**Title:** *“I can't provide what my child needs”*: Early feeding experiences of caregivers of children with craniofacial microsomia.

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**Highlights:**

1. CFM-related feeding challenges had a negative impact on caregiver wellbeing.
2. Some health providers promoted breastfeeding, regardless of the infant’s ability.
3. Many caregivers felt unsupported and lacked reliable feeding information
4. Support should be sensitive, timely, and tailored to suit each family’s needs.

**Abstract**

**Purpose:** Craniofacial microsomia (CFM) is a congenital condition that can be

associated with feeding challenges in infants. As part of the larger ‘Craniofacial

microsomia: Accelerating Research and Education (CARE)’ program, this study

described caregivers’ early feeding experiences.

**Design and Materials:** US-based caregivers of children with CFM (n=34) participated in

remote narrative interviews. Two authors completed inductive thematic analysis in an

iterative process until consensus was reached.

**Results:** Caregivers’ narratives outlined the inherent challenges of feeding an infant

with special healthcare needs. The first theme ‘Navigating Challenges and Managing

Expectations’ describes the distress participants experienced when they were unable

to breastfeed and the negative emotional effect of switching to formula. The second theme

‘Making Adaptations’ outlines the methods participants tried, including breast pumps

and feeding tubes. The third theme ‘Accessing Support’ describes participants’

interactions with healthcare providers and challenges accessing feeding support.

The final theme ‘Growing from Adversity’ recounts participants’ relief once their child

established a feeding pattern and the personal growth gained from their experiences.

**Conclusions:** Caregivers reported several feeding related challenges associated with

CFM, many of which negatively affected their wellbeing. Negative consequences were

particularly pronounced in cases where caregivers’ feeding experiences differed from

their expectations. Participants identified challenges in accessing reliable feeding

information and support. Despite difficult experiences, caregivers cited some positive

outcomes, including increased confidence and resilience.

**Practice Implications:** Holistic feeding information and support for families affected by

CFM should be inclusive of several feeding methods to improve care delivery, child

health, and the caregiver experience.

# **Introduction**

 Craniofacial microsomia (CFM) is a congenital condition associated with asymmetric hypoplasia of the facial structures. CFM has wide clinical variability with prevalence estimates between 1:6,000 and 1:40,000 live births (Barisic et al., 2014; Junaid et al., 2022) and the etiology is unknown for most children. The most common feature associated with craniofacial microsomia (CFM) is microtia (i.e., underdevelopment of external ear). Other features of CFM include mandibular hypoplasia, lateral oral clefts, cleft lip and palate, and facial nerve palsy (Birgfeld & Heike, 2019). The clinical implications of CFM are well-documented and may include difficulties with breathing, hearing, speech, and feeding (Renkema et al., 2022).

**CFM and Feeding Challenges**

 A systematic review on feeding in children with CFM identified a prevalence of feeding difficulties in 42-83% of the population (Caron et al., 2015). In a large multi-center retrospective analysis of 755 patients, 26% of patients with CFM experienced feeding difficulties, which included challenges with suckling, swallowing, chewing, and restricted mouth opening (Caron et al., 2015). Among children with CFM, feeding difficulties were more common in children with lateral oral clefts, cleft lip and/or palate, bilateral mandibular hypoplasia, facial palsy, obstructive sleep apnea, and/or extracraniofacial anomalies (Caron et al., 2018).

 Feeding difficulties can be a direct result of anatomical changes, such as a lateral oral cleft that impairs the ability of the infant to obtain an adequate seal on the breast or bottle. Other causes can be more subtle and require swallow studies to identify sources of feeding difficulties (Miller & Madhoun, 2016). Based on swallow studies, van de Lande et al. (2018) found that patients with CFM had difficulties with bolus formation across ages. Further, those under 6 months of age had problems with nasopharyngeal reflux and aspiration, while some patients over 6 months old had difficulties with swallow trigger and stasis of food after swallowing. Those with mandibular hypoplasia had more difficulties in bolus formation than those without mandibular hypoplasia.

 Mandibular hypoplasia can also be associated with upper airway obstruction and contribute indirectly to feeding difficulties associated with a compromised airway (Heike et al., 2013). In these cases, swallowing can be challenging, and there is a risk for aspiration (Pullens & Streppel, 2021). Similarly, van der Plas et al. (2022) retrospectively assessed outcomes of patients (n=22) with mandibular hypoplasia associated with a syndrome or a unifying craniofacial diagnosis (including CFM) and severe airway obstruction treated with mandibular distraction surgery. Feeding and swallowing difficulties were prevalent in the sample and 59% of the sample had ongoing feeding difficulties following surgical intervention and required a gastrostomy tube (van der Plas et al., 2022).

 Caron et al. (2018) noted that most feeding difficulties present in early infancy (<6 months) (Caron et al., 2018). The possible long-term detrimental effects on physical and psychological wellbeing associated with early feeding challenges are well documented among children in the general population (Kamity et al., 2021), including morbidities associated with infant malnutrition (Corbett & Drewett, 2004; Dewey & Mayers, 2011) and long-term risks of disordered eating (Bergonzini et al., 2022).

