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Ten-year retrospective evaluation of therapeutic choices and related satisfaction in patients with auricular deformities

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ABSTRACT

To adequately counsel future patients with auricular abnormalities, this study aimed to analyze the therapeutic choices patients make after consultation concerning their auricle and/or hearing. All patients who visited the auricle consultation between January 2010 and January 2020 were included. This comprises patients with all types of auricular anomalies of both congenital and acquired etiology. Follow-up was performed by telephone to obtain informed consent and to take a short nonvalidated questionnaire regarding their therapeutic choices and satisfaction with these choices. Patient characteristics and information regarding therapeutic choices after consultation were subsequently obtained from the patients' medical files. The main outcome parameters were the patients' therapeutic choices regarding the auricle and hearing. A total of 134 patients (61.2%) were included with a mean follow-up of 3.85 years. For congenital and acquired etiologies, 72.8% and 18.2% declined reconstruction, 14.0% and 18.2% chose autologous, 5.3% and 0% chose alloplastic, 6.1% and 45.4% chose prosthetic, and 1.8% and 18.2% chose another type of reconstruction, respectively. A total of 54.9% declined hearing rehabilitation, while 38.2% chose percutaneous bone conduction device (BCD), 2.0% transcutaneous BCD, and 4.9% BCD on a softband. The overall satisfaction score was 8.7/10 concerning the auricle and 8.3/10 regarding hearing. In conclusion, most patients with a congenital etiology chose to wait or decided against auricular reconstruction, while patients with acquired etiologies mostly opted for reconstruction by a prosthesis. Patients or their caregivers were very satisfied with their choice, regardless of whether this implies reconstruction or no reconstruction, which emphasizes the importance of shared decision-making and thorough counseling.

1. Introduction

Currently, evaluation of the quality of health care provided for patients is more important than ever. Patient-reported outcome measures (PROMs) provide a better understanding of treatment efficacy, and their implementation is effective in terms of in-hospital complications (Rotter et al., 2012). On the other hand, clinical pathways have provided standardization. Both PROMs and clinical pathways have ensured improvement of outcomes for patients requiring multidisciplinary care, such as patients with (congenital or acquired) abnormalities of the auricle. This multidisciplinary aspect of care is beneficial for these patients, as the auricle, hearing, and appearance should be addressed.

A patient's demand for health care may vary depending on the type or severity of the auricular deformity, and patients seek consultation to explore their possibilities concerning treatment. First, patients can decide to wait and/or decline auricular reconstruction. If patients choose auricular reconstruction, roughly three different techniques are available: autologous, alloplastic, and prosthetic auricular reconstruction (Vijverberg et al., 2021). Second,

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as auricular deformities can be accompanied by conductive hearing loss due to atresia of the external hearing canal and middle ear, some patients might wish to also explore the possibility of hearing rehabilitation. Rehabilitation of hearing is important considering its influence on speech-language development and communication in younger children. Patients can decide against rehabilitation, especially in cases of unilateral hearing loss, preferably after an extensive trial period. Options to rehabilitate hearing in patients with congenital conductive hearing loss as a result of atresia of the ear canal include a percutaneous or transcutaneous bone conduction device, middle ear implant, or in specific cases of surgical (atresia) repair (Zwartenkot et al., 2016; Vogt et al., 2018).

Satisfaction in patients with auricular deformities (and facial anomalies in general) has been reported in the literature. Most studies used either a nonvalidated and self-constructed questionnaire or the Glasgow Benefit Inventory (GBI), which measures health-related quality of life after a procedure (Robinson et al., 1996; Dings et al., 2020). Patients with auricular deformities showed an increase in both quality of life and satisfaction after a procedure (Soukup et al., 2012; Kievit et al., 2013; Vijverberg et al., 2021).

A previous study evaluated the trajectory of health care regarding patients with auricular abnormalities seeking consultation, which aided in organizing the care for these patients more efficiently (Hol et al., 2016). However, in this study and other previously published literature, very limited data are available on the choices these patients make (Steffen et al., 2010). In addition, most studies focus on short-term results and analyze only patients who opt for surgery, neglecting patients who decide against surgery. Analyzing outcomes in patients with auricular deformities who decline auricular reconstruction is important to adequately counsel future patients seeking consultation. An additional and more thorough long-term evaluation is needed.

