CASE REPORT
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Salivary Gland Choristoma of the Middle Ear: Case Report

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ABSTRACT

Choristoma, a salivary gland tumor, may localize at unforeseen locations. Since 1961 when it was first described, roughly 50 cases of choristoma have been accounted for, including 30 cases among pediatric–adolescents. The patient in this current study is a girl of 14 years of age. She was admitted to a tertiary otolaryngology polyclinic with right-sided hearing problems and aural fullness complaint. Her average airway hearing level measured using the pure tone audiometry hearing test was 70 dB, whereas the bone conduction pure tone average was 6 dB. A high-resolution computerized tomography of the temporal bone demonstrated a mass behind the intact tympanic membrane. The tumor was excised entirely over the facial nerves tympanic part of the facial nerve with careful dissection. Histopathological examination revealed the tumor to be a salivary gland choristoma. In this article, we present the case of a 14-year-old girl with unilateral conductive hearing loss caused by salivary gland choristoma.

Keywords: Choristoma, conductive hearing loss, middle ear

INTRODUCTION

Choristoma is the development of mature tissue at an unexpected location. A choristoma may develop in various parts of the body. A salivary gland choristoma located in the middle ear is unusual (1). Since it was first described in 1961, only 30 pediatric–adolescent cases have been reported (2, 3).

Salivary gland choristomas usually occur behind the healthy tympanic membrane, and are associated with unilateral conductive hearing loss. Furthermore, comorbid facial nerve anomalies, and other anomalies, including Mondini dysplasia, alopecia, preauricular pit, ear tag, and situs inversus, have also been reported (4, 5). The present study aimed to present the surgical, clinical, and radiological findings of a case, which is predominantly observed in the pediatric and adolescent age group, in the light of the literature.

Here we describe a case of salivary gland choristoma along with associated surgical, clinical, histological, and radiographic findings. Ethics committee approval was not obtained for this case report. Before the operation, the patient's parents were informed, and a consent form was signed.

CASE REPORT

The patient, a 14-year-old female, presented to a tertiary otolaryngology polyclinic complaining of hearing loss and aural fullness in the right ear. Her mother stated that the hearing loss had persisted for a long time and that she did not have any other otologic complaints, such as ear discharge, dizziness, or ear pain. The patient's family, pregnancy, and delivery histories were unremarkable. The right eardrum was intact during the otologic examination, but a reddish-brown mass was detected behind the posterior upper quadrant.

The air-bone conduction average (PTA) was 70 dB Hearing Level (HL), the bone conduction PTA was 6 dB HL, and the speech discrimination score was normal in the right ear audiometric examinations. The hearing levels were within normal limits in

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the left ear. A type-A curve was obtained in the tympanometric study.

According to high-resolution computed tomography (CT) of the temporal bone, a mass was observed behind the tympanic membrane, which was associated with the tympanic part of the seventh nerve and was in the region consistent with the facial recess, the border of which could not be distinguished (Figure 1).

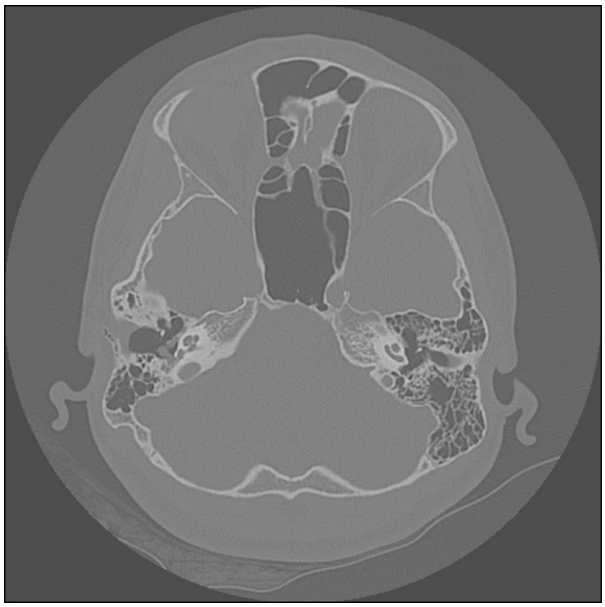

Figure 1: Image of the axial section of the temporal bone and the middle ear mass

Upon the preliminary diagnosis of congenital cholesteatoma or facial nerve neuroma, the patient underwent explorative tympanotomy via an end–aural approach under general anesthesia. The mass was 0.6 cm in diameter and reddish-brown in appearance (Figure 2). The mass was excised entirely over the tympanic part of the 7. nerve with cautious and careful dissection, without inducing any facial nerve stimulation, under

