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ORIGINAL RESEARCH

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Development of an interdisciplinary microtia-atresia care model: A single-center 20-year experience

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Abstract

Objectives: Microtia and aural atresia are congenital ear anomalies with a wide-ranging spectrum of phenotypes and varied functional and psychosocial consequences for patients. This study seeks to analyze the management of microtia-atresia patients at our center over a 20-year period and to propose recommendations for advancing microtia-atresia care at a national level.

Methods: We performed a retrospective analysis of data from patients presenting to the Massachusetts Eye and Ear (Boston, MA) for initial otolaryngology consultation for congenital microtia and/or aural atresia between 1999 and 2018.

Results: Over the 20-year study period, 229 patients presented to our microtia-atresia center at a median age of 7 years. The severity of microtia was most commonly classified as grade III ($n = 87$, 38%), 61% ($n = 140$) of patients had complete atresia, the median Jahrsdoerfer grading scale score was 6 (range 0–10), and 81 patients (35%) underwent surgery for microtia repair. For hearing rehabilitation, 30 patients (64%) underwent bone conduction device implantation and 17 patients (36%) underwent atresiaplasty. The implementation of an interdisciplinary, longitudinal care model resulted in an increase in patient ($r = 0.819$, $p < .001$) and surgical volume (microtia surgeries, $r = 0.521$, $p = .019$; otologic surgeries, $r = 0.767$, $p < .001$) at our center over time.

Conclusion: An interdisciplinary team approach to microtia-atresia patient care may result in increased patient volume, which could improve aesthetic and hearing outcomes over time by concentrating care and surgical expertise. Future work should aim to establish standardized clinical consensus recommendations to guide the creation of high-quality microtia-atresia care programs.

Level of Evidence: 4.

KEYWORDS

atresiaplasty, aural atresia, auricular reconstruction, bone conduction, Microtia

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1 | INTRODUCTION

Microtia (abnormal development of the auricle) and aural atresia (absence or stenosis of the external auditory canal [EAC]) are congenital malformations with a variable spectrum of phenotypes, ranging from minimal abnormalities to major structural alterations. These conditions result in varying degrees of functional and psychosocial consequences for patients and families. The prevalence of microtia has been estimated to be between 1 and 4 per 10,000 births.[1–3] The EAC and auricle are both derivatives of the first and second branchial arches and their intervening branchial clefts and pharyngeal pouches. Given the EAC and auricle share parallel embryonic development paths, most patients with grade III and grade IV microtia also manifest aural atresia.[4,5] Microtia may occur as an isolated condition or can be associated with anomalies such as craniofacial microsomia, renal abnormalities, cardiac defects, and mandibulofacial dysostoses (e.g., Treacher-Collins syndrome and Nager syndrome).[1–3]

The management of microtia-atresia patients is perhaps one of the most challenging clinical scenarios faced by otolaryngologists and plastic surgeons. The auricle is a complex three-dimensional structure that may require multiple reconstructive procedures to achieve an aesthetically favorable outcome. Other options for microtia management include observation, the use of a prosthetic ear or alloplastic implant placement.[6] Each of these options carries its own set of challenges and aesthetic outcomes are highly variable. Hearing rehabilitation in the setting of aural atresia poses a unique challenge due to associated facial nerve and middle ear anatomical abnormalities.[7] A draining or infected post-atresiaplasty ear canal can also threaten a newly implanted microtia rib graft while atresiaplasty or osseointegrated implant dissection can threaten the blood supply of an existing microtia repair. Decision-making regarding the techniques and timeline for microtia and aural atresia repair must therefore be carefully coordinated.

