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Case Report

A Case of Inverted Papilloma of The Mastoid Cavity After Cholesteatoma Surgery

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An inverted papilloma is a benign neoplasm that occurs exclusively in the sinonasal cavity; an inverted papilloma involving the mastoid cavity is extremely rare. We present the case of a patient with an inverted papilloma of the mastoid cavity secondary to cholesteatoma surgery. No case that occurred after acquired cholesteatoma has been reported in literature. A 39-year-old female who was diagnosed with cholesteatoma was treated with a modified radical mastoidectomy in 1988. After recurrence, the patient underwent a canal wall down mastoidectomy in 2006. Four years later, the patient complained of right ear fullness; an examination determined that the right ear canal wall was nearly obliterated by a soft tissue mass. Surgery determined that a papilloma-like mass filled the mastoid cavity. The tumor was surgically resected by revision canal wall down mastoidectomy with canaloplasty. Histologically, it was determined to be an inverted papilloma. There has been no evidence of recurrence to date.

KEYWORDS: Inverted papilloma, mastoid cavity, cholesteatoma

INTRODUCTION

An inverted papilloma is a rare benign epithelial neoplasm. These lesions commonly originate from the mucosa of the nasal cavity. It can spread from the nasal cavity to the paranasal sinuses [1]. Inverted papillomas of the mastoid cavity are classified into two types: one is a primary tumor in the mastoid cavity without any sinonasal disease and the other is a secondary lesion with concurrent or previous sinonasal papillomas [2]. It is rare for inverted papillomas to arise from the temporal bone as a primary lesion or sinonasal papilloma and invade the temporal bone. We present a new case of a patient with primary inverted papilloma of the mastoid cavity after cholesteatoma surgery.

CASE PRESENTATION

A 39-year-old female visited our otolaryngology department with the presenting complaint of right otorrhea 19 years previously. The patient had been diagnosed with cholesteatoma of the middle ear cavity and had received a modified radical mastoidectomy in the same year. Due to recurrence, she underwent canal wall down mastoidectomy 11 years ago. Four years later, the patient visited our outpatient department complaining of right ear fullness. An otoscopic examination revealed that the right ear canal was nearly obliterated by a soft tissue mass (Figure 1). Pure tone audiometry showed total hearing loss with residual hearing of the lower frequencies in the right ear (Figure 2).

Preoperative computed tomographic scans revealed that the patient's right ear was in a post-mastoidectomy state and showed haziness in the right external auditory canal and right mastoid cavity (Figure 3).

Revision canal wall down mastoidectomy with canaloplasty was performed. Intra-operatively, the mastoid cavity that had received surgery was found to be filled with a papilloma-like mass. The mass was completely removed, and a biopsy was performed. The middle ear cavity was clear. Several irrigation procedures were performed to remove residues of bone dust and remnant tissue from the mastoid cavity. External auditory canal stenosis persisted even after removing the mass, and canaloplasty was performed. Histopathological examination revealed an inverted papilloma with chronic inflammation of the mastoid cavity, with no evidence of malignancy (Figure 4).

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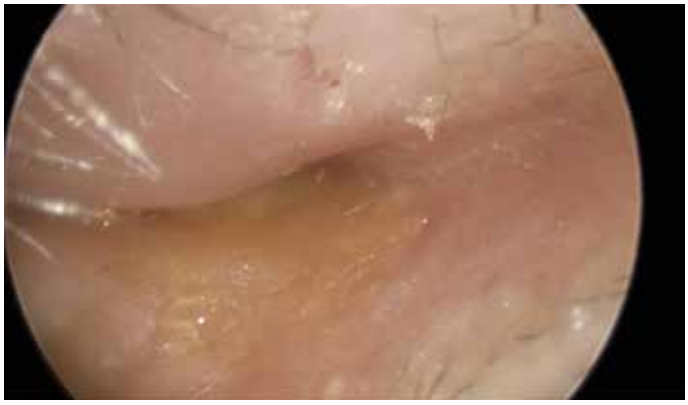

