

# Congenital aural atresia surgery: Transmastoid approach, complications and outcomes

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**Abstract** Repair of complete congenital aural atresia (CAA) could be a challenging procedure due to complications reported with CAA surgery such as facial nerve palsy, canal stenosis, graft lateralization, sensorineural hearing loss or the difficulty involved in the surgical technique. From 2006 to 2009, we used a one stage-modified transmastoid approach for surgical repair of 33 ears with complete CAA via a non-randomized controlled clinical trial. Some modifications in the technique of mastoidectomy, ossiculoplasty, fascia and skin grafting and meatoplasty have been described. Patients were followed up for 12 months to assess audiometric results and post-operative complications. Changes in air-bone gap and need for revision surgery or hearing aids were assessed at follow-up. There were no cases of facial weakness, dead ear or bony canal stenosis. Hearing success in 2 months follow-up was achieved in 72.7% of all patients. Success rate increased to 92.3% in patients with Jahrsdoefer's scores of 8 and above. Overall success rate decreased to 63.6% at 12 months follow-up. There were no significant difference in Jahrsdoefer score of patients with successful first surgical attempt and those who needed revision surgery ( $P$  value  $>0.056$ ). Also patients of lower age (less than 5-years-old) did not have more need for revision surgery when compared with older patients ( $P$  value  $>0.36$ ).

However, being a syndromic patient did increase the need for revision surgery ( $P$  value  $<0.04$ ). Age was not a predictor of meatal/canal stenosis and patients with lower Jahrsdoefer scores could also achieve good results.

**Keywords** Congenital aural atresia · Canal atresia · Aural atresia · Transmastoid approach · Complications · Surgical repair · Audiometry

## Introduction

The incidence of congenital aural atresia (CAA) is about 1 in 10,000 live birth. Unilateral CAA occurs three times more common than bilateral [1]. CAA is characterized by hypoplasia or aplasia of the external auditory canal (EAC) associated with microtia and middle ear abnormalities [2]. CAA repair is a challenging procedure and a thorough knowledge of the surgical anatomy of the facial nerve, oval window, and inner ear as well as their congenital variations is needed for this surgery. The goals in CAA surgery are creation of a patent EAC and meatus in addition to providing hearing [3]. Patients with normal inner ear, normal ossicles, well-developed middle ear space and normal fallopian canal are good candidates for CAA surgery [4]. However, we included patients with small middle ear space and deformed or absent ossicles and also cases with abnormal course of the facial nerve in our study. Recurrent conductive hearing loss, chronic infection and bony canal stenosis have been reported as common post-operative complications [5]. Serious intra-operative complications are facial nerve paralysis and sensorineural hearing loss [1, 6–8]. First explanation of CAA surgery was done by Kisselbach in 1882 [9]. Transmastoid and anterior mastoid approaches are the two widely used techniques. De la Cruz

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and Jahrsdoerfer [1, 7] have modified surgical approach in previous 30 years to obtain better outcomes. We used transmastoid approach with some modifications in surgical steps. This study describes our surgical technique of complete bony atresia repair, its complications and outcomes.

## Material and method

Between 2006 and 2009, 33 CAA repairs were performed on 29 consecutive patients who became the subjects of the study. For the purpose of this non-randomized controlled study, each of the 33 procedures was considered as an individual case and every participants was considered as his/her own control. All the cases with incomplete or soft tissue atresia were excluded. After complete and thorough physical examinations, pure tone audiometry (PTA) and temporal bone high-resolution CT scan were obtained for all participants. Jahrsdoerfer score was calculated for each subject. Follow-up PTA was obtained 2 and 12 months post-operatively. Mean air bone gap (ABG) of 500, 1000, 2000 and 4000 Hz were calculated pre- and post-operatively. All patients were followed 12 months for evaluation of complications such as bony canal stenosis, meatal stenosis, facial nerve weakness, sensorineural hearing loss, granulation tissue formation, graft lateralization, wound dehiscence, ossicular refixation, extrusion of prosthesis, otorrhea, tympanic membrane perforation and cholesteatoma. Hearing success was defined as ABG below 30 dB and being able to communicate without the use of hearing aids at 2 and 12 months post-operatively. Study was approved by the ethical committee of University ENT and HNS research center and all participants completed informed consent.

