

# Transcanal endoscopic treatment for congenital middle ear cholesteatoma in children

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### **Abstract**

# **Background**

To investigate the feasibility and efficacy of transcanal endoscopic treatment for congenital middle ear cholesteatoma in children.

# **Methods**

Eleven children diagnosed with congenital middle ear cholesteatoma were collected at Huazhong University of Science and Technology Union Shenzhen Hospital from January 2016 to December 2020. The retrospective study of their operation process, comparison of pre- and post-operative hearing result, surgical complications through the surgical video.

# **Results**

Eleven children received total ear endoscopic surgery under general anesthesia. One of them received planned second operation to reconstruct the ossicular chain. At six months after operation, 11 children underwent re-examination. The mean bone conduction hearing threshold had no significant change (P>0.05), the mean air conduction hearing threshold was significantly decreased (P<0.05), and the airbone conduction difference was significantly reduced (P<0.05). In 11 children, the airbone conduction difference were all reduced to less than 20 dB, and 7 cases were reduced to less than 10 dB. All the children were followed up so far without sensorineural deafness, facial paralysis and other serious complications, as well as no recurrence.

# Conclusion

Otoendoscope can provide a wide-angle field of vision and advantages in small surgical trauma, quick healing, avoiding repeated dressing changes and high acceptance of secondary surgery. Intraoperative application of 30° and 45° otoendoscope can effectively reduce residuals. Otoendoscope is widely used as a surgical method in the treatment of congenital middle ear cholesteatoma in children.

# **Background**

In 1986, Levenson, *et al.* defined congenital cholesteatoma (CC) as a pearly white mass on the inner side of the normal tympanic membrane with no history of otorrhea, tympanic membrane perforation, or ear surgery. Currently, it is widely recognized and followed by Levenson's revised diagnosis standard [1]. Congenital cholesteatoma is rare in clinic, accounting for 1%-5% of middle ear cholesteatoma [2]. However the actual incidence of CC is more than that because some clinically suspected cases of CC were excluded for secondary otorrhea and perforation of the tympanic membrane. Due to the insidious

symptoms of this disease, it brings great difficulty to early diagnosis. It is also the cause of clinical missed diagnosis and delayed treatment in clinic. Children with congenital middle ear cholesteatoma (CMEC) often delay the optimal treatment time since they cannot describe hearing loss timely. CMEC has a higher incidence and has a greater impact on normal life. It can affect the hearing of children and it can cause intracranial and extracranial complications, which can lead to serious consequences. In this paper, we retrospectively analyzed case data, surgical videos and treatment results of 11 children diagnosed with congenital middle ear cholesteatoma at Huazhong University of Science and Technology Union Shenzhen Hospital from January 2016 to December 2020. We discussed the advantages of otoendoscope in the treatment of children with CMEC though introducing the surgical results and procedures of representative patients.

# **Methods**

#### Case data collection

Thirteen children diagnosed with congenital middle ear cholesteatoma were collected at Huazhong University of Science and Technology Union Shenzhen Hospital from January 2016 to December 2020. Eleven of them received total ear endoscopic surgery. The following information was recorded: age of patients, ear side, surgical procedure of representative cases, pre- and post-operative hearing comparison, etc.

#### **Examination method**

All children patient underwent detailed otolaryngology medical history inquiry, specialist examination and otoendoscope examination. High-resolution temporal bone CT scan was performed before surgery. Pure tone audiometry was performed in children over 6 years old, while auditory brainstem response for airbone conductance was performed in children under 6 years old or with poor coordination. All surgeries were performed by the same group of experienced ear surgeons.

Pure tone audiometry was carried out in a soundproof room according to the national standard (GB/T16403-1996) by GSI audiometer. Tympanometry was measured by GSI Tympstar middle ear analyzer. Auditory brainstem response was measured using international audiometry. Audiological tests were performed pre- and post-operatively. The pure-tone threshold Average (PTA) as well as the pre- and post-operative air-bone gap (ABG) were calculated at 500Hz, 1000Hz, 2000Hz and 4000Hz. 3mm 0° and 30° otoendoscope from STORZ were used in outpatient and intraoperative examination.