Figure 1. Local finding shows obliteration of the external auditory canal

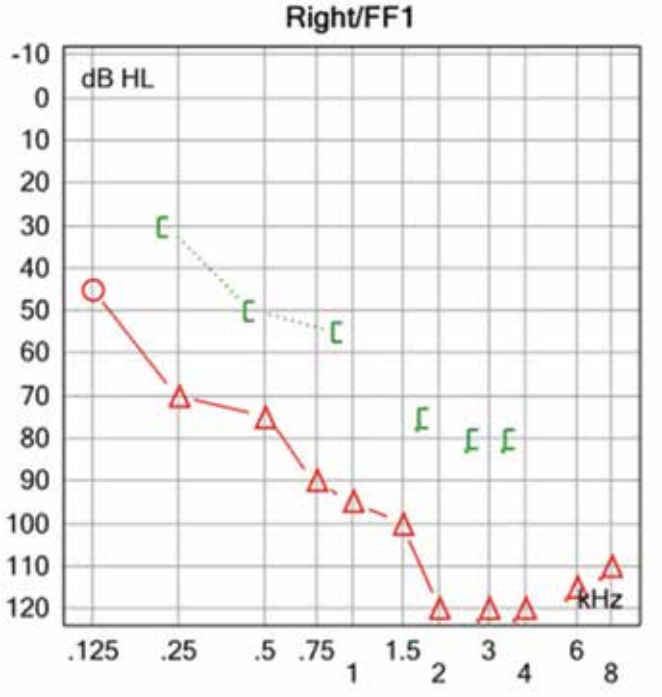

Figure 2. Preoperative pure tone audiometry shows mixed deafness of the right ear Preoperative computed tomographic (CT) scans revealed that the patient's right ear was in a post-mastoidectomy state and showed haziness in the right external auditory canal and right mastoid cavity

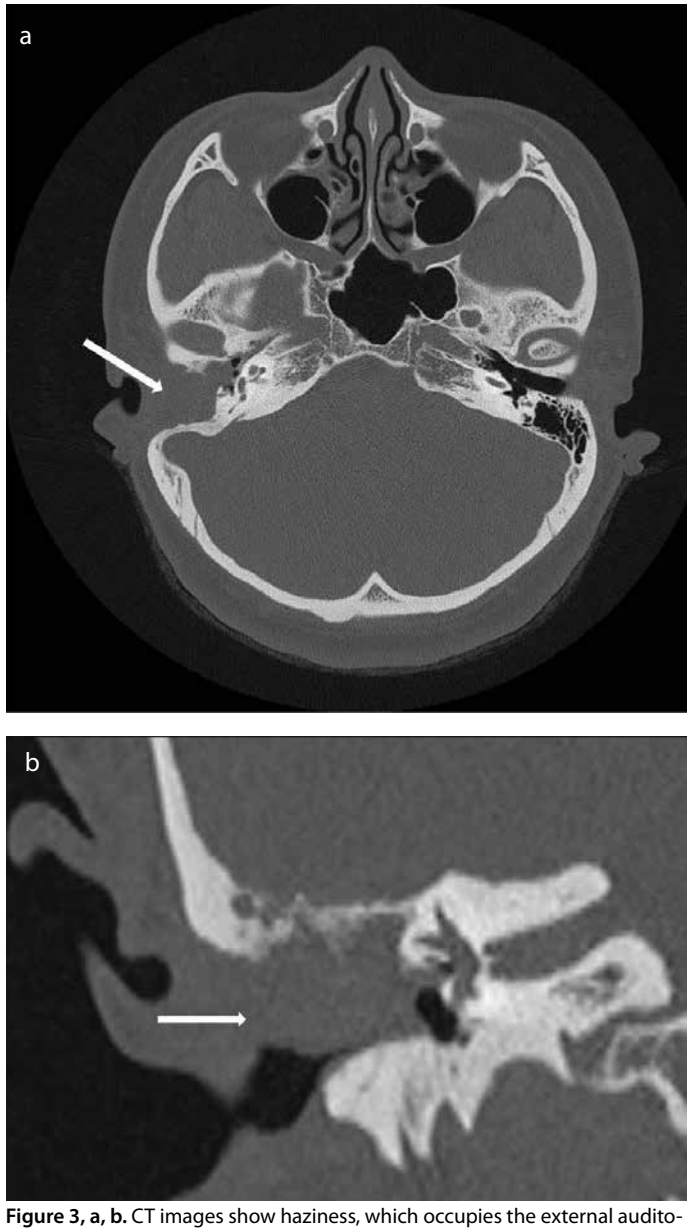

Figure 3, a, b. CT images show haziness, which occupies the external auditory canal and mastoid cavity (arrow), without a mass in the paranasal cavities. Axial view of temporal CT (A); coronal view of temporal CT. (B).

There were no symptoms of ear fullness, although mixed hearing loss persisted after surgery. No signs of recurrence were observed until the 9-month follow-up.

This case report is presented with consent from the patient.

DISCUSSION

Papillomas are classified into three types: inverted, columnar cell, and exophytic papillomas [3]. According to the World Health Organization, an inverted papilloma is derived from the Schneiderian membrane. In 1854, Schneiderian-type papillomas were first described and named by C. Victor Schneider [2]. Inverted papillomas generally arise in the paranasal sinuses and are associated with human papilloma virus infection in approximately 30% of cases. Malignant transformation is estimated to be 7%, and the recurrence is approximately 15% [3].

