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Experiences of Couples Caring for a Child Born with Cleft Lip and/or Palate:

Impact of the Timing of Diagnosis

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KEY WORDS: KEY WORDS: Cleft Lip; Cleft Palate, Couple Relationship, Qualitative

ABSTRACT

Raising a child born with cleft lip and/or palate (CL/P) can be challenging for parents. Few researchers have examined how having a child born with CL/P impacts couples. The purpose of this descriptive qualitative study was to examine how a child's CL/P diagnosis affects couples' adjustment and their relationship. We conducted interviews with 17 couples (10 prenatal and 7 postnatal) caring for children born with CL/P. After conducting thematic content analysis, six overarching themes emerged: (1) relationship growth, (2) challenges, (3) roles and responsibilities, (4) sources of support, (5) talking about cleft, and (6) lessons learned. Findings suggest couples should be routinely assessed for psychosocial issues and prevention programs should be tailored to pre-existing stressors and timing of the diagnosis.

KEY WORDS: Cleft Lip; Cleft Palate, Couple Relationship, Qualitative

Orofacial clefts including cleft lip (CL) with or without cleft palate (CP) are among the most common congenital anomalies in the United States (U.S.) (Parker et al., 2010). The prevalence of orofacial clefts varies by race, occurring most frequently among Asians and Native Americans, followed by Whites, Hispanics, and African Americans (Banales, 2009; Wilkins-Haug, 2012). CL describes when the entire upper lip is divided at birth which can continue up into the nose through the nostril and into the incisive foramen, which is the hard palate immediately behind the incisor teeth. Clefts are classified as unilateral or bilateral based on whether they are located on one or both sides of the cleft lip and/or palate (roof of the mouth).

Cleft lip palate (CL/P) involves a gap in both the lip and palate (Friedman, Wang & Milczuk, 2010). Cleft lip with or without palate (CL/P) impacts one in every 940 (10.63 per 10,000) live births; one in every 1574 (6.35 per 10,000) babies are born with cleft palate without cleft lip (American Cleft Palate Craniofacial Association, 2009; Parker et al., 2010). Although there is no definitive cause for the development of CL/P, chromosomes, genes, advanced maternal age, proteins, and the environment as well as spontaneous genetic mutation all play a role in the development of clefts in utero (Bille et al., 2005; Conrad, Richman, Nopoulos, & Dailey, 2009).

CL/P is associated with speech, hearing, feeding, and dental issues as well as irregularities of the bone and soft tissue which may require surgical reconstruction. Some babies born with CL/P will need ongoing surgical, dental, and speech treatment which can affect their quality of life (Author, 2012). Children are typically treated by multidisciplinary teams of specialists beginning at birth and continuing through early adulthood (American Cleft Palate Craniofacial Association, 2009). Surgeries to repair the lip and palate are first performed in early infancy and continue through adolescence and young adulthood to correct both the form and function of the head and face. Additionally, some school-age children will

experience learning disabilities, poor self-concept, social anxiety, social stigma from peers, and consequently may need additional psychosocial support (Berger & Dalton, 2011; Kramer et al., 2009; Pope & Snyder, 2005).

Over the past several decades, a relatively large body of research has described psychosocial functioning among children with craniofacial conditions and their families. Although many children adjust well, some children with craniofacial conditions experience psychosocial issues, including low self-concept (Abbott et al., 2011; Hunt et al., 2006), learning disabilities (Conrad, Richman, Nopoulos, & Dailey, 2009; Millard & Richman, 2001; Nopoulos, Langbehn, Canady, Magnotta, & Richman, 2007), depression (Hunt, Burden, Hepper, Stevenson, & Johnston, 2006; Hunt et al., 2005; Murray et al., 2010), social anxiety and inhibition (Murray et al., 2010; Sank et al., 2003), and negative social reactions (teasing, peer rejection) from others because of facial disfigurements and speech issues (Berger & Dalton, 2011; Kramer et al., 2009). Researchers have also reported that children with craniofacial issues tend to experience more internalizing and externalizing problems compared to children who are born without them (Hunt et al., 2006).

Although most prior research has focused on the affected children, it has long been recognized that families of children born with CL/P could also experience psychosocial issues (Berger & Dalton, 2011; Hunt et al., 2006; Kramer et al., 2009; Murray et al., 2010). Yet, to date research with parents has been primarily cross-sectional, quantitative, and focused on the emotional, social, and care experiences of mothers (Baker et al., 2009; Despars et al., 2011; Nelson, Glenny, Kirk, & Carress, 2011; Nelson, Kirk, Caress, & Glenny, 2012; Nusbaum et al., 2008). Couples' experiences have been largely neglected and even more sparse are studies that have examined how the timing of a child's diagnosis (prenatal/in utero or postnatal/at birth) affects how couples cope and adapt. A better understanding of the impact of CL/P on couples is needed because it can expand our

understanding of the relational effects of having a baby born with CL/P and lead to the development of couple-based prevention and intervention programs.

In order to fill this gap, this descriptive qualitative study was designed to examine the experiences of both mothers and fathers currently caring for a child born with CL/P, using a dyadic approach to research. The primary purpose of this paper is to describe findings from a descriptive qualitative study (Creswell, 2013) that used content analysis (Braun & Clarke, 2006) to code the data to examine how the timing of a child's CL/P diagnosis (prenatal versus postnatal) affects couples. We conducted semi-structured interviews with 17 couples (10 prenatal and 7 postnatal) currently caring for children (1 to 4 years old) who were born with CL/P from an ongoing quantitative study in a northeastern children's hospital in the U.S. In this paper, we describe six themes that emerged from the couple interviews, clinical implications, and recommendations for future research. First, we review the theory that informed this study.

THEORETICAL FRAMEWORK

The Resiliency Model of Family Stress, Adjustment and Adaptation (McCubbin & McCubbin, 1993) informed the development of this study. McCubbin and McCubbin (1993) identified myriad factors that can affect how couples adjust to an external stressor, like a baby prenatally or postnatally diagnosed with CL/P. McCubbin and McCubbin (1993) reported the following factors can affect adjustment and coping to an external stressor: (1) family's vulnerability, (2) established patterns of functioning, (3) resources, (4) appraisal of the stressor, (5) problem solving, and (6) and coping strategies. These factors help to determine if families are able to effectively cope and adapt to a stressor, resulting in either *maladjustment* (crisis) or *bonadjustment* (growth) (McCubbin & McCubbin, 1993).

If a couple becomes *maladjusted* while facing a crisis or stressor like CL/P in an offspring; this marks the beginning of the adaptation phase to restore stability. *Bonadaptation*

describes coping and adapting to an external stressor at the individual, family and community levels. Factors that play an important role during the adaptation or adjustment phase are: (1) pileup of demands, (2) newly established patterns of functioning, (3) family type, (4) family resources, (5) social support, (6) family appraisal, (7) problem solving, and (8) coping mechanisms.

Based on this theoretical framework, the interview guide used in this study included questions that asked specifically about factors leading to adjustment and adaptation. Rather than categorizing couples as having achieved *bonadaptation* or not, the research team focused on how couples' experiences fit within this framework. Next, we summarize prior research that examined how parents cope with prenatal versus postnatal diagnoses of CL/P.

PRENATAL VERSUS POSTNATAL DIAGNOSIS OF CL/P

In the U.S., an ultrasound examination is routinely conducted on all pregnant women at the end of their first trimester or during the beginning of their second trimester. A prenatal diagnosis of CL/P is becoming more common because 3D ultrasounds are a more routine procedure during pregnancy; CL more than CP (Campbell, Lees, Moscoso, & Hall, 2005; Platt, Devore, & Pretorius, 2006). Despite this improved technology, most cleft cases in the U.S. are still diagnosed at birth (postnatally). Prenatal diagnosis is more common among CL/P and CL cases because a cleft of the lip is more easily visualized in an ultrasound as compared to cleft palate. Additionally, mothers with higher household incomes are more likely to receive a diagnosis prenatally because of the higher frequency and thoroughness of the ultrasound examinations (Johnson et al., 2009).

