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Bilateral automastoidectomy- A rare case report of destructive sequela with challenges in hearing rehabilitation

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Conflicts of Interest: Nil

Abstract

Automastoidectomy is a rare sequela of chronic otitis media characterized by extensive spontaneous erosion of the mastoid and adjacent temporal bone, resulting in a naturally formed canal wall down like cavity. Bilateral automastoidectomy is exceptionally uncommon. We present a 30 year old female with progressive bilateral hearing loss for more than five years and a history of recurrent, scanty, mucopurulent otorrhea during childhood. She had no history of previous ear surgery or otogenic complications. Otoendoscopy demonstrated bilaterally widened external auditory canals communicating with dry, well-epithelialized mastoid cavities, with tympanic membrane retraction and evidence of ossicular erosion. Pure tone audiometry revealed severe-to-profound bilateral conductive hearing loss, with hearing thresholds of 82.5 (decibel) dB in the right ear and 68.75 dB in the left ear. High-resolution computed tomography of the temporal bones showed bilateral mastoid cavities with complete loss of mastoid air cells, communication with the posterior external auditory canals, and near-complete ossicular erosion, producing a characteristic "scooped-out" appearance resembling a postoperative canal wall down mastoid cavity. The patient underwent staged bilateral canal wall down

mastoidectomy with right type 3c cartilage ossiculoplasty and left total ossicular replacement prosthesis ossiculoplasty. Postoperative audiological assessment demonstrated approximately 30 dB improvement in hearing, with stable and dry postoperative cavities. Bilateral hearing aids were subsequently advised for further auditory rehabilitation. This case highlights the rare occurrence of bilateral automastoidectomy following long-standing neglected chronic otitis media and the importance of early recognition and treatment of childhood otorrhea to prevent irreversible temporal bone destruction and hearing loss. Surgical hearing rehabilitation with ossicular reconstruction, supplemented by amplification when required, may provide meaningful functional benefit in appropriately selected patients.

Introduction

Automastoidectomy is a rare pathological phenomenon, characterized by spontaneous erosion and remodeling of the mastoid air cell system, with spontaneous exteriorization of the mastoid cavity through the external auditory canal[1]. It is commonly seen with long-standing chronic otitis media and cholesteatoma. It has been described in neglected cases of keratosis obturans[2]. The progressive destructive process may lead to spontaneous evacuation of cholesteatomatous material and subsequent epithelialization of the cavity thus produced. This condition gives an appearance similar to that of a surgically created canal wall-down mastoid cavity. Bilateral involvement is very rare, although unilateral automastoidectomy has been reported in the literature[3]. Despite fairly inactive clinical disease at presentation, the degree of destruction of the temporal bone and ossicles may be considerable. This entity is important to recognize, as it may be confused with a postoperative mastoid cavity or other destructive pathology of the temporal bone. We present a rare case of bilateral automastoidectomy in a 30-year-old female with a history of recurrent childhood otorrhea and no history of ear surgery, leading to extensive bilateral mastoid and ossicular destruction with severe conductive hearing loss. The case also emphasizes the importance of surgical disease control and subsequent hearing rehabilitation in patients with spontaneous stabilization of destructive middle ear disease[4].

Case Presentation

A 30-year-old female from a rural area of Bihar, India presented with bilateral hearing loss for more than five years. She gave a history of recurrent, scanty, mucopurulent discharge from both ears during childhood, associated with mild intermittent earache and tinnitus. There was no history of previous ear surgery. On examination, the bilateral postauricular regions showed no surgical scars (as shown in Figure 1 and 2).

Otoendoscopy of the right ear demonstrated widening of the external auditory canal, with the posterior canal wall communicating with a well-epithelialized, exposed and dry mastoid cavity. The tympanic membrane was intact and retracted, with a foreshortened handle of the malleus. The left ear showed similar findings, although the mastoid cavity was larger. The handle of the malleus appeared partially eroded [as shown in Figure 3 and 4] Tuning fork examination revealed negative Rinne's test bilaterally at all tested frequencies, a centralized Weber’s test, and absolute bone conduction comparable to that of the examiner bilaterally.

Pure-tone audiometry (PTA) demonstrated severe-to-profound conductive hearing loss bilaterally. The right ear showed an average hearing threshold of 82.5 dB, with an air-bone gap of approximately 55 dB. The left

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ear showed an average hearing threshold of 68.75 dB, with an air-bone gap of approximately 50 dB [as shown in Figure 5]. These findings indicated significant bilateral conductive hearing impairment secondary to extensive middle-ear and ossicular destruction.

