CASE REPORTS

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Coexisting Parotid Cholesteatoma and Temporal Bone Carcinoma: A Case Report

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ABSTRACT

Objective: To report a case of parotid cholesteatoma and concurrent squamous cell carcinoma of the temporal bone in a 51-year-old woman.

Methods:

Design: Case Report
Setting: Tertiary Government Training Hospital
Patient: One

Results: A 51-year-old woman with a left parotid mass and ipsilateral external auditory canal mass and chronic bilateral otorrhea had parotid biopsy histological features of cholesteatoma and temporal bone imaging suggestive of middle ear cholesteatoma and possible parotid region abscess. She underwent subtotal petrosectomy, mastoid obliteration and excision of the parotid mass. Histopathological examination of the parotid mass was consistent with cholesteatoma but tissue from the middle ear cavity showed well-differentiated squamous cell carcinoma.

Conclusion: While cholesteatomas typically arise within the temporal bone, they may occasionally present in atypical or distant sites. In cases where a cholesteatoma demonstrates an unusual location, aggressive behavior, or atypical clinical progression, a high index of suspicion should be maintained for the possibility of an underlying or coexisting temporal bone squamous cell carcinoma.

Keywords: *parotid diseases; cholesteatoma; cancer of ear; squamous cell carcinoma of head and neck*

A cholesteatoma is composed of keratinizing squamous epithelium and keratin debris typically arising within the temporal bone.[1] Although it is a non-neoplastic lesion, it is well known for its locally aggressive behavior, often resulting in serious complications such as meningitis and both intra- and extracranial abscesses.[2] The pathogenesis of such complications can be partially attributed to the directional growth of the cholesteatoma from the middle ear cavity, which

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facilitates erosion of adjacent bony and soft tissue structures.[3] There are limited reports describing cholesteatomas located at a distance from the mastoid region -- referred to as extra-mastoidal cholesteatomas[4-10] -- as well as cases in which cholesteatomas coexist with temporal bone malignancies.[11-17] Both presentations are rare and represent atypical manifestations of otherwise well-characterized disease processes. We present a case of cholesteatoma in the parotid with coexisting squamous cell carcinoma of the middle ear.

CASE REPORT

A 51-year-old woman presented with a slow-growing, painful swelling in the left parotid region that had been present for approximately one year. She also reported a two-year history of recurrent bilateral ear discharge with otalgia, intermittently relieved by antibiotic ear drops and oral analgesics prescribed by her physician. On physical examination, there was a well-circumscribed, tender, erythematous mass in the left parotid region, with ipsilateral House-Brackmann Grade III hemifacial paresis. *(Figure 1)* Otoscopy revealed a perforated tympanic membrane on the right and a polypoid mass occupying the left external auditory canal (EAC). Tuning fork tests demonstrated findings consistent with conductive hearing loss on the right and sensorineural hearing loss on the left. The remainder of the neurologic examination was unremarkable. No cervical lymphadenopathy was palpated bilaterally.

Computed tomography *(Figure 2)* and magnetic resonance imaging scans *(Figure 3)* of the temporal bone showed multiple heterogenous cystic nodules with an enhancing wall in the left infra-auricular region with the largest measuring 1.5 x 1.5 cm. A soft tissue attenuating fluid collection was observed in the left EAC with widening and erosion of the surrounding bone in the middle ear, suggestive of a middle ear cholesteatoma and possible abscess formation in the left parotid region. A biopsy was performed on the left parotid mass, and histopathological analysis confirmed the presence of a cholesteatoma.

With a working diagnosis of bilateral chronic suppurative otitis media with cholesteatoma in the left middle ear cavity and extramastoidal extension into the left parotid space, the patient underwent a subtotal petrosectomy with blind sac closure of the left EAC and mastoid obliteration using an inferiorly based musculoperiosteal flap and temporalis muscle flap. The left parotid mass was excised along with nonviable skin, and the defect was reconstructed using a rotation advancement flap. *(Figure 4)* Intraoperatively, a polypoid, friable mass was identified occupying the EAC ostium involving both its cartilaginous and bony segments. *(Figure 5A)* This mass extended into the middle ear cavity and was