Despite the large number of studies and book chapters detailing techniques for microtia and aural atresia reconstruction, there is limited literature describing consensus recommendations for the longitudinal management of microtia-atresia patients. A 2019 paper by Mazeed et al.[8] describes several recommendations for the development and reform of microtia and atresia services in Egypt using the United Kingdom care standards for microtia-atresia as a model. A survey of 22 plastic surgery centers in Egypt found microtia-atresia patient care to be significantly fragmented with 65% of centers treating less than 10 patients annually. They additionally reported that multiple surgeons perform ear reconstruction in 90% of centers and only 25% utilize a multidisciplinary team approach.[8] We suspect that these results are not unique to Egypt; microtia-atresia patient care is significantly fragmented even within major U.S. cities in addition to across this country. Given the complexity and potential morbidity associated with microtia-atresia surgical repair, our institution has moved towards coordinated management by an interdisciplinary team. This study analyzes the evolution of the patient population and aesthetic and hearing interventions performed at a subspecialty tertiary care center treating microtia-atresia patients over a 20-year period. We also describe the creation of our prospective patient data registry and implementation of an interdisciplinary, longitudinal microtia-atresia care model and its impact on temporal trends in patient and surgical volume.

2 | MATERIALS AND METHODS

2.1 | Study design and data collection

A retrospective cohort study of all patients with congenital microtia and/or aural atresia who presented to the microtia-atresia center at the Massachusetts Eye and Ear (Boston, MA) between 1999 and 2018 was performed. This study was approved by the Mass General Brigham Institutional Review Board (IRB) and written informed consent was obtained for publication of patient photographs. All patients with congenital microtia and/or aural atresia who presented to our center during the specified time frame were included even if they elected not to undergo any intervention for microtia repair or hearing rehabilitation, irrespective of patient age at time of presentation and regardless of prior attempts at microtia or atresia repair at outside institutions. We identified 229 consecutive patients who met the study's inclusion criteria and their medical records were reviewed with attention to pre-operative evaluation and counseling, microtia reconstruction techniques, hearing rehabilitation and longitudinal, interdisciplinary follow-up.

2.2 | Data analysis

Pearson's correlation analysis was used to determine the trend in the number of patients presenting to our microtia-atresia center over time and the number of microtia and otologic surgeries performed between 1999–2018. p values were obtained from two-tailed tests and statistical significance was defined as a p value <.05. All statistical analyses were performed using SPSS version 27 (IBM Inc., Armonk, NY). Categorical variables are presented as numbers (n) and proportions (%) and continuous variables are reported as medians with the range.

2.3 | Prospective patient data registry and clinical photo repository

Meticulous yet efficient collection and organization of patient data including clinical photographs is essential to serially evaluate patient outcomes and improve care. All patients who present to our interdisciplinary microtia-atresia center are asked for their consent to enroll in our IRB-approved prospective study involving the longitudinal collection of microtia-atresia patient information and clinical photographs; clinical, photographic, and demographic data are stored in a secure browser-based data repository.

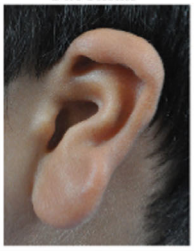
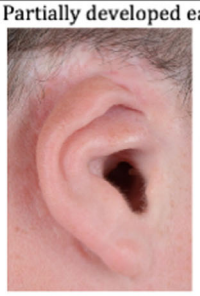
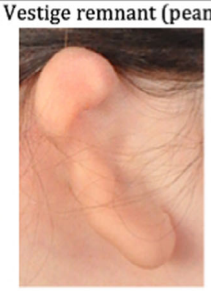
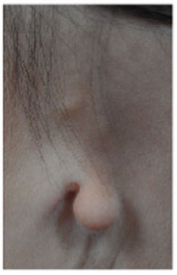
	Microtia Database Intake Form			
Microtia Type	Conchal Type Lobular Type (remnant position: appropriate or inappropriate) Other			
Laterality	Left	Right		
Grade	I: All structures are identifiable but small 		II: Partially developed ear 	
	III: Vestige remnant (peanut) 		IV: Anotia 	
Hairline Position	Favorable	Unfavorable (>20% hair coverage on a new ear)		
Canal Patent?	Yes	No		
Remnant Soft Tissue	Fistula	Pits	None	
STA Pulse	Palpable	Not palpable		
Hemifacial Microsomia	None	Mild	Moderate	Severe
Family History of Microtia	Yes	No	Unknown (adopted)	

FIGURE 1 The microtia intake form utilized for prospective patient data collection. MEEI, Massachusetts Eye and Ear Infirmary; STA, superficial temporal artery

Our facial plastic surgery team created a microtia patient intake form that records specific variables relevant to reconstruction and the proposed repair technique (Figure 1). The form has the advantage of taking less than 1 min for the clinician to complete and is easily entered into the secure electronic database. Likewise, our pediatric otolaryngology and neurotology teams created a smart phrase within our electronic medical record (EMR) system that achieves the same result. The EMR system data has the advantage of permanence in the patient's electronic chart; data are likewise easily transferred into the secure computer database.