**The Experiences of Caregivers**

 Adjusting to their child’s diagnosis of CFM can present a range of psychosocial challenges for caregivers (Johns et al., 2024) such as eliciting emotions of anxiety, shock, grief, and worry (Luquetti et al., 2018). Additionally, caregivers must adapt to new care-related demands, including those related to feeding (Miller & Madhoun, 2016). While feeding difficulties in CFM have been described, the perspectives of caregivers are notably absent. Yet, caregivers play an imperative role in their child’s health and wellbeing and caregiver distress is an established risk factor for adverse medical and psychosocial outcomes in children (El Hamaoui et al., 2006; Helgeson et al., 2012; Klinnert et al., 2008; Noronha & Faust, 2007). Understanding of the experiences and support needs of caregivers of children with CFM in relation to their child’s feeding is a critical next step to improve care delivery and family-centered outcomes. The aim of the present study was to investigate the early feeding-related experiences of US caregivers of children with CFM.

# **Method**

**Design**

 This study is part of a larger research program entitled Craniofacial microsomia: Accelerating Research and Education (Stock et al., 2023), which aims to better understand the psychosocial effects of CFM and the treatment-related experiences of individuals and families of individuals with CFM. For the first phase of the program, caregivers participated in remote narrative interviews, a method that allowed participants to lead the interview according to the information that they deemed to be important in their lived experiences (Riessman, 2008; Ziebland, 2013). Rather than a structured or semi-structured interview design based on predetermined areas of content identified by the researchers, the narrative interview design allowed topics to be entirely based on what participants elected to discuss. In this paper, we focused on the feeding-related experiences that caregivers included as part of their full narratives.

**Study Population**

 Participants were biological English or Spanish-speaking caregivers of children aged 3 to 17 years with a diagnosis of within the CFM spectrum, including microtia, hemifacial microsomia (HFM), oculo-auriculo-vertebral syndrome (OAV), or Goldenhar syndrome. Participants were eligible if their children received care in the US and met the FACIAL research inclusion criteria, defined as having a one or more of the following craniofacial features: 1) microtia; 2) anotia; 3) facial asymmetry and preauricular tag; 4) facial asymmetry and facial tag; 5) facial asymmetry and epibulbar dermoid; 6) facial asymmetry and lateral oral cleft; 7) preauricular tag and epibulbar dermoid; 8) preauricular tag and lateral oral cleft; 9) facial tag and epibulbar dermoid; 10) lateral oral cleft and epibulbar dermoid (Luquetti, et al., 2012). Exclusion criteria included syndromes, such as Treacher-Collins, Townes-Brocks, or Nager, or a major medical condition not associated with CFM that was perceived by parents to have a substantial effect on the child’s health. Participants were recruited across the US through advertisements at pediatric craniofacial clinics, social media posts, CFM-related advocacy events, and outreach to participants in prior CFM studies. Due to the open recruitment methods, we are unable to estimate how many people chose to participate relative to the number of people who had information about the study.

**Procedure**

 Interviews were conducted between 11/23/2020 and 4/20/2023. After completing consent and providing data on sociodemographic variables, participants reported on their child’s medical history over the telephone. This call lasted approximately 90 minutes and included details of their child’s health history, specialty providers, medical treatments, surgeries, therapies, and education. Participants were then asked to reflect on their life experience with CFM and prepare for a narrative interview by dividing their CFM story into “chapters” based on the aspects of their journey that were most meaningful to them.

 Narrative interviews were completed primarily via a HIPAA-complaint videoconference platform and a few participants opted to complete the interview by telephone. Narrative interviews were completed by three females in English and two females in Spanish. Four had master’s or doctoral level training in psychology, one had master’s degree in public health, and all had experience with qualitative methods. Four interviewers were white and one was Latina. At the start of study visit, interviewers established rapport and reviewed the study aims, provided information about themselves, and explained the format (e.g., taking breaks). The duration of narrative interviews included in this study ranged from 40 minutes to three hours (mean=86 minutes). Interviewers asked questions at the completion of each chapter as needed for clarification or elaboration on aspects of their narratives. Participants were asked to consider a “high” point (a particularly positive aspect) and a “low” point (a particularly difficult aspect) of each chapter. Interviewers also asked how participants’ experiences could have been improved. Participants received a gift card to thank them for their time. Audio recordings were transcribed for analysis and identifying information was removed.

**Analysis**

 For this paper, interview content relating to feeding experiences was extracted from the larger dataset. Two white female authors with doctoral level psychology training and experience in qualitative methods and craniofacial research completed reflexive thematic analysis (Braun & Clarke, 2006; Braun & Clarke, 2019). One author had also completed interviews in English. After becoming familiar with the participant narratives through multiple readings of all the transcripts, open inductive coding was completed with frequent meetings to discuss content and refine the codes in an iterative process. Coding discussions included reflecting on the role of the researchers’ perspectives based on their prior understanding and experience with craniofacial care. Themes were described and grouped together and then shared with the larger interdisciplinary group of authors, including craniofacial healthcare providers and a parent of a child with CFM, for further discussion of the themes until consensus was met. Illustrative quotes were selected as examples of the range of experiences reported.

