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CLINICAL REPORT

Clinical Analysis of 22 Cases with Automastoidectomy Caused by Cholesteatoma

SunKyu Lee, Seunggeun Yeo, Moonsuh Park, Jaeyoung Byun

Department of Otorhinolaryngology, Head and Neck Surgery, KyungHee University Hospital at Gandong, Seoul, Korea (SK, MP, JB)
Department of Otorhinolaryngology, Head and Neck Surgery, KyungHee University Hospital, Seoul, Korea (SY)

Objective: Automastoidectomy is an uncommon but unique complication of middle ear cholesteatoma that has been understudied in the current literature. The authors treated patients suffering from automastoidectomy due to cholesteatoma and reported clinical features of this complication.

Materials and Methods: Retrospective study with medical records review was performed in tertiary care academic otolaryngology practice. Patients with chronic cholesteatomatous otitis media who showed evidence of automastoidectomy by temporal bone computerized tomography (TBCT) were included. Preoperative, intraoperative, and postoperative records were analyzed retrospectively.

Results: There were 22 patients with automastoidectomy. Symptoms were otorrhea, hearing disturbance, otalgia, and facial palsy. Procedures performed were as follows: 2 patients underwent canal wall up mastoidectomy, 19 patients underwent canal wall down mastoidectomy, and 1 patient underwent a subtotal petrosectomy. The rates of facial nerve palsy and labyrinthine fistula were 9.1% and 36.4%, respectively. Postoperatively, hearing improved in 1 case and 2 cases resulted in deafness.

Conclusion: Despite relatively minor symptoms, operative findings were severe in automastoidectomy. Precise preoperative evaluation and a thorough explanation to patients are necessary. Additionally, any surgical procedure involving the middle or inner ear must be performed by an experienced surgeon.

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Introduction

Cholesteatoma is a cystic lesion formed from keratinizing stratified squamous epithelium.[1] Since the initial stages of research in cholesteatoma, there have been numerous discussions regarding its classification. Classically, cholesteatoma is divided into several categories: congenital, primary acquired and secondary acquired. There is also a classification of acquired cholesteatoma based on the lesion site: attic, sinus, and tensa cholesteatoma.[2] Classification of cholesteatoma also takes into account the site of origin which is also important for surgical decision making and prognosis.

Complications due to cholesteatoma are usually related to the disease state and are important during medical and surgical management. Representative complications are fistula formation of the lateral semicircular canal (SCC), facial palsy, total sensorineural hearing loss, sinus thrombosis, and intracranial invasion.[3] "Automastoidectomy" is an uncommon and rather unique complication of cholesteatoma of the middle ear.[4] It can be defined as the destruction of the mastoid bone forming the posterior wall of the external auditory canal (EAC) by the adjacent cholesteatoma in patients with no history of a previous operation.

Corresponding address:
Byun JaeYong
Department of Otorhinolaryngology, KyungHee University Hospital at Gangdong,
#892, Sangil-dong, Gangdong-gu, Seoul, 134-727, Korea
Tel; 82-2-440-8160
Fax; 82-2-440-8470
E-mail; otorhino512@naver.com

Few articles concerning "automastoidectomy" are found in the current literature. Some reported CT findings of "automastoidectomy" and compared them with those of post-mastoidectomy.[5,6] Another case-report of "automastoidectomy" was in the setting of keratosis obturans, not cholesteatoma.[7] The authors analyzed the treatment of patients with "automastoidectomy" due to cholesteatoma and reported pre-, intra-, and post-operative findings.

Materials and Methods

Patients diagnosed with chronic suppurative otitis media and cholesteatoma from January, 2003 to February, 2011 were included in this study. Medical records including medical history, otomicroscopic findings, audiologic results, and temporal bone computerized tomography (TBCT) findings were thoroughly evaluated.