#### Diagnosis and grading standards

Diagnostic criteria: (i) white mass behind intact tympanic membrane, (ii) no previous history of otorrhea and tympanic membrane perforation, (iii) no previous ear surgery history, (iv) exclude congenital external or middle ear malformation, (v) do not rule out the history of acute otitis media, (vi) exclude mixed or sensorineural hearing loss.

According to Potsic's cholesteatoma standard [3], Stage I: lesions only exist in a single quadrant, Stage II: lesions involve multiple quadrants but do not invade the ossicular chain, Stage III: lesions involve multiple quadrants and invade ossicular chain, Stage IV: lesions develop to the mastoid process.

#### **Statistics**

SPSS 26.0 statistical software was used for statistical analysis. The data of each group was represented by X±s, and the comparison between the two groups was performed by independent sample t test. P<0.05 was considered as statistically significant difference.

The study compiled with the principles of the Declaration of Helsinki on Biomedical Research Involving Human Subjects and was approved by the Ethical Review Board of Huazhong University of Science and Technology Union Shenzhen Hospital (Nanshan Hospital). Written informed consent was obtained from each child's parent or supervisor.

## **Results**

# Preoperative examination results

Among the 11 children patients, the age was 3-14 years old, with an average age of 7.91±3.11 years old. There were 8 cases of left ear and 3 cases of right ear. 8 cases (72.7%) were hospitalized for conductive hearing loss with different course of disease while 3 cases (27.3%) did not pass the hearing screening in physical examination. All the 11 children patients had conductive hearing loss and the average difference of air-bone gap was 29.09±8.46 dB HL. High-resolution temporal bone CT scan and otoendoscope examination were performed before surgery. Three cases (27.3%) were Potsic stage I, four cases (36.4%) were stage II and four cases (36.4%) were stage III. Five cases in early Potsic stage were preschoolers (< 7 years old) with intact ossicular chain and good prognosis. Four children in stage Potsic III were all over 8 years old with hearing loss as chief complaint. Preoperative data in detail are shown in Table 1.

Table 1
Preoperative case data of 11 children

No.	Age	Location of lesions	ABG	Tympanometry	Chief	Potsic
		(side, quadrant)	(dB HL)		complaint	Stage
1	10	Left, posterior attic + posterior hypotympanum	30	В	Hearing loss	III
2	6	Right, posterior hypotympanum	32	$A_S$	Hearing loss	I
3	3	Left, anterior hypotympanum + posterior hypotympanum	20	В	Physical examination	II
					(-)	
4	8	Right, posterior attic + posterior hypotympanum	55	С	Hearing loss	III
5	7	Left, posterior attic	40	В	Hearing loss	I
6	14	Left, posterior attic + posterior hypotympanum	21	С	Hearing loss	III
7	4	Left, posterior hypotympanum	30	В	Physical examination (-)	Ι
8	11	Left, anterior hypotympanum + posterior hypotympanum	25	С	Hearing loss	II
9	8	Right, anterior hypotympanum + posterior hypotympanum	35	В	Hearing loss	II
10	9	Left, posterior attic + posterior hypotympanum	20	С	Hearing loss	III
11	7	Left, posterior attic + posterior hypotympanum	25	С	Physical examination	II
					(-)	

# **Surgical Procedure And Intraoperative Results**

Eleven children received total ear endoscopic surgery under general anesthesia. All surgeries were performed by the same group of experienced ear surgeons using STORZ 3 mm 0°otoendoscope and 30°otoendoscope for exploratory. The duration of surgery lasted 60-90 minutes and the amount of blood loss was controlled within 5 mL. The surgical procedures of 11 children were shown in Table 2. Surgical procedures of representative case is shown in Figure 1. The rectangular flap incision was made in the external auditory canal 3mm from the drum ring. STORZ curettage or electric drill was used to enlarge

and fully expose the surgical field in the case of posterior wall or lower wall of external auditory canal uplift or ear canal curvature. The drum ring was raised to keep the integrity of the tympanum nerve. After stripping the cholestatoma cyst, eustachian tube, the entrance of tympanic sinus, facial crypt, tympanic sinus and anterior tympanic crypt were explored with 30° otoendoscope in order to guarantee no cholesteatoma epithelial residue. At the same time, hearing reconstruction was performed. Antibiotic NasoPore was filled with the external auditory meatus without pressure dressing after external auditory meatus-tympanic membrane flap reduction. Then the surgery was completed.