**Ethical Considerations**

 Ethical approval was granted by the Institutional Review Board (STUDY00002637) at the primary site and participating sites (SITE00001444 and SITE 00001366). Verbal informed consent was obtained from all participants prior to their participation in the study.

# **Results**

**Participants**

 Thirty-five parents of 34 children with CFM completed interviews that included content on feeding. Individual interviews were completed in English (76%) or Spanish (24%) with mothers (82.4%) and fathers (5.9%) with one joint interview that included a mother and father (2.9%), as shown in Table 1. Caregivers had a mean age of 39.6 years (SD = 12.3) at the time of the interview and were living in 15 US states. Participants were primarily White (64.7%) and non-Hispanic/Latinx (67.6%), married/cohabitating (73.5%), college graduates (50.0%), and employed (73.6%). As seen in Table 2, children of caregivers had a mean age of 9.8 years (SD = 4.7), over half (55.9%) were male, and most had private health insurance (55.9%). Most children had microtia with mandibular hypoplasia (91.2%), and the rest had other combinations of CFM-associated features (8.8%). According to caregiver report, newborns were discharged 18.6 days (SD = 43.7) after birth on average. Neonatal medical problems requiring monitoring or intervention were reported for 55.9% of infants in this sample, such as jaundice, feeding difficulties, and respiratory concerns.

**Themes**

While some participants reported no concerns with their child’s feeding, many families discussed a range of challenges which were the focus of the analyses. We identified the following four themes: 1) Navigating Challenges and Managing Expectations; 2) Feeding Adaptations; 3) Accessing Support; 4) Growing from Adversity. Each theme is discussed in further detail below, alongside illustrative quotes. As suggested by Hill et al the representativeness of this study’s results across participants is described by categorizing findings as general (applied to all or all but one case), typical (more than half of cases), or showing variance (less than half but more than two cases) (Hill et al., 2005). When reporting the findings, these three labels are referred to as all (general), most (typical), and some (variance). In some instances, quotes have been shortened by the authors, indicated by ‘…’, or rephrased slightly for clarity, indicated by ‘[ ]’.

**Navigating Challenges and Managing Expectations**

This theme describes participants’ early feeding challenges, which largely centered around difficulties breastfeeding. All participants reported that they (or their partner) initially attempted to breastfeed their infant, but some experienced significant difficulties. This not only affected the infant’s physical health but also caregivers’ psychological wellbeing. One mother said, “It was [possibly] a combination of [my son] not being able to nurse effectively and me not producing enough [milk], but in any case, he was starving and very skinny. Lots of crying, fitful sleep.” Another mother commented: “It was probably a day and a half of just continuous crying and trying [to breastfeed]. Crying on [my son’s] part and my part and trying to figure it out. And still he was losing weight and he wasn’t getting any milk.” This had a negative influence on participants’ wellbeing. One parent said: “We were having trouble with [my daughter] getting to latch and nobody really had told us what was going on...It was all scary stuff.”

 This was particularly challenging as prior to having their child, most participants held the belief that “breast is best” and had intended to breastfeed. As one mother stated, “This idea [that] breastfeeding is the only way you can do it... It was the end all be all in my mind”. Another mother stated “I had breastfed my other kids... I wanted to breastfeed [my son too].” Participants felt that healthcare providers also strongly enforced the “breast is best” policy and passed judgment on families that weren’t able to breastfeed. One participant related the message was communicated to her that “formula is not good, here are all these reasons... there wasn’t the thought of whatever you do, you’re feeding your child and that’s what matters.” Another mother said:

[Lactation specialists are] supposed to go in and show you how to [breastfeed] and encourage you, which is fine, but they also highly discourage the bottle and almost put you in a guilty situation if you’re bottle-feeding. And it’s like “I don’t need that right now, I have bigger issues!’”

 As a result of not being able to meet their own and/or others’ feeding expectations and/or their child’s nutritional needs, many participants experienced emotional distress. This included feeling “guilty”, “confused”, “frustrated”, and “scared”. Some participants felt they had ‘failed’ their child: *“*My baby was starving and [it was my fault].” One mother asked, “Why can’t I do this and what have I done to my child? I grew him, [but] I can’t feed him and he can’t do these things that babies are supposed to instinctively do. Why isn’t this working?” Another mother said:

 The low point for me was just realizing I couldn’t feed [my son] and realizing I was starving him. I felt really, really bad about that and it was a struggle because I felt a lot of guilt about it...My body is not providing him the nutrients he needs.

 Some participants also cited the negative effect on breastfeeding of frequent medical investigations in the postnatal period. Participants described the lack of private time to connect with their infant:

[I was] trying to learn to breastfeed, which ended up being really hard... because [the healthcare providers] kept taking [my son] to do ultrasounds of his kidneys and his spine and tested his heart and all the various things. So, he keeps getting whisked away and we just didn’t feel like we got to bond with him or be with him all that much in those first few days.

In addition, some participants spent a considerable amount of time in the hospital fixated on feeding and their child’s growth, rather than enjoying their newborn at home. As one mother stated, “The issue to get her out of the hospital became [my daughter’s] eating and gaining weight.”