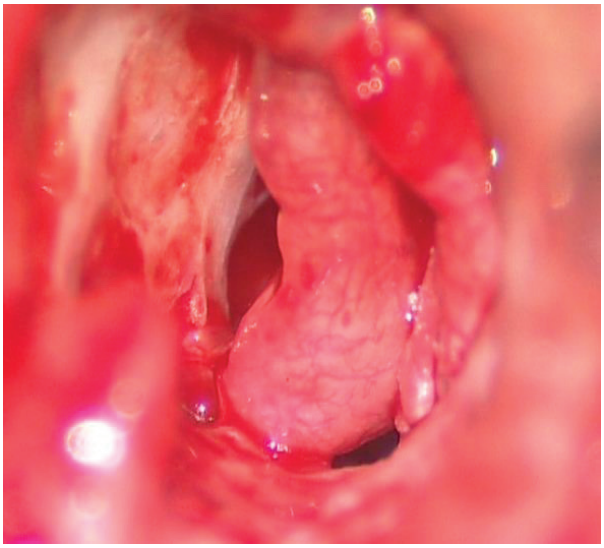

Figure 2: Preoperative image

continuous facial nerve monitoring, and the excised tissue (hereafter, specimen) was sent for further analysis.

The ossicular chain was intact, there was no erosion, and the ossicles were mobile, as observed during the middle ear examination after removal of the tumor.

Facial nerve motor functions were normal during the postoperative period. The pure tone threshold audiograms of the patient with persistent hearing loss at high frequencies in the right ear were performed at the preoperative and postoperative periods and shown in Figure 3.

Based on histopathological examination, the mass was determined to be a salivary gland choristoma, which indicated salivary gland acinar structures in the tissue stroma covered with pseudostratified cylindrical epithelium (Figure 4).

One year after the patient's surgery, the tympanic membrane was intact, and no recurrence was observed as confirmed by postoperative temporal bone magnetic resonance imaging

a

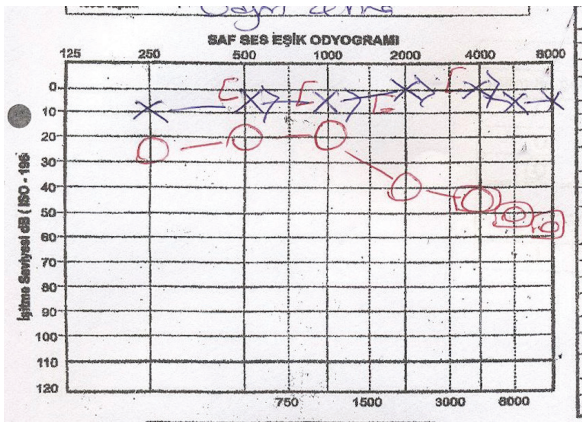

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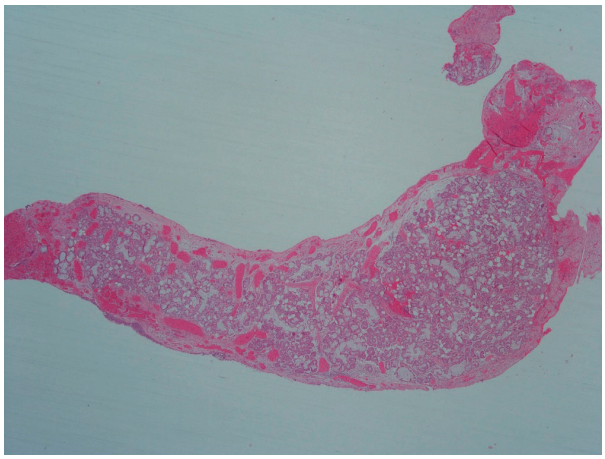
Figure 3: a,b Preoperative (a) Postoperative 2. Month (b)

a



a

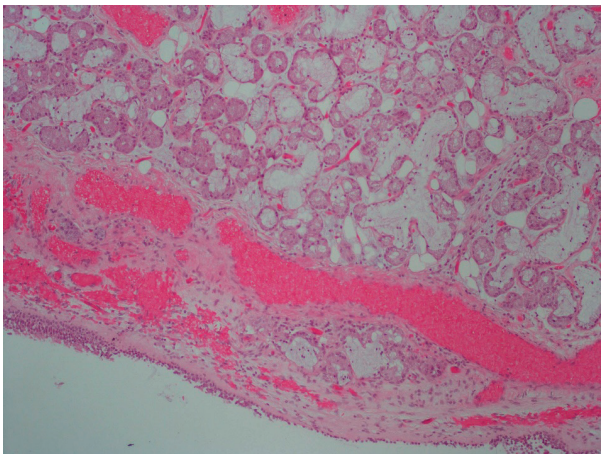


Figure 4: a, b.Polypoid tissue with acinar structures of the salivary gland (H&E×20) (a); Salivary gland acinar structures in the tissue stroma covered with pseudostratified cylindrical epithelium (H&E ×100) (b)

(MRI) (Figure 5); nevertheless, conductive hearing loss at high frequencies continued in the right ear.

