

**HHS PUBLIC ACCESS**

Author manuscript

Health Psychol. Author manuscript; available in PMC 2016 May 01.

Published in final edited form as:

Health Psychol. 2016 May ; 35(5): 474–482. doi:10.1037/hea0000262.

Caregiver responses to early cleft palate care: A mixed method approach

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Abstract

Objective—This study sought to understand caregivers' (CGs') responses to early cleft lip/palate care for their infants.

Method—A prospective, mixed methods multicenter longitudinal study was conducted among CGs (N=118) seeking treatment for their infants' cleft lip and palate or cleft lip only at one of six cleft treatment centers in the United States. Participants were in one of two treatment groups: traditional care only or nasoalveolar molding (NAM) plus traditional care. The CGs completed semi-structured interviews and standardized questionnaires assessing psychosocial well-being and family impact at three time points: the beginning of treatment (~1 month of age), pre-lip surgery (~3–5 months of age), and post-palate surgery (~12–13 months of age). Multi-level modeling was used to longitudinally assess CGs' psychosocial outcomes.

Results—While the first year was demanding for all CGs, NAM onset and the child's lip surgery were particularly stressful times. CGs used optimism, problem-solving behavior, and social support to cope with this stress. Qualitatively, CGs' ability to balance cleft treatment demands with their psychosocial resources and coping strategies influenced family adaptation. Qualitative and quantitative results indicated CGs of NAM-treated infants experienced more rapid declines in anxiety and depressive symptoms and better coping skills over time than CGs whose infants had traditional care.

Conclusion—CGs of NAM-treated infants experienced more positive psychosocial outcomes than CGs whose infants had traditional care. Results from the mixed model support the Family Adjustment and Adaptation Response Model as used in pediatric chronic condition research.

Keywords

Cleft lip and palate; family adjustment; coping; depression

Introduction

The treatment for cleft lip and palate (CLP), the second most common birth defect in the United States (Parker et al., 2010), is long and arduous, starting in infancy and often lasting into young adulthood. The first year of treatment is particularly burdensome as infants are evaluated by multiple specialists and must undergo two surgeries—the first to close the lip at around 3–4 months of age and the second to close the secondary palate at around 12 months of age. Depending on the cleft center, caregivers (CGs) may have the opportunity to choose whether their child undergoes traditional surgical cleft treatment alone or in combination with nasoalveolar molding (NAM). NAM is a nonsurgical therapy that uses molding plates to reduce cleft size and reshape the infant's nose prior to lip surgery (Grayson & Maull, 2004).

Having a child with a chronic condition like CLP is stressful for parents (Cousino & Hazen, 2013). There are a myriad of infant issues (e.g., feeding, smiling, vocalizing, etc.) that are affected by early cleft care (Endriga & Speltz, 1997). Coping with diagnosis, reorganizing the family structure, choosing treatment options and site, as well as managing hospitalizations and changes in the course of illness can be difficult (Meleski, 2002). Parents are responsible for everyday caretaking as well as working with healthcare professionals to coordinate their child's treatment. This increased caretaking burden is reportedly associated with higher levels of depression and anxiety (Hauenstein, 1990).

Yet, some evidence suggests that having a child with a chronic condition can also foster resilience and positive adjustment in parents (Baker, Owens, Stern, & Willmot, 2009). A recent study found that CGs whose infants were undergoing NAM overcame initial anxieties and experienced empowerment, improved self-efficacy, and positive identity construction (Sischo, Broder, & Phillips, 2014). Indeed, a growing movement in health services research has identified the importance of positive psychology for understanding the psychosocial adjustment in families impacted by chronic conditions (Lin, Rong, & Lee, 2013). Moreover, such parental adjustment has been shown to influence the future well-being of the child (Speltz, Morton, Goodell, & Clarren, 1993; Wallander & Varni, 1998) who is reportedly at risk for reduced quality of life during the school-aged years (Broder, Wilson-Genderson, Sischo, & Norman, 2014).

The Family Adjustment and Adaptation Response (FAAR) Model provides a framework to explain how parents cope with having a child with a chronic condition and why some fare better than others (Patterson, 1988). According to the model, family adjustment and adaptation depend on how families balance demands with capabilities, including psychosocial resources and coping behaviors. When families experience a state of crisis (i.e., when demands significantly exceed capabilities), they respond in ways that either foster (resilience) or weaken (vulnerability) family functioning (Patterson, 1988). The birth and care of a child with CLP can be such a crisis.

It is critical to understand psychosocial trajectories in family functioning during early cleft treatment as well as risk (e.g., depression, anxiety) and protective factors (positive coping, optimism) that can influence CG psychological well-being and family/child functioning. Yet, relatively little is known about family adaptation to treatment for infants with CLP. NAM is a relatively new procedure in cleft care that, despite being considered controversial and burdensome by some teams, is now practiced at about one-third of the large cleft centers (Sischo et al., 2012). No study to date has compared caregiver responses to traditional care and NAM. Given this paucity of research, a mixed method design is particularly suited to gather in-depth insights into CGs' responses and behavior (qualitative) while also identifying patterns of psychosocial adjustment such as depression, anxiety, and optimism (quantitative) relative to early cleft care (Harris & Brown, 2010). This study's research questions include: 1) Are there treatment group differences in CGs' psychosocial adjustment over time?, 2) How do CGs cope with their infant's cleft and cleft treatment during the first year of life?, and 3) How do resources and demands affect family adaptation among CGs of infants with cleft over time?