After first learning about the diagnosis, parents in both groups (prenatal and postnatal diagnosis) have reported feelings of shock and searching for a possible cause of the cleft (Nusbaum et al., 2008; Pope et al., 2005). They describe feeling: (1) sadness and grief, (2) anxiety regarding how to care for a child born with CL/P, and (3) guilt about the cleft

condition. At the same time, parents also report experiencing feelings of happiness about having a new baby (Johansson & Ringsberg, 2003; Nelson, et al., 2012; Nusbaum et al., 2008).

There are unique experiences among parents in the two diagnosis groups. Some parents who received the diagnosis prenatally reported having more time to prepare themselves psychologically, and to learn about the condition and course of treatment (Davalbhakta & Hall, 2000; Matthews et al., 1998; Nusbaum et al., 2008). Some parents whose children were diagnosed before birth reported they actually preferred not knowing ahead of time because they were anxious for the rest of the pregnancy, which prevented them from enjoying being pregnant (Baker et al., 2009; Kuttenger, Ohmer, & Polska, 2010).

Parents who receive a postnatal diagnosis of their child reported questioning why CL/P was not diagnosed prenatally, despite having the recommended ultrasound examinations (Davalbhakta & Hall, 2000; Matthews, et al., 1998). Parents have also reported preferring to have an opportunity to adjust *before* the birth of their babies so they could psychologically, financially, and medically prepare themselves (Kuttenger, et al., 2010; Nusbaum et al., 2008). In contrast, some parents in the postnatal diagnosis group reported they would have been more anxious during the pregnancy if they had received the CL/P diagnosis before the birth which was also reported by parents diagnosed prenatally (Davalbhakta & Hall, 2000; Kuttenger, et al., 2010; Matthews, et al., 1998; Nusbaum et al., 2008).

IMPACT OF CL/P ON COUPLES

Few researchers have examined how having a child born with CL/P impacts couples. Most have examined parental stress (Pope, Tillman, & Snyder, 2005), mothers' functioning (Speltz, Arnsden, & Clarren, 1990), parental sensitivity (Pelchat, Bisson, Bois, & Saucier, 2003), adaptation (Pelchat et al., 1999; Pelchat, Lefebvre, Proulx, & Reidy, 2004), and

attachment (Speltz, Endriga, Fisher, & Mason, 1997). Among the few studies with couples, Pelchat, Bisson, Bois and Saucier (2003) evaluated marital distress among mothers and fathers of toddlers born with CL/P and with Down syndrome compared to toddlers who had no disabilities; the timing of the diagnosis (prenatal versus postnatal) was not reported in this study.

Pelchat and colleagues (2003) interviewed 117 18-month-old toddlers and their parents (116 mothers and 84 fathers). Marital stress was measured using the following four statements: (1) In the last six months, I had difficulties to accept that my spouse expresses anger toward our family situation, (2) In the last six months, I had difficulties to accept that my spouse expresses sadness towards our family situation, (3) In the last six months, fights are more frequent between my spouse and me, and (4) In the last six months, we do not have any more activities together (Pelchat, et al., 2003, p 36). Marital stress was significantly associated with fathers' increased levels of insensitivity toward children born with clefts. Fathers who reported less marital stress tended to be more sensitive towards their children born with clefts.

Speltz, Armsden and Clarren (1990) evaluated how having a child born with a craniofacial issue affected marital adjustment at post-infancy. In the CL/P group, mothers and 33 of their offspring between the ages of 1 and 3 with CL/P, CP, or sagittal synostosis (premature closing of the soft spot on the top of a baby's head) were included. Twenty-three of these children were born with CL/P or CP. The demographically matched control group included 22 mothers and their children who did not have any health issues. Speltz and colleagues (1990) evaluated marital satisfaction using the Locke-Wallace Marital Adjustment Scale (leisure time spent together, affection, agreement about finances and decision making). Mothers caring for a child born CL/P or CP tended to report lower marital adjustment compared to the matched control group.

Pelchat, Lefebvre, Proulx and Reidy (2004) implemented an early family prevention program for Canadian parents who recently gave birth to a baby with CL/P or Down syndrome to help new parents adapt and cope. Their program included six to 8 meetings with a nurse educator and each couple to: (1) normalize parents' reactions, (2) increase resources for adaptation, (3) increase self-awareness and competencies, and (4) facilitate mutual support between parents, extended family, and other services. During the first three meetings, nurse educators met with couples at the hospital, right after the birth of their child and until they headed home with their babies; then four to 6 meetings took place in couples' homes (the number of meetings was determined by the family's needs). The goal of this prevention program was to explore factors that affect parents' views of the situation, encourage parents to talk about their emotions and interact positively with their newborn, and to emphasize positive coping and adaptation.

A program evaluation study with couples who completed this program was conducted to assess participant satisfaction (Pelchat et al., 2004). Nurse educators gave parents a satisfaction survey that asked about individual, marital, parental, extended family/other support systems, and printed material during their last meeting. In total, 72 parents (36 couples) participated; out of these 72 parents, 19 mothers and 18 fathers had children born with CL/P. In the marital domain, parents reported that while caring for a child born with CL/P, they often forgot about each other. Some misunderstandings took place, causing them to withdraw from each other. Most parents reported the prevention program was very helpful because they were able to more openly discuss worries about shared parenting responsibilities and how it affected their relationship (Pelchat et al., 2004). Raising a child with CL/P can be challenging for parents. Yet few researchers have examined how having a child born with CL/P impacts couples. The purpose of this study was to examine how the

timing of a child's diagnosis (prenatal versus postnatal) affects couples' adjustment and their relationship

METHODS

A qualitative approach was used because of the limited research available describing couples' experiences caring for young children born with CL/P. We conducted a descriptive qualitative study (Creswell, 2013) with a purposive sample of 17 couples and used thematic content analysis (Braun & Clarke, 2006) to code the interviews.

Researchers' Characteristics

The lead researcher (first author) is a licensed couple and family therapist who was born prematurely and was also diagnosed with CL/P as a newborn. She was a couple and family therapy Ph.D. candidate when the interviews were conducted. During the study, the first author kept a journal to track her own biases and assumptions after each interview. The second author is a licensed couple and family therapist and academic; she conducts clinical research and treats families coping with pediatric and parental illnesses. The third author is a clinical psychologist who works in a hospital with families coping with CL/P, the fourth author is a licensed nurse and academic and works with families coping with disabilities, and the fifth author has her doctorate in marriage and family therapy and has clinical experience working with couples.

First Author's Personal Experiences with Cleft

The first author is a 32-year-old White, upper-middle class, heterosexual female who was born and raised in Turkey. She is engaged and has been in a relationship with her partner for over four years; she is the oldest child in her family and was born prematurely with a cleft lip palate and was diagnosed at birth. The medical provider was very calm when he told her mother the diagnosis; the doctor decided to not place the first author in an incubator because she was a very active and a healthy premature baby.

The first author's father was unable to attend her birth, however, her maternal grandmother was with the first author's mother who at first had a lot of trouble feeding her baby. Nurses tied her mother's breasts to help her milk dry up, because she was unable to breast feed due to the cleft, which was very hard for her mother physically and emotionally. The doctors told her mother, if she was not able feed her baby, she could die which led to a lot of worry and concern.

The first author had her first lip repair surgery when she was three months old. By the age of 19, she had undergone six surgeries to repair her nose, lips, and teeth as well as dental treatment that lasted for approximately 10 years. As a child, strangers often asked her questions about her facial features and some made fun of her nasal speech, and inability to pronounce some letters clearly. She learned that answering questions about her scars, surgeries, and dental treatments was a part of her life she needed to accept. She went through her own struggles in a different country, culture, and time period. For this reason, the first author tried to keep her personal biases in check by writing memos throughout the study and working closely with her research team in order to capture the couples' experiences coping with a child born with CL/P.

Sample Recruitment

Couples were recruited from an ongoing quantitative study at a U.S. cleft center located in a northeastern urban children hospital using a purposive and convenience approach to sampling. This ongoing study was designed to examine how the timing of a CL/P diagnosis affects parents. Within this sample there were 32 couples whose children received the diagnosis prenatally, and 13 couples whose children received the diagnosis at or shortly after birth; their children were between 1 to 4 years old. The specific inclusion criteria for our follow-up qualitative study were: (1) two biological parents of a child diagnosed with CL/P ages 18 years and older, (2) English speaking, and (3) parents are cohabitating or married at

the time of the interview. The exclusion criteria were: (1) inability to read and/or understand English, (2) presence of a cognitive or physical disability that would impair the parents' ability to complete self-report surveys and interviews, and (3) child was diagnosed with other significant health problems (e.g., heart problems), in addition to the cleft condition.

