

Psychosocial and health-related experiences of individuals with microtia and craniofacial microsomia and their families: Narrative review over two decades

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Running Head: Microtia and craniofacial microsomia psychosocial and health-related experiences

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Abstract

Objective This paper describes 20 years of microtia and craniofacial microsomia (CFM) psychosocial and healthcare studies and suggests directions for clinical care and research.

Design A narrative review of papers January 2000-July 2021 related to psychosocial and healthcare experiences of individuals with microtia and CFM and their families.

Results Studies (N = 64) were mainly cross-sectional (69%), included a range of standardized measures (64.1%), and were with European (31%), American (27%), or multinational (23%) samples. Data were generally collected from both patients and caregivers (38%) or patient self-report (35%). Sample sizes were 11-25 (21%), 26-50 (19%), 51-100 (22%), or over 100 (38%). Studies addressed 5 primary topics: 1) Healthcare Experiences, including Medical Care, Hearing Loss/Amplification, Diagnostic Experiences, and Information Preferences; 2) Psychosocial Experiences, including Teasing, Behavioral Adjustment, Psychosocial Support, and Public Perception; 3) Neurocognitive Functioning and Academic Assistance; 4) Pre- and Post-Operative Psychosocial Outcomes of Ear Reconstruction/Canaloplasty; and 5) Quality of Life and Patient Satisfaction.

Conclusions Care involved multiple specialties and was often experienced as stressful starting at diagnosis. Psychosocial and neurocognitive functioning were generally in the average range, with possible risk for social and language concerns. Coping and resiliency were described into adulthood. Satisfaction and positive benefit of ear reconstruction/canaloplasty were high. Care recommendations include increasing: hearing amplification use, microtia and CFM knowledge among providers, efficient treatment coordination, psychosocial support, academic assistance, and advances to minimize surgical scarring. This broad literature overview informs clinical practice and research to improve psychosocial outcomes.

Keywords: craniofacial microsomia; microtia; healthcare experiences; psychosocial adjustment

Introduction

Craniofacial microsomia (CFM) occurs in approximately 1 in 3,000 to 5,600 births and is characterized by underdevelopment of the ear(s), mandible, facial soft tissue, facial nerve, and orbit and can also include vertebral, renal, cardiac, and additional extracranial anomalies.^{1,2} Due to similarities in presentation with disruption in the embryonic first and second pharyngeal arches, microtia can be conceptualized as a minimally impacted point in the CFM diagnostic spectrum encompassing hemifacial microsomia (HFM), oculo-auriculo-vertebral syndrome (OAV), and Goldenhar Syndrome.³ In addition, a substantial proportion of individuals with presumed isolated microtia may have features of CFM identified on further examination.⁴ Along with audiology intervention, treatment for microtia often includes multi-stage ear reconstruction and additional CFM treatment can include surgeries for the mandible and maxilla, facial nerves, and soft tissues with intervention for feeding, breathing, speech, vision, dental, and vertebral anomalies.⁵ The functional, sociocultural, and psychological importance of ears as a shared feature of microtia and CFM has been highlighted throughout history and ear reconstruction dates to 3000 BCE.⁶ Healthcare providers continue to focus on improving treatment for patients with microtia and CFM, such as investigating tissue engineering and 3D-printed frameworks for auricular reconstruction.⁷ Beyond navigating medical care, patients and families also have to adapt to psychosocial challenges.⁸

Psychosocial components of care are increasingly recognized as necessary in integrated microtia and CFM healthcare. For example, care pathways for children with microtia in the UK⁹ and a proposed team model in Egypt¹⁰ both include psychosocial support. In the USA, craniofacial teams often provide care for patients with CFM and follow the American Cleft Palate-Craniofacial Care Parameters (2018),¹¹ which mandates incorporation of psychological and social services into treatment from diagnosis into adulthood. Recent European CFM care guidelines recommend access to psychological support, linkages to family support networks, assistance with school advocacy, psychosocial screening completed at key transitional time points, and incorporation of validated outcome measures into care.⁵

Building on these comprehensive care recommendations, providers can better respond to patient and family needs by gaining a greater understanding of the context of CFM-related healthcare and psychosocial experiences. Since 2000, there have been numerous papers addressing varied aspects of patient and family experiences; however, it can be challenging for

providers and researchers to integrate findings across the wide range of relevant care domains, which can result in a fragmented understanding of the patient experience. As the aims and methodology of systematic reviews can inadvertently exclude studies that have topical relevance,¹² this study allows for a more inclusive design through the use of a narrative review. Following the standards set in the Scale for the Assessment of Narrative Review Articles,¹³ this paper aims to synthesize psychosocial and healthcare findings into a foundational understanding of current knowledge and outline directions for clinical care and future research.

Method

PubMed and PsychInfo were searched for articles published (including pre-press papers published online) between January 2000-July 2021 for all fields with the diagnostic/surgical terms of: CFM, HFM, microtia, anotia, aural atresia, OAV syndrome, Goldenhar syndrome, ear reconstruction, and auricular reconstruction. Searches were paired with topical terms of: psychosocial, psychological, quality of life, teasing, neurodevelopment, academic, special education, patient satisfaction, and patient reported outcomes. Reference lists identified additional studies. After reviewing abstracts and/or the full article texts, studies were included if self, caregiver, and/or proxy reports of psychosocial and/or healthcare experiences of microtia or CFM were reported. If studies had participants across multiple diagnoses, studies were included if the results of those with microtia or CFM were presented separately. Other than being published in a peer-review indexed journal, there were no language or methodology exclusion criteria in order to capture the breadth of the existing literature. Study design, setting, participants, measures, and key findings were summarized and grouped by primary topic.

Results

Of the 1,464 papers identified, 293 were duplicates, 1,116 were not directly related to CFM and/or psychosocial or healthcare experiences (e.g., surgical outcomes), and 9 additional papers were identified from reference lists. The 64 included studies were grouped by main topic into 5 domains with details of key findings reported in supplemental tables. Participants were primarily younger than age 25 years (57%), followed by samples including children and adults (30%), with fewer studies focusing on adults (13%). Study characteristics are shown in Table 1.

Healthcare Experiences

Papers in this domain addressed medical care, hearing loss and amplification, diagnostic experiences, and information preferences (see Table 2 and Supplemental Table 1).

Medical Care

The complexity of CFM medical care was highlighted in an international sample of English-speaking caregivers and adults with CFM ($N = 51$) who reported an average of 7.7 healthcare specialists and a mean of 4 surgeries (range 1-30).¹⁴ In a sample from multiple American centers, young children with CFM ($N = 92-108$) had seen 3 to 9 specialty providers, 28% had at least one surgery before age 24 months, and 70% received early intervention services.¹⁵⁻¹⁷ In an American sample ($N = 20$), 75% had received multidisciplinary team care and 45% participated in at least a year of speech, physical, and/or occupational therapies.¹⁸ In studies tracking a multisite North American sample of children with CFM ($N = 107-121$), 60%-79% received intervention services,¹⁹⁻²¹ including speech therapy (58%) and physical therapy (36%). Although communication difficulties were reported by more than two thirds of those with aural atresia (total $N = 254$), only about half participated in speech therapy.²²⁻²⁴ While not noted in many CFM studies, approximately 10% to 15% of participants had a cleft lip/palate (CL/P) in some of the samples,^{15,21,25} which is consistent with what has been previously reported.²⁶

Hearing Loss and Amplification

Microtia and CFM are associated with hearing loss; however, reports of amplification use vary widely, despite a growing understanding of the benefits of hearing aids even for unilateral hearing loss.^{27,28} Additionally, an American chart review study ($N = 68$) found that out of 13 demographic, surgical, and medical variables, only treatment to improve hearing predicted reports of positive adjustment for the third of pediatric patients with CFM who had psychosocial concerns.²⁹ In a sample of young children with CFM from multiple American centers, over 90% had hearing loss and 51% used hearing aids.¹⁵⁻¹⁷ In a smaller American sample, 90% had hearing loss.¹⁸ This was similar to a sample from the USA, Colombia, and Peru ($N = 169$), with 90% with hearing loss and 54% using hearing aids.³⁰ In a smaller Australian study, all children with aural atresia ($n = 10$) had amplification in place by a median age of 5 months (range 1-58 months) and they all had FM systems.³¹ However, hearing loss was somewhat lower at 70% with only 23% use of hearing aids in a multisite North American study ($N = 107-121$) of children with CFM^{20,21} with 33% receiving hearing services.¹⁹ For a group of American children ($N = 74$), amplification was in place for all children with bilateral aural atresia, but only 4% with unilateral aural atresia had amplification, despite maximal conductive hearing loss in the affected ear.²² Kesser et al. (2013) reported similar patterns of frequent hearing loss with low amplification use

(15%) in another USA group with unilateral aural atresia ($N = 40$).²³ In a follow-up study of individuals with hearing loss due to unilateral aural atresia ($N = 140$), 27% had amplification and 26% had FM systems.²⁴ The ongoing impact of hearing difficulties was reported by caregivers,³² adolescents and young adults,³³ and adults, particularly in work settings ($N = 15$).³⁴

Diagnostic Experiences

Formative experiences of learning of diagnosis, etiology beliefs, and related suggestions for providers have been described in several samples, including Dutch parents ($N = 84$),³⁵ primarily Latinx parents in the USA ($n = 87$),³⁶ mothers in 3 USA states ($N = 20$),¹⁸ and an international online group of English-speaking caregivers ($n = 42$).^{8,32} Across these studies, diagnoses were primarily provided around the time of birth by pediatricians (35%-46%), with the interaction often described negatively by caregivers without specific information discussed. For example, only a third of mothers were satisfied with the information provided at the time of diagnosis and only a fifth felt they received excellent answers about treatment.¹⁸ Parents across these studies described a range of negative emotions, including anxiety, shock, grief, guilt, confusion, and concern for the future. Caregivers' etiology beliefs included perceived medical/genetic explanations, folk beliefs, and religious factors and around a third were uncertain about etiology.^{8,36} Adults with CFM ($n = 9$)¹⁴ were unsure of cause and had similar explanations as caregivers. In a Chinese sample ($N = 410$), children's awareness of microtia was generally at age 3 (37%) or age 4 (19%) years.³⁷ Awareness of CFM diagnosis was also reported at a mean age of 3 years for an English-speaking online sample¹⁴ and mean age of 4.4 years in a multinational study.³⁰ Children with microtia ($n = 62$)³⁶ most often said they didn't know the etiology, described that something was "wrong", or that this was how they were born.

Information Preferences

Caregivers' preferences for topics they would like healthcare providers to discuss with them included hearing, development, reconstructive surgery, maxillofacial surgery, genetics, psychological adjustment, treatment steps, medical specialties involved in care, financial and medical information, as well as clarity regarding surgical options.^{32,35} Parents appreciated providers who used clear communication, patient-focused approaches, and had a positive attitude.³² Suggestions for providers included timely referrals, improved CFM knowledge across providers, and better coordination and communication about CFM.³² Adolescents ($N = 11$) noted they wanted to be provided with treatment information and included in decision making from a

young age.³³ Norwegian adults with Goldenhar syndrome ($n = 7$) discussed experiencing different surgical expectations than the medical team and wanting more information and support before and after surgeries.³⁸ In keeping with caregivers' priorities for CFM research,¹⁴ Dutch providers recently surveyed caregivers ($N = 37$) about attitudes toward ear reconstruction options.³⁹

Psychosocial Experiences

Psychosocial experiences covered teasing, behavioral adjustment, psychosocial support, and public perception (see Table 3 and Supplemental Table 2).

Teasing

As microtia and CFM involve visible differences, multiple investigators have addressed experiences of teasing. Teasing was reported by 61% of a Chinese sample³⁷ and for 41% of a multinational sample starting between ages 5-6, mostly by classmates at school.³⁰ In a young group of American children with microtia ($N = 28$), the mean age of teasing onset was age 3.8 years by parent report and age 4.6 years by self-report.⁴⁰ In that sample, 100% of the 6–10-year-olds reported experiencing teasing before ear reconstruction in contrast to 31% of preoperative teasing for those ages 3-5.⁴⁰ Luquetti et al. (2018) found 43% reported teasing in a CFM sample with teasing onset at a mean age of 6 years, most often at school with name calling.¹⁴ In a UK study ($N = 62$), 85% of participants experienced teasing prior to ear reconstruction.⁴¹

Adolescents and young adults with CFM reported that teasing was related to lower self-esteem and feeling isolated, with females noting greater difficulties in feeling different and some covering their ears with their hair.³³ As adults, ongoing anxiety about their ears being seen in different settings was reported in a UK study.³⁴ Norwegian adults with CFM also reported negative social experiences, such as teasing, and discussed ways they continue to cope with social situations to feel a greater sense of belonging.³⁸

Behavioral Adjustment

Behavioral adjustment studies included parental reports using the Child Behavior Checklist (CBCL)⁴² with some studies also including teacher report and self-report with additional measures. A chart review of CBCL scores included a subgroup with HFM ($n = 47$) with scores in the average range.⁴³

A large sample of individuals in Beijing ages 5-37 ($n = 410$) with microtia and their parents ($n = 356$) participated in a study prior to ear reconstruction.³⁷ In Shanghai, Li et al.