Methods

Treatment Groups

Traditional Cleft Treatment Only—Traditional cleft treatment refers to surgery to close a child's primary palate (lip) at ~three months of age and later secondary palate surgery at ~one year of age. Children with CLP undergoing traditional cleft treatment generally see the cleft team five or six times during early cleft care: before birth (with prenatal diagnosis), soon after birth, and before and after lip and palate surgeries or if CGs experience cleft-related issues (e.g., feeding).

Nasoalveolar Molding (NAM) and Traditional Cleft Treatment—NAM treatment, which starts at about one month of age and lasts for approximately 3–5 months (depending on cleft type and severity), uses surgical tape, an intraoral molding plate, and nasal stents to improve cosmetic and functional outcomes (Grayson & Maull, 2004). CG(s) are required to make daily adjustments to the tape on the infant's face as well as attend substantially more weekly or biweekly clinic appointments (approximately 20–24 appointments prior to lip surgery) than traditional treatment alone. NAM is followed by traditional early cleft surgeries (i.e., lip and palate repairs).

Participants/Procedures

CGs seeking treatment for infants with either cleft lip only (CLO) or CLP were recruited at six geographically diverse cleft treatment centers. As per the IRB-approved protocol, after CGs met with the teams' plastic or reconstructive surgeons to discuss treatment options, they were given a study information letter.

After deciding on treatment, the principal CG (mother or father) provided informed consent. Inclusion criteria included CGs having infants <seven weeks old who had non-syndromic CLO or CLP and no other major medical issues. CGs with major psychiatric disorders were

excluded. CGs were required to speak and understand English or Spanish. Interpreters were used as needed, as were readers for CGs with low literacy proficiency.

The sample included 118 CGs who ranged in age from 17.4 to 45.5 years of age (mean=30.3, sd=6.2). Mothers comprised about 90% of the sample. The overall enrollment rate was approximately 81%. Time constraints and privacy were the two most common reasons for enrollment refusal. The attrition rate was 4%. Eight participants switched from NAM to traditional care prior to visit two (lip surgery) and were analyzed as part of the traditional care group. Data were collected for most participants at three time points in coordination with the infants' regular clinic visits: 1) one week after *NAM insertion* (or ~six weeks of age); 2) at the *pre-surgical evaluation* prior to lip surgery (at 3–5 months); and 3) at the palate surgery *post-operational appointment* for infants with CLP or follow-up clinic visit for infants with CLO (at ~13 months). Two sites were later added that only perform traditional care to ensure an adequate number of these participants. Data were collected at only two time points for these sites: 1) at the pre-surgical evaluation prior to lip surgery, and 2) at the palate surgery post-operational appointment or follow-up clinic visit for infants with CLO. At each observation, CGs completed standardized questionnaires and a semi-structured interview with an experienced interviewer that averaged 45 minutes. They received a \$50 gift card at each visit for their time and effort.

Design

A mixed methods design was utilized in this study because it capitalized on qualitative methodology's ability to address relatively unexplored areas of early cleft treatment and quantitative methodology's use of standard assessments to identify risk/protective factors and family impact of treatment (Padgett, 2012).

Qualitative data collection procedures—Semi-structured face-to-face interviews between the team's trained mental health professional and the CG addressed various factors relevant to pediatric conditions and care (e.g., decision-making, social support, contextual factors in families) based on the pediatric chronic health literature. Questions were open-ended to examine the family impact of cleft habilitation and pediatric chronic conditions. In accordance with grounded theory, interview guides were modified during the study period based on caregiver responses and varied at each data collection point. Interviews were audio recorded and transcribed with all identifiers removed.

Quantitative data collection measures—Standard psychosocial assessment tools and demographic variables were used to address research questions relating to group differences in psychosocial functioning. The Patient Health Questionnaire (PHQ-9) (Martin, Rief, Klaiberg, & Braehler, 2006) and Generalized Anxiety and Depressive symptom scales (GAD-7) (Lowe et al., 2008) were used to measure CGs' anxiety and depression. The Parenting Stress Index (PSI)-short form (36 items), a well-validated measure, was used to assess parent-child relationships and child and parent characteristics (Abidin, 1995). The 10-item revised Life Orientation Test (LOT-R) assessed dispositional optimism by focusing on expectations of good versus bad outcomes (Scheier, Carver, & Bridges, 1994). The Family Environment Scale (FES) (Moos & Moos, 2002) assessed family cohesion among CGs and

has been used in studies examining families with cleft conditions (Crerand et al., 2014; Wyszynski, Perandones, Yannibelli, & Bennun, 2005). The FES consists of 27 items that assess three domains: family cohesion (commitment and support between members), expressiveness (talking and expressing feelings), and conflict (amount of anger, fighting, etc.). The 45-item Coping Health Inventory for Parents (CHIP) measures three main coping patterns in CGs of children with chronic conditions (McCubbin et al., 1983). The coping subscales include: family (maintaining family integration, cooperation, and optimism), social (maintaining social support, self-esteem, and psychological stability), and medical (understanding the medical situation through communication with other parents and medical staff). While higher scores on the PHQ9, GAD7, and PSI indicate worse outcomes, higher scores on the LOT-R, FES and CHIP reference positive outcomes. The only exception is that the conflict subscale of the FES is specified so that higher scores indicate worse conflict, though this subscale was reoriented for inclusion in the FES total score. Other potential covariates (e.g., age and sex of CGs, child's age, cleft type, birth order, etc.) were also recorded (Simon et al., 2010). Instruments took ~15 minutes to complete and were administered after the qualitative interview.