The first author contacted all couples who fit the inclusion criteria and asked if they wanted to volunteer for the study. The data collection continued until saturation was reached. The first author wrote detailed memos before and after each interview which helped the team track thematic saturation. The first author conducted three additional interviews; two for the prenatal and one for the postnatal diagnosis group after saturation was reached to ensure saturation was reached. The final sample included 17 couples (ten couples who received the diagnosis prenatally and seven couples who received the diagnosis postnatally). While the primary aim of this qualitative study included understanding the experiences of parents individually and dyadically as a couple, data reported here are for the seventeen couples who were interviewed together as a couple. The couples' children at the time of the study were between the ages of one and four; all children had non-syndromic CL/P.

Procedure

After first receiving approval from the Institutional Review Board at the northeastern pediatric hospital and university where the first author was a doctoral student, a recruitment letter was mailed to all eligible couples. Data were collected using: (1) a self-report demographic survey; (2) the Revised Dyadic Adjustment Scale (RDAS) which can range from 0 to 69. Higher scores indicate better marital adjustment; non-distressed is >48 ; moderately distressed is 32–47; and severely distressed is < 32 (Busby, Christensen, Crane, & Larson, 1995); and, (3) in-depth semi-structured interviews (father and mother interviewed separately, then together as a couple).

After couples provided written informed consent, completed the demographic self-report questionnaire (e.g., age, race, education level, years married or partnered, number of children at home, and current age of child born with CL/P) and the RDAS, a semi-structured interview was conducted in the following three-part sequence: (a) mother individually for 30 minutes, (b) father individually for 30 minutes, and (c) couple together for 30 minutes. Partners were interviewed first apart and then together for two reasons: (1) to triangulate the individual interviews with couple interviews and, (2) to ensure participants could describe experiences without undue pressure to conform to partners' experiences.

Face-to-face interviews occurred whenever possible, but participants were also offered the option of a telephone or a web-based interview when in-person interviews were not feasible. These options were offered to reduce participant burden, as parents of young children often need to balance numerous home and work commitments, which can make meeting face-to-face challenging. Three couple interviews were conducted face-to-face (two prenatal and one postnatal). One couple from the prenatal diagnosis group was interviewed using the Internet. The remaining 13 couple interviews, 7 from the prenatal diagnosis group and 6 from the postnatal diagnosis group, were conducted over the telephone. The telephone interviews were helpful for recruitment because it gave couples flexibility regarding the timing and location of the interview.

Even though the telephone interviews did not make it possible to track facial expressions and body language, the first author paid close attention to changes in participants' tone of voice as experiences were described. The face-to-face interviews may have led to a closer relationship between the interviewer and participants, especially because the interviewer shares the same visible difference with their children. Yet, there were no noteworthy differences between the lengths of interviews or the depth of the experiences shared among couples interviewed in-person versus over the telephone.

All interviews followed the same semi-structured interview guide, were audio-taped, and then transcribed verbatim. The semi-structured interview guide (see Table 1) asked about parents' experiences at diagnosis (e.g., feelings, concerns), birth experiences (e.g., parents' preparedness), first year (e.g., struggles, relationship with the child, how it impacts family life), reactions from their families, and impact on their relationship. One more question was added to the interview guide, "Is cleft a concern for your next child?" after the first interview.

Insert Table 1 here

Interviews lasted between 50 to 145 minutes; the average length was 90 minutes. Couples were given gift cards to thank them for their time; each parent received a twenty-dollar gift card. The interviewer took detailed memos during and after each interview to record impressions that could not be captured on the audiotape (e.g., tone of voice, facial expression, body language). The interviews, notes, and memos were all transcribed verbatim and coded as qualitative data. Participants used pseudonyms which they chose to ensure complete anonymity.

Data Analysis

Thematic conventional content analysis (Braun & Clarke, 2006) in which categories are derived directly from the text, was used to analyze the qualitative data. Hsieh and Shannon (2005) noted that conventional content analysis is ideal for describing experiences, particularly when existing literature or theory is scarce. The recorded interviews were transcribed verbatim and stored as electronic audio files in a password-protected computer. Data analysis included an initial reading and re-reading of interviews for content. MAXQDA (2010) qualitative software was used to organize the data. Codes, categories, and themes emerged from text segments of all lengths, from a single word to several sentences of text

(Braun & Clarke, 2006). Major themes and subthemes were described by more than half of participants. Data were analyzed independently by the three researchers (first, second, and fourth authors), and as a team. When disagreements occurred, we discussed them until consensus was reached.

Credibility, Transferability, Dependability, Conformability, and Trustworthiness

The following three strategies were used to increase creditability, transferability, dependability, conformability and trustworthiness (Creswell, 2013): (1) triangulation, (2) peer debriefing, (3) inquiry audit and, (4) member checking. Findings were triangulated in the following three ways: (1) examining the relational distress measure for husbands and wives before the interview, (2) multiple coders (first, second, and fourth authors), and (3) triangulation of mothers and fathers individual data with their couple data. The relational distress measure was used as a descriptive measure to assess if couple's descriptions of their current relationship were credible. While conducting the interviews, the first author tracked changes in partner's descriptions when interviewed alone and together and included her impressions in detailed memos. Furthermore, while coding the data the authors noted differences between the individual and couple interviews. Among all couples, the experiences shared when alone and together were similar.

Lincoln and Guba (1985) defined peer debriefing as explaining the research process to a disinterested peer who can explore and challenge the researcher's biases, meanings, and explanations. The de-briefer challenges the researcher about his/her hypotheses, encouraging deeper exploration and for planning the next steps. In this descriptive qualitative study, the de-briefer was the first author's peer, another doctoral student who was working on her qualitative dissertation at the same time.

Lincoln and Guba (1985) stated that ensuring credibility may be sufficient for ensuring dependability, however, they also described additional techniques that can increase

dependability of the findings. Inquiry audit was the technique used in this study. An auditor examined the research process and findings of the study to ensure its accuracy. The second and fourth authors audited the research methods and codes that emerged.

An inquiry audit is not possible without an audit trail. An audit trail, a technique for establishing conformability, describes how records are kept during each step of data collection and analysis. These records can include: (1) raw data of the interviews (electronic files), (2) field notes, (3) survey results, (4) data reduction and analysis trails, (5) products, (6) data reconstruction and synthesis trails and products, (7) reflexive memos, (8) notes describing the trustworthiness process, and (9) notes describing the theoretical frameworks (Lincoln & Guba, 1985). In this study, all of the steps leading to the research findings were carefully detailed; documentation was available for examination at any time.

Member checking was also used to increase credibility of the findings. Once the data analysis was completed, the first author contacted all participants (n=17 couples) via e-mail and asked if the themes generated captured their experiences. After waiting for two weeks, the first author sent an additional email reminder. In the email reminder, the first author noted that if she did not hear back from couples in two weeks, she would assume they were in agreement with codes that emerged. Thus, there was no need to re-interview couples after the coding or member-checking.

Two couples wrote back to confirm the findings of study who were very invested in the topic and findings. One couple was the first one to volunteer for the study and the second couple continuously expressed their enthusiasm about the study throughout their interview.

FINDINGS

Characteristics of Prenatal Diagnosis Group

Mothers' ages ranged from 32 to 45 (\bar{x} =39 years old); fathers' ages ranged from 30 to 52 (\bar{x} =41.20 years old) (see Table 2). Most couples had college or graduate degrees. All

fathers were employed full time; eight mothers were employed full or part-time. None of the fathers reported receiving psychological or psychiatric help during the last 12 months; one mother reported receiving psychiatric help and two mothers reported previously participating in support groups. The length of relationships ranged from six to 25 years; the mean length was 11.41 years. All couples were married and living together at the time of the interview, ranging from 3 years 8 months to 18.5 years. Half of the couples in the prenatal diagnosis group had two children; the other half had only one child. For seven out of 10 couples, their first child was born with CL/P. All couples in the prenatal diagnosis group had only one child born with CL/P.