Therefore, the aim of the current study was to analyze all patients with auricular abnormalities and evaluate which therapeutic choice they made concerning their auricle and/or hearing and to assess their associated satisfaction with these choices.

2. Materials & methods

2.1. Study design

All patients who visited the auricle consultation between January 2010 and January 2020 at the Radboudumc, Nijmegen, the Netherlands, were included in the current study. This comprises patients with both congenital and acquired etiologies and all types of severity. All patients were followed-up with a phone call to obtain informed consent and to complete a short nonvalidated questionnaire. This questionnaire was administered to the patient's caregiver if the patient was underage (<18 years).

After informed consent was obtained, the questionnaire was taken, and subsequently, the patients' medical charts were retrospectively reviewed. Information regarding therapeutic choices at the time of consultation was retrieved from the medical charts and supplemented with the information the patient (or caregiver) provided at the time of follow-up. Demographic and surgical characteristics were noted (age, sex, etiology, syndrome, laterality, microtia and atresia classification, follow-up time, and if applicable, type of surgery). Microtia was categorized using Nagata's classification (Nagata, 1993).

Patients are usually eligible for autologous reconstruction when they are 10–12 years old (Siegert et al., 2009). Alloplastic reconstruction is feasible from four years of age, as is prosthetic reconstruction. Before deciding on a final decision on hearing rehabilitation, patients were offered a trial with a bone conduction device for at least a few weeks, as the added value in unilateral congenital conductive hearing loss is not always clear within a few days. Patients younger than four years old are not considered eligible for percutaneous treatment as a definite hearing solution and were offered a bone conduction device on a softband as a temporary solution until the preferred age for implantation (4–6 years) is reached (Verhagen et al., 2008). All surgeries were performed at the Radboudumc, except for the alloplastic auricular reconstruction. If patients chose alloplastic auricular reconstruction, they were referred to a different center.

This clinical investigation was performed in accordance with the current version of the Declaration of Helsinki and Good Clinical Practice (International Conference on Harmonization Good Clinical Practice) and was approved by the local ethical committee (file number 2020–6183).

2.2. Outcome parameters

The primary outcome parameter was the patients' therapeutic choices regarding the auricle and hearing. The possible options regarding the auricle were as follows: 1. To wait and accept the current situation, 2. Auricular reconstruction using autologous rib cartilage, 3. Auricular reconstruction using an alloplastic framework, 4. Auricular reconstruction using an external prosthesis, or 5. Other (in case of more limited surgery, e.g., in trauma or cup-ear). Regarding hearing, the possible options were as follows: 1. To wait and accept the current situation, 2. Percutaneous bone implant, 3. Transcutaneous bone implant, 4. Middle ear implant, or 5. Surgical (atresia) repair. The secondary outcome parameter was the patients' satisfaction regarding their choice, which was scored on an NRS-scale from 0 (most dissatisfied) to 10 (most satisfied). Patient substantiation for this score was noted to further improve the auricular consultation.

2.3. Statistical analysis

All data were analyzed using descriptive statistics with IBM SPSS statistics, version 25 (IBM Corp., Armonk, NY, USA). For continuous variables, medians, standard deviations, and ranges were reported. For dichotomous variables, frequencies were reported. The follow-up time was defined as the time between the first visit and the date of assessment. Logistic linear regression was used to analyze the relationship between etiology (congenital or acquired) and outcome (wait or reconstruction), independent of age.

3. Results

3.1. Patients

A total of 219 new patients visited the Nijmegen auricle consultation between January 2010 and January 2020 and were eligible for inclusion. From these patients, 134 patients (61.2%) were reached and provided informed consent and were included in the current study. The remaining patients could either not be contacted or did not want to participate in the study.