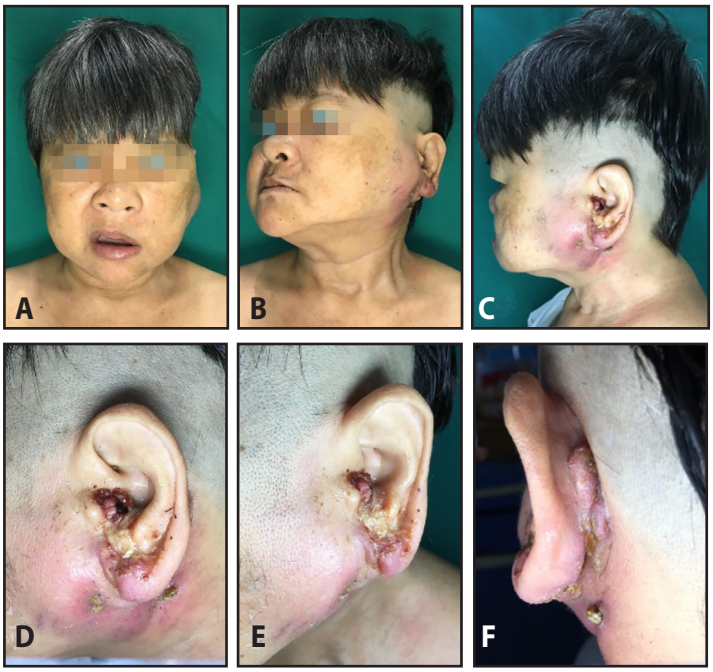

Figure 1. The profile of the patient in **A.** frontal, **B.** oblique, and **C.** lateral views showing a shallow left nasolabial fold and left parotid mass; Close up images of the left ear and parotid area in **D.** lateral, **E.** oblique and **F.** posterior views showing the circumscribed, erythematous parotid mass and ear discharge. Photos published with permission.

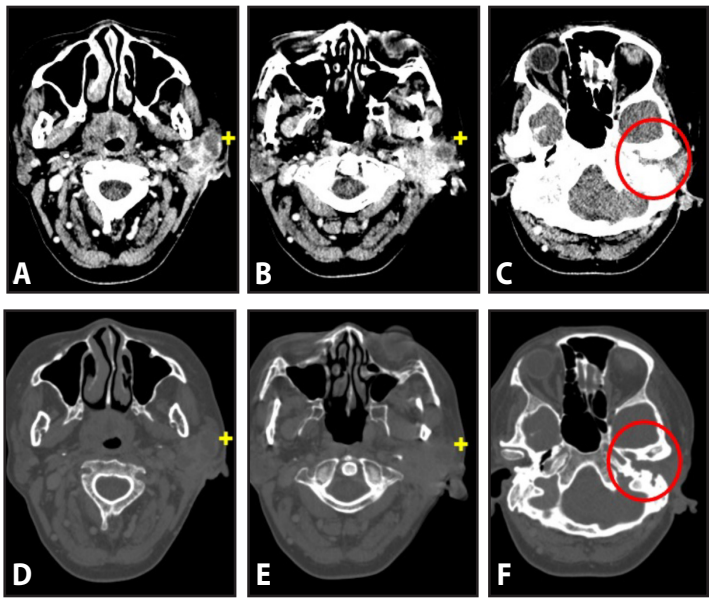

Figure 2. Computed Tomography Scans, axial high resolution: **A.-C.**, soft tissue and **D.-F.,** bone windows revealing a multiple heterogenous cystic nodule with enhancing wall seen in the left infra-auricular region (+) and soft tissue attenuating fluid collection seen in the left internal auditory canal with associated widening and erosion of the surrounding bone in the middle ear. (encircled)

contiguous with the parotid mass via a fibrous stalk, which passed through the dehiscent bony floor of the EAC. *(Figure 5B)* Upon widening of the mastoid bowl, white, caseous material encased in a sac—consistent with cholesteatoma—was encountered. *(Figure 5C,*

D) The vertical segment of the facial nerve was encased in the mass. *(Figure 5E, F)* The excised left parotid mass was carefully dissected along with the previously identified fibrous stalk.

Specimens from the external ear canal, middle ear, and parotid region were sent for histopathological examination. On cut section, the parotid mass revealed a thick, white, non-foul-smelling caseous material encased by a thick wall with areas of necrosis. *(Figure 6A, B)* The tissue samples from the parotid region confirmed the presence of a cholesteatoma. *(Figure 7A, B)* Histopathological analysis of the external and middle ear specimens showed anastomosing sheets of squamous cells diffusely infiltrating the surrounding fibrous tissue with significant keratin pearl formation, consistent with well-differentiated squamous cell carcinoma. *(Figure 7C, D)*

Using the Pittsburgh TNM staging system as modified by Moody et al.[18] and Ariaga *et al.*,[19] we classified the tumor as Stage IV (T4N0M0). This staging was based on the extensive soft tissue involvement, osseous destruction, and facial paresis, with no evidence of regional lymph node involvement or distant metastases.