(2010) included individuals ages 5-50 ($n = 170$) with microtia before ear reconstruction and parents ($n = 91$) in comparison to controls matched by age and gender ($n = 264$) and their parents ($n = 97$).⁴⁴ For both studies, elevated concerns were reported relative to controls and by cut-off scores (20%-37%), particularly for males; however, standard scores were not reported to interpret clinically. Females comprised approximately a quarter of the sample in these studies and Li et al. (2010) stated this reflected both the higher rate of microtia in males and a lower likelihood to seek ear reconstruction for females.⁴⁴ A risk factor identified by Jiamei et al. (2008) was parental report of the family impact of the diagnosis, which was rated as moderate (46%) or severe (35%) and was mirrored in the high rates of self-reported maternal distress of cases compared to control mothers.³⁷ In a more recent study in Beijing with 53 young children ages 4-7 with bilateral microtia who began using a bone conduction hearing device in infancy, there were no group differences on the CBCL when compared to local norms.⁴⁵

A study in the Netherlands of individuals ages 6-20 years with congenital aural atresia ($N = 29$) found that all scores on the CBCL and the Youth and Adult Self-Report forms were in the average range with few differences by age.⁴⁶ The remaining studies included comparisons of North American cases and controls accounting for sociodemographics from early childhood to adolescence. Caregivers of children with CFM ($n = 89$) and matched controls ($n = 72$) 36-42 months completed the CBCL.⁴⁷ Caregivers and teachers reported on children with HFM ($n = 136$) and matched controls ($n = 568$) drawn from 26 craniofacial centers in the USA and Canada at ages 5-12.²⁵ This sample was followed up at ages 10-17 with caregiver, teacher, and self-report.⁴⁸ Across these 3 studies, mean scores by parent report for internalizing and externalizing behaviors were average and generally no different or sometimes indicated fewer concerns than measure norms and controls. In contrast to parents, teachers of school-age children reported greater internalizing and total problems and lower social competence for cases compared to controls.²⁵ For young children with CFM, parents of children reported more concerns with anxiety, stress, and getting along with peers.⁴⁷ For school age children, parents reported more social problems than controls.²⁵ Similarly, adolescents self-reported more social problems relative to controls.⁴⁸

Psychosocial Support

To help cope with the range of potential diagnosis-related stressors, caregivers identified sources of support in their spouses, families, healthcare providers, self-reliance, religious beliefs,

and online resources.^{32,36} Participation in mental health services was reported for 14% of children with CFM in a large multisite study.¹⁹ Adolescents and young adults stated they felt their diagnosis and related experiences had an overall positive impact on their character development and advised being confident and open about diagnosis as supports.³³ Adults also emphasized openness as supportive and developing an identity separate from their diagnosis, with some noting that downward comparisons helped their coping.³⁴ Adults with CFM reported on the importance of family support and friends to be open and authentic with.³⁸ Advice offered by adults as supportive included ensuring informed medical decision making, having self-confidence, reframing when teased, and open communication about their diagnosis.³²

Studies also document that families frequently seek support and information online. A study of the top 100 searched microtia websites found similar quality ratings given by physicians and families.⁴⁹ Social media is a large source of online medical information as illustrated in a content analysis of 254 posts from online Facebook groups, which found that half of the posts were related to seeking medical guidance.⁵⁰

Public Perception

Three studies focused on general public perception of microtia and CFM using online recruitment from the general public. One study examined the perceived burden of microtia and found microtia without hearing loss was rated as healthy, while microtia with unilateral hearing loss had a similar trade-off score as monocular blindness.⁵¹ Another public perception study used visual analogue scale ratings of photos of ears without microtia, with microtia, and after ear reconstruction on social characteristics.⁵² There were trends showing areas of lower ratings in friendliness, health, intelligence, and success for those with ears with microtia and no differences between reconstructed ears and the ears without microtia.⁵² Almadani and Gilardino (2020) reported on ratings based on photos of children with CFM before and after mandibular distraction with improved utility, psychosocial acceptance, and cost effectiveness after distraction.⁵³

Neurocognitive Functioning and Academic Assistance

Neurocognitive Functioning

Seven studies included standardized assessment of neurocognitive functioning (see Table 3 and Supplemental Table 2). A Swedish study of children with OAV ($N = 20$) included several measures of cognitive and adaptive functioning as part of a study focusing on Autism Spectrum

Disorder (ASD) and reported that about half of the sample had average intelligence.⁵⁴ They found that 10% met diagnostic criteria for ASD and, of those with radiologic imaging, 64% had structural cerebral anomalies or abnormalities in white or gray matter, which was posited to be linked to embryonic brain development.⁵⁴ An Australian study comparing children with aural atresia ($n = 10$) who had amplification since infancy with peers ($n = 10$) reported both groups had a median score in the 82nd percentile for nonverbal cognitive functioning with no group differences in reading or language measures.³¹

Two series of longitudinal studies examined cognitive functioning domains in children with CFM. These studies analyzed results by case and control comparisons and by CFM phenotype accounting for sociodemographic variables. The first series was completed in 6 American craniofacial centers with matched controls for cognitive, motor, and language development. Cases ($n = 108$) and controls ($n = 84$) were evaluated between 12-24 months with average range mean scores and no group mean differences, with an area of delay found in 21% of children with CFM.¹⁶ (While there were no differences in facial expressiveness between children with and without craniofacial microsomia when scored by observers, computer vision-based scoring identified less expressiveness in cases with microtia with mandibular hypoplasia and other associated features of CFM.^{55, 56}) At follow-up between 36-42 months, cases ($n = 92$) and controls ($n = 76$) had average range cognitive and motor mean scores; however, children with CFM had 2 lower language subtest scores and 39% had an area of delay.¹⁷

The second study series was based on a sample described in Werler et al. (2009) of 230 case participants with HFM and 678 matched controls from 26 craniofacial centers in the USA and Canada.⁵⁷ The first study of cases ($n = 136$) and controls ($n = 568$) was conducted at ages 5-12 and measured receptive language and visual motor integration, as well as parent and teacher report of academic competency.⁵⁸ All participants had average range mean scores; however, children with CFM had lower scores relative to controls.⁵⁸ The second study included 121 cases and 315 controls at ages 11-17 and utilized several brief measures of cognitive functioning and achievement; scores were in the average range with relatively lower reading and writing scores for children with CFM than controls.²⁰ In a third study, children ages 11-17 ($n = 107$ cases and $n = 306$ controls) completed measures of language, articulation, intelligibility, and parent and teacher reported communication.²¹ Disordered speech was found for 35% of the sample with CFM along with lower expressive language scores than controls.²¹

Academic Assistance

The relatively lower performance in neurocognitive skills than their peers suggests that academic assistance is needed for a higher proportion of children with CFM. In a multinational study of CFM, about half of American children had Individualized Education Programs (IEPs) with resource program support (16%) or special education classes all day (17%) compared to only 4% of South American students placed outside of general education.³⁰ Among a North American sample of children with CFM, 31% received special education academic services.¹⁹ Academic supports were provided for a third of American children with aural atresia, which corresponded to the 23% of parents who reported learning concerns.²² In an American sample with microtia, 30% reported receiving learning interventions.¹⁸ In another USA study with unilateral aural atresia, 48% had IEPs and 20% received special education academic services.²³ Rates were somewhat lower in an aural atresia sample, likely due to inclusion of preschool children, with IEPs for 36% and special education for academic services for 16%.²⁴ In Australia, children with aural atresia were all in general education placements.³¹ In a group of individuals with congenital aural atresia ($N = 29$) in the Netherlands, a third of the sample had been retained a year in school despite receiving classroom accommodations (79%), speech therapy services (61%), and hearing amplification (45%).⁴⁶ In a Dutch study of parents of children with HFM ($N = 31$), parental stress, although lower than general population norms, was associated with children's learning difficulties.⁵⁹ School advice offered by caregivers of children with CFM included addressing concerns about hearing, teasing, and difficult social interactions and increased peer awareness of CFM.³² A pair of papers examined caregiver reports of school and community activities participation.^{19,60} While there were few differences between caregivers of children with CFM and healthy peers who had a history of participating in intervention services, there was a pattern of less involvement in school and community activities compared to peers with no service history, including less participation in organized physical activities, trips, clubs, and spending time with peers informally.^{19,60}

Pre- and Post-Operative Psychosocial Outcomes of Ear Reconstruction/Canaloplasty

A few studies aimed to measure both baseline and postoperative outcomes with different methodologies (see Table 4 and Supplemental Table 3). Two cross-sectional studies asked participants at a single time point to respond about what they remembered prior to ear reconstruction as well as how they felt postoperatively. In a UK sample ($N = 62$) aged 4-86 with

a range of medical diagnoses who primarily had autologous ear reconstruction, participants reported feeling less self-conscious and experiencing less teasing, improved social life, increased self-confidence, and rated their ears positively.⁴¹ In a similarly designed German study of participants ($N = 68$) aged 12-58 primarily with microtia who had autologous ear reconstruction, there was no change after surgery based on recall of preoperative functioning on a standardized measure of performance ability, self-esteem, and psychosocial attitude.⁶¹ Questions specific to perioperative experiences identified concerns for ear appearance, surgical complications, chest scarring, and length of hospital stay. Around a quarter of participants reporting postoperatively stated their thoracic scars were not acceptable.⁶¹

The remaining studies were prospective with relatively small samples, including 2 from Germany with wide age ranges. Participants ($N = 21$) with microtia before and after alloplastic ear reconstruction reported improvements postoperatively on several standardized measures, including physical functioning, health perception, social functioning, mental wellbeing, mood, self-perception, and social acceptance.⁶² Steffen et al. (2010) reported on participants with microtia who either had autologous ear reconstruction ($n = 16$) or did not have surgery ($n = 23$) and replicated the Steffen et al. (2008) cross-sectional result of no changes on a standardized psychological measure postoperatively.⁶³ Steffen et al. (2010) reported that the required hospital length of stay for healthcare guideline and billing purposes (e.g., admission time of 15-16 days across 3 hospital stays) was perceived as a stressor for 77% of those who decided against surgery and 50% of the group who proceeded with surgery.⁶³ Those who did not pursue surgery had a more positive psychosocial attitude than both the pre- and postoperative scores of the surgical group,⁶³ which the authors noted as important knowledge for parents of children who decline surgery.

Several prospective studies focused on children, adolescents, and young adults. Two studies included younger children ages 3-10 with microtia and their caregivers before and after alloplastic ear reconstruction. The first study found average range scores on a standardized behavioral measure for participants ($N = 23$) before and after surgery with decreased anxiety and depression and increased postoperative social skills.⁶⁴ However, the older group had higher negative emotions preoperatively than the younger group and parents tended to underestimate younger children's negative emotions and diagnostic awareness.⁶⁴ The second study focused on teasing, which was associated with feeling sad, worried, and mad before ear reconstruction and

with hiding their ear(s) after surgery.⁴⁰ By self-report, teasing decreased by half postoperatively for those ages 6-10 and from 31% to 8% for those ages 3-5, with parents also reporting lower rates of postoperative teasing than children.⁴⁰ A study of Saudi Arabian adolescents with microtia ($n = 20$) who had autologous ear reconstruction and matched peers ($n = 20$) found higher body image concerns and lower emotional and social functioning and life satisfaction for those with microtia preoperatively.⁶⁵ Postoperatively, adolescents with microtia showed significantly improved scores across all areas and improvement in body image explaining 52% of the variance in life satisfaction.⁶⁵ In a South Korean study on the impact of canaloplasty, Cho et al. (2020) reported that self-consciousness in participants with microtia and aural atresia ($N = 34$) was higher in adolescents preoperatively and that all ages showed improvements after surgery.⁶⁶ They found no relationship between postoperative audiological improvement and psychosocial functioning.⁶⁶

Quality of Life and Patient Satisfaction

Multiple papers addressed health-related quality of life and satisfaction with treatment and surgery (Table 5 and Supplemental Table 4). Using the Pediatric Quality of Life Scale 4.0,⁶⁷ a study of children ages 5-12 with CFM ($n = 136$) and controls ($n = 568$) and their mothers found no group differences based on self-report; further, 22% of mothers of children with CFM reported scores below clinical cutoff compared to 15% of control mothers.⁶⁸ Using the Hearing Environments and Reaction on Quality of Life Questionnaire,⁶⁹ children with aural atresia in Australia were significantly lower for the Total, Environment, and Feelings scores, indicating more negative feelings about hearing loss across settings.³¹

The Glasgow Benefit Inventory (GBI)⁷⁰ and Glasgow Children's Benefit Inventory (GCBI)⁷¹ were used in multiple studies, which is scored from adverse effects (-100) to positive effects (+100) with 0 representing no benefit. In a Chinese sample of children ($N = 53$) with bilateral microtia, the parent-reported GCBI of using soft-band bone conducted hearing devices was positive ($M = 33$).⁴⁵ Similarly, in another Chinese sample of children with bilateral microtia ($N = 12$), the GCBI of a bone conduction hearing aid implant with or without auricular reconstruction was also positive ($M = 40$).⁷² A UK sample ages 9-21 with isolated microtia ($n = 40$) or as part of a syndrome ($n = 15$) reported benefit on the GBI from ear reconstruction ($M = 48$), with no variation by sex or age at surgery.⁷³ GBI scores were higher for those who had

microtia as part of a syndrome and there was a moderate correlation between surgical outcome ratings and benefit scores.⁷³

Several German samples reported benefit using the GBI and GCBI after ear reconstruction. Adolescents and adults ($n = 89$) as well as parents of children ($n = 24$) who had alloplastic ear reconstruction noted overall satisfaction in more than 75% of the sample; there was benefit by patient self-report ($M = 17$) and caregiver-report ($M = 19$) with no differences by sex.⁷⁴ In a sample of parents of children ($n = 20$) and individuals ages 16-65 ($n = 45$) with a combination of isolated, syndromic, and acquired microtia or similar auricular differences, the mean GBI/GCBI scores were similar for adults ($M = 21$) and parents ($M = 28$) for ear reconstruction, with no differences by sex.⁷⁵ Although 73% of adults and 85% of parents were satisfied with aesthetic outcomes, the authors noted that no benefits on the GBI/GCBI were reported for those that were unsatisfied with postoperative ear appearance.⁷⁵ In smaller samples of individuals with microtia ($n = 15$)⁷⁶ and HFM ($n = 6$),⁷⁷ the mean GBI/GCBI scores for ear reconstruction ranged from 24 to 40. Similar to the recommendation to minimize thoracic scarring in autologous ear reconstruction,^{61,78} Braun et al. (2013) recommended that surgeons minimize scars resulting from full-thickness skin grafts taken from groin and scalp scars from temporoparietal fascia flaps and skin from behind the contralateral ear.⁷⁶