To track aesthetic outcomes after microtia reconstruction, we also created a database of standardized photographic documentation, including photos of the external ear at first presentation, then annually until preoperative evaluation, and each postoperative visit (Figure 2). This photographic database is searchable via diagnosis code (i.e., microtia grade, bilateral microtia etc.) and via reconstruction technique (i.e., porous polyethylene framework, rib graft etc.)—thereby generating a list of patients meeting specific diagnostic or therapeutic criteria to facilitate future research studies.

3 | RESULTS

3.1 | Patient characteristics and interdisciplinary care model

Two-hundred and twenty-nine patients presented to our microtia-atresia center at a median age of 7.1 (range 0–56) years between 1999 and 2018. Their demographic and clinical characteristics are listed in Table 1. Fifty-seven percent ($n = 131$) of patients were male and the most common comorbid congenital syndrome was Goldenhar syndrome ($n = 5$, 2.2%). Fifty-four patients (24%) had undergone some form of previous attempt at microtia repair. The severity of microtia was most commonly classified as grade III ($n = 87$, 38%). Sixty-one percent of patients ($n = 140$) had complete atresia and 11% ($n = 26$) of patients presented with bilateral microtia. The median Jahrsdoerfer grading scale score[9] based on preoperative computed tomography (CT) of the temporal bones was 6 (range 0–10).

All patients with congenital microtia-atresia who present to our center are evaluated in a specialized microtia-atresia clinic by an interdisciplinary otolaryngology team, including a pediatric otolaryngologist,

FIGURE 2 Preoperative (A1-4) and postoperative (B1-4) clinical photographs 6 months status-postsecond stage left microtia repair. Photos retrieved from our prospective repository of patient data

neurotologist, audiologist and facial plastic surgeon. The intake forms and data questionnaires are filled out by each provider at time of the initial visit. Monthly microtia-atresia conferences are hosted at which time the interdisciplinary team discusses all patients evaluated during the preceding 4 weeks to develop a personalized management plan for each patient. Subsequent to each monthly conference, a letter is mailed out to the patients' families and their primary care physicians, detailing the microtia-atresia team's consensus recommendations and the importance of consistent follow-up. All patients are encouraged to follow-up in our interdisciplinary microtia-atresia clinic at least once per year to assess their growth and hearing until they are more than a year status-post the conclusion of their final reconstructive surgery.

When microtia presents with additional mild abnormalities (such as skin appendages, accessory or remnant soft tissue), these are addressed surgically as early as 6 months of age (Figure 3). Minor forms of microtia requiring unilateral or contralateral otoplasty, or maneuvers to improve symmetry may be undertaken as early as age five, at which time the contralateral pinna is approximately 80%–90% the size of an adult auricle.[10] It is also around age five that children will attend school and may need an auricle to support eyeglasses or a hearing aid and may also be subject to the psychosocial effects of bullying. For these reasons, minor abnormalities are often corrected around age five. If a rib graft is determined to be required, typically for reconstruction of grade III and IV microtia, we delay ear reconstruction until age 10–12 with exact timing of repair determined by development of the thorax in so much as it will provide adequate costochondral stock. As patients approach the age for rib graft auricular reconstruction, families are counseled regarding this complex reconstructive endeavor that will require full social and family support due to the need for inpatient hospitalization, several postoperative visits and multiples stages of surgery.