The patient was advised definitive treatment including lateral temporal bone resection and post-operative radiotherapy. However, she and her family made an informed decision to refuse further management.

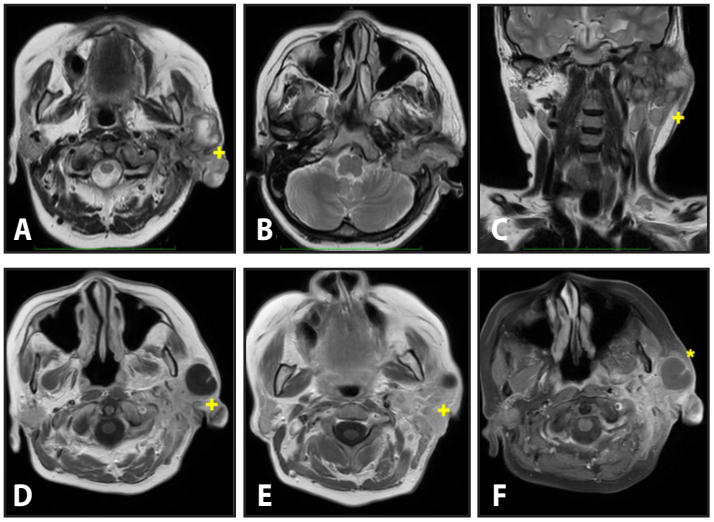

Figure 3. Magnetic Resonance Imaging; **A., B., D., E., F.,** axial and **C.**, coronal sections showing areas of hyperintensity signal on DWI in the left mastoid region with **D.-F.**, T1 hypo intensity and **A.-C.**, mild T2 hyperintensity signals and enhancement on post contrast administration. (*) Multiple, heterogenous fluid collections are seen in the left infraauricular region, with the largest measuring 2.7 x 2.2 x 2.0 cm. (+) These exhibit T1WI hypointense signal, T2/DWI hyperintense signal and rim enhancement post contrast administration.

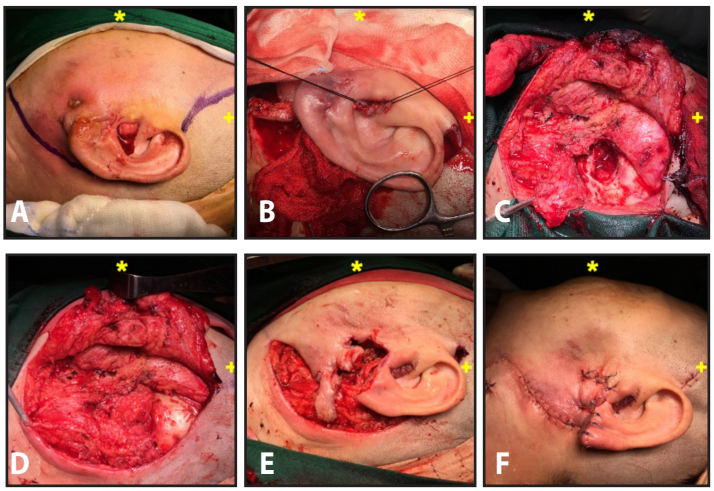

Figure 4. Intraoperative images showing **A.** the incision, **B.** blind sac closure, and **C.** mastoid bowl before obliteration with **D.** temporalis muscle flap; the **E.** skin defect is also shown after excision, and **F.** reconstruction. (* anterior, + posterior)

DISCUSSION

Cholesteatomas located outside of the mastoid cavity have been reported in the literature, though they are few. These extramastoidal cholesteatomas have been documented in various locations, including the upper neck,[20] infratemporal fossa,[8] maxillary sinus,[21] and the parotid space.[4,6,7,10,22] Previous reports of parotid cholesteatomas often describe a history of chronic ear discharge and prior ear surgery for cholesteatoma ipsilateral to the parotid growth, hypothesizing that extramastoidal cholesteatomas could be iatrogenic, resulting from surgical implantation.[4,6,7,10,22] However, in our present case, the patient had no history of ear surgery, suggesting an alternative mechanism for migration of the cholesteatoma.