## Surgical technique

All procedures were performed by the same surgeon (senior author). Post-auricular incision was made slightly posterior than usual to allow room for wide drilling of the mastoid and also to avoid injury to the facial nerve in the mastoid area in younger patients. A large piece of temporalis fascia was harvested and left to dry. The skin and periosteum were incised and raised as one flap. The TMJ was recognized anteriorly.

We started drilling posterosuperior to the TMJ leaving a thin layer of bone intact over the TMJ. All mastoid air cells (when present) were drilled. The cortical bone was saucerized superiorly and posteriorly. When mastoid air cells were not present, we drilled spongiotic bone posteriorly toward the sigmoid sinus and superiorly up to the level of middle fossa plate. In our opinion, wide drilling of the

mastoid and creation of an EAC with a diameter of at least 25 mm will lower chance of bony stenosis. Similarly, saucerizing the mastoid could reduce the length of the newly created EAC and, therefore, further reduce possibility of bony stenosis.

Meatoplasty was done prior to placement of the fascial graft to prevent displacement of the fascial graft while performing the meatoplasty. A posterior based incision was made in the skin over the newly created EAC (Fig. 1). The cartilage and soft tissue was removed under this skin flap and the skin was reflected posteriorly. At the end of the procedure the subcutaneous tissue under created skin flap was sutured to the subcutaneous tissue at the posterior edge of the incision.

The atretic plate was removed completely. Drilling was continued inferiorly toward the hypotympanum and anteriorly until sufficient room was created down to the level of eustachian tube for deep anterior placement of the fascial graft. The facial nerve was positively identified at this point and fallopian canal was skeletonized in order to make maximal room for placement of TORP/PORP and fascia and skin graft. The drilling was also continued posterior to the lateral and posterior semicircular canals and deep to the level of semicircular canals in the sino-dural angle so that we could “tuck” the fascial graft deeply posterior and thus avoid lateralization of the graft in the post-operative period. At this point mobility of the ossicular chain was checked. We left the ossicular chain intact only if the ossicles were completely mobile. In most cases where the ossicles were missing or defective and also when mobility of the ossicular chain was in question, reconstruction with PORP/TORP was preferred. The incudostapedial joint was separated as soon as feasible to prevent drill contact with the ossicles and thus avoid sensorineural hearing loss.

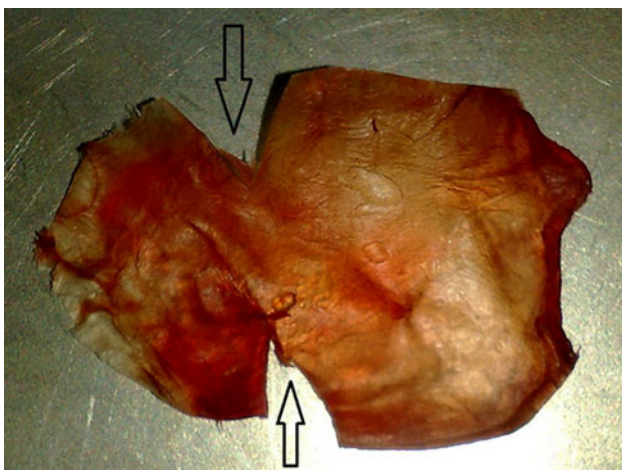


**Fig. 1** Posterior based flap incision according to dashed line

Two triangular wedges were made in the fascial graft as seen in Fig. 2. This allows the graft to be placed easily and fit in the newly created EAC and mastoid and helps avoid folding of the fascial graft. A thin split thickness skin graft (STSG) was taken from the thigh area. We made several parallel incisions in this STSG (Fig. 3). This could allow the fluid to escape and thus prevent accumulation of fluid and blood underneath the skin graft. The fascial graft was placed in a way that the center of the graft was located over the ossicles or TORP/PORP. Care was made so that the fascia over PORP/TORP was at least 5 mm higher than the rest of the fascial graft. The “meshed” STSG was then placed over the fascia and firmly kept in place using gel-foam. We tried to overlap the STSG slightly (1–2 mm) over the skin edges of newly created meatus. We did not suture the STSG to the skin of the meatus because in our opinion this could cause lateral displacement of the skin graft. Facial nerve monitoring was done during the entire procedure. We used antibiotic soaked gauze over the gel-foam in the created EAC and meatus and left it in place for 3 weeks.