Hearing rehabilitation in patients with microtia and aural atresia is also nuanced and similarly requires longitudinal follow-up and a personalized approach tailored to each patient and family. Hearing function in pediatric patients is closely monitored during the first few years of life, beginning with the newborn hearing screen and subsequent non-sedated auditory brainstem response (ABR) testing in the first 3 months of life. A behavioral audiogram is typically obtained at 9–12 months of age and every 6 months thereafter, with close monitoring of language development. Referral to Early Intervention programming occurs at the

TABLE 1 Patient demographics and clinical characteristics

Characteristic	No. of patients (%)
Total No.	229
Sex	
Male	131 (57%)
Female	98 (43%)
Ethnicity	
Caucasian	92 (40%)
Hispanic	39 (17%)
Asian	27 (12%)
Syndromes	
Goldenhar syndrome	5 (2.2%)
Treacher-Collins	3 (1.3%)
Fanconi anemia	2 (0.9%)
S/P prior microtia repair	54 (24%)
Microtia type	
Lobular	59 (26%)
Conchal	31 (14%)
Microtia grade	
1	32 (14%)
2	41 (18%)
3	87 (38%)
4	6 (2.6%)
Bilateral microtia	26 (11%)
Complete atresia	140 (61%)
Median Jahrsdoerfer score	6 (range 0–10)
Median preop PTA	64 (range 22–100) dB
Microtia surgery type	
Autologous rib graft	49 (21%)
Medpor	4 (1.7%)
Other techniques	28 (12%)
Otologic surgery type	
BCD implantation	30 (13%)
Atresiaplasty	17 (7.4%)
Median age at time of presentation (years)	7.1 (range 0–56)
Median age at time of first microtia surgery (years)	10.3 (range 2–54)
Median age at time of first otologic surgery (years)	7.8 (range 3.8–44)
Median follow-up period (months)	24 (range 0–200)

Abbreviations: BCD, bone-conduction hearing device; dB, decibel; preop, preoperative; PTA, pure-tone average; S/P, status-post.

time of the confirmatory ABR. Amplification with a soft-band or adherent bone conduction device (BCD) (ADHEAR, Baha or Ponto) is offered by 6 months of age. Trial of a BCD is strongly recommended for children with bilateral atresia and is also recommended for patients with unilateral conductive hearing loss (CHL) given recent studies demonstrating use-dependent neural plasticity in the setting of chronic CHL.[11] For children approaching school-age, we discuss appropriate accommodations to optimize the classroom listening environment, including strategic seating towards the source of instruction and classroom modifications to reduce ambient noise, such as carpeting on the floor and felt covers on chairs. Use of FM or Bluetooth-based hearing assistive technology to improve signal-to-noise ratio is also encouraged.

CT imaging of the temporal bones is obtained around 4–5 years of age for children with complete atresia; in cases of canal stenosis or where risk of canal cholesteatoma exists, imaging is performed earlier. Older age at time of CT imaging is advantageous as it often allows successful imaging without sedation, the mastoid and calvarium are closer to adequate growth to support osseointegrated BCDs and risks related to radiation-exposure are lower.[12] BCD surgery is typically performed after age five whereas atresiaplasty is generally delayed until after microtia surgery is completed. When microtia surgery is limited to an existing auricular superstructure without the need for costal cartilage grafting, atresiaplasty may be performed prior to microtia repair. However, patients and caregivers are also given the option to employ both hearing rehabilitation techniques in a staged fashion such that a BCD is implanted at age five and then atresiaplasty is performed after the conclusion of all stages of microtia reconstruction. At times, atresiaplasty is also performed concurrently with the final stage of microtia surgery. Notably, the continued evolution of various types of BCDs requires frequent interdisciplinary adaptation of the microtia-atresia reconstructive plan as each type of device necessitates distinct surgical approaches.

We also discuss with patients and caregivers the advantages and disadvantages of BCD implantation versus atresiaplasty based on the status of the ossicles and facial nerve on the CT scan. Given fewer intraoperative risks, lower rates of postoperative complications and favorable hearing outcomes with BCD implantation in comparison to atresiaplasty in our experience, we encourage most patients with complete atresia to trial a BCD before proceeding with atresiaplasty. Patients and their families are also counseled regarding differences in complications and hearing thresholds for BCD transcutaneous magnetic mounting, which results in fewer wound complications, and percutaneous abutment mounting, which offers superior hearing thresholds but has higher rates of postoperative surgical site issues. Given the multitude of BCDs now available, including the recently FDA-approved Osia® System,[13] which utilizes digital piezoelectric stimulation to transmit sound vibrations to the inner ear, we counsel patients regarding the advantages and disadvantages of each type of device. Over several years of counseling regarding the different techniques for microtia repair and hearing rehabilitation, our patients and their families form a longitudinal bond with the interdisciplinary microtia-atresia team and are able to make an informed decision regarding their personal preference for future microtia-atresia repair.