Patient postoperative satisfaction was the focus of several studies, generally with samples with microtia who had autologous ear reconstruction. In a UK sample ages 9-19 ($N = 69$), about 84% were satisfied with their ear and areas of concern for about a third of the sample were included chest scars and inadequate pain control.⁷⁸ In a Swedish sample ages 9-23 ($N = 59$), 73% expressed aesthetic satisfaction and 24% felt happier, with no differences by sex.⁷⁹ While 90% reported satisfactory pain management, a fifth reported problems with thoracic donor region, cleaning the reconstructed ear, and wearing glasses.⁷⁹ A Chinese study with parents of children ($n = 37$) and individuals with microtia ages 13-45 ($n = 34$) reported that mean satisfaction with ear substructures was 74%, with higher satisfaction among parents and participants under age 12 years and no differences by sex.⁸⁰ The authors noted lower patient satisfaction for those with higher Body Mass Index as possibly related to blunted auricular contour and more hypertrophic thoracic scarring.⁸⁰ The authors also noted higher ratings for left-sided microtia matching general higher ratings of the left side of the face.⁸⁰ Satisfaction with a bone-anchored prosthetic ear implant in a UK sample ages 17-56 ($N = 20$) found aesthetic appearance was rated positively by

85%; however, skin problems were reported by 75%, most commonly granulation tissue around abutments.⁸¹

Several studies described the development and/or translation of measures for different aspects of patient satisfaction and quality of life, which offer insight into provider and research topics of interest internationally and identify areas that are not yet covered in existing measures. Cui et al. (2018) developed a measure rating postoperative satisfaction using a photograph for 11 parts of the ear and overall satisfaction with individuals with microtia ($N = 180$) in Shanghai.⁸² Hearing was the primary focus in developing a quality of life measure titled the Congenital Aural Atresia Questionnaire with youth with atresia ($N = 140$) after ear reconstruction in Beijing.⁸³ A German translation of the Youth Quality of Life – Facial Differences Questionnaire by Patrick et al. (2002) was validated with youth with microtia.⁸⁴ Bradford et al. (2020) described developing psychosocial, aesthetic, and decision-making items that was piloted with 25 patients with microtia or CFM and their caregivers.⁸⁵ Results were reported in terms of the agreement between patients and caregivers with a trend of children with microtia expressing interest in ear reconstruction earlier than parents and children with CFM wanted to address facial asymmetry sooner than ear reconstruction.⁸⁵

Klassen et al. (2018) described the process developing the EAR-Q⁸⁶ by interviewing youth ($N = 25$) and noted about a third of concerns were related to ear appearance, with positive comments made after ear reconstruction.⁸⁷ Physical concerns were less frequent (9%) and focused on hearing and adverse effects shortly after surgery.⁷⁸ The remaining items (63%) were related to psychological, social, and school quality of life with trends noted for generally negative psychological and school items and more positive social items.⁷⁸ Field testing of translations of EAR-Q scales reported consensus was met in 4 languages.⁸⁸ The psychometric properties of the EAR-Q self-report of Appearance and Adverse Effects scales were acceptable with a large international sample ($N = 863$) and showed a pattern of higher quality of life with higher appearance scores as well as mean post ear reconstruction appearance scores that were twice as high compared to those who were pre-surgery.⁸⁹

Discussion

This narrative review documents the breadth of microtia and CFM-related psychosocial and healthcare studies over 20 years and provides a synthesis of information across domains to inform CFM care and research.

Summary of Care

The complexity of care reported reflected the range of features associated with microtia and CFM, for example, one sample had an average of 4 surgeries ranging from 1-30 and had been seen by an average of 7 specialty providers.¹⁴ Care was often experienced as stressful starting from the time of diagnosis with little information provided about what families identified as important. Child awareness of their diagnosis generally took place at ages 3-4, corresponding to normative development of self-awareness.⁹⁰ Teasing began between ages 4-6 for a third to all individuals, mostly by classmates. Parent-reported behavioral and psychosocial functioning was generally in the average range clinically and when compared to peers. An exception was seen for up to a third of school-age to adult Chinese participants with microtia who reported clinical symptoms, such as anxiety, before ear reconstruction. While clinical psychosocial distress was generally low, social concerns were consistently reported. In North American samples, parents had concerns about their child not getting along with peers around age 3 years, with elevated parent- and teacher-reported social problems at school age and self-report of social concerns in adolescence. This was reflected in public perception studies noting areas of negative personal and social preoperative appraisals that improved after ear reconstruction or mandibular distraction. Along with some ongoing social concerns in adulthood, positive growth from diagnosis-related experiences and coping skills were described, often highlighting the benefits of open communication, diagnosis acceptance, and building confidence. Online content was noted as a growing source of support and information.

From ages 12 months to 17 years, individuals with CFM typically had average neurocognitive functioning. This pattern was within the context of high rates of intervention services and hearing aid use that may have supported development in those samples. Despite overall average performance, about 20% to 40% of children with CFM had greater relative risk for delay compared to peers and/or were below the 25th percentile (i.e., cut-off used to define a delay). With the exception of visual motor integration, delays were primarily reported in speech and language-based skills, including receptive language, intelligibility, articulation, reading, and writing. There were no consistent patterns across studies based on CFM phenotype. As in the general population, the role of socioeconomic status (SES) was identified as risk factor for delays.^{91,92} Although nearly all participants had hearing loss, there was a wide range of hearing aid use and utilization of intervention and academic support services.

Postoperative ear reconstruction concerns were often related to scarring on the chest, groin, scalp, and contralateral ear depending on the surgery. Otherwise, there was a general trend of postoperative improvement in self-image and social interactions across types of surgery, appearance satisfaction, and positive benefit ratings of varying magnitude. Parents and younger children tended to be somewhat more satisfied with ear reconstruction than adolescents and adults. However, there were a few exceptions with German findings of no differences postoperatively as well as higher psychosocial functioning reported by ear reconstruction candidates who elected not to have surgery. These findings may underscore the importance self-perception determining psychosocial adjustment.⁹³

Future Directions: Implications for Care

The inclusion of psychosocial input in recent treatment guidelines is promising and the past 20 years of CFM and microtia research suggest several avenues to improve care. Recommendations include increased psychosocial support, microtia and CFM knowledge among all providers, clear communication with a positive and reassuring attitude, and coordinated care among specialties. Given consistent difficulties reported during the initial diagnosis experience, it appears that medical training curricula and continuing education for pediatricians, delivery room providers, and other early healthcare professionals should be offered by experienced clinicians on how to appropriately inform and support families. Treatment should be coordinated across providers with frequent communication about care plans. Transparent discussions from a young age are needed to address surgical options and expectations, including use of postoperative photographs representative of the spectrum of surgical outcomes. Further, patients, families, and surgeons expressed interest in ongoing treatment innovations and refinements, including minimization of ear reconstruction scarring. Comprehensive CFM and microtia information should also be widely and easily available in multiple formats online for families and patients across developmental phases, healthcare providers, and professionals within systems of care, such as school districts.

Academic risk factors for unilateral hearing loss^{27,28} suggest that amplification and school-based assistance is likely needed for most children with microtia and CFM. Providers should ensure appropriate hearing aid use, which may include joining in healthcare policy advocacy, and classroom accommodations, such as preferential seating close to teachers. Assessment of and intervention for language-based skills is also recommended. This may include

early intervention services, language and speech therapy, and IEP academic resource specialist instruction for reading and writing. Based on overall cognitive functioning, general education instruction is appropriate for most individuals with CFM; however, up to a fifth of students may require the support of an all-day special education classroom.

Screening across disciplines for psychosocial concerns should begin in early childhood using both caregiver and self-report (along with teacher reports when possible) with a focus on the highest risk area of social functioning. Either through community referrals or as part of interdisciplinary team care, mental health services should address social skills building, coping with teasing, and self-image concerns across development. Greater access both within and independent of healthcare systems is needed for social support programs, including online and in-person peer and caregiver groups. As the CL/P literature addresses several similar concerns, there may opportunities to build on identified patient/parent-reported outcome measures,^{94,95} educational resources for medical providers,⁹⁶ and proposed intervention frameworks.⁹⁷ Experienced providers can assist in informing school-based interventions to increase diagnosis understanding among peers and empathy building. Similarly, for older adolescents and adults, healthcare providers can collaborate on workplace education programs, advocacy for accommodations, and countering possible discrimination.

Summary of Methodology

Studies in the last 20 years were largely conducted in plastic surgery and craniofacial centers in Europe and North America, which limits their generalizability for a global population given differences in healthcare systems. For example, providing the context of treatment beyond operative steps was highlighted by the contrast of alloplastic ear reconstruction in the USA as an outpatient surgery compared to 15-16 day admission over 3 stays, which was specifically identified as a stressor in a German sample.⁶³ Many studies did not provide information about participants' health and intervention history to assist in meaningfully interpreting study findings. While some studies described and accounted for SES and contextual factors in analyses, many studies did not report on these basic variables that impact psychosocial outcomes.^{91,92} Similarly, the generalizability of findings is further limited by wide variability and minimal reporting of key variables within biopsychosocial^{98,99} and ecological systems¹⁰⁰ models that heavily influence psychosocial experiences. A cultural context example was provided by Li et al. (2010) who

noted Chinese families may be less likely to seek ear reconstruction for females with microtia compared to males.⁴⁴

As previously noted in cleft and other craniofacial research, the studies in this review were largely cross-sectional and generally had small samples of convenience without multiple informants.¹⁰¹ Inherent to smaller samples making multiple comparisons is the concern for possible false positives in the findings and greater attention is needed in to how to address this concern in study design and analyses.^{102,103} Relative to the larger number of cross-sectional treatment satisfaction and benefit papers, fewer studies gathered both preoperative and postoperative assessments of functioning to help address recall bias.¹⁰⁴ Although a significant proportion of individuals with CFM undergo orthognathic surgery, there were no studies specific to this population. While about a quarter of papers included comparison groups and two thirds of studies incorporated standardized measures, the lack of appropriate comparison samples, inconsistent reporting of standard scores, and the variety of measures used makes directly comparing findings challenging. This mirrors the call in the larger craniofacial literature for additional diagnosis-specific measures^{105,106} and has begun with instruments like the EAR-Q.⁸⁹ Mixed method designs can help address methodology challenges, such as the absence of widely-used standardized measures; however, fewer than 10% of studies utilized mixed methods.

This review has limitations, including only evaluating articles indexed in PubMed and PsychInfo since 2000 and therefore may not be representative of global CFM and microtia research. Due to differences in study methods as well as the healthcare systems and participants' sociocultural contexts, definitive conclusions cannot be drawn from the data reported. Additionally, the domains included may not have captured all patient and family experiences and the comprehensive scope precluded focusing on specific conclusions in favor of broad inferences.

Future Directions: Implications for Research

This review demonstrates increasing interest in psychosocial outcomes, consistent with a recent bibliometric analysis of microtia studies.¹⁰⁷ However, additional research is needed to inform the complexities of care as well as implementation of psychosocial assessment and intervention. As previously noted in cleft and other craniofacial research, longitudinal designs with larger representative samples across informants are recommended¹⁰¹ and small sample designs and analyses need to better address possible false positives. Greater global representation

in CFM research is essential. Detailed descriptions of study participants' phenotype, service utilization, healthcare systems, SES variables, and cultural context are important to report to aid meaningful interpretation of findings.^{91,92,98-100}

As Steffen and Frenzel (2014) noted, improvements in quality of life evaluations are needed for individuals with ear anomalies, including use of standardized measures validated in multiple languages to allow for international research along with practical utility for busy clinicians to inform their practice.¹⁰⁸ As has been recommended in craniofacial research, it is useful to include both diagnosis-specific and general measures along with appropriate comparison groups, depending on the study aims.¹⁰⁹ It is important for authors to report effect sizes and descriptive statistics that allow for interpretation of clinical significance beyond statistical significance.¹¹⁰ Additional research is needed on the feasibility and efficacy of psychosocial screening and interventions for CFM. Comparisons of the psychosocial literature published in CL/P and other craniofacial conditions suggest that similar adjustment domains may apply, indicating that outcome measures and interventions could be trialed across diagnoses with modifications made as appropriate for diagnosis-specific needs.¹⁰¹

Several areas of need identified in the literature are incorporated into the Craniofacial microsomia: Accelerating Research and Education (CARE) program funded by the National Institute of Dental and Craniofacial Research with a multidisciplinary team based in the USA, UK, and Europe.¹¹¹ Building on advances in CFM and CL/P, this international research is utilizing a mixed method and multi-informant approach including individuals with CFM ages 12-45, caregivers of children 3-17, healthcare providers, and advocates. Participation will be offered globally through online recruitment. CARE aims to evaluate psychosocial concerns and key points of risk in the treatment pathway, identify predictors of psychosocial distress and resiliency, and assess current psychosocial screening and intervention provision. The program is also establishing a registry facilitating data sharing with an international community of participants and aims to provide an online dissemination platform.