High-resolution computed tomography of the temporal bones demonstrated bilateral mastoid cavities with complete loss of the normal mastoid air-cell system and communication with the posterior wall of the external auditory canals. The ossicular chains were not visualized except for a portion of the head of the malleus on either side. The overall appearance was described as a "scooped-out" mastoid cavity resembling a postoperative canal wall-down mastoidectomy cavity [as shown in Figure 6A and 6B].

In view of the characteristic clinical and radiological findings, together with the absence of previous otologic surgery, a diagnosis of bilateral automastoidectomy secondary to longstanding chronic otitis media with cholesteatomatous disease was made. The patient underwent staged surgical management. The right ear underwent canal wall down mastoidectomy with cartilage ossiculoplasty. The left ear subsequently underwent canal wall-down mastoidectomy with Total Ossicular Replacement Prosthesis (TORP) ossiculoplasty (as shown in Figure 7). The objectives of surgery were to establish a safe and accessible cavity, address residual pathological changes, stabilize the disease, and attempt hearing rehabilitation [as shown in Figure 8 and 9].

Postoperative audiological assessment demonstrated stabilization of hearing with a modest functional improvement of approximately 30 dB reduction in air-bone gap. The patient was subsequently counselled regarding bilateral hearing-aid rehabilitation to further optimize auditory function and quality of life.



Figure 1: Postauricular scarless right ear (as shown by red arrow) The image has been taken by the authors



Figure 2: Postauricular scarless left ear (as shown by blue arrow) The image has been taken by the authors

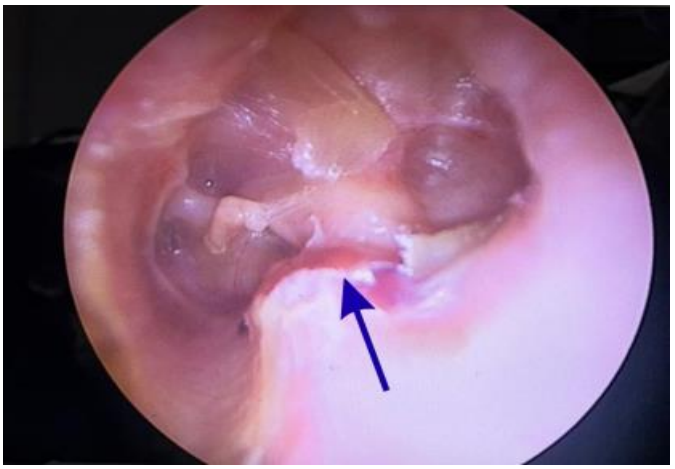


Figure 3: Pre-operative otoendoscopy of left ear canal showing widening of the external auditory canal with a

well-epithelialized, exposed and dry mastoid cavity, tympanic membrane retracted and the handle of the malleus eroded (as shown by blue arrow) The image has been taken by the authors



Figure 4: Pre-operative otoendoscopy of right ear showing widening of the external auditory canal, with the posterior canal wall communicating with a well-epithelialized, exposed and dry mastoid cavity, tympanic membrane was intact but retracted, and the handle of the malleus appeared foreshortened (as shown by red arrow) The image has been taken by the authors

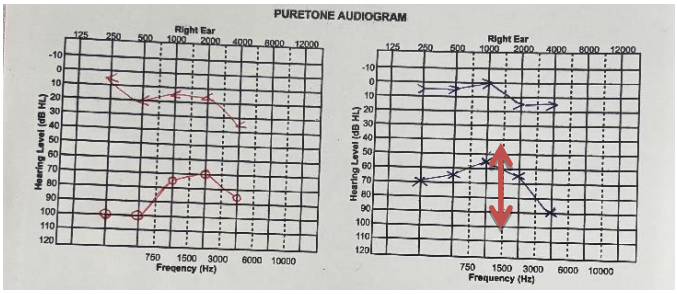
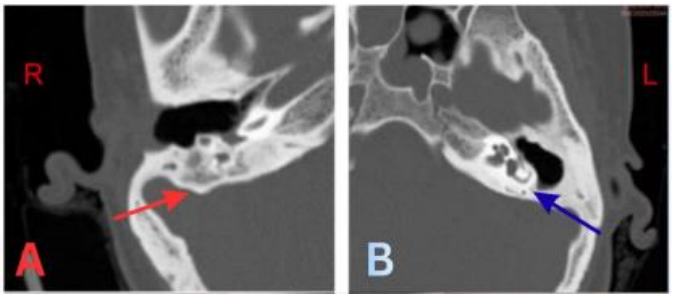


Figure 5: Image showing Pure Tone Audiometry (PTA) where red arrow indicated air bone gap of right ear and blue arrow shows air bone gap of left ear. The image has been taken by the authors