## Results

Thirty-three ears with CAA underwent surgical repair (22 right and 11 left, 29 patients total, because four patients had bilateral repair). Patients were between 3- and 23-years-old (mean  $11.06 \pm 6.58$ ); eight patients (24.2%) were younger than 5, eleven patients (33.3%) between 5 and 10, and 14 subjects (42.4%) were older than 10 years. Eight patients had syndromic anomalies such as Goldenhar’s and Treacher Collins syndromes. Mean Jahrsdoerfer’s score was  $6.6 \pm 1.7$ , (minimum 2, maximum 9). Also Mean Jahrsdoerfer’s score for syndromic patients was 5.2 and for non-syndromic patients was 7.0. Scores were more than seven



**Fig. 2** Temporalis fascial graft, two wedges shown by arrows



**Fig. 3** Meshed split thickness skin graft

in 13 patients (39.4%), six or seven in 12 subjects (36.4%) and eight patients (24.2%) had Jahrsdoerfer score of 5 or less.

## Surgical findings

The malleus was absent in one case (3%), deformed in 22 patients (66.7%), fixed in nine (27.3%), and appeared normal only in one case (3%). The incus was absent in one case (3%), deformed in 25 (75.8%), fixed in five (15.2%), and appeared normal in two patients (6.1%). Stapes was normal in 24 patients (72.7%), deformed in eight (24.2%), and fixed footplate was observed in one patient. Ossicular chain reconstruction was performed in 22 cases (66.6%) (19 with PORP and three with TORP). In one case stapedectomy was necessary for placing the TORP (Table 1).

The facial nerve was in its normal position in 26 cases (78.8%); it was partially overlapping the footplate in three (9.1%), and in four cases (12.1%) the course of fallopian canal was entirely abnormal. We continued with surgery even in cases where the facial nerve was partially overlapping the footplate. The oval window was obliterated in two ears (6.1%), and we could not find an apparent round window in two cases (6.1%). In one patient, cholesteatoma was found intra operatively in both ears.

## Hearing results and complications

Air-bone gap improved from a pre-operative mean of  $53.5 \pm 7.6$  up to  $28.78 \pm 10.23$  dB 2 months after surgery. When all patients were considered, long-term post-operative ABG after 12 months was  $30.15 \pm 8.8$  dB. Nine of 33 cases underwent revision surgery during the first

**Table 1** Audiometric and surgical results, pre- and post-operation

Case no. <sup>a</sup>	Age (years)	Side <sup>e</sup>	Syndromic association	Jahrsd-oefer score	Pre-op ABG (2 months)	Post-op ABG (12 m)	OCR	Ossicular deformity	Course of facial nerve	OW	RW	Complications	Revision
1	6	L	No	8	60	30	PORP	M: def I: def	NL	NL	NL	Otorrhea	No
2	6	L	No	8	60	30	No	M: fix I: def	NL	NL	NL	No	No
3	4	R	No	8	60	25	No	M: fix I: def	NL	NL	NL	No	No
4	8	B(R)	No	8	60	20	PORP	M: def I: def	NL	NL	NL	No	No
5	5	L	No	8	40	30	PORP	M: def I: def	NL	NL	NL	Otorrhea	No
6	8	R	No	6	60	25 <sup>b</sup>	PORP	M: def I: def	NL	NL	NL	Graft lateralization, granulation tissue	Yes
7	13	B(L)	No	7	50	25 <sup>b</sup>	No	M: fix I: def	NL	NL	NL	Ossicular refixation, granulation tissue	Yes
8	21	R	No	8	45	35	PORP	M: fix I: fix	NL	NL	NL	Granulation tissue, otorrhea	No
9	4	B(L)	Yes	2	60	30 <sup>b</sup>	No	M: def I: def	NL	OB	OB <sup>c</sup>	TM perforation, granulation tissue	Yes
10	4	B(R)	Yes	4	60	35	No	S: def M: def I: def	NL	NL	NL	No	No
11	21	R	No	7	60	20	PORP	S: def M: def I: def	NL	NL	NL	No	No
12	8	R	Yes	8	45	25 <sup>b</sup>	PORP	M: def I: def	NL	NL	NL	Meatal stenosis, granulation tissue, otorrhea	Yes
13	4	R	No	8	60	25 <sup>b</sup>	No	M: fix I: fix	NL	NL	NL	Meatal stenosis, ossicular refixation, TM perforation	Yes
14	6	R	No	8	40	25	PORP	M: def I: def	NL	NL	NL	No	No
15	9	R	No	8	45	35	PORP	M: fix I: fix S: def	NL	NL	NL	Otorrhea	No