The demographic characteristics of the patients are presented in Tables 1 and 2, & 3. The median age at the time of consultation was 12.5 years (min 0 – max 84; SD 18.4). The majority of patients who presented at the auricle consultation were between 0 and 19 years old (73.3%), which indicates that the majority of questionnaires were answered by (one of) the patient's caregivers. The mean follow-up time was 3.9 years (0.4–9.9 years; SD 2.3). A total of 35 patients (26.1%) were syndromic, which included craniofacial microsomia (n = 16), Goldenhar (n = 13), Treacher Collins (n = 3),

Table 1
Demographic characteristics of the total population.

Variable	Total population (n = 134)	
Gender		
Male	88	(65.7%)
Female	46	(34.3%)
Age		
Total		
0–1	7	(5.2%)
2–10	47	(35.1%)
11–20	49	(36.6%)
21–30	9	(6.7%)
31–40	5	(3.7%)
41–50	6	(4.5%)
51–60	3	(2.2%)
60+	8	(5.9%)
Surgical history		
Regarding auricle[a]	25	(18.6%)
Regarding hearing[a]	14	(10.4%)
Radiotherapy	1	(0.7%)
None	95	(70.9%)
Syndromic	35	(26.1%)

[a] One patient underwent both previous auricular reconstruction and atresiaplasty surgery.

Brachio-Oto-Renal (n = 1), CHARGE (n = 1), and Pierre Robin sequences (n = 1).

All patients (n = 134) were divided into congenital and acquired etiologies. Congenital causes (n = 120, 88.8%) included a type of microtia (n = 102 patients, 76.9%), helical rim deformity (n = 9, 6.7%), cup-ear (n = 5, 3.7%), and protruding ears (n = 3, 2.2%). Acquired causes (n = 12, 9.0%) included malignancy (n = 7, 5.2%), traumatic (n = 4, 3.0%), and chondritis of the auricle (n = 1, 0.7%).

Table 4
Choices regarding auricle.

Congenital (n = 114)		
Autologous	16	(14.0%)
Alloplastic	6	(5.3%)
Prosthetic	7	(6.1%)
Wait/do nothing	83	(72.8%)
Other[a]	2	(1.8%)
Acquired (n = 11)		
Autologous	2	(18.2%)
Alloplastic	0	(0.0%)
Prosthetic	5	(45.4%)
Wait/do nothing	2	(18.2%)
Other[b]	2	(18.2%)
Not applicable[c] (n = 9)		

[a] One patient underwent otoplasty, and one underwent cup-ear correction.
[b] Two patients underwent reconstruction without cartilage.
[c] Either no auricular deformation or no subject for the patient.

3.2. Choices regarding auricle

Table 4 and 5 show that the majority of patients did not opt for a surgical solution at the time of follow-up in the group of patients presenting with a congenital auricular abnormality. In the group of patients with an acquired etiology, almost half chose an auricular prosthesis. In Figs. 1 and 2, the therapeutic choices divided by age category are presented for the group of patients with a congenital etiology and acquired etiology, respectively. Logistic regression showed that etiology did not predict the outcome (B = −.111, p.907). In other words, no specific outcome was preferred when comparing congenital patients with acquired patients.

Table 2
Demographics of population concerning auricles.

Auricle	Total population (n = 134)		Distribution laterality (R/L/Bi; in %)	Age
Etiology auricle				
Congenital	120	(88.8%)	60.0/27.5/12.5	11.5 (min 0 – max 62; SD 12.14)
Of which microtia	102	(76.9%)	65.0/24.3/10.7	12.0 (min 0 – max 62; SD 12.82)
Acquired	12	(9.0%)	66.7/33.3/0.0	61.5 (min 16 – max 84; SD 22.40)
None	2	(2.2%)	–	1
Type of microtia				
Lobular	65	(63.7%)		
Conchal	13	(12.8%)		
Small conchal	9	(8.8%)		
Atypical	5	(4.9%)		
Anotia	4	(3.9%)		
Other[a]	4	(3.9%)		
Unknown	2	(2.0%)		

[a] In bilateral patients: 3x lobular and conchal, 1x lobular and anotia.