Insert Table 2 here

Characteristics of Postnatal Diagnosis Group

Six out of 7 mothers and five out of 7 fathers returned their demographic surveys (see table 2). Mothers' ages ranged from 23 to 41 (\bar{x} =34.3 years old), and fathers' ages ranged from 24 to 43 (\bar{x} =34.4 years old). Three out of 6 mothers and three out of 5 fathers completed college and/or graduate school. Four out of 5 fathers and two out of 7 mothers were employed full time; two mothers and one father were employed part time and two mothers were homemakers. One father and one mother reported receiving psychological help during the last 12 months. Six out of the 7 couples were married and living together. One couple reported they were living together but planning to separate. The length of relationships ranged from five to 18.5 years; mean relationship length was 11.06 years. Four couples had two children and three couples had one child. For five out of 7 couples, their first child was born with CL/P. All couples in this group had only one child born with CL/P.

Revised Dyadic Adjustment Scale (RDAS)

The RDAS was scored for prenatal (n=10 couples) and postnatal couples (n=5 couples) who returned the RDAS (fathers and mothers separately). On average the prenatal diagnosis group was not distressed (48 or higher). In contrast, average scores of individuals in the postnatal diagnosis group were within the moderately clinically distressed range (32 to 47). In the prenatal diagnosis group, only two couples scored below the distress cut-off score of 48. Specifically, the average individual RDAS scores for each partner and average couple RDAS score (\bar{x} =53.4) for the prenatal diagnosis group were both well above the total cutoff score of 48. Additionally, the mean consensus (\bar{x} =24.8), satisfaction (\bar{x} =16.1) and cohesion (\bar{x} =12.5) scores indicated they were not distressed in these areas.

Four out of 5 couples in the postnatal diagnosis group were within the moderately clinically distressed range (32 to 47). The average individual RDAS score and the average couple score (\bar{x} =42.8) were also in the moderately distressed range, indicating couples who received a postnatal diagnosis were more relationally distressed at the time of the interview. The mean consensus (\bar{x} =20.6), satisfaction (\bar{x} =12.8) and cohesion (\bar{x} =9.4) scores also indicated distress in these areas.

Qualitative Interviews

The following six dominant themes (more than half of couples reported the theme) describe the 17 couples' experiences with CL/P: (1) *relationship growth*, (2) *challenges*, (3) *roles and responsibilities*, (4) *sources of support*, (5) *talking about cleft*, and (6) *lessons learned*. In the next section, the six themes are described and illustrated with anonymous quotes using pseudonyms couples chose during the interviews.

Dominant Theme One: Relationship Growth

In both diagnosis groups, most couples (nine prenatal and six postnatal) reported raising a child born with CL/P made their relationship stronger and increased faith in their

partnership. Together they worked as a team and supported each other. Overall, they were able to overcome challenges without turning on each other.

For example, Francis and Rachel from the prenatal diagnosis group said their ability to communicate improved under stress:

Rachel: We got through this because we – we got through it together. We can get through anything together.

Francis: Yeah, I would say we learned that our priorities are exactly the same. We are a great partnership.

Jill and Larry who learned about their daughter's CL/P diagnosis at birth, similarly said they learned how to depend on each other because they had more reason to rely on each other, especially during the first year:

Larry: I think we just learned to depend on each other for a lot of things, because it was a trying time for the first few months, after she was born. Everything we had to go through before the surgeries and a little bit after the surgeries, so I think we just really learned to depend on each other, maybe more so than we had previously.

Jill: It made our relationship stronger

Ann and Eric reported their relationship improved after the CL/P diagnosis postnatally, even though they were having problems throughout the pregnancy. Although they were in the process of separation at the time of the interview, both shared that raising a child with cleft made them feel closer to each other:

Ann: It helped our relationship because it just brought us together because there was like a bigger issue than just our own personal issues.

Interviewer: Okay. How did these issues bring you together?

Eric: It's important for me and her to stay civil with each other for our daughter's sake.

Even though most couples in both diagnosis groups reported this experience brought them closer, couples in the postnatal diagnosis group tended to report more pre-existing stressors prior to the birth of their child. For example, Diane and Jack said they received fertility treatments for two years before finally getting pregnant which was hard on their

relationship. Additionally, Jill and Larry had a difficult time scheduling the couple interview because they worked reverse shifts to balance work and taking care of their children.

Similarly, a couple in the postnatal diagnosis group, Minnie and Junior, reported more conflict because Minnie struggled with her mother-in-law because she was taking over the child care:

We ended up fighting, my husband and I. Then my mother-in-law and I ended up having a humongous fight after her surgery. It was never resolved. My husband says I was making it up, like it was all in my head. I'm being treated for postpartum depression now, but it's underlying depression and everything else because his mom and I always had issues. I guess her having cleft, it brought everything else out.

Darth and Vader (postnatal) as well as, Minnie and Junior (prenatal) were the only couples in their diagnosis groups who did not report relational growth. For Minnie and Junior, having a child with cleft exacerbated their pre-existing problems whereas Darth and Vader reported it did not impact their relationship either way.

Dominant Theme Two: Challenges

All couples reported experiencing some challenges. During the pregnancy, deciding whether they should have an amniocentesis or abortion was reported as a salient challenge for couples in the prenatal diagnosis group. It was risky to receive the amniocentesis because there was a chance of losing the baby. Jane and Mitch had a challenging time deciding; Jane said:

We'd missed the window for a CVS [Chorionic villus sampling (CVS) is a prenatal test that diagnoses chromosomal abnormalities such as Down syndrome, as well as other genetic disorders], so the question was, do you want to have an amniocentesis? Originally, I was afraid to do it. What if we lost the pregnancy? I think Mitch was concerned about that too. If you had the test for me and we lose the pregnancy because we had the test, will you ever be able to forgive me? For me it was if we don't do the test and we have a very sick baby that you might've wanted to not have, how will you ever forgive me?

The cleft diagnosis took the “pure joy” away from having a baby for many couples in the prenatal diagnosis group. They had to cope with the emotional, physical, and financial

impact of caring for a child born with CL/P. Consequently, it was difficult to keep their emotions “*in check*” when dealing with feelings of anxiety because of the “*unknown*”. At times, they were tired of “*bracing people*” and answering questions about the baby. Bill and Laurie shared:

Bill: I guess just the fact what I kept reiterating before is that we constantly have to feel like we have to compensate or brace everybody for because everybody is used to the everything is fine and dandy, perfect; perfect little person, and some people just don't – some people just have never had to deal with something like that.

Laurie: I think part of it was having to worry about everybody else. You know, I think that was difficult. I don't think it was as difficult between us, I think it was more us making sure that everybody else understood.

The first few months after the birth was reported as the most challenging for couples regardless of the timing of the diagnosis, especially for first-time parents (12 out of 17 couples). Couples were learning how to take care of a newborn, and attending many doctors' appointments. They had to decide on a treatment team and struggled when there were medical differences of opinion.

Surgery was the most challenging treatment consideration for couples in both diagnosis groups. For example, Rebecca and Ben were worried about possible complications and became very anxious when the surgery took longer than they expected. After the surgery, it was tough witnessing their child in pain and seeing any positive changes because of the swelling right after surgery. Ben said:

I guess the biggest challenge, now that I think back on it that was cleft-related, it was the day of the surgery, both in terms of when we said goodbye to the baby and hoping everything was going to be okay and nothing went wrong with the surgery. When things went a little longer than expected, but then also after the surgery when everything was swollen and it wasn't looking like what we thought it would. We imagined a perfectly healed baby at that time. Then also dealing with the idea that the baby is in pain and having to deal with that pain, and having to share that burden between the two of us.

Two couples in the prenatal diagnosis group, Francis and Rachel as well as Abby and Murray reported spending time as a couple became a challenge after their child was born.

When they became parents, priorities significantly changed; they had difficulty finding time to connect as a couple.

Couples in the postnatal diagnosis group reported they sometimes took their frustrations out on each other, experienced difficulty managing the division of labor, feedings, felt guilty about the cleft, financial struggles, and working as a team. For example, Jill and Larry whose daughter had colic in addition to having cleft lip palate said they often got upset with each other when they felt overwhelmed taking care of their daughter. At times, one of them felt like they were doing more childcare than the other person. They shared:

Jill: I think that she was so colicky, it was hard with that too. It just seemed like we would both get attitudes more, even with each other.