3.2 | Surgical management techniques

Between 1998 and 2018, 81 patients (35%) underwent surgery for congenital microtia at our center with 49 of these patients (60%)

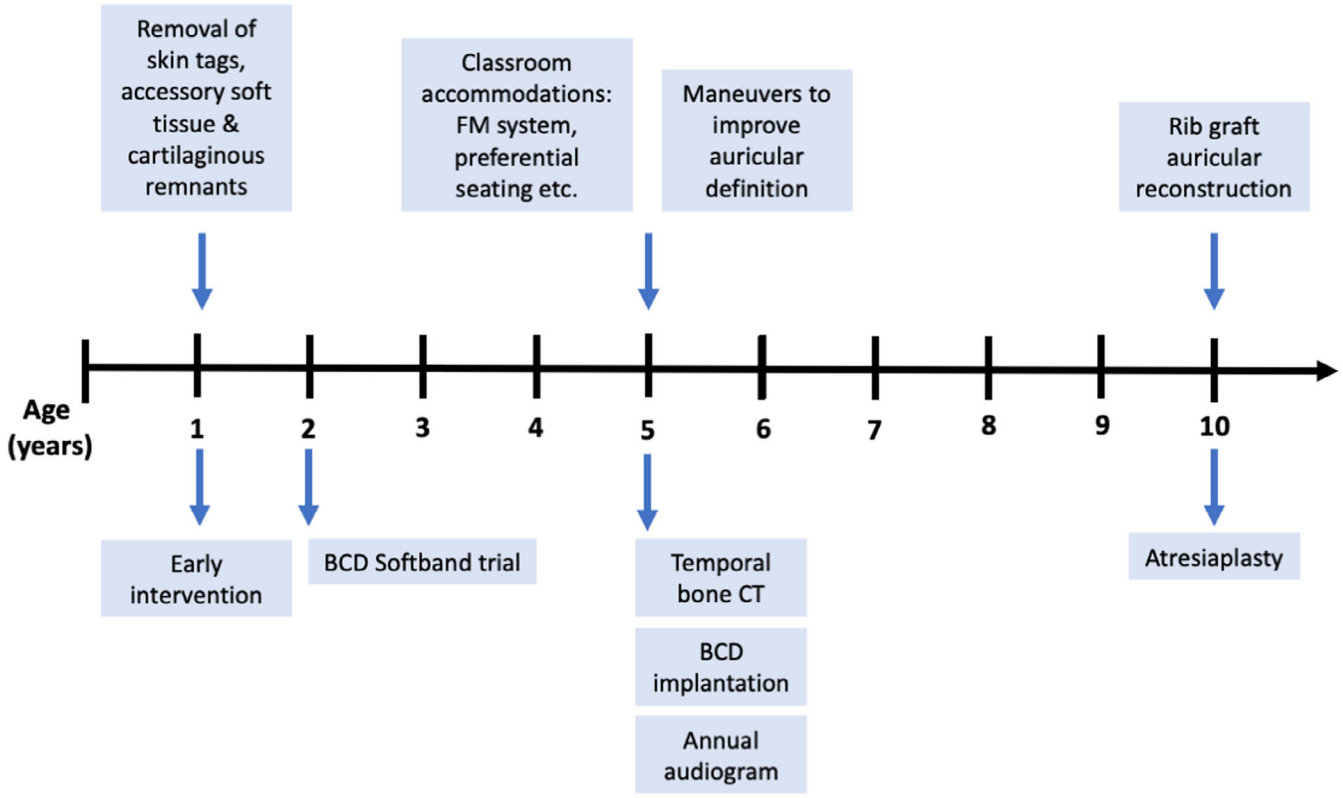


FIGURE 3 Timeline of diagnostic and therapeutic interventions for microtia and atresia patients. BCD, bone-conduction hearing device; CT, computed tomography; FM, frequency modulation