Table 3
Demographics of population concerning hearing.

Hearing	Total population (n = 134)		Distribution laterality (R/L/Bi; in %)	Age
Etiology ear canal				
Congenital	113	(84.3%)	61.9/27.4/10.6	11.0 (min 0 – max 62; SD 12.81)
Acquired	0	(0.0%)	–	–
None	21	(15.7%)	–	21.0 (min 0 – max 84; SD 29.29)
Type of hearing loss				
Conductive	107	(79.9%)		
Sensorineural	1	(0.7%)		
Mixed	4	(3.0%)		
None	19	(14.2%)		
Unknown[a]	3	(2.2%)		

[a] No audiometry was available.

Table 5
Choices regarding reconstruction categorized by severity.

Microtia grade (n = 106[a])		Choice regarding auricle		
Lobular	69	Autologous	14	(20.3)
		Alloplastic	3	(4.3)
		Prosthetic	4	(5.8)
		Wait/do nothing	48	(69.6)
Conchal	16	Autologous	0	(0)
		Alloplastic	0	(0)
		Prosthetic	0	(0)
		Wait/do nothing	16	(100)
Small conchal	9	Autologous	0	(0)
		Alloplastic	0	(0)
		Prosthetic	0	(0)
		Wait/do nothing	9	(100)
Atypical	5	Autologous	1	(20)
		Alloplastic	0	(0)
		Prosthetic	0	(0)
		Wait/do nothing	4	(80)
Anotia	5	Autologous	0	(0)
		Alloplastic	2	(40)
		Prosthetic	1	(20)
		Wait/do nothing	2	(40)
Unknown	2	Autologous	0	(0)
		Alloplastic	0	(0)
		Prosthetic	2	(100)
		Wait/do nothing	0	(0)

[a] Total patients with microtia were 102 patients, of which 4 were bilateral.

3.3. Choices regarding hearing

Table 6 shows the therapeutic choices regarding hearing. In Fig. 3, the therapeutic choices divided by age category are presented regarding hearing. Out of the 134 patients who visited the consultation, 19 (14.2%) patients did not have any type of hearing loss. Of the 102 patients who visited the consultation with questions regarding hearing rehabilitation, 80 patients completed a trial with a bone conduction device (BCD), while 22 patients declined this trial or started this trial during follow-up. After completing this trial, 39 patients (48.8%) opted for a percutaneous implant, and 34 (42.5%) decided against hearing rehabilitation either with a percutaneous implant or a BCD on a softband. The remaining seven patients chose a temporary solution as the softband due to age <4 years (n = 5, 6.2%) or a passive transcutaneous option (n = 2, 2.5%), such as nonsurgically implanted devices. At the time of follow-up, three pediatric patients were using a BCD on a softband. Two of these patients were within the trial period of the BCD at the time of follow-up. The other patient was a 6-year-old with *unilateral* hearing loss who declined a percutaneous implant at this timepoint and chose to continue hearing rehabilitation by a BCD on a softband.

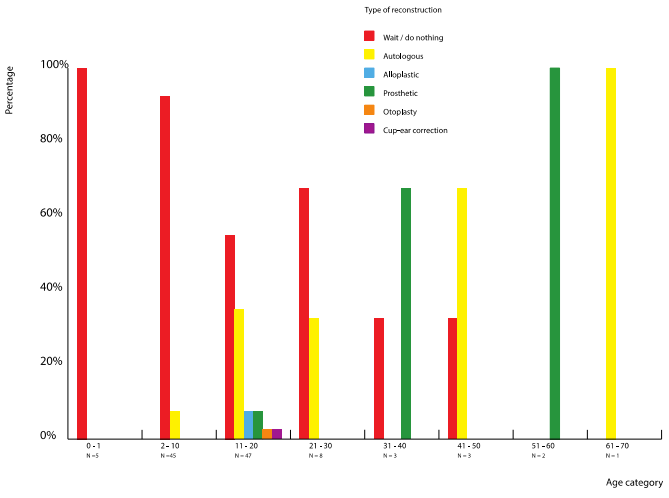

Fig. 1. Choices regarding auricles categorized per age category for the congenital group.

