

# Surgical Decision-Making Regarding Hearing and Ear Reconstruction in Craniofacial Microsomia: Exploring Caregiver Narratives

Nicola M. Stock (DPhil)<sup>1</sup> – Corresponding author

Beth Herring (PhD)<sup>1</sup>

Leanne Magee (PhD)<sup>2</sup>

Alexis L. Johns (PhD, ABPP)<sup>3</sup>

Canice E. Crerand (PhD)<sup>4</sup>

Carrie L. Heike (MD, MS)<sup>5</sup>

Amy Schefer<sup>5</sup>

Amelia F. Drake (MD)<sup>6</sup>

Melissa Tumblin<sup>5</sup>

Kristin Billaud Feragen (Clin. Psychol., PhD)<sup>7</sup>

<sup>1</sup>Centre for Appearance Research, University of the West of England, Bristol, United Kingdom

<sup>2</sup>Children's Hospital of Philadelphia, Philadelphia, PA, USA

<sup>3</sup>Division of Plastic and Maxillofacial Surgery, Children's Hospital Los Angeles, Los Angeles, CA, USA

<sup>4</sup>Departments of Pediatrics and Plastic Surgery, The Ohio State University College of Medicine, Columbus, OH, USA

<sup>5</sup>Center for Clinical and Translational Research, Seattle Children's Research Institute, Seattle, WA, USA

<sup>6</sup>University of North Carolina at Chapel School of Medicine, Chapel Hill, NC, USA

<sup>7</sup>Centre for Rare Disorders, Oslo University Hospital, Oslo, Norway

## Highlights

- Caregivers may experience internal conflict about surgical decisions for their child with craniofacial microsomia (CFM).
- Caregivers and children may experience psychological distress related to their CFM treatment experiences.
- Families' experiences of CFM treatment can influence future treatment decisions.

- Surgeons and other healthcare providers can offer families a thorough understanding of all CFM treatment pathways and options.
- The use of effective shared decision-making practices is strongly encouraged.

## **Abstract**

Treatment decision-making is an integral but complex part of healthcare, particularly in the context of craniofacial surgeries. The aim of the current study was to explore caregiver narratives to inform future surgical care delivery and best practice. 'Life Story' narrative interviews were conducted with US English- and Spanish-speaking caregivers ( $n=62$ ) of children aged 3-17 years with craniofacial microsomia (CFM). Extracts relating to treatment decision-making were inductively coded using Reflexive Thematic Analysis. Four themes were identified: 1) 'Grappling with Difference' exemplifies how participants dealt with having a child who was different; 2) 'Seeking Authoritative Guidance' illustrates how participants proactively pursued information about treatment options over several years; 3) 'In the Driving Seat' describes participants' beliefs about whether and how much to involve their child in treatment decisions; and 4) 'Post-Treatment Reflections' depicts participants' reflections of the decision-making experience. Surgeons and other healthcare providers are encouraged to use neutral and accessible language, to ensure families and children have a thorough understanding of all treatment pathways, and to engage in effective shared decision-making practices. Content predominantly focused on surgeries for ear reconstruction and hearing amplification. Future studies would benefit from examining other treatment decisions that caregivers are required to make.

**Key words:** microtia; OAVS; Goldenhar; hemifacial microsomia; psychosocial; surgery

## **Introduction**

Treatment decision-making is an essential yet complex part of healthcare, particularly when multiple treatment choices are available. Healthcare providers and patients must weigh the benefits and risks of a procedure and consider each option in the context of what is best for the individual (Stiggelbout et al., 2015). Surgeons are often tasked with guiding individuals and families through treatment planning and consent and therefore play a crucial role in managing the decision-making process.

Treatment decisions are especially challenging when there is clinical heterogeneity, the evidence-base for an intervention is weak (Stiggelbout et al., 2015), and/or when decisions are being made on behalf of a child (Jackson et al., 2008). Additional ethical questions are raised if a treatment decision involves an irreversible procedure, and/or alters a child's appearance (Parens, 2006).

Each of these challenges frequently occur in craniofacial care (Aspinall, 2010). In particular, the rare and heterogenic nature of craniofacial microsomia (CFM) presents a variety of complex clinical needs. CFM is a congenital condition characterised by unilateral or asymmetric bilateral underdevelopment of the facial structures (Birgfeld and Heike, 2019), most often of the ear(s) and mandible. Facial appearance may be affected, alongside hearing loss, upper airway obstruction, and feeding difficulties. Many individuals also have extracranial anomalies (Birgfeld and Heike, 2019). Long-term interdisciplinary care is therefore recommended.

Caregivers of children born with medical needs are at higher risk of elevated stress, anxiety and depression (Cousino & Hazen, 2013; Pinguart, 2018; Cohn et al., 2020). Stressors may relate to difficult experiences at the time of diagnosis, ongoing monitoring and/or treatment burden, developmental transitions, and/or fluctuations in the child's health or need for hospitalizations (Melnick et al., 2001). To mitigate stress and enhance caregiver coping, interventions focused on psychoeducation, problem-solving, and caregiver-provider communication can be beneficial. Yet, and despite the development of treatment guidelines for CFM (Henderson et al., 2015; Mazeed et al., 2019; Renkema et al., 2020), widespread consensus regarding clinical standards is lacking and care remains highly variable (Stock et al., 2023). In addition, access to reliable medical information can be limited, which poses difficulties for patients and families trying to navigate healthcare services (Johns et al., 2018a; Luquetti et al., 2018).

During childhood, it is necessary for caregivers to make a wide variety of treatment decisions. While treatment options vary depending on the child's clinical needs and the availability of resources, common treatment options presented to families include surgeries for microtia and treatments to address hearing loss associated with aural atresia. Three proposed interventions for microtia are auricular prostheses, alloplastic reconstruction (which uses a porous polyethylene implant such as Medpor®), and autologous costal cartilage reconstruction (which uses cartilage harvested from the ribs via a two- or three-stage process; Henderson et al., 2015). Treatment options for conductive hearing loss include the use of external hearing aids, canaloplasty to create or widen the external auditory

canal, and surgical placement of a bone-conduction device (Rooijers et al., 2022). Treatment options are complex, and some surgical interventions may provide a suboptimal situation for future intervention, and/or may alter the anatomy in a way that precludes future treatment choices (Truong et al., 2022). While all interventions can be associated with complications, those specifically linked to ear reconstruction and conductive hearing loss include framework exposure or extrusion, graft loss, framework reabsorption, wire exposure, scalp and auricular scarring complications, and infection (Rooijers et al., 2022; Truong et al., 2022; Ronde et al., 2024).

Patient and family treatment choices may be affected by biomedical variability (such as phenotype, medical history, and treatment eligibility), contextual variability (including treatment availability and sociocultural environment), and individual variability (such as patient/family values, treatment goals and expectations, and past treatment experiences; Lipstein et al., 2012; Ronde et al., 2024). Studies have found associations between unilateral hearing loss and an increased likelihood of speech delays, learning concerns, and poorer academic performance among children and adolescents (Kesser et al., 2013). These adverse impacts can be compounded by additional challenges with speech production for a subset of individuals with CFM (Kinter et al., 2023) and may act as a driving force for treatment. Prior research has also indicated elevated rates of teasing among children and adolescents with CFM (Johns et al., 2017; Hamilton et al., 2018), as well as a prevalent fear of stigma and teasing among caregivers (Feragen et al., 2022). Patients and families may therefore be motivated to pursue treatment that reduces the visibility of the condition to decrease the likelihood of negative social interactions.

Despite known psychosocial concerns in families affected by CFM and the complexity of treatment decisions surrounding the condition, research examining the breadth of caregiver experiences is scarce (Johns et al., 2023). In addition, caregivers' views on the long-term outcomes of treatment decisions remain largely unknown (Ronde et al., 2021). A comprehensive understanding of the barriers and facilitators to effective treatment decision-making from the caregiver perspective would allow surgeons and other healthcare providers to make relevant improvements in information provision and care delivery, leading to enhanced treatment satisfaction and long-term well-being in patients and families. The aim of the current study was to explore caregiver narratives of treatment decision-making in CFM to inform future care delivery and best practice.