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Table 1. Characteristics of 64 papers included in review of microtia and craniofacial microsomia (CFM)

Characteristic	N (%)
Geographic Area	
USA	17 (26.5%)
Multinational	15 (23.4%)
Germany	8 (12.5%)
China	7 (10.9%)
UK	5 (7.8%)
Netherlands	4 (6.3%)
Sweden	2 (3.1%)
Canada	2 (3.1%)
Australia	1 (1.6%)
Norway	1 (1.6%)
Saudi Arabia	1 (1.6%)
South Korea	1 (1.6%)
Study Setting	
Plastic Surgery	16 (25.0%)
Craniofacial Centers	15 (23.4%)
Otolaryngology/Otorhinolaryngology, Head, and Neck Surgery	13 (20.3%)
Multiple settings (e.g., online, Craniofacial Center, and Genetic clinic)	12 (18.8%)
Online	6 (9.4%)
Family Conference	1 (1.6%)
Orthodontic	1 (1.6%)
Sample size^a (range 11-863)	
11-25	13 (20.6%)
26-50	12 (19.0%)
51-100	14 (22.2%)
101-200	11 (17.5%)
201-500	7 (11.1%)
501-900	6 (9.5%)
Participants^a	
Individuals with CFM and parents/caregivers (including teachers)	24 (38.1%)
Individuals with CFM	22 (34.9%)
Caregivers of individuals with CFM	13 (20.6%)
Others (eg, members of the public)	4 (6.3%)
Analysis Design	
Cross-sectional (including of longitudinal samples)	44 (68.8%)
Prospective pre- and post-surgery	6 (9.4%)
Mixed methods (cross-sectional and qualitative)	6 (9.4%)
Qualitative	5 (7.8%)
Chart review	3 (4.7%)
Measurement Tools	
Standardized measures	27 (42.2%)

Non-standardized measures	23 (35.9%)
Both standardized and non-standardized measures	14 (21.9%)

Note: ^aa study of online posts was excluded as it was not possible to determine the number or identity of participants

Table 2. Healthcare experiences

Citation	Study Design	Setting	N	Participants	Mea
Hamilton et al. (2018) ³³	qualitative analysis ^a	online in USA, UK, and Australia and Craniofacial clinic, Stanford, CA, USA	11	ages 12-22 with craniofacial microsomia (CFM); 36% post autologous ear reconstruction	study interview of me groups, impact of d and socially, teasing adv
Hamlet et al. (2020) ³⁴	qualitative analysis ^a	online through Microtia UK and Center for Appearance Research, UK	15	ages 20-62 with microtia ($n = 12$) or CFM ($n = 3$); 47% post autologous ear reconstruction and 20% with ear prosthesis	study interview o mic
Jensen et al. (2013) ²²	chart review ^a	Craniofacial clinic, St. Louis, MO, USA	74	ages 2-12 with microtia and/or aural atresia, including CFM	chart review of hearing intervention services learning and beh
Johns, Im, et al. (2018) ³⁶	qualitative analysis ^b	Plastic and Maxillofacial Surgery clinic, Los Angeles, CA, USA	149	ages 3-17 ($n = 62$) and caregivers ($n = 87$) of children with microtia; 65% preoperatively and 35% after alloplastic ear reconstruction 6-48 months prior to study	no
Johns, Luquetti, et al. (2018) ³²	cross-sectional and qualitative analysis ^a	online survey and Craniofacial centers in Seattle, WA and Los Angeles, CA, USA	51	ages 24-76 ($n = 9$) and caregivers ($n = 42$) of children ages 0-17 with CFM	study questionnaire home, school, med settings, what has bee
Johns et al. (2021) ⁴⁷	cross-sectional ^a	online in USA, 4 Craniofacial centers in USA, 4 Genetic departments in Colombia and 1 in Peru	169	caregivers of children ages 3-18 with microtia and/or at least 2 CFM features	study questionnaire o awareness,
Kancherla et al. (2009) ¹⁸	cross-sectional ^a	National Birth Defects Prevention Study, Arkansas, New York, and Iowa, USA	20	mothers of children with microtia	study questionnair concerns, and pa
Kesser et al. (2013) ²³	cross-sectional ^a	attendees of New York Microtia/Atresia Conference or Otolaryngology-Head and Neck Surgery clinic in	40	ages 5-31 with unilateral atresia with or without hemifacial microsomia (HFM)/Goldenhar Syndrome or their caregivers	study questionnaire and behavior

Charlottesville, VA, USA					
Luquetti et al. (2018) ¹⁴	cross-sectional and qualitative analysis ^a	online survey and Craniofacial clinics in Seattle, WA and Los Angeles, CA, USA	51	ages 24-76 (<i>n</i> = 9) and caregivers (<i>n</i> = 42) of children ages 0-17 with CFM	study questionnaire on communication about beliefs, family communication, diagnosis awareness, for re
Luquetti et al. (2019) ¹⁵	cross-sectional ^a	Craniofacial centers and Otolaryngology clinics at 5 sites in USA	108	caregivers of children ages 12-24 months with microtia and/or 2 CFM features	study questionnaire serv
Mandelbaum et al. (2017) ²⁹	chart review ^a	Plastic and Reconstructive Surgery clinics, Los Angeles, CA, USA	68	ages over 13 with CFM who had autologous ear reconstruction	chart review for psycho aesthetic, and m
Myhre et al. (2021) ³⁸	qualitative analysis ^a	Norwegian Craniofacial Association Craniofacial Clinics, Norway	7	ages 19-42 with Goldenhar syndrome who had at least one surgery that altered their appearance	semi-structured experiences of appea
Otto et al. (2020) ³⁹	cross-sectional ^a	microtia family day and Plastic Surgery clinic visits, Utrecht, Netherlands	37	parents of children with microtia	study questionnaire toward different treatment
Reed et al. (2016) ²⁴	cross-sectional ^a	attendees of New York Microtia/Atresia Conference or Otolaryngology-Head and Neck Surgery clinic in Charlottesville, VA, USA	140	ages 1-31 with unilateral atresia with or without HFM/Goldenhar Syndrome or their caregivers	study questionnaire and behavior
van Hovell tot Westerflie et al. (2018) ³⁵	cross-sectional ^a	Microtia and Atresia Conference, Netherlands	84	caregivers of children ages 0-17 with isolated microtia (<i>n</i> = 59), Goldenhar (<i>n</i> = 4), OAV (<i>n</i> = 9), or Treacher Collins (<i>n</i> = 6)	study questionn experiences and pref informing c

Note: ^aNon-standardized measure(s)

Table 3. Psychosocial experiences, neurocognitive functioning, and academic assistance

Citation	Study Design	Setting	N	Participants	Comparison Group	
Almadani et al. (2020) ⁵³	cross-sectional public perception ^{a,b}	online survey from Montreal, Canada	463	ages 18 or older from general population	none	ratings of microsomia of mandible on photos; analog scale; Test, psych quality-ad; co
Byun et al. (2016) ⁵¹	cross-sectional public perception ^{a,b}	online survey from Halifax, Canada	104	ages 20 or older from general population	none	ratings of h or without and binoc blindness scale, time gamble sco
Collett et al. (2011) ⁵⁸	cross-sectional (of longitudinal sample) ^a	26 Craniofacial centers in USA and Canada	704	ages 5-12 (<i>n</i> = 136) diagnosed with CFM, microtia, oculoauricular vertebral syndrome (OAV), or Goldenhar syndrome or at least 2 CFM features	ages 5-12 (<i>n</i> = 568) with no health issues and matched mainly by age and geography	Peabody Test-Third Beery-Buk Test of Vis -Fifth Academic Child B (CBCL) From (TR repo
Collett et al. (2019) ²¹	cross-sectional (of longitudinal sample) ^a	26 Craniofacial centers in USA and Canada	413	ages 11-17 (<i>n</i> = 107) diagnosed with CFM, microtia, OAV, or Goldenhar syndrome or at least 2 CFM features	ages 11-17 (<i>n</i> = 306) with no health issues and matched mainly by age and geography	Clinical Ev Fundame (CELF-4), Commu Second I parent and t Intelligibil scoring; Te Articulat
Collett et al. (2021) ¹⁷	cross-sectional (of longitudinal sample) ^a	6 Craniofacial centers in USA	168	ages 36-42 months (<i>n</i> = 92) diagnosed with microtia/antia and/or a combination of at least 2 CFM features	ages 36-42 months (<i>n</i> = 76) with no health issues and matched by age, sex, SES, and language	Bayley S Toddler I Edition (B Clinical Ev Fundament Edition (C

Dufton et al. (2011) ²⁵	cross-sectional (of longitudinal sample) ^a	26 Craniofacial centers in USA and Canada	704	caregivers and teachers of children ages 5-12 (<i>n</i> = 136) diagnosed with CFM, microtia, OAV, or Goldenhar syndrome or at least 2 CFM features	caregivers and teachers of children ages 5-12 (<i>n</i> = 568) with no health issues and matched mainly by age and geography	CBCL for teacher); Scale - Parent teacher V Acceptance
Fan et al. (2020) ⁴⁵	cross-sectional (of longitudinal sample) ^a	Department of Otolaryngology, Head, and Neck Surgery, Beijing, China	53	caregivers of children ages 4-7 with bilateral microtia and use of soft-band bone conduction hearing aid	none; comparison to norm sample	CBCL; C Benefit
Hyland et al. (2020) ³¹	cross-sectional ^a	Early intervention for hearing loss and local schools, Brisbane, Australia	20	caregivers, teachers, and children ages 7-12 with aural atresia (<i>n</i> = 10), 80% had microtia, who had early amplification at a mean age 5 months	caregivers, teachers, and children ages 6-10 (<i>n</i> = 10) with no health issues and matched for gender, age, and nonverbal intelligence	Ravens P; Clinical Ev Fundame New Zea (CELF-5 Vocabulary (PPVT-4); Reading f Australia Hearing Reflection Questionn CCC-2; careg qu
Jiamei et al. (2008) ³⁷	cross-sectional ^a	Auricle Center, Plastic Surgery Hospital, Beijing, China	766	ages 5-37 (<i>n</i> = 410) with microtia prior to ear reconstruction and parents (<i>n</i> = 356) reporting on children and their families	none; comparison to norm sample	CBCL for Symptom C for ages q
Johansson et al. (2007) ⁵⁴	cross-sectional ^a	multidisciplinary study team, Göteborg, Sweden	20	ages 8 months-17 years with at least 2 of 4 OAV features: orocraniofacial, ocular, auricular, and vertebral	none; comparison to norm sample	Wechsler I Children Griffiths D or Vinelan Scales to functioning Autism

Johns et al. (2021) ⁴⁷	cross-sectional (of longitudinal sample) ^a	6 Craniofacial centers in the USA	161	caregivers of children ages 36-42 months (<i>n</i> = 89) diagnosed with microtia/anotia and/or a combination of at least 2 CFM features	caregivers of children ages 36-42 months (<i>n</i> = 72) with no health issues and matched by age, sex, SES, and language	
Kaelin et al. (2021) ¹⁹	cross-sectional (of a longitudinal sample) ^a	26 Craniofacial centers in USA and Canada	396	caregivers of children ages 11-17 (<i>n</i> = 120) diagnosed with CFM, microtia, OAV, or Goldenhar syndrome or at least 2 CFM features	caregivers of children ages 11-17 receiving services (<i>n</i> = 140) or not receiving services (<i>n</i> = 136) with no health issues matched by age and geography	Participat Measure fo (PEM-C along with comple careg participatio special
Kaelin et al. (2022) ⁶⁰	cross-sectional (of a longitudinal sample) ^a	26 Craniofacial centers in USA and Canada	396	caregivers of children ages 11-17 (<i>n</i> = 120) diagnosed with CFM, microtia, OAV, or Goldenhar syndrome or at least 2 CFM features	caregivers of children ages 11-17 receiving services (<i>n</i> = 140) or not receiving services (<i>n</i> = 136) with no health issues matched by age and geography	PEM-CY, along w Environ Resour children's participatio special
Li et al. (2010) ⁴⁴	cross-sectional ^a	Plastic Reconstructive Surgery, Shanghai, China	622	ages 5-50 (<i>n</i> = 170) with microtia before ear reconstruction and parents (<i>n</i> = 91) reporting on children and themselves	ages 5-50 (<i>n</i> = 264) matched for age and gender in general population and parents (<i>n</i> = 97) reporting on children and themselves	CBCL for a Children's (PHCSS) f fo
Nuyen et al. (2020) ⁵²	cross-sectional public perception ^b	online survey from Stanford, CA, USA	631	adults in Qualtrics LLC participant database with mean age of 39	none	visual ana photos of e with mic reconstruct ch

Ongkosuwito et al. (2018) ⁵⁹	cross-sectional ^{a,b}	Department of Orthodontics, Rotterdam, Netherlands	31	parents of children ages 3-19 with microtia	none	Nijmegen Index Kor Cognitive Questionnaire
Sepehripour et al. (2017) ⁴⁹	webpage review ^a	100 top microtia websites on Google	18	9 physicians with expertise in microtia and 9 adults with microtia	none	DISCERN scoring including
Smit et al. (2021) ⁴⁶	cross-sectional ^a	Department of Otolaryngology, Head, and Neck Surgery, Utrecht, Netherlands	29	ages 7-19 with congenital aural atresia with hearing loss and/or caregivers	none; comparison with norm sample	CBCL for Report (Y Adult Self 18-20; Kid for age 11 report age Spatial and Scale (SSQ and self- CCC-2; e
Snyder and Pope (2010) ⁴³	chart review ^a	Department of Plastic Reconstructive Surgery, New York, USA	47	caregivers of children ages 2-3 ($n = 12$) and ages 4-11 ($n = 35$) with hemifacial microsomia as part of larger study	none; comparison with norm sample	
Speltz et al. (2017) ²⁰	cross-sectional (of longitudinal sample) ^a	26 Craniofacial centers in USA and Canada	436	ages 11-17 ($n = 121$) diagnosed with CFM, microtia, OAV, or Goldenhar syndrome or at least 2 CFM features	ages 11-17 ($n = 315$) with no health issues and matched mainly by age and geography	Wechsler Intelligence Wide Range Fourth Edition (C Woodcock Achievement II
Speltz et al. (2018) ¹⁶	cross-sectional (of longitudinal sample) ^a	6 Craniofacial centers in USA	192	ages 12-24 months ($n = 108$) diagnosed with microtia/antia and/or a combination of at least 2 CFM features	ages 12-24 months ($n = 84$) with no health issues and matched by age, sex, SES, and language	Bayley-III, Language

Umbaugh et al. (2020) ⁵⁰	qualitative analysis of Facebook group posts ^b	microtia and CFM Facebook groups in USA and UK	n/a	individuals with microtia or CFM, caregivers, administrators, and others who posted on 2 Facebook group pages	none	content a
Wallace et al. (2018) ⁴⁸	cross-sectional (of longitudinal sample) ^a	26 Craniofacial centers in USA and Canada	436	caregivers, teachers and self-report of children ages 10-17 (<i>n</i> = 121) diagnosed with CFM, microtia, OAV, or Goldenhar syndrome or at least 2 CFM features	caregivers, teachers, and self-report of children ages 10-17 (<i>n</i> = 315) with no health issues and matched mainly by age and geography	CBCL for teachers; PEDIATRIC QUALITY OF LIFE (PedsQL) parent report; CCC-2 for

Note: ^aStandardized measure(s); ^bNon-standardized measure(s)

Table 4. Pre- and post-operative psychosocial outcomes of ear reconstruction/canaloplasty