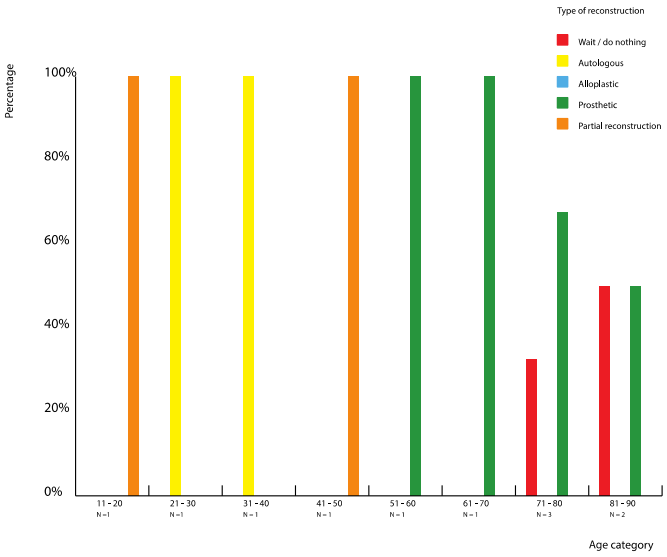

Fig. 2. Choices regarding auricle categorized per age for the acquired group.

3.4. Patient satisfaction

From the population that participated in this study, 112 patients (83.6%) rated their satisfaction with their choice concerning the auricle. Their overall satisfaction score was 8.7 (min 0 – max 10; SD 1.4). Satisfaction scores did not differ significantly between patients choosing either autologous, alloplastic, or prosthetic reconstruction or who decided to wait or decline reconstruction. Two patients (1.8%) were dissatisfied (score <6) with their choice concerning their auricle after consultation. One patient was dissatisfied that alloplastic auricular reconstruction was not possible in the Netherlands, and the other patient underwent partial reconstruction after trauma of the auricle and had higher expectations regarding the postoperative result. Eight patients who visited the consultation only had questions regarding their hearing, and 14 patients found it too difficult to express their satisfaction on an NRS score.

Table 6
Choices regarding hearing.

Congenital (n = 102)		
Percutaneous BCD	39	(38.2%)
Transcutaneous BCD	2	(2.0%)
BCD on softband	5	(4.9%)
Wait/do nothing	56	(54.9%)
Not applicable[a] (n = 32)		
Unilateral (n = 93; 91.2%)		
Percutaneous BCD	31	(33.3%)
Transcutaneous BCD	2	(2.2%)
BCD on softband[b]	4	(4.3%)
Wait/do nothing	56	(60.2%)
Bilateral (n = 9; 8.8%)		
Percutaneous bilateral BCD	8	(88.9%)
Bilateral BCD on softband[b]	1	(11.1%)

[a] Either no hearing loss, or no subject for the patient.
[b] Currently in trial period, and not eligible for percutaneous solution due to age.

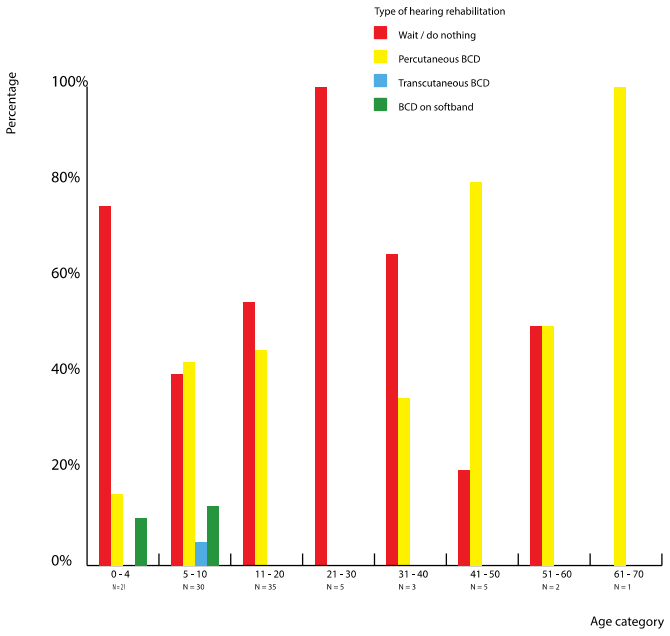

Fig. 3. Choices regarding hearing categorized per age category.