Interviewer: Because she was colicky, you were both agitated and you were both getting at each other?

Jill: Yes.

Interviewer: What about for you Larry, would you agree or do you have anything to add?

Larry: I agree with that. I think that we got frustrated, at times, and we would sort of take it out on each other.

Minnie and Junior, who also disclosed pre-existing relational issues shared that becoming a team was their biggest challenge. For Brandon and Sarah, making sure they had enough money for their son's treatment was a significant challenge. In order to afford the treatment, they had to make financial adjustments until they paid off the hospital bills. This situation was difficult because they got "*frustrated quickly over money.*"

In the postnatal diagnosis group three couples described delays in their children's speech and one couple in their child's development. These ongoing problems were described as additional stressors. In summary, couples in both diagnosis groups described experiencing relational challenges while caring for their children, especially during the first few months

after birth. Yet, couples in the postnatal diagnosis group described more relational and financial difficulties.

Dominant Theme Three: Roles and Responsibilities

In the prenatal diagnosis group, mothers reported doing most of the research, evaluating treatment teams, and scheduling medical appointments. In some cases, both mothers and fathers did research individually and then “*compared notes.*” One mother, Laurie, was also born with CL/P and was a speech pathologist. She took on more of the “*educator role*” at the time of the diagnosis and during the pregnancy because of her previous personal and professional experiences. One husband, Francis, described himself as the “*chauffeur and the part-time psychologist*” throughout the pregnancy. His wife, Rachel agreed that he was her “*rock*” and “*he held the ground*”. Francis also made sure their older daughter was not behind on her activities and took care of the finances.

During the birth, all prenatal couples were both present; husbands wanted to make sure mothers were doing well after the labor and hospital staff was attentive to their babies. Couples either tag-teamed, had more traditional gender roles or like Bill and Laurie tried to evenly divide up responsibilities:

Bill: We each took turns feeding him, putting him to sleep, and playing with him; responsibilities that a parent would require.

Laurie: Yeah, I think it was I think we divvied things up equally because my husband wanted to be a part of it also.

All fathers in the prenatal diagnosis group had to go back to work much earlier than the mothers did. For this reason, all 10 mothers diagnosed prenatally became the primary caregivers while fathers made sure “*bills were paid*” and “*financial resources were available*” for care and treatment.

Fathers continued to provide emotional support to their partners, despite going back to work soon after their child’s birth. Mothers pumped their breast milk because breast feeding

is often challenging for babies born with CL/P, did the diaper changing, NAM tapings [Nasoalveolar molding (NAM) is a nonsurgical method of reshaping the gums, lip and nostrils before cleft lip and palate surgery, lessening the severity of the cleft; within the first few weeks after birth, babies are fitted with a custom-made molding plate that looks like an orthodontic retainer], and made all medical appointments. Yet, no mothers in the prenatal diagnosis group identified their more traditional relationships as problematic.

Both mothers and fathers agreed that fathers shared the responsibilities of childrearing when they were home in the evenings and on weekends. For two couples, the fathers became more involved over time. For example, Jane and Mitch described changes in their relationship:

Jane: Now we are much more 50/50, whereas when we have a newborn, which we've been through twice, I'm much more lead on the process. Certainly, when we had our firstborn, who was the child with cleft.

Mitch: Everything is less defined. I think it just comes more natural. Initially, it was like okay, you are going to do this; I am going to do that. Now there's no discussion on who is doing what. It just kind of happens.

Even though the more traditional division of labor has not changed for the remaining eight couples, mothers did not report this as a current problem in their relationships.

In the postnatal diagnosis group, five out of 7 couples shared child care. Two couples had more traditional gender roles and for one of those couples it seemed to be a problem (Sarah and Brandon). After their child's birth, Sarah took care of their son while Brandon worked full time:

Sarah: I took care of the baby all of the time and my husband didn't change too many diapers or give too many baths. I was constantly with the baby. I got up in the middle of the night all of the time. He slept through the night peacefully and got up for work and went to work in the morning.

Brandon: I worked 12-hour days too, every single day, being self-employed, so it was kind of tough.

At the time of the interview, three mothers in the postnatal group described feeling frustrated with the division of labor at home. Sarah still hoped her husband would become more involved. Minnie said even though they tried to share responsibilities, she still feels it is unfair. The main difference between the two diagnosis groups seems to be more feelings of resentment among postnatal mothers regarding their partners' lack of involvement in child care.

Dominant Theme Four: Sources of Support

Couples in both diagnosis groups identified each other, friends, and family members as main sources of support. They supported each other by having open conversations about the cleft, going to medical appointments together, trying to share childcare, being affectionate towards each other, and giving each other respite, as needed. Yet how they supported each other varied depending on what their partner needed at the time. For example, Rachel and Francis shared:

Interviewer: How do you support each other?

Rachel: Communication.

Francis: Sometimes it's just hugging.

Rachel: The unspoken; emotional, like you said, a hug.

Francis: Sometimes it's giving the other one some space.

Some family members took care of their children when couples were at work or cared for older siblings when couples needed to take their babies to medical appointments. Some family members even learned how to feed their children and provided emotional support. For some couples, seeing their family members accepting their children was an important reassuring and a source of emotional support.

Friends were another important source of support for the couples in both diagnosis groups, especially if they had children the same age. Couples described friends visiting and

embracing their children as they would embrace any other child, being sympathetic, and even bringing over meals when they did not have time to cook. In addition to friends, co-workers and neighbors provided support; they received words of encouragement and prayer cards.

Chip said,

“I could think of a lady at work who I really only have limited contact with, but she kept sending me, whether it was a prayer card or whether it was information about the cleft or hooking me up with another parent that she wanted me to talk to who had this same problem with her child.”

For Abby and Murray, church was an important source of support:

Murray: She’s the church’s child.

Abby: The church’s child, not just ours. It’s like they take ownership of her along with us. So we really felt that love surrounding us. We didn’t really need them to do anything for us. Just knowing that they were there rooting us on, I think, being cheerleaders for us.

Furthermore, couples in both diagnosis groups identified their treatment teams as important sources of support. Francis and Rachel shared:

Francis: We can pick up the phone and call somebody at any given time and they’ll be happy to talk to us. We’ve sent a half a dozen emails to Meg and she gets back to us in 24 hours. We walk around here; we know everybody.

Rachel: Yep. We were greeted by [name of doctor] from day one, we were introduced to these doctors by their first name.

Francis: We know everybody here.

Rachel: We’re not a number to anybody. Everybody is a special person to each and every of these nurses and doctors.

The hospital staff gave them contact information of parents who had been through the same experience and invited them to annual picnics where they could meet similar families, which reduced their feelings of isolation. Additionally, couples used cleft-specific web sites and online support groups. Even though couples in both diagnosis groups reported similar sources of support, couples in the prenatal diagnosis group tended to identify more sources of support compared to couples in the postnatal diagnosis group.

Dominant Theme Five: Talking About Cleft

For couples in both diagnosis groups, cleft was no longer an issue they discussed regularly, even though they reported openly discussing it with each other during the first year of their child's life. Cleft was no longer a topic of everyday conversation because, *"it's not a concern"* anymore.

If their children had residual issues such as dental or speech issues, couples talked about how they could resolve them. For example, Mo stated; *"Her teeth maybe were a little bit of a concern for us too because she has a couple teeth that are a little out of line. I guess that's more the conversation we might be having now, not meaning today in particular but in general: What do you think they're going to do with her mouth?"*

In the postnatal diagnosis group, six out of seven couples worried about their next child being born with a cleft. Yet cleft was not described as a *"game changer"* regarding their decision to have another child. For Paul and Pam, cleft was a concern *"at the back of their minds"* but this did not stop them from having a second child. Yet, they wanted additional testing to rule out a cleft diagnosis for their second baby. Similarly, for Jack and Diane, cleft was a concern for them when they thought about having another child. They said now that they were older compared to when they had their daughter, they were nervous about more severe problems that can co-occur with cleft rather than the cleft itself. Yet, this did not stop them from actively trying to have a second child.