Middle ear and mastoid cavity involvement of inverted papillomas was first described by Stone et al. [4] in 1987. The middle ear and mastoid cavity as the primary involved sites is extremely rare. To date, less than 30 cases of inverted papilloma arising primarily from the middle ear and mastoid cavity have been published [3]. The origin of an inverted papilloma of the middle ear and mastoid is controversial; three hypotheses have been proposed [3]. The first hypothesis is the origin of the inverted papilloma by the migration of paranasal sinus-inverted papilloma cells through the Eustachian tube. The second hypothesis is that of migration abnormality of the ectopic Schneiderian membrane into the middle ear mucosa. The last hypothesis is that the inflammatory cells of chronic otitis media stimulate the development of Schneiderian mucosa. In our case, inverted papilloma of the mastoid cavity occurred after surgery for acquired cholesteatoma. It is possible that chronic inflammation after surgery was converted to the inverted papilloma. Our case matches the third

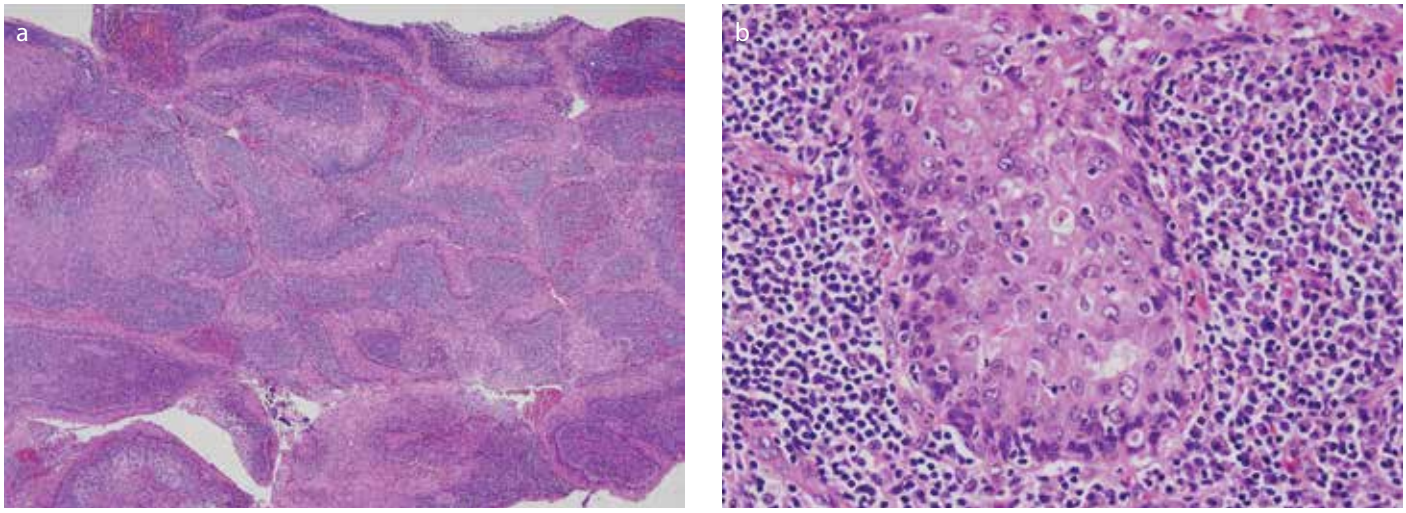

Figure 4, a, b. Lesion is composed of multiple layers of epithelial cells (hematoxylin and eosin stain; original magnification, 40×) (A); lesion as visualized under high-power microscopy (hematoxylin and eosin; original magnification, 400×) (B).

hypothesis. Previously, a case of congenital cholesteatoma, developed as an inverted papilloma, has been reported [5].

Surgery is primarily used for the management of inverted papilloma of the middle ear and mastoid cavity. Inadequate resection is associated with higher rates of recurrence. Nathan et al. [6] reported at least one recurrence in 10 of 21 cases (48%). Recurrence was reported in all three cases (100%) initially treated with tympanoplasty and simple excision, compared with 39% following more aggressive surgery, such as mastoidectomy or temporal bone resection [6].

Long-term follow-up is integral in all patients with inverted papilloma of the middle ear and mastoid cavity because of the possibility of recurrence and the high rate of malignant transformation [7]. Detailed clinical observations during outpatient visits are important to reduce the recurrence and malignant transformation. In addition to clinical examination, magnetic resonance imaging should be performed during the follow-up [8].

Postoperative radiotherapy is not routinely performed because of the risk of osteoradionecrosis. Nonetheless, postoperative radiotherapy should be considered in cases in which malignant disease foci are present in inverted papillomas. Postoperative radiotherapy may also be performed when the tumor cannot be completely removed or when it has relapsed several times, even in the absence of malignancy [9].

Papilloma of the mastoid cavity is an extremely rare disease, and its clinical characteristics remain unknown. However, Because of the high recurrence rate and the possibility of a change to cancer, thorough outpatient follow-up is required.

CONCLUSIONS

Our case differs from other cases in that inverted papilloma of the mastoid cavity occurred after cholesteatoma surgery. In most cases, after cholesteatoma surgery, the patient is seen due to the possibility of cholesteatoma recurrence. However, because inverted papilloma may occur rarely, as in the case described here, it should be considered among the various diseases if recurrence is suspected during follow-up.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

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