## **Methods**

## ***Design***

This study is part of the larger CARE research program (Stock et al., 2023) which aims to understand the psychosocial needs and treatment experiences of individuals with CFM and their families. The initial phase of this program involved individual, narrative interviews which were predominantly self-structured and led by the participant. A summary of broad themes to arise from these interviews is presented in a conceptual thematic framework (Stock et al., 2024). Additional analyses using the narrative data are also being performed to explore specific pertinent topics in greater depth. The current paper focuses on caregivers' self-led reflections on treatment decision-making in CFM. To ensure a comprehensive reporting of all study aspects, the Consolidated Criteria for Reporting Qualitative Studies (COREQ) was followed (Tong et al., 2007).

## ***Procedure***

### Ethical Considerations

Ethical approval was granted by the Institutional Review Board at (*redacted for anonymity: hospital; IRB ID number*). All documents were subsequently ratified by the Faculty Research Ethics Committee at (*redacted for anonymity: university, ethics ID number*). Before any study procedures were performed, informed consent was collected from participants over the phone, via videoconference, or in-person, and in a location that allowed for privacy. IRB approval included a waiver of documentation of consent, and therefore all participants consented verbally. The date of consent was documented in the tracking database.

### Recruitment

Craniofacial microsomia is a broad term that includes the following clinical diagnoses: microtia, hemifacial microsomia, Goldenhar syndrome, and Oculo-Auriculo-Vertebral Spectrum (Luquetti et al., 2012; Stock et al., 2023). The research criteria for CFM established by the Facial Asymmetry Collaborative for Interdisciplinary Assessment and Learning (FACIAL) network (Birgfeld and Heike, 2019) were used. English and Spanish-speaking caregivers with children who met the inclusion criteria were eligible to participate in the study. Exclusion criteria included a known syndrome, such as Treacher Collins, Townes- Brocks, or Nager, and/or a major medical condition not associated with CFM that participants felt had a more substantial impact on their health, such as cancer. Participants were recruited across the United States (US) through online advertisements, and in-person recruitment at craniofacial clinics and advocacy events. As recruitment information was made widely available, an estimate of how many participants expressed an interest in

participating in relation to how many families saw recruitment materials is not available. Of the potential participants who initiated participation in the larger study, one was ineligible and two withdrew without completing study activities. Enrolment was actively monitored to ensure the inclusion of caregivers of children with CFM that represented US geographic regions, the diverse range of healthcare needs associated with CFM, and the full spectrum of ages (3-17 years) selected for this study.

### Data Collection

Participating caregivers provided demographic information and details of their child's medical and surgical history by telephone. Separate appointments were scheduled for the narrative interviews. Participants also submitted standardised 2D facial photographs of their children.

Narrative interviews were conducted in English or Spanish via telephone ( $n=7$ ) or a teleconference platform ( $n=55$ ), according to the participants' choice. The narrative interview method used an adapted version of the participant-led 'Life Story' interview (McAdams et al., 2001), which involves asking participants to divide their CFM story into "chapters" based on experiences that were meaningful to them. In contrast to semi-structured interviews, which predetermine areas of content to be discussed, the "Life Story" approach allows participants to freely identify and discuss the subject areas they deem to be salient in their lived experiences (McAdams, 2001; Riessman, 2008). Interviewers asked questions at the completion of each chapter to clarify and/or elaborate on aspects of participants' narratives, as well as how their care-related experiences might have been improved. Narrative interviewers were four female researchers with Master's level training. One of these researchers was bilingual. A senior interviewer was a bilingual clinical psychologist with many years' experience in the field of craniofacial conditions. All narrative interviewers were trained in qualitative interviewing techniques and most completed a minimum of two practice interviews, which were checked for quality and fidelity prior to study commencement, in addition to receiving feedback from caregiver advocates. Informal field notes were kept throughout, and the interview approach was reviewed regularly in team meetings. Sample size was determined by information power (Malterud et al., 2016), which considered representation of the full clinical spectrum, the child's age, and primary language, among other key characteristics.

### **Data Analysis**

#### Medical, Surgical and Phenotypic Data Integration

All phenotypic coding was performed by a pediatric craniofacial specialist using the previously published protocol (Stock et al., 2024). Data from caregiver interviews and ratings from photographs were integrated to establish the phenotype by feature for each participant.

### Narrative Interview Data

A total of 62 narrative interviews with caregivers were completed, including 50 in English (80.6%) and 12 in Spanish (19.4%). Interviews ranged from 30 to 191 minutes in length (M=83 minutes). Interviews conducted in English were transcribed by an external individual transcriptionist. Interviews conducted in Spanish were transcribed and translated by a professional transcription company.

Analysis was performed by two female senior investigators with qualitative experience and clinical and research expertise in the field of craniofacial conditions. Reflexive Thematic Analysis was chosen for its flexibility in developing, analysing, and interpreting patterns across qualitative datasets (Braun and Clarke, 2022). No prior framework was applied to the data. A hermeneutic-phenomenological approach was used to give primacy to the participants' experiences while acknowledging the influence of our own perspectives on the analysis (Laverty, 2003). Both researchers became familiar with caregiver narratives through multiple readings of all transcripts. Data were then extracted from the larger narrative interviews that could be broadly relevant to treatment decision-making. Extracts were methodically coded for content relevant to the study aim. Initial themes were generated based on central and distinctive organising concepts, and rich descriptions of each theme were produced. Each theme was then reviewed and adjusted as necessary to provide a coherent, nuanced, and robust narrative of the overall dataset. Themes were named, and exemplar quotations were selected. The two senior investigators worked closely while generating and reviewing themes. They regularly reflected (individually, together) on the analytical process, considering their subjective positioning and discussing any concerns or discrepancies. They consulted with the team's Psychology Committee (an international group of leading clinical psychologists with specific expertise in craniofacial conditions) as the themes developed. They also discussed preliminary findings with the caregiver representatives and other multidisciplinary professionals (plastic/maxillofacial/ENT surgeons, epidemiologists and paediatricians) on the wider Advisory Committee. All co-authors reviewed and approved the written content.

## **Results**

### ***Participant Characteristics***

As presented in Table 1, participants included 57 mothers (91.9%) and 5 fathers (8.1%). Caregivers had a mean age of 40.2 years (SD=11.9), were primarily White (61.3%), married/cohabiting (77.4%), and had completed college (54.8%). Families' health insurance was private (56.5%) or public (43.5%). As illustrated in Table 2, the mean age of participants' children at the time of interview was 10.4 years (SD=4.4), and just over half of the children were male (51.6%). Most children had microtia (98.4%), aural atresia (82.3%), and/or some degree of mandibular hypoplasia (72.6%). As shown in Table 3, the median number of surgeries per child was 4 (range 1-21). The most common craniofacial surgeries were ear reconstruction (38.7%), removal of preauricular and/or facial tags (38.7%), and placement of tympanostomy tubes (32.3%). A full sample description is available elsewhere (Stock et al., 2024).

### **Qualitative Synthesis**

When offered the opportunity to self-lead their CFM narrative, participants in this study predominantly spoke about surgical decision-making related to ear reconstruction and hearing aid abutment. Four core themes were identified: 1) Grappling with Difference; 2) Seeking Authoritative Guidance; 3) In the Driving Seat; and 4) Post-Treatment Reflections. Each theme is outlined below using exemplar quotations. In some instances, quotes have been shortened by the authors to enhance clarity and focus. As suggested by Hill and colleagues (2005), the findings are categorised 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). When reporting the findings, these three labels are referred to as all (general), most (typical), and some (variance).

#### **Theme 1: Grappling with Difference**

The first theme exemplifies how participants dealt with having a child who was born different. This internal struggle, combined with other people's reactions, had an influence over participants' motivations for pursuing treatment and the strategies they used to cope with their child's difference while they waited for treatment to become available.