Most participants had to stop breastfeeding or needed to supplement breastmilk with formula after their infant failed to thrive. This was generally upsetting and felt to participants like “giving in”. One mother said, “I remember going to the store and I bought formula. I was so, so ashamed of myself and I felt so - like a failure, because I couldn’t do what my body was supposed to do.” Another noted, “[The] moment of defeat, standing in the supermarket, holding this can of formula and thinking ‘You didn’t do it.'”

Being around other caregivers who had managed to breastfeed successfully was also upsetting for some participants. As one mother described, “The group of individuals I was around, they were all able to breastfeed... It was all very ‘pro’ breastfeeding”. Similarly, another mother commented that “The trigger of [other] moms breastfeeding, that was really hard for me for a long time. There was almost a bitterness... it would just hit that sore spot… A deep envy, like ‘I wish I could have had that.’”

For those who were able to breastfeed, participants also described the physical consequences breastfeeding had on their own bodies, including infections in the breasts and chapped/bleeding nipples, such as “my nipple was so chapped and just destroyed from trying to breast feed” and “when I did get home from the hospital my breasts got an infection in them.”

**Feeding Adaptations**

This theme describes the adaptations that most participants made to feed their child, including bottle and formula feeding, pumping, and home remedies. Some participants used nasogastric (NG) and gastrostomy (GT or G-tube) tubes. Collectively, the journey to finding a successful feeding method was described by most participants as extremely challenging: “I was doing all the things that I could (…) to make things better for [my son]. But even that was a lot too, because...everything was just so exhausting.”

Participants made many adaptations to their feeding routines and tried several different methods to feed their child. For those able to produce breast milk, this involved pumping milk from the breast. This method required considerable effort from caregivers, for example, “I was pumping around the clock – all night, all day - and my husband was trying to feed [our daughter] those bottles and we were trying to keep up so that she could eat enough.” Another mother reported that while her daughter was admitted she pumped “up to 80 bottles or more of milk every week.””

These caregivers also cited challenges related to equipment access and use. One mother reported that “The pump that I had bought hadn’t worked...so I had to go to a different hospital and get a hospital-grade pump...so that I could feed [my son] that way.” A father noted that “[My wife was] working four days a week, 10-hour shifts, lugging around a breast pump, having to pump in the bathroom, the locker room.”

 Some caregivers reported limited breast milk production:“I also had a hard time producing enough breast milk.”, and in some cases, tested home remedies: “I tried Milk Thistle, I tried everything.” One participant used social media to source breastmilk donations when their own production was no longer sufficient:

I reached out to this group of women and I asked for breast milk donations...By that evening I was able to give [my son] his first really solid meal since he was born, and it changed his whole…he became a relaxed baby for the first time.

Despite apprehensions tied to the perception of negative appraisals of formula, most participants ultimately used formula feeding. Some participants cited “needing to trial multiple formulas” to find one that worked, particularly if their child had special dietary needs: “Every tummy is so different, and it shouldn’t be a challenge to be able to get a child a soy-based formula or a non-dairy-based.” Similarly, another mother reported: “We had problems almost from birth until about two years because his body didn’t accept the nutrients and they changed his formula every so often until the last one they gave him.” For these caregivers, the financial burden of purchasing formula was also often noted, as one participant noted, “We went through...probably six different formulas...and the cost was an absolute fortune.”

Some participants described bottle-feeding as difficult, given persistent challenges with latching: “Even the bottle-feeding was just really challenging, because of [my son’s] poor suck.” Others made adaptations to feeding strategies to provide their infant with breastmilk, such as a mother who said, “We were supplementing with a syringe and feeding [our daughter] that way.”

Some participants described digestive issues, as one mother said “he was throwing up a lot, he could almost never keep any food down in his stomach.” In addition to influencing their child's health, these instances were described as stressful, embarrassing, and caused practical challenges to daily life. One participant stated, “As soon as you’d feed him, he would just basically vomit. We would probably go through about six or seven outfits a day.”

 A few participants reported the use of nasogastric and gastric feeding tubes. Generally, most participants were reluctant for their child to have a feeding tube, and for those who had to have one, were motivated to remove it as soon as possible: ”I’m always worried about him, that they’re not going to take his tube out and he’s going to eat like that all his life.” Similarly, another participant stated:

We would see some parents talking about feeding tubes, and then we also spoke with our speech therapist about it and she had heard of kids who developed oral aversions or who would have the tubes until they were teenagers, or their whole lives, and we just…we didn’t really know...we wanted a plan. We wanted this to be a temporary thing and we wanted to know that.

 Participants whose child required tube feeding reported having to learn new skills to facilitate tube feeding for their child. For example, one mother noted “The first time I tried to [change the tube] myself, [the nurse] was there, but she made me do it, and I almost passed out... I couldn’t do it... It was awful.” Similarly, another participant reported “Every time our son gagged, I gagged. I’d be almost throwing up putting the NG tube in him... and he’d cry, I’d cry, he’d gag, I’d gag. I’m like ‘I am incapable of this.’”Particularly in the early stages, feeding tubes were a source of considerable stress for participants:

[My son] ended up coming home with a g-tube which was all new to me too, and it scared the hell out of me because... I’m not a nurse! And then when they tell us that “You’re going to have to change this every three months”, [we were] like “What? I don’t know what I’m doing. You just gave me this baby that needs all these things and now this, and I don’t know what I’m doing!”