"Automastoidectomy" was defined as the presence of posterosuperior wall destruction of the bony EAC or attic seen on otomicroscopic examination or when a bony defect surrounding the mastoid antrum was connected to the EAC as seen by TBCT.[4] Patients meeting criteria for "automastoidectomy" underwent surgery for removal of the lesion or improvement of hearing were included to this study. Postoperatively, all patients were followed for more than 1 year. Patients were excluded from the study if they had a history of previous surgery or trauma (traumatic tympanic membrane perforation, temporal bone fracture) on the side of the lesion.

Preoperative medical history, pure tone audiometry (PTA) results, physical examination findings, and TBCT findings were analyzed. Operations for removal of cholesteatoma and hearing improvement and not improvements were analyzed in addition to intraoperative findings. Postoperative otomicroscopic findings and PTA results were examined. Postoperative PTA was performed at 1 month, 3 months, 6 months, and 12 months postoperatively and then annually after the first year.. In this study, final PTA results 1 year postoperatively were evaluated.

Mean thresholds at frequencies of 0.5, 1, 2, and 3 kHz were used for PTA averages according to recommendations made by the Committee on Hearing and Equilibrium.[8] Using air and bone conduction values an air-bone gap (ABG) was calculated. The postoperative air-bone gap value was used to estimate postoperative hearing improvements according to the Committee on Hearing and Equilibrium: 0-10 dB, 11-20 dB, 21-30dB, and >30dB.[8]

This study was approved by the local ethics committee (KHNMC IRB 2012-084).

Results

Preoperative findings

A total of 22 patients were included to this study with 10 male and 12 female patients. There were 14 cases of right ear and 8 cases of left ear involvement. The mean age was 48.1 years old (Standard deviation: ± 13.2 years and range: 17 – 66 years). Major symptoms were 10 cases of otorrhea, 7 cases of hearing disturbance, 3 cases of otalgia, and 2 cases of facial palsy on the ipsilateral side of the involved ear. The mean duration major symptoms was 21.3 years (± 20.8 years). While most patients complained of a long duration of suffering, 2 cases of facial palsy complained of a short duration of a major symptom (4 days and 4 months, respectively) compared to other patients.

Results of otomicroscopic examination at the outpatient-clinic were as follows: All 22 patients showed destruction of the attic or posterosuperior bony EAC. 17 cases posterosuperior bony EAC destruction , 14 cases of attic destruction, 12 cases of retraction or adhesion of the tympanic membrane (pars tensa and/or flaccida), and 4 cases of tympanic membrane perforation.

Intraoperative findings

Bony dehiscence of the facial nerve was observed intraoperatively in several patients. Bony dehiscence of the tympanic segment (including the second genu) was found in 9 cases and 2 cases showed mastoid segment dehiscence. Both tympanic and mastoid segments were exposed during 6 cases and facial nerve herniation and ischemic change was observed in 1 case within these cases.

The malleus was absent in 14 cases, the malleus head was destroyed but with an intact handle in 4 cases, and an intact malleus was present in 4 cases. An absent incus was seen in 19 cases, partial incus was found in 1 case, and 2 cases showed an intact incus. The whole stapedial superstructure, excluding the footplate, was absent in 15

cases and in 1 case only a membranous structure overlying the oval window was found. An intact stapes was found in 6 cases.

Lateral SCC fistula was observed in 8 cases and 2 cases of them showed involvement of the membranous labyrinth. There were 4 cases with defect of tegmen: 2 cases showed a defective tegmen tympani and exposed dura of the middle fossa, 1 case showed a defect in the mastoid tegmen, and 1 case showed defects in both the tegmen tympani and mastoid tegmen. Laceration of the dura or cerebrospinal fluid leakage was not observed in any of the 4 cases.

Operative procedure

The mastoid operation was performed for removal of cholesteatoma and control of inflammation in 22 cases. We performed a canal wall down (CWD) mastoidectomy in 19 cases, canal wall up (CWU) mastoidectomy in 2 cases, and subtotal petrosectomy in 1 case. Attic reconstruction with a cortical bone chip was performed in 2 cases of canal wall up mastoidectomy. Of the 19 cases of canal wall down mastoidectomy, 1case was an intact bridge mastoidectomy.