Gayle and Joey decided not to have another child for reasons that were not related to worries about cleft. Gayle stated she had complications during her last labor, so their decision not to have more children was related to her health and their financial situation. When talking about the possibility of their next child being born with cleft, Sarah and Brandon disagreed. Sarah stated that it would be a slight concern for her whereas Brandon said it would not be a

concern for him. They were also more worried about possible problems that could occur in the future. For example, Diane and Jack describe,

But like dental... She didn't have a cleft palate and a hard palate so we didn't really have anything concerned about with dental but it was a question that we asked the dentist. Like we're always curious if something that she's experiencing... if it could lead to something else, you know.

Most couples said they do not currently discuss the cleft with each other unless someone comments on their children's appearance or asks about the next stage of cleft treatment. Most couples said they were proud of their children because of their strength and resilience.

Dominant Theme Six: Lessons Learned

The sixth dominant theme, *lessons learned*, describes lessons learned while raising a child born with cleft. Couples in both diagnosis groups identified cleft as a “cosmetic” and “manageable” problem. They described the importance of doing research, learning about cleft and how to work with providers, finding the best treatment team and checking for additional anomalies once their child was born.

Most couples in the prenatal diagnosis group said they were happy they knew before the birth, because it gave them time to grieve the loss of the perfect child. Couples also described the importance of knowing about the severity of the cleft, and getting additional testing done to get a more definitive diagnosis. For example, Jane and Mitch said:

Jane: I think when you've got that diagnosis and it could be so bad, the more you can tell a couple about what their baby actually likely will experience, the better. So more imaging and better guidance on what they're seeing will mean in terms of—for me particularly it was feeding—but just so you can talk more specifically before you can find out exactly what's involved.

Mitch: I guess the more testing you can do and the more definitive you are in what situation you have, the easier it is to deal with and educate yourself rather than just reading a bunch of things on the internet that aren't really applicable to your situation.

For couples in the postnatal diagnosis group, this experience taught them to put things into perspective, the importance of developing a good support team, openly communicating with health care providers, and going to a hospital with the expertise to provide CL/P care. Larry and Jill realized their main priorities in life were being emotionally and physically present for their children; Larry said:

I've learned to kind of take a step back and realized how important it is to be there for the kids and support them. I knew that before, but going through this, it really hits home what you are capable of dealing with and how much you love your kids. I mean I don't want her to be born with that, but in a way it's been a positive experience just the challenges we've gone through. It's made me stronger and I'm sure it's made her stronger, too.

Minnie and Junior said having a child with speech and developmental delays helped them put things into perspective. Junior said, *"It's hard to say with the surgery or just having kids in general just that all the small things you worry about before that and how much it really didn't matter."* Couples in both diagnosis groups described the importance of getting educated about CL/P and having a plan as well as keeping things in perspective so they could work as a team and be responsive and loving parents.

DISCUSSION

Prior research conducted with parents of children born with cleft have primarily included mothers (Baker et al., 2009; Johansson & Ringsberg, 2003). This descriptive qualitative study filled a gap by eliciting detailed, first-person descriptions from couples currently caring for a child born with CL/P. During the semi-structured interviews, most couples in both diagnosis groups reported raising a child born with CL/P made their relationship stronger. Our findings suggest a couple's vulnerability, pre-existing patterns of functioning, practical and financial resources and problem appraisal are all important factors that can impact adjustment which supports the Resiliency Model of Family Stress (McCubbin & McCubbin, 1993).

For twelve out of 17 couples who participated in this study, their babies born with cleft were their first children. Thus, they were coping with the normative stressor of having a new baby and the non-normative stressor of having a baby born with cleft (McCubbin & McCubbin, 1993). Couples whose RDAS scores were in the moderately distressed range tended to be postnally diagnosed but also described more pre-existing stressors (e.g., fertility treatments, financial issues, struggles with role distribution) prior to having a child born with CL/P. It is possible these couples were relationally distressed because of the postnatal diagnosis or they could have been relationally distressed before the diagnosis because of these additional stressors. Another possibility is that couples diagnosed prenatally had more time to develop a treatment plan, find social support and resources and overall were better prepared for the birth of their child, leading to better couple adjustment when they were interviewed. Since this was a retrospective study, understanding these differences is limited until more prospective studies are done in the future.

Even though couples in the postnatal diagnosis group tended to report more stress when they gave birth and during the first few months, most couples were not struggling at the time of the interviews, unless their child had ongoing delays in speech and development, which are not uncommon among children born with CL/P (Pope et al., 2005; Speltz et al., 1990). For example, three out of the seven children diagnosed postnatally had speech problems; two children were in speech therapy at the time of the interview. Even though couples generally identified cleft as a “*manageable*” and “*cosmetic*” problem, their appraisal of cleft varied if their children were still struggling with speech and development issues. They had to find the appropriate treatment and care for their children, find time and the financial resources for the treatment and attend to speech and development issues. Prior studies suggest that having a child with medical issues can affect family adjustment (Baker et

al., 2009; Berger & Dalton, 2011). Thus, it is possible that couple functioning could be affected when dealing with these ongoing stressors.

Yet, most couples in this study reported this experience provided an opportunity to work as a team and to be supportive of each other, which suggests relationship growth was experienced. Even though most cleft literature has examined parent-child and patient-provider relationships, understanding couples' adjustment is an understudied area (Gassling et al., 2014; Johansson & Ringsberg, 2003; Stock & Rumsey, 2015). Furthermore, despite the existing literature on marital discord and women's employment (Schoen, Astone, Rotherd, Standish, & Kim, 2002), in our study only three women (postnatal) who worked outside of the home reported difficulties sharing child care. Although more prenatal mothers were stay at home caregivers, they did not seem resentful of this child care arrangement.

Similar to other studies, couples who received the diagnosis prenatally had more time to process their feelings and educate themselves about cleft and the course of treatment before their babies were born (Speltz et al., 1990; 1997). For this reason, these couples were more prepared to care for their children, while parents who learned of their child's condition at birth tended to understandably experience more feelings of shock and confusion while at the same time needing to quickly learn how to care for their child (Kuttenberger et al., 2010; Nelson et al., 2011; 2012). Initially, this affected couples' appraisal of the problem since they had to manage caring for a new baby who had feeding issues and develop a treatment plan. Nusbaum et al. (2008) reported that parents who received a postnatal diagnosis in their study eventually adjusted; they were able to find resources and social support quickly. In our study, most couples in the postnatal diagnosis group described feeding issues, guilt about not being able to breastfeed and giving birth to a visibly different child as well as struggling to at first find the right resources but they also eventually adjusted.

Noteworthy prior research on cleft suggests that guilt and self-blame are common experiences among mothers (Nelson, Kirk, & Caress, 2011; Nusbaum et al., 2008). In our study, three mothers and one father from the postnatal diagnosis group reported experiencing self blame. Two mothers were concerned about not being able to breastfeed their children, one mother felt guilty for giving birth to a visibly different child despite her husband's good looks, and the one father was concerned genetically he contributed to the cause of the cleft.

Clinical Implications

The time period right after a CL/P diagnosis and the first few months after the birth were described as the most challenging for parents in both diagnosis groups and beyond that time period, if their children had ongoing developmental and speech delays. This is also when couples were in closer contact with the cleft treatment team (Nelson et al., 2011; 2012). Thus, providers should conduct routine psychosocial assessments and provide education about the impact of cleft on couple functioning (Author, 2012; Pelchat et al., 2003; 2004). Hospital staff and treatment teams should be prepared to help couples targeting factors that would promote resiliency. Although CL/P is not uncommon, some couples reported hospital staff did always not know how to help them, suggesting that all front line providers need to be educated so they can connect families to local cleft treatment teams and resources such as Cleft Palate Foundation (American Cleft Palate Craniofacial Association, 2009). It is important that medical providers are knowledgeable about the cleft since their tone and demeanor while delivering the diagnosis can impact couples' appraisal of the problem. Furthermore, the medical team's knowledge and demeanor can facilitate the development of trust between the treatment team and the family; this relationship is an important resource for couples throughout their child's treatment.

Our findings also suggest mental healthcare providers and family therapists should assess couples' existing patterns of functioning and problem appraisal especially during the

first year. Couples who had pre-existing conflicts and significant relational distress before their children's diagnosis or birth may need interventions tailored to improve their relationship, for example, couples therapy or a couple's support group. Furthermore, mental healthcare providers should meet more often with parents whose children are experiencing ongoing difficulties.