Table 1 continued

Case no. <sup>a</sup>	Age (years)	Side <sup>e</sup>	Syndromic association	Jahrsd- oerfer score	Pre-op ABG (2 months)	Post-op ABG (12 m)	OCR	Ossicular deformity	Course of facial nerve	OW	RW	Complications	Revision
16	3	B(R)	No	5	60	20	25	PORP	NL	NL	NL	No	No
17	13	B(R)	No	7	40	20	20	PORP	Abnormal	NL	NL	No	No
18	7	R	No	8	55	20	30	No	NL	NL	NL	Granulation tissue	No
19	17	B(R)	No	3	55	30	35	TORP <sup>d</sup>	NL	OB	OB <sup>c</sup>	No	No
20	21	R	No	5	60	60	40 <sup>b</sup>	PORP	Overlap	NL	NL	Meatal stenosis, granulation tissue	Yes
21	18	B(L)	Yes	6	60	30	35	PORP	NL	NL	NL	No	No
22	18	B(R)	Yes	6	45	25	25 <sup>b</sup>	PORP	NL	NL	NL	Meatal stenosis, granulation tissue, otorrhea	Yes
23	13	R	No	9	55	35	35	No	NL	NL	NL	No	No
24	7	R	No	8	45	20	30	PORP	NL	NL	NL	Meatal stenosis, granulation tissue	Yes
25	23	B(R)	No	6	50	30	15 <sup>b</sup>	No	NL	NL	NL	Ossicular refixation, SNHL	Yes
26	23	B(L)	No	8	45	15	20	PORP	NL	NL	NL	No	No
27	4	L	No	6	45	20	20	TORP + staped- ectomy	NL	NL	NL	Otorrhea	No
28	5	L	No	6	60	30	35	PORP	NL	NL	NL	No	No
29	14	B(R)	Yes	5	60	45	60	No	Abnormal	NL	NL	Meatal stenosis granulation tissue	Yes
30	14	B(L)	Yes	3	60	45	40 <sup>b</sup>	No	Abnormal	NL	NL	Meatal stenosis, granulation tissue	Yes



Table 1 continued

Case no. <sup>a</sup>	Age (years)	Side <sup>b</sup>	Syndromic association	Jahrsdoerfer score	Pre-op ABG	Post-op ABG (2 months)	Post-op ABG (12 m)	OCR	Ossicular deformity	Course of facial nerve	OW	RW	Complications	Revision
31	10	B(L)	Yes	4	60	35	45	TORP	M: def I: def S: def	Abnormal	NL	NL	Meatal stenosis, granulation tissue, otorrhea	Yes
32	7	R	No	4	45	20	40	PORP	M: def I: def S: def	Overlap	NL	NL	Meatal stenosis	Yes
33	21	B(R)	No	7	60	30	30	PORP	M: def I: def	Overlap	NL	NL	Meatal stenosis	Yes

RW Round window, OW Oval window, L Left, R Right, B Bilateral, NL normal, M Malleus, I Incus, S Stapes, def deformed, fix fixed, abs absent, OB obliterated, OCR ossicular chain reconstruction

<sup>a</sup> Cases number 9 and 10, 21 and 22, 25 and 26, 29 and 30 are two ears in the same patients

<sup>b</sup> ABG after second stage surgery

<sup>c</sup> No apparent round window was seen intraoperatively

<sup>d</sup> At first obliterated oval window was drilled and then TORP was placed over a piece of perichondrial graft

<sup>e</sup> Letter in () shows the side that surgery was performed

post-operative year, so the PTA after their second surgery was considered in their 12 months ABG results. Mean pre-operative ABG was similar between each Jahrsdoerfer score group ( $P$  value 0.198), whereas 12 months post-operative ABG was better in patients with score of 6 and above ( $P$  value  $<0.001$ ) (Table 2). Hearing success in 2 months follow-up was achieved in 72.7% of all patients and in 92.3% of patients with Jahrsdoerfer's scores of 8 and above (Table 1). Overall success rate was 63.6% after 12-month follow-up. At 12-month post-operation three patients needed hearing aids to communicate. Although pre-operative ABG was similar between syndromic ( $56.2 \pm 6.9$  dB) and non-syndromic ( $52.6 \pm 7.7$  dB) patients ( $P$  value 0.24), 12-month post-operative ABG was 36.9 dB for syndromic patients and 28.0 dB for non-syndromic subjects ( $P$  value 0.01).