While treatment decisions for ear reconstruction occur later in childhood, most participants were informed about this treatment option when their child was an infant. Surgery was rarely presented as a choice, but as an explicit step to anticipate. During early interactions with healthcare providers, microtia was typically described as a 'physical anomaly' that was 'easily fixed' through surgery: "*There was a plastic surgeon in the room less than five hours*



*after [my son] was born, saying... 'When he gets older, we can easily just reconstruct his ear and fix everything'" (caregiver of 12-year-old).*

These initial interactions had the potential to influence not only how participants felt about their child's differences, but also formed the presumption that surgery was 'the answer': *"[The plastic surgeon] said 'when [your son] has his ear reconstructed, he's going to be...like any other boy'... I was overjoyed to hear those things... Even though he was only 3 months [at the time]... I'm like 'Oh my God, yes!'... One day his ear will be normal"* (caregiver of 13-year-old).

Participants were more likely to consider ear reconstruction from an early stage if they were struggling to come to terms with their child's condition. Focusing on surgery was one way for some participants to allay their own feelings surrounding their child's differences: *"[Surgery] was a fixation for me from the beginning... It made me feel much better about it all...because I felt confident that 'we're taking action. We're doing something... I've got control of this'... And that made me feel much less shame"* (caregiver of 11-year-old).

Most participants were preoccupied by the visible aspects of their child's condition. Real and/or imagined social reactions from others, and the potential impact of these reactions on their own or their child's well-being were key motivations for pursuing surgery: *"I was extremely worried about bullying and that was something that we wanted to think about and change for [our son] in the future"* (caregiver of 9-year-old). Participants were also prompted to pursue treatment if they believed their child's hearing difficulties would impact academic achievement and/or the ability to participate effectively in social activities.

Some participants saw surgery as a milestone to be achieved, and anticipated feeling relieved once surgery was 'over'. Having been told that certain treatment options would not be available until their child was much older, these participants expressed frustration at needing to wait so long for a resolution: *"It's been several years and I'm still wanting and waiting for [my son's] surgery"* (caregiver of 6-year-old).

While waiting for treatment, some participants identified alternative ways to minimise their child's differences to reduce the likelihood of unfavourable reactions: *"[I] kept [my son's] hair long enough to cover his right ear... Just to not bring that negative attention... The hearing device...was almost identical to the colour of [my son's] hair. So, it blended quite well"* (caregiver of 7-year-old). Other participants felt more conflicted about pursuing treatment to 'fix' their child's differences and debated what was the 'right or wrong' course of action. For those participants experiencing doubt, the knowledge that treatment decisions were on the

horizon was a source of stress: *“That’s when it started, that kind of feeling of foreboding, and just worrying in general that...there were major decisions down the road”* (caregiver of 6-year-old).

Some participants worried that *“my choice to have surgery [would be] an indication to [my son] that I didn’t like the way he looked”* (caregiver of 12-year-old). Participants who were cognisant of their role in shaping their child’s attitude towards their appearance were less likely to conceal their child’s differences, and placed more emphasis on helping their child to develop their personality, abilities, and characteristics other than physical appearance: *“Hiding it just tells [our son] that who he is, is not OK...[and] that defeats the purpose of what we’re trying to instil in him”* (caregiver of 7-year-old).

## **Theme 2: Seeking Authoritative Guidance**

The second theme illustrates how participants proactively pursued information about treatment options over a long period of time, often years. Information-seeking was a crucial part of the decision-making process and was seen as a way of gaining some control over a stressful situation. Participants sought authoritative guidance from healthcare providers, the Internet, and other parents of children with CFM.

Despite *“no lack of effort to find answers”* (caregiver of 5-year-old), some participants found it difficult to access reliable and consistent evidence from healthcare providers to guide or validate their treatment decisions. Participants also frequently commented on the challenges of managing health insurance and the psychological toll this could take on the caregiver: *“It was so frustrating...exhausting... [Health insurance] became basically a second job”* (caregiver of 4-year-old).

Often in response to these perceived barriers, many participants sought information from additional sources, including non-profit organisations and peers. Participants valued hearing families’ experiences, sharing advice, and seeing the results of other children’s treatments. Contact with other parents also brought a deep sense of being understood and a normalisation of the thoughts and emotions involved in treatment decision-making that differed from interactions with healthcare providers: *“[Through this network] I met other families...and every question...I wanted to ask...I had a place to go... The best person that can give you advice is the person that’s been through the same as you”* (caregiver of 4-year-old). Some participants had had the opportunity to attend a specialist conference for families affected by CFM. These educational events gave participants a chance to talk to other

families and healthcare providers, allowing them to gather a large amount of information about treatment options, costs, and pathways in a short space of time.

It was often through their own research that participants learned about the range of treatment options and healthcare providers available. This independent information-seeking could lead participants to discover treatment options of which they had not previously been aware. The impression that their existing healthcare provider had withheld information and/or having a feeling of having been deliberately misled evoked distress for some participants: *“I started researching [and] finding all this information tied to these surgeries that we had talked about for so long... I learned that one, the [rib graft] wasn’t the only option and two...that if it was the option we were aiming for, there were specialists across the country who did that... I just was floored... I thought ‘I let my child down’”* (caregiver of 14-year-old). Some participants had subsequently chosen to leave their healthcare provider: *“We never went back to them because...even if they didn’t believe in [a particular treatment], they didn’t even educate us”* (caregiver of 12-year-old).

A small number of participants reported feeling pressured by healthcare providers into making decisions that they were not completely comfortable with. In contrast, a handful of participants expressed a preference for more directive recommendations from their healthcare provider: *“It would have been nice if somebody had told me exactly what to do...because I trust [the healthcare providers] so much... But instead, they [gave] me...two very different options, so it definitely took some research on my end, to decide what I felt was best. And then circling back to them and saying ‘OK, this is what I came up with’, and that’s when I got the reassurance... ‘If you knew this from the beginning, why did you put me through that?’”* – caregiver of 4-year-old.

Participants also described positive interactions with healthcare providers, which were most often associated with engagement with a specialist craniofacial team. These experiences had reassured them their child was receiving good medical care and alleviated feelings of stress and overwhelm. Participants appreciated healthcare providers taking a calm approach, expressing empathy, sharing clear and comprehensive information, and making appropriate referrals. Some participants highlighted healthcare providers who had stated *“there is also a ‘do nothing’ option, and there’s no harm in waiting”* (caregiver of 11-year-old).

### **Theme 3: “In the Driving Seat”**

The third theme describes participants' beliefs about whether and how much to involve their child in treatment decisions. Participants tried to navigate their own beliefs about what would be best for their child in the long-term, alongside whether and how to listen to and respect their child's treatment wishes.

As participants gathered information about the treatment options available, most described an internal conflict in which they debated 'back-and-forth' about what was best for their child. Participants not only questioned which surgical option was most suitable, but whether to opt for surgery at all: "*We were just...learning our different options, and what do we do? Do you wait and let [your daughter] decide? Do we make the decision? Do we do anything? Do we leave it? How do we manage all these decisions?*" (caregiver of 14-year-old). This conflict was amplified if the parents disagreed on the best course of action.

For some participants, being a 'good' parent meant actively pursuing all options available. These participants believed the responsibility for the decision should reside with the caregiver: "*I wouldn't ever want [my daughter] to say 'look at me, my ear is like this because my parents [did nothing]'. For people to say 'hey, honey, why didn't you get your ear fixed?'"* (caregiver of 13-year-old).

Other participants saw the theoretical benefit of involving their child in treatment decisions, but as a result of believing it was 'best' to perform surgery at an early age, felt their children's developmental level would not allow them to understand what was at stake and/or to coherently express their wishes: "*I couldn't imagine myself having a conversation with [my son] when he was...three and four, when we were really launching on the path. He wasn't yet of the age of rationality where you would turn this decision over to him"* (caregiver of 11-year-old).