electing to undergo autologous costal cartilage microtia reconstruction at a median age of 8.3 (range 5–32) years. Only four patients (5%) underwent alloplastic porous polyethylene implant placement and two patients (2%) elected for placement of an auricular prosthesis. For hearing rehabilitation, 17 patients (36%) elected to undergo atresiaplasty while 30 patients (64%) underwent BCD implantation. Among these 30 patients, 19 (63%) patients elected to undergo Baha (Bone-anchored hearing aid) Attract implantation, 10 (33%) patients received a Baha Connect and one (3%) patient received a Ponto device. Decisions regarding choice of hearing rehabilitation technique were based upon an interdisciplinary discussion with the microtia-atresia team, the patients and their families and depended on multiple factors including: prior attempts at repair at outside institutions; the severity of microtia and/or aural atresia; significant sensorineural hearing loss; the anatomical status of the middle ear, inner ear and facial nerve; presence of comorbidities such as craniofacial syndromes, developmental delay and autism spectrum disorder; and patient/ family preference.

3.3 | Temporal trends in patient and surgical volume

From 1999 to 2018, there was an overall increasing trend in the number of patients (Pearson's coefficient = 0.819; $p < .001$) presenting to our microtia-atresia center for initial consultation (Figure 4). There was additionally a statistically significant rise in the number of microtia surgeries (Pearson's coefficient = 0.521; $p = .019$) and BCD or atresia surgeries (Pearson's coefficient = 0.767; $p < .001$) performed at our center over this 20-year period (Figure 5). Our interdisciplinary, longitudinal approach to microtia-atresia patient care was implemented in 2013. We postulate that the increase in patient and surgical volume after 2013 may be attributed to the introduction of this new care model. The rise in microtia-atresia surgical volume at our center, we speculate, may result in improved patient aesthetic and hearing outcomes over time.

4 | DISCUSSION

The surgical management of congenital microtia and atresia is perhaps one of the most challenging endeavors undertaken by otolaryngologists and plastic surgeons. There is extensive literature detailing surgical techniques for microtia-atresia repair, but an interdisciplinary care model and prospective patient data collection have not been well described. A 2019 review by Zhang et al.[14] aimed to provide international recommendations for functional ear reconstruction for patients with microtia and aural atresia. The review outlines the definition and classification of microtia and atresia, reconstructive options and future research directions. Although the abstract mentions that all patients