Tympanoplasty without ossicular reconstruction (so called type 0 tympanoplasty) and columella over stapes footplate with total ossicular replacement prostheses (TORP; made of polycel) was the most frequently performed procedure for tympanoplasty (6 cases and 6 cases, respectively). Other procedures include columella over stapes footplate with cartilage block in 4 cases, columella over stapes head with partial ossicular replacement prostheses (PORP; made of polycel) in 3 cases, tympanoplasty type 1 (with fascia of tempolaris muscle) in 1 case, tympanoplasty type 3 (with fascia of tempolaris muscle) in 1 case, and middle ear obliteration in 1 case.

Postoperative findings and hearing results

All patients were followed for 1 year after surgery and 20 cases showed no evidence of disease recurrence. A large perforation of the tympanic membrane without otorrhea was observed in 1 case and retraction of the tympanic membrane (Sade grade 1) was observed in 1 case. These 2 cases have been followed without the need for additional surgical procedures.

Results of postoperative PTA are shown in Table 1. There was 1 case of intact bridge mastoidectomy with tympanoplasty type 1 which showed a successful

Table 1. Summary of hearing results according to operative procedure.

	Number	Mastoidectomy		Preop PTA		Postop PTA		Preop ABG	Postop ABG	Postop ABG ≤10dB	Postop ABG ≤20dB	Postop ABG ≤30dB	Postop ABG >30dB	Comment
		CWDM	CWUM	AC	BC	AC	BC							
Tympanoplasty type 1	1	1 (IBM)		15	5	15	5	10	10	1/1				
PORP over stapes head	3	1	2	48 ± 7	15 ± 6	46 ± 24	18 ± 15	33 ± 2	28 ± 9		1/3	1/3	1/3	
TORP over stapes footplate	6	6		71 ± 12	29 ± 4	66 ± 12	32 ± 8	42 ± 10	34 ± 12		1/6	2/6	3/6	
Columella over stapes footplate	4	4		75 ± 29	36 ± 15	83 ± 27	41 ± 20	39 ± 15	41 ± 12				4/4	
Tympanization deaf	6	6		80 ± 33	41 ± 18	93 ± 42	51 ± 22	40 ± 17	42 ± 21				6/6	1 case total
Tympanoplasty type 3	1	1		36	10	40	14	26	26			1/1		
Obliteration deaf	1	1		70	39								1/1	1 case total

postoperative ABG of less than 10 dB. A postoperative ABG of less than 20dB was seen in 2 cases; 1 case of columella over stapes head with PORP (made of polycel) and 1 case of columella over stapes footplate with TORP (made of polycel). There were 17 cases with an unimproved postoperative ABG according to recommendations of the Committee on Hearing and Equilibrium.[8] There were 2 cases that resulted in postoperative deafness. Complete hearing loss occurred in 1 case of subtotal petrosectomy due to middle ear obliteration and 1 case of CWD mastoidectomy with tympanization due to rupture of the lateral SCC during dissection of the cholesteatoma sac over the lateral fistula SCC.

Specific cases

While most cases involved the CWD mastoidectomy procedure, 2 cases required a CWU mastoidectomy. Of the 2 cases, one was a 33-year-old female patient who suffered from recurrent otorrhea beginning 20 years prior. Under otomicroscopic examination, large, healed perforations of the tympanic membrane were observed along with attic destruction and perforation of the pars flaccida. TBCT showed a blunted scutum, widened Prussak space, an "automastoidectomy" state, and a previous antro-cutaneous fistula at the posterolateral portion of bony EAC. The epitympanum and middle ear cavity was relatively clear (Fig 1.). The opposite ear was also had evidence of CSOM. Cholesteatoma was observed in the mastoid cavity and granulation tissue was found in the epitympanum. Considering that the patient was a young female and had bilateral chronic otitis media, we performed a CWU mastoidectomy with a PORP insertion. Preoperative air conduction, postoperative air conduction, and postoperative ABG were 43 dB, 25 dB, and 12 dB, respectively. Hearing improved and cholesteatoma recurrence has not yet been observed.