Table 2 Intraoperative findings in 11 children patients

No.	Location of lesions	Cavity condition	Hearing reconstruction
1	posterior attic + posterior hypotympanum	Long process of incus is damaged, nerve of tympanic cord is severed, tympanic mucosa is smooth, stapes is intact	Implantation of 1.0 mm PORP
2	posterior hypotympanum	Tympanic mucosa is smooth, ossicular chain is intact	/
3	hypotympanum	Tympanic mucosa is smooth, ossicular chain is intact	/
4	posterior attic + posterior hypotympanum	Long process of incus is damaged, tympanum mucosa is smooth, stapes superstructure is absent, soleplate moves well	Implantation of 4.5 mm TORP
5	posterior attic	Tympanic mucosa is smooth, ossicular chain is intact	/
6	posterior attic + posterior hypotympanum	Long process of incus is damaged, tympanum mucosa is smooth, stapes is intact, low position of facial nerve	Second surgery for hearing reconstruction
7	posterior hypotympanum	Tympanic mucosa is smooth, ossicular chain is intact	/
8	hypotympanum	Tympanic mucosa is smooth, ossicular chain is intact	/
9	hypotympanum	Tympanic mucosa is smooth, ossicular chain is intact	/
10	posterior attic + posterior hypotympanum	Intraoperative eardrum fracture, long process of incus is damaged, tympanum mucosa is smooth, stapes is intact,	Myringoplasty, implantation of 1.0 mm PORP
11	posterior	Tympanic mucosa is smooth, ossicular chain is intact	/

# **Postoperative Situation**

11 children took food 4-6 hours after surgery and sleep well at night. No postoperative fever, dizziness, tinnitus, sensorineural deafness, facial paralysis and other serious complications were observed. For case 1, the nerve of tympanic cord was severed during the surgery and no aberrations of taste was found. The second examination was performed three weeks after surgery. The packing of the ear canal was removed and the otoendoscope was performed for checking up. The incision of the external ear canal healed well, the tympanic membrane was intact, and there was no ear discharge or stenosis. Six months after surgery, as shown in Table 3 there was no significant change in bone conductance threshold (F=2.7, P>0.05), air conductance threshold and air-bone gap were significantly decreased (F= 0.05 and 0.039, respectively, P<0.05). One year after surgery, otoendoscope examination and temporal bone CT showed good healing of tympanic membrane without invagination and cholesteatoma recurrence. The pre- and post-operative results of representative case 4 were shown in Figure 2-5.

Table 3

Pre- and post-operative pure tone audiometry

No.	Pre-AC (dB HL)	Pre-BC (dB HL)	Pre-ABG (dB HL)	Post-AC (dB HL)	Post-BC (dB HL)	Post-ABG (dB HL)
1	37.5	7.5	30	20	6.25	13.75
2	38.25	6.25	32	10	5	5
3	25	5	20	11.25	6.25	5
4	61.25	6.25	55	10	10	0
5	45	5	40	18.75	5	13.75
6	25	5	20	35	5	30
7	37.5	7.5	30	15	6.25	8.75
8	30	5	25	12.5	5	7.5
9	41.25	6.25	35	16.25	6.25	10
10	25	5	20	17.5	5	12.5
11	31.25	6.25	25	10	5	5
Average	36.19±10.75	5.90±0.98	30.27±10.41	17.84±14.67	5.45±0.63	12.39±14.76

Pre-AC: preoperative air conductance, Pre-BC: preoperative bone conductance, Pre-ABG: preoperative air-bone gap, Post-AC: postoperative air conductance, Post-BC: postoperative bone conductance, Post-ABG: postoperative air-bone gap.

### **Discussion**

In recent years, more congenital middle ear cholesteatoma cases have been reported in children due to the widespread application of outpatient otoendoscopy and the improvement of clinicians' awareness of the disease. However, due to the characteristics of "short, flat and straight" eustachian tube in children, some children were diagnosed with CMEC only when tympanic membrane perforation and ear discharge occurred. Other children might be diagnosed when they visited hospital for secretory otitis media which were excluded by Levenson's criteria[4–6]. Since the lack of timely and accurate expression of unilateral hearing loss in children at a younger age, they are often sent to specialists when failed the hearing screening. In this study, 3 children did not pass the hearing screening and then were diagnosed with CMEC. Gilberto[7] suggest that routine hearing screening should be carried out for children entering kindergarten and school. For children who fail the hearing screening, otoendoscopic temporal bone CT should be performed. In this paper, 11 children underwent outpatient otoendoscope examination and all of them were found to have white localized mass shadows on the inner side of intact tympanic membrane. Therefore, the early detection rate of CMEC can be improved by using otoendoscope as a routine examination method for children with hearing loss or hearing screening failure in outpatient department. In addition, for children with secretory otitis media if Symptoms do not improve for a long time after treatment, temporal bone CT should be considered to perform to exclude CMEC.