Data analysis

Qualitative analyses were informed by Burawoy's (2009) "extended case method," which allows for examining wider social processes involved in each case. Moving from a specific situation (cleft treatment) to a general process (parental and family adjustment) is reliant on theory. Thus, theory guided the analysis by informing interview questions, coding, and memo writing (see below). Coding was a two-step process: initial coding was open to analytic ideas, while later coding was more focused on important theoretical categories (Charmaz, 2006). To provide inter-rater consistency in interview coding, two investigators independently reviewed and coded randomly selected transcripts and met regularly to discuss codes, achieve consensus, and resolve differences. This analytical triangulation improved the confirmability and credibility of the findings (Graneheim & Lundman, 2004; Patton, 1999).

As investigators coded interviews, they also began writing theoretical memos (Charmaz, 2006). The memos were used to reflect on and develop emerging concepts and themes in ways that moved the analysis from individual CG experiences to larger themes related to family functioning, parental stress, and coping in relation to caring for a child with cleft and cleft treatment. Writing theoretical memos and making changes to the interview guide increased dependability (Graneheim & Lundman, 2004). Theoretical saturation was achieved when gathering new data no longer revealed new theoretical insights nor new aspects of theoretical categories (Charmaz, 2006). *Atlas.ti* was used to aid data analysis (ATLAS.ti, 2012).

Quantitative analyses used longitudinal multi-level modeling (MLM) to examine change over time. MLM allows assessment of within-person changes (Hoffman, Hofer, & Sliwinski, 2011) while accounting for repeat measurement bias, and is robust to data that are missing at random (Rabe-Hesketh & Skrondal, 2008). The outcomes used represent count-data, thus following a Poisson process. Because of the small sample size, generalized MLM was used

to model the count data using a Poisson distribution and a log-linear modeling approach (Cameron & Trivedi, 2013). Depression, anxiety, PSI and PSI subscales, FES and FES subscales, and the LOT-R and its optimism subscale had skewness and kurtosis supporting the use of generalized MLM for these data (not shown).

Time was specified using child's age in years at each visit. Psychosocial risk and protective factors (e.g., coping, anxiety, depression) were examined at baseline and over time to identify possible treatment group differences. To account for individual differences in CGs' psychosocial status, individual-specific intercepts and random slopes were estimated over time, and an unstructured covariance matrix was assumed to account for regression to the mean. Risk ratios (RRs), 95% Confidence Intervals (95% CIs), and exact p-values derived from two-tailed significance tests were provided. Model fit was assessed using Pseudo-R². Analyses were done using Stata 13.1/IC (StataCorp, 2013).

Mixed methods analyses involved examining both the qualitative and quantitative data and assessing the research questions (Creswell & Clark, 2011). A convergent design was used that merged the two types of data to compare results. However, the qualitative data were given priority over the quantitative data given the nature of the research questions (Brennan, Hugh-Jones, & Aldridge, 2013).

Results

Data were collected at three time points: 1) at NAM insertion or when the infants were ~1.5 (SD=0.72) months old; 2) at the pre-surgical visit at ~4.9 (SD=1.80) months; and 3) at the post-surgical visit at ~13.16 (SD=3.01) months. CG characteristics at first visit (Table 1) show that most CGs were women and that treatment was almost always covered by insurance. More children were treated with NAM and approximately 75% had CLP. Average scores for each scale and averages derived from published results are presented in Table 1. These comparisons show that CGs had, on average, elevated levels of anxiety (GAD7), but lower average levels of parental stress (PSI), reported less difficult children (PSI-DC), greater expressiveness (FES) and less conflict (FES), as well as better scores on CHIP subscales.

Analyses of the associations between covariates and treatment type (Table 2) suggest that CLP, insurance coverage, and birth order were associated with treatment type. Those with CLP versus CLO were more likely to be treated with NAM and those having traditional care were more likely to have their treatment covered by insurance. Finally, those children who were third or higher in birth order were more likely to receive traditional treatment. Results are presented based on components of the FAAR model: demands, capabilities, and family adaptation.

Demands

Based on interview data, CGs of infants with cleft reported experiencing stressful situations and life events that caused negative affect, including depression and anxiety. Overwhelmingly, CGs identified two events (or "demands") in their child's early cleft treatment as being particularly stressful: 1) onset of NAM treatment (for those whose

children had NAM) and 2) the surgery to repair their children's lips. CGs' experiences at each of these events are described, followed by quantitative findings and trends in psychosocial risk factors over time.