Even after having made the decision to pursue treatment on their child's behalf, some participants expressed ongoing doubt: "*In my mind and in my heart, I was going back and forth. 'Are you making the right decision? You're making a huge decision for somebody else... What if [your son] would have preferred to just be who he was at birth, and you changed that?' There were many different things going through my mind and...I had a lot of anxiety about that"* (caregiver of 14-year-old). Participants were therefore relieved if their child communicated a treatment decision, either to pursue surgery or to wait, that aligned with their own.

Some participants expressed their wish to involve their child in treatment decisions, yet also shared their intention to encourage their child to pursue a particular pathway. These

participants often had a clear idea of what they thought was best for their child and were prepared to weigh in on their child's decision if their child held a different opinion to theirs: *"[Our son] says 'no surgery, no surgery, no surgery'...but the reality is we're going to do what we need to do to make sure he's getting the best care and the best chance possible in life, even if that means 'OK, you're going to get a magnet put in your head. It's not a serious surgery, you're going to be fine'"* (caregiver of 5-year-old).

In contrast, some participants felt strongly that the decision to pursue surgery should ultimately rest with their child and were more able to respect their child's choices and potential reluctance towards treatment. These participants had introduced the idea of surgery to their children but had put treatment on hold until their child was able to engage more fully with the process: *"This is [our son's] body. We want to have these conversations with him, so he is aware of what's going on...[but] we're leaving it up to him"* (caregiver of 6-year-old).

Instead of making a decision on their child's behalf, or influencing them in a certain direction, these participants placed the most emphasis on developing their child's self-confidence and decision-making skills. In particular, participants wanted their child to *"make these life-changing decisions for the right reasons, not because [they're] being teased"* (caregiver of 4-year-old) and emphasised the need to *"support [our daughter] emotionally and developmentally... Helping her understand her choices and...getting checked out every year...so she has the best information...[and] the latest technology"* (caregiver of 4-year-old).

#### **Theme 4: Post-Treatment Reflections**

The fourth and final theme depicts participants' reflections of the decision-making process, which was broadly based on whether they perceived treatment outcomes to be positive or negative. These reflections seemed pivotal in the context of future treatment decisions, with the potential to either strengthen or displace participants' trust in their own decision-making abilities.

Once a treatment had taken place, participants were reassured in their decision-making if they perceived the treatment to have been successful. "Success" could be defined as a satisfactory aesthetic outcome, an improvement in hearing, a lack of complications, a perceived reduction in social reactions, and/or a perceived increase in the child's self-esteem: *"I was very happy with the results. Everything looked great, [my son] did really well*

*with it, we didn't have the complications. So, we were very, very happy overall with the surgery, with the surgeons, and how the whole process went"* (caregiver of 9-year-old).

Other participants felt disappointed by the outcome of surgery, which led them to experience regret over the choices they had made: *"We were all excited... 'Finally, [our son] is going to have a normal ear!'... And then he had surgery, and it was a let-down because...his ear didn't look anything like...the pictures we'd seen before. He still looked pretty deformed"* (caregiver of 13-year-old). While this disappointment was immediate for some participants, others' satisfaction with the treatment outcome had reduced over time: *"The ear which looked so amazing at the end of the surgical process [is] no longer protruding at all... That's when the regret started, really... I flat out wish we hadn't done it"* (caregiver of 11-year-old). Participants also described how their initial satisfaction with their child's hearing device contrasted with the child's lack of acceptance, due to complaints of pain and disturbing sounds.

Some participants described post-surgical complications that caused their child significant discomfort. In several cases, this led to unanticipated follow-up treatment and monitoring, and for some, the treatment had ultimately failed. These complications were extremely distressing for both participants and their children: *"[The surgical site] was infected... [My daughter's] ear was bleeding... They had to take the Medpor out... That was when we really started to struggle a lot... To almost lose everything we had done, achieved, what she'd suffered - in a moment it came to nothing... I should have left it alone, because she struggled in vain"* (caregiver of 13-year-old). Participants who experienced complications did not believe these risks had been adequately explained prior to surgery.

As a result of treatment and/or the complications that followed, some participants' children began to exhibit emotional distress, a fear of medical settings, and/or a strong wish to avoid any future treatment: *"Last week we were at an eye appointment...and [my son had] a little mental breakdown. [He] was crying and thought something bad was going to happen to him... He doesn't want to have any more surgery"* (caregiver of 4-year-old). These experiences could also influence the relationship between the participant and their child: *"I told [my daughter] 'you have to understand, [this treatment is] for your own good'... And she told me... 'you always lie to me... You make them do this to me, and it hurts me...I don't want to see you here... No, no, no, you're not my real mom'... The idea that a real mom doesn't make her children suffer"* (caregiver of 13-year-old).

Such experiences were emotionally demanding and impacted future treatment decisions from the participants' perspective as well as the child's: "*We didn't realise the emotional impacts the surgery...would have on [our son]... A month or two later, [I asked] 'what would you tell another kid who was considering this?' He told me 'I'd tell them not to do it because I really wish I didn't do it.' And that was hard*" – caregiver of 14-year-old.

Participants were more likely to question their decision if they had experienced complications, were unsatisfied with the treatment outcome, or perceived the negative impact on their child to outweigh the benefits of the surgery itself. This led some participants to take a temporary or permanent step back from treatment: "*We are quitting while we're ahead. We're not doing anything else*" (caregiver of 12-year-old).

Reflecting on the decision-making process overall, some participants cautioned against making decisions too quickly or basing decisions on unfounded concerns and assumptions. Some appeared more confident than others to choose a 'wait and see' approach, and letting the child's behaviour, experiences, and well-being guide their choices along the way: "*I do think it would be helpful, for aesthetic decisions in particular, to recognise that the world is not always going to be cruel and that there is a place in the world for just keeping differently developed bodies as they are... It's OK to choose to fix them but it's also OK to leave them alone*" (caregiver of 12-year-old).

Some participants seemed to have found comfort in the view that there is no "right" decision, and trusted life to go well for their child despite their child's differences: "*Everyone's journey is different... There's not one right way to do anything. Everyone does what seems to fit best for them and their family and...it's not one size fits all*" (caregiver of 6-year-old).

## **Discussion**

This study sought to explore caregiver narratives of treatment decision-making in CFM to inform future care delivery and best practice for surgeons and other healthcare providers. The findings provide evidence of helpful clinical and community-based support for families, alongside significant challenges and unmet needs.

From the moment their child with CFM is born, caregivers begin a "waiting game". There are often many years between caregivers first learning that treatment is possible and the child being old enough for certain treatments to become optimal. During this time, new techniques and products may also emerge, ensuring that caregivers must continually update their knowledge of what is possible. Caregivers may seek information and guidance from surgeons, other healthcare providers, the Internet, and peers to aid their decision-making.

Caregivers may also experience internal conflict about whether to pursue treatment, which treatment option offers the best fit for their family, and the degree to which they should involve their child in the decision-making process. Caregivers' post-treatment reflections can vary according to immediate and long-term treatment outcomes, any medical complications, the impact of treatment on the well-being of the child, and/or the influence of the treatment process on the parent-child relationship. Consequently, caregivers may feel reassured they have made the 'right' decision or may experience a degree of decisional regret. Experiences of early decision-making and treatment can influence future treatment decisions, both for caregivers and their children.