The pathogenesis of CMEC is still unclear. Currently, there are epidermoid theory, drum ring development disorder theory, metagenesis theory, ectoderm implantation theory, etc. Among them, the epidermoid theory has been widely accepted. Although the accumulation of keratoid epithelium is benign, it can grow progressively and destroy normal surrounding tissue[7]. Congenital cholesteatoma in the posterior attic quadrant of the eardrum is often detected late in routine ear examinations because the eardrum is more opaque. Due to the adjacent ossicular chain, facial nerve and other organizational structures, congenital cholesteatoma occurring in the posterior quadrant is more invasive and can cause serious intracranial and external complications. In this study, 4 children with stage III CMEC all occurred in the posterior quadrant. Therefore, early detection and early treatment of CMEC is particularly important to avoid the occurrence of serious complications.

Currently, surgical treatment is the only treatment for CMEC[8]. The main objectives of surgery are to completely remove lesions, prevent recurrence, preserve or rebuild hearing. For patients with CMEC, the classical surgical methods are complete wall mastoid radical resection and tympanoplasty according to the lesion scope. For localized lesions of stage I and stage II, some scholars also used enlarged tympanum exploration [9]. CMEC children's mastoid gasification is good and lesions are limited. Radical mastoidectomy is more traumatic and children needs to be cleaned regularly in the clinic which cause children poor cooperation. Moreover, microscopic exposure of the attic tympanum, facial recess and tympanum sinus is limited which may cause incomplete removal of the lesion and recurrence. We chose the surgical treatment of CMEC under otoendoscope based on the literature report that CMEC within stage III can be treated through the ear canal approach under the microscope. This approach can avoid damage to mastoid bone and air chamber with mild lesions [10]. For stage III CMEC, it could also be completed by ear canal approach when lesion is localized and mastoid process is not invaded before surgery. The stage III cases in this paper were all underwent surgeries by otoendoscope. The application of angle scope also helped to preserve the bone wall of the external auditory canal and the lateral wall of the upper drum. External auditory canal exudation and dressing time decreased significantly.

It is reported that the recurrence rate of CMEC in children is 10.5%-45%[11, 12]. At present most reports on CMEC are small-sample reports[7, 11, 13-16], so there is no data to prove that otoendoscope can reduce the recurrence rate of CMEC. With the development of ear endoscopy, ear endoscopy with a diameter of 2.7 mm and 1.9 mm can pass through the narrow ear canal, increase the surgical field of vision, improve the clarity of the surgical field and observe tissue from multiple angles at close range[17]. Otoendoscope can provide a clear vision for the superior tympanum, facial crypt, tympanum sinus, eustachian tube and other concealed anatomical sites. The incision of otoendoscopic surgery is small and it is easier for families to accept even the children who plan to have a second surgery. Another difficulty of CMEC children is the stenosis of the ear canal. When preoperative CT indicates that the anteroposterior diameter of middle part of external auditory canal (midpoint of ear canal opening and umbilical point of eardrum) ear bone wall exceeds 4.5 mm, an otoendoscope with a diameter of more than 2.7 mm can be used for operation [18]. The anteroposterior diameter of the isthmus of the external auditory canal in Chinese children is 3.27±0.75 mm for 0-5 years old and 4.01±0.75 mm for 6-15 years old. If the external auditory canal bulge affects the surgical field, curettors or electric drills can be used to expand the external auditory canal to increase the surgical field of vision. Due to the thin skin of children's ear canal, protection of ear canal flap is important which can avoid postoperative scar stenosis.