NAM Treatment—CGs often felt uncertain and anxious about their role at the beginning of NAM treatment. CGs explained that NAM onset was very stressful, stating: “Actually learning all the little mechanics of it, putting it on, and taping it to his face and holding him down while I’m having to do it and he’s screaming at the time [was stressful]...The first month was horrible” [1–010].

Mastery was an important qualitative theme—as CGs began to master the tasks associated with NAM, their stress generally decreased over time. As one caregiver explained: “[My stress with NAM decreased] definitely after the first week. Just familiarity and comfort in doing it. It wasn’t as foreign. It was a little less scary in that regard...[and] I became less nervous” [2–029]. Parental stress also decreased once they saw improvement in their child’s cleft, which made them feel like NAM was working and the burden of care associated with NAM was “worth it.”

Lip Surgery—Facing the children’s initial surgery at 3–5 months of age was an emotionally complicated experience for CGs in both treatment groups—many experienced a range of positive and negative feelings about their child’s lip surgery. In relation to the latter, most CGs felt anxious about this surgery, including the risks (particularly anesthesia), the appearance outcome and its emotional impact, and caring for their child post-surgery. Risks associated with anesthesia were especially daunting for CGs. As one traditional care CG explained:

“[I’m feeling] a little anxiety, just about him—and not so much about his lip, but just, he’s a tiny, little, three-month-old baby, and the idea of him going under anesthesia and having surgery makes me just a little nervous and makes me feel sad for him.” [5–003]

CGs also expressed nervousness about caring for children after surgery, particularly pain management and feeding problems. This NAM CG’s sentiments were typical of both treatment groups: “[The lip surgery is] stressful. I’m thinking we’re just getting to a really good place, and he’s feeding really good, finally...We’re at such a good place that now I’m just terrified of what’s to follow the surgery.” [1–002]

Finally, concerns about the surgical outcome also weighed on the CGs’ minds. As one NAM CG stated, “You want everything to be perfect, and you’re worried that maybe it’s not going to be as perfect as you think” [2–004]. Some CGs were also apprehensive about their children’s new appearance since they had grown accustomed to their children’s appearance with the cleft. Thus, having their children’s cleft corrected was a bittersweet experience for some CGs.

Not all CGs experienced negative affect at their children’s primary surgery: some experienced anticipatory excitement. For NAM CGs, surgery signaled the end of NAM, which meant children no longer had to wear the appliance and the caregiving burden would

decrease. According to one CG, “I think I’m more excited about him not wearing the NAM and all that tape on his face anymore” [1–009]. Virtually all CGs were eager to put the lip surgery behind them. As one traditional CG stated, “I just wanted [the lip surgery] done...I want us all to really focus on just [my son] and his development as a baby and not just his cleft” [1–018].

To determine if differences in psychosocial adjustment existed between treatment groups, quantitative results indicated that CGs in NAM and traditional groups had similar levels of anxiety and depressive symptoms at baseline (Table 3, Models 1 & 2). To clarify, temporal estimates of anxiety and depressive symptoms are provided in Figure 1. These exhibit divergence in trends over time as CGs in the NAM treatment group experienced more rapid declines in the likelihood of anxiety and depressive symptoms between the first and second visits coupled with more rapid declines in the risk of anxiety and depressive symptoms over time when compared to those in the traditional care group. This decline in anxiety for NAM CGs supports similar qualitative findings for this group over time. The groups did not differ in stress levels at baseline, and both groups’ stress decreased over time, though the NAM group reported lower levels of stress over time (Model 3). Results were similar in subscale analyses.

Capabilities/Coping

CGs from both treatment groups dealt similarly with the stressful events in their lives, and common strategies included: being optimistic, engaging in problem solving, talking to others about cleft, and meeting with the cleft team. Generally CGs tried to stay positive about their situation, stating, for example, “the most important thing is just to keep a positive attitude and see him like this is something natural, and then after the surgery, he is gonna be a perfect baby, and he won’t have any disabilities” [1–503]. Some CGs also put their child’s cleft into perspective by comparing it to other, often life-threatening, chronic conditions. Unlike cancer or Down’s syndrome, for example, CGs asserted that CLP is just a “cosmetic defect” without any long-term effect on their child’s life. One NAM CG explained, “[I feel happy] just knowing that [his cleft] could be fixable, is correctable, and nothing else is wrong with him” [1–007].

Despite CGs’ optimism about their children’s early cleft treatment, many also identified concerns about their child’s future, including anxiety about bullying, teasing, cost of care, more surgeries, speech problems, appearance, and social functioning. In fact, 83% of NAM and 50% of traditional CGs expressed one or more such concerns, which suggests that many recognized the reality that issues and concerns about cleft and cleft treatment will likely arise in the future.

CGs also used problem solving as a coping strategy by learning or researching about cleft and cleft treatment. Since most CGs knew little about cleft before their child’s diagnosis, most did their own research into cleft, its causes, and treatment. Learning about CLP often helped CGs cope with their child’s condition. A traditional CG explained: “We were just prepared. We did the research before we went into any of this and we felt good overall” [3–015]. Other than doctors and cleft teams, the Internet was the most common source of information for CGs.