Consistent with caregiver experiences described in prior research (Johns et al., 2023), the findings of this study are indicative of opportunities for improvement in CFM care delivery. These findings have been translated into considerations for surgeons and other healthcare providers (Table 4). First, all healthcare providers are encouraged to consider the language they use when describing CFM to caregivers. Instead of terms which emphasise the child's differences and/or pathologize CFM ('defect', 'malformation', 'abnormality', 'disfigurement'), the use of neutral language ('diagnosis', 'condition', 'small ear', 'microtia') can retain meaning and precision without having stigmatising or negative connotations (Stock et al., 2020). Similarly, using accessible descriptive terms, such as 'surgery' or 'treatment' instead of 'repair' or 'fix' conveys an accurate message without unintentionally implying that a child is 'broken', and that surgery is the solution (Stock et al., 2020). Awareness among healthcare providers that families may be grappling with complex feelings about their child's differences and navigating the social reactions of others (Johns et al., 2017; Thornton et al., 2021) can help to ensure providers do not inadvertently add to the stigma already felt by caregivers. Consensus within craniofacial teams that choosing not to pursue elective treatment is a valid option and ensuring this option is presented to all families (Renkema et al., 2020) can offer reassurance to caregivers and may act as a powerful counter to societal stigma. Routinely screening for psychological distress in caregivers alongside input from a psychologist where indicated may also help caregivers adjust to their child's differences and reduce the likelihood of families pursuing treatment for reasons which they later call into question. In addition to direct guidance from healthcare providers, caregivers may seek out support from advocacy groups and peers. Peer support for caregivers and patient-centred educational events have been shown to positively impact diagnosis and treatment knowledge, coping skills, resiliency, empathy, self-care, social comfort, parenting skills, self-efficacy, and mental health (Bogart et al., 2016; Johns et al., 2018b; Lancaster et al., 2023). Referring



families to verified advocacy groups and peer services may therefore reduce parental distress and favourably inform the decision-making process.

Offering families comprehensive and reliable information about their child's diagnosis and the available treatment options is paramount in supporting families to make confident and informed treatment decisions. Existing CFM information, including that available online is often disjointed and of variable quality (Alamoudi et al, 2015; Johns et al., 2018a; Luquetti et al., 2018). Given the additional lack of referrals for reconstruction assessments (Long et al., 2023), difficulties accessing care (Jovic et al., 2021), and the inherent challenges of addressing all potential health needs in one source, the development of an authoritative evidence-based resource(s) may be helpful. Ideally, information should be offered in a variety of formats (e.g. verbal explanations and/or printed/online written materials and diagrams) to suit families' preferences, learning styles, and literacy levels. Clinically, healthcare providers can take steps to ensure that families understand the implications of their child's diagnosis and that they accurately communicate the risks and likely outcomes of each treatment option, including any possible complications and post-treatment monitoring requirements. Healthcare providers should aim to stay up to date with the latest research and clinical advances across disciplines, in order to offer families the full scope of best multidisciplinary practice. Given the time constraints in many subspecialty clinics, better tools and processes for clinicians to address these needs would be beneficial.

The findings of this study imply that shared decision-making practices are not being effectively and/or consistently implemented, which has implications for surgical planning. Shared decision-making (SDM) recognises the ethical imperative for individuals and families to be involved in healthcare decisions and is defined as a collaborative process in which patients and providers identify a mutually agreed-upon treatment plan (Légaré et al., 2018; see Figure 1 for an example). SDM supports families to make decisions based on their personal preferences, which in turn enhances adherence to evidence-based treatment regimes, manages expectations, and improves patient satisfaction (Murray et al., 2005). SDM strategies may be most effective in situations where there is a lack of clear scientific evidence to demonstrate the superiority of one treatment choice over another (Stiggelbout et al., 2015), as is arguably the case in CFM (Stock et al., 2023). SDM guidelines place an emphasis on the healthcare provider adopting a consulting style that is curious, supportive, non-judgemental, and un-biased (Coulter and Collins, 2011). This involves building trust and an authentic rapport with families, and utilising essential interpersonal skills, such as active listening, empathy, patience, flexibility, and confirmation of understanding (O'Toole, 2024).

Guidelines also stipulate that a formal system for documenting, communicating, and implementing shared decisions should be incorporated into routine practice (Coulter and Collins, 2011). Decision aids (such as informative leaflets and videos; O'Connor et al., 2009), decision support tools and interventions (e.g. a set of recommended steps; Clarke et al., 2021), and decision coaching (based on motivational interviewing techniques; Rollnick et al., 2008) can guide surgeons and other healthcare providers effectively through the SDM process. These evidence-based decision aids may help to reduce health inequities (King et al., 2008), which may be especially pertinent for healthcare systems that are not universally accessible (Lu et al., 2022).

Finally, support for caregivers to involve their child in developmentally appropriate decision-making may be beneficial. Caregivers and children may disagree about whether to pursue treatment and/or treatment preferences, which can lead to emotional distress and fractured relationships (Miller et al., 2009). The struggle to know when and how to include their child in decisions may also result in caregivers trying to filter information or influence their child's decisions (Daniel et al., 2005). Although children may not fully comprehend the nature, purpose, and possible consequences of treatment, they hold a strong interest in what happens to their bodies (Lipstein et al., 2012). Surgeons and other healthcare providers can support children to be involved in decisions by communicating information in ways that are appropriate to the child's developmental level, respecting the child's wishes to be involved in decision-making (or not), and providing them with opportunities to share their concerns and opinions (NICE, 2023). It may be helpful for all healthcare providers to view family and patient education as a process that occurs over time, with increasing involvement of the child in conversations about treatment options, risks, benefits, and outcomes. Including the child in the SDM process may also provide parents with a model of how to communicate with their child about CFM and/or its treatment, which is important for the child's self-confidence and the development of self-advocacy.

Limitations of this study include the largely well-educated, employed, White/European American sample, which does not represent the experiences of all families affected by CFM. Fathers were also underrepresented in the sample, reflecting ongoing challenges to engage fathers in paediatric research (Ferreira de Moura and Philippe, 2023). Concerted efforts were made to include caregivers of children with a range of CFM phenotypes, as well as Spanish-speaking participants, yet further efforts to represent these families in research are needed. CFM is associated with wide clinical heterogeneity, and exploration of decision-making for children with more complex clinical needs is warranted. Similarly, and given the

average age of participants' children at the time of interview, the views of caregivers of older children may not be entirely reflected. Experiences of later surgeries and other types of surgeries not discussed in this paper therefore require further investigation. Finally, patients' own experiences of decision-making were not included in the current paper. Future research should seek to elicit the views of children, adolescents, and young adults, as well as healthcare providers, in order to understand the complexities of the decision-making process from the perspectives of all parties.

The narrative approach used in this study relied on caregivers' ability to identify, recall and report on the aspects of their experiences they considered to be most salient. While this approach allows participants the freedom to direct their own story, it does not permit more structured or focused questioning about a given topic. As a result, it is likely that not all pertinent aspects of treatment decision-making were highlighted by the current study. What is clear is that treatment decision-making is a source of significant stress for families and a critical area of focus for future research. Targeted studies directly addressing the experiences and unmet needs of families representing the broad spectrum of CFM are strongly encouraged.

## **Conclusions**

This study provides an understanding of the treatment decision-making experiences of a large US cohort of caregivers of children with CFM. The findings have been translated into recommendations that may improve the caregiver experience. Surgeons and other healthcare providers are encouraged to communicate using neutral and accessible language, to ensure families and children have a thorough understanding of all treatment pathways, and to engage in effective shared decision-making. The field would also benefit from the development of decision aids and informational resources to support families through this complex and challenging process.

**Declarations of interest:** None.

## **References**

Alamoudi, U., Hong, P. Readability and quality assessment of websites related to microtia and aural atresia. *Int J Pediatr Otorhinolaryngol.* 2015;79(2): 151-6. doi: 10.1016/j.ijporl.2014.11.027.