Citation	Study Design	Setting	N	Participants	Comparison Group	
Awan et al. (2018) ⁶⁵	prospective pre- and post-surgery ^a	Plastic and Reconstructive Surgery, Jeddah, Saudi Arabia	40	ages 13-18 (<i>n</i> = 20) with grade III microtia 2 weeks before and a year after autologous ear reconstruction and their parents	ages 13-18 (<i>n</i> = 20) control group and their parents matched on age, sex, nationality, education, and family income	Students' Life Satisfaction (SLSS), global life satisfaction, Quality of Life by parent, Image, Questionnaire report (me Arabi ps
Cho et al. (2020) ⁶⁶	prospective pre- and post-surgery ^a	Department of Otorhinolaryngology-Head and Neck Surgery, Seoul, South Korea	34	ages 9-24 with microtia and aural atresia who had canaloplasty in previous year	none	Derrifon (DAS-24
Hempel et al. (2014) ⁶²	prospective pre- and post-surgery ^a	Department of Otorhinolaryngology-Head and Neck Surgery, Munich, Germany	21	ages 4-62 (<i>n</i> = 21) with microtia before alloplastic ear reconstruction and group of 17 after surgery; children were ages 4-12 (<i>n</i> = 7) and adolescents/adults were ages 13-62 (<i>n</i> = 14)	none	Glasgow Inventory, 36 Hea Child, Ques
Horlock et al. (2005) ⁴¹	cross-sectional (post-surgery with recall of pre-surgery) ^{a,b}	Regional Center for Plastic and Reconstructive Surgery, UK	62	ages 4-15 (<i>n</i> = 36) and ages 16-86 (<i>n</i> = 26) with microtia (<i>n</i> = 44), traumatic amputation (<i>n</i> = 14), malignancy requiring amputation (<i>n</i> = 3), or ear loss after otoplasty (<i>n</i> = 1) who had autologous (<i>n</i> = 52) or alloplastic (<i>n</i> = 10) ear reconstruction in the prior 3-5 years	none; comparison with a norm sample	CEQ
Johns et al. (2015) ⁶⁴	prospective pre- and post-surgery ^{a,b}	Plastic and Maxillofacial Surgery clinic, Los Angeles, CA, USA	23	ages 3-6 (<i>n</i> = 11) with microtia who had alloplastic ear reconstruction and caregivers report	ages 7-10 (<i>n</i> = 12) with microtia and caregivers report	Behaviora for Child (BASC) questi

Johns et al. (2017) ⁴⁰	prospective pre- and post-surgery ^b	Plastic and Maxillofacial Surgery clinic, Los Angeles, CA, USA	28	ages 3-5 (<i>n</i> = 13) with microtia who had alloplastic ear reconstruction and caregivers report	ages 6-10 (<i>n</i> = 15) with microtia and caregivers report	study qu
Steffen et al. (2008) ⁶¹	cross-sectional (post-surgery with recall of pre-surgery) ^{a,b}	Department of Otolaryngology, Lubeck, Germany	68	ages 12-58 with microtia (<i>n</i> = 60) or traumatic ear defects (<i>n</i> = 8) who had autologous ear reconstruction in the prior 1-6 years	none	Frankfurt (FSKN);
Steffen et al. (2010) ⁶³	prospective pre- and post-surgery ^{a,b}	Department of Otolaryngology, Lubeck, Germany	44	ages 12-28 (<i>n</i> = 21 Time 1; <i>n</i> = 16 Time 2) with microtia who had autologous ear reconstruction in the prior 3-4 months	ages 12-32 (<i>n</i> = 23) with microtia who did not have surgery	FSKN;

Note: ^aStandardized measure(s); ^bNon-standardized measure(s)

Table 5. Quality of life and patient satisfaction

Citation	Study Design	Setting	N	Participants	Comparison Group	
Akter et al. (2017) ⁷⁸	cross-sectional post-surgery ^{a,b}	Plastic Surgery centers, London and Edinburgh, UK	69	London sample ages 9-14 (<i>n</i> = 42) and Edinburgh sample ages 11-19 (<i>n</i> = 27) who had first stage autologous ear reconstruction for microtia between 8-108 months prior	none	Questionnaire general specific and detailed questions psychosocial aesthetic satisfaction
Bradford et al. (2020) ⁸⁵	cross-sectional pre-surgery or post-surgery ^b	Craniofacial clinic, Chicago, IL, USA	25	ages 7-20 with microtia (<i>n</i> = 9) or craniofacial microsomia (CFM) including microtia (<i>n</i> = 16) who participated preoperatively (<i>n</i> = 9) or postoperatively (<i>n</i> = 16) and their caregivers (<i>n</i> = 25)	none	developmental questionnaire social psychosocial decision-making 13 items additional
Braun et al. (2010) ⁷⁵	cross-sectional post-surgery ^{a,b}	Otorhinolaryngology, Head and Neck Surgery clinic, Munich, Germany	65	ages 16-65 (<i>n</i> = 45) and parents of children ages 4-15 (<i>n</i> = 20) with isolated microtia (<i>n</i> = 35), microtia as part of a syndrome (<i>n</i> = 20), or acquired auricular defects (<i>n</i> = 10) who had alloplastic ear reconstruction 0.5-6 years prior	none	Glasgow (GBI) composite Glasgow Inventories by children questionnaire satisfaction
Braun et al. (2013) ⁷⁶	cross-sectional post-surgery ^{a,b}	Otorhinolaryngology, Head and Neck Surgery clinic, Munich, Germany	15	ages 15-42 (<i>n</i> = 9) and parents of children ages 10-14 (<i>n</i> = 6) with microtia who had alloplastic ear reconstruction 3-7 years prior	none	GBI composite GCBI composite parents; satisfaction

Cui et al. (2017) ⁸⁰	cross-sectional post-surgery ^a	Plastic and Reconstructive Surgery, Shanghai, China	72	ages 13-45 (<i>n</i> = 34) and parents of children ages 7-12 (<i>n</i> = 37) with microtia who had autologous ear reconstruction at least four months prior	none	Pat Question rating pat a label parts o sa
Cui et al. (2018) ⁸²	qualitative interviews and cross-sectional post-surgery ^b	Plastic and Reconstructive Surgery, Shanghai, China	180	ages 7-18 (<i>n</i> = 10) for initial interviews; ages 7-55 for field test (<i>n</i> = 76); ages 7-44 (<i>n</i> = 94) for psychometric analysis who had microtia and completed first stage ear reconstruction at least four months prior	none	deve questio rating pat a label parts o
Fan et al. (2017) ⁷²	cross-sectional post-surgery ^a	Otolaryngology Department, Beijing, China	12	ages 6-18 with bilateral microtia who had autologous ear reconstruction and bone-conduction hearing aid implant (<i>n</i> = 9) or hearing aid implant only (<i>n</i> = 3) 6 months prior and parents	none	auricu satisfac Profile of Inter Invento GCBI co
Hempel et al. (2013) ⁷⁷	cross-sectional post-surgery ^a	Otorhinolaryngology, Head and Neck Surgery clinic, Munich, Germany	28	ages 26, 27, and 35 and parents of children ages 4, 6, and 9 at time of alloplastic ear reconstruction surgery with hemifacial microsomia (HFM)	mean age 26 years (<i>n</i> = 12) and parents of children mean 7 (<i>n</i> = 10) with isolated microtia who had ear reconstruction	GBI con GCBI co
Khetani et al. (2013) ⁶⁸	cross-sectional (of longitudinal sample) ^a	26 Craniofacial centers in USA and Canada	704	mothers of children ages 5-12 (<i>n</i> = 136) diagnosed with CFM, microtia, oculoauricular vertebral (OAV), syndrome or	mothers of children ages 5-12 (<i>n</i> = 568) with no health issues matched by	Pedia (PedsQ Vocabula

				Goldenhar syndrome or at least 2 CFM-features by a craniofacial physician and caregivers report	age and geography	
Klassen et al. (2018) ⁸⁷	qualitative interviews and cross-sectional pre surgery or post-surgery ^b	Plastic Surgery clinics in Canada, Australia, USA, and UK	44	ages 8-21 (<i>n</i> = 25 initial interviews) with prominent ears (<i>n</i> = 9), microtia (<i>n</i> = 9), oromandibular syndrome (<i>n</i> = 1), hemangioma (<i>n</i> = 1), Goldenhar syndrome (<i>n</i> = 2), CFM (<i>n</i> = 2), or Treacher Collins syndrome (<i>n</i> = 1) pre or post-surgery; ages 8-21 (<i>n</i> = 17 cognitive interviews) 9 same participants and 6 new participants with similar diagnoses; 13 clinical experts reviewed scales	none	developmental Appearance Effect
Klassen et al. (2021) ⁸⁹	cross-sectional pre surgery or post-surgery ^a	21 collaborating sites in Australia, Brazil, Canada, China, Ireland, Spain, UK, and USA; online through Microtia UK	863	ages 8-29 with microtia (<i>n</i> = 607), prominent ears (<i>n</i> = 145), or other, including trauma (<i>n</i> = 111)	none	psychometric EAR-Q and, if valid, surgery, Psycho School
Kristiansen et al. (2013) ⁷⁹	cross-sectional post-surgery ^b	Plastic and Reconstructive Surgery, Malmo, Sweden	59	ages 9-23 with unilateral microtia who had autologous ear reconstruction 0-10 years prior	none	study questionnaire aesthetic psychometric re

Ren et al. (2012) ⁸³	qualitative interviews and cross-sectional post-surgery ^{a,b}	Otolaryngology Head and Neck Surgery clinic, Beijing, China	140	ages 6-18 with congenital aural atresia who underwent ear reconstruction <i>Note:</i> demographics of patients, caregivers, and medical staff initially interviewed not reported	none	developmental Aural Awareness for physical item functional social function Adolescents Events
Schrötzlmair et al. (2021) ⁷⁴	cross-sectional post-surgery ^{a,b}	Otorhinolaryngology, Head and Neck Surgery clinic, Munich, Germany	113	individuals who had who had alloplastic ear reconstruction at a mean age of 19 (range 4-68 years) that was 1-11 years prior or their caregivers; most had congenital auricular dysplasia ($n = 104$)	none	GBI; questionnaire satisfaction postoperative and a
Soukup et al. (2012) ⁷³	cross-sectional post-surgery ^{a,b}	Plastic Surgery centers, London UK	55	ages 9-21 who had autologous ear reconstruction 4 months-4 years prior for isolated microtia ($n = 40$) or microtia as part of a syndrome ($n = 15$) and their parents; surgical outcomes also rated by their surgeon, an adolescent, and a medical student	none	GBI; s measuring of ear, s of e
Steffen et al. (2012) ⁸⁴	cross-sectional post-surgery ^{a,b}	Department of Otorhinolaryngology, Lubeck, Germany	56	ages 10-20 ($n = 50$) with microtia and other congenital ear diagnoses who had ear reconstruction within 3 years prior; adolescents with head and neck diagnoses ($n = 6$) for initial German translation	none	German Quality of Life (YQOL- of Life Child Invento

Tsangaris et al. (2019) ⁸⁸	cross-sectional ^a	Plastic Surgery or Ear, Nose, and Throat (ENT) clinics in Doha, Qatar; Shanghai, China; Paris, France; Madrid, Spain	28	ages 5-29 with prominent ears (50%), microtia (43%), or constricted ears (7%)	none	translation Ear Appearance Adverse items) i Fren
Younis et al. (2010) ⁸¹	cross-sectional post-surgery ^b	Plastic Surgery clinic, Northwood, UK	20	ages 17-56 with microtia who had congenital ear diagnosis ($n = 10$) or traumatic ear loss ($n = 9$) who had surgery for a bone anchored prosthetic ear implant 7-108 months prior	none	study qu satisfac aesthe prob

Note: ^aStandardized measure(s); ^bNon-standardized measure(s)

Supplemental Table 1. Healthcare experiences key findings

Citation	Key Findings
Hamilton et al. (2018) ³³	1) overall positive impact of diagnosis on character development reported, including empathy, i open-mindedness; 2) negative psychosocial impacts reported included teasing that led to lower s feeling isolated, with pattern of greater difficulty of being different for females; 3) frequent deci difference with hair style and not discuss diagnosis or hearing loss to avoid embarrassment, que 4) advice was given to be open about diagnosis and have confidence; 5) desire expressed to be p decision making and participants appreciated being included in decision making even at young g difficulties were reported due to hearing loss, especially in group and loud settings
Hamlet et al. (2020) ³⁴	1) participants reported frequent ability to hide microtia; 2) noted feeling anxious ear(s) may be settings and relationships; 3) hearing loss was difficult in work settings; 4) surgery and outcome were generally seen positively; 5) process of surgery noted to have some difficulties; 6) some p comfortable without surgery; 7) psychosocial support was noted as a need; 8) emphasis placed o development independent from diagnosis; 8) greater diagnosis awareness was seen as positive; comparisons helped some participants cope
Jensen et al. (2013) ²²	1) all children with bilateral aural atresia received amplification; 2) despite maximal conductive affected ears, only 4% with unilateral aural atresia had amplification (1 FM system; 2 soft band participants had speech therapy at average start age of 4.3 years; 4) based on in-clinic speech ev articulation errors and 31% had language errors; 5) parental reports of problems in school inclu concerns (23%), discipline issues (11%), and behavioral concerns (12%); 6) 31% of participants academic support (e.g., resource program, tutoring, special education classrooms)
Johns, Im, et al. (2018) ³⁶	1) diagnosis given most often by pediatrician (42%), delivery nurse (34%), or obstetrician/gyne most often a physical description (40%) or no specific information (30%); 2) initial responses w negative emotions (73%), concern for future (29%), and positive responses (24%); 3) etiology v