Concerning hearing, 93 patients (69.4%) rated satisfaction regarding their choice after the auricle consultation. Their overall satisfaction score was 8.3 (min 4 – max 10; SD 1.4). Satisfaction scores did not differ significantly regarding their choice concerning hearing solution. Seven patients (7.5%) with unilateral hearing loss were dissatisfied with their choice regarding hearing. These patients opted for a percutaneous bone implant after an extensive and positively evaluated trial period and reported nonuse afterward (average follow-up 6.0 years) due to either no long-term benefit in hearing or postoperative complications (implant loss or skin reaction). Twenty-nine patients did not visit the consultation with questions regarding hearing, and 12 patients found it too difficult to express their satisfaction on an NRS score.

4. Discussion

4.1. Main findings and clinical applicability

This study evaluated the choices regarding the auricle and/or hearing of patients who visited the auricle consultation within a time span of ten years.

Most of the patients who presented at the auricle consultation had a congenital etiology. Notably, the majority of these patients (72.8%) did not opt for surgical reconstruction at the time of follow-up. Some caregivers of the patients in this study motivated their choice by stating that they wanted to let their child decide when he or she reached adulthood. Consequently, a part of this group might still opt for auricular reconstruction in the future. In contrast, in the acquired group, most patients chose surgical reconstruction. The sample size of this acquired group is remarkably smaller. There was strong heterogeneity between the groups in terms of age; the median age was 61.5 years, compared to 11.5 years in the congenital group. Instinctively, it could be that patients who experience a sudden change in facial appearance have more difficulties coping with social responses and are therefore more predisposed to opt for surgical reconstruction. However, studies failed to demonstrate a significant difference in psychosocial functioning between congenital and acquired facial disfigurement (van den Elzen et al., 2012; Versnel et al., 2012). At first sight, it seems that patients with an acquired etiology are more inclined to opt for surgical reconstruction. However, with logistic regression analysis (which corrected for patient age), no statistically significant relationship between etiology and outcome was found in the current set of data. An important point to mention is that a substantial portion of patients who presented at the consultation were too young to opt for auricular reconstruction. Therefore, these patients might make a different choice when they reach an older age, which could have influenced our results at the time of follow-up. No analysis was performed if patients changed in outcome regarding auricle or hearing between the time of consultation and the time at follow-up. In addition, the choice these patients made at the time of consultation is likely to have been influenced by their caregivers. Another explanation could be that the stress an auricular deformity generates for the patient, which could drive them to opt for a surgical solution, is similar between congenital and acquired deformities and varies depending on patient age (which itself could be related to the social network of patients). Another explanation could be that no differences were found due to the small sample size of the acquired etiology group.

The proportions are closer together regarding hearing, as just more than half of patients (56.9%) chose to wait or decide against hearing rehabilitation. Most of the other patients chose hearing rehabilitation by means of a bone conduction hearing solution. Various options are currently available (Reinfeldt et al., 2015). Most of the patients who opted for hearing rehabilitation chose percutaneous BCD. It is recommended to rehabilitate hearing in children with congenital atresia of the ear canal due to an increased risk of delayed speech-language development, especially in bilateral cases (Lieu, 2004, 2015). Unilateral hearing loss affects children differently, as some children can manage adequately with one functioning ear. Therefore, shared decision-making, information at the right time, and monitoring of the patient are essential. Moreover, the possible benefits of binaural hearing can be more subtle than the patients' subjective benefit, as higher repeating rates and increased fatigue have been reported in these patients (Lieu, 2015; Bakkum et al., 2022). The trial with a BCD should be conducted in a multidisciplinary team with knowledge and expertise regarding other aspects of hearing impairment (such as school, psychological, and speech language development), in addition to the hearing itself. In case patients decide against hearing rehabilitation after an extensive trial (in unilateral hearing loss), it remains important to explain that they are always welcome to revise their decision. Additionally, it is important to inform them about preferential seating in a classroom and other important aspects of hearing with one ear, such as safety in traffic.