Since cleft treatment requires ongoing appointments with multiple providers, we recommend having a medical family therapist on staff who can administer psychosocial assessments, provide ongoing support and inform the treatment team about the specific dynamics and needs of the families.

Limitations

Although a purposeful and criterion-based sampling strategy was used in this study, participants still self-selected to be interviewed. There may have been sampling bias in which only couples who had more positive experiences raising a child with CL/P decided to participate. Yet, it is important to note that three families in the postnatal diagnosis group had ongoing stress because of their children's developmental delays. Most couples in both diagnosis groups reported they had a "*positive experience*" overall.

The couples provided retrospective descriptions of their experiences regarding the timing of the diagnosis, birth, and initial stages. Thus, it is possible they did not accurately remember some parts of the process such as specific experiences, feelings, and concerns. In order to overcome this limitation, each parent was first interviewed individually before interviewing the couple together, to get a more systemic understanding of their experiences.

This sample of couples was not racially diverse and did not include non-English speakers, which may add additional stress when interacting with the health care system. There could also be differences in responses among couples who had experienced their child's birth in the last year versus responses among couples whose children were three or

four years old. Due to the small, non-representative sample, findings from this study cannot be generalized, a fundamental assumption of qualitative research.

Finally, telephone interviews may have limited capturing participants' non-verbal communication during the interviews. Nonetheless, this option facilitated the participation of both partners, given most couples were balancing the demands of work and caring for their young families. Phares, Lopez, Fields, Kamboukos and Duhig (2005) reported that engaging parents in research can be challenging because of work and family commitments; conducting interviews on the telephone or using the Internet can be an effective strategy for including parents in health-related research.

Recommendations for Future Research

Future studies should be designed to examine couples' experiences in real time. More studies should be designed to understand the impact of the type of cleft (cleft lip, cleft palate, or cleft lip palate) on child development and family functioning because two out of three children who were experiencing delays in development and speech were born with cleft palate. Future research should focus on intervening with couples who report more distress; couple-based approaches delivered by a multi-disciplinary treatment team are a promising area of future clinical research. For example, the relationship distress of couples can be routinely evaluated at diagnosis (in utero and at birth) and then assessed again during the first year. Couples who are more distressed should be routinely assessed to better understand challenges they are experiencing in their relationships. Finally, there is a dearth of diverse samples in the cleft literature. It is important to understand the experiences of minority couples and non-English speakers who are raising a child born with CL/P so that linguistically and culturally sensitive interventions can be developed.

CONCLUSION

Findings from this descriptive qualitative study suggest that regardless of the timing of diagnosis, couples coping with a child born with CL/P could benefit from: (1) an initial information session with both parents at diagnosis (e.g., describing the course of treatment; feeding, how to work as a team, screening for relational distress); (2) peer support from other couples to reduce feelings of isolation; and, (3) ongoing screening and referrals for couple-based interventions, especially for couples who have pre-existing stressors (e.g., marital distress, financial concerns). Finally, future research should include more racially and economically diverse samples of couples to develop culturally sensitive intervention and prevention programs.

REFERENCES

Author (2012).

American Cleft Palate Craniofacial Association. Parameters for Evaluation and Treatment of Patients with Cleft Lip/Palate or Other Craniofacial Anomalies-Revision. 2009.
Available at: http://www.acpa-cpf.org/uploads/site/Parameters_Rev_2009.pdf.
Accessed September 11, 2014.

Abbott, M. M., Kokorowski, P. J., & Meara, J. G. (2011). Timeliness of surgical care in children with special health care needs: delayed palate repair for publicly insured and minority children with cleft palate. *Journal of Pediatric Surgery*, 46(7), 1319-1324. doi: 10.1016/j.jpedsurg.2010.10.002

Baker, S. R., Owens, J., Stern, M., & Willmot, D. (2009). Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. *Cleft Palate-Craniofacial Journal*, 46(3), 229-236. doi: 10.1597/08-075.1

Banales, J. E. (2009). Cleft Lip/Palate more prevalent in Asians. Retrieved from U.S. Department of Health and Human Services Office of Minority Health website: <http://minorityhealth.hhs.gov/templates/content.aspx?ID=7590&lvl=3&lvlID=287>

Berger, Z. E., & Dalton, L. J. (2009). Coping with a cleft: Psychosocial adjustment of adolescents with a cleft lip and palate and their parents. *The Cleft Palate–Craniofacial Journal*, 46, 435–443. doi: 10.1597/08-093.1

Bille, C., Skytthe, A., Werner, V., Knudsen, L. B., Andersen, A. N., Murray, J. C., & Christensen, K. (2005). Parent's Age and the Risk of Oral Clefts. *Epidemiology*, 16(3), 311-316.

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, 3, 77-101. doi: 10.1191/1478088706qp063oa

- Conrad, A. L., Richman, L., Nopoulos, P., & Dailey, S. (2009). Neuropsychological functioning in children with non-syndromic cleft of the lip and/or palate. *Child Neuropsychology*, 15, 471–484. doi:10.1080/09297040802691120
- Creswell J.W. (2013). *Qualitative Inquire and Research Design*, 3rd edition. Thousand Oaks, California: Sage.
- Davalbhakta, A. & Hall, P. N. (2000). The impact of antenatal diagnosis on the effectiveness and timing of counseling for cleft lip and palate. *British Journal of Oral and Maxillofacial Surgery*, 53, 298-301. Doi: 10.1054/bjps.2000.3330
- Despars, J., Peter, C., Borghini, A., Pierrehumbert, B., Habersaat, S., Muller-Nix, C., Ansermet, F., & Hohlfeld, J. (2011). Impact of a cleft lip and/or palate on maternal stress and attachment representations. *Cleft Palate-Craniofacial Journal*, 48(4), 419-424. doi: 10.1597/08-190
- Friedman, O., Wang, T. D., Milczuk, H. A. (2010). Cleft Lip and Palate. In P. W. Flint, B. H. Haughey, V. J. Lund, J. K. Niparko, M. A. Richardson, K. T. Robbins, & J. R. Thomas (Eds.), *Cummings Otolaryngology: Head & Neck Surgery: Volume 3. Pediatric Otolaryngology* (pp. 2659-2675). Philadelphia, PA: Mosby Elsevier.
- Hunt, O., Burden, D., Hepper, P., & Johnston, C. (2005). The psychosocial effects of cleft lip and palate: a systematic review. *The European Journal of Orthodontics*, 27(3), 274-285. doi: 10.1093/ejo/cji004
- Hunt, O., Burden, D., Hepper, P., Stevenson, M., & Johnston, C. (2006). Self-reports of psychosocial functioning among children and young adults with cleft lip and palate. *The Cleft Palate-Craniofacial Journal*, 43(5), 598-605. doi: 10.1597/05-080
- Hsieh, H. F., & Shannon, S. E. (2005). Three approaches to qualitative content analysis. *Qualitative health research*, 15(9), 1277-1288. doi: 10.1177/1049732305276687

- Johansson, B., & Ringsberg, K. C. (2003). Parents' experiences of having a child with cleft lip and palate. *Issues and Innovations in Nursing Practice*, 47(2), 165-173. doi: 10.1111/j.1365-2648.2004.03075.x
- Johnson, C. Y., Honein, M. A., Hobbs, C. A., Rasmussen, S. A., & the National Birth Defects Prevention Study. (2009). Prenatal diagnosis of orofacial clefts, National Birth Defects Prevention Study, 1998 – 2004. *Prenatal Diagnosis*, 29, 833-839. doi: 10.1002/pd.2293
- Kramer, F. J., Baethge, C., Sinikovic, B., Schliephake, H. (2007). An analysis of quality of life in 130 families having small children with cleft lip/palate using the impact on family scale. *International Journal of Oral and Maxillofacial Surgery*, 36, 1146-1152. doi: 10.1097/SCS.0b013e3181be8892
- Kuttenberger, J., Ohmer, J. N., & Polska, E. (2010). Initial counseling for cleft lip and palate: Parents' evaluation, needs and expectations. *International Journal of Oral and Maxillofacial Surgery*, 39, 214-220. doi: 10.1016/j.ijom.2009
- Matthews, M. S., Cohen, M., Viglione, M., Brown, A. S. (1998). Prenatal counseling for cleft lip and palate. *Plastic and Reconstructive Surgery*, 101(1), 1-5. doi: 10.1097/00006534-199801000-00001
- Murray, L., Arteche, A., Bingley, C., Hentges, F., Bishop, V. M. D., Dalton, L., Goodacre, T., & Hill, J. (2010). The effect of cleft lip on socio-emotional functioning in school-aged children. *Journal of Child Psychology and Psychiatry*, 51(1), 94-103.
- MAXQDA. (2010). Software for qualitative data analysis. Distributed by VERBI Software. Marburg, Germany. Available for purchase at <http://www.maxqda.com/>