The other case requiring a CWU mastoidectomy was a 23-year-old female, who suffered from a right-sided hearing disturbance for 1 year. Destruction of the attic and retraction of the tympanic membrane (Sade grade 1) was observed. TBCT showed a blunted scutum with widened Prussak space (Fig 2). Chronic cholesteatomatous otitis media of the opposite ear was

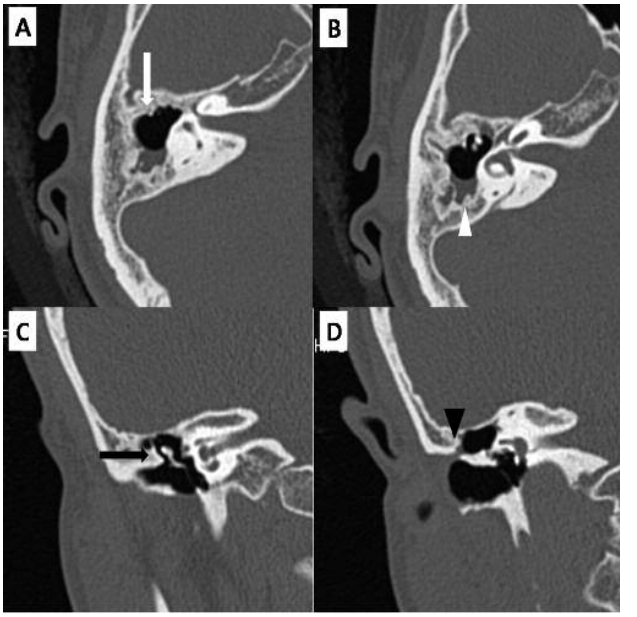

Figure 1. Temporal bone computed tomography. Axial view; **A, B:** Widened aditus and an example of the automastoidectomy state (white arrow) with cholesteatoma material remaining in the automastoidectomy cavity (white arrow head). Coronal view; **C:** Blunted scutum with widened Prussak space (black arrow). **D:** Possible previous antro-cutaneous fistula (black arrow head).

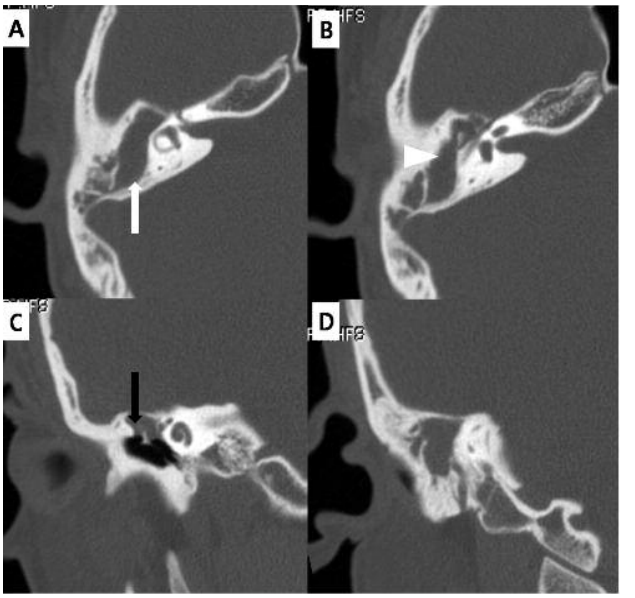

Figure 2. Temporal bone computed tomography. Axial view; **A, B:** An automastoidectomy state (white arrow)and cholesteatoma in the automastoidectomy cavity and epitympanum (white arrow head). Coronal view; **C:** Blunted scutum with a widened Prussak space (black arrow). **D:** Automastoidectomy cavity filled with cholesteatoma.

also found. Preoperative air and bone conductions of the right ear were 50 dB and 15 dB and those of left ear were 19 dB and 10 dB, respectively. The right side was operated on first and due to her bilateral chronic cholesteatomatous otitis media, we decided to perform a hearing-preserving procedure. Intraoperatively, cholesteatoma was found to be located at the mastoid cavity, aditus and epitympanum. The malleus head and body of the incus were destroyed and the middle ear cavity was relatively clear. CWU mastoidectomy with PORP insertion was performed. Postoperative air conduction, bone conduction, and ABG measurements were 30 dB, 6 dB, and 24 dB, respectively. After the first operation, the opposite side was corrected 3 years later with an intact bridge mastoidectomy. Both sides have been followed and are without evidence of disease recurrence.