In this study, all 11 children were treated with transcanal endoscopic surgery. In 1 case, cartilage connection was implanted because of low position of facial nerve and the hearing was not recovered after surgery. The second surgery to reconstruct hearing was considered. In another case, tympanic membrane fissure occurred during surgery separation. The eardrum is repaired with perichondrium at the same time. The healing was good 3 weeks after surgery. Up to now, hearing of all cases has been significantly improved. The 7 cases of stage I and stage II were young and prompt diagnosis. Their external auditory canal and shield plate were not expanded during the surgery. The ABG was reduced to within 10 dB HL after surgery and recovered dry ear in about 2 months. The 4 cases of stage III were all over 7 years old which may be related to the lack of early birth examination and hearing screening. 1 case planned to undergo the second surgery for hearing reconstruction, the hearing was significantly improved in other 3 cases. In order to clarify the lesion, there were different degrees of external auditory canal enlargement. All the children recovered dry ears within 3 months after surgery and no auditory ossicle discharge, displacement and recurrence were observed.

## **Conclusions**

Transcanal endoscopic treatment for congenital middle ear cholesteatoma in children is feasible, minimally invasive and functional. It is also worth popularizing in the treatment of CMEC in children. However, due to the limited number of children with CMEC and the short follow-up time of some children, further research is needed.

## **Declarations**

Ethics approval and consent to participate

The study compiled with the principles of the Declaration of Helsinki on Biomedical Research Involving Human Subjects and was approved by the Ethical Review Board of Huazhong University of Science and Technology Union Shenzhen Hospital (Nanshan Hospital). Written informed consent was obtained from each child's parent or supervisor.

Consent for publication

Not applicable.

Availability of data and materials

The datasets and analysis used during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

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Not available.

Authors' contributions

QY and SL are acting as the submission's guarantors, take responsibility for the integrity of the work, and substantially contributed to the conception and design of the study; NZ and ML were involved in the acquisition of data; NZ, SY and LZ performed the analysis and interpretation of the data; NZ and SY wrote the first draft of the paper; QY and SL revised it critically for important intellectual content. All authors have read and approved the manuscript.

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# **Figures**

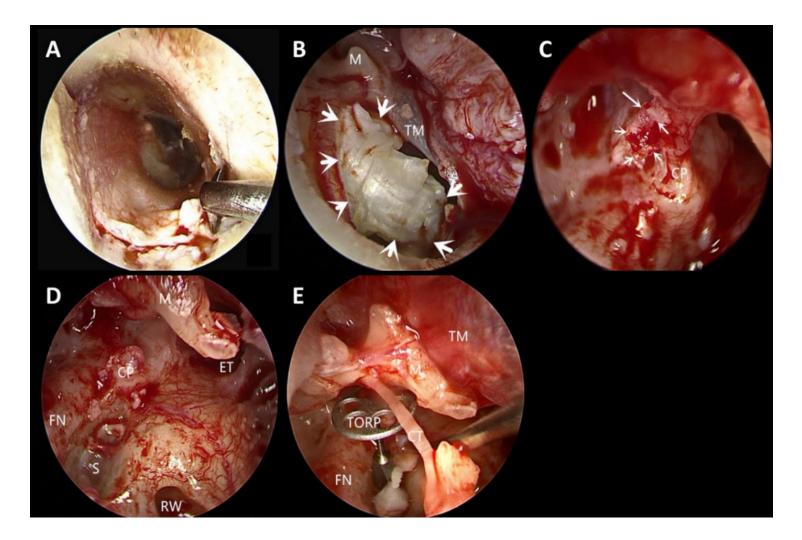


Figure 1

Case 4 surgical procedure: (A) rectangular flap was opened under otoendoscope, (B) exposure of cholesteatoma cysts, (C) replace 30° otoendoscope to reveal the cholesteatoma epithelium in the medial side of tympanic tensor tendon, (D) residual cholesteatoma was observed under 30° otoendoscope, (E) total ossicular replacement prosthesis . M: malleus, TM: tympanic membrane, CP: spoon process, ET: eustachian tube, FN: facial nerve, S: stapes, RW: round window, CT: chorda tympani nerve, TORP: total ossicular replacement prosthesis.