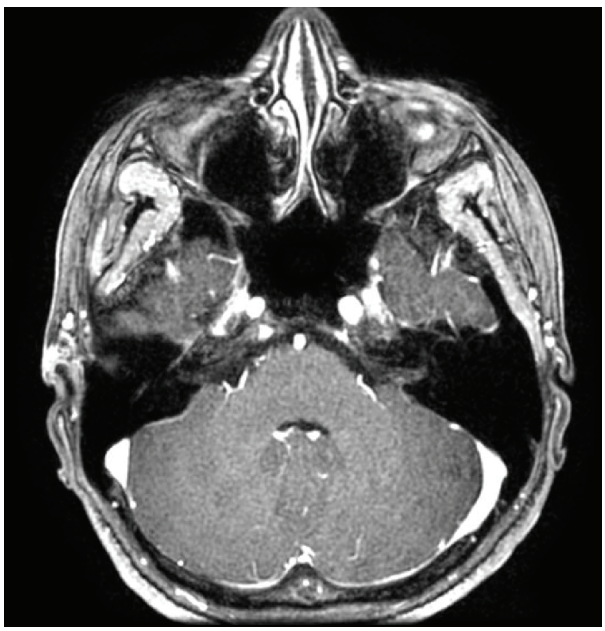


Figure 5: Axial T1-weighted MR images Temporal bone

DISCUSSION

A choristoma is the development of histologically normal mature tissue in an unexpected region. As a well-defined example of heterotopic salivary glands, choristomas are prevalent in the ear-nose-throat region but are rarely located in the middle ear (6, 7). The exact cause of this anomaly remains unknown.

Cases of choristoma observed in the ear-nose-throat region have been reported (8). Only 30 pediatric–adolescent cases have been reported since 1961 when Taylor and Martin first described the condition. (2). Approximately 50 cases have been reported, including 30 cases in the pediatric–adolescent age group (3).

The mechanism of salivary gland tissue development in the middle ear remains to be elucidated. Relevant literature suggests that salivary gland tissue that has been compressed during the process fusion of the temporal bone and remained in the middle ear, tissues without sufficient embryological resolution, and the second branchial arch defects that develop before the 4th intrauterine month may account for the condition (4).

Left ear involvement is more frequent in salivary gland choristomas and more prevalent in children and young adults (10 months – 52 years) (8). Varnetta et al. (9). reported that sensorineural hearing loss following labyrinthitis development is a choristoma case with a round window anomaly.

Furthermore, choristomas were reported with comorbidities, such as short cochleas, inner ear anomalies, such as Mondini dysplasia, and other abnormalities, including temporal alopecia, conchal bands, facial asymmetry, branchial cyst, and situs inversus totalis (4, 10, 11). There was no additional anomaly in the present case.

Relevant studies in the literature reported choristoma cases in the middle ear with Branchiootorenal syndrome induced by an autosomal dominant inheritance due to a mutation in the 8th chromosome (12). The family tree of our case was unremarkable.

Characteristically, a choristoma is a benign and slowly growing lesion. CT renders better results in detecting small masses in the middle ear. CT provides a better view, especially of the ossicular system structures, and allows the detection of small erosions and dislocations (13). CT cross-sectional images facilitate the differential diagnosis of choristomas from other benign middle-ear masses, including hamartoma, teratoma, dermoid cyst, epidermoid cyst, and congenital cholesteatoma

(14). The present case was followed up using MRI during the postoperative period, although CT was the preferred procedure during the preoperative period.

The nature of the surgical operation varies depending on location of the mass, the size and the erosion it creates in the ossicular system (15). Total excision of the mass by means of explorative tympanotomy is usually sufficient. Nevertheless, in rare cases, mastoidectomy, canal wall-up mastoidectomy, and ossiculoplasty may be required (3). In the present case, mastoidectomy and ossiculoplasty were not preferred as there was no destruction or dislocation of the ossicular chain.

In addition, the risk of iatrogenic seventh nerve injury is 12% during these types of surgical operations (4). Therefore, closeness to the facial nerve should be considered during the procedure, and accordingly, an intraoperative facial nerve stimulator should be used, and electrocauterization of the mass should be avoided (3). The postoperative motor functions of the facial nerve were normal.

Choristomas in the middle ear may cause erosions on the ossicular system due to the mass effect. It was reported that ossicular system reconstructions through primary and secondary surgeries significantly benefit the patient's hearing function (8). In the present case, no erosion was observed in the preoperative ossicular system, and therefore, no reconstruction was performed. However, a second look surgery intended for conductive hearing loss was recommended to the parents, and we decided to follow the patient closely.

A previous study reported that preoperative potassium titanium phosphate (KTP) laser had facilitated the excision of a mass without inducing any damage to the ossicular system. KTP laser option was not available in our clinic; therefore, it was not used and successful excision was achieved using classical otologic surgical instruments (16).

CONCLUSION

In the middle ear, salivary gland choristomas are very rare. The location of the mass behind the intact eardrum should be taken into consideration in the initial diagnosis, especially its proximity to the facial nerve, and its association with comorbidities, such as erosion, anomalies, and syndromes, in the ossicular system should be kept in mind in patients presenting with unilateral hearing loss.

Informed Consent: Written informed consent was obtained.

Peer Review: Externally peer-reviewed.

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