The most severe case was in a 39 year-old male patient with left facial palsy and a House-Brackmann grade of 5 that occurred 4 days before admission. His primary complaints on presentation were otorrhea and otalgia. Destruction of the attic and posterosuperior EAC, cholesteatoma matrix filling of the bony defect site, and large perforations of the tympanic membrane were observed. An "automastoidectomy" state with cholesteatoma, labyrinthine fistula, and facial canal dehiscence was observed using TBCT (Fig 3). Cholesteatoma was located in the mastoid cavity, aditus, and epitympanum and extended into the petrous apex. Tympanic and mastoid segments of the facial nerve were herniated causing ischemia to the tissue. During the operation the lateral SCC was ruptured and caused leaking of a large amount of perilymph. Transmastoid facial nerve decompression, subtotal petrosectomy, and obliteration of the middle ear and mastoid using an abdominal fat were performed. Preoperative air bone conduction measurements were 70 dB and 39 dB, respectively. The patient lost hearing postoperatively.

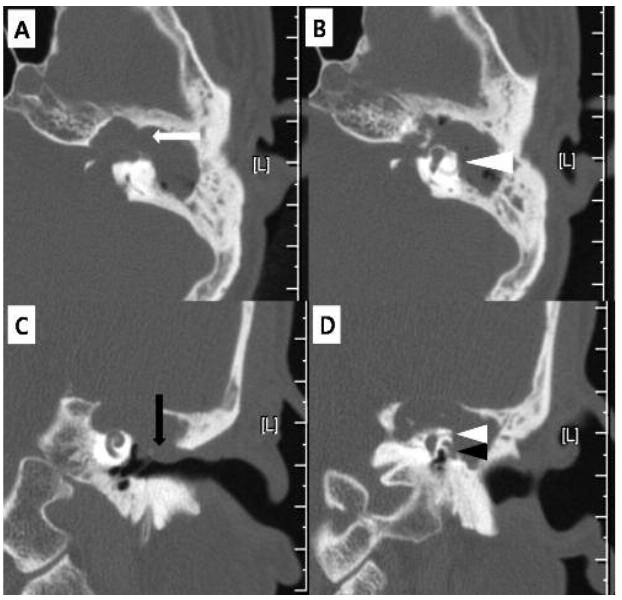

Figure 3. Temporal bone computed tomography. Axial view; **A:** Extensive cholesteatoma and automastoidectomy state with extension into the petrous apex (white arrow). **B:** Destructed bony wall of the lateral semicircular canal (white arrow head). Coronal view; **C:** Destroyed posterosuperior bony external auditory canal wall and cholesteatoma extending to the external auditory canal (black arrow). **D:** Destroyed facial nerve canal (white arrow head) and possible labyrinthine fistula (black arrow head).

Discussion

Automastoidectomy is defined as the extensive destruction of the middle ear cavity and mastoid. Occasionally, a cholesteatoma will drain after spontaneous destruction of the posterosuperior wall of the EAC causing the appearance of a post-mastoidectomy state.[4,9,10] This condition may arise as a complication of cholesteatomatous chronic otitis media or of keratosis obturans.[7,11] However, only a few characteristics, regardless of disease etiology (especially radiologic findings), have been described regarding automastoidectomy in the literature.