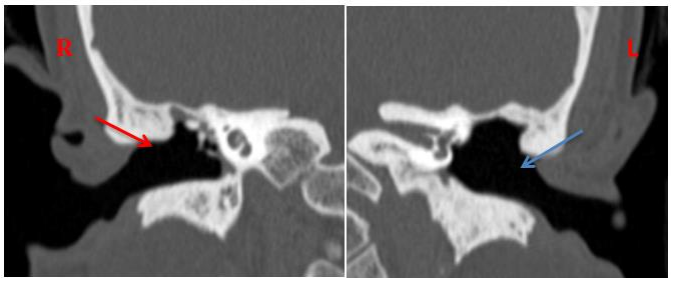


Figure 6: High-resolution computed tomography (axial & coronal sections respectively) of the temporal bone shows bilateral mastoid cavities with complete loss of the normal mastoid air-cell system and communication with the posterior wall of the external auditory canals. The ossicular chains were not visualized except for a portion of the head of the malleus on either side. The overall appearance was described as a scooped-out mastoid cavity resembling a postoperative canal wall-down mastoidectomy cavity (as shown by red and blue arrow)

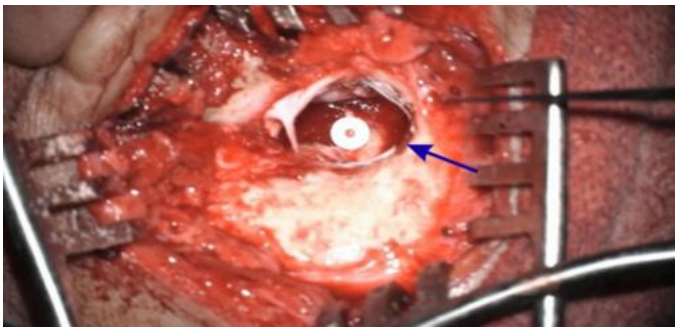


Figure 7: Intraoperative image showing placement of TORP (total ossicular replacement prosthesis) in left ear (as shown by blue arrow) The image has been taken by the authors

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Figure 8: Post-operative left ear otoendoscopy showing canal wall-down mastoidectomy with TORP (total ossicular replacement prosthesis) ossiculoplasty (as shown by blue arrow). The image has been taken by the authors

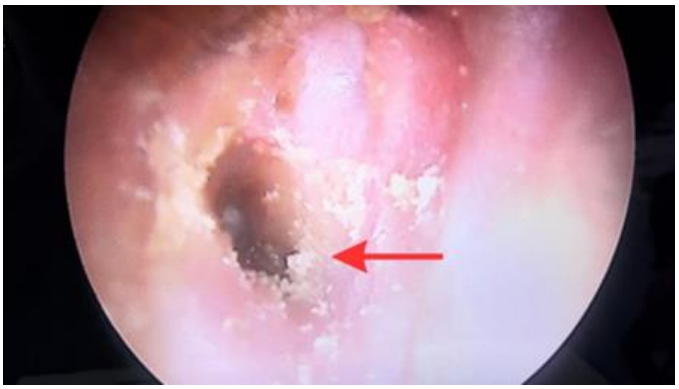


Figure 9: Post-operative right ear otoendoscopy showing canal wall-down mastoidectomy with cartilage ossiculoplasty (as shown by the red arrow) The image has been taken by the authors

Discussion

Automastoidectomy represents an uncommon endpoint of chronic destructive middle-ear disease in which the disease process itself produces spontaneous erosion and exteriorization of the mastoid cavity. The resulting anatomy may closely resemble that of a surgically created canal wall-down mastoid cavity, creating an important diagnostic challenge in patients without a history of previous ear surgery. The condition has most frequently been associated with chronic otitis media and cholesteatoma. Progressive inflammatory osteolysis and cholesteatomatous bone erosion may lead to destruction of the mastoid air cells and posterior external auditory canal. Once the disease establishes communication with the external auditory canal, spontaneous drainage of pathological material may occur, followed by epithelialization of the cavity. This process can result in apparent spontaneous stabilization of the disease[5].

Hawke and Shanker described automastoidectomy associated with keratosis obturans, demonstrating that extensive spontaneous temporal bone remodeling may occur in chronic external canal disease as well. Other reports have described automastoidectomy as a rare sequela of chronic suppurative otitis media and cholesteatoma[6].

The present case is notable because of the bilateral involvement with extensive ossicular erosion. While chronic otitis media may affect both ears, simultaneous extensive spontaneous mastoid exteriorization with marked ossicular destruction is distinctly uncommon. The patient had a history of recurrent ear discharge during childhood but presented decades later predominantly with hearing impairment. This illustrates an important feature of automastoidectomy: the extent of anatomical destruction may be disproportionate to the current clinical activity of the disease[7]. Lee et al. reported a clinical series of 22 patients with automastoidectomy caused by cholesteatoma. Their findings demonstrated extensive destruction of the attic and/or posterosuperior external auditory canal, with many patients requiring canal wall down mastoidectomy. The study emphasized that automastoidectomy may represent an advanced stage of cholesteatomatous disease despite apparently limited

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clinical symptoms[8].