The final two coping strategies reveal the importance of social support and medical coping for facilitating their cleft-related challenges. As a NAM CG explained:

“It’s important for [CGs] going through this to have somebody to talk to who’s been through it because it’s not black and white. It’s not that he has a cleft lip and palate, and it’s gonna be fixed. There’s so much that goes on in the middle that people don’t realize.” [1–002]

Finding sources of social support was an emerging qualitative theme. When confronted with stressful events, CGs turned to significant others, family members, and close friends for support. Families of children with cleft were also a valuable source of support for many CGs. This finding was especially true for NAM CGs who often forged friendships in the waiting room during their weekly or biweekly appointments. As one NAM CG explained: “Making friends, having that weekly Monday appointment was a God send as far as the support system goes because being in the waiting room, you meet other families that are going through the same thing” [3–014].

While all CGs generally felt supported by the cleft teams, NAM CGs often felt closer to cleft team members than did traditional CGs. Coming in for weekly appointments made some CGs consider the cleft team as “extended family.” A NAM CG explained the special relationship she found with the team this way, “[The cleft team is] awesome...Once you come in, you’re like *[sigh]*. Everything’s great. Everyone understands. You don’t have to explain anything when they’re here. Yeah, it’s very comfortable dealing with the team that’s here” [1–011].

CGs’ reliance on social support occasionally varied over time, however. For example, some CGs were initially unable to talk about their child’s cleft. This NAM CG explained:

“Probably at the beginning I was trying to just bear it all myself, I guess. So after a little while it’s like, okay. I kinda need—a release, I guess—instead of keeping everything bottled up—kind of showing that vulnerability, like, this is hard.” [4–002]

Conversely, others relied on social support at the beginning of cleft treatment. For example, “When you’ve got more craziness going on you need to vent more than when everything kind of settles down. At the beginning of the process I needed more support than I do now.” [2–025] NAM CGs often used NAM as a problem-focused coping strategy. While initially CGs experienced anxiety and stress, as they continued through the NAM process, they often reported positive feelings, including self-efficacy, for successfully completing NAM. Participating in their child’s cleft treatment was often an empowering experience that helped them construct a positive identity and cope with their child’s cleft. Indeed, empowerment emerged as an important theme for NAM CGs. The qualitative findings addressed how CGs coped with their infants’ cleft and cleft treatment.

Treatment groups did not differ in baseline levels of life orientation, or with changes in LOT-R over time (Model 4). Results were similar in optimism and pessimism subscales (not shown). While there was no difference in baseline coping, CGs in the traditional treatment group reported an increase in likelihood in difficulty coping at the primary surgical visit

(Model 5) that were not evident for CGs in the NAM group (derived $RR = 0.90 \times 1.14 = 1.028$, 95% CI [0.97, 1.09], $p=0.385$). Analyses of family, social, and medical coping subscales show similar patterns but with less statistical power (Table 4). Specifically, we found slight overall declines in family coping over time, but found further that the NAM treatment group reported significantly improved coping over time and at pre-surgical visits in comparison to CGs in the traditional treatment group (Table 4; Model 1). While no results were evident for social coping (Model 2), results do show that NAM was associated with marginal improvements in medical coping over time in comparison to CGs in the traditional care group (Model 3). While these data support qualitative findings that CGs from both groups engaged in coping behaviors, they also show some CGs were more successful in using these strategies than others.

Family Environment/Adaptation

Family impact and adaptation were important themes in the qualitative analyses. Some CGs indicated that cleft and early cleft treatment had a negative impact on their family functioning. As one CG stated, “[Not having my husband involved] was really hard at times because I expected to have that person to just talk to and be understanding. He really didn’t even wanna talk” [5–002]. The CG believed her son’s cleft contributed to her marital problems. However, other CGs claimed their child’s cleft had either no effect or a positive effect on their family functioning. For these CGs, cleft treatment-related stress strengthened their familial bonds. For example, one NAM CG said: “[Cleft treatment] hasn’t really...even though you do get stressed with each other sometimes, but it’s not, it hasn’t really changed us. Almost in a way made us closer too.” [1–002].

Quantitative analyses show that, at baseline (a particularly stressful time for NAM CGs), NAM CGs had poorer family functioning (Table 3, Model 6). According to subscale results for the FES (significant results provided), expressiveness was poorer among NAM CGs ($RR=0.64$ [0.44, 0.93], $p=0.021$). Similarly, NAM CGs reported worse baseline cohesion (NAM treatment, $RR=0.45$ [0.22, 0.92], $p=0.028$), but differences were reduced at the primary surgical visit (NAM*Pre-surgical visit, $RR=2.13$ [1.12, 4.00], $p=0.020$). No significant results were found for the conflict subscale. These analyses demonstrate how resources and demands can impact family adaptation over time. At a time of high stress (baseline/NAM onset), NAM CGs had lower family functioning. However, over time their stress decreased and their family functioning improved.

