



Case Report

An latrogenic Cholesteatoma of the Squamous Part of the Temporal Bone

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A cholesteatoma is a relatively common condition within the middle ear cavity, whereas a cholesteatoma of the squamous part of the temporal bone is an exceptionally rare entity. A case of an iatrogenic cholesteatoma located in the squamous part of the temporal bone is presented, which was revealed by an intermittent purulent discharge from an opening above the right ear 20 years after retroauricular myringoplasty. The diagnosis of an iatrogenic cholesteatoma is often made after several years of evolution, sometimes even at the stage of complications. This set the importance of the otoscopic and radiological regular monitoring of patients who have undergone a surgery of the temporal bone.

KEYWORDS: Cholesteatoma, temporal bone, diffusion magnetic resonance imaging

INTRODUCTION

A cholesteatoma is a relatively common condition within the middle ear cavity $^{[1]}$, whereas a cholesteatoma of the squamous part of the temporal bone is an exceptionally rare entity $^{[2-4]}$.

CASE PRESENTATION

A 29-year-old woman who had undergone right retroauricular myringoplasty (for the repair of a tympanic membrane perforation that was causing repeated discharge) 20 years ago presented with a 2-month history of an intermittent purulent discharge from an opening above her right ear. She underwent exeresis for a suspected supra-auricular cyst. However, the early recurrence of the cutaneous purulent discharge led to imaging tests being performed.

Computed tomography (CT) revealed an 18×11-mm expansile lytic lesion in the squamous part of the right temporal bone (Figure 1a), with an extension to Macewen's triangle (also called the suprameatal triangle or mastoid fossa, Figure 1b); a 2-mm bone defect of the skull base and a 15-mm lateral bone defect of the squama were identified. Diffusion-weighted magnetic resonance imaging showed a lesion with low signal intensity on unenhanced T1-weighted images and an apparent diffusion coefficient value of 0.11 on diffusion-weighted images (Figure 1c). Otoscopy showed intact eardrums; however, another CT scan image showed a communication of 2 mm into the external auditory canal, which had been used for the surgical approach 20 years ago (Figure 1d).

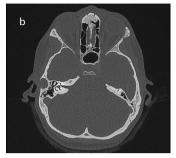
This time, the patient underwent a more extensive supra-auricular surgical approach. A cholesteatomatous lesion was identified, which caused erosion of the squamous part of the temporal bone until it exposed 2 mm of the dura. The entire lesion was excised, and the bone defect was covered with OtoMimix Bone Cement (Olympus, Center Valley, PA, USA). Subsequently, the cavity was filled with temporalis muscle. Finally, written informed consent was obtained from the patient for the publication of this paper and the accompanying images.

DISCUSSION

Depending on its origin, a cholesteatoma can be classified as congenital or acquired [5]. There are several theories to explain an acquired cholesteatoma [6], e.g., implantation, invasion, metaplasia, and invasive hyperplasia. Of these, the implantation theory explains an iatrogenic cholesteatoma. Surgical procedures in which the tympanic membrane or epithelium is trapped beneath grafted materials would promote the formation metaplasia and a cholesteatoma [7].

Although the possibility of this tumor to be a congenital cholesteatoma cannot be excluded, the presence of the tumor in Macewen's triangle, and the presence of a communication from the cholesteatoma into the external auditory canal provides evidence for this tumor to be iatrogenic. A cholesteatoma in a subject with history of same-side retroauricular myringoplasty suggests iatrogenic implantation, even when the time interval is long. It is important to take into account this potential risk when performing otologic surgical procedures.





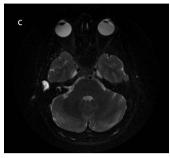




Figure 1. a-d. Image tests. (a) A coronal high-resolution computed tomography scan shows a lytic lesion in the squamous part of the right temporal bone, with a 2-mm bone defect of the skull base and a 15-mm lateral bone defect of the squama. (b) An axial high-resolution computed tomography scan showing extension to Macewen's triangle. (c) Diffusion-weighted magnetic resonance imaging shows evident diffusion restriction in the right temporal bone (hyperintensity), compatible with a cholesteatoma. (d) A coronal high-resolution computed tomography scan shows a lytic lesion in the squamous part of the right temporal bone, with a 2-mm bone defect into the external auditory canal.

Informed Consent: Written informed consent was obtained from patient who participated in this case.

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