Overall, patients were very satisfied with their choices regarding the auricle and hearing after visiting the auricle consultation. Interestingly, there were no significant differences in patient satisfaction between different outcomes, which could indicate that providing complete patient-centered counseling and shared decision-making regarding the patients' choice is of greater importance to patient satisfaction than the choice itself. This high patient satisfaction could be achieved through its multidisciplinary aspect, providing complete information in one visit by taking extensive time in counseling the patient and its caregivers to its choice with neutrality. In the total population of 134 patients, nine patients were dissatisfied with their choice, of which two concerned auricle and seven concerned hearing. The lesson learned, again, is that thorough meticulous patient counseling and careful patient selection are essential. This patient-centered and multidisciplinary approach is advocated when counseling the patient through the process of deciding for either of the treatment options.

4.2. Comparison with other studies

To the best of our knowledge, no research has been previously published about the implementation of a clinical pathway regarding the absent auricle or microtia. However, there is one paper that analyzed the current practice in Egypt and compared this to the UK standards, resulting in five key recommendations in reforming microtia service: 1. Establishing nationally designated centers to concentrate expertise, 2. Assigning fewer high-volume surgeons to improve outcome, 3. Providing treatment by experienced multidisciplinary teams; 4. Using validated tools of PROMs, 5. Collecting and keeping standardized records of all aspects of microtia treatment (Henderson et al., 2015; Mazeed et al., 2019). Further research and evaluation are needed to evaluate and constantly improve the implementation of the clinical pathway and to objectify the outcome for patients with microtia specifically and patients with a missing auricle in general (e.g., in terms of PROMs and quality of life).

Previous research on auricular reconstruction was generally focused on surgical options and thereby does not address the option to wait or decline auricular reconstruction. To the best of our knowledge, only one study in the literature has addressed this aspect (Steffen et al., 2010). The authors suggest that patients who decline auricular reconstruction have higher self-esteem and coping skills than patients who opt for surgery preoperatively. Moreover, patients generally do not regret their decision against auricular reconstruction later on. The latter is underlined by the current study. This, again, emphasizes the necessity to thoroughly counsel caregivers and patients through the process. Future studies should consider including the option to decide to “accept and wait” or even against auricular reconstruction as a valid alternative to the mentioned treatment options.

4.3. Strengths and limitations

The current study provides a holistic view of the therapeutic options in patients with a missing auricle from a patients’ perspective with long-term follow-up. A limitation of the study is the retrospective character of the subsequent medical chart review, although the frequency of missing information was reduced to a minimum due to the follow-up by phone. All patients who presented at the auricle consultation were included in this study. As not all treatment options for patients with a missing auricle were available in the studied center (e.g., alloplastic reconstruction; although information is provided and if wished for a referral), it is possible that the population that presented at the auricle consultation slightly deviates from the total population of patients with a missing auricle. Another limitation of this study is that the questionnaire used has not been validated, which affects the comparability of the data. Additionally, response bias could have been introduced in this study, as patients who are either satisfied or dissatisfied might be more inclined to participate. Furthermore, patient satisfaction scores could have been influenced by confirmation bias. Patients are inclined to justify their own choices and might also want to satisfy their caregiver or physician, which could result in higher patient satisfaction scores.

5. Conclusion

Patients and/or their caregivers were generally satisfied with their choice after auricle consultation, regardless of whether this implies reconstruction or no reconstruction, hearing rehabilitation or no hearing rehabilitation. This emphasizes the importance of shared decision-making and thorough counseling of the patient by a multidisciplinary team about the ear, hearing, and appearance.

Declaration of competing interest

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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