	<p>within perceived medical framework (65%), no known cause (32%), folk explanation (27%), re (3%) for parents while children mostly had no known cause (53%), something wrong (18%), or born (16%); 4) Parental coping included family support (58%), consulting providers (47%), self and religious coping (29%); 5) caregivers reported talking to their children about ear reconstruction descriptions (37%), and providing reassurance (25%), while children reported discussing ear reconstruction positive descriptions (19%), not remembering discussions (18%), or being born with microtia (1</p>
Johns, Luquetti, et al. (2018) ³²	<p>1) between a fifth to a third of sample did not have difficulties in home, school, community, and settings; 2) in home and community settings, difficulties included reported hearing concerns, special treatment burden for caregivers and hearing concerns, social comparisons, and lack of understanding communication for adults; 3) supports caregivers identified included online content, partners/spouse providers and the most frequent caregiver advice given was to offer reassurance; 4) advice offered CFM included making informed medical decisions, having self-confidence, reframing when teaching communication; 5) school concerns identified were hearing, teasing, accessing services, and difficult interactions; 6) a third to half of the sample received school accommodations and/or services; 7) school setting included addressing hearing needs, increased peer awareness of CFM, and addressing healthcare difficulties reported included provider lack of CFM knowledge, lack of empathy, lack of guidance, difficulty accessing treatment, conflicting medical recommendations, and difficulty completing treatment; 9) healthcare supports identified included clear communication, patient-focused approach attitudes, reassurance, taking time with families, and coordinated care; 10) advice offered to improve was increasing treatment coordination, providing clear CFM data, making appropriate referrals, advocacy, offering reassurance, and linking to others with CFM</p>
Johns et al. (2021) ⁴⁷	<p>1) 90% of children had hearing loss and 54% used hearing aid; 2) for the total sample, 79% were in special education classes, 10% in general education with resource program supports, and 10% in special education all day; 3) more services were reported for children in the US with 67% in general education, 16% in special education program supports, and 17% with special education classes all day, while 96% of South American children were in general education classes without academic support; 4) in the US, 48% had an Individualized Education Program (IEP), most often for hearing impairment (62%) and speech or language impairment (43%); 5) 24% had speech therapy, 11% deaf/hard of hearing services, 10% occupational therapy, 7% physical therapy, and 7% mental health services; 6) diagnostic awareness was reported at a mean age of 4.4 years; 8) teasing was reported by 41% starting mostly between ages 5-6 and generally at school (86%) by classmates (86%); 9) teasing was reported as taking place almost never (39%), some of the time (45%), half of the time (3%), most of the time (10%), and almost always (6%)</p>
Kancherla et al. (2009) ¹⁸	<p>1) 68% of mothers were very happy with child's appearance and 75% felt need for additional options for appearance; 2) 35% of mothers were very satisfied with information at time of diagnosis and 21% were very satisfied with answers for treatment options with 50% very satisfied with support provided at time of diagnosis; 3) 90% of children were seen by a multidisciplinary treatment team; 4) 90% of children had hearing loss and 80% had a hearing aid; 5) cognitive delays reported for 10% of children and emotional or behavioral problems for 10%; 6) children's services included 60% speech therapy, 30% learning interventions, and 5% behavioral interventions</p>
Kesser et al. (2013) ²³	<p>1) all participants had hearing loss; 2) 15% had used amplification (bone-conducting or conventional); 3) 73% of participants reported some difficulties with communication, mostly in groups or noisy environments; 4) 73% had received intervention services; 5) 45% participated in speech therapy; 6) 48% of participants had received special education academic support; 8) 13% were reported to have a behavioral diagnosis</p>
Luquetti et al. (2018) ¹⁴	<p>1) most individuals with CFM had been seen by a craniofacial clinic (77%) and had an average of 12 surgeries, most often audiology (98%), otolaryngology (73%), plastic surgery (71%), and dentistry (67%); 2) 80% had one surgery, with a mean of 4 and range of 1-30 surgeries; 3) hearing loss was reported by 80% of participants and 80% currently used hearing aids; 4) therapies provided included speech therapy (53%), physical therapy (13%), and occupational therapy (13%)</p>

	<p>occupational therapy (20%), and mental health services (10%); 5) diagnosis was most often given by a pediatrician (46%), delivering physician (27%), or delivery nurse (17%); 6) common caregiver concerns about diagnosis were concern/anxiety (79%), surprise/shock (64%), sadness (64%), guilt (55%), and confusion (55%); 7) adults with CFM most often first heard of their diagnosis from a parent (67%), plastic surgeon (11%), or through their own research (11%); 8) etiology beliefs were generally unsure for a third of caregivers and adults with CFM, with explanations including genetics, random occurrence, circulation issues during pregnancy, or religious explanations; 9) positive family communication about diagnosis was reported by 36% by caregivers; 10) caregivers and adults with CFM both reported diagnosis awareness at mean age of 6 years and first teasing at mean age of 6 years; 11) caregivers reported teasing took place for 43% of children by 10 years and was most often at school (92%) consisting of name calling; 12) caregiver priorities for child development included etiology, understanding the diagnosis and treatment, hearing concerns, while adults with CFM valued hearing, research on teasing, social concerns, and communication about diagnosis by healthcare providers</p>
Luquetti et al. (2019) ¹⁵	<p>1) CFM was diagnosed at birth for 79% of participants; 2) 59% had been seen in a craniofacial clinic; 3) 9 medical specialists seen within an average of 3 medical settings; 4) 64% had received early intervention; 5) 28% had at least one surgery by age 2 years</p>
Mandelbaum et al. (2017) ²⁹	<p>1) 63% of patients were noted to have no concerns reported in their annual team meeting; 2) 31% of patients were noted in their charts to have a concern in school, psychosocial functioning, and/or peer interactions; 3) positive psychosocial outcome was predicted only by treatment of hearing loss (OR = 4.9, $P = .01$) with no variation based on gender, microtia severity or laterality, mandibular involvement, hearing loss, age of surgeries, complications, or aesthetic ratings</p>
Myhre et al. (2021) ³⁸	<p>Themes drawn from interviews about appearance-altering surgery experiences included: 1) striving for normalcy, including feeling like they were more than their diagnosis and struggling with belongingness; 2) managing appearance, including hopes, expectations, and disappointments, challenging processes and dialing back surgery, questioning how much can be changed, and wondering how much change is enough; 3) navigating family and friends, including close family and difficulties with openness in friendships</p>
Otto et al. (2020) ³⁹	<p>1) majority positive attitudes were reported for tissue engineering, tissue engineered cartilage, and autologous chondrocyte transplantation; 2) positive and receptive attitudes were reported for autologous chondrocytes, autologous stem cells, natural materials, decellularized tissue, and cell-seeded Medpor; 3) more negative attitudes were reported for harvesting cells from child, implantation of engineered cartilage, and participating in clinical trials; 4) parents expressed concern with long term outcomes, child pain, and participating in early clinical trials; 5) established evidence of success</p>
Reed et al. (2016) ²⁴	<p>1) all participants had hearing loss in their affected ear; 2) 27% of participants with aural atresia had surgery; 3) 4% had been retained to repeat another year in the same grade; 4) 36% of participants had IEPs; 5) 16% had received speech therapy; 6) 16% had academic special education services; 7) 66% were reported to have communication problems; 8) 14% were reported to have behavioral problems; 9) 5% were noted to have emotional diagnosis; 9) there was no consistent pattern of significant differences based on laterality of affected ear</p>
van Hovell tot Westerfliet et al. (2018) ³⁵	<p>1) diagnosis was given at time of birth for 74% of caregivers; 2) initial information about microtia was provided by pediatricians (35%), caregiver internet searches (26%), or ENT surgeon (26%); 3) initial information consultations were rated as terrible (43%), bad (21%), moderate (16%), reasonable (12%), good (12%), or excellent (1%); 4) topics caregivers rated as very important to be informed about included hearing, development, reconstructive surgery, maxillofacial, genetics, psychological, medical team, financial, and information; 5) open-ended responses from caregivers highlighted the importance of clarity of surgical options (which varied to vary by country), dissatisfaction with the lack of knowledge in some providers, a need for coordinated multidisciplinary teams, wanting more psychological guidance, linking to financial support, and the need for information to be provided both at time of diagnosis and across developmental phases; 6) children</p>

caregivers had mostly had no ear reconstruction (79%) with 18% who had alloplastic ear reconstruction, 1% who had autologous ear reconstruction, and 1% had an ear prosthesis

Supplemental Table 2. Psychosocial experiences, neurocognitive functioning, and academic assistance

Citation	Key Findings
Almadani et al. (2020) ⁵³	1) health ratings of 0 (death) to 1 (perfect health) made online by members of the general public of brief descriptions and photos for mandibular hypoplasia in participants with craniofacial microsomia (CFM) before mandibular distraction ($M = 0.48$) was between binocular blindness ($M = 0.31$) and monocular blindness ($M = 0.53$) with higher scores for CFM after distraction ($M = 0.63$); 2) there was no statistical significant difference in the time trade-off of mandibular distraction ($M = 0.83$) compared to a surgery to achieve perfect health ($M = 0.79$); 3) ratings of psychosocial acceptance across roles improved after mandibular distraction; 4) ratings reflected distraction was considered cost effective (<\$50,000/quality-adjusted life years) if health benefits lasted at least 3 years
Byun et al. (2016) ⁵¹	1) health ratings of 0 (death) to 1 (perfect health) made online by members of the general public of brief descriptions and photos for microtia without hearing loss was rated as healthy ($M = 0.90$) and microtia with deafness was rated lower ($M = 0.80$), which were both higher than monocular blindness ($M = 0.65$); 2) while monocular blindness was rated as more of a concern, microtia with hearing loss was similar to monocular blindness on the trade-off scores, with scores indicating giving up five years of life and risking a 9% chance of death to have a surgery that would achieve perfect health; 3) the authors noted the public perception of the burden of microtia may not match the experience of people with microtia
Collett et al. (2011) ⁵⁸	1) Both craniofacial microsomia (CFM) group and control group scores were in the average range across all measures; 2) CFM group was lower on Peabody Picture Vocabulary Test-Third Edition (PPVT-III) ($P < .001$) and Beery-Buktenica Developmental Test of Visual Motor Integration-Fifth Edition (VMI-5) ($P = 0.004$) with adjusted effect sizes (ES) = -0.31 to -0.43; 3) CFM group was lower on Child Behavior Checklist (CBCL) ($P < .001$) and Teacher Report Form (TRF) ($P = .008$) with adjusted ES = -0.27 to -0.45; 4) CFM group was 3 times as likely to be in the at-risk range on PPVT-III; 5) CFM group twice as likely to be in at risk range on VMI-5; 6) CFM group was twice as likely to be in at risk range for lower academic competence by parent and teacher report; 7) larger differences were observed CFM group among males and with mothers ≤ 25 years old at time of child's birth; 8) larger differences were observed for CFM group with speech problems for parent-reported academic competence
Collett et al. (2019) ²¹	1) 94% of CFM group and 99% of controls had correct articulation (adjusted ES = -0.98, $P < .001$); 2) 35% of CFM group and 3% of controls had disordered speech; 3) 93% of CFM group and 97% of controls were intelligible (adjusted ES = -0.61, $P = .001$); 4) CFM group had lower Clinical Evaluation of Language Fundamentals-Fourth Edition (CELF-4) Recalling Sentences (adjusted ES = -0.27, $P = .02$); 5) by parent report on the Children's Communication Checklist-Second Edition (CCC-2), 1 of 6 subtests was lower for CFM group; 6) by teacher report on the CCC-2, 3 of 6 subtests lower for

	CFM group; 7) children with microtia with mandibular hypoplasia ($n = 46$) had lowest scores by phenotype; 8) 70% of group with CFM and 1% of controls failed hearing screening; 9) 23% of group with CFM used hearing aids; 10) 60% of CFM group received speech therapy
Collett et al. (2021) ¹⁷	1) average scores for CFM group and controls for cognitive and motor development on the Bayley Scales of Infant and Toddler Development – Third Edition (Bayley-III) and most subtests of the Clinical Evaluation of Language Fundamentals-Preschool, Second Edition (CELF-P2), except children with CFM were in the below average range for the CELF-P2 Recalling Sentences subtest ($M = 7$); 2) CFM group was significantly lower on CELF-P2 subtests of Recalling Sentences (adjusted $ES = -0.68$, $P = .02$) and Concepts and Following Directions (adjusted $ES = -0.58$, $P = .01$); 3) 39% of CFM group and 15% controls had developmental delay; 4) little difference among children with CFM identified based on phenotype, with some larger differences for extracranial anomalies; 5) 82% of CFM group had hearing loss; 6) 53% of CFM group had hearing aids; 7) 70% of CFM group and 12% of controls received intervention services
Dufton et al. (2011) ²⁵	1) all CBCL mean scores for CFM group and controls were in the average range by parent and teacher report; 2) by parent report, there were no CBCL composite differences with a subscale difference was for more Social Problems for children with CFM (adjusted $ES = 0.30$, $P = .002$); 3) by teacher report, children with CFM had higher internalizing (adjusted $ES = 0.27$, $P = .011$) and total problems (adjusted $ES = 0.25$, $P = .005$) as well as significantly more concerns on 6 of 8 subscales; 4) there were no group differences on Social Competence Scale - Parent Version; 4) children with CFM were lower on Social Competence Scale - Teacher Version total (adjusted $ES = -0.24$, $P = .019$); 5) on the Peer Acceptance Ranking completed by teachers, children with CFM were ranked lower (52%) than controls (42%) (adjusted $ES = 0.43$, $P < .001$); 6) teachers reported children with additional CFM features had more concerns; 7) authors noted protective socioeconomic (SES) factors of the sample
Fan et al. (2020) ⁴⁵	1) participants started wearing bone conducted hearing devices (BCHD) between 2-24 months at an average age of 7.5 months; 2) participants had worn BCHD for an average of 4.6 years at time of evaluation for an average of 7 hours a day; 3) raw CBCL mean scores did not differ from raw scores of Chinese norm group 4) there were 4% ($n = 2$) of participants above a clinical cutoff score; 4) the final average rating on the Glasgow Children's Benefit Inventory (GCBI) was 33 with a range of 4 to 96
Hyland et al. (2020) ³¹	1) all 10 children with aural atresia (AA) received early amplification at median age of 5 months (range 1-58) months and had FM systems in their classrooms; 2) 80% of children with AA received Auditory-Verbal Therapy intervention at median age of 5.5 months (range 1-58) for a duration of a median of 62 months (range 13-76); 3) all participants were in general education classrooms; 4) compared to controls, who had median scores all above 95, children with AA reported significantly lower scores on the Hearing Environments and Reflection on Quality of Life Questionnaire (HEAR-QL-26)