Some participants perceived feeding tube interventions to have negative effects, including their child’s wellbeing, which further contributed to negative perceptions of and anxiety about feeding tubes. For example, “Since day one really, [our daughter] has just had so much personality, so that’s why it was so hard to see her so lethargic on the feeding tube. [Before that] she was very alert and was starting to smile.”

 Participants described a range of effects that feeding tubes had on daily family life, including sleep, work, and social connections. One participant explained, *“*For a time, you feel alone, because your kid constantly needs things. And people don’t understand that you don’t have time to go out... It was a lot.” Another commented, “It took us a really long time to try to leave the house... because [my daughter] had so much stuff and I was just so worried about tube feeding her in public.” Similarly, a mother said:

The NG tube holds a flap of the stomach open and so we have to start feeding [our son] every hour on the hour, which means me and my husband are up around the clock taking turns, while we’re both trying to work.

 Participants also cited financial concerns associated with feeding tubes. One participant noted, *“*The feeding supply kit is important because we have to pay for that out of pocket... We ended up probably spending close to $2,000.” Another commented, “I think we’ve gotten pretty lucky. We have good insurance... [but] we’re on these Facebook groups where people are trying to crowdfund for a blender to feed their kid.”

 Participants described adaptations made by the wider family to accommodate tube feeding. One participant noted “[Child’s grandmother] was learning how to tube feed. So, that was good.” Participants also prepared siblings: “We just showed [our child’s siblings] this book. So, they had a picture of...what a feeding tube is. So, it wasn’t such a shock when they got in there, which was great.”

**Accessing Support**

 This theme focuses on caregivers’ broad experiences of medical care and their interactions with healthcare providers in relation to their child’s feeding. Generally, caregivers were apprehensive about medical tests and interventions: “Every single specialist that I can think of came in and it was very overwhelming as a parent.” Some participants reported not feeling listened to by healthcare providers, which caregivers described as impeding progress in their child’s treatment as well as having adverse psychological consequences they experienced. One participant said, “He was vomiting and not eating, and we felt that there [was] something more... No one was helping us.” Another commented, “Our experience of [health providers] was they weren’t hearing what we were saying and listening to what we were telling them.” Similarly, one participant said:

We kept on telling them: “There’s something wrong with him, he’s not gaining weight. We’re feeding him, we’re doing everything [you] are telling us to and he’s still not gaining weight.” So, we were really frustrated... we didn’t know what to do or say.

 Participants also reported concerns that additional medical issues were adversely affecting feeding and were missed by medical staff. One mother reported:

 [My son] would have never been able to latch to me. Breast would have never been best, unless he got a medical procedure to get his tongue and lip tie cut, which didn’t happen well until months and months later, because no-one had looked.

Another participant stated:

The [otolaryngologist] didn’t know [my daughter] had a cleft palate and her muscles weren’t suctioning properly. So, we never got an adapted bottle for her [and milk] would just go out her nose and nobody had any idea... Nobody told us anything... It was just “Do your best at home.”

 Some infants spent time in the neonatal intensive care unit (NICU). Some caregivers felt apprehensive leaving their child in the care of others, and one noted “When your child has all these machines and suction and feeding tubes you don’t trust just anyone to her care.” However, others felt reassured by the presence of the NICU staff, as one mother said about her daughter “being in the NICU for so long was hands-on caregiver training, because the nurses were able to give me so much information that I would have never known and was able to really set her up for success.” Similarly, healthcare providers who were supportive of caregivers’ needs were much appreciated by participants, such as one mother who said, “Before we left [the hospital] there was a doctor that came [and] we explained everything. I think [that doctor] was the one that was the most knowledgeable person.”

 Yet, many participants felt they did not receive the information that they needed about treatment before leaving the hospital with their infant. A participant stated, “We were struggling with [our daughter] having a feeding tube, because we really didn’t feel like we were given all the information. We felt like there might be other options... especially as she was struggling to swallow.” Another participant explained:

 We got to go home on Christmas Eve...and we were told to keep her away from anybody that was sick, given that she had this feeding tube, and she was vulnerable. Still not a lot of information. Basically, it was just fear, living in fear, and being scared and wondering...What’s going on?

 Due of the emphasis on breastmilk, some participants did not feel they were receiving the support they needed, as one mother said, “No one was helping me because [the hospital has] a hands-off policy.” Additionally, many participants encountered challenges when trying to access local support for their child, including long wait-times:

 When [our son] got his NG tube...placed, [we asked] "Who’s going to help us get a plan to get him off the tube?”’ and they said Developmental Pediatrics would call us... I called them when we got home [and] we were told there was a year’s wait to get in.