- McCubbin, H. I., & McCubbin, M. A. (1993). Families coping with illness: Resiliency Model of Family Stress, Adjustment and Adaptation. In C. B. Danielson, B. Hamel-Bissell, & P. Winstead-Fry (Eds.), *Families, Health & Illness: Perspectives on Coping and Intervention* (pp. 21-63). St Louis, Missouri: Mosby-Year Book, Inc.
- Millard, T., & Richman, L. C. (2001). Different cleft conditions, facial appearance, and speech: relationship to psychological variables. *The Cleft Palate-Craniofacial Journal*, 38(1), 68-75. doi: 10.1597/1545-1569(2001)038<0068:DCCFAA>2.0.CO;2
- Nelson, P., Glenny, A. M., Kirk, S., & Caress, A. L. (2011). Parents' experiences of caring for a child with a cleft lip and/or palate: a review of the literature. *Child: Care, Health and Development*, 38(1), 6-20. doi:10.1111/j.1365- 2214.2011.01244.x
- Nelson, P. A., Kirk, S. A., Caress, A., & Glenny, A. (2012). Parents' emotional and social experiences of caring for a child through cleft treatment. *Qualitative Health Research*, 22(3), 346-359. doi: 10.1177/1049732311421178
- Nopoulos, P., Langbehn, R. D., Canady, J., Magnotta, V., & Richman, L. (2007). Abnormal Brain Structure in Children with Isolated Clefts of the Lip or Palate. *Archives of Pediatrics and Adolescent Medicine*, 161(8), 753-758.
- Nusbaum, R., Grubs, R. E., Losee, J. E., Weidman, C., Ford, M. D., & Marazita, M. L. (2008). A qualitative description of receiving a diagnosis of clefting in the prenatal or postnatal period. *Journal of Genetic Counseling*, 17, 336-350. doi: 10.1007/s10897-008-9152-5
- Parker, S. E., Mai, C. T., Canfield, M. A., Rickard, R., Wang, Y., Meyer, R. E., Anderson, P., Mason, C. A., Collins, J. S., Kirby, R. S., & Correa, A. (2010). Updated National Birth Prevalence estimates for selected birth defects in the United States, 2004–2006. *Birth Defects Research (Part A): Clinical and Molecular Teratology*, 88, 1008-1016. doi: 10.1002/bdra.20735

- Pelchat, D., Bisson, J., Bois, C., & Saucier, J. (2003). The effects of early relational antecedents and other factors on the parental sensitivity of mothers and fathers. *Infant and Child Development*, 12, 27-51. doi: 10.1002/icd.335
- Pelchat, D., Lefebvre, H., Proulx, M., & Reidy, M. (2004). Parental satisfaction with an early intervention program. *Journal of Perinatal and Neonatal Nursing*, 18(2), 128-144. doi: 10.1097/00005237-200404000-00008
- Phares, V., Lopez, E., Fields, S., Karnboukos, M. A., & Duhig, A. M. (2005). Are fathers involved in pediatric psychology research and treatment? *Journal of Pediatric Psychology*, 30(8), 631-643. doi:10.1093/jpepsy/jsi050
- Pope, A. W. & Snyder, H. T. (2005). Psychosocial adjustment in children and adolescents with a craniofacial anomaly: age and sex patterns. *Cleft Palate – Craniofacial Journal*, 42(4), 349-354. doi: <http://dx.doi.org/10.1597/04-043R.1>
- Pope, A. W., Tillman, K., Snyder, H. T. (2005). Parenting stress in infancy and psychosocial adjustment in toddlerhood: A longitudinal study of children with craniofacial anomalies. *Cleft Palate-Craniofacial Journal*, 42(5), 556-560. doi: <http://dx.doi.org/10.1597/04-066R.1>
- Ricard, N., Bouchard, J-M., Perreault, M., Saucier, J. F., Berthiaume, M., & Bisson, J. (1999). Adaptation of parents in relation to their 6-month-old infant's type of disability. *Child Care, Health and Development*, 25(4), 377- 397. doi: 10.1046/j.1365-2214.1999.00107.x
- Sank, J. R., Berk, N. W., Cooper, M. E., & Marazita, M. L. (2003). Perceived social support of mothers of children with clefts. *The Cleft Palate-Craniofacial Journal*, 40(2), 165-171. doi: [http://dx.doi.org/10.1597/1545-1569\(2003\)040<0165:PSSOMO>2.0.CO;2](http://dx.doi.org/10.1597/1545-1569(2003)040<0165:PSSOMO>2.0.CO;2)

- Schoen, R., Astone, N. M., Rothert, K., Standish, N. J., & Kim, Y. (2002). Women's Employment, Marital Happiness and Divorce. *Social Forces*, 81(2), 643-662. doi: 10.1353/sof.2003.0019
- Speltz, L. M., Armsden, G. C., & Clarren, S. (1990). Effects of craniofacial birth defects on maternal functioning post-infancy. *Journal of Pediatric Psychology*, 15(2), 177-196. PMID: 2374074
- Speltz, L. M., Endriga, C. M., Fisher, P., & Mason, A. C. (1997). Early predictors of attachment in infants with cleft lip and/or palate. *Child Development*, 68(1), 12-25. PMID: 9084121
- Wilkins-Haug, L. (2012). Etiology, prenatal diagnosis, obstetrical management, and recurrence of orofacial clefts. Up to Date website: <http://www.uptodate.com/contents/etiology-prenatal-diagnosis-obstetrical-management-and-recurrence-of-orofacial-clefts>

Table 1. Semi-structured Interview Guide for Couples

Interview Questions
1. If you were talking to someone as a parent, what would advise them?
2. What did you learn from your experience that was most helpful to you?
3. What was the biggest challenge for you in this process?
4. What were the roles or responsibilities that you and your spouse took on surrounding the time of your child's diagnosis? How were decisions made for your family at that time?
5. Did you tell friends in addition to family members about the cleft before your child was born? (Prenatal only)
6. How did you explain your child's condition to people after birth? (Post-natal only)
7. Should parents meet with another parent who has had a child born with a cleft?
8. How do the two of you manage the care of your child?
9. How did this experience impact your relationship?
10. How do you arrive at decisions regarding a) treatment b) parenting?
11. Does your child have trouble with a) making friends b) appearance concerns c) speech d) development? How do you deal with it? Does this impact your social life?
12. How do you talk about cleft lip palate a) among yourselves b) within your family c) to other people
13. How are you talking to your child about CLP?
14. Where do you get your support from?
15. Is there anything else important that you think we should know about your experience?

Table 2. Demographic Profile of Couples (N=17)

Demographic Variable	Prenatal Group of Parents	Postnatal Group of Parents
Race		
White	18	11
Other (Asian)	2	0
Missing	0	3
Total	20	14
Education		
High School Diploma	2	1
Some College	1	1
College/Graduate Degree	17	9
Missing	0	3
Total	20	14
Occupation		
Full Time	17	7
Part Time	1	2
Homemaker	2	2
Missing	20	3
Total	20	14
Mothers' Age Range (years)	32-45	23-41
Fathers' Age Range (years)	30-52	24-43
Mean Length of Relationship (years)	11.41	11.06
Child's diagnosis		
Cleft Lip Palate (CL/P)	8	1
Cleft Palate (CP)	0	4
Cleft Lip (CL)	2	2
CL/P Child's Age		
1	1	0
2	5	4
3	3	2
4	1	1
Total Number of Children		
1	5	3
2	5	4
Birth Order of CL/P Child		
1 st	7	5
2 nd	3	2