Cholesteatoma could be classified according to the anatomical site or origin: attic cholesteatoma, sinus cholesteatoma, and tensa cholesteatoma.[2] Attic cholesteatoma is defined as a retraction of the pars flaccida or Shrapnell's membrane which extends into the attic and eventually into the antrum, mastoid, or tympanic cavity.[2] Combination of the retraction and proliferation theories have been suggested as an explanation for the pathogenesis of an attic cholesteatoma.[12] Sinus cholesteatoma is defined as a posterosuperior retraction or perforation of the pars tensa extending to the tympanic sinus or beyond the posterior tympanum.[2] Tensa cholesteatoma is defined as the retraction and adhesion of the entire pars tensa involving the tympanic orifice of the Eustachian tube.[2]

All 22 cases of this study had no history of surgery or trauma to the involved ear. Destruction of the attic or posterosuperior EAC wall was found in all cases and matrix or cholesteatoma sac was found in the antrum, aditus, and epitympanum in all cases, intraoperatively. This suggests that all cases could be considered as primary acquired cholesteatoma and extension of attic cholesteatoma.

Mean duration of symptoms was 21.3 years, except in cases of sudden catastrophic symptoms such as facial palsy, which may be due to disease progression without adequate treatment. Progression of disease during a long period may cause destruction of the mastoid bone and the state of "automastoidectomy." Tympanic and/or mastoid segments of the facial nerve were dehisced in 17 cases (77.3%), which were observed intraoperatively. Clinical facial palsy developed on the side of cholesteatoma in 2 cases. Exposed tympanic and mastoid segments with ischemic change and a House-Brackmann grade of 5 was seen in 1 case. Subtotal petrosectomy and transmastoid facial nerve decompression was performed in this case. Postoperatively, facial palsy was aggravated as House-Brackmann grade of 6. The other case showed exposure of the tympanic and mastoid segments without neural change and a House-Brackmann grade of 4. CWD mastoidectomy and transmastoid facial nerve decompression was performed in this case and facial palsy was improved to House-Brackmann grade of 1. Fistula of the lateral SCC was observed in 8 cases (36.4%). Cholesteatoma was found to be firmly attached to the membranous labyrinth in 2 cases of lateral SCC fistula. Subtotal petrosectomy was performed in 1 of them. Rupture of the membranous labyrinth occurred during dissection of the sac of cholesteatoma over the fistula site in another case. Both cases became deafness, postoperatively.

Only a few studies have reported incidences of facial palsy after treatment of cholesteatoma.[13–16] According to previous studies, the frequency of facial palsy in chronic suppurative otitis media (CSOM) ranges from 0.16 to 14.3%.[17] Labyrinthine fistula develops from an inflammatory response restricted to a discrete portion of the bony labyrinth and endosteum without involvement of the membranous labyrinth. Common causes of pathologic fistulae of the labyrinth are cholesteatoma, granulation tissue development, and cholesterol granulomas. The most common site is the lateral SCC, however, fistulae of the cochlear or vestibular walls may occur. Labyrinthine fistula was reported in 3-12% of total CSOM cases[18-20] and 7% of chronic cholesteatomatous otitis media cases.[21] While cases from this study showed similar rates of facial palsy, the rate of labyrinthine fistula (36.4%) was much higher than that of previous reports and may be the result of higher disease severity as evidenced by the presence of the automastoidectomy state in our study patients.

Preoperative air conduction, bone conduction, and ABG of total patients were 66.9±26.7 dB, 30.2±15.6 dB, and 36.8±13.2dB, respectively. Postoperative air conduction, bone conduction, and ABG were 66.4±29.9 dB, 32.3±18.7 dB, and 34.0±13.9 dB, respectively. When patients were classified according to recommendations made by the Committee on Hearing and Equilibrium,[8] hearing improved in 3 patients (3/20, 15%). 3 patients were as follows; one was performed IBM, another one was performed CWD mastoidectomy with ossiculoplasty with PORP on the stapes head, and the other one was performed CWD mastoidectomy with ossiculoplasty with TORP on the stapes footplate. Patient inserted TORP showed preoperatively 65dB air conduction and 24 dB bone conduction threshold and postoperatively 40dB air conduction threshold and 25 dB bone conduction threshold. The incus was the most frequently involved ossicle and was damaged or missing in 20 cases (90.9%). The stapes was abnormal in 16 cases (72.7%). Presence of cholesteatoma, ossicular status, middle ear granulation, and otorrhea are known to be important prognostic factors of postoperative hearing threshold.[22] Because all automastoidectomy cases were caused by cholesteatoma and had poor ossicular status and middle ear environment, result of ossicular reconstruction was generally poor.