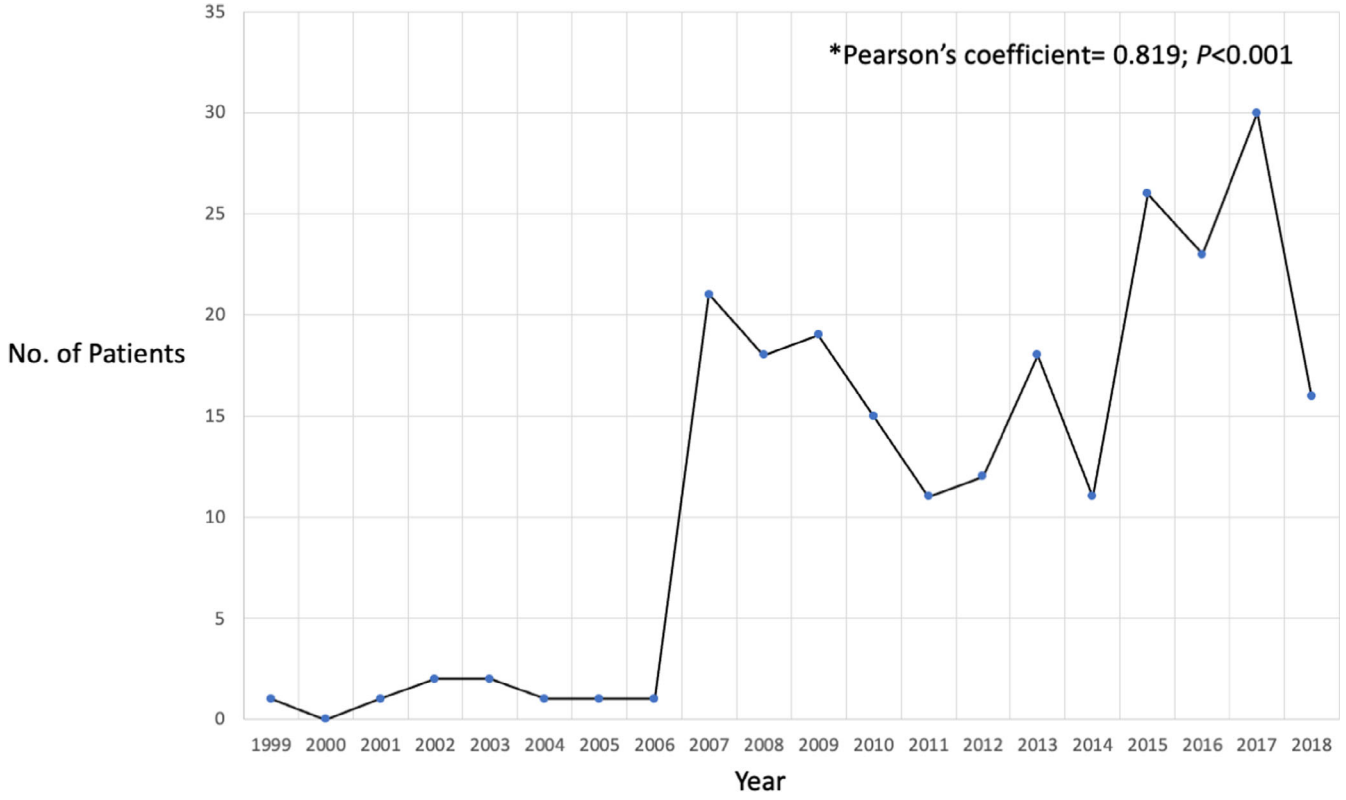


FIGURE 4 Rise in the number of patients presenting to our center over 20 years (Pearson's correlation coefficient = 0.819; $p < .001$)