- Aspinall, C. Anticipating benefits and decreasing burdens: The responsibility inherent in pediatric plastic surgery. *J Craniofac Surg.* 2010;21(5): 1330-1334. doi: 10.1097/SCS.0b013e3181ef2a5b.
- Birgfeld, C., Heike, C. Craniofacial microsomia. *Clin Plast Surg.* 2019;46: 207-221. doi: 10.1016/j.cps.2018.12.001.
- Bogart, K.R., Hemmesch, A.R. Benefits of support conferences for parents of and people with Moebius syndrome. *Stigma Health.* 2016;1(2): 109–121. doi: 10.1037/sah0000018.
- Braun, V., Clarke, V. *Thematic analysis: A practical guide.* Sage; 2022.
- Clarke, A., Paraskeva, N., White, P., Tollow, P., Hansen, E., Harcourt, D. PEGASUS: the Design of an Intervention to Facilitate Shared Decision-making in Breast Reconstruction. *J Cancer Educ.* 2021;36(3): 508-518. doi: 10.1007/s13187-019-01656-6.
- Cohn, L.N., Pechlivanoglou, P., Lee, Y., Mahant, S., Orkin, J., Marson, A., Cohen, E. Health Outcomes of Parents of Children with Chronic Illness: A Systematic Review and Meta-Analysis. *J Pediatr.* 2020;218:166-177.e2. doi:10.1016/j.jpeds.2019.10.068.
- Cousino, M.K., Hazen, R.A. Parenting stress among caregivers of children with chronic illness: A systematic review. *J Paediatr Psychol.* 2013;38(8):809-828. doi:10.1093/jpepsy/jst049.
- Coulter, A., Collins, A. *Making shared decision-making a reality.* The Kings Fund, London: 2011.
- Daniel, E., Kent, G., Binney, V., Pagdin, J. Trying to do my best as a mother: Decision-making in families of children undergoing elective surgical treatment for short stature. *Br J Health Psychol.* 2005;10(Pt 1):101-14. doi: 10.1348/135910704X14609.
- Feragen, K.B., Myhre, A., Stock, N.M. "Will You Still Feel Beautiful When You Find Out You Are Different?": Parents' Experiences, Reflections, and Appearance-Focused Conversations About Their Child's Visible Difference. *Qual Health Res.* 2022;32(1): 3-15. doi: 10.1177/10497323211039205.
- Ferreira de Moura, A., Philippe, K. Where is the father? Challenges and solutions to the inclusion of fathers in child feeding and nutrition research. *BMC Public Health.* 2023;23. doi:10.1186/s12889-023-15804-7.
- Hamilton, K.V., Ormond, K.E., Moscarello, T., Bruce, J.S., Bereknyei Merrell, S., Chang, K.W., Bernstein, J.A. Exploring the medical and psychosocial concerns of adolescents and young adults with craniofacial microsomia. *Cleft Palate Craniofac J.* 2018;55(10): 1430-1439. doi: 10.1177/1055665618768542.
- Henderson, R., Moffat, C., Stewart, K., Clapperton, K. *UK care standards for the management of patients with microtia and atresia.* 2015. Available at: [MICROTIA and ATRESIA- CARE STANDARDS \(bapras.org.uk\)](https://www.bapras.org.uk/microtia-and-atresia-care-standards). Accessed December 2023.
- Hill, C.E., Knox, S., Thompson, B.J., Williams, E.N., Hess, S.A., Ladany, N. Consensual qualitative research: An update. *J Counsel Psychol.* 2005;52: 196–205. doi: 10.1037/0022-0167.52.2.196.

Jackson, C., Cheater, F.M., Reid, I. A systematic review of decision support needs of parents making child health decisions. *Health Expectations*. 2008;11(3): 232-251. doi: 10.1111/j.1369-7625.2008.00496.x.

Johns, A.L., Gutierrez, Y., Nicolaou, D.C., Garcia, L., Cespedes-Knadle, Y., Bava, L. A support group for caregivers of children with craniofacial differences. *Social Work With Groups*. 2018b;41(3): 211-226. doi: 10.1080/01609513.2017.1333480.

Johns, A.L., Lewin, S.L., Im, D.D. Teasing in younger and older children with microtia before and after ear reconstruction. *J Plast Surg Hand Surg*. 2017;51(3): 205-209. doi: 10.1080/2000656X.2016.1222294.

Johns, A.L., Luquetti, D.V., Brajcich, M.R., Heike, C.L., Stock, N.M. In their own words: caregiver and patient perspectives on stressors, resources, and recommendations in craniofacial microsomia care. *J Craniofac Surg*. 2018a;29(8):2198-2205. doi:10.1097/SCS.0000000000004867.

Johns, A.L., Stock, N.M., Costa, B., Feragen, K.B., Crerand, C.E. Psychosocial and health-related experiences of individuals with microtia and craniofacial microsomia and their families: Narrative review over two decades. *Cleft Palate Craniofac J*. 2023;60(9): 1090-1112. doi: 10.1177/10556656221091699.

Jovic, T.H., Gibson, J.A.G., Griffiths, R., Dobbs, T.D., Akbari, A., Wilson-Jones, N., Costello, R., Evans, P., Cooper, M., Key, S., Lyons, R., Whitaker, I.S. Microtia: A Data Linkage Study of Epidemiology and Implications for Service Delivery. *Front Pediatr*. 2021;26(9):630036. doi: 10.3389/fped.2021.630036.

Kesser, B.W., Krook, K., Gray, L.C. Impact of unilateral conductive hearing loss due to aural atresia on academic performance in children. *Laryngoscope*. 2013;123(9): 2270-2275. doi: 10.1002/lary.24055.

King, J.S., Eckman, M.H., Moulton, B.W. The potential of shared decision making to reduce health disparities. *J Law Medicine Ethics*. 2008;39:s30–3. doi: 10.1111/j.1748-720X.2011.00561.x.

Kinter, S., Kotlarek, K., Meehan, A., Heike, C.L. Characterizing speech phenotype in individuals with craniofacial microsomia: A scoping review. *Am J Speech-Language Pathol Audiol*. 2023;33:485-504. doi: 10.1044/2023\_AJSLP-23-00152.

Lancaster, K., Bhojti, A., Kern, M.L., Taylor, R., Janson, A., Harding, K. Effectiveness of peer support programmes for improving well-being and quality of life in parents/carers of children with disability or chronic illness: A systematic review. *Child Care Health Dev*. 2023;49(3): 485-496. doi: 10.1111/cch.13063.

Laverty, S. M. Hermeneutic Phenomenology and Phenomenology: A Comparison of Historical and Methodological Considerations. *Int J Qualit Methods*. 2003;2(3):21-35. doi: 10.1177/160940690300200303.

Légaré, F., Adekpedjou, R., Stacey, D., Turcotte, S., Kryworuchko, J., Graham, I.D., Lyddiatt, A., Politi, M.C., Thomson, R., Elwyn, G., Donner-Banzhoff, N. Interventions for increasing the use of shared decision making by healthcare professionals. *Cochrane Database Syst Rev*. 20187(7):CD006732. doi: 10.1002/14651858.CD006732.pub4.

Lipstein, E.A., Brinkman, W.B., Britto, M.T. What is known about parents' treatment decisions? A narrative review of pediatric decision making. *Med Decision Making*. 2012;32(2): 246-258. doi: 10.1177/0272989X11421528.

Long, A.S., Junn, A.H., Shah, H.P., Almeida, M.N., Rivera, J.C., Nguyen, H.P., Persing, J.A., Alperovich, M. Gaps in Access to Reconstruction Compared with Audiologic Care among Children with Microtia. *J Pediatrics*. 2023;262: 113340. doi: 10.1016/j.jpeds.2023.01.014.

Lu, Y., Elwyn, G., Moulton, B.W., Volk, R.J., Frosch, D.L., Spatz, E.S. Shared Decision-making in the U.S.: Evidence exists, but implementation science must now inform policy for real change to occur. *Z Evid Fortbild Qual Gesundheitswes*. 2022;171:144-149. doi: 10.1016/j.zefq.2022.04.031.

Luquetti, D.V., Brajcich, M.R., Stock, N.M., Heike, C.L., Johns, A.L. Healthcare and psychosocial experiences of individuals with craniofacial microsomia: patient and caregivers perspectives. *Int J Pediatr Otorhinolaryngol*. 2018;107:164-175. doi: 10.1016/j.ijporl.2018.02.007.

Luquetti, D.V., Saltzman, B.S., Vivaldi, D., Pimenta, L.A., Hing, A.V., Cassell, C.H., Starr, J.R., Heike, C.L. Evaluation of ICD-9-CM codes for craniofacial microsomia. *Birth Defects Res: A Clinical and Molecular Teratology*. 2012;94(12): 990-995. doi: 10.1002/bdra.23059.