Discussion

In our mixed model longitudinal study of CG responses to early cleft care, treatment group differences were observed. Despite the greater burden associated with NAM therapy, NAM CGs experienced better psychosocial outcomes than traditional CGs. Specifically, NAM CGs expressed more rapid declines in depressive symptoms and anxiety than traditional CGs. While qualitative analyses found that most CGs engaged in common coping strategies, quantitative analyses identified group differences in coping ability, particularly medical coping. This difference is likely linked with NAM CGs’ increased contact with other families with children with cleft and the cleft team as a result of their weekly appointments

(Barbarin & Chesler, 1986; Kerr & McIntosh, 2000). Indeed, higher levels of anxiety in the traditional care group at the primary surgical visit was observed, yet decreases in anxiety throughout the first year of life among NAM CGs were found, which may reflect greater team interaction and increased trust. It is unknown whether these coping differences continue beyond early cleft treatment and if the positive effect of increased team exposure for NAM CGs continues to be protective over time. Since children with cleft are evaluated yearly and additional surgeries are likely during childhood and adolescence, positive medical coping is key.

Family functioning and adaptation were indicative of some CGs' ability to balance the demands of early cleft care with their capabilities. At baseline when demands were high, NAM CGs had elevated levels of stress and lower levels of family functioning. Over time, however, their stress decreased and their family functioning improved, which is likely due to coping strategies, including the ability to actively participate in their child's treatment, a strategy not available for the traditional care CGs. These results supported the Family Adjustment and Adaptation Response (FAAR) Model by showing that positive family adaptation can occur when capabilities and resources outweigh demands (Patterson, 1988). The findings are similar to other studies of parents with chronically ill children that have used the FAAR model, including studies of parents of children with cancer (Miedema, Hamilton, Fortin, Easley, & Matthews, 2010) and autism and communication disorders (Bristol, 1987). However, this study is the first to examine the model using a mixed methods analysis. The present study shows that the FAAR model can also be applied to family adjustment to having an infant with cleft and cleft treatment and underscores the importance of examining positive factors in health psychology and sociomedical research.

CGs' ability to cope with demands may be important as children grow and continue the CLP treatment process, which includes team evaluations and the likelihood of future surgery(ies). While their children's initial surgeries are behind them, most CGs expressed concerns about the future. Yet group differences also existed, with more NAM CGs expressing such future concerns than traditional CGs, which may reflect group differences in medical knowledge. It is unknown whether coping strategies CGs used during early cleft care will be effective as children develop and families experience new demands related to the child's CLP. The psychosocial well-being of parents is deemed critical since the family directly affects the treatment decisions, adherence, and clinical outcomes for children with cleft (Paynter, Jordan, & Finch, 1990; Zeytinoglu & Davey, 2012). Indeed, families can act as integral members of the CLP team (Pannbacker & Scheuerle, 1993).

While innovative, this study is based on a convenience sample that may not represent all CGs whose infants undergo early cleft care. As such, this study warrants replication. Second, the sample size is relatively small for quantitative analyses, limiting statistical power. Although the analytic methods are robust with respect to data missing at random, the effect on the results of including two traditional care only centers with a shortened data collection protocol (pre lip and post palate surgery) is unknown. Lastly, while the study was prospective, CGs' mental health prior to their children's diagnosis is unknown.

This study highlights the benefits of using mixed methodology in health psychology research. The qualitative data fostered in-depth understanding about CGs' subjective responses to having a child with cleft and cleft treatment, and the standardized survey instruments identified trends among CGs in relation to risk and protective factors and the family impact of cleft treatment using MLM. Although treatment group differences were found, longitudinal research is needed on this CG population given the scant data available on family impact and preschool children's quality of life. Providing supportive counseling services, in particular around hospitalizations, may be useful for families.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgment

This research was supported by the National Institute of Dental and Craniofacial Research (grant DE021853), Hillary L. Broder, PI. The caregivers' participation is greatly appreciated. We also thank our research collaborators, Drs. John Riski from Children's Healthcare of Atlanta; Jeffrey Marsh from Mercy Children's Hospital, Judah Garfinkle from Oregon Health & Science University; Barry Grayson from New York University Langone Medical Center; Roberto Flores from Riley Hospital for Children, as well as the teams' interviewers (Ms. Allison Pelletieri, Dr. Mina Nguyen-Driver, and Dr. Margot Stein) and clinical and research staff for facilitating data collection and their dedication to patient care.