 As a result of this lack of information and support, some participants carried out independent research into their child’s feeding, including searching online. One mother said, “We weren’t being given any information, so of course we’re going to Google it.  We hadn’t been given... the pros and cons of [the feeding tube], so we really tried to just look things up.” Others, acted against medical advice:

I’m feeding [my son] whatever he will eat. I’m giving him netted foods... He can’t swallow them, but he can taste them. And I’m feeding him, against the orders of the physicians [to have nothing by mouth], because I feel like we’re not getting any help... So, I’m feeding [my son] now... foods that he can’t choke on. I’m just sitting there watching him eat and seeing how it’s going, because I feel like we have to make our own plan, so [we can] get him off this feeding tube.

Participants who were able to access support from healthcare providers for their child’s feeding typically received occupational therapy and support with a dietician: *“*At some point I got some advice on feeding from a dietician.” Another participant said, “Somebody referred us to an occupational therapist at a different hospital and they taught me some exercises to do and different things to try to help [with feeding].”

**Growing from Adversity**

This theme outlines the positive elements of participants’ narratives, including joy associated with improvements in their child’s feeding, reframing experiences from negative to positive, and increased confidence and resilience. Collectively, participants expressed joy and excitement when feeding finally improved. Feeding progress had a positive influence on caregiver wellbeing and day-to-day life, such as: “When [my daughter] first latched on and started breastfeeding...it was incredible, when all of a sudden it clicked.” Similarly, a parent said “Eventually, I think, [we] found the right bottle and [our son] probably learned to co-ordinate that suck/swallow... I think we just both gradually learned how to do it better.”

While experiences of feeding tubes were often described negatively, growth was demonstrated by participants’ reframing of narratives, for example, highlighting the positives including opportunities from less caregiver involvement in feeding: “Our son got a PEG tube placed...under his [clothes] and he can’t...pull it out...Now we can start feeding him more with less feedings, and [we] can both start to work, and [his] daycare can feed [him] too.”

 Participants experienced an enormous sense of relief when their child’s weight reflected the feeding progress they had made:

We had to go...[to] a doctor’s appointment, and I put [my son] on the scale and I was like “He probably has only gained a little bit” and he had gained 5lbs! And it was just in that moment that I was, like “OK!”

 Some participants reflected that their feeding journey resulted in feeling more confident and resilient as individuals and as caregivers. As one mother stated, “I can change a G-tube now without passing out. I’ve come a long way in my growth as well.” Another participant said:

Pretty much everything in my mind that I thought going into having a baby went out the door seconds after [my daughter] was born...You can think and plan as much as you want, but it’s not [always] going to happen that way. So, even if you have a healthy baby to start with, things are going to be different than you think. We just kind of learned to adapt.

Further, participants often cited innovative ways to adapt to their child’s feeding needs, for example, adapting a traditional celebration with cake: “We had done a cake smash with her older brother, we let him just destroy a cake. She couldn’t have cake because she wasn’t cleared to have it, so we made her a pudding smash instead”.

Despite difficulties with breastfeeding and with some healthcare providers, some participants demonstrated assertiveness: “For me being a third time mother, at 35 years old, I finally just said ‘Look, I need to feed my child, just bring me the bottle.’” One participant reframed the common ‘breast is best’ narrative to “fed is best”.

# **Discussion**

 These study results provide insight into the caregiver’s perspectives about the challenges that infants with CFM and their caregivers may encounter. Study narratives powerfully describe the caregiver experience and complement prior publications focused on the prevalence and types of feeding challenges that are common in infants with CFM (Caron et al., 2015; Caron et al., 2018). For new caregivers, feeding difficulties in their infants may pose several care-related and psychosocial adjustment challenges. Yet, the perspective of caregivers of children with CFM has been largely absent from the literature. While not all participants from our larger cohort reported difficulties, current findings show the extensive efforts that some caregivers must make to feed their infants with CFM.

 Participants observed several types of feeding difficulties and contributing factors, including facial nerve weakness and cleft palate, that are consistent with previously published research in children with CFM (Caron et al., 2018; Renkema et al., 2022). Less common challenges identified in this sample and reported in other studies included reflux and aspiration (van de Lande et al., 2018), which can increase the risk of malnutrition and have negative consequences for physical development (Dorise et al., 2019). Results from the current study are consistent with similar studies in infants with feeding difficulties and provide further evidence that such challenges can contribute to caregiver emotional distress about their child’s feeding and growth (Silverman et al., 2021; Vizzari et al., 2023).

 Caregivers’ narratives included an emphasis on the importance of breastmilk and perceived internal and external pressure to breastfeed. These expectations had negative outcomes for caregivers who were not able to breastfeed their child. Many who ultimately chose to use formula milk cited negative emotions such as guilt, shame, and feelings of failure. While breastfeeding is possible for some infants with CFM (Agnol et al., 2016), it can be challenging, and an emphasis on breastfeeding is detrimental to caregivers who may feel negatively about being unable to breastfeed. Further, caregivers’ description of ‘breast is best’ and ‘hands off’ policies by medical teams demonstrated the harmful consequence of a complete focus on breastmilk – that is, a lack of support for any other type of feeding (e.g., formula).