On physical examination, all previously reported cases presented with masses in the parotid region and an ipsilateral pathologic ear—findings that were also observed in our present case.[4,6,7,10,20,22] However, not all cases exhibited associated symptoms such as pain, facial paresis or vestibular disturbances.[4,6,7,10,22]

More recent studies have utilized MRI or CT scans as diagnostic modalities in cases of extramastoidal cholesteatomas. These reports identified lesions in the parotid region and ipsilateral temporal bone.[6,7,10,22] A report by Tornari *et al.* noted a breach in the mastoid bone wall through the fissures of Santorini.[22] Similarly, our patient had a dehiscent inferior wall of the bony EAC with a contiguous lesion extending from the EAC to the parotid region. This finding was evident on preoperative imaging and confirmed intraoperatively. We hypothesize that this dehiscence provided the pathway through which the mastoid cholesteatoma extended into the parotid region. In contrast, Tovi *et al.* proposed a different mechanism, suggesting that residual cholesteatoma may migrate along the facial nerve through the stylomastoid foramen, eventually reaching the parotid gland.[8]

Due to the unusual presentation of our patient, we performed a biopsy to determine the etiology of the parotid mass. This led to the

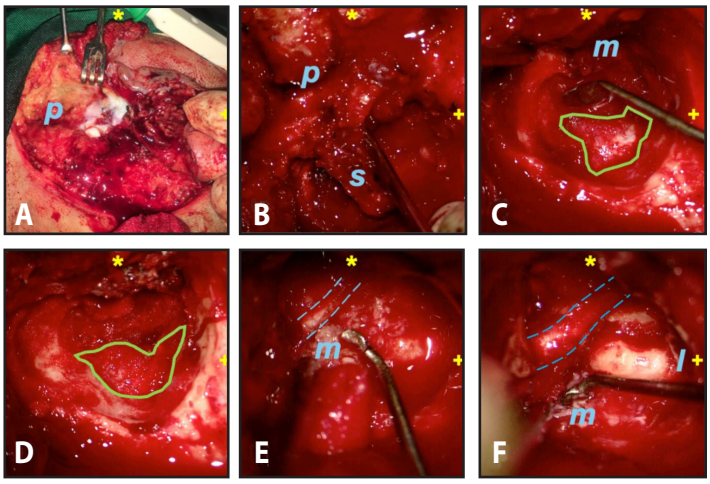

Figure 5. Intraoperative photos showing: **A.** caseous material from the parotid mass (p) upon dissection; **B.** dehiscence of bone, external bony canal with a fibrous stalk (s) connecting the middle ear mass (m) and parotid mass (p); **C., D.,** cholesteatoma (solid line) occupying the middle ear cavity was identified during mastoid sequestration; **E., F.,** Vertical segment of the facial nerve (broken lines) enveloped by the middle ear mass (m) as it courses inferiorly; the lateral semicircular canal is also seen (l). (* anterior, + posterior)

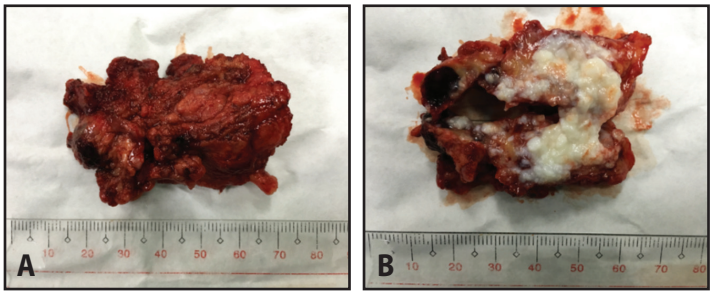

Figure 6. The excised parotid mass: **A.** gross specimen; and **B.** cut section