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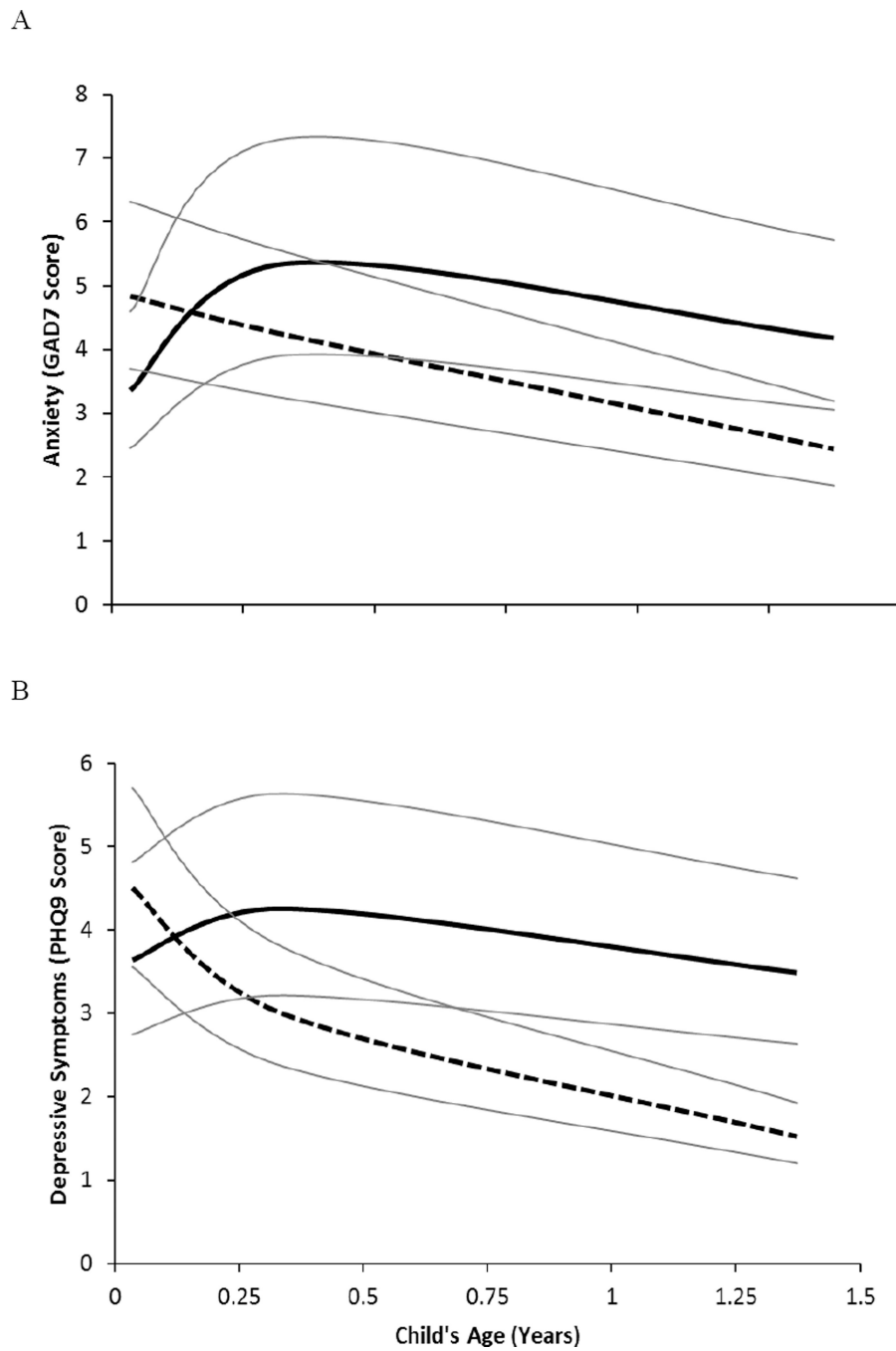


Figure 1. Non-linear association between child's age at each visit and caregiver anxiety separated into NAM (dashed lines) and traditional (solid lines) treatment. 95% confidence intervals are provided (gray lines). Panel A provides results for anxiety, panel B for depression.

Table 1

Sample characteristics among CGs of children with CLP (N=118)

	N (%)
Male CGs	11 (9.7)
Father employed	106 (90.1)
Mother employed	60 (50.5)
Traditional treatment	53 (45.1)
Insurance covers treatment	107 (90.5)
Birth Order First	53 (45.0)
Second	29 (24.8)
Third or higher	36 (30.3)
Cleft Lip and Palate	88 (74.3)
Bilateral Cleft Lip	26 (22.1)

Subscales	Mean	SD	Published Norms			
			Mean	SD	P	Citation
Caregiver age	30.3	6.2				
Depressive Symptoms	3.6	3.4	3.6	4.1	0.919	Rief et al. ¹
Anxiety	4.2	4.1	3.0	4.0	0.002	Lowe et al. ²
Parenting Stress (PSI)	67.2	19.6	73.4	25.6	0.026	Reitman, Currier, & Stickle ³
Parental Distress	24.6	8.5	24.7	9.1	0.938	Reitman, Currier, & Stickle
Difficult child	16.2	5.7	26.6	9.7	<0.001	Reitman, Currier, & Stickle
Parent-Child	22.4	7.2	22.2	8.9	0.836	Reitman, Currier, & Stickle
Family Environment (FES)	4.4	3.3				
Cohesion	8.1	1.3	8.0	1.3	0.435	Crerand et al. ⁴
Expressiveness	6.5	1.6	6.2	1.7	0.016	Crerand et al.
Conflict	1.2	1.4	2.2	1.8	<0.001	Crerand et al.
Coping Health Inventory for Parents (CHIP)	108.3	16.4				
Family	46.4	6.3	40.0	15.0	<0.001	Chip training manual ⁵
Social	44.2	7.8	28.0	12.0	<0.001	Chip training manual
Medical	17.7	3.7	15.0	7.0	<0.001	Chip training manual
Life orientation (LOT-R)	16.3	4.1	15.6	3.9	0.040	Glaesmer et al. ⁶
Optimism	8.6	2.2	8.8	2.2	0.363	Glaesmer et al.
Pessimism	7.7	2.6	5.2	2.8	<0.001	Glaesmer et al.

¹(Rief, Nanke, Klaiberg, & Braehler, 2004)²(Lowe et al., 2008)³(Reitman, Currier, & Stickle, 2002)⁴(Crerand et al., 2014)⁵(University of Wisconsin & Maccubbin, 1987)⁶(Glaesmer et al., 2012)

Table 2

Predictors of traditional care versus NAM treatment groups at CG's first visit.