 Caregivers reported making many adaptations to overcome feeding challenges, including the use of bottles with formula or pumped breastmilk, though these approaches were not without their own challenges (e.g., access to the necessary supplies) and the effectiveness of some of these interventions are not well documented in infants with breastfeeding difficulties (Mohandas et al., 2023). While more conservative feeding assistance (e.g., use of a cleft bottle/nipple) may be sufficient for infants with mild difficulties with feeding, other infants may require more intensive interventions such as the use of tube feeding (Caron et al., 2018) particularly in cases of upper airway obstruction (Miller & Madhoun, 2016). Some participants discussed the use of feeding tubes, which required caregivers to learn new skills and were described as particularly demanding. The current findings align with previous research indicating the significant effect of feeding tubes on the entire family unit, including challenges for children’s health and happiness, caregiver stress, and logistics (Nelson et al., 2015). Unmet expectations in mothers who must rely on tube feeding their child can have adverse consequences, particularly on the development of a caregiver identity (Wilken, 2012). This research highlights the importance of robust familial education and support with tube feeding, including access to information on all aspects including practical, social and emotional factors (Syrmis et al., 2019).

 In the current study, caregivers described the support they had received from the healthcare providers at the birth hospital and/or neonatal intensive care unit and highlighted positives, such as building feeding skills prior to discharge. Caregivers also discussed receiving support from feeding therapists and diet/nutrition services. Yet, some highlighted negative experiences, including being distracted during feeding consultations due to their focus on their infant’s other medical concerns and overwhelmed by visits from multiple providers. Some caregivers cited a lack of quality information, particularly from non-specialists, and this is consistent with prior studies (Johns et al., 2018; Johns et al., 2024). This further illustrates the need for additional resources for non-specialists who are likely to interact with families of infants with rare conditions in the early postnatal period (Johns et al., 2024). Broadly, caregivers reported a desire to be more informed and involved in their child’s long-term care plan, and in some cases, dissatisfaction with providers led caregivers to disengage (e.g., going against NPO order). In the absence of trustworthy sources of support and information, they may seek out information online and make independent decisions against medical advice (Umbaugh et al., 2020). Accurate information and support reduces ambiguity for caregivers and contributes to improvements in psychosocial outcomes as well as potentially increasing treatment adherence (Crerand et al., 2018).

 Caregivers also cited positive aspects of feeding-related experiences. These included experiencing joy when feeding improved, increased confidence and resilience, and pride at gaining new skills (e.g., tube feeding). Evidence of reframing was also noted in the data, for example highlighting the practical benefits of feeding tubes - including less frequent need for feeding and greater opportunities to receive support from others. In line with previous research exploring personal growth in caregivers of children with craniofacial conditions (Feragen et al., 2020), these discussions indicate adaptive coping and offer further insights into how care providers can facilitate positive adjustment.

**Implications for Practice**

 The experiences reported by this sample illustrate the importance of the recommendation that all infants with CFM should be screened for feeding difficulties and have their growth monitored (Renkema et al., 2022). Screening for caregiver wellbeing is also indicated. Nurses serve an essential role in optimizing maternal and child health. Nurses providing care to infants during the birth hospitalization, primary care follow up, home health visits and through community programs provide critical assessments and interventions to optimize infant feeding and health. Nurses who identify challenges with feeding can facilitate timely referrals to craniofacial teams, which may mitigate the challenges that some caregivers described. The interdisciplinary craniofacial care model typically includes feeding support from providers experienced in caring for infants with craniofacial conditions, and nurses serve essential functions on such teams (American Cleft-Palate Craniofacial Association, 2018; Miller & Madhoun, 2016; van de Lande et al., 2018; van der Plas et al., 2022). Providers can assess relevant functions including oral sensorimotor strength and skills (movement, sucking). They can use the information gained from this assessment to collaborate with other medical professionals and contribute to feeding treatment plan. This may be particularly key in the first year of life which is paramount for the development of swallowing skills (van der Plaset al., 2022). As feeding challenges can change during treatment, such as mandibular distraction, feeding and growth should be continuously monitored (van der Plas et al., 2022). However, current standards for treatment or information about the efficacy of feeding interventions in this population is limited (Miller & Madhoun, 2016), such that van der Plas et al. (2022) posed that more research about feeding in children with CFM is warranted. Additional research would allow for a greater understanding of what interventions are effective, including long-term outcomes.

 A further challenge highlighted by caregivers was the long wait-times to access services, and poor communication between healthcare providers – an issue previously reported in the CFM literature (Miller & Madhoun, 2016). While a craniofacial multidisciplinary team should always be involved (van de Lande et al., 2018), other broader and universally accessible sources of support are also indicated. There is a pressing need for user-friendly online resources, collaboratively developed by healthcare professionals working with families of children with craniofacial conditions. Resources should include information about possible feeding challenges and interventions, as well as practical support for breast, bottle and tube feeding. Resources should be holistic and inclusive of the family unit, in recognition of the wide-reaching effects of CFM on the family. In this study, some caregivers noted the involvement of siblings and grandparents in tube feeding, which highlights the wider familial role in caring for children with craniofacial conditions (Guest et al., 2019; Stock et al., 2016). Involving the broader family unit in care also alleviates the burden that may be solely felt by primary caregiver(s).