In our patient, both ears demonstrated wide external auditory canals communicating with large, dry and well-epithelialized mastoid cavities. Importantly, there were no postauricular surgical scars and no history of previous ear surgery, supporting a spontaneous rather than postoperative origin[9]. HRCT played an important role in defining the extent of disease. The complete loss of mastoid air cells, communication between the mastoid cavity and external auditory canal, and near-complete ossicular destruction produced a characteristic "scooped-out" appearance. Such findings may mimic a previous canal wall-down mastoidectomy. Correlation with clinical history is therefore essential. The hearing loss in this patient was predominantly conductive, with air-bone gaps of approximately 55 dB on the right and 50 dB on the left. The near-complete absence of the ossicular chain on imaging provides a structural explanation for the large conductive component. Extensive ossicular erosion is a recognized consequence of longstanding cholesteatomatous and chronic inflammatory middle-ear disease.[10]

Management of automastoidectomy needs to be individualized. Spontaneous exteriorization may produce a dry and apparently stable cavity; however, residual disease, inaccessible areas, persistent soft tissue, or significant functional impairment may warrant surgery. In the present case, staged canal wall-down mastoidectomy was performed to provide adequate disease control and a safe, accessible cavity. Ossicular reconstruction was undertaken simultaneously as an attempt at hearing rehabilitation[11]. The choice of reconstructive technique depends on the remaining ossicular anatomy and integrity of the stapes. In the present case, cartilage ossiculoplasty was performed on the right, while a TORP was used on the left. Postoperative assessment demonstrated approximately 30 dB of functional improvement, indicating that meaningful hearing rehabilitation may still be possible despite extensive spontaneous temporal bone destruction[12].

However, reconstructive surgery cannot always restore normal hearing in such severely damaged ears. Hearing aids therefore remain an important adjunct, particularly in patients with bilateral disease. Our patient was consequently counselled for bilateral amplification following surgical stabilization[13].

The most important implication of this case is the potential preventability of such extensive disease. Recurrent childhood otorrhea should not be regarded as a trivial condition, particularly when persistent or associated with hearing impairment. Early diagnosis and appropriate management of chronic middle-ear disease may prevent progressive ossicular erosion, cholesteatoma formation and irreversible temporal bone destruction[14]. The exact mechanism responsible for the bilateral occurrence in this patient remains uncertain. Genetic susceptibility, environmental factors, chronic inflammatory responses and differences in temporal bone anatomy may potentially contribute, but the available clinical information in this case does not permit a definitive conclusion.

Conclusions

Bilateral automastoidectomy is a rare and unusual sequela of longstanding chronic middle ear disease, characterized by spontaneous destruction and exteriorization of the mastoid cavity in the absence of previous mastoid surgery. The combination of a history of chronic childhood otorrhea, absence of previous ear surgery, a dry epithelialized cavity communicating with

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the external auditory canal, extensive mastoid destruction on HRCT, and marked ossicular erosion should raise suspicion for automastoidectomy or nature's mastoidectomy. Although spontaneous exteriorization may result in clinical stabilization of the disease, patients may be left with extensive anatomical destruction and severe conductive hearing loss. Canal wall-down mastoidectomy with individualized ossicular reconstruction can provide disease control and may offer useful functional hearing improvement in selected patients. Adjunctive hearing-aid rehabilitation remains important when surgical hearing restoration is incomplete. This case emphasizes the importance of early recognition and treatment of chronic ear disease during childhood, which may prevent irreversible temporal bone destruction and long-term hearing disability.

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Isha Sahai, Unnati Watal, Divya Aggarwal, Puneet Gupta

Acquisition, analysis, or interpretation of data: Isha Sahai, Unnati Watal, Divya Aggarwal, Puneet Gupta

Drafting of the manuscript: Isha Sahai, Unnati Watal, Puneet Gupta

Critical review of the manuscript for important intellectual content: Isha Sahai, Unnati Watal, Divya Aggarwal, Puneet Gupta

Supervision: Isha Sahai, Unnati Watal, Divya Aggarwal, Puneet Gupta

Ethics Statement and Conflict of Interest Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. Northern Railway Central Hospital New Delhi issued approval nil. Institutional Ethics Committee approval was obtained/waived in accordance with the institutional policy for publication of individual case reports. Written informed consent was obtained from the patient for publication of the clinical details and images. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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