Malterud, K., Siersma, V.D., Guassora, A.D. Sample size in qualitative interview studies: Guided by information power. *Qual Health Res*. 2016;26(13):1753-1760. doi: 10.1177/1049732315617444.

Mazeed, A.S., Saied, S., Abulezz, T., Youssef, G., Bulstrode, N.W. Recommendations for the development and reform of microtia and atresia services. *J Craniofac Surg*. 2019;30(4):1135-1139. doi:10.1097/SCS.00000000000005209.

McAdams, D.P. The Psychology of Life Stories. *Rev Gen Psychol*. 2001;5(2): 100-122. doi: 10.1037/1089-2680.5.2.100.

Melnyk, B.M., Feinstein, N.F., Moldenhouer, Z., Small, L. Coping in parents of children who are chronically ill: strategies for assessment and intervention. *Pediatr Nurs*. 2001;27(6):548-558.

Miller, V.A. Parent-child collaborative decision making for the management of chronic illness: A qualitative analysis. *Fam Syst Health*. 2009;27(3):249-66. doi: 10.1037/a0017308.

Murray, E., Burns, J., See, T.S., Lai, R., Nazareth, I. Interactive Health Communication Applications for people with chronic disease. *Cochrane Database Syst Rev*. 2005;19(4):CD004274. doi: 10.1002/14651858.CD004274.pub4.

National Institute for Health and Care Excellence. *Babies, children and young people's experience of healthcare*. NICE guideline: ng204. Available at: [www.nice.org.uk/guidance/ng204](http://www.nice.org.uk/guidance/ng204). Accessed December 2023.

O'Connor, A.M., Bennett, C.L., Stacey, D., Barry, M., Col, N.F., Eden, K.B., Entwistle, V.A., Fiset, V., Holmes-Rovner, M., Khangura, S., Llewellyn-Thomas, H., Rovner, D. Decision aids for people facing health treatment or screening decisions. *Cochrane Database Syst Rev*. 2009;3:CD001431. doi: 10.1002/14651858.CD001431.pub5.

O'Toole, G. *Core interpersonal skills for healthcare professionals*. 2024; Elsevier: Australia.

- Parens, E. (Ed). *Surgically shaping children: Technology, ethics, and the pursuit of normality*. John Hopkins University Press: 2006.
- Pinquart, M. Parenting stress in caregivers of children with chronic physical conditions: A meta-analysis. *Stress Health*. 2018;34:197-207. doi: 10.1002/smi.2780.
- Renkema, R.W., ERN CRANIO working group on craniofacial microsomia. European guideline: Craniofacial microsomia. *J Craniofac Surg*. 2020;31 Suppl 8:2385-2484. doi: 10.1097/SCS.0000000000006691.
- Riessman, C. *Narrative methods for the human sciences*. Sage; 2008.
- Rollnick, S., Miller, W.R., Butler, C.C. *Motivational Interviewing in Health Care: Helping patients change behaviour*. 2008. London: The Guilford Press.
- Ronde, E.M., Esposito, M., Lin, Y., van Etten-Jamaludin, F.S., Bulstrode, N.W., Breugem, C.C. Long-term complications of microtia reconstruction: A systematic review. *J Plast Reconstr Aesthet Surg*. 2021;74(12): 3235-3250. doi: 10.1016/j.bjps.2021.08.001.
- Ronde, E.M., Esposito, M., Lin, Y., van Etten-Jamaludin, F.S., Bulstrode, N.W., Breugem, C.C. Long-term aesthetics, patient-reported outcomes, and auricular sensitivity after microtia reconstruction: A systematic review. *J Plast Reconstr Aesthet Surg*. 2021;74(12): 3213-3234. doi: 10.1016/j.bjps.2021.08.004.
- Ronde, E.M., van der Lucht, V.A.P., Lachkar, N., Ubbink, D.T., Breugem, C.C. Stakeholder views on information needed in a patient decision aid for microtia reconstruction. *Cleft Palate Craniofac J*. 2024. doi: 10.1177/10556656221146584.
- Rooijers, W., Tio, P.A.E., van der Schroeff, M.P., Padwa, B.L., Dunaway, D.J., Forrest, C.R., Koudstaal, M.J., Caron, C.J.J.M. Hearing impairment and ear anomalies in craniofacial microsomia: A systematic review. *Int J Oral Maxillofac Surg*. 2022;51(10): 1296-1304. doi: 10.1016/j.ijom.2022.01.005.
- Stiggelbout, A.M., Pieterse, A.H., De Haes, J.C. Shared decision making: concepts, evidence, and practice. *Patient Educ Couns*. 2015;98: 1172-1179. doi: 10.1016/j.pec.2015.06.022.
- Stock, N.M., Costa, B., Parnell, J., Johns, A.L., Crerand, C.E., Feragen, K.B., Stueckle, L.P., Mills, A., Magee, L., Hotton, M., Tumblin, M., Schefer, A., Drake, A.F., Heike, C.L. A conceptual thematic framework of psychological adjustment in caregivers of children with craniofacial microsomia. *Cleft Palate Craniofac J*. Online ahead of print; 2024. doi: 10.1177/10556656241245284.
- Stock, N.M., Crerand, C.E., Johns, A.L., McKinney, C.M., Koudstaal, M.J., Drake, A.F., Heike, C.L. Establishing an international interdisciplinary research network in craniofacial microsomia: The CARE program. *Cleft Palate Craniofac J*. 2023. doi: 10.1177/10556656231176904.
- Stock, N.M., Marik, P., Magee, L., Aspinall, C.L., Garcia, L., Crerand, C.E., Johns, A.L. Facilitating positive psychosocial outcomes in craniofacial team care: Strategies for medical providers. *Cleft Palate Craniofac J*. 2020;57(3): 333-343. doi: 10.1177/1055665619868052.
- Thornton, M., Harcourt, D., Deave, T., Kiff, J., Williamson, H. "Have we done enough?" A cross-condition exploration of the experiences of parents caring for a child with an

appearance-affecting condition or injury. *Developmental Neurorehabilitation*. 2021;24(6): 418-428. doi: 10.1080/17518423.2021.1901150.

Tong, A., Sainsbury, P., & Craig, J. Consolidated criteria for reporting qualitative research (COREQ): A 32-item checklist for interviews and focus groups. *International J Quality Health Care*. 2007;19(6), 349–357. <https://doi.org/10.1093/intqhc/mzm042>.

Truong, M.T., Liu, Y.C., Kohn, J., Chinnadurai, S., Zopf, D.A., Tribble, M., Tanner, P.B., Sie, K., Chang, K.W. Integrated microtia and aural atresia management. *Policy Practice Rev*. 2022;9 doi:10.3389/fsurg.2022.944223.