 While caregivers of children with CFM will benefit from support around feeding, healthcare providers should consider the larger context for the families (Luquetti et al., 2018), particularly in the early postnatal period, and tailor information/communication in accordance. Information that is presented in a sensitive and timely manner facilitates processing and positive adjustment. Caregivers have additionally expressed a desire for information to be presented clearly and positively (Johns et al., 2022). Ideally, healthcare providers and caregivers would partner to assess the infant’s feeding abilities, discuss options, and co-develop plans to optimize the infant’s feeding and nutrition along with caregiver health.

**Limitations**

 While we sought to include English and Spanish-speaking participants with children who represented the clinical spectrum associated with CFM (including children with more complex needs such as gastrostomy tubes and tracheostomy tubes), geographic diversity in the US, and variation in access to craniofacial teams, the study sample included a higher proportion of individuals with characteristics that are typically associated with higher socioeconomic status and may not fully reflect experiences of all infants and caregivers with CFM. Fathers were also underrepresented within the sample, which aligns with recruitment trends in broader craniofacial research (Stock & Rumsey, 2015). It is thus imperative that future research focuses on how to broaden and diversify research cohorts, so that the needs of all affected, particularly those most vulnerable, can be considered in support provision.

 We used a narrative interview method in order to collect rich data that was guided by the priorities of each participant. However, this method may be influenced by recall bias, particularly for caregivers of older children (range 3-17 years) recounting their early feeding experiences. In addition, the interviews were carried out remotely in English or Spanish. We continually strive to address barriers with access to an appropriate device/internet and to design future studies that include individuals who speak additional languages. The advantage of the narrative approach was to ensure that the content was guided by caregivers, who were able to select topics most meaningful to them. Given that we did not prompt specifically about feeding during the interviews, our study may under-represent all feeding difficulties experienced by caregivers who participated in this study.

# **Conclusion**

 Caregivers play a pivotal role in their children’s health and wellbeing. The current study explored the experiences and support needs of caregivers of children with CFM, with a focus on early feeding. A significant psychosocial effect of feeding challenges for caregivers of infants with CFM was identified. Based on this study, we recommend proactive screening and timely interventions for feeding challenges that includes an empathetic approach to the possible psychosocial stressors felt by caregivers of infants with CFM.

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| --- |
| **Table 1.** Characteristics of caregiver participants |
|  | **N (%)** |
| Total N | 34\* |
| Relation to child w/CFM |  |
| Mother | 31 (91.2) |
| Father | 2 (5.9) |
| Both mother and father | 1 (2.9) |
| Mean age at interview (mean, (SD)) | 39.6 years (12.3) |
| Relationship status |  |
| Married/cohabiting | 25 (73.5) |
| Single | 8 (23.5) |
| Preferred not to answer | 1 (2.9) |
| Education |  |
| <12 yrs | 5 (14.7) |
| High school degree or equivalent | 2 (5.9) |
| Some college | 9 (26.5) |
| College degree or higher | 17 (50.0) |
| Preferred not to answer | 1 (2.9) |
| Employment |  |
| Working full-time | 16 (47.1) |
| Working part-time | 9 (26.5) |
| Not working outside the home | 8 (23.5) |
| Preferred not to answer | 1 (2.9) |
| \*34 interviews were completed with 35 people. Demographic characteristics are presented for only one parent for the pair that participated in a joint interview.  |

|  |
| --- |
| **Table 2**. Characteristics of Children of Caregiver Participants |
|  |  **N (%)** |
| Number of children of participants | 34 |
| Mean age at interview (mean, SD) | 9.8 years, 4.7 |
| Gender |  |
| Male | 19 (55.9) |
| Female | 15 (44.1) |
| Duration of hospitalization after delivery (mean, SD) | 18.6 days, 43.7 |
| Craniofacial diagnosis (can be more than one) |  |
| Microtia | 30 (88.2) |
| Hemifacial microsomia (HFM) | 20 (58.8) |
| Craniofacial microsomia (CFM) | 18 (52.9) |
| Goldenhar Syndrome | 10 (29.4) |
| Oculo-auriculo-vertebral spectrum (OAVS) | 4 (11.8) |
| Other CFM diagnosis | 1 (2.9) |
| Age at time of clinical diagnosis (mean, SD) | 3.1 months, 10.5 |
| < 6 mos | 29 (85.3) |
| ≥6 mos | 5 (14.7) |
| Seen at a craniofacial clinic |  |
| Yes | 27 (79.4) |
| No | 5 (14.7) |
| Unknown | 2 (5.9) |
| Age at first clinic visit (mean, SD) | 0.7 years, 1.1 |
| Phenotype Classification |  |
| Microtia with mandibular hypoplasia | 31 (91.2) |
| CFM-associated features without microtia | 3 (8.8) |
| Extracranial anomalies |  |
|  Congenital heart anomaly | 12 (35.3) |
|  Spinal anomaly | 10 (29.4) |
|  Kidney anomaly | 4 (11.8) |
|  Other anomaly | 8 (23.5) |
| Health Insurance |  |
|  Private | 19 (55.9) |
|  Public | 15 (44.1) |