study if you don’t want to.

We won’t be mad at you if you decide you don’t want to do this.

Your doctor will still take care of you even if you don’t want to do this.

If you have any question while you are in the study, please ask your parents or call Dr. Charles R. Neal Jr. at 808-983-6000, or Sandra Sinclair, his nurse, at any time. Her office number is 808-983-6247.
APPENDIX N: ASSENT FORM (11 TO 17 YEARS OLD)
ASSENT INFORMATION SHEET
(Ages 11 to 17 years old)

TITLE: The Impact of a Multidisciplinary Approach on Growth, Development, Hearing and Speech, and General Quality of Life in Children born with Cleft Lip and/or Palate in the State of Hawaii

PROTOCOL NO.: 2008-01-Neal-OCC
WIRB® Protocol #20090567

STUDY-RELATED PHONE NUMBERS:
Charles R. Neal Jr., M.D., Ph.D.
808-983-6000 (hospital operator – pager)

Sandra Sinclair, R.N., M.S.N.
808-983-6247

You are being asked to participate in a clinical research study. Your decision to be in this study is voluntary. You do not have to participate in this study if you do not want to.

This information sheet will give you information about the risks and benefits of this study so that you can make a better decision about whether you want to take part or not.

Study Purpose (Why the study is being done)
The purpose of this study is to learn more about children who were born with cleft lip and/or palate. You are being asked to be in this study because you have a cleft lip and/or palate. The study doctor will look at how the Oral Cleft Clinic helps children and their families. The study will also look at how you are doing physically in growth, hearing, and speech. The study will look at how you feel overall, and how you feel about your life in general.

Study Procedures (This is what will happen during the study visit)
We will keep information, in a research database, about your visits to the Oral Cleft Clinic and all of your study visits.

Once each year, you will be asked to answer questions on paper that will help us learn more about you. There are 3 different forms that take from 5 to 15 minutes each to complete. Some of the questions are about your life, your physical health, your feelings, how you get along with others, and how you are doing in school. Other questions are about how other people treat you, how you see yourself, and how you deal with certain situations.

You will be in this study until you are 18 years old. You may stop being in the study at any time.
Risks and Discomfort
Some of the questions may be personal. If you feel uncomfortable with any of the questions, you do not have to answer the questions. If you have any questions or concerns about the topics, please talk about it with your parents or the doctor or the study staff.

Benefits
Participating in this study may not help you, but your participation will help us to learn more about what children experience when they are born with cleft lip and/or palate. It may help us understand and better care for children with clefts and their families.

Compensation for Participation
You will receive a teddy bear or a $10 gift card to a book store for your time in the study. Some of our bears have sewn on “stitches” to look like a cleft lip repair.

It’s Your Choice
You don’t have to be in this study if you don’t want to. Also, you can decide to be in the study now, and change your mind later and stop being in it. Your doctor will still take care of you and nothing will change about your care.

For more information about this study, please see the consent form for this study that was discussed with you and your parents.

Questions
If you have any question while you are in the study, you may call Dr. Charles R. Neal Jr. at 808-983-6000 (hospital operator – pager), or Sandra Sinclair, the nurse who works with Dr. Neal, at any time. Her office number is 808-983-6247.
References


American Cleft Palate Association (ACPA) (2009). Parameters for evaluation and treatment of patients with cleft lip/palate or other craniofacial anomalies.


Cleft Palate Foundation (2008). The First Four Years.


Scaling and Scoring of the Pediatric Quality of Life Inventory. (2008). Mapi Research Trust.


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