Our goal was one stage surgery, but 14 cases (nine in the first year post-operative and five cases later on) needed revision surgery due to meatal stenosis, graft lateralization, ossicular re-fixation or TM perforation. Ten patients (30.3%) developed soft tissue meatal stenosis. There were no cases of bony canal stenosis. Lateralization of the graft was seen in one patient, and ossicular chain re-fixation in three patients (9.1%). TM perforation was seen in two patients (6.1%). Eight cases (24.2%) had post-operative otorrhea which was managed successfully using steroid and antibiotic drops. Granulation tissue was seen in 11 cases (33.3%) and was managed effectively by using topical steroid drops and/or chemical cautery in the office. Sensorineural hearing loss was detected in one patient (3%). It averaged 30 dB in four consecutive frequencies and was more noticeable in higher frequencies. There were no cases of facial nerve palsy, acquired cholesteatoma, extrusion of prosthesis or wound dehiscence after surgery. Among 14 patients with post-operative complications, six patients were syndromic and eight were non-syndromic. The soft tissue meatal stenosis was the main reason for revision surgery in both syndromic and non-syndromic patients (Table 1). We compared mean age of patients who developed soft tissue meatal stenosis ( $12.4 \pm 6.1$ ) with patients who did not have post-operative meatal stenosis ( $9.9 \pm 6.3$ ). There was no statistically significant difference between these two groups ( $P$  value  $>0.31$ ).

The patients' age played no significant role in their need for revision surgery ( $P$  value  $>0.36$ ). Mean age in patients who needed revision surgery was  $12.3 \pm 6.4$ , and in patients with successful primary surgery it was  $10.2 \pm 6.7$  years. Also, we could not find a significant difference of Jahrsdoerfer score in the group of patients who needed revision surgery when compared to those who did not need a revision. Patients with revision surgery had mean Jahrsdoerfer score of  $5.9 \pm 1.7$  and patients with successful primary surgery had mean Jahrsdoerfer score of  $7.1 \pm 1.5$

**Table 2** Short-term and long-term post-operation mean air bone gap of 500, 1000, 2000 and 4000 Hz frequency

	Mean ABG pre-operative (dB)	Mean ABG after 2 months (dB)	Mean ABG after 12 months (dB)
Syndromic association			
Yes	56.2 ± 6.9	35.0 ± 8.1	36.9 ± 11.6
No	52.6 ± 7.7	26.8 ± 10.1	28.0 ± 6.7
Jahrsdoerfer score			
5 or less	57.5 ± 5.3	38.7 ± 11.8	40.6 ± 9
6 or 7	52.9 ± 7.8	28.3 ± 8.6	25.8 ± 6.6
8 or more	51.5 ± 8.2	23.0 ± 5.2	27.7 ± 4.8

( $P$  value >0.056). Syndromic association could be considered a risk factor for needing revision surgery (revealed by Pearson  $\chi^2$  test;  $P$  value <0.04).

## Discussion

In the field of CAA surgery, proper timing and selection of surgical approach is controversial. There is some controversy in choosing BAHA surgery versus reconstructive surgery in unilateral CAA [10]. An excellent hearing result after atresiaplasty is the most favorable possible outcome; in this situation, cosmesis is acceptable to the patient because there is no external device, spontaneous learning is possible and hearing improvement is continuous [11]. BAHA is not the best cosmetic choice, although hearing result is excellent. In developing countries such as ours, surgical reconstruction is less expensive than BAHA. The philosophy of our approach is influenced by the economic condition in our country, the total cost of an atresia repair surgery in a university hospital is much less than the cost of BAHA implant alone.

As Lambert mentioned, bilateral hearing is very important in communication [4], therefore, we prefer surgical reconstruction of unilateral cases in pre-school age. The patients and/or their parents report better understanding of speech and improved verbal communication skills especially in noisy environments. This is similar to BAHA surgery, although in both surgeries bilateral hearing result could be asymmetric [12].

Our surgical approach is a one stage-modified transmastoid surgery. We modified the technique of fascia and skin grafting, ossiculoplasty and meatoplasty. We changed the design of fascial graft by creating two wedges in the temporalis fascia (Fig. 2) before putting it over the ossicles or prosthesis. Also in our opinion “Meshing” the split thickness skin graft by making parallel incisions in the STSG (Fig. 3) could lead to better drainage and prevent fluid accumulation and hematoma formation underneath

the skin graft. This could prevent thickening of the TM and in turn result in better post-operative hearing.