Normal ossicles and their interactions were normal in 1 case. In this case, the patient visited the hospital for recurrent otorrhea, which began 4 years ago. Destruction of the attic and posterosuperior EAC wall as well as retraction of the pars tensa (Sade grade III)

was observed under otomicroscopic examination. TBCT showed normal ossicles, a blunted scutum, and destruction of the posterosuperior bony EAC. Mastoid air cells were partially exteriorized and the EAC was widened (Fig 4.). Preoperative air conduction was 10 dB. Spontaneous drainage of cholesteatoma through a bony defect is suspected in this case. We suspected the cause of the chief complaint, otorrhea, would be due to a cavity problem. Intact bridge mastoidectomy with attic reconstruction was performed to remove remnant mastoid air cells and preserve hearing. Mastoid obliteration and meatoplasty was also performed to solve the cavity problem. Postoperative air conduction was maintained within normal limits (10 dB) and the mastoidectomy site remained normal.

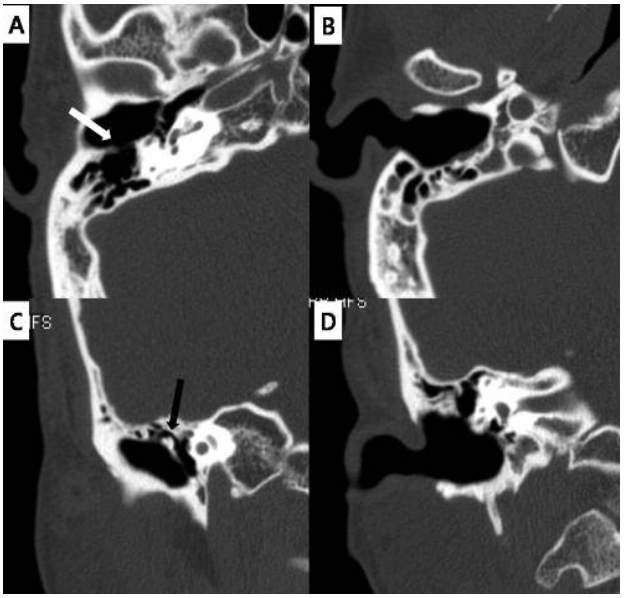

Figure 4. Temporal bone computed tomography. Axial view; **A:** Destruction of the posterosuperior bony external auditory canal (white arrow). **B:** Coronal view of a widened bony external auditory canal with partial exteriorization of mastoid air cells. Coronal view; **C:** Intact malleus head with shallow epitympanum (black arrow). **D:** Widened bony auditory canal with partial exteriorization of mastoid air cells.

Conclusion

Automastoidectomy is an uncommon complication of chronic cholesteatomatous otitis media. It shows similar features to post-mastoidectomy on TBCT. Most cases usually have a long duration of symptoms, such as otorrhea and hearing disturbances, which are generally not severe. While symptoms may be minor, complications are frequently encountered due to the severity of the lesion. The main goal of surgery is eradication of cholesteatoma and a radical procedure is usually acceptable. Occasionally, function-preserving procedures, such as CWU mastoidectomy and intact bridge mastoidectomy, can be performed when the condition of the opposite ear and severity of the lesion is considered. Unpredicted accidents can occur during an operation and resulting in poor postoperative hearing. Adequate preoperative analysis of each patient and a thorough explanation about complications and surgical results are essential. Most importantly, the operation must be performed by an experienced surgeon.

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