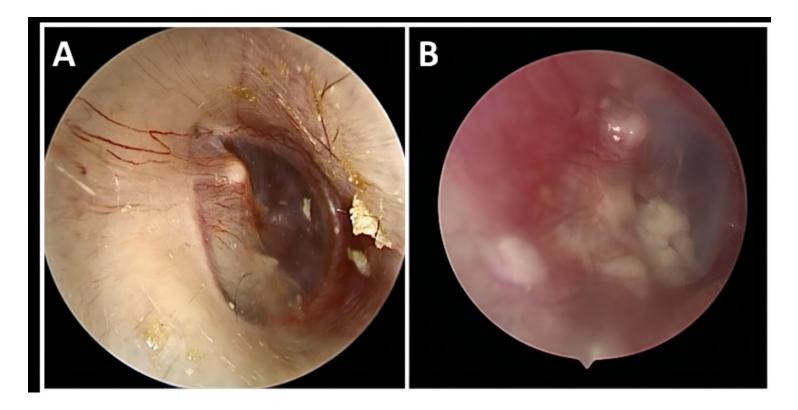


Figure 2

Otoendoscope examination in case 4: (A) white patchy shadow behind intact tympanic membrane was observed before surgery, (B) the tympanic membrane was intact and cartilage was visible inside after surgery.

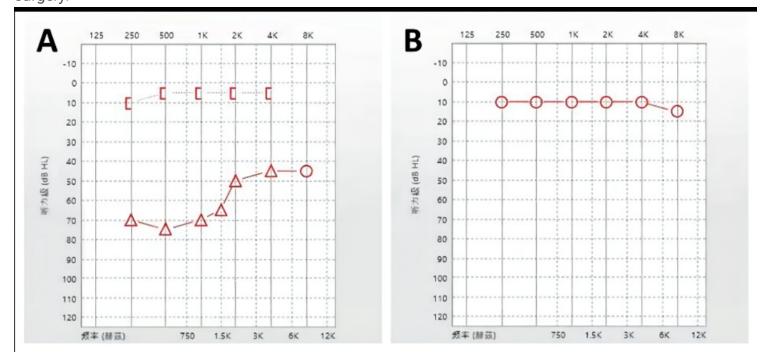


Figure 3

Pure tone audiometry in case 4: (A) severe conductive hearing loss before surgery, (B) hearing recovery after surgery.

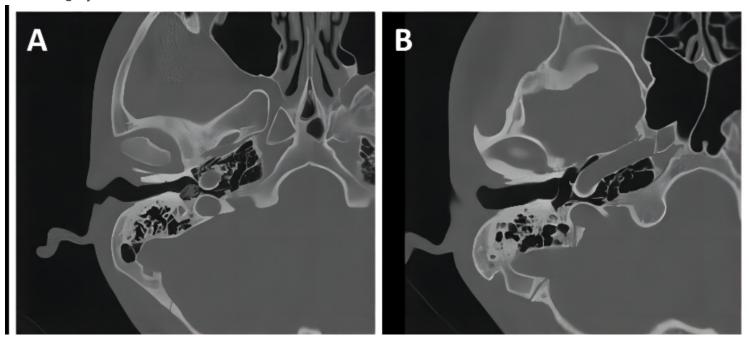


Figure 4

CT images in horizontal view in case 4: (A) hypodensity shadow was observed in the inferior tympanum horizontally before surgery, (B) tympanic hypodensity shadow disappeared and posterior wall of external auditory canal was intact after surgery.

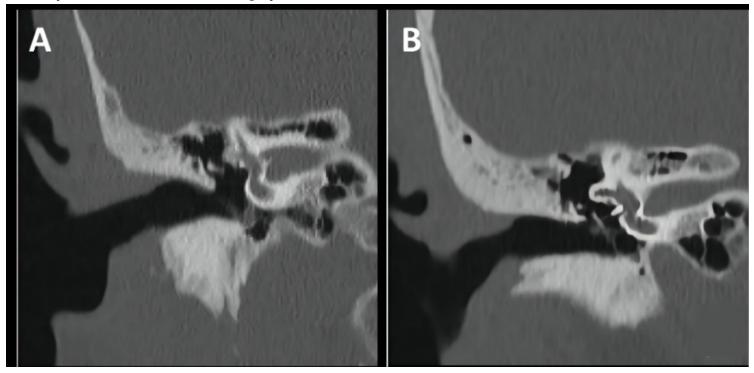


Figure 5

CT images in coronal view in case 4: (A) intact scute, hypodensity shadow of inferior tympanum, incomplete ossicular chain before surgery, (B) the scute was intact, hypodensity shadow of the inferior tympanum disappeared, artificial ossicles were present in the vestibular window.