In previous studies, successful hearing results were defined as ABG reduction to 30 dB or lower [8]. Success rate varied from 27 to 63.1% for short-term follow-up and 47 to 50% for long-term follow-up [5, 13]. Our short-term (2 months) success rate for all patients was 72.7%. Overall success rate was reduced to 63.6% in 12 months follow-up. However, in their long-term follow-up only three patients needed use of hearing aid for their communication. They attended school without any problems. Facial nerve palsy, a catastrophic complication of CAA surgery has been reported to be about 1.5% in expert hands [14]. Even though we have included syndromic patients and patients with documented abnormal course of facial nerve in this study, we did not encounter any cases of facial nerve palsy in our series. In our opinion, positive intraoperative identification of the facial nerve in all cases could help prevent injury to the facial nerve.

Meatal stenosis is a common post-operative problem which has been reported between 3.8 and 29.3% [5, 8, 13]. Although we did not have any cases of bony canal stenosis post-operatively, meatal stenosis was seen in 30.3% of our cases. Meatal stenosis was mostly due to anterior rotation of the cartilaginous remnant of the auricle. We learned that removing more of the cartilaginous remnant could help reduce this problem.

Chang [14] found an inverse correlation between meatal stenosis and age of the patients, and in his study meatal stenosis was higher in younger patients, but we could not find a significant correlation between patient’s age and development of meatal stenosis ( $P$  value >0.31).

Graft lateralization with gradual worsening in hearing level was reported in 3.4% of patients in previous studies [5] and it was seen in one of our reconstructed patients. In our opinion this problem can be avoided by “tucking” the anterior part of the graft deep in the eustachian tube area and “tucking” the posterior end of the graft deep to the posterior semicircular canal in the sinodural angle. We

measured the length of the prosthesis and left its top 3–5 mm higher than the remainder of the fascial graft to avoid separation of the graft from the top of the prosthesis.

Ossicular re-fixation was seen in three patients in our study. Since the ossicular movement is not normal in most cases of complete atresia, we believe it is preferable to remove the incus and malleus and use PORP/TORP when feasible (mean post operative ABG was 27.3 dB when prosthesis was used and 31.8 dB for cases without prosthesis). Opening the middle ear space down to the level of the eustachian tube and hypotympanum and drilling the mastoid air cells widely will allow more room for the TORP/PORP and the fascia and skin graft. In our opinion this could prevent adhesion of the prosthesis to the surrounding tissues and could possibly result in better post-operative hearing.

Sensorineural hearing loss in high frequencies was reported in about 7.5% of patients in other studies [5]. In our series, one patient (3%) developed sensorineural hearing loss after surgery. Separating the incudostapedial joint as soon as feasible even before completing the drilling, could prevent sensorineural hearing loss resulting from contact of the drill with the ossicles.

We included patients as young as 3 years old in this study. We could not find a significant correlation between patient's age and the need for revision surgery. The ideal age for atresiaplasty has been reported at 6 years or older in the literature [15]. However, in the current study post-operative results seem to be independent of age and satisfying results were achieved in patients as young as 3 years old. BAHA implantation has been preferred to atresiaplasty because patients in lower age could be implanted safely [12, 14]. In our opinion, working with patients of lower socioeconomic status or where BAHA is not available, as an alternative atresia repair could be done safely for patients as young as 3 years old.

We did not find a significant difference between Jahrsdoerfer score and the need for revision surgery. However, there was a remarkable difference between syndromic and non-syndromic patients in their need for revision surgery, i.e., the syndromic patients needed revision surgery more often than non-syndromic patients.

The suboptimal results in syndromic patients could be attributed to their restricted and severely abnormal temporal bone anatomy. However, it is of note that even in syndromic patients and patients with low hearing gain, the EAC remained patent after revision surgery. The parents

reported that having patent EAC helped these children to better cope psychologically with their ear problem. Having a patent EAC also enabled these children to use conventional hearing aids instead of using bone conduction hearing aids when needed [10].

In conclusion, it seems that this procedure could be done safely with relative success in patients as young as 3 years old and patients of low Jahrsdoerfer score when BAHA is not available. There is a need for further studies with larger number of patients and longer periods of follow-up to better evaluate the surgical results.

**Conflict of interest** The authors declare that they have no conflict of interest.

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