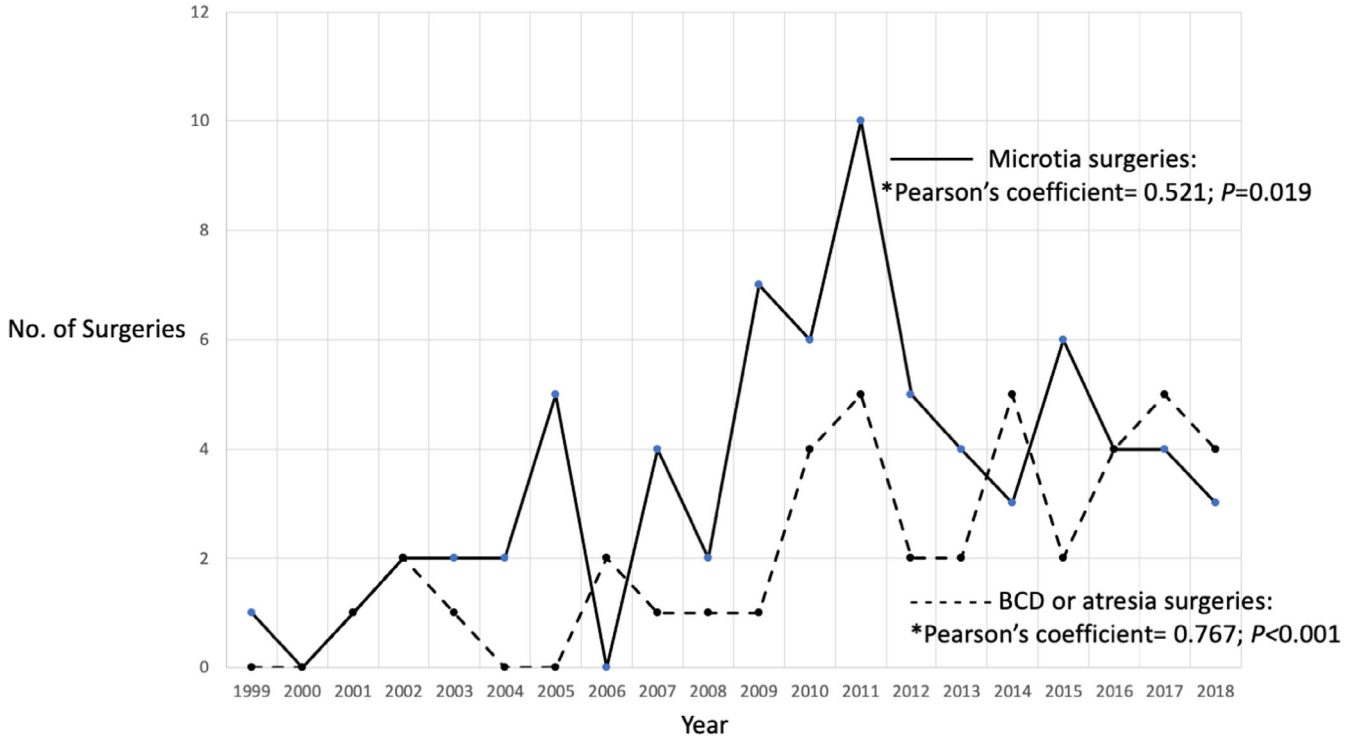


FIGURE 5 Increase in the number of microtia surgeries (Pearson's correlation coefficient = 0.521; $p = .019$) and atresiaplasty or BCD implantations (Pearson's correlation coefficient = 0.767; $p < .001$) performed over 20 years. BCD, bone-conduction hearing device

should be seen in the setting of a multidisciplinary team, the details of such a care model are not described in the body of the paper. In this report, we outline our center's approach to providing personalized, longitudinal and interdisciplinary care to microtia-atresia patients and the development of a prospective data registry of clinical information and photographs to facilitate future research. Analysis of the cohort of patients presenting to our microtia-atresia center over a 20-year period demonstrates that implementation of an interdisciplinary, longitudinal care model may result in increased patient and surgical volume, which in turn, may improve patient outcomes over time by concentrating care and surgical expertise. Although there are several conceptual advantages to interdisciplinary team management of microtia-atresia patients, demonstrating a direct positive impact on aesthetic and hearing outcomes is limited by confounding factors linked to substantial changes in microtia repair techniques and BCD advancements during the study period.

Many societies and panels have successfully developed consensus recommendations

to standardize and optimize the care of patients with rare or challenging conditions such as congenital cleft lip and palate,[15,16] pediatric aerodigestive disorders[17] and Pierre Robin sequence.[18] Multidisciplinary, team-based care for patients with cleft and craniofacial abnormalities, for example, has been the standard practice of care in North America and Europe for more than 30 years.[15] Although many cleft teams have been in existence for several decades, new centers are encouraged to apply for certification through the American Cleft Palate—Craniofacial Association (ACPA), the largest international organization of clinicians that care for patients with orofacial clefts.[19] Additionally, to assist surgeons interested in starting a cleft team, the organization has previously developed a list of action items to guide the assembly of a high-quality multidisciplinary center.[19]

To further the goal of advancing microtia-atresia patient care, microtia and atresia experts should review the existing literature and develop international consensus recommendations. The establishment of standardized guidelines for microtia-atresia centers may foster both interdisciplinary clinical care and patient data collection via prospective registries, thereby facilitating future multicenter outcome studies needed to advance microtia-atresia care. The creation of such a clinical consensus report would provide the initial framework necessary to resolve the current fragmentation of microtia-atresia patient care and help guide institutions in the establishment of high quality, interdisciplinary care models, which may improve aesthetic and hearing outcomes.

5 | CONCLUSION

The management of patients with congenital microtia and aural atresia is perhaps one of the most challenging clinical scenarios faced by otolaryngologists and plastic surgeons. Implementation of an interdisciplinary care model may result in increased patient and surgical volume, with the potential to lead to improved aesthetic and hearing outcomes over time via concentration of care and surgical expertise. Patient care could therefore be advanced by the development of clinical consensus recommendations for microtia-atresia centers via a future meeting or international survey of otolaryngology and plastic surgery experts.

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CONFLICT OF INTEREST

Michael S. Cohen: Med El Corporation, sponsored research agreement.

Alicia M. Quesnel: Frequency Therapeutics, sponsored research agreement; Grace Medical, sponsored research agreement, licensed patent.

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