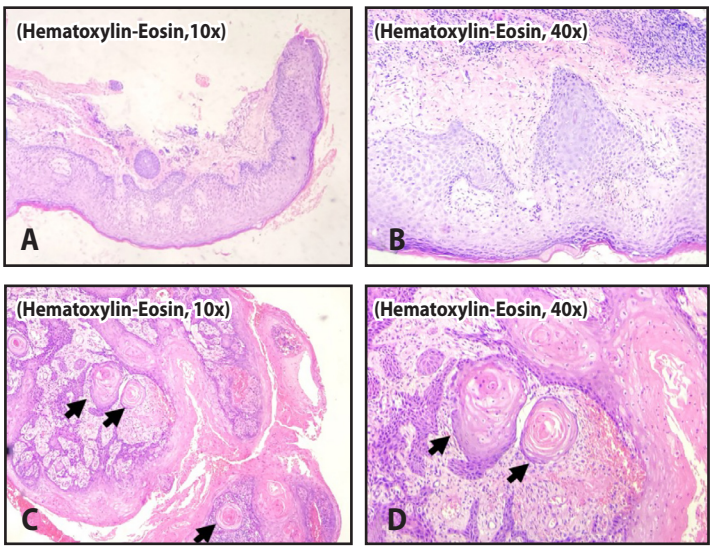

Figure 7. Hematoxylin/eosin-stained slide of cholesteatoma of the parotid at **A.** low power (10x magnification) and **B.** high power objective (40x magnification); Hematoxylin/eosin-stained slide of **C.** external and middle ear mass at low power (10x magnification) and **D.** high power objective (40x magnification) showing well-differentiated squamous cell carcinoma; note keratin pearls (black arrows).

diagnosis of a parotid cholesteatoma which guided our decision to proceed with excision. Our approach differs from previous reports in the literature where a definitive histopathologic diagnosis was only made after surgical excision of the parotid mass.[4,6,7,10,22] In all reported cases, including ours, excision of the parotid cholesteatoma was performed during mastoid exploration. To the best of our knowledge, there have been no reported recurrences of parotid cholesteatomas following primary excision in the literature.[4,6,7,10,22]

What about the temporal bone carcinoma? Primary malignant tumors of the temporal bone are rare, with an estimated incidence of 6 per 100,000 population representing less than 0.2% of head and neck carcinomas with squamous cell carcinomas accounting for 60-80% of cases.[23-25] However, the disease is still associated with significant morbidity and mortality that continues to evade current advances in its diagnosis and management.

Temporal bone tumors typically present with nonspecific signs and symptoms affecting hearing and balance, often leading to delays in diagnosis.[26] The most common symptoms in decreasing order of incidence include otorrhea, otalgia, hypoacusis, facial nerve palsy, preauricular swelling and the presence of a mass in the EAC.[24, 25, 27] These symptoms can also be seen in patients with chronic ear infections and cholesteatomas.[3] Reported cases of TBSCC and coexisting cholesteatoma, including our own, have presented with similar equivocal clinical features[11-17] which can often lead to misdiagnoses and subsequent delays in treatment.[28]

Given the nonspecific nature of symptoms and potential inconsistencies on otoscopic examination, imaging modalities such as MRI and CT and tissue biopsies are critical for accurate diagnosis of temporal bone malignancies.[25,26] We performed both CT and MRI scans which revealed lytic lesions in the temporal bone. Chee *et al.* identified such lytic bony lesions as radiologic red flags for malignancy.[24] Nonetheless, a definitive diagnosis required histopathological confirmation and in our patient's case—as with other cases reported in the literature—this confirmation was only achieved following surgical intervention[11,15,17]

Conventional treatment modalities for temporal bone malignancies include surgical resection with or without chemotherapy and/or radiotherapy with varying results. Generally, patients with early stages of disease fared better than those with advanced stages.[29]

The coexistence of cholesteatoma and malignancy is a rare occurrence, leading several authors to speculate that cholesteatoma could potentially serve as a premalignant lesion for temporal bone carcinoma.[11-17] Research has identified mediators involved in the osteolytic activity seen in cholesteatoma,[30] which may play a role in the neoplastic process. Notably, matrix metalloproteinases have

been identified in both cholesteatoma and squamous cell carcinoma tissues. Other biomarkers examined in these specimens include the activation of c-myc, angiogenesis, tumor necrosis factor-alpha, as well as an increase in the expression of p63, p53 tumor suppressor genes,[31, 32] and telomerase activity.[23] These factors have been associated with hyperproliferation and/or the survival of neoplastic cells.

Our case highlights two distinct and unusual presentations of cholesteatomas, underlining the necessity for careful evaluation and independent consideration of each presentation. Physicians should be alert to the variations in presentation, as observed in these cases, as they may help in identifying similar situations in future patients. For parotid cholesteatomas, a history of a chronically discharging ear with the development of new tissue growth in the ipsilateral parotid area suggests the possibility of extra mastoidal migration of cholesteatoma. On the other hand, the presence of an EAC mass in the context of a chronically discharging ear, especially when imaging studies reveal extensive bone and soft tissue involvement, should heighten suspicion for malignancy.

In conclusion, while cholesteatomas typically arise within the temporal bone, they may occasionally present in atypical or distant sites. In cases where a cholesteatoma demonstrates an unusual location, aggressive behavior, or atypical clinical progression, a high index of suspicion should be maintained for the possibility of an underlying or coexisting temporal bone squamous cell carcinoma.

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