	OR	95% C.I.	P
Male caregiver	0.64	0.13, 3.27	0.596
Father employed	1.15	0.25, 5.27	0.860
Mother employed	1.05	0.38, 2.91	0.931
Caregiver age	0.94	0.86, 1.03	0.191
Birth Order Second	1.10	0.34, 3.5	0.874
Third or higher	5.58	1.35, 23	0.017
Insurance covers treatment	30.97	1.97, 487.25	0.015
Cleft Lip and Palate	0.09	0.02, 0.35	0.001
Bilateral Cleft Lip	2.26	0.68, 7.48	0.181
Pseudo-R ²		0.22	<0.001

Note: Analyses adjusted for child's age.

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Table 3

Longitudinal associations between treatment type and caregivers' mental health.

	Model 1: Depressive Symptoms			Model 2: Anxiety			Model 3: Stress			Model 4: Life Orientation			Model 5: Coping			Model 6: Family Functioning		
	RR	95% C.I.	P	RR	95% C.I.	P	RR	95% C.I.	P	RR	95% C.I.	P	RR	95% C.I.	P	RR	95% C.I.	P
Traditional txt	Reference			Reference			Reference			Reference			Reference			Reference		
NAM txt	1.27	0.78, 2.09	0.34	1.47	0.86, 2.52	0.16	0.92	0.61, 1.39	0.691	0.86	0.65, 1.13	0.273	0.96	0.69, 1.32	0.780	0.74	0.54, 1.00	0.053
Child Age	0.97	0.59, 1.60	0.903	1.18	0.70, 1.98	0.54	0.71	0.47, 1.08	0.112	0.91	0.74, 1.11	0.352	0.77	0.60, 0.98	0.033	0.88	0.68, 1.15	0.351
Traditional txt	Reference			Reference			Reference			Reference			Reference			Reference		
NAM txt x Child Age	0.46	0.25, 0.85	0.014	0.51	0.27, 0.96	0.04	1.10	0.65, 1.87	0.722	1.05	0.82, 1.34	0.710	1.51	1.09, 2.09	0.014	1.36	0.98, 1.88	0.069
Pre-surg Visit	1.18	0.94, 1.49	0.156	1.52	1.21, 1.89	<0.001	0.88	0.78, 0.98	0.019	0.96	0.82, 1.12	0.599	0.9	0.83, 0.99	0.026	1.02	0.82, 1.26	0.875
Traditional txt	Reference			Reference			Reference			Reference			Reference			Reference		
NAM x Pre-surg Visit	0.72	0.53, 0.97	0.031	0.67	0.51, 0.89	0.005	1.01	0.89, 1.15	0.897	1.12	0.92, 1.36	0.260	1.14	1.02, 1.26	0.02	1.18	0.9, 1.55	0.243
Pseudo-R ²		0.24	<0.001		0.27	<0.001		0.51	<0.001		0.11	<0.001		0.44	<0.001		0.16	0.003

Note: It is assumed that outcomes are distributed Poisson. Child age is measured in years at each visit. Models incorporate random intercepts and slopes and estimate an unstructured covariance matrix. For models 1–3, higher numbers reference worse outcomes and thus an RR over 1 would be interpreted as increasing the risk of poor mental health, while for models 4–6, higher scores are indicative of better outcomes and therefore an RR over 1 should be interpreted as providing support for improved psychosocial health.

Txt = Treatment
 Child Age = Child Age in Years
 Surg = Surgical

Table 4
Longitudinal associations between treatment type and CGs' coping strategies, by coping subscales.

	Model 1: Family Coping			Model 2: Social Coping			Model 3: Medical Coping		
	RR	95% C.I.	P	RR	95% C.I.	P	RR	95% C.I.	P
Traditional treatment		Reference			Reference			Reference	
NAM treatment	0.89	0.66, 1.20	0.462	0.93	0.68, 1.26	0.622	0.90	0.68, 1.19	0.459
Years	0.80	0.62, 1.04	0.098	0.95	0.74, 1.22	0.705	0.87	0.70, 1.08	0.209
Traditional treatment		Reference			Reference			Reference	
NAM treatment x Ch									
Years	1.39	1.02, 1.89	0.039	1.20	0.90, 1.61	0.219	1.29	0.99, 1.68	0.062
Pre-surgical Visit	0.90	0.79, 1.02	0.110	0.91	0.80, 1.03	0.124	0.91	0.77, 1.07	0.272
Traditional treatment		Reference			Reference			Reference	
NAM x Pre-surgical Visit	1.20	1.02, 1.42	0.028	1.11	0.95, 1.30	0.172	1.15	0.93, 1.41	0.203
Pseudo-R ²		0.24	<0.001		0.27	<0.001		0.11	<0.001

Note: It is assumed that outcomes are distributed Poisson. Years measures child age in years. Models incorporate random intercepts and slopes and estimate an unstructured covariance matrix. For all outcomes, higher scores are indicative of better outcomes and therefore an RR over 1 should be interpreted as providing support for improved psychosocial health.

Ch Yrs = Child Years