<b>Table 1. Characteristics of Caregivers of Children with Craniofacial Microsomia</b>	<b>N (%) (N=62)</b>
Age at interview in years (mean, SD)	40.2 (11.9)
Gender	
Mothers	57 (91.9)
Fathers	5 (8.1)
Interview language	
English	50 (80.6)
Spanish	12 (19.4)
Race/ethnicity*	
White (not Hispanic/Latinx)	38 (61.3)
Hispanic/Latinx	16 (25.8)
Multi-racial	6 (9.7)
Other race	1 (1.6)
Marital status	
Married	46 (74.2)
Divorced	5 (8.1)
Never married (single)	5 (8.1)
Living with partner	2 (3.2)
Widowed	2 (3.2)
Separated	1 (1.6)
Unknown	1 (1.6)
Education status	
Unknown	1 (1.6)
<12 years (no high school diploma)	8 (12.9)
12 years high school/diploma/GED	4 (6.5)
Some college/associate degree	15 (24.2)
Completed university/college	34 (54.8)
Insurance Status	
Private	35 (56.5)
Public	27 (43.5)

\*listed as “other” in order to preserve privacy

<b>Table 2. Characteristics of the Child</b>	<b>N (N=62)</b>	<b>%</b>
Age at interview in years (mean, SD)	10.4 (4.4)	
Gender		
Male	32	51.6%



Female	30	48.4%
Phenotype features (may have more than one)		
Microtia	61	98.4%
Atresia	51	82.3%
Mandibular hypoplasia	45	72.6%
Facial nerve palsy	15	24.2%
Epibulbar dermoid	8	12.9%
Lateral oral cleft	7	11.3%
Eyelid coloboma	4	6.5%
Cleft palate	4	6.5%
Cleft lip	1	1.6%
CFM diagnosis terms used by caregivers (can be more than one)		
Microtia	55	88.7%
Hemifacial microsomia (HFM)	28	45.2%
Craniofacial microsomia (CFM)	27	43.5%
Goldenhar Syndrome	13	21.0%
Oculo Auriculo Vertebral Syndrome (OAVS)	5	8.1%
Other CFM diagnosis	2	3.2%
Seen at a craniofacial clinic		
No	13	21.0%
Yes	46	74.2%
Unknown	3	4.8%
Hearing aid use (ever)		
Yes	49	79.0%
Age at first use of hearing aid in years (mean, SD)		
	2.5 (3.5)	
Seen by a subspecialty provider		
	61	98.4%
Number of providers seen (mean, SD)		
	9.6 (4.8)	

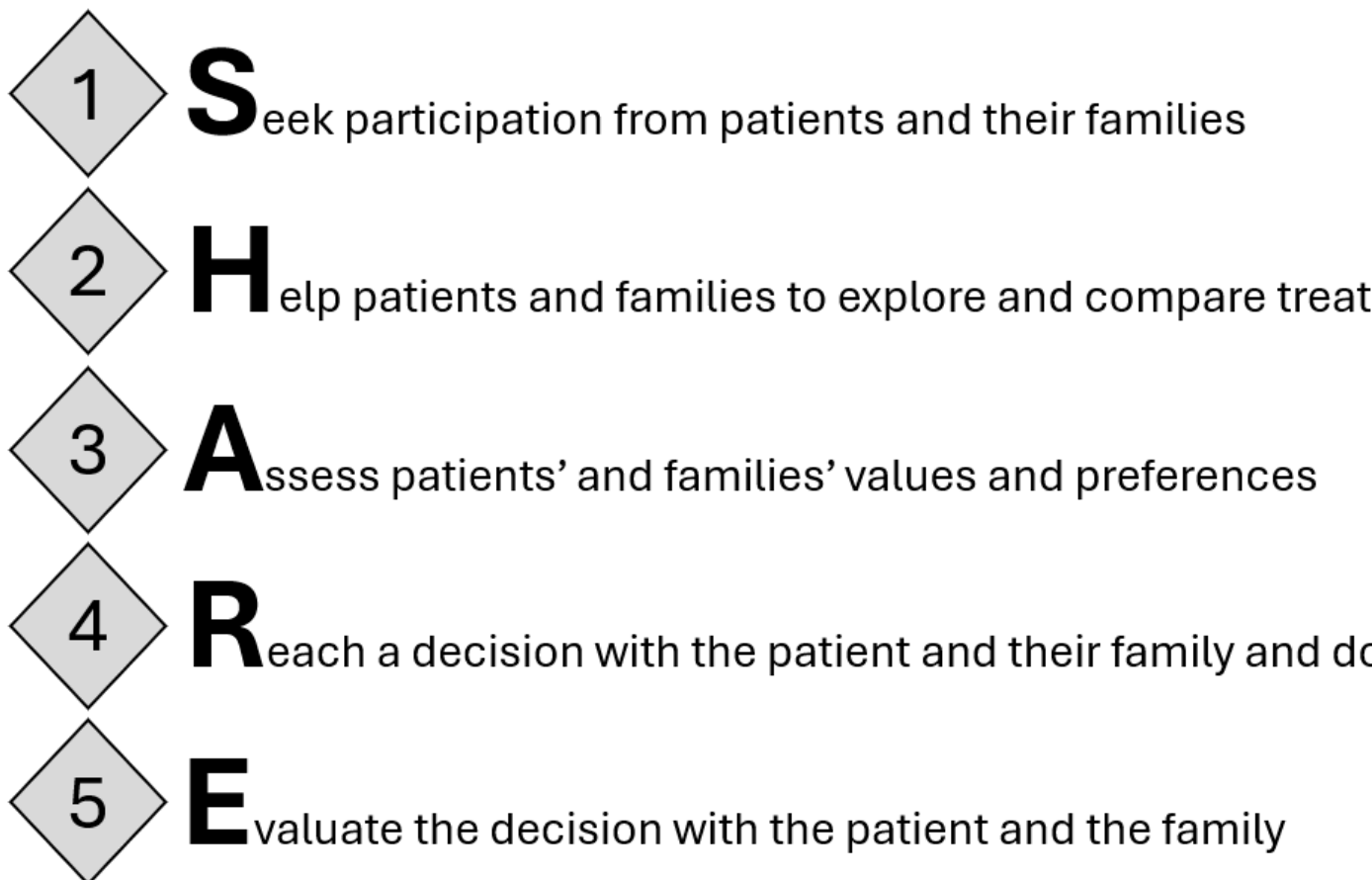
**Table 3. Craniofacial surgeries reported by caregivers for their child ne by participants' children**

	N= 62	%
Children who had undergone surgery	60	96.8%
Number of surgeries per participant (min-max)	5.0 (1-21)	
Ear reconstruction	24	38.7%
Skin tag removal	24	38.7%
Tympanostomy tubes and tympanoplasty	20	32.3%
Adenoidectomy and/or tonsillectomy	17	27.4%
Dental restoration/extraction	16	25.8%
Bone anchored hearing aid abutment surgery	13	21.0%
Aural atresia repair	8	12.9%
Lower jaw surgery	6	9.7%
Lateral oral cleft surgery	6	9.7%
Other ophthalmologic surgery	6	9.7%
Removal of epibulbar dermoid	5	8.1%
Cleft lip and/or cleft palate surgery	4	6.5%
Tracheostomy surgery	3	4.8%
Coloboma surgery	2	3.2%

<b>Barriers</b>	<b>Recommendations</b>
Stigmatising or negative comments which emphasise the child's differences and/or increase the stigma already felt by caregivers	Use of neutral language; offering reassurance that many families have similar feelings about their child's diagnosis; screening for high level caregiver concerns
Implicit or explicit presumption that surgery is 'the only solution' to caregivers' concerns	Avoiding terms such as 'repair' or 'fix'; ensuring that 'no treatment' is presented as a valid option
A lack of accessible information about all available treatment options/healthcare providers	Provision of evidence-based, comprehensive, and unbiased information about all available treatment choices and (if applicable) options for families to other sources of information, healthcare providers, and support resources; staying up to date with the latest multidisciplinary research
Limited engagement with patient and family preferences about treatment and limited communication of potential risks and outcomes	Enactment of shared decision-making principles, including patient and family input, setting and prioritising, risk management, supporting families with decision-making aids, tools/interventions, and/or decision-making processes
Lack of opportunities to connect with other families affected by CFM	Referring families to verified advocacy groups, online support groups, and peer-led services
Lack of engagement with child treatment preferences	Provision of developmentally appropriate information; facilitating family input to share their opinions and concerns
Lack of documentation of decisions and short- and long-term outcomes	Utilisation of a formal system for documenting, communicating, and reviewing decisions; publishing outcomes to inform the broader CFM community
Fat graft surgery	2 3.2%
Nerve surgery	2 3.2%
Speech surgery	2 3.2%
LeFort I advancement	1 1.6%
Rhinoplasty or septoplasty	1 1.6%

**Table 4:** Barriers and Recommendations for Effective CFM Treatment Decision-Making

**Figure 1:** The 'SHARE' Model of Shared Decision-Making



Adapted from the D