	<p>with an AA group Total score median = 79 (ES = 0.77, $P < .001$), for Feelings subscale with AA group median = 75 (ES = 0.74, $P = .001$), and for the Environments subscale with AA group median = 73 (ES = 0.79, $P < .001$) and there were no group differences on the Activities subscale; 5) there were no significant differences between AA and control groups on any of the remaining 5 measures; 6) nonverbal cognitive functioning on Raven's Progressive Matrices for children with AA was a median percentile of 82 (range 39-87) and children without AA also had a median percentile of 82 (range 61-97); 7) Clinical Evaluation of Language Fundamentals – Fifth Edition (CELF-5) core language median for children with AA = 106 (range 76-118) and without AA = 105 (range 86-129); 8) Peabody Picture Vocabulary Test – Fourth Edition (PPVT-4) AA group median = 113 (range 70-130) and controls = 118 (range 99-134); 9) York Assessment of Reading for Comprehension – Australian Edition (YARC) Accuracy scale AA group median = 104 (range 70-113) and controls = 111 (range 93-130), YARC Rate scale AA group median = 108 (range 85-113) and controls = 108 (range 85-127), and YARC Comprehension scale AA group median = 109 (70-119) and controls = 110 (range 99-130); 10) CCC-2 AA group median = 33 (range 1-92) and controls = 59 (range 16-95).</p>
Jiamei et al. (2008) ³⁷	<p>1) awareness of microtia took place between ages 1 to 7 years, with most at age 3 (37%) or 4 (19%); 2) teasing about microtia was reported by 61% of participants; 3) families rated impact of microtia as severe (35%), moderate (46%), or minimal (20%); 4) depression was reported in 20% of participants, interpersonal sensitivity/social withdrawal in 37%, and hostility/aggression in 26%; 5) presence of depression was related to being age 15 years or older, over age 5 years when microtia was perceived, greater severity grade microtia, being teased, and negative impact on family; 6) interpersonal sensitivity/social withdrawal related to being 15 years or older, over age 5 years when diagnosis was perceived, being teased, and a negative impact on family; 7) hostility/aggression related to being 15-20 years old, being teased, and negative impact on family; 8) authors noted females (23%) were underrepresented in the sample; <i>Note:</i> standard scores were not reported to interpret clinical ranges</p>
Johansson et al. (2007) ⁵⁴	<p>1) Based on a range of measures including direct assessment and caregiver report, 45% had average intelligence (IQ >85), 10% had below average intelligence (IQ = 70-85), 20% had mild Intellectual Disability (IQ = 50-69), 15% had moderate ID (IQ = 50-69), and 10% had severe ID (IQ = 20-49); 2) Autism Spectrum Disorder (ASD) criteria was met for 10% of children, ASD-like for 5%, ASD traits for 25%, possible ASD traits for 15%, and no ASD for 40%; 3) for children with cerebral radiological imaging ($n = 11$), 64% had structural cerebral anomalies/abnormalities in white/gray matter, which was proposed to be related to embryonic brain development and possibly related to ASD</p>
Johns et al. (2021) ⁴⁷	<p>1) CBCL mean scores were all in the average range for children with CFM and controls; 2) there were no significantly higher odds ratio (OR) of being within the clinical range on any CBCL scale for children with CFM, with the highest area of concern seen for 19% of the CFM group within clinical range for</p>

internalizing problems; 3) there were no differences on CBCL composite scales between CFM and control groups; 4) among subscales, the CFM group was higher than controls group for Anxious/Depressed (adjusted ES = 0.35, $P = .04$), Stress Problems (adjusted ES = 0.40, $P = .04$), Anxiety Problems (adjusted ES = 0.34, $P = .04$), and Autism Spectrum Problems (adjusted ES = 0.41, $P = .02$); 5) specific items that were elevated for CFM group were for being too dependent, not getting along with other children, and speech problems; 6) by phenotype, additional CFM features were generally associated with more concerns; 7) concerns for males with CFM were higher relative to females with CFM as well as to males in the control group; 8) 82% cases had hearing loss

Kaelin et al. (2021)¹⁹ 1) children with CFM had lower participation rates in school occupations than peers without service use (ES = -0.14, $P = .029$) and were no different from peers who had a history of service use (ES = -0.02, $P = .82$); 2) compared to peers without service history, children with CFM had a 41% increase in mean number of school occupations for which change was desired and no differences from peers who used services; 3) the specific activities parents of children of CFM wanted increased were field trips/school events (OR = 1.77, $P = .045$), spending time with peers outside of class (OR = 2.39, $P = .003$), and special roles at school (OR = 1.86, $P = .014$); 4) perceived school supports were similar across caregivers; 5) 79% of children with CFM participated in at least one service, including speech-language therapy (58%), physical therapy (36%), hearing services (33%), special education academic services (31%), occupational therapy (26%), mental health services (14%), and vision therapy (8%)

Kaelin et al. (2022)⁶⁰ 1) children with CFM had lower participation rates in community activities than peers without a history of service use (adjusted ES = -0.52, $P < .001$), particularly in organized physical activities and spending time with peers in the community; 2) there were no differences compared to peers with a history of service use; 3) the level of involvement was lower for children with CFM compared to peers with no service use and no differences compared to peers with service use; 4) there was a 35% increase in mean number of activities parents of children with CFM desired change, including overnight trips, spending time with children in the community, group/club activities, and organized physical activities; 5) there were no significant group differences in perceived environmental supports in the community

Li et al. (2010)⁴⁴ 1) among males with microtia ages 5-16 years, adolescents generally had more clinical concerns on CBCL; 2) compared to controls, males with microtia age 8-10 and 14-16 years were higher for social problems and aggressive behavior; 3) compared to controls, males with microtia ages 17+ had higher interpersonal sensitivity, depression, anxiety, and hostility on Symptom Checklist-90 (SCL-90) and females with microtia 17+ years old had higher depression; 4) there were no differences between females with microtia and the control group on the CBCL; 5) on Piers-Harris Children's Self-Concept Scales (PHCSS), gregariousness was lower in the males with microtia ages 14-16 and females with microtia ages 11-13; 6) mothers of children with microtia had more

	<p>concerns than fathers; 7) compared to control mothers, mothers of children with microtia had higher interpersonal sensitivity, depression, and anxiety on the SCL-90; 8) authors noted the low number of female participants (26%) was likely due to both higher male incidence of microtia and that due to some traditional cultural values that place higher importance on male children, surgery may not be sought for females with microtia and were thus not available to be in study; <i>Note:</i> standard scores were not reported to interpret clinical ranges</p>
Nuyen et al. (2020) ⁵²	<p>1) no differences in ratings by general public based on three ear photos (no microtia, preoperative microtia, and postoperative microtia) were found for athleticism, confidence, leadership, or popularity; 2) no differences between the reconstructed ear and ear without microtia photos; 3) microtia photos were rated as less friendly than photos with no microtia, less healthy than photos of a reconstructed ear and no microtia, less intelligent than reconstructed ear photos, and less successful than no microtia photos</p>
Ongkosuwito et al. (2018) ⁵⁹	<p>1) mean parental stress of sample was lower than norm values; 2) parental stress was correlated with parent-reported child learning difficulties ($r = 0.71$, $P < .001$) and psychosocial problems ($r = 0.63$, $P < .001$); 3) Cognitive Emotion-Regulation Questionnaire (CERQ) acceptance ($r = 0.50$, $P < .05$) and positive reappraisal ($r = 0.54$, $P < .05$) were correlated with parental stress; 4) in the final regression analysis model, child learning difficulties ($\beta = 0.50$, $P < .001$) and acceptance ($\beta = 0.30$, $P < .05$) explained variance in parental stress; 5) the authors suggested acceptance may represent a passive giving up or reflect variation in this variable based on too wide a range in the age of the children in the sample; 6) authors noted that the degree of parent-reported visibility of diagnosis was unrelated to parental stress</p>
Sepehripour et al. (2017) ⁴⁹	<p>1) microtia websites were mostly from US (77%) and UK (9%); 2) websites were mostly from private healthcare providers (48%) or university or charitable organizations (19%); 3) the overall quality scores ranged from 15 to 80 by physician ratings with a mean of 39; 4) there was no difference between patient and physician quality ratings of the websites; 5) there was little association between Google ranking and quality ratings; 6) the authors noted that although many patients use social media as a primary source of patient information, only two social media sites were in the top 100 and were not highly rated; 7) authors noted barriers in accessing high quality online material from medication education journals that require subscription/payment and that journals are also not interactive</p>
Smit et al. (2021) ⁴⁶	<p>1) school retention for a year was reported for 35% and 7% had special education classroom placements; 2) 61% had speech therapy and 79% had classroom accommodations for hearing needs (e.g., preferential seating by teacher); 3) 45% used hearing amplification; 4) hearing-related quality of life did not differ by age, sex, or use of hearing amplification and was lower compared to previous reports in the literature; 5) quality of life as measured by Kidscreen-27 was all within average range with exception of higher physical well-being for ages 6-10; 6) all CBCL, Youth Self-Report (YSR), and Adult Self-Report (ASR) mean scores were in average range; 7) among participants,</p>

	ages 6-10 had higher attention seeking and ages 16-20 had higher withdrawn/depressed scores
Snyder and Pope (2010) ⁴³	1) all CBCL mean scores were in the average range; 2) children ages 4-11 were significantly less likely to be in clinical range for internalizing problems (3%) compared to norms; 3) there were no other differences from norm sample across CBCL problem or competence scales
Speltz et al. (2017) ²⁰	1) average range scores for CFM and control groups across all cognitive and academic measures; 2) CFM group was lower than controls on reading (adjusted ES = -0.3, $P = .04$); 3) CFM group lower than controls on writing (adjusted ES = -0.3, $P = .01$); 4) 38% of CFM group and 25% of controls had learning problems; 5) learning problems were associated with lower SES, Latino families, and bilingual families; 6) compared to controls, among CFM phenotypes, microtia with mandibular hypoplasia had lower vocabulary, reading, and writing scores and microtia only had lower reading scores; 7) 72% of children with CFM and 33% of control group received intervention services; 8) 70% of the CFM group failed a hearing screening
Speltz et al. (2018) ¹⁶	1) average range scores for cognitive and motor development and language for CFM and control groups with no differences after accounting for demographics; 2) 21% of CFM group and 16% of controls had an area of delay; 3) for CFM group, males were more likely to show an area of delay; 4) there were no consistent differences based on CFM phenotype; 5) for all children, lower SES and public insurance more likely to show an area of delay; 6) for CFM group, 33% had bilateral hearing loss, 62% had unilateral hearing loss, and 5% had no hearing loss; 7) 51% of CFM group used hearing aids; 8) 65% of CFM group and 1% of control group received intervention services
Umbaugh et al. (2020) ⁵⁰	1) of 254 Facebook group posts, posts seeking guidance were most common (43%) and generated 13 times as many comments in response; 2) promotional posts (33%) often focused on building awareness; 3) posts sharing experiences (24%) were generally positive and generated a high rate of 81 "likes" per post; 4) emotional content were seen in 16% of posts; 5) authors noted the Facebook groups functioned primarily in sharing of medical information in the posts and comments
Wallace et al. (2018) ⁴⁸	1) CBCL, TRF, and YSR mean scores all in the average range; 2) adolescents with CFM had lower externalizing problems by parent-report (adjusted ES = -0.3, $P = .02$) and teacher-report (adjusted ES = -0.2, $P = .05$) compared to the control group; 3) although similar to controls and in the average to high range, CFM group had lower social functioning on Pediatric Quality of Life (PedsQL) by self-report (adjusted ES = -0.3, $P = .03$) and parent-report (adjusted ES = -0.3, $P = .04$); 4) there were no differences on CCC-2 by parent or teacher report; 5) clinical level range of concern in any area was 38% for CFM group and 45% for control group; 6) there was no consistent pattern of differences by CFM phenotype; 7) parents of adolescents with CFM reported fewer rule breaking concerns (adjusted ES = -0.6, $P = .001$) and less aggressive behavior (adjusted ES = -0.3, $P = .02$); 8) adolescents with CFM reported fewer rule breaking concerns (adjusted ES = -0.9, $P = .005$) and more social problems (adjusted ES = 0.7, $P = .006$); 9) the social functioning items endorsed by

adolescents with CFM included being left out and being teased; 10) 70% of CFM group failed hearing screening, with no consistent pattern of scores based on hearing status

Supplemental Table 3. Pre- and post-operative psychosocial outcomes of ear reconstruction/canaloplasty key findings

Citation	Key Findings
Awan et al. (2018) ⁶⁵	1) preoperatively, adolescents with microtia had significantly higher scores on Body Image Disturbance Questionnaire (BIDQ), lower scores on Students' Life Satisfaction Scale (SLSS), and lower scores on Quality of Life (Peds-QL) total score and psychosocial, emotional, and social functioning by parent report; 2) preoperatively, there were no differences for Peds-QL physical health or school functioning by parent report; 3) postoperatively, adolescents with microtia showed significantly improved scores compared to control for BIDQ, SLSS, and Peds-QL total, emotional, and social functioning by parent and self-report for physical health or school functioning; 4) BIDQ improvement explained 52% of the variance in SLSS; 5) postoperative scores for the microtia group were similar to the control group scores across measures
Cho et al. (2020) ⁶⁶	1) prior to canaloplasty, children with aural atresia ages 9-11 had lower self-consciousness distress scores than ages 12-24; 2) there were no preoperative differences by gender or history of auricular elevation; 3) postoperatively, self-consciousness distress decreased on 6 items; 4) postoperatively, hearing was improved; 5) self-consciousness scores were not related to improved hearing status
Hempel et al. (2014) ⁶²	1) Glasgow Health Status Inventory (GHSI) total scores before ear reconstruction were $M = 39$ and after reconstruction were $M = 44$; 2) Short Form 36 Health Survey (SF-36) mean scores improved from baseline to after reconstruction for Physical Functioning, General Health Perception, Social Functioning, and Mental Health; 3) no change in the already high functioning three remaining subscales; 3) Childhood Experiences Questionnaire (CEQ) mean scores were baseline $M = 70$ and postoperatively $M = 75$; 4) Kidscreen-52 mean scores improved from baseline to after reconstruction for Moods and Emotions, Self-Perception, and Social Acceptance, without change in the other subscales
Horlock et al. (2005) ⁴¹	1) motivation for ear reconstruction included dissatisfaction with ear appearance (31% of children and 25% of adults) and children also wanted to prevent teasing (25%); 2) preoperative retrospective report of: teasing for children taking place at school (100%), home (14%), or other locations; 3) preoperative report of: confidence (71%), depression (55%), and anxiety (52%); 4) On CEQ, compared to norms, children and adults reported behavioral concerns based on recall of preoperative functioning; 5) postoperatively, 83% children and 83% adults reported experiencing less teasing; 6) postoperatively, self-consciousness improved for 91% children and 91% adults and social life improved for 50% of adults and 42% of children; 7) on CEQ, postop improvement in: less teasing and social concerns; 8) on CEQ, postop improvement in: less teasing and social concerns; 9) postoperative ears rated positively by adults and children
Johns et al. (2015) ⁶⁴	1) all Behavioral Assessment System for Children-Second Edition (BASC-2) mean scores in the areas of social skills and after ear reconstruction; 2) preoperatively, children ages 7-10 had higher negative emotions than children ages 3-5 by parent report; 3) preoperatively, the 7-10 year old group had lower Social Skills than children ages 3-5; 4) children showed decreased negative emotions and microtia awareness after surgery; 5) all ages showed decreased depression and higher social skills after surgery; 6) parents tended to underestimate younger children's negative emotions and microtia awareness compared to children's self-report
Johns et al. (2017) ⁴⁰	1) before ear reconstruction, by self-report, 31% of children ages 3-5 were teased and 100% of children ages 6-10 were teased and, by parent-report, 39% of children ages 3-5 were teased and 93% of children ages 6-10 were teased; 2) postoperatively, by self-report, teasing decreased to 8% for children ages 3-5 and went down to 0% for children ages 6-10 and, by parent report, teasing took place for 8% of children aged 3-5 and 20% of children ages 6-10; 3) the age when teasing started was 4.6 years by self-report and 3.8 years by parent report; 4) before surgery, teasing was associated with self-report of feeling sad, worried, and mad and after surgery teasing was associated with hiding emotions; 5) negative emotions were higher for children ages 6-10 than 3-5 before surgery; 6) negative feelings decreased after surgery by self-report for all ages

Steffen et al. (2008) ⁶¹	1) positive social support was received after ear reconstruction for 94% of participants; 2) postoperative concerns were endorsed for quality of the cosmetic outcome (59%), surgery complications (44%), (28%), and length of hospital stay (22%); 4) no significant change in retrospectively reported pre- and postoperative scores on Frankfurter Selbstkonzeptskalen (FSKN), which were in the neutral to positive range
Steffen et al. (2010) ⁶³	1) positive social support after ear reconstruction for 94% who had surgery and positive support was reported for deciding not to have surgery for 73% of those who did not have surgery; 3) postoperatively, the length of stay was judged to be unacceptable to 31% and chest movement pain reported by 40%; 4) there were no significant changes after surgery for self-esteem on FSKN or differences between the surgical and nonsurgical group; 5) the surgical group improved psychosocial attitude on FSKN from baseline to after surgery; 6) the nonsurgical group showed no change in psychosocial attitude on FSKN compared to both baseline and postoperative scores of surgical group; 7) the study noted the length of hospital stay needed for healthcare guidelines and billing in Germany is 15-16 days, with length of stay considered stressful to 50% of the surgical group and 77% of the group that decided against surgery

Supplemental Table 4. Quality of life and patient satisfaction key findings

Citation	Key Findings
Akter et al. (2017) ⁷⁸	1) most aspects of postoperative ear appearance were rated highly by patients (medians of 4 on a 5-point scale); 2) cartilage bulging out of ribs was the lowest rated area (median of 3 out on a 5-point scale); 3) 84% were satisfied with reconstructed ear and 90% felt the surgeon explained the surgery well; 4) 16% reported inadequate pain control (36%), trouble with chest scar (30%), anxiety in seeing hairdresser (16%), not being conscious about ear (15%), and hiding ear with hat and in photos (13%); 5) the authors recommend postoperative pain management and addressing chest wall deformities
Bradford et al. (2020) ⁸⁵	1) children and caregivers were similar in their responses; 2) trend toward children with microtia wanting reconstruction earlier than parents and children with craniofacial microsomia (CFM) wanting to achieve facial symmetry sooner than ear reconstruction. <i>Note:</i> Scores reported as difference between child and parent
Braun et al. (2010) ⁷⁵	1) following ear reconstruction, adults reported benefits significantly higher than zero on Glasgow Benefit Inventory (GBI) scales of Total ($M = 21$), General ($M = 30$), and Social Support ($M = 9$), with no difference for the Physical scale ($M = -0.8$); 2) 73% of adults were satisfied with aesthetic outcomes; 3) adults with acquired defects had lower scores than congenital diagnoses; 4) by parent report on Glasgow Children's Benefit Inventory (GCBI), children had benefits significantly higher than zero benefit for Total ($M = 28$), Emotion ($M = 35$), and Learning ($M = 17$), with no difference for the Physical Health scale ($M = 5$); 5) 83% of parents were satisfied with children's aesthetic outcomes; 6) no differences were found by sex for adults or parents for their children; 7) adults and parents unsatisfied with aesthetic outcomes rated no benefits above zero; 8) mean postoperative pain rating on scale of 0-100 was 14.2 for adults and 4.6 for children; 9) 85% of children slept on their reconstructed ear; 10) adults were mainly critical of scars (40%), ear shape (36%), and other complaints (29%) related to complications at donor site of free skin flap; 11) scars were also the primary concern of parents (45%) and children; 12) authors recommend refining surgical technique based on areas of concern of patients and providing preoperative counseling about aesthetic outcomes and possible complications
Braun et al. (2013) ⁷⁶	1) parents of all children reported benefit of surgery on the GCBI with (Total $M = 31$); 2) all children reported benefit of surgery on GBI (Total $M = 24$); 3) mean aesthetic ratings of scars from best (1) to worst (5) were scalp ($M = 3$), contralateral post-auricular scar ($M = 3$), and groin ($M = 3$); 4) attraction of public attention was 7% to 20%; 5) sensation disorders around scars were reported for 7% for contralateral post-auricular scar, 47% for both scalp and groin scars; 6) rating scars from 0-10, pain was 0.1 to 1.3, annoyance was 0.1 to 1.3, and limitations in daily life were 0.7 to 1.3, and feelings of shame was 0.5 to 1.5; 6) authors recommend work on minimizing scarring
Cui et al. (2017) ⁸⁰	1) average satisfaction of ear substructures was 74%, with highest scores for the helix and lowest scores for the concha; 2) overall satisfaction was higher among children ages 7-12 (60%) compared to group aged 13-45; 3) satisfaction was lower for higher Body Mass Index patients, which authors suggested may be related to blunted auricular contour and more hypertrophic thoracic scarring; 4) left-sided microtia had higher satisfaction than right-sided, which authors suggested could be related to general higher ratings of left side of face; 5) there were no significant differences by sex, with 21% of the sample identified as female
Cui et al. (2018) ⁸²	1) patient satisfaction measure met psychometric criteria with items including satisfaction of 9 ear substructures, the position of the ear, the upper and lower halves of the ear, and overall impression
Fan et al. (2017) ⁷²	1) auricular reconstruction was rated as highly satisfactory by 89% and basically satisfactory by 11%; 2) sound thresholds were significantly improved with use of bone-conducting hearing aid implant compared to soft-band hearing aid; 3) speech discrimination scores were also significantly higher with implant (90%) compared to soft-band (80%) or unaided (46%); 4) scores on the Abbreviated Profile of Hearing Aid Benefit (APHAB) were significantly higher with implant (90%) compared to soft-band (80%) or unaided (46%); 5) impact of disability with the implant, followed by the soft-band, for ease of communication, background noise, and hearing in noisy situations

	reverberation, and aversiveness of sound; 5) scores on the International Outcome Inventory for similar for the implant and soft-band hearing aid; 6) scores on the GCBI were also similar for the implant and soft-band hearing aid ($M = 33$)
Hempel et al. (2013) ⁷⁷	1) following ear reconstruction, adults with CFM had mean scores on GBI scales of Total $M = 21$, Social Support $M = 6$, and Physical $M = 6$; adults with microtia had GBI total score $M = 21$; 2) following ear reconstruction, parents of children with CFM had mean scores on GCBI of Total $M = 40$, Emotion $M = 43$, Learning $M = 36$, Physical Health $M = 22$; parents of children with microtia had a GCBI total score $M = 33$
Khetani et al. (2013) ⁶⁸	1) parent-reported health-related quality of life (QoL) was below a cutoff of 77 for 22% of children with CFM and 15% for control group; 2) there were no group differences in mean QoL scores by self-report; 3) QoL was lower for CFM group on physical (adjusted ES = 0.26, $P = .004$), social (adjusted ES = 0.32, $P = .001$), and school (adjusted ES = 0.32, $P = .001$), and total functioning (adjusted ES = 0.31, $P = .001$); 4) PPVT-III mean scores were in the average range; 5) PPVT-III scores were higher for cases ($M = 97$) than controls ($M = 106$) without changes in the QoL analyses when accounting for hearing level
Klassen et al. (2018) ⁸⁷	1) item pool was within 3 main areas, starting with appearance concern items (29%) that covered part, position, qualitative judgement (e.g., good, different), scenario (e.g., up close, in mirrors), symmetry, and visibility with generally negative comments (78%) and positive comments were 22%; 2) physical concern items (9%) covered hearing and adverse effects after surgery with 10% in the past week, including itchy, tingling, pain, numbness, discoloration, activity limitations, and sleep issues; 3) Health-Related Quality of Life (63%) items covered psychological, social, and school specific concerns; 4) psychological items were negative (67%) focusing on ear concealment, confidence, self-consciousness, and normal; 5) social items were generally positive (58%) focusing on support provided, as well as teasing, staring, and feeling judged; 6) school items were negative (75%) and focusing on missing items
Klassen et al. (2021) ⁸⁹	1) final scales met psychometric criteria; 2) lower Appearance scores significantly correlated with Psychological ($r = 0.53$), Social ($r = 0.41$), and School ($r = 0.32$) scales; 3) when comparing the preoperative (39%) participants, mean scores were significantly lower across scales preoperatively; 4) the Appearance scale mean twice as high postoperatively; 5) the most frequently reported adverse effects were sensitivity, itchiness, and sleep issues
Kristiansen et al. (2013) ⁷⁹	1) 69% participants said they wanted to have ear reconstruction, 58% said their parents wanted to have ear reconstruction, and 15% reported the surgeon wanted the surgery; 2) following ear reconstruction, aesthetic satisfaction was similar with no differences by age, sex number of surgeries, or years since completing surgery; 3) aesthetic satisfaction was lower for those who had surgery done earlier 2000-2005 (62%) than from 2006-2010 (83%), which was attributed to improved surgical technique; 4) functional difficulties included troubles with wearing glasses (29%), problems cleaning ear (29%), and problems in thoracic donor region (21%); 5) compared to before surgery, patients felt happier; 6) 42% hid their ear before surgery and 37% hid reconstructed ear with hair after surgery; 7) 42% of patients for having surgery were wanting two identical ears (81%), thinking their ear looked strange (42%), asking questions about ear (37%), wanting to wear glasses (36%), and being teased (12%); 8) pain management after surgery was satisfactory for 90%; 9) 91% felt the reconstructed ear was part of them
Ren et al. (2012) ⁸³	1) measure met psychometric criteria; 2) item wording was based on language of the participants: "I cannot tell the direction of a sound," "I care about whether others notice my ears," and "Ear problems affect my studies"
Schrötzlmair et al. (2021) ⁷⁴	1) postoperative ear reconstruction complications (e.g., extrusion) were reported for 25% of patients; 2) aesthetic deficits (e.g., scar) were reported for 39%; 3) on a scale from -100 to +100, patient self-reported satisfaction ($M = 46$) and caregiver-reported satisfaction ($M = 48$) were similar with 82% of patients and 82% of parents reporting overall satisfaction with surgery; 4) GBI mean scores by adolescents and adults were Total ($M = 17$), General ($M = 25$), Physical Health ($M = -0.2$), and Social Support ($M = 5$); 5) GCBI scores by adolescents were Total ($M = 19$), Physical Health ($M = 11$), Emotion ($M = 26$), Learning ($M = 20$), and Vitality ($M = 17$)

	<p>patient's surgery-related benefit was correlated with aesthetic satisfaction ($r = 0.57$); 6) surgery not influenced by postoperative complications or surgical revision, but was significantly lower for those who had framework extrusion ($M = 6$) than patients ($n = 76$) who didn't have an extrusion ($M = 10$); 7) frequent complaints were for scars (33%), ear shape (27%), and feeling of ear (24%); 8) there were no differences based on sex; 9) patients younger than 15 were more satisfied ($M = 65$) than those older than 15 ($M = 58$); 10) Physical Health was rated higher for those who had simultaneous hearing rehabilitation ($M = 20$) than those who did not ($M = -9$)</p>
Soukup et al. (2012) ⁷³	<p>1) following ear reconstruction, significantly higher benefit than zero on GBI scores for Total ($M = 48$), Social Support ($M = 78$), and Physical ($M = 27$); 2) there were no differences by sex; 3) there were no changes based on age at time of surgery or time that had passed since surgery; 4) a relatively greater benefit was reported among those with microtia as part of a syndrome compared to isolated microtia; 5) across all ages, mean total integration of the ear was 3.5 on a 5-point scale, with significantly lower scores from 1 to 2 for adolescent; 6) moderate significant correlation ($r = 0.50$) between GBI total and surgical outcome</p>
Steffen et al. (2012) ⁸⁴	<p>1) German translation met psychometric criteria; 2) there was convergent and discriminate validity for all measures; 3) following ear reconstruction, 21% of participants found thoracic scar unacceptable; 4) 15% of participants were above the clinical cut-off on the Depression Inventory of Children and Adolescents</p>
Tsangaris et al. (2019) ⁸⁸	<p>1) linguistic and cultural adaptations completed for two scales of EAR-Q in Arabic, Chinese, French, and Spanish with conceptual equivalency after going through rounds of forward and back translation</p>
Younis et al. (2010) ⁸¹	<p>1) following bone-anchored ear prosthesis was placed, skin problems reported by 75% (30% mild, 45% moderate, and 25% severe); 2) daily use of the prosthetic was daily (70%), infrequently (20%), or never (10%); 3) stability was described as stable by 60% and unstable by 20%; 4) aesthetic appearance was rated positive by 60%, terrible by 5%; 5) cleaning abutments was rated as easy for 60% and difficult for 20%; 6) overall satisfaction was positive (60%), neutral (25%), or negative (15%); 7) authors noted dissatisfaction with outcome due to skin problems, most commonly granulation tissue around abutments, which can be related to smoking; 8) authors recommend careful patient selection and pre and postoperative education</p>